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Research Paper

Soft tissue release and osteotomies in the treatment of patients with spastic diplegic cerebral palsy
軟組織鬆解和截骨術治療痙攣型雙癱性腦癱患者

Ng Bobby Kin-Wah*, Chau Wai-Wang, Hung Alec Lik-Hang, Lam Tsz-Ping, Cheng Jack Chun-Yiu

Department of Orthopaedics and Traumatology, Chinese University of Hong Kong, Hong Kong

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ABSTRACT

We aim to study the outcome of soft tissue releases by tendon elongations and osteotomies in fixed joint contractures by clinical examination and patient self-reported assessment on 20 patients (14 males and 6 females) with spastic diplegic cerebral palsy treated with single-event multilevel surgery (SEMLS) between 2000 and 2012. A questionnaire was used to collect information on problems encountered before and after surgery and decision on surgery. Comparing patients with Gross Motor Function Classification System class I/II, (N = 8), III (N = 8) and IV/V , patients of classes IV/V showed much slower mean recovery time than I/II group (14.00 vs. 4.38 months, $p < 0.01$). SEMLS in the treatment of patients with spastic diplegia had good mid-term results in most patients. The patients who had unfavourable outcomes are associated with mental retardation, general or local complications and previous selective dorsal rhizotomy surgery. Patient selection and good rehabilitations preoperation and postoperation provided the most favourable outcomes of SEMLS.

中 文 摘 要

我們的目的是通過臨床檢查和患者自我報告，評估20例（男14例，女6例）痙攣型雙癱性腦癱患者在2000年至2012年之間進行單次多層手術治療（SEMLS），研究以肌腱延長和截骨術等軟組織鬆解治療固定關節攣縮弛的結果。使用問卷收集手術前後遇到的問題以及手術決定的信息。比較粗大運動功能分類系統運GMFCS I / II級（N = 8）、III（N = 8）和IV / V組患者的平均恢復時間比I / II組慢（14.00比4.38個月，p <0.01）。SEMLS治療痙攣型雙癱性腦癱在大多數患者中有良好的中期療效。不良後果的患者與弱智、全身或局部並發症以及先前選擇性背側神經根切斷術相關。患者選擇、手術前後的良好康復提供了SEMLS最有利的結果。

Introduction

Spasticity at their quadriceps often occurs in children with cerebral palsy (CP), making their bodies difficult to walk.[1] In children with CP, knee flexion deformity is common resulting from spastic and contracted hamstring muscles.[2] Soft tissue releases by tendon elongations and osteotomies in the treatment of fixed joint contractures are the standard orthopaedic procedures in the management of patients with cerebral palsy aiming to improve joint motion and gait efficiency.[3] The results after surgery, however, are contradictory.[4] Some patients showed great improvement in gait, whereas others show little improvement.[5] These procedures may weaken hip extension and knee flexion.[6] Gait analysis of these patients form the objective assessment of these patients, but it is time-consuming. Gait analysis was usually carried out for a selected group (moderately to severely suffering patients), and those with milder involvements might never have a chance to have a gait study, which incur a selection bias in gait studies involving children with CP.[7] We aim to study the outcome of these procedures by clinical examination and patient self-report assessment to provide the patient-centred outcome assessment of the treatment effect.

* Corresponding author. Department of Orthopaedics and Traumatology, Chinese University of Hong Kong, Hong Kong.
E-mail address: bobng@ort.cuhk.edu.hk (B.K.-W. Ng).

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Methods

We review all patients with spastic diplegic cerebral palsy treated with single-event multilevel surgery (SEMLS) between year 2000 and 2012 to look at the functional change, clinical outcome and complications of these standard orthopaedic procedures, approved by the local institution review board. Using the Clinical Data Analysis and Reporting System (CDARS) of the Clinical Management System (CMS) database, we retrieved patients who had been treated for more than the 10 years.

A questionnaire aiming at collecting information on surgery, follow-up and the outcomes on problems encountered before surgery, improvements after surgery and decision on surgery (yes or no, with reasons) were designed, and the questionnaire survey was carried out for every participant (Appendix 1). They were interviewed over the telephone by the chief medical practitioner (BKW Ng). The information collected was transformed to either numeric or categorical variables and was stored in a secured database.

Data analysis

We reviewed the case records and X-ray reports to obtain data on age at operation, joint involvement, extent of surgery, length of stay, pre- and post-Gross Motor Function Classification System (GMFCS) class, recovery time to normal or maximal function and complications, and the analysis of the dataset from the questionnaire was also carried out. Variables were expressed using student *t* test for continuous variables or analysis of variance for categorical variables where appropriate. The patients were then grouped by their preoperative GMFCS classes (I/II, III and IV/V), and the mentioned demographic and outcome variables from the questionnaire were compared. All statistical analyses were performed using IBM SPSS version 24.0 (IBM Corp. Released 2016. IBM SPSS Statistics for Windows, Version 24.0. Armonk, NY: IBM Corp.). A two-sided p value ≤ 0.05 was considered statistically significant.

Results

A retrospective review of 22 operated records from 20 patients (14 males and 6 females, two patients had repeated operations) was carried out. The mean age of patients at operation was 12.65 ± 3.14 years. Their mean age at this study was 18.11 ± 3.15 years, and the mean follow-up interval was 5.45 ± 3.06 years (Table 1). There were 36.3% (N = 8) patients who had obtained GMFCS classes I and II, and it was decreased to 31.8% (N = 7) after surgery. On the other side, eight (36.4%) patients had preoperative joint fixed flexion contracture at knee, followed by hip/knee (27.3%), hip knee ankle (18.2%), knee ankle (13.6%), and knee ankle foot (4.5%). Nine (40.9%) patients had soft tissue release at level 1, 31.8% at level 2, 22.7% at level 3 and 4.5% at level 4. The details of soft tissue releases are designated by Hp (psoas release at pelvic brim), Ha (adductor releases), K (hamstring muscle elongation) and A (elongation of Tendo Achilles).

Comparing patients with GMFCS classes I/II (N = 8), class III (N = 8) and IV/V (N = 6), patients of classes IV/V had much slower mean recovery time (14.00 vs. 4.38 months, $p < 0.01$) (Table 2). Patients from both groups had similar self-rated percentages of postoperative limb power ($p = 0.370$), stamina ($p = 0.246$) and self-rated improvement scores ($p = 0.707$). After surgery, one patient had a change in the GMFCS class from III to II, and one patient from II to IV. For questions on their choices of operation if they could choose again, one patient of class IV/V replied not to have the surgery, and three replied "not sure" (1 each from class I/II, class III and class IV/V groups). Among patients who had left comments on their choices, one patient choosing "Yes" commented "some gain some loss", and two patients who chose "not sure" commented "one patient because of stiff knee post SDR and one patient yes because it helped to relax, not sure because walking ability is worsened".

Table 1
Demographic characteristics of the patients with CP.

Demographics	Number (%)
Gender	N = 20
Male	14 (70.0)
Female	6 (30.0)
Age at operation (years)	N = 22
Mean ± SD (range)	12.65 ± 3.14 (5.64–18.26)
Median	12.63
Age at FU (years)	N = 22
Mean ± SD (range)	18.11 ± 3.15 (10.94–24.74)
Median	18.41
FU interval (years)	N = 22
Mean ± SD (range)	5.45 ± 3.06 (1.48–13.11)
Median	5.99
GMFCS (preoperation)	N = 22
I	5 (22.7)
II	3 (13.6)
III	8 (36.4)
IV	5 (22.7)
V	1 (4.5)
GMFCS (postoperation)	N = 22
I	5 (22.7)
II	3 (9.1)
III	7 (36.4)
IV	6 (27.3)
V	1 (4.5)
Preoperative joint FFC (HKA)	N = 22
HK	6 (27.3)
HKA	4 (18.2)
K	8 (36.4)
KA	3 (13.6)
KAF	1 (4.5)
STR levels	
1	9 (40.9) (9 K)
2	7 (31.8) (4 KA, 2 HpK and 1 HaK)
3	5 (22.7) (3 HpHaK, 1 HpKA and 1 HaKA)
4	1 (4.5) (1 HHKA)

A = elongation of Tendo Achilles; CP = cerebral palsy; FFC = fixed flexion contracture; FU = follow-up; GMFCS = Gross Motor Function Classification System; Ha = adductor releases; HKA = hip knee ankle; HK = hip knee; Hp = psoas release at pelvic brim; K = hamstring muscle elongation; KA = knee ankle; KAF = knee ankle foot; SD = standard deviation; STR = soft tissue release.

Table 2
Gender, age at operation and outcome variables by grouped GMFCS ratings.

Demographics/Outcomes	GMFCS classes			p
	I/II (N = 8)	III (N = 8)	IV/V (N = 6)	
Gender				
Male, N (%)	5 (62.5)	7 (87.5)	4 (66.7)	0.493
Female, N (%)	3 (37.5)	1 (12.5)	2 (33.3)	
Age at operation (years)				
Mean ± SD	11.12 ± 2.84	14.10 ± 3.28	12.77 ± 2.84	0.167
Recovery time (months)				
Mean ± SD	4.38 ± 2.20	8.25 ± 4.17	14.00 ± 3.10	<0.01
Percentage of limb power postoperation				
Mean ± SD	95.00 ± 10.69	100.00 ± 0.00	96.67 ± 5.16	0.370
Percentage of stamina postoperation				
Mean ± SD	95.00 ± 10.69	100.0 ± 0.00	100.0 ± 0.00	0.246
Improvement score				
Mean ± SD	83.34 ± 35.63	95.00 ± 14.14	83.33 ± 40.82	0.707
Have the operation again?				
Yes	7 (87.5)	7 (87.5)	4 (66.7)	0.566
No	0 (0.0)	0 (0.0)	1 (16.7)	
Not sure	1 (12.5)	1 (12.5)	1 (16.7)	

GMFCS = Gross Motor Function Classification System; SD = standard deviation.

Discussion

SEMLS has been shown by gait laboratory assessment to have good long-term results at more than 10 years follow-up. Over

lengthening of hamstrings weakens hip extension, and when coupled with persistent spastic psoas, it leads to secondary hip flexion and stiff knee deformity, causing sagittal plane imbalance.[8] Intrapelvic intramuscular psoas tenotomy produced an improvement of hip flexion deformity without the loss of muscle power to initiate the swing phase. In this series, most patients responded well to the SEMLS treatment. This study is the first study comparing the recovery time, preoperative/postoperative limb power and attitude on surgical decision given a second thought between patients with low (I/II), moderate (III) and high (III/IV/V) GMFCS ratings. Patients with CP with GMFCS ratings I and II showed much shorter time for them to recover. One case of GMFCS class drop from II to IV resulted from the patient developing a sore heel postoperation (Figure 1). This patient suffered from mental retardation, and the complication delayed rehabilitation which had a great impact on outcome of this patient. Two patients who had SDR before SEMLS also reported unfavourable results. Other patients who reported no reduction of lower limb power and regained good walking ability reported that they would have the same surgery again. One patient who migrated from GMFCS III to II had a very supportive and highly motivated mother who personally trained the patient before and after surgery (Figure 1).

Patients with CP with higher GMFCS ratings IV/V were older than patients with lower GMFCS ratings, which means that relatively older patients had poorer functional abilities. The ideal situation is that they would have received operations when their functions would be comparatively promising. Through the findings from the present study, the patients with GMFCS ratings IV/V required longer time to recover. Medical professionals should take these observations into account and try to offer earlier and effective treatments for these patients.

Patients from the three groups had similar self-rated percentages of postoperative limb power, stamina and self-rated improvement scores. Sung et al carried out a long-term outcome study on the use of SEMLS in spastic diplegia, and the results showed that the ankle dorsiflexion during stance was maintained for 5 years after a 1-year increase.[9] This is a very important observation on the fact that SEMLS does offer good power restoration on sagittal plane kinematics regardless of their motor function level. Moreover, general factors of personality, age and sex were important for patients with spastic diplegia to have a successful outcome. Patients in these two groups showed promising results, showing their underlying willingness to improve their degree of mobility.

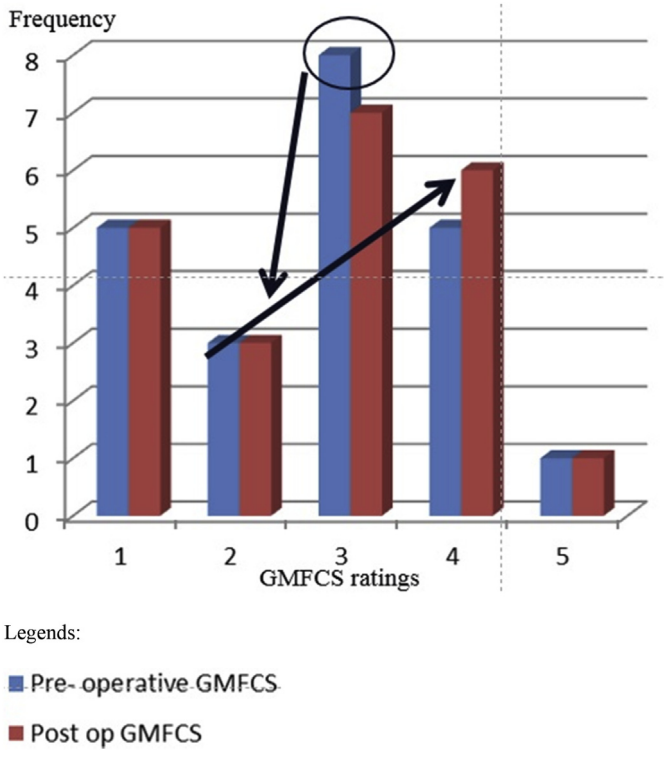

Figure 1. Changes of GMFCS ratings after surgery.
GMFCS = Gross Motor Function Classification System.

Few patients changed their surgical decisions if they were offered a second chance, either not operated or difficult to make the decision. This finding is very important on establishing a good communication between the surgeon-in-charge and patients, explaining the pros and cons clearly before the surgery. This is often neglected in surgical decision-making which might incur legal actions from the patients and their family members.

Limitation of this study

There are several limitations of this study that may contribute to bias in the results, common to retrospective studies with this population. First, this is a retrospective review study in which selection bias in the selection of patients is inevitable. Second, the study is prone to recall bias when asking the patients to fill out questionnaires on their ability levels, personal feelings and problems encountered after surgery, which was taken place years ago. The small number of patients participated in this study might limit the data generalisability for this study although the number of study patients was pretty similar in related studies.[10]

In conclusion, this is the first study comparing the recovery time, preoperative/postoperative limb power and attitude on surgical decision given a second thought between patients with low and high GMFCS ratings. Our results showed that most patients responded well to the SEMLS treatment. Patients with CP with GMFCS ratings I and II showed much shorter time for them to recover. Patients with CP with GMFCS rating I/II, III and IV/V had similar postoperative limb power, stamina and self-rated improvement scores. Support from family members or caregivers was definitely helpful on improving patients' GMFCS level.

SEMLS in the treatment of patients with spastic diplegia has good mid-term results in most patients. These patients reported no weakness of their lower limb muscle power. The patients who had unfavourable outcomes are associated with mental retardation, general or local complications and previous selective dorsal rhizotomy surgery. Patient selection and good rehabilitations preoperation and postoperation provided the most favourable outcome of SEMLS.

Conflict of interest

None declared.

Appendix

Appendix 1. Questionnaire on postoperative assessment for patients with CP

Patient details:
Date of operation:
Procedures:
Date of assessment:
Follow-up interval:
What problem has your child developed from the cerebral palsy before operation?

1 Pain from persistent spasticity
2 Limited joint motion from fixed spastic contracture
3 Tripping during walking from excessive in-toeing/out-toeing
4 Poor standing posture or walking pattern
5 Poor walking ability
- Slow walking/stiffness/easy fatigue/frequent fall

Has the operation improved the problems your child had before surgery?

1 Pain	Eliminated/Reduced/no change/worsened
2 Joint motion range	Improved/no change/worsened
3 Foot progression	Normalised/No change/worsened
4 Standing posture	Improved/No change/Worsened
5 Walking ability	Improved/No change/Worsened

If you were in the same position again in future, would you choose to have surgery again?
Yes.
Why?
No.
Why?

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