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Multiple gait patterns within the same Winters class in children with hemiplegic cerebral palsy

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ABSTRACT

Background: Previous literature hypothesized that Winters type I are mainly characterized by a hypo-activation of dorsiflexors and type II by hyperactivation of plantarflexors around initial contact. However, it is currently not known if hemiplegic children belonging to the same Winters class really share the same muscle activation patterns, although this information might have relevant clinical implications in the patient management.

Methods: Gait data of 38 hemiplegic cerebral palsy children (16 Winters type I, 22 Winters type II) were analyzed, focusing on the foot and shank. A 2.5-minute walk test was considered, corresponding to more than 100 gait cycles for each child, analyzing the muscle activation patterns of tibialis anterior and gastrocnemius lateralis. The large stride-to-stride variability of gait data was handled in an innovative way, processing separately: 1) distinct foot-floor contact patterns, and for each specific foot-floor contact pattern 2) distinct muscle “activation modalities”, averaging only across gait cycles with the same number of activations, and obtaining, in both cases, the pattern frequency-of-occurrence.

Findings: At least 2 representative foot-floor contact patterns within each Winters group, and up to 4-5 distinct muscle activation patterns were documented.

Interpretation: It cannot be defined a predominant muscle activation pattern specific for a Winters group. For a correct clinical assessment of a hemiplegic child, it is advisable to record and properly analyze gait signals during a longer period of time (2-3 minutes), rather than (subjectively) selecting a few “clean” gait cycles, since these cycles may not be representative of the patient’s gait.
Keywords: Gait; gait analysis; spastic hemiplegic children; cerebral palsy; foot-floor contact; muscle activation patterns; EMG.

1. Introduction

In children, hemiplegia is a common consequence of cerebral palsy (CP) and causes altered selective motor control, weakness and spasticity. Gait analysis is frequently used in the management of CP children to support clinicians in surgery planning and outcome evaluation, botulinum toxin treatments, or in the choice of ankle-foot orthoses. Gait analysis is often combined with an electromyographic (EMG) analysis of the muscle activation patterns, or of the muscle synergies, to study CP dynamic motor control.

A correct classification of children with CP is important to assist diagnosis and clinical decision-making. The classification of spastic hemiplegia proposed by Winters et al. is widely accepted in literature. It is based on sagittal joint kinematics and introduces four classes of patients, with a progressive distal-proximal involvement of the paretic lower limb. Type I is defined by the presence of drop foot in swing, type II by the persistence of equinism throughout the gait cycle, with a possible knee hyperextension in stance, type III also shows a reduced knee flexion in swing and type IV, in addition, shows a reduced motion of the hip. However, the most common hemiplegic forms observed in CP are the mildest forms Winters type I and II.

In spite of the popularity of Winters classification, it is currently not known if children belonging to the same Winters class share the same EMG patterns, although this information might have relevant clinical implications in the patient management.

A recurring difficulty in the study of dynamic EMG patterns is dealing with the inherent variability of human gait. EMG patterns change from stride-to-stride and the study of multiple strides is
necessary, requiring the correct determination of the beginning and the end of each stride. This problem is traditionally handled collecting repeated trials where the subject hits one or more force plates arranged along the pathway. At each passage, 1-3 consecutive strides are recorded. Innovative techniques based on wearable sensors allows for recording long periods of uninterrupted walking. Independent from the technique used to handle intra-subject variability, the EMG signal is usually time-normalized with respect to the gait cycle to obtain ensemble averages from multiple strides.

A first problem encountered in this process is the “segmentation” of the EMG signal into separate gait cycles, i.e. the identification of the start and end of individual gait cycle. Frequently, in the clinical environment the EMG signal is segmented manually. However, this segmentation is dependent on the subjective judgment of the operator and even very experienced users may lack repeatability. Furthermore, manual segmentation is unpractical when evaluating long periods of walking. Hence, a fundamental but critical aspect of the EMG analysis is the evaluation of the gait cycle timing and the segmentation process. Foot contact event detection is particularly challenging in children with CP due to their initial contact with the forefoot. Sometimes the analysis is carried out selecting “clean hits” obtained from the force plate or by using pressure footswitches. The work by De Stefano et al. proposed (automatic and semi-automatic) methods as an alternative to the more traditional manual segmentation, and analyzed how the gait cycle selection affects the EMG analysis. They found that, in CP children, the segmentation method used for gait cycle selection has a major influence in the study of the EMG signal.

Furthermore, gait event detection is usually limited to initial contact and toe-off, thus neglecting the sub-phases of stance, which proved to be important in the evaluation of pathological subjects. In previous studies an algorithm for the segmentation of the foot-switch signal and the gait cycle classification was described and validated also in pathological gait and in particular in hemiplegic
children locomotion.\textsuperscript{27} It automatically provides the foot-floor contact sequences and complete gait phase timing of an uninterrupted walk of 2-3 minutes.

In the mildest forms of hemiplegia (Winters type I and II), preliminary results showed that there are at least 2 gait cycle “classes” characterizing each Winters group.\textsuperscript{27} Notice that a single child may contact the floor in different ways, while walking, and this may correspond to different EMG patterns. This holds especially for the muscles directly involved in the ankle joint movements. These findings suggested us that the EMG signal must be averaged across cycles belonging to the same foot-floor contact class.\textsuperscript{27,28}

However, even using this precaution of processing the EMG signal separately for each foot-floor contact class, there are other sources of EMG intra-subject variability that must be accounted for. More specifically, a single subject’s muscle does not show a single “preferred” pattern of activation, but up to 4-5 distinct EMG patterns, each characterized by a different number of activation intervals occurring within the gait cycle.\textsuperscript{28,29} This was demonstrated on a sample of 100 typically developing children.\textsuperscript{28} Similar findings were obtained, by other researchers, in healthy adults,\textsuperscript{29} indicating that “ensemble averaging” over different EMG activation modalities is not appropriate. Hence, the different EMG activation patterns relative to a specific class of gait cycles, and to a specific activation modality, must be considered separately.

The aim of this work was to study the dynamic EMG patterns of tibialis anterior (TA) and gastrocnemius lateralis (GL) correctly managing the intra-subject variability, and evaluating the percentage of occurrence of each pattern in CP children. In particular, it was explored the hypothesis that type I children are characterized by a hypoactivation of dorsiflexors (TA), and type II by a prolonged activation of plantar-flexors around initial contact (GL).\textsuperscript{16}

2. Materials and methods
2.1 Subjects

Gait analysis data were obtained retrospectively including CP children that were referred to the Gait Analysis Laboratory of our Rehabilitation Unit from 2005 to 2012. The laboratory database was searched for children with Winters’ type I and II spastic hemiplegia, that were the most common forms recorded in the database. The children GMFCS-level was between I and II. Cases that underwent previous lower limb orthopedic surgery, or botulinum toxin injections in the six months preceding the gait examination, were excluded from the study. Two raters (A.N., A.G.) examined independently the video-recording and kinematic data of children and selected a total of 38 CP patients matching the inclusion criteria: 16 type I (10 males/6 females, 11 right/5 left hemiplegia, age: mean 8.9 (SD 2.8) years, height: mean 131.9 (SD 15.6) cm; weight: mean 29.3 (SD 9.9) kg) and 22 type II (12 males/10 females, 13 right/9 left hemiplegia, age: mean 7.9 (SD 3.4) years, height: mean 122.8 (SD 19.4) cm; weight: mean 27.7 (SD 13.3) kg). Gait data from 100 normal developing children were available from a previous study, and have been used as reference. The research reported in this paper was undertaken in compliance with the ethical principles of the Helsinki Declaration.

2.2 Instrumentation and experimental protocol

Patients were equipped bilaterally with: a) foot-switches under the heel, the first and the fifth metatarsal heads, b) ankle electro-goniometers (sagittal plane), c) surface EMG probes positioned over TA and GL. The multichannel recording system used for the acquisitions was Step32. The size of foot-switches was 10 mm×10 mm (thickness: 0.5mm). Foot switches were fixed under the barefoot sole by double-sided adhesive tape. Single differential (SD) probes constituted by Ag-disks were used (diameter: 4 mm, interelectrode distance: 12 mm, gain: 1000, high-pass filter: 10 Hz, 2 poles). EMG signals were further amplified and low-pass filtered by the recording system (450 Hz, 6 poles). An overall gain from 1000 to 50,000 could be chosen to suit the need of the specific
muscle observed (input referred noise: ≤1 µVrms). Details on the crosstalk issue were described in a previous study on normal children.\textsuperscript{28} CP children walked barefoot at self-selected speed, back and forth over a 10-m walkway for 2.5 min. A video was captured synchronously to the gait signals. This was recorded by a single camera (part of the STEP32\textsuperscript{a}) focusing on the sagittal plane, acquiring 25 frames per second. This video provided a clinical record of a patient’s gait, but no quantitative data were obtained from it.

2.3 Data analysis: foot-floor contact

Time events were identified from the 4-level footswitch signal and the following gait phases were determined: heel contact (H), flat-foot contact (F), heel-off/push-off or “forefoot contact” (P), swing (S).\textsuperscript{22} HFPS is the normal sequence of gait phases, H-F-P being the normal sequence of the sub-phases of stance. To study the foot-floor contact sequences of CP children it was used an algorithm that segments the foot-switch signal and classifies the gait cycles even in presence of forefoot strikes or other abnormal cycles.\textsuperscript{22} Only the gait cycles corresponding to the child walking straight along the walkway were considered in this analysis, while the cycles corresponding to changes of direction, accelerations and decelerations (in proximity of the turns) were automatically removed by a multivariate statistical filter (included in the STEP32 software\textsuperscript{a}).

For each CP child a mean of 128 (SD 36) gait cycles for each lower limb were analyzed. The total number of gait cycles analyzed in the hemiplegic population was 9773.

The sequences of gait phases observed in the foot-floor contact analysis were named as follows:

- HFPS: normal sequence of gait phases (heel contact, flat-foot contact, push-off, swing)
- PFPS: after forefoot initial contact (P), the heel also touches the ground - hence the entire foot is in contact with the ground (F) - then the heel raises for push-off (P), and swing follows (S)
- **PS**: the heel never touches the floor, only the forefoot contacts the ground (P) during stance, and swing follows (S)
- **FPS**: the cycle initiates with flatfoot strike (F), then the heel raises for push-off (P), and swing follows (S)
- **OTHERS_P**: cycles initiating with a forefoot strike (P) but different from PFPS and PS, e.g. PFPSPS
- **OTHERS**: cycles not matching any of the previous categories.

The number of cycles belonging to each class: HFPS, PFPS, PS, FPS, OTHERS_P, and OTHERS was calculated.

### 2.4 Data analysis: EMG

The muscle activation intervals were obtained by a robust technique based on a double-threshold statistical detector.\(^{30}\) The input parameters of the detector (background noise, signal-to-noise ratio and duty cycle) were adaptively estimated every 30 seconds of signal.\(^{31}\) Then, for each child, muscle on/off instants were averaged considering, separately, each class of gait cycle and each modality of activation observed for that specific class, following “statistical gait analysis”.\(^{25,28}\)

“Modality of activation” means an EMG pattern characterized by a specific number of activation intervals within a single gait cycle: in the nth-activation modality the muscle is active in n intervals within the gait cycle. STEP32 software\(^a\) and custom Matlab®\(^b\) routines were used for EMG statistical analysis.

### 2.5 Statistical analysis: foot-floor contact

The frequency of occurrence of each class was calculated for type I and type II. Previous results on controls were reported for reference. Both the hemiplegic and non-hemiplegic sides of CP children were considered. Differences between the CP populations were assessed by two-sample t-tests (confidence level $\alpha=0.05$, 2 tails). The duration of the gait phases for the most representative
classes of each population was calculated. For the sake of simplicity, only the hemiplegic side of CP children was reported in this case. The duration of the gait phases among the populations was compared by two-sample $t$-tests (confidence level $\alpha=0.05$, 2 tails).

3. Results

The ankle kinematics of the two groups is reported in Fig. 1, for completeness.

3.1 Foot-floor contact

The frequency of occurrence of each foot-floor contact sequence is reported in Fig. 2, for hemiplegic and typically developing children. Controls were mainly characterized by HFPS cycles (88% of strides), type I by HFPS (35%) and PFPS (44%) cycles, and type II by PFPS (43%) and PS (40%) cycles. Notice that a single CP child may either show a prevalent sequence (observed in 80-90% of the strides) or more sequences equally representative of his/her gait (i.e., with a comparable frequency of occurrence). Table I reports the frequency of occurrence of the gait cycle classes for each child (hemiplegic side). A schematic representation of the foot-floor contact timing characterizing type I, type II and controls is shown in Fig. 3. The duration of the gait phases among the three populations was compared as follows.

3.1.1 HFPS: controls vs. type I

In the “normal” sequence (HFPS), type I showed differences in timing with respect to controls. The most significant difference was a shortened H-phase (mean 2.2 (SD 0.8) vs. mean 5.9 (SD 1.8) % of the gait cycle (GC), $p<0.001$). Moreover, push-off was slightly prolonged (mean 28.3 (SD 11.9) vs. mean 22.1 (SD 5.4) % GC, $p=0.006$), and swing was shortened (mean 33.8 (SD 12.4) vs. mean 39.5
The flat-foot contact (F) phase was not significantly different from controls (mean 35.7 (SD 8.0) vs. mean 32.5 (SD 5.6) %GC, p=0.11).

3.1.2 PFPS: type I vs. type II
In PFPS cycles, initial toe-contact (P) duration was not different between type I and II (mean 4.1 (SD 2.6) and 3.7 (SD 1.4) %GC, p=0.5), but the durations of the other sub-phases of stance were markedly different between groups. In particular, in type II F was significantly shortened (mean 12.1 (SD 7.3) vs. 36.6 (SD 8.1) %GC, p<0.001) and the push-off (P) was increased (mean 24.0 (SD 10.5) vs. 46.5 (SD 8.0) %GC, p<0.001). The whole stance phase (P+F+P) was not different between type I and II (mean 64.6 (SD 7.9) and 62.2 (SD 4.0) %GC, respectively, p=0.3).

3.1.3 PFPS vs. PS cycles in type II
In type II, stance was not different between PFPS and PS cycles (mean 62.2 (SD 4.0) and 63.1 (SD 4.5) %GC, respectively, p=0.6).

3.1.4 Stance: type I vs. type II
Considering HFPS and PFPS cycles for type I, and PFPS and PS cycles for type II, stance was not globally different between the CP groups (mean 65.3 (SD 9.6) vs. 62.6 (SD 4.2) %GC, p=0.17).

3.2 Muscle activation patterns
Figure 4 represents the EMG patterns characterizing the three populations. Due to the richness and complexity of these patterns, only their main features will be analyzed in the following. A first observation is that even selecting a specific type of foot-floor contact, e.g. type II-PS cycles, CP children showed more than one pattern of activation. Focusing on the last plot of TA, four different EMG patterns are clearly distinguishable: a pattern with a single activation occurred in 22% of the cycles, a pattern with 2 activations (32% of cycle), 3 activations (34% of cycles) and 4 activations (12% of cycles).

3.2.1 Tibialis anterior
Overall, hemiplegic children showed a curtailed activity during terminal swing and a lack of
activity at loading response, with respect to normal subjects. Controls showed a missing activity of TA, at initial contact, only in 1% of the strides. In type I children, this percentage raised to 9% in HFPS cycles, and further raised up to 63% (30 plus 33%) in PFPS cycles.

3.2.2 Gastrocnemius lateralis

Especially the type II group showed abnormal activity both at initial contact and in terminal swing. In controls, the activity of GL was completely absent in late swing (in preparation to initial contact) in 61% (27 plus 34%) of strides. In type I, this percentage decreased to 38% in HFPS cycles and to 27% in PFPS cycles. In type II, this percentage further decreased to 13% in PFPS cycles and to 9% in PS cycles.

4. Discussion

Traditionally, gait analysis considers only “standard” cycles for a specific subject under study. As an example, it implicitly assumes that a normal child walks using the sequence: heel contact, flat-foot contact, push, swing (HFPS). Possible “non-standard”, “unusual” or “atypical” cycles are commonly not studied, although they can represent more than 10% of the total gait cycles in normal children (Fig. 2). Similarly, it may be assumed that a hemiplegic child first contact the floor with the forefoot rather than the heel on the basis of the analysis of a few cycles, establishing that this is the “standard” behavior on his affected side. However, depending on the child, heel striking may be observed from time-to-time or very often (Table I). Knowing how often a pattern is observed is essential to determine if there is a single, prevalent, foot floor contact pattern, or if there are more than one pattern actually characterizing the child gait. On the other hand, neglecting this information may lead to erroneous interpretations of EMG gait data, especially for what concern the shank muscles. Hence, in general, classical gait analysis implicitly limits its investigation to a single gait cycle class, i.e. a predetermined “template” of gait phases that is considered “characteristic” of the subject under study. Similarly, this approach fails to recognize the presence of different muscle activation patterns in a subject’s walk. The other patterns are usually discarded, because they do not
match the “characteristic pattern” expected, or, even worse, averages are carried out over different (unrecognized) cycle types, and different (unrecognized) activation patterns.

In this study, the prevalent foot-floor contact patterns of CP children were evaluated with a portable and low-cost system based on simple foot-switches. Considering long periods of walking, the different foot-floor contact patterns of hemiplegic children were studied and the percentage of occurrence of each cycle type was calculated, through an automatic analysis of more than 100 gait cycles for each child (“statistical gait analysis”). The duration of the gait phases for each cycle type was also calculated. Furthermore, by this approach it was explored the richness of the EMG patterns in the two groups of hemiplegic children, revealing information that can be relevant in the clinical management of CP.

4.1 Foot-floor contact

Type I children showed two main classes of gait cycles: cycles where the heel-rocker was preserved (HFPS), and cycles in which forefoot initial contact was followed by flat-foot contact, push-off and swing (PFPS). Type II children also showed PFPS cycles, but with a much shortened F-phase. In addition, they showed cycles in which the F-phase was completely missing (PS). Hence, two different foot-floor contact patterns characterized each Winters group, confirming previous results.\textsuperscript{27} Within each group, some of the children mainly adopted one of the two patterns, but some others showed a higher stride-to-stride variability, alternating both patterns during their walk. This should point out the importance of analyzing long and uninterrupted walking trials instead of selecting a few “clean” gait cycles. A few “selected” cycles may not be representative of a child’s real gait, and do not allows for evaluating stride-to-stride variability.

Foot contact event detection is usually limited to initial contact and toe-off.\textsuperscript{20,21} However, this kind of gait timing does not allow for distinguishing between type I and II. In the present study, stance was not different for the two CP groups. Usually, the ankle kinematics in the sagittal plane is used for the Winters’ classification in type I and II.\textsuperscript{10,13} Previous works pointed out some of the
difficulties that may arise in applying the Winters’ classification, particularly in the milder forms, which are the most frequently observed in clinics. Foot-contact event detection, extended to the sub-phases of stance, may be explored as an alternative (or complementary) method to perform the classification of CP children in the milder forms of hemiplegia. Hence, another message derivable from the present study is that it is important to consider also the sub-phases of stance, since they allow for discriminating type I and II, while analyzing stance and swing only wouldn’t have shown any difference between the CP groups.

4.2 EMG patterns

The results showed that it cannot be defined a predominant EMG pattern, specific for a Winters group. Although in type I it was mainly evident a reduced activation of dorsi-flexors with respect to controls and in type II a pre-activation of plantar flexors, our findings highlights the high variability of the muscle activation patterns. This should stress the importance to analyze separately the muscle activation patterns belonging to: a) different gait cycle classes; b) different EMG activation modalities.

More specifically, the EMG patterns of TA were useful especially in the interpretation of the foot-floor contact results concerning type I children. The shortening of the H-phase in cycles with a preserved heel-rocker, and the presence of forefoot contact in PFPS cycles, were associated with a progressively diminished activity of TA around initial contact. On the other hand, especially the type II group showed abnormal GL activity both at initial contact and in terminal swing. This observation is in agreement with the common clinical finding of spasticity in this group, causing equinus at initial contact and throughout the stance phase. However, the EMG patterns of GL were useful in the interpretation of the gait abnormalities of both CP groups. The progressively anticipated activity of GL may explain the progressive worsening in the foot-floor contact sequences, ranging from the “milder” cycles of type I, showing a preserved heel-rocker (HFPS cycles), up to the “more severe” cycles of type II, where the F-phase is completely lost (PS cycles).
The fact that it cannot be defined a predominant muscle activation pattern has clinical implications. In fact, while it has already been demonstrated that a clinical evaluation in static conditions is not sufficient to assess gait functionality,\textsuperscript{32} this study suggests that a kinematic evaluation of gait for Winters classification may not be sufficient to orient the rehabilitative choices. The EMG investigation in dynamic conditions may be an important element to define a personalized rehabilitation program.

4.3 Study limitations

EMG data of the non-hemiplegic side were not complete across all CP children and hence they were not reported. For some of the children knee kinematics and EMGs of thigh muscles (rectus femoris, lateral hamstrings, vastus lateralis) were also available, but again they were not complete across CP children and hence they were not mentioned in this study. A more complete gait protocol could have allowed for a better understanding of variability/diversity and individuality of gait in CP children.

5. Conclusions

In conclusion, this work showed that it cannot be defined a predominant muscle activation pattern, specific for a Winters group. This highlighted that a dynamic EMG study of the single child during a 2-3 minutes “real” walk may help a personalized approach to patient’s care. In particular, the pattern percentage-of-occurrence may be used to quantify how often a child exploits a specific gait pattern, and, consequently, to what extent this pattern is representative of his/her gait. This also provides information on the patient intra-subject variability, an important aspect to be considered in the longitudinal follow-up of patients. Furthermore, the presence of several distinct patterns of foot-floor contact and EMG activation in the same Winters type, may be used as a new or at least a supplementary classification for distinguishing hemiplegic CP children, possibly aiding
individualized therapeutic care or intervention decision.

References


Suppliers

a. STEP32; Demitalia, sold by Medical Technology, Via Bogetto 8, Torino, IT 10144.
http://www.medicaltec.it/STEP32.html

b. MATLAB®; The MathWorks Inc, 3 Apple Hill Dr, Natick, MA 01760-2098.
Figure captions

Fig. 1 - Ankle kinematics of the hemiplegic and non-hemiplegic sides are displayed for the two groups. Mean (solid lines) and mean plus/minus one standard error (dashed lines) are displayed.

Fig. 2 - Classes of gait cycles with different foot-floor contact sequences and their percentage of occurrence for Winters type I and II (mean and standard error are displayed for each population). Controls are also reported for reference. Upper plot: hemiplegic side of CP children; lower plot: contralateral side. The average between left and right sides is reported for controls, in both plots. Significant differences between type I and II are indicated with an asterisk ($p<0.05$). For the sake of simplicity, no statistical results are reported for the differences between controls and the two CP groups.

Fig. 3 - Gait-phase timing for the main foot-floor contact sequence/s of each population. The hemiplegic side is reported for CP children and the average between left and right sides for controls.

Fig. 4 - Muscle activation intervals for tibialis anterior and gastrocnemius lateralis: for each group (controls, type I and type II) and each foot-floor contact sequence characterizing the group (HFPS for controls, HFPS and PFPS for type I, PFPS and PS for type II). Five muscle activation modalities are represented (with 1 to 5 activations occurring during the gait cycle). Horizontal bars are grey-level coded in order to represent the number of children whose muscle was active. Black: all the children activated the muscle, white: none of the children activated the muscle. On the right-hand side of each plot the frequency of occurrence of each modality was reported. The timing of the gait phases is shown superimposed, marked by dashed vertical lines.
Figure 1
Figure 2
Figure 3
Figure 4
Table 1

<table>
<thead>
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<th>PS (%)</th>
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Percentage of occurrence of the foot-floor contact sequences for each child (hemiplegic side).
Highlights

- Population: 38 hemiplegic children (16 Winters type I, 22 type II)
- EMG patterns of tibialis anterior and gastrocnemius lateralis are analyzed
- Different foot-floor contact leads to distinct EMG patterns within the same group
- For each specific gait cycle class, there are up to 4-5 distinct EMG patterns