

Pain and rehabilitation problems after single-event multilevel surgery including bony foot surgery in cerebral palsy

A series of 7 children

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Background and purpose — Surgical correction of foot deformities as part of single-event multilevel surgery (SEMLS) to optimize postoperative training is sometimes indicated in ambulatory children with cerebral palsy. We have, however, experienced excessive postoperative pain and rehabilitation problems in a number of these patients. We therefore investigated children who underwent such procedures regarding postoperative rehabilitation and pain, gait parameters 1 year after surgery, and mobility 5 years after surgery.

Patients and methods — 9 children with diplegic cerebral palsy who had also undergone bony foot surgery were identified from a cohort of 70 children treated with SEMLS according to a standardized protocol. 2 children were excluded due to mental retardation and atypical surgery, and 7 patients (4 of them boys) were included. The children and their parents underwent a semi-structured interview on average 5 (3–7) years after the surgery. Gait parameters preoperatively and 1 year postoperatively were compared.

Results — 5 children had experienced regional pain syndrome and considerable sociopsychological problems during the first postoperative year. 5 years after surgery, 4 of the 5 children still had hypersensitive and painful feet, 2 had lost their ability to walk, 1 child was no longer self-reliant in daily care, and 3 were wheelchair bound. There were, however, no clinically significant differences in functional mobility scale (FMS) or gait parameters preoperatively and 1 year postoperatively.

Interpretation — We found troublesome postoperative rehabilitation and poor outcomes in this series of children who had undergone simultaneous multilevel surgeries and bony foot corrections. Caution is warranted when treating marginally ambulatory children with bilateral spastic cerebral palsy and foot deformities.

Foot deformities are frequently seen in ambulatory children with moderate to severe cerebral palsy. Surgical correction of foot deformities is sometimes indicated in order to improve gait or to enable the child to use orthoses to optimize postoperative training after single-event multilevel surgery (SEMLS). According to Gage et al. (2009) all corrective surgeries, including foot corrections, can be performed simultaneously in a “single-event multilevel surgery to improve gait”, and this concept is now widely accepted and used (Schwartz et al. 2004). Computer-based movement analysis is used for preoperative diagnostic evaluation and outcome assessment (Gough et al. 2004, Lofterød et al. 2007).

In our clinic, we use a standardized multilevel protocol in treating ambulant children with cerebral palsy, including pre- and postoperative 3D gait analysis (Vicon Motion Systems) and an extensive and standardized protocol for postoperative rehabilitation (Lofterød et al. 2007, Lofterød and Terjesen 2008).

We have experienced excessive postoperative pain and rehabilitation problems in a number of our diplegic SEMLS patients who had simultaneous bony foot surgery. We therefore investigated how children who had undergone simultaneous multilevel soft tissue and corrective bony foot surgeries performed regarding postoperative rehabilitation and pain, gait parameters 1 year after surgery, and mobility 5 years after surgery.

Patients and methods

9 ambulant children with diplegic cerebral palsy—all classified as GMFCS III—who had undergone bony foot surgery as part of SEMLS treatment were identified from a cohort of 70

Table 1. Description of the cohort of 70 SEMLS patients from which the patients were selected

Diagnoses	
Unilateral cerebral palsy	4
Bilateral cerebral palsy	64
Other diagnoses	2
GMFCS level	
Level 1	4
Level 2	39
Level 3	27
Surgical procedures, total = 399	
Psoas tenotomies	73
Adductor tenotomies	39
Rectus femoris transfers	91
Hamstrings tenotomies	94
Gastrocnemius slides	40
Achilles lengthenings	15
Various surgeries	6
Proximal femoral osteotomies	23
Dega acetabuloplasties	2
Bony foot surgeries	15

children treated with SEMLS during the 5-year period January 2005 to January 2010 (Table 1). 1 severely mentally retarded girl and 1 boy who was considered not to be truly SEMLS (as

he had had atypical surgeries: foot corrections and bilateral simple tenotomies of rectus femoris) were excluded. Thus, 7 children were selected for inclusion. None of the children had experienced any significant foot pain preoperatively.

The patients were followed up 5 (3–7) years postoperatively with a semi-structured interview. The postoperative 1-year follow-up gait analysis was compared to the preoperative one.

All the children had an extensive number of soft tissue surgeries done, ranging in number from 5 to 8, in addition to the bony foot surgeries. All, except for 1 girl, had surgical treatment before the SEMLS. Thus, gait patterns could not be determined and evaluated further. However, preoperatively all patients had significantly increased flexion in their hips and knees (> 2 SD), as typically seen in a crouch-like gait pattern.

Table 2 lists the types of surgeries performed on the patients prior to and at the SEMLS. Mean age at surgery was 13 (10–17) years. Mean time between preoperative gait analysis and surgery was 11 (6–24) months and mean time between surgery and postoperative gait analysis was 16 (13–20) months.

Single-event multilevel surgeries

All surgeries were carried out according to a standardized protocol (DeLuca et al. 1998, Gough et al. 2004, Schwartz et al. 2004, Gupta et al. 2008). All patients were given acetamino-

Table 2. Surgeries performed on the patients

Patient	Sex	Age at SEMLS	Surgeries performed prior to SEMLS	Surgeries at SEMLS ^a	Foot corrections at SEMLS ^a
1	Female	10	None	Psoastenotomies over the brim Rectus femoris transfers to gracilis Medial hamstring lengthenings Gastrocnemius recessions	Grice extra-articular arthrodeses
2	Male	15	Medial hamstring lengthening	Ileopsoastenotomies Adductor tenotomies Rectus femoris transfers to gracilis Medial hamstring lengthenings (second time)	Triple arthrodeses
3	Female	14	Adductor tenotomies Ileopsoas tenotomies Medial hamstrings lengthenings Gastrocnemius recessions	Psoastenotomies over the brim Adductor tenotomy right side (second time) Rectus femoris transfers to gracilis Medial hamstring lengthening (second time)	Calcaneal lengthening osteotomies
4	Male	16	Tendon achilles lengthenings Medial hamstring lengthening	Psoastenotomies over the brim Rectus femoris transfers to gracilis Medial hamstring lengthenings (second time) Gastrocnemius recessions	Triple arthrodeses
5	Female	10	Proximal femoral derotational osteotomy, left side	Psoastenotomies over the brim Rectus femoris transfers to gracilis Medial and lateral hamstring lengthenings Gastrocnemius recessions	Calcaneal lengthening osteotomy, right foot
6	Male	9	Adductor tenotomies Ileopsoas tenotomies Medial hamstring lengthening	Rectus femoris transfers to gracilis Medial hamstring lengthening, right side Gastrocnemius recessions	Calcaneal lengthening osteotomies
7	Male	10	Medial hamstring lengthening Tendon achilles lengthening	Psoastenotomies over the brim Rectus femoris transfers to gracilis Medial hamstring lengthenings	Grice extra-articular arthrodesis, right foot

^a Bilateral procedures unless noted otherwise

phen and epidural analgesic catheters with Bupivacain (1 mg/mL), Phentanyl ($\mu\text{g/mL}$), and adrenalin (2 $\mu\text{g/mL}$) for postoperative pain treatment. In addition, antiflogistics and opioids were given when needed.

Bony foot correction procedures

All the children had valgus foot deformities, and the choice of foot correction was according to the surgeon's preference. Triple arthrodeses were performed in a standardized manner after resection of the joint surfaces and correction of the deformities, if any (Ireland and Hoffer 1985). Subtalar arthrodeses were performed according to the technique of Grice-Green (Hoiness and Kirkhus 2009). Calcaneal lengthening osteotomies were performed according to Mosca (1995), except for the plication of the talonavicular joint capsule and shortening of the posterior tibial tendon. Bone grafts were harvested from the ipsilateral iliac crest. In all children, a circular cast was applied for 6–10 weeks, after which weight bearing and rehabilitation started.

Postoperative rehabilitation

Postoperative rehabilitation followed a standardized protocol. Passive physiotherapy in the form of joint mobilization was started 4–7 days after surgery. All patients had new ground reaction ankle foot orthoses (GRAFO) for postoperative training and rehabilitation. Immediately after surgery, the patients had rigid knee extension braces to maintain knee extension during resting periods.

Approximately 6 weeks after the foot corrective surgery, the plaster casts were replaced with GRAFOs during walking to provide passive knee extension in the stance phase of gait.

The GRAFOs were routinely used during walking the first postoperative year. The knee extension braces were used until muscle balance at the knee was stabilized and the patients were able to actively extend the knees fully.

Hospital stay in the rehabilitation unit ranged from 4 to 6 weeks, during which daily physiotherapy and strength- and gait training were provided.

Semi-structured interviews

Qualitative, semi-structured interviews of the 7 children and their parents were performed and based on 1 main research question: In what way did the children and their parents experience the rehabilitation process after multilevel surgery (SEMLS)?

The interviews were carried out with the parents and child together in the patient's home.

A low-structured interview guide was developed covering experience of pain, coping with training and physiotherapy after hospitalization, experience of family coping and school acceptance, social participation, and evaluation of outcome compared with the efforts during rehabilitation.

The interviews were recorded and transcribed by one of the authors (HC) for later evaluation. The transcribed data were

analyzed according to the method of qualitative research analysis described by Kvale et al. (2009).

The parents and the patient together were asked to rate the degree to which the patient's pain affected his/her daily activities on a visual analog scale (VAS) ranging from 1 to 10 where 1 signified "no effect on daily activities and mobility" and 10 signified "maximum effect on daily activities and mobility".

Gait analysis

All the children underwent pre- and postoperative clinical examination and gait analysis including sagittal and coronal plane video recording, registration of time and distance parameters, and kinematic and kinetic data. The same multidisciplinary team (a child neurologist, an orthotist, a physiotherapist, and an orthopedic surgeon) performed and assessed the pre- and postoperative gait analyses over the whole study period. A 6-camera Vicon System (612) (Oxford Metrics, Oxford, UK) and 2 AMTI force plates (Advanced Mechanical Technology, Watertown, MA) were used to collect motion analysis data. To compare kinematic and kinetic data obtained from gait analysis, a representative trial from each subject was selected for analysis. If all trials showed similar patterns based on visual evaluation of the plots, the initial trial was selected. If 2 of 3 trials were similar, the first trial of the 2 was selected (Ounpuu et al. 2002). All the children performed their gait analyses without shoes or braces. 3 children needed support from walking aids to complete their gait analyses.

Ethics

The patients and their parents gave their written consent before inclusion. The study was evaluated and accepted by the local ethics committee (2010/1592).

Results

The semi-structured interviews revealed 2 very different groups depending on how the patients and their families experienced the first postoperative year after SEMLS. 2 patients had an uneventful postoperative year with little or moderate pain, sufficient rehabilitation, and an overall satisfactory experience of SEMLS.

The remaining 5 patients experienced extensive problems, mainly due to severe pain and hypersensitivity in their feet. 1 of these patients underwent a triple arthrodesis, 2 patients had Grice arthrodeses, and 2 patients had undergone calcaneal lengthening osteotomies. All these children developed a chronic regional pain syndrome (CRPS) and required highly specialized multidisciplinary care.

2 children had pain during the cast period, while 3 had pain on removal of the cast. The pain lasted more or less a year in all cases. The situation worsened due to random medication routines throughout the postoperative year and generally poor coping with pain due to insufficient specialized pain

Table 3. Comparison of preoperative and postoperative gait parameters

	Pre-operatively mean (SD)	Post-operatively mean (SD)	p-value (t-test)
In stance (°)			
Maximum ankle dorsiflexion	15 (7)	19 (7)	0.1
Mean foot progression	-8 (17)	-12 (10)	0.4
Minimum knee flexion	37 (16)	29 (15)	0.001
Maximum hip extension	15 (9)	18 (13)	0.2
Mean femur rotation	2 (3)	3 (4)	0.4
Mean anterior pelvic tilt (°)	10 (10)	6 (5)	0.08
Cadence (strides/min)	111 (29)	95 (26)	0.4
Stride length (cm)	76 (18)	65 (24)	0.2
Velocity (cm/s)	70 (23)	53 (28)	0.1

care—both locally and in the operating hospital. During the first postoperative year these families received little support to cope with pain.

Pain at night resulted in substantial sleeping problems for the children and their parents, who struggled to keep up with their work. Chronic pain led to poor attendance at school, at training programs, and at physiotherapy, and made it difficult to have everyday contact with peers.

All 5 families characterized this year as being a “nightmare”. On a VAS (ranging from 1–10) describing how pain had affected the patient’s daily activities and mobility during the first postoperative year, 4 families gave a rating of 10 and 1 family gave a rating of 8.

5 years after surgery, 4 of the 5 children still had hypersensitive and painful feet, 2 had lost their ability to walk, 1 child was no longer self-reliant in daily care, and 3 were wheelchair bound.

4 patients regretted the surgeries. Despite having multiple postoperative problems, 1 girl did not regret the surgeries because her knees were straighter postoperatively.

All the patients had 1-year postoperative gait analysis except for 1 girl who used a wheelchair because of problems in walking postoperatively. Statistically significant changes were found in minimum knee flexion in stance (Table 3). There were no clinical differences in FMS when we compared the 1-year postoperative score with the preoperative score (Table 4).

Discussion

Postoperative pain, discomfort, and psychosocial problems are infrequently discussed and evaluated in reports on orthopedic surgical treatment of children with cerebral palsy. Scarcely any reports have focused on postoperative pain after foot corrections.

We believe that incorporation of pain parameters, quality of life, and psychosocial parameters are highly relevant in

Table 4. FMS evaluated at gait analysis preoperatively and 1 year postoperatively

	Preoperatively				1 year postoperatively			
	Min	Max	Mean	SD	Min	Max	Mean	SD
FMS preop 5 m	2	5	4.2	1.2	3	5	4.2	1
FMS preop 50 m	2	5	3.5	1.4	3	5	3.5	0.8
FMS preop 500 m	1	4	1.8	1.3	1	4	1.5	1.2

clinical studies on surgical treatment of children with cerebral palsy. To address this, it seemed useful to combine qualitative and traditional quantitative methods in this study.

The foot pain in our patients was most likely caused or initiated by the foot surgery, as none of the patients had experienced significant foot pain preoperatively. No specific foot correction was associated with postoperative pain and disability. Over-stretching of the knees during medial hamstrings lengthening tenotomy is known to cause traction injury to the sciatic nerve, which may cause long-lasting pain and dysesthesia in the feet that mimic or induce CRPS. This may have contributed to pain in this study, as all the patients had medial hamstrings tenotomies. Moreover, traction injury to the sciatic nerve may also have been inflicted by over-enthusiastic knee stretching during the postoperative period.

Several authors have highlighted that children with cerebral palsy are generally at risk of pain, especially following surgical procedures (Oberlander et al. 1999, Hadden and von Baeyer 2002). All 5 children with serious chronic pain are within the definition of chronic pain (Turk and Melzack 1992). The 4 children with persistent foot pain and dysesthesia after 5 years match the revised IASP general definition of CRPS (Harden and Bruehl 2006, Harden et al. 2007).

In a population of 406 adults with cerebral palsy, Jahnsen et al. (2004) found that nearly one-third had chronic pain, as compared to 15% in the normal population. The feet were a common location for pain in children with diplegia (54%), exceeded only by back pain (64%). Turk et al. (1997) reported a high incidence of chronic pain (84%) in women with cerebral palsy, and they also found an association between pain and mental retardation. Other investigators have commented that pain is associated with spasticity, deformity, scoliosis, joint stress, overuse, and rehabilitation procedures such as bracing (Ireland and Hoffer 1985, Kibele 1989, Broseta et al. 1990, Cassidy et al. 1994). In a long-term follow-up study of 24 individuals with cerebral palsy who had had triple arthrodeses performed as children, Tenuta et al. (1993) found 9 patients (38%) who still reported pain in their feet, which was the predominant factor related to patient satisfaction.

Cuomo et al. (2007) found that “improved functional well-being after SEMLS does not imply improved psychosocial well-being, and patients and their families should be counseled accordingly”. Our study confirms this observation.

Extensive surgery and postoperative rehabilitation can be expected to have a great impact on the patient and the patient's family in their everyday life. The negative effect upon the families during rehabilitation cannot be underestimated. The fact that parents had to stay away from work for 4–9 months to care for their sick children, had to take sleep medication, and described the first postoperative year as “terrible” and “a nightmare” underscores the serious implications for the families involved. The risk of bringing families into such situations should be considered in the preoperative decision-making process.

The moderate preoperative maximum ankle dorsiflexion in stance may have been due to the fact that the children had severe foot deformities, which make assessment of ankle movement challenging. It is, however, likely that they in fact had equinus deformities at clinical examination, as they all required gastrocnemius recessions in order to achieve satisfactory ankle dorsiflexion after correction of their valgus deformities. According to our treatment protocol, we aim for not more than 0–10° of dorsiflexion after foot corrections to enhance knee extension and to avoid crouch gait. Significant improvement in minimum knee flexion during stance was found 1 year postoperatively, indicating improved knee extension, most likely due to the medial hamstrings lengthenings and foot corrections.

Gait function and parameters may continue to change in CP children as part of the natural history, especially during the growth spurt, but Thomason et al. (2011) also found significant functional improvements after 2 years that were not found at 1 year. The gait function found at 1 year in the present study may thus have changed later both due to the natural progression and as an effect of the SEMLS performed. Mean age at surgery in this study was 13 years, and thus gait changes may not have been an effect of the SEMLS alone, but may in part be due to the fact that most of these children had not yet reached adolescence at the time of surgery.

Comparing preoperative values and values 1-year postoperatively, there was no clinically significant change in FMS, but in the longer term 2 children with excessive pain and rehabilitation problems lost their ability to walk and 1 child was no longer self-reliant in daily care.

Being a retrospective study, all the relevant data may not have been recorded and important information may have been lost. Pre- and post-treatment quality-of-life scores were lacking, and such parameters, or scores, would have been able to describe and quantify the patients' well-being and psychosocial environment to a better degree.

From the same cohort of 70 SEMLS-operated children, we identified 7 patients (5 of them boys) who were comparable with respect to age, gender, CP type, GMFCS level, FMS, and types of SEMLS surgeries, except they had not undergone any foot surgery. The selection of these control patients was performed by the staff at our gait laboratory, who did not participate in the study, thus blinding the investigators to the selection.

5 of these patients were selected for evaluation, as 1 boy was excluded due to mental retardation and 1 girl refused to participate. These patients were quite similar to the study patients in all respects except for they had not had foot corrections. They were followed up and examined in an identical fashion to the study patients, including the 1-year gait lab analysis and the semi-structured interviews.

Notably, none of these control patients had any experiences of severe pain or troublesome postoperative rehabilitation, and none of them regretted the treatment. There were no clinically significant differences in gait lab parameters compared to the foot-operated children at the 1-year gait analysis.

Our findings indicate that caution is warranted when treating marginally ambulant children with severe bilateral spastic cerebral palsy and foot deformities. A 2-stage procedure in which the foot deformities are corrected first and SEMLS is performed after healing of the foot would possibly reduce the risk of postoperative pain and rehabilitation problems.

PRH organized the study, retrieved patients for inclusion, evaluated the data, and drafted the protocol and the manuscript. HC took part in writing of the protocol, performed and evaluated the interviews, and co-wrote the manuscript. BL took part in writing of the protocol, performed the evaluation of the gait lab parameters, and co-wrote the manuscript. All the authors read and approved the final manuscript.

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No competing interests declared.

Broseta J, Garcia-March G, Sanchez-Ledesma M J, Anaya J, Silva I. Chronic intrathecal baclofen administration in severe spasticity. *Stereotact Funct Neurosurg* 1990; 54-55: 147-53.

Cassidy C, Craig C L, Perry A, Karlin L I, Goldberg M J. A reassessment of spinal stabilization in severe cerebral palsy. *J Pediatr Orthop* 1994; 14: 731-9.

Cuomo A V, Gamradt S C, Kim C O, Pirpiris M, Gates P E, McCarthy J J, Otsuka N Y. Health-related quality of life outcomes improve after multilevel surgery in ambulatory children with cerebral palsy. *J Pediatr Orthop* 2007; 27: 653-7.

DeLuca P A, Ounpuu S, Davis R B, Walsh J H. Effect of hamstring and psoas lengthening on pelvic tilt in patients with spastic diplegic cerebral palsy. *J Pediatr Orthop* 1998; 18: 712-8.

Gage J R, Schwartz M H, Koop S, Novacheck T F. The Identification and treatment of gait problems in cerebral palsy. Mac Keith Press, London 2009.

Gough M, Eve L C, Robinson R O, Shortland A P. Short-term outcome of multilevel surgical intervention in spastic diplegic cerebral palsy compared with the natural history. *Dev Med Child Neurol* 2004; 46: 91-7.

Gupta A, Srivastava A, Taly A B, Murali T. Single-stage multilevel soft-tissue surgery in the lower limbs with spastic cerebral palsy: Experience from a rehabilitation unit. *Indian J Orthop* 2008; 42: 448-53.

Hadden K L, von Baeyer C L. Pain in children with cerebral palsy: common triggers and expressive behaviors. *Pain* 2002; 99: 281-8.

- Harden R N, Bruehl S P. Diagnosis of complex regional pain syndrome: signs, symptoms, and new empirically derived diagnostic criteria. *Clin J Pain* 2006; 22: 415-9.
- Harden R N, Bruehl S, Stanton-Hicks M, Wilson P R. Proposed new diagnostic criteria for complex regional pain syndrome. *Pain Med* 2007; 8: 326-31.
- Hoiness P R, Kirkhus E. Grice arthrodesis in the treatment of valgus feet in children with myelomeningocele: a 12.8-year follow-up study. *J Child Orthop* 2009; 3: 283-90.
- Ireland M L, Hoffer M. Triple arthrodesis for children with spastic cerebral palsy. *Dev Med Child Neurol* 1985; 27: 623-7.
- Jahnsen R, Villien L, Aamodt G, Stanghelle J K, Holm I. Musculoskeletal pain in adults with cerebral palsy compared with the general population. *J Rehabil Med* 2004; 36: 78-84.
- Kibele A. Occupational therapy's role in improving the quality of life for persons with cerebral palsy. *Am J Occup Ther* 1989; 43: 371-7.
- Kvale S, Brinkmann S, Anderssen T M, Rygge J. Det kvalitative forskningsintervju. Gyldendal akademisk, Oslo 2009.
- Lofterod B, Terjesen T. Results of treatment when orthopaedic surgeons follow gait-analysis recommendations in children with CP. *Dev Med Child Neurol* 2008; 50: 503-9.
- Lofterod B, Terjesen T, Skaaret I, Huse A B, Jahnsen R. Preoperative gait analysis has a substantial effect on orthopedic decision making in children with cerebral palsy: comparison between clinical evaluation and gait analysis in 60 patients. *Acta Orthop* 2007; 78: 74-80.
- Mosca V S. Calcaneal lengthening for valgus deformity of the hindfoot. Results in children who had severe, symptomatic flatfoot and skewfoot. *J Bone Joint Surg (Am)* 1995; 77: 500-12.
- Oberlander T F, O'Donnell M E, Montgomery C J. Pain in children with significant neurological impairment. *J Dev Behav Pediatr* 1999; 20: 235-43.
- Ounpuu S, DeLuca P, Davis R, Romness M. Long-term effects of femoral derotation osteotomies: an evaluation using three-dimensional gait analysis. *J Pediatr Orthop* 2002; 22: 139-45.
- Schwartz M H, Viehweger E, Stout J, Novacheck T F, Gage J R. Comprehensive treatment of ambulatory children with cerebral palsy: an outcome assessment. *J Pediatr Orthop* 2004; 24: 45-53.
- Tenuta J, Shelton Y A, Miller F. Long-term follow-up of triple arthrodesis in patients with cerebral palsy. *J Pediatr Orthop* 1993; 13: 713-6.
- Thomason P, Baker R, Dodd K, Taylor N, Selber P, Wolfe R, Graham H K. Single-event multilevel surgery in children with spastic diplegia: a pilot randomized controlled trial. *J Bone Joint Surg (Am)* 2011; 93: 451-60.
- Turk D, Melzack R. The measurement of pain and the assessment of people experiencing pain. Guilford Press, New York, 1992.
- Turk M A, Geremski C A, Rosenbaum P F, Weber R J. The health status of women with cerebral palsy. *Arch Phys Med Rehabil* 1997; 78: S10-S17.