ISCoS The International Spinal Cord Society

www.nature.com/sc

REVIEW ARTICLE

Check for updates

Treatment of shoulder pain in people with spinal cord injury who use manual wheelchairs: a systematic review and meta-analysis

Matthew Wellisch[1] ✉, Kim Lovett[1], Meg Harrold[1], Carsten Juhl[2], Birgit Juul-Kristensen[2], Leanda McKenna[1] and Camilla M. Larsen[2,3]

STUDY DESIGN: Systematic review and meta-analysis

OBJECTIVES: The objective was to summarise prior research regarding the efficacy of active physiotherapy interventions and prevention strategies on shoulder pain, decreased physical function and quality of life in people with a spinal cord injury (SCI).

METHODS: A systematic literature search was conducted in CENTRAL, EMBASE (via Ovid), CINAHL and MEDLINE (via Ovid). Randomised controlled trials investigating effects of active physiotherapy interventions on shoulder pain, physical function and quality of life were included. Further, prospective cohort studies investigating effects of active physiotherapy interventions in prevention of shoulder pain and reduced physical function were included. Mean difference (MD) for pain (15 items on a 0–10 scale) and standardised mean difference (SMD) for physical function were summarised in a random effects meta-analysis.

RESULTS: Four studies on treatment (totalling 167 participants), and no studies on prevention were included. Significant and clinically meaningful improvements on shoulder pain (MD 19.06, 95% CI 5.72–32.40; $I^2 = 65\%$) (scale 0–150) and physical function (SMD 0.61, 95% CI 0.27–0.94; $I^2 = 0\%$) were found for active physiotherapy interventions. Only one study included quality of life, making meta-analysis inappropriate.

CONCLUSIONS: Evidence from a sparse number of studies supports active physiotherapy interventions to decrease shoulder pain and increase physical function in people with SCI who use a manual wheelchair. No studies met the criteria for prevention, highlighting a lack of research investigating prevention of shoulder pain and decreased physical function and quality of life.

Spinal Cord (2022) 60:107–114; https://doi.org/10.1038/s41393-021-00673-x

INTRODUCTION

The incidence of traumatic spinal cord injuries globally in 2007 was estimated to be 23 cases per million people, with an estimated prevalence between 236 and 1298 per million [1, 2]. In Australia alone, spinal cord injury (SCI) has an estimated cost of 2 billion dollars annually and people with less physical function require increased assistance and consequently incur greater associated costs [3, 4]. The shoulder is one of the common sites of chronic musculoskeletal pain in people with SCI [5]. The prevalence of shoulder pain in people with SCI has been reported to range from 36 to 76% [6–8], and a recent large study reported a 3-month prevalence of 63% [9]. Shoulder pain in people with SCI is a pervasive condition with wide reaching implications reducing independent function and quality of life (QoL) [10, 11].

People with SCI using manual wheelchairs have a greater reliance on their upper limbs to maintain their level of independence through daily activities, such as wheelchair propulsion and transfers. Although shoulder pain has a multifactorial aetiology in this group, shoulder pain may be a result of the increased biomechanical load imposed on their upper limbs [12], especially during wheelchair transfers [13, 14] and propulsion [15]. Those experiencing greater mechanical loads are more likely to have shoulder pain [16]. However, the evidence regarding associations between physical activity and the risk of shoulder pain is equivocal. Previous studies have shown conflicting results reporting that wheelchair athletes have lower prevalence of shoulder pain than non-athlete wheelchair users [17], that there is no group difference [18], and that overhead sport is a risk factor for shoulder pathology in people with SCI [8].

Physiotherapy interventions such as therapeutic exercise and movement optimisation focus on increasing capacity and function and reducing biomechanical load. Therapeutically administered exercise including resistance exercises can decrease shoulder pain, improve function and QoL in people with SCI using manual wheelchairs [19, 20]. In addition, optimisation of wheelchair propulsion and transfers have resulted in reduced biomechanical loading of the shoulder [21, 22]. Previous systematic reviews investigating active physiotherapy interventions on people with SCI have found encouraging results regarding the effect of resistance training on shoulder function [23], and resistance combined with stretching exercises on shoulder pain [24]. However, no systematic review has examined the effects of the

[1]School of Physiotherapy and Exercise Science, Faculty of Health Sciences, Curtin University, Perth, WA, Australia. [2]Department of Sports Science and Clinical Biomechanics, University of Southern Denmark, Odense, Denmark. [3]Health Sciences Research Centre, UCL University College, Odense, Denmark. ✉email: matthew.wellisch@postgrad.curtin.edu.au

Received: 31 December 2020 Revised: 12 July 2021 Accepted: 14 July 2021
Published online: 9 August 2021

entire range of active physiotherapy interventions on shoulder pain, decreased physical function and QoL. In addition, since the publication of previous reviews, new findings have been presented, and prior to this study no systematic review has succeeded with meta-analysis. Finally, there is no consensus of the most effective prevention strategy for shoulder pain in this group.

Therefore, the aim was to provide an updated systematic review of the available evidence regarding the efficacy of active physiotherapy interventions in the (1) treatment and (2) prevention of shoulder pain, decreased physical function and QoL in people with SCI using manual wheelchairs.

METHODS

Protocol and registration

This systematic review was conducted in accordance with the Cochrane Handbook for Systematic Reviews of Interventions Version 6.1 [25] and reports according to the Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) guidelines [26]. The proposal was registered in PROSPERO (CRD42019136693), an international database of systematic review protocols on health-related topics via their website; https://www.crd.york.ac.uk/prospero.

Eligibility criteria

Types of studies. For objective one, we included randomised controlled trials (RCTs) investigating the effect of active physiotherapy interventions on shoulder pain, decreased physical function or QoL. For objective two, we included prospective cohort studies that investigated the effect of active physiotherapy interventions on primary prevention of shoulder pain or decrease in physical function in people with SCI using manual wheelchairs. Included studies had to be published in English and available in full text.

Types of participants. For objective one and two, we included studies with participants who had complete or incomplete SCI at any level, of an age between 18 and 70 years, who had at least 1 year since their SCI and whose primary means of mobility was a manual wheelchair. For objective one, we included studies whose participants had musculoskeletal pain localised to the shoulder for at least 3 months. For objective two, we included studies with participants who had no shoulder pain at the commencement of the study.

Types of interventions. For objective one, we included studies that had administered an active physiotherapy intervention aimed at reducing shoulder pain, improving physical function or QoL in people with SCI using manual wheelchairs. For objective two, we included studies that had administered an active physiotherapy intervention aimed at primary prevention of shoulder pain or decreased physical function. Active interventions include any active strategy that aims to increase the capacity or function of a shoulder or that reduce biomechanical loading. Examples include exercise prescription, muscle re-education and optimisation of wheelchair transfer and propulsion.

Types of comparison. For both objectives, we included studies that compared active physiotherapy interventions to control treatment (no treatment or passive interventions). Passive interventions include surgery, corticosteroid injections and medications such as nonsteroidal anti-inflammatory drugs (NSAIDs).

Types of outcome measures. The primary outcome for both objectives was pain; secondary outcomes include physical function and QoL. We accepted a variety of methods for measuring these outcomes, including both specific to the population of interest and those generalisable to the wider population.

Information sources and search

Initially, the PROSPERO database and the Cochrane database were searched for registered protocols on this topic. Databases were searched for studies published from inception until November 2020 in CENTRAL, EMBASE (via Ovid), CINAHL and MEDLINE (via Ovid) [27]. We used a search strategy with the domains of 'shoulder pain' and 'SCI'. The combined search strategy for both objective one and two is outlined in the PROSPERO registry platform (https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42019136693) and was modified as required for each database. We searched reference lists of eligible studies, systematic reviews from the past 10 years, grey literature and conference proceedings for the past 2 years of the Australian & New Zealand Spinal Cord Society and the European Spinal Cord Federation. We downloaded the results of each search to Endnote (X9.1, Clarivate Analytics) and deleted any duplicates found in the combined search results. The remaining studies were exported to Covidence (Covidence systematic review software, Veritas Health Innovation) where remaining duplicates were identified and removed prior to screening.

Study selection

Firstly, two authors independently screened the title and abstract of the identified studies for eligibility. Following consensus on initial screening, the full text of the remaining studies were independently screened for inclusion. Disagreement at either stage of screening was resolved through discussion, or—if required—with the assistance of the group of authors.

Data extraction and management

Data were independently extracted by two authors from the included studies using a standardised form in Covidence. Disagreement was resolved through discussion or—if required—with the assistance of the group of authors. Study authors were contacted to request additional data if study data were missing or unclear. Data extraction included study design, participants (sample size, participant characteristics), interventions (type, prescription, adherence), outcome measures and results (baseline and follow-ups). Data extraction forms were stored securely in Covidence and are available upon request from the authors.

Summary measures

For objective one, we extracted the number of participants allocated to intervention and control treatment, mean and standard deviation (SD) for relevant outcome measures from each group immediately after cessation of treatment. If between-group-differences were evident at baseline, change scores from baseline to post-intervention were used [28]. Where change scores were used and a change SD was not provided by the authors, baseline SDs were used for meta-analysis. For comparison to the use of baseline SDs, sensitivity analysis was performed using an imputed change-from-baseline SD applying a correlation coefficient of 0.6,

$$SD_{change} = SD^2_{baseline} + SD^2_{final} - (2 \times Corr \times SD_{baseline} + SD_{final})$$

[29]. Continuous outcome measures using the same scale were entered into the meta-analysis using mean differences (MD) with 95% confidence intervals (95% CIs) [28]. Where studies reported the outcome using different scales, standardised mean differences (SMD) with 95% CIs were used to measure the effect size [28]. Studies using outcome measures of differing directions were standardised to a common direction [29]. SMD's were measured as Cohens d and were adjusted to Hedges' g [30]. The clinical relevance of SMD values were interpreted as: 0.2 represents a small effect, 0.5 a moderate effect and 0.8 a large effect [31]. Where outcome measures had no established minimal clinical important difference (MCID) for comparison to the pooled MD

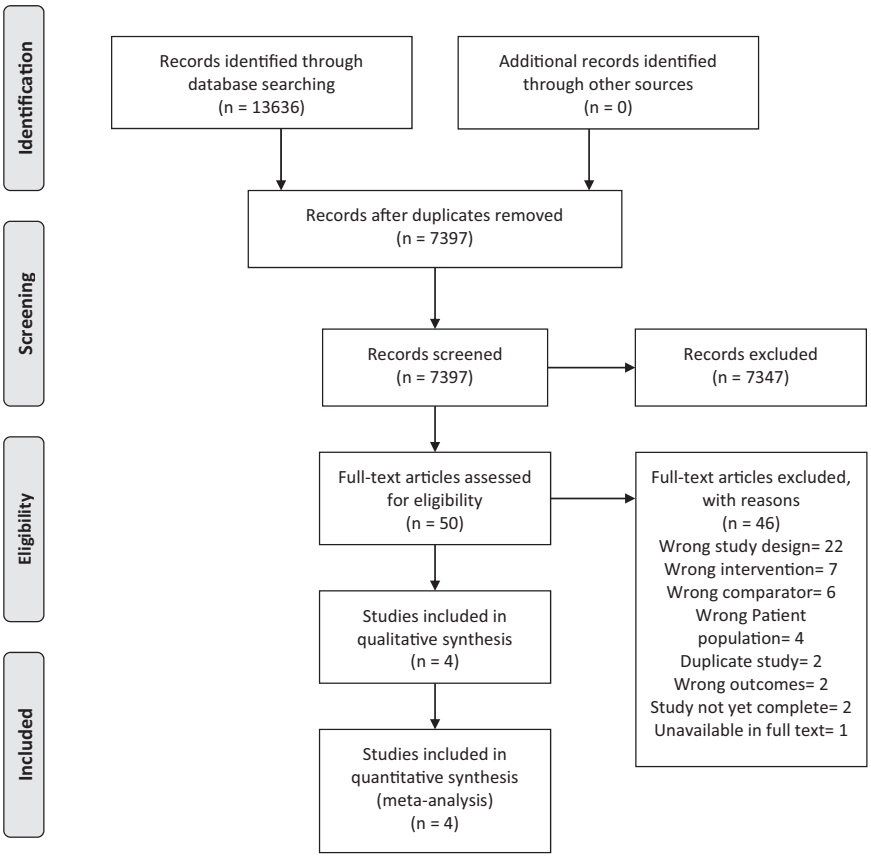

Fig. 1 PRISMA flow diagram of the study search and selection process, including reasons for exclusion.

between groups, clinical relevance of results were interpreted recalculating the outcome to SMD and using the above-mentioned interpretation of SMD.

Assessment of heterogeneity

Heterogeneity was evaluated by calculating the I^2 statistics to quantify the percentage of variability attributable to heterogeneity. I^2 values between 0 and 40% may not be important; 30–60% may represent moderate heterogeneity; 50–90% may represent substantial heterogeneity; 75–100% may represent considerable heterogeneity [28].

Data synthesis

As heterogeneity was expected due to differences in participants, intervention and outcomes, a random-effect model was used as default. We used RevMan (Review Manager version 5.2, The Cochrane Collaboration) to perform the statistical analysis.

Risk of bias across studies

Two authors independently assessed the risk of bias of each RCT using the Cochrane Risk of bias 2.0 tool [32]. Risk of bias was assessed in the domains of bias arising from the randomisation process, deviations from intended intervention, missing outcome data, measurement of the outcome and selection of the reported result, answering a number of signaling questions as described in the Revised Cochrane risk-of-bias tool for randomised trials (RoB 2) guide [32]. Overall risk of bias judgement was categorised as either 'low risk of bias' if all categories were judged to be low risk of bias; 'some concerns' if at least one domain was judged as some concern but no domain is judged high risk of bias; or 'high risk of bias' if one domain is judged as high risk of bias or multiple domains are judged as some concern [32].

RESULTS

Study selection

After removing studies through screening and eligibility assessment, four studies met the inclusion criteria for objective one—treatment and were included in the quantitative analysis (see Fig. 1). No studies met the inclusion criteria for objective two—prevention. Reasons for exclusion at each stage of the study selection process are summarised in Fig. 1, with full details available in Appendix 1. Attempts to contact authors for clarification on data regarding three studies [19, 33, 34] resulted in all three studies being excluded: in two studies addressing objective one, one study used duplicate data from another already included study [19], and one study's author was contacted without success [33]; one study addressing objective two was embargoed prior to publication [34].

Study characteristics

In total, 167 participants were recruited (Table 1). The Wheelchair User's Shoulder Pain Index (WUSPI) (15 items on a 0–10 scale) was used in all four studies to measure pain. Three studies assessed physical function using different outcome measures, including the Constant–Murley Shoulder Outcome Score [35], SF-36 physical function component [36] and Disabilities of the Arm, Shoulder, and Hand (DASH) questionnaire [37]. One study assessed QoL using the Social Interaction Index and the Subjective QoL Scale [36].

According to the FITT (frequency, intensity, type and time) principles, the resistance programs were prescribed at a frequency of one to two times daily or three times weekly (Table 1). The intensity was dosed at 8 or 15 repetition maximum for three sets. Types of exercise included resistance and stretching exercises, consisting of some or all of the following exercise types: shoulder abduction, shoulder external rotation and scapular retraction and

Table 1. Descriptive characteristics and quality assessment of studies included in the meta-analysis of differences between active physiotherapy interventions and controls.

Study	Participants	Assessment	Intervention	Control	Results (Final mean (SD))	Risk of bias assessment
Cardenas et al. [36]	32 wheelchair users with shoulder pain (NRS ≥ 4, duration > 3 months), SCI of duration ≥ 1 year, manual or powered wheelchair use ≥ 50% of time and ability to independently transfer, age > 20 years	Administered pre and post intervention at 12 week follow up: WUSPI (pain) DASH (physical function)	12 weeks, 3x per week shoulder exercise program: stretches of anterior and posterior shoulder joint + upper trapezius, warm up including 4 non-resistive active movements and 4 resistance exercises using resistive bands and hand weights including shoulder abduction and external rotation (3x8 repetition maximum) + scapular plane elevation and scapular retraction (3x15 repetition maximum) Initial exercise instruction and 4 week review of technique and resistance level	One hour educational video and handout on shoulder anatomy, mechanisms of injury and pain management, generalised with no recommendations for specific exercises	WUSPI: Intervention = 53.60 (30.49) Control = 39.23 (28.62) DASH: Intervention = 37.85 (17.46) Control = 31.99 (14.94) P values for mean differences unavailable.	Randomisation process = 🙂 Deviations from intended interventions = 🙂 Missing outcome data = 🙂 Measurement of the outcome = 😐 Selection of the reported result = 🙂 *Overall Risk of bias judgement* = 😐
Curtis et al. [37]	25 manual wheelchair users with shoulder pain, SCI C6 or below, SCI of duration ≥ 1 year, used manual wheelchair for > 3 hours per week	Administered pre and post intervention at 2 month intervals till 6 months: WUSPI (pain)	Initial 60 minute instructional session, including education on functional shoulder anatomy and exercise purpose, exercise demonstration and individual instruction of 5 seated home exercises. 6 months home exercise program: 2 static stretches (5 repetitions 20-30s hold, 2x per day) for anterior shoulder musculature (pec and biceps) and 3 resistance exercises (3x 15 repetitions, 1x per day) for posterior shoulder musculature (scapular retraction, shoulder external rotation and abduction) using stretchable exercise bands, biweekly phone call review + in person review at 2 and 4 months	Continue daily activities as usual	WUSPI: Intervention = 17.16 (20.35) Control = 19.74 (20.85) P value for mean differences unavailable.	Randomization process = 🙂 Deviations from intended interventions = 🙂 Missing outcome data = 🙂 Measurement of the outcome = 😐 Selection of the reported result = 🙂 *Overall Risk of bias judgement* = 😐
Dondal et al. [34]	30 manual wheelchair users with shoulder pain, complete or incomplete SCI below T1 and manual wheelchair used as primary source of mobility for > 1 year	Administered pre and post intervention at 4 week follow up: WUSPI (pain) CMS (physical function)	4 weeks, 45 minutes 3x per week shoulder strengthening and stretching exercises	No intervention	WUSPI: Intervention = 57.7 (13.68) Control = 69.9 (19.82) CMS: Intervention = 76.60 (11.53) Control = 67.53 (9.32) P values for mean differences unavailable.	Randomization process = 😐 Deviations from intended interventions = 🙂 Missing outcome data = 🙂 Measurement of the outcome = ⚫ Selection of the reported result = 🙂 *Overall Risk of bias judgement* = ⚫
Mulroy et al. [35]	80 manual wheelchair users with unilateral or bilateral shoulder pain, SCI below T2 of duration ≥ 5 years and manual wheelchair used for mobility ≥ 50%	Administered pre and post intervention at 12 week follow up: WUSPI (pain) SF-36-FC (physical function) SII and SQLS (QOL)	12 weeks, 3x per week shoulder exercise program: stretches of anterior and posterior shoulder joint + upper traps, warm up including 4 non-resistive active movements and 4 resistance exercises using resistive bands and hand weights including shoulder abduction and external rotation (3x8 repetition maximum) + scapular plane elevation and scapular retraction (3x15 repetition maximum) Initial educational handout, exercise instruction and on movement optimisation strategies regarding transfers, depression raises, and wheelchair propulsion, 4 week review of technique and resistance level	One hour educational video and handout on shoulder anatomy, mechanisms of injury and pain management, generalised with no specific recommendations to change behaviour	WUSPI: Intervention = 14.90 (14.00) Control = 45.60 (35.20) P value (mean difference) = P < .001 SF-36-FC Intervention = 39.90 (6.50) Control = 35.70 (7.30) P value (mean difference) = P < .05 SII Intervention = 53.30 (30.60) Control = 40.80 (16.60) P value (mean difference) = P = .14 SQLS Intervention = 5.3 (0.90) Control = 5.0 (1.40) P value (mean difference) = P < .05	Randomization process = 🙂 Deviations from intended interventions = 🙂 Missing outcome data = 🙂 Measurement of the outcome = 😐 Selection of the reported result = 🙂 *Overall Risk of bias judgement* = 😐

🙂 = low risk of bias, 😐 = some concerns, ⚫ = high risk of bias, Abbreviations: WUSPI, Wheelchair User's Shoulder Pain Index; DASH, Disabilities of the Arm, Shoulder, and Hand questionnaire; CMS, Constant-Murley Shoulder Outcome Score; SII, Social Interaction Inventory; SQLS, Subjective Quality of Life Scale; SF-36-FC, SF-36 Function component

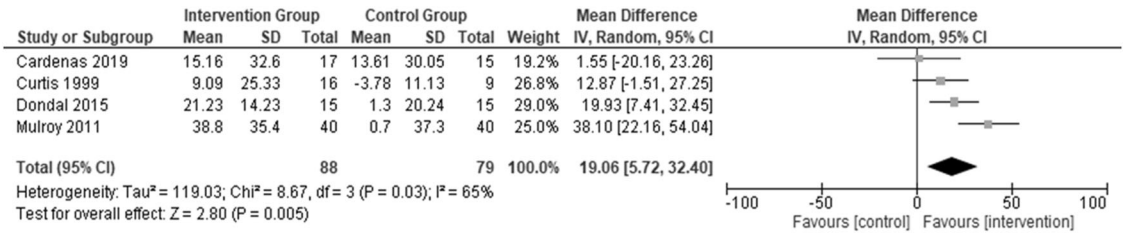

Fig. 2 Forest plot for shoulder pain outcome. Mean difference in Wheelchair User's Shoulder Pain Index (WUSPI) at cessation of intervention between active physiotherapy interventions and controls (*X*-axis shows 100 of 0–150 scale).

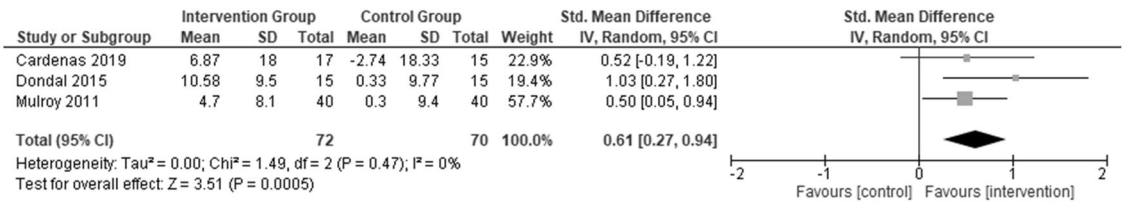

Fig. 3 Forest plot for physical function outcome. Standardised mean difference in physical function at cessation of intervention between active physiotherapy interventions and controls.

Table 2. Sensitivity analysis of shoulder pain and physical function outcome measures in the meta-analysis of differences between active physiotherapy interventions and controls.

Outcome	Sensitivity analysis	MD	95% CI	I^2
Shoulder pain	Using baseline standard deviation	19.06	[5.72–34.40]	65%
	Imputed SD correlation coefficient 0.6	19.00	[5.20–32.81]	73%
	Curtis 2-month scores	15.11	[−2.32–32.53]	80%
	Excluding Cardenas et al.	23.15	[9.45–36.85]	64%
	Excluding Mulroy et al.	14.29	[5.27–23.32]	6%
Outcome	**Sensitivity analysis**	**SMD**	**95% CI**	I^2
Physical function	Using baseline standard deviation	0.61	[0.27–0.94]	0%
	Imputed SD correlation coefficient 0.6	0.70	[0.36–1.04]	0%
	Excluding Cardenas et al.	0.67	[0.18–1.16]	29%
	Excluding Mulroy et al.	0.75	[0.23–1.27]	0%

scapular plane elevation. One study [36] also included education in movement optimisation. Time or duration of the exercise interventions were prescribed within a range from 4 weeks to 6 months. Comparison groups included either no intervention [35], usual care [38] or a non-specific educational video on shoulder pain and management designed as a 'sham' intervention [36, 37].

One study included participants using a manual or powered wheelchair, but the participants were required to transfer independently [37]. Further data on the number of participants that used a powered wheelchair in this study, and data specific to those that only used a manual wheelchair, were sought from the study authors, but was not provided.

Risk of bias

Three studies [36–38] were judged as raising 'some concerns' and the fourth study [35] was judged to include a 'high risk of bias' (Table 1). The raising of 'some concerns' from the three studies [36–38] resulted from the domain 'measurement of the outcome' being judged as having 'some concerns', because participant self-reported outcome measures were used and there was an inability to blind participants to group allocation. The judgement of 'high risk of bias' in one study [35] showed a lack of clarity regarding randomisation, allocation and blinding of assessors with only three of five domains considered 'low risk of bias'.

Primary outcome

Meta-analysis of active physiotherapy interventions in the treatment of shoulder pain in people with SCI who use manual wheelchairs showed a superior outcome compared with control interventions on the WUSPI scale (MD = 19.06 (95% CI 5.72–32.40, $I^2 = 65\%$)) (Fig. 2). No MCID value is currently available for the WUSPI scale to allow comparison of the pooled MD. Thus, a SMD was also generated in Revman for the pooled difference in WUSPI between intervention and control groups (SMD = 0.72 (95% CI 0.23–1.21, $I^2 = 0\%$)), indicating a moderate effect size that is likely to be clinically relevant [31].

For physical function, active physiotherapy intervention provided significantly greater improvement compared with controls (SMD = 0.61 (95% CI 0.27–0.94, $I^2 = 0\%$)) (Fig. 3), interpreted as moderate effect size that is likely to be clinically relevant [31].

Meta-analysis was unable to be performed for QoL as only one study [36] measured QoL. Mulroy et al. [36] found a greater improvement in Subjective QoL Scale mean score following active physiotherapy intervention involving exercise and movement optimisation compared with controls (intervention 5.3 (0.9) vs. control 5.0 (1.4), $P < .05$), but no significant difference in post-test scores between intervention and controls when measured by the Social Interaction Index (intervention 53.3 (30.6) vs. control 40.8 (16.6), ($P = .14$)).

Additional analysis

Only two studies [36, 37] recorded adverse events for a total of six events related to the active physiotherapy interventions. All recorded adverse events related to the intervention were non-serious and involved either onset of neck or elbow pain or increased shoulder pain.

Sensitivity analysis

Sensitivity analysis was performed to determine the effect of imputed SD, different timepoints for outcome measures, inclusion criteria differences and a significant difference in drop-out rate within one study (Table 2). Sensitivity analysis was performed using an imputed SD as a comparator to using baseline SD in change score in the meta-analysis. For both pain and physical function, using the imputed SD had minimal impact on the result. Curtis et al. [38] collected WUSPI data at 2-months interval during the intervention period, up until 6 months, therefore, sensitivity analysis was performed using 2-months data from this study in comparison with the 6-months post-intervention measure. Using 2-months data reduced the MD for shoulder pain.

As a result of the difference in inclusion criteria, that is, the inclusion of powered wheelchair users able to independently transfer, sensitivity analysis was performed on the exclusion of Cardenas et al. [37]. Exclusion of Cardenas et al. [37] had little influence on the results for both shoulder pain and physical function. Sensitivity analysis for the exclusion of Mulroy et al. [36] due to higher drop-out rate resulted in reduced heterogeneity for shoulder pain from substantial to may-not-be-important and reduce the MD slightly.

DISCUSSION

This systematic review and meta-analysis supports the use of active physiotherapy interventions in the treatment of shoulder pain and decreased physical function in people with SCI using manual wheelchairs. The meta-analysis found significant and clinically meaningful improvements in both shoulder pain and function resulting from active physiotherapy interventions compared with controls. An insufficient number of studies including QoL outcomes resulted in the inability to pool data for meta-analysis. However, the only study [36] including QoL found an improvement as a result of active physiotherapy intervention. No studies on prevention met the inclusion criteria for the current study.

Objective one—Treatment

An important finding of the current review was the significant and clinically relevant reduction in shoulder pain as measured by WUSPI from active physiotherapy interventions compared with controls. Active physiotherapy interventions included movement

optimisation strategies and stretching and resistance exercises using resistance bands or weights, prescribed either daily or three times weekly for between 4 weeks and 6 months. However, the result is limited to short- to medium-term effects of active physiotherapy interventions with none of the included studies performing long-term follow up assessment. Our review shows an MD of 19.0 points between groups in the WUSPI and supports results from a previous systematic review by Cratsenberg et al. [24], that narratively synthesised seven studies (both RCT and observational), finding a decrease in WUSPI scores between 8.3 and 37.0 points.

Furthermore, our review shows an SMD of 0.61 for a positive effect on physical function between intervention and control groups, which was also a clinically meaningful improvement due to active physiotherapy intervention. This is consistent with a previous systematic review of Kloosterman et al. [39], that in a narrative synthesis of eight studies concluded that therapeutic exercise has a positive effect on upper limb motor control and function. That prior review differs from our review, as it only included participants with tetraplegia and manual wheelchair use, and shoulder pain was not a specified inclusion criterion.

Our review aimed to conduct a meta-analysis of QoL. However, only one study [36] included QoL measures and therefore data were not pooled. As reported, Mulroy et al. [36] found significant improvements on a self-reported QoL outcome ($P < .05$) after 12 weeks active physiotherapy intervention compared with controls. This is consistent with the findings of Hicks et al. [40] in a previous RCT investigating the effects of exercise in people with SCI; however the inclusion criteria did not contain manual wheelchair use or having shoulder pain. Hicks et al. [40] concluded that 9 months twice weekly of supervised progressive resistance exercises (90–120 min, two sets 50% RM, increasing to 70–80% RM at week 4) and endurance exercises (arm ergo at 70% maximum HR, Borg 3–4, two bouts 5–10 min increasing to 15–20 min) resulted in significantly increased self-reported QoL.

Only four treatment studies met the inclusion criteria for our study, demonstrating a large research gap in knowledge on active physiotherapy interventions in this population. Similarly, Mason et al. [41] in their recent scoping review summarising the knowledge on conservative treatment of shoulder pain in manual wheelchair users (not only those with SCI), found only 21 studies meeting their criteria that included any study with conservative treatment either longitudinal or within subject measure design; with 12 of those studies including active physiotherapy interventions. They concluded that the evidence investigating conservative treatment of shoulder pain in this population is low and recommended further research on a multidisciplinary approach with active physiotherapy interventions underpinning treatment. Their study did not provide meta-analysis on outcome measures.

Objective two—Prevention

Surprisingly, no prospective cohort studies met our review's inclusion criteria addressing the prevention of shoulder pain and reduced function and QoL in people with SCI using manual wheelchairs. Of note is, that there were four studies investigating the prevention of shoulder pain or the maintenance of function, but they were excluded from this review, due to either study design (a prospective non-randomised controlled trial or a non-controlled trial) [42, 43], inclusion of participants with pain at commencement of the trial [44] or the unavailability of the full text article [34].

Nevertheless, results from these excluded prevention studies have shown encouraging results in both preventing shoulder pain and decreased physical function. In one of the studies, people with paraplegia had significantly reduced shoulder pain onset compared with controls at both 18 months and 3 years, following a home exercise program combined with education in movement optimisation [34]. In another study, people with incomplete tetraplegia participating in a once-weekly 2-year progressive wheelchair rugby training program starting with a strength and endurance training phase, through a skill acquisition phase, and finally to a 140–150 min wheelchair rugby training phase, showed improved shoulder function compared with controls at 2-year follow-up [42]. In the third and fourth studies, wheelchair basketball athletes improved shoulder range of motion after resistance and stretching exercises prescribed three times per week for either 6 or 10 weeks [43, 44]. Despite all these studies not being generalisable to the wider population with SCI, as they focused on either people with paraplegia [34], or tetraplegia [42] or included participants without SCI [43, 44], the results may inspire further investigation into the efficacy of active physiotherapy interventions in preventing shoulder pain and maintaining shoulder function in people with SCI using manual wheelchairs.

Strengths, limitations and generalisability

There were several limitations to this study. Firstly, only a small number of studies were included, giving a small total number of participants. Clear descriptions regarding exercise intensity, and when and how to progress exercises, were not given in all the included studies. Another limitation of our study was the substantial heterogeneity ($I^2 = 65\%$) detected in the meta-analysis of change in pain scores for objective one—treatment. In addition, only 16 (10%) of the included participants were classified as having tetraplegia, and therefore our review's results are less generalisable to populations with cervical injuries. Finally, none of the included studies considered the potential differences in the level of SCI. Level of SCI has been demonstrated to significantly impact shoulder muscle recruitment during manual wheelchair propulsion [45], and also impact the prevalence of rotator cuff disorders [46].

Strengths of our review include the comprehensive and rigorous methodology used, and included studies investigating interventions over relatively large time periods, with selection criteria that preferentially targeted active physiotherapy interventions with outcomes meaningful to people with SCI. In addition, only RCTs were considered for objective one and prospective cohort studies for objective two [47].

Direction of future research

This systematic review highlighted a large knowledge gap, with only four RCTs meeting the selection criteria for treatment of shoulder pain, reduced physical function and QoL, and no cohort studies meeting the inclusion criteria for investigating prevention. Given the high prevalence of shoulder pain in this population, and that QoL is highly related to functional independence, treatment and prevention of pain and reduced function require further research [7, 10]. Furthermore, high-quality studies considering level of SCI, complete vs. incomplete SCI and other individual participant characteristics such as baseline pain and level of physical function, could guide individualised intervention prescription with improved outcomes. Furthermore, we recommend that future studies include a detailed description of the intervention using the FITT principles [48] according to a recommended guideline [49], which will better enable future replication of the therapeutic exercise interventions.

CONCLUSION

Despite a sparse number of studies, the results of this review provide evidence to support the clinical use of active physiotherapy intervention in the treatment of shoulder pain and reduced physical function and QoL in people with SCI using manual wheelchairs. Unfortunately, only one study included QoL but reported a significant improvement in QoL as a result of active physiotherapy intervention. There were no prospective cohort studies identified addressing prevention of shoulder pain and

reduced physical function. Given the potential implications of shoulder pain in this group, more high-quality studies are needed on the effects of active treatment and preventing intervention strategies on shoulder pain, reduced physical function and QoL.

DATA AVAILABILITY

We applied to the Information Department at Curtin University to receive access to R: drive, a shared drive which allowed us to store data securely, minimised the risk of data loss and was accessible by all members of the research team. Data will be available upon request to the authors.

REFERENCES

1. Lee B, Cripps RA, Fitzharris M, Wing P. The global map for traumatic spinal cord injury epidemiology: update 2011, global incidence rate. Spinal Cord. 2014;52:110–6.
2. Furlan JC, Sakakibara BM, Miller WC, Krassioukov AV. Global incidence and prevalence of traumatic spinal cord injury. Can J Neurol Sci. 2013;40:456–64.
3. Hamilton BB, Deutsch A, Russell C, Fiedler RC, Granger CV. Relation of disability costs to function: spinal cord injury. Arch Phys Med Rehabil. 1999;80:385–91.
4. The Victorian Neurotrauma Initiative. The economic cost of spinal cord injury and traumatic brain injury in Australia. Access Economics Pty Limited; Australia 2009.
5. Widerström-Noga EG, Felipe-Cuervo E, Yezierski RP. Relationships among clinical characteristics of chronic pain after spinal cord injury. Arch Phys Med Rehabil. 2001;82:1191–7.
6. Bossuyt FM, Arnet U, Brinkhof MWG, Eriks-Hoogland I, Lay V, Muller R, et al. Shoulder pain in the Swiss spinal cord injury community: prevalence and associated factors. Disabil Rehabil. 2018;40:798–805.
7. Richardson A, Samaranayaka A, Sullivan M, Derrett S. Secondary health conditions and disability among people with spinal cord injury: a prospective cohort study. J Spinal Cord Med. 2021;44:19–28.
8. Akbar M, Brunner M, Ewerbeck V, Wiedenhofer B, Grieser T, Bruckner T, et al. Do overhead sports increase risk for rotator cuff tears in wheelchair users? Arch Phys Med Rehabil. 2015;96:484–8.
9. Larsen CM, Juul-Kristensen B, Kasch H, Hartvigsen J, Frich LH, Boyle E, et al. The Danish Spinal Cord Injury Shoulder (DanSCIS) cohort: methodology and primary results. Spinal Cord. 2021;59:821–31.
10. Gutierrez DD, Thompson L, Kemp B, Mulroy SJ. The relationship of shoulder pain intensity to quality of life, physical activity, and community participation in persons with paraplegia. J Spinal Cord Med. 2007;30:251–5.
11. Samuelsson KA, Tropp H, Gerdle B. Shoulder pain and its consequences in paraplegic spinal cord-injured, wheelchair users. Spinal Cord. 2004;42:41–6.
12. Figoni SF. Overuse shoulder problems after spinal cord injury: a conceptual model of risk and protective factors. Clin Kinesiol. 2009;63:12–22.
13. Gagnon D, Nadeau S, Noreau L, Dehail P, Gravel D. Quantification of reaction forces during sitting pivot transfers performed by individuals with spinal cord injury. J Rehabil Med. 2008;40:468–76.
14. Gagnon D, Nadeau S, Noreau L, Dehail P, Piotte F. Comparison of peak shoulder and elbow mechanical loads during weight-relief lifts and sitting pivot transfers among manual wheelchair users with spinal cord injury. J Rehabil Res Dev. 2008;45:863–73.
15. Kulig K, Rao SS, Mulroy SJ, Newsam CJ, Gronley JK, Bontrager EL, et al. Shoulder joint kinetics during the push phase of wheelchair propulsion. Clin Orthop Relat Res. 1998;354:132–43.
16. Ferrero G, Mijno E, Actis MV, Zampa A, Ratto N, Arpaia A, et al. Risk factors for shoulder pain in patients with spinal cord injury: a multicenter study. Musculoskelet Surg. 2015;99:53–6.
17. Fullerton HD, Borckardt JJ, Alfano AP. Shoulder pain: a comparison of wheelchair athletes and nonathletic wheelchair users. Med Sci Sports Exerc. 2003;35:1958–61.
18. Finley MA, Rodgers MM. Prevalence and identification of shoulder pathology in athletic and nonathletic wheelchair users with shoulder pain: a pilot study. J Rehabil Res Dev. 2004;41:395–402.
19. Kemp BJ, Bateham AL, Mulroy SJ, Thompson L, Adkins RH, Kahan JS. Effects of reduction in shoulder pain on quality of life and community activities among people living long-term with SCI paraplegia: a randomized control trial. J Spinal Cord Med. 2011;34:278–84.
20. Nash MS, van de Ven I, van Elk N, Johnson BM. Effects of circuit resistance training on fitness attributes and upper-extremity pain in middle-aged men with paraplegia. Arch Phys Med Rehabil. 2007;88:70–5.
21. Requejo PS, Mulroy SJ, Ruparel P, Hatchett PE, Haubert LL, Eberly VJ, et al. Relationship between hand contact angle and shoulder loading during manual wheelchair propulsion by individuals with paraplegia. Top Spinal Cord Inj Rehabil. 2015;21:313–24.
22. Tsai CY, Boninger ML, Hastings J, Cooper RA, Rice L, Koontz AM. Immediate biomechanical implications of transfer component skills training on independent wheelchair transfers. Arch Phys Med Rehabil. 2016;97:1785–92.
23. Aravind N, Harvey LA, Glinsky JV. Physiotherapy interventions for increasing muscle strength in people with spinal cord injuries: a systematic review. Spinal Cord. 2019;57:449–60.
24. Cratsenberg KA, Deitrick CE, Harrington TK, Kopecky NR, Matthews BD, Ott LM, et al. Effectiveness of exercise programs for management of shoulder pain in manual wheelchair users with spinal cord injury. J Neurol Phys Ther. 2015;39:197–203.
25. Higgins JPT, Thomas J, Chandler J, Cumpston M, Li T, Page MJ, et al (editors). Cochrane Handbook for Systematic Reviews of Interventions, version 6.2 (updated February 2021). Cochrane, 2021. Available from https://www.training.cochrane.org/handbook.
26. Moher D, Liberati A, Tetzlaff J, Altman DG. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. BMJ. 2009;339:b2535.
27. Michaleff ZA, Costa LOP, Moseley AM, Maher CG, Elkins MR, Herbert RD, et al. CENTRAL, PEDro, PubMed, and EMBASE are the most comprehensive databases indexing randomized controlled trials of physical therapy interventions. Phys Ther. 2011;91:190–7.
28. Deeks JJ, Higgins JPT, Altman DG (editors). Chapter 10: Analysing data and undertaking meta-analyses. In: Higgins JPT, Thomas J, Chandler J, Cumpston M, Li T, Page MJ, Welch VA (editors). Cochrane Handbook for Systematic Reviews of Interventions version 6.2 (updated February 2021). Cochrane; 2021. Available from https://www.training.cochrane.org/handbook.
29. Higgins JPT, Li T, Deeks JJ (editors). Chapter 6: Choosing effect measures and computing estimates of effect. In: Higgins JPT, Thomas J, Chandler J, Cumpston M, Li T, Page MJ, Welch VA (editors). Cochrane Handbook for Systematic Reviews of Interventions version 6.2 (updated February 2021). Cochrane; 2021. Available from https://www.training.cochrane.org/handbook.
30. Deeks JJ, Higgins JPT (editors). 10.S1 Supplementary material: Statistical algorithms in review manager 5. Statistical Methods Group of The Cochrane Collaboration. In: Higgins JPT, Thomas J, Chandler J, Cumpston M, Li T, Page MJ, Welch VA (editors). Cochrane Handbook for Systematic Reviews of Interventions version 6.2 (updated February 2021). Cochrane; 2021. Available from https://www.training.cochrane.org/handbook.
31. Cohen J. Statistical Power Analysis for the Behavioral Sciences. Hillsdale (New Jersey): Lawrence Erlbaum; 1988.
32. Higgins JP, Sterne JA, Savovic J, Page MJ, Hróbjartsson A, Boutron I, et al. A revised tool for assessing risk of bias in randomized trials. Cochrane Database Syst Rev. 2016;10:29–31.
33. Gironda R. An interdisciplinary, cognitive-behavioral shoulder pain treatment program for individuals with paraplegia. SCI Psychosocial Proc. 2004;17:247–52.
34. Mulroy S, Haubert LL, Eberly V, Conners S, Weiss W. Effectiveness of Two Intervention Programs to Prevent Shoulder Pain after Spinal Cord Injury. Arch Phys Med Rehabil. 2017;98:174.
35. Dondal K, Kulkarni V, Patole R, Rairikar S, Shyam A, Sancheti P. Effect of shoulder exercises on functional performance in paraplegic wheelchair users having shoulder pain. Indian J Physiother Occup Ther- Int J. 2015;9:83–6.
36. Mulroy SJ, Thompson L, Kemp B, Hatchett PP, Newsam CJ, Lupold DG, et al. Strengthening and optimal movements for painful shoulders (STOMPS) in chronic spinal cord injury: a randomized controlled trial. Phys Ther. 2011;91:305–24.
37. Cardenas DD, Felix ER, Cowan R, Orell MF, Irwin R. Effects of home exercises on shoulder pain and pathology in chronic spinal cord injury: a randomized controlled trial. Am J Phys Med Rehabil. 2020;99:504–13.
38. Curtis KA, Tyner TM, Zachary L, Lentell G, Brink D, Didyk T, et al. Effect of a standard exercise protocol on shoulder pain in long-term wheelchair users. Spinal Cord. 1999;37:421–9.
39. Kloosterman MG, Snoek GJ, Jannink MJ. Systematic review of the effects of exercise therapy on the upper extremity of patients with spinal-cord injury. Spinal Cord. 2009;47:196–203.
40. Hicks A, Martin K, Ditor D, Latimer A, Craven C, Bugaresti J, et al. Long-term exercise training in persons with spinal cord injury: effects on strength, arm ergometry performance and psychological well-being. Spinal Cord. 2003;41:34–43.
41. Mason B, Warner M, Briley S, Goosey-Tolfrey V, Vegter R. Managing shoulder pain in manual wheelchair users: a scoping review of conservative treatment interventions. Clin Rehabil. 2020;34:741–53.
42. Furmaniuk L, Cywińska-Wasilewska G, Kaczmarek D. Influence of long-term wheelchair Rugby training on the functional abilities of persons with tetraplegia over a two-year period post-spinal cord injury. J Rehabil Med. 2010;42:688–90.
43. Wilroy J, Hibberd E. Evaluation of a shoulder injury prevention program in wheelchair basketball. J Sport Rehabil. 2018;27:554–9.
44. García-Gómez S, Pérez-Tejero J, Hoozemans M, Barakat R. Effect of a home-based exercise program on shoulder pain and range of motion in elite wheelchair basketball players: a non-randomized controlled trial. Sports. 2019;7:180.

45. Mulroy SJ, Farrokhi S, Newsam CJ, Perry J. Effects of spinal cord injury level on the activity of shoulder muscles during wheelchair propulsion: an electromyographic study. Arch Phys Med Rehabil. 2004;85:925–34.
46. Sinnott KA, Milburn P, McNaughton H. Factors associated with thoracic spinal cord injury, lesion level and rotator cuff disorders. Spinal Cord. 2000;38:748–53.
47. Oxford Centre for Evidence-Based Medicine Levels of Evidence Working Group. "The Oxford 2011 Levels of Evidence". Oxford Centre for Evidence-Based Medicine, Oxford, United Kingdom 2015. Available from http://www.cebm.net/index.aspx?o=5653.
48. American College of Sports Medicine. ACSM's Guidelines for Exercise Testing and Prescription. Philadelphia: Lippincott Williams & Wilkins, 2010.
49. Slade SC, Dionne CE, Underwood M, Buchbinder R. Consensus on exercise reporting template (CERT): explanation and elaboration statement. Br J Sports Med. 2016;50:1428–37.

ACKNOWLEDGEMENTS

Acknowledgement to Curtin University for providing funding for the purchase of Covidence, to Curtin University librarian Diana Blackwood for assistance with search terms and sourcing of full text articles, to Cindy Davis for assistance with screening and Gretta Adolph for assistance with search terms and administration.

AUTHOR CONTRIBUTIONS

All authors (CML, CJ, KL, BJK, LM, MH and MW) were responsible for designing the review protocol. MW and KL were responsible for conducting the initial search and screening for eligible studies by title and abstract; screening the full text paper for inclusion, with all authors providing arbitration on some of the final decisions; assessing the qualifying articles for quality; extracting and analysing the data, with all authors contributing to the analysis process and assisting to interpret the results. KL and MW wrote the report with all remaining authors contributing and providing feedback.

FUNDING

All facilities and resources were provided by Curtin University, including funding for the purchase of Covidence at a cost of $240 AUD. In addition, Camilla Marie Larsen received grant funding from the Danish Society of Polio and Accident Victims, the Danish Physiotherapy Research Foundation, the Danish Jascha Foundation, and the Danish Vanføre Foundation for the initial planning and writing of this work.

COMPETING INTERESTS

The authors declare no competing interests.

ADDITIONAL INFORMATION

Supplementary information The online version contains supplementary material available at https://doi.org/10.1038/s41393-021-00673-x.

Correspondence and requests for materials should be addressed to M.W.

Reprints and permission information is available at http://www.nature.com/reprints

Publisher's note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.