Determinants of hip pain in adult patients with severe cerebral palsy

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The purpose of our investigation was to study the relationship between radiographic results of the femoral head and pain in people with severe cerebral palsy. We conducted a cross-sectional study on hip radiography results and pain in 160 patients with severe cerebral palsy. Eighteen percent of our patients had hip pain in hip-loading situations. Migration and deformity were closely related. There was a significant association with hip pain (odds ratio, 2.79; 95% confidence interval 1.01–7.70). There is a high prevalence of hip pain after unsuccessful femoral bone surgery. Migration and deformity of the femoral head are strongly interrelated, and are associated with pain. *J Pediatr Orthop B* 14:120–125 © 2005 Lippincott Williams & Wilkins.

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Introduction

In severe cerebral palsy (CP), the femoral head tends to migrate from the acetabulum, and approximately 60% of the hips of non-ambulant CP patients will subluxate [1]. According to the method described by Reimers [2], subluxation is defined as a migration of 30% or more [3]. One in six of these hips will progress to dislocation (migration of 100%) [3]. Above a migration of 60%, dislocation will follow, unless prevented by surgery [1,4,5]. Finally, prevalence of dislocation in non-ambulant patients is about 50–60% [6,7]. Migration can be visualized on a radiograph of the pelvis from the age of 18 months [8]; dislocation occurs at the age of 5–7 years [9–12].

Subluxation and dislocation of the hip can be treated by conservative and operative procedures with the purpose of preventing severe problems in adulthood, such as pain, difficulties with care, seating problems, decubital ulcers and fractures [13]. Pain in patients with CP is a frequent phenomenon, also in the hip region: in a sample of CP patients, 39% reported chronic pain in the hip region with a mean duration of 20 years [14]. However, there is still debate in the literature as to whether hip disorders are the cause of the pain. This leads to differences in opinion concerning the necessity of (preventive) surgery [15–17]. Some authors state that pain is related to migration or dislocation in itself [18]. Others consider osteoarthritis to be an additional factor [3], while pain is also explained by deformity of the femoral head [19,20]. Furthermore, although deformity of the femoral head is related to migration, it also occurs in non-migrated hips [21].

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Unfortunately, the methods used to assess pain in relation to migration are sometimes not clearly or accurately defined in the literature. Moreover, no author has yet used a method which enables CP patients to assess their own pain. The pain experienced by these patients, who are often mentally handicapped and are frequently verbally impaired, is difficult to assess. In the literature, pain is often assessed by proxy, which makes reports of the relationship between migration and pain difficult to interpret [1,10,22,23].

The aim of this study was to investigate the relationship between radiographic results for the femoral head (migration, deformity, osteoarthritis) and pain in a group of patients with severe CP. Use was made of an instrument that was specifically designed for the self-assessment of pain in this group of patients: a CP Pain Assessment Instrument (PAICP), based on the Faces Pain Scale (FPS) developed by Bieri *et al.* [24]. This instrument had been validated in an earlier study in healthy children and adults with CP [25].

Materials and methods

Patients

A cross-sectional study was conducted to investigate the relationship between radiographic results (migration, deformity and osteoarthritis) of the femoral head and pain in a group of 160 patients with severe tetraspastic CP. The study was approved by the Medical Ethics Committee of the VU University Medical Center. The study population consisted of patients with CP, who were not able to walk independently with or without walking

aids, with a minimum mental age of 4 years, and able to use an FPS. The ability to use the PAICP (vision, ability to recognize and select) and the mental level of the patients was assessed in advance with the Columbia Mental Maturity Scale (CMMS) [26], a non-verbal mental development test which has been validated for use with adults and young children with CP [27,28]. To demonstrate ability to use the PAICP, a minimum score of 25 points is needed, which indicates a minimum mental age of 4 years. Patients were recruited in homes for the severely handicapped and in rehabilitation centers in The Netherlands. Informed consent was obtained from the patients themselves or from their representatives.

Methods

The PAICP contains 21 drawings of daily occurring situations. It includes six situations that are usually not painful ($P-$). They result in a mean score of 1.4 or under. Six drawings represent situations that are usually painful ($P+$), resulting in a mean score of 4.0 or above. There are four drawings introducing the method. It also contains five hip loading situations (H) possibly painful for those with hip problems. The latter category consists of: putting on trousers, being lifted from bed, lying in bed, physiotherapy of the legs and sitting in a wheelchair. The drawings were shown in random order (available on request from the first author), and the patient assessed the amount of pain experienced in the situations represented by the drawings with a score ranging from one to seven (one, no pain; seven, severe pain) and located the pain on a picture of the human body (male or female, according to the sex of the patient).

Because the perception of pain is a subjective matter, a method was sought with which to relate the potentially hip-related pain to situations that definitely cause pain and those that do not. Therefore, the scores for the drawings representing hip-loading situations were related to the scores for the other groups of questions, usually non-painful and painful. Hip pain was mathematically related to the scores for the drawings of usually painful situations ($P+$) and the drawings of usually non-painful situations ($P-$).

Patients were considered to suffer hip pain if the result of the formula $(H-(P-))/((P+)-(P-))$ was 0.33 or greater, and H was greater than $P-$, where H is the median of answers concerning situations possibly painful for people with hip pain; $P+$ is the median of usually painful situations; $P-$ is the median of usually not painful situations. $P-$ has a minimal value of 1; $P+$ has a maximum of 7. Therefore, $P+ - P-$ has a maximal value of 6. The difference between H and $P-$ must be at least 2 in order to consider the amount of pain as positive.

Scores of 0.33 and above as outcome of the formula represent patients that experience pain in daily returning situations such as putting on trousers, being lifted from bed, lying in bed, receiving physiotherapy for legs and sitting. In this way, the patients were divided into two groups: those with hip pain and those with no hip pain. In the subsequent analyses, pain was considered as a dichotomous variable. Patients were interviewed and files were checked about surgery of the hip in the past and its indication.

An anterior–posterior radiograph of the pelvis was then made, unless there was already a radiograph available that was not made more than 5 years before.

The migration percentage (MP) [2] was measured on an anterior–posterior photograph. This widely used method is known to be reliable in assessing displacement of the femoral head from the acetabulum [8,29]. The asymmetry of migration was assessed by calculating the difference between the migration percentages on each side. It was divided into four groups (Table 2). Femoral head deformity was scored on a three-point scale, with the categories absent, moderate and severe.

Osteoarthritis was graded according to the Tönnis classification [30]. Grade 1 osteoarthritis is increased sclerosis of the femoral head and acetabulum, a slight decrease in the height of the cartilage and slight osteophytes. Grade 2 refers to small cysts in the femoral head or acetabulum, a marked decrease in the height of cartilage and slight deviation from the round form of the femoral head. Grade 3 represents large cysts in the femoral head and acetabulum, a severe decrease in cartilage up to complete absence of the joint cleft, severe deviation of the round form of the femoral head and avascular necrosis.

One author (M.J.) conducted all the interviews, and another author (E.B.) scored all the radiographs, without any knowledge of patient characteristics. Logistic regression analysis using SPSS version 10.0 (SPSS, Chicago, Illinois, USA) was performed with pain (yes or no) as dependent variables and migration and its difference, deformity and osteoarthritis of the femoral head as independent variables. Surgery was considered to be successful when the result was in concordance with the primary indication and no adverse side-effects, such as pain, luxation or subluxation, occurred.

Results

Patients

The age of the patients varied between 16 and 84 years, with a mean of 36 years. Of the 160 patients, 87 (54%) were male and 73 (46%) were female; 19 (12%) were not

able to speak, and 11 (7%) of them used a scanning device for communication.

Prevalence of pain

Thirty patients (18%) had hip pain in hip-loading situations according to the study criteria. Pain occurred more frequently in women (12 of 63) than in men (six of 77), but there was no differentiation in age.

Radiography results

The radiographs were subdivided, on the basis of the maximal migration percentage, into four groups, according to the standard method [7]. In 20 cases the radiograph showed an abnormal configuration of femur and pelvis, in which the migration percentage could not be calculated. On those radiographs the femoral head was either absent, obviously totally disintegrated, or impossible to identify. Direct aberrant contact between parts of the femur and the pelvis was also classified as an abnormal configuration. It was decided to consider these patients as a separate fifth group (group V) (Table 1). The remaining 140 patients (groups I–IV) had a migration of less than 100% or a dislocation with a femoral head that was clearly recognizable (group I, MP < 30%; group II, MP 30–59%; group III, MP 60–89%; group IV, MP > 90%).

Relationship between radiographic results and pain

In cross-tab analysis of the five sub-groups, the relationship between pain and the situation of the femur head was significant (Spearman's ρ 0.367; $P = 0.000$) (Table 1). No relationship was found between pain and sex.

Groups I–IV (migration less than 100% or dislocation)

In the 140 patients in groups I–IV, deformity proved to be a concomitant factor for pain (Table 2). Migration and deformity of the femur head were significantly interrelated and combined for univariate logistic regression analysis ($r_s = 0.46$; $P < 0.01$). Also, pain increased with the quantity of asymmetry and osteoarthritis, respectively (Table 3). Migration percentage and difference in migration put together were also closely related ($r_s = 0.64$; $P < 0.01$). When arranged in one table, pain showed to be a factor dependent on multiple factors (Table 4).

Table 1 Determinants of pain: relationship between situation of the femoral head and pain

Group	Migration of femoral head	*n*	With pain
I	<30%	89	8 (9%)
II	30–59%	35	5 (14%)
III	60–89%	9	2 (22%)
IV	>90%	7	3 (42%)
V	Abnormal configuration	20	12 (60%)
Total		160	30 (19%)

Spearman's ρ=0.37; $P<0.01$.

Table 2 Determinants of pain: relationship between deformity, migration and pain

Deformity	Migration	Pain		Total
		No	Yes	
None	<30%	29	3	32
	30–59%	7	1	8
	90–100%	1	0	1
Total		37	4 (9.8%)	41
Moderate	<30%	43	3	46
	30–59%	8	1	9
	60–89%	3	0	3
Total		54	4 (6.9%)	58
Severe	<30%	9	2	11
	30–59%	15	3	18
	60–89%	4	2	6
	90–100%	3	3	6
Total		31	10 (24.4%)	41

Data for groups I–IV; $n=140$. Spearman's ρ=0.46; $P<0.01$.

Table 3 Determinants of pain: relationship between asymmetry, osteoarthritis and pain

	n	With pain
Asymmetry of femoral head, groups I–IV		
None (difference 0–10)	79	7 (9%)
Mild (difference 11–30)	35	4 (11%)
Moderate (difference 31–60)	17	3 (18%)
Severe (difference >60)	9	4 (44%)
Total	140	18
Osteoarthritis of femoral head, groups I–IV		
None	57	6 (10%)
Mild	60	8 (13%)
Moderate	11	1 (9%)
Severe	12	3 (25%)
Total	140	18

Table 4 Relationship between osteoarthritis, migration, deformity and pain; groups I–IV

Osteoarthritis	Migration	Deformity	Pain	
			No	Yes
None	<30%	None	19	1
		Moderate	17	1
		Severe	2	1
	30–59%	None	4	1
		Moderate	1	1
		Severe	1	1
	60–89%	Moderate	2	
		Severe	3	
	90–100%	None	1	
		Severe	1	
Mild	<30%	None	10	2
		Moderate	23	2
		Severe	4	1
	30–59%	None	2	
		Moderate	7	
		Severe	6	
	60–89%	Severe		1
	90–100%	Severe		2
Moderate	<30%	Moderate	2	
		Severe	2	
	30–59%	None	1	0
		Severe	3	1
	90–100%	Severe	2	
Severe	<30%	Moderate	1	
		Severe	1	
	30–59%	Severe	5	1
	60–89%	Moderate	1	0
		Severe	1	1
	90–100%	Severe		1
Total			122	18

In univariate logistic regression analysis of groups I–IV, the migration and deformity factors were combined into two groups, because of their close inter-relationship: group 1 with migration less than 33% and no or moderate deformity, the second with migration > 33% or severe deformity. The analysis showed a significant relationship ($P < 0.05$) between the combined factor migration over 33% or severe deformity and pain, with an odds ratio of 2.79. Also asymmetry of migration showed a significant relationship with pain (odds ratio 8.23), indicating an increased relative risk. Osteoarthritis was a factor that was not significantly related to pain (Table 5). If adjusted for the other factors, the odds ratios remained positive but not statistically significant, which illustrates their close interdependence.

Group V (abnormal configuration of the femoral head and pelvis)

This abnormal configuration was strongly associated with pain: 12 of the 20 patients (still) suffered from hip pain. In this subgroup only patients with ankylosis of femur and pelvis and a previous resection of the femoral head at the level of the minor trochanter currently had less than 100% pain (Table 6).

Results of surgery

Of the 160 patients 132 had surgery for three major indications: pain, reduction of the femur head and others/unknown. The results of this surgery are shown in Figure 1. Soft tissue surgery was most successful with reduction as indication (80%); result for indication pain was poor (54%). Pain was best cured by derotation varus osteotomy (71%; Fig. 1).

Table 5 Crude and adjusted odds ratios (OR) and 95% confidence intervals for determinants of hip pain

Migration/deformity	Crude OR	Adjusted OR[a]
Migration <33% and no/ moderate deformity	1.00 (reference)	1.00 (reference)
Migration ≥ 33% and/or severe deformity	2.79 (1.01–7.70)*	1.70 (0.39–7.34)
Osteoarthritis		
None	1.00 (reference)[b]	1.00 (reference)[b]
Mild	1.31 (0.42–4.04)	1.29 (0.40–4.15)
Moderate	0.85 (0.09–7.85)	0.54 (0.05–5.62)
Severe	2.83 (0.60–13.44)	1.60 (0.28–9.12)
Asymmetry of migration		
None	1.00 (reference)[c]	1.00 (reference)[b]
Mild	1.33 (0.36–4.86)	1.19 (0.30–4.81)
Moderate	2.20 (0.51–9.57)	1.45 (0.23–9.03)
Severe	8.23 (1.79–37.87)*	5.43 (0.82–35.8)

Data for groups I–IV; $n = 140$. [a]Adjusted for the other variables in the table. [b]P for trend not significant. [c]P for trend 0.011. *$P < 0.05$.

Discussion

The overall prevalence of hip pain in patients with severe CP found in the present study (18%) is in agreement with the prevalence of pain in patients with CP found in other studies [31,32], but less than reported by Hodgkinson *et al.* [22], who found a percentage of 47%. However, their interviews were partly held with care-givers and not with the patients themselves, as took place in the present study. Furthermore, they used a pain scale that had not been validated for this group of patients. The prevalence of osteoarthritis found in the present study was also in concordance with findings reported in the literature [33].

A relationship between deformity and pain had already been found [19]. The results of the present study show that there is an association between migration of the femoral head, its deformity and pain. However, the design of the study did not make it possible to assess the temporal relationship between these factors and pain.

Migration and deformity are closely related, as was found in the analysis, and also in other studies [21,23,33], but they do not seem to be completely interdependent. Although deformity generally increases with migration, it is not the result of migration alone. Eleven out of 89 patients with a migration percentage of less than 30% already had severe deformity of the femoral head (Table 3). At least one of the patients without osteoarthritis, migration and deformity suffered from pain; on the other hand there was a patient with severe osteoarthritis and deformity and subluxation without pain (Table 4). Also the relationship we found between asymmetry of the migration percentage and pain confirms that in people with CP the pain apparently is a multifactorial phenomenon that cannot be explained by migration, deformity or asymmetry alone.

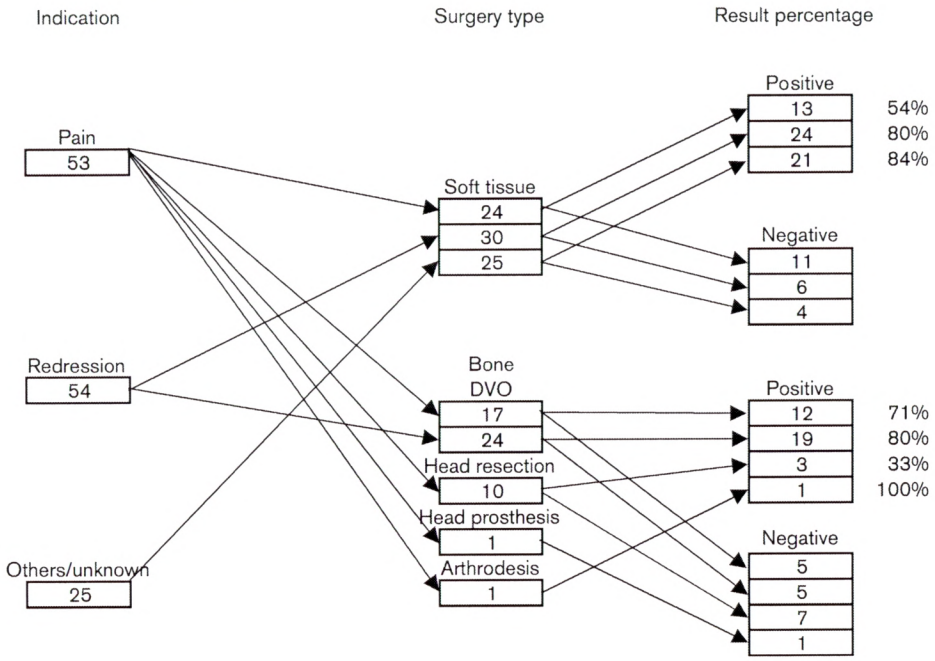
Several patients in this study had undergone surgery to reduce the femoral head or to reduce pain. A number of these patients were in groups I–IV and their surgery had been successful (Fig. 1). However, a number of operations had obviously not been successful: 20 of 160

Table 6 Patients with abnormal configuration of femoral head and pelvis

Situation	Number	History of bone surgery	Pain as indication for bone surgery	Residual hip pain
After proximal resection of the femoral head (sub-capital)	5	5	4	4
After distal resection of the femoral head (level minor trochanter)	3	3	2	1
Ankylosis of femur and pelvis	4	4	2	2
Aberrant contact between (rest of) femur and pelvis	8	5	5	5
Total	20	17	13	12

Data for group V; $n = 20$.

Fig. 1



Indication, type of surgery and result. DVO, derotation varus osteotomy.

patients had an abnormal configuration of femur and pelvis, mostly after bone surgery, which was associated with (residual) pain. Only distal resection of the femoral head at the level of the minor trochanter was found to have no association with pain (Table 6). This in concordance with the findings of Cooperman *et al.* [19]. The results of the present study suggest that deformity, asymmetry and migration of the femoral head should ideally be prevented or treated, but the question that rises is how and at what price? Soft tissue or bone surgery to prevent or correct migration can be quite successful [3,10], but surgery has a great impact on these already very disabled patients, and if soft tissue surgery fails the decision to perform bone surgery should be even more carefully considered. Surgery in itself does not cure asymmetry [34], and may even aggravate it when performed unilaterally [35]. Furthermore, unsuccessful bone surgery creates a high risk for residual, severe pain. In the literature, the distal resection of the femoral head at the level of the minor trochanter is reported to be a successful salvage treatment in curing pain when other measures fail [19,32]. Our findings support that view.

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