Multilevel Surgery for Children With Cerebral Palsy: A Meta-analysis

Noor Amirah Amirmudina, Grace Lavelle, PhD,b Tim Theologis, PhD,c Nicky Thompson, PhD,c Jennifer M. Ryan, PhD,a,b

abstract

CONTEXT: Multilevel surgery (MLS) is standard care for reducing musculoskeletal disorders among children with spastic cerebral palsy (CP).

OBJECTIVE: To summarize the literature examining effects of MLS and satisfaction with MLS for children with CP.

DATA SOURCES: Medline, Embase, Cumulative Index to Nursing and Allied Health Literature, and Cochrane Central Register of Controlled Trials were searched.

STUDY SELECTION: Studies in which authors reported effects of or satisfaction with MLS in children with CP were selected.

DATA EXTRACTION: Two authors screened and extracted data on gross motor function, gait speed, gait (eg, Gait Profile Score), range of motion, strength, spasticity, participation, quality of life, satisfaction, and adverse events.

RESULTS: Seventy-four studies (3551 participants) were identified. One was a randomized controlled trial (RCT) (n = 19); the remainder were cohort studies. Pooled analysis of cohort studies revealed that MLS did not have a long-term effect on gross motor function (standardized mean difference [SMD]: 0.38; 95% confidence interval [CI]: −0.25 to 1.01) or gait speed (SMD: 0.12; 95% CI: −0.01 to 0.25) but did improve gait (SMD: −0.80; 95% CI: −0.95 to −0.65). The RCT also revealed no effect of MLS on gross motor function but improvements in the Gait Profile Score at 1 year. Participation and quality of life were reported in only 5 studies, and adverse events were adequately reported in 17 studies.

LIMITATIONS: Data were largely from cohort studies.

CONCLUSIONS: Findings reveal that gait, but not gross motor function, improves after MLS. RCTs and improved reporting of studies of MLS are required.
Cerebral palsy (CP) is characterized by abnormal fine and gross motor functioning. The incidence of CP is ∼2 to 3 per 1000 live births worldwide. Musculoskeletal disorders, including contractures of muscle-tendon units and bony deformities, are a secondary impairment of CP and contribute to restricted mobility. Multilevel surgery (MLS) followed by intensive rehabilitation is considered standard care for reducing musculoskeletal disorders among children with spastic CP. To date, the literature on MLS for children with CP has been examined in 2 systematic reviews. The first, conducted in 2010, included 31 studies, and the authors concluded that there was a trend toward improvements in passive range of motion, gait kinematics and kinetics, and gait efficiency but little evidence for improvements in gross motor function or quality of life (QoL). However, study quality was variable, and a meta-analysis of data was not performed. The authors also identified variability in the quality of reporting of surgical procedures, rehabilitation, and adverse events. In the second review, the authors examined the effect of MLS specifically on gait parameters. The authors concluded from a narrative synthesis that there was a trend toward improvements in gait parameters but reported variability in study quality.

The World Health Organization’s International Classification of Functioning, Disability, and Health (ICF) framework is useful for considering the impact of CP on the individual. Developed in 2001, the ICF is used to classify health-related functions and structure, activity, and participation. The ICF replaced the International Classification of Impairments, Disabilities, and Handicaps (ICIDH), which was used to conceptualize a health condition leading to impairment, disability, and handicap in a linear manner. The ICF places greater emphasis on the role of the social and physical environment on functioning and conceptualizes that functioning results from a complex interaction between the person with the health condition and his or her context (consisting of personal and environmental factors). When developed, it was envisaged that 1 use of the ICF would be the evaluation of interventions. However, the review in 2010 revealed that authors of few studies evaluate the effect of MLS across multiple domains of the ICF.

Given the impact of MLS on children with CP and their families, a summary of the current literature on MLS and a systematic evaluation of the effects of MLS are required. Thus, our aim for this review was to summarize the current literature on MLS for children with CP, including the effects of MLS and satisfaction with outcomes after MLS. We also aimed to examine the use of the ICF and ICIDH by authors when reporting outcomes of MLS.

METHODS

Study Selection Criteria

We included studies meeting the following criteria: (1) included children with CP (we chose to define a child as a person aged 0 to 20 years to be inclusive of all studies in which MLS in children was evaluated because we expected variation in the age range across studies); (2) reported outcomes before and after MLS or reported satisfaction with MLS; and (3) additional interventions, such as Botulinum toxin injections, were not performed simultaneously to surgery. We defined MLS as “2 or more soft-tissue or bony surgical procedures at 2 or more anatomical levels.” Original peer-reviewed articles published in English were included. To provide a comprehensive overview of the current literature, we did not limit studies to randomized controlled trials (RCTs). However, qualitative studies, reviews, commentaries, conference abstracts, and case reports were excluded.

Search Strategy

We searched Ovid Medline, Embase Ovid, Cumulative Index to Nursing and Allied Health Literature, and Cochrane Central Register of Controlled Trials from January 2000 to June 2018. Search terms relating to children, CP, and surgery were combined. Each search was adapted for the respective database (see Supplemental Information). Additionally, reference lists of previous reviews were searched.

Two investigators screened titles and abstracts independently against eligibility criteria. When articles met eligibility criteria or when there was doubt over inclusion, full texts were obtained. Any disagreement regarding the inclusion of an article was resolved through discussion.

Data Extraction

Two authors (N.A.A. and J.M.R.) extracted data independently using a standardized piloted data extraction form. Data on study design, study population (eg, Gross Motor Function Classification System [GMFCS] level), intervention (eg, procedures performed), rehabilitation received, duration of follow-up, adverse events, and outcomes were extracted. We noted if outcomes were reported according to ICIDH or ICF frameworks in studies.

We extracted data on gross motor function as measured by the Gross Motor Function Measure 66 (GMFM-66) or Gross Motor Function Measure 88, the Functional Mobility Scale (FMS), and the Gillette Functional Assessment Questionnaire (FAQ). Although
gros motor functionwas assessed in some studies by using the GMFCS, we did not consider the GMFCS to be a measure of gross motor function because it is primarily a classification system. We extracted data on gait speed and summary statistics of gait (ie, the Gait Deviation Index, Gait Profile Score [GPS], or Gillette Gait Index)\textsuperscript{10–12}. When \textgreater{}\textasciitilde{}1 summary score was reported in studies, we extracted data on the GPS. We chose to extract data on gait summary scores rather than individual kinematic or kinetic variables because authors of many studies reported a large number of variables obtained from three-dimensional gait analysis. In addition, we extracted data on participation, QoL, and satisfaction with surgery. Since the publication of the ICF, there has been a lack of consensus regarding measurement tools to assess participation. For the purpose of this review, we extracted data on measures that mapped to the recently developed family of participation-related constructs.\textsuperscript{13} Finally, we extracted data on passive range of motion, muscle strength, and spasticity because these are commonly evaluated before and after MLS as part of a clinical examination and are used to inform clinical decision-making on surgery. We extracted data on passive knee extension and passive dorsiflexion only because these were the most commonly reported joints. Because authors typically reported muscle strength and spasticity for \textgreater{}\textasciitilde{}1 muscle group, we ranked muscle groups in order of frequency of reporting across all included studies. For each study, we then extracted data for strength and spasticity, respectively, for the most frequently reported muscle group. If data were not reported for the most frequently reported muscle group, we extracted data for the next most frequently reported muscle group, and so on. If data were reported on both limbs, data on the most affected limb or the right limb were extracted. Data on short-term (\textless{}6 months postsurgery), intermediate- (6 months to 1 year postsurgery), and long-term (>1 year postsurgery) effects were extracted. If \textgreater{}1 assessment was performed during any of these time periods, the last measurement taken during that period was extracted. When 2 groups that received MLS were compared in studies, we extracted pre- and postsurgery data for the whole sample, if available, or pre- and postsurgery data for the 2 groups separately. In these cases, we report results according to the number of groups included in the meta-analysis.

**Quality Assessment**

The Methodological Index for Nonrandomized Studies (MINORS) checklist was used to assess the quality of studies on the basis of 8 or 12 items\textsuperscript{14} (items related to the description of the aim, selection, prospective data collection and sample size calculation, appropriateness of the follow-up period, and loss to follow-up). In addition, the adequacy of reporting of surgical procedures, previous surgery, adverse events, and rehabilitation was rated as 0 (not reported), 1 (reported but inadequate), or 2 (reported adequately).

**Data Analysis**

When sufficient data were obtained, we conducted separate meta-analyses for each outcome at each time point.

Weighted effect sizes were calculated by using the inverse-variance method. A random effects model was used because there was large variation between studies in terms of procedures, populations, and context.\textsuperscript{15} Effect sizes were calculated as Hedges’ $g^*$ to correct for bias associated with a small sample size.\textsuperscript{16} The SD for the change score (SD\textsubscript{change}) (ie, mean difference [MD] between baseline and follow-up values) was used to calculate Hedges’ $g^*$. When this was not provided, SD\textsubscript{change} was estimated by using statistics provided (eg, $t$ values, confidence intervals [CIs], SEs, and $P$ values) or by using SDs for the baseline and follow-up score and the correlation between baseline and follow-up scores ($r$). When $r$ was not provided, we estimated $r$ using either individual data or using SDs for the baseline and follow-up scores and SD\textsubscript{change}. If this was not possible, we imputed $r$ as 0.7 for the analysis of short-term effect on gross motor function and imputed $r$ as 0.3 for all other analyses on the basis of the strength of correlations observed in studies with similar samples, outcome measures, and follow-up periods.\textsuperscript{17} However, sensitivity analyses were performed by imputing $r$ as 0.1, 0.3, and 0.7 to examine the robustness of the findings to this assumption.\textsuperscript{17} Finally, the SE of Hedges’ $g^*$ was calculated by using the provided estimated or imputed $r$. We conducted a subgroup analysis of studies that included children with bilateral CP only. We assessed statistical heterogeneity and its impact using the $\chi^2$ test and the $I^2$ statistic.\textsuperscript{18} To aid interpretation, we proposed an effect size of 0.2, 0.5, and 0.8 to represent a small, moderate, and large effect, respectively,\textsuperscript{19} although this was used cautiously.

**RESULTS**

There were 2364 articles identified in database searches; 2 articles were obtained from reviewing reference lists (Fig 1). After removal of duplicates, 1823 titles and abstracts were screened. We obtained 164 full-text articles. Of these, 75 articles in which findings from 74 studies were reported were included. A description of each study is presented in Table 1. MINORS score ranged from 4\textsuperscript{20} to 21\textsuperscript{15} (Supplemental Table 3).
Study Design

We identified 1 RCT in which MLS was compared with progressive resistance training at 1 year. The authors of this study also reported outcomes for the MLS group at 2 years. Fifty-two studies were retrospective cohort studies. In studies that included a comparison group, results between male and female patients, between participants with diplegia and hemiplegia, between participants with more or less than a 5-degree increase in anterior pelvic tilt or between patients who received different surgical procedures as part of MLS were compared.

Seventeen studies were prospective cohort studies, of which a single-subject AB design was used in 1 and 2 surgical procedures as part of MLS were compared in 2. Four studies were RCTs in which 2 surgical procedures as part of MLS or 2 types of rehabilitation after MLS were compared. We treated these as cohort studies in pooled analyses because they did not include comparisons of MLS with controls.

Participants

In total, 3551 participants were included in 74 studies. Sample sizes ranged from 7 to 314. We considered 58 studies (78%) that were small (n < 50), 8 that were medium (n = 50–100), and 8 that were large (n > 100). Participants were 3 to 20 years of age at the time of surgery (mean age 6.0–14.0 years). When sex was reported, the percentage of male patients ranged from 20% to 100%; overall 62% were male patients.

When type was reported (76% of included studies), all participants had spastic CP. Anatomic distribution was reported in 86% of studies. Two studies included children with unilateral CP only, and 12 studies included children with unilateral and bilateral CP. The remaining studies included children with bilateral CP only.

Forty-seven studies included children in GMFCS levels I to III. One study included children in level IV only, and the remainder included participants in levels I to IV, I to V, or II to IV. GMFCS level was not reported in 17 studies; all children were described as ambulatory with or without a walking aid except for those in the study by Khan, who reported all participants were nonambulatory.

Description of Surgery and Rehabilitation

Description of surgery was rated adequate in 48 studies (65%). Soft-tissue procedures only were performed in 7 studies. Bony- and soft-tissue procedures were performed in 64 studies. The authors of 3 studies did not state the type of procedures performed. When reported, the number of procedures conducted per participant ranged from 2 to 18; the mean per patient was 2.2 to 12.9. Provision of previous surgery was adequately reported in 33 studies.

The authors of 19 studies (26%) did not report any information regarding rehabilitation provided after surgery. An inadequate description of rehabilitation was provided in 25 studies (34%). The authors of 30 studies (40%) reported the frequency and/or duration of rehabilitation provided. When reported, the content of rehabilitation included active and passive movements, strengthening
<table>
<thead>
<tr>
<th>Author</th>
<th>Design</th>
<th>Participants</th>
<th>Surgery Type</th>
<th>Procedure per Patient, Mean ± SD (Range)</th>
<th>Duration of Follow-up, y</th>
</tr>
</thead>
<tbody>
<tr>
<td>Adolfsen et al</td>
<td>Retrospective</td>
<td>31 (NS)</td>
<td>Soft tissue</td>
<td>4.4</td>
<td>Mean (range): 1.9 (0.7–6.4)</td>
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<tr>
<td>Akerstedt et al</td>
<td>Prospective single subject</td>
<td>11 (10, 1)</td>
<td>Mixed</td>
<td>NS</td>
<td>2</td>
</tr>
<tr>
<td>Ancillao et al</td>
<td>Unclear</td>
<td>9 (7, 2)</td>
<td>Mixed</td>
<td>NS</td>
<td>Mean ± SD: 1.2 ± 0.4</td>
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<td>Bernthal et al</td>
<td>Prospective</td>
<td>23 (NS)</td>
<td>Soft tissue</td>
<td>4.2 (2–8)</td>
<td>1</td>
</tr>
<tr>
<td>Blumetti et al</td>
<td>Retrospective (RFT, non-RFT)</td>
<td>216 (NS)</td>
<td>Mean ± SD: 3.4 ± 2.7; non-DFT: 3.4 ± 2.7</td>
<td>20 ± 0.2</td>
<td></td>
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<tr>
<td>Böhm et al</td>
<td>Retrospective (PT, PN)</td>
<td>32 (NS)</td>
<td>Mixed</td>
<td>NS</td>
<td>2</td>
</tr>
<tr>
<td>Braitz et al</td>
<td>Prospective</td>
<td>30 (18, 12)</td>
<td>Mixed</td>
<td>6.4</td>
<td>Mean ± SD: 1.1 ± 0.2</td>
</tr>
<tr>
<td>Buckon et al</td>
<td>Prospective</td>
<td>25 (20, 5)</td>
<td>Mixed</td>
<td>NS</td>
<td>0.5</td>
</tr>
<tr>
<td>Chang et al</td>
<td>Prospective</td>
<td>57 (NS)</td>
<td>Soft tissue</td>
<td>NS</td>
<td>Mean (range): 1.3 (0.8–2.5)</td>
</tr>
<tr>
<td>Desai et al</td>
<td>Retrospective (PTS, no PTS)</td>
<td>41 (NS)</td>
<td>Mixed</td>
<td>NS</td>
<td>Mean ± SD: 2.3 ± 0.8</td>
</tr>
<tr>
<td>Dreher et al</td>
<td>Prospective</td>
<td>30 (22, 8)</td>
<td>Mixed</td>
<td>10.5</td>
<td>Mean ± SD: 11.3 ± 0.7</td>
</tr>
<tr>
<td>Dreher et al</td>
<td>Prospective (RFT, non-RFT)</td>
<td>32 (19, 13)</td>
<td>Mixed</td>
<td>7.1</td>
<td>Mean ± SD: 10.6 ± 0.6; non-DFT: 12 ± 0.2</td>
</tr>
<tr>
<td>Dreher et al</td>
<td>Retrospective</td>
<td>39 (26, 13)</td>
<td>Mixed</td>
<td>10.1</td>
<td>Mean ± SD: 8.1 ± 1.8</td>
</tr>
<tr>
<td>Dreher et al</td>
<td>Retrospective (C-DRFT, P-DRFT)</td>
<td>53 (56, 17)</td>
<td>Mixed</td>
<td>8.8 ± 0.8</td>
<td>Mean ± SD: 8.8 ± 2.3</td>
</tr>
<tr>
<td>Dreher et al</td>
<td>Retrospective</td>
<td>44 (26, 18)</td>
<td>Mixed</td>
<td>8.6 ± 0.6</td>
<td>Mean ± SD: 8.6 ± 2</td>
</tr>
<tr>
<td>Dreher et al</td>
<td>Prospective (CBM, MTL)</td>
<td>42 (28, 14)</td>
<td>Mixed</td>
<td>12.9</td>
<td>Mean ± SD: CBM: 9.2 ± 2.5; MTL: 9.1 ± 2.6</td>
</tr>
<tr>
<td>Dreher et al</td>
<td>Retrospective</td>
<td>42 (27, 15)</td>
<td>Mixed</td>
<td>12.9</td>
<td>Mean ± SD: 3.2 ± 2.6</td>
</tr>
<tr>
<td>Dreher et al</td>
<td>Retrospective</td>
<td>231 (142, 89)</td>
<td>Mixed</td>
<td>8 ± 3</td>
<td>Mean ± SD: 9.1 ± 3.0</td>
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<tr>
<td>Feger et al</td>
<td>Retrospective (O, MTL)</td>
<td>48 (27, 21)</td>
<td>Mixed</td>
<td>0.30 ± 1.5 (1–5); MTL: 2.2 ± 1.3 (1–4)</td>
<td>Mean ± SD (range): 0.17 ± 0.6 (1.0–2.9); MTL: 19 ± 0.6 (10–30)</td>
</tr>
<tr>
<td>Firth et al</td>
<td>Retrospective</td>
<td>40 (25, 15)</td>
<td>Mixed</td>
<td>9.1 (5–18)</td>
<td>Minimum: 4</td>
</tr>
<tr>
<td>Gannotti et al</td>
<td>Retrospective</td>
<td>20 (15, 1)</td>
<td>Mixed</td>
<td>3.6</td>
<td>Mean (range): 7.5 (4.4–14.6)</td>
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<tr>
<td>Gannotti et al</td>
<td>Retrospective</td>
<td>11 (5, 6)</td>
<td>Mixed</td>
<td>NS</td>
<td>Mean (range): 13 (11–15)</td>
</tr>
<tr>
<td>Author</td>
<td>Design</td>
<td>Participants</td>
<td>Surgery Type</td>
<td>Procedure per Patient, Mean ± SD (Range)</td>
<td>Duration of Follow-up, y</td>
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<tr>
<td>Godwin et al43</td>
<td>Retrospective</td>
<td>84 (35, 49)</td>
<td>5.99 ± 3.3 (5.5–15.3)</td>
<td>Diplegia (51), hemiplegia (21), quadriplegia (27)</td>
<td>5</td>
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<tr>
<td>Gough et al44</td>
<td>Retrospective</td>
<td>12 (8, 4)</td>
<td>6.4 (5.5–8.9)</td>
<td>Diplegia (100)</td>
<td>Mixed</td>
</tr>
<tr>
<td>Gough et al45</td>
<td>Retrospective</td>
<td>19 (13, 6)</td>
<td>12.1 ± 3.1 (6–16)</td>
<td>Diplegia (100)</td>
<td>Mixed</td>
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<tr>
<td>Graham et al46</td>
<td>Prospective</td>
<td>17 (14, 3)</td>
<td>10 ± 2.5 (6–16)</td>
<td>Diplegia (100)</td>
<td>Mixed</td>
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<tr>
<td>Harvey et al47</td>
<td>Retrospective</td>
<td>53 (191, 88)</td>
<td>8.0 ± 3.1 (5.6–18.2)</td>
<td>Diplegia (64), T (5), hemiplegia (18), quadriplegia (14)</td>
<td>Mixed</td>
</tr>
<tr>
<td>Khan49</td>
<td>Prospective</td>
<td>85 (53, 32)</td>
<td>8.5 (5–12)</td>
<td>Diplegia (100)</td>
<td>Mixed</td>
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<tr>
<td>Lofterød and Terjesen59</td>
<td>Retrospective</td>
<td>28 (16, 12)</td>
<td>12 ± 3 (7–19)</td>
<td>Diplegia (100)</td>
<td>Mixed</td>
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<tr>
<td>Metaxiotis et al61</td>
<td>Retrospective</td>
<td>20 (13, 7)</td>
<td>11.5 (5.6–17.0)</td>
<td>Diplegia (100)</td>
<td>Mixed</td>
</tr>
<tr>
<td>Ng et al62</td>
<td>Retrospective</td>
<td>20 (30, 18)</td>
<td>15 ± 3.1 (5.6–18.2)</td>
<td>Diplegia (100)</td>
<td>Mixed</td>
</tr>
<tr>
<td>Ounpuu et al63</td>
<td>Prospective</td>
<td>20 (NS)</td>
<td>8.1 ± 2.9 (5–15)</td>
<td>Diplegia (64), T (5), hemiplegia (18), quadriplegia (14)</td>
<td>Mixed</td>
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<tr>
<td>Ounpuu et al64</td>
<td>Retrospective</td>
<td>22 (11, 11)</td>
<td>8.0 ± 2.7</td>
<td>Diplegia (64), T (5), hemiplegia (18), quadriplegia (14)</td>
<td>Mixed</td>
</tr>
<tr>
<td>Author</td>
<td>Design</td>
<td>n (Male Patients, Female Patients)</td>
<td>Age, Mean ± SD (Range), y</td>
<td>Distribution (%)</td>
<td>GMFCS Level (%)</td>
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<td>------------------------</td>
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<tr>
<td>Patikas et al&lt;sup&gt;5&lt;/sup&gt;</td>
<td>Prospective (EG, CG)</td>
<td>39 (27, 12)</td>
<td>10.6 ± 3.2; C: 8.9 ± 1.9 (6–16)</td>
<td>Diplegia (100)</td>
<td>I (31), II (46), III (23)</td>
</tr>
<tr>
<td>Patikas et al&lt;sup&gt;6&lt;/sup&gt;</td>
<td>Prospective</td>
<td>34 (22, 12)</td>
<td>10.1 ± 3.0 (6–16)</td>
<td>Diplegia (53), hemiplegia</td>
<td>NS</td>
</tr>
<tr>
<td>Rodda et al&lt;sup&gt;8&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>10 (7, 3)</td>
<td>12.0 (7.9–16.2)</td>
<td>Diplegia (100)</td>
<td>II (30), III (70)</td>
</tr>
<tr>
<td>Rutz et al&lt;sup&gt;9&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>107 (61, 48)</td>
<td>10.8 ± 2.7 (6–17)</td>
<td>Bilateral (100)</td>
<td>II (69), III (51)</td>
</tr>
<tr>
<td>Rutz et al&lt;sup&gt;10&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>121 (73, 48)</td>
<td>10.7 ± 2.7</td>
<td>Bilateral (100)</td>
<td>II (68), III (54)</td>
</tr>
<tr>
<td>Rutz et al&lt;sup&gt;11&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>14 (10, 4)</td>
<td>13 (7–18)</td>
<td>Diplegia (100)</td>
<td>I (7), II (71), III (21)</td>
</tr>
<tr>
<td>Saraph et al&lt;sup&gt;12&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>22 (NS)</td>
<td>12.6 (7.4–16.6)</td>
<td>Diplegia (100)</td>
<td>NS</td>
</tr>
<tr>
<td>Saraph et al&lt;sup&gt;13&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>12 (NS)</td>
<td>12.7 ± 3.3</td>
<td>Diplegia (100)</td>
<td>NS</td>
</tr>
<tr>
<td>Saraph et al&lt;sup&gt;14&lt;/sup&gt;</td>
<td>Retrospective (diplegia, hemiplegia)</td>
<td>22 (NS)</td>
<td>11.9 (9.2–15.5)</td>
<td>Diplegia (38), hemiplegia</td>
<td>NS</td>
</tr>
<tr>
<td>Saraph et al&lt;sup&gt;15&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>25 (NS)</td>
<td>13.6 (6.0–15.5)</td>
<td>Diplegia (100)</td>
<td>NS</td>
</tr>
<tr>
<td>Saraph et al&lt;sup&gt;16&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>32 (NS)</td>
<td>11.1 (8.7–13.3)</td>
<td>Diplegia (100)</td>
<td>NS</td>
</tr>
<tr>
<td>Saraph et al&lt;sup&gt;17&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>11 (NS)</td>
<td>12.4 (9.5–17.2)</td>
<td>Diplegia (27), hemiplegia</td>
<td>NS</td>
</tr>
<tr>
<td>Schranz et al&lt;sup&gt;18&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>14 (9, 5)</td>
<td>12.1 ± 3.3 (6–17)</td>
<td>Unilateral (100)</td>
<td>I (45), II (57)</td>
</tr>
<tr>
<td>Schwartz et al&lt;sup&gt;19&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>176 (99, 77)</td>
<td>100 ± 3.4</td>
<td>NS</td>
<td>I (14), II (48), III (80)</td>
</tr>
<tr>
<td>Seniorou et al&lt;sup&gt;20&lt;/sup&gt;</td>
<td>Prospective (EG, CG)</td>
<td>20 (10, 10)</td>
<td>12.5 ± 2.5 (7–16)</td>
<td>Diplegia (100)</td>
<td>I (15), II (65), III (20)</td>
</tr>
<tr>
<td>Steinwender et al&lt;sup&gt;21&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>16 (NS)</td>
<td>10.2 (6–14)</td>
<td>Diplegia (100)</td>
<td>NS</td>
</tr>
<tr>
<td>Stephan-Carlier et al&lt;sup&gt;22&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>12 (10, 2)</td>
<td>14.0 ± 2.5 (11–18)</td>
<td>Diplegia (83), hemiplegia</td>
<td>I (25), II (25), III (60)</td>
</tr>
<tr>
<td>Sung et al&lt;sup&gt;23&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>29 (18, 11)</td>
<td>8.3 ± 2.6 (5.4–16.3)</td>
<td>Diplegia (100)</td>
<td>I (24), II (66), III (10)</td>
</tr>
<tr>
<td>Sung et al&lt;sup&gt;24&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>314 (198, 116)</td>
<td>7.9 ± 3.7 (3.4–20.0)</td>
<td>Diplegia (100)</td>
<td>I–III</td>
</tr>
<tr>
<td>Svehlik et al&lt;sup&gt;25&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>32 (17, 15)</td>
<td>10.3 ± 3.1 (5.7–15.5)</td>
<td>Bilateral (100)</td>
<td>II (58), III (62)</td>
</tr>
<tr>
<td>Svehlik et al&lt;sup&gt;26&lt;/sup&gt;</td>
<td>Retrospective</td>
<td>39 (22, 17)</td>
<td>10.3 ± 3.1 (5.7–15.5)</td>
<td>Bilateral (100)</td>
<td>II (51), III (49)</td>
</tr>
<tr>
<td>Taylor et al&lt;sup&gt;27&lt;/sup&gt;</td>
<td>Retrospective (DFEO, PKC)</td>
<td>31 (NS)</td>
<td>13 (7–18)</td>
<td>Diplegia (87), hemiplegia</td>
<td>I (6), II (52), III (58), IV (53)</td>
</tr>
<tr>
<td>Thomason et al&lt;sup&gt;28&lt;/sup&gt;</td>
<td>RCT</td>
<td>19 (12, 7)</td>
<td>9.7 (6–12)</td>
<td>Diplegia (100)</td>
<td>II–III</td>
</tr>
<tr>
<td>Thomason et al&lt;sup&gt;29&lt;/sup&gt;</td>
<td>Prospective</td>
<td>18 (11, 7)</td>
<td>10.2 ± 1.7 (7–17)</td>
<td>Bilateral (100)</td>
<td>II (72), III (28)</td>
</tr>
<tr>
<td>Thompson et al&lt;sup&gt;30&lt;/sup&gt;</td>
<td>Prospective (MI-SFMLS, SEMLS)</td>
<td>20 (12, 8)</td>
<td>11.0 ± 1.9 (7.8–14.3)</td>
<td>Diplegia (100)</td>
<td>I (15), II (65), III (20)</td>
</tr>
<tr>
<td>Truong et al&lt;sup&gt;31&lt;/sup&gt;</td>
<td>Retrospective (control, postas)</td>
<td>87 (NS)</td>
<td>NS (3–8)</td>
<td>NS</td>
<td>I and IV (86), III and IV (54)</td>
</tr>
</tbody>
</table>
exercises, stretching, balance, and gait training.

Outcome Assessment

The duration of follow-up varied widely between studies. When described, the mean duration of follow-up ranged from 1.089 to 21.3 years.84 We extracted data on short-term outcomes from 3 studies,27,28,79 on intermediate-term outcomes from 26 studies, and on long-term outcomes from 45 studies. Outcomes in terms of the ICIDH framework were described in no studies. Authors of 7 studies described outcomes in terms of the ICF framework, but there was inconsistency in the measures used to assess each domain.9,22,27,41,47,48,87 Authors reported assessing the collective domain of "activity and participation," using the PedsQL, Pediatric Evaluation of Disability Inventory (PEDI),27 time spent in upright positions measured with an activity logger,9 and self-reported walking ability.22

Authors of 15 studies assessed gross motor function; the GMFM-66 or Gross Motor Function Measure was used in 10 studies,9,22,27,28,39,79,86–89,93 the FAQ was used in 3 studies,29,56,89 and the FMS was used in 2 studies.47,48 Gait speed was assessed in 29 studies, and gait was assessed by using kinematic data in 35 studies.

Participation was assessed in 4 studies,27,29,56,78 QoL was assessed in 3 studies,9,22,29 and satisfaction with surgery was reported in 4 studies.22,56,57,81 Passive range of motion (knee flexion [n = 17] and ankle dorsiflexion [n = 15]) and spasticity were assessed in 22 studies, muscle strength was assessed in 14 studies, and satisfaction with surgery was reported in 4 studies.27,29,56,78 QoL was assessed in 3 studies,27,29,56,78 and satisfaction with surgery was reported in 4 studies.27,29,56,78 Passive range of motion (knee flexion [n = 17] and ankle dorsiflexion [n = 15]) and spasticity were assessed in 22 studies, muscle strength was assessed in 14 studies, and satisfaction with surgery was reported in 4 studies.27,29,56,78 QoL was assessed in 3 studies,27,29,56,78 and satisfaction with surgery was reported in 4 studies.27,29,56,78

TABLE 1

<table>
<thead>
<tr>
<th>Author</th>
<th>Designa</th>
<th>Participants (n (Male Patients, Female Patients))</th>
<th>Age, Mean ± SD (Range), y</th>
<th>Distribution (%)</th>
<th>GMFCS Level (%)</th>
<th>Surgery Typeb</th>
<th>Procedure per Patient, Mean ± SD (Range)</th>
<th>Duration of Follow-up, y</th>
</tr>
</thead>
<tbody>
<tr>
<td>Van Drongelen et al90</td>
<td>Retrospective</td>
<td>14 (8, 6)</td>
<td>9.3 ± 2.0 (6.6–12.0)</td>
<td>Diplegia (100)</td>
<td>I (14), II (64), III (21)</td>
<td>Mixed</td>
<td>NS</td>
<td>Mean (range): 2.0 (1.3–3.2)</td>
</tr>
<tr>
<td>Zwick et al91</td>
<td>Retrospective</td>
<td>17 (NS)</td>
<td>11.2 (5.7–16.4)</td>
<td>Diplegia (100)</td>
<td>NS</td>
<td>Mixed</td>
<td>NS</td>
<td>Mean (range): 3.8 (2.6–5.7)</td>
</tr>
<tr>
<td>Zwick et al92 (male patients, female patients)</td>
<td>Retrospective</td>
<td>34 (19, 15)</td>
<td>9.9 ± 3.2; female patients: 11.3 ± 2.8</td>
<td>(\text{GMFCS I} = 100)</td>
<td>(\text{II–III})</td>
<td>Mixed</td>
<td>Male patients: 4.09 ± 2.85; female patients: 3.92 ± 2.38</td>
<td>Minimum: 10</td>
</tr>
</tbody>
</table>

CBM, conversion of biarticular muscles; CG, control group; C-DRFT, distal rectus femoris transfer to correct decreased peak knee flexion in swing phase; DFEO, distal femoral extension osteotomy; DRFT, distal rectus femoris transfer; EG, exercise group; FDO, femoral derotation osteotomy; IMPL, intramuscular psoas lengthening; MI-SEMLS, minimally invasive single-event multilevel surgery; MTL, muscle-tendon lengthening; NS, not stated; O, osteotomy with or without muscle-tendon procedure; P-DRFT, prophylactic distal rectus femoris transfer; PKC, posterior knee capsulotomy; PN, no anterior pelvic tilt group; PT, anterior pelvic tilt group; PTS, patellar tendon shortening; RFT, rectus femoris transfer; SEMLS, single-event multilevel surgery — not applicable.

a Comparison groups are in parentheses if applicable.
b Mixed: soft-tissue and bony surgery.
c The group that had C-DRFT and the group that had P-DRFT were compared.
d The O group was compared with the group that had muscle-tendon-procedure only (MTL).
e Patients treated before year 2000 (group 1) were compared with patients treated after 2000 (group 2).
f Patients who had psoas lengthening as part of MLS (psoas) and patients who did not have psoas lengthening as part of MLS (control) were compared.
removal for pain, superficial wound infection, nerve palsy, avascular necrosis, permanent reflex sympathetic dystrophy syndrome, and occurrence of genu recurvatum. Authors of 3 studies reported adverse events according to the Clavien-Dindo classification.38,53,77 In these studies, 68 grade I, 52 grade II, 9 grade III, and 1 grade IV complications were reported among 265 children.

Effects of MLS

Data from 54 studies were included in the meta-analysis; data from 1 RCT are reported separately.87 Results from a further 8 studies are reported descriptively because insufficient information was provided to include them in the meta-analysis.29,47,48,51,56,57,81,85 Data on 12 studies are not reported because authors of 8 studies assessed individual gait variables or GMFCS level20,31,43,44,52,54,59,62 and authors of 4 studies did not report results from statistical analyses to allow interpretation of findings.22,40,49,70

Gross Motor Function

One RCT of 19 children revealed that gross motor function, as measured by the GMFMM-66, did not differ between children receiving MLS and children receiving resistance training at 1 year postsurgery (MD between groups: 0.3; 95% CI: −4.5 to 5.0). However, at 2 years postsurgery, the children receiving MLS demonstrated an improvement in the GMFMM-66 score from baseline (MD: 4.9; 95% CI: 0.98 to 8.7). Pooled analysis indicated that gross motor function did not improve in the short-term (standardized mean difference [SMD]: 0.01; 95% CI: −0.48 to 0.50; P = .97; I² = 92%; P < .001; n = 52), intermediate- (SMD: 0.51; 95% CI: −0.56 to 1.58; P = .35; I² = 97%; P < .001; n = 104), or long-term (SMD: 0.38; 95% CI: −0.25 to 1.01; P = .24; n = 103; Fig 2). Authors of 2 studies (n = 118) reported that the FAQ score improved by a mean of 1.06 (<.001) and 0.5 (P = .002), respectively,29,56 and the authors of 1 study (n = 75) reported no change in the FAQ score (P > .05) in the intermediate-term. The authors of 1 study (n = 156) reported that 5 years after MLS, 23%, 18%, and 19% of participants required less assistance at 5, 50, and 500 m, respectively.48 A second study (n = 66) revealed evidence of a poorer rating on the FMS at 6 months compared with presurgery at all distances.47 This study revealed no evidence of change in the FMS at 12 months but revealed evidence of an improvement in rating at 24 months at all distances.

Gait Speed

The authors of 1 study assessed the short-term change in normalized walking speed after surgery and found that it declined (MD: −0.04; P = .002; n = 20).79 There was no evidence that walking speed changed in the intermediate- (SMD: −0.08; 95% CI: −0.22 to 0.07; P = .30; I² = 43%; P = .02; n = 481) or long-term (SMD: 0.12; 95% CI: −0.01 to 0.25; P = .08; Fig 3).

Gait

The GPS improved in children who received MLS compared with those who received resistance training at 1 year (MD between groups: −5.5; 95% CI: −7.8 to −3.4; n = 19). Pooled analysis revealed that gait improved at 1 year after MLS (SMD: −0.78; 95% CI: −0.98 to −0.57; P < .001; I² = 70%; P < .001; n = 497). There was also improvement in gait in the long-term (SMD: −0.80; 95% CI: −0.95 to −0.65; P < .001; Fig 4). The authors of 1 study (not included in the pooled analysis) reported improvements in the Gait Deviation Index in the intermediate- (MD: 12.1; P < .001; n = 39) and long-term (MD: 10.3; P < .001; n = 39)85

Passive Range of Motion

Passive ankle dorsiflexion was greater among children who received MLS compared with those who received resistance training at 1 year (MD between groups: 7; 95% CI: 3 to 12; P < .001; n = 19). Knee extension declined in both groups.

FIGURE 2

Long-term effect of MLS on gross motor function. df, degrees of freedom; DFEO, distal femoral extension osteotomy; IV, inverse variance; MTL, muscle-tendon lengthening; O, osteotomy with or without muscle-tendon procedure; PKC, posterior knee capsulotomy.
(MD: –1° for the MLS group; MD: –3° for the control group), but a comparison between groups was not conducted.

There was evidence of intermediate-term improvements in knee extension (SMD: 0.60; 95% CI: 0.43 to 0.77; P < .001; I² = 0%; n = 228) and dorsi flexion (SMD: 0.48; 95% CI: 0.10 to 0.86; P = .01; I² = 49%; P = .12; n = 96) after MLS. Long-term improvements in knee extension (SMD: 0.43; 95% CI: 0.24 to 0.61; P < .001; Fig 5) and dorsiflexion (SMD: 0.47; 95% CI: 0.17 to 0.76; P = .002; I² = 76%; P < .001; n = 324) were also observed. The authors of 1 study (not included in the pooled analysis) reported an improvement in dorsiflexion in the intermediate-term (P < .001; n = 19).51

**Strength**

Plantar flexor strength improved by a larger amount in children after MLS compared with resistance training at 1 year (MD between groups: 1.9 kg; 95% CI: 0.01 to 3.9 kg; n = 19).9 Although quadriceps strength, hip extensor strength, and hip abductor strength were also measured in the RCT, comparisons between groups were not conducted.

One study (n = 20) revealed evidence of a decline in strength of the hip flexors (MD: –0.2 Nm/kg; P < .001), hip extensors (MD: –0.5 Nm/kg; P < .001), hip abductors (MD: –0.1 Nm/kg; P = .002), knee flexors (MD: –0.6 Nm/kg; P < .001), and knee extensors (MD: –0.5 Nm/kg; P < .001) at 6 months postsurgery.79 In the second study, an increase in general muscle strength was reported at 6 months postsurgery (median: 2 vs 3 on a scale of 0–4; P = .020; n = 25).28 Pooled analysis revealed no evidence for change in muscle strength in the intermediate-term (SMD: –0.22; 95% CI: –0.50 to 0.06; P = .12; I² = 51%; P = .08; n = 117). We were unable to pool data from 2 studies; no change was reported in 1 study,63 and a decline in muscle strength was reported in the second study.35

Pooled analysis revealed no evidence of change in muscle strength in the long-term (SMD: –0.39; 95% CI: –0.79 to 0; P = .05; I² = 74%; P < .001; n = 165). We were unable to include 3 studies in the pooled analysis; 2 revealed no change,63,64 and 1 revealed an improvement (P < .05).35

**Spasticity**

All studies in which short- (n = 25)28 and intermediate-term (n = 145)35,37,53,93 changes in spasticity were assessed revealed reduced spasticity (P < .05). Authors of 2 studies reported reductions, and the authors of 1 study reported no change in spasticity in the long-term (n = 118).25,35,37

**Participation and QoL**

Evidence for improvement in participation was observed at
6 months and at 127,29,56 and 2 years after MLS (Table 2). Intermediate- and long-term improvements in QoL were also observed (Table 2).

**Satisfaction**

Satisfaction with the outcome of surgery was rated as a mean (SD) of 7.9 (2.0) on a scale of 0 to 10 (with 10 being completely satisfied) by 279 parents.57 Sixty-one parents rated satisfaction with functional and cosmetic outcomes as means (SDs) of 6.8 (2.0) and 6.6 (2.0) of 10, respectively.56 Of 11 parents, 91% reported that they were satisfied with surgical outcomes.81 When 11 children were asked if the “results from SEMLS and rehabilitation were worth the effort,” 9 and 10, respectively, said yes at 1 and 2 years post-MLS.22

**Sensitivity Analyses**

Imputing the correlation coefficient as 0.7 instead of 0.3 resulted in a change in conclusions for the change in gait speed in the long-term (SMD: 0.17; 95% CI: -0.04 to 0.30; P = .01; I^2 = 78%; P < .001) and strength in the long-term (SMD: -0.58; 95% CI: -1.0 to -0.15; P = .008; I^2 = 91%; P < .001).

**Subgroup Analysis**

When children with bilateral CP only were included in pooled analyses, there was evidence that muscle strength declined in the intermediate-term (SMD: -0.35; 95% CI: -0.64 to -0.06; P = .02; n = 40) and long-term (SMD: -0.49; 95% CI: -0.96 to -0.02; P = .04; n = 134). All other analyses of children with bilateral CP only produced similar effect sizes and identical inference to that obtained from the primary analysis.

**DISCUSSION**

In summary, we found no evidence from a meta-analysis that gross motor function improved after MLS, but we found some evidence of improvements in gait and passive...
The range of motion. It should be noted, however, that there was significant heterogeneity associated with the pooled analysis of gross motor function and inconsistent findings across studies; although many studies revealed no evidence of change in gross motor function after MLS, some revealed evidence of improvement. Conversely, there was some evidence that muscle strength declined in the 6 months after MLS and was not different from presurgery levels at 1 year after MLS. Although participation and QoL were assessed in only 3 and 2 studies, respectively, there was some evidence that participation and QoL improved after MLS. Overall, parents reported being satisfied with outcomes of MLS, although there was considerable variation in satisfaction ratings. Adverse events after MLS appear relatively rare; only 1 adverse event that resulted in a life-threatening complication (including central nervous system complication) requiring intensive care and/or ICU management (grade IV complication according to the Clavien-Dindo classification) was reported. However, adverse-event reporting was incomplete, with adverse events being adequately reported in only 23% of studies.

Our findings support 2 previous reviews of MLS for CP. In both reviews, it was concluded that gait kinematics improved after MLS, although a quantitative synthesis of data was conducted in neither. With a similar search strategy to that used in the current review, McGinley et al included CP.

**TABLE 2** Changes in Participation and QoL After MLS

<table>
<thead>
<tr>
<th>Study</th>
<th>Sample Size</th>
<th>Outcome</th>
<th>Outcome Measure</th>
<th>Domain</th>
<th>Result</th>
</tr>
</thead>
<tbody>
<tr>
<td>Intermediate-term results</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Buckon et al27</td>
<td>7</td>
<td>Participation</td>
<td>PEDI</td>
<td>Social skills</td>
<td>MD: 7.41; 95% CI: 2.56 to 12.26</td>
</tr>
<tr>
<td>Buckon et al27</td>
<td>7</td>
<td>Participation</td>
<td>PEDI</td>
<td>Self-care</td>
<td>MD: 5.5; 95% CI: 0.65 to 10.37</td>
</tr>
<tr>
<td>Cuomo et al29</td>
<td>57</td>
<td>Participation</td>
<td>PODCI</td>
<td>Global function</td>
<td>MD: 8.25; P &lt; .001</td>
</tr>
<tr>
<td>Lee et al36</td>
<td>61</td>
<td>Participation</td>
<td>PODCI</td>
<td>Global function</td>
<td>MD: 2.6; P = .02</td>
</tr>
<tr>
<td>Cuomo et al29</td>
<td>57</td>
<td>QoL</td>
<td>PedsQL</td>
<td>—</td>
<td>MD: 10.03; P &lt; .001</td>
</tr>
<tr>
<td>Long-term results</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Buckon et al27</td>
<td>7</td>
<td>Participation</td>
<td>PEDI</td>
<td>Self-care</td>
<td>MD: 8.17; 95% CI: 2.35 to 13.99</td>
</tr>
<tr>
<td>Buckon et al27</td>
<td>7</td>
<td>Participation</td>
<td>PEDI</td>
<td>Mobility</td>
<td>MD: 7.34; 95% CI: 0.39 to 14.2</td>
</tr>
<tr>
<td>Buckon et al27</td>
<td>7</td>
<td>Participation</td>
<td>PEDI</td>
<td>Social skills</td>
<td>MD: 7.87; 95% CI: 3.10 to 12.24</td>
</tr>
<tr>
<td>Thomason et al9</td>
<td>11</td>
<td>QoL</td>
<td>CHQ</td>
<td>—</td>
<td>MD: 22; 95% CI: 4 to 39</td>
</tr>
</tbody>
</table>

CHQ, Child Health Questionnaire; PedsQL, Pediatric Quality of Life instrument; PODCI, Pediatric Outcomes Data Collection Instrument; —, not applicable.
identified 31 studies in which the effect of MLS for children with CP was examined. Despite including twice as many studies in the current review, we identified similar issues with the evidence base to those identified in 2010. There was consistently inadequate reporting of participant characteristics, surgical interventions, previous surgery, and rehabilitation. MLS is a complex intervention, consisting of interacting components that are particularly difficult to evaluate. For example, there are difficulties standardizing the design and delivery of MLS and rehabilitation, and the causal association linking the intervention and outcome is lengthy, complex, and likely context dependent.95 The results of this review reveal that a pattern of initial deterioration in gross motor function occurs in the year after MLS, followed by a return to baseline at 1 to 2 years post-MLS, and potentially further improvements in the longer-term. However, it is difficult to comment on the long-term impact of MLS on gross motor function given the relatively small number of patients who were assessed beyond 1 year after surgery. This complex and lengthy interaction between the intervention and outcome reveals that the contents of MLS and rehabilitation are likely crucial to explaining effectiveness or otherwise. Improved reporting of these factors is therefore essential to improving the evidence base for MLS. Authors of future studies should use reporting guidelines, such as the Consolidated Standards of Reporting Trials statement96 and the Template for Intervention Description and Replication checklist,97 to ensure that the study and the intervention are described in sufficient detail to appraise and replicate.

A lack of RCTs is a significant limitation to the evidence base. We identified just 1 RCT of 19 children. All other evidence was from cohort studies, with many being retrospective reviews of clinical records. As a result, patients were often excluded from studies without routine follow-up assessments, suggesting that the findings are subject to significant selection bias. Although effect estimates may be biased as a result of excluding participants from the analysis,98 the extent of attrition bias is unknown because the proportion of eligible participants included in analyses was not reported in the majority of studies. In addition, authors of few prospective cohort studies reported loss to follow-up. In addition, establishing causality is difficult without a comparison group to control for the potential impact of known and unknown confounding variables on the outcome. It should be noted, however, that findings from the RCT largely align with pooled analyses of data from cohort studies. In particular, the MD of −5.5 in the GPS, observed between the intervention and control groups at 1 year,9 was similar to the improvement in the GPS observed in cohort studies (−3.7 to −7.1).53,76,77,87

The pilot RCT reveals that it is feasible to conduct RCTs of MLS. However, researchers still face several barriers to conducting well-designed RCTs. These include ethical concerns regarding allowing people to experience progression of musculoskeletal deformities without intervening, difficulty recruiting patients given the relatively small number of children that undergo MLS annually, and difficulties obtaining funding to conduct a multicenter trial with sufficient duration of follow-up to evaluate the impact of MLS. In addition, when designing future studies, careful consideration should be given to the comparator to overcome ethical concerns and provide clinically meaningful results. Although the current review reveals that MLS improves gait, whether MLS is the most appropriate approach to improving gait cannot be established.

We found few studies in which authors assessed outcomes according to ICF domains despite all but 3 studies being published after the introduction of the ICF in 2001. The lack of prospective studies may explain why outcomes across ICF domains are assessed in so few studies. Lack of consensus regarding how to assess each ICF domain is also a barrier to using this framework. Regardless of whether a framework was used to evaluate MLS, it is concerning that authors of only 10 studies assessed activity using the Gross Motor Function Measure, which is considered the criterion measure of gross motor function. Additionally, authors of only 4 studies assessed participation, and authors of 3 studies assessed QoL. Passive range of motion, strength, and spasticity were assessed in more studies, although these are arguably less meaningful outcomes to participants. Indeed, passive range of motion and spasticity are associated with no change or small changes in gross motor function.99 To provide comprehensive information on the effects of MLS to families and professionals, authors of future studies need to prospectively plan to collect data on a range of outcomes. Although the ICF provides a standardized framework for the evaluation of interventions, it may be unrealistic to expect MLS to improve activity and participation given that activity and participation result from a complex interaction between the individual and the environment. However, even if MLS does not improve activity or participation, it may prevent further activity limitations and participation restrictions by preventing deterioration in musculoskeletal deformities.
The lack of prospective data collection may explain why authors of ~70% of studies did not report information on adverse events. We can therefore not make conclusions regarding the safety of MLS in this review. Standardized recording and reporting of adverse events should be implemented in future studies to ensure consistent and deliberate reporting of safety. Additional limitations of the evidence base are that all included studies are at a high risk of bias for blinding of the outcome assessment, and the majority had small sample sizes. Lack of blinding may exaggerate the effect of MLS, particularly when outcomes are assessed by using self-report measures, such as participation and QoL.100 Sample sizes may have impacted results by either inflating effect sizes or resulting in statistically nonsignificant findings.

There are also limitations to this review. We did not include gray literature or studies published in any language other than English. We included studies in which outcomes before and after MLS were reported, even if the primary aim for the study was not to assess the effect of MLS. To include some data in meta-analyses, we imputed correlation coefficients. However, sensitivity analyses revealed that our findings were generally robust for different imputed values. There was evidence of at least moderate heterogeneity in all models, which may be explained by the large amount of clinical and methodologic diversity in included studies.

CONCLUSIONS

This review represents the most comprehensive summary of the evidence on MLS for children with CP to date. Findings reveal that MLS is associated with changes in gait but not gross motor function in children with CP. However, in the review, we identified considerable limitations to the evidence base that need to be addressed in future trials. Specifically, there is a need for RCTs and improvements in reporting of trials.


Multilevel Surgery for Children With Cerebral Palsy: A Meta-analysis
Noor Amirah Amirmudin, Grace Lavelle, Tim Theologis, Nicky Thompson and Jennifer M. Ryan
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