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REVIEW



Factors associated with joint contractures in adults: a systematic review with narrative synthesis

Hina Tariq^a, Kathryn Collins^a, Desiree Tait^a, Joel Dunn^b, Shafaq Altaf^c and Sam Porter^a

^aFaculty of Health and Social Sciences, Bournemouth University, Bournemouth, UK; ^bDorset Healthcare University Foundation Trust, Poole, UK; ^cShifa Tameer-e-Millat University, Islamabad, Pakistan

ARSTRACT

Purpose: The primary objective of the review was to collate the available evidence on factors associated with joint contractures in adults.

Methods: A systematic literature search was conducted on MEDLINE, CINAHL, AMED, and EMBASE. Studies that involved participants aged ≥18 and assessed joint contracture as a primary or secondary outcome were included. Two independent reviewers screened studies against the eligibility criteria, performed data extraction, and assessed the quality of evidence. A narrative synthesis by domain and subdomain was undertaken. The protocol was registered on PROSPERO: CRD42019145079.

Results: Forty-seven studies were included in the review. Identified factors were broadly classified into three major domains: sociodemographic factors, physical factors, and proxies for bed confinement. Sociodemographic factors were not associated with joint contractures. Functional ability, pain, muscle weakness, physical mobility, and bed confinement provided the most consistent evidence of association with joint contractures. The evidence regarding the relationship between spasticity and joint contractures remains unclear. Other factors might be important, but there was insufficient evidence to make inferences.

Conclusions: The review identified and collated evidence on factors associated with joint contractures, which can be utilised to develop effective prevention and management strategies.

> IMPLICATIONS FOR REHABILITATION

- Clinical interventions based on the timely identification of risks related to joint contractures in vulnerable adults have the potential to prevent or ameliorate their development or progression.
- Quality and consistency of care for vulnerable adults would be enhanced by developing effective joint contracture prevention and rehabilitation strategies based on the evidence presented in this review.
- As many vulnerable adults are located in the community or non-acute care settings, strategies should target these loci of care.
- Structured risk assessments that can support non-physiotherapy staff working in these loci of care to identify risks related to joint contractures would provide an important resource for risk management.

ARTICLE HISTORY

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KEYWORDS

Contracture; range of motion; PROM; joint; adult; associated factor; risk factor

Introduction

Joint contractures, commonly defined as a limitation in the passive joint range of motion (PROM), usually develop following structural alterations within the periarticular connective tissue(s) [1]. The connective tissue changes prevent movement of the involved joint(s) through its full available range of motion (ROM). Both intra-articular tissues involving bone, cartilage, and capsules, as well as extra-articular tissues, such as muscles, tendons, and skin, can restrict a joint from moving through its full available ROM [2]. The type of connective tissue involved in joint movement restriction usually defines the type of contracture developed; however, multiple tissues can have underlying involvement, and it is often difficult to identify a single origin of joint restriction [2]. As a result, contractures can further increase the risk of physical impairments; consequently, there is difficulty in performing self-care, restrictions in physical mobility, and social activities [3]. This, in turn, leads to a vicious cycle of further immobility, exacerbation of existing or formation of new joint contractures, and decreased quality of life (QOL) [4].

There are three different types of joint contractures based on the underlying tissue involved: myogenic, arthrogenic, and soft tissue contractures. Myogenic contractures denote a reduction in muscle length leading to a limitation in both active and PROM [2], commonly seen in neurological conditions, e.g., brain and spinal cord injury (SCI), multiple sclerosis (MS), and cerebral palsy (CP) or after a prolonged period of immobility such as bed confinement in the intensive care unit (ICU). Arthrogenic contractures

CONTACT Hina Tariq 🔯 htariq@bournemouth.ac.uk 🖻 Faculty of Health and Social Sciences, Bournemouth University, Bournemouth, UK



are usually coupled with pain and involve prominent changes in bone, cartilage, and the joint capsule [5]. This may result from damage and/or tightening of connective tissue, such as in osteoarthritis (OA), systemic sclerosis (SSc), osteochondritis, and intraarticular fractures [6]. Finally, soft tissue contractures, also known as scar contractures, involve cutaneous, subcutaneous, and loose connective tissue around the joint [2]. These are frequently seen in soft-tissue injuries like burns and open wounds. Regardless of tissue involvement, all types of contractures significantly impact functional ability and physical mobility [7].

Depending upon the underlying pathology, joint contractures can also be classified as progressive or non-progressive. Progressive contractures are acquired, associated with chronic conditions like an injury to the brain, or spinal cord, arthritic diseases, and surgical repair procedures such as total knee arthroplasty (TKA), and are usually associated with extrinsic factors. Examples of extrinsic factors encompass restricted joint ROM, reduced physical mobility, muscle weakness, spasticity, impaired cognition, and pain [8-10]. In contrast, non-progressive contractures are usually congenital, affect multiple joints and limbs and are associated with genetic causation, e.g., arthrogryposis multiplex congenita.

This review will focus on progressive myogenic and arthrogenic joint contractures and their associated factors.

Epidemiology of contractures

Data regarding the epidemiology of contractures are underreported and record a wide range of prevalence and incidence. It is likely that this large variation is partially artifactual and attributable to the lack of a universally accepted definition of contractures, poorly understood aetiology, and/or lack of a standardised measure for the screening and assessment of contractures [8]. However, there is also evidence of an objective variance in prevalence related to different conditions [11].

The development of contractures is a commonly reported secondary impairment associated with chronic neurological and musculoskeletal conditions. The prevalence of contractures in brain injuries ranges from 16.2% up to 67% [12-16]; overall incidence of contractures in at least one joint in SCI was reported to be 66% [17]; 56% in MS [18], and 24% in Alzheimer's disease [19]. Ritter et al. [20], in their large retrospective cohort study, reported that 93% of patients with knee contractures who presented for the TKA had a diagnosis of OA, 5% had rheumatoid arthritis (RA), and 1.1% had osteonecrosis [20]. The prevalence of joint involvement in SSc, including the occurrence of contractures, also ranges widely between 46% and 97% [21].

Impact of joint contractures on function and quality of life

The presence of joint contractures is a self-limiting problem that leads to sequelae of further decline in mobility, function, and complications such as abnormal positioning, pain, pressure sores, skin breakdown, depressive symptoms, osteoporosis, and fractures, ultimately affecting the overall QOL [22,23]. In addition, upper limb joint contractures decrease the ability to perform selfcare activities such as eating, dressing, and bathing, whereas lower limb joint contractures might limit one's ability to walk independently, entailing a higher risk of fall and bed confinement [7,22,24].

A study by Heise et al. [4] on 294 older individuals residing in geriatric settings demonstrated a significant association between functioning, disability, and QOL among individuals living with joint contractures. Recent studies have identified several domains of limitations relevant to contractures using the biopsychosocial model provided by the International Classification of Functioning (ICF) [3,4,8]. The most frequently identified problems with joint contractures were associated with activity limitation, participation restriction [4], mobility, muscle power, and pain [25]. Fischer et al. [26] described the impact of joint contractures from the patients' perspective on multidimensional components of functioning and disability. These included pain, emotional distress, difficulty in performing activities of daily living (ADLs) like walking, climbing stairs, house chores, shopping, etc., and increased dependency on assistive devices and caregivers [26].

Rationale for the systematic review

Although the primary literature on joint contractures is growing, it still lacks an in-depth understanding of the role of proximate, ultimate, and associated factors [8]. Previous reviews have explored the risk factors associated with joint contractures; however, they were limited in the scope of the search restricted to one database [11] or restricted to exploring only the elderly population [24]. The lack of evidence limits the ability of caregivers and health care clinicians to identify the risk of joint contracture development in a timely fashion and thus early diagnosis and initiation of early intervention [24].

This systematic review aims to identify and collate the factors associated with progressive myogenic or arthrogenic contractures. It is hoped that its findings will aid the identification of individuals at risk of contracture development or progression. Earlier identification and management of contractures may impact an individual's ability to maintain independence with ADLs and functional mobility contributing to improved QOL.

Methods

This systematic review conforms to the updated guidance on Preferred Reporting Items for Systematic Reviews and Meta-Analyses-Protocols (PRISMA-P) checklist [27]. The protocol of this review was registered on the International Prospective Register of Systematic Reviews (PROSPERO) database: CRD42019145079.

The following bibliographic electronic databases were searched: MEDLINE, CINAHL, EMBASE, and AMED from January 1999 to January 2022. Individual search strategies were developed for each database to account for differences in Thesaurus terminology and indexing. A complete search strategy for MEDLINE can be found in Supplementary information (A). Restrictions were applied for date of publication, age (adults), and humans where applicable. The reference lists of all relevant papers and documents were also screened for references not identified in the database search.

Study eligibility

The selection of studies was based on the Population, Exposure, and Outcome (PEO) framework [28].

Population

Studies involving human participants aged 18 years or above who developed progressive arthrogenic or myogenic contracture(s) as a secondary consequence of a primary condition (e.g., brain and SCI, OA, etc.), orthopaedic surgery, or a period of prolonged immobility were included. Studies involving children, non-



progressive contractures, and scar contractures were excluded as the underlying aetiology in these populations is mostly congenital and intrinsic, and do not fall into the scope of this review. Studies that included both children and adults as participants were only considered if the results for adults were presented separately. Moreover, studies on conditions in which contractures formed a part of the primary diagnostic criteria, especially in idiopathic conditions such as Dupuytren's disease and adhesive capsulitis, were also excluded.

Exposure of interest

The exposure of interest was factors associated with joint contractures, for example, physical mobility, pain, cognition, or functional ability. Intrinsic or disease-specific factors, genetic, congenital, pharmacological, and surgical factors were excluded.

Outcome

Studies which included joint contractures as a primary or secondary outcome were included. Joint contractures in this review are operationally defined as a limitation or reduction in the joint PROM.

Types of studies

The review included prospective and retrospective cohort studies, case-control studies, and cross-sectional analytical studies. Secondary analysis of interventional studies where baseline data were obtained were also included. Biological and qualitative studies were excluded, as were case series, individual case reports, theses, conference abstracts, letters, commentaries, or books without primary data or quantitative outcomes. Studies published in languages other than English were also excluded due to a lack of resources for the translation of data.

Study selection

Studies retrieved from the electronic search were collated and uploaded into Endnote reference manager v9 (Clarivate Analytics, Philadelphia, PA), and duplicates were removed. A database record was also maintained using Microsoft Excel (Redmond, WA), detailing each review stage. Two independent reviewers then screened the titles (HT and KC/JD), abstracts, and full texts (HT and SA) based on the inclusion criteria. Where necessary, any discrepancies or disagreements were resolved through discussion or using a third reviewer (SP/KC/JD). Reasons for exclusion at the full-text stage are documented in the PRISMA flowchart (Figure 1).

Assessment of methodological quality

The methodological quality of the eligible studies was assessed by two independent reviewers (HT and SA) using methodologically appropriate critical appraisal checklists from the Joanna Briggs Institute (JBI) [29]. These included checklists for cohort studies, case-control studies, analytical cross-sectional studies, randomised controlled trials, and quasi-experimental studies. The discrepancies and disagreements were resolved through discussion or using a third reviewer where necessary (KC/JD).

Data extraction and synthesis

Data from the included studies were extracted by two independent reviewers (HT and SA) in an excel sheet in accordance with the PRISMA guidelines [30]. The extracted information included but was not limited to the following: author names, publication date, country of origin, study characteristics (e.g., study design, setting), participant characteristics (e.g., sample size, age, gender), the definition of contractures, methods of assessment used for outcomes and exposures, and relevant study findings. If consensus was not reached, the discrepancies or disagreements were resolved through discussion or using a third reviewer (KC/JD). A meta-analysis of the data was not possible because of a lack of homogeneity between the studies regarding the study population, setting, and outcomes; therefore, a narrative synthesis was conducted.

Results

A step-by-step process of study screening and selection and the reasons for exclusion are given in the PRISMA flow diagram (Figure 1).

The electronic database searching retrieved 10 026 citations: An additional 16 citations were identified from other sources, such as hand-searching reference lists of included studies, relevant systematic reviews, and book chapters. After removing duplicates, title, and abstract screening, full texts of 183 studies were assessed for inclusion in detail, of which additional 136 studies were excluded, with reasons recorded. Forty-seven studies met the inclusion criteria and were included in the review [9,12,15–18,21,31–70].

Characteristics of included studies

A summary of the characteristics of the included studies is presented in Table 1. Of the 47 studies included, 20 studies used a cross-sectional design [18,31,33,36,37,39-41,44,47,48,51, 53-55,59, 62,64,66,68], 13 were prospective cohort studies [12,16,17,32, 34,35,38,45,50,57,58,69,70], six were retrospective registry reviewbased cohort studies [42,43,49,52,56,61], and three were case-control studies [15,21,65]. The remaining five publications were a secondary analysis of previously conducted studies [9,46,60,63,67].

The included studies were conducted in various countries. Sixteen studies were conducted in North America [9,38-44,47, 49,51,52,61,62,68,69], 12 in the UK and Europe [15,21,34–37, 45,55,60,63,65,67], six in Australia [12,17,18,31,32,66], eight in Asia [33,48,50,53,54,56,58,64], one in Brazil [16], and the remaining four had participants from different parts of the world [46,57,59,70].

Participants

A total of 275,631 participants were included in 47 studies; the sample size ranged from 21 to 254 519, the participants' age ranged from 18 to 93, and had both male and female participants. It is important to note that three papers [42-44] included the same cohort of participants in their studies. Therefore, these papers were treated as one study to avoid spurious multiplication of the number of participants. The findings of the papers, however, are reported separately as each of them evaluated different factors.

The study patient population included a variety of specific diagnostic groups. Twenty-two studies included patients with neurological conditions, of which 12 were on brain injuries [12,15,16,31-34,46,50,60,63,65], six were on SCI [17,36,45,47,51,68],

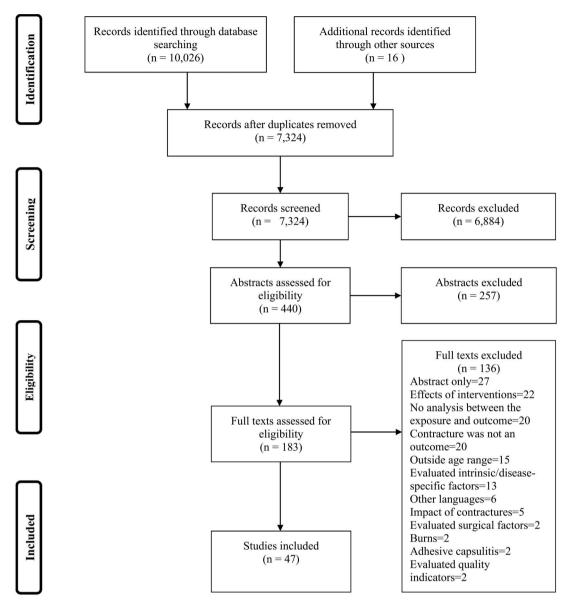


Figure 1. PRISMA flow diagram.

three were on CP [37,53,62], and one study was on MS [18]. Seventeen studies included participants with different musculo-skeletal conditions out of which seven studies included patients with SSc [21,35,38,57,59,69,70], five studies evaluated patients who underwent orthopaedic surgery [49,52,54,56,61], four studies examined patients with OA [39,40,55,66], and one study included transtibial amputees [48]. The remaining eight studies targeted patients with mixed diagnoses [9,41–44,58,64,67].

Joints assessed

studies joints Twenty-one assessed single studies [15,31-34,36,39,40,45,48-50,52,54,56,60-63,65,66], 21 multiple joints [9,12,16–18,21,35,37,38,42–44,46, 47,51,55,57,58,68-70], and the remaining five studies did not specify the number of joints assessed [41,53,59,64,67]. The most commonly assessed single joint was knee (eight studies) [39,40,48,49,52,54,56,65], followed by the shoulder (four studies) [15,33,36,45], elbow (three studies) [31,32,61], wrist [60,63], ankle [34,50], and hip [62,66] (two studies each). Sixteen studies assessed joints of the upper extremity [15,16,21,31-33, 36,38,45,46,51,57,60,61,63,70], 14 studies assessed joints of the lower extremity [34,37,39,40,48–50,52,54–56,62,65,66], 12 studies assessed both joints of upper and lower extremities [9,12,17,18,35,42–44,47,58,68,69], and the remaining five did not specify the joints assessed [41,53,59,64,67].

Contracture definition

Out of the total 47 studies, 37 studies provided an operational definition of contracture [12,15–18,31–38,40–52,55–62,65,66]. Of these, 12 studies categorised them according to their severity [12,15,17,18,33,35,36,40,44,51,57,59]; eight studies used a different term for contractures: limitation in joint ROM [33,66], limited PROM [37,47], impaired ROM [45], and arthrofibrosis [49,52,61]. Ten studies did not specify any operational definition for contractures [9,21,53,54,63,64,67–70].

Contracture identification and documentation

The most common method to identify contractures was goniometry which measured the PROM (14 studies) [16,33,37,39,

Table 1. Characterist	Table 1. Characteristics of included studies.							
Reference	Sample	Design	Joints assessed	Measurement/ assessment of contracture	Operational definition for contracture	Included factors	Excluded factors	Overall quality
Ada and OʻDwyer [31]	N = 24 Age range = 36–87 years Diagnosis, stroke Setting: rehabilitation units	Cross-sectional	Elbow	Photographic measurement	Loss of elbow extension range	Associated reactions	1	37.5%
Ada et al. [32]	N = 27 Mean age = 63 years Diagnosis: stroke Setting: hospital	Prospective cohort	Elbow	Photographic measurement	Quantified as the intact minus the affected angle in degrees	Loss of muscle strength; spasticity; upper limb activity	ı	55.6%
Aras et al. [33]	N = 85 Mean age = 58.7 years Diagnosis: stroke Setting: hospital Country: Turkey	Cross-sectional	Shoulder	PROM by goniometer	Limitation in the PROM mild, moderate, severe: <50%, 50–67%, of the normal for flexion and <22.2%, <22.2–33.3%, or >33.3% of the normal for extrotation extrotation	Pain	1	62.5%
Avouac et al. [21]	N = 162 (120 SSc. 42 controls) Age range = 20-90 years Diagnosis: SSc Setting: hospital Country: France	Case-control	Hand joints (wrist; MCP; PIP; DIP)	Radiographs	ı	Functional disability	Type of SSc; pulmonary fibrosis; fibrosis severity; positive ATA	40%
Baagøe et al. [34]	N = 33 (19: 14 controls) Mean age = 48 years Diagnosis: ABI Setting: hospital Country: Denmark	Prospective cohort	Ankle	Custom-built handheld device which measured ROM with a gyroscope	Reduced ROM	Spasticity	ı	72.7%
Balint et al. [35]	N = 131 Mean age = 56 years Diagnosis: SSc Country: Hungary	Prospective cohort	PIP, MCP, wrist, elbow, shoulder, hip, knee, ankle	Assessment of ROM (tool unspecified)	Contracture: limitation of ROM > 25% of the normal ROM Severe contracture: limitation of >50% of the normal ROM	Gender; dominant vs. non-dominant side; skin hypo/ hyperpigmentation; functional status; upper limb disability	SSc specific skin manifestations; SSc specific laboratory measures; disease duration; pulmonary/cardiac involvement; effect of drug therapies; steroid use	77.8%
Bossuyt et al. [36]	N = 1549 Mean age = 52.3 years Diagnosis. SCI Setting: community Country: Switzerland	Cross-sectional	Shoulder	3-point ordinal scale	Limited ROM of a joint (mild-infrequent, moderate-occasional, and significant-chronic problem)	Musculoskeletal shoulder pain	Lesion level (paraplegia vs. tetraplegia)	62.5%
Brantmark et al. [37]	N = 102 Median age = 20 years Diagnosis: CP Setting: community Country Śweden	Cross-sectional	Hip, knee, ankle	PROM by goniometer	Limited PROM: hip abduction <44°, hip int. rot <40°, hip ext. rot <40°, hip extension <0°, popliteal angle <140°, knee extension <0°, foot df with flexed knee <20°, foot df with extended knee <10°.	Mobility	ı	75%
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Reference	Sample	Design	Joints assessed	Measurement/ assessment of contracture	Operational definition for contracture	Included factors	Excluded factors	Overall quality
Fheodoroff et al. [46]	N=456 Mean age = 57 Diagnosis: stroke Countries: 22 countries in Europe, Pacific Asia, and South America	Secondary analysis	Shoulder, elbow, wrist, hand	Composite contracture score – severity of contractures rated on a scale of 1–12.	>3 quarters limitation of movement in at least one segment	Age; pain	Primary goals in active and passive function; onset of stroke	54.5%
Frye et al. [47]	N = 29 Mean age = 42.3 Diagnosis: SCI Setting: hospital Country: USA	Cross-sectional	Shoulder, elbow, forearm, wrist, hip, knee, and ankle	PROM by goniometer	the value, ions in the value, ions in the value, ions in the day as a little cked from on or 0°). This the value is to reach the isted as	Independence, ADLs	1	62.5%
Ghazali et al. [48]	N = 50 Mean age = 55.4 Diagnosis: transtibial amputees Setting: community Country: Malaysia	Cross-sectional	Knee	Unspecified	a negative on the stumble to fully extend the stump. Degree of contracture was measured as the remaining angle for the stump to achieve a full extension. Significant contracture	Ag e	Diabetes; length of stump	62.5%
Haller et al. [49]	N = 186 Mean age = 46.4 Diagnosis: post-operative tibial fracture Setting: trauma centre Country: USA	Retrospective cohort	Knee	PROM by goniometer	defined as or an gical or restore ROM on or ROM for	Age; male gender	Diabetes; tobacco use; infection; high energy fracture; surgical factors; CPM	71.4%
Hamzah et al. [50]	N = 70 Mean age = 38 Diagnosis: ABI Setting: NICU Country: Malaysia	Prospective cohort	Ankle	PROM by goniometer	°0 ×	Age; gender; ethnicity; spasticity, dystonia; clonus; duration of IMV; LOS in hospital	Cause of brain injury; severity of brain injury; sepsis	77.8%
Hardwick et al. [51]	N=38 Median age = 52 Diagnosis: SCI Setting: medical centre Country: USA	Cross-sectional	Shoulder, elbow, forearm, wrist, fingers, thumb	PROM by goniometer	at knee extension 1. No contracture: no loss in range of motion 2. Mild contracture: loss of up to 1/3rd range	Age; muscle strength; innervation status; functional independence	Injury level; time post-injury	75%
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Reference	Sample	Design	Joints assessed	Measurement/ assessment of contracture	Operational definition for contracture	Included factors	Excluded factors	Overall quality
Lee et al. [59]	N=2157 Mean age = 54.8 Diagnosis: SSc Setting: community Country: Australia, Canada, France, Mexico, Spain,	Cross-sectional	Unspecified	Dichotomised as: none- mild and moderate-severe	Presence of joint contractures: none/ mild (<25%) and moderate/severe (>25%) limitation in ROM	Pain	1	87.5%
Malhotra et al. [60]	UK, and USA $N=30$ Median age $=70.5$ Diagnosis: stroke Setting: hospital	Secondary analysis	Wrist	PROM and stiffness measured using a custom-built device	An increase in stiffness and a reduction in ROM	Arm function	1	69.2%
Marchand et al. [61]	Country: UK N=390 Mean age = 44.3 Diagnosis: post-operative elbow fracture	Retrospective cohort	Elbow	ROM documentation (goniometer)	Arthrofibrosis: elbow flexion contracture >45'or flexion- extension arc of	Duration of immobilisation; age; gender	Tobacco use; diabetes; surgical factors; types of fractures	81.8%
Matozinho et al. [16]	Country: $0.5A$ $N = 76$ Median age = 66 Diagnosis: stroke Setting: hospital	Prospective cohort	Shoulder, elbow, wrist	PROM by goniometer and gravity inclinometer	motion < 100° Minimum loss of 10° between measures obtained within the first 4 weeks and at	Upper limb function; muscle strength; pain, manual dexterity; spasticity	1	88.9%
Noonan et al. [62]	Country: Brazil N = 77 Mean age = 40 Diagnosis: CP Setting: residential long-term care	Cross-sectional	Ню	Hip ROM (tool unspecified)	3-month follow-up Hip FC >30°	Pain; pressure ulcers	1	%05
Pandyan et al. [63]	N = 22 Nean age = 64 Diagnosis: stroke Setting: hospital	Secondary analysis	Wrist	PROM measured using a custom- built device	1	Functional recovery	1	69.2%
Pohl and Mehrholz [15]	N=110 (50:60) Mean age (patients)=58.2 Mean age (controls)=50 Diagnosis: ABI Setting: rehabilitation centre Country: Germany	Case-control	Shoulder	Manual measurement with a ruler in cm	1 - severe contracture: hand in neck is not possible 2 - moderate or clinically relevant contracture: the hand in the neck position is possible, but the mean distance between the olecranon and underlay is > age- and sex-related referent values of the control group 3 - no or clinically nonrelevant contracture: the hand in the neck position is possible, and the mean distance between the olecranon and underlay is > to the	Spasticity	duration; diagnosis	%04
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Reference	Sample	Design	Joints assessed	Measurement/ assessment of contracture	Operational definition for contracture	Included factors	Excluded factors	Overall quality
Pohl et al. [65]	N = 55 (45:10) Mean age = 41 Diagnosis: ABI Setting: unspecified	Case-control	Knee	Photographic measurement	referent values of the control group Knee joint contractures were defined as clinically relevant if normal PROM was	Involuntary muscle activity	ı	%06
Pua et al. [66]	N=100 Mean age = 62 Diagnosis: OA Setting: community Country: Australia	Cross-sectional		PROM by digital inclinometer	Decreased hip PROM	Physical function; muscle strength; pain	T	87.5%
Takai et al. [64]	N = 171 Mean age:85.4 Diagnosis: mixed Setting: nursing homes Country: Japan	Cross-sectional	Unspecified	Unspecified	1	Pain	ı	42.9%
Vanderwee et al. [67]	N = 235 Median age = 87 Diagnosis: mixed Setting: nursing homes Country: Belgium	Secondary analysis	Unspecified	Dichotomised as: yes/no	1	Pressure ulcers	ı	53.8%
Vogel et al. [68]	N = 216 Age range = 24–37 Diagnosis: SCI Setting: community Countries: USA and Ganada	Cross-sectional	Elbow, ankle, hip	Interviews	1	Ethnicity, gender; functional independence; spasticity, muscle weakness	Age at injury; age at follow up; duration of injury; level of injury	90%
Wagner et al. [9]	N = 273 Mean age = 83.7 Diagnosis: mixed Setting: nursing homes Country: USA	Secondary analysis	Shoulder, elbow, wrist/hand, hip, knee, ankle/foot	Unspecified	1	Age; gender; ethnicity; functional status; mobility; fall risk; pain; cognition; behavioural symptoms; urinary incontinence; physical restraint; healthcare insurance; nursing home LOS	Stroke; arthritis; fracture; use of pain medication; psychoactive drug use	71.4%
Wiese et al. [69]	N = 200 Mean age = 50 Diagnosis: SSc Setting: scleroderma centres	Prospective cohort	Shoulders, elbows, wrists, fingers, knees	Unspecified	1	Functional disability; QOL	Physician and patient global health; tendon friction rubs; CPK	%9'25
Wojeck et al. [70]	N=1903 Mean age = 54.8 Diagnosis: SSc Setting: community Countries: Australia, Canada, France, Mexico, Spain, UK, and USA	Prospective cohort	DIP, MCP, PIP	Dichotomised as: moderate or severe	No	Pain	1	44.4%

PROM: passive range of motion; SSc. systemic sclerosis; MCP: metacarpophalangeal; PIP: proximal interphalangeal; DIP: distal interphalangeal; ATA: anti-topoisomerase; ABI: acquired brain injury; ROM: range of motion; SCI: spinal cord injury; CP; cerebral palsy; USA: United States of America; OA: osteoarthritis; KFC: knee flexion contracture; BMI: body mass index; FC: flexion contracture; WOMAC: Western Ontario and McMaster Universities Arthritis Index; ICU: intensive care unit; IMV: invasive mechanical ventilation; LOS: length of stay; DM: diabetes mellitus; NM: neuromuscular; APACHE: Acute Physiology and Chronic Health Evaluation; QOI: quality of life; ADL: activities of daily living; MUA: manipulation under anaesthesia; CPM: continuous passive motion; NICU: neurosurgical intensive care unit; TKA: total knee arthroplasty; MS: multiple sclerosis; AKS American knee society; UK: United Kingdom; CPK: creatine phosphokinase.

40,45,47,49-51,54-56,61]. Of these, one also utilised a gravity inclinometer along with the goniometer [16]. The second most common mode of identification of contractures was utilisation of either an ordinal or nominal scale to establish the presence and/ or severity of contractures [12,17,18,36,41,44,46,57–59,67,70] (12 studies). Four studies utilised photographic or radiographic measurement method to identify contractures [21,31,32,65]; three studies used a custom-built device to measure the PROM [34,60,63]; two studies utilised physical examination to assess the PROM [35,62]; two studies used a manual measurement method [15,38]; one study used a digital inclinometer to quantify PROM [66]; one study in addition to using an ordinal scale also assessed PROM of specific joints at standardised torque [17]. Seven studies documented contractures through the patient medical records [42,43,52,61], filled survey responses of participants [44,48], and telephonic interviews of patients [68]. Four studies did not specify the methods used to identify or document contractures [9,53,64,69].

Methodological quality

Table 2 provides the risk of bias and quality assessment of the included studies with scores according to the research design. The overall score was calculated for each checklist and expressed in percentage. Ten studies were rated with an average score between 80 and 100% (excellent) [16,39-41,55,56,59,61,65,66], 22 studies were rated between 60 and 79% (good) [9,12,17,18,33-37,43,45-54,60,63], 13 were rated between 40 and 59% (fair) [15,21,32,42,44,57,58,62,64,67-70], and two studies were rated below an average score of 40% (poor) [31,38]. The most common area for high risk of bias for studies with low average scores was the lack or unclear use of valid and reliable tools for outcomes and exposures and the lack of identification of the confounding factors.

Identified factors

A detailed list of factors evaluated in the included studies with their statistical findings is provided in the Supplementary information (B). The identified factors were broadly grouped into three main domains: sociodemographic factors, physical factors, and proxies for bed confinement. A summary of the review findings according to the evidence is presented in Table 3.

Domain 1: sociodemographic factors

Age

Out of a total of 47, 18 studies included ageing as a potential factor [9,12,17,38-40,42,44-46,48-51,53,56,58,61] of which two reported a significant association of age with contractures [40,46]. In two studies, the association depended on specific planes of movement [17,45], e.g., significant association with shoulder flexion and abduction but not external rotation [45]. One study demonstrated no significant association between age and the severity of contracture but a moderate positive correlation with any contracture [51]. One study demonstrated that older age was an independent risk factor for the development of a new contracture for participants who already had a contracture [58]. The remaining 12 studies failed to establish any significant association of contractures with age [9,12,38,39,42,44,48-50,53,56,61].

Gender

Fourteen studies included gender as a possible associated factor [9,35,38-40,42,45,49,50,54,56,58,61,68]. Of these, 12 studies found no association of either gender with contractures. One study demonstrated that males are more likely to develop new joint contracture through univariate analysis, but multivariate analysis showed that the male gender was not an independent risk factor for contractures [58]. One study found no correlation of contractures with gender after TKA; however, it demonstrated that the recurrence rate of flexion contracture (FC) after TKA was significantly higher in males than in females [54].

Ethnicity

Five studies evaluated ethnicity as a potential factor [9,38,40,50,68]. One study identified a statistically significant association between the occurrence of contractures among African American participants compared to white American participants [9]. The remaining four studies found no association with the ethnic backgrounds of participants [38,40,50,68].

Weight, height, and body mass index

Three studies investigated weight, height, and body mass index (BMI) as potential factors [39,40,56]. One study reported that participants with FC were heavier, taller, and had a greater BMI than those without FC [40]. The remaining two studies reported no association with contractures [39,56].

Education and employment status

One study evaluated the association of education status (illiterate, literate, primary school, high school, or university graduate) and employment status (unemployed, working part-time, or working full-time) with joint contractures and reported moderate correlations for both [53].

Accommodation

One study evaluated accommodation (whether the participants lived alone, with their family, spouse, or caregiver) as an associated factor for joint contractures and reported no significant correlation [53].

Laterality

One study evaluated laterality (dominant vs. non-dominant side) as a potential factor and reported a significant positive association with small hand contractures [35].

Healthcare insurance

One study evaluated healthcare insurance as a potential factor in nursing homes and reported that residents with healthcare insurance (Medicaid) were significantly more likely to have contractures [9].

Domain 2: physical factors

Functional ability

Functional ability in this review is operationally defined as the ability to perform basic and instrumental ADLs. Nineteen studies evaluated the association of functional ability with contractures [9,12,16,21,32,35,38,44,46,47,51,55-57,60,63,66,68,69].studies reported significant association of poor or reduced overall ability with contractures [9,16,21,32,35,38, 46,47,51,55,57,60,63,66]. Among others, Kwah et al. [12] reported a significant association of contractures with poor combined upper limb motor function and "sit-to-stand" activity but did not

Table 2. Quality of evidence of included studies.

	Q2. E	Q3. Q4. Baseline Participant similarity blinding	Q5. Int Therapist g blinding	Q6. th Accessor blinding	Q7. Identical treatment of both groups Foll	Q8. Inte	Q9. Q10. Intention Outcomes in to treat both groups	Q11 Valid and es in reliable oups outcomes	nd Q12. e Statistical es analysis	Q13. :al Trial is design	Total score	% Score
	> Z >	Z Z Z	ZZZ			> Z >	>>> > Z Z	z > z	z >> >	>->-	9/13 7/13 9/13	69.2% 53.8% 69.2%
Q1. Similarity of groups	Q2. Similarity of exposures	Q3. Valid and reliable exposures	Q4. Confounding Factors	Q5. Strategies to deal with confounding factors	Q6. Free of outcome at the start of study	Q7. Valid and reliable outcomes	Q8. Sufficient follow up t	Q9. Reasons to loss i	Q10. Strategies to address incomplete	Q11. Statistical analysis		
	NA	> 1	> 1	z	> 1	>- 1	> 1	z	z	z	6/5	25.6%
	≻ ≨	>- >	>- >	≻ z	⊃ Z	>- >	> - >	>- >	z >	z >	8/11	72.7%
	K K	- >-	- Z	zz	z ⊃	- ⊃	- >-	- z	- Z	- >-	3/6	33.3%
	N S	· > -	:>	z))) ⊃	· > -	:≻	z	· >-	5/6	55.6%
	> 2	>- >	>- Z	zz	⊃ z	> >	>- >	>- >	z>	>- >	6/2	77.8%
	4 4 2 2	- >-	≥ ≻	zz	zz	- >-	- ≻	- >-	- >-	- >-	6/2	90.7% 77.8%
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	Y Z	>- >	>>	ZZ	⊃ >	>- >	> - >	≰>	¥=	>- >	5/7	71.4%
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	≻ 8	>- >	>- >	z z	>- >	> z	>- >	>- >	> >	>- >	10/11	90.9% 77.8%
	A N	- >-	- >-	z	. ⊃	z	· > -	· ⊃	- z	· > -	4/9	44.4%
	>-	Π	z	z	>	z	>-	NA	N A	>	6/5	25.6%
	≻ ≨	> >	>->	- >	> >	>- >	> >	> >	z 2	>- >	9/11	81.8%
	Z Z	- >-	- >-	- z	- z	- ⊃	- >-	- >-	zz	- >-	5/9	55.6%
	NA	>	>	n	n	O	>	O	z	>	4/9	44.4%
	Q2. Matching	Q3. Same criteria for cases and controls	ر Q4. Valid and reliable exposures	Q5. Exposure measured same on way for cases ures and controls		Q6. Q7 ding factorsconf	Q6. Q7. Strategies for Q8. Valid and Confounding factors reliable outcomes	Q8. Valid and eliable outcomes	Q9. Length of Exposure period		Q10. Statistical analysis Total score % Score	Total score
1	z	z	> >			n	n :	z) 		> :	4/10
	z >-	z >	>- > -	>- >-		z >-	z >-	>- > -	> >		> >	4/10 9/10
1 2	Q2.	Q3. Similarity of treatment/care other		Q5. Multiple		200	Q7. Outcomes measured in same	e Q8. Valid and		99.	,	3000
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Checklist for analytical cross-sectional studies	ross-sectional studies									
	Q1. Clearly defined inclusion criteria	Q2. Description of subjects and setting	Q3. Valid and reliable exposure	Q4. Standard criteria for measurement of condition	Q5. Confounding factors	Q6. Strategies for confounding factors	Q7. Valid and reliable outcomes	Q8. Statistical analysis	Total score	% Score
Ada and O'Dwyer [31]	>	>	n	>	z	Z	z	n	3/8	37.5%
Aras et al. [33]	Z	>	Z	>	>-	Z	>-	>-	2/8	62.5%
Bossuyt et al. [36]	>-	>-	Z	>	>-	Z	Z	>-	2/8	62.5%
Brantmark et al. [37]	>-	>	>-	>-	Z	Z	>-	>-	8/9	75%
Campbell et al. [39]	>-	>	>-	>-	>-	>-	>-	Z	2/8	87.5%
Campbell et al. [40]	>-	Z	>-	>-	>-	>-	>-	>-	2/8	87.5%
Castle and Engberg [41]	>-	>	Z	NA	>-	>-	>-	>-	2/9	85.7%
Clavet et al. [44]	>-	>	>-	NA	Z	Z	Z	>-	4/7	57.1%
Frye et al. [47]	>-	>-	>-	n	Z	>-	>-	z	2/8	62.5%
Ghazali et al. [48]	>-	>	Π	>-	>-	Z	Z	>-	2/8	62.5%
Hardwick et al. [51]	>-	>	>-	>-	Z	Z	>-	>-	8/9	75%
Hoang et al. [18]	>-	>	>-	>-	Z	Z	>-	>-	8/9	75%
içağası ğ lu et al. [53]	>-	>	>-	>-	Z	Z	D	>-	2/8	62.5%
Kinoshita et al. [54]	>-	>	>-	>-	Z	Z	>-	>-	8/9	75%
Kocic et al. [55]	>-	>	>-	n	>-	>-	>-	>-	2/8	87.5%
Lee et al. [59]	>-	>	>-	>-	>-	>-	Z	>-	2/8	87.5%
Noonan et al. [62]	>-	>	>-	>-	Z	Z	n)	4/8	%09
Pua et al. [66]	>-	>	>-	>-	>-	>-	Z	>-	2/8	87.5%
Takai et al. [64]	Z	>	>-	NA	Z	Z	Z	>-	3/7	42.9%
Vogel et al. [68]	>-	>	Z	>-	Z	Z	Z	>-	4/8	%09
Average % score										%8'99
:										

Y: yes; N: no; U: unclear; NA: not applicable.



Table 3. Summary of review findings.

Evidence for association with contractures	Factors
Consistent evidence	Bed confinement
	Functional ability
	Muscle weakness
	Pain
	Physical mobility
Inconsistent evidence	Age
	Spasticity
Weak evidence	Gender
	Ethnicity
	Height
	Weight
	BMI
	Laterality
	Education and employment status
	Healthcare insurance
	Level of cognition
	Clonus
	Spastic dystonia
	Ethnicity
	Urinary incontinence
	Pressure ulcers
	Manual dexterity
No evidence	Accommodation status
	Involuntary muscle activity
	Anxiety
	Behavioural symptoms
	Ouality of life

report any significant relationship between reduced walking function and contracture. Two studies reported differences between the types of joints affected by lack of function; Vogel et al. [68] reported a significant association of poor functional ability with hip and elbow contractures but not with ankle contractures. The remaining three studies did not report any significant association of functional ability with contractures [44,56,69].

Pain

Of the 18 studies that examined the relationship of pain with contractures [9,12,16,17,33,36,39,44–46,52,56,58,59,62,64,66,70], reported significant association between pain and contractures [9,16,33,36,39,45,46,56,59,62,64,66,70]; one reported that the association depended on specific plane of movement, i.e., significant association with ankle dorsiflexion but not with wrist and elbow extension [12]. The remaining four studies did not report any significant association of pain with contractures [17,44,52,58].

Muscle weakness

Nine studies evaluated the association of muscle weakness with contractures [12,16-18,32,45,51,66,68]. Eight out of nine studies reported significant association of contractures with muscle weakness [12,16-18,32,51,66,68]. Of these, one reported a significant association of contractures with muscle weakness but not with innervation status [51]. The remaining study did not report any significant association between the two [45].

Muscle tone

Spasticity: Nine studies evaluated the association of spasticity with the development of contractures [12,15,17,32,45,50,68]. Six studies reported significant associations with contractures [12,15,17,32,45,50]; however, two of these only identified jointspecific positive associations: a significant association between spasticity with elbow and wrist contractures was identified in one study [17] but was non-significant in another study [12]. Interestingly, one study reported an inverse relationship between spasticity and contractures [34]. The remaining two studies

reported no significant association of spasticity with contractures [16,68]. Spastic dystonia and clonus: One study evaluated spastic dystonia and clonus as potential factors and reported a statistically significant association with contractures [50].

Physical mobility

Physical mobility in this review is operationally defined as an individual's ability to move independently and safely in different environments to perform ADLs. Eight studies evaluated the association of reduced physical mobility with contractures [9,18,37,40,43,44,53,58], and all of them reported a significant association.

Skin chanaes

Three studies evaluated the association of changes in the skin with the occurrence of contractures [35,62,67]. Balint et al. [35] included skin hypo/hyperpigmentation as a possible associated factor; however, no statistically significant association was found. Noonan et al. [62] and Vanderwee et al. [67] evaluated the association of pressure ulcers with the development of contractures, and both studies demonstrated a significant association.

Involuntary muscle activity

Two studies investigated the association between contractures and involuntary muscle activity/associated reactions and showed no significant association [31,65].

Psycho-coanitive functions

Cognition: Wagner et al. [9] evaluated the association of level of cognition with contractures; univariate analysis demonstrated a significant association of cognitive decline with contractures. Learning disability: İcağası ğılu et al. [53] evaluated the association of mental retardation with contractures in adults with CP and found a moderate correlation between the two. Anxiety: Clavet et al. [44] assessed the association of anxiety with contractures and demonstrated no significant association. Behavioural symptoms: Wagner et al. [9] included behavioural symptoms as a possible associated factor for contractures, but the findings did not reach significance.

Urinary incontinence

Wagner et al. [9] included urinary incontinence as a potential associated factor. Univariate analysis revealed a significant association with contractures, but multivariate analysis showed no significant association.

Manual dexterity

Matozinho et al. [16] evaluated manual dexterity as a potential risk factor. It was reported to be an independent predictor for the development of joint contractures.

Domain 3: proxies for bed confinement

In this review, proxies for bed confinement are operationally defined as any extrinsic factors limiting an individual's mobility or confining them to bed.

Seven studies evaluated different proxies for bed confinement potential associated factors for joint contractures [9,41-43,50,58,61]. Marchand et al. [61] evaluated the association of length of immobilisation with contractures and demonstrated a statistically significant association. Clavet et al. [43] investigated the difference in contracture occurrence between patients mobilised in the ICU and those who were not; they reported a

significant difference between them. Lam et al. [58] evaluated dependency for bed mobility as a possible risk factor and demonstrated it to be an independent predictor of new upper limb contractures. Three studies investigated physical restraints as a potential factor associated with joint contractures in long-term care residents [9,40,57]. All of them reported that physically restrained residents have a significantly higher chance of developing contractures. Wagner et al. [9] identified nursing home length of stay (LOS) as a potential factor. There was a statistically significant difference between the mean LOS of residents who developed contractures and those who did not. Two studies evaluated the association of the LOS in ICU with contractures identifying a significant association between the two [42,43]. Two studies investigated the association of the LOS in hospital with the development of contractures [42,50]; one of which [42] reported a weak association, and the other reported no significant association [50]. Two studies evaluated the association of the duration of invasive mechanical ventilation with the development of contractures within the ICU setting [42,50]. One study demonstrated that the odds of developing contractures were 7.7 times higher in mechanically ventilated patients for more than two weeks than those who were mechanically ventilated for two or less weeks [50]. The other study showed a weak association between contractures and the duration of mechanical ventilation [42].

Discussion

The identified factors in this review were broadly categorised into three main domains: sociodemographic factors, physical factors, and proxies for bed confinement. The factors which provided the most consistent evidence for association with contractures were poor functional ability, pain, muscle weakness, reduced physical mobility, and bed confinement.

Methodological quality

The overall methodological quality of the studies was rated good, with an average of 66.8%. The most frequent area of the potential risk of bias in the low-quality studies was the unclear use of goldstandard tools to assess contractures and the failure to identify confounding factors. The assessment of contractures was variable across the studies because no gold standard assessment tool exists to identify their presence. However, most studies defined and documented contractures as a limitation in the joint PROM. Therefore, the variability of the assessment methods does not directly influence the identification of factors associated with contractures. On the other hand, confounding factors play an essential role in a multifactorial condition such as joint contracture. Therefore, it is difficult to make inferences about the direct causal implications of most of the factors associated with contractures when other potential influences are not accounted for. Therefore, the findings of the review should be interpreted with caution.

Factors with consistent evidence of association

Findings across studies on reduced functional ability, pain, muscle weakness or paralysis, physical mobility, and bed confinement were generally consistent and found strong correlations with joint contractures. In theory, the relationship of contractures with these factors could be explained by the notion that the presence of any of the above-mentioned factors reduces the overall functionality and places the joint(s) in a static position for extended periods

due to a lack of active movement. This leads to a reduced number of sarcomeres, decreasing the overall muscle mass and length. The connective tissues, in turn, lose their elasticity and undergo fibrosis, potentially predisposing the muscle to shorten and form a contracture [7,22,24,71]. In case of pain, the patients tend to adopt a particular position that relieves the associated discomfort. For instance, patients with anterior knee pain tend to adopt a position of partial flexion for prolonged periods to avoid pain and discomfort, potentially leading to the reduced functional use of the knee joint and the potential development of knee FCs [56].

Factors with inconsistent evidence of association

Age

Evidence regarding age was inconsistent across the studies. The associations identified in five studies [17,40,45,46,51] could be attributed to other musculoskeletal disorders and typical degenerative changes associated with increasing age leading to reduced joint ROM rather than contractures occurring as a part of the normal ageing process [24].

Spasticity

Spasticity was identified as a potential factor for contractures in six out of nine studies that evaluated the relationship between the two variables. This is supported by the underlying theory that abnormal muscle activity associated with spasticity could lead to abnormal posturing resulting in muscle and soft-tissue shortening and, consequently, forming a contracture [72]. However, the evidence provided by these studies is questionable because the methods utilised to measure or evaluate the presence of spasticity were not consistent. There is sufficient evidence that while conventional clinical scales are easy to administer, they also lack clinical sensitivity and have limited validity and reliability to document the abnormal muscle activity associated with spasticity [73,74]. On the contrary, neurophysiological measurements provide a direct measure of muscle activity to quantify spasticity according to the existing definition [74]. In this review, the only study that utilised a neurophysiological measure to identify the presence of spasticity and found a significant association with contractures lacked statistical power (n < 30) [32]. In addition, the study failed to consider the evaluation of confounders alongside spasticity and weakness, such as upper limb function, which could be the primary factor for the development of contractures [60].

Additionally, because current neurophysiological measures are not feasible as assessment tools for clinicians in everyday practice, the relationship between spasticity and contractures remains inconclusive. Further evidence is required, and this is likely to depend on developing a practical assessment method of spastialso differentiate between contractures city that can and spasticity.

Factors with weak or no evidence of association

Sociodemographic factors like gender, ethnicity, height, weight, BMI, laterality, education and employment status, healthcare insurance, and accommodation status showed either no or insufficient evidence of association with contractures. Among physical factors, involuntary muscle activity, anxiety, QOL, and behavioural symptoms also failed to provide any evidence of association with contractures. Other physical factors like clonus, dystonia, manual dexterity, pressure ulcers, urinary incontinence, and level of cognition demonstrated correlations with contractures, but there was insufficient evidence to make any inferences.



Strengths and limitations of the review

This is the first systematic review to identify the factors associated with joint contractures targeting adults aged 18 and above.

The development and progression of joint contractures usually involve a complex interplay of factors. Once developed, contractures are followed by a chain of physical impairments such as loss of function, limited mobility, pain, and deconditioning. This potentially leads to further immobility, predisposing to exacerbating existing or developing new contractures resulting in a vicious cycle [75]. For this reason, this review was not just limited to longitudinal studies which aim to establish the risk factors and temporal relationships. Rather, all types of studies that addressed ultimate, proximate, or associated factors linked with progressive joint contractures were included, regardless of their temporal occurrence. The associations found in this review, therefore, could arise from factors either contributing to or occurring as a consequence of joint contractures.

A comprehensive search strategy was employed as part of this review to capture most of the evidence. However, it may be subjected to retrieval bias as the search was limited to the year 1999 and the English language only. This might have led to the exclusion of substantial evidence published before this date and in other languages.

There was a lack of consistency in the definitions and outcome measures used for contractures and the associated factors; the lack of a uniform definition and assessment methods made it difficult to compare the findings across the studies and understand the direction of the relationship.

Conclusions

The factors which provided consistent evidence on association with joint contractures in this review were poor functional ability, pain, muscle weakness, reduced physical mobility, and bed confinement. These factors do not necessarily qualify as independent predictors for the development of joint contractures. However, considering the multifactorial aetiology of joint contractures, the evidence for different associations can be used to design targeted and effective prevention and management strategies to reduce the incidence of joint contractures.

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