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Review

The effect of femoral derotation osteotomy on transverse plane hip and pelvic kinematics in children with cerebral palsy: A systematic review and meta-analysis



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ABSTRACT

The purpose of this study was to systematically review the current literature to determine the effect of a femoral derotation osteotomy (FDRO) on hip and pelvic rotation kinematics during gait compared to no intervention in children with spastic cerebral palsy (CP). We performed a systematic search for prospective and retrospective cohort studies of children with CP, who were treated with a FDRO, and were assessed with pre and post surgery three-dimensional gait analysis. Medline, CINAHL, EMBASE, the Cochrane Library and Web of Science were searched up to December 2013. Data sources were prospective and retrospective studies. Mean differences were calculated on pooled data for both pelvic and hip rotation kinematics. Thirteen of 196 articles met the inclusion criteria (5 prospective, 8 retrospective). All included studies were of sufficient quality for meta-analysis as assessed using a customised version of the STROBE checklist. Meta-analysis showed that FDRO significantly reduced pelvic retraction by 9.0 degrees and hip internal rotation by 17.6 degrees in participants with unilateral CP involvement and hip internal rotation by 14.3 degrees in participants with bilateral CP involvement. Pelvic symmetry in children with unilateral spastic CP is significantly improved by FDRO. Patients with bilateral involvement do not improve their transverse plane pelvic rotation profiles during gait as a result to FDRO, although this result should be interpreted with caution due to the heterogeneous nature of these participants and of the methods used in the studies assessed.

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1. Introduction

Internal hip rotation (IHR) is common in children with cerebral palsy (CP) and is a major contributor to an intoed gait [1]. The consequences of IHR include knocking or rubbing of the knees, increased occurrence of trips and falls, and altered foot pressure distribution, which may result in pain and excessive shoe wear [2,3]. The underlying mechanism of IHR in children with CP is a combination of dynamic and static factors. Dynamic factors are due to spasticity, abnormal tone, contracture, and/or muscle imbalance in the adductor, hamstring, gluteal and tensor facia lata muscles

[4–11]. The static factor is an excessive femoral anteversion angle that reduces the mechanical advantage of muscles that cross the hip joint (i.e. hip abductors and glutei) leading to less efficient muscle contribution to forward propulsion during gait [6,12]. The reduction in mechanical advantage is commonly referred to as lever arm deficiency because the increased femoral anteversion results in a reduced coronal plane hip abductor moment arm. To compensate, patients commonly internally rotate the hip to maximise the hip abductor moment arm and subsequently the contribution of these muscles during gait.

The established orthopaedic intervention to address IHR in children with CP is single event multilevel surgery (SEMLS), which involves simultaneous correction of the dynamic and static contributors to the IHR. The accepted orthopaedic intervention to correct excessive femoral anteversion in children with CP is a femoral derotation osteotomy (FDRO), which can be performed at a proximal (intertrochanteric) [13] or distal (supracondylar) [3] level with comparable post surgical gait outcomes [3,14]. The literature

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provides good evidence for correction of IHR during gait one to three year post SEMLS [3,14–18], emerging evidence that correction is sustained in the long term [16,17,19], and some suggestion that children who have surgery after the age of 10 years have better retention [16,20].

Internal hip rotation can develop unilaterally (spastic hemiplegia or asymmetrical bilateral involvement) or bilaterally (participants with bilateral involvement). Children with unilateral involvement have been reported to compensate for IHR by retracting their pelvis on the impaired side to normalise the foot progression angle [21], however, other authors have found no significant change in pelvic rotation postoperatively and suggest that pelvic rotation may be a primary deformity caused by pelvic obliquity, spinal deformities and/or muscle imbalance at the hip and pelvis [22]. Currently there is no consensus in the literature regarding the effect of FDRO on pelvic rotation during gait in children with unilateral and/or bilateral CP involvement. This lack of agreement is due to limited participant numbers, different inclusion criteria and different analysis techniques across previous studies. The purpose of this study was to systematically review the current literature to determine the effect of FDRO (unilateral or bilateral) on hip and pelvic rotation kinematics during gait compared to no intervention in children with spastic CP. We hypothesise that FDRO will be affective in addressing internal hip rotation and transverse plane pelvic asymmetry in children with unilateral and bilateral spastic CP.

2. Materials and methods

2.1. Search strategy

In order to identify the key papers on this topic, a comprehensive search was undertaken of the following computerised databases: Pubmed (1980-December 2013), CINAHL (1982-December 2013), EMBASE (1980-December 2013), the Cochrane Library (1993-December 2013) and Web of Science (1980-December 2013). The search strategy used included MeSH terms and text words for 'cerebral palsy' AND 'osteotomy' AND '(biomechanics OR gait OR locomotion OR kinematics)'. References

from key papers were also scanned to ensure that all key studies were included.

2.2. Inclusion criteria

Inclusion criteria stipulated that studies incorporate a pre and post surgery three-dimensional gait assessment of children with specified unilateral or bilateral CP who were treated with a femoral de-rotation osteotomy (Population - CP patients, Intervention - femoral derotation osteotomy, Comparison - pre and post surgery, Outcome - hip and/or pelvic kinematics during gait). For studies that were excluded see the study flow diagram (Fig. 1).

2.3. Study collection and quality evaluation

The titles and abstracts of papers retrieved in the initial searches were screened independently by the three authors (CC, TP and JE) after removing duplicates. Assessments were included following agreement by all three raters, and any conflicting viewpoints were discussed until a consensus was reached. Full text articles were then sought and the independent screen process was repeated by the three authors. Quality assessment was undertaken independently by three authors (CC, TP and JE) using a customised version of the STROBE checklist for cohort studies [23], whereby questions 6b, 12c, 12d and 22 were removed due to irrelevance to the studies assessed. The STROBE checklist provides recommendations on the reporting of observational research. Items in the checklist relate to title, abstract, introduction, methods, results and discussions sections of articles providing best practice guidelines [23]. Studies scoring above 70% on the checklist were considered to have adequate internal validity for quantitative meta-analysis. Conference abstracts were excluded.

2.4. Data extraction

Data extracted from each study included population demographics, surgical details including type of femoral de-rotation, indication for FDRO and any potential surgical procedures that may confound the kinematic relationship between FDRO and pelvis

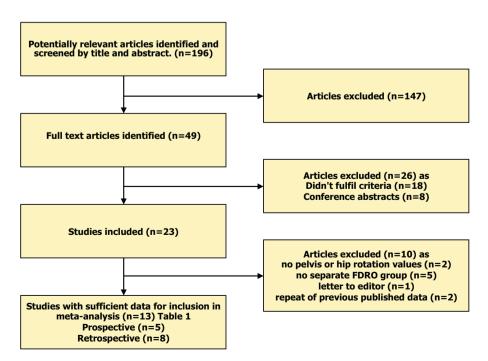


Fig. 1. Systematic search strategy results.

Table 1Study design and population characteristics.

Study	Study design	Subgroups	Participants (n)	Sex (m/f)	Age (years)	•		Follow (years)	up		Unilateral/ bilateral CP	Unilateral /bilateral FDRO	
					Mean	SD	Range	Mean	SD	Range			
Aminian et al. (2003) [24]	R		9	_	8.5	-	4-13	1.1	-	-	9/0	9/0	
Chung et al. (2008) [22]	R		34	21/13	8	_	5-11.6	1.8	-	1-2.3	34/0	34/0	
de Morais Filho et al. (2013) A [27]	R	Below LT	24	12/11	9.2	_	_	3.4	-	_	1/23	12/12	
de Morais Filho et al. (2013) B [27]	R	Above LT	29	17/12	12	_	_	3.7	-	_	4/25	23/6	
Dobson et al. (2005) [25]	P		17	14/3	12.9	4.4	7.1-17.1	2.9	0.9	2.0 - 5.5	17/0	17/0	
Dreher et al. (2007) A [15]	P	↑ affected	30	22/8	10	2.9	_	0.9	0.7	_	0/30	3/27	
Dreher et al. (2007) B [15]	P	↓ affected	27	20/7	10	2.9	_	0.9	0.7	_	0/27	0/27	
Kay et al. (2004) A [21]	R	↑ affected	19	6/13	8.3	2.5	_	1.5	0.6	_	3/16	6/13	
Kay et al. (2004) B [21]	R	↓ affected	16	4/12	8.2	2.3	_	1.5	0.7	_	0/16	3/13	
Kim et al. (2005) [16]	R		30	14/16	9.2	_	4.8 - 17.2	1 ^a	-	_	0/30	15/15	
Kwon et al. (2013) [29]	R		25	16/9	6.8	1.5		1.0	0.2		0/25	0/25	
Ounpuu et al. (2002) [17]	P		20	_	8.1	2.9		1 ^a	-	_	2/18	13/7	
Pirpiris et al. (2003) A [3]	P	Prox FDRO	14	9/5	11.9	2.1	9-14.6	1 ^a	_	-	0/14	0/14	
Pirpiris et al. (2003) B [3]	P	Dist FDRO	14	10/4	12.9	2.6	7.6-16.2	1 ^a	-	-	0/14	0/14	
Rutz et al. (2012) [26]	R		11	6/5	11.1	2.7	7-16	1 ^a	-	_	11/0	11/0	
Saraph et al. (2002) A [28]	R	Diplegia	8	_	12	_	_	3.2	-	_	0/8	8/0	
Saraph et al. (2002) B [28]	R	Hemiplegia	14	_	11.4	_	_	3.1	-	_	14/0	14/0	
Thompson et al. (2010) A [30]	P	MI FDRO	10	8/2	10.6	_	7.1-13.9	1 ^a	-	_	0/10	0/10	
Thompson et al. (2010) B [30]	P	FDRO	10	4/6	11.4	-	7.9-14.4	1 ^a	-	_	0/10	0/10	
Total			361	183/127	9.36 ^b			1.64 ^b			95/266	168/193	

^{- =} not reported, FDRO = Femoral derotation osteotomy, CP = cerebral palsy, R = Retrospective, P = Prospective, MI = minimally invasive, LT = lesser trochanter, ↑ affected = more affected lower limb. □ affected lower limb.

rotation kinematics, age at surgical intervention, follow up time, mean hip rotation during gait and mean pelvic rotation during gait. For the purpose of categorising CP involvement, hemiplegic participants were classified as having (predominately) unilateral CP involvement and participants with spastic diplegia, triplegia or quadriplegia were classified as having bilateral CP involvement.

2.5. Data synthesis

Further quantitative analysis was conducted in Review Manager (RevMan), version 5.2 for Windows, (The Nordic Cochrane Centre, The Cochrane Collaboration, Copenhagen, Denmark). Pooled data for treatment effect were calculated across trials with a variable effect model. Data were analysed with effect sizes, mean differences and 95% confidence intervals (CI). Heterogeneity was evaluated by using the I^2 statistic, with larger percentage scores representing greater heterogeneity.

 Table 2

 Methodologic quality assessment of include studies: STROBE checklist.

3. Results

3.1. Search Results

With our search we retrieved 196 articles, with 49 remaining after deletion by title and abstract. Included articles and reasons for the exclusion of 36 of the 49 articles for which full text was obtained are listed in Fig. 1. Thirteen empirical articles were included. The study participants, study design and surgical interventions are summarised in Table 1. There were 5 prospective cohort studies and 8 retrospective cohort studies. Six studies were split into distinct subgroups as denoted in Table 1 by an 'A' or 'B' and defined in the subgroup column.

3.2. Qualitative Analysis

The methodological quality of included studies is presented in Table 2. STROBE checklist scores ranged from 21.5 to 26.5

• . •																						
Study	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	Total
	/2	/1	/1	/ 1	/2	/1	/2	/2	/1	/1	/ 1	/2	/1	/3	/1	/1	/ 1	/ 1	/1	/ 1	/1	/28
Aminian et al. (2003) [24]	1	1	1	0.5	1.5	1	1.5	1.5	0.5	0.5	1	2	1	2	1	1	1	1	0.5	1	1	22.5
Chung et al. (2008) [22]	2	1	1	1	1.5	1	1.5	1.5	0.5	1	1	2	1	2	1	1	1	1	1	1	1	25
de Morais Filho et al. (2013) [27]	1.5	1	1	1	2	1	2	2	0	1	1	2	1	3	1	1	1	1	1	1	1	26.5
Dobson et al. (2005) [25]	1	1	1	1	2	1	2	2	0	1	1	1.5	1	3	1	1	1	1	0.5	1	1	25
Dreher et al. (2007) [15]	0.75	1	1	1	1.5	1	2	2	1	0.5	1	2	0.75	1.75	1	1	1	1	0.5	1	1	23.75
Kay et al. (2004) [21]	2	1	1	1	1.5	1	2	1.5	0	1	1	2	1	3	1	1	1	1	0	1	1	25
Kim et al. (2005) [16]	1.25	1	1	0.5	2	1	2	2	0	1	1	2	1	3	1	1	0.5	1	0.5	1	1	24.75
Kwon et al. (2013) [29]	2	1	0.75	1	2	1	1.5	2	0.5	1	1	2	1	1.75	1	1	1	1	1	1	1	26.5
Ounpuu et al. (2002) [17]	1	1	1	1	2	1	1.5	2	0.5	1	1	2	1	3	1	1	1	1	0.5	1	1	25.5
Pirpiris et al. (2003) [3]	1.5	1	0.5	1	1.5	1	1.5	1.5	1	1	1	2	1	3	1	1	1	1	1	1	1	25.5
Rutz et al. (2012) [26]	1.5	1	1	1	1.5	1	1.5	1.5	0	1	1	1.5	1	3	1	1	1	1	1	1	1	24.5
Saraph et al. (2002) [28]	1	1	0	0.5	1	1	1	1	0.5	1	0.5	2	1	3	1	1	1	1	1	1	1	21.5
Thompson et al. (2010) [30]	1	1	0.5	1	1.5	1	1	1.5	1	0.5	1	2	1	2.5	0.5	1	0.5	1	0.5	1	1	22

The modified STROBE checklist criteria (1) title and abstract; (2) background/rationale; (3) objectives; random allocation; (4) study design; (5) setting; (6) participants; (7) variables; (8) data sources/measurement; (9) bias; (10) study size; (11) quantitive variables; (12) statistical methods; (13) reporting of participants; (14) descriptive data; (15) outcome data; (16) main results; (17) other analyses; (18) key results; (19) limitations; (20) interpretation and (21) generalisability.

^a Approximate value.

^b Weighted mean adjusted for number of participants.

Table 3Type of femoral derotation osteotomy (FDRO) and percentage of participants that underwent additional soft tissue and bony procedures at time of FDRO.

Study	Type of FDRO	Calf lengthening ^a	Tib post lengthening	Adductor lengthening	Hamstring lengthening	Psoas lengthening	Rectus transfer	Tib A/P transfer	TDRO	Other ^b
Aminian et al. (2003) [24]	Prox	56	33	67	56	22	11	0	0	22
Chung et al. (2008) [22]	Prox	90	0	10	70	10	30	0	0	60
de Morais Filho et al. (2013) A [27]	Prox	42	17	3	53	0	11	3	8	25
de Morais Filho et al. (2013) B [27]	Prox	63	14	11	63	22	46	6	11	20
Dobson et al. (2005) [25]	Prox	76	18	47	76	12	71	0	0	0
Dreher et al. (2007) A [15]	Prox/Dist	79	_	2	86	11	95	40	14	50
Dreher et al. (2007) B [15]	Prox/Dist	79	_	2	86	11	95	40	14	50
Kay et al. (2004) A [21]	_ `	42	0	32	58	32	63	0	0	0
Kay et al. (2004) B [21]	_	_	_	_	-	-	_	0	0	0
Kim et al. (2005) [16]	Dist	60	13	50	60	13	37	20	23	0
Kwon et al. (2013) [29]	Prox	100	0	0	100	0	100	0	0	0
Ounpuu et al. (2002) [17]	Prox/Dist	85	0	37	100	26	100	0	4	0
Pirpiris et al. (2003) A [3]	Prox	85	0	29	100	29	36	0	0	0
Pirpiris et al. (2003) B [3]	Dist	43	0	29	100	43	7	0	0	0
Rutz et al. (2012) [26]	Prox	45	0	100	27	9	27	0	18	0
Saraph et al. (2002) A [28]	Dist	90	c	40	100	100	100	100 ^c	40	30
Saraph et al. (2002) B [28]	Dist	100	c	30	100	100	100	100 ^c	50	50
Thompson et al. (2010) A [30]	Prox (MI)	70	10	100	100	0	90	0	10	50
Thompson et al. (2010) B [30]	Prox	100	20	20	100	50	50	0	0	50

^{- =} not reported, TDRO = tibial derotation osteotomy, MI = minimally invasive.

(maximum score: 28). All the studies were deemed to be of sufficient quality to be considered for meta-analysis according to the quality assessment (all studies scoring >70%). In general lower scores were in reference to the title and abstract, description of the participants and setting, efforts to avoid bias and description of statistical methods.

3.3. Study participants

Of the 13 included studies, 6 studies divided participants into defined groups enabling 19 separate cohorts to be entered into the meta-analysis. Four studies [22,24–26] included participants with unilateral involvement (hemiplegia), four studies [17,21,27,28] included participants with both unilateral and bilateral involvement (diplegia, triplegia) and five studies [3,15,16,29,30] included participants with bilateral involvement. Mean age of the participants ranged from 6.8 to 12.9 years and mean follow up time post surgery ranged from 0.9 to 3.1 years.

3.4. Types of intervention

Of the 13 studies, five [22,24–26,28] included participants that received a unilateral FDRO, five [15–17,21,27] included patients that received unilateral or bilateral FDRO and three [3,29,30] studies included participants that received bilateral FDRO. Information regarding the indication for FDRO was varied across studies with some studies providing quantifiable thresholds for FDRO indication [3,15,16,25,27], some studies reporting measures (without indication of thresholds) that were used to guide FDRO indication [26,28–30] and some not reporting indications for FDRO at all [17,21,22,24]. In general the indication for FDRO was increased internal hip rotation range and decreased external hip rotation range during physical exam, increased internal hip rotation on gait analysis and increased femoral anteversion on physical exam or MRI/CT scan. Additional concomitant surgical procedures performed at time of FDRO are reported in Table 3.

3.5. Quantitative analysis

All of the 13 studies retrieved and tabulated had adequate data for meta-analysis on the effect of femoral derotation osteotomy on hip rotation during gait (Figs. 2 and 3). Only 9 of the 13 studies retrieved had adequate data for meta-analysis on the effect of femoral derotation osteotomy on pelvic rotation during gait (Figs. 2 and 3).

3.6. Outcome measures

Of the 13 studies, eight [16,22,24–28,30] reported transverse plane kinematic data across the entire gait cycle and five [3,15,17,21,29] reported data confined to the stance phase. In the meta-analysis data were combined. Meta-analysis showed that femoral de-rotation osteotomy significantly reduced pelvic retraction by 9.0 degrees and hip internal rotation by 17.6 degrees in participants with unilateral CP involvement and hip internal rotation by 14.3 degrees in participants with bilateral CP involvement (Table 4).

4. Discussion

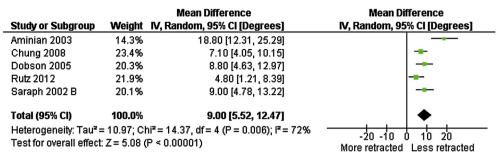
Internal hip rotation in children with CP is a common cause of an intoed gait. The mechanism of IHR is multifaceted and includes a combination of dynamic (i.e. spasticity, abnormal tone, and contracture) and static (i.e. femoral anteversion) contributions. Three-dimensional gait analysis in combination with a clinical examination can inform whether surgical intervention is indicated, and if so, which sources of dynamic or static contributions to the IHR need to be addressed. The purpose of this systematic review and meta-analysis was to determine the effect of FDRO on hip and pelvic transverse plane kinematics during gait in children with unilateral and bilateral spastic CP. Five prospective and eight retrospective cohort studies met the a-priori inclusion criteria. Data extraction revealed a heterogeneous population within and

^a Calf lengthening refers to either gastrocnemius recession or lengthening of tendo Achillis.

b Other refers to either lengthening of flexor hallucis longus, lengthening of flexor digitorum, abductor hallucis release, plantar fasciotomy, fibular brevis lengthening, lateral column lengthening.

^c Tibialis posterior transfers and lengthening grouped together for this study.







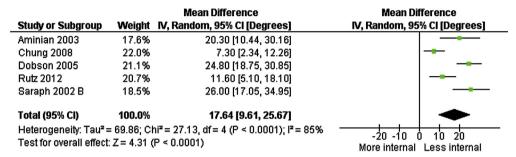


Fig. 2. Meta-analysis of the effect of femoral derotation osteotomy on (A) transverse plane pelvis kinematics and (B) hip rotation kinematics, in children with unilateral spastic cerebral palsy.

between reported studies, suggesting caution should be taken when interpreting the results of any of the studies in isolation. Overall, the results indicated that FDRO improved hip rotation profiles in both unilateral and bilateral involved patients and pelvic rotation profiles in unilaterally involved patients only.

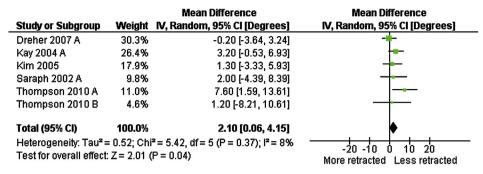
4.1. Effect of FDRO on hip kinematics

In agreement with our first hypothesis, FDRO significantly improved hip rotation during gait in children with unilateral and bilateral CP involvement. Given anteversion in children with CP is unlikely to decrease with age [22,31-33], and the response to conservative management is poor [27], we interpret these results to indicate that FDRO is an appropriate treatment option to correct IHR in children with spastic CP. Data extraction revealed that the magnitude of rotational correction during surgery was consistently greater than the observed kinematic changes observed during gait at follow up [15]. Factors that may account for this difference include persistence of dynamic contributions to IHR or differences in surgical versus gait analysis measurement of IHR. If dynamic contributions to IHR, which may include spasticity of the internal hip rotators, hip abductor weakness or excessive knee flexion (which increases the moment arm(s) of the internal rotators of the hip) are not identified and treated at time of FDRO or during postoperative rehabilitation, their contribution to IHR may persist [7,34]. Conventional three-dimensional gait analysis does not allow measurement of changes in hip rotation proximal to the osteotomy site, rather hip rotation is calculated about the long axis of the femur as defined by a line connecting the hip joint centre and the knee joint centre. The resulting rotation is the angle between the sagittal orientation of the femur (projected into a plane perpendicular to the long axis of the femur) and the knee joint axis, hence hip rotation during gait is determined by the orientation of the knee flexion axis, which is distal to the femoral osteotomy site [24]. Furthermore, it is prudent to comment that conventional three-dimensional gait analysis is sensitive to imprecise marker placement and joint centre location and as a result, hip rotation measurement error between clinicians and between laboratories can be significant [35]. Other factors that that could account for differences in the magnitude of IHR between documented surgical correction and gait analysis findings might include overestimation of the amount of surgical correction achieved at the time of surgery or recurrence of anteversion during the follow up period. Finally, it is quite possible that differences in the magnitude of IHR between documented surgical correction and gait analysis findings may be due to the amount of compensatory internal rotation adopted by patients with increased femoral anterversion prior to surgery. In many cases compensation may not be entirely necessary to overcome the lever arm deficiency of the hip abductors because some patients have enough strength, selective control and or balance to overcome the reduced hip abduction moment arm induced by the increased femoral anterversion.

There were differences between studies in the choice of FDRO technique. Some studies reported on the outcomes of proximal FDRO, some on the outcomes of distal FDRO and some reported on both. Proximal FDRO's are preferred by some surgeons as they have the potential biomechanical advantage of being closer to the deformity, whereas other surgeon's prefer the distal approach as it is simpler and the use of tourniquet can reduce blood loss [36,37]. One study that included both techniques reported similar kinematic outcomes in IHR for patients with CP [3].

Long term follow up results were presented in a number of studies and indicated a higher IHR recurrence rate when surgery was performed before the age of 10 [16,20]. Nonetheless, Ounpuu et al. [17], who followed up participants with a mean age of 8.2 years at time of surgery, reported no IHR recurrences at 1 and 5 year follow up intervals. Furthermore, Rutz et al. [26] cautioned against delaying surgery for patients with evidence of acetabular dysplasia as surgery at a younger age (i.e. before 12) may encourage more normal acetabular development and enhance opportunities for pelvic osteotomy before skeletal maturity. Both of these factors need to be considered during intervention planning.





(B)

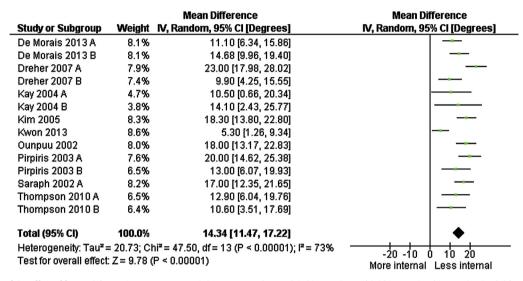


Fig. 3. Meta-analysis of the effect of femoral derotation osteotomy on (A) transverse plane pelvis kinematics and (B) hip rotation kinematics, in children with bilateral spastic cerebral palsy.

Table 4Meta-analysis results describing the effect of femoral de-rotation surgery on hip and pelvis rotation kinematics in unilateral and bilateral involved cerebral palsy patients (CI = confidence interval).

Kinematic variable	Participants included in model	Mean angle difference (deg)	95% CI	P	Hetrogeneity (I^2)
Unilateral CP involvement					
Reduction in hip internal rotation Reduction in pelvic retraction Bilateral CP involvement	85 85	17.6 9.0	9.6–25.7 5.5–12.5	<0.01 <0.01	85% 72%
Reduction in hip internal rotation Reduction in pelvic retraction	365 140	14.3 2.1	11.5–17.2 0.1–4.2	<0.01 0.37	72% 8%

4.2. Effect of FDRO on pelvic retraction

In partial agreement with our second hypothesis, FDRO significantly improved pelvic kinematics in children with unilateral involvement, but not in children with bilateral involvement. Meta-analysis results of the 5 included studies that assessed unilaterally involved patients provide strong evidence for the correction of pelvic retraction following FDRO, and suggest that preoperative pelvic rotation is most likely a compensation to maintain the knee flexion-extension axis close to the line of gait progression and/or a more typical foot progression angle. In contrast, meta-analysis results for bilaterally involved participants

showed little evidence of pelvic rotation improvement following FDRO. The meta-analysis model result for bilaterally involved patients suggests that (1) preoperative pelvic rotation is not a compensation for IHR in bilaterally involved patients, but rather a primary deformity caused by pelvic obliquity, spinal deformities, altered neurological input and/or local muscle imbalance [22,26] or (2) that bilaterally involved patients do not have the ability to change the dynamic contribution to pelvic rotation after FDRO due to impaired motor control on both sides. Nonetheless, there was disagreement across the 5 included studies in the meta-analysis for the bilaterally involved patients. Some studies reported a significant improvement [21,30] while others did not (see Fig. 2a).

To understand the differences in findings between these studies a number of methodological considerations need to be taken into account. First, the major contributing factor that determined the amount of pelvic rotation change for both unilateral and bilaterally involved patients was the preoperative magnitude of pelvic rotation [38], with patients that had less asymmetry preoperatively being less likely to experience change. It follows that most of the included studies assessing bilaterally involved patients had more symmetrical pelvic rotation profiles preoperatively. Furthermore, most of these studies included participants that underwent bilateral FDRO, in which case the presence of preoperative compensation would be impacted by the magnitude of the intervention on both sides, not just the most affected side. Another consideration in interpreting the results is that Kim et al. [16] and Thompson et al. [30] included participants that underwent bilateral FDRO, and included the rotation of the pelvic for both sides into their analysis. The problem with this approach is the external rotation of one hemipelvis would cancel the internal rotation of the other side in the overall model [15,38]. In light of these methodological issues, a major consideration for intervention planning in bilaterally involved patients is that the magnitude of transverse plane pelvic change following FDRO is dependent on preoperative asymmetry and concurrent contralateral intervention. In support of the findings of Kay et al. [21] who found reduced pelvic asymmetry post FDRO in asymmetrical spastic diplegic patients our clinical laboratory results have revealed a number of asymmetrical spastic diplegic patients who showed marked correction of pelvic asymmetry following unilateral FDRO [39].

4.3. Confounding variables and conflicts between papers

Data extraction revealed a heterogeneous population within and between reported studies. Some studies included both unilaterally and bilaterally involved patients and patients that had unilateral or bilateral FDRO, which would have contaminated the overall kinematic results. Furthermore, within and between studies there were differences in concomitant soft tissue and bony procedures at the time of FDRO, which may also have confounded the kinematic results. A number of these additional procedures were aimed at addressing the foot progression angle in conjunction with the FDRO, and secondary analysis of our results confirmed that the foot progression angle was improved following surgical intervention in the included studies that additionally presented foot progression angle as an outcome measure (see Supplementary fig. S1). Although muscle-tendon lengthening procedures are primarily aimed at addressing fixed deformities and gait abnormalities in the sagittal plane [25], there is speculation that spasticity of the gluteus medius [40], gluteus minimus, medial hamstrings [6,11], and adductors may contribute to an internally rotated femur during gait, suggesting that lengthening procedures may have a secondary (positive) affect on dynamic hip rotation during gait. However, simulated musculoskeletal models have indicated that the medial hamstrings, adductors and gracilis have negligible rotational moment arms in children with CP who walk with a crouched, internally rotated gait, indicating that lengthening would not provide any change to hip rotation during gait [41]. Not withstanding evidence from simulated models, soft tissue surgery in the absence of FDRO has been shown to have a modest (positive) effect on pelvic and hip rotation profiles during gait [21,27,38] and therefore, should be considered as appropriate in conjunction with FDRO. Furthermore, in unilaterally involved participants there is evidence to suggest that calf lengthening procedures, which occurred in more than half the participants in each study, has a significant impact on pelvic and hip kinematics in the transverse plane [42]. This is an important consideration for the treatment of hemiplegic patients that undergo FDRO as persistent equinus has been shown to contribute to IHR recurrence [42]. In addition to concomitant soft tissue procedures the majority of studies also included patients who underwent concomitant bony procedures at the time of FDRO. Concomitant bony procedures were presumably necessary as one of the major considerations in performing rotational bony surgery is the foot progression angle, which is slightly external in typically developing children. Although internal foot progression can be a result of rotation at the pelvis and hip, it may also be caused by knee rotation, tibial torsion, hindfoot-tibia rotation and/or forefoot-hindfoot adduction [21]. With the exception of a few studies [2,3,21,24,29], who specifically excluded tibial and foot procedures, the studies in this review included various bony procedures that may have contaminated the transverse plane kinematic results.

4.4. Potential limitations

There are a number of considerations that should be taken into account when interpreting the results of the meta-analysis: (1) only papers written in English were included, (2) of the 13 included papers 8 were retrospective, 5 were prospective (0 RCTs), (3) the participant pool was heterogeneous within and between studies due to different selection criteria for FDRO, variability in the duration of follow up and lack of information of patients lost to follow up and varied efforts to control bias, (4) reporting of hip and pelvis rotation was across the gait cycle in some studies and confined to the stance phase in other studies and, (5) many articles did not clearly document indication for FDRO raising a concern that selection bias may exist between studies. Finally, although the prepost design of the included studies precluded an adequate control group the evidence that anteversion in children with CP is unlikely to decrease with age over such a short period [22,31–33] supports the inclusion of the pre-surgery data a quasi-control group in the meta-analysis.

4.5. Consideration for future prospective assessments

Evidence for the effect of muscle-tendon lengthening on transverse plane rotation is still contentious. Ethically, it's not viable to assess the effect of soft tissue surgery in isolation when there is indication for bony re-alignment, or when soft tissue procedures are concomitantly required to address sagittal plane deformities. Advances in musculoskeletal-modelling that incorporate torsional deformity [41,43] and are informed by muscle activity [44] in generating a forward dynamic simulation may provide an avenue for further insight. Such models may also assist in determining the optimal amount of required derotation when performing FDRO [22]. Future prospective studies assessing the effect of FDRO on pelvic and hip transverse plane kinematics should: (1) include only the side with more external pelvic rotation in their analysis [21,38], (2) subdivide bilaterally affected participants into symmetrical and asymmetrical groups based on pelvic asymmetry, (3) exclude participants that undergo tibial and foot procedures [3,21,22,24], (4) document the amount of rotation performed during surgery [15], (5) document the amount of rotation performed on the contralateral femur if the participant underwent bilateral FDRO, and (6) limit the calculation of mean transverse plane kinematic measures to the single support subphase of the gait cycle.

5. Conclusion

The results of this systematic review confirm that pelvic asymmetry in children with unilateral spastic CP can be significantly improved by FDRO, although there is ambiguity

regarding the optimal degree of rotation required during surgery and the impact of concomitant soft tissue lengthening procedures. Meta-analysis findings suggested that patients with bilateral involvement do not improve their pelvic rotation profiles during gait as a result to FDRO, although this result should be interpreted with caution due to the methodological considerations of included studies.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at http://dx.doi.org/10.1016/j.gaitpost. 2014.05.066.

Conflict of interest

The authors declare that there are no conflicting interests.

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