

Exploration of the artificial eye process in children with Retinoblastoma: addressing the psychological impact and potential for technological advancement.

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Submitted in partial fulfilment of a PhD to be awarded by Bournemouth University

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## **AUTHOR'S DECLARATION**

This thesis is submitted in partial fulfilment of the requirements for a Doctor of Philosophy degree at Bournemouth University. I declare that this is my own work that meets the academic regulations held by the University. All work of others has been fully referenced and acknowledged throughout the thesis.

Parts of the thesis have been published in journals, books and as posters for conferences. A full list of the publications can be found in Appendix A.

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## ABSTRACT

**Authors name:** Holly Chinnery

**PhD Title:** Exploration of the artificial eye process in children with retinoblastoma: addressing the psychological impact and potential for technological advancement.

**Background:** Retinoblastoma (Rb) is the result of genes becoming mutated and can be hereditary (predominately unilateral and unifocal) or non-hereditary (predominately bilateral and multifocal). 70% of unilateral Rb requires enucleation and thus a lifetime supply of artificial eyes.

**Aim:** To explore the artificial eye process in children with a diagnosis of Rb to address the psychological impact and potential technological improvements that can be made to the current process.

**Methods:** A qualitative approach was used consisting of one study and three components. Firstly, a qualitative questionnaire of artificial eye prosthetists (AEP) perspective of the process. Secondly, an Interpretative Phenomenological Study (IPA) to understand the lived experience of the fitting process of artificial eyes in 13-16 year olds and parents of children with a diagnosis of Rb. Thirdly, a transfer of knowledge between the equipment and tools used in the assessment stage by AEP's and maxillofacial prosthetists (MP).

**Findings:** Component 1 highlighted the distress of the process experienced by the child patient and their parents as well as the role of parents and the AEP which can act as a barrier and facilitator to the process. Component 2 revealed a potential link between the way artificial eyes are fitted and the psychological wellbeing of the patient and their parents. Component 3 suggested that the tools and equipment used by MP's have the potential to be utilised in the artificial eye process.

**Conclusion:** This thesis demonstrates an original and significant contribution to knowledge in exploring the psychological impact of the artificial eye process in children with a diagnosis of Rb and the continual need to investigate the capabilities of technology for its potential incorporation into the process. The common theme running throughout this thesis was that of an intrinsic link between human and technological factors in creating an effective service. The findings contribute to both the Rb and ophthalmology literature: highlighting the needs and requirements for the progression of artificial eye services and the treatment and care of children with a diagnosis of Rb.

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## Glossary

Rb	Retinoblastoma
IPA	Interpretative Phenomenological Approach
AEP	Artificial eye prosthetists
MP	Maxillofacial prosthetists.
PWP	Psychological Wellbeing Practitioner
UK	United Kingdom
MRI	Magnetic Resonance Imaging
CT	Computerised Tomography
ICR	International Classification of Retinoblastoma
IRSS	The International Retinoblastoma Staging System
TMN	Tumour, Nodes and Metastasis Classification
EBRT	External Beam Radiation Therapy
NAES	National Artificial Eye Service
CHECT	Childhood Eye Cancer Trust
WWII	World War II
MDT	Multi-disciplinary team
RP&M	Rapid prototyping and manufacturing
3D CAD	Three-Dimensional Computer Aided Design
3D Scanning	Three-Dimensional Scanning.
IPA	Interpretative Phenomenological Approach
ENQ	Experience Near Question
CBT	Cognitive Behaviour Therapy
QoL	Quality of Life

## Terminology

Language is a communication tool that helps express feelings, desires and queries to the world around us. This enables us to harness our innate ability to form bonds with other people. Thus, the use of language plays an important role in how we interact, perceive and are perceived by others.

With connotations associated with certain words, we need to be aware of the terminology we use and how it affects others. Often people with or who have had a diagnosis of cancer are bombarded with war metaphors such as *survivor* or are referred to as either *winning* or *losing* their battle with cancer. Such terms can be viewed as offensive and have been reported as being the most challenging aspects of the cancer experience. Furthermore, when people whom have not experienced cancer use these terms, they can provoke what is known as *survivor's guilt*.

As not to cause any potential distress via the use of terminology, I will refer to participants within this thesis as either children with Retinoblastoma (Rb) or parents of children with Rb.



## PREFACE

The small incidence rate of Rb and the low requirement of artificial eyes in the UK, have resulted in both being under-researched areas. With literature on cancer showing negative psychological impacts and with technological advancements being utilised in healthcare, exploration of the artificial eye process in children diagnosed with Rb is warranted. This research was undertaken to create an in-depth body of knowledge of the process from the perspective of those involved, aiding future development. This thesis has significantly contributed to our knowledge through: (1) identifying and beginning the process of addressing the absence of knowledge regarding the psychological impact of the artificial eye process in children with Rb; (2) identifying variations in impact of the artificial eye process in AEP's, Rb survivors and their parents, creating suggestions for an improved process to enhance satisfaction to service delivery and product outcome; (3) establishing a need for further research and development into the incorporation of technology of the artificial eye process; (4) highlighting the need to treat human and technological factors as a continuum in improving the artificial eye process, and, (5) highlighting a need for better uniformity of artificial eye services and clarity of processes used as well as for the services to be recognised in their own right.

Before embarking on this research journey, I was working as a Psychological Wellbeing Practitioner (PWP) in NHS Adult Primary Care Mental Health. Within this role, I assessed and referred clients with various mental health difficulties, whilst treating adults with mild-moderate depression and anxiety (including social phobias, health anxiety, generalised anxiety disorder and simple phobias). I worked with clients with long—term conditions where mental health difficulties were a secondary by-product of the health condition. Working with this clientele, showed me the impact of long-term conditions not only by the sufferer but also their close family and friends. Furthermore, I became aware of the extent of this population whereby any health condition can lead to emotional and mental health difficulties. This ignited a keen interest to extend my understanding and clinical experience of the psychological impact of under-researched health conditions.

The PhD envisioned a thorough investigation into the design and fitting of artificial eyes including an in-depth exploration of current technologies with possible improvements to the current method being considered. This led to an extensive background review on Rb, the development of artificial eyes and the psychological impact of childhood cancers, disfigurements and prosthetic use.

The background reviews highlighted a lack of information and evidence into the psychological impact of the fitting of artificial eyes in children diagnosed with Rb from those affected by the process. Consequently, it became apparent that first and foremost, we need to understand the phenomenon from the perspective of those involved in the process before assessing the capability of technology to meet the needs required in artificial eye development.

With understanding the phenomenon being key, this thesis has employed a qualitative methodology consisting of 1 study with three components. The first component sought to gain an insight into the manufacturing and fitting process of artificial eyes from the perspective of AEP's. The second component sought to understand the lived experience of 13-16 year olds and parents of children wearing an artificial eye as a result of Rb. The third component sought to create a transfer of knowledge between the equipment and tools used in the assessment stage by AEP's and MP's.

Though the overarching aim of this thesis was to explore the artificial eye process in children with a diagnosis of Rb, the individual components provide their own independent and significant contribution to knowledge. Component 1 and 2 are the first of their kind to explore the perspectives of AEP's, parents of children and 13-16 year olds regarding the manufacturing and fitting process of artificial eyes following a diagnosis of Rb. Understanding the process better by those affected by it will lead to improved patient satisfaction and better outcomes through identifying future research and development opportunities. Component 3's assessment of the capabilities of technology for its incorporation into the artificial eye process was yet to be investigated. The findings from this component can lead to its application or suggestions for its future development.

As well as having clinical and service based implications for Rb and artificial eye services, this thesis provides an opportunity to further examine the psychological factors associated with the artificial eye process and its possible technological development.

# **Chapter 1 - Background Chapter**

The small incidence rate of Rb and the low requirement of artificial eyes in the UK, have resulted in both being under-researched areas. With literature on cancer showing negative psychological impacts and with technological advancements being utilised in healthcare, exploration of the artificial eye process in children diagnosed with Rb is warranted.

This chapter presents background information for the research presented in this thesis. It starts by examining the current literature regarding Rb: specifically, its definition, prognosis, clinical manifestations, diagnosis, staging and treatment options. In the following section we review the development of artificial eyes throughout the years using a scoping review and PRISMA framework. A literature review of the psychological impact of childhood cancers, disfigurements, amputations and prosthetic use follows before an overview of this topic is presented.

The contents of this chapter have led to two publications (see Appendix A for publication references).

## **1.1 Retinoblastoma**

### **1.1.1 Anatomy of the eye**

The eyeball is set in a protective cone-shaped cavity in the skull called the orbit or socket (Snell & Lemp, 1998). The average newborn's eyeball is 18 mm in diameter, growing to 19.5 millimeters (mm) in infancy to 25 mm in adulthood (Garnart & Lakshminarayanan, 2015). The orbit is surrounded by layers of soft, fatty tissue, which protect the eye and enable it to turn easily. Six extraocular muscles regulate the motion of the eye: the superior rectus (moves the eye upwards), the inferior rectus (moves the eye downwards), the medial rectus (moves the eye towards the nose), the lateral rectus (moves the eye outwards), the superior oblique (rotates the eye inward and also moves the eye downwards), and the inferior oblique (rotates the eye outwards and also moves the eye upward) (Garnart & Lakshminarayanan, 2015; Snell & Lemp, 1998). The tendons of these muscles are attached to the sclera. The sclera is the white outer wall of the eye and is made of collagen fibres (Parver, 1999). The conjunctiva, a transparent mucus membrane, covers the visible part of the sclera as well as lining the inner surface of the eyelids. The conjunctiva helps to keep the eye moist. Sitting on the outer wall of the eye is the cornea. This

is where the first step of turning light rays into visual images takes place (Parver, 1999; Snell & Lemp, 1998).

Located in the anterior chamber, the cornea is a transparent dome shape surface that covers the front of the eye. It is half a millimetre thick and it comprises of five layers: epithelium, bowman's membrane, stroma, decemet's membrane and the endothelium, all of which provide nourishment, clarity and protection from injury (Garhart & Lakshminarayanan, 2015). The cornea refracts the light entering the eye onto the lens before reaching the retina. Behind the cornea is a coloured ring shaped membrane called the iris. The iris has an adjustable circular opening called the pupil that controls the intensity of light permitted to strike the lens. In bright light situations, the iris constricts (the sphincter muscle), to limit the amount of light. Conversely, in dim light situations, the iris dilates (the dilator muscle) to increase the amount of light that enters the eye. A clear fluid called the aqueous humour fills the space between the cornea and the iris (Parver, 1999). The aqueous humour is divided into two sections called the anterior chamber (in front of the iris) and the posterior chamber (behind the iris). Situated behind the pupil is a colourless, transparent bi-convex structure called the crystalline lens. The lens measures 10mm and is used to fine tune vision. Cillary muscles surround the lens, and are attached by zonule fibres. When the muscles relax, they pull on and flatten the lens allowing the eye to see objects that are far away. To see closer objects, the muscles contract, thus thickening the lens (Snell & Lemp, 1998). After passing through the lens, light rays travel through the vitreous humour, a jelly-like tissue, before striking the retina. In early life, the vitreous humour is attached to the retina but may detach with age. The vitreous humour is separated from the aqueous humour by the hyaloid diaphragm.

The retina is a light sensitive layer that lines the interior of the eye. The retina receives its nutrients and oxygen from blood vessels in the choroid, which is located between the retina and the sclera. The retina is composed of light sensitive nerve endings known as photoreceptors. Photoreceptors contain chemicals that change when they are hit by light causing an electrical signal. There are two forms of photoreceptors: rods and cones. There are 100 million rods in the retina, which are necessary for seeing in low light, known as scotopic vision (Comer & Gould, 2012). They contain a pigment called rhodopsin, also called visual purple, which is broken down in the light and regenerated in the dark. The breakdown of rhodopsin gives rise to nerve impulses in bright light resulting in the rods being non-functional, thus activating the cones. As well as functioning best in bright light (photopic vision), cones are essential for acute vision. The eye contains three types of cones: red, green and blue, each sensitive to a different range of colours. There are 7 million cones in the retina with the greatest concentration being found in the fovea (Comer & Gould, 2012). The fovea is located at the centre of the macula, which is a yellow spot

in the retina at the back of the eye responsible for central vision. Nerve fibres, known as axons, branching from the rods and cones travel to the visual cortex: the part of the brain that controls our sense of sight, via the optic nerve. The optic nerve is a continuation of the retina, leaving the eye at the optic disk. At the point on the retina where the axons converge on the optic nerve, there are no rods or cones. This is called the papilla or the blind spot, and thus is insensitive to light. When the axons reach the brain, the light rays are turned into visual images (Garhart & Lakshminarayanan, 2015; Parver, 1999; Snell & Lemp, 1998). See Figure 1 for an illustration of the anatomy of the eye.

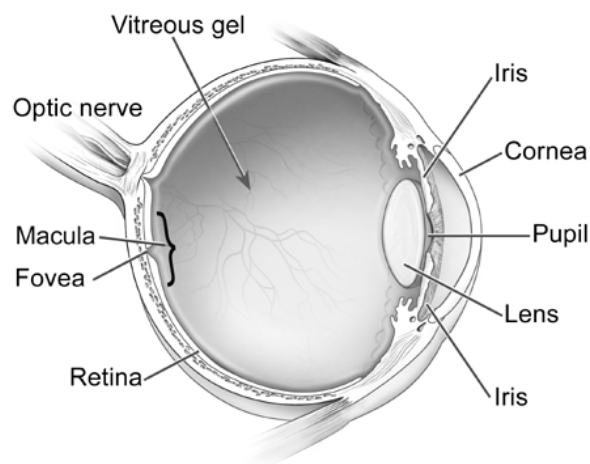


Figure 1: Anatomy of the eye. American Foundation for the Blind (2017). Eye Health: Anatomy of the eye. Retrieved from: <http://www.visionaware.org/info/your-eye-condition/eye-health/anatomy-of-the-eye/125>

### 1.1.2 What is retinoblastoma?

Rb is a rare type of eye cancer but the most common intraocular malignancy of early childhood (Dyer, 2016; Goel, Jain, Goel, & Juneja, 2012; Huang et al., 2013; MacCarthy et al., 2009; Meel, Radhakrishnan, & Bakhshi, 2012; Ray & Gombos, 2012; Sengupta, Pan & Khetan, 2016), forming 3% of all childhood cancers (Shields, 2008). Although almost exclusively a cancer of childhood, it has been detected in the foetus and can also occur in older children (Balmer, Zografos, & Munier, 2006; Shields, 2008). Shields, Shields and Shah (1991) found that 8.5% of 400 patients with Rb were older than 5 years of age at the time of initial diagnosis: median age of 6 years and the oldest age of 18 years. Misdiagnosis before referral was common in this population with the majority having unilateral disease (Karcioglu, Abboud, Al-Mesfer, Al-Rashed, & Pilapil, 2002).

Rb most often presents from birth to five years of age, typically being diagnosed in children before the age of three (Chintagumpala, Chevez-Barrios, Paysse, Plon, & Hurwitz, 2007; Meel et al., 2012; Willard et al., 2014). Its prevalence is equal in regard to age, sex, and right or left eye (Carty, 2009).

Between 40 and 50 children are diagnosed with Rb each year in the United Kingdom (Childhood Eye Cancer Trust, 2014). Data for the period of 2006 to 2010 show a five-year survival rate of 100%, in the UK. Survival rate in developed countries averages at 95% compared to 50% worldwide (Chauhan & Sharma, 2016; Meel et al., 2012; Shields, 2008; Yan, Zhang, & Li, 2013). Delay in presentation, diagnosis and access to resources are the most common explanations for this difference (Ramírez-Ortiz et al., 2014).

Rb develops in the retina, which is made up of nerve tissues that senses light as it comes through the front of the eye, before sending signals via the optic nerve to the brain where the signals are interpreted as images (Childhood Eye Cancer Trust, 2014). During the early stages of eye development, retinoblasts (cells) divide into new cells and fill the retina. At a certain point the cells stop dividing and develop into mature retina cells that detect light. In rare cases retinoblasts continue to divide and grow out of control forming Rb.

### **1.1.3 Genetics of retinoblastoma**

Rb arises due to a change (mutation) on the Rb 1 (Rb1) gene (Delhiwala, Vadakkal, Mulay, Khetan & Wick, 2016). The change in the Rb1 gene stops it working as it should. Depending on when and where the change in the Rb1 gene occurs, two different types of Rb can result: hereditary or non-hereditary. Rb can either affect one eye (unilateral) or both eyes (bilateral) and can lead to one (unifocal) or multiple (multifocal) tumours.

The Rb gene is located on chromosome 13, position 14.2 and is a tumour suppressor gene (Ray & Gombos, 2012). Each cell normally has 2 Rb1 genes that enable proliferation (growth and multiplication of cells) and suppress apoptosis (cell death). The Rb1 gene produces a tumour suppressor protein called pRB which stops proliferation, and thus regulates cell cycle (Xu et al., 2015). In healthy individuals, the pRB attaches to two other proteins XRCC5 and XRCC6, which mend broken strands of DNA. When the Rb1 gene is dysfunctional, more stranded DNA breaks leading to chromosomal abnormalities, which can evolve to become aggressive tumours (Cook et

al., 2015). As long as a retinal cell has 1 working Rb1 gene, it will not form Rb. However, when both Rb1 genes are mutated or lost, abnormal proliferation occurs leading to cells dividing uncontrollably and eventually become cancerous (Carty, 2009; Ray & Gombos, 2012; Shields, 2008).

Within the human body there are trillions of cells, two of which are somatic and germ cells. Somatic cells contain DNA arranged in chromosomes: of which each cell carries 23 pairs. Each pair of chromosomes comprises of one chromosome inherited from the father and one inherited from the mother. By contrast, germ cells give rise to gametes, which are cells that fuse during sexual reproduction (sperm and egg). Gametes contain half as many chromosomes. Thus, somatic cells have two copies of the Rb1 gene whereas gametes have 1 copy of the Rb1 gene. When two gametes meet during conception they fuse together creating a zygote (fertilized egg cell). If a mutation is present in the gametes a child with Rb is said to have the hereditary form. This is also referred to as a germline mutation. Conversely, if a mutation occurs in the somatic cells, a child with Rb is said to have the non-hereditary form. This is also referred to as sporadic Rb.

Whereas affected individuals of non-hereditary Rb are born with two normal copies of the Rb1 gene, those with hereditary Rb are born with one copy of the Rb1 gene mutated. For Rb to develop, mutation of the remaining gene in hereditary cases or mutation or loss of both genes in the non-hereditary form is needed. The loss of the second functional Rb1 gene can be due to a separate somatic mutation (Knudson, Hethcote & Brown, 1975). Knudson's 'two hit' hypothesis developed in 1971 explains this. In patients with hereditary Rb, the first mutation is acquired via the germline and thus is present in every cell. The second hit required in the predisposed patient is a somatic mutation. This second mutation typically occurs early in life in retinal cells. More specifically Balmer et al., (2006) stated that between the third month of post conception and 4 years old where retinoblasts reach maturity. In non-hereditary cases of Rb, both hits are acquired via somatic mutations by a precursor cell.

It is estimated that 40% of children with Rb have a germline mutation in 1 Rb1 gene, thus all the cells in the body have a defective Rb1 gene (Balmer et al., 2006; McCarthy et al., 2013). In 25% of these cases, the child has inherited it from one of their parents. The remaining 75% is the result of a mutation developed after conception while in the womb (McCarthy et al., 2013). Hereditary Rb is diagnosed on average at one year of age and the child is more likely to have multifocal and bilateral disease (McDaid, Hartley, Bagnall, Ritchie, Light, & Riemsma, 2005). Having the

genetic form of the disease also increase's the child's risk of developing secondary primary cancers. The most common secondary primary tumours are osteosarcomas, brain tumours and soft-tissue sarcoma (McDaid et al., 2005). Patients with bilateral disease are at a 5% chance of developing another cancer during the first 10 years of follow-up, 18% within the first 20 years and 26% within the first 30 years. This makes secondary primary tumours the greatest cause of death for patients with Rb (Balmer et al., 2006).

If a child has the hereditary form of Rb, yet they do not have an affected parent (as the mutation happened in the womb), the chance that the parents will conceive another child with Rb is less than 5%. However, the patient with hereditary Rb has a 50% chance of passing the abnormal gene onto their offspring (Chintagumpala et al., 2007).

The remaining 60% of children with Rb do not have the Rb1 gene mutation in all cells of the body. Instead, the Rb1 mutation occurs early in life in a single retinal cell by chance, sometime after conception (Shields, 2008). As of yet, it is not known how or why this occurs. Patients with non-hereditary Rb have unilateral, unifocal disease (Meel et al., 2012). The average age at diagnosis is two years and the disease cannot be passed to the patient's offspring (Chintagumpala et al., 2007). The disparity in age of onset and tumour number and location between the hereditary and non-hereditary form of Rb supports Knudsons 'two hit' hypothesis (Carty, 2009; Ray & Gombos, 2012).

If a patient with unilateral or bilateral Rb has relatives with the disease, it can be assumed that the patient has the inherited form of Rb. However, it cannot be assumed that a patient without a family history of the disease has the non-hereditary form. In other words, whilst all bilateral cases are hereditary, not all unilateral cases are non-hereditary. 10% of patients with unilateral disease have underlying germline mutation and are at risk of developing the disease in the uninvolved eye (Shields, 2008). It is therefore necessary to have all patients with Rb genetically tested to determine whether the Rb is (non) hereditary (Chintagumpala et al, 2007). The current techniques that are used can detect 90% of mutations in patients with bilateral Rb and 85% in patients with unilateral Rb

Approximately 5% to 8% of patients with Rb have a chromosomal abnormality involving the Rb1 gene that can be detected microscopically via a blood sample (Chintagumpala et al., 2007). If the chromosomal abnormality is detected in the patient, analysis of the parent's chromosome should



be performed as this increases the chance of the parent's having other offspring with Rb, (Chintagumpala et al., 2007). Where a chromosomal abnormality is not detected, specialised DNA tests are performed to identify small Rb1 gene changes in blood cells. If an Rb1 gene is deleted or changed in all the blood cells tested, the patient is presumed to have been born with a changed/deleted Rb1 gene in all cells. Although most of the time this change/deletion has been inherited from a parent, occasionally, it can occur spontaneously de novo. If the same Rb1 gene change/deletion is identified in one parent, it can be assumed the Rb is inherited. Conversely, if the Rb1 gene change/deletion is not detected the Rb is likely to be not inherited (Chintagumpala et al., 2007).

Rb is characterised by a high incidence of sporadic cases. 80% of newly diagnosed bilateral cases are sporadic without a family history and caused by de novo germline mutations in the Rb1 gene (the 75% of inherited cases not passed on from a parent). In contrast, 87% of unilateral Rb is caused by post-zygotic mutation occurring at the early stages of embryo development that can lead to mosaicism (Chen et al., 2014). Mosaicism is evident when a mutation is detected in some but not all cells. Somatic mosaicism involves germ cells when the mutation occurs prezygotic (prior to conception). Isolated germinal mosaicism happens when the initial mutation occurs in a progenitor germ cell that continues to divide (Rushlow et al., 2009). Mosaicism can only be detectable in the deficit gene if a family is known and the family members are available for analysis (Chen et al., 2014). Sanger sequencing and lymphocyte DNA are techniques currently used for detection (Chen et al., 2014).

#### **1.1.4 Trilateral retinoblastoma**

Trilateral Rb occurs in 5% to 15% of patients with heritable retinoblastoma: usually bilateral but can also be unilateral (McDaid et al., 2005). Trilateral Rb results from the development of an independent brain tumour that forms in the pineal gland (McDaid et al., 2005). Trilateral Rb is relatively uncommon, does not result from the spread of intraocular retinoblastoma and is usually fatal (McDaid et al., 2005; Ortiz & Dunkel, 2016). The condition is usually diagnosed 2 years following diagnosis of retinoblastoma and has a median survival time from diagnosis of the disease of 9 months (McDaid et al., 2005). Given the poor prognosis and the short interval between diagnosis of Rb and the occurrence of trilateral disease, imaging tests are recommended every six months for five years for those with the hereditary disease or unilateral disease and a positive family history (McDaid et al., 2005).

### **1.1.5 Prognosis**

The prognostic outcome of life and vision in patients with Rb has significantly improved in recent years. Factors such as improved methods for diagnosis and treatment, and age and stage of tumour(s) as well as type of Rb, have a contributing role (Meel et al., 2012; Willard et al., 2014; Yan et al., 2013). Patients diagnosed before the age of two are thought to have a higher survival rate than those diagnosed at two years or older. Reasons for this include advanced presentation and an increased risk of extraocular extension and metastasis in those who are diagnosed at a later age (Chintagumpala et al., 2007). Consequently, more intensive treatment is required which may adversely affect vision and survival of the patient.

Patients with intraocular Rb have a five-year survival rate of 98% compared to 10% in patients with extraocular Rb. Furthermore, patients with the hereditary form of Rb have a higher risk of developing secondary primary cancers and are susceptible to the development of trilateral Rb (McDaid, 2005). Those with trilateral Rb have an average survival rate of eight months when treated compared to one month in those untreated (Provenzale, Gururangan, & Klinworth, 2004).

### **1.1.6 Clinical manifestations of retinoblastoma**

Clinical manifestations of Rb vary dependent on the stage of the disease (Carty, 2009; Shields, 2008). The most common presenting sign at 60% is leukocoria (Balmer & Munier, 2007; Ortiz & Dunkel, 2016; Selistre et al., 2016). When a bright light is shone into an eye, blood vessels will reflect a red colour back. In those with Rb the pupil will appear white or yellow. This is also known as 'Cat's eye' and is often noticeable when a photograph is taken using the flash (Meel et al., 2012). As a late manifestation of Rb, the tumours are likely to be large (Chintagumpala et al., 2007). However, prognosis remains high at 88% of the five-year survival rate (Balmer & Munier, 2007).

The second most common sign at 20% is strabismus. As an early sign, prognosis and eye salvage is high. Strabismus (misalignment of the eye) results from a loss of central vision in one or both eyes (Chintagumpala et al., 2007). It involves a lack of coordination between the extraocular muscles preventing the gaze of each eye at the same point. Thus, one eye may look straight ahead, upward, downward or outward. Other presenting symptoms include reddish pupil, larger than normal pupil, poor or decreased vision, wandering eyes (nystagmus) and parental history of Rb (Goel, 2012).

### **1.1.7 Diagnosis**

If Rb is suspected, a red reflex test needs to be conducted (Carty, 2009). This test is used to view both reflexes at the same time in order to determine if the patient has normal ocular alignment. The test is conducted in a dimly lit or dark room using an ophthalmoscope at 50 centimeters (cm) away from the child. This allows simultaneous illumination of both eyes making it easier to compare the reflex. Pupillary dilation has been found to be helpful when performing this test. To be considered normal, a red reflex will be systematic and shown in both eyes. Dark spots in the red reflex, a diminished reflex and the presence of a white reflex indicate an abnormality (Childhood Eye Cancer Trust, 2014). Thus, an urgent referral to an ophthalmologist for further evaluation is required (NICE, 2005). It is advised that children with a positive family history of Rb are screened soon after birth. This needs to be repeated every four to six weeks up to the age of one year and then every two to three months until three years of age (NICE, 2005).

Once Rb has been diagnosed, imaging tests are conducted to find out if the cancer has spread within or beyond the eye. This process is known as staging. Staging not only depicts how far the cancer has spread, it also provides prognosis of survival, outlook for saving vision, size and location of the tumour(s) and likelihood that certain treatments will be effective (Meel et al., 2012; Ray & Gombos, 2012; Willard et al, 2014). Imaging tests used in the detection and diagnosis of Rb are an ultrasound, Magnetic Resonance Imaging (MRI) and Computerised Tomography (CT) scans (Goel et al., 2012).

#### **1.1.7.1 Ultrasound**

An ultrasound (echography) examination helps determine the thickness or height of the tumour (Chintagumpala et al., 2007). Sound waves are used to penetrate and outline structures in the eye. By placing a small ultrasound probe against the eyelid or eyeball, echoes of the sound wave that bounce off the tissues inside and around the eye are detected and displayed on a computer screen.

#### **1.1.7.2 Magnetic Resonance Imaging**

The Magnetic Resonance Imaging (MRI) scan provides detailed images of the eye and surrounding areas such as the spinal cord and brain (Meel et al, 2012). Antennas within the MRI machine pick up the radio waves and feed them into a computer to create an image. Whereas most children with Rb will have one MRI scan, those with bilateral Rb may have scans for several

years after treatment to ensure no further spread. The procedure takes up to an hour and requires the patient to lay still. General anaesthetic may be given to help with this.

#### **1.1.7.3 Computerised Tomography**

A Computerised Tomography (CT) scan is a specialised x-ray test that creates detailed cross-sectional images of parts of the body, particularly soft tissues (Chintagumpala et al., 2007). The CT scanner takes a number of small pictures as it rotates around the patient. The images produced are called tomograms which helps to determine the size of a Rb tumour and its spread within the eye and to nearby areas. An injection of the contrast medium dye may be given beforehand in order to make the scan clearer. Although CT scans give off radiation making MRI scans a preferred choice, calcification (deposits of calcium in the tumour) is detected better using this test (Chintagumpala et al., 2007). This is particularly helpful when the diagnosis of Rb is not clear.

#### **1.1.7.4 Bone scan**

In cases where there is a strong reason to think that Rb has spread to the skull or other bones, a bone scan is undertaken. A very small amount of radioactive material is injected into the vein intravenously and travels through the bloodstream. The radioactive material settles in areas of damaged bone and is detected by a camera. The areas with cancer show up brighter. However, it is important to be aware that the areas may be a result of other bone changes.

#### **1.1.7.5 Additional tests**

Other types of tests that are carried out when there are additional symptoms (weight loss, vomiting), or abnormal findings associated with the diagnosis of Rb include blood tests, lumbar puncture and bone marrow aspiration and biopsy. Blood tests check for problems with the liver and kidneys and for changes in chromosome 13. A lumbar puncture, also called a spinal tap, is a procedure that removes cerebrospinal fluid (CSF) from the lower back that surrounds the brain and spinal cord. CSF is a clear liquid that delivers nutrients to the brain. The removal of CSF allows for examination of cancer cells. Bone marrow aspiration and biopsy determines whether any Rb cells have spread to the bone marrow, which is the soft inner part of certain bones. The samples are taken from the back of the pelvic bone. Bone marrow aspiration is conducted first where a thin needle is inserted into the bone and a syringe is used to suck out a small amount of liquid bone marrow (aspirate). For the biopsy, a small piece of solid tissue is removed. Both samples are then looked at under a microscope to identify any potential tumour cells.

### **1.1.8 Differential diagnosis**

Due to a number of ocular disorders in children clinically resembling Rb, it is important that the diagnosis of Rb is fully confirmed prior to the start of treatment. Shields et al. (2013) found that 42% of patients who were referred with possible Rb had lesions that simulated Rb. In this study, a total of 23 different conditions were accounted for the pseudo-Rb including: persistent hyperplastic vitreous (blurred vision due to scarred vitreous); Coats disease (abnormal development of blood vessels behind the retina) and presumed ocular toxocariasis (rare infection caused by roundworms). Patients with these diseases often present with leukocoria: the most common presentation in Rb. Other less common conditions that may mimic Rb are congenital cataract, retinopathy of prematurity, Norrie disease and familial exudative vitreoretinopathy, (Kiss, Leiderman, & Mukai, 2008). A careful clinical examination using the red reflex test as described above is usually sufficient to determine the correct diagnosis.

### **1.1.9 Staging of retinoblastoma**

As previously mentioned, Rb is staged on the results of eye exams, imaging tests and in rare cases, biopsies. The stage of Rb determines best treatment option and can help predict a patient's prognosis. Rb is staged on the basis of whether it is intraocular, extraocular or recurrent.

Intraocular Rb is the earliest stage of the disease where the cancer is still within the eye. It may be confined to the retina or extend to other structures such as the optic disk, choroid or anterior chamber, however, does not extend beyond the eye. If left untreated for 12 months, metastasis is likely to occur and can become fatal within a few years (Carty, 2009). Extraocular Rb is an advanced stage of disease where the cancer has spread outside of the eye. In these cases, cells break away from the main tumour on the retina and float through the vitreous to reach other parts of the eye. The cancer can spread either to the tissues around the eye (orbital Rb), the central nervous system (CNS) or to the bone marrow or lymph nodes (metastatic Rb). In the majority of cases, extraocular Rb occurs in developing countries due to late detection, limited access to resources and thus delayed diagnosis (Leander et al., 2007). Recurrent Rb is where the tumour has recurred or progressed following initial treatment. The recurrent tumour may be confined to the eye, tissues surrounding the eye or other parts of the body.

In the UK, Rb is primarily detected and diagnosed before it has spread outside of the eye. Therefore, staging systems that apply only to intraocular Rb are used most often in this country.

#### **1.1.9.1 Intraocular staging of retinoblastoma**

There are two main staging systems for intraocular Rb: the Reese-Ellsworth staging system and the International Classification for Intraocular Retinoblastoma.

##### **1.1.9.1.1 The Reese Ellsworth Staging System**

In the 1960's the Reese-Ellsworth classification was created based on intraocular tumour staging and globe salvage after External Beam Radiotherapy (EBRT) (Balmer et al., 2006; Chintagumpala et al., 2007). The system divides eyes into 5 groups (I to V) and 10 subgroups ('a' and 'b' for each group) according to location, focality and the size of the tumours. Group I have the lowest risk of treatment failure and group V has the highest risk of enucleation. Tumour control for group I – III is reported to be 78% compared to 20% in group III-V (Hernandez et al., 1995).

During this era, EBRT was the treatment modality of choice, however it had little success with multifocal and large tumours due to technical difficulty (Ramasubramanian & Shields, 2012). Consequently, these tumours were presumed more aggressive and were given a higher ranking in the classification system, implying a worse ocular prognosis (Shields, 2013).

With radiation being linked to disfigurement and increasing the risk of developing secondary cancers, particularly in those with hereditary Rb, Chemoreduction combined with focal treatments became the primary treatment modality (Kiss et al., 2008). This shift in treatment modality found that multifocal and large tumours do not have a worse prognosis than small, solitary macular tumours. What emerged as the difficulty in treating Rb was the management of vitreous and subretinal seeds. By not addressing seeding, the Reese-Ellsworth classification was found to be a poor predictor of Chemoreduction success (Kiss et al., 2008). This led to the International Classification of Retinoblastoma (ICRB) being developed in 2003, which is predominately based on the extent of subretinal and vitreous seeding with minor consideration of tumour size and location (Shields, 2013).

### **1.1.9.1.2 Intraocular Classification of Retinoblastoma**

The International Classification of Retinoblastoma (ICR) (2003) is a staging system that divides intraocular Rb into 5 groups labelled A to E. It is based on the natural history of Rb (early stage, group A to late stage group E) and on the likelihood of saving the eye following primary treatment of chemotherapy and adjuvant focal therapy (Balmer et al., 2006; Kiss et al., 2008). Each eye is staged independently and the tumour with the higher grade is used to classify the eye

- Group A: The tumour is confined to the retina and is less than 3 mm across. The tumours are not near important structures such as the foveolar and the optic disc. There is no vitreous or subretinal seeding.
- Group B: The tumour is confined to the retina, larger than 3 mm and is close to the optic disc and foveolar. No vitreous or subretinal seeding but the subretinal fluid is less than 5 mm from the base of the tumour.
- Group C: Tumours with a small amount of subretinal spread.
- Group D: Large tumours with widespread vitreous or subretinal seeding.
- Group E: Very large tumours that have destroyed the eye.

Group A has the lowest risk of treatment failure whereas group E eyes are rarely salvageable, requiring additional EBRT or enucleation (Shields et al., 2006). The chances of retaining useful vision decreases from group A to E (Kiss et al., 2008).

### **1.1.9.2 Extraocular staging of retinoblastoma**

Several classification systems have been developed for extraocular Rb, and some for both intraocular and extraocular Rb. Extraocular staging applies to cancer that has spread outside of the eye and takes into account the degree of local extension, intracranial metastasis and haematological metastasis.

#### **1.1.9.2.1 International retinoblastoma staging system**

The International Retinoblastoma Staging System (IRSS) encompasses both intraocular and extraocular Rb. The IRSS is based on the extent of the disease (Meel et al., 2012). The staging system is split into 5 sections ranging from stage 0 to stage I

- Stage O: The tumour is in the eye only. The eye has not been enucleated and the tumour was treated without surgery.
- Stage I: The tumour is in the eye only. The eye has been removed and no cancer cells remain.
- Stage II: The tumour is in the eye only. The eye has been removed and there are cancer cells that can be seen microscopically.
- Stage III: This stage is divided into two sub-stages:
  - IIIa: The cancer has spread to tissues around the eye socket.
  - IIIb: The cancer has spread to lymph nodes near the ear or in the neck.
- Stage IV: This stage is divided into two sub-stages:
  - IVa: The cancer has spread to the blood where there are one or more tumours.
  - IVb: The cancer has spread to the brain or spinal cord.

#### **1.1.9.2.2 The Tumour, Nodes and Metastasis classification**

The Tumour, Nodes and Metastasis (TMN) classification is particularly useful in describing the extent of Rb. It takes into account the size of the primary tumour and how far it has grown within and outside of the eye; whether or not the cancer has reached the lymph nodes and whether it has metastasised to other sites such as the bone marrow and brain

#### **1.1.9.3 Recurrent retinoblastoma**

Recurrent Rb is cancer that has returned or continues to grow after treatment. The cancer may come back in the same place (local recurrence), nearby (regional recurrence), or in another place (distant recurrence). Further testing is undertaken in order to restage the cancer. The cancer is restaged depending on whether it is intraocular or extraocular. Intraocular and extraocular recurrences have very different prognoses and are treated in distinctly different ways.

#### **1.1.10 Treatment**

The goals of treatment for a patient with Rb are to save the patient's life, to save at least one eye and to prevent vision loss (Delhiwala et al., 2016; McDaid, 2005). The management of Rb needs a multidisciplinary team approach including an ocularist, pediatric oncologist, genetist and an ophthalmic oncologist (McDaid, 2005).



Treatment of Rb is highly individualised and is dependent on whether the cancer is intraocular or extraocular, unilateral or bilateral and unifocal or multifocal. Additional features that affect treatment choice include the age of the child, family and societal perception, overall prognosis, cost effectiveness of treatment and the size, location and number of tumours (Goel et al., 2012, Willard et al., 2014). Treatment of Rb in the UK is primarily based on intraocular disease due to its high percentage at diagnosis. There are several methods to manage intraocular Rb including focal therapy (photocoagulation, thermotherapy, cryotherapy and brachytherapy), local therapy (external beam radiation and enucleation), and systemic therapy (chemotherapy). While primary focal therapy treats small tumours, local and systemic therapy treats advanced tumours. Combinations of treatment may be required to achieve tumour control (McDaid, 2005; Meel et al., 2012; Willard et al., 2014).

#### ***1.1.10.1 Treatment of intraocular retinoblastoma***

##### **1.1.10.1.1 Focal Therapy**

Focal therapy refers to a group of treatments that are applied directly to the eye. Focal therapy is performed under general anaesthetic and is repeated numerous times until all remaining signs of active tumour have disappeared. A noticeable side effect of focal therapy is scarring of the retina thus affecting vision. There are four main types of focal therapy used to treat Rb: photocoagulation, thermotherapy, cryotherapy and brachytherapy. Focal therapy is generally used to treat small to medium tumours that fall into group A-C on the International Classification for Intraocular Retinoblastoma and groups I to IV on the Reese Ellsworth Classification for Intraocular Retinoblastoma.

##### **1.1.10.1.2 Photocoagulation (laser therapy)**

Photocoagulation, also known as laser therapy, is used for small tumours, measuring less than 3.5 mm in thickness, residual tumour after chemotherapy and recurrence after chemotherapy (Ray & Gombos, 2012). Photocoagulation uses heat in the form of a laser to physically destroy tumour cells. The laser is directed to the affected areas of the retina through the pupil. Pharmaceutical drops are used to dilate the pupil. The heat of the laser cauterizes the blood vessels that surround and supply the tumours (Carty, 2009). On average, photocoagulation is delivered 2 or 3 times with a month between treatments (Ray & Gombos, 2012). Photocoagulation is a painless procedure although the eye may be red following treatment. In rare cases photocoagulation can damage the retina, which can lead to blind spots or temporary retinal detachment (Chintagumpala et al., 2007).

#### **1.1.10.1.3 Thermotherapy**

Thermotherapy is recommended for small tumours, measuring 4 mm in diameter and 2 mm in thickness, close to the fovea or optic nerve, where other therapies might create greater visual loss (Carty, 2009). Thermotherapy involves the use of heat to help shrink tumour cells. The heat often used in treating Rb is infrared rays and is directed either on the whole eye or localised to the tumour area (Balmer et al., 2006; Chintagumpala et al., 2007; Ray & Gombos, 2012; Yan et al., 2013). The tumour is heated at a temperature between 40 and 60 degree Celsius until it turns a subtle grey, in order not to damage the retinal vessels (Chintagumpala et al., 2007). Complete tumour regression can be achieved in 85% of tumours using 3 to 4 sessions. Common complications include retinal detachment, clouding of the lens, shrinking off the iris and damage to the retina.

#### **1.1.10.1.4 Cryotherapy**

Cryotherapy is primarily used to treat small tumours, measuring up to 4 mm in diameter and 2 mm in thickness, located on the front part of the retina (Balmer et al., 2006; Yan et al., 2013). Cryotherapy uses liquid nitrogen to destroy the Rb cells by freezing them (Carty, 2009). A small metal probe is placed on the outer surface of the eyeball next to the tumour, which is frozen and thawed several times. The thawing process kills the tumour cells as ice crystals pierce the tumour membranes destroying the cells. Cryotherapy is delivered 2 or 3 times with a month between treatments. Cryotherapy may cause the eye and eyelid to swell for a few days and can damage the retina leading to blind spots and temporary retinal detachment (Chintagumpala et al., 2007).

#### **1.1.10.1.5 Brachytherapy**

Brachytherapy, also known as plaque radiotherapy, is used for small to medium sized tumours less than 15 mm in diameter and 10 mm in thickness that are situated away from the optic nerve and centre of vision. Brachytherapy can also be used as a secondary measure after prior failed treatment such as cryotherapy, or in the case of recurrence (Balmer et al., 2006; Carty, 2009).

Brachytherapy involves the application of radioactive material to the outer surface of the eye (sclera) at the base of the tumour (Chintagumpala et al., 2007). The radioactive material is put into a small carrier known as a plaque, which delivers a low dose of radiation directly to the tumour. The plaque is surgically attached to the outer part of the eyeball, directly over the tumour.

The plaque is left in place for between two and five days before being surgically removed. Brachytherapy has a 90% tumour control rate but can lead to the development of a cataract. However, this can be corrected with surgery once the Rb is under control.

#### **1.1.10.1.6 Local therapy – External Beam Radiation Therapy**

External Beam Radiation Therapy (EBRT) was the preferred form of Rb management in the 1990's. However, the long-term complications such as damage to nearby body tissue, the increased risk of developing secondary cancers, particularly in those patients with hereditary Rb, stunting of the orbital growth, dry eye, and cataract, led to the development of newer chemotherapy protocols (Chintagumpala et al., 2007).

External beam radiation therapy is most often used in advanced cases of Rb such as advanced bilateral cases or recurrence (Balmer et al., 2006; Carty, 2009; Ray & Gombos, 2012). It is also indicated in eyes that contain one or more tumours that involve the optic disc, a tumour that is larger than 16 mm in diameter, where there is subretinal seeding, and for eyes where primary chemotherapy and local therapy has failed. It may also be used to treat the eye socket after enucleation if there is extension behind the area (Chintagumpala et al., 2007).

External beam radiation therapy uses an invisible form of high energy (like x-rays) to kill cancer cells or keep them from growing or dividing. A linear accelerator machine directs radiation to the precise area of the eye needing treatment. However, if the cancer is extensive, radiation treatment of the entire eye may be necessary. The treatment is given in doses measured in units called centigrays (cGy). Ray and Gombos (2012) reported a dosage of 45-50Gy given in 1.5-2 Gy/fraction. Radiation is usually given 5 days a week for several weeks with the delivery only taking a few minutes (Chintagumpala et al., 2007).

#### **1.1.10.1.7 Enucleation**

As a result of earlier tumour detection as well as improved use of more conservative eye-sparing treatments such as focal therapy, there has been a significant decrease in the use of enucleation in patients with Rb over the last 40 years (Ray & Gombos, 2012). However, enucleation remains a frequent treatment for Rb and is indicated for all unilateral tumours that fill over half the eye, where there is extensive seeding, retinal detachment, high risk of metastasis and tumour invasion into the optic nerve (Balmer et al., 2006; Goel et al., 2012; Ray & Gombos, 2012). In patients

with bilateral Rb, the eye with the more advanced tumour will be enucleated and the less affected eye managed by other therapies (Chintagumpala et al., 2007). If neither eye has useful vision because of damage already caused by the cancer, enucleation of both eyes will be undertaken to make sure that all the cancer is gone. Thus, enucleation primarily occurs in groups D-E of the International Classification for Intraocular Retinoblastoma and group V of the Reese-Ellsworth Classification of Intraocular Retinoblastoma.

Enucleation is the surgical removal of the eyeball, leaving the 6 extraocular muscles and the contents of the eye socket intact. Enucleation is performed under general anaesthesia with the procedure taking 60 to 90 minutes. Immediately after the eyeball has been removed, an orbital implant is inserted into the socket. The eye muscles are attached to the implant to improve motility and the implant is covered externally with the conjunctiva (pink surface tissue that lines the eyelid) and sutured in place. The patient is then fitted with a conformer shell made of clear acrylic resin offering a smooth curved surface over which the eyelids blink without rubbing the suture line. The shell maintains the shape of the eye socket and helps stop infection. It has a small hole to allow drainage and is marked with a letter to indicate its size. A pressure pad is worn over the empty socket for 12 to 48 hours after the operation to help reduce tissue swelling in the socket.

Side effects of enucleation include bruising, ptosis (droopy eyelid), haemorrhage, infection, scarring of the socket and extrusion of the implant (Chintagumpala et al., 2007).

#### **1.1.10.1.8 Systemic therapy – chemotherapy**

Chemotherapy is an effective treatment for large tumours or tumours that are located near the optic nerve or centre of vision (Chintagumpala et al., 2007). Chemotherapy is the use of anti-cancer drugs to destroy cancer cells by stopping them from growing or multiplying. The chemotherapy drugs most commonly used for treating Rb are etoposide, carboplatin and vincristine. Often two or more drugs are given at the same time depending on the extent of the tumour (Chintagumpala et al., 2007)

Chemotherapy can be given in several different ways, but is most often given intravenously (IV) through a vein or orally (per oral – PO). This is known as systemic chemotherapy. Systemic chemotherapy is where the drugs travel through the bloodstream to eradicate the cancer rather than being applied directly to the cancer. When given intravenously, the chemotherapy drugs

travel through a device called a port-o-cath. The port-o-cath is surgically inserted under the skin of the patient's chest and is attached to large blood vessels. The chemotherapy drugs travel through the bloodstream to enter the blood supply of the tumour where they begin to destroy it. Direct insertion into the bloodstream reduces the risk of harm to the surrounding tissues.

Systemic chemotherapy is given in cycles of treatment followed by a rest period. Each cycle lasts a few weeks with the total length often being several months; though this is dependent on how well the tumour responds.

These methods can expose the entire body to significant doses of chemotherapy, thus can result in unwanted and sometimes long-lasting side effects such as hair loss, mouth sores, damage to the heart, kidneys and lungs, nausea and vomiting, fatigue and increased chance of infections (Chintagumpala et al., 2007).

Intra-arterial chemotherapy is a new treatment for advanced Rb. The chemotherapy drug (melphalan and/or topotecan) is delivered through the blood vessels (Yan et al., 2013). A thin catheter is inserted into a large artery on the inner thigh and threaded through blood vessel into the ophthalmic artery. The chemotherapy drug is then infused into the artery. This method is designed to minimise the drug's exposure to the rest of the body and to reduce side effects often seen in systemic chemotherapy. The average number of treatment sessions is three for each eye with each session being delivered at four-week intervals. Due to its recent development, the long-term effects of this treatment are yet unknown. However, early reports indicate up to 100% globe salvage for group C and D of the International Classification of Intraocular Retinoblastoma.

Typically, chemotherapy is used in addition to other therapies in order to either shrink a tumour before other therapies, (known as neo-adjuvant therapy or Chemoreduction), to destroy cancer cells that remain after therapy, for example post-enucleation, (called adjuvant chemotherapy) or to help destroy cancer if it spreads or recurs (Chintagumpala et al. 2007).

#### **1.1.10.1.9 Chemoreduction**

Chemoreduction has become an important therapeutic tool in treating Rb (Shields & Shields, 2004). Chemoreduction is a method of using intravenous chemotherapy to reduce the size of the

tumours so that residual tumours can be eradicated with focal treatment methods. This approach results in 85% globe salvage of eyes classified as Reese-Ellsworth groups I to IV, compared to 47% in group V (Shields & Shields, 2004). Chemoreduction coupled with focal therapies can minimise the need for enucleation or external beam radiation therapy.

Chemoreduction is used in nearly all children with bilateral Rb and about 25% of children with unilateral Rb (Chintagumpala et al., 2007; Yanagisawa, 2004). It is most successful for tumours without subretinal fluid or vitreous seeding.

#### ***1.1.10.2 Treatment of extraocular retinoblastoma***

Few patients in developed countries present with extraocular Rb. Extraocular disease may be localised to the soft tissues surrounding the eye or to the optic nerve, brain, or central nervous system.

The standard treatment options for when the cancer has spread within the eye, known as orbital and locoregional metastasis, is chemotherapy and radiation therapy; with the cure rate standing at 60% to 85% (Chantada, Fandino, Casak, Manzitti, Raslawski & Schwartzman, 2003). Orbital Rb occurs as a result of progression of the tumour through the sclera and occurs in 60% to 70% of patients with extraocular disease.

Patients with stage I to III extraocular Rb have had an eye removed yet there is some microscopic spread to the optic nerve, lymph nodes or bone cavity. Patients with spread to these areas require adjuvant chemotherapy and possibly radiation therapy after enucleation (Chintagumpala et al., 2007). Adjuvant chemotherapy destroys microscopic cells in order to prevent a possible cancer recurrence.

Stage IV encompasses extraocular spread to distant areas of the body outside of the eye such as the central nervous system. This stage of extraocular Rb is treated with high dose chemotherapy and/or radiation therapy to destroy as many cancer cells as possible, before being given a stem cell transplant. A stem cell transplant is where hematopoietic stem cells are given to a patient to replace bone marrow that contains cancer. These cells grow into healthy blood cells to replace the ones the patient lost (Chintagumpala et al., 2007).

### ***1.1.10.3 Treatment of recurrent retinoblastoma***

Recurrent Rb is cancer that has returned after it has been treated. The cancer may recur in the eye, (intraocular), in tissues around the eye, or in other places in the body (extraocular). Furthermore, the cancer may come back in the same place (called a local recurrence), nearby (regional recurrence), or in another place (distant recurrence). When this occurs, tests will be undertaken to determine location, size and how aggressive the new tumour is. Treatment for recurrent intraocular Rb may include enucleation, external beam radiation therapy, systemic chemotherapy and/or focal therapy (Yanagisawa, 2004). Treatment failure necessitates additional external beam radiation in 10% of patients and enucleation in 15% of patients at five-year follow-up (Yanagisawa, 2004). Recurrent extraocular Rb can be treated with systemic chemotherapy, radiation therapy and stem cell transplant. Long term monitoring will detect possible recurrence at an early stage, thus be controlled with further salvage measures (Shields & Shields, 2004).

### ***1.1.10.4 Treatment of trilateral retinoblastoma***

The optimal therapy for patients with trilateral Rb is not known, however treatment have progressively resulted in increased survival of patients in the last 20 years (Mouratova, 2005). Before 1995 the five-year survival rate of trilateral Rb was 6% compared to 44% (de Jong et al., 2014). Current research suggests that successful treatment of trilateral Rb includes screening at diagnosis and treating with high-dose chemotherapy with stem cell transplant (Carty, 2009; de Jong et al., 2014).

### ***1.1.10.5 Treatment summary***

Chemoreduction with or without focal therapy is the most commonly employed conservative treatment for Rb (Meel et al., 2012). Although an effective treatment, tumour and associated vitreous and subretinal seeds can recur within the first 3 years (Shields & Shields, 2004). Thus, Chemoreduction is most successful for tumours without subretinal and vitreous seeding: group A-B on the International Classification for Intraocular Retinoblastoma and groups I-IV on the Reese-Ellsworth Classification for Intraocular Retinoblastoma. Enucleation stands alone as the treatment of choice for patients with tumours that fill more than 75% of the eye. High-dose chemotherapy with stem cell transplant is the treatment of choice following enucleation for patients with extraocular Rb stage I to IV and for trilateral Rb.

### **1.1.11 Research directions**

The relatively small numbers of Rb cases coupled with non-comparable staging systems, for example the Reese-Ellsworth, had resulted in little research into this prolific disease (NHS England, 2013). However, over the last twelve years' collaboration between national and international centres has resulted in an increase in the evidence base of Rb management. Current research is directed towards genotype-phenotype relationship, local drug delivery methods, minimising side effects of treatments, developing better animal models and exploring biological treatment such as growth factors (NHS England, 2013).

## **1.2 The development of artificial eyes throughout the years: A scoping literature review**

### **1.2.1 Introduction**

Human understanding of expressions and what they mean emerges very early on in life. Confronted with and revealing a whole host of emotions when we look into one another's eyes gives credence to the saying 'our eyes are the windows to our soul'. Our eyes act as our navigational tools: we adjust and modify our behaviour and act in accordance to our interpretations, whether it is related to our physical surrounding, social interactions or emotional contact. With many aspects of human functioning relying on our eyes, a loss of one or both can have a significant impact on a person.

The loss of an eye(s) can be caused by a congenital defect or be acquired. Examples include malignancies, intraocular and extraocular infections, trauma, orbital tumours and painful blind eyes (Manoj, 2014). Following the loss of an eye, patients are fitted with an artificial eye. Today's artificial eyes are a thin hard acrylic shell that covers the surface of the eye, fitting over an orbital implant and under the eyelids (Manoj, 2014).

Research into the impact that the loss of an eye has, highlights medical, psychological, social, emotional and physical difficulties (Ayanniyi, 2013; Benson, 1977; Cerullo, 1990; Deacon, 2008; Goiato et al., 2014; Manoj, 2014; Raizada & Rani, 2007). From a medical perspective, the loss of one eye results in a change from binocular vision to monocular vision leading to initial feelings of disorientation and clumsiness (Cerullo, 1990). Although adjustments by turning the head can overcome the defects in depth perception and field of vision, some activities of daily living such



as playing sports, driving and work requiring prolonged visual vigilance will continually be impacted.

Psychological adjustment to the loss of an eye comes with emotional acceptance in all areas of one's life: including personal, professional and societal. In circumstances where a person has not adjusted to the loss, difficulties such as depression, anxiety, low self-esteem, low self-image and feelings of hopelessness may arise. This can lead to maladaptive coping mechanisms impacting social functioning and independence (Deacon, 2008). With the potential of psychological maladjustment impeding the artificial eye process, attention needs to be paid to the way artificial eyes are designed, manufactured and fitted. The focus of health technology can then begin to enhance the current process not only benefiting the recipients but also the prosthetists that produce the artificial eyes.

Whilst research has documented the factors associated with the loss of an eye (Benson, 1977; Cerullo, 1990; Deacon, 2008; Goiato et al., 2014), knowledge surrounding the impact of the artificial eye process is still in its infancy. Furthermore, despite its progression in related fields, such as prosthetic limbs, technological development regarding artificial eye design and manufacturing has stagnated. These two key facets are essential in identifying future research and development in order to create better outcomes for healthcare professionals and artificial eye wearers alike.

In order to achieve this, a strong knowledge base needs to be created: often done through reviews. The recent drive in evidence-based practice has resulted in various types of reviews of literature being developed. This includes systematic reviews, traditional literature reviews and scoping reviews. Traditional literature reviews critique and summarise a body of literature about the topic. With no clear methodological approach, information is collected and interpreted with subjective summaries of findings. In comparison to a systematic review which answers a defined research question by collecting and summarising empirical evidence that fits pre-specified eligibility, scoping reviews gather and map the research in a given area rather than involving an assessment of the quality of the studies. With the aim to portray current knowledge regarding the development of artificial eyes in order to aid future progression, the methodological approach chosen was a scoping review.

This review has led to a publication (See Appendix A for publication reference).

### 1.2.2 Method

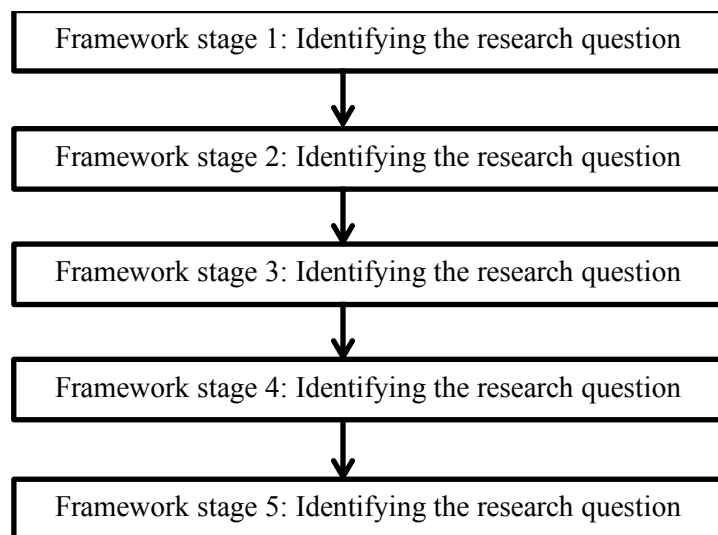
A scoping review methodology was chosen as it adds a narrative integration of the relevant evidence. Being of particular use when a topic has not yet been extensively reviewed, this scoping review describes the development trajectory of artificial eyes. Furthermore, being commonly undertaken to examine the extent, range and nature of research activity, this scoping review can highlight and create understanding regarding the period of stagnation. By identifying gaps in the literature, suggestions and recommendations for improvement to the current process can be made.

#### 1.2.2.1 Searching

#### 1.2.2.2 Screening and selection

This scoping review followed the five framework stages outlined by Arksey & O'Malley's (2005) (see Table 1).

Table 1: Arksey & O'Malley's (2005) scoping review framework stages



#### 1.2.2.3 Framework stage 1: Identifying the research question

Identification of the research question was based on two facets: firstly, the design and manufacturing of artificial eyes remaining the same since the 1960's despite rapid technological advancement; and, secondly, people's needs evolving over time. This led to the development of

the research question: ‘*What is known from the existing literature about the development of artificial eyes throughout the years?*’ It is envisioned that this review aids in creating an understanding of where and if technology can provide the tools required by artificial eye wearers.

Guiding the way search strategies are built, it is important to identify what aspects of the research question are important. To gain a rounded understanding of artificial eyes, a search strategy involving not only the historical and current development of artificial eyes, but also what influences the development (for example technological knowledge and the impact upon artificial eye wearers) was deemed necessary. Although these terms are ambiguous which can result in large numbers of references, gaining a sense of the volume and scope of the field is required before parameters are set (Arksey & O’Malley, 2005).

#### **1.2.2.4 Framework stage 2: Identifying relevant studies**

When conducting a search strategy, it is important to make decisions about time span and the language used in the search terms (Arksey & O’Malley, 2005). As artificial eyes are a relatively small field in comparison to others such as prosthetic limbs, it was decided that no time span or foreign language material would be applied at this stage with the view of amending this following completion of the search. By employing this approach, potentially relevant papers were not missed

In order to locate a wide variety of research relating to artificial eye development, a threefold search strategy was adopted. This involved developing a search strategy for electronic databases, internet searches of key sites and a bibliographic list search of all articles identified.

Electronic databases generated the majority of studies included in the scoping review (see Table 2). The majority of references were found on the electronic database MedLine Complete. The internet search was the least fruitful search strategy approach. The search strategy was devised to maximise the number of relevant studies reflecting three main areas of interest. These included: articles relating to the history of artificial eyes; articles related to the impact of wearing artificial eyes and articles related to the technological development of artificial eyes. A list of synonyms for these areas of interest was used to ensure that all possible articles were retrieved.

Table 2: Distribution of references by electronic bibliographic source

Database	Dates covered	Date Search	Hits
Science Direct	All years	06/08/16 – 16/08/16	71,190
PubMed	All years	16/08/16 – 20/08/16	6047
MedLine Complete	All years	22/08/16 – 23/08/16	2,190
Web of Science	All years	23/08/16 – 25/08/16	3,078
Scopus	All years	25/08/16 – 29/08/16	8,729
Google Scholar	1950 - current	29/08/16 – 31/08/16	8,070

The search was performed between August 6<sup>th</sup> and August 31<sup>st</sup> 2016 using six databases: Scopus; PubMed; MedLine Complete; Web of Science and Science Direct and Google Scholar. The search strategy used terms to denote ‘artificial eyes’ combined with a range of terms denoting its development (see Table 3 for search terms).

Table 3: Search Terms

Search Terms
Artificial eye(s)
AND
History OR development OR technology OR creation OR improvement OR techniques OR fabrication OR manufacturing OR design OR impression OR rehabilitation OR custom made OR design OR process OR fitting OR materials OR rapid prototyping OR computer aided design OR computer aided manufacturing OR digital imaging OR digital photography
AND
Enucleation or exenteration or evisceration or genetics or trauma or injury or infection or disease

The second and third stage of the search strategy was reference checking and internet search. Despite reaching a saturation point, 51 articles were identified. See Table 4 for the number of studies included according to source.

Table 4: Publications regarding the psychological impact of wearing artificial eyes and facial disfigurement

Bibliographic Source	N	%
Electronic databases	189	79
Reference checking	30	12
Internet search	21	9

### 1.2.2.5 Framework stage 3: Study selection

A total of 240 references were identified from the search strategy. A vast number of studies were generated that were not relevant to the main research question. Possible reasons for this include it being a small field (consequently few articles being produced) and various eye conditions requiring different surgeries and types of artificial eyes i.e., orbital prostheses. For these reasons, inclusion and exclusion criteria were applied to all studies that represented a best fit with the research question (see Table 5).

Table 5: Inclusion/exclusion criteria

Inclusion and exclusion criteria	
Inclusion	Exclusion
1950 onwards	Stock eyes, eye shells, implants, orbital prosthesis
Enucleation, evisceration, exenteration	Clinical and case reports
English language articles (including translations)	Reports on the condition and aftercare of artificial eyes
	Pre 1950

To assist in the study selection a PRISMA framework (Moher, Liberati, Tetziaff & Altman, 2009) was included (see Figure 2). The inclusion and exclusion criteria were applied to all 240 references using abstracts or the full articles where available. At stage one 81 articles were excluded for reasons including not being relevant (such as clinical reports, type of prosthesis and reports on aftercare). This left 87 articles for review in stage two. At this stage, a further 60 articles were excluded due to reasons including being unavailable through library resource and predating 1950. In the third stage the 26 articles included in the review were synthesised using charting as described in framework 4 of Arksey & O'Malley's (2005) scoping review.

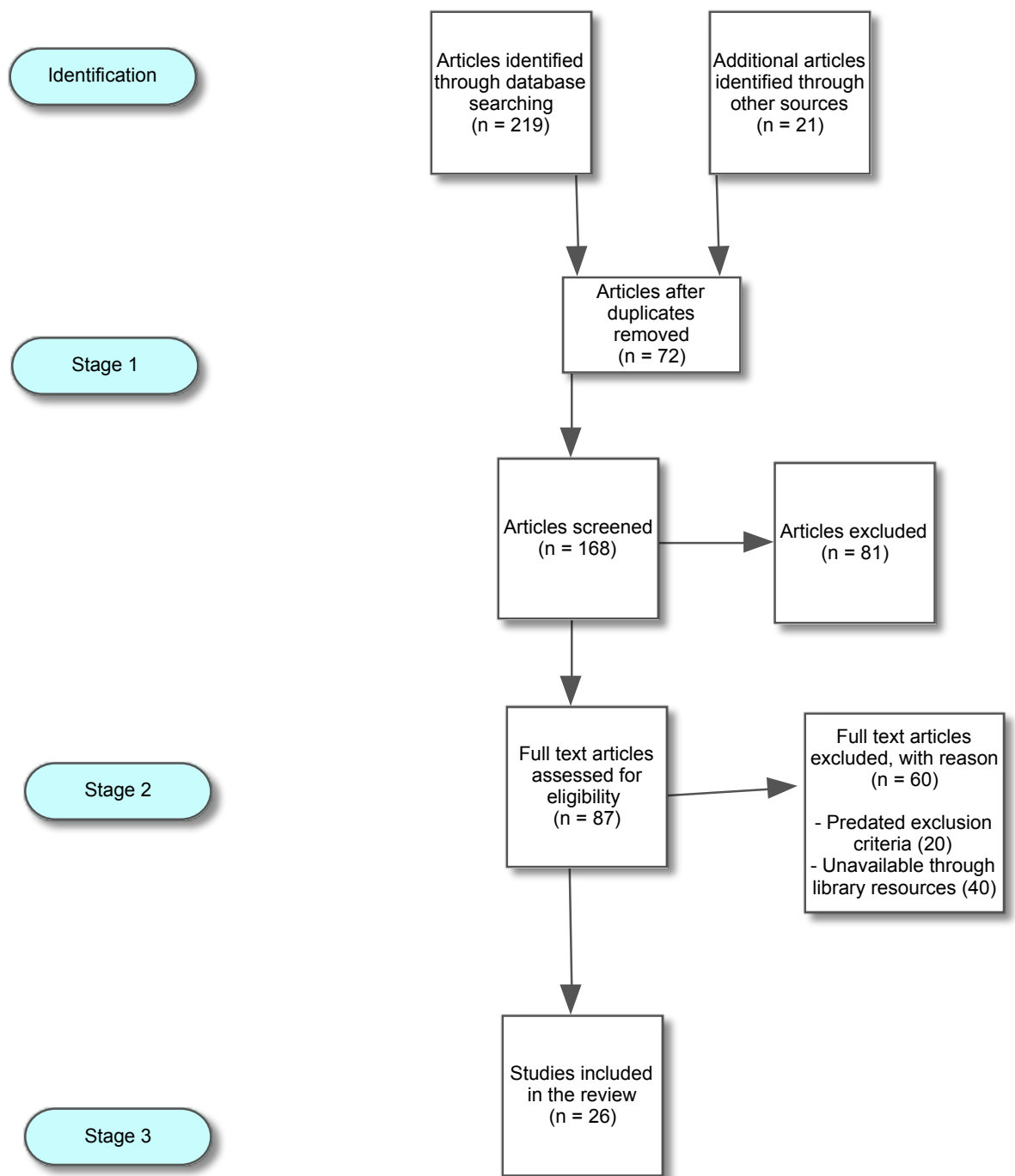


Figure 2: PRISMA 2009 Flow Diagram

#### 1.2.2.6 Framework stage 4: Charting the data

Charting is a technique that synthesises and interprets data by sorting key issues and themes (Ritchie & Spencer, 1994). A structured data charting form was created using the database programme Excel to systematically review the identified articles. All 26 articles were entered into the form that contained the following information: author(s), year of publication, study location; aim; typology, and, findings. These data formed the basis of the analysis.

### **1.2.2.7 Framework stage 5: Collating, summarising and reporting the results**

Rather than assessing the quality of articles, a scoping study aims to ‘map’ or identify literature in the field (Mays et al., 2001). Consequently, this study mapped the literature, reporting on articles most relevant to the research question rather than seeking the best evidence. This was done in two stages: firstly, basic numerical analysis and secondly, thematic analysis. Two themes around artificial eyes were extracted: the history of artificial eyes, and, the current design and manufacturing process.

### **1.2.3 Findings**

This section presents reflections on the study characteristics of the articles included in the review and the exploration of the two themes identified (the history of artificial eyes, and, the current design and manufacturing process).

#### **1.2.3.1 Study Characteristics**

The study characteristics of the twenty-six articles were: fifteen narrative inquiries reporting on the fabrication of artificial eyes; ten articles were historical reviews reporting on the development of artificial eyes, and one literature review article that described the different techniques for fabricating an artificial eye. The majority of the articles were derived from the USA (57% n = 15). In comparison, a far smaller proportion were derived from the UK (15% n = 4). New Zealand, Australia and Portugal accounted for 4% (n = 1 for each country) with articles from India and Brazil accounting for 8% (n = 2 for each country).

All articles related to the development of artificial eyes were historical reviews: 70% (n = 7) derived from the USA and 10% (n = 1) each from the UK, Australia and New Zealand. All narrative inquiries related to the fabrication of artificial eyes: 53% (n = 8) of articles derived from the USA, 20% (n = 3) derived from the UK, 13% (n = 2) from India and 7% (n = 1) each from Brazil and Portugal. Furthermore, one literature review (100%) derived from Brazil was also in regards to the fabrication of artificial eyes.

### 1.2.3.2 History of artificial eyes

#### 1.2.3.2.1.1 Ancient Egypt in the Dark/Middle Ages (3100BC – 1500AD)

Anecdotal reports and relics from ancient civilizations point to the first artificial eyes being made by the Egyptians in the 4<sup>th</sup> Dynasty (2613 – 2392 BC) and the ancient cultures of Babylon and Jericho (Artopoulou, Montgomery, Wesley & Lemon, 2006; Danz, 1990; Dyer, 1980; McCord, 1965; Raizada & Rani, 2007; Reisberg & Habakuk, 1990; Roman, 1994; Sobrinho, de Lima, da Glória, Mattos & Watanabe, 1986).

The Egyptians, Greeks and Romans embellished important statues, mummies and animals with artificial eyes (McCord, 1965; Sobrinho et al., 1986; Wilson, 1972). They were known as ‘art eyes’ (see Figure 3) and were made from precious stones such as earthenware, enamelled bronze, copper and gold. The orbit was filled with wax or plaster on top of which the precious stone was inserted to represent the iris of an eye (Danz, 1990; Deacon, 2008; Dyer, 1980; Kelley, 1970; McCord, 1965; Sobrinho et al., 1986). It is thought that these ‘art eyes’ were used for cosmetic purposes as a symbol of light and life in their religious beliefs (Artopoulou et al., 2006; Dyer, 1980).



Figure 3: “Art-eyes’ made by precious stones in Ancient Egypt. The College of Optometrists (2015). Early Artificial Eyes. Retrieved from [http://www.college-optometrists.org/en/college/museyeum/online\\_exhibitions/artificialeyes/early.cfm](http://www.college-optometrists.org/en/college/museyeum/online_exhibitions/artificialeyes/early.cfm)

With variations in techniques for fabricating artificial eyes, Alfred Lucas of Cairo developed a classification system (Wilson, 1972). Class I artificial eyes dated from the fourth Dynasty (Old Kingdom: 2900 - 2750 BC) to the thirteenth Dynasty (Middle Kingdom: 1803 – 1649 BC). They displayed a strong resemblance of the natural eye. The margins of the eyelids and sclera were



wedge-shaped with the former constructed from a narrow rim of thin copper and the latter being cut out of opaque white quartz or white crystalline limestone which was painted pink to represent the caruncle. The cornea was cut out of transparent quartz and fitted into a flat bottom circular hole in the stone inlay. A small shallow circular recess was drilled in the middle-back of the cornea, which was filled with dark brown/black resin to obtain the effect of the pupil (Wilson, 1972).

Class II were the largest and most common classification of artificial eyes covering the Old Kingdom (2980 – 2475 BC) to the Late Period of Ancient Egypt (663 – 525 BC) (Wilson, 1972). The cornea, iris and pupil were manufactured as one item, which was attached to the sclera. The sclera was made from opaque white glass and the fitting of the cornea and eyelids to the sclera followed the same procedure as Class I artificial eyes (Wilson, 1972).

Class III artificial eyes date from the Ptolemaic Dynasty of 30 BC and was only found in Roman mummy masks (Wilson, 1972). The sclera was wedge shaped and consisted of white crystalline limestone. A cornea hole was cut into the sclera where a corneal clear colour glass plug and a small cone shaped plug of black glass was inserted to represent the cornea, iris and pupil (Wilson, 1972). Despite both plugs being separated by a thin layer of copper foil, both suffered from corrosion and multiple internal fractures thus presenting a moth-eaten appearance (Wilson, 1972).

Class IV artificial eyes were similar in technique to Class I artificial eyes. The inlay consisted of one piece of transparent rock crystal. Whereas the front surface was polished and curved, the back surface was left unpolished and flat. The caruncle, iris, pupil and sclera were painted on the back surface of the inlay. With a poorer quality and design, this class of artificial eyes suffered from surface corrosion (Wilson, 1972).

Class V and VI artificial eyes dated from the late Egyptian period (26<sup>th</sup> Dynasty: 663 – 525 BC) to the Graeco-Roman periods (332 BC – 395 AD). The artificial eyes consisted of one piece of wood, glass or limestone that was painted to represent the natural features of a human eye. However, the representation of the human eye was of poor quality (Wilson, 1972).

From the 26<sup>th</sup> Dynasty through the Dark/Middle Ages (500 – 1500 AD), the making of artificial eyes was largely abandoned (McCord, 1965; Reisberg & Habakuk, 1990). It was not until the Renaissance period (1400 – 1700 AD) that artificial eyes saw a resurgence (Sobrinho et al., 1986).

#### 1.2.3.2.1.2 16<sup>TH</sup> Century

In the 16<sup>th</sup> Century, a French army surgeon, Ambroise Paré, described the use of artificial eyes to fit an eye socket. Paré described two types of artificial eyes: the ‘Hypoblephara’, which fitted underneath the eyelid, and the ‘Eklephara’ which fitted externally (see Figure 4) (Danz, 1990; Raizada & Rani, 2007). The ‘Eklephara’ consisted of a metal band covered with leather or cloth. One end of the band had a small oval plate covering the orbit, which was covered in leather and painted to resemble an eye including eyelids and brow. The other end of the band gripped behind the head (Danz, 1990; Kelley, 1970; McCord, 1965; Reisberg & Habakuk, 1990). Both types of artificial eyes were expensive, heavy, painful; to wear and lacked the moist quality of a normal eye (Martin & Cloudis, 1976).

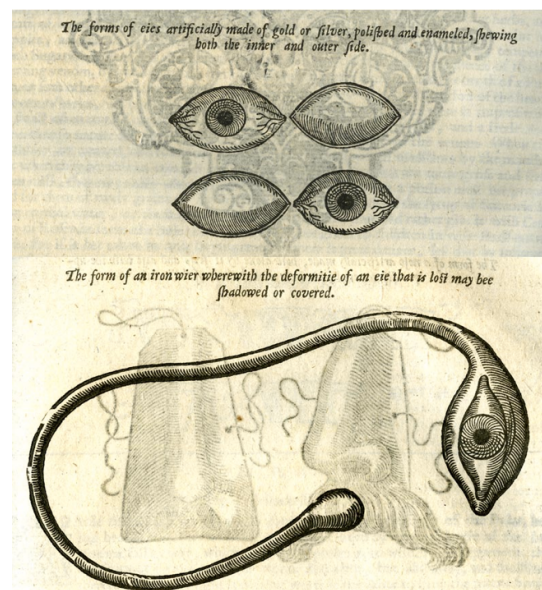


Figure 4: Ambroise Paré’s Hypoblephara (top) and Eklephara (bottom) eye. Llieva, P (2015). History, Science and Art of Ocular Prosthetics. Retrieved from <https://blogs.library.ucsf.edu/broughttolight/2015/04/24/1751/>

Although soda glass was much more durable and compatible to the tissue, erosion often occurred within 18 – 24 months resulting in the surface becoming rough, irritating the socket which would cause mucus discharge (Erpf, 1953). Artificial eyes made from soda glass were also susceptible to temperature change and having thin walls meant that they often fractured for no particular

reason. Despite these shortcomings, glass was better tolerated by the orbital tissue (Danz, 1990) and was seen as being able to eliminate the problems associated with metal artificial eyes (gold and silver) such as its weight and expense (Martin & Clodius, 1976).

The development of glass in manufacturing artificial eyes saw their shape change from spheres to a shell form (Artopoulou et al., 2006). Although the shell form caused irritation to the socket, it was less harmful than the metallic sphere form (Reisberg & Habakuk, 1990). Other failures of the shell form included not being able to adequately replace lost orbital volume thus trapping secretions in the void behind the prosthesis and being thin and fragile with sharp edges which put patients at risk of bruising the conjunctival bed (Kelley, 1970; Phillips, 1950; Reisberg & Habakuk, 1990).

#### **1.2.3.2.1.3 18<sup>th</sup> and 19<sup>th</sup> Century**

In the 18<sup>th</sup> Century, the importance of a custom-fit eye to the socket to overcome the above problems was being realised (Reisberg & Habakuk, 1990). The artistic and technical development of fabricating artificial eyes from glass saw it transitioning from having its monopoly in Venice where the making and fabrication of glass eyes were kept secret for many years, before moving to France, Italy and Germany (Danz, 1990; Goiato, Bannwart, Haddad, dos Santos, Pesqueira & Miyahara, 2014; Kelley, 1970; McCord, 1965; Raizada & Rani, 2007). In 1851 Mager and Gougelman established the first artificial eye service in America (Kelley, 1970; McCord, 1965).

Its relocation to Germany can be credited to a glassblower, Ludwig Müller-Uri, who produced dolls eyes that had good human-like qualities (Dyer, 1980). This led to a local physician commissioning him to supply eyes for his patients (Benson, 1977; McCord, 1965). Experimenting with new materials and methods for making human glass eyes, Müller-Uri first replaced lead glass with bone glass to produce the scleral shade. Using a clear crystal glass rod, he melted and applied thin strips of coloured glass evenly to create the iris colour. This was followed by twisting the heated rod and drawing the glass out to a thin pencil form that produced the stroma effect when applied to the iris thus creating a semi-transparent depth to the iris when the crystal cornea was melted in place (Danz, 1990; Raizada & Rani, 2007).

Müller-Uri then developed a new Cryolite glass made from arsenic oxide and Cryolite from sodium-aluminium fluoride which produced the whiteish grey sclera colour (Danz, 1990). This glass is hard and light, thus did not irritate the conjunctiva (Danz, 1990; Martin & Clodius, 1976; Reisberg & Habakuk, 1990).

Teaching his nephew the art of artificial eye making, Friedrich A Müller was later credited with developing the double wall glass prosthetics: a two-layered shell with a space between the layers. In 1894, the eye was named as the Snellen Reform Eye after Herman Snellen who introduced the eye to the main stream (see Figure 5) (Raizada & Rani, 2007; Reisberg & Habakuk, 1990). It was created by separating the anterior part of the glass ball that contained the iris and cornea from the posterior part (Martin & Clodius, 1976; Sobrinho et al., 1986). The anterior of the glass was blown to the desired shape whilst the posterior section was drawn back towards the anterior form until the desired thickness was achieved (Danz, 1990). The three most common Snellen Reform Eye shapes were the ovoid shape, the triangular shape to fit a deep hollow space at the 12 o' clock position of the socket, and one that had a prominent cut in the upper nasal area (Allen & Webster, 1969).



Figure 5: Glass shell eye prosthesis (left) and Snellen Reform eye prosthesis (right). Kelley, J. J. (1970), History of Ocular Prostheses. *International Ophthalmology Clinics*, 10(4), 713 – 719

By providing support at the anterior of the socket, creating lightness of weight and displacing soft tissues of the orbit around the prosthesis to fill volume thus eliminating a sunken appearance, the Snellen Reform Eye was comfortable for the wearer and offered excellent appearance (Allen & Webster, 1969; Danz, 1990; Reisberg & Habakuk, 1990). Furthermore, by eliminating many of the disadvantages of the shell eye such as replacing sharp edges with rounded edges, the Snellen Reform Eye did not suffer from surface tears and facilitated eye movement and eye retention (Kelley, 1970; McCord 1965; Phillips, 1950). The standard of the Snellen Reform Eye was at such a high standard that it replaced the shell eye for the majority of cases expect fittings over the atrophied or eviscerated globes (Kelley, 1970).

Despite the Snellen Reform Eye's vast improvements over the shell eye, they were uncomfortable in terms of pressure points causing irritation and discharge from large quantities of trapped tears and mucus. This was partially due to being unable to achieve best fit due to the abstract nature of the Snellen Reform Eye and without the help of an impression (Allen & Webster, 1969). This led to Hazard-Mirault in 1818 outlying the proper standards to shape the prosthesis allowing for the most comfortable fit (Danz, 1990).

Other disadvantages associated with glass eyes include fragility, surface erosion, imperfect fit and difficulty in moulding (Raizada & Rani, 2007; Schneider, 1986). Spontaneous breakage or implosion can occur in glass eyes as a result of faulty tempering of the glass during fabrication or sudden changes in temperature (Charlesworth, 1966; Erpf, 1953; McCord, 1965). The difficulty in handling and adjusting the material once the blowing is complete makes it essential to establish accuracy in visual alignment to reduce the requirement of remakes (Charlesworth, 1966; Dyer, 1980; Erpf, 1953; Sobrinho et al., 1966). With moisture wetting only the surface, it will evaporate quickly leaving the surface dry, tacky and uncomfortable to lid movements (Erpf, 1953). Furthermore, glass eyes are susceptible to mishandling and shock and superficial staining which over a period of time tends to darken areas where the pores of the glass have been exposed by erosion (Charlesworth, 1966; Erpf, 1953; McCord, 1965).

These defects led to experimentation with more durable materials such as aluminium, ivory, bone, vulcanite and celluloid was undertaken in the 19<sup>th</sup> century.

In 1881 Neiden experimented with grey vulcanite, a hard rubber product consisting of 48 parts of rubber, 24 parts of sulphur and 48 parts of zinc oxide (McCord, 1965; Reisberg & Habakuk, 1990). When the artificial eye was formed, the grey vulcanite would be softened in hot water then placed in a mould or manipulated by hand (McCord, 1965). In 1884 Van Duyse provided a detailed method of moulding vulcanite into an artificial eye (Reisberg & Habakuk, 1990). However, the material was still too grey, thus the use of vulcanite was discarded as a viable material (Dyer, 1980).

Having similar properties to vulcanite, celluloid was also a less than adequate replacement for the glass eye. Both vulcanite and celluloid were vulnerable to socket secretion, had a lifespan of a

few months due to rapid deterioration, was uncomfortable and lacked the realism of the glass eye (Kelley, 1970; Reisberg & Habakuk, 1990; Sobrinho et al., 1986). Van Duyse and Froelich also experimented with ivory, aluminium and horn, recommending its use as it was unbreakable and less costly than glass. However, these materials were heavy and replication of the natural eye was hard to achieve (McCord, 1965; Sobrinho et al., 1986).

The ineffectiveness of these new materials saw the fabricating of glass eyes using sand with a low iron oxide content being exclusively used until 1939 when resin was introduced (Benson, 1977; Deacon, 2008; Kelley, 1970).

#### **1.2.3.2.1.4 20<sup>th</sup> Century**

With the turn of the 20<sup>th</sup> century artificial eye services were becoming more regulated. In 1917, the artificial eye industry came under the Defence of the Realm Regulations where the number and material of all eyes in use had to be submitted (Roman, 1994). During this time, artificial eyes were made of glass and supplied in standard sizes and colours that could not be altered (Bartlett & Moore, 1973).

With the advent of World War I, glass artificial eyes were extended from military use to general population use (Deacon, 2008; Roman, 1994). Entering a Second World War in 1939, supplies of artificial eyes and glass became unobtainable in Germany resulting in the United States temporarily relying on stock eyes to meet the need of the military and civilian population (Artopoulou et al., 2006; Kelley, 1970; Martin & Clodius, 1976; Sobrinho et al., 1986). In 1946, the Veterans Administration based in America, initiated the 'Plastic Artificial Eye Program' to train artificial eye prosthetists to make artificial eyes for a large number of disfigured and disabled veterans (Cerullo, 1990).

The scarcity of imported glass during this time led to a search for a more suitable material such as acrylic resin, polyvinyl and urethane (Bartlett & Moore, 1973; Sobrinho et al., 1986). The materials were assessed for aesthetic quality, retention of luster when polished and subjected to use as well as being biocompatible, thus being non-toxic and not produce inflammatory reaction (Erpf, 1953; Sobrinho et al., 1986). With good translucence and intrinsic and extrinsic colouring capabilities as well as its tolerance to human tissue (as observed in its use in dentistry), saw methyl methacrylate being deemed as the most suitable material for making artificial eyes (Artopoulou

et al., 2006; Danz, 1990; Cerullo, 1990; Deacon, 2008; Goiato et al., 2014; Kelley, 1970; Martin & Clodius, 1976; Raizada & Rain, 2007; Schneider, 1986).

Acrylic resin is a polymerised form of methyl methacrylate. It is lightweight, easy to mould, fit and adjust; durable; has good colour permanence; is easily fabricated and is inert in nature (Artopoulou et al., 2006; Goiato et al., 2014; Raizada & Rani, 2007; Reisberg & Habakuk, 1990). Furthermore, acrylic resin does not erode in socket secretions, is resistant to rough handling; easy to work with and provides an aesthetic appearance (Benson, 1977; Deacon, 2008; Schneider, 1986; Sobrinho et al., 1986). With moisture to the surface of the eye contained within the plastic; thus, acting as a tissue lubricant, the plastic eye is superior to the glass eye (Erpf, 1953; Sobrinho et al., 1986).

The few disadvantages associated with plastic artificial eyes such as not being fully scratch resistant, not capturing iris depth and colour fading, are all related to poor restoration. With the right equipment and materials, these obstacles were overcome (Erpf, 1953).

#### **1.2.3.2.2 Fabrication of artificial eyes**

Initially, acrylic artificial eyes were developed based on the shell and reform form used for glass eyes (Kelley, 1970). Thus, similar problems such as soreness due to ill fit, lack of mobility and lid distortion due to incorrect shape occurred (Dyer, 1980). Through extensive research conducted by the United States Naval Dental and Medical Schools, it was realised that due to the individuality of each enucleated socket, artificial eyes need to be custom made (Artopoulou et al., 2006; Bartlett & Moore, 1973; Kelley, 1970; Martin & Clodius, 1976; Matthews, Smith, Sutton & Hudson, 2000; Reisberg & Habakuk, 1990; Sobrinho et al., 1986).

Although custom made artificial eyes necessitates the work of a skilled artist making it a time consuming and costly process, they permit accurate colouring of the iris, veining and tinting of the sclera making it more cosmetically satisfactory to the patient, their family, friends and the wider society (Cerullo, 1990). Accurate fitting of the socket, minimising tissue distortion, improved facial contours and increasing the degree of motility are also achieved in custom made artificial eyes (Allen & Webster, 1969; Cerullo, 1990; Jain et al., 2010; Matthews et al., 2000). This allows even distribution of volume and weight in the socket reducing long-term discomfort and producing better cosmetic outcomes (Dyer, 1980; Sobrinho et al., 1986). Furthermore, by providing close contact with the soft tissue, the artificial eye allows normal tear secretion, volume

replacement, adequate orbital fat and absence of socket inflammation (Allen & Webster, 1969; James, Ellis & Foulds, 1976; LeGrand & Hughes, 1990; Raizada & Rani, 2007). These advantages result in the artificial eye moving like a natural eye following almost simultaneously with the patient's natural eye (Dyer, 1990).

In order to approximate the natural eye, accurate records of the posterior wall and its relation to the palpebral and the extent of the superior and inferior fornices of the palpebral is required in order to provide sufficient support (Matthews et al., 2000). Without sufficient support, the eyebrow can fall nasal-wards, the eyelid muscles can become weaker leading to entropion (where the eyelid is rolled inwards towards the socket) and the edge of the eyelids can become inverted (Dyer, 1990). Currently, this is achieved by a method known as the impression technique. The fabrication of an artificial eye using the impression method establishes the defect contour and the iris and sclera can be individually characterised offering better aesthetic and more precise outcomes (Allen & Webster, 1969; Artopoulou et al., 2006; Goiato et al., 2014; Manoj, 2014).

The initial impression technique in the mid-1940s reported partial success in fitting artificial eyes (Allen & Webster, 1969). However, by pressing the paste (which was often heavy) into the socket to fill all spaces and smoothing out the conjunctiva folds and wrinkles often led to overfilling (Allen & Webster, 1969). Furthermore, by only testing the wax mould of the impression for a short period of time, thus not giving enough time for the orbicularis muscle to reflex from the foreign object, a good fit was not always achieved (Allen & Webster, 1969; Goiato, dos Santos, Moreno, Haddad & Turico, 2013). These techniques often caused discomfort and ignited fear in the patient as a result of the process, materials and equipment used (Goiato et al., 2013).

Advancements in this method include the direct/external impression technique and the modified impression technique. (Artopoulou et al., 2006; Goiato et al., 2013; Goiato et al., 2014). Whereas the former involves injecting low viscosity alginate or reversible hydrocolloid material directly into the socket, the latter places a stemmed tray into the eye socket for the material to be inserted into. Variations to the modified impression technique include attaching a solid suction rod to an existing prosthesis and investing it in an irreversible hydrocolloid mould before replacing it with clear acrylic resin (custom ocular tray impression); applying ophthalmic alginate to a suitable stock eye which is inserted into the socket for a definitive impression (stock ocular tray modified technique); trimming/polishing a stock eye or using alginate or soft wax which is invested and processed (ocular prosthesis modification technique); and adapting baseplate wax around half an appropriately sized steel ball to create a wax blank which is tested in a socket and adjusted as



required (wax scleral blank technique) (Allen & Webster, 1969; Bartlett & Moore, 1973; Benson, 1977; Cerullo, 1990; Deacon, 2008; Manoj, 2014; Matthews et al., 2000; Raizada & Rani, 2007).

Despite these advancements, all impression techniques are unable to design the front of an artificial eye, thus is unable to manipulate the eyelids to their proper opening and shape (LeGrand & Hughes, 1990). Furthermore, the effectiveness of these techniques is often dependent on the prosthetists expertise and availability of materials (Goiato et al., 2014).

Following an impression of the socket, a plaster mould is created using molten wax to produce a replica (Deacon, 2008; Raizada & Rani, 2007). Whilst the wax is being cured, the iris is produced. The iris disc is sized based on the observations of the natural eye and measuring the distance from the facial midline and pupillary light reflex allowing a guide for a centrally placed iris ensuring correct alignment (Deacon, 2008). Several methods such as paints, pigments and papers have been used: all of which rely upon the artistic skills of the skilled technician. The iris can be painted either on paper discs, ethyl-cellulose discs or directly on the acrylic resin sclera using watercolours or oil paints (Benson, 1977; Cerullo, 1990; Goiato et al., 2014). Relying upon the artistic skills of the skilled technician led to the development of creating an iris using digital imaging.

Digital imaging involves taking a digital photograph of a patient's natural iris. The photograph and the natural iris are compared where adjustments are made to the colour, brightness, contrast or hue using graphics software, if required (Artopoulou et al., 2006). Once aesthetically pleasing, the image is covered with 3 light coats of water resistant spray before attaching it to the ocular disc and then the wax pattern (Artopoulou et al., 2006).

Digital imaging provides aesthetic quality as it closely replicates the natural iris with minimal colour adjustment and modifications needed. Furthermore, this method is simple and practical as well as being less time consuming than the traditional method as it requires minimal artistic skills (Artopoulou et al., 2006; Goiato et al., 2014). However, special digital photography equipment and computer software is needed (Artopoulou et al., 2006; Goiato et al., 2013). Furthermore, the light instability of photographic dyes results in the coloured iris fading quickly (Cerullo, 1990).

Following insertion of the iris into the wax pattern, it is cast in clear acrylic resin and cured under compression (Deacon, 2008). Once cooled, red cotton thread is adhered to the sclera using liquid monomer and polymer clear powder (Benson, 1977; Cerullo, 1990; Goiato et al., 2014). The artificial eye is then coated with a layer of clear acrylic resin before being cured under heat and pressure, cooled and polished (Bartlett & Moore, 1973; Benson, 1977; Cevik et al., 2012; Deacon, 2008; Goiato et al., 2014). See Figure 6 for a custom made artificial eye.



Figure 6: A custom-made artificial eye showing fine detail. Erickson Labs Northwest (2015). Artificial Eye. Retrieved from [http://www.ericksonlabs.com/v/Artificial\\_Eyes/our\\_process.asp](http://www.ericksonlabs.com/v/Artificial_Eyes/our_process.asp)

The current methods used in the fabrication of artificial eyes are labour intensive and time consuming with the end result being heavily dependent on the experience of the prosthetist. The need for both functional and aesthetic consideration makes the fabrication of artificial eyes a challenging task in prosthetics.

### **1.2.3.3 Limitations of the scoping review**

‘Mapping’ the literature produces a vast amount of research, thus creates difficulty in deciding breadth over depth. However, the framework of a scoping review allows prioritisation of certain aspects of the literature. Whilst this can keep the scope of the research question tight, it may also result in other important aspects being missed. The development and application of the inclusion and exclusion criteria after the search strategy allows for the most relevant articles to be included based on familiarity with the literature. From the twenty-six articles included, two themes were developed: (1) history of artificial eyes, and, (2) fabrication of artificial eyes.

Most of the studies were narrative inquiries. Whilst the majority of articles were related to the current design and manufacturing of artificial eyes, there has been minimal changes in their

technological development. Variations of the impression technique have been developed through trial and error by the prosthetists, rather than specific research. As this specialty area grows, quantitative studies should be undertaken that would further enhance our knowledge and lead us closer to incorporating technological advancements into the design and manufacturing process.

Though this scoping review found articles relating to recent developments, the majority were published by AEP's showing techniques they have developed. Whilst this allows for transferability, its subjectivity means the process has not been validated from the governing bodies, further increasing variations in product outcomes (NICE, 2014). With the need to meet patient expectations and the ever-increasing availability of medical technology (Eggbeer, 2008; Evans et al., 2004), a resource drive is needed in this field to produce artificial eyes that are in keeping with the latest technological advancements whilst producing the best possible outcome for the patient.

By not being discriminate in the location of where articles and/or studies were undertaken resulted in the review highlighting various techniques employed in the manufacturing and fitting process of artificial eyes. Consequently, some of these techniques may not be applicable or used in other countries or services. Through continuous research recommendations and guidelines may be produced to create a standard of care for all wearers within and between countries.

### **1.3 Psychosocial implications of childhood cancer and its secondary affects**

#### **1.3.1 Introduction**

When an individual faces the diagnosis of cancer, they are not only subjected to changes in their physical health as a consequence of the disease, treatment and experiences (von Blanckenburg et al., 2014; de Vries & Friedrich, 2014; Mellblom et al., 2014; Patenaude & Kupst, 2005; Van Dongen-Melman, 2000), but also changes to self-perception and social relationships. This can lead to maladaptive coping mechanisms such as denial and self-blame (de Vries & Friedrich, 2014; Patenaude & Kupst, 2005). Although this may result in adverse lifelong psychological distress (Van Dongen-Melman, 2000), it is often underdiagnosed in the oncology setting (van Dongen-Melman, 2000). Unfamiliarity of the symptoms, misinterpretation of them, individual's not subscribing to the pre-conception of their experience, shortcomings of the measuring instruments used or lack of time and/or resources are common reasons for this (Deshields & Nanna, 2010; Patenaude & Last, 2001). With improved treatment and outcomes resulting in

increased survivorship, there has been a recent change of focus from the medical to the psychosocial aspects of the illness: encompassing the experiences of those directly and indirectly affected (Ross, Lipper, Abramson & Preiser, 2001). This section will provide an overarching appraisal of the significant factors that have been found to play a role in those (in)directly by cancer and its secondary affects.

A systematic search was conducted using Bournemouth University's iteration of the EBSCO Discovery Service tool. A literature review was undertaken that focused on the psychological effects (wellbeing, psychological impact, mental health and clinical implications) of the main childhood cancers (Rb, Leukaemia, Neuroblastoma, Brain and Spinal cord tumours, Wilms tumour, Lymphoma and Rhabdomyosarcoma) and on disfigurement, amputation and prosthetic use (facial disfigurement, limb loss and enucleation).

The literature presented here is by no means a comprehensive review of the research available in this area. Rather, it is intended to highlight pertinent areas related to the rationale for this thesis. This review presents the need to give a voice to the experiences of those directly and indirectly affected by cancer and its on-going implications.

### **1.3.2 Childhood cancers**

Over the last twenty years, a plethora of research has examined the experiences and psychosocial outcomes of children with cancer. Findings point to a correlation between increased survival rates and an increase in associated psychosocial difficulties (Li, Chung, Ho, Chiu & Lopez, 2012). Potential explanations include, delays and/or difficulties in achieving developmental tasks as a result of the cancer; vulnerability to stress of illness, and, an inability to understand the reasons for their illness (Li et al., 2012). The distress experienced can manifest in anxiety, depression, fear, uncertainty and feelings of hopelessness (Deshields & Nanna, 2010; Mellblom et al., 2014; Li et al., 2012; Van Dongen-Melman, 2000; Wiener et al., 2014). These symptoms are often exacerbated by: loss of independence; reduced energy levels; a lack of enjoyment in once enjoyable activities; communication and emotional difficulties; discrepancies between anticipated and achieved goals and maintenance of unhelpful coping mechanisms (von Blanckenburg et al., 2014).

McDougal & Tsonis (2008) undertook a systematic review examining the quality of life of children with cancer. Articles included in their search strategy were those published between 2001-2008, were in English and used quantitative measures and statistical tests that compared health-related quality of life (HQRL) or Quality of Life (QoL) of children with cancer with population norms or matched comparisons group. Their findings showed similarity in scores between both population groups in the psychological, social and physical domains of the HRQL and QoL: with the greatest difference being in the physical wellbeing. Furthermore, personal and environmental factors including older age at diagnosis, longer time since diagnosis, and certain cancer and treatment types, were negatively correlated with the psychological, physical and social domains for children with cancer.

Giving meaning to these findings, Han et al (2011) explored the experiences and feelings of 29 7-14 year old Chinese children with Leukaemia during the treatment stage. Face to face in-depth interviews were undertaken and analysed using content analysis. Two main themes emerged: (1) experience and feelings in the early stage (0-3 months), and, (2) experiences and feelings in the intermediate and later stage (>3 months). Their findings point to a series of negative experiences and feelings within the first three months of treatment. These included not adapting to hospital life, missing family and friends, enhancing dependence, and, feelings of worry, grief, irritability and unhappiness. As the child progressed through the treatment, adaption occurred, however this was depended on personality and age factors. Extroverted and younger children were found to adapt to hospital life more easily than older children and those with more introverted characters. Potential explanations for this finding include motivation towards learning and understanding of friendships. Being more aware of the impact of treatment interruptions in their personal and educational learning as well as the effect on maintaining current and forming new friendships, older children (7-14 year olds) were found not to be as adaptable to hospitalisation.

Alongside adapting to their treatment, psychosocial difficulties in children with cancer are often related to developmental changes. Using an Interpretative Phenomenological Analysis (IPA), Griffiths, Schweitzer and Yates (2011) interviewed 9 families where a child was diagnosed with cancer, on two separate occasions over a 12 month period in Australia. With experiences being pertinent to the stage of treatment, changes over time were found and formed 5 themes. These were: (1) experience of illness; (2) upside of being sick; (3) refocusing on what is important; (4) acquiring a new perspective, and, (5) experience of returning to wellbeing.

Examining the psychosocial needs of adolescents with Acute Lymphoblastic Leukemia (ALL), Kahalley et al., (2012) examined the occurrence of cognitive, behavioural and emotional concerns. Measures used included The Behaviour Rating Inventory of Executive Function (BRIEF), The Diagnostic Interview for Children and Adolescents IV, parent version (DICA-IV), Wechsler Abbreviated Scale of Intelligence (WASI), Wechsler Processing Speed Index (WISC-IV) and the Delis-Kaplan Executive Function System. Quantitative analysis found 82% of children with ALL exhibited cognitive, emotional and behavioural concerns with the latter being more prominent. The concerns raised were found to be in multiple domains of functioning with particular overlap between behavioural and cognitive domains. A potential explanation for this is as a child with cancer ages, they start to experience cancer related concerns including the (experience and consequence of) physical, psychological and social changes. These concerns can manifest behaviourally and are expressed indirectly to their care team and more often when family members are not present.

As well as occurring during the treatment phase, psychosocial difficulties have been evidenced in children in remission. Reinfjell (2009) assessed the mental health and psychological adjustment of 40 children in remission from ALL. Employing a cross sectional design, participants completed the Child behaviour Checklist (CBCL), the Youth Self Report (YSR) and the Strengths and Difficulties Questionnaire (SDQ). Results showed significant behaviour symptoms in children with ALL compared to healthy controls. The finding that problems can be seen several years after diagnosis demonstrates the need for close awareness of adjustment related to mental health and psychosocial functioning in children in remission from ALL.

This is supported by Ellertsen, Rannestad, Indradavik and Vik (2011) whom explored the psychosocial health in 50 Norwegian children and adolescents three years post diagnosis compared with healthy controls from the perspective of the children, their parents and teachers. Findings from the Strengths and Difficulties Questionnaires and the Achenbach System of Empirically Based Assessment Questionnaire revealed that children with cancer are at a higher risk of experiencing emotional problems and have poorer academic performance when compared to their peers. The emotional problems were consistently reported by parents, teachers and children themselves: particularly in children with brain tumours and those with late effects.

Alongside the normal developmental changes such as development of own identity, decision making and intimate relationships, children with cancer are also at risk of (chronic) health

problems and may be affected by academic achievement, impaired or decreased social relationships and low self-esteem.

Low self-esteem, also known as self-concept is multi-dimensional and is built from the environment, interpersonal relationships, emotionality, bodily experience, family life and academic life (Tremolada, Tavern, Bonichini, Basso & Pillion (2017). Facing additional burdens as a result of cancer, psychological adjustment needs to take place to create an identity and self-esteem and to reduce the negative impact on schooling and forming relationships. Undertaking a cross sectional study, Li, Chung, Ho, Chiu and Lopez (2012) described the psychosocial wellbeing and QoL of Hong King Chinese children with cancer. 137 9-16 year olds completed various psychosocial Questionnaires including The Center for Epidemiologic Studies Depression Scale For Children (CES-DC), the State Anxiety Scale for Children (CSAS-C), the Rosenberg Self-esteem Scale (RSES) and The Paediatric Quality of Life Inventory (PedsQL). Their findings show a significant number of children with cancer had low self-esteem and high levels of depression: with greater symptoms of depression being associated with higher levels of anxiety, low self-esteem and poor QoL.

Screening for risk in self-esteem perception and schooling difficulties, Tremolada et al., (2017) recruited 25 (pre)adolescent children and their parents five years following Leukaemia treatment. Results from the Multidimensional Self Esteem Test and schooling questionnaire showed that 43.5% of parents reported their child as having difficulties at school, with children undergoing high risk treatment being at a higher risk of experiencing low self-esteem.

This is supported by Weintraub et al., (2011) who found that children with Rb perceive their quality of life at school as lower compared to their healthy peers. Examining the developmental functioning of infants and young people with Rb through the first five years of their life, Willard et al., (2014) found a low-level range of functioning which led to a significant decline over time. With educational environment, time away from school, relationship and support from peers and teachers being crucial factors in children with cancer quality of life at school, continual assessment on their functioning as they age is required (Bassell, 2001; Willard et al., 2014).

Undertaking an exploratory study, Tougas, Jutras, Bigras and Tiurigny (2015) examined 49 children with Leukaemia examining their perceptions of benefits and burdens in relation to their illness on school engagement. Analysis of variance on a (self-developed) questionnaire

measuring school engagement and an interview focusing on the impact of cancer on their lives found that school engagement was greater for those who perceived the presence of benefits. The majority of (older) participants reported benefits from having cancer, specifically at an interpersonal level. The burdens reported by half of the participants revolved around physical and psychological concerns. The latter was found to be correlated to the amount of time elapsed since their diagnosis.

Physical and psychological functioning was also found to be affected by sleep and fatigue. Wu et al (2008) undertook a phenomenological study to explore experiences of fatigue reported by 14 Chinese children with Leukaemia (aged 7-18 years). Alongside impaired physical and psychological functioning, reduced cognitive welling and negative effects on the child's family, social interactions and schooling were reported.

Psychological wellbeing is maintained by having a good social support network. For children, this inevitably will revolve around their schooling and family. In regards to schooling, Katz, Leary, Breiger and Friedman (2008) used an observational method to assess the quality of peer relationships in 51 7-12 year olds with ALL compared to healthy children. Results from an audiotaped free-play session between dyads: (1) the child with cancer and their best friend, and, (2) both children without cancer; indicated that the dyad with a child with cancer were more likely to experience disengagement than dyads with healthy participants. This finding suggests a compromise in some specific areas of the relationship between a child with cancer and their best friend.

The other risk factor for poor psychological functioning in children with cancer are changes in family dynamics (Rodriguez, Dunn, Zuckerman, Vannatta, Gerhardt & Compas, 2011). This includes both the families view of the child with cancer functioning and the functioning of the family themselves as a result of having a child with cancer.

Parents often underestimate their child's understanding about their illness and their resilience with assumptions being made on what a good quality of life for their child is. Morris, Blount and Cohen (1997) investigated differences in child adjustment and family functioning of 33 children with Leukaemia and 32 healthy children. Results indicated that families of children with Leukaemia rated themselves as less cohesive and more conflicted than families of healthy children. Although no differences were found in child adjustment between children with



Leukaemia and healthy children, different associations between family functioning and child adjustment for the two groups were found. Greater family conflict and less encouragement of autonomy were associated with greater externalising symptoms by the child, whereas lower family cohesion, less open expression and higher levels of control within the family were associated with more child internalising problems.

Chao, Chen, Wang, Wu & Yeh (2003) explored the psychosocial impact of cancer on 24 8-17 year old Taiwanese children and their parents as well as the degree of depression experienced by these children. The results showed that whereas the children did not perceive significant changes in their psychological adjustments, parents indicated significantly lower mood and a slight decrease of number of friends in their child. This finding is supported by Weintraub et al. (2011) whom found that when asked to rate their child who has Rb quality of life, parents perceived their child's general and emotional health as poor and reported more emotional and behavioural problems.

Litzelman, Catrine, Gangnon and Witt (2011) finding that the child's clinical characteristics appeared to be related to poor parental QoL could account for the discrepancy between the parents view of their child's functioning and the child's view of their level of functioning. This relationship was found to be mediated by the parent's burden and stress. With parents of children with cancer exhibiting higher levels of physiological symptoms of stress than parents of healthy children (Pollock, Litzelman, Wisk & Witt, 2012), interventions that can reduce burden and stress may mitigate the detrimental effects of caregiving (Litzelman et al., 2011).

Bougea, Darviri and Alexopoulos (2011) conducted a systematic review to summarise the existing evidence regarding interventions towards reducing stress in parents with a child with Leukaemia and their effect in child and family wellbeing. Various cognitive behavioural and problem skills training programs were used on different platforms including group therapy, education, CBT and (un)structured counselling. The behavioural, psychological and physiological measures, self and observer reports highlighted CBT as being able to provide both medium and long term benefits in managing distress in parents of, and children with cancer.

With the majority of parents of children with cancer acting as their caregiver, they are at an increased risk of experiencing psychological distress due to changes in family dynamics, socioeconomic status, daily routine, employment and financial issues (Dahlquist et al., 1993; Fotiadou, Barlow Powell & Langton, 2008; Lou & Chan, 2002; Sawyer et al., 1993; Patenaude

& Last, 2001; van Dijk et al., 2008; Yamazaki, Sokejima, Mizoue, Eboshida & Fukuhara, 2005). Malpert et al. (2014) found that perceived caregiver strain was significantly associated with parent emotional distress. This distress is heightened at the time of diagnosis with reactions such as shock, disbelief, despair, sorrow and anger being experienced (McGrath 2002, Kars et al. 2003; Nagayoshi et al., 2016; Patistea et al. 2000; Tarr & Pickler 1999; Varni et al. 1999). This is often followed by anxiety, depression, denial, anger, and low self-esteem which often occur in response to fear of a future relapse or uncertainties about the child's future development (Fotiadou et al., 2008; Van Dongen-Melman, 2000; Williams, McCarthy, Eyles & Drew, 2013).

The impact of the illness and treatment on the child's current functioning, and, how parents live day to day with the threat of future relapse or further long-term physical and psychological complications, emerged as key themes in Vance, Eiser & Horne, (2004) IPA study. Parents were found to actively attempt to provide replacements or alternatives for the gaps in their child's life.

The fear of uncertainty over their child's future health status often results in parents becoming overwhelmed and misunderstanding what is being asked of them. This can result in delayed treatment decisions that may lead to bad effects or put the child with cancer at a disadvantage (Panton et al., 2009). General and disease specific information that is presented at a level each parent is able to understand improves parental adjustment, increases their understanding of risk, potential outcomes and enhances compliance with treatment (Beaudin, Intille & Morris, 2006; King, King, Rosebaum & Goffin, 1999). This finding is supported by Panton et al. (2009) who found that parental comprehension of treatment and risk in Rb treatment is multifactorial and involves language fluency, age, life experience and education attainment. Furthermore, they found visual information more effective than the spoken word when communicating to individuals under stress to make medical decisions. Developing support networks, whether that is with their family, friends or with other people who have/are going through the same experience at the start of their journey is vital for psychological adjustment.

Aiming to gain an insight into the lived experience of parenting a child with Leukaemia during treatment, Kars, Duijnste, Pool, van Delden & Grypdock (2007) conducted a grounded theory study. From a one-time individual interview with 12 mothers and 11 fathers of children with Leukaemia, the core concept of 'being there' was developed. This refers to the parental response to the perceived vulnerability of the child and their need to give meaning to parenthood via protection (guarding their child from the negative affect of illness and treatment) and preservation (the way the parent influences their child's perception of their own life).

Psychosocial difficulties continue to be experienced by parents following their child's remission of cancer. Ljungman, Cernvall, Gronqvist, Ljotsson, Ljungman & Essen (2014) conducted a systematic review on the positive and negative long-term psychological late effects for parents of children with cancer 5 years post diagnosis and 2 years after treatment. Whilst findings from 15 papers (a mix of quantitative, qualitative and mixed methods) showed psychological functioning, coping and family functioning as being in the normal range, a subgroup reported a clinical level of general psychological distress including PTSD symptoms, worry, disease-related thoughts and feelings as well as Post-Traumatic Growth (PTG). Factors associated with the long term effects include parents' maladaptive coping during earlier stages of their child's disease, and the child's current poor adjustment.

Evaluating prospectively the association between parental anxiety during treatment for childhood Leukemia and Posttraumatic Stress Symptoms (PTSS) after treatment ends, Best, Streisand, Catania and Kazak (2001) conducted a longitudinal follow-up study of 113 parents of children treated for Leukemia. Using hierarchical multiple regression, data from self-reported questionnaires completed during and after treatment found anxiety to be a significant predictor of later PTSS for mothers but not fathers. Furthermore, anxiety self-efficacy, PTG and length of time since treatment ended were associated with parental avoidance.

This finding is supported by Turner-sack, Menna, Setchell, Mann, and Cataudella (2015) whom found that both parents and siblings had similar levels of PTG. Alongside the use of more active coping mechanisms, younger parents with high life satisfaction and older siblings where there is a longer time since their brother or sister was diagnosed report less psychological distress.

With the aim of reducing PTSS Kazak (2004) conducted a randomized wait-list control trial of a newly developed 4-session, 1-day intervention called the Surviving Cancer Competently Intervention Program (SCCIP). 150 adolescent survivors (aged 11-19) and their mothers, fathers, and adolescent siblings (150 families) attended the program which was based on CBT treatment with family therapy. The significant reduction in intrusive thoughts among fathers and in arousal among the children in cancer supports the use of brief interventions to reduce PTSS in this population group (Kazak, 2004).

Reflecting on parents experiences of their child's cancer journey can offer another dimension to the psychosocial impact they may experience. Cox (2018) examined 38 Australian caregivers of children with cancer experiences of their child's cancer diagnosis and early treatment period retrospectively 5 years post diagnosis. Through analysing interviews using a grounded constant comparison approach, the concept of neo-normal was developed. This concept referred to the threat of cancer, loss of control and certainty in protecting their child's wellbeing and reliance on medical interventions. The initial experience of loss was found to be quickly redefined through immersing themselves in their child's care. Participants specifically spoke of acquiring new knowledge and skills through the treatment process, the importance of developing and maintaining routines to normalise treatment and of having a positive relationship with nursing and clinical staff.

Normality has been found to be an essential part of caring for a child with cancer. Earle, Clarke, Eiser & Sheppard's, (2006) longitudinal cross section study of 32 mothers of children recently diagnosed with ALL provides an understanding of how normal family life is compromised. The findings from their thematic analysis showed mothers understanding of the importance yet difficulty in achieving a normal life. The differences were found in how mothers perceived lack of normality: from frustration and disappointment to managing and acceptance; showed that striving for normality in order to facilitate their child's adjustment can be both helpful and damaging when it proves elusive.

The potential elusiveness of normality combined with compromises to their day to day living not only requires adjustment on behalf of the parents but also a level of resilience. Eilertsen, Hjemdal, Le, Diseth, Reinfjell (2015) assessed resilience factors and mental health among parents of children surviving ALL and parents of healthy children. Results from the Resilience Scale for Adults and the General Health Questionnaire showed parents of children with ALL had significantly lower levels of resilience than parents of healthy children. Furthermore, resilience factors of family cohesion, good self-perception and future planning were positively associated with mental health.

An individual's level of resilience often dictates how they will cope with stressful life situations. In terms of children with cancer, Van Dijk et al. (2009) reported that Rb survivors who use task and avoidant orientated rather than emotion orientated mechanisms in stressful situations have a positive view of their functioning. Those who partake in emotion orientated coping strategies were likely to experience behavioural problems such as self-blame, anger and isolation resulting

in maladjustment (Van Dijk et al., 2009). Furthermore, these behavioural problems were found to be associated with level of social support, life events and acceptance of the disease (van Dijk et al., 2009).

Reduced level of social support and engagement, life dissatisfaction and communication problems were found to impact effective coping mechanisms in parents (Fotiadou, Barlow Powell & Langton., 2008). Hoekstra-Webbers, Jaspers, Kamps & Klip (1998) investigated the differences in psychological distress and coping styles between fathers and mothers of pediatric cancer patients, over a 1-year time period: at diagnosis, 6 month and 12 month follow-up. Although distress declined significantly with time, fathers and mothers reported experiencing higher levels of psychiatric symptomatology and psychological distress at diagnosis than men and women of a norm-group. By being able to separate their thoughts and emotions parents were better able to cope, thus had an improved quality of life (Hamama-Raz, Rot & Buchbinder, 2012).

### **1.3.3 Disfigurement, amputation and prosthetic use**

Disfigurement and/or the loss of a body part and the artificial replacement can evoke emotional, behavioural and psychosocial responses in the affected individual, their family and friends as well as the wider society (Ayanniyi, 2013; Lekay-Adams, Sykes & du Plessis, 2014; Willard et al., 2014). Although the field of prosthetics is becoming more encompassing, many studies still focus on factors that either facilitate or impede the physical adjustment of the prosthetic (Desmonds & MacLachlan, 2002; Gutterfleisch, 2003; Messinger, 2009). Thus, little consideration has been given to the psychosocial impact that wearing a prosthetic and its cause has on the patient. With the reasons behind the loss/disfigurement of a body part impacting upon a person's psychological adjustment (Gupta & Padmanabhan, 2012), understanding and sensitivity is required to provide the best overall care for the individual (Ayanniyi, 2013).

With a scarcity of research in this field, this section will draw upon literature addressing disfigurements and amputation (eye and limb) and their associated prosthetic use. A recent surge in understanding the psychological, social and behavioural components of post amputation and prosthetic use (termed Psychoprosthetics), has resulted in a shift from understanding the variables related to an individual's adjustment to the amputation and successful use of the prosthetic, to recognition of the need to understand the perspective of those (in)directly affected. This section will draw on both quantitative and qualitative studies of Psychoprosthetics.

Limb amputation can stem from either a traumatic injury or due to biological factors (Atala & Carter, 1992; Smith & Campbell, 2009) and can be partial or full severance (Marcovitch, 2005). Loss of a limb through amputation presents a significant life change for the individual. From a functional and physical perspective, the loss of a limb is a permanent change of an individual's ability to interact with their environment. The visible changes following an amputation and the fear of stigmatisation from a social group can become a stressor (Loucas et al., 2017). Consequently, the loss of a body part may impact the individuals emotional and social functioning (Loucas, Brand, Bedoya, Muriel & Wiener, 2017). Often individuals will opt to wear a prosthetic to mask their amputation and to maintain some normality in terms of aesthetic and functional use.

Positive adjustment to amputation and the prosthetic plays an important role in the rehabilitation process. Sinha, van den Heuvel & Arokiasamy (2014) reported adjustments to amputation and a prosthetic as intricate psychological, physical and social processes. This includes allowing the body to adjust to the amputation and potential phantom limb pain, readjust their functional expectations with the prosthetic to be able to perform activities of daily living (ADL) in relation to their roles and responsibilities, acceptance of the changed body image, and, be able to maintain their social role and social contacts (Sinha et al., 2014).

Maintaining both ADL such as cleaning and personal care, as well as physical activity has been found to contribute to body satisfaction (Murray & Fox, 2002). Tatar, (2010) and Wetterhahn, Hanson and Levy, (2002) found that the maintenance of physical activity is correlated with decreased body image anxiety. With an individual's ability to return to the tasks they previously did being dependent on the nature of the amputation and the prosthetic abilities, some individuals will experience decreased psychosocial adjustment (Coffey et al., 2009; Williamson et al., 1994). This is further affected by functional and social restrictions (Coffey et al., 2009; Williamson et al., 1994). By being able to adjust expectations and adapt goals to their own needs, individuals can counteract these symptoms, resulting in increased satisfaction and improved adjustment (Coffey et al., 2009; Hamill et al., 2010).

The primary psychological disorders associated with amputations and prosthetic use is depression and anxiety (Jaraway, Perry, Phillips, Ziegler & Wolgemuth, 2013). The presence of depressive and anxiety symptoms have been found to vary based on age, gender, maturity, support and time (Jaraway et al., 2013; Singh et al, 2009). With depression and poorer rehabilitation post amputation being correlated with adjustment (Jaraway et al., 2013), the individual needs to be psychologically prepared in order to reduce long-term distress and complications (Andrews,

Anderson, Fairbain & Downing, 2012; Jaraway et al., 2013; Loucas, Brand, Bedoya, Muriel & Wiener 2017). Singh et al's. (2009) longitudinal study highlights the importance of this. They found that whilst depression and anxiety symptoms reduced quickly following amputation, they increased two years following discharge. The reduction of support at home compared to the inpatient setting was found to increase low self-esteem related to an individual's ability to manage (Coffey et al., 2009; Desmonds & MacLachlan, 2006; Jaraway et al., 2013).

Potentially acting as a confounding variable, the reason for the amputation can in part explain the above findings. Whereas an amputation as a result of a traumatic event is unanticipated, amputation as part of cancer treatment is part of a care package, thus giving the individual time to come to terms with the event. Using a cross-sectional design, Sinha et al. (2014) investigated the different facets of adjustments to amputation and prosthetic wear. Findings from a Stepwise regression analysis of face to face interviews and semi-structured questionnaires revealed average satisfaction and moderate psychosocial adjustment with the functioning of the prosthesis. Age, employment, gender and daily use of the prosthesis were found to be the most important factors. The unemployed and women were found to be less socially adjusted, with functional and social restrictions also being found in the unemployed. Potential explanations for these findings include those individuals who are adjusted to the prosthesis having more chance of being employed which in itself creates independence thus these individuals tend to be more physically active.

In regards to age, children and young people, in addition to experiencing the above difficulties, will have specific needs related to their social and emotional personality development (Najam & Ijaz, 2012). To facilitate adjustment, introductory interventions are required that promote positive coping mechanisms that enhance their psychosocial wellbeing (Loucas et al., 2017). Andrews et al. (2011) explored the pathways of care for children who underwent lower limb amputation from pre-surgery to rehabilitation. Undertaking a literature review to confirm the best practice of care for this population group, four stages were identified. These were: (1) multidisciplinary pre-operative care; (2) multidisciplinary post-operative care; (3) mid-term rehabilitation, prosthetics, limb casting and fitting, and, (4) multidisciplinary discharge planning. Having to learn to cope with a disability and its effects on mobility, pain and discomfort, as well as a significant change to body image, the first stage highlights the need for multidisciplinary pre-operative care to aid in the adjustment of the child/young person and their family. This includes tailoring play sessions and providing psychological care and interventions at the earliest opportunity. The second stage, multidisciplinary post-operative care, is primarily focused on the physical side of recovery including pain control, prevention of infection and promotion of wound healing. Once the individual is physically recovered, prosthetic limbs are fitted to restore maximum functioning,

cosmesis and symmetry as well as to reduce the duration of dependence and to restore body image. An individualised physiotherapy program that involves the individual and their family follows with the aim to optimise functioning. This makes up the third stage of mid-term rehabilitation, prosthetic limb casting and fitting. The fourth and final stage is multidisciplinary discharge planning. Being complex and requiring co-ordination of many services, the planning should begin as soon as the decision to amputate is taken. Once discharged the individual is referred to community-based health care and an outpatients specialist rehabilitation centre (Andrews et al., 2011).

Whereas Andrews et al. (2011) focused on the care pathways for children undergoing lower-limb amputation, Loucas et al. (2017) examined the type and degree of psychosocial preparation provided to the child and their family prior to their lower limb amputation via a systematic review. From the five studies identified, five themes were developed. The first theme: interdisciplinary and family communication highlights the necessity of informing the parents of young people 21 years and under of the amputation prior to informing the patient. The reasons for this includes initial information gathering prior to obtaining consent; allowing the parents to experience their emotional reaction in a safe environments, and, coming to a joint decision on how to inform the patient. The second theme: psychosocial screening and assessment refers to the preoperational factors that need to be taken into consideration when preparing the patient for surgery. This include child-specific variables (patient's age, temperament, cognitive capacity, developmental stage and location of the planned amputation), and, external variables related to family dynamics (parental reaction to illness and coping styles). Pre-operative education was the third theme and referred to a patients understanding of the impact following surgery. Tebbi et al. (1989) found that whilst 88% of patients considered themselves independent, only 58% indicated a good pre-operative understanding of the functional limitations that they would face following surgery. The fourth theme of child interventions highlighted the use of play therapy, emotive imagery, and behavioural techniques as improving the child's coping skills both before and after the amputation. The final theme of family intervention highlighted family specific therapies that are designed to identify and address discrepancies in coping style and stages of grief are an effective component of pre-surgical psychosocial involvement.

Providing psychosocial intervention for both the child patient and their parents increases overall adjustment and allow for psychological growth whereby the individual redefines themselves with positive support of others (Masnari et al., 2013). Najam & Ijaz (2012) investigated the relationship between parental acceptance-rejection and social adjustment in congenitally amputee children. Findings from the Parent Acceptance-Rejection Questionnaire and the Social Adjustment



Questionnaire found that children who perceived themselves as accepted by their parents are more socially adjusted than children who perceived themselves as rejected by both parents. Supporting these findings, Jaraway et al.'s (2013) retrospective study found that family-centred pre-operative care and preparation helps to decrease the child's anxiety which positively affects post-operative recovery.

The studies by Loucas et al. (2017), Andrews et al. (2011), Najam & Ijaz (2012), and Jaraway et al. (2013) emphasised the importance of pre - and post - operative preparation in obtaining psychosocial adjustment. Adjustment is achieved through multidisciplinary and individual and family centred preparation (Andrews et al., 2011; Jaraway et al., 2013). The parents and family's reaction to the child's surgery can influence the child's psychosocial adjustment, thus being able to process the amputation and what it means before informing the child allows them to properly prepare and support their child (Jaraway, et al., 2013; Loucas et al., 2017). This support results in acceptance of the amputation and contributes to the child's social adjustment (Najam & Ijaz, 2012).

In addition to the social adjustment and restrictions found in both adult and child amputees, correlations have been found between depression and anxiety with appearance related beliefs (Atherton & Robertson, 2006; Williamson, Schulz, Bridges & Behan, 1994). These beliefs come from both the self and the perceived perspectives and reactions of others (Atherton & Robertson, 2006; Rybarczyk, Elliot, Nicholas & Nyenhuis, 2002; Rybarczyk & Behel, 2008). Regarding the self, Rybarczyk et al. (1995) found body image to be a significant predictor of depression and quality of life. Not only do amputees have to reconcile the changes of their appearance (including self-image pre and post amputation with or without a prosthesis and their image of themselves), they have to renegotiate changes to functional ability (Rybarczyk et al., 1995). Maladjustment to the amputation and/or prosthesis can lead to psychosocial and prosthesis related difficulties. Research conducted by Breakey, (1997), Coffey et al. (2009), and Rybarczyk et al. (1992) using self-reported questionnaires found body image anxiety in this population group being statistically correlated with psychological variables including increased depression and anxiety, decreased prosthesis satisfaction and decreased self-esteem and quality of life.

Clinical levels of anxiety, depression and raised levels of appearance related distress and social avoidance were also found in individuals wearing an artificial eye and those with facial disfigurements (Clarke, Rumsey, Colin and Williams, 2003; Fu et al., 2011; James, Jenkinson, Harrad & McBain, 2011). Psychological adjustment has been shown to be worse for patients with

acquired facial disfigurement than congenital facial disfigurement. Whereas females with facial disfigurements were found to have significantly higher levels of anxiety compared to their male counterpart (Atay et al., 2013; Fu et al., 2011), gender, alongside age of acquisition, duration of prosthetic wear and type of prosthesis were not found to be related to psychosocial adjustment in artificial eye wearers (Ayanniyi, 2013; James et al., 2011; McBain et al., 2014). Instead, older age, having children and the belief that the prosthesis influences social and interpersonal relationships (how accepted they felt by society) were found to lead to increased levels of distress. This could be partly accounted for by an artificial eye being a replacement to a disfigurement, thus providing some level of normality.

Individuals with an artificial eye or with facial disfigurements reported difficulties in social settings when meeting someone for the first time and in forming friendships. This leads to high levels of anxiety, low self-esteem and creates expectations about life chances (Clarke, 1999; de Melo et al., 2012). Reasons for this include, perceived visibility of the condition, reactions of other people to the disfigurement, impact of treatment and self-perception of their illness (Atay et al., 2013; de Melo et al., 2012; Fu et al., 2011; Hill et al., 2011).

Following removal of the natural eye, the condition of the socket, movement of the muscles and the patient's psychological status will all impact on the acceptability of the prosthetic replacement (Lekay-Adams et al., 2014). Most (non) verbal communication with other people are initiated with eye contact, thus discrepancies in eye colour, movement and position will be immediately noticed (Lekay-Adams et al., 2014). If a normal appearance of the artificial eye is not achieved, cognitive process such as how the individual thinks about themselves and their prosthesis, as well as their perceptions of what the wider society think about them, are likely to arise and lead to psychosocial difficulties (Gupta & Padmanabhan, 2012; Lekay-Adams et al., 2014; McBain et al., 2014). Clarke, et al. (2003) found that alongside the physical and functional difficulties disfiguring eye conditions causes, a large proportion of patients experienced psychosocial difficulties associated with the appearance of their eye. However, in contrast, McBain et al. (2013) reported individuals being satisfied with the shape, colour, motility, comfort and fixation of their artificial eye. This disparity indicates there not being a relationship between physical appearance and psychosocial adjustment. Thus, an individualised approach to the assessment and provision of support is required.

Providing social support that matches the patient's needs and challenging beliefs about the disfigurement can lead to successful outcomes such as acceptance (Clarke, 1999; Deno et al.,

2012). Self-acceptance comes from finding meaning of the distress whereby the patient looks beyond their physical disability. In turn, this makes them feel more accepted by others and increases their level of confidence and independence (Threader & McCormack, 2016). Self-acceptance comes from a patient's level of self-efficacy which can reduce the level of social distress, depression and anxiety (Deno et al., 2012).

The perceived social stigma from others were found to also produce higher levels of body image anxiety and psychosocial adjustment difficulties in amputees (Atherton & Robertson, 2006). This suggests that the adjustment process may be more weighted towards the perceived reaction of others than their own perception of themselves. Whilst this finding is supported by Saradjian et al. (2008), who found that individuals who reported personal satisfaction with their body image will cover up their amputation as not to cause others distress, this was found not to always be the case. Murray (2009) reported that rather than hiding their amputation, some individuals would show their prosthetic which they may have decorated or personalised. However, it could be argued that the individual's attention to the aesthetic of the prosthesis could be further evidence of their attempt to detract others attention away from the functional ability of the prosthetic. If this is the case, the need to prioritise function over aesthetic further supports both the importance of others' reactions and the individual's need of the prosthetic to closely replicate the ability of the natural body part.

Gallagher & Maclachlan (2001) and Senra et al. (2012) reported that of paramount importance to individuals with a prosthesis is the desire to achieve a normal appearance. This normal appearance centres on two factors: firstly, the individual's self-perception, and, secondly, an individual's ability to continue to function to the same social level as they did prior to the amputation. Gallagher & Maclachlan (2001) and Hamill et al. (2010) found that individuals with a prosthetic reported difficulty coping with the reaction of others when their amputation and physical differences were made apparent in a social situation. Hamill et al. (2004) found that *both* the actual and perceived reaction of others (including, negative judgements on ability, patronising and awkwardness) resulted in individuals wanting to hide their amputation.

With importance being placed on being treated as an individual rather than being defined by their disability (Hamill et al., 2010), individuals have to redefine their previous held perceptions of ability and disability. This enables better integration of their pre-amputation self-identity with their post-amputation experience (Murray, 2009; Saradjian et al., 2008). Redefining the role of a prosthetic is a good example of this. Saradjian et al. (2008) reported that the worth of an item to

an individual leads it to be redefined. For example, prior to the amputation, individuals would see a prosthetic as belonging to a 'disabled' person. However, once this became a need for them, that prosthetic would be labelled as 'enabling.'

Redefining the role of a device allows an individual to take charge and be in control of their new identity. To further push away from socially inferred identities and potential negative perceptions, individuals would 'prove' their ability and worth by challenging assumptions through achievements. Primarily this involved returning and pursuing their interests and role prior to the amputation such as leisure activities and work (Hamill et al., 2010; Murray, 2009). For an individual to achieve this, the prosthesis needs to be functional and be able to do all, if not most the things the natural body part was able to do.

## **1.4 Overview**

Rb is a cancer of the retina affecting between 40 and 50 children in the UK each year. It is not discriminate in terms of gender, right or left eye but primarily occurs before the age of 5 years due to this being the critical developmental period of the retina. Survival rate in developed countries stands at 100% and 50% in under-developed countries. Survival rate is dependent on early diagnosis and treatment.

Forty per cent of patients diagnosed with Rb will have a positive family history therefore will have the hereditary form. The remaining sixty per cent of patients will have no family history: seventy-five per cent of which will have the non-hereditary form with the remaining twenty-five percent having the hereditary form. Patients with the hereditary form are more likely to have multifocal tumours affecting both eyes, whereas patients with the non-hereditary form are likely to have unifocal tumours primarily affecting one eye.

Rb occurs due to a mutation of both Rb1 genes. In the hereditary form, patients are born with one mutated Rb1 gene whilst in non-hereditary cases both genes are normal. To develop Rb, mutation must occur in the remaining gene or both genes post birth.

The two most common symptoms of Rb are leukocoria at 60% and strabismus at 20%. Imaging tools such as an ultrasound and MRI are used for diagnosis purposes. Following a confirmed

diagnosis, Rb is staged according to whether it is intraocular, extraocular or recurrent: this guides treatment options. In cases of unilateral Rb where the tumour fills over half of the globe and there is retinal detachment, enucleation is the treatment of choice. Patients with bilateral Rb often undergo a combination of Chemoreduction with the aim of salvaging at least one eye with the most vision. Often, enucleation occurs in the eye where there is little or no vision.

Following enucleation, patients are fitted with an artificial eye. From their innovation in ancient Egypt, artificial eyes have undergone noteworthy advancement (Danz, 1990). The 16<sup>th</sup> Century saw a resurgence in the manufacturing and design process where artificial eyes were being fitted in the socket and experimentation with materials lead to the use of cryolite glass; changing the shape from a sphere to a shell form then later to the Snellen Reform Eye (Deacon, 2008; Dyer, 2000). Despite investigation with other materials, it was not until WWII that methyl methacrylate replaced glass as the main material (Cerullo, 1990; McCord, 1965). Its advantages included being lightweight, easy to mould, fit and adjust, durability, resistant to rough handling and having good colour permanence (Erpf, 1953; Goiato, 2014). The few disadvantages associated with this material are largely related to poor restoration as a result of inadequate equipment and materials; all of which can easily be overcome (Charlsworth, 1966).

The advent of WWII also highlighted the importance of obtaining an accurate impression of the socket for optimal fit. With custom made eyes came research into effective ways to fabricate an artificial eye (Benson, 1977; Goiato, 2014; Phillips, 1950). Being created from dental material, the process of manufacturing methyl methacrylate artificial eyes has largely followed the same procedures employed in manufacturing dentures (Danz, 1990). Whilst adaptations have been made, this has largely been through trial and error by the prosthetist throughout the years (Cerullo, 1990; Phillips, 1950; Roman, 1994; Wilson, 1972).

The existing evidence regarding recent developments in the design and manufacturing have been produced by prosthetists. This includes digital imaging the iris and varying impression techniques (Artopoulou et al., 2006; Cevik et al., 2012; Jain et al., 2010; Prithviraj et al., 2013). Many of the changes and varying techniques are likely to be service based, thus resulting in variation in outcome and service delivery.

The secondary effects of cancer including Enucleation and prosthetic use, as well as the cancer journey itself often leads to psychosocial difficulties for both the child with cancer and their

parents. Psychosocial difficulties including anxiety, depression and PTSS have been found to occur from diagnosis to remission. For children, the type and level of psychosocial difficulties have been found to be related to developmental changes with age and personality influencing the child's ability to adapt and cope with the effects of cancer (Griffiths et al, 2011; Han et al., 2011). Cognitive, emotional and behavioural difficulties have consistently been reported with impact on the child's academic performance and social relationships noted (Kahalley et al., 2012; Weintraub et al., 2011). This impact has been associated with higher levels of anxiety, depression and low self-esteem (Tremolada et al., 2017). Children who perceive the presence of burdens (Tougas et al., 2015) and those who use emotion orientated rather than task orientated coping mechanism show higher levels of behavioural problems such as anger which often results in impaired social relationships and schooling (Tremolada et al., 2017; Weintraub et al., 2011).

Whilst children with cancer report a normal level of functioning, parents often perceive a decrease in adjustment and social interaction in their child with cancer (Chao et al., 2003). As their primary caregiver, parents are under a high level of stress and burden resulting in psychosocial distress. Grief, anger, shock and disbelief in the early stages of their child's disease is replaced by anxiety, low self-esteem and PTSS during and after the treatment journey (Deshields & Nanna, 2010; Mellblom et al., 2014; Li et al., 2012; Van Dongen-Melman, 2000; Wiener et al., 2014). By actively attempting to provide replacement or alternatives to cover the gap in their child's life and normalise the child's experience (Ellertsen et al., 2015; Vance et al., 2004), parents are able to redefine their initial feelings of loss (Cox, 2018). Although parents have been found to have lower levels of resilience than parents with healthy children (Ellertsen et al., 2015), the use of active coping mechanisms can see a decrease in their level of psychosocial distress.

Similar to those (in)directly affected by cancer, psychosocial distress such as anxiety and depression have been found in patients with disfigurements, amputees and those with prosthetics (Jaraway, Perry, Phillips, Ziegler & Wolgemuth, 2013). Furthermore, variations in symptoms have also been found to be based on age, gender, maturity, support and time (Jaraway et al., 2013; Singh et al, 2009). Appearance related beliefs (perceived visibility of the condition, reactions of other people to the disfigurement, impact of treatment and self-perception of their illness) leads to high levels of anxiety, depression, low self-esteem, decreased prosthesis satisfaction and decreased quality of life (Breakey, 1997; Clarke, 1999; Coffey et al., 2009; de Melo et al., 2012; Fu et al., 2011; Hill et al., 2011; Rybarczyk et al., 1992).

Rehabilitation programs that promote positive coping mechanism and enhance psychological wellbeing are imperative for those affected by cancer, disfigurement and prosthetic use (Loucas et al., 2017). Psychological preparation, early intervention, an MDT approach and continual psycho-education for the patient and their families need to be at the centre of treatment (Andrews et al., 2011). This increases overall adjustment and allow for psychological growth whereby the individual redefines themselves with positive support of others (Masnari et al., 2013; Murray, 2009; Saradjian et al., 2008; Singh et al., 2009).

Across the studies of parents of children with cancer, the child with cancer and/or disfigurement and prosthetic use, recent evidence suggests implications on social functioning, relationships and academic performance. This has been found to lead to symptoms of depression, anxiety, low self-esteem and PTSS. Yet, the degree of, and even existence of distress has been found to vary. This heterogeneity of findings may be due to a number of factors. Katz et al. (2011) stated that social problems may be relatively uncommon and more prevalent in a subgroup of survivors. Furthermore, studies vary in terms of methodology and population groups. Many studies have included participants with a range of types of cancer and varying ages of one group, for example children from aged 6-16 years with Leukaemia, Rb and bone cancer (Katz et al., 2011; Westbrook et al., 2016)

As a complex construct, quality of life is hard to measure. This is evidenced in this review which, although refers to both quantitative and qualitative methodologies, has seen a plethora of different instruments being used in individual studies. Though effective in capturing psychological symptoms such as depression, the Hospital Anxiety and Depression Scale (HADS) and the Brief Symptom Inventory (BSI-18) are time consuming (Jacobsen et al., 2005). The Quality of Life (QoL) and the Health-Related Quality of Life (HRQL) measurements take into consideration the social, cognitive, emotional and physical functioning of the patient. However, by assessing only the negative aspects of health, not being able to distinguish between related concepts (quality of life, wellbeing, health status) and variations in the sources of information (doctors overestimating physical symptoms, parents focusing on long-term effects and the child focusing on immediate effects), results in these questionnaires' inability to capture the whole picture (Vance, Morse, Jenney & Eiser, 2001).

In addition, much of the research in terms of the cancer journey has relied upon teacher and parents reports of the child with cancer. Focusing on general dimensions of functioning results in a failure to capture the subtle components processes that reflect wellbeing, particularly as these

groups may have reduced opportunity to observe and evaluate the child with cancer overall functioning (Katz et al., 2011).

Without a fully functional assessment measure, the practical application of clinical interventions that addresses specific concerns of patients and their families full short: mostly due to a lack of information and guidance (Deshields & Nanna, 2010, Wiener et al., 2014). Patients and their families whose psychological needs are not met are less likely to tolerate treatment and late effects (Oeffinger & Wallace, 2006). This in turn will increase psychosocial distress.

Due to the intensity of treatment and time availability/resources, interventions should be delivered on a least intrusive most effective basis often starting at the first session (Deshields & Nanna, 2010). Eliciting the person's story, their current resources for coping and support mechanisms through normalising and validation are essential to provide them with practical, educational and social support (Deshields & Nanna, 2010). Specific treatment options for patients experiencing clinical symptoms such as depression and anxiety include CBT and relaxation: both of which have a positive long-term effect on quality of life (Osborn et al., 2006).

In spite of the disparities mentioned above, the findings of this review supports the notion that not only is psychosocial care needed, it also needs to incorporate the child and their families' strengths and limitations in order to enhance certain qualities that make the whole family better at coping with adversity (Deshields & Nanna, 2010; Van Dongen-Melman, 2000; Weiner et al, 2014).

The need for a fuller understanding from an idiographic perspective requires the use of qualitative methodologies. This would provide understanding of how the psychological and social variants of experience are understood by those (in)directly affected and the impact that they are likely to have on their adjustment to the cancer diagnosis and secondary affects. The current study intends to address this by attempting to gain a subjective understanding of the experience of the artificial eye process from the perspective of children diagnosed with Rb, their parents and AEP's.



## Chapter 2 - Methodology

### 2.1 Research rationale and aim

The background chapter has identified various findings and related issues that need to be addressed. This includes stagnation of the artificial eye process and its impact. Regarding the latter, the shift from the medical perspective as a result of increased survival to psycho-oncology has resulted in a focus upon the impact of the disease not only on the patient, but also those affected at a secondary level, for example, the patient's family (Carty, 2009; de Vries & Friedrich, 2014).

The uncertainties associated with Rb (such as the risk of secondary cancers and the social, physical and emotional impact on the patient) have been shown to have a negative impact on the parents. Literature has shown that parents overrate their child's behavioural and emotional problems despite the child having a positive view of their functioning (van Dijk et al., 2008; Weintraub et al., 2011). Furthermore, parents, caregivers and healthcare professionals were found to underestimate a child's resilience and understanding of their illness (van Dijk et al., 2008; Weintraub et al., 2011). This is partly due to parents coping mechanisms which were found to be influential in the child's acceptance and adjustment to treatment. The discrepancies between a patient's view of their functioning and their perceived functioning by others involved in their care emphasise how essential it is to listen and learn from the patient's personal experiences in order to improve the care offered.

A further finding from the background chapter is stagnation of the artificial eye process in terms of technological development. The consistency of artificial eye development from their innovation in Ancient Egypt up to the end of WWII has now stalled. Being a small healthcare sector where there is a lack of funding and investment may account for this.

With patient satisfaction being dependent on product outcome, exploration in improving the current process is warranted. Thus, the aim of this thesis is to explore the artificial eye process in Rb survivors: addressing the psychological impact and potential for technological advancement. This thesis is framed as one study consisting of three components: exploring the artificial eye process as it relates to the experience of (1) AEP's; (2) Rb survivors and their

parents, and (3) AEP's and Maxillofacial Prosthetists (MP's). Within each chapter attention is paid to both the impact of the process and the potential for technological incorporation.

## **2.2 Methodology**

In making a decision as to which methodological approach to employ, the research aim as well as the researcher's practice and interests should be taken into account (Koch, 1995). For an inquisitive academic with a background in mental health, a methodology that explores, listens, and understands the views of others holds importance. With literature on the artificial eye process, specifically the psychological impact and potential for technological improvements, being sparse, it is necessary to obtain a rich understanding of the complexities associated with this phenomenon. Based on these factors, this thesis employed a qualitative methodology. Thus, this chapter will discuss in detail the methodological approach undertaken in each component of the study, with its application being demonstrated within each component.

### **2.2.1 Qualitative research**

Qualitative research has its roots within sociology dating back to the 1920's. It has since been applied to psychology, anthropology, philosophy, and most recently healthcare such as nursing research (Silverman, 2000). Qualitative research is an interpretative or constructivist paradigm based on the assumption that there are multiple realities that are socially constructed (Braun & Clarke, 2013; Yardley & Bishop, 2008). From an epistemological perspective, the researcher and the participant interact to generate and interpret data about the meaning of what is being studied (Sale, Lohfield & Brazil, 2002).

Being exploratory in nature, qualitative methods are used to gain an understanding of underlying reasons, opinions, and motivations. They can generate new, richer insights which can open up meaning of areas such as social norms, behaviours and phenomena that were have not been previously understood. Thus, qualitative research contributes to growth of understanding (McLeod, 2001; Finlay, 2006; Willig, 2008).

Qualitative research involves in-depth exploration, description and interpretation of the subjective meanings of phenomena and experiences given by the research participant (Finlay, 2006). This

seems consistent with the constructivist position of this study which believes there is no single reality, with the researcher eliciting participants' views of reality

Qualitative research uses an iterative style of eliciting and categorising responses to questions. It allows issues to be explored from the participant's perspective and to study the nature and structure of peoples' attitudes, feelings, ideas, and thoughts (Kairuz, Crump & O'Brien, 2007; Wilig, 2008). There are a number of different qualitative methods available such as grounded theory, ethnography, and phenomenology. Although the methodologies differ on their philosophical underpinnings and data collection techniques, they all explore the 'what', 'why' and 'how' questions with an emphasis upon understanding subjective experience. It is a systematic and rigorous form of inquiry that seeks to capture the complexities and idiosyncrasies of experience (Buston, Parry-Jones, Livingston, Bogan & Wood, 1998; Pasick et al., 2009).

### **2.2.2 Study specific rationale and methodology**

Whilst all 3 components are qualitative in nature, they vary in terms of their approach and methods. Whereas methodology is the study of how research is done, methods refer to the tools, techniques and processes used in research. Component 1 and 3 draw upon Thematic Analysis (TA) as a method, and component 2 utilises Interpretative Phenomenological Approach (IPA). Whilst the processes involved in both TA and IPA will be discussed in detail within this chapter, their specific applications will be detailed in the relevant chapters.

The choice of methodology and methods used depends on the research question(s) being examined (Teherani, Martimianakis, Stenfors-Hayes, Wadhwa & Varpio, 2015). Being suited to questions regarding individual experiences, views, and opinions made TA a suitable method to use in component 1 and 3. With the background chapter highlighting stagnation of the artificial eye process in terms of methods, tools, and equipment used, component 1 sought to understand the process as it currently stands including its psychological impact upon the patient and the potential for technological development. The need to evolve the process led to component 3 which sought to create a transfer of knowledge between the equipment and tools used in the assessment process by AEP's and MP's. The objective of this was to explore both AEP's and MP's perceptions of the tools currently used and potential for technological development in their field. Being a similar field to artificial eyes and having access to more technologically advanced tools, it was envisioned that the tools used by MP's may have the potential to be applied to the artificial eye process.

Whereas TA allows flexibility in terms of epistemological and ontological position, sampling size and methods as well as data collection methods, IPA stipulates the above. Focusing on a particular phenomenon; IPA allows an in-depth analysis of the sense-making process and the meaning an individual attaches to it (Cassidy, Reynolds, Naylor & De Souza, 2011; Holloway & Todres, 2003). Seeking to understand the *lived* experience of the fitting of artificial eyes from the perspective of young Rb survivors and their parent's, made IPA a suitable methodology to utilise in component 2.

### 2.2.2.1 *Thematic Analysis*

Although TA has recently been recognised as a methodological approach (Guest et al., 2012; Joffe, 2011), its theoretical flexibility also allows it to be identified as an analytic method (Braun & Clarke, 2013). Viewing TA as an approach in identifying and analysing patterns in qualitative data means that it is not wed to any particular theory of framework. Consequently, TA was approached in an inductive way in components 1 and 3 of this study, where coding and theme development are directed by the content of the data. In keeping with utilising TA as a method, both studies followed Braun and Clarke's (2013) six stages (see Table 6).

Table 6: Stages of Thematic Analysis

Stages of thematic analysis	Description of processes within each stage
1. Familiarisation	Listening to the interview recording, transcribing the data and noting initial observations about data items and data sets.
2. Generating initial codes	Codes are developed across the data items that relate to the research question
3. Searching for themes	Codes are reviewed to identify similarity
4. Reviewing themes	Re-read data items to ensure the themes capture the meaning of the dataset in relation to the research question
5. Defining and naming themes	The scope and content of each theme is labelled and described.
6. Producing the report	Consideration of what the theme means, what are the assumptions underpinning it and what are the implications.

The first stage is familiarization with the depth and breadth of the content through repeated reading of the data in an active way (searching for meanings and patterns). This is followed by generating initial codes to identify words and/or phrases that capture the essence of the research question. The third stage is searching for themes. Themes can be generated at either a semantic level, whereby the statements are taken as an accurate representation of the participant's beliefs, or a latent level, where the researcher looks at the underlying conceptualisations (Braun & Clarke, 2013). Reviewing the themes follow where the themes in the previous stage are refined: firstly,

at the coded level, then at the data set level. This is to ensure that they fit and that the meanings are accurately reflected. A thematic map is often developed to help with this process.

Stage five is defining and naming themes. This is where the essence of each theme is identified and labelled accordingly. The scope and content of each theme is described, identifying the story it tells and how it fits into the overall story about the data in relation to the research question. The final stage is producing the report where the story of the data is told within and across themes. Throughout the data analysis stage, the data is checked and rechecked to enhance conformability.

#### **2.2.2.2 *Interpretative Phenomenological Approach***

IPA is a methodology in its own right, based within a phenomenological epistemology and relativist ontology where there is a relationship between what people say and how they think about their experiences (Smith & Osborn, 2003). IPA sees individuals as ‘self-interpreting beings’ where they are actively engaged in interpreting events, objects and people: known as ‘sense making’. As such the way people think, what they say, and what they do can be seen as a central analytical concern. Although this is in keeping with cognitive psychology and social cognition, IPA differs in that the methodology it employs is an in-depth qualitative analysis rather than a quantitative and experimental methodology that the former two employ (Smith & Osborn, 2003).

IPA involves the detailed examination of participant’s experiences of a particular phenomenon, how they have made sense of it and the meanings they attach to them (Smith & Osborn, 2003). IPA is used to inform understanding of a novel or under-researched topic enabling an experience to be expressed in its own terms rather than pre-defined categories (Cassidy, Reynolds, Naylor & De Souza, 2011; Smith, Flowers & Larkin, 2012).

IPA was developed by Smith, (1996) as an alternative to established quantitative and qualitative methodologies in psychology (Cassidy, Reynolds, Naylor & De Souza, 2011), and was further developed by Smith (2004) and Larkin, Watts & Clifton (2006). IPA has two primary aims, to make sense of people making sense of their lived experience and to say something in detail, about the perceptions and understandings of a particular group. Its aim is to gain an ‘insider perspective’ (Conrad, 1987) in understanding the meaning people attribute to their experiences.

IPA is an approach often used in clinical settings to inform practice and development of services. This occurs from the perspective of individuals with a particular health condition and/or those indirectly affected (Cassidy, Reynolds, Naylor & De Souza, 2011; Smith, 1996; Smith, 2004; Willig, 2008). As an under-researched topic, this study's focus on the fitting process of artificial eyes in parents of children, and, 13-16 year olds with a diagnosis of Rb, makes it highly suitable to an IPA methodology.

IPA is a method that is both descriptive and interpretative. Descriptive as it is concerned with how things appear and letting things speak for themselves, whereas interpretative offers insight into how a given person in a given context makes sense of a given phenomenon (Smith & Eatough, 2012). The interpretative element allows exploration of idiographic subjective experiences and thoughts (Biggerstaff & Thompson, 2008). With an idiographic focus, IPA is suitable to a smaller sample size and can be used to analyse individual cases or to generate themes across a small group of participants (Smith, Flowers & Larkin, 2012).

The key theoretical perspectives of IPA are phenomenology, hermeneutics (interpretation) and idiography (Smith, 2004; Smith et al., 2009). Each of these is discussed below.

#### **2.2.2.2.1 Phenomenology**

Phenomenology originated in the 1900's by Edmund Husserl (1859-1938) who was concerned with putting the meaning individuals ascribe to events at the forefront of discovery before any scientific account can be undertaken (Biggerstaff & Thompson, 2008; Langdrige, 2007; Smith, Flowers & Larkin, 2012). Husserl's phenomenology is descriptive in nature. He believed that it is important to know how someone comes to know their own experience. Husserl believed that we are always conscious of something whether that is something 'out there' or something inside of us such as a feeling or thought. Husserl referred to this as 'intentionality' which led to the development of a 'phenomenological method' investigating the features of human experience. This is done by bracketing (also known as *epoché*): focusing on how we perceive the objects and not on what the objects actually are (Langdrige, 2007; Smith et al., 2012). Bracketing takes place through a series of reductions in which our perceptions are peeled off layer by layer leading towards the essence of the experience of a given phenomenon (Smith et al., 2012). Through repeated reflection layers of meaning in the phenomenon is uncovered which is then verified by checking the text and whether it makes sense in the context (Langdrige, 2007).

One approach to this is 'eidetic reduction'. This approach establishes the essence where consideration of different possible instances of the phenomenon takes place. Building on this, Husserl described a process known as 'transcendental reduction' which looks at the nature of consciousness. This is where the individual moves away from what is experienced (noema) and the way it is experienced (noesis) to viewing the process from the outside (Langdrige, 2007). Following reduction, Husserl believed that 'imaginative variation' takes place: where the phenomenon is approached from different perspectives so the essence may come into view (Langdrige, 2007). Essences are the culmination of bracketing, phenomenological reduction and imagination variation.

Husserl's theories emphasise the self-reflective nature of the researcher and the participant and through his concepts developed a systematic study of our lived experience (Smith et al., 2012). Criticisms of Husserl's approach mainly focused on internal processes which are based on his own introspection and with a lack of detail in the steps involved in the concepts described; particularly the concept of 'essences' (Smith et al., 2012). Furthermore, whilst there was agreement that human existence is intentional, the ability to bracket off all our presuppositions was debated. Whereas Husserl believed that bracketing can be done, existentialists believed that humans are too engaged with the world to make such an abstraction (Langdrige, 2007).

Husserl's former student, Martin Heidegger, continued the existential turn rejecting the transcendental view in favour of interpreting the meaning of things in their appearing (Langdrige, 2007; Smith et al., 2012). Heidegger believed that phenomenology should be concerned with ontological questions of existence not just the processes such as perception and awareness (Heidegger, 1927).

Believing people are inseparable from the world, Heidegger argued that you cannot bracket off to see the essence of a phenomenon. Instead you need to take into consideration the historical and cultural context of the experience (Smith et al., 2012). Heidegger referred to our being-in-the-world from our perspective which is temporal and always in relation to something as 'inter-subjectivity'. IPA encourages researchers to embrace our own subjective and intersubjective experience or interpretation of the participants.

Building on Heidegger's work were Satre and Merleau-Ponty. Whereas Heidegger emphasised the worldliness of our experience, Merleau-Ponty believed that as humans we see ourselves as different to everything else as we are focused on looking at the world rather than being absorbed within it (Smith et al., 2012). Merleau-Ponty believed that freedom comes from our position in the world with our actions making ourselves and the world meaningful (Langdrige, 2007). As each experience is related to our own position in the world, we cannot fully share others' experiences. According to Merleau-Ponty, this is where phenomenology should be focused.

Satre also echoed Heidegger's concept of the worldliness of our experience but develops the point in context of personal and social relationships, thus our experiences are affected by our relationships to other people (Smith et al., 2012). Satre also believed consciousness is created through our lived experience which gives us freedom to choose who we want to be within our own existence therefore the past and present does not determine who someone is (Langdrige, 2007). Yet, people evade the choice due to people's concerns of what we will be rather than what we are. Satre described the self as an on-going project rather than a pre-existing unity which creates a fundamental way of seeing ourselves that often goes unchallenged (Smith et al., 2012).

Husserl and Heidegger's philosophical viewpoints translated into two different approaches descriptive and hermeneutics: also known as interpretative. The former is concerned with what is experienced and how it is experienced where the researcher is free of assumptions. In contrast, the latter is concerned with the interpretation of existence where the researcher cannot be free of prejudices. Component 2 of this study utilises the interpretative approach to phenomenology. Explanation for its use can be found in Chapter 4.

IPA is grounded in phenomenological theory which stipulates that there is no one true reality (Langdrige, 2007; Smith, Flowers & Larkin, 2009). Thus, the focus is on the perception of the world and how it is experienced recognising that this is different to different people (Langdrige, 2007). With an object entering our reality only when it is perceived, individuals' experiences will be shaped by society, culture, relationships with others, and history (Eatough, Smith & Shaw, 2008).



#### **2.2.2.2.2 Hermeneutics**

Hermeneutics is the theory of interpretation where the researcher interprets what the participant has revealed about their experience to understand that experience (Smith et al., 2012). Acknowledging that the researcher cannot directly access a participant's world, the researcher uses their own knowledge to make sense of what the participant is trying to make sense of. This is known as double hermeneutics (Smith et al., 2012). Three key theorists in this field are Schleiermacher, Gadamer and Heidegger.

Schleiermacher believed interpretation is based on both grammatical (concerned with exact and objective textual meaning) and psychological (refers to the individuality of the author/speaker) concepts. Schleiermacher emphasised the need to take a holistic stance to interpretation where the participant's experience is also related to the wider community or context they are situated in. With the participant's experience going through a systematic and detailed analysis where connections emerge as a result of a larger data set, Schleiermacher believed that the researcher can gain a better understanding of the participant than they understand themselves (Smith et al., 2012).

Heidegger was keen to mark phenomenology as an interpretative process. As previously mentioned, Heidegger believed that bracketing our preconceptions and beliefs (referred to as fore-structures) was not fully possible as they not only influence our encounter with a phenomenon but also allows us to understand our fore-structures more clearly, (Smith et al., 2012). Heidegger also stated that an awareness of these fore-structures can emerge either in advance or through the process of interpretation, therefore our preconceptions need to be adjusted where new questions arise from the text resulting in new meanings emerging (Moran, 2000; Smith et al., 2009).

Gadamer also believed that interpretation can both help and hinder the understanding of the participant's experience emphasising the importance of culture and history in its process (Smith et al., 2012). In the construction of knowledge, our history and what influences the way we experience the world cannot be suspended. Gadamer believed that understanding is at the core of human existence and is determined by our pre-judgments which is an accumulation of knowledge. Language is seen as the means of gaining an understanding of the world often revealing something that was previously hidden (Langdrige, 2007). Gadamer saw language as not only reflecting our experience but also humanity, giving an understanding of the self and our culture (Langdrige, 2007).

#### **2.2.2.2.1 Hermeneutic circle**

IPA follows an iterative process where different ways of thinking about the data takes place (Smith et al., 2012). It involves a dynamic and non-linear relationship with the whole and the part where you cannot understand one without looking at the other, (Smith et al., 2009). This is an important part of IPA as it offers reflective practice where our unknown pre-conceptions may come to light and be influenced/alterd by every new idea or text encountered (Smith et al., 2012).

#### **2.2.2.2.3 Idiography**

Smith (2009) describes idiography as being concerned with the particular rather than the universal. This is a move from making claims about the wider population to a systematic analysis of a particular group of people in a particular context and understanding how they make sense of their experience (Eatough & Smith, 2008; Smith et al., 2012). With a focus on uncovering individual experiences, idiosyncrasies and expectations that can be found in general datasets can be explored and understood.

#### **2.2.2.2.4 IPA data analysis**

The crux of IPA is to gain an ‘insiders’ perspective to the lived experience of the participant. It does not look for a cause-effect relationship instead allowing participants to discuss their lived experience (Shinebourne, 2011). With no fixed process of analysis in IPA, its flexible approach allows the researcher to analyse the data in a way that is focused on how the participant makes sense of their experience (Smith et al., 2009). Smith et al’s (2009) six stages were employed in analysing the data (see Table 7)

Table 7: Stages of IPA data analysis

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1. Reading and re-reading
2. Initial noting
3. Developing emergent themes
4. Searching for connections across emergent themes
5. Moving to the next interview
6. Looking for patterns across interviews

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The first stage involves reading and re-reading of the data for full immersion in the data. The second stage involves initial note-taking of an interview chosen at random. This is in-keeping with the ideographic nature of IPA. With a clear and open mind, an examination of semantic content and language use where key words and phrases are highlighted and commented upon in

the left-hand margin of the transcript page takes place. Attention is paid to the meaning of imagery and metaphor with sections that appeared significant being noted.

The third stage involves developing emerging themes. Whereas the earlier stage is aimed at producing the 'phenomenological core' of the account, this stage is where the interpretation takes place (Larkin et al., 2006). Notes are made in the right-hand margin that captures the meaning the participant was trying to convey and the ways in which they were trying to make sense of their experiences. The importance of maintaining a link between what participants said and the comments made about them means that this process needs to be repeated several times.

Stage four involves creating a list of emerging themes which is then interrogated for similar or overlapping themes and rearranged to represent the new connections/clusters: termed *master themes*. Clustering allows data reduction whilst remaining grounded in it. A table of master themes is created and theme titles generated capturing the experience of the participant.

Stage five involves the repeating of stages 1-4 for all the transcripts. This stage requires an open and clear mind as not to let our pre-conceptions and previous knowledge influence analysis thus altering our ability to recognise emerging themes.

Stage six involves looking for patterns across all interviews. Master themes for each interview are arranged side by side and re-sorted as an entire data set to identify similarities and overlaps. Clusters of related master themes are termed *superordinate themes*. Superordinate themes encapsulate something of the meaning of the experience across some if not all of the participants. Some themes may be dropped at this stage if they do not fit well with the emerging structure. Resultant superordinate themes are then put into a table that captures the essence of lived experience across all or most participants.

### **2.3 Criteria for assessing qualitative research**

With a different philosophical underpinning to quantitative research, qualitative research should be measured and evaluated by different means. Yet, many of the criticisms of qualitative research comes from it being evaluated using the same criteria as quantitative research. From a positivist approach, quantitative research states that research should be objective in nature where there is

no personal involvement from the researcher. Contrary to this is qualitative phenomenology research which states that researchers always bring something of themselves to the research process thus an acknowledgement of these influences is required. Howitt (2010) points out that a qualitative study that fulfils quantitative criteria will exclude important aspects of a qualitative research.

With a focus on establishing validity and reliability rather than accepting the value of the experience which lies within the words of the participants, qualitative research has faced criticism regarding its scientific rigour. Various methods of assessing the quality in qualitative research have been constructed (e.g., Elliot, Fischer & Rennie, 1999; Mays & Pope, 2000). Although similar, three different methods for assessing qualitative research was used in this study: Lincoln & Guba's (1985) criteria; Yardley's (2000) criteria, and, Smith's (2004) IPA method. Explanation of how this study met the criteria can be found below, with a summary of how it was ensured in each relevant component.

### 2.3.1 Lincoln and Guba's evaluative criteria

At the centre of evaluating a research study's worth is trustworthiness. This is achieved through establishing credibility (confidence in the truth of the findings), transferability (showing the findings are applicable in other contexts), dependability (findings are consistent and can be repeated), and, confirmability (the extent to which the findings are shaped by the respondents). See Table 8 for Lincoln & Guba's (1985) criteria for assessing qualitative research.

Table 8: Lincoln & Guba (1985) criteria for assessing qualitative research

Criteria	Definition
Credibility	Refers to the believability and trustworthiness of the findings from the perspective of the participant.
Transferability	The degree to which the results can be transferred to other contexts or settings
Dependability	The consistency to which the results could be repeated and result in similar findings.
Confirmability	The degree to which the results could be confirmed or collaborated by others.

These four criteria were met through employing a series of techniques. By spending time with AEP's, understanding of the artificial eye process was gained through shadowing prosthetists in the fitting, design and manufacturing stages. Observing, speaking with and developing a relationship with AEP's and the services facilitated appreciation of the work and the context they

work in. This is known as prolonged engagement and was the technique used for establishing credibility.

With the focus on qualitative research being on depth rather than breadth, producing thick descriptions is essential to validating the findings. Thick descriptions allow a phenomenon to be described in detail allowing for evaluation of the extent to which the conclusions drawn are transferable to other times, settings and people (Lincoln & Guba, 1985). Transferability was achieved in this study by describing the phenomenon in detail within each component of the study, including accounts of the research methods, the context and the raw data (Houghton, Casey, Shaw & Murphy, 2013). By reporting clear descriptions of the phenomena being studied and the context in which the study took place, the person wishing to 'transfer' the results can judge its suitability.

The third criteria of dependability were established via inquiry audit. The researcher's supervisors examined the data collection, analysis and results of each component of the study to confirm the accuracy of the findings and to ensure the findings are supported by the data collected (Houghton et al., 2013). This approach goes some way to ensuring that the findings are consistent and repeatable as well as ensuring that there was not anything missed in the study.

In addition to inquiry audit (which can also be used for the final criteria) triangulation was used to establish confirmability. Triangulation uses two or more methods of data collection or sources of data to produce rich, robust and comprehensive accounts (Cohen & Crabtree, 2006). Various types of triangulation can be used, with each component of this study using analyst triangulation. By using multiple analysts (the researcher's supervisors) to review the findings, blind spots in the analysis were highlighted and refined (Braun & Clarke, 2013).

### **2.3.2 Yardley's evaluative criteria**

Yardley (2000) proposed four principles that acknowledge the different conceptual framework underlying qualitative research and a variety of ontological and epistemological viewpoints and methodologies. For Yardley (2000) it is important the checklist highlights quality issues rather than a rigid checklist that can restrict the freedom and flexibility of the researcher. The four principles are: (1) sensitivity to context; (2) commitment and rigour; (3) transparency and coherence; and (4) impact and importance.

Sensitivity to context is concerned with the analysis and interpretation being sensitive to not only the data but also the social context and the relationship from which it emerged. It can also be established by the existing literature on the topic and to the underpinnings of the research methodology (Smith et al., 2012). This was attained by understanding the research literature on the young person patient experience of Rb, appreciating already formed relationships with purposively recruited participants and valuing the data gathered from the interviews.

Commitment and rigour are demonstrated by the thoroughness of a study and the degree of attentiveness to the data collection and analysis. By placing the participant at the heart of the interview which was treated as a unique experience where the participant could make sense of their experience as the researcher made sense of the participant's narrative, produced commitment and rigour to the process.

Transparency and coherence refers to consistency and a detailed and logical description across the research question, method, philosophical perspective and analytical and interpretative choices. This will provide an open and honest account of this component of this study undertaken. A step by step and detailed guide of the research process which represented the phenomenon with the selection of appropriate participants was outlined in the methodology section allowing for transparency and coherence.

Impact and importance relates to its practical and theoretical utility: whether it tells the reader something important and/or useful. Smith et al., (2009) states that the effectiveness of an IPA study should be based on the light it sheds in a broader context, a rich data-set and being sufficiently related to current literature. This research has a high impact and importance factor as it highlights the gap in understanding the impact of the fitting process of artificial eyes from the perspective of parents of children and 13-16 year olds with a diagnosis of Rb.

### **2.3.3 Assessing IPA research**

IPA has faced criticism regarding its scientific rigour. Smith (2004) argued that validation is not appropriate for IPA research due to being an interpretative process. As an interpretative process, the researcher is required to have an awareness of their own fore-structures and the bias this can have if not correctly managed during data collection and analysis (Parker, 2005). Too little

interpretation can result in the true experience of the population group not being fully revealed, whereas too much interpretation can take the researcher away from the lived experience of the participant resulting in inaccurate conclusions.

Smith (1996) replaced the discussion of validity to internal coherence. This is the extent to which a theme is consistent and driven by the data. With it being essential that IPA research remains grounded in the data, themes need to be representative of the lived experience of the participants. Moving back and forth between the participant's accounts and the researcher's levels of interpretation is key in maintaining truth to participant's experiences.

As a qualitative method, IPA is seen as more of a descriptive form of analysis (Larkin, Watts & Clifton, 2006; Smith, 2004). However, the need to maintain reflexivity and epoché in utilising this approach makes it just as challenging as other approaches. Furthermore, with a flexible nature, the complexity of IPA is often overlooked by many.

Language plays an important role in phenomenological research allowing description of people's own realities. However, it has been argued that language may simply describe reality rather than constructing it (Willig, 2001) and in some contexts words can add unintentional meaning to the description of an experience (Potter & Wetherall, 1987). However, individual's ability to self-reflect means that there will be some truth to our internal thoughts expressed through language (Smith, Flowers & Osborn, 1997).

Other criticisms of IPA include its assumption that the participant is fully aware of their motivations and have the skill set to express them. Madill & Gough (2008) referred to this as the psychological naivety of IPA. As a subjective research methodology, the truth is individualised to the participant expressing it. Therefore, whether or not what the individual says is true, their description is part of their identity which holds its own meaning. IPA has also been criticised for its lack of replication (Giorgi, 2010). However, the dynamic and complex nature of this approach makes it very difficult for replication to take place, (Smith, 2010).

## **2.4 Thesis design**

Typically, qualitative research does not prescribe to a specific order in which data is collected. However, taking into consideration the paucity of research on the artificial eye process, specifically the psychological impact and potential for technological improvements, there was a need to apply a sequential order to the studies undertaken. Therefore, each component was designed and executed based on the literature reviews and the findings of previous studies.

## **2.5 Summary of methodology**

Employing a qualitative methodology, this thesis explores the artificial eye process in children with a diagnosis of Rb: addressing the psychological impact and potential technological improvements. Details regarding the methods of each component and their findings will be discussed accordingly. An overall review and recommendations for future research will be put forward in the discussion chapter.



## **Chapter 3 - Artificial eye prosthetists' perspective on the manufacturing and fitting process**

### **3.1 Introduction**

The previous chapter examined three key areas in relation to this thesis: (1) Retinoblastoma; (2) development of artificial eyes, and, (3) the psychological impact of childhood cancers, amputation, disfigurement and prosthetic use. Findings highlighted stagnation in the artificial eye process post WWII and limited research into the psychological impact the process has upon the patient and their parents. Potential reasons for this include, gaps in the evidence base regarding the impact of after-care services, and, as a small healthcare sector, there are limited resources, funding and investment, meaning that research is not a priority.

Emotional and behavioural effects were found across the board in both the child patient and their parents (Ayanniyi, 2013; Lekay-Adams, Sykes & du Plessis, 2014; Willard et al., 2014). Whilst the psychological effects of having an artificial eye fitted has not been explored, sensitivity and understanding still needs to be given to the experience of those affected to ensure acceptance and to decrease psychological difficulties (Ayanniyi, 2013; Garg, Garg, Bansal & Suresh, 2012; Gupta & Padmanabhan, 2012; Lekay-Adams et al., 2014; McBain, Ezra, Rose & Newman, 2014). Furthermore, whilst there has been experimentation with the techniques used in the process, this has been prosthetists led, based on their skills and expertise.

Consequently, this component of the study sought to understand the current artificial eye process, its psychological impact upon the child patient and their parent(s) and the potential for technological development. Being at the forefront of the process as it currently stands, this aspect of the study sought the perspective of AEP's. AEP's can shed light on aspects of the process and how it impacts themselves as well as their patients and their families. Thus, this aspect of the study acts as a starting point (within this thesis and further ahead) for exploring various aspects of the process from the perspective of those (in)directly involved whereby suggestions for its improvements can be made.

Therefore, the aim of this component was to gain an insight into the manufacturing and fitting process of artificial eyes in children with a diagnosis of Rb from the perspective of AEP's. The

specific objectives were to design a questionnaire that explored the factors involved in the artificial eye process; to conduct a survey of AEP's using this questionnaire; and to produce qualitative analysis on the data collected.

This component of the study has led to a publication (see Appendix A for publication reference).

## **3.2 Method**

### **3.2.1 Data collection and sampling**

Questionnaires are a commonly used method of collecting information about a population of interest. Seeking the experiences of AEP's, open-ended questions were used. The sample size for a qualitative study needs to be appropriate to the research aims and assumptions and provide an adequate amount of data to fully analysis the topic being studied (Braun & Clarke, 2006). Braun & Clarke (2006) suggest a sample size of 15-20 for qualitative surveys. Due to the small population size and taking into consideration the potential of non-response, all potential participants identified were included.

Based on the aim of generating insight and understanding of the current manufacturing and fitting process of artificial eyes, the sampling method chosen was purposive sampling. As a non-probability sampling method suiting small populations, a wide range of perspectives from specific characteristics of the population will be attained, allowing for greater insights into the phenomenon being investigated (Braun & Clarke, 2006).

### **3.2.2 Participants**

In identifying potential participants, three factors were taken into consideration: job role, experience and population size. For the purpose of this aspect of the study, potential participants needed to be AEP's and have experience of fitting artificial eyes in children particularly those with a diagnosis of Rb. Within the UK, there are 3 NHS services and limited private services that provide artificial eyes for the country. Due to the small population size within the UK and a need to represent the diversity of the phenomenon under study (Jansen, 2010), parameters were extended to include participants in Europe and Canada. The benefits to these territories include Europe having a similar culture and medical system to the UK, and Canada as a leading expert in Rb research, both of which increases the knowledge of the phenomenon.

### 3.2.3 Materials

An online questionnaire was used because they are easy to distribute, ideally suited to sensitive topics as they offer privacy and anonymity and have the potential for quick data collection (Braun & Clarke, 2006). However, due to the questions being set in advance, online surveys lack flexibility, responses are constrained as they cannot be probed and risks excluding marginalised groups who do not have computer access (Braun & Clarke, 2006). This is particularly problematic for qualitative research.

The questionnaire was designed to gain an insight into the current process used in the manufacturing and fitting of artificial eyes in children and whether this can be improved. Research from the literature and discussion with the population group helped develop suitable topic areas (see Table 9)

Table 9: List of topics for the survey

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-	Current practice of manufacturing and fitting artificial eyes
-	Advantages and disadvantage of how artificial eyes are manufactured and fitted
-	The psychological impact of the process on the child
-	Influence of the parent' on the child's adjustment
-	Factors that make the process easy or hard for the ocularist
-	Technological advancements of the process

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The topics were then grouped into sections that would form the structure of the questionnaire. These were:

- Section 1: Demographics
- Section 2: Manufacturing and fitting of artificial eyes in children
- Section 3: Impact of the process on the child patient
- Section 4: Influence of parents on fitting artificial eyes in children
- Section 5: Current practice of fitting artificial eyes in children
- Section 6: Technological advancements in the manufacturing and fitting of artificial eyes

Whilst the aim suiting open-ended questions, the data collection method is more suited for questions that are short, clear and concise. For this reason, the survey used a combination of both open-ended (29) questions and closed ended (12) questions. The closed-ended questions

consisted of multiple choice options: the majority of which were demographic in nature and related to the difficulties fitting artificial eyes in children with Rb.

Affecting the result by influencing answers to later questions and as a result of habituation, question order also plays a key role in designing a questionnaire (Braun & Clarke, 2013). The early questions in a survey need to be easy and pleasant to answer as they encourage participants to continue the survey. Questions that are difficult to answer or sensitive in nature should be placed at the end of the survey. Consequently, questions regarding general information about the respondent and their job role were put at the start of the questionnaire, with questions relating to the incorporation of technology into the process being asked last. (See Appendix E for a copy of the questionnaire).

#### **3.2.4 Ethical approval**

Ethical approval for this aspect of the study was obtained from Bournemouth University Ethics Board (Reference ID: 8842) on 30/07/2015 (see Appendix D).

Participation was voluntary and anonymous. No personal information was collected or accessed. Consent for anonymous data to be used was indicated by ticking their agreement to participation before starting the survey. Participation burden was minimised by the use of online surveys allowing the participant to reply in their own time and in their own environments. No remuneration was provided for participants.

#### **3.2.5 Pilot study**

The questionnaire was pilot tested (see van Teijlingen & Hundley, 2001, for importance of this process) with five individuals consisting of academics from the field of prosthetics, a parent of a child with an artificial eye as a result of retinoblastoma, and practitioners from the field of manufacturing and fitting artificial eyes. The pilot testers were asked to read and give feedback on a printed copy of the questionnaire and the information sheet. Pilot testers were specifically asked to pay attention to the language, presentation, content and ease of completing the form.

Following pilot feedback, the ordering of questions was altered to reflect the chronology of the fitting process and minor amendments were made to some questions.

### **3.2.6 Recruitment**

Following the pilot study and identification of potential participants, an email invitation introducing the study and asking for their participation was sent. The email contained an information sheet detailing purpose, the inclusion criteria, their role, confidentiality and their right to withdraw during and up to one week after completion (see Appendix E). Potential participants who met the inclusion criteria and were happy to take part were asked to use the link contained in the email to participate. On opening the link to the survey, participants were asked to re-read the information sheet and to tick a box conveying an understanding of their rights and agreeing to their participation (see Appendix F).

The online survey was conducted using the online platform 'Survey Monkey' (Survey Monkey Inc., 2018). Initially, participants were given five weeks to complete the survey. After two weeks, an email reminder was sent to all participants. Due to a low response rate, it was decided by the researcher and supervisors' that the survey invitation would be extended by another two weeks. All participants were informed of the change in timeline via email. Five days before the end of the survey, a follow-up email was sent to all participants expressing the importance of their contribution to this survey.

### **3.3 Data analysis**

Data was analysed qualitatively using Braun and Clarke's (2013) thematic analysis procedure in order to identify themes and patterns within the data set. Thematic analysis allows rich detail and exploration of participant accounts. Although not wed to any pre-existing theoretical framework, the thematic analysis undertaken was approached in an inductive way where coding and theme development were directed by the content of the data. Thematic analysis is used to analyse most types of qualitative data where the focus is on patterned meaning across the data set (Braun & Clarke, 2013), and is therefore the most suitable method of analysis for this aspect of the study,

In order to become familiar with the data, reading and re-reading of the data took place in the first instance. Identification of words and/or phrases that were in keeping with the research question followed. From here the initial codes were then reviewed to identify similarity. Re-reading of these themes took place and a thematic map developed (see Figure 7) to ensure that they fit with

the meaning and accurately reflect the data. The themes that were found to be in keeping with the research question were then identified and labelled. From here, the story of the data was told.

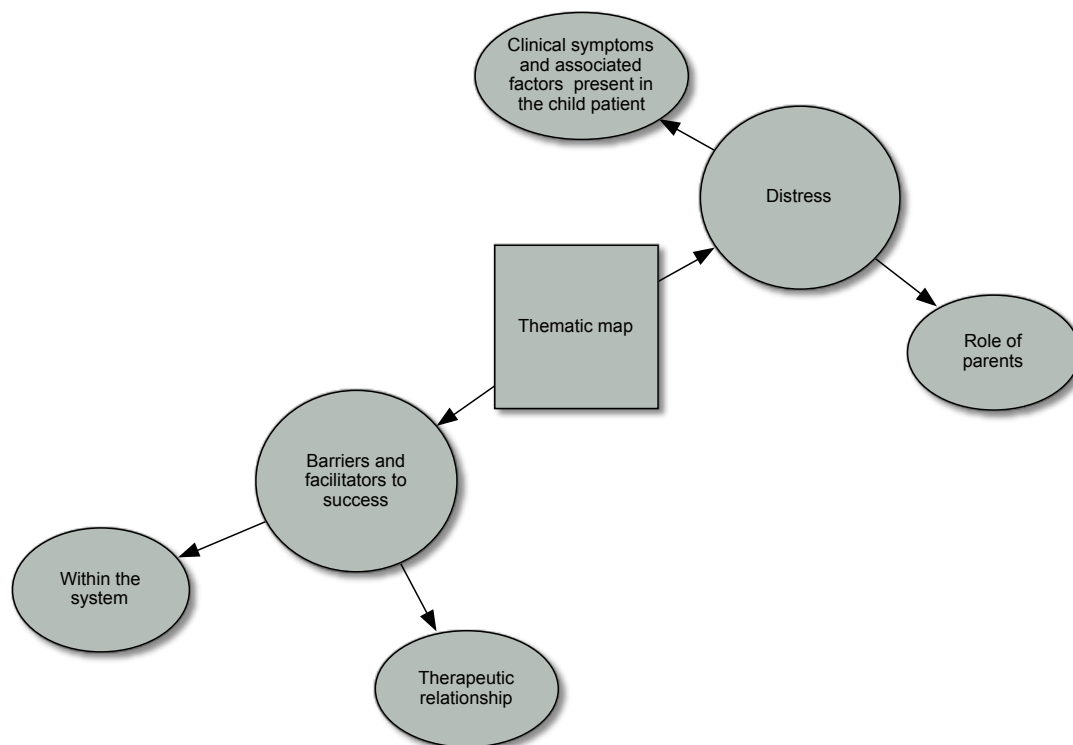


Figure 7: Thematic map of AEP's perspective of the manufacturing and fitting process of artificial eyes

## 3.4 Findings

### 3.4.1 Response rate

A total of 39 email invitations containing the link to the online survey were sent to AEP's in 14 countries across Europe and Canada. A response rate of 46% (n = 17) replies was achieved from 6 countries. The UK and Canada had the highest response rate, (UK: 47%; Canada: 29%), with the number of respondents from these countries being 62%, (n = 8) participants and 50% (n = 5) participants, respectively. Spain had a 50% response rate (2 participants invited) with the remaining 3 countries (Belgium, Ireland and Romania) having a 100% response rate (1 participant per country invited).

### 3.4.2 Participant demographics

The sample comprised of ocularists and AEP's, a ratio of approximately 70:30. Two of these participants also identified themselves as 'lead tutors', one as a dental technician specialising in maxillo-facial prosthesis, and one as an optician and technical orthopaedic. 71% of participants were from Europe with the remaining 29% of participants working in Canada. Although a wide

variation in years working as an ocularists or AEP was found (ranging from one to five years to thirty one years plus), the majority have worked within their field for eleven to twenty years (41%). The variations in years of services did not appear to be country specific.

With regards to training, participants reported having an apprenticeship (50%) and dentistry qualification (29%) as a job requirement with 21% stating no job requirement was necessary. Previous experience includes dentistry (64%), a background in art (9%) and none (27%). See Table 10 for breakdown of the demographic information.

Table 10: Participant demographics

<b>Participant by Country</b>	<b>Years Service</b>	<b>Job Title</b>	<b>Job Qualification</b>	<b>Previous experience</b>	<b>Training opportunities</b>
Belgium	31+	Ocularist	Apprenticeship	Dentistry	N/A
UK	31+	Ocularist	Dentistry	Dentistry	None
Canada	11-20	Ocularist	Apprenticeship	Art	ASO
UK	31+	Ocularist	Dentistry	Dentistry	Teach at NTU
Romania	11-20	Dental technician specialized in maxilla-facial prosthesis	Dentistry	Dentistry	Conferences
UK	11-20	Ocularist	BTEC Diploma in Dental Technology	Dental Technology	NHS training, mandatory courses
Spain	11-20	Prosthetist	Optician, Technical Orthopedic		Share experiences with other ocularists
Canada	6-10	Ocularist	Apprenticeship	Dentistry	ASO/NEBO education program
Canada	21-30	Ocularist	Apprenticeship	Medical Lab Research technician	N/A
Canada	6-10	Ocularist	Apprenticeship	Commercial pilot	ASO/NEBO exams
UK	11-20	Ocularist	Dentistry	Dentistry	In-house
Ireland	31+	Ocularist	Apprenticeship	Surgical Prosthetics	None
Canada	31+	Ocularist	None	None	Art scholarship
UK	1-5	Prosthetist	Bachelor's degree	Pathology & Microbiology lab	NHS training, Conferences, joint clinics
UK	21-30	Prosthetist	None	N/A	N/A
UK	11-20	Prosthetist	Apprenticeship	None	Conference, training, teaching skills course
UK	11-20	Prosthetist	None	Dentistry	Conferences, mandatory training, national study days

Demographic data showed similarities and variations in the way participants from different countries responded. The sample from Europe and Canada were equivalent in terms of previous experience being dentistry. Furthermore, Canada and 60% of countries from Europe identified themselves as ocularists. Respondents that identified as AEP's (Spain and 62% of UK respondents) are primarily from services that split the fitting and manufacturing process. Differences in the demographic data mainly exist between Europe and Canada and are in relation

to training and guidelines followed. In comparison to Canada where training to become an ocularist/AEP involves attendance to the American Society of Ocularists where formal examinations are undertaken and guidelines followed, there are no formal qualifications to become an ocularist/AEP in Europe. Most of the training and guidelines followed are a result of in-house training, teaching skill courses, NHS training (UK only), conferences and sharing experiences with other ocularists/prosthetists.

### **3.4.3 Item non-response rate**

The survey was split into four sections: demographic information; current process of manufacturing and fitting artificial eyes in children; psychological impact of the process, and, the incorporation of technology. Numerical data had a non-response rate of 15% whereas qualitative data had a non-response rate of 38%. This is in keeping with the evidence suggesting surveys are more suited to quantitative methods.

Demographic information had a non-response rate of 21% with questions centred on qualifications needed for the job, previous job experience and training opportunities receiving the fewest responses.

Non-response rate for questions regarding the current manufacturing and fitting process was 22% with a range of 35% and SD of 15.2. Questions with the highest response rate were the material used in manufacturing of artificial eyes and the reasons for artificial eyes being fitted in children. Questions regarding changes and disadvantages to the current process had the highest non-response rate of 41% respectively.

The psychological impact of the manufacturing and fitting process had the second lowest non-response rate of 28% with a range of 12% and SD of 5.2. Questions within this section with the highest non-response rate were related to the influence of parents/guardians on the fitting process.

The highest non-response rate (57% with a range of 35% and SD off 11.6) was related to the incorporation of technology into the current process. The disadvantages and advantages of incorporating current technology into the manufacturing and fitting process and how it's impacted by current regulations/guidelines had the highest non-response rate. This finding may suggest a



lack of understanding of technology and/or how it can aid the artificial eye field, as well as potential resistance to a process seen by AEP's as bespoke and custom made.

#### **3.4.4 Findings of the thematic analysis**

The thematic analysis generated two themes with each theme containing two sub-themes. Theme 1 related to distress and consisted of two sub-themes: (1) clinical symptoms and associated factors present in the child patient and (2) role of parents. The second theme centred around barriers and facilitators to success. Two sub -themes were identified: (1) therapeutic relationship, and, (2) within the system. Detailed interpretation and explanation of the findings are reported in the discussion section of this chapter.

##### **3.4.4.1 Theme 1: Distress**

The diagnosis of cancer and its secondary effects is an overwhelming life experience for both the child patient and their parents. If not identified and managed, psychological distress as a result of the diagnosis and treatment process can escalate. Not only will this negatively impact treatment success, the child with cancer and their parents' wellbeing is at further risk of deterioration.

With the goal of healthcare services being to protect and improve the health of individuals and populations, it is imperative that we gain an understanding of potential psychological distress. Thus, this theme explores distress in terms of clinical symptoms and associated factors in the child with Rb, and their parents.

##### **3.4.4.2 Subtheme 1: Clinical symptoms and associated factors present in the child patient**

The psychological difficulties experienced by children with cancer particularly those who undergo amputation/enucleation can be accounted for by a variety of factors. This includes the potential experience of multiple life-threatening traumas, invasive medical procedures, reason for the loss and age of the child. Whereas the removal of a body part as a result of a traumatic experience is unanticipated, removal as a part of an on-going care package, for example, individuals with cancer, is anticipated. This allows the individual to prepare for the surgery and begin the acceptance and adjustment process. The importance of the reason for the loss on an individual's wellbeing is emphasized by Sean.

“This (psychological distress) depends on the reason for the loss of the eye; trauma injuries or painful disease like glaucoma is more distressing than having an eye fitted, but for congenital problems the child has not experienced any distress from the eye itself.” (Sean, UK)

In the case of Rb, the removal of the eye occurs within one week of the treatment decision being made. Therefore, whilst it can be argued that it is an anticipated surgery, the time-period for adjustment is limited. With the age of the child undergoing enucleation occurring from birth up to 5 years old, it cannot be unequivocally claimed that distress is or is not experienced. This was evident in this aspect of the study with various factors including type of distress, age of the patient and amount of time the child has had an artificial eye for, playing a key role in perceived distress.

“It depends; physical distress is caused by losing the eye, but fitting the artificial eye generates psychological distress, as children remember the previous stress and pain, and anticipate similar things from fitting.”  
(Bella, Romania)

Bella distinguishes between the physical and psychological distress experienced in relation to specific stages in treatment. The physical distress experienced by the loss of an eye can present significant life changes in the patient. This includes changes from binocular vision to monocular vision which can lead to feelings of disorientation, thus impacting on the individual’s ability to interact with the environment and those within it. Although physical distress as a consequence of loss of a body part can lead to reduced emotional functioning, the psychological distress was viewed by Bella as being a consequence of the fitting process only. With patients being fitted with an artificial eye six to eight weeks post-enucleation, psychological distress as a result of the loss may not off manifested to an identifiable level.

Contrasting findings in terms of no perceived distress can also be accounted for by the short time period between enucleation and fitting. Attending to patients at an early stage may reduce any (simmering) negative psychological responses that the patient may be experiencing. As described by Kylie, ensuring the fitting process runs smoothly can also help negate potential distress.

“I think on the whole children recover from the loss of an eye fairly quickly, most also don't have a problem with the fitting if tackled correctly. Some do fear the fitting process but this is more the thought of it beforehand.” (Kylie, UK)

It is worth noting that both Bella and Kylie reported cognitive processes as contributors to distress. The experience of distressing and/or unwanted thoughts can emerge as a result of uncertainty or can be based on prior memories. Current situations and events can trigger upsetting memories leading an individual to believe that the danger is here and now rather than in the past. As highlighted above, a child with Rb may have undergone traumatic medical procedures therefore any treatment particularly to do with the eye can evoke an emotional reaction. In addition, a child's inability to understand the reasons for their illness and what their continual treatment will involve can lead to thoughts of uncertainty which manifest in anxiety, fear and anger.

These psychological reactions can develop in behavioural responses which can be seen as a way for the child to exert some form of control in what is happening to them.

“Fear about potential discomfort; often, initially, a desire to have things happen their way (i.e. they decide when or what treatment to have as a way of exerting some control over the process).” (Sean, UK)

Becoming accustomed to the artificial eye process can reduce the cognitive and emotional distress. Sean states that this is related to the number of medical interventions and its delivery (traumatic versus non-traumatic)

“If a child has had lots of medical interventions in their life then they often find a fitting 'not so bad' and can become fine with it at a very young age, as young as 20 months. Other children (especially where the prosthesis is not discussed or maintained at home) may not become accustomed to the process until adulthood.” (Sean, UK)

Primarily being an emotional response as a result of cognitive processes, means that a young child, though unable to effectively communicate how they are feeling, can nevertheless experience distress.

The age at which a child is able to communicate verbally is concurrent with their ability to understand what is happening to them.

“Starting 4 years old. They begin understanding more things and better control themselves.” (Bella, Romania)

Guss reports that although communication and understanding reduces the child’s instinctual emotional response, awareness of the loss becomes more prevalent, which can put the child at risk of (continual) psychological distress. It is at this stage that Guss reports the need to impart knowledge that the child with an artificial eye can still do what a healthy child can do.

“After age 3, when they start communicating vocally. They can quantify what’s going on, eliminating the flight or fight reaction ... After that (2-3 years of age) they are conscious of the fact eye loss is prevalent. When monocular best to communicate that you are not disabled, and can do most everything.” (Guss, Canada)

#### **3.4.4.3 Sub-theme 2: Role of parents**

The above sub-theme touched upon changes to the individual’s ability to interact with the environment and those within it as a result of enucleation and the fitting of an artificial eye. Being the first social group of children, family (particularly parents) play a key role in shaping their psychological and social development. The threat imposed by cancer and its secondary effects escalates parent’s view of their role and obligations. Becoming their child’s advocate, leads to increased demands on the parents which can result in distress. Persistent parental distress may in turn have a negative impact on their child’s as well as their own adjustment: therefore, compromising the success of on-going and future treatment.

The role of parents was consistently reported by AEP's as being intrinsically tied to the success of the artificial eye process.

“They (parents) make or break the whole process. I understand they are concerned for their child, but they sometimes have trouble trusting a stranger. But then again we are the experts.” (Guss, Canada)

Parental concern for their child can be exhibited in various forms: the main two being physical and verbal. Physical affection includes cuddling, smiling and hugging. Verbal affection consists of saying positive things and complementing a child. Whilst the intention may be to protect their child, it may have the opposite effect. The difference between effective and ineffective communication by parents towards their child during the fitting process is described by Eddie.

“Some are too soft or squeamish to adequately help in dealing with their children causing a negative experience - others take it in a very matter of fact way and are very relaxed with their children which help enormously and make the experience positive.” (Eddie, Canada)

By engaging in a care giver role and adapting to stress caused by a traumatic event, the presentation of stress related symptoms in parents is often incongruent with their perceived levels of stress. The (unintentional) communication of stress, whether that is verbally and/or behaviourally, can lead to negative outcomes in terms of the treatment process and the child's psychological wellbeing.

“Most of them (parents) contribute positively, but some of them are too stressed about their child situation and transmit this stress to the child, even it is unintentional.” (Bella, Romania)

In addition to managing their own distress, parents may need to manage their child's behaviour, particularly when attending health appointments. Some parents may adopt a more lenient approach whilst others will treat their child as they would have done before diagnosis. In order to aid normality, participants reported the need for parents to be, matter of fact, calm and consistent with their child.

“If parents/guardians are quiet and calm with their child it often leads to a better outcome overall. Parents who show their fear/disgust do not encourage their children at all and lead to a disappointing outcome all round.” (Chloe, UK)

“Parents can positively reinforce the experience by being happy to practice consistently with removing and replacing the prosthesis; problems often arise when parents do not want to do this, making the child anxious whenever it does need to come out.” (Sean, UK).

#### **3.4.4.4 Theme summary**

This theme uncovered various factors related to distress in both the child with cancer and their parents. The short time frame from treatment decision to surgery and surgery to prosthetic treatment does not provide a sufficient adjustment period. Thus, when they present to the artificial eye service, distress symptoms may appear. Unless identified and supported, the distress that is often based on unwanted thoughts and/or memories; can escalate.

Communication and understanding were found to be imperative for both the child with cancer and their parents in reducing distress. In regards to the child, increased age was found to counteract psychological difficulties. However, this was reported to be dependent on their parent's reactions. Engaging in a care giver role, parents experience high levels of stress which is often unintentionally communicated to their child. Parents *need* to protect and provide for their child results in emotional rather than rational based decisions.

#### **3.4.5 Theme 2: Barriers and facilitators to success**

Theme 1 explored the psychological factors of the artificial eye process upon the child patient and their parents. In order to understand their implications and assess how they may be

incorporated into clinical practice an understanding of the fitting process from within is needed. A lack of understanding of the experience of the child patient and their parents on part of the clinical team may have the potential to lead to determinantal outcomes for all those involved. As an on-going interactive experience, exploration of the understanding and experience of AEP's is needed. Thus, this theme explores the barriers and facilitators to success in terms of the therapeutic relationship and from within the system.

#### **3.4.5.1 Subtheme 1: Therapeutic relationship**

The unintentional communication of stress by parents has already been highlighted above as having the potential to lead to negative outcomes: particularly when an interdependent relationship exists between the child patient and their parent(s). The reliance on one another will often push the parent to make treatment decisions on what they perceive their child's quality of life to be. As an emotional response, these decisions are not always appropriate. To better prepare parents to make the most rational decisions regarding their child's care and treatment as well as enhancing their child's quality of life, parents need support from health care professionals that rely on open, clear communication and accurate, unambiguous information.

Limited communication of information may increase parents' preference not to delegate care, instead getting overly involved in the treatment process themselves. This can create an interdependent relationship between parent and child, making the AEP's job harder to do.

“The worst of these negative situations in my opinion relate to parents who coddle their children. They reinforce the negative with their children and create a dependency on the parent. This slows down my process of gaining the child's trust by several years and can adversely affect the overall cosmesis.” (Victor, Canada).

Acquisition of knowledge plays a key role in parents making treatment decisions that are rational and in the best interest of their child. Not only can a lack of information hamper parents' ability to contribute to the decision-making process, it may also make them feel that they have no choice but to place 'blind trust' in the AEP. More often than not, imparting knowledge will see parents transition from feeling disempowered and cautious at diagnosis to confidence in their child's AEP as treatment progresses. Eddie emphasizes the power of knowledge in getting the parents on board.

“At the consultation stage I try to impart to the parents that they must impress upon the child that when we say the prosthesis is going to be inserted/ removed it is going to happen.” (Eddie, Canada).

Imparting knowledge requires a skill set whereby the parents feel listened to and understood. Having witnessed and gone through their child’s cancer journey with them, parents may either expect or hope that the artificial eye process will provide a quick and easy solution which will restore the defect. Through information giving AEP’s are able to manage expectations that are more in keeping with what they are confidentiality able to provide. The importance of getting parents to agree on a course of action was expressed by both Kylie and Tony.

“Getting the parents on board in the first place, if they understand the difficulties they can help you achieve the best possible result.” (Kylie, UK)

“It is important that parents feel they have complete access to you at all times within surgery hours and when they feel they have your support it gives them enough confidence not to actually access the process.” (Tony, Ireland)

Often there are discrepancies between the AEP and parents in terms of perceptions of the same issue. Whereas parents focus will be on the psychological impact of the process (including and being based on past experiences), AEP’s attention will be focused on creating the best outcome for the child patient. The conflicting viewpoints can lead to frustration and assigning of blame which can negatively influence the therapeutic relationship and process. Pre-operative education for the parents and child patient about the psychological and social effects of the process can enhance adjustment and pre-empt elements of distress experienced. Furthermore, understanding the perceptions, opinions and experiences of both the child patient and their parents can afford the AEP insight into how best to manage expectations. Understanding, compassion and involvement are a few therapeutic skills highlighted by AEP’s as contributing positively to the artificial eye process.



“Compassion, skill, understanding parents’ and children’s concerns and an ability to reach realistic goals with a view to achieving the best fitting and cosmesis.” (Amy, UK).

“By being involved in the initial removal of the eye and becoming confident without portraying any fear to the child.” (Tony, Ireland)

With time, parental involvement in the process will reduce and/or eliminate their and their child’s distress, allowing the AEP to achieve the best outcome for the patient. This outcome as a result of the therapeutic role of the AEP’s was reported as a benefit of the process by Bella.

“Eliminate/lower children's fear, and sometimes the pain (if there is any).” (Bella, Romania)

The six week time frame from surgery to first appointment with the AEP allows for clinical interventions including structured therapeutic strategies such as play therapy being developed. As a custom made process with various factors such as age of the child, temperament, cognitive functioning and developmental stage impacting success, interventions will often be tailored to need. Awareness of the interdependent relationship between parent and the child patient, Sean and Tim point to the need of both to work in tandem from the initial appointment onwards.

“Depending on the child’s age, helping the child to understand what is going to happen before the appointment and not to make too much fuss about it. Children also react badly if the parent is distressed about the treatment too.” (Tim, UK)

“I typically introduce children to the materials in play first, often by making a model of their finger in the materials we will use for the eye mould. Working with parents is usually about demonstrating both the advantages and the limits of the material, so that they do not feel their child is not getting the best possible treatment.” (Sean, UK)

As noted earlier, the lack of information may result in the parent placing ‘blind trust’ in the prosthetist. This can result in both the parent and child patient taking on a passive role with the AEP as expert. This primarily occurs in the initial stages of treatment. The lack of knowledge and/or confidence can hamper their ability to engage in the process due to being unable to communicate their views. Involvement from the outset was highlighted by many AEP’s as being essential to creating a therapeutic relationship where no ‘power differences’ occur. As an individualised treatment process, tailoring the way the AEP engages with the child patient is of paramount importance. The need to tailor treatment according to age and psychological wellbeing was discussed by Victor and Chloe.

“Taking time to gain their confidence and being as gentle as possible in your approach. If the child will absolutely not co-operate then speed and accuracy are your key.” (Victor, Canada)

“If they are of an age that they can understand what is happening I let them ask questions and get involved. It is very important that they trust me in what I am doing. I try to be calm and gain their trust. Too much noise and excitement is, in my opinion, is a negative thing.” (Chloe, UK)

Interactions between the parent, child patient and the AEP is key to physical and emotional rehabilitation success: with decreased anxiety and depression, improved adjustment and better coping mechanisms being reported. The use of open dialogue creates trust and confidence in the AEP and process, thus increasing co-operation.

“Gaining their confidence is a key factor to success with kids. Having the confidence in fitting a child also goes a long way.”  
(Victor, Canada)

As mentioned above, disparity in focus between the AEP (functional aspects of the prosthesis) and the child patient and their parents (psychosocial outcome) can occur. Not only was awareness of these interrelated factors highlighted, methods to overcome them were discussed.

Understanding is created through the use of objects and language, with adjustment to the process being enabled through regular appointments.

“Using toys, pictures etc. Sometimes we need to use anaesthesia but I discourage this at all costs.” (Amy, UK)

“Often terminology like 'special' eye or even just 'new' eye can be helpful, as long as they understand they will not be able to see through it (this is normally only an issue in older children).” (Sean, UK)

“Children generally have difficulty with the procedure for a brief period of time (a few months) - we have the parents bring the children in regularly so that they get used to the environment- once they realize that it is not a painful procedure they behave really well in general.” (Eddie, Canada)

#### **3.4.5.2 Subtheme 2: Within the system**

The above sub-theme highlighted measures taken by AEP's to create an effective therapeutic relationship whereby there is a joint focus on the psychological impact and on creating an aesthetically pleasing and functional artificial eye. As an individualised program whereby the artificial eye is custom-made, creating that balance between wellbeing and product outcome becomes challenging: being further hampered by system complexity. This sub-theme will explore system barriers and facilitators to success in regards to outcome. Particular attention will be paid to current process and potential changes around psychological and technological expansions.

Following removal of the natural eye, the condition of the socket, movement of the muscles and the patient's psychological status will all impact on the acceptability of the prosthetic replacement. Whilst best attempts will be made to produce an aesthetically and functional artificial eye, the role of the AEP in enabling physical rehabilitation and function may at times mean that they prioritise obtaining a good fit over cosmesis.

“An accurate fit! This generally requires fitting under anesthetic for young children. Failure to recognize this can result in serious complications, especially when fitting over a live globe.” (Victor, Canada)

Enabling function and comfort: which are often seen as the two cornerstones that dictate an individual’s experience of the process; has resulted in various protocols being implemented by services and AEP’s alike. Mixed opinion in the use of anaesthesia was reported with some AEP’s using it as a first port of call, and others using it as a last resort in terms of medical, and, at times, psychological need. Justification for its use centred around reducing (physical) complications and in creating a therapeutic relationship.

“Sometimes under light sedation for the initial impression and once you get a comfortable fit the child develops trust and it becomes much easier. Having the parents on side by even sacrificing a couple of initial appointments is worth it in the end. So impression and a trial shape for 2 - 4 weeks before making up an eye.” (Tony, Ireland).

“I will take a light impression in the office if possible, sometimes I fit empirically and impression at a later date. If an impression is imperative but not possible in the office I will set up with the patients doctor to arrange an impression under anaesthesia.” (Frank, Canada)

Although artificial eyes are custom made, giving them the best chance of fit and cosmesis, its success is not always guaranteed. This is often dependent on co-operation of the child patient.

“Every eye is custom made and therefore has the best chance of optimum fit and cosmesis therefore requiring less visits for alteration.” (Chloe, UK)

“The general fitting and painting process when the child is un-cooperative. Trying to get correct shape, iris colour and iris position when you might only get a brief look at the socket.” (Kylie, UK)

As well as being measured in terms of the process, success is also based on long term outcomes. Creating dual meaning of the artificial eye between positive enabling (when working as it is meant to) and negative control (when it does not work, i.e., lose fit and discolouration), the child patient and their parents are required to adjust to difficulties that they are likely to face in prosthesis use. In addition to providing information and consultation, expectations need to be managed in order to retain psychological wellbeing. As noted in theme 1 sub-theme 2, a lack of preparation and information with regards to the process of rehabilitation and learning to use the artificial eye is problematic and the root of many of the difficulties encountered in the initial stages of recovery. If psychological distress is apparent and impeding the treatment process, some patients and/or their parents may require further support.

Mixed responses were evident regarding (specialist) psychological support and referral on behalf of the child patient and their parent(s). Whereas some AEP’s reported making referrals on an individual basis, others reported being prohibited due to regulations.

“Done on an individual basis. Close links with children’s hospital for services if needed or passed on to charity support.” (Tim, UK)

“No, Ocularists are not allowed to refer according to OMH/ADP rules/contracts.” (Guss, Canada)

Regulations provide standardization of service process including product design, development and outcome as well as onward referrals. They communicate desired outcomes and act as a fundamental guideline to help make decisions. With the custom-made nature of artificial eyes requiring process flexibility, AEP’s reported following various guidelines in the creation of artificial eyes including national and service specific.

“The only regulations are MDA rules. I follow the Hospital method which I am partly responsible for creating.” (Amy UK)

“(The guidelines impact) a lot. Any changes in techniques practices have to be approved by the OMH/ADP. Although with that said, not afraid to try new things in hopes of improving the final product.” (Guss, Canada)

As identified by Guss, stringent regulations can impact the speed and progress of improving the service and product outcomes. With advancements in healthcare capabilities due to significant progress in available technologies, their integration can result in improved service and patient outcomes. Whilst suggestions of new technologies that can be incorporated into the artificial eye process were made, refinement of current techniques was seen as sufficing.

“Most promising is 3D scanning is most appealing but think this is limited as it cannot show you soft tissue displacement like conventional fitting.” (Guss, Canada)

“Levelling the playing field in certain areas of skill sets to overall improve the quality of service offered to our patients.” (Victor, Canada)

The view by some AEP’s that the current process is a skill and craft that needs to be preserved, saw attention move from technological improvements to service recognition.

“Personally, think the methods employed by the ASO are sufficient. Case of, if it’s not broken don't fix it.” (Guss, Canada)

“Regulations and proper certification - Understanding and recognizing the need for certification in this field.” (Victor, Canada).

### **3.4.5.3 Theme summary**

The barriers and facilitators to success reported by AEP's in this theme suggest the need for a review of clinical practice in regard of the child patient/parent and AEP relationship and service provision. The need for an artificial eye has an unmistakable impact on the child patient and their parent(s) in terms of their psychological and social wellbeing: both of which are intricately related to the child patients use of the artificial eye and the meaning they both attach to it. Although AEP's have a primary focus on the functional aspect of the artificial eye, their reports highlight an attempt to understand the impact of the process and strive to ensure that both the child patient and their parents play an active role in the care process. This is obtained through the acquisition of new knowledge and skills. Developed through the treatment process increases parents co-operation, whereby they work alongside the health care provider to ensure their child receives the best outcome. This in turn reduces the psychological distress of both the child and the parents.

The attempt to create a shared understanding as a way to manage expectations and to increase compliance is dependent on service provision. Whilst flexibility is an essential requirement of the artificial eye process, variations in regulations and guidelines followed, results in outcome inconsistency. The individualised nature of the job saw AEP's report the need for certification and regulation to create a level playing field, rather than incorporate new technology into the process. Consistency in outcomes increases confidence and trust in the AEP thereby enhancing their relationship with the child patient and their parent(s).

## **3.5 Discussion**

### **3.5.1 Summary**

The aim of this aspect of the study was to gain an insight into the manufacturing and fitting process of artificial eyes in children diagnosed with Rb from the perspective of AEP's. By understanding the current process from the perspective of AEP's, areas were identified for further exploration within and outside of this PhD.

The findings highlighted the psychological distress experienced by the child patient and their parent(s) as well as the barriers and facilitators of the system. Negatively affecting obtaining a good impression, the child patient's distress can be reduced by the AEP and their parents by providing encouragement, reassurance, practicing the insertion and removal of the artificial eye and being matter of fact. With technology being viewed as not being able to meet the

requirements to produce aesthetically pleasing artificial eyes, AEP's focus was on creating equality in their work practice through regulations and certification.

The above points to a potential relationship between the fitting process and the emotional wellbeing of the child patient and their parent(s): with its management revolving around both human and technological factors. AEP's, the child patient's parents and the consistency in the process were reported as positively contributing to the patient's acceptance and adjustment to having an artificial eye fitted.

This suggests further exploration of the fitting process and its effects on the child patient and their parents. In addition to reduced psychological distress, the benefits of this exploration include improvements to the service process and delivery.

### **3.5.2 Relationship to the literature**

#### **3.5.2.1 Theme 1: Distress**

Although previous literature has predominately focused on the experience of having a cancer diagnosis and the impact of loss of a body part, the findings may hold relevance to the artificial eye process in Rb survivors. Shaikh et al. (2014) highlighted a relationship between the loss of an eye and anxiety, depression, and stress. These psychological difficulties, as well as cosmetic disorders experienced by the child patient can make behaviour management difficult (Goel et al., 2012). In keeping with Goel et al.'s (2012) findings, this aspect of the study highlighted trust, patience, reassurance, and understanding by the healthcare professional as important aspects in managing the child patients distress. By conducting research on the psychological impact of the artificial eye process, management programs can be developed to best manage a patient's emotional upset. It can also highlight areas for improvement which can have a positive impact on the patient's acceptance of the process.

Showing a potential relationship between the patient's distress and its management by their parents, exploration of the impact of the artificial eye process upon the parents is essential for improving acceptance and adjustment. As their child's primary caregiver, Patenaude & Last (2001) reported that parents are more likely to experience changes in their lifestyle, family dynamics, and emotional wellbeing. This may result in parents experiencing feelings of loss and uncertainty (Van Dongen-Melman, 2000). Consequently, parents are likely to feel overwhelmed, impacting on their coping mechanisms regarding their child's diagnosis, treatment, appointments,



and after-care support. Whilst there is no literature on the role parents play in the impact of, and how they manage their child's distress of the fitting process, research on the effect of caring for a child with a cancer diagnosis highlights poorer quality of life, anxiety, depression, increased vulnerability, guilt and sleep disorders (Holm, Patterson & Gurney, 2003; Jurbergs, Long, Ticona & Phipps, 2009; Klassen et al., 2008). Successful improvements to the current artificial eye process can only be made from understanding the impact of the process from the perspectives of those involved and the role they play in managing the distress of the patient. This includes understanding the process in the context of a family to enable post-traumatic growth.

### **3.5.2.2 Theme 2: Barriers and facilitators to success**

Alongside parents, AEP's were highlighted as being pivotal in managing the impact upon the patient and helping them adjust to the process. Satisfaction with the process and the end product was found to be enhanced when healthcare professionals work successfully within a multi-disciplinary team (MDT) (Cancer Australia, 2017; Covinsky et al., 1998). Just as important as treating the patient, healthcare professionals need to build a successful relationship with the patient and their families. Goold & Lipkin (1999) found that the patient-practitioner relationship is crucial for vulnerable patients as they experience heightened reliance on the healthcare professional's competence and skills. This is also the case for families whom rely on the practitioner's skill-set and expertise. As a vital component to the success of treatment and satisfaction, exploration of the role of the AEP, specifically in children with a diagnosis of Rb is needed. This would offer further guidance in how to manage the artificial eye process and how to manage patients and their families experiencing any emotional upset as a result of their treatment.

Another area highlighted that has the potential for improving the current artificial eye process is the incorporation of technology. An efficient and effective process can reduce the distress experienced, thus improving the patient's emotional wellbeing and making the fitting easier for the AEP. AEP's highlighted non-uniformity in the equipment/tools used and the procedures followed. This results in varying outcomes of an artificial eye within and between patients: often negatively contributing to the patient's psychological wellbeing and acceptance of the process. Whilst the results of this aspect of the study showed that the fitting process can be distressing for the child, it cannot be solely attributed to the non-uniformity of the process. The distress highlighted, may be related to other factors such as coming to terms with the loss of the eye, relationship with the AEP, patient's own coping mechanisms as well as factors outside of having an artificial eye. These areas are yet to be explored.

Although experimentation of the application of technology in smaller prosthetic fields has begun, AEP's reported that technology is not yet accurate enough to meet the requirements of creating an aesthetically pleasing artificial eye. Areas currently under investigation include orbital prosthesis and maxillofacial prosthesis with three-dimensional surface capture (3D scanning), three-dimensional Computer Aided Design (3D CAD) and layer additive manufacturing processes/rapid prototyping and manufacturing (RP&M) being explored (Bi et al., 2013; Bibb et al., 2013; Evans et al., 2004; Li et al., 2015; Wu et al., 2009). Whilst refinement of the techniques is needed to ensure correct soft tissue placement, good fit and cosmesis, the evolutionary nature of technological development means that its investigation into artificial eyes needs to begin. Due to the technological limitations highlighted above, the majority of AEP's were in favour of creating system wide regulations to level the playing field in artificial eye outcome. Consistently receiving high standard artificial eyes will contribute to adjustment and acceptance of the prosthesis by the child patient and their parent(s).

### **3.5.3 Strengths and limitations**

#### **3.5.3.1 Participants**

A small population group, AEP's were recruited across Europe and Canada: with access to participants coming from a contact at the NAES and through an online search. Suiting small population groups, the sampling method chosen was purposive. The sample size achieved of 17 participants created enough depth to produce sufficient findings in keeping the research question (Patton, 1990).

#### **3.5.3.2 Research design**

Qualitative research is exploratory in nature usually involving direct interaction with an individual(s) (Braun & Clarke, 2013). Having identified the sample, the recruitment method needed to be amenable to the participants. Consequently, face to face methods were ruled out in favour of an online method.

Online methods are seen as equivalent or in some cases superior to face to face methods (Campbell et al., 2001; Hinchcliffe & Gavin, 2009; Kenny, 2005; Reid & Reid, 2005), especially when trying to access small population groups (Adler, Zarchin, 2009; Sweet, 2001; Thorsten, Isabelle, Alexander & Roediger, 2008). Online methods benefit from a flexible design, are less time consuming, are convenient for respondents, have a low cost, and offer greater anonymity. The perceived anonymity of online methods which is particularly beneficial when collecting information about sensitive topics, as well as instant access to transcripts and having a higher

level of group homogeneity which increases participants' willingness to disclose personal experiences, makes online methods beneficial (Ayling & Mewse, 2009; Campbell et al., 2001; Joinson, 2005; Mann & Stewart, 2000). However, this study experienced difficulties in using this method. In keeping with the findings of Opdenakker (2006) and Wright (2005), limitations of using online methods were highlighted in this study. This included data errors due to question non-responses, different interpretation of the questions by respondents leading to unclear data and reduced sense of responsibility leading to potentially less trustful information or half-hearted answers. The latter was particularly prominent in this study with open-ended questions having the highest non-response rate. Potential explanations for this include time constraints and not having a strong opinion regarding the question asked. With face to face interviews being more likely to elicit detailed responses due to the setting and the use of follow-up questions, consideration of its future use needs to be given to aid and expand the findings of this study.

The online method used in this study was that of a survey. The recency of online surveys in health-related research means its successes and failures are continually being updated (Eysenbach & Wyatt, 2002). Biases of online surveys are similar to those found in all online methods. They include its non-representative nature (selection bias), technical issues and ethical considerations. By ensuring the topic is suitable to be conducted online and being transparent in terms of how the research was conducted and the difficulties and benefits experienced can help overcome some of these limitations. Whilst, approaches were employed to maximise response rates, such as utilising pre-existing contacts with AEP's, the promise of survey results, anonymity and transparency in the study's aim, response rates varied overall and within and between questions.

Literature concerning response rates to online surveys are inconsistent. Converse, Wolfe and Oswald (2008) reported a response rate for electronic surveys at 25%-30%. However, utilising multi-mode approaches such as postal surveys and follow-ups can increase the response rate to above 60% (Yun & Trumbo, 2000). Based on these findings and the study sample, a response rate of 40% was deemed satisfactory. Although the questionnaire achieved this target, individual questions had a response rate varying from 24% to 100%. Low response questions were primarily those that asked for in-depth responses. Improved response rate to the questions may have been achieved by other qualitative methods such as interviews and focus groups. However, for reasons already stated, this will not be feasible.

### **3.6 Conclusion**

This aspect of the study has explored the current manufacturing and fitting process of artificial eyes in children with Rb. Following a thematic analysis two themes were identified. Firstly, distress which included the clinical symptoms and associated factors present in the child patient, and, the role of the parents. The second theme reflected upon the barriers and facilitators within the system. This theme included the therapeutic relationship and within the system.

Intrinsically linked to both human and technological factors, the process can be seen as an interaction rather than linear in nature as its success is dependent on the child patient's (and their parent's) acceptance and adjustment which is dependent on how the process is conducted. As a multi-faceted process involving both psychology and technology factors, an understanding of the requirements and needs of those (in)directly involved in the process is needed. This would allow recommendations to be made and developments to begin for its improvement.

# **Chapter 4 - Children with Retinoblastoma and their parents**

## **perspective of the fitting process**

### **4.1 Introduction**

Rb is a rare type of intraocular childhood eye cancer typically occurring in children before the age of five years (Childhood Eye Cancer Trust, 2014; Goel et al., 2011; Huang et al., 2013; MacCarthy et al., 2009; Meel et al., 2012; Ray & Gombos, 2012). Prognosis is dependent upon early diagnosis, accurate classification of the tumours and early treatment. Unilateral Rb is normally non-hereditary compared to bilateral Rb which is typically hereditary where secondary tumours are more common (McCarthy et al., 2013). Whilst survivorship is much better for unilateral compared to bilateral incidences, 70% of patients diagnosed with unilateral Rb will require enucleation and consequently a lifetime supply of artificial eyes (Childhood Eye Cancer Trust, 2014).

The psycho-oncology of Rb literature has predominately focused on the impact of the disease rather than the after-care following successful treatment. One area highlighted within this research is the discrepancy between the patient's perceived level of functioning and that as perceived by other's: most prominently their parents, but also healthcare professionals and other caregivers. Whereas parents reported their child experiencing behavioural problems associated with level of social support, life events and acceptance of the disease, the affected child reported normal functioning in their everyday life apart from school where there were lower levels of quality of life (Willard et al., 2014). With assumptions made at the disease level by the parents, questions are raised as to whether these assumptions are extended to after-care support.

Findings from component 1 highlighted the fitting process as being more distressing for the patient than the loss of an eye, with parents being an influencing factor in the child's adjustment and acceptance of the process. With an emerging link between the human and technological factors in regards to the artificial eye process, this component sought to explore the fitting process in terms of its impact and potential for improvement from the perspective of children with Rb and their parents. Exploring the impact of the fitting of artificial eyes in both population groups has the potential to shed light on the current process including its advantages, disadvantages and areas for growth. With technological developments having not yet been investigated in the artificial eye process combined with a mixed reaction from AEP's regarding its incorporation (as

highlighted from component 1) assessing children with Rb and their parents' requirements may provide weight to future research and development (Buhler, 1996; Ram, Grocott & Weir, 2007).

## **4.2 Research aims and objectives**

The aim of this component of the study is to understand the lived experience of the fitting process of artificial eyes from the perspective of parents of children, and, 13-16 year olds with a diagnosis of Rb.

The aim has been achieved by employing an Interpretative Phenomenological Approach (IPA) in order to understand the feelings, values and perceptions of both population groups. This allows suggestions to be made for its improvement both in terms of its psychological impact and potential technological advancements.

## **4.3 Methodology**

### **4.3.1 Rationale**

Exploring the fitting of an artificial eye(s) from the perspective of parents of children, and, 13-16 year olds with Rb presents a number of methodological challenges. As previously identified, literature on the impact of the fitting process is sparse. Therefore, there are no established theories or proposed psychological constructs of importance. Consequently, a qualitative approach is necessary to obtain a rich understanding of the complexities associated with this process.

Being a poorly defined and conceptualised phenomenon, IPA is concerned with how people perceive and make sense of their experience (Langdrige, 2007). Consequently, IPA aims to generate rich and detailed descriptions of the phenomenon under investigation. IPA's concern with the participant's sense making generates questions that tap into emotive issues as well as allowing self-reflection (Smith & Eatough, 2012).

Committed to the detailed analysis of a participant's experience of a particular process (Smith, Flowers and Larkin, 2009), this approach fits with the research question and aim of this

component of the study. Thus, as a thorough and detailed methodological process which purports rich data, IPA can help in producing better outcomes for those directly or indirectly experiencing the phenomenon. Please refer to Chapter 2 for details of IPA.

### **4.3.2 Participants**

This component of the study involved two population groups of participants: parents of children, and 13-16 year olds wearing artificial eyes as a result of Rb. Smith et al. (2009) stated that having two different groups within IPA can make the process of analysis more demanding. However, with more than one viewpoint required in understanding the phenomenon in question, it is essential to have two groups that can grant access to different perspectives. Both populations are being targeted, as the parent(s) are critical to the care their child receives as well as their adjustment to the process (as found in component 1); and 13-16 year olds (as the primary receiver of the treatment), are best placed to share their experience of the current process and ways in which it can be improved.

The need for a certain level of emotional reliance and an ability to clearly articulate their experiences (due to interpretation being a key factor in the methodological approach of this component), resulted in the child participants being recruited that are aged between 13-16 years old.

Whilst there were no age restrictions for the parent participant, the exploration of the fitting process in children with a diagnosis of Rb, required that the parent has a child aged between 0-16 years.

The inclusion and exclusion criteria for this study were as follows:

Inclusion criteria:

- The young person has a confirmed diagnosis of Rb
- The young person has 1 or both eyes enucleated as a result of Rb
- The young person has had 1 or more artificial eye(s) fitted
- The young person is aged between 13 and 16 years of age
- Enucleation and the fitting of artificial eye(s) has taken place in the UK.
- The parent has a child whom wears an artificial eye(s) as a result of Rb aged between 0 and 16 years.

Exclusion criteria:

- The young person has an eviscerated or exenterated socket(s)
- The young person does not permanently live in the UK

### **4.3.3 Sampling method and size**

Participants were selected through purposive sampling in order to gain a diverse range of information-rich experiences from topics of interest (Patton, 1990). Purposive sampling is a type of non-probability sampling where the focus is on particular characteristics of a population of interest that can best answer the research question.

With the aim being to capture a wide range of perspectives of the given phenomenon, a maximum variation (heterogeneous) sampling method was employed (Langdrige, 2007). This sampling method fits well with the purpose of the study, where there are two population groups: parents of children, and, 13-16 year olds who wear an artificial eye(s) as a result of Rb.

The sampling method can result in selection bias where the sample is not representative of the populations being explored. Rather than being seen as a weakness, this sampling method is seen as a choice whereby the participants are selected based on their varying perspectives. Whilst, this potential bias cannot be overcome, awareness of it and how it affects the results will help draw reasonable conclusions.

IPA studies tend to be conducted on very small sample sizes with depth being more important than breadth (Smith & Eatough, 2012). Smith and Osborn (2003) state that there is no right sample size with the decision on sample size being based upon the research question and the phenomenon in question. Englander (2012) goes on to say that the focus should be based on how many times the phenomenon makes its presence in the description, not on how many have had the experience. Whereas Larkin and Thompson (2012) reported that IPA studies lend itself to single case analysis, Langdrige (2007) and Smith et al. (2012) reported a sample size of three to six participants for student projects. With this in mind, a sample size of four participants from each population group was sought.



#### **4.3.4 Recruitment**

Recruitment occurred in two stages with data being collected following each recruitment stage. Stage one recruited 13-16-year-old participants between April and May 2017. Parent participants made up stage 2 recruitment which occurred during June 2017.

##### **4.3.4.1 Stage one**

Participants were recruited via the Rb outpatient clinic based at Birmingham Women's and Children's NHS Foundation Trust. The Rb nurse specialist identified potential participants that met the study inclusion criteria and sent them and their parents a study invitation letter, participant information sheet, and a parent information sheet (see Appendix F). The parent information sheet asked them to consider the wishes of their child in taking part in the study and advised them to discuss the research with their child. All information sheets and the study invitation letter clearly stated that a parent needs to bring their child to the interview so they can sign a consent form agreeing to their child's participation.

Both information sheets provided details about this component of the study. The information sheet can increase recruitment success through providing a clear explanation of a study, the importance of their participation and feedback and ensuring anonymity and confidentiality. Participants' right to withdraw before, during and up to two weeks' post interview was clearly stated as well as what will happen to their information following their participation.

Participants in collaboration with their parents were given one week to decide whether to participate in this component of the study. Although response to the recruitment strategy was high, with the two participants contacted agreeing to participate, the number of participants recruited was low. This was due to the majority of patients attending the Rb Clinic at Birmingham Women's and Children's NHS Foundation Trust being aged between 5 and 10 years, therefore not meeting the inclusion criteria of this component of the study. Access to other participants that met the inclusion criteria were sought via the Rb clinic at Royal London/St Barts Hospital and CHECT. However, access was denied, therefore recruitment of this population group came to a natural end. Whilst the quality of IPA research is based on depth rather than breadth, the low sample size has created strong limitations which will be examined in the discussion section of this chapter.

#### **4.3.4.2 Stage two**

Parent participants were recruited via an online support group for parents of children with a diagnosis of Rb. A brief description of this component of the study was posted on the 'closed' online support group. The brief description provided information regarding the study's aim, purpose, inclusion criteria and information on what the study entailed and the timeframe for completion. Those whom expressed interest in the study were sent a link via e-mail that contained a participant information sheet, consent form as well as demographic questions and the main research question (see Appendix F). The latter two could only be accessed once the participant had signed the consent form agreeing to their participation.

#### **4.3.5 Data collection**

##### **4.3.5.1 Experience near question**

IPA data collection needs to be flexible giving experience a central place whilst recognising various influences on that experience such as cultural and historical factors (Smith & Eatough, 2012). Common data collection methods in IPA research include face to face interviews, focus groups and diaries (Brocki & Wearden, 2006; Smith & Eatough, 2012; Smith & Osborn, 2003).

Question(s) are developed that address the topic of interest but are used as a guide rather than dictating the interview. In order for a researcher to understand and have an 'insider's perspective' to that experience, it is necessary to develop an experience near question (ENQ) allowing the participant to re-explore their experience. Employing an ENQ allows modification of and additional questions to be asked in a dynamic nature during explorative discussions (Smith & Osborn, 2003). An ENQ is a question asked that gets to the crux of the phenomenon being investigated (Churchill & Wertz, 2001). They are conducted in real-time where the researcher can follow up important and unexpected issues encountered during the interview. Regarded as experts in their own experiences an ENQ gives participants freedom to voice their experiences in a safe and secure environment. Due to these reasons, the data collection method for this component of the study was in-depth interview using an ENQ.

The ENQ were designed to elicit and explore participant's views and experiences. Developing the ENQ before the interview allows advance consideration of any difficulties that may be encountered and how they might be dealt with (Smith & Osborn, 2003). Furthermore, given the

sensitive nature of the interview, the ENQ allowed careful consideration of the wording of the question to avoid misunderstanding and misinterpretation.

Development of the ENQ began by identifying the area of interest and the issues contained within it. For this component of the study, this included how the fitting process impacted the participant; how the fitting process can be improved; and the positive and negative experiences of the process. With the aim of seeking to understand the lived experience of the fitting process of artificial eyes, the initial questions were reformatted into one question that would get to the crux of the phenomenon being investigated (see Table 11 for the ENQ).

In addition to the ENQ, prompts and probes were considered and noted to assist the researcher in delving deeper into the participant's answers, encourage the participant to elaborate and to check the meaning of the participant's response (see Table 11 for the prompts and probes used).

Table 11: ENQ, prompts and probes and closing question

ENQ
- Can you tell me about your new eye, what's it like having it fitted?
Prompts and probes
- Can you expand on that/Can you tell me a bit more about...
- What do you mean by...

#### **4.3.5.2 Conducting the interviews**

Whilst the ENQ was used for both population groups, its delivery varied due to access to participants. Face to face interviews were conducted for 13-16 year old participants whereas parent participants completed the ENQ online. The differences between population groups as a result of data collection methods is explored in the discussion section of this chapter.

##### **4.3.5.2.1 Face to face interviews: 13-16 year old participants**

On arrival to the interview, small talk was made with the participant and their parents about recent events in order to develop rapport. Prior to the start of the interview, the purpose of this component of the study was explained and any questions answered. Consent was then sought: initially from the participant's parent(s) who assessed their child's ability to partake in the study in accordance to the Fraser guidelines (level of maturity to make decisions and understand the

implications of those decisions), then from the participant themselves. The consent forms stated what this component of the study involves, that they have had the opportunity to ask any questions, that their participation is voluntary and they have the right to withdraw before, during and up to two-weeks post-interview, and that the interview would be digitally audio-recorded and that the data will be stored securely.

A brief demographic questionnaire was given to the participant intended to provide social and personal context for their responses. Before the interview began, all participants were asked whether they wanted to be interviewed on their own or have their parent(s) sit in with them. There was an even split with one participant having their parent sit in with them, and the other one not.

The interview style was non-directive, narrative and participant led: starting with the ENQ and being followed up with prompts and probes. Attention was paid to the language used by participants with the researcher using their language when clarification was needed. Verbal and non-verbal skills (including frequent eye contact, nodding, using open body language and an interested tone of voice, smiling, reassuring the participant, being responsive to the body language of the participant, pacing the interview according to the participant's needs and permitting contemplative silences), was used by the interviewer to create rapport particularly in cases when it is noted participants are struggling in recounting emotive experiences: known as 'existential fog' (Van Maanen, 1988).

When the interview was drawing to a natural end, the interviewer provided a summary of what the participant had said to facilitate further reflection and to give the participant the opportunity to add any more information they so wish (Collins & Nicolson, 2002).

All participants were thoroughly debriefed at the end of the study where the purpose of the research and their role within it were re-stated (see Appendix F). Participants had the opportunity to ask the interviewer any questions and raise any concerns. All questions were answered as honest and fully as possible. Information about services that participants can contact if they needed any emotional support was given. Furthermore, participants were reminded that they could speak to the researcher and/or the Rb team in the weeks following the end of the interview.

#### **4.3.5.2.2 Online interviews: parent participants**

Although face to face interviews are the preferred method of choice in IPA studies due to their real-time dialogue and flexibility (Smith & Osborn, 2003), access to parent participants meant that their data was collected online. As a relatively new approach to collecting data, Wilkerson, Iantaffi, Grey, Bockting & Rosser's (2014) best practice guidelines in conducting online qualitative data was followed.

Following the expression of interest, participants were sent a link to the online component of this study. The link was to the online platform 'Survey Monkey.' Upon accessing the link, participants were asked to read an information sheet detailing the aim and purpose of their participation as well as explaining what was required (see Appendix F). The email address of the researcher was provided to answer potential questions or concerns. Participants were informed that they could return to the link after taking time to consider their participation.

Following the information sheet, the link contained a consent form that all participants had to tick to show their agreement before taking part (see Appendix F). Participants were then provided with a series of 11 demographic/pre-interview questions. Once completed, the participant was asked to answer the ENQ. To ensure participants' responses reflected their own experience and were detailed in nature, participants were asked to spend time reflecting on the fitting process and various aspects involved. Discussion points were provided which were in keeping with the prompts and probes used in the face to face interview. These were included to help participants recall aspects of the fitting process that they may not have considered.

After completion of the ENQ, participants were provided with a debrief (see Appendix E) that restated the purpose of this component of the study and thanked them for their participation.

#### **4.3.6 Ethics**

##### **4.3.6.1 Ethical concerns**

The main ethical concern in this component of the study was the possible distress caused by discussion of the fitting process of their (child's) artificial eye, particularly in cases where the process may not off have gone smoothly. Participants are likely to find discussing sensitive topics confronting and a stressful experience (Brannen, 1988). Therefore, as researchers, it is important

to create a safe and supportive environment (Willig, 2008). If any form of distress is not picked up on, trust and openness can be affected where the intellectualised data will replace free-flowing data (Hollway & Jefferson, 2000; Willig, 2008). Therefore, an environment allowing free expression that seeks to explore distress in a safe and contained manner along with researcher and participant responsibility is essential in managing such situations (Hollway & Jefferson, 2000; Willig, 2008).

As this is the crux of the study being conducted, questions regarding the process and its impact on the participants needed to be asked. This was addressed by informing participants about the risk and asking questions in a sensitive, clear, concise and age appropriate manner in order to reduce possible misinterpretation and misunderstandings. Furthermore, all participants were given a list of support services they could contact if required. This included contact details of the Rb service and the researcher. Informed consent confirming participation under these conditions was obtained from all participants.

Other ethical considerations relating to this study include confidentiality, anonymity and informed consent. Participants were fully informed of the research procedure and were informed in advance of the interview that they could withdraw their participation at any time during the process and up to two-weeks post interview without any judgement or repercussions (Willig, 2008).

Confidentiality and anonymity were maintained at all times. All data collected was made anonymous using a unique identification number. Following transfer of data into an electronic database, all paper records and recordings were disposed of in accordance with Bournemouth University's procedure for disposing of confidential information.

Ethical standards not only need to be maintained for the participant, but also for the researcher. In order to maintain professional accountability, regular supervision from Bournemouth University and support from the Rb team were sought. This provided a space to review how the interviews were going and allow discussion of any concerns experienced regarding all aspects of this component of the study. An additional measure employed was placing self-imposed boundaries between the role as a researcher and an individual being, in order to maintain good mental health.

#### **4.3.6.2 Ethical approval**

Ethical approval was sought on two different occasions in keeping with the recruitment stages.

##### **4.3.6.2.1 13-16 year old participant**

Ethical approval was first sought from Bournemouth University's Ethics Committee. However, as the research participants were identified due to their use of a NHS service this study required NHS Ethical approval.

NHS Ethical approval was first sought on 07/06/2016 from the South-Central NHS Research Ethics committee. The outcome of the review was an 'unfavorable opinion'. The points raised were addressed and a new application was submitted and reviewed on 15/11/2016.

A provisional opinion was granted with the recommendation of adjusting the child participant group from 5-16 year olds to 13-16 year olds. This was based on the researcher's experience of working with children in a research capacity and the workload of two population groups. Following these adjustments, a favourable opinion from the NHS Ethics Committee was granted on 17/01/2017 and the Health Research Authority on 20/01/2017 (see Appendix D).

##### **4.3.6.2.2 Parent participants**

Due to the low sample of 13-16 year olds and the importance of gaining various perspectives of a given phenomenon, a joint decision between the researcher and supervisors were made to include parent participants in this component of the study.

With participants being recruited via an online support group, ethical approval was sought from Bournemouth University Ethics Board. Ethical approval was granted on 13/06/2017, reference number 16622 (see Appendix D).

### 4.3.7 Transcription

Transcription is taking data from spoken word to written form for analysis (Stuckey, 2014). Consequently, transcription is only required for the face to face interviews of this component of the study.

The digitally recorded interview files were transferred to a secure university services for storage before being deleted from the electronic device. The interviews were then transcribed verbatim by the researcher with care taken to include all interview questions, comments and verbal utterances on behalf of the researcher as well as all speech from the participant. To preserve anonymity all identifiable information from the text was removed during the transcription process and each participant was allocated a unique identification number which has subsequently been used in all aspects of data analysis and reporting.

A transcription notation system was developed by the researcher that enabled a clear and concise understanding of all utterances (see Table 12).

Table 12: Transcription notation

Notation	Meaning
*	Pause
!	Long pause
-	Indecipherable speech
<b>Bold</b>	Emphasis
(Laughs)	Laughs lightly
(Laughs!)	Laughs a lot
+	Sentence interrupted

### 4.3.8 Data analysis

Best suited to questions regarding experiences and perceptions, data was analysed using the flexible IPA approach described by Smith et al., (2009). Please refer to chapter two for details of the analysis process.

## 4.4 Findings

### 4.4.1 Introduction

Data was collected from 2 13-16 year old participants' and 9 parent participants. As there were 2 population groups, data was analysed based upon which group they belonged to.



For the purposes of this analysis and confidentiality, all names and reference to locations were changed. Extracts from the transcripts are referenced in the following order: page number, line number.

The analysis of 13-16 year old participants identified 4 superordinate themes (see Table 13). Each superordinate theme had between 2 and 3 subordinate themes. The 4 superordinate themes were: *Artificial Eye and the Self*; *The Artificial Eye Process*; *Coping with the Impact of the Process*, and, *Role of Parents*. *Artificial Eye and the Self* had 3 subordinate themes. These were: *Artificial Eye and Sense of Identity*; *Age-related Changes to Self-perception of having an Artificial Eye*, and, *Presenting the Self to Others*. *The Artificial Eye Process* superordinate theme had 2 subordinate themes. These were: *Outcomes of the Fitting Process*, and, *Trusting the Prosthetist*. The third superordinate theme of *Coping with the Impact of the Process* had 2 subordinate themes. These were: *Physical and Emotional Implications*, and *Resilience After Process Outcome*. Finally, the superordinate theme of *Role of Parents* had 2 subordinate themes. These were: *Reliance on Parents*, and, *Change of Role Between the Patient and their Parent*.

Table 13: Table of Superordinate and Subordinate themes of 13-16 year old participants

<b>Super-ordinate theme</b>	<b>Subordinate theme</b>
1, Artificial Eye and the Self	A. Artificial Eye and Sense of Identity B. Age-related Changes to Self-Perception of having an Artificial Eye C. Presenting the Self to Others
2. The Artificial Eye Process	A. Outcomes of the Fitting Process B. Trusting the Prosthetist
3. Coping with the Impact of the Process	A. Physical and Emotional Implications B. Resilience After Process Outcome
4. Role of Parents	A. Reliance on Parents B. Change of Role Between the Patient and the Parent

The analysis of parent participants identified 3 superordinate themes (see Table 14). Each theme had between 2 and 3 subordinate themes. The 3 superordinate themes were: *The Onus is on the Parent(s)*; *Process Repercussions on the Patient and How this can be Managed*, and, *Service Experience*. The Onus is on the Parent(s) had 3 subordinate themes. These were: *Impact of*

*Having a Child with Additional Healthcare Needs; Support and Recognition from Others*, and, *Normalising the Experience*. The second superordinate theme of *Process Repercussions on the Patient and How this can be Managed* contained 2 subordinate themes. These were: *Patient Impact of Having an Artificial Eye Fitted*, and, *Role of Prosthetist*. The final superordinate theme, *Service Experience*, had 2 subordinate themes

Table 14: Table of Superordinate and Subordinate themes of parent participants

<b>Super-ordinate theme</b>	<b>Subordinate theme</b>
The Onus is on the Parent(s)	Impact of Having a Child with Additional Healthcare Needs Support and Recognition from Others Normalising the Experience.
Process Repercussions on the Patient and How this can be Managed	Patient Impact of Having an Artificial Eye Fitted Role of Prosthetist
<i>Service Experience</i>	Service Delivery Outcomes Artificial Eye Outcomes

In classifying a theme, this component of the study aimed to follow Smith’s (2011) suggestion that a theme must reoccur in at least half of the participant sample size. Classifying themes this way not only demonstrates a commonality among participants where sharing of a similar experience or feelings towards a particular experience is expressed, but also increases the validity of the results (Smith et al., 2009). However, due to the mixed level of detail provided by parent participants, some of the themes presented did not occur in over half of the sample. Table 15 and 16 presents the themes along with the reoccurring patterns under each participant.

Table 15: Identified patterns of reoccurring themes in 13-16 year old participants

<b>Superordinate Theme</b>	<b>Polly</b>	<b>Wendy</b>	<b>Theme present in over half of the participant sample?</b>
Artificial Eye and the Self	Yes	Yes	Yes
The Artificial Eye Process	Yes	Yes	Yes
Coping with the Impact of the Process	Yes	Yes	Yes
Role of Parents	Yes	Yes	Yes

As can be seen from Table 16, the superordinate theme of ‘*The Onus is on the Parent(s)*’ is only present in 4/9 of participant’s responses. In addition, the superordinate theme of ‘*Process*

*Repercussions on the Patient and How this can be Managed*' is only present in 3/9 of participant's responses. Whilst this does not indicate that it cannot be classified as a theme, it demonstrates how the analysis has evolved where the commonalities of the participants' experiences were negotiated with the idiographic stance of IPA.

Table 16: Identified patterns of reoccurring themes in parent participants

Superordinate Theme	Emily	Rachel	Claire	Sarah	Leah	Rebecca	Andre	Lucy	Naomi	Theme present in over half of the participant sample?
The Onus is on the Parent(s)	Yes	No	No	Yes	Yes	No	Yes	No	No	No
Process Repercussions on the Patient and How this can be Managed Service Experience	Yes	No	No	Yes	Yes	No	No	No	No	No
	No	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes

## 4.4.2 Demographic information

### 4.4.2.1 13-16 year old participants

The sample comprised of 2 (100%) female participants. Participants were aged between 15 and 16 (M = 15.5). Both participants were in full time education currently studying for their GCSE's. The nationality of both participants was British with their ethnicity being White and Black or African. Although both participants had the non-hereditary form of Rb, Polly's was unilateral whereas Wendy's was bilateral (see Table 17).

Table 17: Demographic information of 13-16 year old participants'

Participant Name	Diagnosis	Age	Gender	Ethnicity	Age of first artificial eye	Number of artificial eyes	Number of artificial eyes fitted
Polly	Non-hereditary unilateral Rb	16	Female	Black or African	3-4 years	1	7
Wendy	Non-hereditary bilateral Rb	15	Female	White	1-2 years	1	10

#### **4.4.2.2 Parent participants**

A total of 21 participants responded to the online interview. All 21 respondents completed the demographic questions: however, only 9 answered the ENQ. This gave a response rate of 42.9%. Although the data from those who did not respond was excluded from analysis, a brief review of both the ‘*non-response*’ and ‘*response*’ participants is given below for interest purposes. Following the demographic section, only data from “*response participants*’ are analysed and thus, will be referred to as ‘*parent participants*.’

##### **4.4.2.2.1 Non-response participants**

All 12 non-response participants were female and White, (M = 100%). The majority were aged between 30-39 years (66.7%, N = 8), with 3 participants being aged 40-49 years (25%) and 1 participant being aged 50 years plus (8.3%). 75% (N = 9) of non-response participants classed themselves as employed compared to 25% (N = 3) who classed themselves as homemakers.

All but 1 participant had a child with a diagnosis of non-hereditary unilateral Rb (91.6%, N = 11). The other diagnosis was non-hereditary bilateral Rb. In 58.3% of responses, the child was male, with the remaining 41.7% being female. The majority of children had their first artificial eye at age 1-2 (66.7%, N = 8). This was followed by ages 3-4 (16.7%, N = 2) and 0-1 years (8.3%, N = 1).

All non-response participants reported their children wearing only 1 artificial eye. 58.3% of the participants’ children have had between 4-8 artificial eyes fitted. This is followed by 33.3% (N = 4) whom have had between 9-12 artificial eyes and 8.3% (N = 1) whom had between 1-3 artificial eyes fitted. Interestingly, the latter is the child with non-hereditary bilateral Rb whom had their first artificial eye at a later age of 5-6 years compared to the other children as reported by the participants.

##### **4.4.2.2.2 Response participants**

All 12 non-response participants were female and White, (M = 100%). The majority were aged 30-39 years (44.4%, N = 4) followed by 40-49 years (22.2%, N = 2), 18-29 years (22.2%, N = 2), and 50 years plus (11.1%, N = 1). 77.8% (N = 7) of participants classed themselves as employed compared to 22.2% (N = 2) who classed themselves as homemakers (see Table 18).

Table 18: Demographic information of parent participants'

Participant Name	Child's Diagnosis	Age of Child	Gender of Child	Number of artificial eyes	Age of first artificial eye	Number of artificial eyes fitted
1	Hereditary bilateral retinoblastoma	4.5	Female	1	1-2	4-8
2	Hereditary bilateral retinoblastoma	7 & 9	Male x2	1	3-4	4-8
3	Non-hereditary unilateral retinoblastoma	16	Male	1	1-2	13-16
4	Non-hereditary unilateral retinoblastoma	4	Male	1	1-2	9-12
5	Non-hereditary unilateral retinoblastoma	5	Male	1	0-1	4-8
6	Non-hereditary unilateral retinoblastoma	5 years 9 months	Male	1	0-12	13-16
7	Non-hereditary unilateral retinoblastoma	5	Female	1	0-1	13-16
8	Hereditary bilateral retinoblastoma	8	Female	2	1-2	9-12

Six (67.7%) participants have a child with a diagnosis of non-hereditary unilateral Rb. The remaining 33.3% (N = 3) of participants has a child with a diagnosis of hereditary bilateral Rb: 1 participant of which reported having two children (both male) with this diagnosis. The age of participant's children ranged from 4 – 16 years. In 33.3% of responses, the child was female, with the remaining 67.7% being male.

Whereas 88.9% (N = 8) of participants children had 1 artificial eye, 11.1% (N = 1) have 2 artificial eyes. The average age of having their first artificial eye fitted was 1-2 years (55.5%, N = 5), followed by 0-1 years (33.3%, N = 3) and 3-4 years (11.1%, N = 1). 33.3% (N=3) had between 4-8 and 13-16 artificial eyes fitted respectively. 22.2% (N = 2) had between 9-12 and 11.1% (N = 1) had 17 plus artificial eyes fitted.

#### **4.4.3 Findings from 13-16 year old participants**

##### **4.4.3.1 Superordinate theme 1: Artificial eye and the self**

This theme relates to how a person views themselves and the role that their artificial eye plays in this. In other words, whether their artificial eye is part of, or separate to their self. This theme also represents changes to perception of the self. How we view ourselves and present to others is based on various constructs such as our social relationships, familial relationships, abilities, beliefs and disabilities. Evolving with age, our sense of self is often subject to changes, related to both situation and time. This is more apparent in children and young people whom learn what is acceptable and how to function in various situations through interactions with others.

Having a disability can affect an individual's sense of self including their identity and self-worth. Whilst some individuals will have a positive sense of self in relation to their disability (known as *disability identity*), others will have a negative sense of self. With sense of self being transient, it is common that an individual's sense of self in relation to their disability can change. Hence, the theme of *artificial eye and the self* represents how one identifies themselves to the self and others and how this evolves as a result of age changes.

##### **4.4.3.1.1 Subordinate theme: Artificial eye and sense of identity**

The first subordinate theme that emerged from 13-16 year old participants presents as a prominent subordinate theme that encapsulates how an artificial eye(s) relates to one's identity. Wendy clearly states that her artificial eye is an integral part of who she is. By not separating her artificial eye from her identity, Wendy reflects that her artificial eye does not change how she is as a person.

**Wendy:** "I don't think it changes how I am, like how I present myself because, I've always, it's always been, it's it's, part of me." (p. 1, lines 71-72)

There were similarities among both participants regarding seeing their artificial eye as part of their identity.

**Polly:** "... coz of I had it since I was little, I see it as just something that I've got to get done so. It's not something that like, oh gosh I got to go get it done." (p. 1, lines 39-40)

**Wendy:** "For me, it's, it is part of me. It is, because I've had it for so long and obviously, I was very little when it happened and I don't remember seeing out of two eyes or. So, I think if I remember it might be a bit different, but because I don't remember, it's just it's all I know and I don't remember anything different from it." (p. 3, lines 101-104)

Wendy reflects that the age of eye loss is a crucial factor in perceiving her artificial eye as part of who she is. Her response suggests that by having her eye removed before she recalled seeing from it eases any potential disturbance and distress during the transition as well as requiring a reduced adjustment and adaptation period. Consequently, the participants' day to day life is not adversely affected.

**Wendy:** "I'd still carry on, I'd still go to school, I'd still do my dance which I do outside of school and carry on with everything." (p. 3, lines 140-141)

#### **4.4.3.1.2 Subordinate theme: Age related to self-perception of having an artificial eye**

This subordinate theme is concerned with increased awareness of issues or concerns regarding participants' artificial eye in relation to age changes. Whereas subordinate theme 1 referred to this in a general sense, this subordinate theme marks out a very specific widening of awareness that relates to the association made between the artificial eye and how it looks during the course of aging. This experience was predominately expressed by Wendy. Reflecting on her experience, Wendy reported more noticeable differences between her artificial eye and *normal* eye as she got older.

**Wendy:** “But like, now, whereas, when it was first fitted it was fine and it looked the same as, as the other one. But I think as, I think my other eye, I don’t know what happened, but it’s just like, it’s become more obvious to me now that they aren’t the same shape or they aren’t the same.” (p. 1, lines 47-50)

It is apparent from Wendy’s experience that differences become more apparent the older she gets. Self-awareness comes with age, therefore as Wendy ages, she will not only become aware of how having an artificial eye may separate her from others, but also the differences between her artificial eye and her *normal* eye. Although Wendy reported that she did not know the reasons for the difference, further exploration signalled the growth of her socket due to the size increment of her artificial eye, as a contributing factor.

**Wendy:** I think it’s just like how, ermmm, open it is and I think that’s still to this day now, I still get self-conscious about my other eye being too small. Whereas, ermmm about a year ago ermmm my artificial eye was very closed so I had a new one ermmm and they made it a bit bigger which lifted my eyelid to make it appear more open. Whereas now, where it is more open, it’s made me self-conscious that my ermmm seeing eye is a lot smaller compared to it.” (p. 1, lines, 21-30)

Wendy’s experience seems to point to a potential relationship between awareness of differences between ones’ artificial and *normal* eye and their psychological wellbeing. Wendy’s reflection that “*still to this day now, I get self-conscious about my other eye being too small,*” points to an ongoing impact on her emotional wellbeing. By taking into consideration the natural growth of the eye socket, this impact does not appear to be debilitating.

**Wendy:** “I think having the new eyes, it’s quite, its quite, like, I don’t know,. It’s good experience because it’s like having a new change, but in a good way because you are, moving on and it, it has to be done because as your body still grows and your eye, your eyes are changing.” (p. 5, lines 193-196)



On the surface, there appears to be conflict between how Wendy views the fitting process. Whilst Wendy states that it is a good experience as it signifies moving forward, with it also comes feelings of self-consciousness and heightened awareness. Although the latter has the potential to lead to a low emotional wellbeing, this is not evidenced in Wendy's narrative. It appears that the balance between the two is acceptance of the process. Both Wendy and Polly stated that acceptance of the process comes from it being a normal part of their life.

**Polly:** "I guess it just goes back to the routine factor, it's just routine for me, I'm just used to it." (p. 2, lines 50-51)

**Wendy:** "So, for me now it is, its normal. But I think before it wasn't, whereas now I am used to having them remade and refitted and putting them in and taking them out." (p. 1, lines 10-12)

#### **4.4.3.1.3 Subordinate theme: Presenting the self to others**

Acceptance of the fitting process is often related to how the artificial eye and the person themselves are perceived by others. As a result, the way an individual presents themselves to the world is crucial to their sense of being. Through normalising their experiences, both participants felt that they should be treated the same as others whom do not wear an artificial eye.

**Wendy:** "I am lucky that my friends are supportive of and don't treat me differently and are open about it in like, talking to me about it... I don't want to be treated differently because it's just, it's just an eye that has, that I don't see out of anymore and like, apart from that I am, I am the same as everyone else." (p. 6, lines 260-261 & p. p. 2, lines 89-91)

Although Wendy distinguishes herself from others due to having an artificial eye, she does not see this distinction as warranting the need to be treated any differently to others. Being treated the same can imply that adjustments are not wanted based on an individuals' specific needs. Interestingly, and in parallel to being treated the same, Wendy highlights that her friends are supportive of her regarding her artificial eye. Rather than being a contradiction, it is likely that

Wendy is referring to being included in activities as anyone else would be. In other words, not to be singled out as a result of her artificial eye.

Being treated differently and potentially being excluded from activities was seen as partly being due to a lack of awareness and knowledge from others leading to assumptions being made. Polly reported encountering remarks from others due to a lack of understanding.

**Polly:** “I guess like there’s remarks sometimes when people see it. It’s like, I guess it’s a normal part. If people don’t know then, they’re just going to think, they - as much as they know so there is always going to be like those remarks so. I usually just like, if I hear something I will just address it. Actually, this is what is going on.” (p. 4, lines 157-161)

Both Polly and Wendy express the importance of being open and honest to friends, family and peers. Sharing their experiences with others helps normalise the situation which some may find difficult to approach.

**Wendy:** “I am very open about my eye and I do tell like whoever, if its ever bought up, I do tell. I don’t think I should hide it from other people or not tell other people ... if they are going to be so involved in my life, then they deserve to know.” (p. 2, lines 7071 & p. 6, lines 255-256)

**Polly:** “I prefer telling them so like ...I always get my friends to like, got any questions ask me like. I don’t mind, like, when I go on trips, and then I do my eye when I go away. Sometimes my friends are like, oh can we see the socket and I’m like go ahead because it’s something they don’t know about and it’s something I got so I am going to like share it with them so.” (p. 4, lines 143-146)

#### **4.4.3.2 Superordinate theme 2: The artificial eye process**

This superordinate theme represents participants’ experience of having an artificial eye fitted. Engaging with participants in this dialogue and then revisiting this experience through the

transcripts highlighted the importance in how the process is conducted and the participant's sense of identity. Trusting the prosthetist was seen as key in producing a functional and cosmetically pleasing artificial eye that was acceptable to the participant.

#### **4.4.3.2.1 Subordinate theme: Outcomes of the fitting process**

Both participants spoke about how they perceive the fitting process affects them and what changes they would like to see to provide future benefits. This particular subordinate theme was explored through additional prompt questions regarding the positives and negatives of the process. Whilst these sub-topics did not form part of the ENQ, the discussion of the impact of the fitting process came up in both interviews. With the nature of IPA research directed towards participants' experiences and their perceptions, there was a need to further explore the dialogue of the participants.

Reflecting on having an artificial eye fitted, both Polly and Wendy show an understanding of the need of some aspects to be uncomfortable to achieve a good outcome.

**Wendy:** "I wouldn't say it's a bad experience coz there's nothing, apart from the uncomfortable, ermmm, the gritty substance ... that's to get the shape to make it, so they know how wide it is gonna be or how close it will be which, that is a bit uncomfortable but as soon as that's removed its fine. It's just like a normal thing for me to have it done." (p. 6, lines 277-280)

**Polly:** "when I get tired and then it will get like sticky and then it will like, when it gets sticky, it might move a bit, like normally move to one side and then I got to pull my eyelid over it. And then where it - to like, - and then I'm back to normal ... Sometimes it will water and, or like there is something in the corner of my eye. I feel no way awkward about it because it is something I've just got to deal with so." (p. 3, lines 103-105 & p. 3, lines 125-127)

Both participants use the word *normal* to describe the process. The possibility of this indicating acceptance of the process as it currently stands was evident by the few suggestions for improvements.

**Polly:** “No, it’s like, it’s quite comfortable, but ermmm, it matches my real eye and it’s quite good ... the only thing really is like maybe if it could move like the real eye.” (p. 1, lines 12-14)

#### **4.4.3.2.2 Subordinate theme: Trusting the prosthetist**

This subordinate theme refers to the trust the participant places and has in their AEP. Not only is trust an essential prerequisite for the effectiveness of treatment, it is an important condition for a good relationship, therapeutic success and compliance. Wendy’s extract highlights how trust is linked to her psychological state of mind as well as her physical appearance.

**Wendy:** “Touching something like, you are messing with the, like, my face because if like something went wrong with it, it’s quite noticeable coz it, as it is part of my face, so I think for me I got to have the trust in them because that is, it’s a big part of my life and I think if it didn’t look right I’d be even more self-conscious about the fact and wouldn’t, wouldn’t want it to be as obvious as I think it would be.” (p. 4, lines 184-188)

Wendy emphasises how paramount trust is given the importance she places on having an artificial eye that is similar if not exactly the same as her normal eye. With trust being built as the relationship develops, a change in AEP will often place the patient-prosthetist relationship back at square one. In this case, the skills and expertise of the prosthetist is crucial in re-building trust.

Reflecting on their experience of having a new prosthetist, Polly and Wendy’s extracts highlight the importance of the prosthetist knowing their socket and what will and will not work. The AEP’s competency in their job and knowing what the patients’ needs and wants are contribute to a therapeutic relationship built on trust and resulting in patient compliance.

**Wendy:** “I used to have ermmm the same person, who I had I think had for about till I was about nine and then I had, I had a new person recently who has done this eye and she like was like really lovely and it’s been really good with fitting it and made the right shape and it was really good with the whole, the whole

experience and didn't seem awkward in doing it and wasn't, like she just, she said what she needed to do and that's what, like what happened and was, yea, she was really good, how she did it." (p. 4, lines 174-179)

**Polly:** "I think, the first appointment there was bit more instruction and she had to do like a new fitting. But. After that, she kind of knows what really I'm looking for so like, how I need it fitted and then, how my socket is. I, think it's the person knowing how your socket is and how it fits - So, her first ones obviously going to be like, 'oh this needs to be done or something like... 'Expect for that, she has done fine." (p. 6, lines 236-240)

#### **4.4.3.3 Superordinate theme 3: Coping with the impact of the process**

This superordinate theme relates to the way in which the fitting process and having an artificial eye impacts the daily life of the 13-16 year old participants'. Bringing with it physical and emotional implications, participants demonstrated the capacity to self-soothe and project a sense of resilience via the adoption of coping mechanisms. The sense of resilience and determination in coping with the artificial eye process was shown by participants' putting their situation in context and by living a *normal* life.

##### **4.4.3.3.1 Subordinate theme: Physical and emotional implications**

This subordinate theme represents the emotional impact of the artificial eye process for 13-16 year olds. Both Polly's and Wendy's narratives show the importance of using compensatory behaviours to adjust to the difficulties experienced as a result of having an artificial eye.

**Polly:** "It's difficult at times, but yea it is quite difficult at times coz like when you look at it, you're like, 'oh'. When I'm sitting down and watching films - and like, coz there is snapchat now, I use, like, it can knock my confidence a bit as I usually just take snaps on one side of my face." (p. 7, lines 310-313)

**Wendy:** "It took me about, a few days just to get to, just to like, when I look in the mirror to think oh, it's more open now because as I was getting more aware that it was a lot smaller, I was very, I'll be becoming self-conscious about that.

So, when it was new, it was like having it a bit bigger it was like having, it was, I don't know how to explain it. It was like trying to get used to that which took days and then it was just like, it's always been like that ... as the days go on its like having the one you had before in because it just feels normal.” (p. 3, lines 130-135 & p. 6, lines 272-273)

Although both Polly and Wendy were able to rationalise and manage changes to their appearance, heightened awareness of the limitations of having an artificial eye resulted in feelings of self-consciousness and lapses in confidence. Further exploration showed that this awareness was linked to the self rather than being a direct response from others' opinions. In other words, adaptations associated to having an artificial eye is more noticeable to those whom wear one than those who do not.

**Wendy:** “I think, more obvious to me because it is my own eye and I notice it more and because I know about it and I'm observing it in that way and taking note of it whereas like everyone who I'm, who I meet I tell, and they go oh you never know ... So, I think that it's probably more in my mental. It is in my mind but it's not, that it is, \* so obvious to other people.” (p. 2, lines 57-59 & p. 2, lines 65-66)

Wendy's comment that “*it is in my mind*” may imply that it is imagined, that what she is feeling is not real. The concern here is the risk of dismissing valid emotions that are essential in being able to adapt and adjust to the fitting process and of having an artificial eye. At various points during the interviews, both participants reflected on the times when they felt sad and frustrated about their situation.

**Polly:** “I find it just like a struggle because like, I can't see this side of my eye, get it right and it just won't look the same as this side would look. It's just, getting used to that. You can't exactly see over that side of your face to do your make-up exactly the same. So, it's just like, I spend a lot of time trying to perfect it, then I'm like forget it ... It depends on like the day, and the mood really as it can affect me more then it usually would so... Just the fact that I do have one eye. It's like maybe why did it happen to me or even if it did, couldn't it off been

removed without me having a fake one like.” (p. 8, lines 350-353 & p. 7, 317-18 & p. 4, lines 81-82)

**Wendy:** “Every so often there will be a time when we will be in the car and I’m looking ahead and I realise I can’t see from a certain distance, whereas other people have that freedom to do so. And I think that makes me feel a bit different from others, but that’s not on a day to day basis. I don’t go to school and think of they got that and I don’t. It’s not something that happens every day.” (p. 5, lines 208-212)

Exploring these extracts at a deeper level, participants appear to easily shift their perspective from feeling low due to the differences between themselves and others to accepting their situation and getting on with their lives. Not dwelling on having an artificial eye highlights the participants’ resilience and effective coping mechanisms.

#### **4.4.3.3.2 Subordinate theme: Resilience after process outcomes**

Resilience is the process of adapting and managing in adverse situations. It can involve using both internal and external resources such as adaptive coping mechanisms and social support. By putting her situation into context Wendy is able to see the positives of her experience.

**Wendy:** “I think I carry on because it could have been worse, I could off not been able to see, I could off had both my eyes removed. So, I did come out of the situation in a positive way and I was able to see and I am going to be able to see. Whereas like, I know people who have had their eyes removed and can’t see. So, I think, they have had it worse off than me and they deal with it amazingly. So, I think why should I dwell on it if they can deal with a worse situation in a better way.” (p. 5. 217-220 & p.5 224-225)

Recalling and hearing others experiences of past events can have a negative impact on an individual’s coping mechanisms. By focusing on the present and thinking about the future, Wendy is further able to contextualise her experience allowing her to carry on with her day to day life.

**Wendy:** “When I hear stories about my treatment and like, how it did affect my mum and dad, that can upset me a bit sometimes. But at the same time, I’ve heard it a lot and I think it is just, it’s happened now and I think as much as it’s happening now, it’s in the past, that happened in the past so I think I will carry on with what I got and I like I said, I am lucky with what I have got and what I did come out with.” (p. 7, lines 296-300)

Being able to self-soothe improves emotional regulation where an individual is more accepting of their situation. Whilst self-soothing also reduces reliance on others, both participants stated that a social support network is valuable.

**Polly:** “They do like, it’s just the fact that they like, yea, as a support and like, I’ll ermmm, it’s just that check-up and just like that kind of support to say okay your artificial eye it will fit, it just, that comfort, confidence and stuff. That kind of helps like.” (p. 8, lines 361-363)

**Wendy:** “I have got a very supportive family who know everything and if I, if it is ever brought up they don’t \* shut down the conversation, or \* aren’t afraid to bring it up like. If someone brings it up I will happily talk about it. And it’s like the same with my friends, my friends are very open about it and they’ll think of something and will just say to me, well why did this happen, and will ask some questions about it.” (p. 6, lines 247-251)

#### **4.4.3.4 Superordinate theme 4: Role of parents**

This superordinate theme represents 13-16 year old participants describing how their parents help them in attending appointments and looking after their artificial eye. We saw in the subordinate theme of *presenting self to others*, the importance others play in the participant’s sense of being. Now we will see what role parents play in supporting the participant. Demonstrating a need for parents’ continual contribution illustrates the reliance participants have upon their parents. This theme also represents participant’s sense of being restricted in their involvement during appointments and how this impacts their sense of control over the process.



#### 4.4.3.4.1 Subordinate theme: Reliance on parents

Health appointments for children and young people require the parents to attend. Parent's level of input in these appointments often varies. Reflecting upon their parent's contribution to the fitting process, participants spoke of their involvement in terms of ensuring the artificial eye is cosmetically accurate.

**Polly:** "I guess, I don't actually do too much for myself, it's kind of like my mum will come with me and then she will tell me, oh it's not lined quite right or something ... will say that the colour could be a bit better or could be more lined up better." (p. 2, lines 79-80)

With the focus being predominately on the cosmetic element, Polly sees her contribution to the process as minimal. As a result, there is a reliance on parents to ensure that their artificial eye is as good as it can be. In contrast to Polly's experience, Wendy expresses contributing to the process as much as possible.

**Wendy:** "I'd say if it's not right but then my mum does help me if like I can't put it into words right, or mum was to say, extra little details that I might off missed out and. That's, that is with any appointment I have whether it's to do with my eye or not. Like I'll say what's happened or, if I'm feeling, if I've got pains or anything. But then if I've missed anything out my mum will just like fill in the, the little bits that I might off missed out or say the little things." (p. 6, lines 235-240)

Both Polly's and Wendy's extracts highlight that the contribution of their parent's is not related to the level or depth of their own contribution. In other words, there is a need to have someone present with the patient to offer additional guidance that best reflects the patient's wants and needs. In this situation, as the patient is a child, the parents are best placed to provide this support. The reliance on their Mum is seen as beneficial to the participant.

**Polly:** “I think it’s really helpful coz then, erm, I know that they want it to be fitted so people don’t actually notice it, that it’s fake. So, it’s like, I know that they do it, I probably won’t realise something and think that eye, yea it’s fine. But when I get home be like actually no its not and then, it’s just like ‘oh maybe it’s a bit too pointy and stuff like that’. So, my Mum would realise that and she knows what she is looking for whereas I’m kind of just like, ‘it’s okay.” (p. 5, lines 215-220)

Reliance on parents was not just fixed to the fitting process. Polly reflects that her parents will always play a role in situations that involves her artificial eye.

**Polly:** “Sometimes my Mum will have to ring up and say we ... want it polished.” (p. 6, lines 260-261)

**Polly:** “I don’t think I will go to appointments by myself, my mum and dad would drive me.” (p. 5, line 201)

#### **4.4.3.4.2 Subordinate theme: Change of role between the patient and their parents**

Whereas the subordinate theme *age related changes to self-perception of having an artificial eye* focused on age changes related to the self, this subordinate theme will address how age may change the role between parent and child. A noticeable change as a child and young person ages is that of independence. Polly reported that as a child her Dad took on more of a role in looking after her artificial eye.

**Polly:** “When I was younger my Dad would like tell me I need to go and do it. My dad would stay on top of it and that but he’s now at the point where, so it’s like it’s up to me and what I do.” (p. 2, lines 91-92, p.5, lines 207-208)

Delving deeper into Polly’s extract, there appears to be an element of responsibility associated with increased age. Whilst Polly reported that she has taken on this responsibility, how she looks after her artificial eye is different to how her Dad looked after her artificial eye.

**Polly:** “I do it but recently I don’t do it a lot just because ermmm, I don’t know, like, I like to feel like I am living a normal life. So, when in the morning when I wake up, it’s not the thing I think off: I need to go and do something extra.” (p. 2, lines 89-91)

Polly’s statement that *I like to feel I am living a normal life* is provided as a reason for the discrepancy between what her Dad did for her artificial eye and what she now does. Importance of fitting in with peers and social groups often overrides care and responsibility of other tasks as an individual age.

#### **4.4.4 Findings from parent participants**

##### **4.4.4.1 Superordinate theme 1: The onus is on the parents**

This superordinate theme relates to the responsibility placed on the parent in caring for their child who wears an artificial eye as a result of Rb. Furthermore, this theme represents the impact the fitting process has on the parent. As the child’s primary caregiver, parents are at increased risk of experiencing emotional distress due to changes in their family dynamics, socioeconomic status and their own coping strategies.

##### **4.4.4.1.1 Subordinate theme: Impact of having a child with additional healthcare needs**

With added responsibility of looking after a child with healthcare needs, parents experience additional pressures often resulting in reduced emotional wellbeing. Examples of these pressures were given by Sarah and Leah.

**Leah;** “We spend a fortune on his glasses and prescription glasses and I know this is because I want them to be extra cool and so people notice them and not necessarily his eye.” (p. 1, lines 36-38)

**Sarah** “We live in Andover and we travel to (healthcare centre) for Peter’s appointments. It takes about an hour and a half in the car.” (p. 1, lines 4-6)

In spite of these financial and time implications, the need to put their child first and for them to ‘*fit*’ in is of upmost importance. This was further evidenced by Leah whom went on to disclose how it is impacting her psychologically.

**Leah:** “It's hard taking so much time off work for appointments and I always feel really guilty for this. But I refuse to miss one as they are so important and I want his eye to be as perfect as it possibly can. I am going through a very difficult time at the moment and am suffering from anxiety and depression. It has all recently happened and I have been put on fluoxetine. I am seeing a Counsellor and it all came out that I am feeling guilty about John and his eye removal that I am to blame and should have done something sooner.” (p. 1, lines 18-24)

Leah’s extract clearly shows how her son’s diagnosis and on-going treatment and care have led to feelings of self-blame and guilt. The gravity of such feelings appears to have a profound influence on Leah’s own mental health and sense of wellbeing. These feelings are particularly strong when they are backed up by beliefs such as they had ‘*faulty*’ genes which they have passed onto their child. Given the situation, these parents find themselves in, it is hard to imagine how such experiences would not have some kind of negative impact on these individuals, most likely questioning their abilities as a person and as a parent.

Yet, in spite of its toll upon the parent, participants exhibited an unwavering drive and passion for their child to receive the best care and treatment. As a consequence, acceptance of the situation they find themselves in often takes place. Whilst this was evidenced by Sarah, she goes on to express how this does not prevent her from desiring a different outcome.

**Sarah:** “As for myself and my husband we accepted the fact Peter would have to have an artificial eye from the beginning, as removing his eye was the only way to get rid of the cancer ... Of course, I wish he had been able to keep his right eye and have the eye sight in it, But, he is safe and free from cancer and to us that is what matters most.” (p. 1, lines 25-27 and p. 1, lines 37-39)

Throughout the online interviews, participants expressed continual worries of their child's current and ongoing wellbeing. Psychosocial worries, and fears around social relationships and progress made in the fitting process were most prominent. These concerns were particularly well expressed by Sarah.

**Sarah:** "At his last appointment, he did for the first time sit on his own in the chair and let Louise put his eye back in. She quickly popped it back in and I think he was taken aback a bit as he wasn't really expecting it. But it was a massive step for him. I just hope that our next visit in August will go that well but from the start." (p.1, lines 19-23)

**Sarah:** "He starts school in September so this will be a new step for him. The only thing that worries me is there is obviously older children and they are more likely to notice and say something (as children do). I just hope my thought of his, don't care attitude is right." (p. 1, lines 30-32)

Both of Sarah's extracts emphasis's *hope*: not only for her child to have positive experiences, but also that her perception of her son is right. At the opposite end of the spectrum to despair, hope is the expectancy of good in the future. It plays a role in the successful coping with illness and/or long-term treatment as well as improving the quality of an individual's life. Sarah's use of the words '*I hope*' intimates the vulnerability of her situation. With Sarah's potential wellbeing being dependent on her son's experiences, she is at risk of extreme thought processes rooted in fear and negativity which can make *bad* situations worse. Being able to separate the needs and experiences of her child from her own is essential in creating a healthy balance.

#### **4.4.4.1.2 Subordinate theme: Support and recognition from others**

Often, parents view looking after their child's additional healthcare needs as an isolating experience. This in itself can result in dependency, not only from the child onto the parent(s), but also from the parent(s) onto the child. Participants highlighted the importance of support from within the family unit, particularly when they need to juggle their family life in being able to attend appointments.

**Andrea:** “Me and my husband attend with our daughter.” (p. 1, line 1)

**Emily:** “Now I usually attend by myself with Anna, or sometimes I have to bring one or two of her siblings with us.” (p.1, lines 8-9)

Support from others was stated by participants as being pivotal to not only their wellbeing, but also that of their child’s. Involving those whom play an important role in the child’s life, not only creates understanding of the process and the child’s needs, but will also goes some way in reducing the onus upon the parent(s).

**Leah:** “We have had Johns nursery and school teachers attend sessions with us to see how to take out, care for and replace eye if needed.” (p. 1, lines 4-5)

**Emily:** “I found having the play specialist with us was important not just to Anna but to me as well. I remember after one of our first appointments she sent me a certificate in the post - not for Anna, but for me! We had been talking about how it felt like a big achievement to have got to the stage where I could help Anna take her eye in and out and I didn’t feel like the prosthetist recognised it, so she obviously decided to recognise it instead!” (p.1, lines 13-18)

Alongside, and closely linked to support, is recognition from others. As Emily clearly states, often the patient is the one who gets supported and recognised for achieving milestones, rather than the parent. However, as the primary caregiver, parents not only look after their child’s medical and physical needs, but also their emotional needs and the impact that ongoing treatment has on them, their family and the wider community/social network. With the little things often making the biggest difference, both support and recognition of the parent as well as the patient is vital.

#### 4.4.4.1.3 Subordinate theme: Normalising the experience

Parents use a variety of methods to help normalise their child's experiences of having an artificial eye(s). This includes, watching videos, seeing pictures of other children with an artificial eye(s) and celebrating the anniversary of their eye removal.

**Emily:** "We found it really helpful to show Anna photos/videos on YouTube and the rb parents group of the fitting process before we go ... It's also good for her to see all the pictures of other children with 'special eyes' and feel that it's something that is not just experienced by her." (p.1, lines 40-41 and p. 2, lines 60-61)

Through normalising the experience, patients are more likely to accept the process, and for some, get involved with it.

**Leah:** "We are extremely lucky as John has always happily been involved in the appointments and sat still so they have always gone really quickly. I always take pictures and talk to him and we are very open about his eye and celebrate a happy eye day each year on the anniversary of its removal." (p. 1, lines 14-17)

However, despite attempts by parents to normalise the process, it can still remain a daunting experience for the child patient. With the way children react to treatment depending on several factors such as their personality, their family and their developmental stage, it is possible that as the child ages and the more experience they have of the process, the more adjusted and accepting of it they will become.

**Sarah:** "I have shown Peter videos of children having their artificial eye removed and I've read him books about it, I got a DVD from Birmingham Children's Hospital and a little book from the artificial eye clinic. But it seems as though he has convinced himself it's going to hurt." (p. 1, lines 16-19)

#### ***4.4.4.2 Superordinate theme 2: Process repercussions on the patient and how it can be managed***

This superordinate theme relates to the how the artificial eye process affects the patient and how this can be managed. More specifically, the role of the AEP in helping/enabling the patient to accept and adjust to the fitting process and living with an artificial eye. Demonstrating a need for the prosthetists to understand and manage the patients' emotional wellbeing, illustrates the multi-faceted role of, and the responsibility and expectations placed upon the prosthetist.

##### **4.4.4.2.1 Subordinate theme: Patient impact of having an artificial eye fitted**

This subordinate theme represents parents' perspective of the impact the fitting process and having an artificial eye has on their child's daily life. This includes the emotional impact of attending appointments and the physical implications of wearing an artificial eye. Leah reported that the physical implications of having an artificial eye has made her aware and protective of her child's remaining eye.

**Leah:** "We are constantly mindful that he has to protect his remaining good eye ... He got hurt in school this week and that was scary and we really need to consider sports goggles for him instead of his glasses (he is long sighted like me) for play time as it will protect better. It's the only thing he has said no too and they were awful." (p. 1, lines 26 and p. 1, lines 33-36)

Alongside adaptations made as a result of having an artificial eye, attending the appointments were seen as being distressing for the patient.

**Sarah:** "Every appointment Peter will have to sit on his Dads knee and be held so his eye can be removed. He hates it!! Crying and trying to escape! But it has to be done else we would never get his eye out to be able to clean it or change it." (p. 1, lines 14-16)

In addition to the distress, Sarah's extract highlights the necessity of the process and the role of the parent in supporting their child in coping with the appointments. Whereas Sarah's child



experienced distress due to having an eye fitted, the distress in Emily's child was a result of having her artificial eye taken away.

**Emily:**” She finds it difficult when they take her eye away to polish it and is always very eager to have it back in - I think it helps her to value it when she sees it being painted, etc.” (p. 2, lines 58-60)

Emily saw the difficulty experienced by her child as a positive. By valuing something you hold it in high regard, therefore you remove the negative connotations and feelings such as embarrassment that often comes with having something that marks you as different to others. Emily goes on to say that positive reinforcements can help in a patient's ability to cope with attending appointments.

**Emily:** “I think kids respond really well to positive reinforcement and yet the service doesn't even give them stickers after an appointment, like you would get at the dentist. Having some 'rewards' would work really well with some of the little ones - balloons/stickers/cards/small toys.” (p. 1, lines 44-47).

Positive reinforcement allows children to grow more confident, self-reliant and independent. It can be particularly beneficial when children are struggling to come to terms with a situation. By addressing specific thoughts or behaviours their child has, parents are able to convey their belief in their child's ability which will help them internalise a positive identity.

**Leah:** “John has recently said some negative things about his eye and that was very upsetting but we talked it through and he seems back to his usual happy self about it all.” (p. 1, lines 31-33).

#### **4.4.4.2.2 Subordinate theme: Role of prosthetist**

Participants reported the importance of the AEP in helping manage their child's adjustment and acceptance of having an artificial eye. Consistency of process and AEP was seen as paramount.

**Sarah:** “Firstly, we saw a lady called Samantha. She was great with Peter and I could tell she had lots of experience. After seeing Samantha for about a year, we were told she had left. I was really disappointed as she was very good at her job and I she was just a lovely lady ... A new lady called Louise took her place ... After seeing Louise for the past year, she has come to know Peter and what he's like. She is very patient and professional.” (p. 1, lines 7-10, p. 1, line 10 and p. 1, lines 23-24)

**Leah:** “We used to see Samantha but she moved areas and now see Louise. Luckily, they have both been amazing and very supportive and happy to listen to our opinions about the look of the eye and I feel that this is an important part of the process.” (p. 1, lines 7-9)

Participants further reflected on how familiarity with the AEP and both their child and themselves being involved in the process helps normalise the experience.

**Emily:** “Now that we have got to know the prosthetist we look forward to seeing him. Anna always talks about her appointments with him and has warmed to him over time. She'll give him ‘high fives’ at her appointments and is less reluctant to let him make adjustments ... .. the prosthetist is happy for me to be involved and usually I will take the prosthetic/wax impression, etc in and out as Eliza would rather I do it.” (p. 2, lines 66-68 and p. 1, lines 31-32)

#### **4.4.4.3 Superordinate theme 3: Service experience**

The two superordinate themes explored above have highlighted the impact having an artificial eye fitted has on both the patient and their parents. With distress associated with negative outcomes such as poor satisfaction with care and poorer quality of life across various domains, this superordinate theme reflects parents’ views of the service as a whole and the processes employed.

#### 4.4.4.3.1 Subordinate theme: Service delivery outcomes

Healthcare service delivery outcomes are commonly focused on quality of care. Outcomes need not only meet the requirements of the patient, but also be in keeping of realistic expectations of the patient's family members. This subordinate theme addresses parent's views of how the artificial eye service is delivered and how it can be improved from the perspective of parents.

Although the subordinate theme "*Role of the Prosthetist*" highlighted patient's satisfaction with their work, participant's reflection of the service revealed a level of discontent. Service cuts, lack of training guidelines and the process being time consuming contributed to participant's feelings of a stressful and at times, traumatic experience.

**Rebecca:** "Poor and traumatic experience because many prosthetists are not specifically trained, having come from dentistry for example. Seems to be trial and error in fitting procedure with little technology. Some are good and most are caring but whole process is so stressful, time-consuming and expensive for my child especially since there are only two proper services in the UK." (p. 1, lines 3-6)

**Leah:** "The appointments vary in length depending on what is needed to be done. Recently there has been a considerable more amount of time in between fittings and getting a new eye and appointments due to the much higher demand for the service and cuts. This is extremely frustrating especially when they need a new eye as I strongly feel you should be able to get one straight away." (p. 1, lines 9-14)

Naomi reflected on the combining of services. Whilst this can improve efficiency not only for the patient and their families, but also the service, it can also result in a lack of support and guidance for those experiencing complications or difficulties.

**Naomi:** "At the beginning of our process we had a fab relationship with our prosthetist and clinic appointments were friendly and no pressure. Then the services joined and care for the eye was done at the hospital at clinic

appointments. This was initially brilliant because it meant we had one appointment instead of 2 but then when things were not going great medically we couldn't ask for the second opinion of the prosthetist.” (p. 1, lines 4-7)

#### **4.4.4.3.2 Subordinate theme: Artificial eye outcomes**

Having an eye removed is a traumatic experience not only for the patient, but also their parents. It is therefore imperative that the artificial eye is as aesthetically pleasing as possible. Dissatisfaction with the outcome often occurs when expectations are not met, whether they are realistic or not. This subordinate theme represents parent's satisfaction with the fitting process and outcome of the artificial eye. Although satisfied with the artificial eye, Leah reported a preference for it to be more lifelike.

**Leah:** “His eye is great and very realistic and the majority of people would never know it was artificial. However, I wish it could be even more realistic and move a little more.” (p. 1, lines 38-40)

Whilst Leah's experiences appear to be positive, the majority of participants reported the outcome as being unpredictable and at times, substandard.

**Lucy:** “We have always had issues with the prosthetics turning whilst she has them in.” (p. 1, line 4)

**Andrea:** “Frustrating that our daughter is so compliant yet outcome of good or bad fit is so unpredictable.” (p. 1, lines 3-4)

The frustration experienced by Andrea can often impact the child's adherence to the treatment programme and can result in blame being placed on the service and/or the healthcare professional. With the ability of a prosthetist to fit an artificial eye being heavily dependent on the condition of the socket, not all outcomes will be positive or as expected. Naomi reflected on a surgery that did not go to plan resulting in her child not being able to wear an artificial eye.

**Naomi:** “A plastic surgery op went very wrong and ... at present our son has been unable to wear an artificial eye for 5 years.” (p. 1, line 8 and p. 1, line 1)

Rachel and Claire reflect further on the complications they have experienced relating to the fitting process and the best approach to overcome these difficulties.

**Claire:** My son had so many problems with getting eyes that fitted him correctly (at two different services) leading to bleeding and cysts and an operation. I have made numerous complaints, wasted appointments where the eyes were wrong/poorly made and numerous visits to A&E.” (p. 1, lines 3-5)

**Rachel:** “I feel better fitting of socket is done under anaesthetic so less movement of muscles. Have had both boys experience prosthetist fitting and this gives to more infections.” (p. 1, lines 3-4)

These extracts highlight a potential disconnect between expectations and experiences. Our expectations of our experiences dramatically colour not just how we experience waiting for them but the experiences themselves. It is therefore vital that the prosthetist clearly states what is and what is not feasible, adjusting their perspective as the process continues. This will help manage the impact of the process for both the patient and their parent(s).

## **4.5 A reflexive account**

Two key components of qualitative research are conducting pilot studies and providing a reflexive account (Kim, 2011; Ortlipp, 2008). Whilst pilot studies are common place in qualitative research, researching a small population often means that there is limited scope for formal piloting. To ensure that the researcher is asking the relevant questions and they are getting the data needed, Braun & Clarke (2013) recommends thoroughly reviewing the interview guide for the first couple of interviews. With questions often developing and evolving when issues arise, constantly reviewing the data allows questions to be reworked, removed or new questions added (Braun & Clarke, 2013). Therefore, in place of a pilot study, a reflexive account is provided that includes review of the data in terms of its development, collection and analysis.

Willig (2008) states that there are two types of reflexivity; *personal reflexivity* whereby you reflect on your own experiences, and *epistemological reflexivity* whereby you question the construct of the research and the assumptions generated through the course of the research. In order to provide (accurate) reflections for both, my thoughts and experiences were collated through the research process: from its development, through to write up. This not only contributed to continual learning of the process, but also allowed for modifications and adjustments based upon the methodology and through the experiences encountered.

When reflecting on one's own research, it is always important to reflect on the researcher's professional background. With a background as a mental health practitioner, I was aware of how my training could play both a conscious and sub-conscious role in the development of the ENQ, collecting data and analysing the data. Bracketing off my pre-conceptions (a vital component of IPA research) was found to be difficult, specifically in the early stages of data collection and analysis. Although Dahlberg et al. (2001) reported that it is impossible to bracket off all our pre-conceptions, I did notice that as the face to face interviews progressed, my pre-conceptions reduced. Whilst my pre-conceptions played less of a role in the parent participants' interviews due to them being conducted via an online platform, I was aware that the discussion points put forward may of guided the participant to answer in a particular way. Whilst initial pre-conceptions are instinctive, I believe my curiosity in the varying perspectives of this phenomenon made me open to new insights, therefore resulting in my pre-conceptions reducing (LeVasseur, 2003).

Whilst I have conducted face to face assessments before, I had not done this in a research interview capacity or with young people. Attendance to relevant workshops, seeking guidance and advice from my supervisors with such experience and referring to key literature on interviewing young people, contributed to the practical knowledge required. However, having an experience led learning style, I found that it was not until the second interview that I was able to actively engage with the participant with confidence. For example, by the second interview, I was able to really get to the crux of the ENQ more effective probing questions and by listening to the participant's verbal and non-verbal cues. Not only has this enabled me to get a rich data set, it has also provided me with a new skill set that I endeavour to enhance and build upon in future research-led interviews with children and young people.

Reviewing the questions as data is collected allows the standard procedures of obtaining consent, collecting data, debriefing and managing confidentiality and anonymity. The face to face interviews highlighted these areas were dealt with successfully. The areas that needed slight amendment were associated with the delivery of the face to face interview, particularly leaving longer gaps after asking the question to allow the participant enough time to think about the question and answer it a way that best suits them.

Keeping on track was another difficulty initially experienced. This was particularly prominent in participants that did not have much to say regarding the fitting process: due to the positivity of the experience or due to the participant not being forthcoming during the interview. As the face to face interviews progressed, I was more able to draw the participant's focus back to the fitting process through expanding on what they said and how that relates to the fitting process.

Furthermore, I often found that discussion turned to the process as a whole rather than just focusing upon the fitting aspect. This is likely due to being a novice researcher and not having interviewed young people before. However, as the interviews progressed, I found myself more able to stick to the topic through using prompts and probes. This not only produced thicker and richer descriptions, but also increased my confidence as a health researcher interviewing young people.

Having worked in a mental health capacity, I noticed that some of the questions raised in the face to face interview evoking responses in me. This included thoughts such as 'This sounds interesting', 'I wonder what led to these feelings', to 'How has that impacted the way you deal with everyday situations?' This self-awareness led me to be mindful and cautious not to project my personal stance onto the participant during the interview.

Whilst the limited input into the responses of parent participants' can be seen as a benefit of the IPA methodology as it is more participant led reflecting their own experiences, I noticed that the majority of responses were not as detailed as I had hoped for. Furthermore, areas that I had assumed would have been discussed were not mentioned. Whilst, these can be seen as limitations of the data collection method, it may also be a reflection of the information provided by the researcher. Being able to clearly express the information and level of detail required without influencing the direction of participant's answers is a difficult task and one that requires a lot of

training and experience in. As a novice researcher, this is something that I envisage will develop over time.

Regarding the data analysis stage, and in keeping with the systematic method by Smith et al. (2009), I analysed each transcript as soon as it was transcribed (in the case of face to face interviews) and analysed the data from the online interviews as soon as it was completed. Although I analysed the data using the step by step guide by Smith et al., (2009), at times I found this process difficult due to my pre-conceptions.

My interpretations were particularly obvious during stage three, where the essence of what was being said as well as explicit reference to a key term or word was used in the development of emergent themes. To ensure that this represented the participants experience as much as possible, my preconceived notions and knowledge attained from experience in the field and existing literature were put to one side, allowing the experience of the (individual) interviews to guide the data analysis stage.

Furthermore, being new to phenomenological research, specifically IPA, I was overtly aware of how I handled and engaged with the data to create the themes as not to base them upon my subjectivity, but rather with an open-mind. Although, I found that I discussed the experiences of all participants, I found that some extracts overlapped, making it difficult to place within a theme. Through the process of checking back through the transcripts, I was able to differentiate between extracts, placing them into the relevant themes.

Whilst the data from both population groups were analysed using Smith et al.'s (2009) IPA method, I was concerned by the potential influence the different data collection methods may have had. By ensuring that both population groups were asked the ENQ at the start of the interview and the discussion points offered to the parent participants were similar to those used in the face to face interviews of 13-16 year old participants, the effect of the different data collection methods were minimised.

Reflecting on this component of the study as a whole, I have found it to be a positive experience whereby I have learnt copious amounts regarding the IPA methodology and myself as a qualitative researcher. As a person-centered individual that has an unwavering interest in understanding an experience from varying perspectives, I feel that as a researcher I sit well within an IPA framework



This component of the study has highlighted my strengths and weakness as a researcher. Whilst I have strengths in being able to develop rapport with participants enabling me to draw out responses, getting to the crux of the research question by keeping on track had proved difficult, and is therefore in need of further development. My knowledge of IPA has increased considerably from its theoretical to practical application. However, as this has been the first time I have used IPA, there is a lot of knowledge that I am yet to obtain through experience. Being in the early stages of my career, these skills are likely to develop and evolve as I progress as a researcher.

## **4.6 Discussion**

### **4.6.1 Introduction**

This section discusses the findings of this component of the study from each population group with their generated superordinate and subordinate themes. The interpretative element of IPA includes engagement with existing theories and evidence that are relevant to the phenomena under investigation and which may contribute to the secondary sense-making process (Larkin et al., 2006). In line with this, the discussion section draws on a range of evidence and theories in relation to the research findings. Implications of the findings and a reflection of the limitations of this component of the study will follow future recommendations for improving the fitting process of artificial eyes in children with a diagnosis of Rb can be found in the discussion chapter of this thesis.

### **4.6.2 Summary of findings from 13-16 year old participants**

The lived experiences of Polly and Wendy presented the reader with four superordinate themes that both participants reoccur in. In other words, the analysis of the findings highlighted that there was homogeneity among 13-16 year old participants experience of having an artificial eye fitted. A prominent similarity shared by Polly and Wendy was their resilience and acceptance as they described their lived experiences. Manne et al. (2015) state that resilience enables positive coping mechanisms which promote positive emotions: one of which is acceptance of the self.

#### ***4.6.2.1 Superordinate theme 1: The artificial eye and self***

In this first superordinate theme, participants shared how they see themselves in relation to their artificial eye. Children with cancer transition from childhood to adolescence can result in

increased vulnerability at a time when they are already vulnerable due to natural age changes (McClellan et al., 2013). This vulnerability was not evident in either Polly's or Wendy's narratives, suggesting that having an artificial eye does not change how they are as a person. As a congenital disfigurement, both participants are likely to be better equipped to adjust than individuals with an acquired disfigurement (Atay et al., 2003; Fu et al., 2011).

The primary reason for the stable sense of self is the age of the participants' first artificial eye. As Rb typically occurs in patients before the age of 5 years, limited changes to self-perception will occur. Consequently, patients will adjust to having an artificial eye without too much difficulty. This is in contrast to de Vries & Friedrich (2014) whom state that changes to self-perception are common following a diagnosis of cancer.

Whilst a relationship between age and self-perception was not observed in this component of the study, Wendy's narrative suggests a link between increased age and awareness of her artificial eye. With awareness comes a feeling of reduced emotional wellbeing. For Wendy, this took the form of feeling self-conscious. Worries, fears and stress are common in individuals with a cancer diagnosis (Deshields & Nanna, 2010; Mellblom et al., 2014; Van Dongen-Melman, 2000; Wiener et al., 2014).

Alongside onset of wearing an artificial eye, an individuals' level of self-efficacy plays a role of self-acceptance which in turn reduces levels of social distress (Deno et al., 2012). By addressing any questions or comments as they come up, both Wendy and Polly's level of self-efficacy can be perceived as quite high. As a result, they are able to present themselves well to others through normalising their experience.

By looking beyond their disability Polly and Wendy appear to have accepted their artificial eye as part of their identity. Findings from this component of the study support Ezra and Newman (2011) and Clarke et al. (2003) research which state that self-acceptance helps individuals feel accepted by others and increases independence and self-confidence.

#### **4.6.2.2 Superordinate theme 2: The artificial eye process**

The second superordinate theme refers to participants' experience of having an artificial eye fitted. Receiving a good fitting was important for both participants as it directly relates to their emotional wellbeing and coping mechanisms. This is in keeping with Gallagher and MacLachlan (1999) findings of a direct link between treatment outcomes and patient satisfaction.

The attempt to make sense of the fitting process in relation to a life-changing event is important in constructing identity. Exploration of this varied somewhat between participants. Whereas Wendy emphasised the importance of placing trust in the AEP, Polly was more ambivalent. Of particular concern to Wendy was the potential noticeability of her artificial eye. Hill et al. (2011) found that the visibility of a condition rather than the severity of a disfigurement increases levels of distress. As noted in the previous superordinate theme, Wendy experiences heightened awareness of her artificial eye, particularly when the artificial eye is new.

Patient satisfaction and health outcomes are often seen as the catalyst for change (Covinsky et al., 1998). Although Polly and Wendy described parts of their artificial eye or the process as somewhat bothersome, this did not equate to their satisfaction levels. Being seen as a “*normal thing*,” or something that they “*have to deal with*,” meant that they accepted the process and outcome as it currently is. Furthermore, the skill and expertise of the AEP (which is vital for outcome success), as well as the need to have an artificial eye appeared to counteract this. This is in keeping with Magnusson and Ahlström (2017) findings that despite (minor) faults with a prosthesis, satisfaction is high as a result of need.

#### **4.6.2.3 Superordinate theme 3: Coping with the impact of the process**

This superordinate theme discusses the impact of the artificial process upon 13-16 year olds and how they cope with it. Psychological adjustment to a disability comes with emotional acceptance in all aspects of the patient's life including personal, professional, and societal (Gutterfleisch, 2003). Although both participants were well adjusted, they reported that having an artificial eye can be a *struggle* at times. This leaves them at risk of experiencing difficulties such as depression, anxiety, low self-esteem and low self-image (Deshields & Nanna, 2010; Gutterfleisch, 2003; Horgan & MacLachlan, 2004; Melbolm et al., 2014). This may lead to maladaptive coping mechanisms (Patenaude & Kupst, 2005). However, Polly and Wendy reported that the times they

feel low are short lived. Wendy reflected that whilst it will take her a few days to adjust to a *new* artificial eye, this does not stop her from carrying on with her day to day life.

The impact of the process and having an artificial eye accounted for both the physical and emotional implications. Unsurprisingly, the physical implications of wearing an artificial eye bought on the emotional response. For example, Polly reported that watching films and using snapchat can knock her confidence due to having to compensate for a lack of peripheral vision.

Clarke et al. (2003) found that alongside the physical and functional difficulties disfiguring eye conditions causes, a large proportion of patients experienced psychosocial difficulties associated with the appearance of their eye. Wendy's heightened awareness of her artificial eye and consequently her feelings of self-consciousness supports their findings. Whilst Polly reported emotional implications of having an artificial eye, less attention was paid to it during the course of the interviews. The varying levels of impact indicate that there is not a relationship between physical appearance and psychosocial adjustment.

The second subordinate theme of resilience complemented van Dijk et al. (2008) finding that children with Rb have a positive view of their functioning. Support from others (primarily their family and friends) was highlighted as a valuable asset to both participants. With behavioural problems being associated with low level of social support (van Dijk et al., 2009), Wendy's and Polly's support network is a likely contributor to their positive attitude to their experience. Addressing the emotional, psychosocial and physical impact experienced by the artificial eye wearer results in their increased emotional wellbeing (Ayanniyi, 2013; McBain et al., 2014). Adaptive coping mechanisms are essential for emotional regulation (Thompson et al., 2010). By putting their experiences in context, both participants were able to self soothe and carry on with their daily activities.

#### ***4.6.2.4 Superordinate theme 4: Role of the parents***

The final superordinate theme for 13-16 year old participants refers to the role of their parents in attending appointments and looking after their artificial eye. It is common practice that a parent attends healthcare appointments with their child: more so when the child has healthcare needs. Furthermore, it is often assumed that (on their own accord) parents take on an active role in their child's healthcare (McKenna, Collier, Hewitt & Blake, 2010). Interestingly, the first

subordinate theme highlighted that the participant needs their parent to be pro-active in their treatment and care of their artificial eye.

Polly's and Wendy's narratives points to reliance on their parents. However, there are differences to the level of reliance participants have on their parents. Whereas Polly solely relies on her mum to attend appointments and to make sure her artificial eye is cosmetically correct, Wendy will contribute as much as possible to the appointments, only relying on her Mum to add details that she has missed.

Research on psychological adjustment in individuals with prosthetics point to the importance of the patient feeling that they have control and autonomy (Butler, Turkal & Seidi, 1992; Desmonds & MacLachlan, 2002; Messinger, 2009). Polly reflected on increased responsibility as she has aged. Whilst she has taken on more of a role in looking after her artificial eye, its level has dropped compared to when her parents were more active in her care. Shakespeare (1996) state that individuals with an impairment often go through a process whereby they negotiate their lives as to be as ordinary as possible to retain some contacts with desired life-worlds. Therefore, it is possible that Polly is adapting (assimilating) to her social world to live her life like a non-disabled person (Murugami, 2009).

#### **4.6.3 Summary of the findings from parent participants**

The lived experiences of the 9 parent participants presented the reader with three superordinate themes. With only the final superordinate theme being present in over half of the sample, there was heterogeneity among parent participants experience of their child having an artificial eye fitted. A prominent similarity shared among participants was the need to improve the service and fitting process. Coulter, Roberts and Dixon (2013) state that there is a need to develop a practical, robust, reproducible and transferable delivery system which can improve the treatment and management of long-term conditions.

##### ***4.6.3.1 Superordinate theme 1: The onus is on the parents***

In this first superordinate theme, participants shared the role they play in, and, the impact of looking after their child who wears an artificial eye(s) as a result of Rb. Nagayoshi et al. (2016) found that mother's experienced high levels of stress particularly around the child's treatment due to psychosocial and medical worries. Furthermore, Van Dongen-Melman (2000) and Williams et al. (2013) reported that parents are more likely to worry on a daily basis about their child's

school performance, lack of friends and side effects. These findings were evident in Leah's narrative whereby she expressed emotional distress due to the guilt and blame she assigns to herself regarding her child's diagnosis and ongoing treatment.

In comparison to Leah's experience, Sarah reflected that both she and her husband have accepted that their son needed his eye removed, and thus, a lifetime supply of artificial eyes. According to Hamama-Raz et al. (2012), the difference between Leah's and Sarah's experiences may be due to an ability to separate thoughts and emotions whereby those able to do this are better equipped to cope and thus, have an improved quality of life.

Coping mechanisms are essential for being able to accept and adjust to difficult situations (Weintraub et al., 2011). Andrea, Emily and Leah reported how having a good support network (for themselves and their child) can improve their own emotional wellbeing.

To help their child adapt and adjust to having an artificial eye fitted, parents reported normalising their child's experience. Normalisation involves parents in promoting normal experiences for their child whilst assessing and adapting to potential problems or limitations resulting from their child's condition (Valentine & Lowes, 2007). Emily, Leah and Sarah reported using pictures, DVDs and celebrating the anniversary of their child's eye removal as effective techniques. With a parent's emotional wellbeing being closely tied to that of their child (particularly those with additional healthcare needs), normalising their child's experience is also beneficial for parents own adjustment and acceptance (Valentine & Lowes, 2007).

#### ***4.6.3.2 Superordinate 2: Process repercussions on the patient and how this can be managed***

This superordinate theme discusses parent's perspective of the impact the artificial eye fitting has on their child. Literature on disability has shown that these individuals often experience stress, worries, anxiety and depression (Van Dongen-Melman, 2000). However, the perceived impact (specifically having an artificial eye fitted) upon a patient from a family member has received little attention.

Parent's perspective of their child's functioning following a diagnosis of Rb has highlighted their general and emotional health as poor with more emotional and behavioural problems (Weintraub

et al., 2011). Whilst Leah's extract regarding John's negative self-talk about his artificial eye potentially supports the above finding, the use of positive reinforcement where specific thoughts and behaviour are addressed appear to negate parents negative view of their child's functioning. Emily, Sarah and Leah reported using positive reinforcement to support their child and reduce the impact that the artificial eye process has upon them. A possible explanation for this difference comes from Sheppard et al. (2005) whom found that the data collection method influences parent's views of their child's quality of life. Whereas interviews resulted in Mothers viewing their child's quality of life as good, standardised measures (which asked the same questions) showed that Mothers rated their child's quality of life as poor.

Participants' highlighted the difficulty of the process rather than the emotional and behavioural distress as contributing to their child's wellbeing. Consequently, these findings do not support previous research that found parents overrate the behavioural problems of their child as a result of their own coping strategies (van Dijk et al., 2008).

The second subordinate theme of '*role of prosthetist*' complemented Ross' (2013) finding that establishing rapport is essential to any healthcare professional-patient relationship. Emily revealed that by having gotten to know the AEP, her daughter is much more willing for him to make necessary adjustments. Developing a positive relationship whereby both the patient and parent can be involved in the process has resulted in better patient adherence, satisfaction and outcomes.

#### **4.6.3.3 *Superordinate theme 3: Service experience***

The final superordinate theme refers to parent's experience of the service, specifically service outcomes and the fitting process. Confronted with information that is difficult to understand and poorly communicated such as treatment process and outcomes, parents can become easily overwhelmed and misunderstand what is being said. Consequently, expectations may be unrealistic and distress can arise, putting both the parent and the patient at a disadvantage (Panton et al., 2009).

Both subordinate themes, "*Service Delivery Outcomes*" and "*Artificial Eye Outcomes*" highlighted parents' dissatisfaction. Rebecca, Leah and Naomi reported service cuts, ineffective

training programme, the subjective nature of the process and being time consuming as having a negative impact on both themselves and their child.

Lucy and Andrea reported that the subjective nature of the process makes the outcome unpredictable, causing frustration and the potential of impacting adherence to the treatment programme. Low adherence increases medical complications, contributes to poorer quality of life and an overuse of the health care system (Jack, McLean, Moffett & Gardiner, 2010). Managing mental health issues appropriately, building a strong relationship, customising the treatment programme, providing information and ensuring family and peer support can improve adherence in children and young people (Taddeo, Egedy & Frappier, 2008).

Dissatisfaction with the outcome is often directed at the professional that is unable to meet expectations, rather than the cause of the negative outcome. Naomi's extract highlights complications experienced prior to the fitting process, resulting in socket damage and an inability of her child to wear an artificial eye. With patient outcomes and satisfaction being dependent on not only the outcome of the product, but also the process, healthcare professionals need to manage expectations. This is often done successfully within an MDT environment, resulting in improved patient care and outcomes, enhanced coordination of care and improved mental well-being of patients and staff (Cancer Australia, 2017).

#### **4.6.4 Strengths and limitations of the study**

The main strengths and limitations of this component of this component of the study revolve around four areas: the participants, research design, researcher factors and reflection on quality in IPA research. Each one will be discussed below.

##### **4.6.4.1 Participants**

Although there is no prescription on the number of participants in an IPA study, the sample size achieved of 11 is seen as acceptable. However, there was a disparity between the number of participants in each population group. Whilst a sample size of 9 was achieved for parent participants, only 2 participants were achieved in the 13-16 year old participant population group. Emmel et al. (2007) reported that researchers face challenges in recruiting enough numbers of



participants in studies that are explicitly designed to reach small populations and ‘hard to reach’ groups.

With no pre-existing ties to 13-16 year old participants or Rb groups, access to this population group was based on their use of services for which the UK health departments are responsible for. With only one of three Rb services (2 NHS and 1 Rb charity) within the UK agreeing to participate in this component of the study, contributed to the small sample size achieved from this population group. Gatekeepers who restrict access to health research have been identified as a barrier for research participation (Bonevski, 2013).

Limited access has led to a sample size that is neither large enough nor sufficiently representative of the population to achieve results that can be generalised across the population group being explored (Mason, 2002; Sadler, Lee, Lim, & Fullerton, 2010). Whilst the same topics that were discussed by both participants can be seen as providing enough depth, it may be the case that the low sample contributed to a lack of opportunity to discuss other factors imperative to the fitting process of artificial eyes. Additionally, details of other factors mentioned may not off been explored to their full potential. For example, whilst participants briefly referred to the role their parents play in the fitting process, the impact of this upon the patient was not discussed. Consideration of, and the impact of where the participants were recruited needs to be examined. With both participants attending the same prosthetic clinic, it is possible that their experiences are similar to one another. Patton (1990) suggests that the sample should be judged on the context and rationale of the study and sampling strategy used to achieve the study’s purpose rather than the number of participants.

As a clinically relevant PhD that provides recommendations to improving the artificial eye process in children with a diagnosis of Rb, it was imperative that an adequate sample size was achieved. Ritchie et al. (2003) suggests that data can continually be added as participants are recruited. However, this was not possible due to the practical and time limitations inherent in a PhD. Therefore, further research into the fitting process of artificial eyes in 13-16 year olds is needed outside of this study component.

Tuckett and Stewart (2004) suggest applying different techniques of data collection to tackle problems stemming from sample size. This provides the researcher the opportunity to account for similarity and differences of the information collected (Greene & McClintock, 1985).

Consequently, other methods of accessing and recruiting participants were considered. This included accessing participants from artificial eye services, recruiting via (online) advertisements and employing various sampling strategies including those for 'hard to reach groups.' Accessing participants via artificial eye services would require agreement from gatekeepers, identification of participants that meet the inclusion criteria and substantial changes to the NHS Ethics Approval. With artificial eye services serving patients with various acquired and congenital eye loss or deformities, identifying relevant participants within confidentiality and anonymity boundaries would be challenging. Whilst placing advertisements may have negated the need for NHS Ethics, knowing where to advertise to reach the targeted population proved to be problematic. Sampling strategies such as snowball sampling are often used when potential participants are hard to find (Shaghghi, Bhopal & Sheikh, 2011). It involves recruiting one or two participants who can recruit other participants' that meet the criteria. Whilst this method would be advantageous in this component of the study, it was not until data had been collected from the two participants that the researcher was informed that there was no other participants. Therefore, involving those participants in recruiting other participants was not viable.

As a consequence of the above, recruitment of this population group came to a natural end. Whilst the low sample size has created strong limitations of this component of this study, the reflection by Jonathan Smith (the originator of IPA) that researchers are adopting very small sample sizes, and even single case studies, as well as the inclusion of the parent population group in this study component, provides reasonable justification for its inclusion.

For qualitative research, it is argued that the representation of diversity is often important (Allmark, 2004). The parent population group benefited from a diverse sample of participants including a range of ages of children wearing artificial eyes as a result of Rb.

However, a limitation of this component of the study was the gender of participants. Both population groups consisted solely of female participants. Consequently, it cannot be said whether the lived experience of the fitting process from the perspective of parents and 13-16 year olds are similar or different for male participants. In spite of these factors, this component of the study was able to generate some potentially transferable insight about the fitting process.

Furthermore, whilst there was an even split in the ethnicities of 13-16 year old participants (White and Black), all the participants in the parent population group were White and from the UK. As

a multi-cultural country, experiences from different ethnicities and cultures is worthy of future investigation.

#### **4.6.4.2 Research design**

Focusing on how participants perceive and makes sense of experiences which are happening to them, most IPA studies collect data via interviews. The type of interview mainly used is semi-structured where the researcher is guided by the set of questions they have developed rather than being dictated by them. This approach allows the participant to lead the direction of the interview and discuss a topic the researcher may not have thought of (Braun & Clarke, 2013).

Interviews have been criticised as they rely heavily on retrospective recollection. When discussing sensitive topics participants may experience self-preservation concerns thereby fabrication or withholding of information may take place (Brannen, 1988). However, it has been argued that sensitive topics can make interviews effective. This is because these topics often stand-out in the participant's mind therefore are recalled with greater accuracy than more mundane experiences. To ensure that participants felt able to discuss any part of the fitting process in the way that best suited them, a more flexible approach whereby the participant had control over the interview was chosen. Utilising an ENQ gave participants free-range over how they wanted to answer the question, giving them space to mention specific things about the topic that was particularly significant for them. Prompts and probes were developed for participants that either needed guidance in answering the ENQ and for enquiring more about an issue or topic a participant had raised. This study utilised both face to face and online interviews: the reasons for which have been stated in the 'data collection' section of the 'method.'

Whilst there are disadvantages associated with face to face interviews, such as time and money costs as well as the potential for interviewer bias, these are outweighed by the benefits. The benefits include building rapport, having the undivided attention of the participant, keeping the participant on track and being able to capture their emotions and behaviours as well as other (non) verbal cues (Braun & Clarke, 2013). This is particularly important when interviewing young people whom at the same time as showing maturity, process questions differently than adults (Sacks, 2003). Both the interviewer and participant being able to ask for clarification of the question and responses, as well as being able to directly react on what is said are further advantages of this method.

In comparison to face to face interviews, which are the longest serving qualitative data collection method, online interviews have only existed since the 1990s (Wilkerson, Iantaffi, Grey, Bocking & Rosser, 2014). The benefits of using an online method are discussed in component 1 of this study. This component of the study experienced difficulties in ensuring all participants answered the ENQ. A total of 21 participants responded to the online interview, but only 9 answered the ENQ. Furthermore, the level of detail provided in the ENQ varied greatly. Whereas some participants provided detail at great length, others only wrote a couple of sentences to a few paragraphs. Although it would have been more beneficial to interview parent participants face to face, this was not feasible due to the reasons already stated.

The decision to explore two population groups can be seen as either a strength or a limitation. Whilst the inclusion of both groups gives an added dimension to the phenomenon, it comes with a risk of interpreting data in light of what has been found in the other population group. Having to address the data with ‘two separate heads’ can be a challenging and demanding task where mistakes can be easily made. This was kept to a minimum by following Smith et al.’s (2009) IPA guidelines as well as Yardley’s (2000) appraisal of qualitative research. With improvements being created by understanding various perspectives of a given product, procedure or treatment, the inclusion of two population groups in this component of the study is seen as a strength. Yet, it must be understood that if this study was to be replicated using different participants (particularly due to the low sample size of 13-16 year-old participants’), different qualitative results would be produced.

#### ***4.6.4.3 Researcher factors***

As a novice researcher in phenomenology, this component of the study may have lacked the level of integrity of more experienced phenomenologists. By studying the methodology and seeking out support and guidance from researchers whom have utilised this method contributed to the essential skills required. Further development of the required skills was attained during the data collection stage where rich data and descriptions were produced.

Central to IPA research is the focus of understanding the lived experience of the participant. Consequently, it is essential that the researcher reflects upon their preconceptions about the data, leaving them to one side during the analysis stage. Although producing a reflexive account brought awareness of any preconceptions to the forefront, it was difficult to distinguish between these and what the data was telling me during the data analysis stage. Consistently checking the interview

transcripts throughout the analysis process and taking short and regular breaks (which gave the opportunity to refocus), went some way to ensure only relevant or significant themes were noted. Furthermore, by analysing the data of one population group at a time minimised overlapping and misinterpreting what was presented.

#### **4.6.5 Conclusion**

This component of the study has explored the lived experience of 13-16 year olds and parents of children who wear an artificial eye(s) as a result of Rb. This component of the study has not only equipped me with knowledge about the topic, but also my role as a researcher and the impact my preconceptions, assumptions and biases may have had on the way participants answered the experience near question and how their responses were interpreted during the data analysis stage. It is hoped that I have done justice to the experiences of all the participants interviewed with the sole purpose of having their voices heard. For my own ontological sense of self, this study is viewed as being the start of my research journey, thus will influence my present and develop my future in health and clinical psychology.

# **Chapter 5 - Transfer of knowledge in the assessment of prosthesis**

## **5.1 Introduction**

The previous chapters have highlighted two issues. The first is the impact the artificial eye process has on the AEP and the parents of, and, children with Rb, and the second is the need to consider development of the process in terms of technological advancement. The scoping review and component 1 of this study showed that recent advancements in this field have been AEP led, based on their experience and expertise. This has mainly involved technique variations in creating an impression of the eye socket and digitally creating the iris.

Regarding the literature and findings from component 1, high demand healthcare needs such as heart related conditions and diabetes often attract investment and funding compared to small healthcare sectors such as artificial eye services. With reduced resources, research and development in small healthcare sectors is often slow, sometimes remaining stagnant for a number of years. However, the need to meet patient expectations within healthcare and increased accessibility to technologies has led to technologies' recent application in small sectors (Eggbeer, 2008; Martin, Murphy, Crowe & Norris, 2006).

Although the use of medical technologies in artificial eye development is an area yet to be explored, its investigation in the similar field of maxillofacial prosthetics has begun. This has included scanning technologies that can capture the contours and fine details of the eye socket, 3D CAD software that can accurately represent the surface of the object, and rapid prototyping and manufacturing (RP&M) processes that use wax-based patterns to produce the prosthesis (Bibb, Eggbeer & Evans, 2013; Ciocca, Fantini, Marchetti, Scotti, & Monaco, 2010; Evans, Eggbeer & Bibb, 2004; Li et al., 2015; Peng, Tang, Li & Peng, 2015; Wu et al., 2009).

With the outcome of the prosthesis being dependent on the accurate representation of the deficit site, there is a need to focus upon the techniques used in the assessment process. Scanning technologies such as CT scans, MRI's laser and white light can accurately capture, represent,

manipulate and reproduce soft and hard tissue (Bi et al., 2013; Li et al., 2015; Wu et al., 2009). However, the level of investment required is often a contributing factor to lack of investigation, application and further research and development in small healthcare sectors (Eggbeer, 2008).

With the assessment of the eye socket being done manually, current methods are labour intensive, time consuming and skill/experience dependent (Bi et al., 2013; Dixit et al., 2005). The need for both functional and aesthetic considerations makes the application of technology into the artificial eye process important. Examining the technologies being explored in a similar field is a starting point to incorporating them into the artificial eye process.

### **5.1.1 Research aim and objective**

This component of the study aimed to create a transfer of knowledge between how AEP's and MP's conduct the assessment stage with a focus on the tools and equipment used. The objective is to see whether the equipment and tools used in maxillofacial prosthetics can be incorporated into the artificial eye process. With the limitations of using an online method incurred in component 1, this component used face to face interviews to elicit more detailed responses of the assessment process in both population groups.

## **5.2 Method**

### **5.2.1 Ethics**

Ethical approval was obtained from Bournemouth University Ethics Committee, reference number 14010 on 24/01/2017 (see Appendix D)

Ethical concerns related to this component of the study included anonymity, confidentiality and the voluntary nature of this component. The component of the study followed the Data Protection Act (1998) whereby all participant information is protected. Furthermore, participants were informed regarding the use of their information and were given a choice in how their information was disclosed or used in particular ways. By using audio recordings voice recognition was possible, thus, initially, the data was not anonymous. Two weeks following the interview, all responses were transcribed and the recording deleted ensuring anonymity of all data. All participants were clearly informed about this via the information sheet. Participants were given

the opportunity to ask questions and asked to sign the consent form once they understood the situation.

All information collected as part of this component of the study was kept strictly confidential in a secure place with access being restricted to the researcher. Participants were assigned a unique number for identification purposes only. This number was used for recording and transcribing purposes.

### **5.2.2 Sampling**

The sampling method used was purposive. Purposive sampling allows participants to be selected according to the needs of the study. It provides insight and ‘information-rich’ accounts of the topic of interest as well as a diverse range of views and experiences (Patton, 2002). Requiring opinions of people with a relatively high level of skill and knowledge in maxillofacial and artificial eye prosthetics, resulted in the subtype of expert sampling being used.

Expertise is not fixed; it is a dynamic relationship between the demands imposed by the environment and the resources of a particular person at a particular time (Olsen & Rasmussen, 1989). With no fixed way of defining an expert sample, it is often seen as problematic (Sackman, 1975; Skulmoski et al., 2007).

When deciding on the criteria for an expert sample, job qualifications and time in service are often used. However, as highlighted in component 1, there are no specific job qualifications to become an artificial eye prosthetist in the UK and years of service often centre around the higher end of 20 years plus. In order to get a wide range of perspectives from the population, this component of the study defined ‘an expert’ based on their current job title, experience and location of work. Thus, the expert sample consists of MP or AEP’s. All participants needed to be currently working in the field with a minimum of four years of experience. This ensured that all participants had up-to-date knowledge of the current processes utilised.

To ensure the widest range of processes utilised in maxillofacial prosthetics and artificial eye development, views were sought from these services throughout the UK. Consequently, a



moderate to large sample size was needed to provide enough data to fully explore the research aim (Braun & Clarke, 2013).

Taking into consideration the small population being explored and that the overall study is part of a PhD thesis where there are practical and logistical issues, a sample size of 12 was estimated to be sufficient. Data collection and analysis was run concurrently, stopping at 10 participants (4 AEP's and 6 MP's).

### **5.2.3 Recruitment**

Recruitment took place between January and April 2017. Potential participants were identified via an online search of maxillofacial laboratories and from the NAES within the UK. Telephone calls and email invitations were used as the main recruitment method. The email contained an information sheet (see Appendix G). The email requested participants to contact the researcher by telephone or email if they were interested in participating. A reminder email and telephone calls were made to participants two weeks after the initial contact to ascertain whether their interest and willingness to participate. Following agreement in partaking in this research, a date and time was scheduled at the participant's workplace.

### **5.2.4 Data collection**

Semi-structured interviews, best suited to qualitative methods, were carried out to create a transfer of knowledge between the methods employed in the maxillofacial prosthetic and artificial eye process. They were used to assess whether improvements can be made in the technological development of artificial eyes.

Semi-structured interviews are used to find out individual's experiences, perceptions, opinions and values. They combine structure and flexibility, which enables the researcher to obtain data that will answer the research question, while allowing unanticipated issues to be explored with the interviewee addressing topics in the manner and order most suited to them (Lewis et al., 2009; Willig, 2008).

All interviews were carried out at the participant's place of work to keep the level of inconvenience to a minimum. No incentives or remunerations were offered for taking part. Whilst there were no immediate benefits to participants from taking part, the results have the potential to improve the process of artificial eyes.

The average length of interview was approximately 32 minutes, ranging from 18.40 minutes to 43.01 minutes.

### **5.2.5 The interview guide**

An Interview Guide (see Appendix G) was developed containing open-ended questions designed to elicit key aspects of the participants' views and experiences. The questions used in the interviewing process were developed from the literature review and the findings from component 1 and 2. The key objective, while conducting the interviews, was to understand the processes and technologies employed at the assessment stage in maxillofacial prosthesis and the artificial eye process.

Questions were developed to ensure they were precise, singular, non-leading, non-assumptive, linguistically appropriate and empathetic (Braun & Clarke, 2013). The Interview Guide was organised so that questions were presented in a logical order, starting with the broadest issues, least difficult and most engaging for the participants. Latter questions were clustered around the key topics necessary in answering the research question. A summary of the topics included in the Interview Guide is provided in Table 19 in the order in which they were usually presented to the participants.

Table 19: Topics included in the interview guide

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1. The equipment/tools used in the assessment for a prosthetic fitting
2. The effectiveness of the equipment used
3. Complications associated with the equipment/tools used
4. Future improvements to the assessment process.

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Prompts and probing questions, as well as follow-up questions, were also included in the Interview Guide to assist the researcher in obtaining detailed responses related to different aspects of the research topic and to check the understanding of the participant's meaning (Kvale, 1996).

### **5.2.6 Pilot testing**

In order to assess how easily the questions could be understood, they were tested on colleagues and by refining them in the process of conducting the first few interviews with participants. Feedback from colleagues focused upon the question structure and layout. Questions were funnelled with the easiest and open questions at the beginning of the Interview Guide and the hardest and more complex questions being placed at the end. Furthermore, double barrelled questions were split into two questions.

Feedback from participants who produce artificial eyes highlighted differences in the assessment process. This varied between simply looking at the condition of the socket where no tools were used to taking an impression where tools are used. As this study is primarily looking at the tools and equipment used, it was decided by the researcher and supervisors that the assessment stage will also include taking an impression.

### **5.2.7 Conducting the interview**

On arrival for the interview, the researcher introduced themselves, provided explanation to why the study was being undertaken, went through the information sheet and gave participants the opportunity to ask any questions. Prior to questioning all interviewees gave written informed consent for their participation and completed a written demographic questionnaire. The interviews were recorded on a portable digital audio recorder which was placed between the interviewer and interviewee (See Appendix G for interview materials). Verbal and non-verbal communication tactics were employed to maintain engagement. This included repeating key words spoken by the participant to ascertain meaning and to elicit more of a response, as well as nodding and smiling to encourage the participant to continue.

To signal that the interview was coming to an end and to ensure all topics were covered, interviewees were asked if there was anything else they would like to add. Interviewees were then thanked for their participation and given a study debrief sheet which restated the purpose of the research, described how and when the results would be available and provided the researcher's contact information (see Appendix G).

### **5.3 A reflexive account**

Semi-structured interviews consist of key questions that define the topic, allowing the researcher to pursue a response in more detail and the participant to answer the questions freely. Reflecting on this component of the study enhances understanding of the results, can limit potential biases through awareness and can have a positive impact on future interviews.

I was aware of how my professional background in mental health and interest in the psychological aspects of a process can play both a conscious and sub-conscious role, from design to implementation to analysis and write-up. Utilising pre-prepared probes and prompts as well as taking brief notes during the interview helped ensure that I stayed focused on the topic area being explored. Furthermore, reference from the literature (and its gaps) and service visits, as well as receiving feedback from my supervisors and from the first few interviews were used in developing the Interview Guide and enhancing the questions.

Another area worth reflecting on during the data collection and analysis stages was my pre-existing knowledge of the equipment/tools used in the assessment stage of creating artificial eyes. By being mindful and cautious of the possibility to project my personal opinions and views onto the participant, I relied on open questions and allowed the participant to expand on points they saw as necessary. However, points raised numerous times throughout the interview that needed expanding on were asked in an open manner either at a convenient time during the interview or at the end.

To ensure that the data analysis and write up was a true reflection of the participant's experiences and perspectives, Braun & Clarke's (2013) thematic analysis was followed closely. In addition, checking and re-checking the data during the analysis process, as well as reviewing the themes with my supervisors, kept the analysis and write-up within the correct parameters.

### **5.4 Data analysis**

#### **5.4.1 Data management and transcription**

The digitally recorded interview files were transferred to a secure computer server. The interviews were then transcribed verbatim by the interviewer before being deleted from the audio

recorder. Transcripts were anonymised by removing all identifiable information and allocating each interviewee a unique identification letter which was used in the data analysis stage.

#### 5.4.2 Thematic analysis

Data was analysed using thematic analysis described by Braun and Clarke (2013) ensuring that analytical stages are carried out comprehensively, transparently and rigorously (see Table 6 in chapter 2). Thematic analysis is a systematic approach for identifying, analysing and reporting patterns or themes across a dataset (Braun & Clarke, 2013). A thematic map was developed to show how the themes and subthemes were developed (see Figure 8).

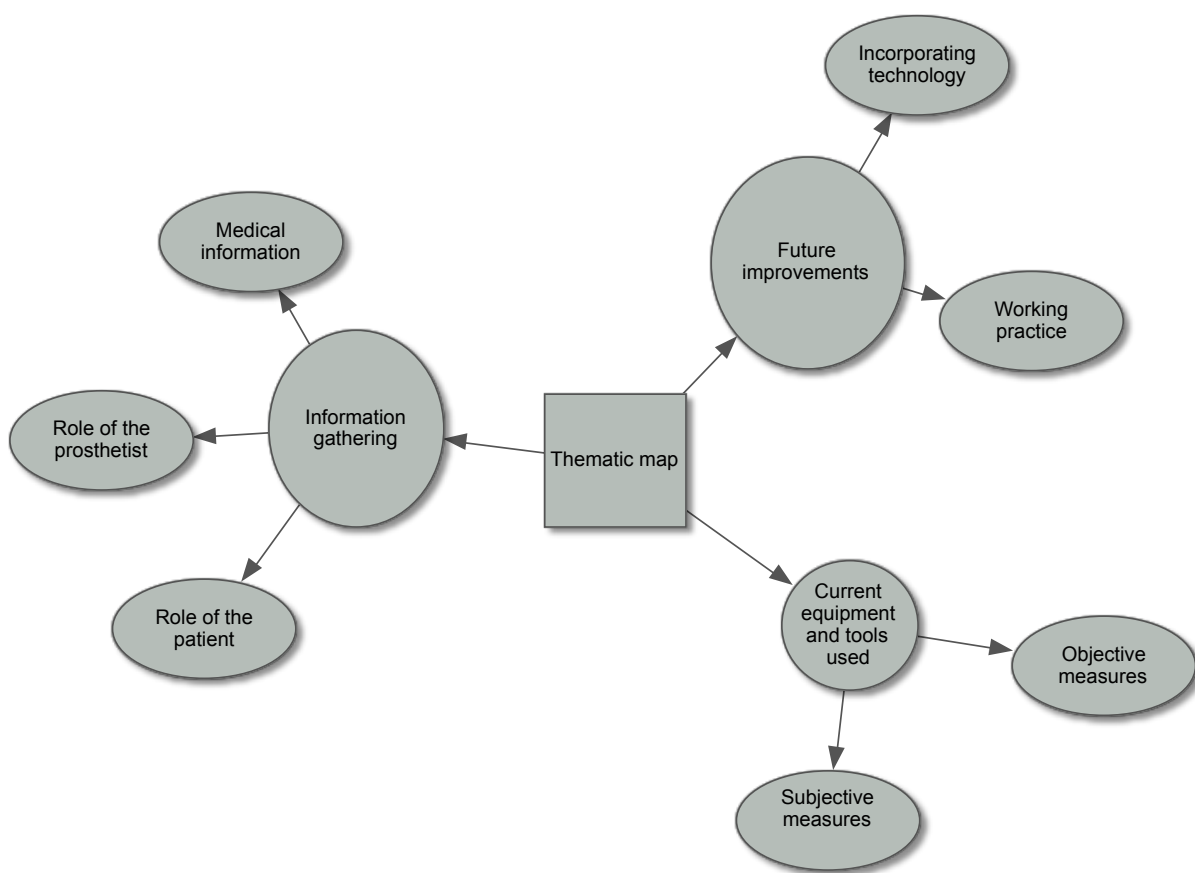


Figure 8: Thematic map of the transfer of knowledge in the assessment of prosthesis

### 5.5 Findings

#### 5.5.1 Response rate

23 MP's and 12 AEP's were contacted during the recruitment stage of this study. A response rate of 26.1% (n = 6) was achieved for MP's: of which 4 participants came from 2 services. A response

rate of 33.3% (n = 4) was achieved for AEP's. All AEP's came from the NAES but were based at various locations throughout England.

### **5.5.2 Participant demographics**

The sample comprised of 6 (60%) females and 4 (40%) males. The majority of the sample was between the ages of 40 and 50 (n = 6, 60%), though 20% (n = 2) were aged 39 years and younger, and 20% (n = 2) were aged 51 years or older. Out of the 4 artificial eye prosthetists, 75% (n = 3) were MP's were female and 50% (n = 3) were male with a mean age of 47.5 years.

Length of service ranged from 13 to 40 years (M = 18 years) with the majority (n = 6, 60%) having worked in their field for 20 years plus. The mean length of service of MP's was 23 years compared to 14.25 years for AEP's.

Regarding education, 10% (n = 1) had a Further Education qualification, 80% (n = 8) had a Higher Education qualification: 50% (n = 5) of which was related to their field and 1 participant (10%) did not answer this question. MP's had the following qualifications: undergraduate degree in Dental Technology (33.3%, n = 2); ONC/HNC/ HND in Maxillofacial Prosthetics & Trauma/Maxillofacial Prosthetics and Orthodontics (33.3%, n = 2) and a City and Guilds qualification (33.4%, n = 1). Artificial eye prosthetists qualifications consisted of A-Levels (25%, n = 1); unrelated BSc (25%, n = 1), and a HND in Art and Illustration (25%, n = 1).

All AEP's and MP's were equivalent in terms of employment status (full time) and ethnicity (White British). See Table 20 for a summary of interviewees' demographics

### **5.5.3 Findings of the thematic analysis**

During the initial stages (stage 1 – 3) of data analysis, commonalities between MP's and AEP's responses became apparent. Consequently, themes were developed, defined, and named that encapsulated both population groups. Differences and similarities between the two population groups are displayed within each theme and reported in the discussion.

The findings of this component of the study centre on conducting an assessment for a prosthetic fitting. An assessment identifies the *need*, *demand* and *supply* of the service provided. *Needs* in healthcare refer to the capacity to benefit; *demand* centres around what the patient needs, and, *supply* is the health care provided. With all three elements overlapping, their relationship is important to consider when assessing healthcare needs. Furthermore, consideration needs to be given to the interpretation of healthcare needs. Whereas healthcare professionals will consider needs in terms of healthcare services they can supply, patients' needs focus on the social and environmental determinants of health in addition to their medical needs.

Table 20: Interviewees' demographics

Job title		Length of Service	Gender	Ethnicity	Qualification	Age	Employment
Artificial Prosthetist	Eye	13 years	Male	White British	3 A-Levels	35	Employed Full time
Artificial Prosthetist	Eye	24 years	Female	White British		47	Employed Full time
Maxillofacial Prosthetist		17 years	Male	White British	BSc Dental Technology; PG(Cert)Maxillofacial prosthetics	45	Employed Full time
Maxillofacial Prosthetist		20 years	Male	White British	HND Maxillofacial prosthetics & Trauma	47	Employed Full time
Maxillofacial Prosthetist		21 years	Female	White British	BSc(Hons) Dental Technology	44	Employed Full time
Maxillofacial Prosthetist		40 years	Male	White British	City & Guilds	56	Employed Full time
Artificial Prosthetist	Eye	4.7 years	Female	White British	BSc Biomedical Science	27	Employed Full time.
Artificial Prosthetist	Eye	15 years	Female	White British	HND in Illustration (Art & Design)	53	Employed Full time
Maxillofacial Prosthetist		20	Female	White British	OND, HNC, HND, Maxillofacial prosthetics & orthodontics	43	Employed full time
Maxillofacial Prosthetist		20 years	Female	White British	HNC in Maxillofacial & orthodontics; Advanced City & Guild in Prosthodontics; BTEC in Dental Technology	50	Employed Full time

These three elements and their individual interpretation are embedded within the three themes generated from the analysis. These are: (1) information gathering; (2) current equipment and tools used, and (3) future improvements. Each of these themes is described in detail with illustrative quotes and extracts from the data supporting the findings. The population group that stated each quote and extract will be reported to enable comparisons between the two. Further interpretation and explanation of the findings will be reported in the Discussion section of this chapter.

### **5.5.3.1 Theme 1: Information gathering**

Assessments need to be appropriate and proportionate. They need to gather information of the health needs of the patient they are treating, including medical needs and needs relating to the wider determinants of health. By taking into account the patient's wishes, preferences, and desired outcomes, a healthy psychological wellbeing of the patient will be attained increasing the success of treatment and enabling the patient to achieve their goals in their day to day life. At the same time, accurate and detailed medical notes of the patient is important to produce an aesthetically pleasing and functional prosthetic. Thus, this theme encapsulates both the medical information and the role of the patient.

#### **5.5.3.1.1 Sub-theme 1: Medical information**

The need for a prosthetic varies from patient to patient: the most common being congenital and acquired disease(s), trauma and injury. The prosthetic assessment comes at the end of an often long and individualised medical journey. Medical journeys involve consultations and treatments: examples of which include amputation, chemotherapy, and enucleation. Thus, detailed and accurate information about the patient's medical history is essential for an aesthetically pleasing and functional outcome. Participants highlighted the importance of receiving a detailed referral of the patient in terms of previous treatment and how far post surgery they are.

“... referral letter of the clinician telling me exactly what has been done ... (this) will give us a rough idea of ... what we might have to do, how far post surgery they are, whether we got to wait for a couple of months to let the wound heal.”  
(Matt, MP)

Other information reported as being essential to know prior to the initial assessment include reason for loss, the patient's surgical history and whether the patient has any allergies or inflammations. This information can help the prosthetist make reasonable adjustments.



“Need to know the reason for, either the loss of the eye or the reason the eye is a shrunken globe.” (Isobel, AEP)

“... (in the case of implant retained prosthesis) ... whether they have had radiotherapy ... as you wouldn't suggest this to the patient if you knew their bone quality was too weak to take it.” (Matt, MP)

“Any allergies ... so we can determine the materials we would use.” (Owen, MP)

Participants reported that they do not solely rely on the information contained in the referral letter. Reasons for this include, a lack of, or missing information, the time period from referral to assessment resulting in changes to the deficit site as well as changes to need/suitability of patients for a prosthetic.

“... see whether patient would be suitable for a prosthesis because sometimes it requires some manual dexterity to actually get the prosthesis to stay on so you got to evaluate whether the patient is suitable for a prosthesis.” (Matt, MP)

Howard and Jacob reported the importance of evaluating the socket post-operatively to make sure the deficit site is healing correctly and that there are no inflammations or infections.

“... evaluating how things are post-operatively ... (looking for) socket condition so whether it's healed ... whether there is an implant ... and the implant type.” (Howard, AEP)

“... have a look and see if anything has become redder, sore or moved out of position because you do need to check if there have been any flows of tissue in unexpected areas.” (Jacob, AEP)

Participants reported the importance of patient feedback at all stages of their treatment cycle. Practicing patient centred care brings many benefits such as enabling the patient to be heard and their ideas, concerns and expectations addressed. Gathering information from the patient regarding their medical history was seen as the first step in conducting an assessment.

“It’s important to get the information you need from the patients end so the patient is happy and aware of what you are doing all the way through the process.” (Howard, AEP)

Participants reported their work is more than gathering medical information: it centres around understanding the patients journey. Specific parts of the patient’s narrative that was expressed as important by the participants is whether they have experienced any problems with the prosthetic in terms of comfort and the way it looks. Understanding the patients experience and their satisfaction with the deficit site and/or the prosthetic, allows the prosthetist to create an individualised treatment plan to achieve the best outcome.

“Is the patient experiencing any discomfort there? Is there anything we should know about?” (Lauren, MP)

“Main concern would be the patients’ discomfort. One or two patients may have no discomfort and the next patient may find it very uncomfortable but it’s not apparent why.” (Howard, AEP)

“... (asking the patient) how comfortable (the prosthesis) has been; whether they’ve had any problems with it; how it looks in the socket ... You are assessing from that what you’ll going to be doing in terms of improvement of the bespoke mould.” (Owen, MP)

Alongside understanding the patient’s experience of the treatment leading up to and including the prosthesis, further information about the patient’s overall health and their demographic details is required. Participants stated that this information often dictates how the assessment is undertaken and how and who the information is directed to. Specific information about the patient that was

reported as being required by the participant includes the patient's age and whether they have any mental health or learning difficulties.

“For a very young child you would have to associate it maybe to the parent. If the child was a bit older I would mainly speak to the child. Get the child to answer and ask me questions to start with and run through what we are going to do and how we are going to do.” (Isobel, AEP)

“If they are elderly and they bring people in, it's usually because they don't have a good memory themselves ... so it is important to take notes on the age of the patient and who they come with.” (Kim, AEP)

“If they got dementia: Alzheimer's, do they know they've had an eye removed.” (Isobel, AEP)

“... (The information needed): Any mental health problems.” (Howard, AEP)

Following information gathering, participants reported involving the patient in the creation of the prosthetic: specifically in testing it out to see whether it is correctly made and fitted.

“... get the patient to give a fair range of tests to the shapes so not just to look at the aesthetically but also look left right, up, down, see how it sits.” (Howard, AEP)

Placing the patient at the centre of treatment makes them feel involved in the process, allows them to take more responsibility, can help manage expectations as well as increase the patient's confidence in the service and the prosthetist.

### **5.5.3.1.2 Sub-theme 2: Role of the patient**

A key element in conducting an assessment is involving and understanding the patient's role throughout the treatment process. The previous sub-theme highlighted the patient's experience as playing a key role in conducting the assessment of a prosthetic fitting. Listening to the patient's narrative is a key requirement of the prosthetist role as it improves the outcome of the prosthetic as well as the patient's emotional wellbeing.

A good therapeutic relationship has been found to improve patient satisfaction and professional fulfilment as well as saving time and increasing compliance. Participants from this study reported that developing a therapeutic relationship is at the heart of the work they do.

“One of the most important things in our job is building the relationship with the patient because if you don't have a good relationship with the patient that's 50% of the prosthesis that won't work as the patient won't accept them.” (Nigel, MP)

“...to get to know the patient before you start any treatment for them ... get to know me, to understand me.” (Isobel, AEP)

Therapeutic relationships are based on developing rapport. Rapport is a state of amicable relationships between people: in this case the patient and the healthcare professional. It relates to how people connect, behave, and engage with each other. Rapport is seen as a starting point for building trust. Participants reported that trust is enhanced by understanding each patient's journey both in terms of their medical and personal background.

“... that we build that rapport with them so they completely trust us. We have to demonstrate that they can trust us.” (Lauren, MP)

“We are very aware of where they have come from, their past, their journey they got to when they came to us. So, I think all the prosthetists I know who do it are very understanding, very empathetic.” (Kim, AEP)

MPs reported that the prosthetist-patient relationship is enhanced the earlier a patient is seen and through time flexibility. Enabling trust, MP's reported that the patient's confidence in the service and the work conducted begins to be built.

“If we know we are seeing a patient we don't book another appointment afterwards. There is no time frame that we have got to keep. We want the patient to feel completely at ease and happy, that we have basically dedicated all our time to make sure what they got is right.” (Matt, MP)

“Introducing the patient at that early stage so at least they know us ... So at least when they do see us in clinic then we would be familiar with them already so we can build confidence with the patient.” (Nigel, MP)

Whilst trust and confidence building was also emphasised by AEP's to enable smooth running of the prosthetic service, the importance of seeing a patient as early as possible was not highlighted. This is due to the difference in working practice. Whereas MP's will see patient's pre-surgery (as sometimes an impression needs to be made of the facial anatomy that will be removed), AEP's only see patients 6-weeks post-operatively once the eye socket has had time to heal.

“We would normally see the patient before they have the surgery (to) explain to them what prosthesis is. We would give them ways to how it would be retained and ... (tell them that it is) going to be a temporary prosthesis.” (Owen, MP)

#### **5.5.3.1.3 Sub-theme 3: Role of the prosthetist**

Prosthetists assess and treat the physical and functional limitations of people resulting from illnesses and disabilities. As well as being dependent on the prosthetist skills, the prosthetic outcome relies heavily on the patient's capacity to follow through with medical recommendations, self-manage, and adopt preventive health behaviours. With patient compliance relying heavily on the prosthetist-patient relationship, the role of the prosthetist is more than producing an acceptable outcome.

At the centre of this subtheme is the prosthetist. Surrounding the prosthetist in terms of their job role are the following: (1) impact of the process upon the patient, (2) managing expectations, and (3) helping the patient make an informed decision. This sub-theme focuses on the *need* of healthcare which has the potential to improve services through development.

Through developing rapport with the prosthetist where confidence and trust is enhanced, the patient is better placed to make an informed decision on their treatment. With the treatment plan being decided by the patient, participants reported the importance of making sure that the patient has the correct information. Participants reported that patients may come in with the wrong information, whether from other healthcare professionals or based on their own research and/or opinions.

“... (knowing) what the patient has been told before. Sometimes they are given strange information before they see us.” (Owen, MP)

“...in their mind (they) have an idea of what it looks like ... The patient will often say my nose wasn't that big, even though you know it was ... it's just perception.” (Lauren, MP)

With the potential for patient expectations to strongly affect postoperative satisfaction, participants reported the need to manage their expectations. This is particularly the case when patients do not know exactly what the treatment process involves.

“... patients' expectations can be quite high ... they don't know exactly what they are going to get.” (Matt, MP)

Making the patient aware of what is possible in relation to their own situation as well as the limitations of prosthesis in general can help manage expectations.

“... you can't always do exactly what they want, for whatever reason, or some reason it is not as good as you would like it to be. It is as good as you can get it

to be for that particular patient for it to work and do what you want it to do.”  
(Lauren, MP)

“... the limitations of the prosthesis. I think that’s the most important because often the patient can come with inflated expectations which can lead to problems later on when you actually fit the prosthesis. If it’s not what they expected, so I think it’s important that they know those limitations to begin with.” (Howard, AEP)

Through managing expectations, participants reported that the patient can make an informed decision on their treatment plan. Informed decision making takes place once a patient is aware of their choices, the potential outcomes of these choices and when their personal views have been considered both by themselves and the healthcare team. By giving the patient a realistic opinion on what is and is not achievable, the patient is able to make a decision that is right for them at that time.

“You can give the patient a realistic opinion of what is available and what is achievable.” (Owen, MP)

“It’s very important you don’t force prosthetics onto people because, particularly with paediatric or younger patients, you put them off for life.” (Nigel, MP)

Participants stated that the patient needs to be given time to think about their options and have the opportunity to ask questions as and when they occur.

“That the patient can talk about those issues ... and they will think, oh that’s not for me, which is fine. It’s their decision: they need to make an informed decision.” (Lauren, MP)

“Let them think about it and to get back to us before committing to anything so they have the time to take it on. We may give them information leaflets and ask any questions before they agree.” (Nigel, MP)

Alongside giving the patient time and opportunities to ask questions, offering reassurance regarding the process and outcome helps manage potential distress. Reassurance occurs within the interaction between the healthcare professional and the patient. The healthcare professional has the intention to reduce the worry as experienced by the patient.

“...to reassure them. To give them a chance to ask any questions from the fitting point of view rather than the Doctor, medical side of views.” (Isobel, AEP)

Kim highlighted that in order to offer reassurance, they try to place themselves in their patient's shoes. By understanding the patient's worry the prosthetist is able to directly respond to these concerns without the patient having to explicitly state them. Kim reported that this enhances their relationship with the patient.

“... when someone comes along to the assessment process, I think their worries are: what is going to happen; what on earth will this look like when it's finished; do they know what they are doing ... So, I think that if we can cover all those bases with the patient so they are confident with us and we've got their trust.” (Kim, AEP)

Reducing patient's distress at a time when they are (more) vulnerable is a challenge for all healthcare professionals. Behaviour management strategies have often been used to manage distress. Participants reported using this technique (specifically play) for young patients.

“Talking to them, playing with them. (in the case of children) we use play ... It's all age relevant. If they are older, then sometimes I might put them in touch with a child that has had a similar sort of thing or someone if they need to talk to someone about it.” (Isobel, AEP)



As a child's primary caregiver, participants reported that alongside assessing and managing the distress experienced by the patient, they need to be aware of how the patient's parents are coping. This is because the coping mechanisms of the parent directly affects how the child will adjust to having an artificial eye. This, in conjunction with a child's illness requiring parental participation and supervision in managing the daily treatment and after-care demands, means there is a critical need for a careful assessment of the parent(s) wellbeing.

“I need to find out how (the parents) are dealing with it, because if they are not doing very well, the child's not going to be a very easy patient.” (Isobel, AEP)

Assessing the needs of both the patient and their parents allows the prosthetist to tailor treatment to the needs of that particular family. This was reported by participants in terms of extra support or making referrals to specialist services such as counselling.

“... whether they are just going to take it on board or if they are going to need some extra help at the beginning to get used to it.” (Isobel, AEP)

“One patient we brought in a mental health team to help them come to terms with the loss of their nose.” (Liam, MP)

#### **5.5.3.1.4 Theme summary**

The findings presented in theme one highlighted the information needed in conducting an assessment for a prosthetic fitting. Three elements that addressed the *needs* and *demands* of healthcare services were evidenced. This includes the patient's medical information, the role of the patient and the role of the prosthetist. Whilst a detailed referral letter (reason for loss, need for a prosthetic and the surgical procedures performed regarding the deficit site) was highlighted as important, it was not seen as vital. Both AEP's and MP's stated that at every assessment they evaluate the deficit site to determine the healing process and to assess suitability.

Contributing to the success of the process is both the prosthetist and patient. Listening to the patient's narrative of their experiences, needs and wants enables the prosthetist to manage patient's expectations, make the correct adjustments necessary, and reduce any potential negative impact associated to the process and having a prosthetic. Working from a patient-centred approach allows a therapeutic relationship between the prosthetist and patient to be built, where rapport, a mutual understanding, trust and confidence is developed.

#### **5.5.3.2 Theme 2: Current equipment and tools used**

The third theme presents findings related to the tools and equipment used by the participants in the assessment process and their perceptions and experience of them. The use of tools by healthcare professionals transforms many parts of clinical practice. This includes early diagnosis, treatment/product outcome, patient satisfaction and healthcare professionals working practice. When tools become common place, rapid growth occurs, advancing service delivery. Responsible for providing health services for patients, families and communities, health service delivery systems that are of high quality are required. With the aim of both healthcare and health technology being to improve the health of specified populations, technological incorporation is inevitable. Understanding how a service is conducted is the first step in addressing the needs highlighted.

As a custom-made process, participants reported the use of both subjective and objective measures in the assessment process of a prosthetic. Objective measures are primarily tools that involve an impartial measurement where no bias or prejudices exist. Conversely, subjective measures are based on the users/observer's personal judgement of what is needed and/or how the skill is performed. Whilst both are seen as imperative to their work, participants placed more emphasis on the subjective tools.

“I think in the majority of cases you are probably using your subjective visual assessment, probably more than the tools which is probably why we've got hardly any tools that do that kind of work.” (Jacob, AEP)

Participants reported that the use of objective (tools) or subjective (personal judgement) measures is decided on various factors. This includes their own experience and on the patient and their presentation. Early career healthcare professionals often display a high level of reliance on

objective tools to help with decision making and to ensure that they achieve the best outcome. For the same reasons, participants reported using objective tools in more complex cases.

“When I was trained ... I basically had a lot of pre-defined shapes and kind of working through those and seeing what ones were the best and fitting my half sphere to that. Actually after a few years and help from colleagues I switched to kind of doing it by eye and seeing that probably the shape is going to be like this: this is probably the shape I need to make it to. So, you kind of switch off that reliance on samples and pre-edited shapes. You can visualise the end shape because you have a better understanding of what it is going to be like, what the socket needs to fit it in the right way.” (Howard, AEP)

“How you visualise it and how it looks and probably the subjective element is mainly what you would go by. But then when things are more difficult, the more complex the case, the more use you can make of the measurements, the tools and the additional techniques.” (Kim, AEP)

#### **5.5.3.2.1 Sub-theme 1: Subjective measures**

Subjective measures in this context can be described as prosthetists reliance on their own knowledge and skill-set which has been gained through experience. Defined as dependence on, or trust in someone or something, participants have shown a reliance and preference for using their own judgement in creating, manufacturing and fitting prosthetics. With the goal of healthcare being to achieve the highest quality of service for their patients, participants emphasised the importance of their experience in attaining this.

“The more experience you have the better at seeing not only the socket as it is but you can visualise what the end results needs to be more effectively.” (Howard, AEP)

“I just have a look at the socket, get an idea how I’m going to make it and from experience whether you are going to mould it or whether you are going to make a temporary eye or whether you are going to have to go to theatre.” (Isobel, AEP)

Rationale for this preference was given by Matt who stated that the custom made-nature of prosthetics means that each patient's treatment needs to be individualised. With continual adjustments and tweaks to the prosthetic needed, a subjective approach is best suited.

“You are constantly re-designing the prosthesis and that only comes with experience - you know of what will work and what won't work. That comes with, you had failures, and you had to get around it by deciding something else.”  
(Matt, MP)

Whilst working subjectively can help in foreseeing potential problems, there is a chance that something may be missed, particularly when a prosthetist is lone working.

“... it's somewhat more proactive when you think you know what's it going to be like anyway which doesn't mean that you are always right but it gives you a good start.” (Howard, AEP)

“... it's easy to potentially miss something just because you look the way you look. You don't necessarily spot something, particularly if it's something you have not seen before.” (Jacob, AEP)

#### **5.5.3.2.2 Sub-theme 2: Objective measures**

Subjective measures often invite patient feedback, thus is seen as patient focused. In comparison, objective measures focus on the healthcare professional that may be seen as a product of the service they work for. Whereas the subjective focuses on satisfaction, the objective focuses on the evidence-base. High levels of satisfaction are attained when needs are met. Both patient and healthcare professionals needs are met when the best treatment and care is given and received. This often comes from utilising the latest technological advancements. Aligning both subjective and objective measures can increase productivity and satisfaction for both the patient and the healthcare professional. Having reviewed the subjective measures in sub-theme 1, sub-theme 2 will address the objective measures, specifically the tools and equipment participants use in

assessing patients for prosthesis. Howard stated that objective tools can be a helpful guide in the assessment stage of creating a prosthetic as it provides a more concrete measurement.

“It’s helpful to have something objective to measure against ... it gives you something a little more solid to work upon that differs from your own perspective.” (Howard, AEP)

In the context of this study, objective tools refer to medical equipment that have been designed to aid in the monitoring and treatment of those needing a prosthetic. Alongside increased service and product outcome satisfaction, the tools used improve quality of care, enhances time management, creates standardisation and increases efficiency. The use of tools varied between participants with only half of MPs having access to technologically advanced tools and AEP having minimal access to any form of objective measures. The tools available included diagnostic imaging (CT, MRI scans) and 3D printers.

“I’ve made prosthesis of a 3D model ear where we have taken the actual scans of the CT and printed a model and have used that to make the prosthesis.” (Matt, MP)

Diagnostic imaging creates visual representations of the body for clinical analysis and medical intervention. The images produced inform healthcare professionals about the anatomic organisation and functional working of the inner structures of a patient’s body. This information allows healthcare professionals to make decisions on type of treatment required and when this is needed. In the context of this study, diagnostic tools provide prosthetists with information regarding whether the deficit site has healed, any scarring, inflammation or evidence of further disease, as well as the size and shape of the prosthesis that needs to be made. The use of diagnostic imaging tools were seen as being dependent on the prosthetic being made.

“... if they are going to be implant retained we would use a CT scan and get a Stereolithography model done.” (Nigel, MP)

“(diagnostic imaging) is essential if the assessment suggests you will be using implants or you need to look at bone quality, bone depth...” (Nigel, MP)

In reviewing the effectiveness of diagnostic tools used, MP’s highlighted their potential and viability. The need to capture the contours of the deficit site increases the difficulty in enabling an accurate fit. However, imaging tools were reported as not being able to achieve accuracy. Matt reported that this allows the prosthetic to be printed, eliminating the need for an impression to be taken. Yet, being expensive, these objective tools were not seen as viable.

“... when the scans were taken, obviously, the patient was laying down so the tissues wouldn’t have been in the position where they would be naturally. But I was really surprised at how accurate it was.” (Matt, MP)

“... we can CT scan the patient pre-op then what we would have to do is to scan them again post-op once the healing process has taken place. That would allow me to take the images and make the 3D printed model from it. So that is another way of actually getting the model made.” (Liam, MP)

“... it’s an expensive way of ... getting hold of the images we need.” (Matt, MP)

Medical photographs were identified by MP’s as being another objective measure they use in their work. Medical photographs are used to accurately and objectively record images of the process of medical procedures. Participants reported using the service pre-surgery to enable them to create an accurate prosthetic post-surgery.

“Photographs are very important ... if there are photos available pre-op, that would make it a bit quicker.” (Lauren, MP)

“... we would use photographs of the patient before the surgery and try to use that to be able to create what they had pre-surgery.” (Paul, MP)

Although AEP's did not have access to medical photographers, in complex cases, the prosthetists took a photograph of a patient's socket and artificial eye using an iPad.

“... you had almost a measurable subjective opinion of how things were working and looking and that was quite a useful tool because otherwise you are having to try and hold in your mind how the older one looked and that can be quite difficult to do if there is not a massive difference. So, as an extra tool that was useful to have a record of what is going on because historically we haven't had that, we've just had written notes which are obviously useful, but there are some things which are easy to express photographically.” (Howard, AEP)

AEP's reported having minimal access to technologically advanced tools. Potential reasons for this include reliance on subjective measures or, as a smaller healthcare field, a reduced and/or a lack of funding. AEP's reported using tools such as an exophthalmometer, a pen torch, a conformer, the existing prosthesis and tissue tests. These tools were mainly used in more complex cases.

“...an exophthalmometer ... it's useful when you know you've got a major deficit so you can see an eye and it looks like it's fairly sunken then you measure it to find that actually you need a prosthesis that is maybe two three centimeters deep to get the eye at the same prominence.” (Howard, AEP)

“I have a pen torch that helps me look at the socket ... Sometimes I use a conformer, or I might need a different shape conformer ... sometimes if the patient has a globe sometimes I will run a tissue test.” (Isobel, AEP)

“The patients existing prosthesis is one of the best guides ... even if its rubbish it can be helpful to see how it is measuring up and what sort of things you need to do next.” (Howard, AEP)

Participants who use objective measures reported few complications with the equipment and tools. Reasons for this include, identifying potential difficulties at an earlier stage, measurements being taken further along the treatment process and well maintained equipment.

“I have had situations where things could have been problems. It was only because the surgeon informed me of this conjunctivitis scaring up the side to the top lid of a Phthisic eye that I knew to look for them. If I hadn’t, because they are right at the back I might not off looked far enough because they are quite unusual.” (Howard, AEP)

“if the equipment is maintained properly then we shouldn’t have any problems. If any of the equipment is not fit for purpose then we wouldn’t use it.” (Owen, MP)

In spite of the benefits of using objective measures, participants stated that they will rely more on their visual judgement. This is because whilst the tools can help in the process, they are not always effective. As reported by Owen, technology can only help up to a certain point.

“You can use a little tool which can make the pupil distance from the nose exactly the same as the existing eye ... Sometimes when you use that particular measure, you can see visually that it doesn’t look right so you adjust it.” (Isobel, AEP)

“Some of the tools we got do not quite pre-determine the shape ... even the big impression trays are either too tall, or not big enough even at maximum volume.” (Howard, AEP)

“I think the technology can only go so far because you know when you got a patient sitting there, for an ear for example, and you ask them to open and close their mouth. The technology can’t tell you how much the soft tissue is going to move or how soft the soft tissue is, or if there is scar tissue, how hard that is. So, it can only help so far.” (Owen, MP)



### **5.5.3.2.3 Theme summary**

Theme two reflects upon how prosthetists conduct the assessment stage. Two elements that addressed the *supply* of healthcare services were evidenced. These were the subjective (skill-set) and objective (tools) measures prosthetists utilise in their work. Given the custom-made nature of prosthesis, all prosthetists felt that their knowledge, experience and expertise are just as, if not more, important as the tools used. With the purpose of supply being to meet the demands and needs of patients, a mix of both subjective (patient focused) and objective (healthcare professional focused) is required.

Having highlighted the potential of objective tools such as diagnostic imaging being able to produce accurate outcomes, their viability in both maxillofacial and artificial eye services are worthy of exploration. Catering for a larger population group, MP's were found to have more access to technologically advanced tools. Consequently, participants from this group were more open to their work heading down a digital path.

### **5.5.3.3 Theme 3: Future improvements**

The focus on meeting the *needs* and *demands* of healthcare patients has led to innovations in their treatment regime and aftercare needs (*supply*). Combined with increased accessibility, the patient-centred approach has seen technological advancements being applied to healthcare sectors (as evidenced by MP's in theme 2). Yet, a lack of resources and investment in some healthcare fields, for example artificial eye services, means that its application and/or continual development is not promised. With an evolutionary element where processes are continually revised and re-developed until the end product results in excellent patient outcome and satisfaction, makes understanding technological capabilities as they currently stand of upmost importance. Thus, the third and final theme from this study reflects upon participant's perspectives of incorporating technology into the assessment process.

#### **5.5.3.3.1 Sub-theme 1: Incorporation of technology**

Due to factors such as fragmentation, access problems, changing social and disease type demographics and unsustainable costs, healthcare services are going through a phase of transformation, of which technology plays a crucial role. In keeping with the findings from theme 2, the potential of technology was more evident from the perspective of MP's whom envisioned their work as taking more of a digital route.

“I see the biggest reform being digitally scanning and 3D modelling.”  
(Nigel, MP)

In terms of incorporating medical technology currently on the market, both AEP’s and MP’s saw potential in using 3D hand-held scanners. 3D scanners generate high accuracy data, which can then be used to construct digital 3D models through using a 3D printer.

“I think something along the lines of 3D hand-held scanners.” (Matt, MP)

“I’ve been to take impressions of patient who’ve had skin grafts and where I’ve got to hold the skin graft in place. Now if we have a hand-held scanner we could ‘ve scanned the patient, printed the model of, so there is no actual physical impression involved.” (Liam, MP)

Whilst technology initially increases healthcare costs, overtime costs decrease and efficiency and quality of care increases. With the purpose of incorporating technology into healthcare to improve service delivery and patient satisfaction, technology needs to be able to meet the needs and demands of both the healthcare professional and the patients. Whilst both AEP’s and MP’s reported the potential of 3D hand-held scanners, accuracy and costs were given as reasons for it not currently being viable.

“A scan of the back of the socket (may help) but you wouldn’t know where the lids need to be to give you roughly the shapes.” (Howard, AEP)

“The accuracy of some of these scanners are not that great ... they can scan to 1 millimetre but that is still quite large ... For any type of prosthesis, you need an accurate model. But the scanners are getting better and better every year and the prices will drop.” (Matt, MP)

Further tools identified by prosthetists as having the ability to contribute and improve their working practice include intra-oral cameras (where images are taken of an outside surface from various angles) and 3D printers (a process used to create 3D objects). The concept behind the use of these tools centre around manipulating fine detail from which a prosthetic mould can be printed.

“The intra-oral camera that gets used by dentists. That could be useful, something that you can then manipulate.” (Lauren, MP)

“... we have a 3D printer (which) is just the beginning of what we are hoping will become a digital treatment.” (Matt, MP)

The need for technology to capture fine detail was emphasised by the majority of participants. Whilst used by some MP's (as highlighted in theme 2), AEP's reported benefits of using medical photographs such as colour matching. Furthermore, it was suggested that a tool needs to be created which can give direct dimensions of the elements within the body part that is removed. This would make the prosthetic more realistic and as a by-product improve the outcome and increase satisfaction.

“A really good photographic system would be helpful ... I think that the disadvantage for us is we don't have that kind of system, quality system in place ... I do think it's quite good if you've get a very good close up of the iris and the rest of the eye and for the painter to be able to see in an overall way how the eye looks and the distance in a normal conversational distance.” (Howard, AEP)

“If we had a tool that would give the exact dimension and diameter of an iris would be useful. I look at one of those smart phones when you focus in on something and it has a little ring when it appears. I think there must be something you can develop along those lines. Changes size to find the exact diameter of an iris – that would be really useful.” (Owen, MP)

### 5.5.3.3.2 Sub-theme 2: Working practice

Working practice is central to the success of a service. It incorporates the method, process, and guidelines healthcare professionals follow. This ensures its success both in terms of outcomes and patient satisfaction whilst meeting set targets of the individual service and organisation as a whole. This sub-theme reflects upon what participants feel are essential for a successful service, addressing both patient and healthcare professional's needs and demands.

With the prosthetic process being subjective in nature and individualised, it was suggested that having a verbal conversation with the patient about the surgical removal and prosthetic fitting and developing a way to measure a patient's perspective of the process from pre-to post treatment would be beneficial. This would also provide further information on satisfaction levels and suggestions for potential improvements.

“I think you need to have a verbal conversation, but to tick boxes, they won't know.” (Liam, MP)

“... (it) would be nice to measure the patient's expectations in a quantifiable way ... if you could have a questionnaire that would assess what they would expect from it and a similar thing at the end. If you just ask them it's difficult to compare to others and to compare to the outcome and to quantify that.” (Nigel, MP)

Identified as being crucial to the success of the process, participants reported the need to work successfully within a MDT. Effective MDT working is seen as the key to high quality care. It involves a group of workers from different disciplines providing specific services to the patient. Care, compassion, competence, communication courage and commitment are at the heart of MDT working.

Within this sub-theme disparities were found between and within both population groups regarding MDT working. Those whom worked within an MDT found it a positive experience for themselves and their patients. Whereas these participants cited enhanced communication, those whom reported not working within an MDT stated there is lack of communication, time and standard protocols and agendas.

“The communication is so much easier, so much better. Sometimes I can actually go grab the consultant in the corridor or the consultant can ask me to come down to their clinic and I work with them and see the patient together.” (Kim, AEP)

“... patients like the fact that we are a team. They are not going off to one, to see one and then having to wait for a referral letter having to go to another. They know we see them together sometimes or they know that I will communicate with the consultants.” (Isobel, AEP)

“... the way sometimes the MDT clinic operates, but that’s basic communication issues like most hospitals these days. Everything is a rush, get everything done. There are so many patients to see and it’s really, you need to find and make time to do it properly.” (Nigel, MP)

Another key factor highlighted as impacting the effectiveness and efficiency of MDT working was access. This includes location and use of centralised computer systems. This was particularly apparent in AEP’s whom often work in isolated clinics, thus do not have access to other members of the MDT including other AEP’s. This in conjunction with a centralised system containing contact details of members of the MDT was reported as making their work easier.

“I think we need to be where the ophthalmic department is ... then the whole process would be so much easier.” (Jacob, AEP)

“It would help if there was something where it is not strictly just left up to the prosthetists. I think it would be better to have something within the management system where it is part and parcel of starting a new job like that to make sure you are known.” (Isobel, AEP)

MDT working requires prescription to a set of guidelines and protocols. Although clinical guidelines can improve the quality of clinical decisions by alerting healthcare professionals to

procedures that are unsupported where attention is drawn to ineffective and wasteful practices, the individualised nature of prosthetic work requires flexibility with an element of trial and error being accounted for. Thus, following a prescribed procedure/guideline in creating a prosthetic was seen by participants as not being viable.

“The care pathway, could be more time consuming. Could be put in care pathways we don’t need. That’s where we need that little bit of flexibility ... Every situation needs to be adapted to the individual’s needs.” (Owen, MP)

“I think it is potentially dangerous, because, I think that people who may wish to embark in this type of work need to be able to undertake the work in the way it is done now not through any step by step guide. The problem with a step by step guide is if one step doesn’t work, there isn’t a plan B and if people work in that way, that’s dangerous.” (Nigel, MP)

Participants reported that while there are no step by step guide in making a prosthetic, certain procedures regarding health and safety are followed. The flexibility this provides in their working practice, allows in-service training programs. Whilst participants reported that their training programs are effective, it was acknowledged that it does not work for other healthcare professionals. This has the potential to impact MDT working in terms of treatment progression, patient and healthcare support as well as communication.

“... there’s not a lot of prescriptive procedures that we use in facial and body prosthetics. There is not a step by step guide. There are certain procedures that we adopt and all tend to follow and there are obvious things you have like patient safety and cross infection and all that sort of things. But in terms of the actual prosthetics. There is very little sort of step by step guide to actually making them.” (Owen, MP)

“I think it is effective ... You are taught to recognise whether the eye is too heavy or whether it is creating extra pressure on the lids or whether it is you know, the way the lid fits is restricting movement. Trainees get to see a number of different prosthetists so they get to work with different people who may have slightly

different approaches or different intuition to how to best manage something like the shape of the lids or the way the socket.” (Howard, AEP)

“... each one works in their own way: sometimes that way is not ideal from our perspective. I can appreciate that it is sometimes from their perspective, but it doesn't necessarily mean that it works for us.” (Kim, AEP)

#### **5.5.3.3.3 Theme summary**

The third and final theme addresses the potential of technological development in prosthetic work, specifically the maxillofacial and artificial eye sectors. This theme encapsulates the three elements vital to healthcare. This is where the *supply* meets the *needs* and *demands* of both the patient and the healthcare professional. Through this, the aim of creating an aesthetically pleasing and functional product is met. This theme has highlighted that supply needs to involve both the use of technology (if capabilities allow), but also to work multi-disciplinary. Participants reported that whilst technology holds potential (for example, 3D hand-held scanners), a lack of accuracy does not make it viable. Furthermore, a conducive working environment was highlighted as ensuring success both in terms of outcomes and patient satisfaction. Working within an MDT with access to healthcare professionals, and information systems was found to increase communication resulting in continuity of care for the patient.

## **5.6 Discussion**

### **5.6.1 Summary**

This component of the study sought to create a transfer of knowledge between the technologies used in the assessment stage of maxillofacial prosthesis and the artificial eye process. Following Braun & Clarke's (2013) thematic analysis steps, three themes were generated which answered the research aim. The major themes and sub-themes are presented in Table 21.

The findings described above cluster around two concepts illustrating the maxillofacial and artificial eye prosthetic assessment (see Figure 9). The first concept relates to the factors involved in the current assessment process. The constructs within this concept include the equipment and tools used, the skills and expertise of the prosthetist, patient history, relationship between the prosthetist and the patient, and benefits of the assessment process. The second concept pertains

to creating change in the assessment process. This comprises the following constructs: the weaknesses of the current assessment process, complications associated with the current assessment process, incorporating technology into the assessment process and improving the working environment.

Table 21: Summary of major themes and subthemes

Theme Number	Theme title and sub-themes
1.	<b>Information gathering</b> Medical information Role of the patient Role of the prosthetist
2.	<b>Current equipment and tools used</b> Subjective measures Objective measures
3.	<b>Future improvements</b> Incorporation of technology Working practice

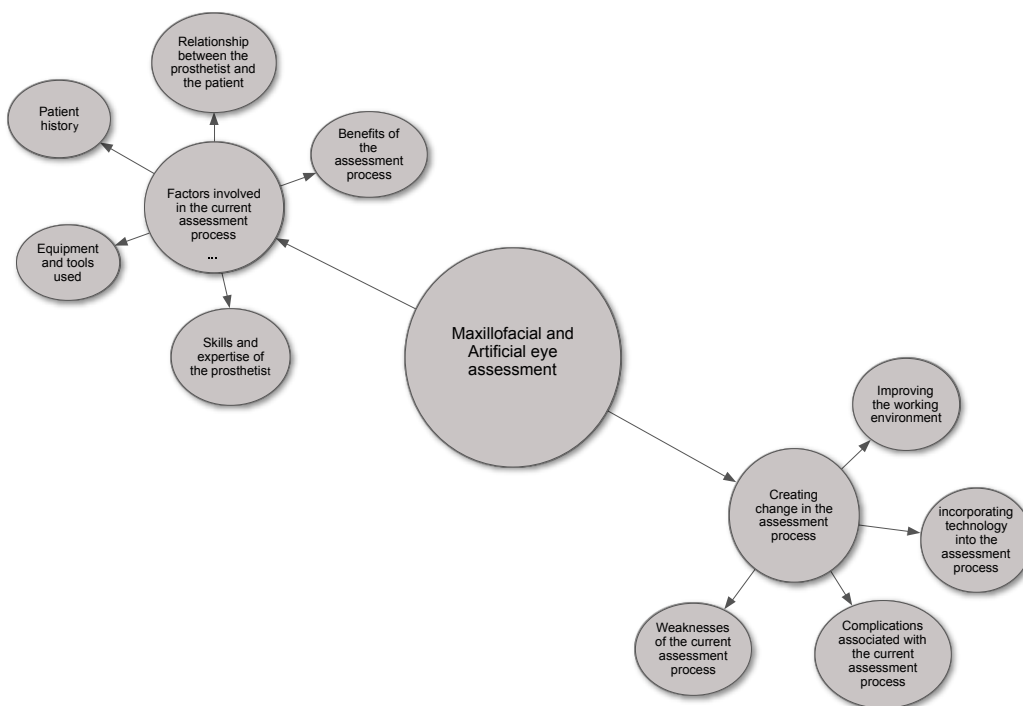


Figure 9: Illustrative diagram of qualitative findings

The ways in which these constructs interrelate are yet to be investigated and it is likely that some of them will be more heavily weighted than others. For example, the skills and expertise of the prosthetist is likely to have more influence on changes to the current process than the current tools and equipment used. Furthermore, the relationship between the prosthetist and the patient may



be a crucial factor in determining change. Despite this study showing the role of these constructs in the current assessment process and its future development, their relationships and weightings to each other are worthy of further investigation.

### **5.6.2 Differences and similarities between artificial eye and maxillofacial prosthesis**

The analysis stage of this component of the study discovered a number of similarities and differences in how MP's and AEP's conduct the assessment process. They occurred both between and within population groups. This was evident in the tools and equipment used. Access to and the types of tools used varied between both population groups. Half of MP's had access to technological advanced tools, such as diagnostic imaging (CT, MRI scans and ultrasounds), 3D printers and medical photographs. The other half of MP's, and the majority of AEP's, used more 'basic' tools, such as the materials used in creating an impression and in the case of AEP's, exophthalmometer, existing prosthesis, pen torch and a conformer. The use of tools was primarily based upon three factors: the type of patient and their presentation (tools being used for more complex cases), access to tools, which is dependent upon funding, and the need of the population the prosthetist is serving. Despite the differences in tools used, all AEP's and MP's felt that their use cannot replace their own knowledge, skills, and expertise; preferring instead to rely on their own visual judgement.

The importance given to their own experience influenced the prosthetists' views on how their field of work will develop. MP's who had access to technological advanced tools were more likely to view their work as heading towards a digital path (3D hand-held scanners, intra-oral cameras and 3D printers). This is in contrast to AEP's, who, although made suggestions for the incorporation of technology (3D hand-held scanners and medical photographs), felt that their work will primarily be reliant on their own skills.

Improving the working practice, specifically MDT working, was seen as an important development for both AEP's and MP's. Whilst the majority of MP's reported working within an MDT, its success was dependent on building a relationship with all members of the MDT, enabling effective communication whereby patients can be easily referred to other departments if required. Whereas the majority of MP's had a clear pathway to referring patients to different services such as mental health teams, AEP's were not aware of any referral process.

Potential differences in MDT working between MP's and AEP's include work location and working practice. MP's are predominately based at one location, AEP's can be based at different locations each working week: some of which are not in an ophthalmology department. This lack of permanent base makes it difficult to build a relationship with the relevant teams. With MP's often taking impressions pre-surgery, they are not only able to build a relationship with various departments, they are also at an advantage in developing an early relationship with the patient. However, AEP's meet the patient 6-weeks post-surgery, once the socket has healed.

Being able to build a relationship with the patient at the earliest opportunity allows the patient to be involved in the process from the start. All MP's and AEP's stipulated the importance of gathering information from the patient regarding what they already know, what they think will happen, what their expectations are and what their concerns may be. By understanding the patient's journey and being open and honest regarding what is possible as well as the limitations of the prosthesis, the patient is able to make an informed decision about their treatment - a considerable strength of the assessment process as highlighted by both population groups.

Further similarities between AEP's and MP's were: the information needed and how the assessment stage was conducted. All participants in both population groups reported a need to obtain a full medical history regarding the surgery and the reason for it. It is vital to assess the deficit site, to see if it has healed and to ascertain whether there are any infections, allergies, inflammation or discomfort experienced by the patient. However, there were disparities in the level of information required both between and within population groups. Whilst some prosthetists felt that they only need to know about the patients' medical history regarding the deficit site, others felt that a more general medical history including learning disability, mental health difficulties, heart conditions and so on, was important to know as this could inform the treatment plan and advice the prosthetist as to the best way to work with the patient. Age was also discussed as an important element regarding working effectively with the patient and their families. This linked to and highlighted the need for improvement regarding MDT working and having access to patient's notes via a central computerised system.

### **5.6.3 Relationship to the literature**

Healthcare interventions are often complex consisting of various components that may act both independently and inter-dependently (Medical Research Council, 2000). With both MP's and AEP's highlighting the importance of the patient relationship in the assessment and in the overall

success of the prosthesis, there is a need to understand their requirements to ensure effectiveness. Furthermore, in the cases of young patients, it would also be beneficial to investigate the views and perceptions of those who care for them. Understanding the requirements and expectations of all those involved in the process can increase success rate and limit the need for modifications and, in some instances, rejection (Martin, Murphy, Crowe & Norris, 2006; Medical Research Council, 2000; Ram, Grocott & Weir, 2008).

In keeping with the literature, findings from this component of the study emphasised the need for the end product to not only be functional, but also aesthetically pleasing, and to be able to fit into the working and living pattern of the patient (Martin et al., 2006). This has been shown to be a crucial element in the assessment stage. With patient outcomes and satisfaction being dependent on not only the outcome of the product, but also the process, particularly its ease, comfort and accessibility, the process of assessing a patient for a prosthetic fitting needs to be as patient friendly as possible. Obtaining subjective feedback from the patient may result in scientific development where their description of a problem or need in non-technical terms may be translated into technical requirements by a technology or material developer.

A focus on addressing the needs of the patient has resulted in the increasingly widespread incorporation of technology in modern healthcare (Eggbeer, 2008). Compared to AEP's, MP's were found to not only be utilising more objective tools and equipment, such as diagnostic imaging in the assessment process, they were also more open to the digital path they saw their work taking in the future. This is not to say that technological developments have not been investigated by AEP's and the services in which they are based. Indeed, the majority of this research has been focused upon the design and manufacturing stages. This has included digital imaging of the iris, varying impression methods and materials and rapid prototyping and manufacturing (Artopoulou et al., 2006; Cevik et al., 2012; Jain et al., 2010; Prithviraj et al., 2013). Limitations with these methods, such as the need for specialist equipment, batch production, where custom-made eyes are required, and limited time and cost saving benefits due to maintenance and updating equipment, have resulted in these techniques never coming to fruition. The need to investigate the capabilities of technology to be utilised in the assessment stage is apparent. This component of the study has, possibly, begun the process. What is needed is attracting significant investment from industry and funding for further research and development (Eggbeer, 2008).

Advancements in technology not only impact the prosthetic form and function, but also create confidence and improved wellbeing in the patient (Padmaja, 2015). One of the examples of new tools, identified in both the literature and in this component, are scanning technologies used in assessing the deficit site of maxillofacial prosthetic patients. They include structured light scanners, laser scanners, stereo-lithography, CT and MRI (Coward, Scott, Watson, Richards, 2007; Sabol, Grant, Liacouras & Rouse, 2011). The prosthesis is designed by taking a 3D scan of the patient's face and then inverting the data from the undamaged side so it overlaps the deficit site (Vale et al., 2016). Whilst these scans are effective in producing a three-dimensional image of facial anatomy (Bibb, Eggbeer & Evans, 2010; Ciocca et al., 2009; Evans et al., 2004; Li et al., 2015; Wu et al., 2009), they are unable to capture areas of shadow and fine details, such as hair and wrinkles (Bibb et al., 2000; Eggbeer, 2008; Evans et al., 2004; Lemperle et al., 2001; Vale et al., 2016). Whilst these limitations can be overcome by using plaster replicas of the deficit site for scanning purposes, it is time consuming. In a field that participants reported as being defined by time constraints, it makes it a non-viable long-term option. Further development of non-surface scanners to enable fine details of the deficit site to be captured (Evans et al., 2004; Li et al., 2015; Peng, Tang, Liu, & Peng, 2015) is essential for these small healthcare sector fields to either start or to further incorporate technology into their process. Whilst the investigation of technology and its future use in these fields is inevitable, technology itself will not be able to replace the experience, knowledge and expertise of the prosthetist.

Literature discussing the success of facial prosthesis has focused on the quality and functional aspects of the process rather than its technical facets (Chang, Garrett, Roumanas & Beumer, 2005; Lowental & Sela, 1982). Furthermore, few studies evaluate the outcome (aesthetic qualities) using quantitative measures, instead focusing on the performance aspects. As suggested by one participant in this component of the study, there is a need to measure quantifiable aspects of prosthesis characteristics at both the technical as well as the social and psychological levels.

Another area highlighted is that of MDT working. Multi-disciplinary working is becoming more prevalent within healthcare and is supported by policies and practices that bring care closer to the patient (Nancarrow et al., 2013). The findings from this study compliment the literature highlighting the benefits of MDT approach in: improved patient care and outcomes, enhanced coordination of care and improved mental well-being of patients and staff (Cancer Australia, 2017). With some prosthetists being placed in isolated locations, the ability to work as part of the MDT is not always viable. Therefore, having better access to patients' notes and their medical history can go some way towards improving the working environment of the prosthetist. This

can lead to a more standardised and equalised system for those working in a large department and those in smaller clinics.

#### **5.6.4 Strengths and limitations**

This qualitative component of the study was the first to examine the equipment and tools used by MP's and AEP's in the assessment process. Furthermore, it provides evidence for the utilisation and transfer of the technologies between these two population groups.

Semi-structured interviews are the most common qualitative method of data collection (Braun & Clarke, 2013). They are flexible and sensitive to the participant giving them the freedom to express their opinions in a way that best suits them. However, their success and validity (often known as *credibility* in qualitative research) is dependent on how well the participants' views and perspectives are reflected. Using non-leading questions and reducing preconceived ideas as much as possible helped in producing a study that best reflects the participants' experience. Furthermore, holding discussions between the researcher and the supervisory team allowed themes to be refined. This not only helped reduce the preconceived notions that may have been applied from the collection to the analysis of the data, but also enhanced confirmability. Checking and rechecking the data throughout the analysis also contributed to the confirmability of the study.

Whilst various analytical methods can be used for analysing interview data, the flexibility of TA and its ability to reflect and unpick reality made it a suitable approach to use (Braun & Clarke, 2006). Identifying themes and patterns of meaning across a dataset in relation to the research question allowed comparison of the two sectors.

Although this component of the study benefited from a diverse sample of participants, including a range of ages and geographical location of participants, it was limited by poor recruitment from both sectors. The response rate from both MP's and AEP's was low. Whilst this study highlighted many factors that are likely to be relevant across both sectors, the findings may be biased towards the opinions of those who were willing to participate in the research, therefore need to be treated with caution. Although the reasons for the low response rate are unclear, it is possible that the potential to change a system that is seen to be working could be an explanation for the low response rate from AEP's. Furthermore, the high demand for two small healthcare sectors may have meant that not all potential participants identified were able to participate.

This component of the study identified subjective and objective measures that are both currently used and have the potential to be utilised in the artificial eye process. Being able to assess the deficit site in an effective and accurate manner allows for the essential components of the design and manufacturing process to be realised whereby a good fit that is functional and aesthetically pleasing is obtained. These findings suggest the possibility of transferring the equipment used in maxillofacial prosthesis to the artificial eye service. Future research is needed to test its feasibility.

## **5.7 Conclusion**

This qualitative component of the study of how MP's and AEP's conduct the assessment process, specifically the tools and equipment used, has provided an overview of the factors involved and those that pertain to creating change. With no existing literature on utilising technological advanced equipment in the assessment stage of the artificial eye process, reference from similar fields that has done this, goes some way in informing the relevant processes.

Key factors in creating change include the importance of the patient, the working environment and the capabilities of technological equipment. Regarding the latter, MP's have access to and have utilised more technological advanced equipment and tools, such as diagnostic imaging, which was reported as having the potential to replace the impression technique. Whilst differences exist between both fields, investigation of whether the tools used by MP's can be applied and utilised by AEP's should be carried out.

Whilst there is potential for future application of technologies into the artificial eye process, direct and indirect costs in a sector where there are reduced resources mean that research and development is often slow and at times non-existent. Yet, with an evolutionary element where processes are continually re-developed until the end product results in excellent patient outcome and satisfaction, makes understanding technological capabilities as they currently stand of upmost importance.

## Chapter 6 - Discussion

### 6.1 Introduction

This thesis employed a qualitative approach to explore the artificial eye process in children with a diagnosis of Rb. With 70% of children diagnosed with unilateral Rb requiring enucleation and thus, a lifetime supply of artificial eyes, exploration of the process was needed to address its psychological impact and potential for technological development. Whilst research exists on the impact of an Rb diagnosis and the loss of an eye(s), literature on the impact of the artificial eye process in this population group was non-existent. Moreover, whilst investigation into technological advancements had begun in prosthetic health sectors, this has not yet reached the field of artificial eyes. This PhD research was therefore necessary to fill gaps in literature regarding the current methods utilised in the artificial eye process, its impact on children with Rb and their parent(s), and to assess the capabilities of current technology to advance the process. As such, this thesis formed one study exploring the artificial eye process in children with Rb, consisting of three components: (1) from the perspective of AEP's, (2) from the perspective of children with Rb and their parents, and, (3) in terms of technological advancement from the perspective of AEP's and MP's.

This chapter presents the opportunity to consider the ways in which individual's experiences and technological factors may impact on the rehabilitation process. By understanding the views of those represented in this thesis, this study offers a unique view point on the potential areas of influence that one's personal perspective may have on both their own experience and that of the others involved in the artificial eye process. The interaction between the child patient, their parent(s) and the AEP, is central to the success of the process. Highlighting potentially problematic areas within clinical practice creates a shared understanding where recommendations for its improvement can be made.

This chapter will summarise the key findings of each study component, present an overview of the key factors related to the artificial eye process in children with Rb, and, suggest recommendations for future research. Throughout this chapter the importance of human and technological factors within the artificial eye process will emerge.

## **6.2 Summary and clinical implications of each study component**

### **6.2.1 Artificial eye prosthetist perspective on the design and manufacturing process**

The aim of this component of the study was to gain an insight into the manufacturing and fitting process of artificial eyes in children with a diagnosis of Rb from the perspective of AEP's. Data was collected via a self-developed questionnaire and analysed using Braun & Clarke's (2013) TA.

The analysis highlighted two themes: (1) distress, and (2) barriers and facilitator's to success. The first theme of distress revolved around two subthemes: (1) clinical symptoms and associated factors present in the child patient, and (2) role of parents. The second theme also consisted of two sub-themes: (1) therapeutic relationship, and (2) within the system.

In conjunction with technological advancements, the role of the child patient's parent(s) and the AEP, were highlighted as key factors in contributing to the patient's acceptance and adjustment to the process. This suggests a potential link between technology and human factors.

The psychological implications from this component of the study predominately revolve around how well the child patient copes with the artificial eye process. Whilst coping mechanisms were not specifically referred to, the child patient's ability to cope with the process was linked to their parents in terms of how they manage their child having an artificial eye during appointments and in everyday situations. When this is not well-managed, the patient is likely to display distressing behaviours such as crying, anger, and refusal to let the AEP near their eye. This negatively affects the smooth running of the appointments which is imperative for obtaining a good fit and an aesthetically pleasing artificial eye(s). Noticeability of a disfigurement can lead to and/or exacerbates the patient's emotional wellbeing. In-depth exploration of the artificial eye process from the perspective of the patient and their parents is essential to create understanding of the difficulties experienced and how this can be improved. This has implications for both service process and delivery. For example, the way the process is conducted and the role of the AEP may be adapted through patient and family management approaches.

The success of the process relies not only on the patient's acceptance of the process, but also the skills and expertise of the AEP. Being bespoke in nature and requiring fine detail makes the



custom-made nature of artificial eyes being seen as not conducive to technological intervention. However, investigation is needed to ascertain whether and how technology can assist in the process, and if not, what adjustments and future developments are needed to make it possible. Suggestions have been made to explore this option through comparisons to other fields and proof of concepts. The specifics of the tools and equipment used were not explored in this study, thus requires examination to assess its success in the overall outcome of the artificial eye.

With the development of the process currently relying on the experience, time, equipment and materials used by AEP's, the absence of technological advancement may be due to a lack of investment and funding opportunities. Being seen as part of a larger prosthetic team, some participants felt that there is a need for recognition and certification of those working in the field. This will not only create potential research and development opportunities, but also produce clearer guidelines and pathways creating uniformity of processes and procedures reducing the variance in the outcome of artificial eyes produced.

### **6.2.2 Children with Rb and their parents perspective of the fitting process**

Whilst having an eye removed is known by healthcare professionals to be a traumatic event for both the patient and (in the case of young children), their parents, there is little understanding of the impact rehabilitation has on them. Reporting physical and emotional implications of the process and the importance of supportive measures made this an area worthy of further exploration. By working collaboratively with the patient from a young age and trying to understand the process from their perspective can help in their own sense-making of the process and how it has and will impact on their own being. This collaboration and the establishment of a therapeutic alliance serves as an indication as to how the patient will get on with the healthcare professional, and consequently, the treatment plan, whereby trust and communication is developed. This is particularly essential for children and young people as their sense of identity will be constantly evolving during their developmental years. As evidenced in the literature and supported by this component of the study, a small proportion of both patients and their parents will experience (short-lived) difficulties. Therefore, it is necessary to develop a pathway whereby both population groups can get specialised support if needed.

The aim of this component of the study was to understand the lived experience of the fitting process of artificial eyes following a diagnosis of Rb from the perspective of parents of and 13-16 year olds. The aim was achieved by employing an IPA approach in order to understand the feelings, values, and perceptions of both population groups. An ENQ was utilised in both

population groups: with 13-16 year olds being interviewed face to face and parent participants being interviewed via an online platform. Data was analysed by population group using Smith et al.'s (2009) IPA 6 stages.

The findings from 13-16 year old participants highlighted four superordinate themes: the artificial eye and the self; the artificial eye process; coping with the impact of the process, and, role of the parent(s). This is in contrast to the parent population whom revealed a need to improve the service and fitting process. Alongside process repercussions on the patient and how this can be managed, other superordinate themes highlighted include service experience and the onus is on the parent(s). The latter superordinate theme can be linked to the other two with parents coping mechanisms being linked to satisfaction level of the service and process.

Supporting and working collaboratively with the patient was seen as a network rather than being down to one or two specific individuals or a unit of people. At the forefront of the network were the child patient's parents' and the AEP's. Other healthcare professionals, siblings and other family members, friends, peers and the school staff were also seen as playing a key role in the patient's sense of being and identity. By having a strong network of people surrounding the patient will help ensure that their experience and feelings are validated and their lived experience is understood: a strength supported by the use of IPA as a method of analysis which attempts to validate lived phenomenon. Both population groups' experiences of the fitting process indicated that there is a genuine therapeutic benefit in a collaborative care approach. Whilst support is provided to parents of this population group, this is often done informally and when they are accessing the service for their child's on-going care and treatment. Consequently, an investigation into the impact of not only the artificial eye process, but also the disease, has on the parent's is needed in order to develop a support care plan for those who need it. This can also be applied to the patient's siblings whom are often affected yet forgotten when it comes to the impact of their sibling's treatment and care.

Whilst there is a therapeutic benefit of working closely with the AEP, parent participants reported dissatisfaction with the process. Service cuts, lack of training guidelines and the process being time consuming contributed to parent participant's feelings of a stressful and at times, traumatic experience. Offering concise instructions on how to provide medical and emotional support, how long patients should wait for treatment, and (in the case of artificial eyes) what methods to employ to produce the outcome, the development of clinical guidelines for this field is worthy of investigation.

Various provisions for children and young people wearing artificial eye(s) as a result of Rb exist: primarily by the two Rb departments in the UK. This includes an artificial eye support group and a program for Rb healthcare professionals to host a talk about Rb and artificial eyes to the child's school and classmates. Whilst these programs have proven successful, their effectiveness is yet to be measured. This would provide suggestions for further improvements to the programs as well as having the potential to identify further needs of the patient that require addressing.

While this component of the study has contributed to understanding the lived experience of 13-16 year olds and parents of children whom wear an artificial eye(s) as a result of Rb, it also serves to highlight a number of areas for further research. As an under-researched area, there are some interesting avenues for future IPA research (of which some have already been mentioned above). This component of the study has focused on the experiences of 13-16 year olds wearing an artificial eye(s) as a result of Rb. Future studies could focus on younger children as well as adults. An analysis of how the results from this component of the study compares to the same population group but with varying ages would be of particular interest. This could shed some light on the potential differing needs, requirements and experiences which may have a direct or indirect impact on a patient's treatment and care plan.

As prominent figures in the patient's life, an IPA study of the impact the process has on the patient's siblings will further explicate how the patient copes with the process as well as understand the impact it has on these population groups. On this note, it would also be beneficial to explore the experiences of wearing an artificial eye in those with acquired disfigurement. This population group are more prone to adjustment and acceptance difficulties; therefore, their needs will vary.

A second area of further research would be to examine in greater detail the relationship between acceptance and quality of life. In light of the fact that acceptance was found to be essential in how participants cope with having an artificial eye fitted, a thorough exploration of its impact on participants' daily life is needed. This has the potential to highlight areas that need addressing that have not been covered here.

Furthermore, concerning the actual artificial eye, some problems remain unsolved (such as the impression material - described as “*gritty*”) and limitations exist regarding the design (fixed iris). It is therefore important to find out what is causing these problems and how they and current limitations can be addressed. A focus group consisting of AEP’s, service managers, artificial eye patients and design and manufacturing specialists will enable a brain-storming approach whereby problems are discussed, ideas are shared, and developments proposed.

Finally, with consistency of treatment and outcome contributing to managing patient expectations, an exploration of how this can be achieved within the artificial eye process is warranted. As a subjective and bespoke discipline, clinical guidelines may not be appropriate. However, the flexibility required can be retained by developing service specific training programme and nationwide patient and family management programme.

### **6.2.3 Transfer of knowledge in the assessment of prosthesis**

Having revealed a potential link between the way artificial eyes are fitted and the psychological wellbeing of the child patient and their parents, the aim of this component of the study was to create a transfer of knowledge between the technologies used by AEP’s and MP’s. Data was collected via semi-structured face to face interviews. Data was analysed thematically, with 3 themes being highlighted: (1) information gathering, (2) current equipment and tools used, and, (3) future improvements.

With participants reporting the need of the assessment process for a prosthetic fitting needing to be patient friendly, the experience, expertise and knowledge of the AEP/MP is vital. Utilising the skills of the prosthetist was found to put the patient at ease, manage their expectations and provide a mutual relationship where the process and the subsequent feedback was a two-way loop. This was found to be particularly important for those prosthetists who work in isolated clinics outside of a MDT, where access to patient information such as medical notes is not easily accessible. Objective tools were used by both MP and AEP’s, however, MP’s had access to more technological equipment than AEP’s and saw their field heading down a more digital path.

With similar assessment procedures to one another, future research into the current state of knowledge of transferring methods and tools from one sector to another is needed to determine its feasibility. Utilising a step wise approach in evaluating the results of future research (from

defining components of the intervention to usability tests and cognitive walkthroughs), as well as holding discussions of the potential success of the product, needs to take place to ensure a good fit between the products technology and the skills of the company producing the product (Campbell et al., 2000; Cooper & Kleinschmidt, 1986; Martin et al., 2006). However, there are a number of factors that need to be considered when developing new techniques and when incorporating new methods into an existing process (Eggbeer, 2008; Ram et al., 2007). These are: financial viability, time saving and changing technology costs (direct and indirect costs), staff lack of technological expertise, and some possible resistance to change. With the incorporation of technology potentially being inhibited by current work practices, a greater awareness of the elements of success is needed to ensure that change only occurs where benefits will be obtained.

Although highlighted as being essential to a successful process, MDT working may prevent the spontaneous need to work case by case, as is often required in the artificial eye process. Nevertheless, there is a need to enhance MDT working within this sector to improve communication, efficiency, and overall a better patient-centred care approach.

Working multi-disciplinary can result in increased involvement in the patient's journey, whereby regular contact is made with the patient from pre-surgery up to their first appointment. In cases where dramatic and traumatic changes take place, such as Rb, contact with the patient and their parents is essential for providing accurate information, managing expectations and offering ongoing support (either from the members of the MDT or via referral to a specialised team if required). Furthermore, by working within an MDT, prosthetists would have access to all relevant patient notes, thereby shortening or replacing the information gathering stage of the assessment appointment with the first stages of creating the prosthesis. Increased rapport and enhanced patient-practitioner relationship are achieved at the start of the patient's journey which not only positively affects the treatment plan, but also allows the patient to remain part of, and contribute to, the wider community they live in.

These qualitative findings highlight the need for two essential developments to the artificial eye process. Firstly, technology transfer, and, secondly, human work practices. Suggestions for future research include further exploration of the (un)successful elements of the artificial eye process and an investigation into if, and where, there is potential resistance to change; the creation of positive links with associated departments for effective MDT working, and to consider creating care pathways for patients requiring prosthesis whilst retaining flexibility in the work process of AEP's.

### **6.3 Comparison and synthesis of findings**

The individual components of the study have explored various aspects of the artificial eye process in children with Rb from the perspective of those directly and indirectly affected. With the aim of addressing the psychological impact and its potential for technological advancement, the individual components will be discussed as a whole leading to the overall significant contribution to knowledge.

One key finding from this study was that stagnation of the technological advancement of artificial eyes. This was evident in the scoping review section of the background chapter; with discussions with artificial eye services during clinic visits, and, in the first component of this study where AEP's shared their views on the current process and the potential for its development. Component 1 highlighted recent developments being AEP led, based on their experience and expertise. This could account for the hesitation reported regarding the incorporation of technology in the artificial eye process. Given the bespoke nature of artificial eyes and that current technology is unable to produce the results needed such as producing a good fit and colour matching, many AEP's felt that the process was sufficient as it currently stands.

Further explanation for both the stagnation and current development is due to varying regulations and guidelines of the process in Europe. Respondents in component 1 highlighted the need to create standards of care across the board that addresses issues such as training and certification. This could then lead to funding and investment whereby investigation into technological advancements can begin.

Another key finding of the background chapter was the psychological impact an artificial eye(s) and/or disfigurement have on the patient. The literature identified predominately focused on the before and after the loss of an eye, highlighting an adjustment period and above average levels of clinical anxiety and depression, maladaptive behaviours and reduced emotional wellbeing in these patients. Further findings from component 1 included the importance of obtaining a good fit, and a need to understand the child patients' concerns as well as their parents whom, alongside AEP's were found to be an influencing factor in the success of the process.

Exploring the fitting process from the perspective of 13-16 year old children with Rb and their parents (component 2) filled in this gap in the literature regarding psychological impact and re-confirmed the above findings of obtaining a good fit to create an aesthetically pleasing artificial eye as well as the importance of the parents in attending the appointments with their child. Whilst the fitting process did not highlight above average levels of distress in 13-16 year olds, distress was found in some parents, primarily due to insufficient coping mechanisms. The stable sense of self experienced by 13-16 year olds can be attributed to their reliance of a good support network, specifically their parent(s) whom through normalising their child's experiences via positive reinforcement allows them to look beyond their disability. Furthermore, the early age of diagnosis, treatment and after-care can account for the disparities in emotional distress experienced by both population groups. The identification of distress occurring at an earlier age in children wearing an artificial eye(s) as a result of Rb (component 1 and 2) is likely to be a result of lasting memories not being formed. This may further account for parents experiencing reduced emotional wellbeing as well as dissatisfaction with the process (component 2).

Enriching component 1 finding of the importance of obtaining a good fit, both 13-16 year olds and parents reported that receiving a good fit directly relates to the patient's emotional wellbeing and ability to cope with the process as well as the daily maintenance of an artificial eye. In contrast to 13-16 year olds whom reported satisfaction with the process and outcome, the majority of parents reported feelings of dissatisfaction. This could provide explanation for the disparity between both population groups perceived own emotional wellbeing. The difficulty of the process (which was perceived as having more of an impact than their child's distress) centred around funding cuts, ineffective training programme, and the subjective nature of the process. Alongside its re-emphasis of developing guidelines to create uniformity in the process as highlighted by component 1, this finding suggests a need for development to how artificial eyes are made.

Whilst component 3 highlighted the potential of technology to be incorporated into the artificial eye process, measures for patient satisfaction which strongly revolved around the relationship with the AEP was seen as more imperative. These findings are consistent with those from component 1 and 2 which found that the patients (and parent's) satisfaction, adherence and outcome increases a positive relationship with the AEP. With AEP's highlighting their role in managing patient's distress (component 1), their ability to understand the impact of the process upon the patient, involve them and their parent(s) in the process, enabling the patient to make an informed decision and managing expectations is essential in improving patient (and parental) satisfaction (component 1 and 3). Furthermore, the dissatisfaction experienced by parents in component 1 was often directed at the professional that could not meet expectations rather than

the cause of the negative outcome, such a treatment and surgery. Having highlighted success as being dependent on building relationships (component 1 and 3), there is a need for AEP's to work in a more conducive MDT whereby access to patient's medical information and relevant healthcare professionals are readily available, enabling effective communication whereby patients can be easily referred to other departments if required, for example emotional support services (component 1 and 3).

Although AEP's own knowledge, expertise, and experience were seen as just as important as the equipment used (components 1 and 2), component 3 highlighted the potential of incorporating the digital tools used by MP's (such as 3D hand-held scanners and intra-oral cameras) into the artificial eye process. Although dependent on funding, the needs of the population group AEP's serve (as highlighted in this thesis), suggests that its investigation is warranted.

## **6.4 Human and technological factors**

This thesis has achieved the stated aim of exploring the artificial eye process in children with a diagnosis of Rb: addressing the psychological impact and potential for technological development. The commonalities and differences identified across all components highlighted an intrinsic link between human and technological factors. The artificial eye process can be seen as linear in nature as its success is dependent on the patient's acceptance and adjustment which is dependent on how the process is conducted (the role of the AEP and the child patient's parents).

Human factors are a promising mechanism with which to improve healthcare delivery. It refers to the understanding of human behaviour and performance. It is about understanding how humans operate, such as understanding ones feelings, cognitive processes and limitations of working memory, in order to improve how humans interact with a given object and/or environment. Using human factors enables expansion beyond the concept of patient satisfaction. It can allow learning of how to improve the experience before patients become part of the process. For this to happen, we need to get feedback from product users from the beginning when designing the new system/product.

With human errors being inevitable, eliminating them is futile. Thus, the focus needs to be on gathering data about human characteristics and human interactions with the environment to design systems and tools that support physical and cognitive abilities of humans and are resilient to



unanticipated events. This approach can foster a culture of safety, promote a learning environment and encourage the development of a culture where unintentional errors are reported and findings used to improve various systems to yield change.

With systems being reported as creating challenges for people, identification of these allows for their modification in aiding those who use and/or work with them. Furthermore, as well as designing systems to support those that use them, understanding the factors related to the broader organisational components are needed to achieve the desired outcome.

By focusing on the elements that influence our performance (for example, tools and technology), theories can be applied to the information to ensure that what people are interacting with is done most effectively and with limited error. Whilst the acceptance and utilisation of technological innovations in healthcare are beneficial for both the healthcare professional and patients, influencing factors may differ between them. While perceived benefits of technological innovations may be the most important factor for the healthcare professional, ease of use is of most importance for the patient. Thus, understanding the interaction between the needs of both patient and the healthcare professional can ensure that technology is used that is of benefit to everyone.

Whilst there is an intrinsic link between human and technological factors, two points need to be remembered. Firstly, patients are the reason for healthcare and they should be at the centre of it. Secondly, technology does not have an agenda of helping healthcare. It develops because of miniaturisation, lowering costs of production and so on. Thus, although technology exists, its use is only justified if it can benefit patient welfare: both in terms of their physical and psychological wellbeing. Through understanding the system we are part of: in terms of its technological and human factors; then they can support us rather than oppose us.

## **6.5 Implications and recommendations for future research**

This thesis has enabled the artificial eye process to move forward by understanding the factors involved in the current process (parent and AEP influences, MDT working and the current tools and equipment used) and those related to creating change (complications and weaknesses of the current process, impact on AEP's and patients in incorporating technology into the process, and improving the working environment through clearer yet flexible guidelines and care pathways).

Through understanding the requirements and needs of those (in)directly involved in the process, recommendations can be made for future research whereby it is hoped a resource drive can begin to produce artificial eyes that are in keeping with the latest technological advancements whilst producing the best possible outcome for the patient.

Further in-depth exploration of the technological and human factors that are inherently linked to the success of the artificial eye process is required. Although intrinsically linked, to get to the crux of the role these factors play, they need to be considered individually and in combination. Thus, this section will make recommendations for future research addressing both the whole and the part of these factors.

With this thesis highlighting the effectiveness of support networks (component 1 and 3) and distress commonly occurring in younger patients (component 1 and 3), investigation into these areas are warranted. Further exploration of the role of family members, particularly parents whom as the primary caregiver attends the appointments with the patient and assists them in caring for their artificial eye, as well as friends whom are often inquisitive and do not differentiate them for others due to their disability, can provide vital information on the needs and requirements of the patient. By understanding the views of others who care for children with Rb, can increase success rates and limit the need for modifications and in some instances, (emotional as well as physical) rejection.

The identified lack of understanding of the impact rehabilitation has on the patient can account for the level of distress experienced. By exploring the impact of the process upon the patient and their parent(s), effective management programs can be developed (components 1 and 3). This may take the form of extra training for AEP's to manage the patient in clinic, or, for those experiencing high levels of distress, referral to emotional support services. An example of this is the application of the Cognitive Behavioural Therapy (CBT) approach to the artificial eye process to teach the patient positive ways to process their emotions and facilitate their overall adjustment.

Creating artificial eye process measures of quality of life (QoL) including the patient's current wellbeing and the potential for psychosocial difficulties can further contribute to creating a behaviour management plan. With acceptance being found as essential to how patient's cope with having an artificial eye fitted, its relationship with a patient's overall QoL is worthy of exploration. It is also imperative to take into consideration other reasons for patient's

experiencing distress, such as loss of eye (component 1), location of appointments (component 3), relationship with the AEP (component 1 and 3), own and those closest to them coping mechanisms (component 1,2 and 3), as well as factors outside of having an artificial eye.

These additional factors point to the importance of AEP's working successfully within a MDT. Often working in isolated locations and clinics, access to patient's notes, medical history and other staff connected to the patient's care are not always available. This can result in differences in service delivery and product outcome: both of which have the potential to reduce patient satisfaction and increase levels of distress. Whilst improved access to patient's notes will create a better patient-practitioner relationship, it may also lead to more clinical guidelines and care pathways that may not be suitable for AEP's given the bespoke nature of their work. Therefore, exploration of creating care pathways that offer flexibility in AEP's working practice is needed. In addition, these guidelines would give AEP's recognition of their work where it is seen as a specialty field in its own right.

With the psychological impact of the artificial eye process being complex in nature, creating a collaborative care model for mental health in medical contexts can be cost-effective and can improve patient experience and the outcomes for patients in clinical practice. In this model, it is likely that oncology nurses and specialists will be the first port of call for those presenting with psychological symptoms relating to the disease and/or artificial eye process and can provide initial psychological support.

Attending appointments and caring for their child's artificial eye, makes understanding parent's distress as important as the patients. Providing parents with concise instructions on how to provide medical and emotional support to their child, involving them in all aspects of the process and managing expectations through adequate information giving at every stage can improve their wellbeing.

With parents of children with Rb being found to have higher levels of dissatisfaction of service delivery and product outcome compared to their child (component 2), primarily due to the subjective nature of the process and no formal training or guidelines followed, investigation into the development of clinical guidelines and care pathways is warranted. As a hand-crafted process that produces bespoke artificial eyes, flexibility in terms of processes is required (component 3).

Through exploration of the impact of the artificial eye process by those (in)directly affected, improvements to the service delivery and product outcomes can begin. Creating a focus groups of AEP's, service managers, patients, family members and technologist specialists can allow for a brain storming session where the description of a problem/limitation or a requirement and need can be translated into technical requirements. This can lead to the creation of prototypes and proof of concepts in assessing its feasibility. The use of diagnostic imaging tools such as 3D scanners, 3D printers and RP&M processes (as used by MP in component 3) can demonstrate whether the design and manufacturing concept has potential and is feasible for continual research and development.

Furthermore, with various techniques, equipment, and processes being used by AEP's coupled with fluctuating levels of patient and familial satisfaction, exploration of the relationship between the two is needed. By understanding the successes and failures of the current tools used, a more standardised process can be developed creating uniformity within and between services as well as equalising patient satisfaction.

In addition to exploring the (un)successful aspects of the process, investigation of AEP's potential resistance to change what is seen as an effective procedure, is needed (component 1). In addition to the artificial eye process being seen as skills based, other underlying reasons may exist for their hesitation to process change. This may include the potential for their skills to become redundant, thus, being replaced by technological equipment and its handlers. By understanding the inhibitors to change, developments that meet patient expectations can begin.

With this thesis focusing on children with a diagnosis of Rb, exploration of the impact of the process of adult survivors is needed: specifically understanding the process from their perspective as well as younger children (<13 years old). With developmental factors playing a role in the patient's adjustment, understanding the process from the perspective of patients of varying ages can shed light on differing needs, requirements and experiences allowing their treatment and care plan to be tailored to their specific needs. Furthermore, with the finding that distress is more common in younger patients (component 1), understanding their experiences can enable more effective treatment and care plans. This could enhance MDT working and provide parents with more effective tools in managing their child's distress inside and outside of the artificial eye appointments.

Furthermore, whilst this thesis focused on the artificial eye process, it would be worthy for future research to delve into other areas related to artificial eyes, not only in patients with a

diagnosis of Rb, but also those whom wear an artificial eye for different reasons. This may include what it is like wearing/living with an artificial eye and how it affects specific parts of the patient's life, for example, school, home, socialising, and work. By gaining a holistic view of the impact artificial eyes has on the wearer, adjustments to the process may be able to take place that will have a positive effect on all areas of the patient's life.

Although the technological factors point to a more investigative approach, for example, the use of RP&M processes to develop artificial eyes, it is important that we first understand its feasibility. Exploring the potential of technology to be used in the artificial eye process can be done by comparing the tools and equipment used to that of a similar field which is currently investing in advanced technological equipment. At the same time, it is imperative to gain an in-depth understanding of the human factors that play a role in the process. Understanding is needed of not only the impact of the current process, but also the needs and requirements of those (in)directly involved in the process. The ability of current technology to meet these requirements can then be assessed. Investigation of current technologies used in the process, or its further development before its application will become the starting point for improvements.

## **6.6 Significant contribution to knowledge**

This thesis significantly contributed to our knowledge in the following ways:

- Identified and began the process of addressing the absence of knowledge regarding the psychological impact of the artificial eye process in children with Rb.
- Identified variations in impact of the artificial eye process in AEP's, children with Rb and their parents: creating suggestions for an improved process to enhance satisfaction to service delivery and product outcome.
- Established a need for further research and development into the incorporation of technology in the artificial eye process.
- Highlighted the need to treat human and technological factors as a continuum in improving the artificial eye process.
- Highlighted a need for better uniformity of artificial eye services and clarity of processes used as well as for the services to be recognised in their own right.

## **6.7 Conclusion**

By using a qualitative approach, this thesis contributes new knowledge to the literature on the psychological and technological impact of the artificial eye process in children with a diagnosis of Rb. A rich and detailed understanding has been obtained of parents of, and the child patient (13-16 year olds) wearing an artificial eye as a result of Rb as well as the AEP views of the impact of the process and how it can be improved. Furthermore, foundations have been set for the technological advancement of the current process.

The common theme identified throughout this thesis is the intrinsic link between human and technological factors, and the role they play in the artificial eye process. To create change in a process that has been stagnated for 60 years, a thorough understanding of the needs of those (in)directly affected and the capabilities of technology to meet these needs is of utmost importance. Taking into consideration the recommendations made above, it is the researcher's perspective that first and foremost an in-depth exploration of the human and technological factors associated with the artificial eye process from the perspective of children with Rb across the developmental age range is needed. From combining these findings with that of this thesis, a step-wise approach in evaluating the feasibility of creating artificial eyes through technological means (such as creating proof of concepts) can begin.

## References

- Allen, L., & Webster, H. E. (1969). Modified Impression Method of Artificial Eye Fitting. *American Journal of Ophthalmology*, 67, 189 - 218.
- Amedi, A., Raz, N., Pianka, P., Malach, R., & Zohary, E. (2003). Early “visual” cortex activation correlates with superior verbal memory performance in the blind. *Nature Neuroscience*, 6(7), 758 - 766. doi: 10.1038/nm1072
- Arksey, H., & O'Malley, L. (2005). Scoping studies: towards a methodological framework. *International Journal of Social Research Methodology*, 8(1), 19 - 32.
- Artopoulou, I. I., Montgomery, P. C., Wesley, P. J., & Lemon, J. C. (2006). Digital imaging in the fabrication of ocular prostheses. *The Journal of Prosthetic Dentistry*, 95(4), 327 - 330. doi: 10.1016/j.prosdent.2006.01.018
- Askins, M. A., & Moore, B. D. (2008). Psychosocial Support of the Pediatric Cancer Patient: Lessons Learned Over the Past 50 Years. *Pediatric Oncology*, 10(6), 469 - 476. doi: 10.1007/s11912-008-0072-1
- Atay, A., Peker, K., Günay, Y., Ebrinc, S., Karayazgan, B., & Uysal, ö. (2013). Assessment of health-related quality of life in Turkish patients with facial prostheses. *Health and Quality of Life Outcome*, 11, 11
- Atkinson., P., & Hammersley., M. (1994). Ethnography and participant observation. In K. N., Denzin., & Y. S., Lincoln (Ed.), *Handbook of Qualitative Research* (pp. 248 - 261). Thousand Oaks: Sage Publications
- Ayanniyi, A. A. (2013). Emotional, Psychosocial and Economic Aspects of Anophthalmos and Artificial Eye Use. *The Internet Journal of Ophthalmology and visual Science*, 7(1). doi: 10.5580/158a
- Bach-y-Rita, P., & Kercel, S. W. (2003). Sensory substitution and the human-machine interface. *Trends in Cognitive Sciences*, 7(12), 541 - 546. doi: 10.1016/j.tics.2003.10.013

- Balmer, A., & Munier, F. (2007). Differential diagnosis of leukocoria and strabismus, first presenting signs of retinoblastoma. *Clinical Ophthalmology*, 1(4). from <http://www.ncbi.nlm.nih.gov/pmc/articles/PMC2704541/pdf/oph-1-431.pdf>
- Balmer, A., Zografos, L., & Munier, F. (2006). Diagnosis and current management of retinoblastoma. *Oncogene*, 25(38), 5341-5349. doi: 10.1038/sj.onc.1209622
- Bartlett, S. O., & Moore, D. J. (1973). Ocular Prosthesis: A Physiologic System. *Journal of Prosthetic Dentistry*, 29(4), 450 – 459
- Batra A., K., M., Paul, R., Patekar, M., Dhawan, D., & Rakhshi, S (2016). Quality of Life Assessment in Retinoblastoma: A Cross-Sectional Study of 122 Survivors from India. *India Pediatric Blood Cancer*, 63, 313 – 317
- Bassell, A. G. (2001). Children surviving cancer: Psychological adjustment, quality of life, and school experiences. *Exceptional Children*, 67(3), 345 - 359. doi: 10.1177/001440290106700304
- Beaudin, J. S., Intille, S. S., & Morris, M. E. (2006). To track or not to track: user reactions to concepts in longitudinal health monitoring. *Journal of Medical Internet Research*, 8(4), 29. doi: 10.2196/jmir.8.4.e29.
- Benson, P. (1977). The Fitting and Fabrication of a Custom Resin Artificial Eye. *Journal of Prosthetic Dentistry*, 38, 532 - 538. doi: 10.1016/0022-3913(77)90029-4
- Bi, Y., Wu, S., Zhao, Y., & Bai, S. (2013). A New Method for Fabricating Orbital Prosthesis with a CAD/CAM Negative Mold. *Journal of Prosthetic Dentistry*, 110(5), 424 - 428. doi: 10.1016/j.prosdent.2013.05.003
- Bibb, R., Eggbeer, D., & Evans, P (2013). Rapid Prototyping Technologies in Soft Tissue Facial Prosthetics: Current State of the Art. *Rapid Prototyping Journal*, 16(2), 130 - 137. doi: 10.1108/13552541011025852
- Biggerstaff, D. L., & Thompson, A. R. (2008). Interpretative Phenomenological Analysis (IPA): A Qualitative Methodology of choice in Healthcare Research. . *Qualitative research in psychology*, 5, 173 - 183.



- Bonanno, A., Esmaell, B. (2012). Facial Disfigurement, Stigma, and Cancer: Interaction Between Patients and Members of Secondary Groups. *Sociological Spectrum*, 32(2), 138156.
- Boynton, P. M. (2004). Selecting, designing, and developing your questionnaire. *British Medical Journal*, 328(7451), 1312 - 1315. doi: 10.1136/bmj.328.7451.1312
- Brannen, J. (1988). Research note The study of sensitive subjects. *The Sociological Review*, 36(3), 552 - 563. doi: 10.1111/j.1467-954X.1988.tb02929.x
- Bowns, L., Kirshner, E. L. & Steinbach, M. J. (1994). Shear sensitivity in normal and monocularly enucleated adults. *Vision Research*, 34, 3389 – 3395
- Braun, V. Clarke., V. (2006). Using Thematic Analysis in Psychology. *Qualitative research in psychology*, 3(2), 77 - 71-71.
- Braun, V., & Clarke, V. (2013). Teaching thematic analysis: Overcoming challenges and developing strategies for effective learning. *The Psychologist*, 26(2), 120-123.
- Brocki, J., & Wearden, A. (2006). A critical evaluation of the use of interpretative phenomenological analysis (IPA) in Health Psychology. *Psychology and Health*, 21(1), 87 - 108.
- Buhler, C. (1996). Approach to the analysis of user requirements in assistive technology. *International Journal of Industrial Ergonomics*, 17, 187 - 192.
- Bull, R., Rathborn, H., & Clifford, B. R. (1983). The voice-recognition accuracy of blind listeners. *Perception*, 12(2), 223 - 226. doi: 10.1068/p120223
- Burton, G. (2000). The role of the sound of tapping for nonvisual judgment of gap crossability. *Journal of Experimental Psychology: Human Perception and Performance*, 25(3), 900 - 916. doi: 10.1037/0096-1523.26.3.900
- Burton, H., Snyder, A. Z., Conturo, T. E., Akbudak, E., Ollinger, J. M., & Raichle, M. E. (2002). Adaptive changes in early and late blind: A fMRI study of Braille reading. *Journal of Neurophysiology*, 87(1), 589 - 607. doi: 10.1152/jn.00129.2002
- Buston, K., Parry-Jones, W., Livingston, M., Bogan, A., & Wood, S. (1998). Qualitative Research. *British Journal of Psychiatry*, 172, 197 – 199

- Butler, D. J., Turkal, N. W., & Seidl, J. J. (1992). Amputation: Preoperative psychological preparation. *The Journal of the American Board of Family Medicine*, 5(1), 69 - 73. doi: 10.1016/j.pmr.2013.09.010
- Campbell, M., Fitzpatrick, R., Haines, A., Kinmonth, A. L., Sandercock, P., Spiegelhalter, D., & Tyrer, P. (2000). Framework for Design and Evaluation of Complex Interventions to Improve Health. *British Medical Journal*, 321(7262), 694 - 696
- Campbell, M., Meier, A., Carr, C., Enga, Z., James, A. S., Reedy, J., & Zheng, B. (2001). Health behavior changes after colon cancer: a comparison of findings from face-to-face and on-line focus groups. *Family Community Health*, 24(3), 88 - 103.
- Cancer Australia. (2017). *Benefits of multidisciplinary care*. Retrieved March 3rd, 2017, from <https://canceraustralia.gov.au/clinical-best-practice/multidisciplinary-care/all-about-multidisciplinary-care/benefits-multidisciplinary-care>
- Carty, C. A. (2009). Retinoblastoma: an overview for advanced practice nurses. *Journal of the American Academy Nurse Practitioners*, 21(3), 149-155. doi: 10.1111/j.1745-7599.2008.00378.x
- Cassidy, E., Reynolds, F., Naylor, S., & De Souza, L. (2011). Using interpretative phenomenological analysis to inform physiotherapy practice: an introduction with reference to the lived experience of cerebellar ataxia. *Physiotherapy, theory and practice*, 27(4), 266 - 277. doi: 10.3109/09593985.2010.488278
- Cerullo, I. (1990). Plastic Artificial Eyes: Overview and Technique. *Advances in Ophthalmic Plastic and Reconstructive Surgery*, 8, 25 - 45.
- Cevik, P., Dilber, E., & Eraslan, O. (2012). Different Techniques in Fabrication of Ocular Prosthesis. *Journal of Craniofacial Surgery*, 23(6), 1779 - 1782. doi: 10.1097/SCS.0b013e31826701bb
- Chang, T. L., Garrett, N., Roumanas, E., & Beumer, J. (2005). Treatment satisfaction with facial prostheses. *Journal of Prosthetic Dentistry*, 94(3), 275 - 280.
- Chantada, G., Fandino, A., Casak, S., Manzitti, J., Raslawski, E & Schwartzman, E. (2003). Treatment of overt extraocular retinoblastoma. *Medical And Pediatric Oncology*, 40(3), 158 - 161. doi: 10.1002/mpo.10249
- Charlsworth, F. (1966). Fitting an Artificial Eye. *Nursing Times*, 62(6), 172 - 174

- Chauhan, A., & Sharma, S. D (2016). Chemotherapy in Retinoblastoma. *International Journal of Therapies & Rehabilitation Research*, 5(1), 55 – 60
- Chebat, D. R., Maidenbaum, S., & Amedi A. (2016). Navigation using sensory substitution in real and virtual mazes *PLoS One*, 10(6)
- Chen, Z., Moran, K., Richards-Yutz, J., Toorens, E., Gerhart, D., Ganguly, T., Shields, C. L., & Ganguly, A. (2014). Enhanced sensitivity for detection of low-level germline mosaic RB1 mutations in sporadic retinoblastoma cases using deep semiconductor sequencing. *Human Genome Variation Society*, 35(3), 384 - 391. doi: 10.1002/humu.22488
- Childhood Eye Cancer Trust. (2014). *About Retinoblastoma (Rb)*. Retrieved September 20, 2014, from <http://www.chect.org.uk/cms/index.php/about-rb>
- Chintagumpala, M., Chevez-Barrios, P., Paysse, E. A., Plon, S. E., & Hurwitz, R. (2007). Retinoblastoma: review of current management. *Oncologist*, 12(10), 1237-1246. doi: 10.1634/theoncologist.12-10-1237
- Churchill, S. D., & Wertz, F. J. (2015). An Introduction to Phenomenological Research in Psychology: Historical, Conceptual, and Methodological Foundations. In 2nd (Ed.), *Handbook of Humanistic Psychology*: Sage.
- Ciocca, L., Fantini, M., Marchetti, C., Scotti, R., & Monaco, C. (2010). Immediate facial rehabilitation in cancer patients using CAD-CAM and rapid prototyping technology: A pilot study. *Support Care Cancer*, 18, 723 - 728. doi: 10.1007/s00520-009-0676-5
- Clarke, A. (1999). Psychosocial aspects of facial disfigurement: problems, management, and the role of a lay-led organization. *Psychology Health & Medicine*, 4, 127–142.
- Clarke, A., Rumsey, N., Collin, J. R. O., & Wyn-Williams, M. (2003). Psychosocial distress associated with disfiguring eye conditions. *Eye*, 17, 35 - 40. doi: 10.1038/sj.eye.6700234
- Collignon, O., Voss, P., Lassonde, M., & Lepore, F., . (2009). Cross-modal plasticity for the spatial processing of sounds in visually deprived subjects. *Experimental Brain Research*, 192(3), 343 - 358. doi: 10.1007/s00221-008-1553-z

- Collins, K., & Nicolson, P. (2002). The meaning of 'satisfaction' for people with dermatological problems: reassessing approaches to qualitative health psychology research. *Journal of Health Psychology, 7*(5), 615 - 629. doi: 10.1177/1359105302007005681.
- Cohen, D., & Crabtree, B. (2006). Qualitative Research Guidelines Project. Retrieved March 5th, 2017, from <http://www.qualres.org/HomeTria-3692.html>
- Conrad, P. (1987). The experience of illness: recent and new directions. *Research in the Sociology of Health Care, 6*, 1 - 31.
- Converse, P. D., Wolfe, E. W., & Oswald, F. L. (2008). Response rates for mixed-mode surveys using mail and e-mail/web. *American Journal of Evaluation, 29*(1), 99 – 107
- Cook, R., Zoumpoulidou, G., Luczynski, M. T., Rieger, S., Moquet, J., Spanswick, V. J., Hartley, J. A., Rothkamm, K., Huang, P. H., & Mittnacht, S. . (2015). Direct Involvement of Retinoblastoma Family Proteins in DNA Repair by Non-homologous End Joining. *Cell Reports, 10*, 1 - 13. doi: 10.1016/j.celrep.2015.02.059
- Cooper, R. G., & Kleinschmidt, E. J. (1986). An investigation into the New Product Process: Steps, Deficiencies and Impact. *Journal of product Innovation Management, 3*(2), 71-85. doi: 10.1016/0737-6782(86)90030-5
- Corner, R., & Gould, E. (2012). *Psychology Around Us* (2nd ed.). United States of America: John Wiley and Sons.
- Coulter, A., Roberts, S., & Dixon, A (2013). Delivering better services for people with long-term conditions Building the house of care. from [https://www.kingsfund.org.uk/sites/files/kf/field/field\\_publication\\_file/delivering-better-services-for-people-with-long-term-conditions.pdf](https://www.kingsfund.org.uk/sites/files/kf/field/field_publication_file/delivering-better-services-for-people-with-long-term-conditions.pdf)
- Covinsky, K. E., Rosenthal, G. E., Chren, M. M., Justice, A. C., Fortinsky, R. H., Palmer, R. M., & Landefeld, S. C. (1998). The Relation Between Health Status Changes and Patient Satisfaction in Older Hospitalized Medical Patients. *Journal of General Internal Medicine, 13*(4), 223-229. doi: 10.1046/j.1525-1497.1998.00071.x
- Coward, T. J., Scott, B. J. J., Watson, R. M., & Richards, R. (2007). A comparison of prosthetic ear models created from data captured by computerized tomography, magnetic resonance imaging, and laser scanning. *International Journal of Prosthodontics, 20*(3), 275 - 285.

- Creswell, J. (1998). *Qualitative inquiry and research design: Choosing among five traditions*. Thousand Oaks, CA: Sage.
- Creswell, J. W. (2012). *Qualitative Inquiry and Research Design: Choosing Among Five Approaches*. London: Sage Publications Inc.
- Dahlberg, K., Drew, N., & Nyström, M (2001). *Reflective lifeworld research*. Lund: Sweden: Studentlitteratur.
- Danz, W. S. (1990). Ancient and contemporary history of artificial eyes. *Advances in ophthalmic plastic and reconstructive surgery*, 8, 1 - 10. from <http://www.ncbi.nlm.nih.gov/pubmed/2248697>
- Davis, K., Drey, N., & Gould, D. (2009). What are scoping studies? A review of the nursing literature. *Nursing Studies*, 46(10), 1386 - 1400. doi: 10.1016/j.ijnurstu.2009.02.010
- Davison, T. L. (2014). Phenomenological Research Using a Staged Multi-Design Methodology. *International Journal of Business, Humanities and Technology*, 4(2).
- de Melo Filho, M. R., Rocha, B. A., de Oliveira Pires, M. B., Fonseca, E. S., de Freitas, E. M., Junior, H. M., & Santos, F. B. G. (2013). Quality of life and self-image in patients with head and neck cancer. *Universitas Psychologica*, 11(1), 13-23.
- Deacon, B. S. (2008). Orbital Implants and Ocular Prostheses: A Comprehensive Review. *Journal of Ophthalmic Medical Technology*. from <http://www.jomtonline.com/jomt/articles/volumes/4/2/orbital.pdf>
- Delhiwala, K. S., Vadakkal, I. P., Mulay, K., Khetan, V., & Wick, M. R (2016). Retinoblastoma: An update. *Seminars in Diagnostic Pathology*, 33(3), 133 - 140. doi: 10.1053/j.semmp.2015.10.007
- de Jong, M. C., Kors, W. A., de Graaf, P., Castelijns, J. A., Kivelä, T., & Moll, A. C. . (2014). Trialateral retinoblastoma: a systematic review and meta-analysis. *The Lancet Oncology*, 15(10), 1157 - 1167. doi: 10.1016/S1470-2045(14)70336-5
- DeLong, C. M., Au, W. W., & Stamper, S. A. (2007). Echo features used by human listeners to discriminate among objects that vary in material or wall thickness: implications for echolocating dolphins. *Journal of the Acoustical Society of America*, 121, 605 - 617. doi: 10.1121/1.2400848

- Desmonds, D., & MacLachlan, M. (2002). *Psychosocial Issues in the Field of Prosthetics and Orthotics, 14(1)*, 19 - 22. *Journal of Prosthetics and Orthotics, 14(1)*, 19 - 22. doi: 10.1097/00008526-200203000-00006
- Deno, M., Tashiro, M., Miyashita, M., Asakage, T., Takahashi, K., Saito, K., Busuijma, Y., Mori, Y., Saito, H., & Ichikawa, Y. (2012). The mediating effects of social support and self-efficacy on the relationship between social distress and emotional distress in head and neck cancer outpatients with facial disfigurement. *Psycho-oncology, 21(2)*:144-152.
- de Vries, M., & Friedrich, S. (2014). *Psycho-oncological interventions and psychotherapy in the oncology setting*. In U. Goerling (Ed.), *Psycho-Oncology* (pp. 121-135). New York: Springer-Verlag Publishing
- Deshields, T. L., & Nanna, S. K. (2010). Providing Care for the "Whole Patient" in the Cancer Setting: The Psycho-Oncology Consultation Model of Patient Care. *Clinical Psychology in Medical Settings, 17*, 249-257. doi: 10.1007/s10880-010-9208-1
- Dixit, S., Shetty, P., & Bhat, G. S (2005). Ocular prosthesis in Children - Clinical Report. *Kathmandu University Medical Journal, 3(1)*, 81 - 83.
- Draucker, C. B. (1999). The critique of Heideggerian hermeneutical nursing research. *Journal of Advanced Nursing, 30(2)*, 360 - 373. doi: 10.1046/j.1365-2648.1999.01091.x
- Dufour, A., Després, O., & Candas, V. (2005). Enhanced sensitivity to echo cues in blind subjects. *Brain Research, 165(4)*, 515 - 519. doi: 10.1007/s00221-005-2329-3
- Dyer, M. A. (2016). Lessons from Retinoblastoma: Implications for Cancer, Development, Evolution, and Regenerative Medicine. *Trends in Molecular Medicine, 22(10)*, 863 - 876. doi: 10.1016/j.molmed.2016.07.010
- Dyer, N. A. (1980). The Artificial Eye. *Australian Journal of Ophthalmology, 8(4)*, 325 - 327. doi: 10.1111/j.1442-9071.1980.tb00293.x
- Eatough, V., Smith, J. A., & Shaw, R. (2008). Women, anger, and aggression an interpretative phenomenological analysis. *Journal of Interpersonal Violence, 23(12)*, 1767 - 1799.
- Eggbeer, D. (2008). *The Computer Aided Design and Fabrication of Facial Prostheses (Thesis)*. University

of Wales Institute, Cardiff. Retrieved from [https://dspace.lboro.ac.uk/dspace-jspui/bitstream/2134/6419/1/Rapid\\_prototyping\\_technologies\\_in\\_soft\\_tissue\\_facial\\_prosthetics\\_current\\_state\\_of\\_the\\_art%5B1%5D.pdf](https://dspace.lboro.ac.uk/dspace-jspui/bitstream/2134/6419/1/Rapid_prototyping_technologies_in_soft_tissue_facial_prosthetics_current_state_of_the_art%5B1%5D.pdf)

- Ek, U. (2000). Emotional reactions in parents and children after diagnosis and treatment of a malignant tumour in the eye. *Child: Care, Health and Development*, 26(5), 415 - 428. doi: 10.1046/j.1365-2214.2000.00159.x
- Elliot, R., Fischer, C. T., & Rennie, D. L. (1999). Evolving guidelines for publication of qualitative research studies in psychology and related fields. *British Journal of Clinical Psychology*, 38, 215 - 229.
- Emmel, N., Hughes, K., Greenhalgh, J., & Sales, A. (2007). Developing methodological strategies to recruit and research socially excluded groups. *Sociological Research Online*, 12(2). doi: 10.5153/sro.1512
- Englander, M. (2012). The Interview: Data Collection in Descriptive Phenomenological Human Scientific Research. *Journal of Phenomenological Psychology*, 43, 13 - 35.
- Erpf, S. F. (1953). Comparative Features of Plastic and/or Glass in Artificial Eye Construction. *Archives of Ophthalmology*, 50, 737.
- Evans, P., Eggbeer, D., & Bibb, R. (2004). Orbital Prosthesis Wax Pattern Production Using Computer Aided Design and Rapid Prototyping Techniques. *Journal of Maxillofacial Prosthetics & Technology*, 7, 11 – 15
- Eysenbach, G., & Wyatt, J. (2002). Using the Internet for Surveys and Health Research. *Journal of Medical Internet Research*, 4(2), e13. doi: 10.2196/jmir.4.2.e13
- Fink, A. (2003). *The survey handbook*. Thousand Oaks, CA: Sage
- Finlay, L. (2008). Reflecting on 'Reflective practice' Retrieved June 20th, 2017, from Commissioned discussion paper) accessible from: <http://www.open.ac.uk/cetl->

- Fu, L., Bundy, C., & Sadiq, S. A. (2011) Psychological distress in people with disfigurement from facial palsy. *Eye*, 25(10):1322-1326
- Gadmer, H. G. (1988). *Truth and Method*. London: Continuum.
- Gallagher, P., & MacLachlan, M. (1999). Psychological adjustment and coping in adults with prosthetic limbs. *Behavioral Medicine*, 25(3), 117 - 124.
- Ganz, P. A. (2014). Cancer Survivors: A Look Backward and Forward. *Journal of Oncology Practice*, 10(5). doi: 10.1200/JOP.2014.001552
- Garhart , C., & Lakshminarayanan, V. (2015). Anatomy of the Eye. In J. Chen, Cranton, W., & Fihn, M (Ed.), *Handbook of Visual Display Technology* (pp. 1-10). USA: Springer Berlin Heidelberg.
- Garg, P., Garg, S., Bansal, D., & Suresh, S (2012). Prosthetic rehabilitation of a patient with enucleated eye - a case report. *Nepalese Journal of Ophthalmology*, 4(8), 312- 314. doi: 10.3126/nepjoph.v4i2.6550
- Gilbert, C. D., & Wiesel, T. N. (1992). Receptive field dynamics in adult primary visual cortex., *Nature*, 356, 150 - 152. doi: 10.1038/356150a0
- Gillham, B. (2000). *Developing a questionnaire (real world research)*. London: Continuum.
- Glaser, B. G., & Strauss, A. L. (1967). *The Discovery of Grounded Theory: Strategies for Qualitative Research*. Chicago: Aldine Publishing Company.
- Goel, G. K., Jain, D., Goel, D., & Juneja, P. (2012). Rehabilitation after surgical treatment for retinoblastoma: ocular prosthesis for a 6-month-old child. *J Prosthodont*, 21(5), 408-412. doi: 10.1111/j.1532-849X.2011.00840.x
- Goiato, M. C., Bannwart, L. C., Haddad, M. F., dos Santos, D. M., Pesqueirs, A. A., & Miyahara, G. I. (2014). Fabrication Techniques for Ocular Prostheses - An Overview. *Orbit*, 33(3), 229 - 233. doi: 10.3109/01676830.2014.881395



- Goiato, M. C., dos Santos, D. M., Moreno, A., Haddad, M., & Turcio, K. H. L. (2013). An Alternate Impression Technique for Ocular Prostheses. *Journal of Prosthodontics*, 22(4), 338 - 340. doi: 10.1111/j.1532-849X.2012.00945.x
- González, E. G., Steeves, J. K. E., & Steinbach, M. J. (1998). Perceptual learning for motion- defined letters in unilaterally enucleated observers and monocularly viewing normal controls. *Investigative Ophthalmology & Visual Science*, 39 S400
- González, E. G., Steeves, J. K. E., Kraft, S. P., Gallie, B. L., & Steinbach, M. J. (2002). Foveal and eccentric acuity in one-eyed observers. *Behavioural Brain Research*, 128(1), 71 - 80. doi: 10.1016/S0166-4328(01)00310-2
- González, E. G., Steinbach, M. J., Ono, H. & Wolf, M. (1989). Depth perception in humans enucleated at an early age. *Clin. Vis. Sci.*, 4, 173 – 177
- Gougoux, F., Zatorre, R. J., Lassonde, M., Voss, P., & Lepore, F. (2005). A Functional Neuroimaging Study of Sound Localization: Visual Cortex Activity Predicts Performance in Early-Blind Individuals. *PLoS Biology*, 3(2), 0324 - 0333. doi: 10.1371/journal.pbio.0030027
- Graneheim, U. H., & Lundman, B. (2004). Qualitative content analysis in nursing research: concepts, procedures and measures to achieve trustworthiness. *Nurse Education Today*, 24(2), 105 – 112
- Greene, J., & McClintock, C. (1985). Triangulation in evaluation. *Evaluation Review*, 9(5), 523 - 545.
- Griffin, D. R. (1944). Echolocation by blind men, bats and radar. *Science*, 100(2609), 589 - 590. doi: 10.1126/science.100.2609.589
- Guest, G., Bunce, A., & Johnson, L. . (2006). "How many interviews are enough? An experiment with data saturation and variability". *Field Methods*, 18(1), 59-82.
- Gupta, R. K., & Padmanabhan, T. V. (2012). Prosthetic Rehabilitation of a Post Evisceration Patient with Custom Made Ocular Prosthesis: A Case Report. *The Journal of Indian Prosthodontic Society*, 12(2), 108 - 112. doi: 10.1007/s13191-012-0115-z
- Gutfleisch, O. (2003). Peg legs and bionic limbs: The development of lower extremity prosthetics. *Interdisciplinary Science Reviews*, 28(2), 139 - 148. doi: 10.1179/030801803225010368
- Hamama-Raz, Y., Rot, I., & Buchbinder, E. (2012). The Coping Experience of Parents of a Child with

Retinoblastoma-malignant Eye Cancer. *Journal of Psychosocial Oncology*, 30(1), 21 - 40. doi: 10.1080/07347332.2011.633977

Hamilton, R. H., Pascual-Leone, A., & Schlaug, G. (2004). Absolute pitch in blind musicians. *Neuroreport*, 15(5), 803 - 806. doi: 10.1097/01.wnr.0000118981.36602.90

Heffner, H. E., & Heffner, R. S. (2005). The sound-localization ability of cats. *Journal of Neurophysiology*, 94(5), 3653 - 3655. doi: 10.1152/jn.00720.2005

Heidegger, M. (1927). *Being and Time*. London: Harper Perennial Modern Classic.

Hernandez, J. C., Brady, L. W., Shields, J. A., Shields, C. L., DePotter, P., Karlsson, U. L., Markoe, A. M., Amendola, B. E., & Singh, A. (1995). External beam radiation for retinoblastoma: Results, patterns of failure, and a proposal for treatment guidelines. *International Journal of Radiation Oncology, biology, physics*, 35(1), 125 - 132. doi: 10.1016/S0360-3016(96)85020-6

Hill, J. C., Whitehurst, D. G., Lewis, M., Bryan, S., Dunn, K. M., Foster, N. E., Konstantinou, K., Main, C. J., Mason, E., Somerville, S., Sowden, G., Vohora, K., & Hay, E. M. (2011). Comparison of stratified primary care management for low back pain with current best practice (STarT Back): a randomised controlled trial. *Lancet*, 378(9802), 1560 - 1571. doi: 10.1016/S0140-6736(11)60937-9

Hinchcliffe, V., & Gavin, H (2009). Social and virtual networks: Evaluating synchronous online interviewing using instant messenger. *Qualitative Report*, 14(2), 318 - 340.

Ho, C. S., Giaschi, D. E., Boden C., Dougherty, R., Cline, R., & Lyons, C. (2005). Deficient motion perception in the fellow eye of amblyopic children. *Vision Research*, 45(12), 1615 - 1627. doi: 10.1016/j.visres.2004.12.009

Holloway, L., & Tordes, L. (2003). The Status of Method: Flexibility, Consistency and Coherence. *Qualitative Research*, 3(3), 345 - 357. doi: 10.1177/1468794103033004

Holm, K. E., Patterson, J. M., & Gurney, J. G. (2003). Parental involvement and family-centred care in the diagnostic and treatment phases of childhood cancer: Results from a qualitative study. *Journal of Pediatric Oncology Nursing*, 20(6), 301 - 313.

Hoover, A. E. N., Harris, L. R., Steeves, J. K. E. (2012). Sensory compensation in sound localization in people with one eye. *Experimental Brain Research*, 216(4), 565 - 574. doi: 10.1007/s00221-011-2960-0

- Horgan, O., & MacLachlan, M. (2004). Psychosocial adjustment to lower-limb amputation: a review. *Disability and Rehabilitation, 26*(14/15), 837 - 850. doi: 10.1080/09638280410001708869
- Houghton, C., Casey, D., Shaw, D., & Murphy, K. (2013). Rigour in qualitative case-study research. *Nurse Researcher, 20*(4), 12 – 17
- Houtzager, B. A., Grootenhuis, M. A., & Last, B. F. (1999). Adjustment of siblings to childhood cancer: a literature review. *Support Care Cancer, 75*(5), 302 - 320. doi: 10.1111/j.1399-5448.2008.00385.x
- Howitt, D. (2010). *Introduction to Qualitative Methods in Psychology*. London: Pearsons Education Limited.
- Huang, D., Zhang, Y., Zhang, W., Wang, Y., Zhang, P., Hong, L., . . . Zhi, T. (2013). Study on clinical therapeutic effect including symptoms, eye preservation rate, and follow-up of 684 children with retinoblastoma. *European Journal of Ophthalmology, 23*(4), 532-538. doi: 10.5301/ejo.5000245
- Hubel, D. H., & Wiesel, T. N. (1962). Receptive fields binocular interaction and functional architecture in the cat's visual cortex. *Journal of Physiology, 160*(1), 106 - 154. doi: 10.1113/jphysiol.1962.sp006837/pdf
- Hubel, D. H., Wiesel, T. N. & LeVay, S. (1977). Ilasticity of ocular dominance columns in monkey striate cortex. *Philosophical Transactions of the Royal Society of London . Series B, Biological Science, 278*(961), 377 - 409. doi: 10.1098/rstb.1977.0050
- Hughes, B. (2001). Active artificial echolocation and the nonvisual perception of aperture passability. *Human Movement Science, 20*, 371 - 400. doi: 10.1016/S0167-9457(01)00059-8
- Husserl, E. (1970). *The idea of phenomenology*. Nijhoff, Netherlands: The Hague.
- Israel Cancer Association. (2016). *Quality of Life in Children Cured of Retinoblastoma*: Hadassah Medical Organization.
- Jack, K., McLean, S., Moffett, J. K., & Gardiner, E (2010). Barriers to treatment adherence in physiotherapy outpatient clinics: A systematic review. *Manual Therapy, 15*(3-2), 220 - 228.
- Jacobsen, P. B., Donovan, K. A., Trask, P. C., Fleishman, S. B., Zabora, J., Baker, F., Holland, J. C. (2005). Screening for psychologic Distress in Ambulatory Cancer Patients: A multicenter Evaluation of the Distress Thermometer. *Cancer, 103*, 1494-1502. doi: 10.1002/cncr.20940
- Jain, S., Makkar, S., Gupta, S., & Bhargava, A. (2010). Prosthetic Rehabilitation of Ocular Defect Using Digital Photography: A Case Report. *Journal of the Indian Prosthodontic Society, 10*(3), 190 -

- James, H., Jenkinson, E., Harrad, R., Ezra, D. G., Newman, S., & Thompson, A. R. (2011). Appearance concerns in ophthalmic patients. *Eye*, 25(8):1039–1044
- Jansen, H. (2010). The Logic of Qualitative Survey Research and its Position in the Field of Social Research Methods. *Qualitative Social Research*, 11(2)
- Joinson, A. (2005). Internet behaviour and the design of virtual methods. In C. Hine (Ed.), *Virtual methods: Issues in social research on the Internet* (pp. 21 - 34). Oxford: Berg.
- Jones, G. (2005). Echolocation. *Current Biology*, 15(13), 484 - 488. doi: 10.1016/j.cub.2005.06.051
- Jurbergs, N., Long, A., Ticona, L., & Phipps, S. (2009). Symptoms of posttraumatic stress in parents of children with cancer: Are they elevated relative to parents of healthy children? *Journal of Pediatric Psychology*, 34(1), 4-13.
- Kahn, D. M., & Krubitzer, L. (2002). Massive cross-modal cortical plasticity and the emergence of a new cortical area in developmentally blind mammals. *Proceedings of the National Academy of the United States of America*, 99(17), 11429 - 11434. doi: 10.1073/pnas.162342799
- Kenny, A. J. (2005). Interaction in cyberspace: an online focus group. *Journal of Advanced Nursing*, 49(4), 414 - 422.
- Kairuz, T., Crump, K., & O'Brien. (2007). Tools for data collection and analysis. *The Pharmaceutical Journal*, 278, 371 - 377.
- Karcioglu, Z. A., Abboud, E. B., Al-Mesfer, S. A., Al-Rashed, W., & Pilapil, D. H. (2002). Retinoblastoma in older children. *Journal of American Association for Pediatric Ophthalmology and Strabismus*, 6(1) 26 - 32.
- Karns, C. M., Dow, M. W., & Neville, H. J. (2012). Altered Cross-Modal Processing in the Primary Auditory Cortex of Congenitally Deaf Adults: A Visual-Somatosensory fMRI Study with a Double-Flash Illusion. *Journal of Neuroscience*, 32(28), 9626 - 9638. doi: 10.1523/JNEUROSCI.6488-11

- Kellogg, W. N. (1962). Sonar system of the blind. *Science*, *137*(3528), 399 - 404. doi: 10.1126/science.137.3528.399
- Kelley, J. J. (1970). History of Ocular Prostheses. *international Ophthalmology Clinics*, *10*(4), 713 - 719.
- Kelly, K. R., Moro, S. S., & Steeves, J. K. E (2013). Living with One Eye: Plasticity in Visual and Auditory Systems. in Steeves, J. K. E., & Harris, L. R. *Plasticity in Sensory Systems*, (pp. 94 - 111). New York: Cambridge University Press.
- Kim, Y. (2011). The Pilot Study in Qualitative Inquiry: Identifying Issues and Learning Lessons for Culturally Competent Research. *Qualitative Social Work*, *10*(2).
- King, G., King, S., Rosebaum, P., & Goffin, R. (1999). Family-centred caregiving and well-being of parents of children with disabilities: linking process with outcome. *Journal of Pediatric Psychology*, *24*(1), 41 - 53. doi: 10.1093/jpepsy/24.1.41
- Kiss, S., Leiderman, Y. I., & Mukai, S. (2008). Diagnosis, Classification, and Treatment of Retinoblastoma. *international Ophthalmology Clinics*, *48*(2), 135 - 147. doi: 10.1097/IIO.0b013e3181693670
- Klassen, A. F., Klaassen, R., Dix, D., Pritchard, S., Yanofsky, R., O'Donnell, M., Scott, A., & Sung, L. (2008). Impact of Caring for a Child With Cancer on Parents' Health-Related Quality of Life. *Journal of Clinical Oncology*, *26*(36), 5884 - 5889. doi: 10.1200/JCO.2007.15.2835
- Knudson, A. G., Hethcote, H. W., & Brown, B. W. (1975). Mutation and childhood cancer. A probabilistic model for the incidence of retinoblastoma. *Proceedings of the National Academy of Sciences*, *72*(12), 5116-5120.
- Koch, T. (1995). An interpretive process: revisiting phenomenological and hermeneutical approaches. *Nurse Research*, *6*(3), 20 - 34.
- Kolarik, A. J., Cirstea, S., Pardhan, S., & Moore, B. C. J. (2014). A summary of research investigating echolocation abilities of blind and sighted humans. *Hearing Research*, *310*, 60 - 68. doi: 10.1016/j.heares.2014.01.010
- Korte, M., & Rauschecker, J. P. (1993). Auditory spatial tuning of cortical neurons is sharpened in cats with early blindness. *Journal of Neurophysiology*, *70*(4), 1717 - 1721.
- Kratz, K. E., & Spear, P. D. (1976). Effects of visual deprivation and alterations in binocular competition on responses of striate cortex neurons in the cat. *Journal of Comparative Neurology*, *170*(2), 141 - 151. doi: 0.1002/cne.901700202/pdf

- Kvale, S. (1996). *Interviews: An Introduction To Qualitative Research Interviewing*. Thousand Oaks, CA: Sage.
- Langdridge, D. (2007). *Phenomenological Psychology: Theory, Research and Method*. Edinburgh: Pearson Education Limited
- Larkin, M., & Thompson, A. (2012). Interpretative Phenomenological Approach *Qualitative research methods in mental health and psychotherapy: a guide for students and practitioners* (pp. 99 - 116). Oxford: John Wiley & Sons.
- Larkin, M., Watts, S., & Clifton, E. (2006). Giving voice and making sense in interpretative phenomenological analysis. *Qualitative research in psychology*, 3(2), 102 - 120.
- Last, B. F., Grootenhuis, M. A., & Eiser, C. (2005). International Comparison of Contributions to Psychosocial Research on Survivors of Childhood Cancer: Past and Future Considerations. *Journal of Pediatric Psychology*, 30(1), 99 - 113. doi: 10.1093/jpepsy/jsi021
- LeGrand, J. A. J., & Hughes, M. O. (1990). Empirical Impression Technique for Artificial Eye Fitting. *Advances in Ophthalmic Plastic and Reconstructive Surgery*, 8, 118 - 125.
- Lekay-Adams, M., Sykes, L. M., & du Plessis, F. (2014). Prosthetic challenges in a patient with ocular evisceration and global preservation: a case report. *South African Dental Journal*, 69(1), 15 - 17. from <http://www.scielo.org.za/pdf/sadj/v69n1/08.pdf>
- Leander, C., Fu, L. C., Pena, A., Howard, S. C., Rodriguez-Galindo, C., Wilimas, J. A., . . . Haik, B. (2007). Impact of an education program on late diagnosis of retinoblastoma in Honduras. *Pediatr Blood Cancer*, 49(6), 817-819. doi: 10.1002/pbc.21052
- Lessard, N., Pare, M., Lepore, F., & Lassonde, M. (1998). Early-blind human subjects localize sound sources better than sighted subjects. *Nature*, 395, 278 - 280. doi: 10.1038/26228
- LeVasseur, j. J. (2003). The Problem of Bracketing in Phenomenology. *Qualitative Health Research*, 13(3).
- Lewis, T. L., Ellemberg, D., Maurer, D., Wilkinson, F., Wilson, H. R., Dirks, M., & Brent, H. P. (2002). Sensitivity to global form in glass patterns after early visual deprivation in humans. *Vision Research*, 42(8), 939 - 948. doi: 10.1016/S0042-6989(02)00041-X
- Li, S., Xiao, C., Duan, L., Fang, C., Huang, Y., & Wang, L. (2015). CT Image-based Computer-Aided

System for Orbital Prosthesis Rehabilitation. *Medical & Biological Engineering & Computing*.  
doi: 10.1007/s11517-015-1307-6

Lincoln, Y. S., & Guba, E. G. (1985). *Naturalistic Inquiry*. Newbury Park, CA: Sage Publications.

Lowental, U., & Sela, M. (1982). Evaluating cosmetic results in maxillofacial prosthetics. *Journal of Prosthetic Dentistry*, 48(5), 567 – 570

MacCarthy, A., Bayne, A. M., Brownbill, P. A., Bunch, K. J., Diggens, N. L., Draper, G. J., . . . Murphy, M. F. (2013). Second and subsequent tumours among 1927 retinoblastoma patients diagnosed in Britain 1951-2004. *Br J Cancer*, 108(12), 2455-2463. doi: 10.1038/bjc.2013.228

MacCarthy, A., Bayne, A. M., Draper, G. J., Eatock, E. M., Kroll, M. E., Stiller, C. A., . . . Murphy, M. F. (2009). Non-ocular tumours following retinoblastoma in Great Britain 1951 to 2004. *Br J Ophthalmol*, 93(9), 1159-1162. doi: 10.1136/bjo.2008.146035

Madill, A., & Gough, B. (2008). Qualitative research and its place in psychological science. *Psychological Methods*, 13(3), 254 - 271.

Maggs-Rapport, F. (2000). Combining methodological approaches in research: ethnography and interpretive phenomenology. *Journal of Advanced Nursing*, 31(1), 219 - 225.

Mann, C., & Stewart F (2000). *Internet communication and qualitative research*. London: Sage Publications.

Manoj, S. S. (2014). Modified Impression Technique for Fabrication of a Custom Made Ocular Prosthesis. *Anaplastology*, 3(2), 129. doi: 10.4172/2161-1173.1000129

Manne, S., Myers-Virtue, S., Kashy, D., Ozga, M., Kissane, D., Heckman, C., Rubin, S. C., & Rosenblum, N. (2015). Resilience, positive coping, and quality of life among women newly diagnosed with gynecological cancers. *Cancer Nursing*, 38(5), 375 - 382. doi: 10.1097/NCC.0000000000000215

Mansouri, B., & Hess, R. F. (2006). The global processing deficit in amblyopia involves noise segregation. *Vision Research*, 46(24), 4104 - 4117. doi: 10.1016/j.visres.2006.07.017

- Marotta, J. J., Perrot, T. S., Nicolle, D. & Goodale, M. A. (1995). The development of adaptive head movements following enucleation. *Eye*, 9(3), 333 - 336. doi: 10.1038/eye.1995.64.
- Martin, J. L., Murphy, E, Crowe J. A., & Norris B. J. (2006). Capturing User requirements in Medical Device Development: The Role of Ergonomics. *Physiological Measurement*, 27, 49 = 62.
- Martin, O., & Clouds, L. (1979). The history of the artificial eye. *Annual of Plastic Surgery*, 3(2), 168. doi: 10.1097/00000637-197908000-00014
- Martin, J. L., Murphy, E, Crowe J. A., & Norris B. J. (2006). Capturing User requirements in Medical Device Development: The Role of Ergonomics. *Physiological Measurement*, 27, 49 = 62.
- Masnari, O., Sxhiestl, C., Rossler, J., Gutlein, S. K., Neuhaus, K., Weibel, L., Meuli, M., & Landolt, M. A. (2013). Stigmatization Predicts Psychological Adjustment and Quality of Life in Children and Adolescents With a Facial Difference. *Journal of Paediatric Psychology*, 38(2), 162-172.
- Mason, J. (2002). *Qualitative Researching*. London: Sage Publications.
- Matthews, M. F., Smith, R. M., Sutton, A. J., & Hudson, R. (2000). The Ocular Impression: A Review of the Literature and Presentation of an Alternate Technique. *Journal of Prosthodontics*, 9(4), 210 - 216. doi: 10.1053/jpro.2000.21157
- Mays, N., Roberts, E. and Popay, J. (2001). Synthesising research evidence *Studying the Organisation and Delivery of Health Services: Research Method* (pp. 188 - 220). London: Routledge.
- McBain, H. B., Ezra, D. G., Rose, G. E., & Newman, S. P. (2014). The psychosocial impact of living with an ocular prosthesis. *Orbit*, 33(1), 39 - 44. doi: 10.3109/01676830.2013.851251
- McCann, T. V., & Clark, E. (2003). Grounded theory in nursing research: Part 1-- Methodology. *Nurse Researcher*, 11(2), 7 - 18.
- McClellan, W., Klemp, J. R., Krebill, H., Ryan, R., Nelson, E. L., Paincker, J., Sharma, M., & Stegenga, K. (2013). Understanding the Functional Late Effects and Informational Needs of Adult Survivors of Childhood Cancer. *Oncology Nursing Forum*, 40(3), 254 - 262. doi: 10.1188/13.ONF.254-262



- McColl, E., & Thomas, R. (2000). *The Use and Design of Questionnaires*. England: The Royal College of General Practitioners.
- McCord, C. P. (1965). Artificial Eyes: The Early History of Ocular Prostheses. *The Journal of Gnathology*, 85, 61 - 68.
- McDaid, C., Hartley, S., Bagnall, A. M., Ritchie, G., Light, K., & Riemsma, R. (2005). *Systematic review of effectiveness of different treatments for childhood retinoblastoma*. Retrieved October 5, 2014.
- McKee, S. P., Levi, D. M. & Movshon, J. A. (2003). The pattern of visual deficits in amblyopia. *Journal of Vision*, 3(5), 380 - 405. doi: 10.1.1.148.7398
- McKenna, K., Collier, J., Hewitt, M., & Blake, H. (2010). Parental involvement in paediatric cancer treatment decisions. *European Journal of Cancer Care*, 19(5), 621 - 630. doi: 10.1111/j.1365-2354.2009.01116.x
- McLeod, J. (2001). *Qualitative Research in Counselling and Psychotherapy*. London: Sage.
- Medical Research Council. (2000). A Framework for Development and Evaluation of RCTs for Complex Interventions to Improve Healthcare. Retrieved October 31, 2016, from <https://www.mrc.ac.uk/documents/pdf/rcts-for-complex-interventions-to-improve-health/>
- Meel, R., Radhakrishnan, V., & Bakhshi, S. (2012). Current therapy and recent advances in the management of retinoblastoma. *Indian J Med Paediatr Oncol*, 33(2), 80-88. doi: 10.4103/0971-5851.99731
- Mellblom, A. V., Finset, A., Korsvold, L., Loge, J. H., Ruud, E., & Lie, H. C. (2014). Emotional concerns in follow-up consultations between paediatric oncologists and adolescent survivors: a video-based observational study. *Psycho-Oncology*, 23, 1365 - 1372. doi: 10.1002/pon.3568
- Merabet, L. B., Hamilton, R., Schlaug, G., Swisher, J. D., Kiriakopoulos, E. T., Pitskel, N. B., Kauffman, T., & Pascual-Leone, A. (2008). Rapid and Reversible Recruitment of Early Visual Cortex for Touch. *PLoS One*, 3(8), 1 - 12. doi: 10.1371/journal.pone.0003046
- Messinger, S. D. (2009). Incorporating the prosthetic: Traumatic, limb-loss, rehabilitation and refigured military bodies. *Disability and Rehabilitation*, 31(25), 2130 - 2134. doi: 10.3109/09638280902943223.

- Miettinen, R., & Hasu, M (2002). Articulating User Needs in Collaborative Design: Towards an Activity-Theoretical Approach. . *Computer Supported Cooperative Work*, 11, 129 - 151.
- Milne, J. L., Goodale, M. A., & Thaler, L. (2014). The role of head movements in the discrimination of 2-D shape by blind echolocation experts. *Attention Perception & Psychophysics*, 76(6), 1828 - 1837. doi: 10.3758/s13414-13014-10695-13412
- Moidell, B., Steinbach, M. J. & Ono, H. (1988). Egocenter location in children enucleated at an early age. *Investigative Ophthalmology & Visual Science*, 29(8), 1348 - 1351.
- Moorse, J. M. (2000). Determining Sample Size. *Qualitative Health Research*, 10(1), 3 - 5.
- Moran, D. (2000). *Introduction to phenomenology*. London: Routledge.
- Mouratova, T. (2005). Triateral retinoblastoma: a literature review 1971-2004. *The Bulletin of the Belgian Society of Ophthalmology*, 297. from [file:///Users/Holly/Downloads/297-03%20\(1\).pdf](file:///Users/Holly/Downloads/297-03%20(1).pdf)
- Nancarrow, S. A., Booth, A., Ariss, S. A., Smith, T., Enderby, P., & Roots, A. (2013). Ten principles of good interdisciplinary team work. *Human Resource Health*, 11 - 19. doi: 10.1186/1478-4491-11-19
- Nagayoshi, M., Hirose, T., Toju, K., Suzuki, S., Okamitsu, M., Omori, T., Kawamura, A., & Takeo, N (2016). Parenting stress related to raising infants receiving treatment for retinoblastoma *Psycho-Oncology*, 25(12), 1507 - 1511. doi: 10.1002/pon.4062
- NHS England. (2013). *NHS Standard contract: Retinoblastoma Service (children)*. Retrieved December 10, 2014, from <http://www.england.nhs.uk/wp-content/uploads/2013/06/e04-retino-serv-child.pdf>
- NHS England. (2016). Commissioning Guidance for Rehabilitation. Retrieved 28th July, 2017, from <https://www.england.nhs.uk/wp-content/uploads/2016/04/rehabilitation-comms-guid-16-17.pdf>
- NICE. (2005). *Children's tumours - suspected: Retinoblastoma*. Retrieved September 25, 2014, from <http://cks.nice.org.uk/childrens-tumours-suspected - !scenariorecommendation:8>
- NICE. (2014). Process and methods guide: Developing NICE guidelines: the manual Retrieved 27th July, 2017, from <https://www.nice.org.uk/media/default/about/what-we-do/our-programmes/developing-nice-guidelines-the-manual.pdf>
- Nicholas, J., Heywood, C. A., & Cowey, A. (1996). Contrast sensitivity in one-eyed subjects. *Vision Research*, 36(1), 175 - 180. doi: 10.1016/0042-6989(95)00119-K

- Nodal, F. R., Kacelnik, O., Bajo, V. M., Bizley, J. K., Moore, D. R., & King, A. J. (2010). Lesions of the Auditory Cortex Impair Azimuthal sound Localization and its Recalibration in Ferrets. *Journal of Neurophysiology*, *103*(3), 1209 - 1225. doi: 10.1152/jn.00991.2009
- Oeffinger, A. C., & Wallace, W. H. (2006). Barriers to follow-up care of survivors in the United States and the United Kingdom. *Pediatric Blood and Cancer*, *46*, 135 - 142. doi: 10.1002/pbc.20614
- Olsen, S. E., & Rasmussen, J. (1989). The reflective expert and the prenovice: Notes on skill-, rule- and knowledge-based performance in the setting of instruction and training. In L. Bainbridge, & Quintanilla, S. A. R (Ed.), *Developing skills with information technology* (pp. 9 - 33). Chichester: Wiley: New technologies and work.
- Opendakker, R. (2006). Advantages and Disadvantages of Four Interview Techniques in Qualitative Research. Retrieved 25th July, 2017, from <http://www.qualitative-research.net/index.php/fqs/article/viewArticle/175/391&sa=U&ei=FdsJTdDCGYOnrAer0YjVDg&ved=0CP4BEBYwXg&usg=AFQjCNEsC2J0wILvNuH7LEhQaA2znBkKvw>
- Ortiz, M. V., & Dunkel, I. J. (2016). Retinoblastoma. *Journal of Child Neurology*, *31*(2), 227 - 236.
- Ortlipp, M. (2008). Keeping and Using Reflective Journals in the Qualitative Research Process. *The Qualitative Report*, *13*(4), 695 - 705.
- Osborn, R. L., Demoncada, A. C., & Feuerstein, M. (2006). Psychosocial interventions for depression, anxiety and quality of life in cancer survivors: Meta-analyses. *International Journal of Psychiatry in Medicine*, *36*(1), 13 - 34. doi: 10.2190/EUFN-RV1K-Y3TR-FK0L
- Padmaja, S. (2015). An insight into the future beckons of maxillofacial prosthodontics: Anaplastology. *Journal of Dental Research and Review*, *2*(2), 91 - 96.
- Panton, R. L., Downie, R., Truong, T., MacKeen, L., Kabene, S., Yi, Q. L., Chan, H. S. L., & Gallie, H. (2009). A visual approach to providing prognostic information to parents of children with retinoblastoma. *Psycho-Oncology*, *18*, 300 - 304. doi: 10.1002/pon.1397
- Parker, J. (2005). Nursing Identity and Difference. *Nursing Inquiry*, *12*(2), 65. doi: 10.1111/j.1440-1800.2005.00267.x
- Parver, D. L. (1999). Anatomy of the Eye. *Human Kinetics Journals*, *4*(5), 13-17.
- Pasick, R. J., Burke, N. J., Barker, J. C., Galen, J., Bird, J. A., Otero-Sabogal, R., et al. (2009). *Behavioral theory in a diverse society: Like a compass on Mars*. *Health Education Behavior*, *36*(5), 11S-35S.

- Patenaude, A. F., & Kupst, M. J. (2005). Psychosocial Functioning in Pediatric Cancer. *Pediatric Psychology, 30*(1), 9 - 27. doi: 10.1093/jpepsy/jsi012
- Patenaude, A. F., & Last, B. (2001). Cancer and Children: Where are we coming from? Where are we going? *Psycho-Oncology, 10*, 281 - 283. doi: 10.1002/pon.28518. doi: 10.1002/pon.518
- Patton, M. G. (2002). *Qualitative Research & Evaluation Methods*. Thousand Oaks: SAGE Publications, Inc.
- Peng, Q., Tang, Z., Liu, O., & Peng, Z. (2015). Rapid prototyping-assisted maxillofacial reconstruction. *Annals of Medicine, 47*(186 - 208). doi: 10.3109/07853890.2015.1007520
- Petrus, E., Isaiah, A., Jones, A. P., Li, D., Wang, H., Lee, H. K., & Kanold, P. O. (2014). Crossmodal Induction of Thalamocortical Potentiation Leads to Enhanced Information Processing in the Auditory Cortex. *Neuron, 81*(3), 664 - 673. doi: 10.1016/j.neuron.2013.11.023
- Phillips, W. (1950). The Making and Fitting of Acrylic Resin Artificial Eyes. *The British Journal of Physiological Optics, 7*(4), 217 - 224.
- Pine, K., Sloan, B., Stewart, J., & Jacobs, R. J. (2011). Concerns of anophthalmic patients wearing artificial eyes. *Clinical Experimental Ophthalmology, 39*(1), 47-52.
- Plano Clark, V. L. (2010). The adoption and practice of mixed methods: U.S. trends in federally funded health-related research. *Qualitative Inquiry, 6*(6), 428 - 440.
- Ploeg, J. (1993). Identifying the best research design to fit the question. Part 2: qualitative designs. *Evidence Based Nursing, 2*, 36 - 37. doi: 10.1136/ebn.2.2.36
- Polit, D., & Beck, C. (2006). *Essentials of Nursing Care: Methods, Appraisal and Utilization* (6th ed.). Philadelphia: Lippincott Williams and Wilkins.
- Potter, J., & Wetherall, M. (1987). *Discourse and Social Psychology: Beyond Attitudes and Behaviour*. London: Sage.
- Prithviraj, D. R., Gupta, V., Muley, N., & Suresh, P. (2013). Custom Ocular Prosthesis: Comparison of Two Different Techniques. *Journal of Prosthodontic Research, 57*(2), 129 - 134. doi: 10.1016/j.jpjor.2012.08.006

- Provenzale, J. M., Gururangan, S., & Klinworth, G. (2004). Trilateral retinoblastoma: Clinical and radiological progression. *Journal of the American College of Radiology*, *183*(2), 505 - 511. doi: 10.2214/ajr.183.2.1830505
- Raizada, D., Raizada, K., Naik, M., Murthy, R., Bhaduri, A., & Honavar, S. G. (2011). Custom Ocular Prosthesis in Children: How Often is a Change Required? *Orbit*, *30*(5), 208 - 213. doi: 10.3109/01676830
- Raizada, K., & Rani, D. (2007). Ocular Prosthesis. *Contact Lens and Anterior Eye*, *30*(3), 152 - 162. doi: 10.1016/j.clae.2007.01.002
- Ram, M. B., Grocott, P. R., & Weir, H. C. M. (2007). Issues and Challenges of Involving Users in Medical Device Development. *Health Expectations*, *11*, 63 - 71. doi: 10.1111/j.1369-7625.2007.00464.x
- Ramasubramanian, A., & Shields, C. (2012). *Retinoblastoma*. New Delhi: Jaypee Brothers Medical Publishers Ltd
- Rauschecker, J. P., & Korte, M. (1993). Auditory compensation for early blindness in cat cerebral cortex. *The Journal of Neuroscience*, *13*(10), 4538 - 4548. doi: 10.1111/j.1369-7625.2007.00464.x
- Ray, A., Gombos, D. S., & Vats, T. S. (2012). Retinoblastoma: an overview. *Indian J Pediatr*, *79*(7), 916-921. doi: 10.1007/s12098-012-0726-8
- Reed, M. J., Steeves, J. K. E., Kraft, S. P., Gallie, B. L., & Steinbach, M. J. (1996). Contrast letter thresholds in the non-affected eye of strabismic and unilateral eye enucleated children. *Vision Research*, *36*(18), 3011 - 3018. doi: 10.1016/0042-6989(96)00019-3
- Reid, D., & Reid, F (2005). Online focus groups: An in-depth comparison of computer-mediated and conventional focus group discussions. *International Journal of Market Research*, *47*(2), 131 - 162.
- Reisberg, D. J., & Habakuk, S. W. (1990). A History of Facial and Ocular Prosthetics. *Advances in Ophthalmic Plastic and Reconstructive Surgery*, *8*, 11 - 24.
- Ritchie, J., Lewis, J., McNaughton-Nicholls, C., & Ormston, R. (2003). *Qualitative Research Practice: A Guide for Social Science Students and Researchers*. London: Sage Publications Ltd.

- Ritchie, J., & Spencer, L. (1994). Qualitative data analysis for applied policy research. In R. G. B. A. Bryman. (Ed.), *Analysing Qualitative Data* (pp. 173 - 194). London: Routledge.
- Rodriguez, E. M., Dunn, M. J., Zuckerman, T., Vannatta, K., Gerhardt, C. A., & Compass, B. E. (2011). Cancer-Related Sources of Stress for Children with Cancer and their Parents. *Pediatric Psychology, 37*(2), 185-197. doi: 10.1093/jpepsy/jsr054
- Rojas, J. A. M., Hermosilla, J. A., Montero, R. S., & Espi, P. L. L. (2009). Physical analysis of several organic signals for human echolocation: oral vacuum pulses. *Acta Acustica United Acustic, 95*(2), 325 - 330. doi: 10.3813/AAA.918155
- Roman, F. (1994). The history of artificial eyes. *Journal of Ophthalmology, 78*(3), 222. doi: 10.1136/bjo.78.3.222
- Rosenblum, L. D., Gordon, M. S., & Jarquin, L. (2000). Echolocating distance by moving and stationary listeners. *Ecological Psychology, 12*(3), 181 - 206. doi: 10.1207/S15326969ECO1203\_1
- Ross, L. (2013). Facilitating Rapport through Real Patient Encounters in Health Care Professional Education. *Australasian Journal of Paramedicine, 10*(4).
- Ross, G., Lipper, E. G., Abramson, D., & Preiser, L. (2001). The development of young children with retinoblastoma. *Archives of Pediatrics & Adolescent Medicine, 155*(1), 80 - 83. doi: 10.1001/archpedi.155.1.80
- Rowan, D., Papadopoulos, T., Edwards, D., Holmes, H., Hollingdale, A., Evans, L., & Allen, R. (2013). Identification of the lateral position of a virtual object based on echoes by humans. *Hearing Research, 300*, 56 - 65. doi: 10.1016/j.heares.2013.03.005
- Rushlow, D., Piovesan, B., Zhang, K., Prigoda-Lee, N. L., Marchong, M. N., Clark, R. D., & Gallie, B. L. . (2009). Detection of mosaic Rb1 mutations in families with retinoblastoma. *Human Genome Variation Society, 30*(5), 842 - 851. doi: 10.1002/humu.20940
- Sabol, J. V., Grant, G. T., Liacouras, P., & Rouse, S. (2011). Digital image capture and rapid prototyping of the maxillofacial defect. *Journal of Prosthodontics, 20*(4), 310 - 314. doi: 10.1111/j.1532-849X.2011.00701.
- Sackman, H. (1975). *Delphi Critique*: Lexington Books.
- Sacks, D. (2003). An approach to interviewing adolescents. *Paediatric Child Health, 8*(9), 554 - 556.

- Sadler, G. R., Lee, H. C., Lim, R. S-H., & Fullerton, J (2010). Recruiting hard-to-reach United States population sub-groups via adaptations of snowball sampling strategy. *Nursing and Health Sciences*, 12(3), 369 - 374. doi: 10.1111/j.1442-2018.2010.00541.x
- Salami, A. (2011). *Neuroplasticity in the Auditory Brainstem: From Physiology to the Drug Therapy*. New York: Nova Science.
- Sale, J. E. M., Lohfield, L. H. & Brazil, K. (2002). Revisiting the quantitative-qualitative debate: Implications for mixed-methods research. *Quality & Quantity*, 36, 43 - 53.
- Sapsford, R. (1999). *Survey research*. London: Sage.
- Schmid, L. M., Rosa, M. G. P. & Calford, M. B. (1995). Retinal detachment induces massive immediate reorganization in visual cortex. *Neuroreport*, 6(9), 1349 - 1353 doi: 10.1097/00001756-199506090-00030
- Schneider, R. L. (1986). Modified Ocular Prosthesis Impression Technique. *The Journal of Prosthetic Dentistry*, 55(4), 482 - 485. doi: 10.1016/0022-3913(86)90182-4
- Schörnich, S., Nagy, A., & Wiegrebe, L. (2012). Discovering your inner bat: Echo-acoustic target ranging in humans. *Journal of the Association for Research in Otolaryngology*, 13, 673 - 682. doi: 10.1007/s10162-012-0338-z
- Selistre, S. G. A., Maestri, M. K., Santos-Silva, P., Schüler-Faccini, L., Guimarães, L. S. P., Giacomazzi, J., Evangelista, M. C., & Ashton-Prolla, P (2016). Retinoblastoma in a pediatric oncology. *BMC Pediatrics*, 16(48). doi: 10.1186/s12887-016-0579-9
- Sengupta, S., Pan, U., & Khetan V (2016). Adult onset retinoblastoma. *Indian Journal of Ophthalmology*, 64, 485 - 491.
- Shaikh, S. R., Gangurde, A. P., & Shambharkar, V. I. (2014). Changing Ocular Prostheses in Growing Children: A 5-Year Follow-Up Clinical Report. *The Journal of Prosthetic Dentistry*, 111, 346 - 348.
- Shakespeare, T. (1996 ). Disability, Identity and Difference. In G. Barnes, & Mercer, G (Ed.), *Exploring the Divide: Illness and Disability*. Leeds: Disability Press.
- Shaghghi, A., Bhopal, R. S., & Sheikh A. (2011). Approaches to Recruiting 'Hard-To-Reach' Populations

into Research: A Review of the Literature. *Health Promotion Perspectives*, 1(2), 85 - 94. doi: 10.5681/hpp.2011.009

Sheppard, L., Eiser, C., & Kingston, J. (2005). Mothers' perceptions of children's quality of life following early diagnosis and treatment for retinoblastoma (Rb). *Child: Care, Health & Development*, 31(2), 137 - 142. doi: 10.1111/j.1365-2214.2005.00498.x

Shields, C. L. (2008). Forget-me-nots in the care of children with retinoblastoma. *Semin Ophthalmol*, 23(5), 324-334. doi: 10.1080/08820530802506029

Shields, C. L., & Shields, J. A. (2004). Chemoreduction for retinoblastoma: analysis of tumour control and risks for recurrence in 457 tumors. *Transactions of the American Ophthalmological Society*, 102.

Shields, C. L., Mashayekhi, A., Au, A. K., Czyz, C., Leahey, A., Meadows, A. T., & Shields, J. A. (2006). The International Classification of Retinoblastoma predicts chemoreduction success. *Ophthalmology*, 113(2), 2276 - 2280. doi: 10.1016/j.ophtha.2006.06.018

Shields, C. L., Schoenberg, E., Kocher, K., Shukla, S. Y., Kaliki, S., & Shields, J.A. . (2013). Lesions simulating retinoblastoma (pseudoretinoblastoma) in 604 cases: results based on age at presentation. *Ophthalmology*, 120(2), 311 - 316. doi: 10.1016/j.ophtha.2012.07.067

Shields, C. L., Shields, J. A., & Shah, P. (1991). Retinoblastoma in older children. *Ophthalmology*, 98(3), 395 - 399. doi: 10.1016/S0161-6420(91)32283-8

Shinebourne, P. (2011). Interpretative Phenomenological Analysis. In N. Frost (Ed.), *Qualitative Research Methods in Psychology: Combining core approaches* (pp. 44 - 65): Open University.

Shortell, S. M. (1999). The emergence of qualitative methods in health services research. *Health Services Research*, 34, 1083 - 1090.

Silverman, D. (2000). *Doing Qualitative Research: A Practical Handbook*. Sydney: SAGE Publications Ltd.

Skulmoski, G., Hartman, F., & Krahn, J., (2007). The delphi method for graduate research. *Journal of Information Technology Education*, 6, 1 - 20.



- Smith, J. A., & Eatough, V. (2012). Interpretative Phenomenological Analysis. In G. M. Breakwell, Smith, J. A., & Wright, D. B (Ed.), *Research Methods in Psychology* (pp. 439-461). Thousand Oaks, California: Sage Publications Inc.
- Smith, J. A., & Osborn, M. (2003). Interpretative phenomenological analysis. In J. A. Smith (Ed.), *Qualitative Psychology: A practical guide to research methods*. London: Sage.
- Smith, J., Flowers, P., & Larkin, M. (2009). *Interpretative Phenomenological Analysis: Theory, Method and Research*. London: Sage Publications Ltd.
- Smith, J. A., Flowers, P., & Larkin, M. (2012). *Interpretative Phenomenological Analysis: Theory, Method and Research*. London: Sage Publications Ltd.
- Snell, R. S., & Lemp, M. A. (1998). *Clinical Anatomy of the Eye* (2nd ed.). Oxford: Blackwell Science Ltd.
- Sobrinho, N. A., Garcia de Lima, E., Mattos, M. G., & Watanabe, I. (1986). A Study of Ocular Prostheses. I. Manufacture and Indications for Use. *Revista da Faculdade de Odontologia de Ribeirao Preto*, 23(2), 135 - 143.
- Spradley, J. P. (1979). *The ethnographic interview*. New York: Harcourt Brace Jovanovich College Publishers.
- Starling, N., & Brown, S. (2015). Ocular Prosthetics in the UK: Hope Beyond Trauma. *International Journal of Ophthalmic Practice*, 6(1), 43 - 47.
- Steeves, J. K. E., Gonzales, E. G., & Steinbacg, M. J. (2008). Vision with one eye: a review of visual function following unilateral enucleation. *Spatial Vision*, 21(6), 509 - 529. doi: 10.1163/156856808786451426
- Steeves, J. K. E., González, E. G., Gallie, B. L. & Steinbach, M. J. (2002). Early unilateral enucleation disrupts motion processing. *Vision Research*, 42(1), 143 - 150. doi: 10.1016/S0042-6989(01)00270-X
- Stewart, F. J. (1946). Plastic Artificial Eyes. *Proceedings of the Royal Society of Medicine*, 39(5), 251 - 252.
- Strauss, A., & Corbin, J. (2008). *Basics of qualitative research: Techniques and procedures for developing grounded theory*. Thousand Oaks, CA: Sage.

- Striem-Amit, E., Cohen, L., Dehaene, S., & Amedi, A. (2012). Reading with Sounds: Sensory Substitution Selectively Activates the Visual Word Form Area in the Blind. *Neuron*, 76(3), 640 - 652. doi: 10.1016/j.neuron.2012.08.026
- Stuckey, H. L. (2014). The first step in Data Analysis: Transcribing and managing qualitative research data. *Methodological Issues in Social Health and Diabetes Research*, 2(1), 6 - 8.
- Sweet, C. (2001). Designing and conducting virtual focus groups. *Qualitative Market Research: An International Journal*, 4(3), 130 - 135. doi: 10.1108/13522750110393035
- Tabachnick, B. G., & Fidell, L. S. (2012). *Using Multivariate Statistics* (6th ed.). Boston: Pearson.
- Taddeo, D., Egedy, M., & Frappier, J. Y. (2008). Adherence to treatment in adolescents. *Paediatric Child Health*, 13(1), 19 - 24.
- Teng, S., & Whitney, D. (2011). The acuity of echolocation: spatial resolution in the sighted compared to expert performance. *Journal of Visual Impairment and Blindness*, 105(1), 20 - 32.
- Thaler, L., Arnott, S. R., & Goodale, M. A. (2011). Neural correlates of natural human echolocation in early and late blind echolocation expert. *PLoS One*, 6(5), 20162. doi: 10.1371/journal.pone.0020162
- Thaler, L., & Goodale, M. A. (2016). Echolocation in humans: an overview. *Cognitive Science*, 7(6), 382 - 393.
- Thompson, R. J., Mata, J., Jaeggi, S. M., Buschkuhl, M., Jonides, J., & Gotlib, I. H. (2010). Maladaptive Coping, Adaptive Coping, and Depressive Symptoms: Variations across Age and Depressive State. *Behaviour Research and Therapy*, 48(6), 459 - 466. doi: 10.1016/j.brat.2010.01.007
- Thorsten, G., Isabelle, S., Alexander, E. R., & Roediger, V (2008). Designing and conducting online interviews to investigate interesting consumer phenomena. *Qualitative Market Research: An International Journal*, 11(3), 256 - 274. doi: 10.1108/13522750810879002
- Threader, J., & McCormack, L. (2016). Cancer-related trauma, stigma and growth: the 'lived' experience of head and neck cancer. *European Journal of Cancer Care*, 25(1), 157-169.
- Tuckett, A. (2004). Qualitative Research Sampling-the Very Real Complexities. *Nurse Researcher*, 12(1), 47 - 61.

- Vale, F., Scherzberg, J., Cavaleiro, J., Sanz, D., Caramelo, F., Maló, L., & Marcelino, J. P. (2016). 3D virtual planning in orthognathic surgery and CAD/CAM surgical splints generation in one patient with craniofacial microsomia: a case report. *Dental Press Journal of Orthodontics*, *21*(1), 89 - 100. doi: 10.1590/2177-6709.21.1.089-100.oar
- Valentine, F., & Lowes, L. (2007). *Nursing Care of Children and Young People with Chronic Illness*. Oxford: Blackwell Publishing.
- van Dijk, J., Grootenhuis, M. A., Imhof, S. M., Cohen-Kettenis, P. T., Moll, A. C., & Huisman, j. (2009). Coping strategies of retinoblastoma survivors in relation to behavioural problems. *Psycho-Oncology*, *18*, 1281 - 1289. doi: 10.1002/pon.1507
- van Dijk, J., Oostrom, K. J., Imhof, S. M., Moll, A. C., Schouten-van Meeteren, A. Y. N., Bezemer P. D., & Huisman, J. (2008). Behavioural functioning of retinoblastoma survivors. *Psycho-Oncology*, *18*, 87 - 95. doi: 10.1002/pon.1381
- Van Dongen-Melman, J. E. (2000). Developing Psychosocial Aftercare for Children Surviving Cancer and their Families. *Acta Oncologica*, *39*(1), 23 - 31. doi: 10.1080/028418600430932
- Van Teijlingen, E. R., & Hundley, V. (2001). The importance of pilot studies. Retrieved 24 September, 2016, from <http://sru.soc.surrey.ac.uk/SRU35.html>
- Vance, Y. H., Morse, R. C., Jenney, M.E., & Eiser, C. (2001). Issues in measuring quality of life in childhood cancer: Measures, proxies, and parental mental health. *Journal of Child Psychology and Psychiatry*, *42*(5), 661 - 667. doi: 10.1017/S0021963001007314
- von Blanckenburg, P., Seifart, U., Conrad, N., Exner, C., Rief, W., & Nestoriuc, Y. (2014). Quality of life in cancer rehabilitation: the role of life goal adjustment. *Psycho-Oncology*, *23*, 1149 - 1156. doi: 10.1002/pon.3538
- Voss, P., Lassonde, M., Gougoux, F., Fortin, M., Guillemot, J.P., & Lepore, F. (2004). Early and late-onset blind individuals show supra-normal auditory abilities in farspace. *Current Biology*, *14*(9), 1734 - 1738. doi: 10.1016/j.cub.2004.09.051
- Wallmeier, L., & Wiegrebe, L. (2014). Self-motion facilitates echo-acoustic orientation in humans. *Royal Society Open Science*, *1*(3), 140185 - 140185. doi: 10.1098/rsos.140185

- Walton, J. A., & Madjar, I. (1999). Phenomenology and nursing. In I. Madjar, & J. A., Walton (Ed.), *Nursing and the experience of Illness: Phenomenology in practice* (pp. 1 - 16). London: Routledge.
- Weintraub, N., Rot, I., Shoshani, N., Pe'er, J., & Weintraub, M. (2011). Participation in Daily Activities and Quality of Life in Survivors of Retinoblastoma. *Pediatr Blood Cancer*, *56*, 590 - 594. doi: 10.1002/pbc.22790
- Wiener, L., Viola, A., Koretski, J., Perper, E. D., & Patenaude, A. F. (2014). Pediatric psycho-oncology care: standards, guidelines, and consensus reports. *Psycho-Oncology*, *24*(2), 204-211. doi: 10.1002/pon.3589
- Wiener, I., Oppenheim, D., Breyer, J., Battles, H., Zadeh, S., & Patenaude, A. F. (2012). A worldview of the professional experiences and training needs of pediatric psycho-oncologists. *Psycho-Oncology*, *21*, 944 - 953. doi: 10.1002/pon.3064
- Willard, V. W., Qaddoumi, I., Chen, S., Zhang, H., Brennan, R., Rodriguez-Galindo, C., . . . Phipps, S. (2014). Developmental and adaptive functioning in children with retinoblastoma: a longitudinal investigation. *Journal of Clinical Oncology*, *32*(25), 2788-2793. doi: 10.1200/JCO.2013.53.1996
- Williams, M. (1968). Superior intelligence of children blinded from retinoblastoma. *Archives of Disease in Childhood*, *43*, 204 - 210. doi: 10.1136/adc.43.228.204
- Williams, L. K., McCarthy, M. C., Eyles, D. J., & Drew, S. (2013). Parenting a child with cancer: Perceptions of adolescents and parents of adolescents and younger children following completion of childhood cancer treatment. *Journal of Family Studies*, *19*(1), 80 - 89. doi: 10.5172/jfs.2013.19.1.80
- Willig, C. (2008). *Introducing Qualitative Research in Psychology*. New York: Open University Press.
- Wilson, R. P. (1972). Artificial Eyes in Ancient Egypt. *Survey of Ophthalmology*, *16*(5), 322 - 331.
- Wright, K. B. (2005). Researching Internet-Based Populations: Advantages and Disadvantages of Online Survey Research, Online Questionnaire Authoring Software Packages, and Web Survey Services. *Journal of Computer-Mediated Communication*, *10*(3). doi: 10.1111/j.1083-6101.2005.tb00259.x
- Wu, G., Bi, Y., Zhou, B., Zennick, C., Han, Y., Kong, L., & Zhao, Y. (2009). Computer-Aided Design and Rapid Manufacture of an Orbital Prosthesis. *The International Journal of Prosthodontics*, *22*(3), 293 - 295.

- Xu, X. L., Singh, H. P., Wang, L., Qi, D. L., Poulos, B. K., Abramson, D. H., Jhanwar, S. C., & Cobrinik, D. (2015). Rb suppresses human cone-precursor-derived retinoblastoma tumours. *Nature*, *514*, 385 - 388. doi: 10.1038/nature13813
- Yan, J., Zhang, H., & Li, Y. (2013). Establishment of the relationship between tumor size and range of histological involvement to evaluate the rationality of current retinoblastoma management. *PLoS One*, *8*(11), e80484. doi: 10.1371/journal.pone.0080484
- Yanagisawa, T. (2004). Systemic chemotherapy as a new conservative treatment for intraocular retinoblastoma. *International Journal of Clinical Oncology*, *9*(1), 13-24. doi: 10.1007/s10147-003-0368-y
- Yardley, L., & Bishop, F. (2008). Mixing qualitative and quantitative methods: A pragmatic approach. In C. W. Willig, Stainton-Rogers (Ed.), *The SAGE Handbook of Qualitative Research in Psychology* (pp. 352 - 370). London: Sage Publications Ltd.
- Yun, G. W., & Trumbo, C. W. (2000). Comparative response to a survey executed by post, e-mail, & web form. *Journal of Computer-Mediated Communication*, *6*(1).
- Zebrack, B. J., Corbett, V., Embry, L., Aguilar, C., Meeske, K. A., Hayes-Lattin, B., Block, R., Zeman, D. T., & Cole, S. (2014). Psychological distress and unsatisfied need for psychosocial support in adolescent and young adult cancer patients during the first year following diagnosis. *Psycho-Oncology*, *23*, 1267 - 1275. doi: 10.1002/pon.3533
- Zwiers, M. P., Van Opstal, A. J., & Cruysberg, R. M. (2001). A Spatial Hearing Deficit in Early-Blind Humans. *The Journal of Neuroscience*, *21*, 1 - 5. from <http://www.mbfys.ru.nl/~johnvo/papers/jns2001.pdf>

## Appendices

## Appendix A – Publications

List of publications originating from this thesis

1. Chinnery, H. (2015). Retinoblastoma. In Thompson, S. B. N. (2015) (Eds.), *Psychology of Trauma: Clinical Reviews, Case Histories*.
2. Chinnery, H., Thompson, S. B. N., Noroozi, S., Dyer, B., & Barratt, K (2015). Retinoblastoma: Identifying the Diagnostic Signs for Early Treatment. *International Journal of Neurorehabilitation*, 2(2). Doi: <http://dx.doi.org/10.4172/2376-0281.10001360>
3. Chinnery, H., Thompson, S. B. N. (2015). Sensory Compensation in Children Following Vision Loss after Trauma and Disease. *Journal of Clinical Research and Ophthalmology*, 2(4), 049-053.
4. Chinnery, H., Thompson, S. B. N., Noroozi, S., Dyer, B., & Rees, K (2016). Questionnaire study to gain an insight into the manufacturing and fitting process of artificial eyes in children: an ocularist perspective. *International Ophthalmology*, doi: 10.1007/s10792-016-0383-4
5. Chinnery, H., Thompson, S. B. N., Noroozi, S., & Dyer, B (2017). Scoping Review of the development of artificial eyes throughout the years. *Edorium Journal of Disability and Rehabilitation* 3 1-10 doi: 10.5348/D05-2017-25-RA-1
6. Chinnery, H., Thompson, S. B. N., Noroozi, S., Dyer, B., & Rees, K (2017). The importance of incorporating technological advancements into the artificial eye process: a perspective commentary. *Insights into Ophthalmology*

## Appendix B – Conferences

### Conferences:

#### *Attended:*

1. International Society of Paediatric Oncology (SIOP). Toronto, Canada. 22-25<sup>th</sup> October 2014.
2. Association of European Ocularists (AEO). Manchester, UK. 3<sup>rd</sup> – 4<sup>th</sup> September 2015

#### *Presented:*

1. The 7<sup>th</sup> Annual Postgraduate Researcher Conference. Bournemouth University, UK. *Poster presentation.*
2. The Faculty of Science and Technology Annual Postgraduate Researcher Conference. Bournemouth, UK. *Oral presentation (1<sup>st</sup> prize). 20/05/2015*
3. 3MP Conference. Bournemouth University, UK. *Oral Presentation. 24/02/2016*
4. The 8<sup>th</sup> Annual Postgraduate Researcher Conference. Bournemouth University, UK. *Poster presentation. 9-10/03/2016*
5. The 8<sup>th</sup> Annual Postgraduate Researcher Conference. Bournemouth University, UK. *Oral presentation. 9-10/03/2016*
6. The Faculty of Science and Technology Annual Postgraduate Researcher Conference. Bournemouth, UK. *Poster 18/05/2016. Poster Presentation.*
7. The 9<sup>th</sup> Annual Postgraduate Researcher Conference. Bournemouth University, UK. *Poster Presentation. 8/03/2017.*



## Appendix C – Extra-circular activities

1. I attended various post-graduate researcher's workshops hosted by Bournemouth University (September 2014 – December 2015).
2. I have marked on a range of undergraduate psychology modules (October 2014 – May 2017).
3. I have assisted with the Showcasing Undergraduate Research Conference (SURE) (2015).
4. I have delivered a talk on eating disorders as part of the eating disorders awareness week at Bournemouth University (February 2016).
5. Teaching on the Masters of Science in Foundations of Clinical Psychology

### Lectures given:

1. Retinoblastoma (MSc) 21/10/2015
2. Lived experience of eating disorders (MSc) 11/11/2015
3. NHS Ethics (MSc) 13/01/2016
4. Emotions (MSc) 19/10/2016
5. Retinoblastoma (MSc) 19/10/2016
6. Lived experience of eating disorders (MSc) 16/11/2016
7. NHS Ethics (MSc) 11/01/2017

## Appendix D – Ethics



# Research Ethics Checklist

<b>Reference Id</b>	8842
<b>Status</b>	Approved
<b>Date Approved</b>	30/07/2015

## Researcher Details

<b>Name</b>	Holly Chinnery
<b>School</b>	Faculty of Science & Technology
<b>Status</b>	Postgraduate Research (MRes, MPhil, PhD, DProf, DEng)
<b>Course</b>	Postgraduate Research
<b>Have you received external funding to support this research project?</b>	No

## Project Details

<b>Title</b>	Investigation of the skills and technologies used in the fitting process of artificial eyes in the UK with exploration into potential improvements to this process.
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<b>Proposed Start Date of Data Collection</b>	10/08/2015
<b>Proposed End Date of Project</b>	21/07/2017
<b>Supervisor</b>	Simon Thompson
<b>Approver</b>	Simon Thompson

<b>Summary – no more than 500 words (including detail on background, methodology, sample, outcomes, etc)</b>
<p>Retinoblastoma is a rare type of eye cancer forming 3% of all childhood cancers (Shields, 2008). It is diagnosed in 40 to 50 children below the age of five in the UK each year and is non-discriminate in terms of gender and right or left eye (Willard et al., 2014). Retinoblastoma has a five-year survival rate of 98% in developed countries, though this is dependent on early diagnosis and treatment (Meel, Radhakrishnan &amp; Bakhshi, 2012). When the tumour fills more than half of the globe and there is extensive seeding, enucleation becomes the treatment of choice. Enucleation is the surgical removal of the eye where an orbital implant is fitted into the socket to retain the volume and stop the eyelids from dropping. Six weeks after the surgery, the patient is referred to one of two National Health Services to be fitted with an artificial eye. The design, manufacturing and fitting of artificial eyes has not changed since the 1960s and varies in terms of skill base and technology base between both services. Currently the process is labour intensive, expensive and uncomfortable for the patient and their family in terms of distress. This research aims to explore the skills and technologies currently used in both the fitting procedure and manufacture of artificial eyes. The outcomes include being able to identify potential improvements that can be made to the existing services, create consistency between services in terms of skill and technologies used, reduce time of fitting and to improve comfort for the wearer. This research will be conducted by interviewing 8 to 15 staff from both Moorefield’s Eye Hospital and the National Artificial Eye Service who are involved in the manufacturing and fitting procedure of artificial eyes</p>

External Ethics Review

<b>Does your research require external review through the NHS National Research Ethics Service (NRES) or through another external Ethics Committee?</b>	No
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Research Literature

<b>Is your research solely literature based?</b>	No
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Human Participants

<b>Will your research project involve interaction with human participants as primary sources of data (e.g. interview, observation, original survey)?</b>	Yes
<b>Will your research project involve interaction with human participants as primary sources of data (e.g. interview, observation, original survey)?</b>	No
<b>Does the study involve participants age 16 or over who are unable to give informed consent (i.e. people with learning disabilities)? NOTE: All research that falls under the auspices of the Mental Capacity Act 2005 must be reviewed by NHS NRES.</b>	No
<b>Will the study require the co-operation of a gatekeeper for initial access to the groups or individuals to be recruited? (i.e. students at school, members of self-help group, residents of Nursing home?)</b>	No
<b>Will it be necessary for participants to take part in your study without their knowledge and consent at the time (i.e. covert observation of people in non-public places)?</b>	No
<b>Will the study involve discussion of sensitive topics (i.e. sexual activity, drug use, criminal activity)?</b>	No
<b>Are drugs, placebos or other substances (i.e. food substances, vitamins) to be administered to the study participants or will the study involve invasive, intrusive or potentially harmful procedures of any kind?</b>	No

<b>Could your research induce psychological stress or anxiety, cause harm or have negative consequences for the participant or researcher (beyond the risks encountered in normal life)?</b>	No
<b>Will your research involve prolonged or repetitive testing?</b>	No
<b>Will the research involve the collection of audio materials?</b>	Yes
<b>Is this audio collection solely for the purposes of transcribing/summarising and will not be used in any outputs (publication, dissemination, etc.) and will not be made publicly available?</b>	Yes
<b>Will your research involve the collection of photographic or video materials?</b>	No
<b>Will financial or other inducements (other than reasonable expenses and compensation for time) be offered to participants?</b>	No

**Please explain below why your research project involves the above mentioned criteria (be sure to explain why the sensitive criterion is essential to your project's success). Give a summary of the ethical issues and any action that will be taken to address these. Explain how you will obtain informed consent (and from whom) and how you will inform the participant(s) about the research project (i.e. participant information sheet). A sample consent form and participant information sheet can be found on the Research Ethics website.**

By using audio recordings voice recognition is possible, thus initially data will not be anonymous. One week following the interview, all responses will be transcribed and the recording deleted ensuring anonymity of all

data. All participants will be clearly informed about this via the information sheet, have the opportunity to ask questions and sign the audio recording consent form only if they understand the situation.

Final Review

<b>Will you have access to personal data that allows you to identify individuals OR access to confidential corporate or company data (that is not covered by confidentiality terms within an agreement or by a separate confidentiality agreement)?</b>	No
<b>Will your research involve experimentation on any of the following: animals, animal tissue, genetically modified organisms?</b>	No
<b>Will your research take place outside the UK (including any and all stages of research: collection, storage, analysis, etc.)?</b>	No

Final Review

**Please use the below text box to highlight any other ethical concerns or risks that may arise during your research that have not been covered in this form.**

Ethical issues that need to be addressed include, responsibility, respect, transparency, integrity and confidentiality. Prior to the start of this study, participants will receive an information sheet and have the opportunity to ask questions before they decide whether to participate. The information sheet will give a clear explanation as to the purpose of the study, their role, confidentiality and their right to withdraw during and up to one week after the interview. Participants will be told that initially the data will not be anonymous due to the nature of the study, however the responses will be anonymised and the recordings will be deleted as soon as they have been transcribed. Although data handling will be confidential and participants will be asked to keep confidentiality, it cannot be assumed that they will. By signing the consent forms participants state that they understand this situation. For those agreeing to participate, they will be given two consent forms to sign: one to participate in the study, the other agreeing for the interview to be audio recorded. The full purpose of the study including the procedures involved and foreseeable risks and benefits will be explained, thus creating transparency and eliminating deception. All participants will be thoroughly debriefed at the end of each experiment where the purpose of the research and their role within it will be re-stated. All questions will be answered as honest and fully as possible.



# Health Research Authority

## South Central - Berkshire Research Ethics Committee

Bristol REC Centre  
Whitefriars  
Level 3, Block B  
Lewins Mead  
Bristol  
BS1 2NT  
Telephone: 0207 104 8049

17

January 2017

**Please note: This is the favourable opinion of the**

**REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval**

Miss Holly Chinnery

Post-Graduate Researcher

Bournemouth University

Faculty of Science & Technology, Psychology Research Centre

Bournemouth University, Talbot Campus, Poole House

Fern Barrow, Poole, Dorset, UK

BH12 5BB

Dear Miss Chinnery

**Study title:** A phenomenology study of the lived experience of parents of, and, children whom wear an artificial eye as a result of retinoblastoma

**REC reference:** 16/SC/0588

**IRAS project ID:** 211910

Thank you for responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact [hra.studyregistration@nhs.net](mailto:hra.studyregistration@nhs.net) outlining the reasons for your request.

#### **Confirmation of ethical opinion**

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

#### **Conditions of the favourable opinion**

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for NHS permission for research is available in the Integrated Research Application System, [www.hra.nhs.uk](http://www.hra.nhs.uk) or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

#### Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).



There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact [hra.studyregistration@nhs.net](mailto:hra.studyregistration@nhs.net). The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

**It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).**

### **Ethical review of research sites**

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

### **Approved documents**

The final list of documents reviewed and approved by the Committee is as follows:

The final list of documents reviewed and approved by the Committee is as follows: <i>Document</i>	<i>Version</i>	<i>Date</i>
Covering letter on headed paper [Covering letter regarding previous application (first review)]	v0.2	07 July 2016
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Professional indemnity]	20 July 2015	
Interview schedules or topic guides for participants [Interview guide/Experience Near Question]	V0.3	09 December 2016
Interview schedules or topic guides for participants [Interview guide/Experience Near Question Tracked changes]	V0.3	09 December 2016
Interview schedules or topic guides for participants [Pre-interview questions ]	V0.3	09 December 2016
Interview schedules or topic guides for participants [Pre-interview questions Tracked changes]	V0.3	09 December 2016
IRAS Application Form [IRAS_Form_18102016]	18 October 2016	
IRAS Checklist XML [Checklist_16122016]	16 December 2016	
Letter from sponsor [BU Sponsor Letter]	17 February 2016	
Letters of invitation to participant [Study invitation letter]	V0.3	09 December 2016
Letters of invitation to participant [Study invitation letter Tracked changed]	V0.3	09 December 2016
Other [Unfavourable opinion of first review letter]		27 June 2016

Other [Employer's Professional Liability]		20 July 2016
Other [Support services]	V0.3	09 December 2016
Other [Debrief]	V0.3	09 December 2016
Other [Study protocol Tracked changes]	V0.3	09 December 2016
Other [Support services Tracked changes]	V0.3	09 December 2016
Other [Debrief Tracked changes]	V0.3	09 December 2016
Participant consent form [Young Person's consent form for participation]	V0.3	09 December 2016
Participant consent form [Young Person's consent form for participation Tracked changes]	V0.3	09 December 2016
Participant consent form [Young Person's assent for participation]	V0.3	09 December 2016
Participant consent form [Parents consent for their child's participation]	V0.3	09 December 2016
Participant consent form [Parents consent for their child's participation Tracked changes]	V0.3	09 December 2016
Participant information sheet (PIS) [Young persons PIS]	V0.3	09 December 2016
Participant information sheet (PIS) [Young Persons PIS Tracked changes]	V0.3	09 December 2016
Participant information sheet (PIS) [Parent's information sheet for child's participation]	V0.3	09 December 2016
Research protocol or project proposal [Study protocol]	V0.3	09 December 2016
Summary CV for student [HC CV]		07 July 2016
Summary CV for supervisor (student research) [ST CV]		07 July 2016
Summary CV for supervisor (student research) [KR CV]		07 July 2016
Summary CV for supervisor (student research) [SN CV]		07 July 2016
Summary CV for supervisor (student research) [BD CV]		07 July 2016

### **Statement of compliance**

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

### **After ethical review**

#### Reporting requirements

The attached document “*After ethical review – guidance for researchers*” gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

### **User Feedback**

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: <http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

### **HRA Training**

We are pleased to welcome researchers and R&D staff at our training days – see details at <http://www.hra.nhs.uk/hra-training/>

**16/SC/0588**

**Please quote this number on all correspondence**

With the Committee’s best wishes for the success of this project.

Yours sincerely



**Pp Mr David Carpenter**

**Chair**

Email: [nrescommittee.southcentral-berkshire@nhs.net](mailto:nrescommittee.southcentral-berkshire@nhs.net)

*Enclosures:* “After ethical review – guidance for

researchers” Copy to: Dr Fiona Knight

Mrs Manni Sandhu, Birmingham Children's Hospital





# Health Research Authority

Miss Holly Chinnery  
Post-Graduate Researcher  
Bournemouth University  
Faculty of Science & Technology, Psychology Research Centre  
Bournemouth University, Talbot Campus, Poole House  
Fern Barrow, Poole, Dorset, UK  
BH12 5BB

Email: [hra.approval@nhs.net](mailto:hra.approval@nhs.net)

20 January 2017

Dear Miss Holly Chinnery



**Study title:** A phenomenological study of the lived experience of parents of, and the children whom wear artificial eyes as a result of retinoblastoma.

**IRAS project ID:** 211910

**REC reference:** 16/SC/0588

**Sponsor** Bournemouth University

I am pleased to confirm that **HRA Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications noted in this letter.

### **Participation of NHS Organisations in England**

The sponsor should now provide a copy of this letter to all participating NHS organisations in England.

*Appendix B* provides important information for sponsors and participating NHS organisations in England for arranging and confirming capacity and capability. **Please read *Appendix B* carefully**, in particular the following sections:

- *Participating NHS organisations in England* – this clarifies the types of participating organisations in the study and whether or not all organisations will be undertaking the same activities
- *Confirmation of capacity and capability* - this confirms whether or not each type of participating NHS organisation in England is expected to give formal confirmation of capacity and capability. Where formal confirmation is not expected, the section also provides details on the time limit given to participating organisations to opt out of the study, or request additional time, before their participation is assumed.
- *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* - this provides detail on the form of agreement to be used in the study to confirm capacity and capability, where applicable.

Further information on funding, HR processes, and compliance with HRA criteria and standards is also provided.

It is critical that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details and further information about working with the research management function for each organisation can be accessed from [www.hra.nhs.uk/hra-approval](http://www.hra.nhs.uk/hra-approval).

### **Appendices**

The HRA Approval letter contains the following appendices:

- A – List of documents reviewed during HRA assessment
- B – Summary of HRA assessment

### **After HRA Approval**

The document “*After Ethical Review – guidance for sponsors and investigators*”, issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research

- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

In addition to the guidance in the above, please note the following:

- HRA Approval applies for the duration of your REC favourable opinion, unless otherwise notified in writing by the HRA.
- Substantial amendments should be submitted directly to the Research Ethics Committee, as detailed in the *After Ethical Review* document. Non-substantial amendments should be submitted for review by the HRA using the form provided on the HRA website, and emailed to [hra.amendments@nhs.net](mailto:hra.amendments@nhs.net).
- The HRA will categorise amendments (substantial and non-substantial) and issue confirmation of continued HRA Approval. Further details can be found on the [HRA website](#)

## Scope

HRA Approval provides an approval for research involving patients or staff in NHS organisations in England.

If your study involves NHS organisations in other countries in the UK, please contact the relevant national coordinating functions for support and advice. Further information can be found at <http://www.hra.nhs.uk/resources/applying-for-reviews/nhs-hsc-rd-review/>.

If there are participating non-NHS organisations, local agreement should be obtained in accordance with the procedures of the local participating non-NHS organisation.

## User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please email the HRA at [hra.approval@nhs.net](mailto:hra.approval@nhs.net). Additionally, one of our staff would be happy to call and discuss your experience of HRA Approval.

## HRA Training

We are pleased to welcome researchers and research management staff at our training days – see details at <http://www.hra.nhs.uk/hra-training/>

Your IRAS project ID is **211910**. Please quote this on all correspondence.

Yours sincerely

Steph Macpherson

Senior Assessor

Email: [hra.approval@nhs.net](mailto:hra.approval@nhs.net) Copy Julie Northam, Bournemouth University [Sponsor]  
to:

Mrs Manni Sandhu, Birmingham Children's Hospital [Lead NHS R&D]







**Birmingham Women's  
and Children's  
NHS Foundation Trust**

**Birmingham Children's Hospital**

**Research and Development  
Steelhouse Lane Birmingham B4 6NH  
Tel: 0121 333 8751/8298/8748  
Research@bch.nhs.uk**

10th March 2017

Miss Holly Chinnery  
39 Mills Chase  
Bracknell  
Berkshire  
RG12 9RE

Dear Holly,

**Letter of access for research**

This letter should be presented to each participating organisation before you commence your research at that site. The participating organisation is Birmingham Women's and Children's Hospital NHS Foundation Trust

In accepting this letter, each participating organisation confirms your right of access to conduct research through their organisation for the purpose and on the terms and conditions set out below. This right of access commences on 10th March 2017 and ends on 18th September 2017 unless terminated earlier in accordance with the clauses below.

You have a right of access to conduct such research as confirmed in writing in the letter of permission for research from Birmingham Women's & Children's Hospital NHS Foundation Trust.

The information supplied about your role in research at the organisation(s) has been reviewed and you do not require an honorary research contract with the organisation(s). We are satisfied that such pre-engagement checks as we consider necessary have been carried out. Evidence of checks should be available on request to the organisation(s).

You are considered to be a legal visitor to the organisations premises. You are not entitled to any form of payment or access to other benefits provided by the organisation(s) or this organisation to employees and this letter does not give rise to any other relationship between you and the organisation(s), in particular that of an employee.

While undertaking research through the organisation(s) you will remain accountable to your substantive employer but you are required to follow the reasonable instructions of the organisation(s) or those instructions given on their behalf in relation to the terms of this right of access.

Where any third party claim is made, whether or not legal proceedings are issued, arising out of or in connection with your right of access, you are required to co-operate fully with any investigation by the organisation(s) in connection with any such claim and to give all such assistance as may reasonably be required regarding the conduct of any legal proceedings.

You must act in accordance with the organisations policies and procedures, which are available to you upon request, and the Research Governance Framework.

You are required to co-operate with the organisation(s) in discharging its/their duties under the Health and Safety at Work etc. Act 1974 and other health and safety legislation and to take reasonable care for the health and safety of yourself and others while on the organisations premises. You must observe the same standards of care and propriety in dealing with patients, staff, visitors, equipment and premises as is expected of any other contract holder and you must act appropriately, responsibly and professionally at all times.

If you have a physical or mental health condition or disability which may affect your research role and which might require special adjustments to your role, if you have not already done so, you must notify your employer and each organisation prior to commencing your research role at that organisation.

You are required to ensure that all information regarding patients or staff remains secure and *strictly confidential* at all times. You must ensure that you understand and comply with the requirements of the NHS Confidentiality Code of Practice and the Data Protection Act 1998. Furthermore you should be aware that under the Act, unauthorised disclosure of information is an offence and such disclosures may lead to prosecution.

You should ensure that, where you are issued with an identity or security card, a bleep number, email or library account, keys or protective clothing, these are returned upon termination of this arrangement. Please also ensure that while on the organisations premises you wear your ID badge at all times, or are able to prove your identity if challenged. Please note that the organisation(s) do not accept responsibility for damage to or loss of personal property.

This organisation may revoke this letter and any organisation(s) may terminate your right to attend at any time either by giving seven days' written notice to you or immediately without any notice if you are in breach of any of the terms or conditions described in this letter or if you commit any act that we reasonably consider to amount to serious misconduct or to be disruptive and/or prejudicial to the interests and/or business of the organisation(s) or if you are convicted of any criminal offence. You must not undertake regulated activity if you are barred from such work. If you are barred from working with adults or children this letter of access is immediately terminated. Your employer will immediately withdraw you from undertaking this or any other regulated activity and you MUST stop undertaking any regulated activity immediately.

Your substantive employer is responsible for your conduct during this research project and may in the circumstances described above instigate disciplinary action against you.

No organisation will indemnify you against any liability incurred as a result of any breach of confidentiality or breach of the Data Protection Act 1998. Any breach of the Data Protection Act 1998 may result in legal action against you and/or your substantive employer.

If your current role or involvement in research changes, or any of the information provided in your Research Passport changes, you must inform your employer through their normal procedures. You

must also inform your nominated manager (Maureen McCalla/Theresa Morton) in each participating organisation and the R&D office in this organisation.

Yours sincerely

**Theresa Morton**  
**Head of Research, Development and Innovation**  
**cc: R&D office at Birmingham Children's Hospital**  
**HR department of the substantive employer**



**Birmingham Women's and Children's NHS Foundation Trust  
Confirmation of Capacity & Capability**



**Research and Development Office**

Direct Line: 0121 333 8707 / 8708 / 8298

[research@bch.nhs.uk](mailto:research@bch.nhs.uk)

**Research and Development**

Steelhouse Lane

Birmingham

B4 6NH

10<sup>th</sup> March 2017

Our Ref: TM/RR/R&D Approval

Holly Chinnery  
Birmingham Children's Hospital,  
Steelhouse Lane,  
Birmingham, B4 6NH

Dear Holly

**Re: Birmingham Children's Hospital Confirmation of Capacity & Capability**

**Project Title: A phenomenological study of the lived experience of parents of, and the children whom wear artificial eyes as a result of retinoblastoma.**

**HRA/ IRAS Ref: 211910**

**R & D No.: 1853**

Thank you for informing Birmingham Children's Hospital NHS Foundation Trust's R&D office of the above project.

I can confirm that Birmingham Children's Hospital NHS Foundation Trust has the capacity and capability to deliver the above referenced study and therefore is issuing R&D approval for this project.

Please find attached our agreed Statement of Activities as confirmation.

**We agree to start this study on 10th March 2017.**

R&D Approval of the study is subject to the following conditions:

1. That you inform and send copies of correspondence to the R&D Office, the appropriate regulatory authorities and competent authorities of any amendments.
2. That you provide the R&D Office with annual progress reports and end of study declaration form as well as any acknowledgements.
3. That you conduct the research in conformity with the Research Governance Framework and other legal and regulatory requirements where applicable.
4. That the chief/Principal Investigator and the research team should be familiar with BCH trial Standard Operating Procedures. These SOPs can be found at the following location on the trust intranet. <http://solitaire.zion.matrix.local/corporate/research-and-development/key-documents/r%2526d-useful-documents>

Documents approved:

Documents	Version	Date
Interview schedules or topic guides for participants [Interview guide/Experience Near Question]	V0.3	09 December 2016
Interview schedules or topic guides for participants [Interview guide/Experience Near Question Tracked changes]	V0.3	09 December 2016
Interview schedules or topic guides for participants [Pre- interview questions ]	V0.3	09 December 2016
Interview schedules or topic guides for participants [Pre- interview questions Tracked changes]	V0.3	09 December 2016
Letters of invitation to participant [Study invitation letter]	V0.3	09 December 2016
Other [Support services]	V0.3	09 December 2016
Other [Debrief]	V0.3	09 December 2016
Participant consent form [Young Person's consent for participation]	V0.4	19 January 2017

Participant consent form [Young Person's assent for participation]	V0.4	19 January 2017
Participant consent form [Parents consent for their child's participation]	V0.4	19 January 2017
Participant information sheet (PIS) [Young persons PIS]	V0.4	19 January 2017
Participant information sheet (PIS) [Parent's information sheet for child's participation]	V0.4	19 January 2017
Research protocol or project proposal [Study protocol]	V0.3	09 December 2016

Your research project documents can be found at: V:\R&D Study Head and Neck\Ophthalmology\Chinnery\_H\_1853\_Fitting of artificial eyes in children with retinoblastoma. If you cannot access this please let us know and we will provide this.

Please inform the R&D Data Manager via email [Rakesh.Sheinmar@bch.nhs.uk](mailto:Rakesh.Sheinmar@bch.nhs.uk) when you have recruited your first patient.

Finally, I would like to take this opportunity to wish you well with your research project. If you need any further assistance or guidance, please do not hesitate to contact us.

Yours sincerely



Theresa Morton

Head of Research, Development and Innovation



# Research Ethics Checklist

<b>Reference Id</b>	16622
<b>Status</b>	Approved
<b>Date Approved</b>	13/06/2017

## Researcher Details

<b>Name</b>	Holly Chinnery
<b>School</b>	Faculty of Science & Technology
<b>Status</b>	Postgraduate Research (MRes, MPhil, PhD, DProf, DEng)
<b>Course</b>	Postgraduate Research
<b>Have you received external funding to support this research project?</b>	No

## Project Details

<b>Title</b>	Understanding the lived experiences of the fitting process of artificial eyes in parents' of children with a diagnosis of retinoblastoma

<b>Proposed Start Date of Data Collection</b>	12/06/2017
<b>Proposed End Date of Project</b>	11/09/2017
<b>Supervisor</b>	Simon Thompson
<b>Approver</b>	Bryce Dyer

<b>Summary – no more than 500 words (including detail on background, methodology, sample, outcomes, etc)</b>
<p>Retinoblastoma is a rare type of intraocular childhood eye cancer typically occurring in children before the age of five years. Prognosis is dependent upon early diagnosis, accurate classification of the tumours and early treatment. Unilateral retinoblastoma is normally non-hereditary compared to bilateral retinoblastoma which is typically hereditary where secondary tumours are more common. Survivorship is much better for unilateral compared to bilateral incidences. Increased survivorship across the spectrum of cancers has seen the development of psycho-oncology. With more prominent cancers existing such as breast and lung cancer, the psycho-oncology of retinoblastoma has been relatively untouched. What we do know is that parent's over-estimate their child's behavioural and emotional problems based on their mal-adjustment and coping strategies to the disease and it's after effects in spite of the survivor rating their own functioning as normal. The discrepancy between the survivor's quality of life and that as perceived by their parents appears to be a reflection of the parent's coping mechanism rather than the child's real functioning. With the fitting process of artificial eyes being anecdotal in the literature, there is a need to understand the experience of this process in parents of the affected child. An Interpretative Phenomenological Approach (IPA) will be used in order to gain an in-depth understanding of the lived experiences of parents of the affected child. The sampling method will be purposive with a sample size of approximately 4-6 participants. Participants will be recruited via an online retinoblastoma support group. A study invitation letter, participant information sheet and a consent form will be given to potential participants prior to the start of the study. Following agreement to participate, participants' will sign the consent form and then complete a demographic form and online questionnaire. The questionnaire will consist of one main question and various discussion points. Participants will be asked to provide as much detail as possible to their answer. Data will be analysed using the IPA method developed by Smith et al., (2008).</p>

#### External Ethics Review

<b>Does your research require external review through the NHS National Research Ethics Service (NRES) or through another external Ethics Committee?</b>	No
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#### Research Literature

<b>Is your research solely literature based?</b>	No
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#### Human Participants

<b>Will your research project involve interaction with human participants as primary sources of data (e.g. interview, observation, original survey)?</b>	Yes
--	-----



<b>Does your research specifically involve participants who are considered vulnerable (i.e. children, those with cognitive impairment, those in unequal relationships—such as your own students, prison inmates, etc.)?</b>	No
<b>Does the study involve participants age 16 or over who are unable to give informed consent (i.e. people with learning disabilities)? NOTE: All research that falls under the auspices of the Mental Capacity Act 2005 must be reviewed by NHS NRES.</b>	No
<b>Will the study require the co-operation of a gatekeeper for initial access to the groups or individuals to be recruited? (i.e. students at school, members of self-help group, residents of Nursing home?)</b>	No
<b>Will it be necessary for participants to take part in your study without their knowledge and consent at the time (i.e. covert observation of people in non-public places)?</b>	No
<b>Will the study involve discussion of sensitive topics (i.e. sexual activity, drug use, criminal activity)?</b>	No
<b>Are drugs, placebos or other substances (i.e. food substances, vitamins) to be administered to the study participants or will the study involve invasive, intrusive or potentially harmful procedures of any kind?</b>	No
<b>Will tissue samples (including blood) be obtained from participants? Note: If the answer to this question is ‘yes’ you will need to be aware of obligations under the Human Tissue Act 2004.</b>	No

<b>Could your research induce psychological stress or anxiety, cause harm or have negative consequences for the participant or researcher (beyond the risks encountered in normal life)?</b>	No
<b>Will your research involve prolonged or repetitive testing?</b>	No
<b>Will the research involve the collection of audio materials?</b>	Yes
<b>Will your research involve the collection of photographic or video materials?</b>	
<b>Will financial or other inducements (other than reasonable expenses and compensation for time) be offered to participants?</b>	No

<b>Please give a summary of the ethical issues and any action that will be taken to address these. Explain how you will obtain informed consent (and from whom) and how you will inform the participant about the research project (i.e. participant information sheet).</b>
Ethical considerations relating to this study include confidentiality, anonymity and informed consent. Participants will be fully informed of the research procedure and will be told that they have the right to withdraw their participation at any time before, during and two weeks after the questionnaires. The two week deadline has been given as this is when the data will have been transcribed, therefore, the researcher will not be able to identify which data belongs to which participant. Participants will be asked to sign a consent form prior to the start of the study stating that they understand the aim of the study, their right to withdraw, confidentiality and anonymity concerns and are happy to participate in the study. Confidentiality and anonymity will be maintained at all times. All data collected during the study will be made anonymous using a unique identification number. Following transfer of data into

an electronic database, all paper records and recordings will be disposed of in accordance with Bournemouth University's procedure for disposing of confidential information.

Final Review

<b>Will you have access to personal data that allows you to identify individuals OR access to confidential corporate or company data (that is not covered by confidentiality terms within an agreement or by a separate confidentiality agreement)?</b>	No
<b>Will your research involve experimentation on any of the following: animals, animal tissue, genetically modified organisms?</b>	No
<b>Will your research take place outside the UK (including any and all stages of research: collection, storage, analysis, etc.)?</b>	No

**Please use the below text box to highlight any other ethical concerns or risks that may arise during your research that have not been covered in this form.**

Ethical issues that need to be addressed include, responsibility, respect, transparency, integrity and confidentiality. Prior to the start of this study, participants will receive an information sheet and have the opportunity to ask questions before they decide whether to participate. The information sheet will give a clear explanation as to the purpose of the study, their role, confidentiality and their right to withdraw during and up to one week after the interview. Participants will be told that initially the data will not be anonymous due to the nature of the study, however the responses will be anonymised and the recordings will be deleted as soon as they have been transcribed. Although data handling will be confidential and participants will be asked to keep confidentiality, it cannot be assumed that they will. By signing the consent forms participants state that they understand this situation. For those agreeing to participate, they will be given two consent forms to sign: one to participate in the study, the other agreeing for the interview to be audio recorded. The full purpose of the study including the procedures involved and foreseeable risks and benefits will be explained, thus creating transparency and eliminating deception. All participants will be thoroughly debriefed at the end of each experiment where the purpose of the research and their role within it will be re-stated. All questions will be answered as honest and fully as possible.

**Please use the below text box to highlight any other ethical concerns or risks that may arise during your research that have not been covered in this form.**

There are no other ethical concerns that have not addressed in this form.



# Research Ethics Checklist

<b>Reference Id</b>	14010
<b>Status</b>	Approved
<b>Date Approved</b>	41/01/2017

## Researcher Details

<b>Name</b>	Holly Chinnery
<b>School</b>	Faculty of Science & Technology
<b>Status</b>	Postgraduate Research (MRes, MPhil, PhD, DProf, DEng)
<b>Course</b>	Postgraduate Research
<b>Have you received external funding to support this research project?</b>	No

## Project Details

<b>Title</b>	Creating a transfer of knowledge between the methods employed in the maxillofacial prosthetic and artificial eye process to assess whether the technology incorporated in maxillofacial prosthetics can be applied to the artificial eye process.
	09/01/2017

<b>Proposed Start Date of Data Collection</b>	
<b>Proposed End Date of Project</b>	29/12/2017
<b>Supervisor</b>	Simon Thompson
<b>Approver</b>	Ben Parris

6.

7.

<b>Summary – no more than 500 words (including detail on background, methodology, sample, outcomes, etc)</b>
<p>Application of technology into healthcare has typically been targeted to high demand illnesses and treatments. However, with an increasing need to meet patient expectations combined with increased accessibility and reduced costs, smaller healthcare fields are starting to investigate its function and usability. Although the use of medical technologies in artificial eye development is an area yet to be explored, its investigation in the similar field of maxillofacial prosthetics has begun. This has included scanning technologies that can capture the contours and fine details of the eye socket, 3D CAD software that can accurately represent the surface of the object and RP&amp;M processes that use wax-based patterns to produce the prosthesis. This can start the evolutionary process where products are continually re-designed and redeveloped to achieve excellent patient outcome and satisfaction levels. This study aims to create a transfer of knowledge between the methods employed in the maxillofacial prosthetic and artificial eye process. This will be achieved by creating a model of the processes employed in each field and comparing them to see whether the technology incorporated in maxillofacial prosthetics can be applied to the artificial eye process. The sampling method for this study is purposive. Purposive sampling allows participants to be selected according to the needs of the study. It provides insight and ‘information-rich’ accounts of the topic of interest as well as a diverse range of views and experiences (Patton, 2002). Requiring opinions of people with a relatively high level of skill and knowledge in maxillofacial and artificial eye prosthetics, resulted in the subtype of purposive sampling: expert sampling, being employed for this study. To ensure the widest range of processes utilised in maxillofacial prosthetics and artificial eye development are acquired, views were sought from these services throughout the UK. Consequently, a moderate to large sample size will be needed to provide enough data to fully explore the research aim (Braun &amp; Clarke, 2013). Taking into consideration the small population being explored and that this study is part of a PhD thesis where there are practical and logistical issues, a sample size of 15-20 was estimated to be sufficient to achieve data saturation (Braun &amp; Clarke, 2013). Recruitment will take place between January and March 2017. Potential participants will be identified via an online search of maxillofacial laboratories and artificial eye services within the UK. Email invitations will be used as the main recruitment method for this study. The email will contain an invitation letter and information sheet about the study. Semi-structured interviews will be used to find out people’s experiences, perceptions, opinions and values. All interviews will be carried out at the participant’s place of work to keep the level of inconvenience to a minimum. Whilst there are no immediate benefits to participants from taking part in the study, the results from the study has the potential to improve the process of artificial eyes. Each interview will last approximately 30 minutes.</p>

## External Ethics Review

<b>Does your research require external review through the NHS National Research Ethics Service (NRES) or through another external Ethics Committee?</b>	No
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Research Literature

<b>Is your research solely literature based?</b>	No
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Human Participants

<b>Will your research project involve interaction with human participants as primary sources of data (e.g. interview, observation, original survey)?</b>	Yes
<b>Does your research specifically involve participants who are considered vulnerable (i.e. children, those with cognitive impairment, those in unequal relationships—such as your own students, prison inmates, etc.)?</b>	No
<b>Does the study involve participants age 16 or over who are unable to give informed consent (i.e. people with learning disabilities)? NOTE: All research that falls under the auspices of the Mental Capacity Act 2005 must be reviewed by NHS NRES.</b>	No
<b>Will the study require the co-operation of a gatekeeper for initial access to the groups or individuals to be recruited? (i.e. students at school, members of self-help group, residents of Nursing home?)</b>	No
<b>Will it be necessary for participants to take part in your study without their knowledge and consent at the time (i.e. covert observation of people in non-public places)?</b>	No
<b>Will the study involve discussion of sensitive topics (i.e. sexual activity, drug use, criminal activity)?</b>	No
<b>Are drugs, placebos or other substances (i.e. food substances, vitamins) to be administered to the study participants or will the study involve invasive, intrusive or potentially harmful procedures of any kind?</b>	No
<b>Will tissue samples (including blood) be obtained from participants? Note: If the answer to this question is ‘yes’ you will need to be aware of obligations under the Human Tissue Act 2004.</b>	No

<b>Could your research induce psychological stress or anxiety, cause harm or have negative consequences for the participant or researcher (beyond the risks encountered in normal life)?</b>	No
<b>Will your research involve prolonged or repetitive testing?</b>	No
<b>Will the research involve the collection of audio materials?</b>	Yes
<b>Is this audio collection solely for the purposes of transcribing/summarising and will not be used in any outputs (publication, dissemination, etc.) and will not be made publicly available?</b>	Yes
<b>Will your research involve the collection of photographic or video materials?</b>	No

<b>Will financial or other inducements (other than reasonable expenses and compensation for time) be offered to participants?</b>	No
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**Please explain below why your research project involves the above mentioned criteria (be sure to explain why the sensitive criterion is essential to your project's success). Give a summary of the ethical issues and any action that will be taken to address these. Explain how you will obtain informed consent (and from whom) and how you will inform the participant(s) about the research project (i.e. participant information sheet). A sample consent form and participant information sheet can be found on the Research Ethics website.**

By using audio recordings voice recognition is possible, thus initially data will not be anonymous. One week following the interview, all responses will be transcribed and the recording deleted ensuring anonymity of all data. All participants will be clearly informed about this via the information sheet, have the opportunity to ask questions and sign the audio recording consent form only if they understand the situation.

Final Review

<b>Will you have access to personal data that allows you to identify individuals OR access to confidential corporate or company data (that is not covered by confidentiality terms within an agreement or by a separate confidentiality agreement)?</b>	No
<b>Will your research involve experimentation on any of the following: animals, animal tissue, genetically modified organisms?</b>	No
<b>Will your research take place outside the UK (including any and all stages of research: collection, storage, analysis, etc.)?</b>	No

**Please use the below text box to highlight any other ethical concerns or risks that may arise during your research that have not been covered in this form.**

Ethical issues that need to be addressed include, responsibility, respect, transparency, integrity and confidentiality. Prior to the start of this study, participants will receive an information sheet and have the opportunity to ask questions before they decide whether to participate. The information sheet will give a clear explanation as to the purpose of the study, their role, confidentiality and their right to withdraw during and up to one week after the interview. Participants will be told that initially the data will not be anonymous due to the nature of the study, however the responses will be anonymised and the recordings will be deleted as soon as they have been transcribed. Although data handling will be confidential and participants will be asked to keep confidentiality, it cannot be assumed that they will. By signing the consent forms participants state that they understand this situation. For those agreeing to participate, they will be given two consent forms to sign: one to participate in the study, the other agreeing for the interview to be audio recorded. The full purpose of the study including the procedures involved and foreseeable risks and benefits will be explained, thus creating transparency and eliminating deception. All participants will be thoroughly debriefed at the end of each experiment where the purpose of the research and their role within it will be re-stated. All questions will be answered as honest and fully as possible.

**Please use the below text box to highlight any other ethical concerns or risks that may arise during your research that have not been covered in this form.**

There are no other ethical concerns that have not addressed in this form.

**Appendix E – Information related to component 1**

## Consent form for participation



Faculty of Science and Technology

### Consent Form

**Research project:** Investigation of the skills and technologies used in the fitting of artificial eyes in Europe with exploration into potential improvements to this process.

**Researcher:** Holly Chinnery – Post Graduate Researcher

**Email:** [hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)

**Telephone number:** 01202 965049

Please read the statements below and sign if you are happy to participate in the research.

- I confirm that I have read and understood the research information for this study and have had the opportunity to ask any questions.
- I understand that my participation is voluntary and that I am free to withdraw at any time without giving a reason and without there being any negative consequences.
- I understand that should I wish not to answer any particular question(s), I am free to decline.
- I understand that I am free to withdraw from the study at any time during the interview, but my responses cannot be withdrawn after 1 week because the written conversation will be anonymised.
- I understand that any data or information used in publications, which arise from this study, will be anonymous.
- I understand that all data will be stored securely and is covered by the data protection act.
- I consent to having the interview recorded on a digital audio recorder.
- I consent to the interview being transcribed.
- I agree to take part in the above research project

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Name of Participant

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Date

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Signature



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Researcher

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Date

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Signature

Thank you very much for your interest in the study. You will receive a copy of the signed and dated consent form and the participant information sheet. Another copy of the signed and dated consent form will be kept separated from your responses, in a secure location.



### **Participant Information Sheet**

**Research project:** Investigation of the skills and technologies used in the fitting procedure of artificial eyes in Europe or Canada with exploration into potential improvements to this process.

You are being invited to take part in a research project. Before you participate it is important for you to understand why the research is being conducted and what it will involve for you. Please take your time to read the following information carefully and ask any questions that you may have about this project.

#### **What is the purpose of the project?**

The purpose of this project is to investigate the current skill and technology base procedures used in the fitting of artificial eyes and to explore potential improvements in this process.

#### **Why have I been invited?**

You have been chosen as you fit into the research criteria of fitting artificial eyes for children in Europe or Canada. This research project will interview 30-35 participants.

#### **Do I have to take part?**

Your participation in this research is voluntary. If you decide to take part you will be given this information sheet to keep and can withdraw at any time during the interview and up to 1 week post interview.

#### **What do I have to do?**

The questionnaire will be a one-time occurrence, which can be done at a suitable time to you lasting between 20-30 minutes.

The questionnaire will begin with a short explanation of what the research will involve and your participation in it. If you agree to take part, you will be asked a series of questions consenting to your

participation in the study. The researcher will then ask you to fill out preliminary questions about your work history and experience prior to the start of the study. We will not ask you any personal details such as your name and contact details, thus you will not be identifiable. If you are interested, we will send you the results after the study have been completed.

The researcher conducting the interviews will be Holly Chinnery, a PhD student in psychology at Bournemouth University. She is supervised by Dr Simon Thompson, Dr Siamak Noroozi and Dr Bryce Dyer at Bournemouth University, UK and advised by Ken Barratt from the National Artificial Eye Service, UK.

**Will my taking part in this project be kept confidential?**

All the information that we collect about you during this research project will be kept strictly confidential. After the study you will have one week to inform us if you want to withdraw from the study. The anonymous data will be kept in a secure place and access will be restricted to the researchers involved in this project. If the results of this research are to be published, you will not be identified in any reports or publications.

**What are the possible benefits, disadvantages and risks of taking part?**

There are no immediate benefits for participating in this research but your opinion is highly valued. The results will help us to gain a better understanding of the current fitting process of artificial eyes and identify any potential improvements that can be made to existing services to improve comfort and to reduce distress for the patient and their families.

If you do not mind sharing your understanding and thoughts on the current process of fitting artificial eyes as well as suggestions for possible improvements, there are no foreseeable disadvantages or risks for taking part in this research.

**Who is funding the research?**

This research is funded by Bournemouth University.

**Contact for further information**

If you have any questions, please do not hesitate to contact Holly Chinnery. You can reach her primarily over email ([hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)), or telephone (01202 965049). For any complaints please contact Dr Simon Thompson over email ([simont@bournemouth.ac.uk](mailto:simont@bournemouth.ac.uk)) or telephone (01202 961558).

Thank you for your interest in this research and for reading the information.

**Appendix F – Information related to component 2**

## Study invitation letter for 13-16 year old participants



### Study Invitation Letter

Dear potential participant

#### **Understanding the lived experience of the fitting process of artificial eyes in 13-16 year olds with a diagnosis of retinoblastoma**

My name is Holly Chinnery: I am a research student at Bournemouth University. I am interested in understanding the lived experience of the fitting process of artificial eyes in young people who have a diagnosis of retinoblastoma. As a young person aged between 13 and 16 years of age who wear artificial eyes as a result of retinoblastoma, I am contacting you to invite you to take part in this study.

Your contribution will be a valuable addition to the research. If you decide to take part, I will come to Birmingham Children's Hospital to interview you. The interview will last approximately 45 minutes. I have included a detailed information sheet about the study.

If you are interested in taking part in the study, please let Maureen McCalla (retinoblastoma nurse specialist based at Birmingham Children's Hospital) know. If you meet the inclusion criteria, Maureen will pass your details onto me and I will then contact you and your parent(s) to discuss the study and arrange a convenient time to conduct the interview.

Thank you.

Yours sincerely

Miss Holly Chinnery

(BSc Hons; MSc, PG Cert).



## YOUNG PERSONS PARTICIPATION INFORMATION SHEET

### UNDERSTANDING THE LIVED EXPERIENCE OF THE FITTING PROCESS OF ARTIFICIAL EYES IN 13-16 YEAR OLDS WITH A DIAGNOSIS OF RETINOBLASTOMA

We would like to invite you to take part in a student research study.

Before you decide if you would like participate in the study, it is really important that you understand what the study is about, why the study is being done and what it would involve for you. Please read and think about this leaflet carefully. Also, talk to your family and nurse about it if you want.

If you have any questions you can ask your parents to give me a call and we can discuss it with both you and your parents. Thank you for reading this.

#### PART 1

##### Who is conducting the research?

The study is a student project being conducted as part of a PhD project investigating the psychological impact of the fitting of artificial eyes in children diagnosed with retinoblastoma and whether improvements can be made via technological advancements. This study is being carried out by the PhD student, Miss Holly Chinnery based at Bournemouth University.



##### Why are we doing this research?

This research is being conducted to understand the lived experience of those wearing artificial eyes as a result of retinoblastoma. By understanding the fitting process of artificial eyes from your perspective, it is hoped that we can reduce potential emotional distress and make recommendations to how the process can be improved.

##### Why have I been invited to take part?

You have been invited because you meet the inclusion criteria of the study. The inclusion criteria for this study is as follows:

- The young person has a confirmed diagnosis of retinoblastoma.
- The young person has 1 or both eyes enucleated as a result of retinoblastoma
- The young person has had 1 or more artificial eye(s) fitted

- The young person is aged between 13 and 16 years of age
- Enucleation and the fitting of artificial eye(s) has taken place in the UK.

You will be a mix of up to ten participants helping us with this study.

### **Do I have to take part?**

No, you do not have to take part in this study. If you are unsure whether to take part in the study, you can talk to your parents, the researcher, the retinoblastoma team or anyone else you so wish too. If participation is agreed, we will ask you to attend the interview with one or more of your parent(s) in order for both of you to sign a consent form agreeing to your participation.

You and your parent(s) will be given a copy of the information sheet and both signed consent forms (one from the parent(s) and one from yourself) to keep for your records.

You are free to withdraw from the study at any point during the interview and up to two weeks' post interview. No reason is needed for withdrawing from the study. This will not affect the ongoing and the standard of care you receive. All data collected before withdrawal will be used for data analysis and will be subject to the confidential guidelines stated below.

### **Where will the interview take place, and will my expenses be paid?**

All interviews will be held at the retinoblastoma outpatient clinic at Birmingham Children's Hospital. Expenses will not be paid.

### **What will happen to me if I take part?**

We will invite you to Birmingham Children's Hospital with at least one of your parents.

The entire research visit will take approximately 45 minutes.

At this visit, you and your parents will have the opportunity to ask any further questions.

Your parent(s) will then be asked to sign a consent form agreeing to you taking part in the study. We will then ask you to sign a consent form also agreeing to your participation. The consent form will also state you understand what the interview entails, that it is voluntary, that you have a right to withdraw from the study before, during and two weeks post-interview and that the interview will be digitally audio-recorded.

You will then be asked to fill in a short demographic questionnaire before the interview. The purpose of this is to provide social and personal context for your responses.

At the start of the interview, you will be asked if you want to have one of your parents sit in the interview with you. Your parents will not be able to contribute to the interview.

We will then start the interview. The researcher will ask you about your experience of having an artificial eye fitted. This includes the impact it has had on you and whether you think any improvements can be made to the current process. You are free to answer the questions in any way that best suits you. You do not need to answer any question that you do not feel comfortable with. If the questions, make you feel uncomfortable or you no longer wish to take part in the study you can ask for it to be stopped. In this situation, all data collected will be used, however will not be identifiable to you.

Once the data has been analysed you may be asked to read a summary of the results to make sure that the views captured are accurate.

### **Will taking part in the study help me?**

The study will not help you right now. However, the results from the study have the potential to help gain a better understanding of the fitting process of artificial eyes in children diagnosed with retinoblastoma. In addition, the results can help identify any potential improvements to the existing process to improve comfort and reduce distress for the patient and their families.





### **What do I do if I don't want to take part in the research anymore?**

You are free to withdraw from the study before, during and two week's post-interview. Withdrawal of interview data will not be possible after two weeks because all identifying information will have been removed. You do not need to give any reason for withdrawing from the study. If you decide to withdraw during the interview, what you have already said will be used.

### **Who should I ask if I have further questions?**

If you have any questions you can talk to your parents, the retinoblastoma team, a member of the research team (details to be found at the end of this sheet) or anyone else you so wish too. Contact regarding this study can be made prior, during and after the study has taken place.

### **What if something goes wrong?**

If there is a problem before, during or after the study, you can talk to your parent's, the researcher or the retinoblastoma team. Any issues that are identified will be treated appropriately and you will be informed of all outcomes.

### **What happens when the research project stops?**

At the end of the study we will collect, transcribe and analyse all responses to the interview questions. This will help in identifying key areas that may need to be addressed in order to improve the current fitting process.

**Thank you for reading so far. If you are still interested, please go to Part 2.**



## **PART 2: FURTHER INFORMATION**

**This is more detailed information that you need to know if you are taking part.**

### **What happens if new information comes along?**

Sometimes in the time it takes to do the study new information comes up. If this happens, we will tell you and we will discuss whether you still want to continue with the study. If you are happy to continue in the study, you and your parent(s) will be asked to sign an updated consent form.

### **What personal data will be accessible and who will have access to it?**

Only members of the direct clinical care team will have access to personal data during the screening process. This is where participants are identified based on whether they meet the inclusion criteria.

Participants who are willing to take part in the study will have their personal data shared with the researcher. This includes name, home/email address and telephone number only. Once the study has been completed, all personal details will be destroyed either by a shredder or by wiping the computer's hard-drive.

### **Will my information be kept confidential? Will anyone else know that I am taking part?**

All information collected as part of this study will be kept strictly confidential in a secure location with access being restricted to the researchers involved in this project. You will be assigned a unique number for identification purposes only and will be used for recording and transcribing purposes. As the interview, will be recorded using a digital audio-recorder voice recognition is possible, thus initially data will not be anonymous. However, all data will be transcribed and the recording deleted within two weeks' post-interview ensuring anonymity of all data. As all identifiable information, will be deleted at this point, withdrawal from the study is not possible two weeks onwards post interview.

All data will be encrypted and uploaded onto a secure server. Following completion of the study, all data will be destroyed either by being shredded (manual data) or by the hard-drive being wiped.

The retinoblastoma team, your parents and the researcher will be the only ones who know you are participating in this study, unless you tell anyone else. Information you share during the interview that highlights above minimal risk of harm to self and/or others will be discussed with those who have a need or right to know like your parents and GP.

### **Who is organising and funding the research?**

The study is being organised by the PhD student, Holly Chinnery and her supervisors. Funding for the study is paid for by Bournemouth University. No funding is associated with this study.

### **What will happen to the results of the research study?**

The results of this study will form part of the researchers thesis. In addition, the study may be published in an academic journal. If the results of this research are to be published, you will not be identified.

### **Who has reviewed the study?**

Before any research is allowed to go ahead it has to be checked and reviewed by an NHS Ethics Committee. This study has been approved by South Central-Berkshire NHS Research Ethics Committee and by the Research Department at Birmingham Children's Hospital.

#### Contact details

*Main researcher*

**Name:** Miss Holly Chinnery

**Job title:** Post-graduate researcher

**Email:** [hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)

**Address:** P104 Poole House, Talbot Campus, Fern Barrow, Bournemouth, Dorset, BH12 5BB

*Researcher's supervisor*

**Name:** Simon Thompson

**Job title:** Dr of Clinical Psychology and Neuropsychology

**Email:** [simont@bournemouth.ac.uk](mailto:simont@bournemouth.ac.uk)

**Address:** 104c Poole House, Talbot Campus, Fern Barrow, Bournemouth, Dorset, BH12 5BB

**If agree to participate in the study, you will be given this information sheet and signed consent forms to keep.**

**Thank you for your time and thinking about taking part in the study – please ask any questions if you need to.**



PARENT INFORMATION SHEET FOR CHILD'S PARTICIAPTION

UNDERSTANDING THE LIVED EXPERIENCE OF THE FITTING PROCESS OF ARTIFICIAL EYES IN 13-16 YEAR OLDS WITH A DIAGNOSIS OF RETINOBLASTOMA

We would like to invite your child to take part in a student research study. Before you decide it is important that you understand what the research is about, why the research is being done and what it will involve for your child.

Please read and think about the information carefully and discuss it with your child.

We will go through the information with you and your child and answer any questions you may have. Take your time in deciding whether or not you want your child to take part.

This information sheet is split into two parts. Part 1 explains the purpose of the study and what will happen to your child if they take part. Part 2 will provide you with more detail about the conduct of the study.

**PART 1 – purpose and involvement in the study**

**What is the purpose of the research?**

We know that the fitting of artificial eyes in children following removal of their natural eye is an important step in rehabilitation: both in terms of functionality and for cosmetic purposes. The fitting process can have an impact on the child's psychological and physical wellbeing. We hope to make suggestions to improve the fitting of artificial eyes through understanding the impact it has on the child patient. Thus, the aim of this study is to understand the lived experience of the fitting process of artificial eyes on the child patient following a diagnosis of retinoblastoma.

**Why has my child been chosen?**

Your child has been chosen to take part as they meet the inclusion criteria of the study. The inclusion criteria for this study is as follows:

- The young person has a confirmed diagnosis of retinoblastoma.
- The young person has 1 or both eyes enucleated as a result of retinoblastoma
- The young person has had 1 or more artificial eye(s) fitted
- The young person is aged between 13 and 16 years of age
- Enucleation and the fitting of artificial eye(s) has taken place in the UK.

Your child will be a mix of up to ten participants helping us with this study.

**Does my child have to take part?**

No, your child does not need to take part. It is up to you and your child whether they participate in the study. If participation is agreed, we will ask you to attend the interview with your child so you *both* can sign a consent form agreeing to their participation. This will also give you and your child the opportunity to ask any more questions you both may have.

You and your child will be given a copy of the information sheet and both signed consent forms to keep for your records.

Your child is free to withdraw from the study at any point during the interview and up to two weeks' post interview. No reason is needed for withdrawing from the study. This will not affect the ongoing and the standard of care your child receives. All data collected before withdrawal will be used for data analysis and will be subject to the confidential guidelines stated below.

### **Where will the interview take place, and will our expenses be paid?**

All interviews will be held at the retinoblastoma outpatient clinic at Birmingham Children's Hospital. Expenses will not be paid.

### **What will happen to my child if they take part?**

We will invite you and your child to Birmingham Children's Hospital for the interview which will last approximately 45 minutes. What the study entails will be restated and you and your child will have the opportunity to ask any questions you may have. If both you and your child agrees to their participation, you will each sign a consent form agreeing to the interview taking place. The consent form will also state you understand what the interview entails, that it is voluntary, that you have a right to withdraw your child from the study before, during and two weeks' post-interview and that the interview will be digitally audio-recorded

Your child will be asked to fill in a short demographic questionnaire before the interview. The purpose of this is to provide social and personal context for their responses.

Your child will be asked if they want you to sit in on the interview with them. If your child agrees to this, you will not be able to contribute to the interview. The researcher will begin the interview by asking your child about their experience of having an artificial eye fitted. This includes the impact it has had on them and whether they think any improvements can be made to the current process. If the questions make your child feel uncomfortable or they no longer wish to take part in the study, they can ask the researcher for it to be stopped. In this situation, all data collected will be used, however will not be identifiable to your child.

Once the data has been analysed your child may be asked to read a summary of the results to make sure that the views captured are accurate.

### **What are the possible disadvantages and risks of taking part?**

There are no foreseeable disadvantages or risks of taking part in the study. However, the study will require your child sitting in a chair for the duration of the interview. Rest breaks will be offered to your child.

Talking about the ongoing care following an illness may be quite distressing for all parties involved. If at any point your child feels that it is causing them an uncomfortable level of distress, they can tell the researcher who will terminate the interview. In addition, we will give your child a list of resources that they may find helpful. Your child is free to end the interview at any point.

### **What are the possible benefits of taking part?**

There are no immediate benefits for participating in this research but your child's opinion is highly valued. The results will help us to gain a better understanding of the current fitting process of artificial eyes and identify any potential improvements that can be made to existing services to improve comfort and to reduce distress for the patient.

### **What happens when the research study stops?**

We will collect, transcribe and analyse all responses to the interview questions. This will help in identifying key areas that may need to be addressed in order to improve the current fitting process.

### **What if there is a problem?**

If there are any concerns about the way your child has been treated during the study or any possible harm they might suffer will be addressed. Detailed information on this is provided in Part 2.

**Will taking part in the research project be kept confidential?**

Yes. We will follow ethical and legal practice and all information about your child will be handled in confidence. Detailed information about this can be found in Part 2.

**Contact for further information**

If you would like any further information about this study you can contact:

**Name:** Holly Chinnery

**Job title:** Post-graduate Researcher

**Address:** P104, Poole House, Talbot Campus, Bournemouth University, Fern Barrow, Dorset, BH12 5BB.

**Email address:** [hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)

**If you are interested in participating in this study, please continue to read the additional information in Part 2 before making any decision.**

## **PART 2: Further information you need to know if you still want your child to take part.**

### **What if new information becomes available?**

Sometimes in the time it takes to do the study new information comes up. If this happens, we will tell you and your child and we will discuss whether your child still wants to continue with the study. This will not affect their participation in the study. If your child decides to continue in the study you will be asked to sign an updated consent form.

### **What will happen if we do not want to carry on with the research?**

If your child withdraws from the study, we will destroy your identifiable data forms, but we will need to use the data collected up to the withdrawal.

### **What if there is a problem?**

If you or your child has a concern with any aspect of the study, you both can speak to the researcher who will do their best to answer your questions. Furthermore, you can talk to the researcher's supervisors and/or the retinoblastoma team.

**Name:** Holly Chinnery

**Job title:** Post-graduate Researcher

**Address:** P104, Poole House, Talbot Campus, Bournemouth University, Fern Barrow, Dorset, BH12 5BB.

**Email address:** [hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)

If you remain unhappy and wish to complain formally, you can do this through the researcher's supervisor or the hospitals Patient Advice & Liaison Team:

**Name:** Dr Simon Thompson

**Job title:** Dr of Clinical Psychology and Neuropsychology

**Address:** P104c, Poole House, Talbot Campus, Bournemouth University, Fern Barrow, Dorset, BH12 5BB.

**Email address:** [simont@bournemouth.ac.uk](mailto:simont@bournemouth.ac.uk)

**Department:** Patient Advice & Liaison

**Telephone:** 0121 333 8403/0121 333 8611

### **What personal data will be accessible and who will have access to it?**

Only members of the direct clinical care team will have access to personal data during the screening process. This is where participants are identified based on whether they meet the inclusion criteria.

Participants who are willing to take part in the study will have their personal data shared with the researcher. This includes name, home/email address and telephone number only. Once the study has been completed, all personal details will be destroyed either by a shredder or by wiping the computer's hard-drive.

### **Will taking part in this study be kept confidential?**

All information collected as part of this study will be kept strictly confidential in a secure location with access being restricted to the researchers involved in this project. Your child will be assigned a unique number for identification purposes only and will be used for recording and transcribing purposes. As the interview, will be recorded using a digital audio-recorder voice recognition is possible, thus initially data will not be anonymous. However, all data will be transcribed and the recording deleted within two weeks' post-interview ensuring anonymity of all data. As all identifiable information, will be deleted at this point, withdrawal from the study is not possible two weeks onwards post interview.

All data will be encrypted and uploaded onto a secure server. Following completion of the study, all data will be destroyed either by being shredded (manual data) or by the hard-drive being wiped.

The retinoblastoma team, the child's parent(s) and the researcher will be the only ones who know that your child is participating in this study, unless they tell anyone else. Information your child shares during the interview that highlights above minimal risk of harm to self and/or others will be discussed with those who have a need or right to know. This includes yourselves, the retinoblastoma team and the child's GP.

#### **Who is organising and funding the research?**

The study is being organised by the PhD student, Holly Chinnery and her supervisors. Funding for the study is paid for by Bournemouth University. No funding is associated with this study.

#### **What will happen to the results of the research study?**

The results of this study will form part of the researchers thesis. In addition, the study may be published in an academic journal. If the results of this research are to be published, you will not be identified.

#### **Who has reviewed the study?**

Before any research is allowed to go ahead it has to be checked by South Central-Berkshire NHS Research Ethics Committee. They make sure that the research is fair. This study has been reviewed by the NHS Research Ethics Committee and approved by the Research Department at this hospital.

#### **How can I find out more about the research?**

For further information about taking part in the study, please contact the main researcher in the first instance. Contact details can be found below.

##### Contact details

###### *Main researcher*

**Name:** Miss Holly Chinnery

**Job title:** Post-graduate researcher

**Email:** [hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)

**Address:** P104 Poole House, Talbot Campus, Fern Barrow, Bournemouth, Dorset, BH12 5BB

###### *Researcher's supervisor*

**Name:** Simon Thompson

**Job title:** Dr of Clinical Psychology and Neuropsychology

**Email:** [simont@bournemouth.ac.uk](mailto:simont@bournemouth.ac.uk)

**Address:** 104c Poole House, Talbot Campus, Fern Barrow, Bournemouth, Dorset, BH12 5BB

**If you and your child agree to their participation in the study, they will be given this information sheet and signed consent forms to keep.**

**Thank you for your time and thinking about your child taking part in the study – please ask any questions if you need to.**



13-16 year olds consent form for participation



CONSENT FORM FOR YOUNG PERSON'S PARTICIPATION

**Understanding the lived experience of the fitting process of artificial eyes in 13-16 year olds with a diagnosis of retinoblastoma**

**Name of Researcher:** Holly Chinnery

**Please initial box**

I (name) ..... understand that my parent(s)/guardian(s) have given permission for me to participate in a study concerning the fitting of artificial eyes in 13-16 year olds with retinoblastoma.

I have had the opportunity to ask questions about the study and understand

what is involved. I agree to taking part in the research and the possibility of being asked to read a summary of the results to make sure that the views captured

are accurate

I understand that the study is voluntary and I have been told that I may stop my participation in the study at any time during and up to two weeks post interview without it impacting the care I receive. However, any data collected will

be used but anonymised.

I understand that my personal details (name, home address, email address telephone number) will be given to the researcher for contact purposes only.

and

Once the study is completed all data will be destroyed.

I understand that all data will be stored securely and is covered by the data protection act and the NHS Code of Confidentiality.

protection act and

I consent to having the interview recorded on a digital audio recorder, which will be transcribed. I understand that the recorded responses are not anonymous due to possible voice recognition, but the responses will be anonymised as soon

as the recordings are transcribed (typed out), and the audio recordings will be destroyed.

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Name of participant:

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Date

---

Signature:

---

Name of researcher:

---

Date

---

Signature:

If you would like to know the results of this study, please provide an email or home address where the researcher can contact you.

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Thank you very much for your interest in the study. You will receive a copy of the signed and dated consent form and the participant information sheet. Another copy of the signed and dated consent form will be kept separated from your responses, in a secure location.

Parent of 13-16 year old's participation consent form



PARENTS CONSENT FORM FOR THEIR CHILD PARTICIPATION

**Understanding the lived experience of the fitting process of artificial eyes in 13-16 year olds with a diagnosis of retinoblastoma**

**Name of Researcher:** Holly Chinnery

**Please initial box**

I (name of relative) ..... have been consulted about (name of child) ..... participation in this research project. I have had the opportunity to ask questions about the study and understand what is involved. I agree to their taking part in the research and the possibility of (name of child) ..... being asked to read a summary of the results to make sure that the views captured are accurate.

I understand that the study is voluntary and I can request (name of child) ..... is withdrawn from the study at any time during or up to two weeks post interview, without giving any reason and without their care or legal rights being affected. However, any data collected will be used but anonymised.

I understand that our personal details (name, home address, email address and telephone number) will be given to the researcher for contact purposes only. Once the study is completed all data will be destroyed.

I understand that all data will be stored securely and is covered by the data protection act and the NHS Code of Confidentiality.

I consent to having the interview recorded on a digital audio recorder,

which will be transcribed. I understand that the recorded responses are not anonymous due to possible voice recognition, but the responses will be

anonymised as soon as the recordings are transcribed (typed out), and

the audio recordings will be destroyed.

**Date:**

\_\_\_\_\_  
Name of relative:                                  Name of child                                  Signature:

Relationship to participant: \_\_\_\_\_

\_\_\_\_\_  
Researcher:    Signature:

If you would like to know the results of this study, please provide an email or home address where the researcher can contact you.

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Thank you very much for your interest in the study. You will receive a copy of the signed and dated consent form and the participant information sheet. Another copy of the signed and dated consent form will be kept separated from your responses, in a secure location.

## Debrief for 13-16 year olds participation



### Debrief

#### UNDERSTANDING THE LIVED EXPERIENCE OF THE FITTING PROCESS OF ARTIFICIAL EYES IN 13-16 YEAR OLDS WITH A DIAGNOSIS OF RETINOBLASTOMA

Thank you for agreeing to take part in the interview which looked at understanding the lived experience of the fitting process of artificial eyes in 13-16 year olds with a diagnosis of retinoblastoma. This study will be presented as part of my PhD thesis, may be published in an academic journal and presented during conference's.

This study was supervised by Dr Simon Thompson, Professor Siamak Noroozi, Dr Bryce Dyer and Dr Karen Rees of Bournemouth University and advised by Ken Barratt of the National Artificial Eye Service, Blackpool.

If at any time during If you decide that you wish to withdraw from the study you may do so by contacting me up to two weeks' post-interview.

If you need to contact me, please do so using the following details:

**Name:** Miss Holly Chinnery

**Job title:** Post-graduate Researcher

**Address:** P104, Poole House, Talbot Campus, Bournemouth University, Fern Barrow, Dorset, BH12 5BB.

**Email address:** [hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)



## PARTICIPATION INFORMATION SHEET

### UNDERSTANDING THE LIVED EXPERIENCE OF THE FITTING PROCESS OF ARTIFICIAL EYES IN PARENTS OF A CHILD WITH A DIAGNOSIS OF RETINOBLASTOMA

We would like to invite you to take part in a student research study.

Before you decide if you would like participate in the study, it is really important that you understand what the study is about, why the study is being done and what it would involve for you. Please read and think about this leaflet carefully.

If you have any questions please speak to the researcher. Thank you for reading this.

#### **PART 1**

##### **Who is conducting the research?**

The study is a student project being conducted as part of a PhD project investigating the psychological impact of the fitting of artificial eyes in children diagnosed with retinoblastoma and whether improvements can be made via technological advancements. This study is being carried out by the PhD student, Miss Holly Chinnery based at Bournemouth University.

##### **Why are we doing this research?**

This research is being conducted to understand the lived experience of parents of children who wear an artificial eye(s) as a result of retinoblastoma. By understanding the fitting process of artificial eyes from your perspective, it is hoped that we can reduce potential emotional distress and make recommendations to how the process can be improved.

##### **Why have I been invited to take part?**

You have been invited because you meet the inclusion criteria of the study. The inclusion criteria for this study is as follows:

- You are a parent of a child with a confirmed diagnosis of retinoblastoma.
- Your child has 1 or both eyes enucleated as a result of retinoblastoma
- Your child has had 1 or more artificial eye(s) fitted.
- Enucleation and the fitting of artificial eye(s) has taken place in the UK.

You will be a mix of up to six participants helping us with this study.

**Do I have to take part?**

No, you do not have to take part in this study. If participation is agreed, we will ask you to sign a consent form agreeing to your participation and complete a demographic form and the questionnaire.

You are free to withdraw from the study at any point during and up to two weeks' post study. No reason is needed for withdrawing from the study. All data collected before withdrawal will be used for data analysis and will be subject to the confidential guidelines stated below.

**Where will the study take place?**

As the study is an online questionnaire, you can take part in the study at your home at a time that is most convenient for you.

**What will happen to me if I take part?**

We will email you asking for your participation in this research study. The email will contain the study invitation letter, participation information sheet and consent forms. If you agree to participate we will ask you to sign a consent form. Once you have done this, we will ask you to fill out a brief demographic questionnaire and then the main questionnaire. This will then conclude the end of the study. A debrief of the study will then be sent to you. The entire study should take approximately 15-30 minutes.

**Will taking part in the study help me?**

The study will not help you right now. However, the results from the study have the potential to help gain a better understanding of the fitting process of artificial eyes in children diagnosed with retinoblastoma. In addition, the results can help identify any potential improvements to the existing process to improve comfort and reduce distress for the patient and their families.

**What do I do if I don't want to take part in the research anymore?**

You are free to withdraw from the study before, during and two week's post-study. Withdrawal of interview data will not be possible after two weeks because all identifying information will have been removed. You do not need to give any reason for withdrawing from the study. If you decide to withdraw during the interview, what you have already said will be used.

**Who should I ask if I have further questions?**

If you have any questions you can talk to the researcher (details to be found at the end of this sheet) or anyone else you so wish too. Contact regarding this study can be made prior, during and after the study has taken place.

**What if something goes wrong?**

If there is a problem before, during or after the study, you can talk to the researcher. Any issues that are identified will be treated appropriately and you will be informed of all outcomes.

**What happens when the research project stops?**

At the end of the study we will collect, transcribe and analyse all responses to the questionnaire. This will help in identifying key areas that may need to be addressed in order to improve the current fitting process.

**Thank you for reading so far. If you are still interested, please go to Part 2.**

## **PART 2: FURTHER INFORMATION**

**This is more detailed information that you need to know if you are taking part.**

### **What happens if new information comes along?**

Sometimes in the time it takes to do the study new information comes up. If this happens, we will tell you and we will discuss whether you still want to continue with the study. If you are happy to continue in the study, you will be asked to sign an updated consent form.

### **What personal data will be accessible and who will have access to it?**

Participants who are willing to take part in the study will have their personal data shared with the researcher. This includes name, home/email address and telephone number only. Once the study has been completed, all personal details will be destroyed either by a shredder or by wiping the computer's hard-drive.

### **Will my information be kept confidential? Will anyone else know that I am taking part?**

All information collected as part of this study will be kept strictly confidential in a secure location with access being restricted to the researchers involved in this project. You will be assigned a unique number for identification purposes only and will be used for transcribing purposes.

All data will be encrypted and uploaded onto a secure server. Following completion of the study, all data will be destroyed either by being shredded (manual data) or by the hard-drive being wiped.

### **Who is organising and funding the research?**

The study is being organised by the PhD student, Holly Chinnery and her supervisors. Funding for the study is paid for by Bournemouth University. No funding is associated with this study.

### **What will happen to the results of the research study?**

The results of this study will form part of the researchers thesis. In addition, the study may be published in an academic journal. If the results of this research are to be published, you will not be identified.

### **Who has reviewed the study?**

Before any research is allowed to go ahead it has to be checked and reviewed by an Ethics Committee. This study has been approved by Bournemouth University's Ethics Committee.

#### Contact details

##### *Main researcher*

**Name:** Miss Holly Chinnery

**Job title:** Post-graduate researcher

**Email:** [hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)

**Address:** P104 Poole House, Talbot Campus, Fern Barrow, Bournemouth, Dorset, BH12 5BB

##### *Researcher's supervisor*

**Name:** Simon Thompson

**Job title:** Dr of Clinical Psychology and Neuropsychology

**Email:** [simont@bournemouth.ac.uk](mailto:simont@bournemouth.ac.uk)

**Address:** 104c Poole House, Talbot Campus, Fern Barrow, Bournemouth, Dorset, BH12 5BB



**If agree to participate in the study, you will be given this information sheet and signed consent forms to keep.**

**Thank you for your time and thinking about taking part in the study – please ask any questions if you need to.**

**Consent form for parents participation**



**CONSENT FORM FOR PARTICIPATION**

**Understanding the lived experience of the fitting process of artificial eyes in parents of a child with a diagnosis of retinoblastoma**

**Name of Researcher:** Holly Chinnery

**Please initial box**

I (name) ..... agree to participate in a study concerning the fitting of artificial eyes in parents of a child with retinoblastoma.

I have had the opportunity to ask questions about the study and understand what is involved. I agree to the possibility of being asked to read a summary of the results to make sure that the views captured are accurate

I understand that the study is voluntary and I have been told that I may stop my participation in the study at any time during and up to two weeks post interview.

I understand that my personal details (name, home address, email address and telephone number) will be given to the researcher for contact purposes only.

Once the study is completed all data will be destroyed.

I understand that all data will be stored securely and is covered by the Data Protection Act.



## Debrief for parents participation



### Debrief

#### UNDERSTANDING THE LIVED EXPERIENCE OF THE FITTING PROCESS OF ARTIFICIAL EYES IN PARENTS OF A CHILD WITH A DIAGNOSIS OF RETINOBLASTOMA

Thank you for agreeing to take part in the online questionnaire which looked at understanding the lived experience of the fitting process of artificial eyes in parents of a child with a diagnosis of retinoblastoma. This study will be presented as part of my PhD thesis, may be published in an academic journal and presented during conference's.

This study was supervised by Dr Simon Thompson, Professor Siamak Noroozi and Dr Bryce Dyer of Bournemouth University and advised by Ken Barratt of the National Artificial Eye Service, Blackpool.

If you decide that you wish to withdraw from the study you may do so by contacting me up to two weeks' post-interview.

If you need to contact me, please do so using the following details:

**Name:** Miss Holly Chinnery

**Job title:** Post-graduate Researcher

**Address:** P104, Poole House, Talbot Campus, Bournemouth University, Fern Barrow, Dorset, BH12 5BB.

**Email address:** [hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)

**Appendix G – Information related to component 3**

Consent form for participation



Consent Form

Faculty of Science and Technology

**Research project:** Comparing the processes and technology employed in the assessment stage of the maxillofacial prosthetics and artificial eye development.

**Researcher:** Holly Chinnery – Post Graduate Researcher

**Email:** [hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)

**Telephone number:** 01202 965049

Please read the statements below and sign if you are happy to participate in the research.

- I confirm that I have read and understood the research information for this study and have had the opportunity to ask any questions.
- I understand that my participation is voluntary and that I am free to withdraw at any time without giving a reason and without there being any negative consequences.
- I understand that should I wish not to answer any particular question(s), I am free to decline.
- I understand that I am free to withdraw from the study at any time during the interview, but my responses cannot be withdrawn after 1 week because the written conversation will be anonymised.
- I understand that any data or information used in publications, which arise from this study, will be anonymous.
- I understand that all data will be stored securely and is covered by the data protection act.
- I consent to having the interview recorded on a digital audio recorder.
- I consent to the interview being transcribed.
- I agree to take part in the above research project

_____	_____	_____
Name of Participant	Date	Signature
_____	_____	_____
Researcher	Date	Signature

Thank you very much for your interest in the study. You will receive a copy of the signed and dated consent form and the participant information sheet. Another copy of the signed and dated consent form will be kept separated from your responses, in a secure location.



## Participant Information Sheet

**Research project:** Comparing the processes and technology employed in maxillofacial prosthetics and artificial eye development.

You are being invited to take part in a research project. Before you participate it is important for you to understand why the research is being conducted and what it will involve for you. Please take your time to read the following information carefully and ask any questions that you may have about this project.

### **What is the aim of the project?**

This study aims to create a transfer of knowledge between the methods employed in the assessment stage of the maxillofacial prosthetic and artificial eye process to assess whether improvements can be made in the development of artificial eyes.

### **Why have I been invited?**

You have been chosen as you fit into the research criteria of being an maxillofacial or artificial eye prosthetist. This research project will interview 8 to 16 participants.

### **Do I have to take part?**

Your participation in this research is voluntary. If you decide to take part you will be given this information sheet to keep and can withdraw at any time during the interview and up to 1 week post interview. After one week the audio recordings will be transcribed, thus, the data will not be identifiable to you.

### **What do I have to do?**

The interview will be a one-time occurrence, which will be held at your place of work lasting between 30 – 45 minutes.

The interview will begin with a short explanation of what the research will involve and your participation in it. If you agree to take part, you will be asked to sign a consent form agreeing to your participation in the study and that the interview will be audio-recorded. You will receive a copy of the information sheet and the consent form. The researcher will then ask you to fill out a brief questionnaire prior to the start of the interview. The questionnaire is to ascertain your work history and experience. At the end of the interview you have the opportunity to ask any additional questions you may have. Everything you discuss will be kept confidential.

Please be aware that the responses you give will not be anonymised during the interview, but will be once it has been transcribed. Your name, contact information and consent forms will be kept secured and separated from your responses so no one can retrace specific participants' responses. If you are interested, we will send you the results after the study have been completed.

The researcher conducting the interviews will be Holly Chinnery, a PhD student in psychology at Bournemouth University. She is supervised by Dr Simon Thompson, Dr Siamak Noroozi, Dr Bryce Dyer and Dr Karen Rees at Bournemouth University and advised by Ken Barratt from the National Artificial Eye Service.

**Will my taking part in this project be kept confidential?**

All the information that we collect about you during this research project will be kept strictly confidential. After the interview you will have one week to inform us if you want to withdraw from the study. Once your responses have been transcribed into written form, everything you said will be anonymised and the audio recordings will be deleted. This means that from this point onwards we cannot withdraw your data, as we will be unable to identify your response to you. The anonymous data will be kept in a secure place and access will be restricted to the researchers involved in this project. If the results of this research are to be published, you will not be identified in any reports or publications.

**What are the possible benefits, disadvantages and risks of taking part?**

There are no immediate benefits for participating in this research but your opinion is highly valued. The results will help us to gain a better understanding of whether current technological advancements can be incorporated in the artificial eye process.

If you are able to sit on a chair for 30 – 45 minutes and do not mind sharing your understanding and thoughts, there are no foreseeable disadvantages or risks for taking part in this research.



**Who is funding the research?**

This research is funded by Bournemouth University.

**Contact for further information**

If you have any questions, please do not hesitate to contact Holly Chinnery. You can reach her primarily over email ([hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)), or telephone (01202 965049). For any complaints please contact Dr Simon Thompson over email ([simont@bournemouth.ac.uk](mailto:simont@bournemouth.ac.uk)) or telephone (01202 961558).

**Thank you for your interest in this research and for reading the information.**

## Demographic/Pre-interview questions



Faculty of Science and Technology

## Demographic/Pre-interview questions

Thank you for volunteering to partake in this research. Individual responses/documents will be kept confidential and secure. Results will not be shared with any third parties.

**Please circle all those that apply.**

### Job title:

- Maxillofacial prosthetist
- Artificial eye prosthetist
- Other (please state): \_\_\_\_\_

### Job qualifications:

- Please state: \_\_\_\_\_

### Length of service:

- Please state: \_\_\_\_\_

### Employment status (if applicable):

- Employed
- Self-employed
- Other (please state): \_\_\_\_\_

### Gender:

- Male
- Female

### Ethnicity:

- White
- Hispanic or Latino
- Black or African American
- Native American or American Indian
- Asian / Pacific Islander
- Other (please state): \_\_\_\_\_

**Age:**

- Please state \_\_\_\_\_

**Thank you for your time. Please ask any questions of you need to.**



### Interview Questions

#### **Transfer of knowledge study between the technologies employed in maxillofacial prosthesis and artificial eye processes.**

Thank you for volunteering to partake in this research. Individual responses/documents will be kept confidential and secure. Individual results will not be shared with any third parties.

1. Tell me how you conduct an assessment for a prosthetic fitting?
2. What information is needed in the assessment process?
3. What equipment/tools do you use for assessing the deficit site?
4. What measurements do you take in the assessment process?
5. Have you experienced any complications with the equipment/tools used in the assessment stage?
6. What do you feel are the benefits of the assessment process?
7. What do you feel are the weaknesses of the assessment process?
8. List your priorities (in order) that determine the success of the assessment process.
9. How do you think the equipment/tools used in the assessment affect the overall function of a prosthesis ?
10. How do you think the equipment/tools used in the assessment affect the overall success of a prosthesis?
11. What are usually the reasons for the assessment process to fail (technological factors, patient factors)?
12. Do you perform any 'checking procedures' in the assessment process?
13. Can you think of anything that would improve the current assessment process?

## Debrief



Faculty of Science and Technology

## Debrief

### COMPARING THE PROCESSES AND TECHNOLOGY EMPLOYED IN THE ASSESSMENT STAGE OF THE MAXILLOFACIAL PROSTHETICS AND ARTIFICIAL EYE DEVELOPMENT

Thank you for agreeing to take part in the interview which aimed to create a transfer of knowledge between the methods employed in the maxillofacial prosthetic and artificial eye process to assess whether improvements can be made in the development of artificial eyes.

This study will be presented as part of my PhD thesis, may be published in an academic journal and presented during conference's.

This study was supervised by Dr Simon Thompson, Professor Siamak Noroozi, Dr Bryce Dyer and Dr Karen Rees of Bournemouth University and advised by Ken Barratt of the National Artificial Eye Service, Blackpool.

If at any time during If you decide that you wish to withdraw from the study you may do so by contacting me up to two weeks' post-interview.

If you need to contact me, please do so using the following details:

**Name:** Miss Holly Chinnery

**Job title:** Post-graduate Researcher

**Address:** P104, Poole House, Talbot Campus, Bournemouth University, Fern Barrow, Dorset, BH12 5BB.

**Email address:** [hchinnery@bournemouth.ac.uk](mailto:hchinnery@bournemouth.ac.uk)