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Single Event Multilevel Surgery in children with bilateral spastic cerebral palsy: A 5 year prospective cohort study☆

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ABSTRACT

Background: Single Event Multilevel Surgery (SEMLS) is considered the standard of care to improve gait and function in children with bilateral spastic cerebral palsy (BSCP). We have demonstrated in a randomized controlled trial (RCT) of SEMLS, that gait was improved at 12 months after surgery and gross motor function at 24 months after surgery. The question addressed in this study, was to determine if improvements in gait and function, would be maintained at 5 year follow-up.
Methods: Nineteen children with BSCP, GMFCS levels II (14 children) and III (5 children), mean age 9.7 years (range 7.7–12.2 years) participated in a prospective cohort study following participation in a RCT, with follow-up to 5 years. Outcome measures were Gait Profile Score (GPS), Gillette Gait Index (GGI), Gait Deviation Index (GDI), Gross Motor Function Measure (GMFM66) and Functional Mobility Scale (FMS).
Results: Eighteen children have completed follow-up, with interval analysis at 1, 2 and 5 years post SEMLS. One child was excluded because of neurological deterioration and his diagnosis was revised to Hereditary Spastic Paraparesis (HSP). GPS improved by 5.29° and GMFM66 by 3.3% at 5 years post SEMLS. Differences between outcome measures at 1 versus 5 years and 2 versus 5 years (except GMFM66) were not significant, indicating that improvements in gait and gross motor function were stable over time.
Conclusions: SEMLS results in clinically and statistically significant improvements in gait and function, in children with BSCP, which were maintained at 5 years after surgery.

1. Introduction

Cerebral palsy (CP) is the result of a static encephalopathy or non-progressive lesion of the developing brain but the musculoskeletal deformities in growing children are often progressive [1]. Musculoskeletal deformities may impair both gait and gross motor function. Since the advent of instrumented gait analysis (IGA) emphasis has been placed on the correction of all fixed musculoskeletal deformities at one operative session, usually referred to as Single Event Multilevel Surgery (SEMLS) [2–5]. SEMLS is considered to be the standard of care to improve gait and function in children with CP [2,4–8]. SEMLS improves the likelihood of achieving sagittal plane balance [9] and reduces the need for repeated anaesthetics, reduces episodes of hospitalization and requires only one major period of rehabilitation [2,5].

The findings from our pilot randomized controlled trial (RCT) to evaluate the outcome of SEMLS in children with bilateral spastic CP (BSCP) showed that the *timing* of improvements following SEMLS were different for gait compared to gross motor function. Clinically and statistically significant improvements in gait were found at 12 months after SEMLS but functional improvements (GMFM66) were not found until 2 years after SEMLS [10]. The natural history in CP is for deterioration in gait and function with time, especially during the pubertal growth spurt [11]. Factors responsible for this decline may include progression of the musculoskeletal pathology as well as unfavourable changes in the ratio of body mass to strength [12,13]. The question to be addressed in this medium-term prospective cohort study was to determine if improvements in gait and function would be maintained through the pubertal growth spurt.

Systematic review [14] has shown that there are several well designed *retrospective* cohort studies which report an improvement in gait or function following SEMLS [3,4,6,7,15–19]. There are several *prospective* studies which also report improvements in gait [8,20–23]. The duration of follow-up in these studies ranged from 12 months [21–23], approximately 18–24 months [3,6,7,15,16,20], to approximately 4 year follow-up [8,17]. Only three studies

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reported follow-up of greater than 4 years [4,18,19], with only one reporting 10 year follow-up [18]. However, none of these longer term studies were prospective. Retrospective studies are more subject to bias because the composition of the cohort is not decided at the initiation of the study nor are the outcome measures defined and the testing intervals are usually not standardized. In contrast, a *prospective* cohort study permits the composition of a study cohort according to specified inclusion and exclusion criteria defined a priori. In addition, the outcome measures and testing intervals are standardized. There are ethical and practical problems with retaining children with CP in *randomized* controlled trials of more than 12 months duration because of the progressive nature of the musculoskeletal pathology and gait deterioration. Prospective cohort studies are probably the best design to investigate medium to long term changes in gait and functioning [24].

The aim of this study was to evaluate the outcome of SEMLS on gait; gross motor function and functional mobility 5 years post SEMLS.

2. Participants

2.1. Inclusion criteria

Children with a confirmed diagnosis of BSCP with registration on the Victorian state-wide CP register, GMFCS levels II and III and aged 6–12 years at the time of randomization in the RCT who required SEMLS were included. All children ($n = 19$) who had participated in the RCT phase of the study and who had IGA, on at least two occasions following SEMLS with one being at least 5 years post-surgery were eligible for the prospective phase of the study.

2.2. Exclusion criteria

Children with a diagnosis other than CP. For further details of inclusion and exclusion refer to the RCT phase of this study [10].

3. Sample size

This was a prospective cohort study of children in both the surgery and control groups of a RCT. The sample size was therefore determined by the considerations of sample size for the RCT [10].

4. Methods

This was a single centre, *prospective cohort study*, of children who had participated in a RCT of SEMLS, for a period of 12 months [10]. At the conclusion of the randomized phase of the trial, children who were randomized to the surgery group were followed up in the prospective cohort study. Children who were randomized to the control group proceeded to surgery, following the RCT protocol then continued their follow-up in the prospective cohort study. All children, regardless of group allocation, were followed for a minimum of 5 years with standardized outcome assessments at 1, 2 and 5 years post SEMLS. Each assessment included IGA and functional assessments including GMFCS, FMS and GMFM66. Children had elective removal of implants including blade plates between 12 and 18 months after SEMLS. Additional surgeries were scheduled either at the time of implant removal or at any time during the 5 year follow-up, according to clinical need and based on information from IGA.

The primary outcome measures were the Gait Profile Score (GPS), the Gillette Gait Index (GGI) and the Gait Deviation Index (GDI). The GPS and GGI are summary statistics of gait and are derived from gait kinematics (GPS) and kinematics plus selected temporospatial parameters (GGI). The GDI is a multivariate measure of overall gait pathology [25]. These measures are considered to be valid and reliable tools to describe gait dysfunction as a single variable and to assess change after intervention [10,18,25]. All three measures were included to allow comparison to previously published data. Functional measures namely the GMFM66 and FMS were also completed.

Ethical approval for this study was granted by the Ethics in Human Research Committee of the Royal Children's Hospital, Melbourne, Australia (reference number EHRC 23144). The trial design and reporting follow the CONSORT principles, as far as practically possible [26].

SEMLS was defined as at least one surgical procedure, performed on two different anatomic levels (hip, knee or ankle) on both sides of the body .The surgical prescription did not need to be symmetrical and was not uniform but individually tailored to the child's needs as determined by a comprehensive evaluation which included standardized physical examination, radiological evaluation and IGA. The multilevel surgical program included muscle tendon lengthening, tendon transfer, rotational osteotomy and stabilization of the hip and foot following published guidelines [5]. The basic principles were the identification and correction of all contractures and lever arm deformities, deemed to be interfering with dynamic gait function.

For the purpose of the prospective cohort study, children originally allocated to the control group proceeded to surgery within 4 weeks of the RCT 12 month assessment. The protocol for surgery for this group followed that of the surgical group of the RCT [10] and was carried out by the same surgical team.

Post-surgical review followed standard protocols [27] and the physical therapy protocol followed that of the RCT [10].

IGA was collected using a 50 Hz, 6 or 10 camera Vicon 370 system (Oxford Metrics, Oxford, UK). Reflective markers were applied to the bony landmarks using a standardized procedure [28]. Kinematic data was calculated using Plug-in Gait (Oxford Metrics, Oxford, UK). The Movement Analysis Profile (MAP), GPS, GGI and GDI were calculated for both legs on four individual gait cycles. The median GPS and mean GGI and GDI were calculated for each child using Gaitabase, a web interfaced repository for gait analysis data.

The GMFM66 and the FMS were conducted following the IGA using standard protocols. Assessments were conducted by senior gait laboratory physical therapists, experienced and trained in the use of all measures.

4.1. Statistical analysis

Analysis was carried out for the total cohort that is RCT surgical and control group combined. Linear regression with robust standard errors for comparison between 5 years and pre-surgery, 1 and 2 years post SEMLS were carried out for all outcome measures (except for FMS) using Stata 10.0 Statistical Data Analysis Program (Statacorp, TX, USA). Frequency data for change in FMS scores is reported.

5. Results

Of the 19 children with BSCP who participated in the RCT all have completed 1 and 2 years follow-up and 18 have completed 5 years follow-up. One child was excluded from this analysis as his diagnosis was revised from CP to Hereditary Spastic Paraparesis (HSP).

Fig. 1 shows the progress of children throughout the study from the start of the RCT until completion of the prospective phase of the study. Children's characteristics pre-surgery and 5 years post-surgery are summarized in Table 1. There was a mean increase in height of 29.7 cm and in weight of 25.1 kg during the 5-year study period.

Indications for surgery and surgical procedures performed are summarized in Table 2. The total number of procedures performed