

**Delft University of Technology** 

# Analysis of gait patterns pre- and post- Single Event Multilevel Surgery in children with Cerebral Palsy by means of Offset-Wise Movement Analysis Profile and Linear Fit Method

Ancillao, Andrea; van der Krogt, Marjolein; Buizer, Annemieke I.; Witbreuk, Melinda; Cappa, Paolo; Harlaar, Jaap

DOI 10.1016/j.humov.2017.08.005

Publication date 2017 Document Version Final published version Published in Human Movement Science

# Citation (APA)

Ancillao, A., van der Krogt, M., Buizer, A. I., Witbreuk, M., Cappa, P., & Harlaar, J. (2017). Analysis of gait patterns pre- and post- Single Event Multilevel Surgery in children with Cerebral Palsy by means of Offset-Wise Movement Analysis Profile and Linear Fit Method. *Human Movement Science*, *55*, 145-155. https://doi.org/10.1016/j.humov.2017.08.005

# Important note

To cite this publication, please use the final published version (if applicable). Please check the document version above.

# Copyright

Other than for strictly personal use, it is not permitted to download, forward or distribute the text or part of it, without the consent of the author(s) and/or copyright holder(s), unless the work is under an open content license such as Creative Commons.

Takedown policy

Please contact us and provide details if you believe this document breaches copyrights. We will remove access to the work immediately and investigate your claim. Contents lists available at ScienceDirect

# Human Movement Science

journal homepage: www.elsevier.com/locate/humov

Full Length Article

# Analysis of gait patterns pre- and post- Single Event Multilevel Surgery in children with Cerebral Palsy by means of Offset-Wise Movement Analysis Profile and Linear Fit Method



Andrea Ancillao<sup>a,b,\*</sup>, Marjolein M. van der Krogt<sup>b</sup>, Annemieke I. Buizer<sup>b</sup>, Melinda M. Witbreuk<sup>c</sup>, Paolo Cappa<sup>a,1</sup>, Jaap Harlaar<sup>b</sup>

<sup>a</sup> Dept. of Mechanical and Aerospace Engineering, "Sapienza" University of Rome, Via Eudossiana 18, 00184 Roma, Italy
 <sup>b</sup> Dept. of Rehabilitation Medicine, VU University Medical Center, Amsterdam Movement Sciences, De Boelelaan 1117, 1081 HV Amsterdam, The Netherlands

<sup>c</sup> Dept. of Orthopedic Surgery, VU University Medical Center, Amsterdam Movement Sciences, Van der Boechorststraat 7, 1081 BT Amsterdam, The Netherlands

# ARTICLE INFO

Keywords: Cerebral Palsy Gait analysis GPS Movement Analysis Profile SEMLS Synthetic indices

# ABSTRACT

Gait analysis is used for the assessment of walking ability of children with cerebral palsy (CP), to inform clinical decision making and to quantify changes after treatment. To simplify gait analysis interpretation and to quantify deviations from normality, some quantitative synthetic descriptors were developed over the years, such as the Movement Analysis Profile (MAP) and the Linear Fit Method (LFM), but their interpretation is not always straightforward.

The aims of this work were to: (i) study gait changes, by means of synthetic descriptors, in children with CP that underwent Single Event Multilevel Surgery; (ii) compare the MAP and the LFM on these patients; (iii) design a new index that may overcome the limitations of the previous methods, i.e. the lack of information about the direction of deviation or its source.

Gait analysis exams of 10 children with CP, pre- and post-surgery, were collected and MAP and LFM were computed. A new index was designed as a modified version of the MAP by separating out changes in offset (named OC-MAP).

MAP documented an improvement in the gait pattern after surgery. The highest effect was observed for the knee flexion/extension angle. However, a worsening was observed as an increase in anterior pelvic tilt. An important source of gait deviation was recognized in the offset between observed tracks and reference. OC-MAP allowed the assessment of the offset component versus the shape component of deviation.

LFM provided results similar to OC-MAP offset analysis but could not be considered reliable due to intrinsic limitations. As offset in gait features played an important role in gait deviation, OC-MAP synthetic analysis was proposed as a novel approach to a meaningful parameterisation of global deviations in gait patterns of subjects with CP and gait changes after treatment.

E-mail addresses: andrea.ancillao@hotmail.com (A. Ancillao), m.vanderkrogt@vumc.nl (M.M. van der Krogt), ai.buizer@vumc.nl (A.I. Buizer),

http://dx.doi.org/10.1016/j.humov.2017.08.005

Received 1 March 2017; Received in revised form 16 June 2017; Accepted 8 August 2017 Available online 19 August 2017 0167-9457/ © 2017 Elsevier B.V. All rights reserved.

Abbreviations: GA, gait analysis; SD, standard deviation; RoM, Range of Motion; RMS, root mean square; CP, Cerebral Palsy; GDI, Gait Deviation Index; GPS, Gait Profile Score; GVS, Gait Variable Score; LFM, Linear Fit Method; MAP, Movement Analysis Profile; MCID, Minimally Clinical Important Difference for GPS; OC-GPS, Offset Corrected - Gait Profile Score; OC-GVS, Offset Corrected - Gait Variable Score; OC-MAP, Offset Corrected - Movement Analysis Profile; R, Pearson's coefficient of correlation; SEMLS, Single Event Multilevel Surgery; TD, typically developing children

Corresponding author at: Dept. of Mechanical and Aerospace Engineering, "Sapienza" University of Rome, Via Eudossiana 18, Roma, Italy.

m.witbreuk@vumc.nl (M.M. Witbreuk), j.harlaar@vumc.nl (J. Harlaar). <sup>1</sup> Deceased.

# 1. Introduction

Gait analysis (GA) is a multifactorial and powerful tool that provides a quantitative description of normal and pathological gait patterns. It is therefore widely adopted as a routine exam in specialized clinical centres (Carriero, Zavatsky, Stebbins, Theologis, & Shefelbine, 2009; Whittle, 1996). For instance, clinical GA was used to characterize: Parkinson's disease (Sale et al., 2013), Down syndrome (Galli, Rigoldi, Brunner, Virji-Babul, & Giorgio, 2008), and Cerebral Palsy (CP) (Carriero et al., 2009; van den Noort, Ferrari, Cutti, Becher, & Harlaar, 2013) and it was used to validate novel treatments (Camerota et al., 2015; Sale et al., 2013; Vismara et al., 2016). GA was proved useful especially to aid the selection of optimal treatment in the case of spastic Cerebral Palsy (CP), which may involve different kinds of motor disorders and therefore different gait patterns (Galli, Cimolin, Rigoldi, Tenore, & Albertini, 2010; Piccinini et al., 2011). Moreover, GA allowed the quantification of changes in gait patterns of subjects with CP after treatment such as surgery (Galli, Cimolin, Crivellini, & Albertini, 2009).

GA exams usually consist of the integration of data from different sources, namely: kinematic data, kinetic data, video recording, electromyography, etc. Thus, a single GA exam contains a large volume of data that is processed into a high dimensional space of parameters, such as spatiotemporal parameters, joint/segment angles, forces, moments, etc. All these parameters are usually presented in the form of a clinical report, i.e. a collection of tracks (the time evolution of a variable as a function of the gait cycle) and numerical parameters (Stebbins et al., 2014; Whittle, 1996). A gait report can be difficult to understand and requires specific training of the clinicians. So the need to represent gait by means of a reduced number of parameters (e.g. a classification) emerged. Many studies focused on the validation of synthetic descriptors that could classify the severity of a pathological gait pattern by quantifying the deviation from a normality range. Such synthetic numbers are useful for treatment follow up evaluation or to study the natural evolution of the gait pattern over time (Galli, Cimolin, De Pandis, Schwartz, & Albertini, 2012).

A recently proposed and widely used index is the Gait Deviation Index (GDI) (Schwartz & Rozumalski, 2008). It is an overall, dimensionless, multivariate and comprehensive index that provides an overall measure of gait quality (Esbjörnsson et al., 2014). It was applied to children with CP (Cimolin, Galli, Vimercati, & Albertini, 2011; Molloy, McDowell, Kerr, & Cosgrove, 2010), showing a good repeatability, with an uncertainty of  $\pm$  10% (Massaad, Assi, Skalli, & Ghanem, 2014). Moreover, the GDI was successfully used to quantify gait deviations in subjects with Parkinson's disease (Galli et al., 2012) and rheumatoid arthritis (Esbjörnsson et al., 2014).

The main limitation of GDI is that, even though it is useful to assess the overall gait pattern, being a single number, it is inherently not informative on the location of the impairment (Massaad et al., 2014). This limitation was addressed by a related method, i.e. the Movement Analysis Profile (MAP) (Baker et al., 2009). The MAP is based on the computation of a deviation index, named "Gait Variable Score" (GVS), for nine relevant kinematic variables (joint angles). The GVSs quantify the deviation from normality for each gait feature and they can be averaged into an overall index, named "Gait Profile Score" (GPS). GPS was shown to be strongly correlated to GDI (Baker et al., 2009).

Validity studies showed a GPS Minimally Clinical Important Difference (MCID), i.e. 1.6° (Baker et al., 2012), while several studies were conducted about GPS reliability when applied to subjects with pathology. E.g. GPS was used to study gait deviation in subjects with Ehlers-Danlos Syndrome (Celletti et al., 2013), concluding that the GPS and MAP are appropriate for the evaluation of functional gait limitation in these patients. GPS was also used for the characterization of gait in children with CP and other neurological/ orthopaedic disorders (Beynon, McGinley, Dobson, & Baker, 2010). Results showed a good correlation with other qualitative ratings of kinematic gait deviation. The effects of orthopaedic interventions on gait in children with CP were studied by Rutz, Donath, Tirosh, Graham, and Baker (2013), finding a pre-operative GPS of  $15.5^{\circ} \pm 3.9^{\circ}$  that reduced to  $11.2^{\circ} \pm 2.5^{\circ}$  post orthopaedic intervention. They observed that the degree of improvement was higher in the patients with the worst initial conditions. GPS score was demonstrated of being correlated to the strength of the subject and it was observed that gait kinematics grossly depended on muscle strength (Schweizer, Romkes, Coslovsky, & Brunner, 2014). This finding confirmed that muscle strength influences stability of ligaments and quality of the motor performance in general (Ancillao, Rossi, & Cappa, 2017). Gait performance was also influenced by cognitive load and dual task activities in subjects with Parkinson's Disease and the GPS was able to detect changes in gait, changing from  $9.17^{\circ} \pm 1.18^{\circ}$  of the "normal gait" condition to  $10.30^{\circ} \pm 1.37^{\circ}$  of the "dual task" condition (Speciali et al., 2014). Another study investigated the walking characteristics in individuals with Multiple Sclerosis, concluding that the single measure of GPS can characterize gait kinematics of such patients (GPS =  $9.12^{\circ} \pm 2.28^{\circ}$ ). Moreover a correlation between GPS and the "Expanded Disability Status Scale" was observed (Pau et al., 2014). Strong correlation between GPS and clinicians' ratings was also previously observed by (Beynon et al., 2010).

Even though the MAP allows to localize the anatomical joint or segment whose pattern deviates from normality, it is still limited in describing which is the nature of the deviation, e.g. the offset between curves, the scaling factor or a time-shift. Identifying the source of deviation is clinically important as it allows to more precisely identify which kind of limitation is affecting gait. E.g. crouch gait, that involves persistent knee flexion, is mainly characterized by an offset in knee flex/ext tracks. Changes in gait patterns, due to surgical procedures, are often observed as changes in the offset of some gait features (Sutherland & Davids, 1993). Thus more detailed synthetic descriptors, which take into account the offset and quantify its effects, are likely to be more informative to the clinical user.

A different approach to compare gait features to reference data was proposed by Iosa et al. (2014). The method allows to assess similarity between the observed waveform and reference GA tracks, in terms of shape, amplitude and offset. It consists of the application of a Linear Fit Method (LFM) to two time-normalized datasets. The result of the LFM are: (i) the  $R^2$  regression coefficient, that quantifies the strength of relationship between the tracks; (ii) the *a0* coefficient, i.e. the constant term of polynomial regression that represents the scalar addition (shift or offset) between the compared datasets; (iii) the *a1* coefficient, i.e. the first coefficient of first order polynomial regression that represents the amplitude scaling factor. When LFM is used to compare a GA exam to a control group, the  $R^2$ , *a0*, and *a1*, parameters can be assumed as synthetic descriptors of deviance from normality. Anyway, it was proved that a0 and a1 lose their meaning when  $R^2$  is lower than 0.5 (Iosa et al., 2014). The LFM method was tested on the sagittal kinematics of hip, knee and ankle of patients with cerebrovascular accident, concluding that it is a simple method to implement and, since it takes into account all the data points of GA tracks, it was concluded to be appropriate and reliable to discriminate between subjects with pathology and healthy subjects, with good sensitivity and specificity (Iosa et al., 2014). However, it is unknown whether the LFM method would be a meaningful measure to assess gait after surgery in subjects with CP.

The aims of this work were to: (i) study gait changes, by means of synthetic descriptors, in children with CP and crouch gait that underwent Single Event Multilevel Surgery (SEMLS); (ii) evaluate three different synthetic indices to assess gait patterns pre- and post-surgery. Special attention was paid in studying the offset between the baselines of observed tracks and reference, as crouch gait was expected to induce deviations mainly in terms of offset.

The implemented indices were: (i) the widely used and clinically validated MAP; (ii) a recently proposed index, i.e. the LFM; and (iii) a new index designed to overcome the limitations of the previous methods by separating the pure offset component from the gait deviation due to different curve shape. Outcomes of the three different methods were compared.

## 2. Material and methods

#### 2.1. Subjects

Nine children diagnosed with bilateral CP, age 11.1  $\pm$  2.4 years, 7 males, 2 females were enrolled in this study. All the subjects were patients followed by the Department of Rehabilitation Medicine of VUmc, Amsterdam, NL. They were evaluated by means of Gross Motor Function Classification System, at the time of admission, obtaining rankings from II to III. The subjects had no prior orthopaedic surgery or botulinum toxin treatment within the previous 16 weeks. All subjects underwent Single Event Multilevel Surgery (SEMLS), involving hamstrings release for all the subjects, and femur or tibia rotation and muscle/tendon repositioning in some cases. Gait analysis was recorded before treatment, i.e.  $2.9 \pm 2.1$  months pre-treatment, and after treatment, i.e.  $14.4 \pm 4.5$  months post-treatment, to investigate changes in gait. The control group was composed of 11 typically developing children (TD), aged 8.2  $\pm$  1.8 years.

This study complied with the principles of the Declaration of Helsinki, and it was approved by the Medical Ethics Committee of the VU University Medical Center.

## 2.2. Equipment and procedures

GA data of both groups were collected in the Motion Analysis Laboratory of the VUmc, Amsterdam, NL. Kinematic data were collected by means of an Optotrak Optoelectronic System (Northern Digital, Waterloo, Ontario) composed of 3 sensors, each one holding 3 cameras. Sampling frequency was 100 Hz. Marker protocol used was the CAST model (Cappozzo, Catani, Della Croce, & Leardini, 1995). The protocol required a calibration trial to identify anatomical landmarks. After that, subjects were asked to complete some practice trials on the walkway to ensure they were comfortable with the experimental procedure. Then, at least 5 walking trials were recorded for each subject. In each trial, subjects were asked to walk barefoot, at a self-selected speed, on the lab's walkway. From each trial, a complete stride was obtained for both hemisides.

The data were processed using BodyMech (http://www.BodyMech.nl), a custom-made software based on MATLAB (the Mathworks, USA), to obtain kinematic joint angles and spatiotemporal parameters by solving the CAST model (Cappozzo et al., 1995). The parameters obtained for each subject were averaged across the recorded strides of the same subject. From the GA dataset of each subject, we obtained the nine bilateral gait features that are required to compute the MAP (Baker et al., 2009). The selected gait curves were: pelvic tilt, obliquity and rotation; hip flexion, abduction and rotation; knee flexion; ankle dorsiflexion and foot progression angles, that were previously identified as the most representative features of gait (Baker et al., 2009).

#### 2.3. Movement Analysis Profile

The computation of the GVSs composing the MAP was implemented as indicated by Baker et al. (2009). The normality dataset was obtained by recording the GA of TD group. Gait features were then averaged across subjects and the mean values were assumed as representative of the control subjects. Namely, GVSs are computed as the RMS difference between a normalized *i*-th gait variable and the respective reference data (Eq. (1)):

$$GVS_{i} = \sqrt{\frac{\sum_{t=1}^{T} (x_{i,t} - \bar{x}_{ref,i,t})^{2}}{T}}$$
(1)

where  $x_{i,t}$  is the value of the *i*-th gait feature at the point *t* of gait cycle, T is the number of points in which the gait cycle has been divided (i.e. 100) and  $\overline{x}_{ref.i.t}$  is the average value for reference population.

The GPS is then computed as RMS average of all GVSs:

(8)

$$GPS = \sqrt{\frac{\sum_{i=1}^{N} (GVS_i)^2}{N}}$$
(2)

The GPS represents the overall deviation of a patient's data from the reference dataset. Thus, the higher the GPS, the worst. GVS and GPS indices were computed for both left and right side of each subject separately. Data were pooled in order to obtain one value for pre-surgery and one value post-surgery for each parameter and for each leg. The pelvis parameters were not pooled to avoid doubling the data, and only data from the right side was used. The average deviations from normality and their SDs were represented as bar plots (MAP). The final MAP contained 9 groups of bars representing the examined gait features, pre- and postintervention, plus 1 group of bars representing the overall GPS, pre- and post-intervention.

# 2.4. Offset Corrected Movement Analysis Profile

To take into account the effect of offset on kinematic gait features, we re-computed GVSs after removing the offset from waveforms. New indices were named as offset-corrected (OC)-GVS, OC-MAP and OC-GPS. Offset was defined as the linear distance between the average value of the gait curves over the gait cycle and the average value of the respective gait curves obtained from control group. Namely, the offset for the i-th gait feature,  $x_i$ , was defined as:

$$offset_i = \overline{x}_i - \overline{x}_{ref,i} \tag{3}$$

where  $\overline{x}_i$  represented the average value of the i-th gait feature.

Eqs. (1) and (2) were re-implemented as:

$$OC-GVS_{i} = \sqrt{\frac{\sum_{l=1}^{T} (x_{i,l} - offset_{l} - \overline{x}_{ref,i,l})^{2}}{T}}$$

$$OC-GPS = \sqrt{\frac{\sum_{l=1}^{N} (OC - GVS_{l})^{2}}{N}}$$
(5)

As in the MAP, the OC-GVSs and the OC-GPS were represented by bar plots, named OC-MAP. Also the offsets of gait features were represented by a bar plot, containing 10 groups of bars representing the examined gait features, pre and post intervention, and the overall RMS average of offset.

#### 2.5. Linear Fit Method

The LFM method was implemented as described in Iosa et al. (2014). Coefficients were obtained according to the following equations:

$$a_{1} = \frac{\sum_{t=1}^{N} (x_{ref,t} - \overline{x}_{ref})(x_{t} - \overline{x})}{\sum_{t=1}^{N} \cdot (x_{t} - \overline{x}_{ref})^{2}} x_{ref}$$
(6)  

$$a_{0} = \overline{x} - a_{1} \cdot \overline{x}_{ref}$$
(7)  

$$R^{2} = \frac{\sum_{t=1}^{N} (a_{0} + a_{1} \cdot x_{t} - \overline{x}_{ref})^{2}}{\sum_{t=1}^{N} (x_{t} - \overline{x}_{ref})^{2}}$$
(8)

where  $x_t$  is the value at point t of gait vector; N is the number of data points in the gait vector;

 $R^2$  measures the strength of linear relationship between x and  $x_{ref}$  al represents the amplitude scaling factor; a0 represents the scalar addition (shift or offset). In case of maximum similarity in waveforms, the parameters assume the following reference values:  $R^2 = 1$ ; a1 = 1; and a0 = 0. LFM was computed as "overall" value on all the 9 gait features pooled in a single gait vector and compared to a normality gait vector built in the same way. The LFM analysis was also conducted on each of the 9 gait features separately.

## 2.6. Statistics

Descriptive statistics analysis was run on data from the three methods. To ensure validity of the statistical tests and to choose the



Fig. 1. Movement Analysis Profile containing average values and SDs of GPS and GVS of 9 examined gait features, pre- and post-intervention. \* significant differences (p < 0.05).

most appropriate one, data groups were preliminary tested for normality by means of the Shapiro-Wilk test. The paired *t*-test was then used. Data were tested for differences between means pre-post intervention for all the parameters. Tests were assumed significant with an alpha level of 0.05.

The variations in gait features were quantified by computing pre-post differences for each GVS and OC-GVS. The differences were then compared to the GPS Minimally Clinical Important Difference (MCID), i.e. 1.6° (Baker et al., 2012). The average among subjects for the change post treatment in each GVS was computed. In order to represent the improvement level associated to a certain GP, a regression plot of the pre-post difference vs. GPS pre intervention was computed, as suggested by (Rutz et al., 2013), A correlation analysis was also conducted between the respective MAP, OC-MAP, Offset and LFM parameters to study the relationship between the different indices. To compute correlation, data from pre- and post-treatment analyses were pooled. The results were presented in the form of a correlation table. Statistical analysis was conducted by means of MS Excel software.

## 3. Results

# 3.1. Movement Analysis Profile

The MAP is depicted in Fig. 1, where values pre- and post-intervention are compared. The differences pre-post, their comparison to MCID and the statistical test are reported in Table 1.

Absolute values of pre-post differences were higher than the MCID (i.e. 1.6°) for all parameters, except hip flexion and pelvic rotation. The positive differences suggested an improvement towards normality. A negative variation suggested a worsening in pelvic tilt (Table 1, first column). The highest improvement was observed for the knee flexion and was confirmed by a statistically significant reduction of the index (Table 1). A significant improvement was also observed for the ankle dorsiflexion and foot progression (Fig. 1 and Table 1).

In Fig. 2 the variation of GPS pre-post is plotted against the GPS score pre-treatment for each subject. A linear trend was observed: the most severe initial conditions corresponded to the highest improvements.

Table 1

Mean across subjects of the differences pre-post intervention for MAP, OC-MAP and Offset analysis. Positive differences indicate improvement. § pre-post higher than the MCID (i.e.  $1.6^\circ$ ). \* significant differences (p < 0.05).

	MAP [°]			OC-MAP	[°]		Offset [°]			
	Mean	SD	p-value	Mean	SD	p-value	Mean	SD	p-value	
GPS/OC-GPS/RMS	5.10 §	4.59	≪0.01*	1.14	1.57	0.01*	5.14	4.51	< 0.01*	
Pel. Tilt	-2.92§	6.77	0.26	-0.06	1.02	0.88	-11.53	8.47	< 0.01*	
Pel. Obl.	3.14§	6.31	0.20	0.59	1.52	0.30	3.64	6.71	0.16	
Pel. Rot.	1.46	3.69	0.29	0.63	1.02	0.12	-0.12	4.81	0.95	
Hip Flex	0.66	6.03	0.66	-0.22	2.22	0.69	-5.07	9.92	0.05	
Hip Abd.	3.18§	5.68	0.03*	0.46	1.51	0.23	0.63	7.31	0.73	
Hip Rot.	5.53§	11.46	0.06	1.45	2.15	0.01*	-1.53	15.01	0.68	
Knee Flex.	12.38§	12.94	≪0.01*	1.51	2.47	0.02*	13.55	14.99	< 0.01*	
Ankle Dors.	4.77§	7.54	0.02*	2.41	3.03	< 0.01*	-5.80	9.14	0.02*	
Foot Prog.	5.15 §	9.86	0.04*	1.72	3.24	0.04*	3.87	11.88	0.20	



Fig. 2. Linear regression analysis of level of variation in GPS scores and their values pre intervention.



Fig. 3. Offset-Corrected Movement Analysis Profile containing: (a) average values and SDs of OC-GPS and OC-GVS of 9 examined gait features, pre- and post-intervention; (b) Average values and SDs of measured Offset for the gait features, pre and post intervention. \* significant differences (p < 0.05).



Fig. 4. Knee flexion/extension angle of a children pre- and post-intervention. The offset is the linear distance between the baselines of observed angle and normative data.

# 3.2. Offset-Corrected Movement Analysis Profile

The OC-MAP is depicted in Fig. 3a and the analysis of the Offset Profile is depicted in Fig. 3b. Numerical values of the differences pre-post are shown in Table 1.

OC-MAP showed lower scores than the MAP. Offset scores were higher than the respective OC-GVSs. Significant improvements were observed in OC-GPS, and OC-MAP of hip rotation, knee flexion, ankle dorsiflexion and foot progression (Fig. 3a and Table 1). The highest improvements were at knee flexion, ankle dorsiflexion and foot progression (Fig. 3a) to which corresponded also high improvements in terms of offset (Fig. 3b). The highest variations in Offset were observed at the pelvic tilt and knee flexion. As positive values of the knee flexion/extension angle are associated to the flexion, a positive offset indicated a condition of permanent flexion. More in detail, we observed a positive offset in the pre-intervention that reduced in the post-, indicating that some deviation remained towards flexion. Positive ankle dorsi/plantar flexion angles are associated to dorsiflexion. In this work, the Offset Profile documented a persistent plantarflexion in the pre-intervention, represented by a negative offset. The offset reduced to  $\sim 0^{\circ}$  in the post, documenting an improvement (Fig. 3b). The offset in the pelvic tilt also changed from a negative value, meaning a posterior tilt, towards anterior tilt with a significant difference pre- and post-treatment (Fig. 3b and Table 1). The RMS average showed a significant overall improvement in the offset (Fig. 3b and Table 1).

The effect of the offset on gait tracks is illustrated in Fig. 4. Knee flexion/extension angle of a subject, pre- and post- intervention, is depicted and compared to reference data. The offset is represented by the distance between the baseline of the observed track and the baseline of normative data.

# 3.3. Linear Fit Method

LFM analysis is depicted in Fig. 5. Pre-post differences are shown in Table 2.

The  $R^2$  parameter of LFM indicated an improvement from pre- to post-intervention in the overall kinematics, in the hip rotation, knee flexion, ankle dorsiflexion and foot rotation. The lowest values of  $R^2$  were observed for pelvic tilt and hip rotation (Fig. 5a), suggesting that the associated *a0* and *a1* could not be considered meaningful (Iosa et al., 2014). The *a0* coefficient (Fig. 5b) provided results similar, as a trend, to offset analysis shown in Fig. 3b, but absolute values were different. Statistically significant improvements in terms of Offset were observed for knee flexion and ankle dorsiflexion (Fig. 5b). The knee flexion showed a statistically significant improvement also in terms of scaling (*a1* coefficient), which was closer to 1 in the post (Fig. 5c). LFM parameters for the pelvic tilt showed a very high SD across subjects (Fig. 5b and c).

## 3.4. Correlation between methods

Results of correlation analysis are reported in Table 3, as the Pearson correlation coefficients R, computed between the respective results from MAP, OC-MAP/Offset and LFM components ( $R^2$ , a0 and a1) for each gait feature. The overall indices, i.e. GPS, OC-GPS and RMS were compared to the  $R^2$ , a0 and a1 from the overall LFM computing.

A strong correlation (> 0.70) was observed for: (i) the overall  $R^2$  of LFM and GPS/RMS; (ii) the *a0* and the *Offset* of each gait feature, with exception of pelvic tilt; (iii) all the LFM parameters of knee flexion and the respective MAP and OC-MAP; and (iv) the *a1* of the pelvic tilt and the respective OC-MAP. Moderate correlations were also observed between  $R^2$  and some gait features of OC-MAP. GPS was strongly correlated to OC-GPS and RMS of offset. Strong correlation was observed between the offset of pelvic



Fig. 5. Results of LFM analysis, averaged across subjects. \* significant differences (p < 0.05).

#### Table 2

Mean across subjects of the differences pre-post intervention for LFM parameters. A negative value of  $R^2$  Pre-Post difference means improvement. \* significant differences (p  $\leq 0.05$ ).

	$R^2$	$R^2$			a0			a1			
	Mean	SD	P value	Mean	SD	P value	Mean	SD	P value		
OVERALL	-0.18	0.13	≪0.01*	-0.66	3.84	0.49	0.00	0.25	0.99		
Pel. Tilt	-0.07	0.08	0.37	-3.99	23.63	0.65	-0.49	1.38	0.35		
Pel. Obl.	-0.09	0.25	0.34	2.92	5.79	0.19	-0.49	1.00	0.20		
Pel. Rot.	-0.13	0.19	0.09	-0.58	4.94	0.75	-0.20	0.30	0.09		
Hip Flex	-0.04	0.12	0.16	-5.20	9.81	0.06	0.01	0.18	0.91		
Hip Abd.	-0.08	0.29	0.29	0.69	7.22	0.70	-0.05	0.41	0.62		
Hip Rot.	0.16	0.26	0.02*	-1.44	15.01	0.70	-0.07	0.75	0.71		
Knee Flex.	-0.13	0.23	0.03*	17.83	17.13	< 0,01*	-0.16	0.17	< 0,01*		
Ankle Dors.	-0.10	0.23	0.04*	-6.01	9.02	0,01*	0.13	0.35	0.14		
Foot Prog.	-0.15	0.28	0.04*	2.80	13.05	0.39	-0.15	0.56	0.28		

#### Table 3

Correlation table showing the correlation between (i) the MAP, OC-MAP, Offset and the parameters computed from Linear Fit Method; (ii) MAP and the OC-MAP/ Offset parameters. Pearson R coefficients for each gait feature are shown. Back color indicates strong (> 0.7; dark grey) and moderate (> 0.4; light grey) levels of correlation.

		Pel. Tilt	Pel. Obl.	Pel. Rot.	Hip Flex	Hip Abd.	Hip Rot.	Knee Flex.	Ankle Dors.	Foot Prog.
	GPS	_				MAP				
F	-0.733	-0.219	-0.125	-0.131	-0.187	-0.399	-0.121	-0.525	-0.434	-0.053
а	<b>o</b> 0.398	0.139	0.837	0.548	0.616	0.030	0.570	0.978	-0.425	0.424
а	<b>1</b> 0.162	-0.102	-0.358	-0.245	-0.178	-0.148	-0.169	-0.694	0.156	0.332
	OC-GPS					OC-MAP				
F	<sup>2</sup> -0.598	-0.427	-0.668	-0.750	-0.795	-0.710	-0.090	-0.917	-0.592	-0.572
а	<b>0</b> 0.026	0.693	-0.092	0.123	0.130	-0.209	0.240	0.690	-0.295	-0.153
а	<b>1</b> 0.126	-0.750	-0.669	-0.465	-0.392	-0.006	-0.360	-0.923	0.338	-0.034
	RMS	_				Offset				
F	-0.714	0.229	0.173	-0.450	-0.001	-0.097	0.070	-0.400	0.050	0.482
а	0 0.442	-0.117	0.981	0.995	0.871	0.999	0.999	0.952	0.999	0.974
а	<b>1</b> 0.161	0.417	-0.129	-0.456	-0.194	-0.261	-0.053	-0.593	-0.327	0.511
	GPS					MAP				
ос-ма	P 0.722	0.410	0.406	-0.168	0.114	0.502	0.373	0.597	0.546	0.560
Offse	et 0.992	0.081	0.901	0.551	0.681	0.022	0.563	0.983	-0.424	0.400

obliquity, knee flexion and the respective GVSs.

## 4. Discussion

In this study, we evaluated the changes occurring in the gait pattern for subjects with CP that underwent SEMLS, by using three different synthetic index methods: the MAP, the recently proposed LFM (losa et al., 2014) and a novel index i.e. the OC-MAP.

The three methods showed an overall improvement in the gait pattern after surgery: GPS reduced from pre to post; OC-GPS and RMS of offset reduced as well; the overall  $R^2$  of LFM increased.

The improvement in GPS found in this study was  $5.1^{\circ} \pm 4.6^{\circ}$ , comparable to the value found by Rutz et al. in subjects with CP after a similar treatment, which was  $4.3^{\circ} \pm 3.7^{\circ}$  (Rutz et al., 2013). Even though the GPS reduced significantly from pre- to post-treatment, it remained higher than normality, indicating that the walking pattern was still compromised. As pointed out in other studies (Rutz et al., 2013), we observed that patients who had initial high deviations, representing the worst cases, benefitted more from treatment. The surgery had a strong positive impact on the kinematics of the knee. The improvement observed for hip rotation and foot progression suggested that the surgery improved the kinematics in the horizontal plane as well. The worsening observed in the pelvis kinematics could be explained as a consequence of SEMLS that involved a lengthening of hamstring group. In fact, surgical lengthening of hamstrings may increase hip flexion during stance (Delp, Arnold, Speers, & Moore, 1996) and anterior pelvic tilt (Hoffinger, Rab, & Abou-Ghaida, 1993). As no effect was observed on hip flexion, it meant that the improvement at the knee induced

a postural compensation at the level of the pelvis.

The main limitation of the MAP was its inability to identify the cause of deviation from normality and the absence of information about the direction of the deviation, e.g. towards flexion or extension or information about the cause of deviation (offset, shape, etc.). The implementation of OC-MAP allowed to analyse separately the effect of the offset and the deviation from normality of the tracks after considering their Offset. Measuring Offset between observed gait angles and their reference helps in documenting a persistent deviation across the gait cycle. Such deviation may occur every time that, due to some functional impairment, the angular movement of a joint is somehow limited.

Our Offset analysis demonstrated that offset could explain a larger part of the deviation from normality than abnormality in shape, as well as a larger part of the correction after surgery. The deviation observed by MAP for pelvic tilt, knee flexion and ankle dorsiflexion was mainly due to offset. The observed improvements in the offset suggested a normalization of the sagittal plane kinematics, leading to a better posture. The change in the offset of pelvic tilt was not identified by the other methods. The increase in pelvic anterior tilt was a consequence of the surgery on the hamstring group (Hoffinger et al., 1993). The significant pre-post differences observed in OC-MAP and OC-GPS (Fig. 3a) confirmed the overall improvement in gait. Another advantage of computing the Offset profile is the information provided about the sign of the deviation, which in turn represents the biomechanical direction of the deviation. Based on our data, the Offset profile documented a persistent knee flexion in the pre-intervention that reduced, but was still detectable in the post. A persistent plantarflexion in the pre-intervention that decreased towards normality in the post was also documented.

The previous results were compared to another method that allowed the analysis of effects of offset and scaling, i.e. the LFM method (Iosa et al., 2014). It is important to point out that the OC-MAP and LFM are based on different assumptions and different math procedures, meaning that they provide different kind of information. As for GPS and OC-GPS, the overall  $R^2$  was able to detect the overall improvement of gait and the *a0* parameter confirmed that the offset component played an important role in the improvement of knee and ankle kinematics. Namely, the *a0/a1* index was capable of detecting the pattern improvement, in terms of shape/scaling factor, at the knee flexion. Anyway, the *a0* and *a1* parameters were not reliable to describe the offset of pelvic tilt, as it showed a very high standard deviation (Fig. 5b and c). The SD of these indices was relatively high also for other gait features. Thus, these indices are poorly reliable for interpreting gait data and should be used cautiously. This is a known limitation of the LFM method, as *a0* coefficient is reliable only when the correlation between the tracks is relatively high (Iosa et al., 2014).

The strong correlation observed between the  $R^2$ , the GPS and OC-GPS/offset RMS suggested that these indices provide similar synthetic information in the overall analysis. The negative values of the Pearson correlation coefficient are due to the different mathematical assumption underlying the parameters being correlated. In fact, the MAP/OC-MAP parameters are RMS differences between observed angles and reference, while the  $R^2$  comes from a regression analysis (observed angles vs. reference). Thus, when the tracks are similar, MAP parameters becomes small while  $R^2$  is high (~1). On the contrary, when tracks are different, RMS difference grows, while  $R^2$  approaches 0.

The *a*0 and the *Offset* of all gait features were correlated, except for the pelvic tilt. This was attributed to the high variability of *a*0 observed across subjects and to the very low  $R^2$  found for pelvic tilt. The knee flexion also showed a strong correlation between parameters, indicating that all the indices were able to identify the changes in this feature. The strong correlations between the GVSs of pelvic obliquity, knee flexion and the respective offsets (Table 3) confirmed that the gait deviation, detected by MAP in those gait features, was due mainly to offset.

Based on the results here obtained, the OC-MAP and Offset analysis provided meaningful supplemental information about the direction of the deviation, with respect to the other methods. Anyway, the OC-MAP was not able to identify other sources of deviation, e.g. changes in slope or time-shifts, therefore it should be used cautiously when interpreting clinical data. For the subjects studied in this work, the offset was a significant component of deviation in gait pattern, therefore the OC-MAP method was a useful extension to the MAP method to clinically interpret data. Although  $R^2$  can be considered a good overall index of similarity, when it is low, *a0* and *a1* lose their meaning, making LFM less suitable to assess gait features. Therefore the use of LFM is not recommended for interpreting gait of children with CP.

# 5. Conclusion

The OC-MAP method overcame a MAP limitation, by separating the offset component from the differences in the shape of the joint kinematics. As the offset was a significant component of deviation in gait pattern, the OC-MAP demonstrated being the most clinically meaningful synthetic method to interpret gait data in CP. Further study of the OC-MAP is necessary on larger cohorts of patients with CP, and also on cohorts of patients with different pathologies, in order to prove its clinical usefulness.

#### Acknowledgments

This research was supported by the Seventh Framework Program FP7 (MD-Paedigree, FP7-ICT-2011-9).

The authors would like to thank the clinical staff of the Dept. of Rehabilitation Medicine, VUmc, Amsterdam, for the use of their lab and for the support provided in data acquisition.**Conflict of interests** 

The authors declare to have no conflict of interests.

#### References

- Ancillao, A., Rossi, S., & Cappa, P. (2017). Analysis of knee strength measurements performed by a hand-held multicomponent dynamometer and optoelectronic system. IEEE Transactions on Instrumentation and Measurement, 66(1), 85–92. http://dx.doi.org/10.1109/TIM.2016.2620799.
- Baker, R., McGinley, J. L., Schwartz, M. H., Beynon, S., Rozumalski, A., Graham, H. K., & Tirosh, O. (2009). The gait profile score and movement analysis profile. *Gait & Posture*, 30(3), 265–269. http://dx.doi.org/10.1016/j.gaitpost.2009.05.020.
- Baker, R., McGinley, J. L., Schwartz, M., Thomason, P., Rodda, J., & Graham, H. K. (2012). The minimal clinically important difference for the Gait Profile Score. *Gait & Posture*, 35(4), 612–615. http://dx.doi.org/10.1016/j.gaitpost.2011.12.008.
- Beynon, S., McGinley, J. L., Dobson, F., & Baker, R. (2010). Correlations of the Gait Profile Score and the Movement Analysis Profile relative to clinical judgments. Gait & Posture, 32(1), 129–132. http://dx.doi.org/10.1016/j.gaitpost.2010.01.010.
- Camerota, F., Galli, M., Cimolin, V., Celletti, C., Ancillao, A., Blow, D., & Albertini, G. (2015). The effects of neuromuscular taping on gait walking strategy in a patient with joint hypermobility syndrome/Ehlers-Danlos syndrome hypermobility type. *Therapeutic Advances in Musculoskeletal Disease*, 7(1), 3–10. http://dx.doi.org/10. 1177/1759720X14564561.
- Cappozzo, A., Catani, F., Della Croce, U., & Leardini, A. (1995). Position and orientation in space of bones during movement: Anatomical frame definition and determination. *Clinical Biomechanics*, 10(4), 171–178. http://dx.doi.org/10.1016/0268-0033(95)91394-T.
- Carriero, A., Zavatsky, A., Stebbins, J., Theologis, T., & Shefelbine, S. J. (2009). Determination of gait patterns in children with spastic diplegic cerebral palsy using principal components. Gait & Posture, 29(1), 71–75. http://dx.doi.org/10.1016/j.gaitpost.2008.06.011.
- Celletti, C., Galli, M., Cimolin, V., Castori, M., Tenore, N., Albertini, G., & Camerota, F. (2013). Use of the Gait Profile Score for the evaluation of patients with joint hypermobility syndrome/Ehlers–Danlos syndrome hypermobility type. *Research in Developmental Disabilities*, 34(11), 4280–4285. http://dx.doi.org/10.1016/j. ridd.2013.09.019.
- Cimolin, V., Galli, M., Vimercati, S. L., & Albertini, G. (2011). Use of the Gait Deviation Index for the assessment of gastrocnemius fascia lengthening in children with Cerebral Palsy. Research in Developmental Disabilities, 32, 377–381. http://dx.doi.org/10.1016/j.ridd.2010.10.017.
- Delp, S. L., Arnold, A. S., Speers, R. A., & Moore, C. A. (1996). Hamstrings and psoas lengths during normal and crouch gait: Implications for muscle-tendon surgery. Journal of Orthopaedic Research, 14(1), 144–151. http://dx.doi.org/10.1002/jor.1100140123.
- Esbjörnsson, A., Rozumalski, A., Schwartz, M., Wretenberg, P., Iversen, M. D., & Broström, E. W. (2014). Quantifying gait deviations in patients with rheumatoid arthritis using the gait deviation index. Gait & Posture, 36, S53. http://dx.doi.org/10.1016/j.gaitpost.2011.10.269.
- Galli, M., Cimolin, V., Crivellini, M., & Albertini, G. (2009). Long-term evaluation of isolated gastroenemius fascia lengthening in children with cerebral palsy using gait analysis. Journal of Pediatric Orthopaedics B, 18(5), 228-233. http://dx.doi.org/10.1097/BPB.0b013e32832bf65c.
- Galli, M., Cimolin, V., De Pandis, M. F., Schwartz, M. H., & Albertini, G. (2012). Use of the Gait deviation index for the evaluation of patients with Parkinson's disease. Journal of Motor Behavior, 44(3), 161–167. http://dx.doi.org/10.1080/00222895.2012.664180.
- Galli, M., Cimolin, V., Rigoldi, C., Tenore, N., & Albertini, G. (2010). Gait patterns in hemiplegic children with Cerebral Palsy: Comparison of right and left hemiplegia. *Research in Developmental Disabilities*, 31(6), 1340–1345. http://dx.doi.org/10.1016/j.ridd.2010.07.007.
- Galli, M., Rigoldi, C., Brunner, R., Virji-Babul, N., & Giorgio, A. (2008). Joint stiffness and gait pattern evaluation in children with Down syndrome. *Gait & Posture, 28*, 502–506. http://dx.doi.org/10.1016/j.gaitpost.2008.03.001.
- Hoffinger, S. A., Rab, G. T., & Abou-Ghaida, H. (1993). Hamstrings in cerebral palsy crouch gait. Journal of Pediatric Orthopedics, 13(6), 722-726.
- Iosa, M., Cereatti, A., Merlo, A., Campanini, I., Paolucci, S., & Cappozzo, A. (2014). Assessment of waveform similarity in clinical Gait data: The linear fit method. BioMed Research International, 1–7. http://dx.doi.org/10.1155/2014/214156.
- Massaad, A., Assi, A., Skalli, W., & Ghanem, I. (2014). Repeatability and validation of Gait Deviation Index in children: Typically developing and cerebral palsy. Gait & Posture, 39(1), 354–358. http://dx.doi.org/10.1016/j.gaitpost.2013.08.001.
- Molloy, M., McDowell, B. C., Kerr, C., & Cosgrove, A. P. (2010). Further evidence of validity of the Gait Deviation Index. Gait & Posture, 31(4), 479–482. http://dx.doi. org/10.1016/j.gaitpost.2010.01.025.
- Pau, M., Coghe, G., Atzeni, C., Corona, F., Pilloni, G., Marrosu, M. G., ... Galli, M. (2014). Novel characterization of gait impairments in people with multiple sclerosis by means of the gait profile score. Journal of the Neurological Sciences, 345(1–2), 159–163. http://dx.doi.org/10.1016/j.jns.2014.07.032.
- Piccinini, L., Cimolin, V., D'Angelo, M. G., Turconi, A. C., Crivellini, M., & Galli, M. (2011). 3D gait analysis in patients with hereditary spastic paraparesis and spastic diplegia: A kinematic, kinetic and EMG comparison. European Journal of Paediatric Neurology, 15(2), 138–145. http://dx.doi.org/10.1016/j.ejpn.2010.07.009.
  Rutz, E., Donath, S., Tirosh, O., Graham, H. K., & Baker, R. (2013). Explaining the variability improvements in gait quality as a result of single event multi-level surgery
- in cerebral palsy. *Gait and Posture*, 38(3), 455-460. http://dx.doi.org/10.1016/j.gaitpost.2013.01.014. Sale, P., De Pandis, M. F., Stocchi, F., Domenica, L. P., Sova, I., Cimolin, V., ... Franceschini, M. (2013). Robot-assisted walking training for individuals with Parkinson's
- disease: A pilot randomized controlled trial. *BMC Neurology*, 13(1), 50. http://dx.doi.org/10.1186/1471-2377-13-50.
- Schwartz, M. H., & Rozumalski, A. (2008). The gait deviation index: A new comprehensive index of gait pathology. Gait and Posture, 28(3), 351–357. http://dx.doi. org/10.1016/j.gaitpost.2008.05.001.
- Schweizer, K., Romkes, J., Coslovsky, M., & Brunner, R. (2014). The influence of muscle strength on the gait profile score (GPS) across different patients. Gait & Posture, 39(1), 80–85. http://dx.doi.org/10.1016/j.gaitpost.2013.06.001.
- Speciali, D. S., Oliveira, E. M., Cardoso, J. R., Correa, J. C. F., Baker, R., & Lucareli, P. R. G. (2014). Gait profile score and movement analysis profile in patients with Parkinson's disease during concurrent cognitive load. Brazilian Journal of Physical Therapy, 18(4), 315–322. http://dx.doi.org/10.1590/bjpt-rbf.2014.0049.
- Stebbins, J., Trinler, U. K., Baker, R., Brunner, R., Wren, T., & Theologis, T. (2014). Recommendations for reporting gait studies. Gait & Posture, 10–11. http://dx.doi.org/10.1016/j.gaitpost.2014.11.014.
- Sutherland, D. H., & Davids, J. R. (1993). Common gait abnormalities of the knee in cerebral palsy. *Clinical Orthopaedics and Related Research, NA, 288,* 139–147. http://dx.doi.org/10.1097/00003086-199303000-00018.
- van den Noort, J., Ferrari, A., Cutti, A., Becher, J., & Harlaar, J. (2013). Gait analysis in children with cerebral palsy via inertial and magnetic sensors. *Medical & Biological Engineering & Computing*, 51(4), 377–386. http://dx.doi.org/10.1007/s11517-012-1006-5.
- Vismara, L., Cimolin, V., Galli, M., Grugni, G., Ancillao, A., & Capodaglio, P. (2016). Osteopathic Manipulative Treatment improves gait pattern and posture in adult patients with Prader-Willi syndrome. International Journal of Osteopathic Medicine, 19, 35–43. http://dx.doi.org/10.1016/j.ijosm.2015.09.001.
- Whittle, M. (1996). Clinical gait analysis: A review. Human Movement Science, 15(3), 369-387. http://dx.doi.org/10.1016/0167-9457(96)00006-1.