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A comparison of conventional and minimally invasive multilevel surgery for children with diplegic cerebral palsy

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Title Page

Title:

A comparison of conventional and minimally invasive multilevel surgery for children with diplegic cerebral palsy.

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Tomos Edwards – project lead, data collection, writing and revision of the manuscript

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A comparison of conventional and minimally invasive multilevel surgery for children with diplegic cerebral palsy.

Abstract

Aim

To compare changes in gait kinematics and walking speed 24 months after conventional (C-MLS) and minimally invasive (MI-MLS) multilevel surgery for children with diplegic cerebral palsy (CP).

Patients and methods

A retrospective analysis of 19 children following C-MLS, mean age at surgery 12 years 5 months (range 7y 10m, 15y 11m), and 36 children following MI-MLS, mean age at surgery 10 years 7 months (range 7y 1m, 14y 10m), was performed. The Gait Profile Score (GPS) and walking speed were collected pre-operatively and 6, 12 and 24 months post-operatively. Type and frequency of procedures as part of MLS, surgical adverse events and subsequent surgery were recorded.

Results

In both groups, GPS improved from the pre-operative gait analysis to the 6 month assessment with maintenance at 12 and 24 months post-operatively. While reduced at 6 months in both groups, walking speed returned to pre-operative speed by 12 months. The overall pattern of change in GPS and walking speed was similar over time following C-MLS and MI-MLS. There was a mean of 9.3 and 9.5 procedures per child as part of C-MLS and MI-MLS, respectively. Surgical adverse events occurred in 7 (37%) and 13 (36%) children with 4 (21%) and 13 (36%) patients requiring subsequent surgery following C-MLS and MI-MLS, respectively.

Conclusion

This study indicates similar improvements in gait kinematics and walking speed 24 months after C-MLS and MI-MLS for children with diplegic CP.

Clinical relevance

- First paper to compare gait kinematics and walking speed following C-MLS and MI-MLS 24 months post-operatively
- Indicates similar improvements in gait kinematics and walking speed 24 months after C-MLS and MI-MLS for children with diplegic CP

Introduction

Multilevel surgery (MLS) is considered the standard of orthopaedic care for gait deviations in ambulatory children with diplegic cerebral palsy (CP).^{1,2} Conventional MLS (C-MLS), occasionally referred to as single-event MLS, often includes open femoral and tibial osteotomies, which involve significant soft tissue dissection. Lower limb muscle contractures are corrected by fractional lengthening, most often at the musculotendinous junction, with the intention of lengthening the muscle while minimising the loss of strength. Potential complications following MLS include failure of fixation, non-union, infection and incomplete correction.^{3,4} In addition, reduced muscle strength has been reported 12 months post-operatively.⁵

To overcome some of the limitations of C-MLS, a minimally invasive technique (MI-MLS) has been developed. This combines the use of closed corticotomy with stable intramedullary nail fixation and percutaneous muscle lengthening.⁶ Closed corticotomy and intramedullary nail fixation requires significantly less soft tissue dissection and there is much less periosteal disruption, thereby promoting earlier healing. Percutaneous muscle lengthening is a minimally invasive procedure designed to reduce both internal and external scarring and reduce soft tissue dissection and associated pain.

The only study comparing C-MLS and MI-MLS reported significant improvements in gait after 12 months in both groups.⁶ The study was limited by the inclusion of only 10 children in each group and a follow-up of 12 months. The aim of this study was to compare changes in gait kinematics and walking speed 24 months after C-MLS and MI-MLS for children with diplegic CP.

Patients and Methods

This study was a retrospective cohort study of children with diplegic CP that underwent MLS in a tertiary referral centre between January 2002 and December 2016. The study was approved by the Clinical Governance and Audit Team.

Patients

The gait laboratory database at our institution was screened to identify patients meeting the following criteria: 1) diplegic CP; 2) Gross Motor Function Classification System (GMFCS) I – III;⁷ 3) MLS at age ≤ 18 ; 4) MLS between January 2002 and December 2016. MLS was defined as two or more bone and/or soft tissue procedures at two or more anatomical levels bilaterally during one operative procedure.⁸

Children who underwent open muscle lengthening and open femoral and/or tibial osteotomies (plate and screws) formed the C-MLS group. Children who underwent percutaneous fractional lengthening of the musculotendinous units and percutaneous femoral and/or tibial osteotomies (closed corticotomy and fixation with titanium elastic nails) formed the MI-MLS group. Children undergoing only bone or soft tissue procedures were grouped according to the technique used (open or percutaneous).

Minimally invasive techniques have been the customary practice at our institution since February 2005.⁶ Several considerations affect eligibility for MI-MLS. A known limitation of intramedullary nails is the inability to correct significant sagittal or coronal plane deformities. As such, children with more severe deformities, including significant knee contractures and/or hip dysplasia are not suitable for MI-MLS. Therefore, it was deemed inappropriate to recruit children into the C-MLS group if their surgery was performed after February 2005, as by default, they would not be comparable to the MI-MLS group. This contention is supported by comparison of the pre-operative kinematic data and physical examination of patients that underwent C-MLS and MI-MLS after the introduction of minimally invasive techniques at our institution. Mean pre-operative knee contracture was 12.2° and 5.1°; knee flexion at initial contact was 40.9° and 37.7°; maximum knee extension in stance was 31.8° and 20.4° in patients that underwent C-MLS and MI-MLS, respectively. The C-MLS group therefore included patients meeting the inclusion criteria prior to February 2005, while the MI-MLS

group was formed by patients undergoing MI-MLS after February 2005.

For the C-MLS group, 19 patients met the inclusion criteria. For the MI-MLS group, 42 patients met the inclusion criteria and 6 were excluded as they underwent surgery at a different unit.

Kinematic outcome

Instrumented 3D gait analysis was performed using a motion capture system (Vicon Motion Systems Ltd, Oxford, UK) and a conventional marker set on the pelvis and lower limbs.⁹ Patients walked barefoot where possible, or in orthosis if not, along a 10m walkway at their self-selected speed with their usual assistive device if required. A minimum of six trials were captured and processed using the Plug-In Gait model (Vicon Motion Systems Ltd, Oxford, UK). The Gait Profile Score (GPS)¹⁰ and walking speed were calculated and then averaged across the six trials for each session. This was performed pre-operatively and 6, 12 and 24 months post-operatively. The GPS is a summary statistic used to evaluate overall patterns of gait. Lower GPS scores represent less deviation from normal gait. In addition, mean hip rotation, knee flexion at initial contact and maximum knee extension in stance were calculated from the gait data, and hip flexion, knee flexion and ankle plantarflexion contracture were extracted from the clinical examination. These variables were recorded pre-operatively and 24 months post-operatively.

Surgical procedures

Type and frequency of bone and soft tissue procedures performed as part of MLS was recorded.

Surgical adverse events

A medical record review identified surgical adverse events following the initial MLS. This was performed a mean of 17 years 1 month (range 16y 2m, 18y 4m) and 8 years 11 months (range 2y 11m, 12y 10m) post-operatively for the C-MLS and MI-MLS groups, respectively. All patients in the C-MLS group and 78% (28/36) in the MI-MLS had reached skeletal maturity when the medical records were reviewed. Surgical adverse events were graded according to the modified Clavien-Dindo-Sink (mCDS) classification.¹¹

Subsequent surgery

The medical record review identified any subsequent surgery required following the initial MLS.

Statistical analysis

Changes in GPS and walking speeds were compared between the groups using a repeated measures analysis of variance using an unstructured covariance matrix with the pre-operative level as a covariate. Inclusion of the pre-operative level increases the statistical efficiency of the comparison and allows for a regression to the mean effect, whereby those with higher baseline levels tend to experience a greater absolute reduction post-operatively. In doing so, it also compensates for any differences in baseline levels between the two groups. Group effects compared the average changes and the group by time interaction compared the patterns of change over time. Additionally, this method was used to assess the effects of pre-operative GMFCS, age at surgery and sex on changes in GPS and walking speed and whether these variables influenced the group effect. Statistical analysis of individual kinematic and physical examination results was not performed given concerns regarding multiple testing and small patient numbers, but data is presented to illustrate composition of the groups. The analyses were performed using IBM SPSS Statistics 24 (IBM Corp, Armonk, USA) and SAS 9.4 (SAS Institute Inc, Cary, USA). The level of significance was set at $p < 0.05$.

Results

Demographics for both C-MLS and MI-MLS groups are shown in Table 1.

	C-MLS (n, 19)	MI-MLS (n, 36)
Sex (M; F)	9 (47%); 10 (53%)	19 (53%); 17 (47%)
Pre-operative GMFCS (I; II; III)	4 (21%); 9 (47%); 6 (32%)	1 (3%); 18 (50%); 17 (47%)
Mean age at surgery (range)	12y 5m (7y 10m, 15y 11m)	10y 7m (7y 1m, 14y 10m)

Table 1. Demographics (percent per group and range) of C-MLS and MI-MLS groups. C, conventional; MI, minimally invasive; MLS, multilevel surgery; GMFCS, Gross Motor Function Classification System.

Mean GPS and adjusted change in GPS (change adjusted for pre-operative level) at each

stage of follow-up for both groups is shown in Table 2. The pre-operative GPS did not differ significantly between both groups ($p = 0.12$). Both groups had a significant reduction in GPS from pre-operative to 6 months, and this was maintained throughout each stage of follow-up ($p < 0.001$). There was no significant difference between groups ($p = 0.34$) and the pattern of change during follow-up was similar ($p = 0.65$).

	C-MLS			MI-MLS		
	n	GPS (°)	Change	n	GPS (°)	Change
Pre-operative	19	19.2 (9.6, 29.9)		36	16.8 (9.1, 25.2)	
6 months	19	12.4 (7.6, 20.2)	-5.9 (0.7)	30	12.7 (8.2, 17.2)	-4.9 (0.5)
12 months	19	12.6 (7.2, 20.3)	-5.7 (0.7)	36	12.4 (6.5, 22.7)	-4.9 (0.5)
24 months	15	12.8 (6.9, 21.9)	-5.3 (0.9)	33	12.4 (7.9, 22.2)	-4.7 (0.6)

Table 2. Mean GPS (range) and adjusted change from pre-operative (SE) at 6, 12 and 24 months post-operatively in C-MLS and MI-MLS groups. C, conventional; MI, minimally invasive; MLS, multilevel surgery; GPS, Gait Profile Score.

Mean walking speed and adjusted change in walking speed (change adjusted for pre-operative level) at each stage of follow-up for both groups is shown in Table 3. The pre-operative walking speed did not differ significantly between both groups ($p = 0.70$). Both groups had a significant reduction in walking speed at 6 months ($p < 0.01$), but this returned to speeds comparable to baseline by 12 months. There was no significant difference between groups ($p = 0.22$) and the pattern of change during follow-up was similar ($p = 0.44$).

	C-MLS			MI-MLS		
	n	Speed (m/s)	Change	n	Speed (m/s)	Change
Pre-operative	19	0.86 (0.43, 1.27)		36	0.83 (0.19, 1.35)	
6 months	19	0.74 (0.17, 1.11)	-0.12 (0.05)	30	0.60 (0.15, 1.27)	-0.24 (0.04)
12 months	19	0.82 (0.33, 1.34)	-0.04 (0.05)	36	0.76 (0.11, 1.34)	-0.07 (0.03)
24 months	15	0.87 (0.18, 1.25)	0.02 (0.05)	33	0.83 (0.18, 1.41)	-0.01 (0.04)

Table 3. Mean walking speed (range) and adjusted change from pre-operative (SE) at 6, 12 and 24 months post-operatively in C-MLS and MI-MLS groups. C, conventional; MI,

minimally invasive; MLS, multilevel surgery.

Neither pre-operative GMFCS, age at surgery or their interaction with follow-up visit had a significant effect on the change in either GPS or walking speed (all $p > 0.1$). There was a similar lack of effect of sex on GPS (all $p > 0.1$), but there was an influence of sex on changes in speed. Girls walking speed returned to baseline by 12 months, while for boys it took 24 months ($p = 0.02$). There was no significant interaction between the sex and group effects for GPS or walking speed (all $p > 0.2$).

Individual kinematic variables and physical examination findings from pre-operative and 24 month post-operative assessment are shown in Table 4.

Variable (°)	C-MLS				MI-MLS			
	n	Pre-operative	n	24 months	n	Pre-operative	n	24 months
Mean hip rotation	19	12.9 (-10.5, 34.8)	15	1.0 (-11.2, 15.8)	36	9.4 (-21.1, 31.5)	33	2.1 (-20.2, 25.9)
Knee flex. init. contact	19	44.3 (12.7, 86.5)	15	30.1 (9.7, 60)	36	37.7 (15.8, 66.5)	33	26.3 (5.5, 60.1)
Max knee ext. stance	19	32.7 (-8.8, 76.2)	15	16.9 (-18.3, 55.4)	36	20.4 (-7.6, 56.9)	33	15.5 (-13.1, 56.0)
Contracture (°)								
Hip flexion	18	9.9 (0, 30)	14	7.6 (0, 20)	35	9.3 (0, 30)	33	3.5 (0, 25)
Knee flexion	18	10.8 (10, 50)	14	1.2 (-30, 30)	35	5.1 (-15, 35)	33	2.1 (-15, 25)
Ankle plantarflexion	18	3.1 (-30, 35)	14	-6.4 (-30, 5)	35	10.1 (-20, 60)	33	-4.5 (-20, 10)

Table 4. Mean (range) kinematic variables and physical examination findings pre-operatively and 24 months after C-MLS and MI-MLS. Negative contracture represents range into knee extension and ankle dorsiflexion. C, conventional; MI, minimally invasive; MLS, multilevel surgery; flex, flexion; ext, extension; init, initial.

Surgical procedures

Type and frequency of surgical procedures performed as part of MLS is shown in Table 5. The mean total number of procedures was 9.5 per child in the C-MLS and 9.3 per child in the MI-MLS group.

	C-MLS	MI-MLS
Bone procedures		

Acetabuloplasty	1 (2%)	0 (0%)
Femoral osteotomy	32 (60%)	62 (65%)
Tibial osteotomy	2 (4%)	3 (3%)
Foot stabilisation	18 (34%)	31 (32%)
Total bone procedures	53	96
Soft tissue procedures		
Psoas lengthening	20 (16%)	0 (0%)
Adductor lengthening	9 (7%)	46 (19%)
Hamstring lengthening	35 (28%)	70 (29%)
Rectus femoris transfer	21 (17%)	49 (21%)
Calf lengthening	34 (27%)	58 (24%)
Tibialis anterior split transfer	4 (3%)	3 (1%)
Tibialis posterior lengthening	4 (3%)	12 (5%)
Total soft tissue procedures	127	238
Total number of procedures	180	334
Mean number of procedures per child	9.5 (6, 12)	9.3 (6, 14)

Table 5. Type and frequency (percent per group and range) of surgical procedures performed as part of MLS. C, conventional; MI, minimally invasive; MLS, multilevel surgery.

Surgical adverse events

Surgical adverse events occurred in 7 (37%) and 13 (36%) children following C-MLS and MI-MLS, respectively (Table 6). All adverse events following MI-MLS were mCDS Grade I or II. One patient required a diagnostic arthroscopy and lateral release for anterior knee pain due to patellar mal-tracking following C-MLS (Grade III). There were no Grade IV or V complications in either group.

	C-MLS	MI-MLS
Grade I		
Cast sore	0	3
Pain	2	3
Anxiety	0	3
Rash	0	1

Total	2	10
Grade II		
Partial rupture of tibialis posterior	1	0
Neuropraxia	1	1
Delayed union	2	0
Cast sore infection	0	1
Fungal skin infection	0	1
Total	4	3
Grade III		
Pain	1	0
Total	1	0

Table 6. Type and frequency of surgical adverse events graded by modified Clavien-Dindo-Sink classification. C, conventional; MI, minimally invasive; MLS, multilevel surgery.

Subsequent surgery

Four (21%) and 13 (36%) children required subsequent surgery, other than removal of implants, following C-MLS and MI-MLS, respectively (Table 7). Intramedullary nails were removed in 42% (14/33) of patients that had derotation osteotomies performed as part of MI-MLS. The most common indication for metalwork removal was pain and irritation. Where possible (8/14), removal was performed alongside any other required subsequent procedures.

C-MLS

Tibialis anterior split transfer, tibialis posterior and calf lengthening (L)	4y 9m
Tibial derotation osteotomy (L)	1y 1m
Calcaneal lengthening and cuneiform shortening osteotomy (L) and tibialis anterior split transfer, tibialis posterior, flexor digitorum and hallucis longus lengthening (B)	6y 5m
Distal femur extension and derotation, tibial derotation, calcaneal lengthening and cuneiform shortening osteotomy and calf lengthening (R)	12y 1m

MI-MLS

Tibialis anterior split transfer and tibialis posterior lengthening (B)	1y 9m
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Hamstring lengthening (B)	7y 7m
Distal femur 8-plate fixation (B)	4y 11m
Tibial derotation osteotomy (R) and hamstring lengthening (B). Subsequent tibial derotation osteotomy (R) and hamstring lengthening (L)	2y 11m & 3y 5m
Supracondylar femoral osteotomy and adductor lengthening (B). Subsequent shelf acetabuloplasty (R)	7y 1m & 11y 9m
Supracondylar femoral, tibial, calcaneal lengthening and cuneiform shortening osteotomy and tibialis posterior lengthening (B)	5y 2m
Tibialis anterior split transfer, tibialis posterior, flexor digitorum and hallucis longus lengthening (L)	2y 8m
Distal femur 8-plate fixation (B)	3y 7m
Adductor and hamstring lengthening (B)	4y 8m
Calcaneal lengthening osteotomy and 1 st MTPJ fusion (L)	3y 1m
Distal femur derotation osteotomy (R)	5y 3m
Tibial derotation osteotomy (L)	1y 3m
Supracondylar femoral extension osteotomy (L)	0y 9m

Table 7. Subsequent surgery, and when performed after initial MLS, for four patients following C-MLS and thirteen patients following MI-MLS. C, conventional; MI, minimally invasive; MLS, multilevel surgery; R, right; L, left; B, bilateral; MTPJ, metatarsophalangeal joint.

Discussion

This study is the first to compare gait and walking speed between children undergoing C-MLS and MI-MLS 24 months post-operatively. Both C-MLS and MI-MLS resulted in early correction of gait kinematics, with no significant difference in GPS between the two groups, and improvements maintained over 24 months. The adjusted mean reduction in GPS was 5.3° and 4.7° in the C-MLS and MI-MLS groups, respectively, representing a three-fold improvement with respect to the minimally clinically important difference (1.6°).¹² In both groups, improvement in GPS was comparable to a recent multicentre study that reported a mean reduction in GPS of 5.0° after a follow-up of 12 to 24 months following MLS.¹³

In addition to the GPS, improvements were reported in crouch gait walking pattern following MI-MLS as knee flexion at initial contact decreased from 37.7° to 26.3° and maximum knee extension in stance improved from 20.4° to 15.5°. Church et al¹⁴ reported similar improvements in knee flexion at initial contact (36° to 30°) and maximum knee extension in stance (21° to 17°) after a 12 year follow-up in a cohort of 59 young adults that had previously undergone MLS.

Prior research reported similar walking velocity pre-operatively and at 12 months follow-up in both C-MLS and MI-MLS groups.⁶ The current study found that after an initial deterioration at 6 months, walking speed had returned to pre-operative levels by 12 months, and was maintained at the 24 month follow-up in both groups. A review of changes in temporal distance parameters over varying lengths of follow-up after MLS reports mixed results; however, there was no significant change compared to pre-operative in 21/30 (70%) of the analysed studies.¹⁵ When comparing walking speed between the C-MLS and MI-MLS groups in this study, there was no difference between groups at any time-point and the overall pattern of change was comparable.

This study reported that walking speed had returned to baseline by 12 months in girls and 24 months in boys. In their multicentre study, Dreher et al¹³ report no effect of sex on the change in GPS and a recent review of predictors affecting outcome after MLS reports that sex-specific differences in children with CP is not of the quality to influence the decision-making process.¹⁶ In this study, the estimates of the effect of intervention were virtually unchanged by the addition of sex, and sex by follow-up visit, to the model. In addition, as the significance of sex by follow-up visit interaction was only $p = 0.02$ and we have performed multiple tests (12 in total), it would be reasonable to attribute the change in recovery of walking speed to chance.

The mean number of surgical procedures was 9.5 and 9.3 for the C-MLS and MI-MLS groups, respectively, similar to that reported in a recent large multicenter study (8.7 per child).¹³ Details on percutaneous techniques have been described previously.⁶ In the MI-MLS group, limited incisions of 2cm were involved in lengthening the gastrocnemius, and if necessary, the soleus. A similar technique is involved in transferring the rectus femoris. As the practice at our institution evolved over time, we no longer undertake diaphyseal tibial derotation osteotomies. We favoured stability and rapid healing over the minimally invasive

technique in this area in order to allow prompt mobilisation. We therefore adopted the supra-malleolar technique with stable internal fixation using locking plates. Similarly, we used locking plates to fix any osteotomies within the foot, in order to allow immediate mobilisation with full weight bearing.

A potential critique of MLS is technical imprecision. However, for both C-MLS and MI-MLS, the same method was used for measuring the correction of femoral rotation intra-operatively; the angle between an anteversion wire placed in the femoral neck and a second wire across the femoral condyles. In addition, gait analysis demonstrates improvement in mean hip rotation from 9.4° to 2.1° following MI-MLS. A potential limitation of percutaneous hamstring lengthening is unintended damage to the muscular portion of both semimembranosus and semitendinosus.¹⁷ We do not have any indication that our patients suffered muscle damage. Our previous study reported significantly faster time to mobilisation following MI-MLS, suggesting less pain, presumably attributable to less trauma.⁶ In addition, this study reports no significant differences in walking speed at any time point between C-MLS and MI-MLS, although it is beyond the scope of the current study to discuss the long-term effect of percutaneous muscle lengthening on gait.

This study reports surgical adverse events in 7 (37%) and 13 (36%) patients following C-MLS and MI-MLS, respectively. Most were mCDS Grade I or II (19/20) which, by definition, either do not require treatment or resolve with simple measures. One patient required a diagnostic arthroscopy and lateral release for anterior knee pain due to patellar mal-tracking following C-MLS (Grade III). This was unlikely to be directly related to the previous surgery and the pain settled post-operatively. Our institution has improved its documenting of complications over time, which may introduce bias against the MI-MLS group. However, after a mean of over 17 years following C-MLS and over 8 years following MI-MLS, this study reports comparable Grade II and no Grade III adverse events following MI-MLS.

Four (21%) and 13 (36%) patients required subsequent surgery, other than removal of implants, following C-MLS and MI-MLS, respectively. Dreher et al¹³ report that 39% of patients in their multicenter study required subsequent procedures, other than removal of implants, comparable to both groups in this study. In addition, 42% (14/33) of patients had intramedullary nails removed following MI-MLS. In this study, data on subsequent surgeries

was collated a mean of 17 years 1 month and 8 years 11 months following C-MLS and MI-MLS, respectively. Patients following MI-MLS may still require additional surgery, however, the majority were skeletally mature when their medical records were reviewed (28/36).

Despite often being minor procedures, the increased rate of subsequent surgery and metalwork removal represents an important limitation of MI-MLS and should be discussed in a candid manner with patients and parents prior to surgery.

There are limitations to this study. First, the small study size limits the power of the study. However, for both GPS and walking speed, statistically significant differences were apparent. Second, the lack of randomization introduces the possibility of selection bias. Children in the MI-MLS group were a mean 1 year and 10 months younger at the time of surgery. Although the repeated measures analysis of variance found that age at surgery did not have a significant effect on the change in either GPS or walking speed, the difference in age may be due to older children not being suitable for MI-MLS. Third, the C-MLS group was formed by a historical cohort. Over the 14 year period of this study changes were noted in our surgical practice. For example, psoas lengthening was replaced by adductor lengthening when hip flexor spasticity was a problem. Whilst the use of a historical cohort allows for a more appropriate comparison between the groups, likely changes in pre and post-operative rehabilitation protocols and improvements in the early treatment of spasticity and contractures must be considered when interpreting the results.

In conclusion, this study adds to the literature by indicating similar improvements in gait kinematics and maintenance of walking speed 24 months after both C-MLS and MI-MLS. Important differences between groups, including age at surgery, must be considered when interpreting these results. Further prospective comparison of matched patients is required in order to confirm MI-MLS is as effective as C-MLS in improving gait for children with diplegic CP.

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