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Effectiveness of surgical and non-surgical management of crouch gait in cerebral palsy: A systematic review

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Abstract

Background: Cerebral palsy (CP) is a prevalent group of neuromotor disorders caused by early injury to brain regions or pathways that control movement. Patients with CP exhibit a range of functional motor disabilities and pathologic gait patterns. Crouch gait, characterized by increased knee flexion throughout stance, is a common gait pattern in CP that increases energy costs of walking and contributes to ambulatory decline. Our aim was to perform the first systematic literature review on the effectiveness of interventions utilized to ameliorate crouch gait in CP.

Methods: Comprehensive searches of five medical databases yielded 38 papers with 30 focused on orthopaedic management.

Results: Evidence supports the use of initial hamstring lengthenings and rectus femoris transfers, where indicated, for improving objective gait measures with limited data on improving gait speed or gross motor function. In contrast, evidence argues against hamstring transfers and revision hamstring lengthening, with recent interest in more technically demanding corrective procedures. Only eight studies evaluated alternatives to surgery, specifically strength training, botulinum toxin or orthoses, with inconsistent and/or short-lived results.

Conclusions: Although crouch in CP is recognized clinically as a complex multi-joint, multi-planar gait disorder, this review largely failed to identify interventions beyond those which directly address sagittal plane knee motion, indicating a major knowledge gap. Quality of existing data was notably weak, with few studies properly controlled or adequately sized. Outcomes from specific procedures are confounded by multilevel surgeries. Successful longer term strategies to prevent worsening of crouch and subsequent functional decline are needed.

Level of evidence: Systematic review.

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The authors have no conflicts of interest to report.

Keywords

Cerebral palsy; Crouch gait; Hamstring lengthening; Pediatric orthopedics; Knee flexion

1. Introduction

Cerebral palsy (CP) is a group of motor disorders caused by non-progressive insults to the brain during early development. The resulting neuronal damage leads to various types and severity of motor involvement, and a range of primary symptoms such as spasticity, paresis, and poor motor control along with other secondary symptoms [1]. Pathological gait patterns such as crouch gait (also referred to as crouched or flexed knee gait) are common [2]. While definitions vary among experts and studies, crouch is generally characterized by an overly flexed knee during the stance phase of gait. A crouched posture reduces the capacity of muscles to extend the knee and hip [3], leading to generation of higher than normal muscle forces during gait [4], and is significantly less efficient [5]. Higher muscle forces lead to higher joint reaction forces [6], which may contribute over time to higher rates of joint pain and degenerative arthritis [7,8]. Left untreated or inadequately addressed, children with CP will experience progressive gait deterioration, leading to even greater functional disability [9–12]. Multiple potential causes of crouch have been proposed including hamstrings spasticity and contracture [13–16], gastrosoleus insufficiency [17], psoas spasticity and contracture [15,18,19], and quadriceps weakness [20], among other causes. Various treatments have been utilized to ameliorate crouch gait. While earlier reviews have investigated gait outcomes of interventions in CP [21,22], no systematic review specifically investigating treatments for crouch gait has been published. Our specific research question for this review was as follows: What is the universe of treatments reported in the scientific literature for the treatment of crouch gait in CP and what is the evidence for their effectiveness or efficacy? The purpose here was to comprehensively identify and evaluate outcomes of interventions in those with CP and crouch gait to inform clinical practice and guide future research.

2. Methods

2.1. Inclusion and exclusion criteria

Our goal was to identify all full length, peer reviewed, English language studies on interventions in CP aimed at correcting excessive knee flexion that provided objective quantification of changes in temporal-spatial aspects of gait and sagittal plane knee joint kinematics using 3-dimensional gait analysis as a result of a clinical intervention. Crouch gait may be referred to by other terms in the scientific literature such as “flexed knee” or “crouched gait,” and as such these were included in the relevant database searches. Additionally, studies that may not have called this specific gait pattern any of the above terms but described outcomes of treatments aimed at correcting excessive knee flexion during stance were included. Studies that investigated an intervention and its effect on multiple gait abnormalities must have stratified results by gait pattern to allow for analysis of that intervention’s effects on crouch. Included studies must also have reported 3-dimensional gait analysis (3DGA) data, specifically on sagittal plane knee kinematics, before and after intervention. Any nonhuman or modeling studies were excluded. Any case studies or studies without statistical analyses were likewise excluded.

2.2. Search methods

Databases searched were Pubmed, EMBASE (www.embase.com), Scopus (www.scopus.com), Web of Science (www.webofknowledge.com), and CINAHL up until August 13, 2015. The search term strategy, allowing for syntactical differences between databases, was as follows: “Cerebral Palsy” AND “crouch” OR “crouched” OR “flexed knee” OR “flex knee” OR “stiff knee” OR “knee flexion” OR “hamstring” OR “hamstrings” OR “knee” OR “gait disorders” OR “gait.” All terms were designated to be searched within both titles and abstracts.

2.3. Reference review

After identifying all citations from each database, duplicates were removed. Titles of remaining citations were independently reviewed by two authors to exclude any that were clearly not investigations of interventions for improving crouch in CP. Remaining abstracts were then reviewed by three authors and inclusion/exclusion criteria applied with disagreements resolved by discussion. Reference lists of included citations were reviewed to further ensure all relevant studies were identified. Manuscripts for each citation were obtained and reviewed independently by three authors.

2.4. Data extraction and quality appraisal

Study type, design, sample size, intervention type and follow-up times were extracted from each manuscript. Extracted patient demographic information included gender, age, GMFCS level, and CP subtype, where available. Collected outcomes focused primarily on knee kinematics (all measured in angles) and included pre and post treatment knee flexion at initial contact (KFIC), maximum knee extension in stance (MKESt), maximum knee flexion in swing (MKFSw), total knee excursion (TKE), and mean pelvic tilt (MPT). Additional data extracted included popliteal angle, cadence, walking speed, and stride length as well as any functional outcomes.

The strength of each study design was assessed using Sackett’s Levels I–V of evidence [23]. Since it was immediately apparent that few studies would be Level I–II, the Methodological Index for Non-Randomized Studies (MINORS) was applied to assess scientific rigor of each study [24]. Three authors performed each of these determinations with disagreements resolved by discussion. This review was constructed in accordance with the PRISMA statement and guidelines, a set of evidence-based criteria for reporting in systematic reviews [25].

3. Results and discussion

3.1. Search results

The search strategy yielded 1998 unique citations from five databases. After title and abstract review, 57 remained for full text review. Review of references from these identified 7 additional papers, leaving 64 papers for full text review [13,14,19–21,26–85]. One additional paper was identified and added during the manuscript review process which recommended that we search for additional terms to ensure that our review had not failed to capture the outcomes of interventions that addressed lower limb alignment on crouch. These additional terms included: lever" OR "anteversion" OR "torsion" OR "malalignment". Of note, this additional search did not yield any new unique citations. The one new paper that emerged had been eliminated previously because the title and abstract did not mention crouch or flexed knee gait, but was selected this time for full text review because it appeared under multiple search terms and was an intervention study that included kinematics. Of 65 full papers evaluated, 27 were excluded for not meeting eligibility criteria and 38 papers (including this new paper) comprised the final selection (Fig. 1, Table 1). Relevant data from each were extracted as detailed above (Tables 2 and 3).

3.2. Study type, design, and size

Of the 38 papers, 28 were retrospective cohort studies, 8 were prospective cohort studies 1 was a meta-analysis of three prospective cohort studies, and 1 was a randomized controlled trial (Table 2 and 3). Thirteen studies were comparative or included a control (Table 3). Results from approximately 1250 unique patients with CP across 38 studies were reviewed. Sample sizes ranged from 8 to 111 patients.

3.3. Participant demographics

The mean age across studies was 10.5 years. Of the 28 studies reporting gender, 56.7% of the studied patients were male. CP types varied across studies and included spastic diplegia, hemiplegia, and quadriplegia. Reported GMFCS levels ranged from I–IV.

3.4. Interventions

Orthopaedic surgical interventions were the focus of 30/38 papers with 27 of those addressing effects of hamstring lengthening (HSL) or transfer (HST) with or without associated procedures such as rectus femoris transfers (RFT) or psoas lengthenings. The remaining 3 surgical studies investigated procedures such as distal femoral extension osteotomy (DFEO) and patellar tendon advancement/shortening (PTA/PTS) [38,67,68]. The remaining 8 papers studied the effects of strengthening [20,34,69] botulinum toxin injections [19,31,60], and orthoses [65,85] on crouch gait.

3.5. Follow up and outcome measures

Follow up periods for surgical studies ranged from 6 months [54] to 11.8 years [70] with 7 reporting outcomes at least three years post-surgery. Outcomes included a wide array of clinical, kinematic, spatiotemporal and functional measures. All publications reported at least one knee kinematic outcome of interest obtained via three dimensional gait analysis (3DGA) since that was an inclusion criterion, and 20 included spatiotemporal measures and only 4 reported functional measures (Tables 2 and 3).

3.6. Level of evidence and study quality

Sackett's levels of evidence ranged from level II (n = 1) to level IV (n = 26), with 11 studies of level III [23]. The average MINORs score was 11.9 out of 24 possible points. Most received low scores for being retrospective studies without adequate control or comparative groups (Tables 2 and 3).

3.7. Outcomes and discussion

Crouch gait is a common problem in a condition prevalent in the developed and developing world [2,86]. This review indicates that orthopaedic surgery remains the dominant intervention to treat crouch in CP with hamstring lengthening still the most common approach, as has been the case for decades. Scientific evidence supports some general surgical recommendations, but does not definitively state which procedures among those studied are most effective for improving biomechanical outcomes in specific patients and provides little data on expected functional changes or long term effects. Despite the recognition that crouch in CP often has a complex multijoint, multiplanar and multisymptom etiology, this review failed to yield knee kinematic outcomes data from surgical procedures directed at other joints and planes commonly utilized in clinical practice to improve lower limb alignment. The literature also largely failed to support successful alternatives to surgery.

An apparent issue in CP literature is a lack of consensus on crouch definitions which varied across studies in the considered joints, planes of motion, and phases of gait [32,47,56,67]. In this review, we allowed for a more inclusive view of crouch gait by including all interventional studies that stated they were addressing crouch or flexed knee gait and/or had 3D kinematic data showing that the knee was excessively flexed at initial contact and/or later in stance. Ankle and hip position or rotational abnormalities were neither an inclusion or exclusion criteria for article selection which depended solely on whether the paper presented pre-post intervention kinematic data on a group or subgroup of subjects all of whom had excessive knee flexion in stance. If we had only included papers that explicitly stated they were addressing crouch, 15 papers would have been reviewed. Of those, 10 provided definitions of crouch, with only 3 using the Rodda et al. definition of crouch gait as both increased knee flexion and ankle dorsiflexion throughout stance [54,64,65,87]. The rest had variable definitions, some of which defined crouch as increased KFIC (n = 3) [20,32,34], and others as increased knee flexion throughout stance (n = 4) [38,47,54,69]. We also included studies that addressed “flexed knee gait” without mention of crouch (n = 7) [13,27,38– 40,43,67] because these satisfied the same knee kinematic criterion, although with similar ambiguity in the precise definition of when in stance the flexion occurred. Most of the remaining papers (n = 15) targeted hamstring contracture or spasticity but did not state they were addressing crouch. These were included here since hamstrings procedures are typically reserved for those with increased knee flexion and all had kinematic data supporting this. Any study explicitly stating that the entire sample had jump-knee gait was excluded, even though KFIC was increased. However, given our more inclusive selection criteria, we acknowledge that some subjects across studies may have had jump-knee gait, especially in those studies that used KFIC to define crouch [20,32,34] or those where the sample was described as containing both crouch and jump-knee gait. In the latter case, if data were reported separately for the different gait patterns, we only extracted and reported the outcomes for the subgroup with crouch. Finally, the entire universe of treatments for crouch gait may not be fully represented here, in part because we excluded studies on crouch that did not present 3D kinematic data. This likely restricted the type and range of interventions and may have skewed the interventions that were included towards greater inclusion of surgical procedures. Future studies should aim to investigate intervention effects on specific, well defined gait patterns or stratify patient outcomes by gait patterns rather than present outcomes on heterogeneous group of patients.

Crouch gait can result from variable combinations of muscular, neurologic, and/or bony pathologic processes. Imperative to successful treatment of crouch gait is an understanding to the mechanism and time course of gait disability in children with bilateral CP. While the hamstrings are often targeted in crouch gait, other muscle or joint abnormalities can precipitate the development or progression of crouch such as planovalgus foot deformity [52] or hip flexion contractures [15,18,19]. A patient may initially walk with increased sagittal plane knee flexion due to hamstring or psoas spasticity [13–16,18,19], quadriceps or gastrocsoleus weakness [17,20], and/or lever arm dysfunction from malalignment or rotational bony deformities that develop as a result of imbalanced muscle forces across joints [52,64,88]. With time, shortening and dynamic contracture of the hamstrings may develop into a static contracture, preventing full active and passive knee extension [88]. Finally, rapid bone growth and weight gain during puberty may lead to progressive gait deterioration in multiple joints and planes and loss of mobility in adolescence and adulthood [88,89].

The multifactorial nature of crouch gait complicates the construction and interpretation of a review such as this, as the underlying deficits are likely different in patients undergoing the various interventions studied here. For example, a child with a dynamic but not yet static hamstring contracture and no bony deformity is not a candidate for muscle-tendon lengthening or femoral extension osteotomy procedures, and is more likely to be treated with braces, botulinum toxin, and physical therapy, limiting the utility or validity of a comparison of these interventions [90]. Future studies should aim to provide as much pre-intervention data on the patient population as possible, including but not limited to type and pattern of CP, functional ability, clinical exam measurements, all relevant lower limb kinematics, and radiologic findings in those with bony deformity.

Only eight studies were identified on non-surgical approaches to crouch gait. The mean age of the patients included in these studies was substantially younger than the age of patients in the surgical studies (9.7 years and 11.3 years, respectively). This trend is not surprising given the proposed mechanism of crouch gait development and progression. Strengthening, orthoses, and botulinum toxin are more likely to be deployed earlier in life before the development of static contractures which would then require surgical correction. Strengthening and orthoses may also be utilized post-operatively to optimize surgical outcomes. The reviewed alternative procedures and their outcomes in this study must then be considered relative to the degree and type of contractures as well as relative to patient size and age.

Hamstrings were lengthened or transferred in 27 of 30 surgical studies. Only 1 study had an adequate, non-hamstring lengthened control group [14], and one other reported outcomes after isolated HSL procedures [13]. Diverse surgical methods of hamstring lengthening are reported, including tenotomies, z-lengthenings, fractional lengthenings, and aponeurotic lengthenings which could differentially affect outcomes, yet no comparative studies were identified.

Guidelines for which hamstrings to lengthen are also not well-established. One noted strategy was to lengthen only medial hamstrings to prevent untethering of the pelvis, anterior pelvic tilt (APT) and genu recurvatum (GR), leaving the lateral hamstrings intact to prevent hyperextension. Another was to lengthen lateral hamstrings based on intraoperative examination after medial hamstring lengthening [36,37]. One comparative study showed 9 greater improvement in MKESt in the medial plus lateral versus the medial only group at 1.5 years. However, 30% in the medial plus lateral group developed GR versus 5% in the medial group [53]. Discrepancies in GR rates were not found in a similar study which had a 12% rate at 8 years in both groups [39]. Additional comparative evidence showed greater improvements in KFIC and MKESt in the medial and lateral versus medial group, although they had even greater flexion preoperatively [36]. Importantly, they found that medial and lateral HSL in patients with normal preoperative pelvic positioning produced significantly increased APT compared to those with increased pelvic tilt prior to surgery or who had medial HSL. More extensive medial and lateral hamstrings lengthening carries a significant risk of increasing APT, but may be necessary in those with severe static contractures to adequately increase knee extension.

One concern with HSL or HST is development of limited postoperative knee flexion in swing (stiff-knee gait) as these procedures often shift the sagittal knee kinematics curve into more extension. Multiple groups have investigated the role of rectus femoris transfer (RFT) to the medial hamstrings. Proposed benefits are two-fold: to augment the hamstrings flexion moment while diminishing excessive rectus activity limiting knee flexion in swing. While debate exists on mechanisms potentiating increased peak swing flexion [18,91], previous studies have demonstrated improved MKFSw after RFT as part of SEMLS [39,92,93]. Aiona et al. reported outcomes in 111 patients with HSL without RFT (n = 57), HSL plus RFT (n = 28), and HSL with delayed RFT (n = 26) at another surgical event. Those who had RFT plus HSL showed no decrease in mean peak knee flexion in swing, while the HSL alone group had significantly decreased mean MKFSw at 1 year. Those with later RFT showed improved MKFSw back to baseline, suggesting HSL procedures exacerbate limited knee flexion in swing and RFT may prevent this [27]. Further evidence from non-comparative studies corroborated this [26,30,47,54,76]. One study showed patients with limited knee flexion preoperatively had significantly improved TKE and MKFSw with RFT, while patients treated with RFT prophylactically before developing stiff-knee gait had 15° deterioration of MKFSw at 9 years [94]. Preoperative gait analysis may help predict the likelihood of RFT success in those with knee flexion limitations [95]. Some have cautioned that RFT may further compromise already weakened knee extensor function, increasing the risk of long term crouch recurrence [93].

Since HSL may also further weaken weak muscles [33] and resultant increased hamstring laxity may lead to excessive APT, hyperlordosis, back pain, and other gait problems, hamstrings transfers have been considered as an alternative, e.g. the Eggers procedure introduced in the 1950s which transferred all hamstrings to the femoral condyles to potentially reduce hamstring laxity. While this soon fell out of favor due to concerns about

GR and loss of active knee flexion [96,97], renewed interest has surfaced in procedures that convert bi-articular to mono-articular muscles (CBM), such as HST to the femoral condyles. Besides biomechanical benefits, CBM procedures are theorized to improve neural control in CP as mono-articular muscles require less complex control strategies [98]. A retrospective cohort study reported outcomes after CBM of the gastrocsoleus, rectus femoris and semitendinosus versus outcomes after HSL and GSL. No significant group differences in spatiotemporal measures were found at 9 years. However, GR rates in the CBM group were two times higher at 1 and 9 years, with similar rates of increased anterior pelvic tilt [40]. A study comparing HST to gracilis and hamstring lengthening found no group differences except for increased APT from 15.7° to 22.2° only in the HST group [43] with another study comparing HSL and HST showing nearly identical results [37]. Interestingly, most reported rates of GR between HST and HSL groups have been similar [39,40,56,57]. Despite one study showing slightly superior APT outcomes in the CBM group compared to HSL and GSL [40], the group differences were clinically insignificant. HST fails to demonstrate any superior outcomes to justify use.

Although HSL demonstrates positive effects on knee biomechanics, multiple modeling studies postulate that short hamstrings may not be the primary cause of crouch in many patients. A short iliopsoas may instead be responsible for pulling the pelvis anteriorly and stretching the hamstrings at their origin thus making them appear too short at the knee [18,99,100]. One report on 8 patients with psoas lengthenings and 9 without during SEMLS, with similar kinematics preoperatively, showed improved MKESt for both, but increased APT only in the psoas group [77]. Another found improvements in psoas and no psoas groups in KFIC and MKESt, but instead found no significant changes in pelvic tilt at 1.5 years [36]. Reliance on passive range of motion measures alone which correlate poorly with gait analysis results and hence actual dynamic gait function [101,102] may be especially problematic when assessing true hamstring length in CP for surgical decision making [77].

Available evidence does not support the effectiveness of revision HSL. One study compared outcomes of primary versus revision HSL procedures in 39 patients, and found that primary procedures improved knee kinematics and popliteal angle at 2 years while revision HSL was ineffective [63]. A similar study in 61 patients reported improved KFIC, TKE, and popliteal angle 1 year after primary HSL with revision HSL only improving KFIC [32]. Better alternatives to revision HSL may require different surgical approaches or other less invasive strategies. There is emerging interest in correcting knee flexion deformities through distal femoral extension osteotomy and improving quadriceps extensor function through patellar tendon advance/shortening [68,103]. One retrospective study reported outcomes from DFEO alone, DFEO and PTA, and PTA alone [68]. All groups showed significant improvements in KFIC and mean improvements in MKESt of 9°, 29°, and 17°, respectively. All had increased APT, largest in the combined group. Notably, 61% of these 73 patients had prior HSLs [68]. Two other studies reported similar improvements in MKEST after DFEO during SEMLS in 12 patients [38] and PTS during SEMLS in 24 patients [67]. However, APT increased with these procedures [38,67] and knee flexion deformity recurred in 27% after DFEO [38]. Complications, including persistent pain, neuropathy, postoperative deformity, and infection, were reported in 18–19% in one study [68]. Improved surgical techniques including more advanced locking plates for internal fixation are now available with lower complication rates

[68,104,105]. DFEO procedures involve the removal of a wedge of bone from the femur, and the post-operative benefits reported may be attributable to an effective lengthening of the hamstrings relative to the femur [106]. While these procedures offer a promising alternative, more investigation of positive and negative effects is needed.

Bony deformities such as pes planovalgus, femoral anteversion, and external tibial torsion may decrease the ability of multiple muscles to extend the hip and knee, referred to as lever-arm dysfunction, thereby exacerbating crouch gait [52,88,107]. Rotational osteotomies to correct femoral anteversion and external tibial torsion, were performed in many of studies included that were primarily investigating HSL, HST, and RFT in the setting of SEMLS [14,27,32,37,39,40,43,46,54,59,62,64,70,73,74,76], so the differential effects of bony surgeries on crouch were not discernible. However, examination of these procedures in isolation has previously demonstrated improved knee kinematics [108,109]. Similarly, another study not included in the review because it did not perform a separate statistical analysis on the subset of participants who had crouch, did demonstrate through correlational analyses of the entire sample that a greater improvement in the pes planovalgus deformity and excessive ankle dorsiflexion after surgery was associated with greater knee extension in stance [52].

Three studies investigated the effects of hamstrings botulinum toxin injections [19,31,60]. Modestly improved knee kinematics were seen 2 weeks post injection but disappeared by 12 weeks. Repeated injections are commonly utilized to facilitate muscle stretch during growth and purportedly delay or prevent development of static contractures [110,111]. Three studies investigated effects of short term strength training with inconsistent results. One showed decreased KFIC and improved mobility after a 6 week quadriceps strengthening program, while two others reported no change in any measures. Although modeling studies indicate that adequate strength of all lower limb extensor muscles is important to prevent crouch [4,112], strengthening hip extensor muscles may exacerbate hamstring contracture in those with spasticity [69]. Selective, longer duration strengthening may be effective in augmenting or maintaining outcomes after HSL or botulinum toxin injections if the etiology is multifactorial; however, more research is needed to support this hypothesis.

Orthoses are highly prescribed and utilized interventions in CP [113], with floor reaction ankle foot orthoses designed specifically for crouch gait [64,114]. A study by Rogozinski et al. investigated effects of already prescribed FRAFOs compared to barefoot walking in 27 patients. Patients had an 11° improvement in MKESt with FRAFOs and increased walking speed and step length as well [65]. The only RCT in our review by Abd El-Kafy investigated the effects of a strapping system and GRAFOs on the gait of CP crouch patients after a 12-week training program. Patients were randomized to receive either gait training, gait training + de-rotational strapping (Thera-Togs™), or gait training + de-rotational strapping and GRAFOs during the study. All groups underwent a rigorous 12-week gait training program with their randomized interventions. The group randomized to strapping and GRAFOS showed the largest improvement in knee kinematics from baseline, and was the only intervention group to differ from the control, gait training group, suggesting that the GRAFOs, and not de-rotational strapping devices, were responsible for the kinematic improvement [85]. Effects of AFOs that control excessive dorsiflexion on crouch remain largely unknown but improvements in upright stance seem biomechanically plausible and initial data suggests that GRAFOs do improve knee extension in stance. Conversely, a brace stretching the bi-articular gastrocnemius to achieve a more plantigrade position in mid-stance could exacerbate knee flexion.

Disappointingly, no studies were identified on other potentially effective interventions such as selective dorsal rhizotomy (SDR) or functional electrical stimulation (FES) on crouch. Studies on SDR have shown generalized improvements in functional measures and knee kinematics in CP [115–118]. FES has demonstrated effectiveness in addressing other gait abnormalities in CP so another future research direction could be exploring its role in alleviating crouch.

Another major shortcoming apparent in the current literature is the lack of functional outcomes reported in 31 of 38 studies [32,34,56,60,64,68]. While the authors acknowledge that inclusion criteria may have excluded studies from this review without kinematics that reported functional outcomes, the current imperative in the care and management of patients with CP is to improve current and future functional mobility and degree of participation in the community. While gait analysis data can measure the direct effects of interventions, kinematics have only a weak correlation with functional ability measures [119–121]. Many well-validated patient reported outcome measures of mobility and physical functioning are now available and are strongly recommended for inclusion in future trials evaluating crouch in CP such as the Functional Assessment Questionnaire (FAQ) [119], the Gross Motor Function Measure (GMFM) [122], Functional Mobility Scale (FMS) [123], the Pediatric Outcomes Data Collection Instrument [124], and the Pedi-CAT [125]. Additionally, a previous study by Kondo et al. and a study included in this review by Yngve et al. have demonstrated differential changes in both functional and objective measures of gait based on patient preoperative functional ability [76,126]. While many studies did report the range of patient GMFCS levels studied, only one stratified results by preoperative functional group or ability. Given that these studies have demonstrated strong predictive value of pre-treatment GMFCS, future studies should strongly consider stratification by functional grouping in order to better advise future individualized patient care and management.

Of those studies that did report the type and pattern of CP involvement, few reported outcomes by sub-group or provided individual data, preventing any analysis of intervention effect on patients of different CP diagnoses. Especially relevant to this review, a few studies investigated the outcomes of interventions in a heterogeneous group that included children with both unilateral and bilateral involvement. The definition and mechanism of flexed-knee gait varies between these two diagnostic groups, and crouch gait is not typically described in children with hemiplegia [7]. The varying underlying pathology of these CP involvement patterns likely also alters the effectiveness of the specific interventions between groups, warranting stricter patient selection or reporting stratification.

Nearly all studies reviewed were small, retrospective, and uncontrolled with many surgical studies having inadequate baseline patient characterization and confounding by concomitant procedures. Given its overwhelming prevalence, McGinley et al. called for more rigorous investigation of SEMLS through RCTs [21]. A subsequent RCT in 19 patients demonstrated improved kinematic outcomes for SEMLS [127] compared to strength training with the sample size too small to evaluate specific procedures. While this study demonstrated that it is possible to evaluate SEMLS with this rigorous type of design that temporarily delayed surgery in the control group, randomizing children to surgery or no surgery when they have a static contracture poses ethical challenges. Additionally, large scale RCTs require considerable resources in terms of patient and project staff time as well as cost [128]. Alternative approaches are gaining traction to document clinical outcomes including establishment of patient registries for both ongoing quality improvement and outcomes research and the implementation of large-scale prospective observational trials to compare effectiveness of surgical approaches. These pragmatic clinical trials do not require randomization and instead utilize advanced statistical methods to control for pre-operative patient differences.

This review highlights a few key issues in the current CP literature that need to be resolved to advance the current state of knowledge supporting clinical practice for those with crouch and other gait abnormalities. The joints and planes of motion and points in the gait cycle used to define crouch gait vary widely in the literature, with no clear consensus [129]. We propose here that a minimum requirement for crouch gait includes excessive knee flexion during the period after weight acceptance until terminal stance., It is also evident that in many cases, sagittal as well as other 3D deformities at the ankle-foot complex and at the hip contribute to the crouch posture as well as interfere with optimal lower limb alignment and should be carefully documented before and after any intervention that may affect muscle length or joint position. These data would inform future discussions aimed at achieving consensus on definitions and more clearly delineate different subgroups that may require different types or combinations of treatments.

This review highlights the paucity of kinematic evidence for other non-surgical interventions commonly used to address crouch including rigid AFOs, dynamic stretching devices and stretching exercises, among others. Additionally, although nearly all of the available literature investigated surgical interventions targeted at the knee joint, it is important to view crouch gait as a disorder of the entire lower limb. Patients can have deformities or limitations at the hip joint, or ankle-foot complex, or deformity of the lower-limb long bones that contribute to crouch gait. The lack of knee kinematic data on the various procedures that address those deformities is a major gap in the literature that warrants further study.

In conclusion, the only well-supported intervention for crouch in CP has long been and remains HSL even though surgical practices have evolved beyond this more simplistic approach. Regardless, even with current treatment, rates of ambulatory decline in adults with CP remain staggering [130]. Lengthening procedures also further weaken already weak muscles. Future research should focus on continued non-surgical and surgical innovations that optimally preserve strength while enhancing alignment and motion.

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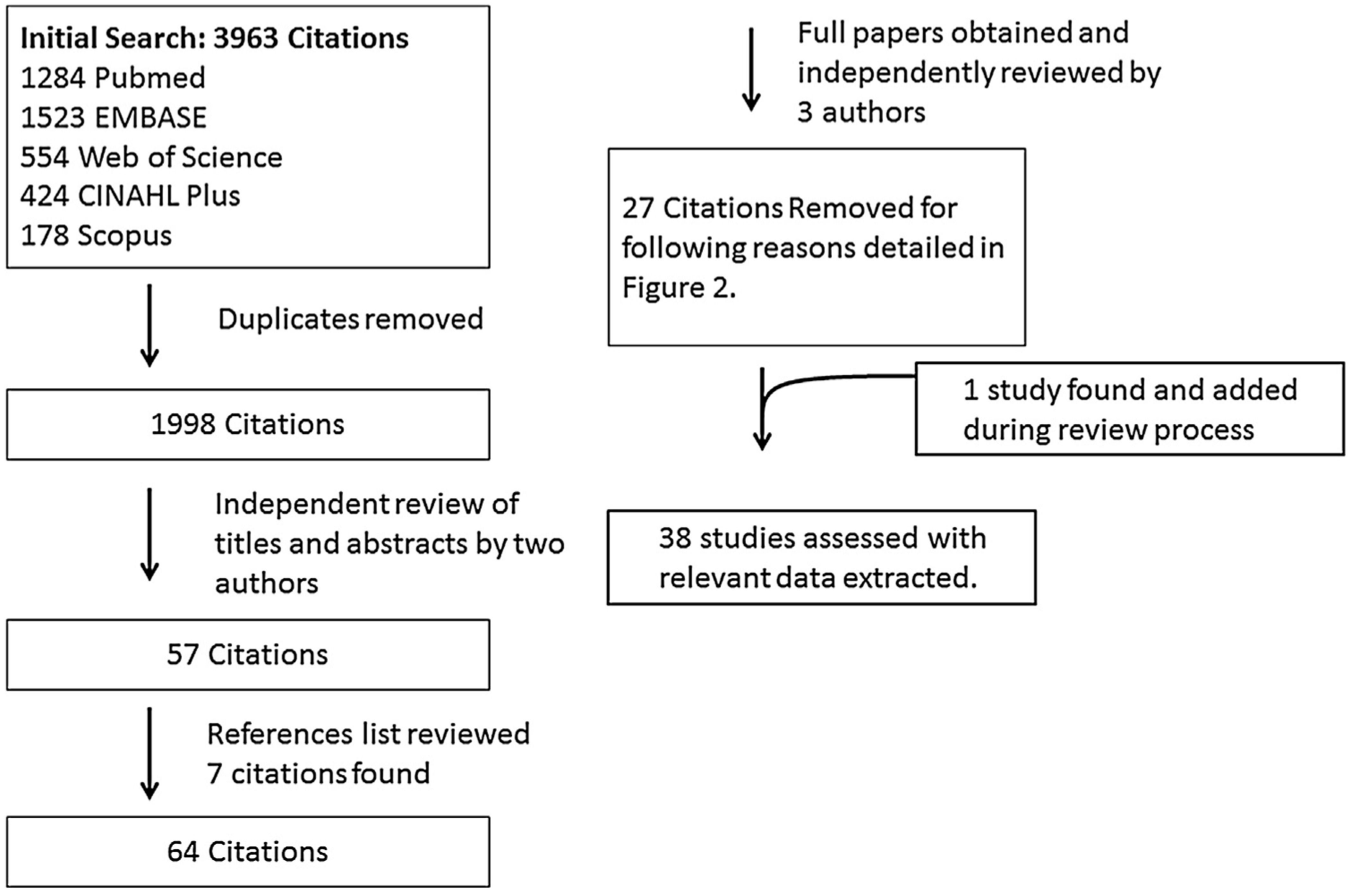

Fig. 1.
Systematic search and review strategy and results.

Table 1

Reasons for exclusion for 27 of 65 publications after full text review. 3DGA – Three dimensional gait analysis. ST-Spatio-temporal.

Reasons for Exclusion	
Baumann [28]	No 3DGA or ST measures
Bozinovski [29]	No 3DGA or ST measures
Das [35]	No 3DGA or ST measures
Drummond [41]	No 3DGA or ST measures
Eek [42]	Not specific to crouch
Gage[44]	Treatment of stiff knee gait, not crouch gait
Ganjwala [45]	No 3DGA
Gannoti [78]	Interventions not specific to crouch gait
Gough [75]	No aggregate group data
Haumont [48]	Not all patients had preop data
Hesse [49]	Not specific to crouch
Hsu [50]	No 3DGA or ST measures
Joseph [51]	No 3DGA or ST measures
Kadhim [52]	Some patients had normal knee flexion, not stratified
Lucarelli [79]	Inadequate statistical analysis/reporting
McGinley [21]	Interventions not specific to crouch
Morton [58]	Intervention not targeted at crouch, no 3DGA
Park [61]	Treatment of jump-knee gait, not crouch gait
Patritti [84]	Case reports with no statistical analysis
Rethlefsen [62]	Not limited to CP population
Roosth [66]	No 3DGA or ST measures
Scholtes [80]	No 3DGA or ST measures
Sutherland [71]	No 3DGA or ST measures
Svhelik 2011 [81]	Inadequate 3DGA reporting
Thometz 1989 [72]	Inadequate statistical analysis/reporting
Unger 2006 [82]	Not specific to crouch
Wesdock 2003 [83]	Not a gait study

Table 2

tracted data from the 24 non-comparative studies reviewed. All data in parentheses is either SD of mean value or range of values. RC – Retrospective
hort, PC – Prospective cohort.

haracteristic and Outcomes of Non-Comparative Studies

udy	Intervention	Study	MINORS Score	# of patients	# of males	Average Age (yrs)	Type of CP	GMFCS Level	Mean #of procedures	Mean follow up (mo)		KFIC (°)	MKES t(°)	MKFSw (°)	TKE (°)	Cadence (steps/ min)	Walking Speed (m/s)	Stride Length (m)	Mean Pelvic Tilt (°)	Popliteal Angle (°)
dolfsen [26]	HSL, RFT, and GSL	RC	8	15		8.5 (2)			3	23	Pre Post	37 (7) 24 (10)	24 (5) 16 (11)	NS NS	NS NS					57 (10) 42 (10)
rney [30]	HSL and RFT, SEMLS	RC	8	23	14	11.75 (2.5)		I-III	3.2	> 12	Pre Post		26 (13) 12 (14)	57 (9) 50 (12)	31 (11) 67 (14)					
rney [13]	HSL alone	RC	8	16	8	12 (8–16)		I-III	1	>12	Pre Post		33 (10) 10 (13)	64 (9) 48 (11)	31 (12) 39 (12)					
rry [31]	Botox injection of Hamstrings	PC	13	10	3	7.2 (4–11)			1	3	Pre Post	NS NS	NS NS	NS NS	NS NS	NS NS	NS NS		20 (6.5) 25 (7.9)	64 (12.8) 47 (7.3)
miano [34]	Extensor Strengthening	PC	15	8	3	7.78 (2.6)	SD	I-III		2	Pre Post		NS NS			NS NS	NS NS	NS NS		
miano [33]	HSL in SEMLS	PC	19	13		9.46 (3.2)				9	Pre Post				30.9 39	NS NS	NS NS	0.71 0.84		
miano [20]	Quadriceps Strengthening	PC	12	14	10	9.1 (2.5)	SD			1.2	Pre Post	32 (10) 27 (15)	NS NS		NS NS	NS NS	NS NS	0.74 (0.2) 0.82 (0.15)		
Morais [38]	DFEO in SEMLS	RC	7	12	5	13.1 (9.1–19.9)	SD	II-IV	5.2	27	Pre Post		43.56 () 22.29 ()						12 21.9	
rdon [46]	Percutaneous medial HSL	RC	8	48		9.45	SD, SH, SQ	I-III		33.8	Pre Post	26.6 (9.9) 20.1 (12.3)	NS NS			NS NS	0.9 (0.34) 1.0 (0.27)	0.86 (0.34) 0.99 (0.2)	NS NS	49.6 (11.9) 41.5 (17)
dley [47]	RFT and HSL in SEMLS	RC	7	24		11 (5–18)	SD, SH			>3	Pre Post		23.2 3.2	NS NS	30.7 46.1					

aracteristic and Outcomes of Non-Comparative Studies

udy	Intervention	Study	MINORS Score	# of patients	# of males	Average Age (yrs)	Type of CP	GMFCS Level	Mean #of procedures	Mean follow up (mo)		KFIC (°)	MKES t(°)	MKFSw (°)	TKE (°)	Cadence (steps/ min)	Walking Speed (m/s)	Stride Length (m)	Mean Pelvic Tilt (°)	Popliteal Angle (°)
ca [54]	RFT and HSL in SEMLS	RC	8	19	9	12.1 (11–27)	SD		5.2	6.3	Pre Post	43.8 (4.1) 20.1 (2.1)	40.5 (NR) 18 (7)	NS NS	14 (2.1) 32 (1.4)	112 87	0.42 (0.1) 0.56 (0.2)	0.41 (0.06) 0.57 (0.08)		61 (5.2) 22 (3.8)
vejoy [55]	HSL during SEMLS (Med and Lat)	RC	9	38	25	12.33 (4.6–19.6)	SD			21.6	Pre Post	43.9 (12.8) 26.8 (10.8)	51.5 (12) 39 (10.6)	41.2 (10.5) 26.6 (9.9)						57.8 (11.1) 37.2 (10.3)
a [56]	HST/HSL	RC	9	19	10	9 (5–14)	SD, SQ	III-IV	6.7	25.2	Pre Post	39 (8) 22 (7)	39 (8) 22 (7)							70 (9.2) 42.6 (9.5)
etaxiotis [57]	CBM (RFT, HST, GST)	PC	14	20	13	11.5 (5.6–17.0)	SD		12.05	37.2	Pre Post	41.5 (15.4) 19.1 (12.8)	28.7 (23.4) 6.2 (14.5)	58.3 (12.9) 51.4 (8.1)	31.9 (15.2) 45.2 (13.9)	118 (25) 103 (24)	NS NS	0.6 (0.14) 0.67 (0.13)		53.6 (13.6) 25 (12.6)
npuu [59]	HSL, GSL, RFT in SEMLS	RC	12	22	11	8 (2.7)		I-III	4.7	132	Pre Post	35 (9) 24 (9)	NS NS		NS NS	139 (23) 115 (15)	NS NS	0.79 (0.22) 0.98 (0.23)	NS NS	
padonikolakis 0]	Botox injection of hamstrings	PC	20	14		11 (217)	SH, SQ, SD			49	Pre Post	NS NS	14 (9) 10 (9)	NS NS	NS NS	NS NS	NS NS	L - 0.70 (.2) L - 0.76 (.2)		
dda [64]	SEMLS for severe crouch	RC	11	10	7	12 (7.9–16.2)	SD	II-III	7	60	Pre Post	52 (7) 26 (10)	44 (9) 17 (11)						14 (12) 24 (9)	NS NS
gozinski [65]	FRAFO	PC	9	27	18	12.4 (2.4)	SD, ST, SQ	II-III			Bare AFO		29 (14.3) 18 (14.4)			NS NS	.72 (.25) .83 (.19)	.84 (.19) 1.00 (.17)		
ssai [67]	PTS in SEMLS	RC	15	24	16	16.1 (5.8)	SD	II-III	4.6	21.8	Pre Post	51.1 (18.2) 32.4 (10.8)	32.4 (22.8) 20.5 (13.9)	71.9 (16.5) 57.8 (10)	55 (19) 37.6 (12.2)				14.1 (6.8) 21.6 (6.6)	
eele [69]	Strength Training	MA	12	30		13.7 (3.8)		I-III		2	Pre Post	NS NS	NS NS							

aracteristic and Outcomes of Non-Comparative Studies

udy	Intervention	Study	MINORS Score	# of patients	# of males	Average Age (yrs)	Type of CP	GMFCS Level	Mean #of procedures	Mean follow up (mo)		KFIC (°)	MKES t(°)	MKFSw (°)	TKE (°)	Cadence (steps/ min)	Walking Speed (m/s)	Stride Length (m)	Mean Pelvic Tilt (°)	Popliteal Angle (°)
ng [70]	HSL in SEMLS	RC	10	29	18	8.3 (5.1–16.3)	SD	I-III	9.1	141.6	Pre Post	31.1 (12.7) 23.6 (8.1)	NS NS	31.4 (12.7) 23 (7.8)	NS NS	NS NS	0.71 (.25) 0.87 (.23)		NS NS	
ompson [19]	Botox injection of hamstrings	PC	11	10		7.2 (4–12)	SQ, SD			0.5	Pre Post	NS NS	26 12.42						NS NS	
n der Linden 3]	HSL during SEMLS (Med and Lat)	RC	8	18		11 (6–20)	SH, SD			13	Pre Post	42 (12) 30 (12)	32 (13) 20 (14)	63 (12) 54 (10)	30 (9) 34 (11)	60 (9) 56 (10)	NS NS	NS NS	NS NS	
estwell [74]	HSL in SEMLS	RC	9	21	9	18.9 (14.2–29.3)	SH, SD	I-IV		13.2	Pre Post	32 (15) 22 (13)			36 (12) 42 (16)					58 (12) 31 (18)

Table 3

Extracted data from the 13 comparative studies reviewed. All data in parentheses is either SD of mean value or range of values. With the exception of Abd El-Kafy 2014, an RCT, and Dreher 2013, a prospective cohort study, all studies in this figure were retrospective cohort studies.

Characteristic and Outcomes of Comparative Studies

Study	Group Description	MINORS Score	Number of patients	Average Age (yrs)	Type of CP	GMFCS Level	Mean follow up (mo)		KFIC (°)	MKESt (°)	MKFSw (°)	TKE (°)	Cadence (steps/ min)	Walking Speed (m/s)	Stride Length (m)	Mean Pelvic Tilt (°)	Popliteal Angle (°)
Abd El-Kafy [85]	PT	20	19	7.5 (1.2)	SD	I-II	3	Pre		35.4 (4.0)			64.5 (3.3)	0.74 (.06)	.69 (.03)		
								Post		31.5 (4.2)			68.6 (4.4)	0.80 (.06)	.77 (.04)		
	PT + TheraTogs		19	7.3 (1.6)				Pre		36.9 (3.2)			63.4 (4.4)	0.76 (.06)	.68 (.05)		
								Post		30.9 (2.6)			69.3 (3.5)	0.81(.05)	.83 (.04)		
	PT + TheraTogs + GRAFO		19	7.1 (1.5)				Pre		36.3 (3.2)			62.5 (4.4)	0.74 (.05)	.71 (.03)		
								Post		24.4 (3.9)			73.4 (3.9)	0.86 (.06)	.89 (.04)		
Aiona [27]	HSL w/ Delayed RFT	9	26	11.6 (3.2)		I-III	12	Pre		26.5 (13.7)	55.7 (11.2)	29.2 (10.5)					
								Post HSL		5.5 (12.7)	41.5 (10.5)	35.9 (13.8)					
								Post RFT		8.8 (11.6)	51 (7.5)	42.2 (11.5)					
	HSL with RFT		57			I-III	12	Pre		27.1 (14.7)	NS	28.6 (13.3)					
								Post		9.8 (11.5)	NS	44 (12.6)					
	HSL only		28			I-III	12	Pre		22.8 (7.1)	56.8 (11.7)	34 (13.2)					
								Post		6.7 (8.2)	47.7 (8.4)	41 (9.6)					
Chang [32]	Primary HSL	14	49	8.7 (4.3)	SD, SH, SQ, SM		14.6	Pre	39.1 (11.1)			42.1 (15.8)				21.3 (5.6)	52 (12)

Characteristic and Outcomes of Comparative Studies

Study	Group Description	MINORS Score	Number of patients	Average Age (yrs)	Type of CP	GMFCS Level	Mean follow up (mo)		KFIC (°)	MKESt (°)	MKFSw (°)	TKE (°)	Cadence (steps/ min)	Walking Speed (m/s)	Stride Length (m)	Mean Pelvic Tilt (°)	Popliteal Angle (°)
								Post	28.0 (11.4)			48.6 (14.5)				23.3 (6.9)	41 (16)
	Repeat HSL		12	12.7 (2.9)			16.4	Pre	35.4 (11.9)			NS				20 (9.2)	NS
								Post	25.0 (8.6)			NS				25.9 (8)	NS
DeLuca [36]	HSL (Med)	15	37	8.9 (4.3)	SD		12	Pre	30 (11)	11 (14)						NS	42 (17)
								Post	17 (12)	3 (12)						NS	30 (17)
	HSL (Med and Lat)		12	14.4 (5.8)	SD		12	Pre	37 (11)	27 (18)						NS	59 (12)
								Post	19 (8)	9 (9)						NS	38 (11)
	HSL (Med) and PL		9	10.6 (4.3)	SD		12	Pre	33 (9)	11 (9)						NS	NS
								Post	21 (9)	1 (11)						NS	NS
	HSL (Med and Lat) and PL		15	10.5 (3.6)	SD		12	Pre	32 (10)	16 (17)						NS	55 (11)
								Post	21 (6)	7 (13)						NS	32 (18)
De Mattos [37]	HSL	13	18	11.6 (3.4)	SD, ST, SQ, SH	I-III	51.6	Pre		22.4 (13.3)						NS	58.9 (14.4)
								Post		10.8 (12.1)						NS	51.1 (12.3)
	HST		32	10.5 (2.9)		I-III	54	Pre								14.1 (7)	57.7 (10.7)
					SD, ST, SQ, SH					24.9 (11.9)							
								Post		12.5 (14.3)						19.3 (7.9)	49.4 (10.1)
Dreher [39]	HSL (Med)	10	30	10.2 (3.5)	SD	I-III	97.2	Pre	36(17)	NS			NS	NS	.82(.21)	NS	NS

Characteristic and Outcomes of Comparative Studies

Study	Group Description	MINORS Score	Number of patients	Average Age (yrs)	Type of CP	GMFCS Level	Mean follow up (mo)		KFIC (°)	MKESt (°)	MKFSw (°)	TKE (°)	Cadence (steps/ min)	Walking Speed (m/s)	Stride Length (m)	Mean Pelvic Tilt (°)	Popliteal Angle (°)
								Post	23(10)	NS			NS	NS	.99(.20)	NS	NS
	HSL (Med and Lat)		9					Pre	45(14)	35(23)			NS	NS	NS	NS	NS
								Post	23(10)	12(16)			NS	NS	NS	NS	NS
Dreher [40]	CBM (HST, RFT, GST)	16	21	11.3 (3.1)		I-III	110.4	Pre	41 (14)	28 (20)	58 (12)	30 (13)	NS	NS	NS	NS	NS
								Post	24 (8)	13(11)	52 (9)	39 (12)	NS	NS	NS	NS	NS
	HSL		21	11.1 (5.4)		I-III	109.2	Pre	41 (16)	28 (21)	61 (13)	33 (13)	NS	NS	NS	14 (8)	NS
								Post	27 (8)	16 (10)	56 (7)	40 (11)	NS	NS	NS	17 (7)	NS
Feng [43]	HSL	14	20	11.9 (3)	SD, SH	I-III	13.1	Pre		31.7 (11.5)						NS	61.4 (13.5)
								Post		23.2 (11.6)						NS	46.2 (9.9)
	HST		18	10.1 (3.4)	SD, ST, SQ	I-III	12.8	Pre		32.6 (11)						15.7 (6.1)	57.8 (11.7)
								Post		18.9 (9.8)						22.2 (6.9)	45.3 (14.7)
Kay [53]	HSL (Med and Lat)	13	21	11.2 (4.5)	SH,SD		17	Pre		27 (15)							53(11)
								Post		12 (16)							33 (13)
	HSL (Med)		16	9.2 (4.8)	SH, SD		18	Pre		20 (12)							51 (12)
								Post		16(17)							41 (11)
Laracca [14]	HSL (Med and Lat)	13	15	14 (11–17)	SD		27	Pre	44.5 (14.5)	NS				NS	NS	NS	49.7 (13.9)
								Post	31.7 (14.9)	NS				NS	NS	NS	42 (14.6)
	No HSL during SEMLS		15	12.6 (10–14)	SD		21	Pre	NS	NS				0.92 (0.32)	.46 (.08)	NS	NS

Characteristic and Outcomes of Comparative Studies

Study	Group Description	MINORS Score	Number of patients	Average Age (yrs)	Type of CP	GMFCS Level	Mean follow up (mo)		KFIC (°)	MKESt (°)	MKFSw (°)	TKE (°)	Cadence (steps/ min)	Walking Speed (m/s)	Stride Length (m)	Mean Pelvic Tilt (°)	Popliteal Angle (°)
								Post	NS	NS				0.68 (0.35)	.40 (.12)	NS	NS
Rethlefsen [63]	Primary HSL	15	21	9.2 (4.3)		II-IV	19.2	Pre	45 (12)	30 (16)							55 (12)
								Post	28 (9)	12 (15)							37 (12)
	Repeat HSL		18	10.8 (3.7)		II-IV	24	Pre	NS	NS							NS
								Post	NS	NS							NS
Stout [68]	DFEO	12	16	14.8 (6)		I-IV	13.2	Pre	43 (18)	40 (17)		NS		NS		NS	19 (9)
								Post	34 (16)	31 (16)		NS		NS		NS	5(11)
	DFEO and PTA		33	14.1 (2.6)		I-IV	14.4	Pre	42 (12)	38 (15)		25 (15)		NS		15(9)	15 (8)
								Post	26 (11)	9 (10)		47 (15)		NS		21(7)	−1 (5)
	PTA Only		24	12.9 (2.5)		I-IV	14.4	Pre	35 (9)	27 (11)		32 (16)		NS		16(6)	3 (7)
								Post	24 (9)	10 (12)		46 (16)		NS		22(8)	−1 (5)
Yngve [76]	Ind. Amb. (DRFT and HSL)	9	39	11	SD,SQ		13.6	Pre		20 (15)	NS	31 (13)	131 (20)	NS	0.9 (0.18)		
								Post		7 (13)	NS	43 (14)	126 (20)	NS	0.94 (0.19)		
	Dep. Amb. (DRFT and HSL)		39	9.8	SD,SQ		13.6	Pre		26 (23)	54 (16)	28 (14)	107 (26)	NS	0.63 (0.18)		
								Post		7 (16)	47 (12)	39 (16)	99 (27)	NS	0.72 (0.18)		
			21	9.8	SD,SQ		13.6	Pre		31 (18)	56 (12)		NS	0.27 (0.11)	0.4 (0.15)		
	Household Amb. (DRFT and HSL)											25 (13)					

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Characteristic and Outcomes of Comparative Studies

Study	Group Description	MINORS Score	Number of patients	Average Age (yrs)	Type of CP	GMFCS Level	Mean follow up (mo)		KFIC (°)	MKESt (°)	MKFSw (°)	TKE (°)	Cadence (steps/ min)	Walking Speed (m/s)	Stride Length (m)	Mean Pelvic Tilt (°)	Popliteal Angle (°)
								Post		12 (13)	45 (9)	33 (15)	NS	0.4 (0.18)	0.55 (0.17)		
Zwick [77]	DRFT + HSL + Psoas tenotomy	18	8	11.3 (2.7)	SD		28.8	Pre	31.8 (4.4)	11.4 (5.6)			NS	NS	0.91 (0.16)	17.8 (7)	52.5 (1.6)
								Post	25.6 (9.8)	1.3 (6)			NS	NS	1.05 (0.30)	29.5 (6.8)	20.5 (9.3)
	DRFT + HSL		9	11.2 (2.7)	SD		28.8	Pre Post	NS NS	NS NS			NS NS	NS NS	0.96 (0.19) 1.15 (0.13)	NS NS	52.5 (8.7) 30 (8.2)