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Original Article

Effect of selective percutaneous myofascial lengthening and functional physiotherapy on walking in children with cerebral palsy: Three-dimensional gait analysis assessment☆

Vasileios C. Skoutelis [a, b, c, *], Anastasios D. Kanellopoulos [d], Stamatis Vrettos [e], Zacharias Dimitriadis [f], Argirios Dinopoulos [a, g], Panayiotis J. Papagelopoulos [a, h], Vasileios A. Kontogeorgakos [a, h]

[a] Medical School, National and Kapodistrian University of Athens, Athens, Attica, Greece
[b] Department of Physiotherapy, Laboratory of Neuromuscular and Cardiovascular Study of Motion, School of Health and Caring Sciences, University of West Attica, Egaleo, Attica, Greece
[c] Department of Physiotherapy, 'Attikon' University General Hospital, Chaidari, Attica, Greece
[d] Department of Orthopaedics, 'Iaso' Children's Hospital, Maroussi, Attica, Greece
[e] 'ENA' Pediatric Physiotherapy Practice, Chalandri, Attica, Greece
[f] Department of Physiotherapy, Health and Quality of Life Research Laboratory, School of Health Sciences, University of Thessaly, Lamia, Greece
[g] Third Department of Paediatrics, 'Attikon' University General Hospital, Chaidari, Attica, Greece
[h] First Department of Orthopaedic Surgery, 'Attikon' University General Hospital, Chaidari, Attica, Greece

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ABSTRACT

Background: Walking is the most affected motor function in children with cerebral palsy (CP). Orthopaedic surgery is regularly used to improve ambulation in children with CP. Selective Percutaneous Myofascial Lengthening (SPML) is considered the state-of-the art technique for surgical lengthening of spastic/contracted muscles in CP. The purpose of this study was to investigate the effect of combined SPML surgery and postoperative functional physiotherapy on gait function and characteristics of children with spastic cerebral palsy (CP).
Methods: Twenty-six children with spastic CP, aged 5−7 years, Gross Motor Function Classification System (GMFCS) levels II (n = 6), III (n = 12) and IV (n = 8) participated in a quasi-experimental one-group pretest-posttest study with a 9-month follow-up. The Global Motion Graph Deviation Index (MGDI) (including MGDI sub-indices of each joint in each plane of motion) and spatiotemporal parameters of a three-dimensional kinematic gait analysis were used to assess the gait function and characteristics, respectively.
Results: Nine months following SPML and functional physiotherapy, statistically significant improvements ($p < 0.05$) were noted in the Global MGDI, the MGDIs of sagittal plane knee and ankle motion analysis graphs, and the four most common spatiotemporal measures of gait: walking velocity, stride length, step length, and cadence.
Conclusion: Children with spastic CP seem to gain better overall gait function following SPML procedure and functional physiotherapy, by achieving higher walking velocity, longer stride length and step length, and faster cadence. Further studies with control group and longer follow-up three-dimensional gait analyses are warranted to validate these positive results.

Abbreviations: CP, cerebral palsy; SPML, selective percutaneous myofascial lengthening; MGDI, motion graph deviation index; GMFCS, gross motor function classification system; 3DIGA, three-dimensional instrumented gait analysis; NSDs, normal standard deviations; GMFM, gross motor function measure; R/L, right and left; non-RCT, non-randomised controlled trial; SEMLS, single-event multilevel surgery.

☆ First Department of Orthopaedic Surgery, National and Kapodistrian University of Athens, 'Attikon' University General Hospital, Postal address: 1 Rimini St, 12462 Chaidari, Attica, Greece.

* Corresponding author. 83 Salaminos St, 18546 Piraeus, Attica, Greece.
E-mail address: vskoutelis@gmail.com (V.C. Skoutelis).

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1. Introduction

Cerebral palsy (CP) is the world's most serious cause of motor disorder in children, which is characterised by atypical neurological and musculoskeletal development [1]. Spastic CP is the most common type of CP, accounting for approximately 80% of all cases [1]. Walking is the most impaired motor function in CP. Ambulant children with bilateral spastic CP demonstrate slower walking velocity, decreased cadence and shorter step length compared to typically developing children [2].

Orthopaedic surgery is one of the most common and popular treatment modalities for improving walking in children with CP [3]. Orthopaedic surgery in terms of single-event multilevel surgery (SEMLS: two or more simultaneous soft-tissue and/or osseous surgical procedures at different levels of the lower limb) [4] is a drastic intervention to simultaneously address the biomechanics of gait [5]. The three-dimensional instrumented gait analysis (3DIGA) has been documented to be a critical measure for an effective surgical decision making and for evaluating the outcomes of an orthopaedic surgery in children with CP [6,7]. However, in order to ensure the optimal functional gain from an orthopaedic surgery, it is necessary to apply an appropriate and effective postoperative physiotherapy programme [3].

Selective Percutaneous Myofascial Lengthening (SPML) represents the newest trend in pediatric orthopaedic surgery [8] for percutaneous lengthening of a spastic shortened muscle [9]. Statistically significant and clinically relevant improvement in gait-related gross motor function were found following SPML procedure and functional physiotherapy in 5-7-year-old children with spastic CP, increasing the amount and level of mobility independence [9]. There are also two-dimensional video-graphic data showed significantly improved ambulatory knee and ankle angle motion following SPML in 3–18-year-old children and adolescents with spastic CP [10,11]. These results are enhanced by the findings of a pilot study suggesting that SPML procedure and functional physiotherapy has the potential to promote overall gait function in 5-7-year-old children with CP, although without demonstrating significant changes in eight of the nine examined spatiotemporal parameters [12]. An expanded study with a larger sample size was consequently needed to better investigate the significance of these preliminary findings and generalise to a larger population [13].

Therefore, the present study was undertaken with a larger sample size of children with spastic CP, in GMFCS levels II to IV, aged 5–7 years, to determine the effects of a combined programme of functional physiotherapy and SPML procedure on gait function and characteristics.

2. Material and methods

2.1. Study design

This was a one-group pretest-posttest study (Fig. 1) of children who participated as experimental group in a non-randomised controlled trial (non-RCT) protocol of SPML procedure and functional physiotherapy [9]. Baseline 3DIGA was performed within 4 weeks prior to orthopaedic surgery and post-operative 3DIGA 9 months after orthopaedic surgery and rehabilitation programme. Nine months was considered a sufficient period of time to observe the changes in gait. 3DIGA data were collected at a gait and motion analysis laboratory. This research has been approved by the institutional review board of authors' affiliated institutions and has been registered at a clinical trial registry. Written informed consent was obtained from the parents of all children prior to enrolment.

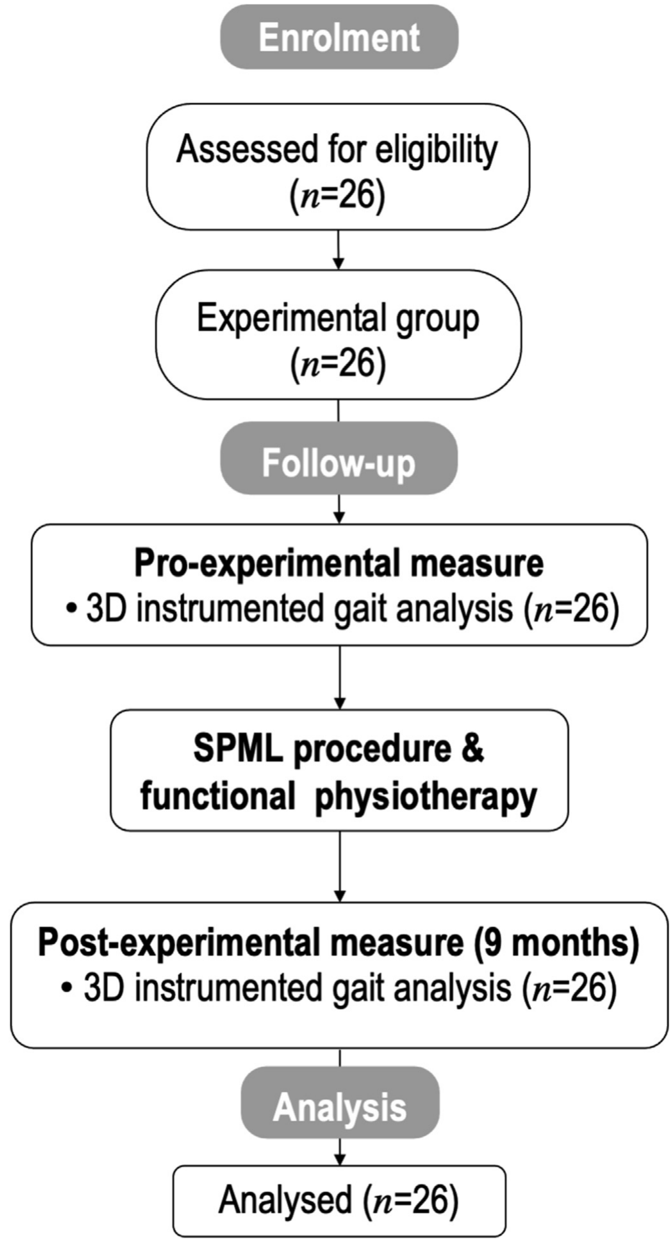

Fig. 1. The flow diagram of the study. 3D, three-dimensional; n, number of participants; SPML, selective percutaneous myofascial lengthening.

2.2. Participants

A G*Power-analysis (power = 80%, significance level = 0.05, effect size = 0.80, two-tailed) revealed that a minimum of 15 participants was needed. To end up with a sufficient number of participants while meeting the needs of the previous non-RCT [9], all children (n = 26) who required SPML intervention and participated in the non-RCT protocol design were included in the one-group pretest-posttest phase of the study. For further details of inclusion and exclusion criteria describe to the non-RCT phase of this research [9].

2.3. SPML procedure and postoperative functional physiotherapy

The combined programme of SPML procedure and a 9-month functional physiotherapy was described in detail previously

[9,12]. Briefly, SPML procedure consisted of lengthening of medial hamstrings (semitendinosus: distal level, semimembranosus and gracilis: middle level) hip adductors, and/or gastrocnemius, as had been indicated. Alcohol blocks (obturator and/or femoral nerves) were an essential part of the SPML procedure in case of excessive reflex-mediated stiffness during passive and active movement of lower limbs. Physiotherapy started on an as-tolerated basis, within a few hours following the SPML procedure, based on an intensive functional strength training programme. The functional training was performed 5–6 times weekly for the first 6 weeks, and then 2–3 times weekly until the end of the 9-month post-operative period. Parent involvement had an active and crucial role during the physiotherapy programme to increase their child's chance for repetitive, varied practice of exercises and activities.

2.4. Kinematic data and processing

3DIGA data were collected and processed with a Vicon three-dimensional motion capture system (Plug-In Gait Lower-Body Marker Set), which has previously been described in detail [12].

Each child was instructed to walk barefoot at self-selected speed along a defined 5 m walkway for five trials (three go and two come back). When the child had not been completed the route, due to falling or stopping walking, the trial would be considered invalid and repeated.

2.5. Outcome measures

Nine spatiotemporal parameters of gait were measured: walking velocity (m/s), cadence (steps/min), step length (m), stride length (m), step time (s), single support time (s), double support time (s), swing time (% of gait cycle) and stance time (% of gait cycle).

The Global Motion Graph Deviation Index (MGDI) [12,14] was used to assess the deviation of overall gait function from normal pattern. The MGDI for each joint-level (pelvis, hip, knee, ankle) in each plane of motion were also calculated. The MGDI, measured in units of Normal Standard Deviations (NSDs), is a valid and reliable measure to quantify gait dysfunction and assess changes following treatment modalities [14].

2.6. Data analysis

Data normality was checked out through Kolmogorov–Smirnov test. Paired sample t-test was applied to compare the pre- and post–experiment mean values of spatiotemporal parameters, Global MGDI, and MGDI of each joint-level in each motion plane.

For each variable, the mean of the five trials from each child was calculated and included. The measurements of the spatiotemporal parameters, obtained from each leg (right, left), were analysed both in the total of lower limbs ($n = 52$), and after dividing the lower limbs into less- and more-affected lower limbs, based on the pre-operative examination of the orthopaedic surgeon. To facilitate the comparison of the examined MGDIs before and after the intervention, the mean of the MGDI for the right and left lower limbs (R/L MGDI) was used.

The level of significance was defined as $p < 0.05$. Statistical analysis was carried out using the SPSS for Mac, version 26.0 (SPSS Inc. Chicago, IL, USA).

3. Results

3.1. Study participants

All the children (16 males, 10 females), mean age 6,15 ± 0,73 years with moderate to severe (GMFCS level II, $n = 6$; level III, $n = 12$; level IV, $n = 8$) spastic CP (tetraplegia, $n = 11$; diplegia, $n = 13$; hemiplegia, $n = 2$), who participated in the non-RCT [9] were completed the 3DIGA measurements.

3.2. Spatiotemporal parameters

Paired sample t-test showed statistically significant ($p < 0.05$) changes in six of the nine spatiotemporal parameters following the intervention, with the significance value differing in some parameters between the total of the lower limbs and the less- or more-affected lower limbs. Specifically, the mean values of walking velocity, stride length and step length increased significantly ($p < 0.05$) both for the total of the lower limbs and separately for the less- and most-affected lower limbs. The mean cadence increased significantly ($p < 0.05$) in the total of the lower limbs but showed a non-statistically significant ($p > 0.05$) increase in the less- and more-affected lower limbs. The mean step time showed a non-statistically significant ($p > 0.05$) decrease both for the total of the lower limbs and separately for the less- and most-affected lower limbs. The mean times of single and double support decreased significantly ($p < 0.05$) in the total of the lower limbs and in the less-affected lower limbs, without showing a statistically significant decrease ($p > 0.05$) in the more-affected lower limbs. The mean values of stance time and swing time showed a non-statistically significant ($p > 0.05$) decrease and increase, respectively, both for the total of the lower limbs and separately for the less- and most-affected lower limbs (Table 1).

3.3. Global MGDI and MGDIs

Paired sample t-test showed that there was a statistically significant ($p < 0.05$) improvement in the mean Global MGDI following the intervention. Apart from statistically significant ($p < 0.05$) improvements in the mean R/L MGDIs of the knee and ankle motion analysis graphs in the sagittal plane and of the ankle in the transverse plane, there were no significant changes ($p > 0.05$) on the mean R/L MGDIs of the remaining joint movements following the intervention (Table 2).

4. Discussion

All children in this study underwent 3D kinematic gait analysis before and after minimal invasive SPML surgery and 9-month functional physiotherapy. This investigation examined the changes in spatiotemporal parameters, which are considered more sensitive measures than the angular kinematics of the joints for the assessment of the degree of motor dysfunction in CP [15]. The overall Global MGDI was also calculated, combined with the changes in the R/L MGDIs for each joint [14]. As demonstrated by the positive results on the most common spatiotemporal measures of gait [16] and the Global MGDI, the combined SPML procedure and functional physiotherapy significantly improved the gait function and characteristics.

4.1. Spatiotemporal parameters of gait

In particular, walking velocity, stride length and step length improved statistically significantly both for the total of the lower limbs, and separately for the less- and more-affected lower limbs. Cadence increased significantly only when calculating the total of the lower limbs. These significant changes in the four most common spatiotemporal measures of gait reflect the increased capacity (using Gross Motor Function Measure [GMFM] dimensions D and E) and performance (using Functional Mobility Scale) in the gait-related gross motor function detected by previous non-RCT [9].

Table 1
Comparison of spatiotemporal parameters before and after the intervention for the total of the legs and for the less- and more affected legs of the participants.

Spatiotemporal parameters	Lower limbs	n	Mean ± SD		Mean difference and 95% CI of the difference			
			Pre	Post	MD ± SD	LCL	UCL	P value
Stride length, m	Totally	52	0.43 ± 0.21	0.53 ± 0.21	0.10 ± 0.12	−0.13	−0.06	**0,001**
	Less-affected	26	0.44 ± 0.21	0.53 ± 0.22	−0.09 ± 0.11	−0.13	−0.04	**<0.001**
	More-affected	26	0.43 ± 0.22	0.53 ± 0.20	−0.10 ± 0.14	−0.16	−0.05	**0.001**
Step length, m	Totally	52	0.22 ± 0.10	0.27 ± 0.10	−0.05 ± 0.65	−0.07	−0.03	**<0.001**
	Less-affected	26	0.23 ± 0.10	0.28 ± 0.11	−0.05 ± 0.06	−0.07	−0.03	**<0.001**
	More-affected	26	0.22 ± 0.11	0.27 ± 0.10	−0.05 ± 0.07	−0.08	−0.02	**0.001**
Walking velocity, m/s	Totally	52	0.34 ± 0.32	0.42 ± 0.27	−0.09 ± 0.21	−0.14	−0.03	**0.004**
	Less-affected	26	0.33 ± 0.32	0.44 ± 0.29	−0.11 ± 0.24	−0.21	−0.02	**0.026**
	More-affected	26	0.32 ± 0.32	0.40 ± 0.26	−0.08 ± 0.17	−0.15	−0.02	**0.019**
Cadence, steps/min	Totally	52	72.32 ± 39.22	83.33 ± 33.45	−11.01 ± 35.80	−20.98	−1.04	**0.031**
	Less-affected	26	73.45 ± 40.05	83.32 ± 33.65	−9.87 ± 36.46	−24.59	4.86	0.180
	More-affected	26	71.18 ± 39.14	83.33 ± 33.91	−12.15 ± 35.82	−26.62	2.32	0.096
Step time, s	Totally	52	1.21 ± 0.84	0.97 ± 0.88	0.25 ± 1.03	−0.04	0.53	0.092
	Less-affected	26	1.25 ± 0.98	0.90 ± 0.62	0.35 ± 0.99	−0.54	0.74	0.087
	More-affected	26	1.18 ± 0.68	1.03 ± 1.09	0.15 ± 1.08	−0.29	0.58	0.497
Single support time, s	Totally	52	0.52 ± 0.21	0.44 ± 0.11	0.08 ± 0.19	0.03	0.14	**0.004**
	Less-affected	26	0.56 ± 0.25	0.45 ± 0.11	0.11 ± 0.24	0.01	0.20	**0.028**
	More-affected	26	0.49 ± 0.13	0.44 ± 0.11	0.05 ± 0.14	0.00	0.11	0.050
Double support time, s	Totally	52	1.31 ± 1.19	0.89 ± 0.83	0.42 ± 1.14	0.11	0.74	**0.010**
	Less-affected	26	1.29 ± 1.20	0.83 ± 0.62	0.46 ± 1.11	0.01	0.91	**0.044**
	More-affected	26	1.34 ± 1.19	0.95 ± 1.01	0.38 ± 1.19	−0.10	0.86	0.111
Stance time, %	Totally	52	72.47 ± 9.12	71.77 ± 8.27	0.70 ± 8.03	−1.54	2.93	0.534
	Less-affected	26	73.43 ± 9.35	72.20 ± 7.97	1.23 ± 8.05	−2.03	4.48	0.445
	More-affected	26	71.51 ± 8.96	71.34 ± 8.70	0.17 ± 8.14	−3.12	3.46	0.917
Swing time, %	Totally	52	27.53 ± 9.12	28.23 ± 8.27	−0.70 ± 8.03	−2.93	1.54	0.534
	Less-affected	26	26.57 ± 9.35	29.91 ± 14.52	−3,34 ± 12.92	−8.56	1.88	0.199
	More-affected	26	28.49 ± 8.96	28.66 ± 8.70	−0.17 ± 8.14	−3.46	3.12	0.917

CI, confidence interval; L/UCL, lower/upper confidence limit; MD, mean difference between pre- and post-experimental measurements; n, number of the lower limbs; SD, standard deviation.
Bold type = statistically significant ($p < 0.05$).

Table 2
Comparison of the Global MGDI and MGDIs of each examined joint (pelvis, hip, knee, ankle) in the three planes (sagittal, frontal, transverse) before and after the intervention.

Deviation index	n	Mean ± SD		Mean difference and 95% CI of the difference			
		Pre	Post	MD ± SD	LCL	UCL	P value
Global MGDI (NSDs)	26	1.95 ± 0.39	1.85 ± 0.31	0.10 ± 0.24	0.01	0.20	**0.037**
R/L MGDI (NSDs) in sagittal plane							
Pelvis motion	26	2.11 ± 1.42	2.48 ± 1.32	−0.37 ± 1.06	−0.79	0.06	0.088
Hip motion	26	1.72 ± 0.86	1.70 ± 0.87	0.02 ± 0.61	−0.23	0.26	0.900
Knee motion	26	3.68 ± 1.40	2.89 ± 0.82	0.79 ± 1.49	0.19	1.39	**0.012**
Ankle motion	26	1.96 ± 0.76	1.66 ± 0.90	0.30 ± 0.69	0.02	0.58	**0.034**
R/L MGDI (NSDs) in frontal plane							
Pelvis motion	26	2.60 ± 1.14	2.40 ± 0.98	−0.20 ± 0.99	−0.20	0.60	0.314
Hip motion	26	1.29 ± 0.28	1.41 ± 0.38	−0.12 ± 0.31	−0.25	0.00	0.054
Knee motion	26	1.35 ± 0.45	1.37 ± 0.62	−0.02 ± 0.78	−0.33	0.29	0.897
Ankle motion	26	0.81 ± 0.33	0.85 ± 0.36	−0.04 ± 0.57	−0.27	0.18	0.686
R/L MGDI (NSDs) in transverse plane							
Pelvis motion	26	3.14 ± 1.17	2.74 ± 1.45	0.40 ± 1.07	−0.02	0.83	0.066
Hip motion	26	1.60 ± 0.65	1.84 ± 0.86	−0.24 ± 1.02	−0.66	0.17	0.234
Knee motion	26	0.71 ± 0.27	0.88 ± 0.44	−0.17 ± 0.49	−0.37	0.03	0.091
Ankle motion	26	2.45 ± 0.63	1.94 ± 0.72	0.51 ± 0.71	0.23	0.80	**0.001**

CI, confidence interval; L/UCL, lower/upper confidence limit; MD, mean difference between pre- and post-experimental measurements; MGDI, Motion Graph Deviation Index; NSDs, (units of) Normal Standard Deviations; n, number of the lower limbs; R/L MGDI, mean value of MGDI of a joint for the right and left lower limbs; SD, standard deviation.
Bold type = statistically significant ($p < 0.05$).

Indeed, walking velocity, cadence and stride length have a moderate to strong positive correlation with the GMFM dimensions D and E [17], while providing a better overview of overall gross motor function in children with CP [15]. This significant improvement in walking velocity has confirmed the research hypothesis of Wild [11], who found a non-statistically significant increase in walking velocity (using video graphic analysis) after SPML surgery (mean postoperative follow-up time: 7.5 months, range: 3–13 months) in 31 children aged 4–18 years (mean age: 8.5 years) with spastic CP and GMFCS level II-IV.

These findings are consistent with the results of Gordon et al. [18] who found a significant increase in walking velocity and stride length, but no significant change in cadence, after percutaneous hamstring tenotomy surgery in 48 ambulatory children with spastic CP (GMFCS I-III), aged 4.5–15.9 years (mean age: 9.5 years). Besides, according to recent systematic reviews [4,16,19,20], varied results have emerged from studies following open or percutaneous muscle-tendon lengthenings as part of SEMLS. The majority of these studies show that walking velocity and cadence remain almost unchanged, with increasing or decreasing trends, at one year postoperatively and improve in the following years, with stride/step length not changing or increasing significantly [16,19,20]. In a systematic review by Lamberts et al. [16] it was found that significant changes in walking velocity, cadence, and stride length occurred in only 31%, 39%, and 46% of studies, respectively. These non-significant results of the majority of studies, which are basically non-comparative studies [16], are confirmed by a non-RCT by Gough et al. [7] who found that walking velocity, cadence, and stride length did not show any significant changes between 3DIGAs (mean time: 16 months) both in ambulatory children with spastic diplegic CP (n = 12, mean age: 10 years and 1 month) who underwent SEMLS, and control children (n = 12, mean age: 9 years and 10 months) who received the usual physiotherapy treatment.

Dreher et al. [21] observed a significant increase in stride length at two to four years after the SEMLS in ambulatory children with spastic CP, aged 6–16 years (mean age: 10.3 years), but this improvement was significant only for the group (n = 60) with medial hamstring lengthening and not for the group (n = 18) with combined medial and lateral hamstring lengthening. In the present study, SPML surgery of the hamstrings, performed on 92% of children, appears to have contributed greatly to the significant improvement in walking velocity, step/stride length and overall gait function based on claims by Laracca et al. [22]. Specifically, Laracca et al. [22] found that walking velocity and step length deteriorated significantly when hamstring lengthening was not included in the SEMLS package in children with CP.

In the current study, lengthening of the medial hamstrings (semimembranosus, semitendinosus, gracilis) was only performed. Such a decision is based on studies that have shown that the combined medial and lateral hamstring lengthening lead to a significantly increased anterior pelvic tilt at one year postoperatively [21,23]. An increased anterior pelvic tilt postoperatively causes a deterioration in “sagittal plane balance” and may lead to increased lumbar lordosis, back pain, spinal instability (spondylolysis, spondylolisthesis), compensatory movements, recurrent pathological gait pattern and, consequently, deterioration of gait function [21,24,25]. The non-statistically significant change of the mean R/L MGDI in the sagittal plane pelvic motion analysis graph following SPML surgery indicates a non-significant increase in anterior pelvic tilt. De Mattos et al. [26] have even argued that “*if the pelvic tilt remains the same or increases mildly, it means that there has actually been an improvement on the pelvic position*” given that when the flexed knee gait is corrected, the femur becomes more vertical position during the stance phase and, thus, anterior pelvic tilt is inevitable.

4.2. Motion graph deviation indices

Furthermore, taking into account the basic principle of the concept of "surgical dose" (that is, the surgical technique that provides the appropriate amount of muscle lengthening to avoid overcorrection) [21,23], the minimally invasive nature of the SPML method appears to have contributed not only to a non-deteriorated or non-recurrent condition, but to an improved gait pattern, as confirmed by the significant improvements of the MGDIs of sagittal plane knee and ankle motion analysis graphs and, mainly, by the significant improvement of the Global MGDI. The MGDI reflects the severity of lower limb joint motion pathology, with the Global MGDI index quantifying the deviation of the overall gait function from normal values. A decrease in the index value means an improvement to a more normal function [14].

These results reinforce the findings of two studies by Wild [11] and her colleagues [27], who illustrated statistically significant improvements ($p < 0.05$) in two-dimensional sagittal plane knee and ankle joint kinematic parameters during walking following SPML procedure in hamstrings, hip adductors, and gastrocnemius. There are many literature reports supporting improvements in sagittal plane knee and ankle kinematics and overall gait quality as well at 12 months following muscle-tendon lengthenings in children with spastic CP, mainly as part of SEMLS [4,16,19].

4.3. Limitations

An important methodological limitation of the present study is that it has no comparison group. Although this one-group pretest-posttest design is weak, threatening the validity of the findings [13], this choice was practically imposed. This is because 3DIGA was performed as part of a standard preoperative evaluation and postoperative follow-up of children underwent SPML surgery. Given the high cost of 3DIGA and also the absence of funding, performing 3DIGA in a control group was not possible. Nevertheless, there is evidence [28] documenting the progressive deterioration of joint kinematics and spatiotemporal characteristics of gait over time in children with spastic CP, who were given conventional rehabilitation therapies (physical, occupational therapies) and no orthopaedic or neurosurgical interventions. Therefore, this evidence justifies and supports [13] the choice of investigating only the changes expected in walking variables following a combined programme of SPML procedure and functional physiotherapy. Besides, there were no repeated measurements over time to record changes in walking variables. Another limitation could also be the use of the Global MGDI, for which its clinical usefulness has not been sufficiently confirmed, due to the limited number of studies using it [12,14,29] in relation to the most used summary measures, the Gait Profile Score and Gait Deviation Index [30].

Institutional ethical committee approval

The Scientific and Ethical Council of the ‘Attikon’ University General Hospital, Chaidari, Attica, Greece (*EBΔ* 2199/14-03-2017) approved the study.

Clinical trial registry

The study has been registered at the Australian New Zealand Clinical Trials Registry (ACTRN12618001535268).

Declaration of competing interest

None.

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