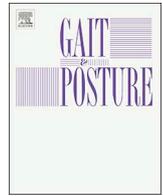




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Pre-operative hamstring length and velocity do not explain the reduced effectiveness of repeat hamstring lengthening in children with cerebral palsy and crouch gait

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ABSTRACT

Background: Hamstring lengthening surgery (HSL) is often performed to correct crouch gait in patients with cerebral palsy (CP). However, crouch can recur over time, and repeat HSL may be ineffective. One possible reason is that the hamstrings in repeat HSL patients are neither short nor lengthening slowly and would therefore not benefit from HSL.

Research question: This study aimed to determine whether the hamstrings are short and/or slow preoperatively only in patients with primary, and not repeat, HSL.

Methods: We compared pre- and postoperative dynamic semimembranosus muscle-tendon lengths for children with CP who had primary (N = 15) or repeat (N = 8) HSL to a group of control participants (N = 10). Outcome measures were compared between visits (pre- vs. postoperative) and groups (control, primary HSL, repeat HSL) using mixed model analysis.

Results: Preoperatively, hamstrings were shorter and slower than normal on average in both HSL groups ($p < 0.001$); all but 3 limbs (primary 26/28, repeat 13/14) had hamstrings that were shorter and/or slower than controls by more than two standard deviations. Postoperative improvements were observed in the primary HSL group for popliteal angle, initial contact knee flexion, minimum stance knee flexion, and dynamic hamstring length ($p \leq 0.001$). The repeat HSL group improved only in dynamic hamstring length ($p = 0.004$) and worsened in passive knee extension ($p = 0.01$) and minimum hip flexion in stance ($p = 0.04$). Hamstrings in both surgical groups on average remained shorter and slower than controls postoperatively ($p \leq 0.001$).

Significance: The fact that repeat HSL is less effective in improving knee motion is not due to a lack of short or slow hamstrings preoperatively. However, in recurrent crouch, short or slow hamstrings do not usually indicate hamstring dysfunction, and correction of other deformities such as rotational malalignment, fixed knee flexion contractures, patella alta, weak calf muscles, and/or loose heelcords should be considered rather than repeat HSL.

1. Introduction

Crouch (flexed knee) gait is a common problem in children with cerebral palsy (CP), progressing in severity with age and growth [1–4]. Surgical lengthening of the hamstrings (HSL) is often performed to correct crouch gait. Previous research has demonstrated the effectiveness of HSL in reducing hamstring contracture and improving knee extension at initial contact and throughout the stance phase of gait [5–12].

Despite the overall effectiveness of HSL in improving hamstring range of motion and knee extension during gait, hamstring contractures

and crouch gait can recur over time in patients with CP. One study showed a need for repeat surgical intervention in 17% of cases [12]. Unfortunately, repeat HSL may not be as effective as primary HSL in improving static and dynamic knee extension. Two studies showed some improvement in knee position at initial contact and in the stance phase of gait after repeat HSL [5,13], while another study reported improvement at initial contact, but not in stance [14]. In contrast, a study done recently at our institution showed improved knee kinematics and passive range of motion only after primary, but not repeated, HSL [15].

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The ability to estimate dynamic muscle-tendon lengths from three-dimensional (3-D) joint kinematic data has provided greater insight into hamstring function during gait in patients with CP. Studies have shown that in ~70–80% of cases of crouch gait in CP the hamstrings are not actually shorter than normal during walking despite excessive knee flexion, owing to the hamstrings' bi-articular structure and the flexed posture of the hip [16,17]. Short muscle-tendon length may reflect hamstring tightness due to contracture, while slow muscle-tendon lengthening velocity may indicate resistance to stretch due to hamstring spasticity. Hamstring lengthening surgery could address the former by reducing passive tension in the muscle-tendon unit and the latter by attenuating the spastic response [17]. However, the absence of short or slow hamstrings may indicate that crouch is due to factors other than hamstring dysfunction and treatments other than hamstring lengthening are needed. In fact, previous research has shown a relationship between preoperative hamstring length and velocity and postoperative changes in hamstring length, hamstring velocity, and knee extension during gait [17–19]. In patients with short or slow hamstrings, short hamstrings tend to become longer, slow hamstrings tend to become faster, and knee extension tends to improve after hamstring lengthening surgery [17,18]. For cases in which the hamstrings are neither short nor slow, however, hamstring lengthening is unlikely to improve knee extension and may worsen pelvic tilt [17,18]. Hamstring length and velocity can therefore serve as predictors of surgical outcome, along with other clinical characteristics including lower extremity rotational alignment and muscle strength [19].

The reasons for recurrence of crouch gait after hamstring lengthening surgery and the lower effectiveness of repeat HSL are unclear. One possibility is that in cases of recurrent crouch the hamstrings are not short or lengthening slowly, and are therefore unlikely to benefit from hamstring lengthening. We hypothesized that patients undergoing primary HSL would have short and/or slow hamstrings preoperatively that would become longer and/or faster after HSL, resulting in improved knee extension, while patients undergoing repeat HSL would not have short or slow hamstrings preoperatively and would therefore not change hamstring length, hamstring velocity, or knee extension after surgery. Therefore, the aims of this study were 1) to determine if the hamstrings were short and/or slow preoperatively only in the primary, and not the repeat, HSL group, 2) to corroborate whether significant improvements in knee range of motion (popliteal angle, knee extension during gait) occurred only in the primary HSL group, and 3) to determine whether improvements in dynamic knee extension were accompanied by increased hamstring length and/or velocity.

2. Methods

A chart review was conducted to identify children with CP under 15 years of age who had primary or repeat hamstring lengthening surgery with pre- and postoperative gait analysis at our institution between November 2005 and March 2016. Limbs were excluded if they had undergone bony correction of knee flexion contractures in addition to HSL (such as anterior distal femoral hemiepiphysiodesis or extension osteotomy with or without patellar tendon advancement). The retrospective patient data were accessed under a waiver of consent granted by our institutional review board (IRB). A convenience sample of controls who had undergone gait analysis for a previous study [20] was also included. Written assent and consent were obtained from control participants and their parents/guardians, and protocols approved by our IRB were followed.

All study participants had undergone gait analysis testing using an 8–10 camera 3-D motion capture system (Vicon 612 or Nexus 2, Vicon Motion Systems Ltd., Oxford, UK). Reflective markers were placed on the patients' legs and pelvis following the Plug-in-Gait implementation of the conventional gait model [21] with a marker on the center of the patella in place of thigh wands [22] and in some cases a marker on the tibia crest in place of tibia wands [23]. Marker position data were

collected at 120 Hz during barefoot walking at a self-selected speed using commercial software (Vicon Workstation or Nexus 2). The marker data were analyzed in the musculoskeletal modeling software OpenSim [24] to calculate joint angles and muscle-tendon lengths. Specifically, a generic model [25] was scaled to each participant based on anatomic marker positions during a static trial. Joint angles during a single representative trial of walking were then calculated using inverse kinematics in which the model was positioned to minimize the difference between the experimental (participant) and model marker positions. Muscle-tendon length of the semimembranosus (SM) was determined across the gait cycle for each surgical and control limb. SM was selected because it is one of the most frequently lengthened hamstring muscles in CP and because SM length correlates strongly with the length of the other hamstring muscles [26,27]. After smoothing with a 2-way 4th order Butterworth filter with a cutoff frequency of 6 Hz, SM length was numerically differentiated to determine SM lengthening velocity. To account for differences in size among participants, SM length and velocity were normalized by SM length with the model positioned in the anatomic position. Walking speed was normalized by $\sqrt{gL_{leg}}$ to calculate non-dimensional walking speed, where g is the acceleration due to gravity and L_{leg} is the participant's average leg length.

Joint range of motion measures were taken at each gait analysis test session by one of three experienced gait laboratory physical therapists using standardized procedures. Measures included passive knee extension, popliteal angle (maximum knee extension with the hip flexed to 90°) to assess hamstring tightness/contracture, and hip extension (maximum hip extension with the opposite hip flexed to flatten the lumbar spine). Hamstring spasticity was measured clinically using the modified Ashworth scale.

All surgeries were performed by the same surgeon except for one case which was performed by another surgeon from the same group. The extent of hamstring lengthening done varied among patients and between groups, depending on the severity of hamstring contracture and the surgeon's common practice at the time of operation (percutaneous HSLs being done more recently than open ones). Details of the hamstring surgeries done are shown in Table 2.

Participant characteristics were compared among the control, primary HSL, and repeat HSL groups using ANOVA with Bonferroni adjusted post-hoc t-tests for continuous variables and Fisher's exact test for categorical variables. Outcome measures were also compared among groups and visits using a linear mixed model with participant as a random effect and side as a fixed effect to account for the inclusion of two sides for some participants. An indicator variable was created for each combination of group (control, primary HSL, repeat HSL) and visit and was included as a fixed effect in the mixed model. Pairwise comparison of means was then performed to examine pre-specified comparisons (comparison of groups pre- and postoperatively, pre- vs. postoperative within each HSL group). Prior to running the mixed model analyses, outcome variables were checked for excessive skewness graphically, and none was found. The primary outcome measures were peak hamstring length and peak hamstring lengthening velocity; secondary outcome measures included hip and knee flexion at initial contact, minimum hip and knee flexion (peak extension) during the stance phase of gait, and passive range of motion of the hip and knee (popliteal angle, hip and knee extension). All statistical analysis was performed in Stata (version 14.0, StataCorp LP, College Station, TX, USA) with a significance level of 0.05.

3. Results

A total of 23 patients and 10 controls were included in the analysis. Fifteen patients had undergone primary HSL (13 bilateral), and 8 had undergone repeat HSL (7 bilateral). One limb was excluded from analysis in the repeat HSL group due to anterior distal femoral hemiepiphysiodesis on one side in a patient with bilateral HSL (i.e., only the side without hemiepiphysiodesis was analyzed). As might be expected,

Table 1
Participant characteristics at baseline.

	Control (N = 10)	Primary (N = 15)	Repeat (N = 8)	P-value
Sex, male	8 (80%)	7 (47%)	5 (63%)	0.29
Age at baseline (yr)	10.6 (2.1)	8.6 (2.4)	12.6 (1.2)†	0.0005
Age at surgery (yr)	—	9.0 (2.5)	13.0 (1.3)†	0.0004
Time baseline to surgery (months)	—	4.6 (2.8)	5.0 (3.0)	0.70
Time surgery to follow-up (yr)	—	1.4 (0.7)	1.9 (1.1)	0.22
GMFCS	—			0.44
I		2 (13%)	0 (0%)	
II		8 (53%)	3 (38%)	
III		5 (33%)	5 (63%)	
Bilateral HSL	—	13 (87%)	7 (88%)	1.00
Walking speed (m/s)	1.3 (0.1)	0.7 (0.3)*	0.8 (0.2)*	< 0.0001
Non-dimensional walking speed	0.36 (0.03)	0.18 (0.07)*	0.21 (0.07)*	< 0.0001

Continuous variables are presented as mean (SD). Categorical variables are presented as N (%). Comparison among groups was performed using ANOVA with Bonferroni adjusted post-hoc tests for continuous variables and Fisher's exact test for categorical variables.

*indicates p < 0.05 vs. Control; † indicates p < 0.05 vs. Primary group.

Table 2
Hamstring lengthening procedures done and concomitant surgeries.

Hamstring Lengthening	Primary (N = 28 sides)	Repeat (N = 14 sides)
Muscles Lengthened		
Medial	16 (57%)	3 (21%)
Medial and Lateral	12 (43%)	11 (79%)
Approach		
Percutaneous	24 (86%)	1 (7%)
Open	4 (14%)	13 (93%)
Concomitant Procedures		
Femur Derotation	19 (68%)	5 (36%)
Hip Adductor Release	18 (64%)	2 (14%)
Tibia Derotation	15 (54%)	2 (14%)
Foot Bony Procedures	8 (29%)	8 (57%)
Gastrocnemius Recession	8 (29%)	2 (14%)
Distal Rectus Femoris Transfer	4 (14%)	2 (14%)
Tendo-Achilles Lengthening	1 (4%)	1 (7%)
Flexor Digitorum Longus Lengthening	1 (4%)	0
Split Posterior Tibialis Tendon Transfer	1 (4%)	0
Split Anterior Tibialis Tendon Transfer	1 (4%)	0
Posterior Tibialis Tendon Lengthening	1 (4%)	0
Hardware Removal	0	2 (14%)
None	4 (14%)	0
Spasticity (modified Ashworth scale)		
0	2 (7%)	5 (36%)
1	18 (64%)	4 (29%)
1+	6 (21%)	5 (36%)
2	2 (7%)	0

the group with repeat HSL was older than the primary HSL group (Table 1). The groups did not differ in terms of sex or time from baseline to surgery and from surgery to follow-up. There was a higher proportion of participants at Gross Motor Function Classification System (GMFCS) levels I and II in the primary HSL group, and at level III in the repeat HSL group, though this difference did not reach the level of statistical significance. Both HSL groups walked more slowly than the controls. The repeat HSL group had a higher proportion of open HSL procedures (p = 0.048) and procedures involving both the medial and lateral hamstrings (p < 0.001) (Table 2). Concomitant surgeries are shown in Table 2.

Compared to controls, both the primary and repeat HSL groups had higher mean values for preoperative initial contact knee flexion and minimum stance knee flexion, with lower peak hamstring length and peak hamstring lengthening velocity (Fig. 1, Table 3). Mean values for baseline range of motion, kinematic variables, and hamstring length/

velocity did not differ between the primary and repeat HSL groups.

Statistically significant pre- to postoperative improvements were observed in the primary HSL group for popliteal angle, initial contact knee flexion, minimum stance knee flexion, and hamstring length (Table 3). All joint angles improved by > 10° on average, which is clinically significant. The repeat HSL group improved only in terms of hamstring length and worsened in passive knee extension and minimum hip flexion in stance. Both groups had minimal knee flexion contracture (restriction of passive knee extension range) both pre- and post-operatively. Knee flexion at initial contact improved by > 10° in 20/28 (71%) of limbs in the primary HSL group compared with 3/14 (21%) in the repeat group (p < 0.001).

At the postoperative visit, both groups still had greater average knee flexion at initial contact and in the stance phase of gait compared to controls, as well as lower average peak hamstring length and lengthening velocity. The repeat HSL group exhibited higher postoperative knee flexion at initial contact and minimum knee flexion angles (less knee extension) than the primary HSL group during gait, along with greater popliteal angles and less passive knee extension.

With regards to classifying hamstrings as short and/or slow, all limbs in the surgical groups had hamstrings that were dynamically shorter and/or slower than controls by at least 1 standard deviation (SD) preoperatively, and all but 3 limbs (2 primary, 1 repeat) had preoperative hamstring lengths and/or velocities > 2 SD below normal (Table 4). Short hamstrings at the 2SD threshold improved to within 2SD of normal for 13/14 limbs (93%) in the primary HSL group and 2/7 limbs (29%) in the repeat HSL group (Table 4). Only 4/25 slow hamstrings (16%) in the primary HSL group and 0/13 in the repeat group improved to within 2SD of normal postoperatively.

4. Discussion

Consistent with our previous study, which had 17 limbs in common with the current study [15], primary HSL surgery produced better outcomes than repeat HSL. The primary HSL group had improvements of 14.3° in knee flexion at initial contact and 11.6° in minimum knee flexion in stance, compared to improvements of only 4.0° and 2.5° for the same measures in the repeat HSL group. In addition, while minimum hip flexion in stance was essentially unchanged in the primary HSL group (change of 0.1°), minimum hip flexion in stance worsened by 6.0° in the repeat HSL group; passive knee extension also worsened by 6.8° in the repeat HSL group.

The improved knee range of motion in the primary HSL group was accompanied by increased hamstring length. Of 14 limbs with short hamstrings before surgery, only 1 remained short after surgery (Table 4). Although the repeat HSL group showed a similar average increase in hamstring length (Table 3), 5/7 short hamstrings remained short postoperatively (Table 4). Neither group showed a significant change in hamstring velocity with surgery (Table 3); 21/25 slow hamstrings remained slow postoperatively in the primary HSL group, and all 13 slow hamstrings remained slow in the repeat HSL group postoperatively. The high percentage of slow hamstrings may be due to the patients' slow walking speed. Previous studies have found that walking speed has a small effect on hamstring length (1% change with doubling of walking speed) but a large effect on hamstring velocity (40% change with doubling of walking speed) [28,29]. We could not classify hamstring velocity using speed-matched norms because our controls only walked at a self-selected speed. While this would make it more difficult for hamstring velocity to improve to within the normal range, it should also be emphasized that hamstring velocity did not change significantly in either patient group postoperatively (Table 3). This may be because our patients had only mild spasticity which was likely not a major cause of slow hamstring velocity or crouch.

The lower effectiveness of repeat HSL does not appear to be related to preoperative hamstring length or velocity, which were similar between the two surgical groups. Approximately half of limbs in both the

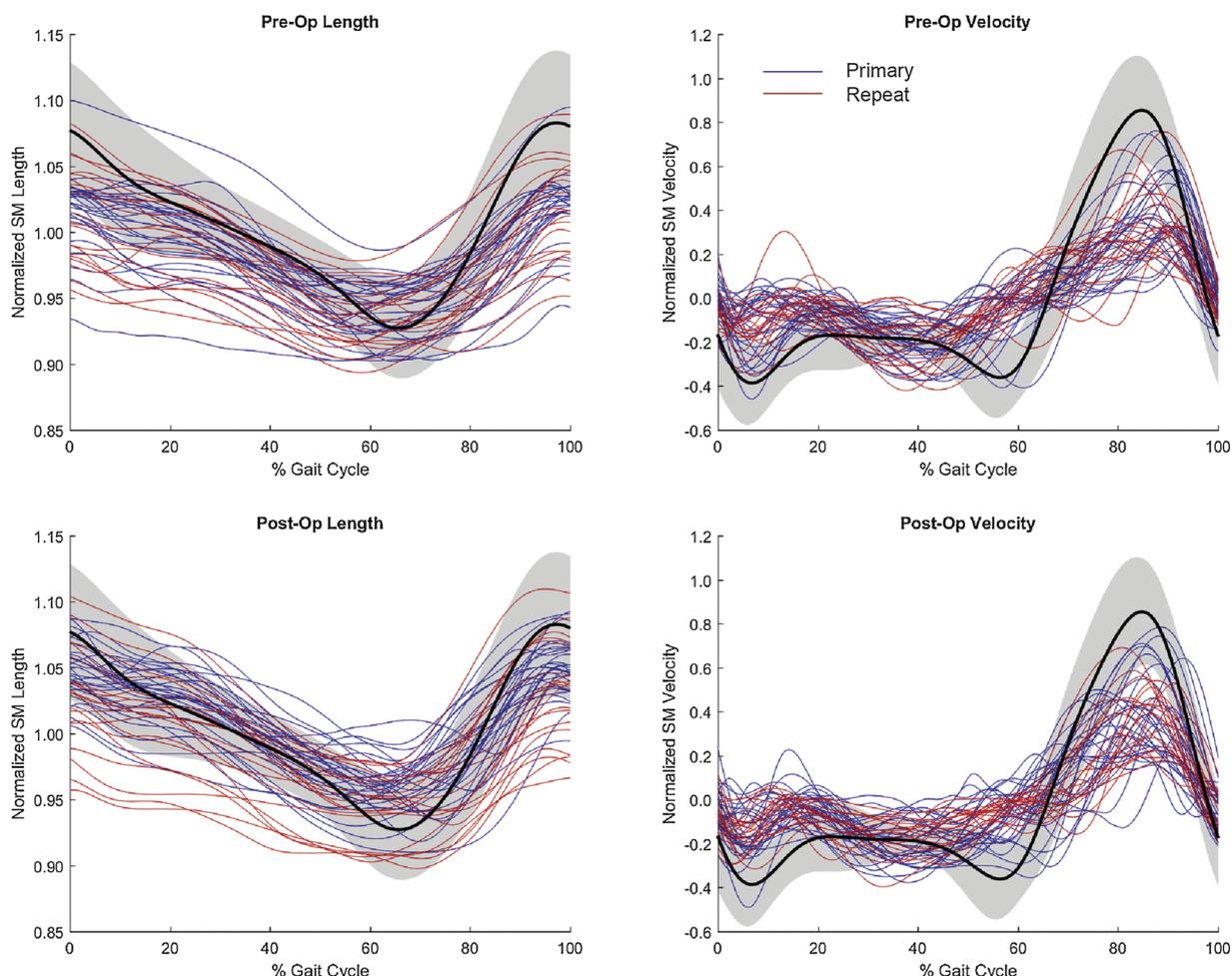


Fig. 1. Normalized hamstring length and velocity for primary and repeat HSL. The black line and grey shaded region represent the mean ± 2 standard deviations of controls.

Table 3
Within-group and between-group comparison of static and dynamic measurements.

	Control (N = 20 sides)	Primary (N = 28 sides)				Repeat (N = 14 sides)			
		Pre	Post	Change	P Change	Pre	Post	Change	P Change
Popliteal Angle (°)	—	53.4 (9.9)	40.5 (10.1)	-12.9 (14.2)	< 0.001	51.4 (9.1)	50.7 (10.5) [†]	-0.7 (11.6)	0.52
Knee Extension (°)	—	-5.4 (5.9)	-4.1 (4.7)	1.3 (5.9)	0.30	-2.9 (4.7)	-9.6 (6.3) [†]	-6.8 (7.2)	0.01
Hip Extension (°)	—	-2.3 (3.7)	-1.4 (3.6)	0.9 (3.9)	0.27	0.0 (0.0)	-2.1 (6.4)	-2.1 (6.4)	0.06
IC Knee Flexion (°)	11.4 (6.2)	40.6 (12.0)*	26.3 (10.4)*	-14.3 (12.6)	< 0.001	41.7 (8.9)*	37.6 (14.3)* [†]	-4.0 (13.6)	0.22
IC Hip Flexion (°)	36.7 (11.5)	40.3 (5.8)	34.8 (16.0)	-5.5 (17.2)	0.056	36.7 (17.4)	40.7 (10.7)	4.0 (10.2)	0.32
Min Knee Flexion Stance (°)	-0.1 (10.0)	27.0 (13.1)*	15.4 (9.7)*	-11.6 (12.8)	< 0.001	28.4 (10.5)*	25.9 (14.0)* [†]	-2.5 (10.5)	0.43
Min Hip Flexion Stance (°)	3.8 (6.9)	3.5 (10.1)	3.4 (10.3)	-0.1 (12.4)	0.95	1.0 (13.9)	7.1 (12.7)	6.0 (13.9)	0.04
Max Hamstring Length ^a	1.08 (0.03)	1.02 (0.03)*	1.05 (0.02)*	0.03 (0.03)	< 0.001	1.02 (0.03)*	1.04 (0.04)*	0.02 (0.03)	0.004
Max Hamstring Velocity ^b	0.87 (0.13)	0.40 (0.16)*	0.45 (0.17)*	0.05 (0.13)	0.08	0.39 (0.14)*	0.41 (0.13)*	0.03 (0.18)	0.47

Results are presented as mean (SD); IC = initial contact; min = minimum; max = maximum.

*indicates p < 0.05 vs. Control; † indicates p < 0.05 vs. Primary.

^aNormalized to length in the anatomic position. Values < 1.0 indicate shorter length than in the anatomic position, > 1.0 indicate longer length than in the anatomic position.

^bNormalized length per second.

primary and repeat HSL groups had hamstrings more than 2SD shorter than normal, and almost all hamstrings in both groups were either short or slow or both. While normal dynamic hamstring length and velocity may be a potential contraindication to surgical hamstring lengthening because they suggest that the hamstrings are not restricting motion abnormally during gait [17], having short and/or slow hamstrings only implicates the hamstrings as a potential, and not necessarily an actual,

cause of crouch. In the case of repeat HSL, it appears that the hamstrings are not usually a primary contributor to crouch gait despite being short and slow. Other factors may play a larger role, and treatments other than hamstring lengthening may be needed.

Other possible causes of recurrent crouch include lever arm dysfunction caused by rotational malalignment between the upper and lower leg, fixed knee flexion contractures, and quadriceps insufficiency

Table 4

Limbs with short hamstring length or slow lengthening velocity (> 2SD below normal) pre- and post-operatively for (a) primary HSL group and (b) repeat HSL group.

		Post-op				Total pre-op
		Neither short nor slow	Short only	Slow only	Both short and slow	
Pre-op	Neither short nor slow	2	0	0	0	2 (7%)
	Short only	1	0	0	0	1 (4%)
	Slow only	1	1	9	1	12 (43%)
	Both short and slow	2	0	10	1	13 (46%)
	Total post-op	6 (21%)	1 (4%)	19 (68%)	2 (7%)	28
Repeat HSL		Post-op				
		Neither short nor slow	Short only	Slow only	Both short and slow	Total pre-op
Pre-op	Neither short nor slow	0	0	1	0	1 (7%)
	Short only	0	0	0	0	0
	Slow only	0	0	6	0	6 (43%)
	Both short and slow	0	0	2	5	7 (50%)
	Total post-op	0	0	9 (64%)	5 (36%)	14

due to patella alta [19,30]. Patella alta and quadriceps insufficiency, in particular, can result from prolonged flexed-knee gait [31], and failure to address over-stretched patellar tendons may contribute to recurrent or residual crouch after hamstring lengthening [32]. These problems can be addressed through surgical shortening of the patellar tendon (patella tendon advancement) combined with distal femoral extension osteotomy in cases of fixed knee flexion contracture [32,33]. Quadriceps insufficiency likely contributed to recurrent crouch in our participants since none had undergone these procedures, which were not routinely performed at our institution at the time these patients were treated.

The percentage of short and slow hamstrings in our cohort is higher than those reported in other literature, which have reported lower percentages. Arnold et al. found that in 35% of cases of children with CP and crouch gait the hamstrings were neither short nor slow [18] compared with only 7% in our study. The reason for the discrepancy in rates is unclear. Participant characteristics were similar, including age, popliteal angle, and knee kinematics at initial contact and in stance. However, our study included participants who walked with ambulatory aids (GMFCS III), and our sample may have had more severe neurologic involvement and greater hamstring dysfunction. There may have been a difference in patient selection criteria for surgical intervention for crouch gait or differences in the technique of the hamstring lengthening procedures between the two institutions affecting hamstring length and velocity in patients with recurrent crouch.

The current study has similar limitations to other retrospective cohort studies. Because of the retrospective nature of the study, we could not control postoperative management (therapy and bracing) or rehabilitation. Given the vagaries of our healthcare system, we are often unable to perform routine postoperative gait studies in patients who are not deemed to be in need of additional surgery. Therefore, some of our postoperative studies were likely done in patients who had recurrent or new gait problems requiring further intervention, which may bias our sample toward patients with poorer outcomes. Additionally, though not statistically significant, the repeat HSL group had a higher percentage of participants with lower ambulatory function (GMFCS III) which could have contributed to their poorer outcome. However, in our prior study [15] the percentage of participants at GMFCS level III was more balanced between the primary (38%) and repeat (50%) groups, and the results were similar to the current study. Crouch is known to increase with age in children with CP [1,2,4,31]. The repeat HSL group was significantly older (average age 13.0 years) than the primary HSL group (average age 9.0 years), and older age could contribute to the lack of improvement in the repeat HSL group. However, age-related differences in body size should not have affected the analysis since the muscle-tendon length data were normalized to subject-specific length in the

anatomic position. Greater muscle weakness and prior surgery, which were not considered in this study, could also predispose the repeat HSL group to worse outcomes. Walking velocity also was not controlled in this study, but was similar between the two HSL groups. Finally, while the musculoskeletal model utilized is well described in the literature [25] and is often used to model children with CP, it is not fully known how well these models represent children with disabilities; we also did not model anatomic differences such as femoral anteversion in a subject-specific manner.

In conclusion, children with CP who underwent repeat HSL showed limited improvement in gait measures postoperatively, but as a group did not have worse preoperative kinematics or hamstring length/velocity than the group of patients undergoing primary HSL who showed greater improvement. This suggests that the poor outcomes after repeat HSL are due to factors other than hamstring dysfunction such as quadriceps weakness or the natural history of CP. Since repeat HSL may not be the most appropriate intervention, other interventions such as distal femoral extension osteotomy and patellar tendon shortening should be considered.

5. Clinical significance

Both patients who underwent primary and repeat hamstring lengthening procedures had shorter than normal hamstrings and/or slow hamstring lengthening velocity, though better functional outcomes were observed following primary HSL. Short or slow hamstrings do not usually indicate hamstring dysfunction in recurrent crouch, leading to the lower effectiveness of repeat HSL.

Conflict of interest statement

The authors have no conflicts of interest to disclose.

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