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ORIGINAL PAPER

Long-term results of multilevel surgery in adults with cerebral palsy

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Abstract

Background Deterioration of gait in adolescent and adult patients with cerebral palsy can be associated with multiple factors. Multilevel surgery (MLS) is one option in adults with cerebral palsy to improve gait function with encouraging short-term results. It is a question whether these improvements are maintained over time.

Methods In a retrospective consecutive cohort study, adults with bilateral spastic cerebral palsy (BSCP) treated with MLS between 1995 and 2011 were scanned for potential inclusion. Patients needed to fulfill the following inclusion criteria: age at MLS > 17, standardized three-dimensional gait analysis (3D-GA) including clinical examination at pre-operative (E0), a short-term follow-up (E1) and at least seven years (E2) after the index MLS. Twenty adults (10 women, 10 men) with a Gross Motor Function Classification Level (GMFCS) I–III and a mean age at MLS of 24.8 years were included in this study. The average long-term follow-up was 10.9 years. The Gait Profile Score (GPS) was used as primary outcome measure.

Results The GPS improved significantly from 13.8° before surgery to 11.2° at short-term ($p = 0.007$) and to 11.3° at long-term follow-up ($p = 0.002$). Mean GPS showed a slight deterioration between E1 and E2 due to a minority of six patients (30%) who showed a significant loss of correction.

Conclusion Surgical treatment in adults with BSCP was feasible and effective in the long-term. Significant improvement of gait and function was maintained in the majority of patients, while some patients were prone to develop crouch gait, hip flexion contractures, or pain.

Keywords Cerebral palsy · Gait · Multilevel surgery · Adults

Introduction

Cerebral palsy (CP) is one of the most frequent causes of child disability in developed countries with the consequence that children, adolescents, and adults need lifelong assistance and care [1]. Transition of care from paediatric to adult health care providers is of general interest because these individuals are surviving now into adulthood [2]. A recent survey among multidisciplinary CP clinics in the USA confirmed that transition of care is not satisfactory to individual physician providers. Furthermore, only 55% of the clinics had a transition program limited to age and therefore a multidisciplinary clinic in an adult care setting with adult providers is desirable [2]. Although no specific treatment can remediate the brain damage that causes complex functional impairment, there are multiple therapies including multilevel surgery and rehabilitation programs that aim at improving patients' activity and participation in daily life [1, 3]. The lack of evidence, underlining that there is no effect of physical exercise on functional mobility in ambulatory adults with CP, shows the need for quality research to determine the best interventions for adults [4]. Short and midterm outcome studies in adults with bilateral spastic cerebral palsy (BSCP) provide promising results after multilevel surgery (MLS) using objective three-dimensional gait analysis and gait indices as primary outcome measure [5–7].

The aim of MLS in adults is to improve gait or to maintain the ability to walk and to decrease energy consumption in

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patients with fixed contractures, lever arm dysfunction, and bony torsional problems [8, 9]. Pain, fatigue, or osteoarthritis are additional factors in aging patients with CP resulting in a decrease of walking ability [10, 11]. Although MLS represents the standard procedure in children with CP to correct multiple deformities of the lower limbs during one surgical intervention, it remains an invasive procedure requiring prolonged rehabilitation in adults [7, 12].

The long-term benefits of MLS in children with CP have been recently documented in a multicentre outcome study [13]. However, there are to our best knowledge no reports on long-term effects of MLS in adults with cerebral palsy. The purpose of this cohort study was to analyze the functional long-term outcome after MLS in ambulatory adults with cerebral palsy.

Methods

A retrospective clinical cohort study of prospectively collected data of a single centre experienced in the conservative and surgical treatment of children and adults with CP was conducted. Based on a previous study [7] that analyzed short-term outcome in 97 adult ambulatory patients with BSCP after MLS, a long-term follow-up study of all patients who underwent MLS in our institution from October 1995 to October 2011 was performed. The inclusion criteria were as follows: (1) Gross Motor Classification Level (GMFCS) I-III [14], (2) multilevel surgery at the age of ≥ 17 years, and (3) long-term follow-up (≥ 7 years). For the present investigation, these patients had to fulfill the following criteria: The presence of a three-dimensional gait analysis (3D-GA) as well as a clinical examination prior to surgical treatment (E0), at a short-term follow-up (E1) and a long-term 3D-GA (E2), defined as ≥ seven years after MLS, was mandatory for inclusion. From 82 patients, 12 patients already had a long-term examination and 60 patients were re-invited to undergo their long-term examination including 3D-GA (Fig. 1). Forty-two subjects were lost to follow-up because they moved away. Overall 28 subjects were asked to participate at the long-term examination, but only eight agreed to return to our hospital to complete the follow-up 3D-GA.

Additional surgical interventions other than removal of metal after MLS were not an exclusion criterion, but were listed separately as well as multilevel surgery in childhood or minor surgical interventions (all surgical interventions other than multilevel surgery [7]). Peri- or post-operative complications were evaluated and classified [15]. These interventions were carried out by three responsible senior orthopaedic surgeons (LD, FB, TD). Furthermore, the need of assistive devices (e.g., crutches) and orthoses was analyzed pre- and post-operatively.

Pre- and long-term clinical examination and 3D-GA were carried out according to standardized gait laboratory protocols [16]. For quantitative 3D gait analysis, a 120-Hz nine-camera system (Vicon ®, Oxford Metrics, UK) and two piezoelectric force plates (Kistler®, Winterthur, Switzerland) were used. Skin-mounted markers were applied to bony landmarks of the patients and kinematics and kinetics were calculated to conventional gait model, implemented in Plug-in-Gait (Vicon, Oxford Metrics, UK). Furthermore, subjects were asked to perform a walk barefoot along a seven metre walkway at a self-selected walking speed with or without assistive devices. All examinations were carried out by an experienced physiotherapist and a study nurse with special training in CP therapy and gait analysis.

The primary outcome measure was the gait index, Gait Profile Score (GPS) [17]. The GPS is an index of overall gait pathology consisting of nine kinematic variables that are calculated as a single number. The study was approved by the local ethics committee (S-270/2006).

Statistical analysis

Basic descriptive statistics including means and standard deviation (SD) were calculated and two-tailed dependent t tests for paired samples and repeated measures (E0, E1, and E2) were conducted to compare pre-, short-, and long-term GPS values. Normal values were obtained from an age-matched reference group of 25 normal subjects. The level of significance was set at $p = 0.05$ for all statistical tests. Statistical analysis was performed using IBM SPSS Statistics for Windows, Version 22.0.

Results

A total of 20 ambulant adults (10 women, 10 men) with BSCP and a Gross Motor Function Classification Level (GMFCS) I–III were included in this study. The mean age at the time of surgery was 24.8 (SD 7.6) years and at the time of long-term follow-up 35.2 (SD 8.8) years. The mean post-operative follow-up period was 10.9 (SD 4.1) years (minimum 7.6–maximum 20.6 years). Demographics are reported in Table 1. There was a significant increase ($p = 0.005$) in weight from baseline to long-term follow-up measurements, from 59.3 kg (10.6 SD) to 65.3 kg (14.4 SD). Height at E0 (164 cm, 9.2 SD) compared to E2 (164 cm, 8.6 SD) was not statistically significant ($p = 0.6$).

Overall 18 patients (90%) maintained the GMFCS between E0 and E2 (GMFCS I in 1, GMFCS II in 14, GMFCS III in 3 patients) compared to 16 patients (80%) between E0 and E1 (GMFCS I in 4, GMFCS 2 in 10, GMFCS III in 2 patients). At E2, two patients (10%) showed a deterioration of the GMFCS (GMFCS I to II and GMFCS II to III) compared to one patient

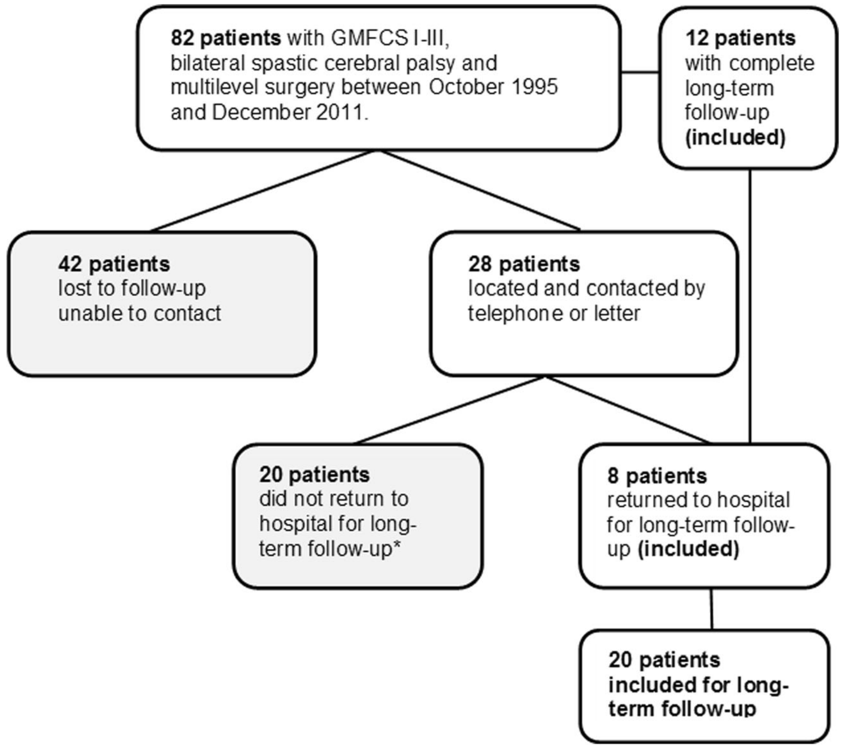

Fig. 1 Flowchart of the 20 patients enrolled in the study. (GMFCS Gross motor function classification level, (Asterisk) six patients declined our invitation, because they lived far away from our hospital and had actually no problems, four patients did not answer, nine patients did not specify the reason, but declined the invitation and one patient had pain)

(5%) at E1 (GMFCS II to III). Three patients (15%) improved at E1: two patients from GMFCS II to I and one patient from GMFCS III to II. At long-term follow-up, three patients (15%) required assistive devices compared to two patients (10%) at baseline. One patient had ankle-foot orthoses and crutches due to pain and a decrease of walking distance.

The number of operative procedures in all patients that was performed in adulthood is depicted in Table 2. Twelve patients (60%) had no surgical interventions in childhood, three patients (15%) had multilevel surgery, and five patients (25%) underwent minor surgery of the lower limbs at one level (Achilles tendon lengthening in three, intramuscular recession of gastrocnemius in one, and lengthening of the hamstrings in one patient).

The total GPS (Fig. 2) improved significantly from 13.8° (3.8 SD) at baseline to 11.3° (3.1 SD) at E1 ($p = 0.007$) and to 11.2° (2.4 SD) at E2 ($p = 0.002$). Means, standard deviations,

Table 1 Demographics of adult patients ($n = 20$) with bilateral spastic cerebral palsy

Characteristics	Preoperative	Long-term follow-up (10.9 years)
Age (years) SD	24.8 SD	35.2 SD
Sex (female/male)	10/10	
GMFCS		
GMFCS I	4	3
GMFCS II	13	13
GMFCS III	3	4
Height (cm)	164.4	164.0
Weight (kg)	59.3	65.3
Orthoses/assistive devices (yes/no)	2/18	3/17

Table 2 Outline of surgical procedures of the study group

Surgical procedures	20 patients ($n = 40$ legs) MLS	20 patients ($n = 40$ legs) additional surgery
Femoral derotation osteotomy (FDO)	11	1
Tibial derotation osteotomy	6	1
Bony foot stabilization*	8	2
Intramuscular psoas lengthening	5	
Rectus femoris surgery	24	
Adductor lengthening	7	
Hamstring lengthening	22	
Posterior capsulotomy (knee)	3	
Calf muscle lengthening	20	
Achilles tendon lengthening	3	
Soft tissue procedures, foot#	3	2
Total	112	6

MLS multilevel surgery

*Consisted of calcaneocuboid distraction arthrodesis, triple arthrodesis, and Evans (lateral calcaneal lengthening) procedure

Consisted of split anterior tibial tendon transfer, split posterior tibial tendon transfer, and plantar fascia release

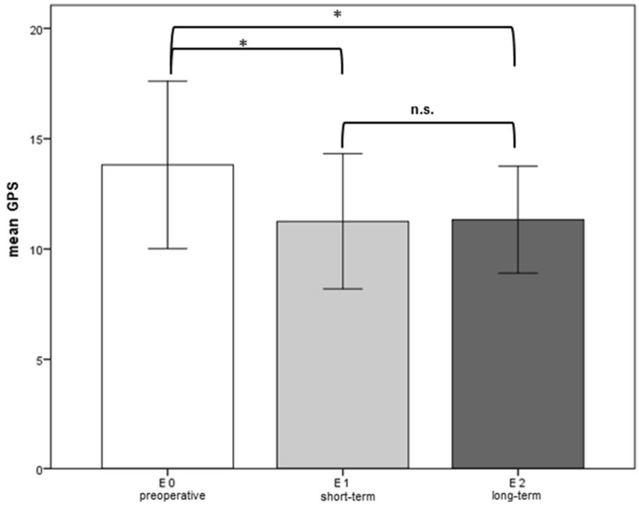

Fig. 2 Bar graphs at E0 (pre-operative), E1 (short-term), and E1 (long-term) including mean GPS (total) values (*y*-axis). *Significant change ($p < 0.05$) E0E1, E0E2. n.s. not significant change E1E2

and statistical significances are depicted in Table 3. For the GPS, no statistical significant change between E1 and E2 was noted ($p = 0.89$). Analyzing the individual course of GPS between E1 and E2, overall six patients (30%) showed a deterioration of ≥ − 1.0 GPS difference (Fig. 3). The ratio between the GPS difference E1E2 and the minimal clinical important difference (MCID) [18] 3.2° was calculated.

The reasons for the deterioration in these individual cases were analyzed for each case: intramuscular lengthening of the hamstrings in combination with rectus femoris surgery in three cases was associated with post-operative hip extensor weakness and a further deterioration of hip flexion contracture as well as deterioration of crouch gait. In one case, recurrence of internal rotation gait was the reason for worsening of the gait indices. In patients four and five, weakening of the calf muscle after intramuscular lengthening of the gastric-soleus muscle was probably too aggressive leading to an increase in dorsiflexion of the ankle and crouch gait. In patient six, the deterioration was due to a progressive pain symptomatic at the level of the knees (retro-patellar pain).

Two patients (10%) had an additional surgical procedure (Table 2). One patient had a Jones procedure and correction of claw toes bilaterally and another patient had unilateral distal femoral derotation and supra-malleolear derotation osteotomy for internal rotation gait.

One patient (5%) had a pressure sore at the level of the Achilles tendon after casting that healed after treatment without consequences.

Table 3 Analysis of GPS values

GPS	Mean	SD	Difference E0, 1, 2 (SD)	Level of significance
GPS_E0	13.8	3.8	E0E2: 2.5 (3.1)	E0E2: 0.002#
GPS_E1	11.3	3.1	E0E1: 2.5 (3.7)	E0E1: 0.007#
GPS_E2	11.2	2.4	E1E2: − 0.1 (2.5)	E1E2: 0.89 n.s.

Level of significance $p < 0.05$, *n.s.* not significant), *SD* standard deviation

Discussion

This cohort study has the longest follow-up (11 years) in ambulant adult patients with cerebral palsy after MLS to date. The main problem in following up these patients is the transition from the pediatric to the adult health care system that induces patients to change the institution [2]. Although in our hospital, cerebral palsy patients can be managed from childhood to adulthood without an age limit; nevertheless, there is a considerable loss to follow-up.

The GPS showed a significant improvement at a mean of 10.9 years post-operatively and additional surgery was necessary in only two patients (10%). The total GPS improved from 13.8° to 11.3° (difference 2.5° SD 3.1°) indicating that MLS is an effective procedure in improving gait in adults with cerebral palsy. In our previous study [7], 97 adults with MLS had a significant improvement in GPS 1.7 years after surgery. The mean values of these results are in accordance with our E1 (short-term) results indicating that these values are representative for the study group.

The overall change of GPS values from 15.9° to 11.9° in the short-term study [7] (19.7 months) compared to our long-term results in adults with CP (13.8° to 11.3°) demonstrates that the final endpoint is nearly the same. However, the slight decrease of baseline GPS 13.8° in the long-term study shows that the included 20 patients were less impaired compared to the short-term study (15.9°). For this reason, our long-term results did not reach the MCID [18] (minimally clinical important difference) of 3.2° between baseline and long-term follow-up. The important contribution of this investigation is that after skeletal maturity, the improvement of gait remained stable over time in the majority of the patients. However, in six patients (30%), a deterioration of more than 1 GPS point between short- and long-term follow-up was noted. The kinematic analysis of these individual cases showed that the occurrence of crouch gait, internal rotation gait, and progressive hip flexion contracture was associated with an excessively weakening of the calf or hamstring muscles.

In contrast to the deterioration of the GMFCS in the short-term study (21%) [7], the pre- and long-term GMFCS in our study showed that 10% of the patients had less deterioration of the GMFCS and 90% maintained their GMFCS. The hypothesis was that after correction of the biomechanical axis, assistive devices were needed after MLS only for a certain time. The present investigation illustrates that pain was a

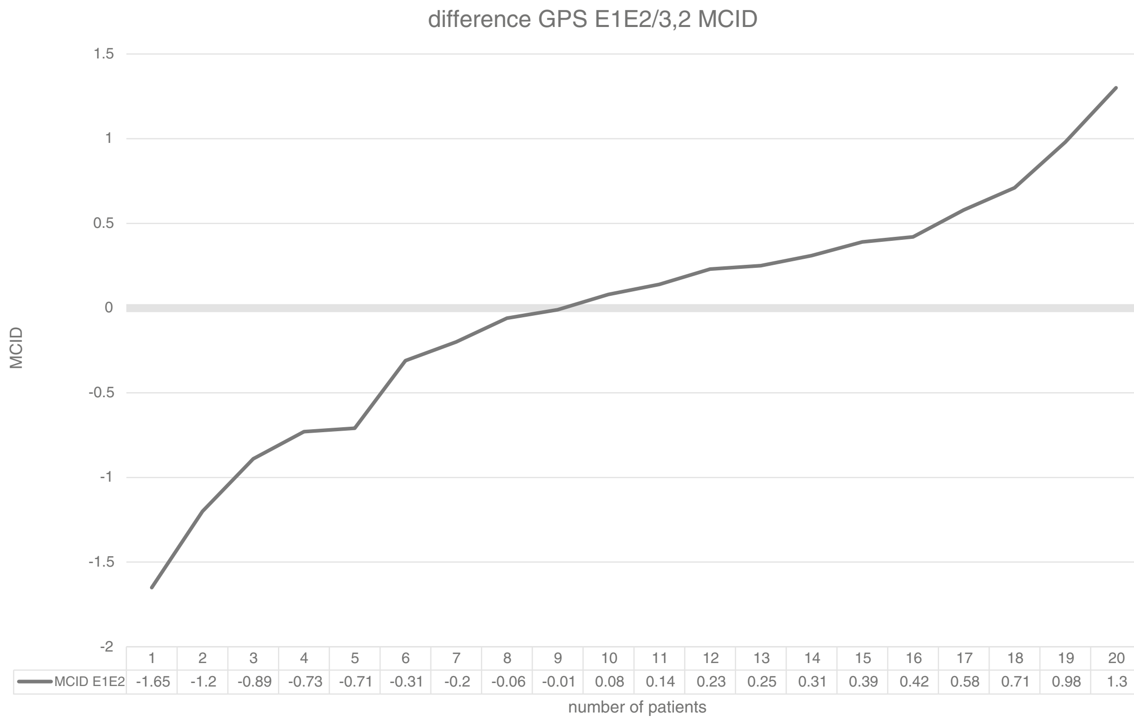

Fig. 3 Individual analysis of GPS difference between short- (E1) and long-term (E2) in the ration to minimally clinically important difference (MCID) of 3.2 in all 20 patients

principal reason for deterioration of GMFCS in the long-term. Careful patient selection for MLS is therefore a principal task. A total of 25% of ambulant CP patients experience gait decline including factors like an inferior initial walking ability and bilateral motor impairment [19]. Musculoskeletal contractures, pain, and fatigue contribute to a decline in gait function and consequently to an inactive lifestyle [20]. A systematic review on health-related fitness in adults with CP indicated that muscular strength and muscular and cardiovascular endurance showed significantly lower values compared to able-bodied controls [20]. Adult patients with CP are limited in their movements due to a compromised reciprocal synchronization of agonists and antagonists, a shortened position of some muscles, a higher ratio of type I to type II muscle, and a 50% lower muscle volume compared to typically developing controls [20]. Russchen et al. [21] pointed out that fatigue is already present at a relatively young age among adults with CP. The CP subtype seemed to be a determinant factor, but no evidence between fatigue and daily life activity and cardiopulmonary fitness was found [21].

In a European multicenter longitudinal study on children with CP, 54% of the children reported that they had pain with the consequence of poorer quality of life [22]. In the follow-up study in adolescence, pain was even more important in 75% of the participants [23]. Alriksson-Schmidt and Hägglund [24] analyzed the Swedish registry data and reported on pain to be a significant problem in children and adolescent with a prevalence of 32.4%. The frequency of pain increased with age and there was an association between the pain site and the GMFCS levels [24]. These results underline that more research on pain in adults with CP is required.

Most studies dealing with the quality of life in adults with CP [20, 21] do not consider the effect of surgery or indicate if patients have undergone surgery. Spasticity remains one dynamic factor after the cessation of growth to deteriorate gait in adult patients with BSCP [20, 21]. Therefore, it remains a question of debate, if the good short- or mid-term results can be maintained over time [5–7]. In our study, adverse events (additional surgery) occurred in 10% of the cases. These surgical interventions are performed to correct persistent deformities and patients and their families should be informed. In comparison to studies in ambulatory children with CP, the effects of MLS in adults are stable over time, but in individual cases, deterioration of the GMFCS and the GPS can occur. Long-term retrospective studies [25] analyzing the effect of single-event multilevel surgery (SEMLS) in children show favorable results, but on the other side, contractures progress during growth resulting in deterioration of function and gait. The largest multicentre long-term outcome study [13] in children with CP and a follow-up of 9 years following multilevel surgery revealed that at short- and long-term follow-up gait and function were stable after the pubertal growth spurt and skeletal maturity. In accordance with our study, the short- and long-term GPS were approximately the same. A total of 4.8% of all children had a deterioration of GPS, compared to 30% in our study. Further, the deterioration of the GPS between E1 and E2 did not include patients with a deterioration of the GMFCS. Our results include a small sample size and there is a possibility that patients with symptoms were over-represented (e.g., deterioration of gait, pain). Our study shows that MLS in adults results in an improvement of gait that is sustained over the long-term.

Limitations of the study

This study was a retrospective study of prospectively collected data preoperatively, at short-term and at long-term follow-up after multilevel surgery, but without a control group that did not undergo surgery. Patients who decided against surgery were not included in the gait laboratory. As in all retrospective studies, there is partial patient selection bias. Statistically, only 24% of the original cohort was analyzed given the fact that overall 51% of the patients were lost to follow-up. This is clearly a major limitation of the study and a problem of transition between paediatric and general orthopaedic institutions treating patients with cerebral palsy. Further prospective investigations are necessary to clarify the effect of multilevel surgery in adults on daily life activities and social and psychological factors.

Compliance with ethical standards

The study was approved by the local ethics committee (S-270/2006).

Conflict of interest The authors declare that they have no conflict of interest.

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