Single-Event Multilevel Surgery in Children with Spastic Diplegia

A Pilot Randomized Controlled Trial

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Investigation performed at the Royal Children's Hospital, Melbourne, Australia

Background: Single-event multilevel surgery is considered the standard of care to improve gait and functioning of children with spastic diplegic cerebral palsy. However, the evidence base is limited. This pilot study is the first randomized controlled trial of single-event multilevel surgery, to our knowledge.

Methods: Nineteen children (twelve boys and seven girls with a mean age of nine years and eight months) with spastic diplegia were enrolled. Eleven children were randomized to the surgical group and eight, to the control group. The control group underwent a program of progressive resistance strength training. The randomized phase of the trial concluded at twelve months. The control group then exited the study and progressed to surgery, whereas the surgical group continued to be followed in a prospective cohort study. The primary outcome measures were the Gait Profile Score (GPS) and the Gillette Gait Index (GGI). Secondary outcome measures were gross motor function (Gross Motor Function Measure-66 [GMFM-66]), functional mobility (Functional Mobility Scale [FMS]), time spent in the upright position, and health-related quality of life (Child Health Questionnaire [CHQ]).

Results: A total of eighty-five surgical procedures were performed, with a mean of eight procedures per child (standard deviation, four). The surgical group had a 34% improvement in the GPS and a 57% improvement in the GGI at twelve months. The control group had a small nonsignificant deterioration in both indices. The between-group differences for the change in the GPS (-5.5; 95% confidence interval, -7.6 to -3.4) and the GGI (-218; 95% confidence interval, -299 to -136) were highly significant. The differences between the groups with regard to the secondary outcome measures were not significant at twelve months. At twenty-four months after surgery, there was a 4.9% increase in the GMFM-66 score and improvements in the FMS score, time spent in the upright position, and the physical functioning domain of the CHQ in the surgical group.

Conclusions: This study provides Level-II evidence that single-event multilevel surgery improves the gait of children with spastic diplegic cerebral palsy twelve months after surgery. Improvements in other domains, including gross motor function and quality of life, were not observed until twenty-four months after surgery.

Level of Evidence: Therapeutic Level II. See Instructions to Authors for a complete description of levels of evidence.

Although cerebral palsy is by definition a static encephalopathy, the associated musculoskeletal pathology is often progressive.3

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During the past twenty years, increasing emphasis has been placed on the correction of all fixed musculoskeletal deformities with single-event multilevel surgery.4–11 The primary

A commentary by Mark F. Abel, MD, and Diane L. Damiano, PhD, PT, is available at www.jbjs.org/commentary and is linked to the online version of this article.
goal of single-event multilevel surgery in children with cerebral palsy is to improve gait\textsuperscript{3-14}. Secondary goals may include improvements in gait efficiency\textsuperscript{3} and appearance, gross motor function\textsuperscript{3,14}, independence\textsuperscript{5,6}, and quality of life\textsuperscript{14-16}. Several well-designed retrospective cohort studies have demonstrated an improvement in gait or functional ability following single-event multilevel surgery\textsuperscript{4,14}. However, there have been few prospective studies, few studies that included a control group, and no randomized clinical trials to our knowledge\textsuperscript{6-14}. Given the tendency for uncontrolled studies to overestimate treatment effects in patients with cerebral palsy, we believed that a randomized clinical trial of single-event multilevel surgery was necessary and appropriate\textsuperscript{7}.

A major issue in the literature is the choice of outcome measures\textsuperscript{17-21}. The International Classification of Functioning, Disability and Health (ICF)\textsuperscript{22} model provides a framework to identify factors affecting disability and should be considered when evaluating an intervention (Fig. 1).

It was our intention to design a large, multicenter randomized controlled trial to evaluate the outcomes of single-event multilevel surgery in children with bilateral spastic cerebral palsy. However, the magnitude and timing of change following single-event multilevel surgery across the multiple ICF domains was at that time largely unknown. Therefore, we considered it appropriate to conduct a single-center randomized controlled trial to determine the treatment size effects and the trajectory of change in outcome measures. The aim of this pilot study was to evaluate the magnitude of change between groups and over time on the basis of gait indices, physical measures, function, activity, mobility, and health-related quality of life following single-event multilevel surgery in children six to twelve years of age who had spastic diplegia (GMFCS [Gross Motor Function Classification System]\textsuperscript{13,14} level II or III).

**Materials and Methods**

We performed a single-center, randomized controlled trial in which single-event multilevel surgery followed by intensive postoperative physical therapy was compared with physical therapy alone. The primary outcome measures were the Gait Profile Score (GPS) and the Gillette Gait Index (GGI), both summary statistics of gait or gait indices\textsuperscript{22-27}. The GPS is a recently described measure of overall gait quality\textsuperscript{23}. It is calculated from the root mean square difference between the gait kinematics for an individual trial and the average kinematic data from children with no gait pathology. The units are degrees, and the larger the GPS the more abnormal is the subject’s gait. A major advantage of the GPS is the associated Movement Analysis Profile (MAP), which provides a graphical display of the specific kinematic deviations giving rise to the overall GPS. The GGI is also a single summary statistic of gait derived from sixteen kinematic and temporospatial measures of gait\textsuperscript{26,27}. Validation studies support its use to evaluate outcomes in individuals who have had three-dimensional gait analysis\textsuperscript{18}. Both measures are considered to be valid and reliable tools to describe gait dysfunction as a single variable and to assess change after intervention\textsuperscript{14,22,23}.

The GPS and the GGI were assessed at baseline and at twelve months postoperatively, at which time the randomized phase of the trial concluded. The children in the control group exited the study after the twelve-month assessment and progressed to surgery (Fig. 2). The children who had been randomized to receive surgery continued to be followed in a prospective cohort study for a minimum of three years.

Ethical approval for this study was granted by the Ethics in Human Research Committee of the Royal Children’s Hospital, Melbourne. The trial design and reporting follow the CONSORT (Consolidated Standards of Reporting Trials) principles. The trial was registered with an online clinical trials registry (Australia New Zealand Clinical Trials Registry ACTRN1260900117280). Without pilot data, a formal sample-size calculation was not possible. We aimed to recruit twelve participants over eighteen months, with the intention of using the data for sample-size estimates for a future definitive study if nonsignificant trends were observed.

**Figs.**

![Fig. 1](image-url)

Outcome measures used according to the International Classification of Functioning, Disability and Health. 3DGA = three-dimensional gait analysis, MAP = Movement Analysis Profile, GPS = Gait Profile Score, GGI = Gillette Gait Index, ROM = range of motion, GMFM-66 = Gross Motor Function Measure-66, FMS = Functional Mobility Scale, CHQ-PF50 = Child Health Questionnaire-Parent Form 50. The activity and participation domains were combined for simplicity as some outcome measures encompass both of these domains.
The inclusion criteria were a confirmed diagnosis of cerebral palsy with registration in the Victorian Cerebral Palsy Register, a spastic movement disorder, an age of six to twelve years, a GMFCS level of II or III, and suitability for multilevel surgery. Prior injection of botulinum neurotoxin A was not a reason for exclusion if more than six months had elapsed since the last injections.

Exclusion criteria were a diagnosis of dystonia and prior orthopaedic surgery, selective dorsal rhizotomy, or intrathecal baclofen therapy. In addition, children were excluded if there was any reason why delaying surgery might cause harm such as hip migration in excess of 25% on radiographs, painful breakdown of the midfoot, and progressive crouch gait. Progressive crouch gait was defined as a loss of knee extension of >10° in late stance.

For the purposes of this trial, single-event multilevel surgery was defined as at least one surgical procedure performed at two different anatomic levels (the hip, knee, or ankle) on both sides of the body. The surgical recommendation was tailored to the child's needs as determined by a comprehensive evaluation, including a standardized physical examination, radiographic evaluation, and instrumented gait analysis. The multilevel surgical program included muscle tendon lengthening, tendon transfer, rotational osteotomy, and stabilization of the hip and foot according to published guidelines.

Informed written consent to participate was obtained from the parents of eligible children following a minimum of two detailed interviews with the treating surgeons and the study coordinator.

The children allocated to the surgical group had surgery, under general anesthesia, performed by two experienced surgeons within four weeks after the baseline assessment. Perioperative antibiotics and epidural infusions of 0.25% bupivacaine were used. The children remained as inpatients for five to seven days following surgery and were discharged wearing below-the-knee plaster casts, with knee immobilizers and use of the appropriate assistive devices as indicated by their GMFCS level.
The children were assessed at three and six weeks postoperatively to check the healing of surgical incisions and osteotomy sites and to provide custom-fitted ankle-foot orthoses. Physical therapy in the first three months was aimed at regaining function lost as a result of the surgery. This was followed by an intensive program performed three times per week for twelve weeks and aimed at improving the range of motion, strength, balance, and function. Funding for the rehabilitation program was provided by the Post Intervention Physical Therapy Program.

The control group underwent a progressive resistance strength training program, to match the intensive physical therapy received by the surgical group, according to protocols that have been described in detail elsewhere. Thus, children in the control group continued their routine physical therapy group, according to protocols that have been described in detail elsewhere.

The Gillette Gait Index, 1 RM values are presented as the mean and standard deviation except where otherwise noted. The Gillette Gait Index is an ordinal scale that describes the level of assistance required by children with cerebral palsy to mobilize over different distances and environments. It is able to detect both improvement and deterioration after single-event multilevel surgery. Isometric muscle strength was measured with use of the Lafayette handheld manual muscle testing system (Lafayette Instrument, Lafayette, Indiana). The time that the child spent in the upright position, standing or walking, was recorded over a four-day period.

### TABLE 1 Results of Between-Group Comparison (with Use of Analysis of Covariance) at Twelve Months for Gillette Gait Index, Gait Profile Score, Physical Examination Measures, Gross Motor Function Measure, Time Spent in Upright Position, and Child Health Questionnaire

<table>
<thead>
<tr>
<th>Measure†</th>
<th>Single-Event Multilevel Surgery Group* (N = 11)</th>
<th>Control Group* (N = 8)</th>
<th>Difference Between Groups in Change at 12 Mo (95% Confidence Interval)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Baseline 12 Mo</td>
<td>Baseline 12 Mo</td>
<td></td>
</tr>
<tr>
<td>Body structure/function</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>GPS‡</td>
<td>13.7 (11.9, 15.2) 9.1 (8.6, 12.6)</td>
<td>14.6 (10.5, 15.8) 15.7 (13.9, 16.2)</td>
<td>-5.5 (-7.6, -3.4)§</td>
</tr>
<tr>
<td>GGI</td>
<td>353 (211) 153 (81)</td>
<td>370 (194) 381 (196)</td>
<td>-218 (-299, -136)§</td>
</tr>
<tr>
<td>Physical examination</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hip extension (deg)</td>
<td>1 (10) 1 (6)</td>
<td>-2 (8)</td>
<td>-5 (7)</td>
</tr>
<tr>
<td>Hip abduction (deg)</td>
<td>30 (7) 30 (7)</td>
<td>27 (6)</td>
<td>24 (7)</td>
</tr>
<tr>
<td>Hip internal rotation (deg)</td>
<td>67 (14) 48 (9)</td>
<td>65 (10)</td>
<td>63 (9)</td>
</tr>
<tr>
<td>Hip external rotation (deg)</td>
<td>21 (14) 30 (8)</td>
<td>21 (11)</td>
<td>20 (19)</td>
</tr>
<tr>
<td>Femoral neck anteversion (deg)</td>
<td>33 (9) 22 (7)</td>
<td>35 (9)</td>
<td>32 (8)</td>
</tr>
<tr>
<td>True popliteal angle (deg)</td>
<td>39 (8) 30 (8)</td>
<td>43 (9)</td>
<td>42 (11)</td>
</tr>
<tr>
<td>Dynamic popliteal angle (deg)</td>
<td>68 (14) 57 (9)</td>
<td>71 (12)</td>
<td>71 (12)</td>
</tr>
<tr>
<td>Knee extension (deg)</td>
<td>0 (3) 1 (3)</td>
<td>0 (8)</td>
<td>-3 (10)</td>
</tr>
<tr>
<td>Dorsiflexion, knee flexed (deg)</td>
<td>12 (15) 21 (7)</td>
<td>12 (11)</td>
<td>13 (8)</td>
</tr>
<tr>
<td>Dorsiflexion, knee extended (deg)</td>
<td>-2 (17) 11 (8)</td>
<td>0 (13)</td>
<td>4 (8)</td>
</tr>
<tr>
<td>Dynamic dorsiflexion (deg)</td>
<td>-19 (14) -4 (9)</td>
<td>-14 (12) -9 (8)</td>
<td>6 (1.11)**</td>
</tr>
<tr>
<td>External tibial torsion (deg)</td>
<td>18 (6) 17 (6)</td>
<td>17 (6)</td>
<td>20 (6)</td>
</tr>
<tr>
<td>Leg press: 1 RM (kg)</td>
<td>116 (33) 110 (40)</td>
<td>109 (61)</td>
<td>116 (53)</td>
</tr>
<tr>
<td>Quadriceps: 1 RM (kg)</td>
<td>14.6 (6.9) 14.6 (7.2)</td>
<td>17.6 (10.3) 17.7 (11)</td>
<td></td>
</tr>
<tr>
<td>Hip extensor strength (kg)</td>
<td>6.4 (2.3) 7.2 (3.3)</td>
<td>8.6 (3.9) 9 (3.4)</td>
<td></td>
</tr>
<tr>
<td>Hip adductor strength (kg)</td>
<td>5.5 (1.5) 6.7 (2.4)</td>
<td>5.8 (2) 6.2 (2.9)</td>
<td></td>
</tr>
<tr>
<td>Quadriceps strength (kg)</td>
<td>11.5 (2.5) 11.2 (4)</td>
<td>11.6 (3.2) 11.9 (3.3)</td>
<td></td>
</tr>
<tr>
<td>Plantar flexor strength (kg)</td>
<td>7 (2.8) 10.3 (4.5)</td>
<td>7.5 (3.1) 8.8 (3.1)</td>
<td>1.9 (0.01, 3.9)</td>
</tr>
<tr>
<td>Activity/participation</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>GMFM-66</td>
<td>65.3 (11.1) 66.1 (8.9)</td>
<td>70.3 (11.3) 69.8 (11.4)</td>
<td>0.3 (-4.5, 5.0)</td>
</tr>
<tr>
<td>Time spent in upright position (hr)</td>
<td>3.7 (1.2)†† 4.1 (0.8)††</td>
<td>5.0 (1.4) 3.9 (1.9)</td>
<td>1 (-0.5, 2.5)</td>
</tr>
<tr>
<td>Quality of life (CHQ·PF50 domain)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Physical function</td>
<td>47 (26) 58 (26)</td>
<td>62 (35)</td>
<td>76 (25)</td>
</tr>
<tr>
<td>Social/emotional</td>
<td>69 (34) 65 (36)</td>
<td>89 (21)</td>
<td>97 (8)</td>
</tr>
<tr>
<td>Family cohesion</td>
<td>72 (20) 83 (13)</td>
<td>69 (20)</td>
<td>69 (20)</td>
</tr>
</tbody>
</table>

*The values are presented as the mean and standard deviation except where otherwise noted. †GPI = Gait Profile Score, GGI = Gillette Gait Index. 1 RM = one-repetition maximum strength, GMFM-66 = Gross Motor Function Measure-66, and CHQ·PF50 = Child Health Questionnaire-Parent Form 50. §The values are presented as the median and interquartile range. †p < 0.001. ††No comparison between the groups was conducted. †*P < 0.05. ††H = 9 for the time spent in the upright position in the single-event multilevel surgery group.
with the use of a positional activity logger (Gornlan ProMed, Melbourne, Australia), attached to the child's right thigh with use of a standard protocol. The time spent in the upright position over a twenty-four hour period was calculated for each day, and then these times were averaged to obtain one value for this outcome. Patient-reported outcomes were assessed with use of the Child Health Questionnaire-Parent Form 50 (CHQ-PF50), Australian authorized adaptation (Fig. 1).

Once suitability was established, written informed consent was obtained from the parents, and assessments were completed, the randomization was performed by the trial statistician, who was unaware of the child's identity and who used a minimization approach to ensure that the groups were well matched. Random allocation was done via a computer program. Minimization was based on the GMFCS level (II or III), age (less than or older than nine years), and type of surgery (osseous only, soft tissue only, or both).

Statistical Analysis
An analysis-of-covariance comparison between the groups at twelve months and a linear regression analysis with robust standard errors for comparison of the baseline and twenty-four-month values within the surgical group were carried out for all outcome measures (except the FMS) with use of the Stata 10.0 Statistical Data Analysis Program (StataCorp, College Station, Texas). Frequency data for the change in the FMS scores are reported.

Source of Funding
In support of this research, funding was received from the Hugh Williamson Foundation, the Murdoch Childrens Research Institute, and the National Health and Medical Research Council, The Centre for Clinical Research Excellence in Clinical Gait Analysis and Gait Rehabilitation.

Results
A consecutive sample of thirty children with spastic diplegic cerebral palsy was assessed for eligibility. The parents of all nineteen children who fulfilled the inclusion criteria agreed to allow their child to participate. Eleven children were excluded from the trial because they were outside the stipulated age range or GMFCS level (four), had progressive crouch gait (one), did not require enough surgical procedures to meet the inclusion criteria (four), or had hip migration of >25% seen on radiographs (two). Eleven children were randomized into the surgical group (single-event multilevel surgery) and eight, into the control group (progressive resistance strength training only). All children completed baseline and twelve-month assessments and thus completed the randomized phase of the trial.

![MAP & GPS SEMLS Group](image)

MAP & GPS Control Group

![MAP & GPS Control Group](image)

![MAP & GPS SEMLS Group](image)

Fig. 3
Movement Analysis Profile (MAP) scores and Gait Profile Scores (GPS) for the group treated with single-event multilevel surgery (SEMLS) and the control group at baseline, twelve months, and twenty-four months (the surgical group only). RMS = root mean square, Pel Tilt = pelvic tilt, Hip Flex = hip flexion, Knee Flex = knee flexion, Ankle dorsiflexion, Pel Obi = pelvic obliquity, Hip Abd = hip abduction, Pel Rot = pelvic rotation, Hip Rot = hip rotation, and Foot Prog = foot progression.
Figure 2 shows the progression of the children throughout the study. The children's baseline characteristics, the indications for surgery, and the types of surgical procedures that were performed are summarized in tables in the Appendix. A total of eighty-five procedures were performed, with a mean of eight procedures per child (standard deviation, four). Adverse events related to surgery were classified as mild if they resolved spontaneously, moderate if they resolved completely following simple treatment, and severe if there was a permanent deficit. Three children had a total of four mild adverse events related to poor postoperative pain management. In two children, this was due to postoperative epidural pain management malfunction. One child had difficulties with pain and excessive consumption of codeine, which was followed by constipation with emesis. Three children had moderate adverse events: two had pain over femoral osteotomy plates, which resolved with implant removal, and one had foot pain following os calcis lengthening, which resolved by six months after the surgery.

The mean measurements at baseline and at twelve months for each group and the between-group-comparison results are summarized in Table I. Measurements of tibial torsion, hip extension, hip abduction, and knee extension showed no change between baseline and twelve months and no further analysis was conducted. The only strength measure indicating a real change was that of the ankle plantar flexors, and this measure was included in the between-group comparisons9.

**Results at Twelve Months: Randomized Phase**

*Table 1 and Figure 3*

The median GPS improved by 4.6° (34%) in the surgical group and demonstrated a nonsignificant trend for deterioration in the control group. The changes in the MAP and the GPS in both groups are shown in Figure 3. The mean GGI improved from 353 to 153 in the surgical group and demonstrated a nonsignificant trend for deterioration in the control group, in which it changed from 370 to 381. (A decrease in either the GPS or GGI is an improvement toward more normal gait.) Analysis of covariance showed that the differences between groups with regard to the change in the GPS (−5.5; 95% confidence interval, −7.6 to −3.4) and in the GGI (−218; 95% confidence interval, −299 to −136) were highly significant at twelve months.

The differences between the groups with regard to the changes in the measures of activity and participation (the Gross Motor Function Measure-66 [GMFM-66] score and
the time spent in the upright position over twenty-four hours) were not significant, although the trends were for improvement in the treatment group and deterioration in the control group.

A significant between-group difference in the social/emotional domain of the CHQ-PF50 was found, with a small deterioration in the surgical group and an improvement in the control group, at twelve months. The family cohesion domain showed a small, nonsignificant improvement in the surgical group. The physical functioning domain improved in both groups, but the difference between the groups was not significant. None of the between-group differences in any of the other domains of the CHQ-PF50 were significant.

In Figures 4-A and 4-B, differences between baseline and twelve-month FMS scores are displayed as the frequency of the patients having the same scores at baseline and twelve months, of their having a better score at twelve months, and of their having a worse score at twelve months.

Results at Twenty-four Months: Nonrandomized Phase (Surgical Group Only, N = 11; Table II)

The principal difference between the twelve-month and twenty-four-month outcomes, when compared with baseline data, was that the trends toward improvements in the GMFM-66 and the CHQ-PF50 physical function domain noted at twelve months had continued and were now significant. The mean increase in the GMFM-66 score from baseline was 4.9% (95% confidence interval, 0.98% to 8.7%).

The frequencies of differences between the baseline and twelve-month FMS scores and between the baseline and twenty-four-month scores in the surgical group are displayed in Figure 4-B. No child had a twenty-four-month score that was worse than the baseline score.

Discussion

The natural history of gait and function of six to twelve-year-old children with bilateral cerebral palsy is progressive deterioration. Given that the cerebral lesion is not progressive, it seems reasonable to implicate the progressive musculoskeletal pathology as the primary reason for gait deterioration.

In this pilot randomized controlled trial, the treatment group had a clinically relevant improvement in gait of 34% as determined by the GPS and of 57% as determined by the GGI. Improvements in gait parameters are expected after single-event multilevel surgery, and the treatment is directed toward improving gait function. The developers of the GGI suggested that a change of > 10% is clinically important and reported a mean improvement of 21% in a retrospective study of multilevel surgery. Although uncontrolled studies may be more prone to overestimate treatment effects, the improvement in GGI in our study is considerably better than that previously reported in retrospective studies and in prospective studies (Table III).

### Table II Results of Linear Regression Analysis of Changes from Baseline to Twenty-four Months After Surgery in the Single-Event Multilevel Surgery Group

<table>
<thead>
<tr>
<th>Measure*</th>
<th>Baseline† (N = 11)</th>
<th>24 Mo† (N = 11)</th>
<th>Difference Between Baseline and 24 Mo (95% Confidence Interval)</th>
</tr>
</thead>
<tbody>
<tr>
<td>GPS‡</td>
<td>13.7 (11.9, 15.2)</td>
<td>9.1 (7.8, 9.6)</td>
<td>-5.4 (-7.5, -3.3)§</td>
</tr>
<tr>
<td>GGI</td>
<td>353 (211)</td>
<td>139 (80)</td>
<td>-213 (-327, -100)§</td>
</tr>
<tr>
<td>GMFM-66</td>
<td>65.3 (11.1)</td>
<td>70.2 (10.1)</td>
<td>4.9 (0.98, 8.7)§</td>
</tr>
<tr>
<td>CHQ-PF50 physical function domain</td>
<td>47 (26)</td>
<td>69 (18)</td>
<td>22 (4, 39)§</td>
</tr>
</tbody>
</table>

*GPS = Gait Profile Score, GGI = Gillette Gait Index, GMFM-66 = Gross Motor Function Measure-66, and CHQ-PF50 = Child Health Questionnaire-Parent Form 50. †The values are presented as the mean and standard deviation except where otherwise noted. ‡The values are presented as the median and interquartile range. §P < 0.05.

### Table III Comparison of Gillette Gait Index Between Present Trial and Previously Reported Studies

<table>
<thead>
<tr>
<th>Study/Intervention</th>
<th>Present trial</th>
<th>Square Root of GGI</th>
<th>% Improvement in Outcome Measure*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Orthopaedic surgery</td>
<td>57</td>
<td>34</td>
<td>34</td>
</tr>
<tr>
<td>No surgery</td>
<td>-3</td>
<td>-1</td>
<td>-8</td>
</tr>
<tr>
<td>Schwartz et al.</td>
<td>21</td>
<td>11</td>
<td>21</td>
</tr>
<tr>
<td>Orthopaedic surgery</td>
<td>Selective dorsal rhizotomy 43</td>
<td>40</td>
<td>23</td>
</tr>
<tr>
<td>Orthopaedic surgery + selective dorsal rhizotomy</td>
<td>38</td>
<td>21</td>
<td></td>
</tr>
<tr>
<td>Gorton et al.</td>
<td>35</td>
<td>19</td>
<td>19</td>
</tr>
<tr>
<td>Orthopaedic surgery</td>
<td>No surgery</td>
<td>-2</td>
<td>-1</td>
</tr>
<tr>
<td>Postans et al.</td>
<td>19</td>
<td>10</td>
<td>10</td>
</tr>
<tr>
<td>Orthopaedic surgery</td>
<td>Selective dorsal rhizotomy</td>
<td>44</td>
<td>25</td>
</tr>
</tbody>
</table>

*GGI = Gillette Gait Index, and GPS = Gait Profile Score.
As a result of our small sample size, it is not possible to relate baseline gait characteristics or outcomes to the number or type of surgical procedures. A table in the Appendix shows the profile of surgical procedures performed, and the GPS and GGI measures at baseline and twelve months after the surgery, for each subject in the surgical group.

The improvements in the GMFM-66 score were not significant at twelve months, but they were at twenty-four months. The 4.9% increase in the GMFM-66 score in this study is greater than that reported in clinical trials of selective dorsal rhizotomy and multilevel orthopaedic surgery. Gait parameters improved much faster and to a much greater degree than did functional parameters after surgery. This finding supports the principle that, although there is a relationship between gait and function, that relationship is not linear.

Previous investigators have reported mixed results with regard to changes in GMFCS scores after single-event multilevel surgery. Kondo et al. reported increases in GMFM-66 scores after soft-tissue procedures in a younger cohort of children with cerebral palsy (GMFCS levels I through IV). Their study was uncontrolled and included children with GMFCS level IV, who are not expected to be capable of long-term ambulation. In a comparative study, Buckon et al. reported increases in GMFCS scores for children who had been managed with selective dorsal rhizotomy or multilevel orthopaedic surgery. However, Gorton et al. found no change in GMFM-66 scores in a multicenter, prospective, controlled trial of children who had undergone orthopaedic surgery. Seniorou et al. conducted a prospective randomized trial of muscle strengthening after multilevel surgery. They found significant deficits in muscle strength and that GMFM-66 scores had not returned to baseline at twelve months after multilevel surgery, despite a strengthening program and improvements in kinematic parameters. However, in our trial, by twenty-four months after surgery, the surgical group had shown a mean improvement of 4.9% in the GMFCS-66 scores, which is clinically and statistically significant. This improvement is better than the expected natural history and 60% greater than what was found in a meta-analysis of three randomized controlled trials of selective dorsal rhizotomy. It is also similar to findings reported by Abel et al., who evaluated changes in GMFCS scores following soft-tissue multilevel surgery in much younger patients. The magnitude and time course of the changes in GMFCS-66 scores in these studies reinforce the need for controlled trials, with stratification by GMFCS level and age. Studies that include younger children with cerebral palsy, who are still on a rising gross motor curve, should be controlled to allow for spontaneous increases in the GMFM-66 score, which may otherwise be erroneously attributed to the intervention. This finding has implications with respect to assessment timing and study duration in terms of both ethical aspects and the feasibility of randomized trials of multilevel surgery.

The relationships among gait, function, and health-related quality of life were found to be even more complex. Physical function as measured with the CHQ increased in both groups in our study at twelve months, although between-group differences were not significant. (All CHQ data showed wide confidence intervals.) There was a greater increase in the control group, which may have been a result of participation in the progressive resistance strength training program. The change in the social/emotional domain of the CHQ differed significantly between the groups at twelve months and may reflect enhanced self-esteem resulting from participation in a strengthening program as well as the stresses of multilevel surgery and rehabilitation in the surgical group. Importantly, at twenty-four months after the surgery, the improvement in the CHQ physical functioning domain in the surgical group was significant and coincided with objective measures of improved function (the GMFM-66 score).

The time spent in the upright position decreased in the control group and increased in the treatment group, but the changes were small and not significant. This may reflect the natural history of gait deterioration of children with cerebral palsy.

A specific goal of single-event multilevel surgery in our center is to reduce the level of support by an assistive device required during walking about the community. Changes in the need for assistive devices are best monitored with the FMS. Previous work has shown that the level of support increases after surgery and decreases slowly back to baseline over the course of about twelve months. This was the case in this study. By twenty-four months after the single-event multilevel surgery, all children had returned to baseline, and no children were using a wheelchair or walker for community distances (500 m) (Fig. 4-B).

The strengths of this study were the randomized controlled design, the relative homogeneity of the study population, the stratification by GMFCS level, and the utilization of reliable outcome measures across all domains of the ICF. Limitations were the small sample size, the fact that all patients were seen at a single center, a lack of blinding, and limitations in the responsiveness of some of the secondary outcome measures. Blinding is appropriate in most clinical trials, to reduce bias. In this trial, the presence of multiple surgical scars, the presence of palpable proximal femoral plates, and the changes in the hip rotation profile meant that attempted blinding would have failed. Blinding may be less relevant when the primary outcome measure is objectively measured with use of equipment and techniques that are largely immune to bias.

Appendix Tables showing baseline characteristics in both groups, the surgical indications and procedures performed in the surgical group, and the surgical procedures performed and the gait measures at baseline and twelve months for each patient in the surgical group are available with the electronic version of this article on our web site at jbjs.org (go to the article citation and click on "Supporting Data").
References


