

CLINICAL ARTICLE

Single event multilevel surgery in cerebral palsy: a review of the literature

Dr FM Bischof, BSc(Physio), MSc(Physio), PhD(Wits)
Honorary Adjunct Professor, School of Therapeutic Sciences, University of the Witwatersrand

Reprint requests:

Dr FM Bischof
PO Box 4105
1411 Germiston South
Gauteng
Phone: (011) 827-6465
Cell: 083-269-1428
Fax: (011) 827-8716
e-mail: faithbischof@gmail.com

Abstract

Single event multilevel surgery (SEMLS) has become an increasing trend in the orthopaedic management of cerebral palsy due to the advantages of one hospital admission and one period of rehabilitation.

This article reviews the studies relating to the outcome of SEMLS published over the last 10 years. Improvements in gait parameters are reported in the literature but the articles provide a low level of evidence and there are limitations in the conduct of the research. Further studies, employing good scientific rigour are indicated to provide higher levels of evidence regarding the efficacy of this intervention for persons with cerebral palsy.

Introduction

Orthopaedic surgery has been a method of intervention in cerebral palsy (CP) for the last century. Many soft tissue and bony procedures have been developed to address the musculoskeletal aspects of this condition. In more recent years, the approach to surgery has changed from performing one or two procedures at a time to addressing all deformities simultaneously. Single event multilevel surgery (SEMLS) refers to the correction of all orthopaedic deformities in one session, which has the advantage of requiring one hospital admission and one period of rehabilitation.^{1,2} A further rationale for SEMLS is the prevention of secondary deformity which can occur when a single deformity is addressed.²

A study by Beals found that a plateau in walking ability in spastic diplegia is reached by the age of 7 years.³ It is generally recommended that SEMLS is performed at about this age.

The growing demand for evidence-based clinical interventions requires the practitioner to integrate clinical skills with research evidence, i.e. published studies of the outcome of the intervention. The purpose of this article is to review literature pertaining to SEMLS. It is not an evidence report, but the analysis of the articles incorporates some of the guidelines provided by the American Academy of Cerebral Palsy and Developmental Medicine for reviewing treatment outcomes.⁴

Method

A Medline search was instituted, and the search confined to articles published in English in the last 10 years. Eleven articles were sourced. These are reviewed individually, followed by a summary of the level of evidence provided, the outcome measures employed and the general conduct of the studies.

Article review

Zwick *et al*⁵ assessed 17 patients with diplegic CP clinically, and by three-dimensional (3D) gait analysis before and after multilevel surgery. An average of 7.5 surgical procedures was performed on each patient.

Surgical intervention was individualised for each patient according to set criteria. A standardised protocol was followed for postoperative physiotherapy. The average follow-up was 3.8 years (range 2.6–5.7). Kinetic parameters showed improved knee extension in stance and more normal ankle motion post surgery. Patients walked faster with increased power generation at the hip in stance. Limitations of this study were non-uniformity of the surgical procedures employed, the small patient numbers, as well as lack of a control group.

Molenaers *et al*⁶ looked at similarities and differences between single event botulinum toxin Type A (BTX-A) treatment and surgery. This was a retrospective study of gait analysis data from two groups of patients: 29 children with CP in the BTX-A group, and 23 in the surgical group. This cannot be labelled a true comparative study, due to the considerable differences in age and pre-treatment condition between groups. Children in the Botox group were younger with primary gait problems in distal joints. The authors state that the benefits of both treatments were confirmed by the study, but due to the differences in improvements in each group, the treatment modalities should be regarded as complementary. No definite conclusions can be drawn from this study, due to the dissimilarity of the groups and other variables.

Gough *et al*⁷ used gait analysis and recording of support needed for walking, as well as parent reports to compare two groups of ambulant children with CP. Twelve children who underwent multilevel surgery were compared with 12 who had no surgical intervention. The latter were labelled the control group, but were not true controls as they continued with their usual physiotherapy and orthotic support. The follow-up period was short (< 2 years). The authors report deterioration in the kinematics of the 'control' group and an improvement in the operated group. Two independent walkers who walked independently before surgery required sticks postoperatively, and the level of walking ability in the other operated patients remained the same. The limitations of this comparative study include small numbers and the short follow-up. The use of the term 'natural' history in the title is inaccurate.

Dobson *et al*⁸ prospectively studied the effects of multilevel surgery in children classified as group IV spastic hemiplegia. They used the classification described by Gage *et al*: increased hip flexion, reduced movement at the hip and knee, and ankle equinus. Seventeen patients fulfilled the study criteria. Ages at surgery ranged from 7–17 years with a mean of 12 years. Again, operative procedures were individualised for each patient. Follow-up ranged from 2–5.5 years with a mean of 2.9 years. Three-dimensional gait analysis was the main outcome measure.

Gait data was statistically analysed and clinically and statistically significant improvements in mean transverse plane kinematic rotations for pelvis, hip and foot progression reported. There was also an improvement in gait symmetry. As in the previous studies, the surgical intervention was not uniform. Numbers were small, and there was no comparison or control group.

Saraph *et al*⁹ retrospectively analysed the gait data of 32 ambulatory children with CP who had SEMLS. The aim of their study was to ascertain whether improvements in gait were maintained after the discontinuation of dynamic ankle-foot orthoses (mean 1 year postoperatively), night splints (mean 2.3 years) and physiotherapy and splints (mean 4.4 years). The average total of surgical procedures per child was 8.1. Clinical examination and gait analyses were employed, and the results statistically evaluated. They found that there was a general decrease in gait function between the first and second postoperative evaluations, followed by a gradual improvement – decreased cadence and increased stride length and velocity. They feel evaluation of SEMLS should be performed a minimum of 3 years post surgery, and acknowledge the need for long-term follow-up.

Rodda *et al*¹⁰ investigated the correction of severe crouch gait in patients with spastic diplegia using SEMLS. This was a retrospective study of a small group of patients. A consecutive sample of 10 diplegics aged 7.9–16.2 years (mean 12 years) at surgery was assessed. Eight of the 10 had previous surgery: six tendo-Achilles lengthenings and two Baker calf lengthenings. The procedures chosen for the multilevel surgery were determined by gait lab assessment. Postoperative rehabilitation consisted of a combination of physiotherapy and hydrotherapy, followed by physical recreational activities. Orthotics were worn for all weight-bearing activities for the first year after surgery. The patients were re-assessed after 5 years. Valid and reliable instruments were used to measure functional mobility and the data was statistically analysed. They reported relief of knee pain, increased extension during gait and improved community walking. The Functional Mobility Scale (FMS) was used and the median score for community mobilisation changed from 1 to 3, i.e. there was improvement. The authors acknowledge the limitations of their study, which include small sample size, lack of controls and variable surgical prescription. A positive aspect of the study is the use of validated and reliable outcome measures.

Khan¹¹ assessed the outcome of SEMLS in 85 diplegic patients who presented as untreated non-walkers at two centres in Pakistan. Their inability to walk could have been due to the presence of fixed contractures of the lower limbs and lack of treatment, rather than their neurological impairment.

The growing demand for evidence-based clinical interventions requires the practitioner to integrate clinical skills with research evidence

They were aged 5-12 years at time of surgery (mean 8.5 years). Soft tissue surgery alone was performed in 79% of the patients. Assessment of the outcome at 2-5 years post-operatively (mean 3.5 years) was by joint assessment and use of a modified walking scale which was originally developed for myelomeningocele. This scale does not indicate the level of support required for walking. The author states that all children became walkers post surgery, with 45.9% becoming household walkers. Physiotherapy and orthotics were provided after surgery.

A study by Harvey *et al*¹² looked at the ability of the FMS to detect changes following SEMLS. This was a retrospective study of 66 walking children with spastic diplegia, with a mean age of 10 years at surgery and who were followed up for 24 months. The FMS documents the use of assistive devices at home, in school and in the community, thus measuring performance in real life (as opposed to performance in a laboratory setting.) Odds ratios showed significant deterioration in mobility at 3-6 months postoperatively, improvement to baseline levels at 12 months and further improvement at 24 months.

Adolfsen *et al*¹³ retrospectively examined gait parameters after identical SEMLS in 31 ambulatory children with CP. This was the only study where the surgical intervention was the same in all the participants. They all had rectus femoris transfer, hamstring lengthening and gastrosoleus lengthening. The study cohort comprised diplegics, hemiplegics and one quadriplegic. All patients were reassessed one year after surgery. Six of the group seen again 4 years post-surgery showed that gains were maintained in the longer term. It was found that children with a jump knee pattern tended to go into knee hyperextension after surgery. The authors also recommend a longer follow-up.

Seniorou *et al*¹⁴ looked at recovery of muscle strength following SEMLS in a prospective randomised controlled trial. They compared the efficacy of progressive resistance strengthening (n=11) to active exercise (n=9) commenced 6 months postoperatively. At 6 months there was a significant reduction of muscle strength in all muscle groups. After 6 weeks of intensive physiotherapy, both groups showed significant improvement in muscle strength, gross motor function measure (GMFM) scores and gait parameters, with resistance training showing some advantages over active exercises. Despite this, strength in some muscle groups did not reach pre-operative values one year after surgery.

In a recent study, Lee *et al*¹⁵ analysed parental satisfaction after SEMLS in ambulatory children with CP using a visual analogue scale. Two hundred and seventy-nine patients were enrolled in the study, which equated to a response rate of 66%. The mean number of surgical procedures per patient was 5.4 (SD 3.0). The most common procedures were distal hamstring lengthening, Z-lengthening of the tendo Achilles, rectus femoris transfer and femoral derotation osteotomy. Mean follow-up after surgery was 6.6 years. Overall mean satisfaction after surgery was 7.9 (out of a possible 10).

Satisfaction was higher for unilaterally involved patients than the bilaterally involved ones ($p<0.001$). Parents of children who were classified as level 1 of the gross motor function classification system were more satisfied than those of children on level II or III of the system ($p=0.029$). The authors found that the level of satisfaction decreased with increasing duration of follow-up. They stated that actual satisfaction might have been lower than reported in the study because of the possibility that the unsatisfied parents did not respond. This study was classified as a level II prognostic study.

Discussion

The outcome studies reviewed all documented improvements in gait following SEMLS. However they measured outcomes before and after the surgical intervention, which provides a low level of evidence (mostly level IV). Except for one study, surgical procedures were not uniform making evaluation of a particular procedure difficult.

Sample sizes were small in studies employing gait outcomes which restricted their power. A meta-analysis of spatio-temporal measures of gait pre- and postintervention¹⁶ suggests that sample sizes for evaluating interventions to improve gait in CP, using individuals as their own controls, should be 50 or 60.

All studies employed more than one intervention (surgery, physiotherapy and orthotics). Consequently it cannot be determined which part of the treatment protocol produced the favourable results.

Reduction in strength in the first year postoperatively was a common observation. This should be explained to parents before surgery is undertaken, and anticipated by the therapy team. It should also be explained that the level of walking ability is unlikely to improve.

Gait analysis was the most frequently used outcome measure. In the more recent studies authors have added functional mobility assessments, which provide a better picture of walking ability in the child's own environment. Only two articles reported parents' perceptions of outcomes. Blinding of the assessors of function in the studies is not documented. Average follow-up after surgery was less than 5 years in most studies. Exceptions were Rodda *et al*¹⁰ where a follow-up evaluation was done after 5 years and Lee *et al*¹⁵ where the mean follow up was 6.6 years. Long-term follow-ups are important in CP interventions to improve gait due to the known deterioration in mobility in CP as the child gets older, especially during adolescence.

In conclusion, further studies employing good scientific rigour are indicated in order to provide higher levels of evidence regarding the efficacy of SEMLS for persons with cerebral palsy. This would guide future surgical recommendations.

Note from the Editor:

It has been quite some time now that single event multi-level surgery in cerebral palsy became popular.

We were anxious to know whether this procedure is safe, whether children could be rehabilitated in a reasonable period of time, whether there were any contra-indications, any combinations of different procedures which are contra-indicated, whether mobility has improved and remains better than it used to be.

First and foremost our aim should be to treat these children conservatively, and taking into account whether the child is bedridden, a sitter or a walker. Surgery is indicated only if there are specific, well-motivated indications geared to the needs of the specific child.

One has to bear in mind that we are dealing with a group of children who do not have the same ability as normal children to be rehabilitated and may find it difficult to reach the status that one hoped for prior to surgery.

Multiple event surgery can be disastrous if, for example, Achilles tendons are lengthened without attending to the hamstrings.

Measuring the outcome is to some extent subjective and difficult to assess. Unfortunately the literature on this problem is sparse. This means that every surgeon must use his or her own judgment as to which procedure will benefit the child and which combinations will be safe and advisable for that specific child. The surgery always has to be tailored to each individual patient's needs.

It may be useful to compare pre-operative DVDs to postoperative DVDs as this is perhaps the most objective way to judge the outcome of our surgery. Due to the big difference in deformities and degree of spasticity, it is very difficult to find a comparable control group.

From the available articles it is clear that follow-up should be at least 3–5 years postoperatively.

Doing SEMLS decreases the number of times that a child will need to be admitted to hospital. However, one can then expect that these children will need a much more specialised, difficult and longer period of rehabilitation.

My own approach would rather be to do fewer procedures in one sitting and to allow for two periods of hospitalisation, especially if bony surgery is also needed. The latter prolongs the period of immobilisation and simultaneously also prolongs the time period before the team can start with mobilisation and strengthening of muscles. Bony surgery and lengthening or transfer of muscles have a different postoperative rehabilitation programme.

Unfortunately, the literature is very sparse on good follow-up studies for a period of 5 years and longer. We could not get answers to all our questions. Therefore, the total responsibility for doing the right surgery rests on the shoulders of the orthopaedic surgeon! Careful decision-making is mandatory to ensure a good result.

References

1. Graham HK, Selber P. Review article. Musculoskeletal aspects of cerebral palsy. *J Bone Joint Surg* 2003;**85-B**(2):157-66.
2. Renshaw TS, Deluca P. Cerebral Palsy in: Morrissey RT, Weinstein SL (eds). *Lovell and Winter's Pediatric Orthopedics*. Philadelphia: Lippincott, Williams and Wilkins;2005: pp 551-603.
3. Beals RK. Spastic paraplegia and diplegia evaluation of non-surgical and surgical factors influencing the prognosis for ambulation. *J Bone Joint Surg (B)* 1966;**48A**:827-46.
4. Butler C, Chambers H, Goldstein M, Harris S, Leach J, Campbell S, Adams R, Darrah J. Evaluating research in developmental disabilities: A conceptual framework for reviewing treatment outcomes. *Dev Med Child Neurol* 1999;**41**:55-9.
5. Zwick EB, Sarah V, Linhart WE, Steinwender G. Propulsive function during gait in diplegic children: Evaluation after surgery for gait improvement. *J Pediatr Orthop* 2001;Part B,**10(3)**:226-33.
6. Molenaers G, Desloovere K, De Kat J, Jonkers I, De Borre L, Pauwels P, Nijs J, Fabry G, De Cock P. Single event botulinum toxin Type A treatment and surgery : similarities and differences. *Eur J Neurol* 2001;**8 Suppl 5**:88-97.
7. Gough M, Eve LC, Robinson RO, Shortland AP. Short-term outcome of multilevel surgical intervention in spastic diplegic cerebral palsy compared with the natural history. *Dev Med Child Neurol* 2004;**46(2)**:91-7.
8. Dobson E, Kerr Graham H, Baker R, Morris ME. Multilevel orthopaedic surgery in Group IV spastic hemiplegia. *J Bone Joint Surg* 2005;**87-B**:548-55.
9. Sarah V, Zwick EB, Aliner C, Schneider F, Steinwender G, Lindhart W. Gait improvement in diplegic children: How long do improvements last? *J Pediatr Orthop* 2005;**25(3)**:263-7.
10. Rodda JM, Graham HK, Nattrass GK, Galea MP, Baker R, Wolfe R. Correction of severe crouch gait in patients with spastic diplegia with the use of multilevel orthopaedic surgery. *J Bone J Surg* 2006;**88-A**:2653-64.
11. Khan MA. Outcome of single-event multilevel surgery in untreated cerebral palsy in a developing country. *J Bone J Surg* 2007;**89-B**:1088-91.
12. Harvey A, Kerr Graham H, Morris ME, Baker R, Wolfe R. The Functional Mobility Scale: ability to detect change following single event multilevel surgery.
13. Adolfsen SE, Ounpuu S, Bell KJ, De Luca PA. Kinematic and kinetic outcomes after identical multilevel soft tissue surgery in children with cerebral palsy. *J Pediatr Orthop* 2007;**27(6)**:658-67.
14. Seniorou M, Thompson N, Harrington M, Theologis T. Recovery of muscle strength following multilevel orthopaedic surgery in diplegic cerebral palsy. *Gait Posture* 2007;**26(4)**:475-81.
15. Lee SH, Chung CY, Moon S, Choi IH, Cho T, Yoo WJ, Lee KM. Parental satisfaction after single-event multilevel surgery in ambulatory children with cerebral palsy. *J Pediatric Orthop* 2009;**29(4)** :398-401.
16. Paul SM, Lohmann Siegel K, Malley J, Jaeger RJ. Evaluating interventions to improve gait in cerebral palsy: A meta-analysis of spatiotemporal measures. *Dev Med Child Neurol* 2007; **49**:542-9.