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Review Article

Integrated hip surveillance pathways for pain, function and quality of life in children with Cerebral Palsy: A systematic literature review

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ABSTRACT

Aim: To determine the effectiveness of integrated hip surveillance pathways on pain, function and quality of life (QOL) in children with Cerebral Palsy (CP).
Method: A systematic literature review, designed, conducted and reported using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses. Inclusion criteria: confirmed CP diagnosis, management under recognised international hip surveillance pathways, outcome measures of hip displacement plus at least one other relevant to pain, function or QOL.
Results: 100 articles were identified. 12 full text articles were screened, and four were included. Reduced range of movement was associated with hip pain in children with CP. Increasing age, Gross Motor Function Classification Score (GMFCS) and migration percentage (MP) were associated with increased hip pain. General health declined with increased age. Increased MP and GMFCS level were associated with interruption to activities of daily living.
Interpretation: Outcomes relating to function and QOL are under-researched in the current integrated hip surveillance pathway evidence-base. Wider outcomes related to function and QOL need to be included to capture the wider impact on children who are at risk of hip dislocation.
What this paper adds: Increased pain was associated with reduced joint range and increased migration percentage. Pain also increased with greater age and Gross Motor Function Classification Score. Early orthopaedic intervention for hip displacement may not successfully mitigate pain. Effectiveness of integrated pathways on function and quality of life is under-evidenced. Studies investigating integrated pathways and holistic outcomes are needed to inform practice.

1. Introduction

Cerebral Palsy (CP) has been defined as an umbrella term for a non-progressive motor impairment resulting from brain lesions or abnormalities occurring in early development [1]. CP affects the developing and immature brain, resulting in a permanent and non-progressive dysfunction of the central nervous system [2]. As a result, CP is established as a movement disorder affecting posture and movement, which impacts upon functional abilities [3].

Despite the presence of a static and non-progressive encephalopathy, the musculoskeletal involvement in children with CP is typically progressive [2]. Children with CP are at risk of secondary musculoskeletal complications leading to pain, reduced function and negative impact on quality of life (QOL) [3–5]. It is often cited that the secondary complications of CP, which lead to pain, reduced participation, and poorer QOL, have a larger impact on everyday life than the primary diagnosis itself [4–6]. Hip dysplasia, leading to hip displacement, is the second most common orthopaedic issue after foot and ankle deformities, for children with CP, resulting in impairment in function. Hip dysplasia occurs in as many as 35 % of children and young people with CP [6,7]. The risk of hip dislocation or displacement increases with increased level of functional deficit, as indicated by their assigned Gross Motor Function Classification Score (GMFCS) [6,8,9]. The GMFCS level is a widely accepted and valid tool for assigning severity of CP in relation to functional ability, with a score of one being least affected and five being most affected by the motor disorder [4,6]. The risk of hip displacement and eventual hip dislocation has been found to be directly correlated with increased GMFCS level [10]. There is an overwhelming consensus in the relevant literature that hip dislocations are painful for those experiencing them, with a subsequent impact on range of movement (ROM)

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and functional abilities [5–9,11–14].

Owing to the complex presentation, secondary complications and impact on pain, function and QOL [4], management of hip displacement or dislocation requires a multidisciplinary approach, including tone management, orthopaedic and physiotherapy intervention and postural management [15–17]. However, the evidence base suggests that dislocation of the spastic hip can be preventable, which can be achieved by implementation of a standardised hip surveillance pathway consisting of standardised follow-up of gross and fine motor function, clinical findings, radiographic imaging and orthopaedic intervention [6,9–11]. A 20 year follow up of a comparison cohort study in Sweden [9] indicated that use of an integrated hip surveillance pathway, resulted in clinically significant reductions in incidence of hip dislocation and displacement in children and young people with CP. However, other clinical outcomes including pain, function and QOL were not assessed. Hip dislocations decreased from 9 % to 0.5 % [9]. This pathway became known as the Quality Registry for Children with Cerebral Palsy (Uppfoljningsprogram for Cerebral Pares, CPUP) and is considered by international experts as the gold standard for follow-up and prevention of musculoskeletal complications in children with CP [12]. As a result, international versions of the integrated hip surveillance model have evolved. Hip surveillance programs for children with CP have been adopted globally as standard care, providing healthcare providers with a framework for obtaining hip radiographs based on patients' age and GMFCS level [18]. The overall aim of the pathway is to improve or maintain physical function and health-related quality of life (QOL) in children with CP by preventing or mitigating secondary conditions, most specifically hip dislocation [9]. Moreover, it is widely agreed that the reduction in incidence of hip dislocation is attributed to the early identification of hip migration and subsequent early surgical and non-surgical intervention [9–14,18]. Early detection and appropriate surgical intervention of hip displacement can prevent progression to hip dislocation and the requirement for more invasive salvage surgery [8]. CPUP and the version adopted in England and Wales (Cerebral Palsy Integrated Pathway, CPIP) are designed as integrated pathways, allowing multidisciplinary management of the musculoskeletal complications of CP. However, a potential criticism is a primary focus on a biomedical approach to clinical decision-making. Although the CPUP and CPIP pathways have evidently been successful in managing the structural component of hip displacement [9,12], they appear to place less emphasis on the person-centred care indicators. As a result of the paradigmatic shift of health from a medical point towards a broader sense, the World Health Organization (WHO) promoted the use of the International Classification of Functioning, Disability and Health (ICF) [19]. The ICF aims to look at human functioning in a multidimensional and interactive way instead of using a more restricted biomedical approach [19]. When assessing paediatric patients, use of the ICF framework encourages clinical reasoning and an improved holistic approach to identifying the patient's problems in context. This, in turn, enables the clinician to create a more appropriate management plan, to the patient's benefit [20]. A survey conducted to explore some of the wellbeing aspects of the Cerebral Palsy Integrated Pathway Scotland (CPIPS) concluded that 90 % of responding parents and carers of children agreed that CPIPS improved their child's overall wellbeing [12]. Despite this, anecdotally, the pain, functional, and QOL outcome measures included in the pathways seem to be limited [10].

This systematic literature review has therefore been conducted to establish whether high quality evidence exists linking valid outcome measures for pain, function and QOL with the hip displacement and dislocation outcomes of international integrated hip surveillance pathways. Such data is crucial for demonstrating the effectiveness and impact of such pathways on holistic patient outcomes.

1.1. Study design

This systematic literature review aims to identify and evaluate the evidence related to the effectiveness of integrated hip surveillance pathways on pain, function and quality of life outcomes for children with Cerebral Palsy.

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) was used to guide the design, conduct and reporting of this review [21]. The review protocol was not prospectively registered (for example, via PROSPERO).

A systematic review of randomised controlled trials (RCTs) and observational cohort studies was conducted. The literature search, screening for eligibility and subsequent critical analysis was completed by the lead researcher (TMcG). Another researcher (SP) validated all aspects of the review. The initial literature search and screening was conducted in May 2020. An updated search was completed in February 2023.

1.2. Search strategy

In line with protocols outlined by PRISMA [21] and Cochrane [22], relevant articles were identified by searching the CINHAHL, AMED, MEDLINE and PEDro databases. To increase search sensitivity in relation to the population group, British Nursing Database (BND) and PsycINFO were also included in the search strategy. The researcher's institutional academic library search engine (University of the West of England, Bristol, UK) was utilised, in addition to Google Scholar and the Chartered Society of Physiotherapy's (CSP) Discovery search engine.

1.3. Search terms

Search terms were developed by the lead researcher following appraisal of similar systematic reviews, consultation with the researchers' institution subject librarian and collaboration with another researcher (SP). The terms were finalised using a modified PICO format, with the 'Comparator (C)' removed. Key words and synonyms related to children, Cerebral Palsy, Cerebral Palsy Integrated Pathway, hip displacement and pain, function and QOL were thus devised (see Table 1). CINAHL, AMED, MEDLINE and PsycINFO databases were searched with the Elton B. Stevens Company Industries (EBSCO) host software with terms cross-referenced to title, subject terms and abstract. Similarly, the BND database advance search was completed using ProQuest software. The PEDro database advanced search function was utilised to increase specificity of the search. Although less specific, the Google Scholar and CSP Discovery search engines' advanced search functions were also employed utilising consistent search terms.

1.4. Eligibility criteria

The use of inclusion and exclusion criteria provided a clear framework for the selection of literature which met the aims of this review.

Papers published in English language (due to resource limitations)

Table 1
– Search terminology.

PICO	Key Word	Synonyms	Wild Cards
Population	Children	Child, young person, school aged, infant, teen, adolescent	Child* Teen*
	Cerebral Palsy	CP	Cere* Pals*
Intervention	Cerebral Palsy Integrated Pathway	CPIP, CPIPS, CPUP, NorCP Hip surveillance	CP*P
Outcome	Hip displacement	Hip dislocation, hip subluxation, hip migration	
	Pain, function, quality of life	QOL	

CP (Cerebral Palsy), CPIP (Cerebral Palsy Integrated pathway), CPIPS (Cerebral Palsy Integrated Pathway Scotland), NorCP (Norwegian CP Follow Up Programme), QOL (Quality of Life).

during or after 1994 were included in the study, aligning with the year of the pilot study for CPUP in Sweden [9]. As the specified intervention, all studies included were required to use a standardised integrated hip surveillance pathway. The population included were children and young people aged under 18 years of age, with a clinical diagnosis of Cerebral Palsy. Outcomes included in this study were hip displacement and/or dislocation, in addition to at least one other outcome measure for pain, function or QOL. This allowed evaluation of the impact of improvements in magnitude of hip displacement/dislocation on the relevant outcome measures for children with a diagnosis of CP. Review articles were not included.

1.5. Study selection

Following online searches, results were screened by the lead researcher (TMcG) in accordance with the inclusion and exclusion criteria. The systematic screening process consisted of screening in order of title and abstract, followed by full text as per the PRISMA flow chart for research selection. This process was repeated two weeks later, in reverse order of the search results, and without reference to the previous results, to check robustness of the selection process. Where doubt existed following review of a full text article, the researcher sought the opinion of another researcher (SP). At the end of this process, the results were identical for the number and identification of specific included studies (see Fig. 1). The search and screening process was updated in February 2023, following the same procedure. Once final articles were confirmed, data extraction was completed in a tabulated format (Table 2) to summarise key methodology principles and significant findings. Critical appraisal of the articles was completed through use of the Joanna Briggs Institute (JBI) checklist for observational cohort studies [23]. Critical appraisal of the included articles was completed by the lead researcher (TMcG) and another researcher (SP) independently, before comparing findings and agreeing on final outcomes (see Table 3). Findings of both the data extraction and critical appraisal informed the narrative data synthesis and discussion. Finally, each reviewer independently applied the Grading of Recommendations, Assessment, Development and Evaluations (GRADE) framework [24] to the strength of the evidence related to pain, function and quality of life. Final GRADE recommendations were agreed by consensus.

2. Results

The number of potential articles identified via the initial search was 110, with an additional three articles identified via the CSP Discovery search engine. Following removal of duplicates 100 articles were screened. 88 articles were excluded in line with the eligibility criteria, leaving 12 full text articles to be screened. During review of the full text articles, two were excluded as an integrated surveillance pathway was not used as the intervention, and six papers did not include an outcome measure for pain, function or QOL. As a result, four papers [25–28] were eligible for inclusion in this review.

2.1. Data extraction

Data was extracted from the included papers (Table 2) to identify evidence linking outcomes in pain, function or QOL for children undergoing integrated hip surveillance pathways. Included studies were published between 2019 and 2021, and involved participants included

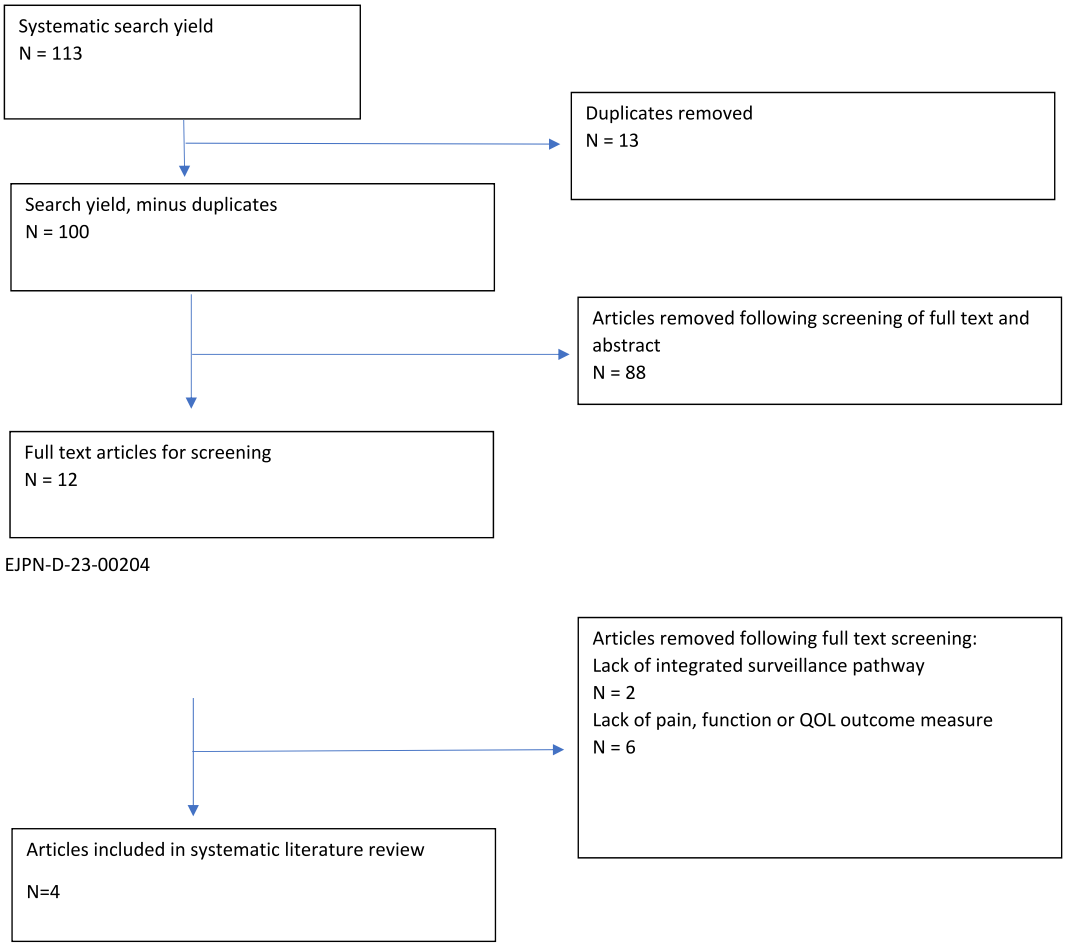

Fig. 1. PRISMA screening flow diagram.

Table 2
– Data extraction summary from included studies.

Citation	Marcstrom, Hagglund and Alriksson-Schmidt [25]	Schmidt, Hagglund and Alriksson-Schmidt (26)	Larsen, Ramstad and Terjesen (27)	Larsen, Terjesen, Jahnsen, Diseth and Ramstad [26]
Country	Sweden	Sweden	Norway	Norway
Year	2019	2019	2021	2021
Population	Whole population of children aged 4–16 with CP in Sweden, registered on the CPUP programme.	Whole population of children aged 2.5–16 with CP in Sweden, registered on the CPUP programme.	Non-ambulatory adolescents aged 12–17 years with bilateral CP, living in South-Eastern Norway.	Adolescents with CP aged 12–17 years. GMFCS III – V.
Number of participants (n)	N = 2890 (1230 females, 1660 males	N = 3256 (1395 females, 1861 males)	N = 67 (28 females, 39 males)	N = 64 (26 females, 38 males)
Study Design	Cross-sectional retrospective registry study	Retrospective population-based registry study	Longitudinal population-based study	Longitudinal population-based study
Intervention	Integrated hip surveillance pathway: CPUP Sweden.	Integrated hip surveillance pathway: CPUP Sweden.	Integrated hip surveillance pathway. Norwegian CP Follow Up Programme (NorCP).	Integrated hip surveillance pathway. Norwegian CP Follow Up Programme (NorCP).
Comparison group	None	None	None	None
Follow-up period	None	None	5 years	5 years
Outcome measures	Spasticity: Modified Ashworth Scale (MAS); Pain: dichotomous yes/no; Gross Motor Function: Gross Motor Function Classification System (GMFCS) I-V; Range of Movement (ROM): in degrees via goniometer and standardised CPUP methodology; Hip displacement: Migration Percentage (MP).	Spasticity: MAS; Scissoring; Hip displacement: MP; Bone and joint complications: ROM: in degrees via goniometer, windswept posture, scoliosis, hip displacement; Mobility: functional mobility scale (FMS), wheelchair use, ability to stand/get up from sitting/use stairs Pain: dichotomous yes/no in hips, knees, feet and back; Gross Motor Function: GMFCS.	Pain: Child Health Questionnaire (CHQ) pain-related questions, recorded as a dichotomous yes/no; Pain: Brief Pain Inventory numerical rating scale to assess interruption to daily activities; Hip displacement: MP; Structural deformities of the hip: pelvic radiographs.	QOL: 5 of the 6 Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) domains: *Activities of daily living and personal care , Positioning, transfer and mobility, Comfort and emotions, General health, and Overall QOL* Pain: CHQ pain related questions.
Findings	The prevalence data indicated trends towards increased hip pain prevalence with increasing age, GMFCS level and MP. Logistic regression confirmed increased odds ratios (OR) for hip pain for the following outcomes: Knee extensor spasticity (MAS level 2) (OR 2.81, 95 % confidence interval 1.18–6.69); Reduced (defined as 5°) hip inward rotation ROM (1.11, 1.01–1.20); Reduced hip flexion ROM (1.18, 1.05–1.32); Reduced hip abduction ROM (1.20, 1.04–1.41); 31–40 % MP (2.11, 1.1–4.06); and 41–100 % MP (2.72, 1.32–5.59).	Structural equation modelling identified that 'bone/joint complications' (incorporating hip abduction ROM, knee extension ROM, windsweeping and scoliosis) had the strongest direct pathway with pain in the lower extremities (standardised regression coefficient = 0.48). 'Mobility' (able to use stairs, able to stand, wheelchair use outside, and FMS 5m, 50m and 500m) was also statistically significantly associated with pain in the lower extremities (standardised regression coefficient = −0.24). Pathways between spasticity and pain, and age and pain were non-significant (p > 0.05).	Age, hip displacement (MP) and GMFCS level were identified as independent risk factors for both hip pain prevalence (p = 0.018, 0.004 and 0.013 respectively) and CHQ hip pain score (p = 0.044, 0.003 and 0.002 respectively). Following multivariable analysis, only GMFCS level remained as a risk factor for hip pain prevalence (p = 0.027), although it should be noted that MP could not be calculated due to a zero value in one of the categories. GMFCS (p = 0.001) and hip displacement (p = 0.005) both remained as risk factors for CHQ hip pain score following multivariable analysis. There was a higher prevalence of hip pain in those with GMFCS level V and MP 50–89 % (as compared to those with no hip pain) The mean CHQ hip pain score was lower (corresponding to worse pain) in patients with MP 50–89 % (mean 33 ± SD 22.5) than those with MP <50 % (77 ± 32.9, p = 0.003). The mean CHQ hip pain score was also lower in those with GMFCS level V (63 ± 35.9) compared to level IV (82 ± 28.1) or level III (96 ± 11.2). GMFCS (p = 0.030) and MP (p < 0.001) were independent risk factors for high interference of hip pain with daily activities but only MP remained following multivariable analysis (p < 0.001). GMFCS (p = 0.003) and MP (p < 0.001) were independent risk factors for high interference of hip pain with sleep and both remained following multivariable analysis (p = 0.011 and < 0.001 respectively). Prevalence of hip pain increased over	Mean CPCHILD scores decreased in *General health*, from 70 ± 23.2 in childhood to 65 ± 23.5 in adolescence (p = 0.021) when all participants were considered. There was also a significant reduction in *General health* scores in those with GMFCS level IV (80 ± 15.8 to 68 ± 23.6 respectively, p = 0.007). There were no changes over time in other CPCHILD domains (all p > 0.05). Mean CHQ pain scores decreased (corresponding to worse pain) from 64 ± 29 in childhood to 43 ± 26 in adolescence (p < 0.001). A final stepwise regression model predicting CPCHILD domain scores in adolescence incorporated age, GMFCS, CHQ pain score in adolescence and the respective domain scores in childhood. This model produced R^2 values ranging from 0.33 for *Overall QOL* to 0.63 for *General Health*. Within that model, statistically significant (p < 0.05) individual predictors were correspondingly low scores in childhood (all domains); lower CHQ pain scores (four domains, with the exception of *Activities of daily living and personal care*); and GMFCS level (*Activities of daily living and personal* care and *Comfort and emotions* domains only).

(*continued on next page*)

Table 2 (*continued*)

Citation	Marcstrom, Hagglund and Alriksson-Schmidt [25]	Schmidt, Hagglund and Alriksson-Schmidt (26)	Larsen, Ramstad and Terjesen (27)	Larsen, Terjesen, Jahnsen, Diseth and Ramstad [26]
			time (27 % of participants 5 years earlier, to 42 % in the present study, p = 0.041), whilst the mean MP remained unchanged (35 ± 22 % and 36 ± 24 % respectively, p = 0.577).	

CHQ (Child Health Questionnaire), CP (Cerebral Palsy), CPCHILD (Caregiver Priorities and Child Health Index of Life with Disabilities), CPUP (Uppfoljningsprogram for Cerebral Pares), FMS (Functional Mobility Scale), GMFCS (Gross Motor Function Classification Scale), MAS (Modified Ashworth Scale), MP (Migration Percentage), NorCP (Norwegian CP Follow Up Programme), QOL (Quality of Life), ROM (Range of Movement).

Table 3
Critical appraisal summary utilising the Joanna Briggs Institute Checklist for Analytical Cross-Sectional Studies (2017).

Citation:	Marcstrom, Hagglund and Alriksson-Schmidt [25]	Schmidt, Hagglund and Alriksson-Schmidt [27]	Larsen, Ramstad and Terjesen [28]	Larsen, Terjesen, Jahnsen, Diseth and Ramstad [26]
Item 1 Were the criteria for inclusion in the sample clearly defined?	Yes	Yes	Yes	Yes
Item 2 Were the study subjects and the setting described in detail?	Yes	Yes	Yes	Yes
Item 3 Was the exposure measured in a valid and reliable way?	Yes	Yes	Yes	Yes
Item 4 Were objective standard criteria used for measurement of the condition?	Yes	Yes	Yes	Yes
Item 5 Were confounding factors identified?	No	No	No	No
Item 6 Were strategies to deal with confounding factors stated?	No	No	Yes	Yes
Item 7 Were the outcomes measured in a valid and reliable way?	No	Unclear	Yes	Yes
Item 8 Was appropriate statistical analysis used?	Yes	Yes	Yes	Yes

on a national hip surveillance register in either Sweden or Norway. All four were population-based cohort studies, with 5 year follow up data included in the Norwegian cohort studies [28,26]. The Swedish cohort studies [25,27] provided analysis of a larger data set with a wider range of presentations (GMFCS I to V) whilst the Norwegian studies [28,26] focused on adolescent participants at GMFCS levels III to V only. Hip pain was reported on consistently in the four included studies, yet function was omitted from three studies [25,28,26] and QOL was also omitted in three of the four studies [25,27,28] included in this review.

2.1.1. Quality appraisal

Critical appraisal of the included studies was completed through use of the Joanna Briggs Institute (JBI) Checklist for Analytical Cross-Sectional Studies [23]. All included studies outlined inclusion and exclusion criteria clearly, detailing the demographics, location and time period of the study. A standardised integrated hip surveillance pathway was utilised in all included studies, with some variation in measurement of spasticity, resulting in inconsistencies in methodology and findings. Identification and mitigation of confounding variables was found to be a limitation across all four included studies. This was of particular interest in determining participant's use of tone management medications and interventions, in addition to data on previous surgical interventions in the Swedish cohort studies [25,27].

The GRADE evaluation of the strength of evidence was 'very low' for pain, function and quality of life. Risk of bias and inconsistency were the primary reasons for downgrading from 'low' to 'very low'.

3. Discussion

3.1. Summary of key findings

This systematic literature review has found evidence of relationships between prevalence of hip pain in children with CP and reduced ROM in the lower limbs, increased GMFCS level, and increased age. CP hip surveillance programmes did not seem to protect participants against developing hip pain during adolescence, although it should be acknowledged that these were observational studies and were not designed to assess effectiveness. There was limited evidence demonstrating relationships between spasticity and pain; and function and QOL outcomes for children undergoing hip surveillance were under-researched. Some limited evidence suggested a general health decline with increasing age, and an association between both increased MP and higher GMFCS level and interruption to activities of daily living. Further investigation is required to inform clinical decision making and planning of interventions for children with CP.

3.2. Migration percentage and range of movement

The findings of Marcstrom, Hagglund and Alriksson-Schmidt [25] indicated an association between degree of hip displacement and prevalence of hip pain. This is in addition to decreased ROM into abduction, flexion and internal rotation of the hip being significantly associated with hip pain prevalence. Similarly, Schmidt, Hagglund and Alriksson-Schmidt [27] highlighted bone and joint deformities, resulting in reduced ROM, as having the strongest direct association with pain in the lower extremities. However, there is some discrepancy in findings relating to the severity of hip displacement and prevalence of pain. Marcstrom, Hagglund and Alriksson-Schmidt [25] concluded that the majority of painful hips were not displaced to an extent requiring salvage surgery, whilst Larsen, Ramstad and Terjesen [28] reported that hip pain was associated most strongly with severe subluxation, as indicated by a MP of between 50 and 89 percent. It is important to note that the cohort studied by Larsen, Ramstad and Terjesen [27] was much smaller (n = 67) than Marcstrom, Hagglund and Alriksson-Schmidt [25]

(n = 2890) and Schmidt, Hagglund and Alriksson-Schmidt [27] (n = 3256). In addition, the focus was on participants with a GMFCS level III-V, of whom a greater proportion had undergone salvage surgeries. Furthermore, the participant cohort is from the Norwegian registry and this may be of significance as the prevalence of hip displacement in the Swedish population is so low that Schmidt, Hagglund and Alriksson-Schmidt [27] were unable to include MP as a variable for statistical analysis.

During statistical analysis, Marcstrom, Hagglund and Alriksson-Schmidt [25] included unilateral hip measurements only, with data for the side with the most spastic muscle group, the largest MP and the lowest recorded ROM included. Postural asymmetries were associated with contractures, hip deformity and inability to change position [29]. Improved internal validity would have resulted from clearer and evidence-based justification of such methodological decisions. Meanwhile Larsen, Ramstad and Terjesen [28] included each hip as a separate entity within their analysis, allowing exploration of differences in bilateral and unilateral presentations. Furthermore, this allowed the exploration of the impact of asymmetry resulting from windswept postures, related to increasing GMFCS level of the population group [6]. Schmidt, Hagglund and Alriksson-Schmidt [28] also detailed bilateral hip presentation information within their analysis with some acknowledgement and discussion of the impact of age on asymmetry in ROM at the hip. They theorised a later emergence of windsweeping postures with increased GMFCS levels as a result of early surgical and non-surgical interventions owing to the surveillance pathways. Significantly, there is a lack focus on functional or QOL outcomes within these studies, in comparison with the ubiquitous assessment of pain. Whilst there can be some implied links with the outcome measures of function and QOL, there is a lack of specific evidence to evaluate the effectiveness of integrated hip surveillance pathways on these outcomes. Larsen, Terjesen, Jahnsen, Diseth and Ramstad [26] began to explore these aspects of patient outcome, but it is clear that further work is required to fully understand the impact of the integrated pathways on biopsychosocial outcomes for children with CP.

3.3. Increasing age and GMFCS level

Larsen, Ramstad and Terjesen [28] and Marcstrom, Hagglund and Alriksson-Schmidt [25] both found a relationship between increasing age and prevalence of hip pain. What remains unclear is the causality of this progression over time. The natural development of CP results in a decline in functional abilities over time and therefore postural deformities increase [27]. Windsweeping and scoliosis are therefore more prevalent in older children and those with a higher GMFCS level. Furthermore, Larsen, Terjesen, Jahnsen, Diseth and Ramstad [26] highlighted increased GMFCS level and increased MP as key indicators for disruption of activities of daily living. In their follow up study, Larsen, Ramstad and Terjesen [28] found that the incidence of hip pain continued to rise despite MP remaining unchanged; and Larsen, Terjesen, Jahnsen, Diseth and Ramstad [26] found that reported general health scores for children with CP declined over time. These are important findings as one aim of integrated pathways is to improve quality of life for children with CP by reducing the incidence of hip displacement and dislocation. Larsen, Ramstad and Terjesen [28] concluded that the integrated surveillance programme did not protect the participants against the incidence of hip pain in adolescence. Furthermore, the effectiveness of the integrated pathway on functional and QOL outcomes for children with CP remains to be fully understood.

3.4. Spasticity

Spasticity has been defined as a velocity dependent increase in muscle tone [30]. Hip dislocation in children with CP has been attributed to increased tone in the adductors and medial hamstrings [6,7]. It has been hypothesised that increased spasticity reduces ROM and function, therefore reducing opportunity for weight bearing and development of typical patterns of articulation, leading to hip migration [6] and muscle contractures [31]. During this systematic literature review, there have been no direct links established between prevalence of hip pain and spasticity in children with CP. Despite referring to the same patient cohort, Marcstrom, Hagglund and Alriksson-Schmidt [25] and Schmidt, Hagglund and Alriksson-Schmidt [27] contradict one another on the relationship between spasticity and pain. Both studies assessed spasticity using the Modified Ashworth Scale (MAS). Systematic review and meta-analysis have concluded that inter and intra-rater reliability of the MAS is satisfactory [32]. However, the ability to differentiate between contracture and higher levels of spasticity, presenting as rigidity has been questioned [33]. Marcstrom, Hagglund and Alriksson-Schmidt [25] combined levels 3 and 4 on the MAS during statistical analysis, whilst Schmidt, Hagglund and Alriksson-Schmidt [27] combine scores of 1 and 1+. The combination of scores by Marcstrom, Hagglund and Alriksson-Schmidt [25] is potentially less valid as it combines MAS scores with reduced ROM with those where ROM is less affected, for the purposes of statistical analysis. It could be interpreted that muscle groups deemed most affected by spasticity might in fact have been presenting with contractures resulting in reduced ROM. Similarly, there were limitations in the measurement of spasticity by Schmidt, Hagglund and Alriksson-Schmidt [27] where scissoring of the lower limbs was simply categorised by the assessor as ‘none’, ‘’mild’ or’’pronounced’. There was a lack of detail described in relation to the criteria for this element of the assessment which may partly explain the contradiction in interpretation of results for this cohort of patients.

3.5. Pain and QoL

The findings of Larsen, Ramstad and Terjesen [28] and Larsen, Terjesen, Jahnsen, Diseth and Ramstad [26] indicate some evidence of a correlation between pain and overall QoL for adolescents meeting GMFCS level descriptors III-V. Utilising the Brief Pain Inventory to assess impact of pain on activities of daily living, Larsen, Ramstad and Terjesen [28] found increased MP and higher GMFCS levels to be independent risk factors for pain interference with sleep and activities of daily living, in addition to being risk factors for CHQ hip pain scores. Similarly, CHQ hip pain scores were used in a regression model to determine indicators for deterioration in general health and overall QoL scores by Larsen, Terjesen, Jahnsen, Diseth and Ramstad [26].

However, it is important to note the findings of Schmidt, Hagglund and Alriksson-Schmidt [27] within this context, who identify reduced ROM, windsweeping and scoliosis as additional indicators for increased incidence of pain in children with Cerebral Palsy. This suggests that pain may arise from areas other than the hip. Nevertheless, the relationship between pain and QoL is not opposed. Despite small population sizes in the Norwegian studies, these findings provide a rationale for greater exploration of the relationship between hip pain, general health, QoL and activities of daily living for children with Cerebral Palsy.

3.6. Limitations of the study

Despite a robust approach to critical appraisal of the included articles, the initial screening process was undertaken by only one reviewer. The included studies were observational, with pain not being the primary endpoint of those studies. Using the Grading of Recommendations, Assessment, Development and Evaluations (GRADE) framework [24], the observational nature of the included studies indicates a low baseline rating for the certainty of evidence included in this systematic review, which was further downgraded to ‘very low’ for evidence relating to pain, function and quality of life resulting from inconsistency and risk of bias. Furthermore, the low yield of included articles did not allow for meta-analysis of results.

Therefore, the main limitations are the low number of studies, observational nature of the available evidence, methodological

inconsistencies and risk of bias, preventing firm conclusions.

3.7. Future considerations

The association between age and hip pain observed in this systematic review is relevant when considering the interventions offered to children with CP, planning for transitions in life and changes in needs over time. There is wider evidence, not identified in the current review, of a general deterioration in activity levels, as shown by the regression of functional abilities in the GMFCS descriptors [34]. And there is also other evidence of the impact of reduced function on participation with age [35]. We can thus begin to reason a changing set of needs for this population over time [35].

Whilst the hip screening pathways have evidently reduced the incidence of hip dislocation in children with CP, there is also evidence of continuing hip pain in this population [5–9,11–14]. Hip pain has been reported to be negatively associated with ROM, function and QOL outcomes [36] yet function and QOL were omitted from evaluation of the hip pathways in three of the four studies included in this review. In addition, positive correlations between spasticity and age have previously been established with prevalence of hip pain [33] yet this review found no direct links between prevalence of hip pain and spasticity in children with CP. Furthermore, the integrated care aspect of these surveillance pathways describes the collaborative working practice between physiotherapists, orthopaedic surgeons and paediatricians in optimising tone management to reduce contractures and deformities and therefore reduce hip displacement [12]. Further work is therefore indicated to investigate the use of valid and relevant outcome measures to inform biopsychosocial outcomes supporting this model of management. Use of a QOL and functional outcome measure within such pathways could enhance the current clinical decision-making frameworks to optimise monitoring, follow up and rationale for the prescription and timing of future interventions for children with CP.

4. Conclusion

Trends associating increased prevalence of hip pain with increasing age and GMFCS level warrant further investigation to develop clinical practice and justify timely interventions received by children with CP.

The overall lack of high quality evidence linking hip displacement (via an integrated surveillance pathway) and outcomes for pain, function and QOL highlights the need for more strategic cohort studies, particularly given the opportunity to access large global data sets. This will facilitate evaluation of effectiveness on outcomes for pain, function and QOL for children with CP through enhanced assessment and outcome measures.

Disclosure

The authors have no conflicts of interest to disclose and no financial disclosure to make.

Conflict of interest

The authors have no conflicts of interest to declare relating to the review titled “Integrated hip surveillance pathways for pain, function and quality of life in children with Cerebral Palsy: A systematic literature review”.

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