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Longitudinal change in foot posture in children with cerebral palsy

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Abstract

Purpose Foot deformities are common in children with cerebral palsy (CP), yet the evolution of such deformities is not well documented. We aimed to observe and analyse changes in foot posture during growth in children with CP.

Methods We followed 51 children (16 unilateral, 35 bilateral; 37 Gross Motor Function Classification Scale (GMFCS) I/II, 14 III/IV) aged two to 12 years in this level II, IRB-approved prospective longitudinal study. Data after bony foot corrections were excluded. Outcome measures included coronal plane pressure index (CPPI) and pressure impulses from the heel, medial midfoot and medial forefoot. Data were LOESS smoothed and resulting models were compared for significant differences across time using a derived FANOVA method.

Results The GMFCS I/II group had more foot valgus than typically developing (TD) children until seven years which normalised thereafter. From two to 12 years, GMFCS III/IV children had more foot valgus than TD children. Heel impulse was significantly reduced in both GMFCS groups compared with TD children, and the III/IV group had less heel contact than the I/II group.

Conclusions Due to early variability and the tendency for resolving valgus foot posture in children with CP, conservative management of coronal plane foot deformity is suggested in early childhood, especially for children classified as GMFCS I and II.

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Introduction

Foot deformities are common in children with cerebral palsy (CP), but the evolution of dynamic foot postures is not well documented.[1] Sixty-one percent of children with CP were observed to have equinus,[2] and foot and ankle deformities account for 25% to 30% of all surgical procedures performed on ambulatory children with CP to improve gait function.[3]

The aetiology of foot deformities in children with CP is complex and multifactorial. The evolution of gait and foot development in children with CP does not follow the patterns seen in typically developing (TD) children.[4] Abnormal foot postures are caused by muscle spasticity and imbalance, soft-tissue contractures, bony torsion and joint instability,[1,2,5,6] which all occur, for the young child, within a dynamic context of neurologic maturation and growth. Bony and soft tissue surgeries,[5,7] physical therapy,[8] foot and ankle orthoses,[6] and Botox injections[9] are all used to manage deformities of the foot and ankle in children with CP. Such a complicated scenario makes it impossible to document the natural history of foot deformity in children with CP, but improved knowledge of foot posture maturation can contribute to more effective treatment paradigms and critically evaluate outcomes.

A few authors have examined gait development and foot deformity statically in children with CP. A study by Bell et al[10] began to describe a natural progression of gait in children with CP, but only included two gait analyses spaced at an average of four years, five months apart. Another study by Renshaw et al suggests that, based on static radiographs, the best time for orthopaedic surgery on the foot is when the child is aged between four and five years or when their adult gait pattern has completely developed.[11] Both of these studies took advantage of observational data but do not include information about the degree of dynamic foot deformity.

A few researchers have used a pedobarograph to study dynamic foot position in children. Pedobarographic data correlate to radiographic data[12] and have been found to be a reliable assessment.[13] The pedobarograph has been used to track the progress of dynamic foot posture in children with clubfoot,[14] and in children with hallux valgus and hallux rigidus,[15] as well as in TD children.[16]

Foot pressure was examined retrospectively in children with CP 26 to 61 months after surgery for valgus correction[17] and for at least seven years after equinus correction.[18] Evaluation of foot pressure in early walkers aged two to four years demonstrated abnormal foot position in children with CP.[6] A 2014 study on midfoot break deformity in children with CP classified midfoot break by severity and recognised it from pedobarographic data.[19] To achieve better understanding of long-term trends in the development of foot posture in children with CP, prospective data must be collected over a longer period. Such a knowledge base could lead to more effective treatment paradigms through critical comparison of outcomes.

The purpose of this study was to identify unique trends in the development of foot deformity using dynamic foot pressure data in children with CP from early walking to adolescence.

Patients and methods

Children aged 17 to 40 months with primarily spastic CP were recruited to participate in this IRB-approved prospective longitudinal observational study. These children were diagnosed with CP based on abnormal muscle tone, limited motor control and developmental delay. All children involved in this study were treated by orthopaedic surgeons from a single centre that specialises in the management of CP. The centre follows a treatment philosophy in which children receive physical therapy during early childhood: children are regularly braced if foot or ankle deformity is present; Botox is sometimes administered to manage dynamic toe-walking; and soft-tissue surgery is performed when fixed deformities emerge that cannot be managed by conservative methods. Therapy, bracing, Botox and soft-tissue surgery were delivered to participants in this study as part of this standard clinical care and were not controlled for or used as exclusion criteria. However, surgery to correct varus or valgus foot deformity including bony foot surgery, tibialis anterior or tibialis posterior tendon transfer or lengthening were exclusion criteria for the analysis, and data following these procedures were excluded because the obvious goal of the surgery was to correct the coronal plane deformity. Additionally, to evaluate the effect of plantar flexor lengthening on coronal plane foot pressure distribution, two groups were compared. Group 1 consisted of all data points from children that did not have a plantar flexor lengthening and all data points from children that did have a plantar flexor lengthening prior to the surgery. Group 2 consisted of data points from children who had a plantar flexor lengthening following the procedure.

The children included in this study were examined every six months until they were aged five years. After that, children were measured once a year until they were aged 12 years. Not all patients were able to attend all study visits, but a child's data were included for analysis if they completed a minimum of five visits or participated to the age of five years. When they were aged four to eight years, children were classified according to the Gross Motor Function Classification Scale (GMFCS)[20] and by distribution of involvement (unilateral or bilateral). Data from children with CP were compared with cross-sectional data from age-matched, TD children. During each visit, three footstrikes on each side were collected with the pedobarograph, and then average values were used for analysis (F-Scan Measurement System; Tekscan, Boston, MA, USA). Children walked with bare feet, independently, with their typical assistive device, or were hand-held. For the children with unilateral CP, only data from the involved limb were used in the analysis.

From the pedobarograph, the foot was divided into five areas—the heel, the lateral forefoot (LLF), the medial forefoot (MFF), the lateral midfoot (LMF) and the medial midfoot (MMF)—as described in Chang et al,[21] and the impulse was measured. Also, the coronal plane pressure index (CPPI) was used as a single objective measure to evaluate the overall distribution foot pressure.[21] The CPPI defines the ratio of medial to lateral pressure impulses in the midfoot and forefoot regions, with higher positive numbers (max = 100) indicating severe valgus and higher negatives (max = -100) indicating severe varus.

For each group, data from each foot segment and CPPI were modelled using R's LOESS function, which implements local polynomial regression fitting.[22] The modelled output included both a smoothed mean fitted curve versus age as well as standard errors at each age. As we were interested in determining where, on average, the groups were similar or different as a function of age, the smoothed curves for each group were then compared for statistical differences based on the FANOVA method introduced by Ramsay et al.[23] Comparisons of curves were initially made at 0.10-year increments using Welch's two-sample t tests. The t statistic at each bin was computed using the formula:

1) $$t = \frac{\bar{X}_1 - \bar{X}_2}{\sqrt{\frac{s_1^2}{N_1} + \frac{s_2^2}{N_2}}}$$

Where X_1 and X_2 were the smoothed values for each curve in the middle of the bin, s_1 and s_2 were the standard deviations of the data for each bin, and N_1 and N_2 were the number of data points in each bin.[24] The number of points per bin was estimated using kernel density smoothing on the age-distributed data. The degrees of freedom ν for each bin were computed using the Welch-Satterthwaite equation.

2) $$\nu \approx \frac{\left(\frac{s_1^2}{N_1} + \frac{s_2^2}{N_2}\right)^2}{\frac{s_1^4}{N_1^2 \nu_1} + \frac{s_2^4}{N_2^2 \nu_2}}$$

Where $\nu_1 = N_1\text{-}1$ and $\nu_2 = N_2\text{-}1$. Bonferonni corrections were made for comparisons between the independent groups so that the corrected alpha level was $\alpha' = 0.05/3 = 0.0166$. No alpha correction was made for the multiple comparisons along the length of the curve since the computed *t* values in adjacent bins were nearly perfectly correlated rather than independent. To check this assumption, we redid the analysis using different bin sizes from one to 11 months. Other than changes to the sharpness of the p-value curves due to larger bins, the magnitudes of the curves and their general shapes did not substantively change as bin size changed. The stability of these results indicated that point-wise Bonferroni corrections would have over-corrected for type I errors.

Since the data were drawn from participants bilaterally, we also analysed the data to determine the degree of between foot dependence so as not to inadvertently increase the chance of type I error. Side-to-side correlations (Pearson's ρ) in CPPI were calculated on a per-patient basis, and from this, a distribution of correlation coefficients was built. Then 100 null distributions of random per-patient correlations were built for comparison to the actual ρ distribution from the data. Distributions of correlations were then compared 100 times using the Welch t-test, and the percent of times p-values were significant was calculated.

To address bilaterality, the effective number of observations per patient was calculated based on within-patient side-to-side correlation coefficient (ρ). This was then used to calculate the effective number of observations per patient in the formula proposed by Hanley et al.[25]

3) $$N_{eff} = N/(1 + \rho)$$

Where N is the actual number of observations per patient. For high correlations, where ρ is close to unity, N_{eff} approaches N/2, but for low correlations, where ρ is close to zero, N_{eff} approaches the full N observations. These effective N values (per patient) were then used to weight the data. An inverse weighting scheme was chosen so as to partition weight patients relatively equally. LOESS fits to these data were then compared using serial Welch t-tests as described previously.

Results

The 96 children from the CP specialty clinics at our hospital were recruited and enrolled in the long-term prospective study. Several patients were excluded from the present analysis, including 17 whose involvement was too severe to record reliable foot pressure and 28 who did not meet the participation requirements. There were 51 children who met the inclusion criteria for this analysis (16 unilateral, 35 bilateral involvement; 86 involved feet; GMFCS: 14 (I), 23 (II), 9 (III), 5 (IV)). The average age at initial visit was two years, eight months (sd, seven months), and 847 evaluations were included in the analysis. The data from children with CP were compared with data from age-matched, TD children (288 evaluations).

Traditional care was provided to patients during the course of the study, and we documented this in our study population. Of 51 patients, 31 underwent orthopaedic lower extremity surgery guided by gait analysis over the course of the study (Table 1). In total, 14 children had one

Table 1. Surgical intervention in ambulatory children with cerebral palsy (CP)

Procedures	≥ 1 Procedure (%) (limb)	≥ 2 Procedures (%) (limb)	≥ 1 Procedures (%) (patient)
Surgeries to address foot varus			
Posterior tibialis tendon transfer	2	0	4
Anterior tibialis tendon transfer	3	0	4
Posterior tibialis recession	1	0	2
Surgeries to address foot valgus			
Calcaneal osteotomy	7	0	8
Subtalar fusion	3	0	4
Medial column correction	3	0	4
Cuneiform osteotomies	2	0	2
Other lower extremity surgeries			
Hamstring lengthening	43	14	45
Gastrocenemius lengthening	39	8	43
Adductor lengthening	27	3	26
Achilles tendon lengthening	15	6	22
Femoral osteotomy	10	2	14
Tibial osteotomy	12	0	14
Iliopsoas lengthenings	7	0	8
Rectus femoris transfer	6	0	6
Knee capsulotomy	4	0	4
Patellar tendon excision and repair	2	0	4
Peri-ilial pelvic osteotomy	1	0	2
Dega pelvic osteotomy	1	0	2
Pemberton osteotomy	1	0	2

surgical event, 17 had two surgical events, and seven had three or more surgical events, with an average of three (SD, 1) procedures completed during each surgical event. Foot pressure data were not excluded following orthopaedic surgery, except where orthopaedic surgery to address foot deformity (bony foot surgery, five children, ten feet) or to address foot position (tendon transfer, four children, four feet) was performed. The excluded data were from children aged six to 12 years (ten years (SD, 1)). The 143 data points were found to be post-plantar flexor lengthening with surgery completed at the ages of three to 11 years.

Conservative management and treatments including Botox injections, orthotics and physical therapy were also a part of the study group's natural history (Table 2). A review of therapy records revealed that physical therapy decreased as the children aged (two to five years, 92%; six to eight years, 80%; nine to 12 years, 60%). Younger children also more commonly reported the use of orthotics (two to five years, 86%; six to eight years, 79%; nine to 12 years, 58%). In all, 43% received Botox injections.

Abnormal foot position was noted in children with CP compared with TD children as reflected in group differences in CPPI and heel impulse (Fig. 1). Children with CP demonstrated significantly greater valgus compared with TD children until the age of seven years. When grouped according to functional ability, the CPPI was significantly more valgus in the GMFCS group I/II compared with TD children aged two to seven years but was not significantly different than the TD group after the age of seven years.

Table 2. **Conservative management in ambulatory children with cerebral palsy (CP)**

Age categories	2 to 5 years	6 to 8 years	9 to 12 years
Botox	43%		
By patient	n = 51	n = 45	n = 38
Private PT	24%	11%	5%
School PT	29%	45%	29%
Both school and private PT	39%	24%	26%
No PT	8%	18%	37%
By involved limb	n = 86	n = 73	n = 61
Solid AFO	33%	4%	8%
Articulated AFO	41%	62%	39%
SMO	9%	8%	8%
Other	3%	5%	3%
No orthotics	14%	21%	43%

AFO, ankle-foot orthosis; PT, physical therapy; SMO, supramalleolar orthosis

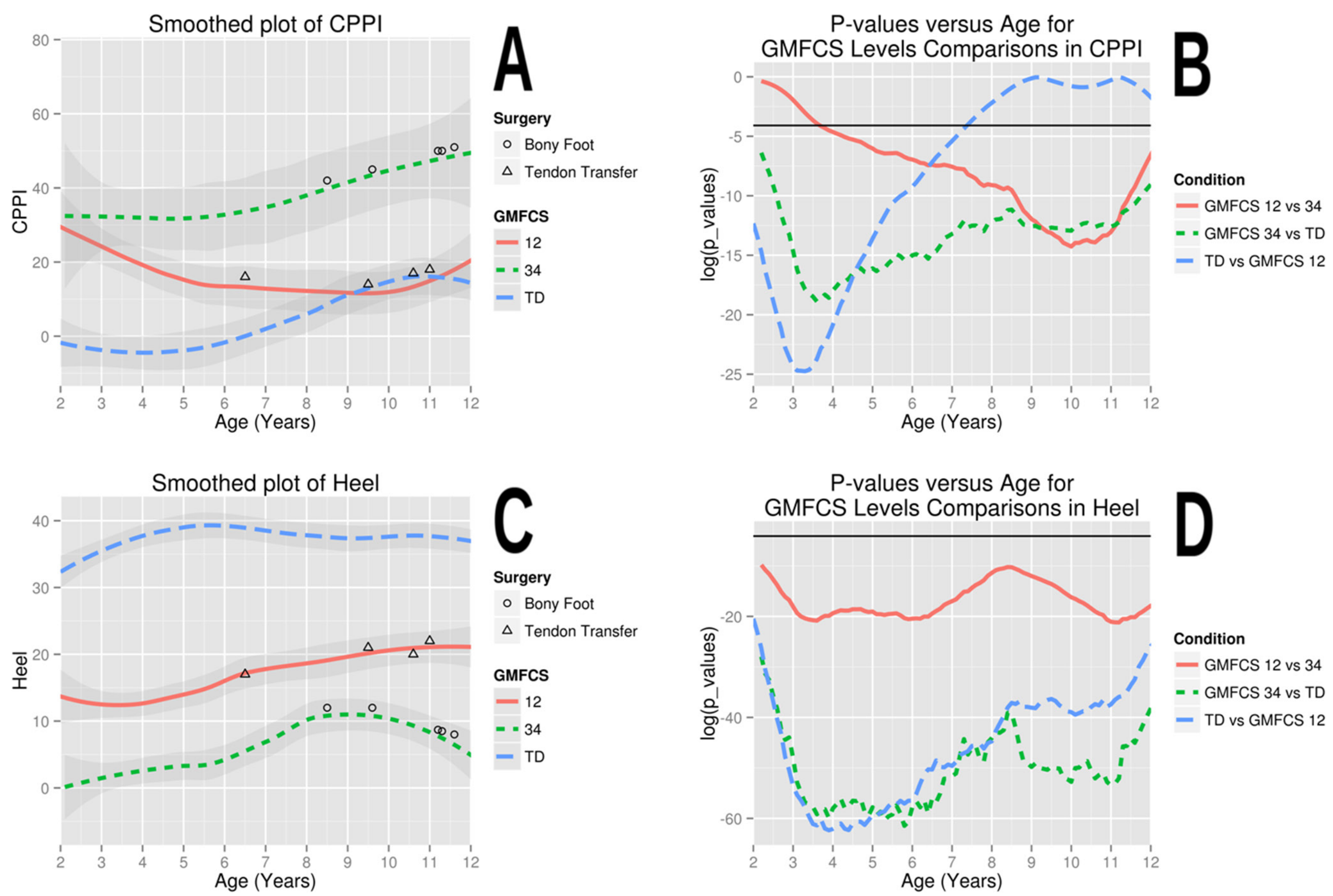

Fig. 1 Longitudinal change in coronal plane pressure index (CPPI) and heel impulse in children with cerebral palsy (CP). (**a, c**) The means and standard errors; (**b, d**) the statistical significance; significant differences are found in values that fall below the horizontal line on the graphs.

The CPPI of the GMFCS level III/IV group was greater in valgus than in TD children aged two to 12 years. The GMFCS I/II and III/IV groups were significantly different between the ages of four and 12 years: the GMFCS III/IV group had a more valgus presentation. Heel impulse was reduced in both the GMFCS I/II and III/IV groups compared with TD children, regardless of age, and the III/IV group had significantly less heel contact than the I/II group at all ages.

Abnormal valgus foot dynamics were reflected by elevated MMF and MFF pressures in children with CP (Fig. 2). MFF pressure was significantly higher in GMFCS I/II children with CP aged two to 12 years compared with TD children. From ages two to 12 years, GMFCS III/IV children had a significantly higher MFF pressure than the TD children. Significant MFF differences between GMFCS I/II and III/IV groups were found at ages two to 12 years, when the III/IV group had higher pressure. In the MMF region, children with GMFCS I/II had a significantly higher impulse compared with TD children aged two to 12 years. GMFCS III/IV children had significantly higher MMF pressure at ages four to 12 years when compared with TD children. MMF pressures in GMFCS I/II and III/IV groups were significantly different after the age of seven years, with children in the III/IV group having higher pressure. There were no significant differences between the groups in LMF or LFF impulse.

Different trends emerge when analysing the data while grouping according to unilateral and bilateral distribution of symptoms. While early foot development is similar in children with GMFCS I/II aged six to nine years, the unilaterally involved children move into significantly less valgus, and several move into a varus foot posture. An associated significant increase in LMF pressure is noted in children with unilateral involvement compared with those with bilateral involvement at ages six to ten years.

Results of the bilaterality analysis indicated that in only four out of 100 comparisons were actual group correlation coefficients significantly different from randomly generated distributions. Therefore, 96% of the time, the actual side-to-side correlations were indistinguishable from random chance. This implies that bilateral correlations in the data are not a concern in these data. Likewise, there was no significant difference in CPPI between groups based on plantar flexor lengthening (Group 1: data from children after plantar flexor lengthening *versus* Group 2: data from children that did not have plantar flexor lengthening and

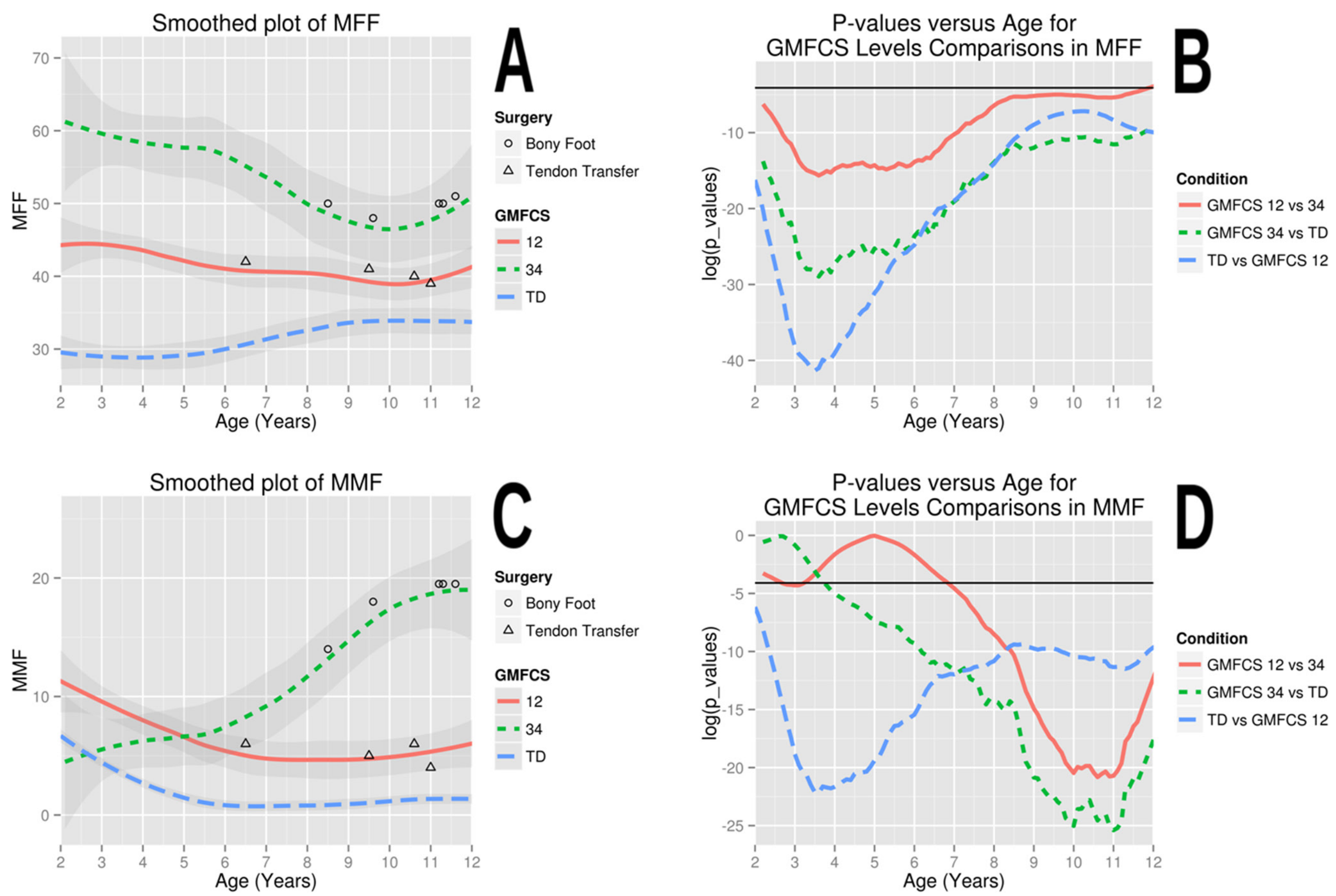

Fig. 2 Longitudinal change in medial midfoot (MMF) and medial forefoot (MFF) in children with CP. (**a, c**) The means and standard errors; (**b, d**) the statistical significance; significant differences are found in values that fall below the horizontal line on the graphs.

data from children that had a plantar flexor lengthening prior to the procedure; Fig. 3).

Discussion

Children with CP are prone to foot deformities that can make ambulating difficult. A variety of treatment options exist, including physical therapy, bracing, Botox injections and orthopaedic surgery. To establish treatment plans, knowledge of how foot posture evolves in this population is necessary.

While a variety of treatments are available for foot deformities, there is little agreement over the timing of intervention, especially the use of surgical intervention. Critical evaluation of the data within this study demonstrates trends of changing foot posture over the course of a child's development. Underlying the trend of resolving valgus foot posture is a great deal of variability in the younger children. While children in this study who used assistive devices (GMFCS II–IV) tended to persist in valgus from an early age, they generally remained in a stable position during the time that only conservative management was used. Several children in this group underwent bony correction in early adolescence. Children with CP who ambulated without an assistive device (GMFCS I/II) often presented with valgus feet in early childhood but tended to resolve with conservative management and growth over the course of development. These findings point to the safety in adopting a conservative approach for the young child with close pedobarographic monitoring to identify progression of the deformities and close clinical follow-up to determine when bracing is no longer tolerated.

There are no published objective data with long-term follow-up of natural history to guide timing of surgical management, so most of the papers in the literature are based on personal opinion.[1,5] Data from our long-term study show the difficulty in predicting eventual foot posture but also reveal the clear trends of GMFCS III/IV-level children, who tended to remain in stable valgus through

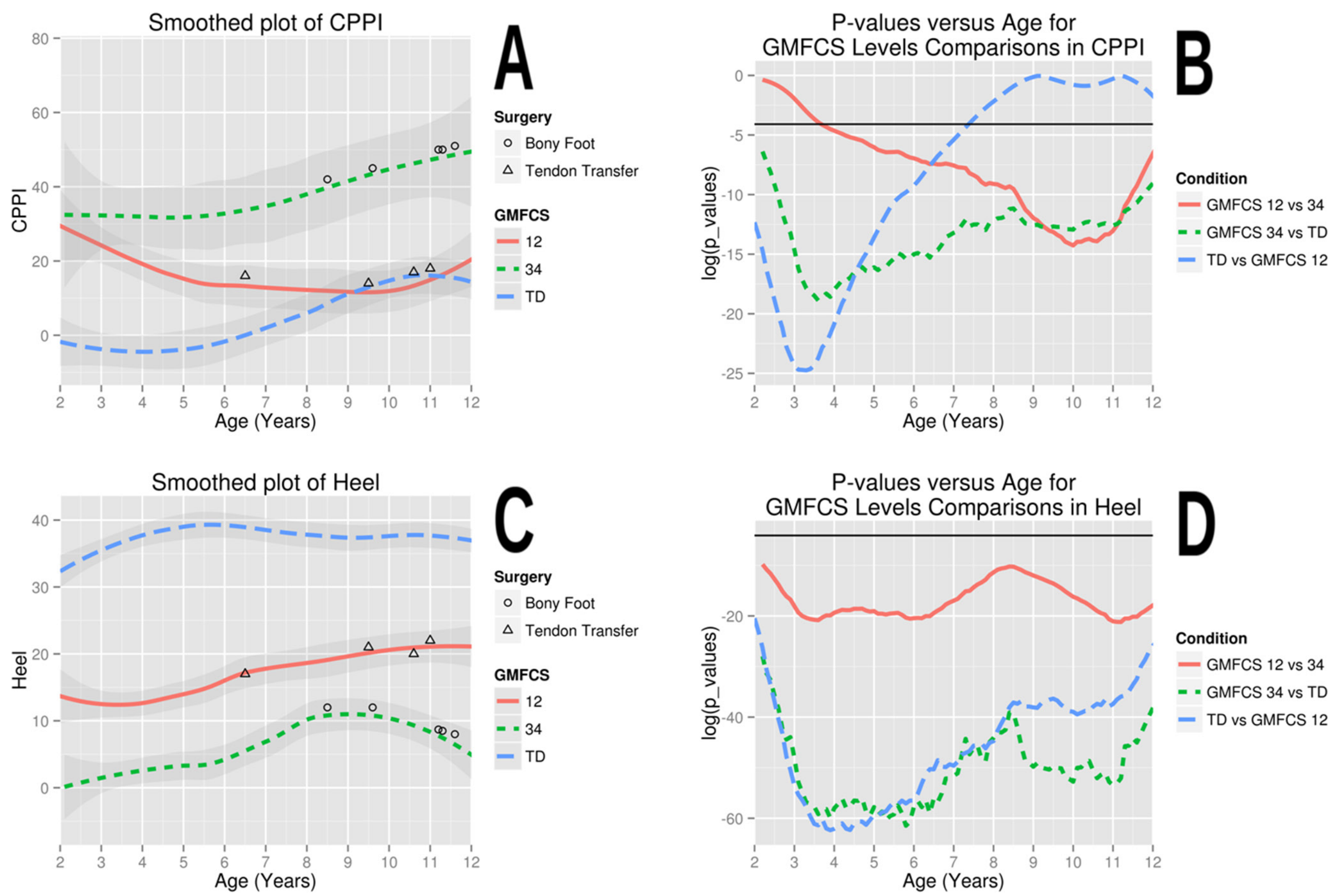

Fig. 3 The effect of plantar flexor lengthening on longitudinal change in coronal plane pressure index (CPPI) in children with cerebral palsy. (**a**) The means and standard errors. (**b**) The statistical significance; significant differences are found in values that fall below the horizontal line on the graphs. The No Surgery group is all data points from children that did not have plantar flexor lengthening and data points from children that had a plantar flexor lengthening prior to the procedure. The Surgery group is all data points from children that had plantar flexor lengthening following the procedure.

middle childhood, and of GMFCS I/II-level children, who tended to improve and move toward normal foot posture or varus.

There is a potential ethical dilemma in attempts to describe the longitudinal development of foot deformity during childhood growth for a population in which several treatments are found to be effective. Conservative management provides clear benefits to gross motor development, flexibility and strength, but its long-term effects on foot posture are unclear.[6,8,9] Withholding treatment with obvious benefits is unethical, but describing development of foot posture in a treated population makes it difficult to determine which trends can be attributed to development and which to intervention. The effect of bony foot surgery or transfer or lengthening of the tibialis anterior or posterior muscles on foot posture are clear and well documented,[3,5,17] so we removed data points following these interventions. While removing these data points eliminates the obvious confounding effect of surgically corrected feet on our outcome measures, it also likely moves the remaining sample mean toward normal by removing feet that were the most abnormal. All bony foot surgeries to address valgus were completed in children in the GMFCS III/IV group aged eight to12 years, so this group may have moved into more severe valgus without intervention. All tendon surgeries to address varus were completed in the GMFCS I/II group of ages of six to 11 years, so this group may have moved into more varus without intervention. Additionally, a closer examination of the effects of plantar flexor lengthening may reveal significant effects, but in our longitudinal study the procedure had no significant effects on group means of the coronal distribution of the plantar pressure.

Some limitations exist in this longitudinal study. It is a lengthy longitudinal follow-up of children with CP who are receiving standard conservative management at a specialised paediatric hospital and therefore is not a pure natural history. Additionally, the trends described may not be relevant to children who might undergo a different course of treatment at other intuitions; however, because the clinical course of the participants represents our institution's broader treatment philosophy, these data provide a benchmark to which future studies can be compared. While this is the longest current report of longitudinal development of foot deformity in children with CP, there would be further benefit in evaluating children through maturity. Foot pressure analysis provides reliable and valuable information, but it can be affected by changes in proximal joint alignment. Changes in hip and knee position affect foot pressure results so change in foot pressure cannot be attributed to change in foot and ankle position alone. Future work should include full-body segmental three-dimensional dynamic analysis including a multi-segment foot kinematic model.

Young children with CP tend to have a valgus foot distribution relative to TD children. Valgus tends to persist in children with GMFCS levels III/IV and to normalise in children with GMFCS levels I/II after the age of six years. Due to variability in the longitudinal development and the tendency for improvement in valgus foot posture in children with CP, conservative management of coronal plane foot deformity is suggested, especially in young children and those ambulating without an assistive device.

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