Variation in kinematic and spatiotemporal gait parameters by Gross Motor Function Classification System level in children and adolescents with cerebral palsy

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ABBREVIATIONS
BSCP Bilateral spastic cerebral palsy
3DGA Three-dimensional gait analysis

AIM The aim of this study was to examine differences in gait kinematics and spatiotemporal parameters in ambulatory children and adolescents with bilateral spastic cerebral palsy (BSCP) among Gross Motor Function Classification System (GMFCS) levels I-III.

METHOD A retrospective review was conducted of individuals with BSCP who had three-dimensional motion analysis (3DGA) at one of seven pediatric hospitals. Means and standard deviations of each gait parameter were stratified by GMFCS levels (I-III) and for a typically developing comparison group.

RESULTS Data from 292 children and adolescents with BSCP (189 males, 103 females; mean age 13y) were compared to a typically developing comparison group (24 male, 26 female; mean age 10y 6mo). Gait patterns differed from typically developing in all GMFCS levels, with increasing deviation as GMFCS level increased in 21 out of 28 parameters. Despite significant differences in selected mean kinematic parameters among GMFCS levels such as knee angle at initial contact of 24°, 29°, and 41° in GMFCS levels I, II and III respectively, there was also substantial overlap among GMFCS levels.

INTERPRETATION GMFCS levels cannot be identified using specific gait kinematics. Treatment decisions should be guided by comprehensive 3DGA that allows measurement of gait impairments at the joint level for each individual.

The Gross Motor Function Classification System (GMFCS) is an important tool that is used routinely to categorize children with cerebral palsy (CP) on the basis of their gross motor functional abilities and limitations. It has been shown that, with increasing GMFCS level, gait function deteriorates in terms of a wide variety of outcome measures used in the clinical setting. It is not known, however, if individual GMFCS levels can be defined by specific gait kinematics of the lower extremities, whether gait patterns at each GMFCS level differ from those who are typically developing and whether gait kinematics decline with increasing GMFCS level. Knowledge of the prevalence of gait pathology by GMFCS level will help clinicians explain to patients and families how a patient’s gait may differ from those who are typically developing and the possible related treatment expectations. This study will also determine whether GMFCS level can define specific gait characteristics or if a more comprehensive motion analysis is needed to determine gait function on a patient by patient basis.

The prevalence of gait deformity at the individual joint level in terms of topographical classifications for CP has been studied by multiple groups. Wren et al.7 reported that the most common problems for patients with diplegia were stiff knee (82%), in-toeing (78%), and crouch (72%). Rodda et al.8 defined diplegia in terms of overall lower extremity patterns at multiple joints such as crouch gait with simultaneous increased knee flexion and ankle dorsiflexion. This study would be the first attempt at determining whether a functional classification system such as the GMFCS provides guidance in terms of understanding gait function by individual GMFCS levels related to prevalence of gait deformity.

The objective of this study was to determine how gait deviations and spatiotemporal parameters in ambulatory children and adolescents with bilateral spastic cerebral palsy (BSCP) vary by GMFCS levels and from those typically developing. It was hypothesized that there would be differences in gait deviations among GMFCS levels and that with increasing GMFCS level (i.e. increasing severity of
impairment) gait deviations and spatiotemporal parameters would worsen with respect to typically developing gait. This study builds upon previous research that examined the relationship between GMFCS level and performance on outcome tools by examining GMFCS levels in terms of gait deviations using data from a large study cohort.

METHOD
This analysis of a large multicenter, prospective, longitudinal cohort includes individuals with BSCP recruited between 2008 and 2011 who were treated according to the local standard of care at one of seven pediatric orthopedic hospitals. The standard of care at each institution included comprehensive motion analysis as an integral part of the orthopedic surgical treatment decision-making process. Inclusion criteria were children and adolescents with a diagnosis of BSCP, GMFCS levels I to III, and age 8 to 18 years who were physically and cognitively able to complete a three-dimensional gait analysis (3DGA). Exclusion criteria were lower extremity orthopedic surgery within the past year, botulinum toxin A injections within the past 4 months, or an operational baclofen pump. A group of typically developing children and adolescents with no diagnosed neuromuscular or orthopedic conditions from one of the seven sites provided a historical reference cohort. Each site obtained Institutional Review Board approval: participant assent, consent, parental permission, and Health Insurance Portability and Accountability Act authorization were obtained as appropriate.

GMFCS level was assessed and recorded by trained examiners at each site following established thresholds by age. Instrumented 3DGA was performed using the conventional gait model to obtain spatiotemporal parameters (step and stride lengths, cadence, walking velocity) and lower extremity kinematics at the pelvis, hip, knee, and ankle. Five sites used Vicon (Oxford, UK) motion capture systems with Plug-in-Gait modeling software (v4.6), while two used Motion Analysis Corporation (Santa Rosa, CA, USA) systems with Orthotrak modeling software (v5.0). Barefoot walking trials at a self-selected speed, using the participant’s typical assistive device were collected using a standardized protocol. Discrete gait parameters were extracted from each trial via custom software and included spatiotemporal measures and local maxima, minima, range, and timing of pelvic tilt, obliquity and rotation, hip flexion, abduction and rotation, knee flexion, ankle dorsiflexion, and foot progression angle. A complete list of gait parameters extracted is shown in Table I (see also Table SI, online supporting information). The left side was arbitrarily selected for analysis a priori. The mean of three trials for all patients was calculated and used in this analysis. Similar data were extracted from the typically developing group using the same methods.

To standardize the study methodology across the seven centers, training was performed by the principal investigator for all study personnel. All study methodology, including marker placement and strength testing, was demonstrated and reviewed to minimize inter-examiner differences. Extensive documentation including illustrations on methodology was also provided. This training followed a protocol developed by Shriners Hospitals for Children and recently published by Gorton.

Statistical analysis
Means and standard deviations of each gait parameter were calculated and stratified by GMFCS level and for the typically developing group. Differences in the means were assessed by single factor analysis of variance (ANOVA) with group as a fixed factor with four levels (typically developing and three GMFCS levels). Significance was assessed using a Bonferroni-adjusted critical \( p < 0.01 \) to adjust for the increased experiment-wise error rate of multiple comparisons. A test for linear monotone trend in the mean across groups in comparison to the typically developing group was completed to assess change with increasing GMFCS level. An analysis of covariance (ANCOVA) was used to assess possible reasons for variance in the results using height as a covariate, site as a random factor, and GMFCS level as a fixed factor. In both cases differences were considered significant at \( p < 0.01 \). Clinical relevance of the differences is reviewed later in the discussion.

Cumulative distribution plots provide information about the variance of a given parameter across all patients within a given group, by graphically comparing the distribution of each parameter in each group (typically developing, GMFCS levels I, II, and III). The \( x \)-axis provides the value of the particular parameter and the \( y \)-axis gives the percentage of people in the group with that value or less for the parameter. The slope of the cumulative distribution function plot reflects the spread of the distribution: a steeper slope reflects a tighter distribution. The \( x \)-value of the graphs when the cumulative percent on the \( y \)-axis is 50 reflects the medians of each group. Probability density graphs calculated using Epanechnikov kernel functions smoothed with a moving window with a width of 5% of the data were plotted to illustrate the distributions at each GMFCS level group and for the typically developing comparison group.

RESULTS
In total, 748 individuals were screened: 273 did not meet inclusion criteria, 80 declined participation, and 18 were excluded for missing data, resulting in 377 participants of whom 292 with bilateral involvement are included in this analysis (Table II). The participation from the seven institutions varied from 9 to 77 patients. The historical typically developing comparison group was younger, shorter, and lighter than individuals with BSCP at each GMFCS level (Table II). There were no differences among
GMFCS levels for age or height, but those at GMFCS levels I and II were heavier than those in level III (Table II). However, there was no difference in body mass index (BMI) between GMFCS levels (I, II, and III) and the typically developing comparison group (Table II).

### Differences among GMFCS level and typically developing comparison group (comparison of means)

Overall, the mean spatiotemporal and kinematic parameters of individuals with CP differed compared to the typically developing comparison group and among GMFCS levels. Differences were present at all GMFCS levels with a monotone trend of increasing deviation from typically developing individuals with CP differed compared to the typically developing comparison group and among GMFCS levels I and II versus III.

Mean joint kinematic parameters (17/23) showed significant differences from the typically developing comparison group at the pelvis, hip, knee, and ankle joints at all GMFCS levels (Table I). The hip showed increased mean anterior tilt and range of motion. The hip showed increased maximum flexion, and decreased minimum flexion and overall range of motion. The knee showed increased flexion at initial contact and increased mean flexion in stance, increased maximum extension in stance, decreased maximum flexion, and overall range of motion. The ankle showed increased peak dorsiflexion in swing, decreased peak plantar flexion in swing, and decreased peak plantar flexion-extension range of motion. The ankle showed increased peak dorsiflexion in swing, decreased peak plantar flexion-extension range of motion. The ankle showed increased peak dorsiflexion in swing, decreased peak plantar flexion-extension range of motion.
tar flexion in swing, and decreased total range of motion. The majority of significant differences were between GMFCS levels I, II, and III (13/23) and levels II and III (11/23). Only five out of 23 kinematic parameters showed differences between GMFC levels I and II.

### Distribution within GMFCS levels and typically developing comparison group

Substantial overlap among GMFCS levels was noted for kinematic and spatiotemporal parameters as shown in the cumulative distribution plots (Fig. 1 and Supplemental online Fig. S1) and the probability distribution plots for the same group of selected parameters (Fig. 2 and Supplemental online Fig. S2). Both plot formats provide a detailed look at the variation of each gait parameter within each GMFCS level and typically developing group, and how each GMFCS level differs from the typically developing comparison group with respect to each parameter. Depending on the kinematic parameter, deviation from the typically developing comparison group is unidirectional, for example, increasing knee flexion at initial contact (Figs 1e and 2e) or bidirectional, for example, increasing internal or external foot progression angle (Figs 1h and 2h). The cumulative distribution plots allow the reader to determine prevalence: the percentage of persons above or below a critical value of a given parameter at each GMFCS level.

The degree of overlap with typically developing varied depending on the parameter. Knee angle at initial contact (Figs 1e and 2e) had the least overlap with the typically developing comparison group, suggesting the greatest amount of pathology for this parameter. The maximum knee angle at initial contact for the typically developing group was 10°. More than 90% of the patients in each GMFCS level I, II, and III had a knee angle at initial contact greater than 10° (Fig. 1e). No other parameter evaluated showed as much difference from typically developing. Mean hip rotation in stance (Fig. 1d) showed overlap with all GMFCS levels and each GMFCS level overlapped the full typically developing range. The highest internal hip rotation in the typically developing group was approximately 10°. GMFCS level I had only 20% of patients greater than 10°; GMFCS level II, 40%; and GMFCS level III, 50%.

Most spatiotemporal and kinematic parameters showed a substantial overlap among GMFCS levels. Values for knee angle at initial contact ranged from 5° to 53° at each GMFCS level (Fig. 1e). Similarly, values for minimum hip extension (Fig. 1c) ranged from −5° to 25° and mean pelvic tilt (Fig. 1b) ranged from 0° to 33° at each GMFCS level. In some cases, there was more overlap between GMFCS levels I and II and not III. Walking speed (Fig. 1a) showed overlap for approximately 90% of patients for GMFCS levels I and II, whereas GMFCS levels II and III only overlapped with 65% of patients. Greater overlap between GMFCS levels I and II was found for mean pelvic tilt (Fig. 1b), minimum hip flexion (Fig. 1c), knee angle at initial contact (Fig. 1e), and timing of peak knee flexion at initial contact (Fig. 1f). These findings are consistent with the statistical comparison of means with greater differences between both GMFCS levels I and II with III.

The cumulative distribution plots also clearly show that gait deviations for certain parameters are bidirectional with respect to the typically developing comparison group. For peak ankle dorsiflexion in terminal stance (Fig. 1g), the majority of patients at each GMFCS level were within the

### Table II: Demographics for the typically developing comparison group and children and adolescents with bilateral spastic cerebral palsy by Gross Motor Function Classification System level

<table>
<thead>
<tr>
<th>BSCP by GMFCS level</th>
<th>TD</th>
<th>I</th>
<th>II</th>
<th>III</th>
<th>Overall p&lt;sup&gt;a&lt;/sup&gt;</th>
</tr>
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<tbody>
<tr>
<td>Age, y</td>
<td>9.5 (3.0)</td>
<td>13.4 (2.5)</td>
<td>12.8 (2.6)</td>
<td>12.8 (2.6)</td>
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<td>Height, cm</td>
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<td>151.9 (14.4)</td>
<td>147.9 (14.1)</td>
<td>142.4 (13.3)</td>
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<tr>
<td>Weight, kg</td>
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<td>46.7 (18.0)</td>
<td>44.8 (15.6)</td>
<td>42.8 (14.2)</td>
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<tr>
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<td>65</td>
<td>71</td>
<td>53</td>
<td>0.010</td>
</tr>
</tbody>
</table>

<sup>a</sup>p value for overall single factor analysis of variance (ANOVA) with levels (typically developing, GMFCS I, II, and III). BSCP, bilateral spastic cerebral palsy; TD, typically developing comparison group; GMFCS, Gross Motor Function Classification System; SD, standard deviation. Bolded values TD-GMFCS I, II, III (ANOVA, p<0.01).
Figure 1: Cumulative distribution plots for selected gait parameters. GC, gait cycle; Post, posterior; Ant, anterior; Ext, extensor; Flex, flexor; Extr, external; Int, internal; Pl, plantar flexion; Dor, dorsiflexion.
typically developing range (75% GMFCS level I, 80% at GMFCS level II, and 55% at GMFCS level III). However, all GMFCS groups show increased peak dorsiflexion (18% GMFCS level I, 15% GMFCS level II, and 30% GMFCS level III) and decreased peak dorsiflexion (9% GMFCS level I, 10% GMFCS level II, and 18% GMFCS level III). Similarly for foot progression angle (Fig. 1h), at least 35% of patients fall within typically developing range. However, all GMFCS levels have patients that show foot progression that is either more internal or external than typically developing.
DISCUSSION

The results of this study confirm that mean kinematic and spatiotemporal parameters of children and adolescents with BSCP, GMFCS levels I to III, differ from typically developing children and become more severe with increasing GMFCS level. This study also found there was a large distribution of parameters within each GMFCS level and overlap between GMFCS levels. Study data were derived from seven geographically separate locations, and were based upon large sample sizes at each GMFCS level, supporting generalizability. The results are highly consistent with the clinical understanding of impairment in BSCP, providing additional support for their validity.

The decline in kinematic parameters with respect to the typically developing comparison group with increasing GMFCS level was expected⁵,¹³ and is consistent with the decline in other outcomes measure scores seen with increasing GMFCS level.⁵ There was a greater kinematic similarity between GMFCS levels I and II as seen on the cumulative and probability distribution plots. This is consistent with the use of assistive walking devices for GMFCS level III, indicating poorer function and associated increased kinematic impairment compared to levels I and II. These differences between GMFCS levels II and III especially are clinically relevant, as in many cases they would surpass the threshold values for treatment.¹⁴

The surprising finding of this study was the wide variation in findings within GMFCS level and the similarities among GMFCS levels while still being diverse from the typically developing group. For example, the prevalence of increased peak knee flexion at initial contact was high: i.e., the vast majority of patients had increased knee flexion at initial contact in comparison to typically developing (Fig. 1e). One of the benefits of cumulative distribution function plots is that they provide an easy visual assessment of these observations and appreciation of the frequency of each parameter at each GMFCS level. Based upon visual inspection of the cumulative frequency plot for knee flexion at initial contact (Fig. 1e) the maximum value for typically developing population was approximately 10°. The majority of patients in each GMFCS level group were greater than 10° of flexion at initial contact: 93% of patients in the GMFCS level I group, 96% of GMFCS level II, and 98% of GMFCS level III. The biomechanical implication of increased knee flexion at initial contact is increased knee flexion in stance. Knowing that this is a very common gait issue at all GMFCS levels is relevant for providers in terms of treatment directions and providing parents with information about prognosis. Also, with an understanding of one’s threshold for treatment, the cumulative frequency plot can provide an estimate of the prognosis for treatment for each GMFCS level.

Also of note was the finding that certain gait parameters can be bidirectional at all GMFCS levels, such as peak ankle dorsiflexion in stance and foot progression angle. Irrespective of the GMFCS level, there are patients who will show increased, and others decreased, peak dorsiflexion in terminal stance. There is a common perception that with decreasing function, as would be described by GMFCS level III, in comparison to levels I or II, there is typically more crouch (increased knee flexion and ankle dorsiflexion in stance). The data in this study show that at GMFCS level III some patients will have crouch and some will be toe walkers. Clinicians should be aware that gait deformities are bidirectional within a given GMFCS level, and therefore may require very different treatment strategies.

The prevalence of gait deformities in patients with CP as a function of topographical classification has been evaluated in a large cohort by Wren et al.⁷ In this study, they found that 72% of patients with BSCP had a crouch gait pattern before surgery. Using the same definition of crouch as defined in the Wren article, (knee flexion greater than 1SD above normal for stance phase), we found a greater evidence of crouch in patients classified by GMFCS level (85% for GMFCS levels I and II, and 93% for GMFCS level III). In this study, 50% of GMFCS level III patients had greater than 40° of knee flexion at initial contact and 30% of GMFCS level III patients had greater than 40° of mean knee flexion in stance. This understanding of the prevalence of gait deformity in patients by GMFCS level is valuable information for clinicians who need to understand the potential for gait deformity, and ultimately treatment of their patients with established GMFCS levels.

A limitation of our study is that the typically developing group was younger and shorter than the individuals with BSCP at each GMFCS level. However, the typically developing group mean age was also well beyond the established age for mature gait patterns.¹⁵ Therefore, this difference in age will not result in a systematic difference in gait kinematics between the individuals with BSCP and the typically developing group because of age. We examined both height and age as covariates, and the results were almost identical. Therefore, we used height as the covariate in the final analysis, as height is likely to have a significant impact on spatiotemporal parameters. The results did not change when accounting for height in terms of the spatiotemporal parameters, nor for the knee kinematics. There were some changes at the pelvis, hip, and ankle kinematics. We could not identify a clinically meaningful basis for these changes. It is possible that the differences at the pelvis noted in the ANOVA are not clinically relevant (limited to a few degrees) and the differences at the ankle were a result of deformity being bidirectional. Another possible limitation is that the motion capture data from seven sites were combined for this analysis. To standardize study methods between sites, all study personnel underwent extensive on-site training by the study’s principal investigator. Slight differences in marker alignment protocols would increase variability within GMFCS groups. The statistically significant differences at p<0.01 were found in nearly all spatiotemporal and kinematic parameters after controlling for height and site, suggest that the differences in protocol as
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Supporting Information

The following additional material may be found online:

- Figure S1: Cumulative distribution plots for additional kinematic and spatiotemporal parameters used commonly to describe pathological gait.
- Figure S2: Probability distribution plots for additional kinematic and spatiotemporal parameters used commonly to describe pathological gait.
- Table S1: Expansion of Table I with confidence intervals and post hoc testing results for all parameters at all Gross Motor Function Classification System levels included.

References