Outcome of intensive rehabilitation following single-event multilevel surgery for crouch gait in children with cerebral palsy

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Background
Crouch gait is one of the most common gait patterns in ambulatory children with cerebral palsy (CP) and is contributed by several factors. The literature reports wide variations in surgical practice and rehabilitation programs following single-event multilevel surgeries.

Objective
To evaluate the outcome of rehabilitation after single-event multilevel orthopedic surgery for crouch gait in children with CP.

Patients and methods
A total of 20 children with bilateral spastic CP and walked with a crouch gait, with gross motor function classification system levels II, III, and IV, were subjected to single-event multilevel surgery. Ten (20 limbs) patients were men and eight (14 limbs) were women. Their age ranged from 5.5 to 19 years. To assess the outcome of our rehabilitation program, we used clinical couch examination parameters, functional mobility scale, and instrumented three-dimensional gait analysis. Rehabilitation program included preoperative and postoperative rehabilitation at 1-year postoperatively.

Results
Highly statistically significant improvements in clinical parameters, which include hip abduction, femoral anteversion, fixed flexion deformity, popliteal angle, and extension lag, were demonstrated (P<0.01), whereas tibial torsion showed a statistically significant improvement (P<0.05). Functional mobility scale at 5, 50, and 500 m improved in 10 (55.6%) cases, 13 (72.2%) cases, and 11 (61.1%) cases, respectively. Instrumented gait laboratory parameters, namely, stride length, crouch angle at initial stance, and peak knee flexion in mid-swing, showed improvement but did not reach statistical significance.

Conclusion
The rehabilitation program we offered improves clinical and functional mobility of children with CP with crouch gait. Thus, it is viewed as an important contributor to the overall outcome together with multilevel orthopedic surgery.

Keywords: cerebral palsy, gait, multilevel, rehabilitation

Introduction
Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to nonprogressive disturbances that occur in the developing fetal or infant brain [1]. Ambulatory children with CP present with different gait patterns caused by spasticity and contractures with subsequent limited range of motion (ROM), leading to loss of functional abilities. Crouch gait is one of the most common gait patterns in children with ambulatory CP [2]. Rodda et al. [3] described five patterns of gait in spastic diplegic patients based on the pelvis, hip, knee, and ankle position during stance. With a definition of crouch gait as knee flexion throughout stance larger than 20°, the ankle is excessively dorsiflexed and the hip is excessively flexed during stance. The pelvis is in the normal range or tilted posteriorly [3]. Children may demonstrate crouch gait because of weakness of hip and knee extensors and ankle plantar flexors as well as contracted hamstrings and hip flexors [4]. Once the crouch gait reaches a certain level of severity in the child, the degree of knee flexion and associated symptoms may progress rapidly because of high stresses at the knee and failure of the knee extensor mechanism. Knee pain, patella alta, and fragmentation or fracture of the inferior pole of the patella all have been documented in this clinical setting [5].
Single-event multilevel surgery (SEMLS) improves the likelihood of achieving sagittal plane balance, reduces the need for repeated anesthesia, episodes of hospitalization, and requires only one major period of rehabilitation with reduction of the cost [6]. A systematic review of SEMLS reported wide variations in surgical practice and rehabilitation. It is the postoperative rehabilitation that makes children functionally ‘better’ [7]. This study aimed to evaluate the effect of rehabilitation after single-event multilevel orthopedic surgery for crouch gait in children with CP.

Patients and methods
This was a cross-sectional prospective study that initially included 20 patients who had bilateral spastic CP and walked with a crouch gait, and who underwent SEMLS; all cases were operated upon and followed up at the authors’ institutions.

Inclusion criteria
The following were the inclusion criteria:

(1) Ambulatory children more than or equal to 5 years of age and adolescents with bilateral spastic CP.
(2) Crouch gait.
(3) Deterioration in walking suggested by a reduction in speed or in walking distance or increased knee flexion in stance phase in the past year and/or pain, especially patellar pain and/or foot pain.

Exclusion criteria
The following were the exclusion criteria:

(1) Botulinum toxin A injections within the preceding 6 months.
(2) Muscle surgery within the preceding 12 months.
(3) Noncommunicable patients.
(4) Patients not attending regular follow-up visits.

All patients gave an informed consent, and the study protocol was approved by the Institutional Review Board of Ain Shams University.

Of the 20 patients, two patients were lost to follow-up, so the total number of patients who completed the study was 18 (34 limbs).

Preoperative evaluation: patients were evaluated for the following:

(1) History taking, which emphasized on, but was not limited to, the following points: perinatal history, developmental history, any recent deterioration of patient activities and anterior knee pain, previous physiotherapy, and previous interventions.

(2) Clinical examination: standard couch examination of each patient was done; goniometric measurements of joint ROM and deformities were done for the hip, knee, and ankle joints bilaterally; muscle power of the lower limb muscles was assessed on a five-grade scale by manual muscle testing; and observational gait analysis. Evaluate expanded and revised gross motor function classification system (GMFCS E and R) for children and youth with CP [8].

(3) The functional mobility scale (FMS): this is a tool for the assessment of patient mobility. FMS was measured by asking each patient or care giver about the level of mobility. FMS uses three distances (5, 50, and 500 m), which represent typical distances walked by children at home, at school, and in the community. For each distance, a rating of 1–6 was assigned depending on the amount of assistance required for mobility (6 being the highest ability) [9].

(4) Instrumented three-dimensional gait analysis: it was done for 15 cases preoperatively; of them, nine patients did postoperative gait analysis as well. Kinematics and dynamic electromyography assessments were done in the same laboratory, and it is done to formulate a surgical plan for each patient individually [10].

Rehabilitation program
Rehabilitation of children with CP after SEMLS was tailored according to each patient condition. We followed the following rehabilitation program [7,11].

Preoperative rehabilitation
It included regular ROM exercises, muscle strengthening exercises (including quadriceps, hamstrings, hip abductors, extensors, and flexors, ankle dorsiflexors, and evertors), muscle stretching to spastic muscles (hamstrings, gastrocnemius, and adductors), gait training, balance, and core exercises. Description of postoperative rehabilitation plan for parents and children is as follows:

Postoperative rehabilitation
From day of surgery to 6 weeks postoperatively
In the operated limb (which was in cast): Assisted active to active ROM exercises for unfixed joints (e.g. hips, toes) as tolerated was recommended along with strengthening of the muscles acting on unrestricted joints, from isometric progressed to isotonic exercises. Best position in bed for hips, knees, and ankles; elevation for edema control; trunk exercises; and assisted transfer education were also recommended.
Non-weight-bearing in bony surgery and weight bearing as tolerated in the case of soft tissue surgery only were initiated.

From 6 to 12 weeks postoperatively
This period was very important for regaining ROM and muscle strength. ROM exercises were started in previously restricted joints. Starting of isometric strengthening exercises in muscles acting on previously restricted joints progressed to isotonic and resistance exercises.

Use of heating modalities on stiff joints was common to decrease pain and improve ROM (e.g. paraffin wax or infrared radiation) with caution to avoid burns to the child.

Electric neuromuscular stimulation was added in cases with muscle recession or transfer (e.g. rectus femoris recession and tibialis anterior transfer).

Stretching exercises were started to maintain muscle length and avoid contractures and deformities of the hip, knee, and ankle flexors and adductors according to each case condition and lengthening interventions. Partial weight bearing was started with the use of orthotic devices and walker and or parallel bar for gait training.

Instructions for use of orthotic devices according to each case condition were as follows: mostly knee ankle foot orthosis was described for night positioning, ankle foot orthosis, or ground reaction ankle foot orthosis for ambulation (in case of quadriceps muscle weakness or lag).

Session frequency: five times per week for 1–2 h. Home program of simple exercises and positioning was described for adolescent children as well as parents.

From 3 to 6 months
Continuation of the orthotic device was done. Progressive ROM, strengthening, and stretching exercises were recommended, along with assisted balance exercises (static to semidynamic balance), and increased ambulation distance.

From 6 to 9 months
Continuation of the previous items, along with continuation of day use of orthotic devices was recommended. Moreover, return to preoperative level of walking and community participation was suggested.

From 9 to 12 months
This period led to progression to better gait, independence, and dynamic activity.

The frequency of sessions was reduced for most children and was replaced with recreational activities, including family walks, bicycle riding, and sports participation.

Postoperative evaluation
The patients were reviewed postoperatively at regular follow-up intervals: 6 and 12 weeks and then 3 monthly during the first year. They were evaluated by the following: standardized physical examination, observational gait analysis, the FMS, and instrumented gait analysis, which were assessed after 1 year. Patients with osteotomies were followed up by serial radiograph till complete healing.

Statistical analysis
All parameters were measured preoperatively and at 1-year postoperatively. The paired Wilcoxon signed-rank test was used to compare the preoperative and postoperative physical examination findings, walking speed, and FMS. The values were given as the median and the interquartile range. The significance level was defined by the probability value: $P$ value less than or equal to 0.05 was considered significant; $P$ value less than 0.01 was considered highly significant; and $P$ value more than 0.05 was considered nonsignificant.

Results
Ten patients were men (20 limbs, 58.82%) and eight were women (14 limbs, 41.18%). Age ranged from 5.5 to 19 years, with a mean age of 12 years.

The 18 patients were classified according to the expanded and revised GMFCS E and R for children and youth with CP [11]. Five (27.8%) were GMFCS II, 10 (55.5%) were GMFCS III, and three (16.7%) were GMFCS IV. All patients have had bilateral spastic diplegic CP. Two of them have had an inborn error of metabolism, namely (well controlled), maple syrup urine disease. Eleven (61%) patients had previous interventions, either surgery or Botox injection (>1 year and 6 months before index procedure, respectively), especially of the calf muscles.

Preoperative and 1-year postoperative values for six physical examination parameters are described in Table 1. Data from both sides were added. Most of the values at 1-year postoperatively were highly significantly different from the preoperative values ($P<0.01$).
Values of FMS at 5, 50, and 500 m are described in Tables 2 and 3. FMS at 5, 50, and 500 m improved in 10 (55.6%) cases, 13 (72.2%) cases, and 11 (61.1%) cases, respectively.

Preoperative and 1-year postoperative values of three gait laboratory parameters, namely, stride length, crouch angle at initial stance, and peak knee flexion in mid-swing, are represented in Table 4.

Example of improvement of cases is represented by a case of a male patient, 13 years old, with GMFCS III. He had a history of obstructed labor and delayed developmental milestones. Botox injections to hamstring and gastrosoleus four times were followed by right hamstring lengthening 2 years before his index surgery. He was subjected to bilateral femoral supracondylar extension derotation osteotomy, bilateral patellar tendon advancement, bilateral gastrocnemius recession, and right supramalleolar derotation osteotomy, and correction of both feet deformities were done by a double team working simultaneously. Rehabilitation program started at day 5 postoperatively and continues till 1 year.

At 1-year postoperatively, the patient improved in terms of clinical parameters, especially femoral anteversion, fixed knee flexion deformity, extension lag, and foot deformity (Fig. 1). FMS was improved at 50 m only. There were improvements in walking speed. Postoperative gait laboratory parameters showed improvement in knee kinematics and hip rotation as compared with preoperative curves (Fig. 2).

Discussion

There is limited information available about post-SEMLS rehabilitation [12]. Evidence was found for large improvements in gait with more equivocal evidence for changes in gross motor function [12]. Orthopedic surgery results in weakness, loss of independence, and decreased gross motor function,
which makes rehabilitation after SEMLS challenging [7].

No previous comparative studies have been conducted to compare different rehabilitation protocols. Ten of our patients were men and eight were women. Their age ranged from 5.5 to 19 years, with a mean age of 12 years. Most children showed a plateau in gait and gross motor function, diminishing response to botulinum toxin injections, and the progression of fixed contractures between the age of 5 and 8 years, and this is the time when serious planning for SEMLS should start [7].

A total of 146 procedures were done to the patients: 92 soft tissue procedures and 54 bony procedures. Apart from the two missed cases, 18 patients were highly compliant to rehabilitation sessions and follow-up visits. The clinical parameters of our patients showed a highly significant improvement in hip abduction, femoral anteversion, fixed flexion deformity of the left knee, popliteal angle, and extension lag of knees, whereas there is significant improvement of tibial rotation and right knee fixed flexion deformity.

In our study, we observed that the pattern of change differed according to the functional level preoperatively. Children with GMFCS II walked without assistive devices at household distances and therefore had improved statistically significantly at greater distances. Children with GMFCS levels III and IV had improved more at short distances and the greatest change was at 5 m. FMS of 5, 50, 500 m improved in 10 (55.6%) cases, 13 (72.2%) cases, and 11 (61.1%) cases, respectively. Our patients reached the preoperative level at 6 months postoperatively. This
agrees with other studies. The world’s first randomized controlled trial of SEMLS was published by the team at the Royal Children’s Hospital in Melbourne. This randomized clinical trial reported a 57% improvement in gait according to the Gillette Gait Index and a 4.9% improvement in gross motor function according to the GMFM-66 [13].

Terjesen et al. [14] studied gait improvements in 34 ambulatory children with spastic diplegia after multilevel surgery. They evaluated the functional level using the FMS and found the same results, as compared with our study, at 1 year. Moreover, 5-year postoperatively, children at GMFCS level III showed improvements at FMS 500 m [14].

Harvey and colleagues examined the ability of the FMS to detect changes in children with CP undergoing SEMLS. Their study was a retrospective one and comprised 66 children. For each FMS distance (5, 50, and 500 m), odds ratios showed a significant deterioration in mobility at 3 and 6 months postoperatively. Mobility then improved to baseline levels by 12 months and improved further by 24 months postoperatively [8].

Regarding the complications in this series, there is a need for additional surgery in four (22.2%) patients. Even if SEMLS is aimed at correcting all existing deviations in gait, additional surgery is sometimes necessary. Kay et al. [15] found a need for such surgery already at the 1-year follow-up. Preoperative and 1-year postoperative values of three gait laboratory parameters, namely, stride length, crouch angle at initial stance, and peak knee flexion in mid-swing, showed improvement but did not reach statistical significance, which may be owing to the small sample size, as preoperative and postoperative instrumented three-dimensional gait analyses were done for only nine cases. It may be also owing to the short follow-up period. Dequeker et al. [16] found a significant increase in the pediatric evaluation of disability, the mobility questionnaire, and FMS at 6 months and 1 year, which agrees with our results. Dreher et al. [17] found that ambulatory children aged 10 years and 7 months with GMFCS levels I, II, and III undergoing multilevel surgery showed a decrease (improvement) in preoperative gait profile score from 16.3° to 11.3° at short-term follow-up, and improvement of 5° at long-term follow-up. A large systematic review [18] on hip dislocation in children with CP has clearly revealed these short comings of retrospective studies, thus the body of evidence on the treatment outcomes of retrospective studies is considerably more limited than for prospective studies.

Our study was prospective, and rehabilitation is the main scope, which is present in few studies and is considered a strength point. We consider the relatively small sample size as important limitations. The generalizability of the findings would also have been improved with a larger study sample and a longer follow-up period.

Conclusion
At 1-year postoperatively, there were significant improvements in all clinical parameters and FMS. These measures reflect clinical improvement of patients and their functional mobility, denoting the essential role of rehabilitation in these patients.

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Conflicts of interest
There are no conflicts of interest.

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