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Original Research Article

Lower extremity soft tissue surgery in spastic cerebral palsy: experience from a government rehabilitation unit

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ABSTRACT

Background: Spastic cerebral palsy (CP) remains the most common type of CP and may be managed surgically or non-surgically depending upon its severity. Recent advances have replaced single-level surgery by the concept of multilevel surgery where multiple levels of musculoskeletal pathology, in one/both lower limbs, are addressed during one operative procedure, requiring only one hospital admission and one period of rehabilitation. This study assessed the outcome of lower limb soft tissue surgery in children with spastic CP in a government rehabilitation unit and measured its feasibility with limited infrastructure facilities and patient compliance.
Methods: The study comprised of 26 patients aged between 2-12 years. Physical examination and GMFCS scores were recorded and evaluation of sitting balance, standing balance and gait were done. Musculotendinous soft tissue lower limb surgery was performed at one or more levels unilaterally or bilaterally and the results were interpreted.
Results: Complete or near complete correction of deformities were attained by all children postoperatively. Significant improvements were noted in the gross motor functional classification system (GMFCS) scores. All parents and children were satisfied with the surgical outcome and reported improvement in functional abilities and locomotion in the follow-up along with better quality of life and mobility.
Conclusions: Lower limb soft tissue surgery is a valuable aid in improving functional abilities and locomotion in children with spastic CP. Surgery should be undertaken depending upon clinical indications and can be successfully carried out in government hospitals with ordinary infrastructure in developing countries as well.

Keywords: Cerebral palsy, Deformity, Surgery

INTRODUCTION

Cerebral palsy (CP), a term considered more descriptive than diagnostic, owes its earliest description to the orthopaedic surgeon William Little in 1862. Cerebral palsy is a syndrome of motor impairment resulting from a lesion occurring in the developing brain; the disorder varies in the timing of the lesion, the clinical presentation, and the site and severity of the impairments.[1] CP is one of the most common causes of physical disability in children, accounting for 2% to 2.5% per 1000 live births.[2]

There are two major subtypes of CP: the spastic form and the non-spastic form which can be further divided as the dyskinetic and ataxic types.[3] Spastic form of CP is highly predominant in preterm infants and remains the leading type in term infants as well.[4]

Management of spastic CP may be surgical or non-surgical (oral medications, injection phenol, intrathecal baclofen, injection botulinum toxin, physical/ occupational therapy, orthoses) or a coupling of both. A multidisciplinary approach is the most successful and is mandatory considering the spectrum of disorders that affect a CP child. Surgical treatments when indicated must focus on improving the degree of motor function and overall quality of life of the patient along with prevention of associated secondary complications rather than aiming at miraculous cures. Orthopaedic procedures have been designed to address the various components of the progressive musculoskeletal pathology including tendon lengthening, tendon transfers, rotational osteotomies, and joint stabilization procedures.[5] Recent advances have replaced single-level surgery by the concept of multilevel surgery in which multiple levels of musculoskeletal pathology, in both lower limbs, are addressed during one operative procedure, requiring only one hospital admission and one period of rehabilitation.[5] First reported by Norlin and Tkaczuk in 1985, it is also known as multilevel surgery, gait-improvement surgery and, most frequently, single-event multilevel surgery (SEMLS) to distinguish it from the 'birthday syndrome' approach of the past.[5] Adequate postoperative rehabilitation is critical in ensuring that results of surgery are sustained.

The objective of this study was to assess the outcome of lower limb soft tissue surgery in children with spastic CP in a government rehabilitation unit and to consider its feasibility with limited infrastructure facilities and patient compliance.

METHODS

The study subjects constituted patients admitted in the Department of Physical Medicine & Rehabilitation (PMR), Karnataka Institute of Medical Sciences (KIMS), Hubballi, Karnataka, India. The study included 26 patients (M:F, 1.8:1) with a mean age of 6.77 years (2-12 years). All patients were admitted for corrective surgery over a period of 24 months (March 2014 to March 2016). Informed consent was obtained from all the parents. Ten patients had hemiplegia, nine patients had diplegia, five patients had quadriplegia, and two patients were afflicted with triplegia.

All children barring quadriplegics had attained good truncal balance, dynamic sitting, and good muscle power in both the upper limbs or in unaffected limb (in hemiplegics). None of the patients had fixed spinal deformity or prior surgery for correction of lower limb deformity.

Exclusion criteria included children with dyskinetic or mixed CP, those requiring bony surgeries, and those with severe or profound mental retardation.

Physical examination and gross motor functional classification scale (GMFCS) scores were used to assess the static and dynamic conditions in the lower extremities.[6]

Evaluation of sitting balance, standing balance and gait were done. Soft-tissue contractures in the lower limbs were assessed with special tests and joint examinations.

All deformities were measured using goniometer according to the neutral 0 method. Hip abduction restriction in extension and in 90° of flexion was attributed to hip adductor muscle contracture. Hamstring tightness was assessed using popliteal angle which was measured with the patient in supine position and the hip in 90° of flexion. Silverskiold test was used to measure equinus deformity at ankle. Gait patterns such as crouching, scissoring, toe-to toe gait, and toe-to-heel gait were see in all the children clinically.

Operative procedures

In cases of hip adductor tightness, open tenotomy of adductor longus and brevis was performed. In static knee flexion deformity, fractional lengthening of hamstrings was performed which included tenotomy of semitendinosus along with fractional lengthening of semimembranosus medially and biceps femoris laterally. In all patients with static equinus deformities, Z-plasty was done for lengthening of Achilles tendon using White's Method.

Postoperative protocol

After hip surgeries, compression bandage was applied & the hips placed in abducted position. All patients were discharged on the 5th postoperative day following completion of a course of intravenous antibiotics. Patients were recalled on the 14th post op day for suture removal and were sent home with groin-to-toe cast in the operated limbs. They were advised rest with cast for 4-6 weeks and recalled for removal of casts. Physiotherapy was started and measurement of orthoses (used as resting splints) was given simultaneously.

In cases of surgery for knee flexion/equinus deformity/both, high above-knee cast was applied at the time of surgery with knee in 5-10° in flexion and ankle in about 5° dorsiflexion. All patients were discharged on the 5th postoperative day following completion of a course of intravenous antibiotics. After removal of sutures on 14th postoperative day, children were discharged with cast and advised to come after 4-6 weeks. Patients were advised rest during this period (walking not permitted). At follow up, cast was cut in bivalved manner on the first day of admission, so that the lower end could be used as shell. Wax bath to both knee and ankle joints was started from the next day (to make joint supple), and passive range of motion started from 3rd day both manually and with knee continuous passive motion (CPM) equipment. Plastic solid ankle foot orthoses (AFO) or plastic knee ankle foot orthoses (KAFO) were prescribed and gait training with the orthosis and assistive device (walker or walking aluminium cane depending upon unilateral or bilateral surgery) was given.

Statistical analysis

Categorical variables were summarized as frequency, percentage and numerical variables were summarized as mean, standard deviation, median and interquartile range. Analysis of pre and post-surgery qualitative data was done using stratified McNemar-Bowker test for internal symmetry and Stuart Maxwell statistic for marginal homogeneity. Quantitative data for change in pre to post surgery GMFCS score was done using non parametric Wilcoxin signed ranked test. Analysis was done using SPSS version 19 and MH Program version 1.2. A P value less than or equal to 0.05 was considered statistically significant.

RESULTS

Preoperatively, all the children had static deformities in the lower limbs. All 10 hemiplegic children and 8 out of 9 diplegics were ambulatory preoperatively although with deformities. Five quadriplegics and both triplegics were non ambulatory pre operatively. The objective of the soft-tissue surgery in hemiplegics, diplegics and triplegics was to correct the deformity to aid patients in standing upright, wear the orthoses, and for ambulation using orthoses with or without assistive device for walking. In quadriplegics, the aim was to improve sitting balance, perineal hygiene and to prevent further hip subluxation and dislocation.

Deformities were recorded in all the joints. There were 29 static equinus deformities, 10 hip adductor deformities, and 22 knee flexion deformity (medial and lateral hamstrings) in 26 patients. Mean hip adduction deformity was recorded at 12.4º, mean knee flexion (Hamstring) deformity was 63.8º and mean equinus deformity was 27.4º preoperatively. Complete or near complete correction of deformities were attained by all children postoperatively (Table 1).

GMFCS scores and physical examination was performed to assess improvement in functional abilities and locomotion preoperatively and postoperatively (3 months) in all 26 patients (Table 2).

Significant improvement in GMFCS score following surgery (P=0.000) was observed. No major complications were noticed at the operated site. However, 5 children developed wound dehiscence at the suture line with loss of skin and were managed with antibiotics as well as daily wound dressing, resulting in wound healing with no further complications.

Table 1: Distribution of patients according to age group, gender, diagnosis and surgical procedure performed.

Parameters		GMFCS	Number (n)	Mean±SD	Median	Wilcoxin Signed rank test
Age group	**2-6 years**	Pre	15	3.67±0.98	3	P=0.001
		Post	15	2.47±1.36	2	
	7-12 years	Pre	11	3.55±0.82	4	P=0.005
		Post	11	2.55±1.21	2	
Gender	**Male**	Pre	17	3.41±0.94	3	P<0.001
		Post	17	2.18±1.33	2	
	Female	Pre	9	4.00±0.71	4	P=0.005
		Post	9	3.11±0.93	3	
Diagnosis	**Diplegia/triplegia**	Pre	11	3.64±0.50	4	P=0.002
		Post	11	2.64±0.67	3	
	Hemiplegia	Pre	10	2.90±0.57	3	P=0.004
		Post	10	1.40±0.70	1	
	Total body involvement	Pre	5	5.00±0.00	5	P=0.083
		Post	5	4.40±0.55	4	
Procedure	**Fractional lengthening of Hamstrings and tendoachilles lengthening**	Pre	10	3.60±0.52	4	P=0.004
		Post	10	2.60±0.70	2.5	
	Tendoachilles lengthening	Pre	11	3.00±0.63	3	P=0.003
		Post	11	1.55±0.82	1	
	Hip adductor tenotomy	Pre	5	5.00±0.00	5	P=0.083
		Post	5	4.40±0.55	4	

[a]GMFCS: Gross motor functional classification scale.

Table 2: Comparison of preoperative and postoperative GMFCS scores.

		Postoperative GMFCS score					Total	Mc Nemar-Bowker test
		1	2	3	4	5		
Preoperative GMFCS score	1	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	P=0.001
	2	2 (7.69)	0 (0)	0 (0)	0 (0)	0 (0)	2 (7.69)	
	3	5 (19.23)	6 (23.08)	0 (0)	0 (0)	0 (0)	11 (42.31)	
	4	0 (0)	1 (3.85)	6 (23.08)	1 (3.85)	0 (0)	8 (30.77)	
	5	0 (0)	0 (0)	0 (0)	3 (11.54)	2 (7.69)	5 (19.23)	
Total		7 (26.92)	7 (26.92)	6 (23.08)	4 (15.38)	2 (7.69)	26 (100)	

[a]GMFCS: Gross motor functional classification scale.

Thirteen children inclusive of 9 diplegics, 2 triplegics and 2 hemiplegics were advised plastic KAFO as a locomotive aid postoperatively. Eight hemiplegic children received plastic solid AFO for ambulation. Five children with quadriplegia were advised plastic bilateral KAFO for use as resting splints and not as ambulatory aids.

Mean duration of follow-up in the study was 4 to 12 months. During the follow up, 7 (26.92%) children were functional/household ambulators, 14 (53.84%) were community ambulators and all 5 (19.23%) quadriplegics were non ambulatory. All parents and children were satisfied with the outcome of surgery and reported improvement in functional abilities and locomotion in the follow-up. Quality of life was better and many children who were unable to stand before surgery were standing and walking with orthoses and assistive devices. Parents of quadriplegic children reported improved sitting balance and perineal hygiene and were content with these results. No episodes of hip dislocation amongst the quadriplegics were reported.

DISCUSSION

Cerebral palsy is one of the commonest disorders of child development and has a major impact on the quality of life of the affected child as well as the parents.[7] Spasticity is the main motor disorder in children with CP and poses a major challenge in management and rehabilitation as well.[5]

Surgical interventions targeting the locomotor system play a pivotal role in management of children with CP. Corrective surgery is indicated when the contractures are static and severe enough to interfere with movements and locomotion.[8] Surgical procedures for lower extremity soft tissue such as tenotomy, tendon transfers, fractional or Z lengthening of the tendon can be performed to correct contractures and deformities of the joint to lengthen the spastic muscle.[9] SEMLS which has replaced the previous concept of single level surgery, has shown improved long term gait outcomes in both prospective and retrospective studies in bilateral CP. [10]

Owing to poor financial statuses of many of the children in the study group and inadequate resources in the rehabilitation unit, surgery could not be performed at all three levels in the present study. However, surgery at two levels was taken up in most of the cases either unilaterally or bilaterally as indicated. A study by Schranz et al analysed the long term outcome over a period of 10 years of unilateral SEMLS in 14 children with spastic hemilplegia.[10] They found sustained improvement of GPS (used as main outcome measure) at 3.73 degree over the follow up period of 9 years and concluded that children with unilateral spastic CP can benefit from unilateral SEMLS and can maintain these benefits as well.[10]

Various deformities recorded in the joints of children in our study resulted in complete or near complete correction postoperatively. This finding is in accordance with several well documented studies in literature that have demonstrated a positive outcome of surgery either at single level or multilevel on deformity correction and subsequent improvements in gait and posture.

Gorton et al studied 75 ambulatory children (aged 4-18 yrs) with spastic CP prospectively who underwent lower extremity musculotendinous surgery to improve gait. The children were individually matched on the basis of sex, gross motor function classification system level, and CP subtype to a nonsurgical cohort. Surgical procedures performed included single-level soft tissue release or multilevel bony and/or soft tissue procedures. The surgical group showed significant improvement in function after 1 year compared with the nonsurgical group as measured by the Gillette gait index, with few significant changes noted in outcome measures.[11]

Thomson et al conducted a randomized controlled trial of single-event multilevel surgery on 19 children (mean age of nine years and eight months) with spastic diplegia. The children were randomized into two groups i.e., surgical group (11 children) and the control group (8 children). The control group underwent a program of progressive resistance strength training, with the randomized phase of the trial concluding at twelve months following which they exited the study and progressed to surgery, whereas the surgical group continued to be followed in a prospective cohort study. The gait profile score (GPS) and the Gillette gait index (GGI) were the primary outcome measures assessed and the gross motor function (gross motor function measure-66 [GMFM-66]), functional mobility (functional mobility scale [FMS]), time spent in the upright position, and health-related quality of life (child health questionnaire [CHQ]) were the secondary measures assessed.

Surgical intervention comprised a total of eighty-five surgical procedures, with a mean of eight procedures per child. The surgical group displayed a 34% improvement in the GPS and a 57% improvement in the GGI at twelve months while the control group had a small non-significant deterioration in both indices. These differences between the groups were found to be highly significant. Postsurgical follow up at twenty-four months revealed an increase in the GMFM-66 score by 4.9% and an improved FMS score, time spent in the upright position, and the physical functioning domain of the CHQ in the surgical group.[12]

Both of these studies support our findings that surgical interventions are beneficial in improving gait and function in children with spastic CP.

It is a commonly held opinion that surgical interventions to improve gait should be postponed until motion patterns are well established. This outlook is further confounded by fears that interventions preformed at a young age may be associated with an increased risk of failure and relapse with less predictable results.[13] Patients in our study ranged between 2-12 yrs with a mean age of 6.77 yrs. Only two patients were aged below 4 years and both were quadriplegic with significant hip subluxation that was detected during radiographic hip surveillance necessitating early surgical intervention.

It has been suggested that surgery should always be considered once other conservative treatment methods have been deemed ineffective, and it should definitely not be delayed if walking function is deteriorating. [13] Svehlik et al stated that though their study showed better long-term outcomes in older children with CP, in practice there is often no luxury of being able to sit back and wait for the adolescent growth spurt in order to achieve these better long-term results.[13]

Significant improvement was seen in the GMFCS scores of patients post operatively. Two patients who were scored at GMFCS grade 2 preoperatively moved to grade 1 post operatively. Out of the 11 patients who were at GMFCS grade 3 preoperatively, 6 patients moved to grade 2 postoperatively and 5 patients moved to grade 1. Amongst the 8 patients who were graded at GMFCS grade 4 pre operatively, 6 patients moved to grade 3, 1 to grade 2 and 1 remained at grade 4. Out of the 5 GMFCS grade 5 patients, 3 moved to GMFCS grade 4 and 2 remained at grade 5 postoperatively.

Zorer et al evaluated the results of single-stage multilevel muscle-tendon surgery performed on the lower extremities for the treatment of contractures in 23 patients with spastic cerebral palsy and found an improvement of at least one GMFCS level in all patients.[14]

Gupta et al studied the effects of SEMLS in the lower extremity in 34 children with spastic cerebral palsy found similar results as that of Zorer et al and noted that results were maintained till the time of last follow-up.[15]

Orthoses play a vital role in the physical management of children with CP in conjunction with medical, surgical, and therapeutic interventions. The aims of lower limb orthotic management of CP include correction/prevention of deformity, provision of a base support, facilitation of training in skills and improvement in gait efficiency. Before prescribing an orthoses, a thorough assessment of the child’s needs is essential based on the severity of their impairment and their individual activity limitations.[16]

Various orthoses prescribed in the study were in accordance to the needs of the patients with the aim of maintaining the surgical correction and to further improve the gait characteristics such as velocity, endurance, and cadence. Patients maintained the improvement at the time of discharge and during the postsurgical follow-up period.

There were no complications noted in the 12 month follow up period in our study and parents were satisfied with the postsurgical outcomes. However, considering the short follow up period in our study, being a major limiting factor, these follow up results warrant caution.

Gough et al studied the short term outcome of multilevel surgical intervention in 24 ambulant children with spastic diplegic cerebral palsy of which twelve children had surgical intervention (treatment group) while the other 12 did not (control group). An interval three-dimensional gait analysis was performed for all the children. The changes between the control group and the treatment group at follow-up were substantial wherein the control group, showed significant increase in minimum hip and knee flexion in stance which were unrelated to age, the interval between analyses, changes in the passive joint range of motion, nor changes in anthropometric measurements. The treatment group on the other hand showed a significant improvement in minimum knee flexion and in ankle dorsiflexion in stance suggesting positive outcomes in gait immediately after surgery.[8]

Thomson et al studied the effects of SEMLS in 19 children with bilateral spastic CP through a prospective cohort study to determine if improvements in gait and function would be maintained at 5 years post follow up. They found that differences between outcome measures (gait profile score [GPS], Gillette gait index [GGI], gait deviation index [GDI]), gross motor function measure [GFM66] and functional mobility scale [FMS]) 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.[17]

Lower extremity soft tissue surgery in children with spastic CP and good trunk control yields favourable results especially more so when performed as a single-stage multilevel procedure as established by several studies in literature. In developing countries like India, where patient turn up for multistage surgery and follow up is poor owing to socio economic limitations, disease related social stigma and minimal knowledge regarding CP prognosis, SEMLS offers a cost effective and rational alternative to patients and their care givers who would otherwise not turn up for multistage surgery.

However, SEMLS could not be undertaken in our study due to inadequate resources at our disposal. Surgical corrections were undertaken according to clinical indications and the type of infrastructure available during the study period. Although all children showed satisfactory results with minimal complications after corrective surgery, and their functional ability improved significantly, small sample size and follow up period were major limitations of the study. Additionally, performing SEMLS with digital gait analysis will lend further credibility to the positive results obtained in this study.

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