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Full Length Article

The Human Body Model versus conventional gait models for kinematic gait analysis in children with cerebral palsy



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ABSTRACT

With the rise of biofeedback in gait training in cerebral palsy there is a need for real-time measurements of gait kinematics. The Human Body Model (HBM) is a recently developed model, optimized for the real-time computing of kinematics. This study evaluated differences between HBM and two commonly used models for clinical gait analysis: the Newington Model, also known as Plug-in-Gait (PiG), and the calibrated anatomical system technique (CAST). Twenty-five children with cerebral palsy participated. 3D instrumented gait analyses were performed in three laboratories across Europe, using a comprehensive retroreflective marker set comprising three models: HBM, PiG and CAST. Gait kinematics from the three models were compared using statistical parametric mapping, and RMSE values were used to quantify differences. The minimal clinically significant difference was set at 5°. Sagittal plane differences were mostly less than 5°. For frontal and transverse planes, differences between all three models for almost all segment and joint angles exceeded the value of minimal clinical significance. Which model holds the most accurate information remains undecided since none of the three models represents a ground truth. Meanwhile, it can be concluded that all three models are equivalent in representing sagittal plane gait kinematics in clinical gait analysis.

1. Introduction

Cerebral palsy (CP) is a common motor disorder affecting 2 in 1000 births in Europe (Johnson, 2002), often leading to an aberrant gait pattern (Davids, Rowan, & Davis, 2007). The gait pattern in CP is often analyzed using three-dimensional gait kinematics to assist clinical decision making and to evaluate treatment outcomes. Furthermore, gait kinematics can be used as parameters in real-time gait specific biofeedback (Booth, Steenbrink, Buizer, Harlaar, & van der Krogt, 2016; Booth, Steenbrink, Buizer, Harlaar, & Van der Krogt, 2017; van Gelder et al., 2017). Such biofeedback can be used for functional gait training. A recent literature review has shown

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that functional gait training outperforms standard physical therapy on several parameters (Booth et al., 2018) including walking speed, walking endurance and gait-related gross motor function. The study further addressed that virtual reality and biofeedback seem promising tools to improve engagement and rehabilitation outcomes in children with CP (Booth et al., 2018). This has also been found for other patient populations (Giggins et al., 2013; Pfeufer et al., 2018). Tate and Milner (2010) specifically reviewed biofeedback on kinematics and found this an effective method to improve functional outcomes like gait speed and symmetry, at least on the short-term. With the rise of biofeedback training, the need for accurate real-time measurements of kinematics is rising.

Multiple models exist which can be used to measure kinematic gait features, including Plug-in-Gait (PiG)(Davis, Ounpuu, Tyburski, & Gage, 1991), the calibrated anatomical system technique (CAST)(Cappozzo, Catani, Della Croce, & Leardini, 1995) and the Gait2392 model as implemented in OpenSim (Delp et al., 2007). The human body model (HBM) is a recently developed model optimized for computing real-time kinematics (van den Bogert, Geijtenbeek, Even-Zohar, Steenbrink, & Hardin, 2013). Within HBM, several features are implemented to enhance the possibility to calculate kinematics in real-time. Technical markers are added to an anatomical marker set to provide redundancy of markers and thereby robustness against marker dropouts. Global optimization is used to further minimize the effects of marker dropout, as well as to limit effects of soft tissue artefacts (Duprey, Cheze, & Dumas, 2010). The global optimization used in HBM is based on a weighted least squares problem to model marker positions: the distance between measured and modeled markers is minimized for the entire model at once, as is further described by Falisse et al. (2018).

So far, only a few studies have assessed the outcomes of HBM. van den Bogert et al. (2013) first published healthy adult gait kinematics computed using an early version of HBM. Modifications have been made since to better match patient characteristics. The main modifications being the inclusion of anatomically defined knee and ankle axes, based on knee epicondyles and ankle malleoli respectively, an improved regression model to calculate hip joint centers (Harrington, Zavatsky, Lawson, Yuan, & Theologis, 2007), and changes in shank and thorax definitions to better match ISB standards (Wu et al., 2002). Falisse et al. (2018) compared the modified HBM with the OpenSim gait2392 model, which is often used for musculoskeletal modeling purposes. Although several assumptions are similar between both models, kinematic outcomes deviated substantially (Falisse et al., 2018). These deviations could largely be attributed to differences in pelvic orientation and hip and knee joint center estimation methods.

Thanks to its real-time performance, HBM is very well suited for gait training purposes, but it can also be used for other purposes such as clinical decision making and treatment evaluation. HBM complies with several recent insights, such as the Harrington hip joint center equation (Kainz et al., 2016) and global optimization (Duprey et al., 2010) and is therefore expected to present accurate results for gait analysis. Obviously, it is beneficial to use the same model in gait analysis as in gait training, to avoid possible confusion due to differences between models. For clinical implementation of HBM, it is important to see how outcomes in terms of gait kinematics compare to marker models currently used in clinical care. Constraints (i.e. a reduction in the six degrees of freedom of a segment) can be a cause of differences between models. A commonly used constraint includes linking two segments together (i.e. limiting translation between segments), thereby constraining the joint to three degrees of freedom. Another common constraint is analyzing the knee as a hinge joint, thereby reducing joint movements to one degree of freedom. Constraints can decrease the effect of soft tissue artefacts and marker placement errors, hence improve reliability (Duprey et al., 2010; Groen, Geurts, Nienhuis, & Duysens, 2012). However, modeled joint constraints should match the anatomy of the patient of interest, and are typically based on nonpathological anatomy. Children with CP often experience bony and joint deformities, which might violate these assumptions. Therefore, it is important to evaluate kinematic models for the specific patient group of interest. While previous comparisons made with HBM are performed using healthy adults, comparison in children with CP adds more to the insight of the differences in eventual care. Therefore, this study aimed to compare gait kinematics from HBM in children with CP with those from two commonly used marker models for clinical gait analysis; PiG and CAST.

2. Method

2.1. Subjects

Twenty-five children with CP participated in this study. Participants were recruited by their doctors from the VU University medical center Amsterdam (VUmc, N = 10), Children's Hospital Bambino Gesù (OPBG, N = 6) and the University of Leuven (KUL, N = 9). No significant differences existed in patient characteristics between participant groups from each measurement site (Table 1).

Mean values and standard deviations for patient characteristics separately for each measurement site, including statistical outcomes.

	VUmc	OPBG	KUL	P-values ANOVA	
GMFCS level	6 x II, 3 x I	6 x I	6 x II, 2 x I		
Age	$11.4y \pm 2.1$	$11.0y \pm 2.8$	$9.2y \pm 2.6$	0.410	
Height	1.5 m ± 0.1	$1.4 \text{ m} \pm 0.1$	$1.3 \text{ m} \pm 0.1$	0.138	
Weight	$41.1 \text{ kg } \pm 9.2$	$34.2 \text{ kg} \pm 8.3$	$31.3 \text{ kg} \pm 9.0$	0.266	
Leg length	74.6 cm ± 9.2	$76.8 \text{ cm} \pm 9.5$	$69.0 \text{ cm} \pm 7.8$	0.269	
Foot length	$22.7 \text{ cm} \pm 2.2$	$23.3 \text{ cm} \pm 3.8$	$20.0 \text{ cm} \pm 2.6$	0.437	
Foot off	$65.4\% \pm 1.5$	$66.1\% \pm 2.0$	$65.1\% \pm 1.9$	0.630	
Walking speed	$1.1 \text{ m/s} \pm 0.3$	$0.9 \text{ m/s} \pm 0.2$	$1.0 \text{ m/s} \pm 0.1$	0.196	
Stride length	$1.1 \text{ m} \pm 0.2$	$0.9 \text{ m} \pm 0.2$	$1.0 \text{ m} \pm 0.2$	0.143	

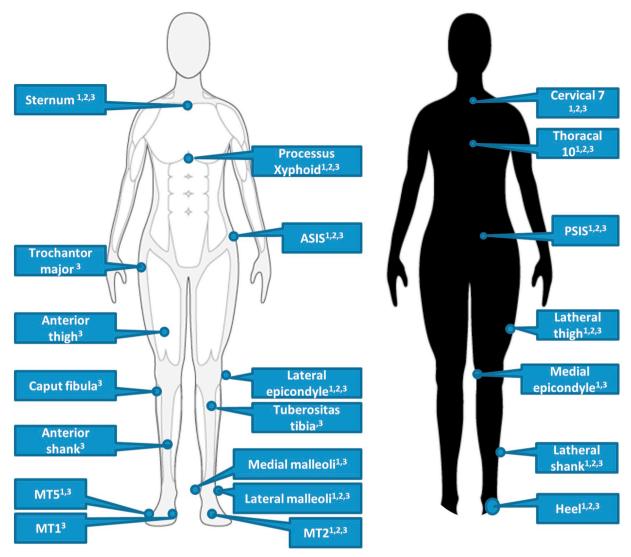


Fig. 1. Overview of used marker set and the location of the markers. Numbers indicate which models include this marker, ¹ being HBM, ² PiG and ³ CAST. MT = Metatarsal phalangeal joint, ASIS = anterior superior iliac spine, PSIS = posterior superior iliac spine.

Inclusion criteria were: diagnosed with spastic CP, GMFCS level I-II, aged between eight and fifteen years, no orthopedic lower limb surgery or selective dorsal rhizotomy in the last year, nor chemo-denervation in the last six months prior to the assessment, and the ability to follow simple instructions. Participants aged twelve and older and all parents provided written informed consent before participation. The study was approved by each of the three local ethics committees.

2.2. Protocol

Forty-two retroreflective markers were placed on each subject, covering HBM, PiG and CAST (Fig. 1, Appendix A). A medial ankle marker was added in PiG to account for tibial torsion as often present in CP (Kerr Graham & Selber, 2003). This is further addressed in the Appendix. As inter-observer reliability increases by marker placement training (Gorton, Hebert, & Gannotti, 2009), all examiners were trained by internal experts and involved examiners came together several times to standardize protocols. At each measurement site, two or three experienced gait analysts were in charge of applying markers and performing the measurements.

After marker placement, a static trial for subject calibration was captured with participants standing upright. At VUmc, participants walked on an instrumented treadmill at a self-paced (Sloot, van der Krogt, & Harlaar, 2014), comfortable walking speed wearing thin flexible gym shoes. One participant was not capable of performing self-paced walking, therefore a self-selected fixed speed was used. All children got several minutes to familiarize to treadmill walking. Participants at KUL and OPBG walked barefoot at comfortable walking speed on a ten meter walkway. At least three gait cycles were collected for each participant and three cycles per subject were used for analysis. Differences in kinematics between over-ground and self-paced treadmill walking are generally small

for children with CP (van der Krogt, Sloot, & Harlaar, 2014) and therefore neglected in this study. Although footwear might cause different marker movement artefacts, the effect on differences between models is thought to be limited since similar foot markers were used, with the exception of the MT1/2/5 markers. Marker capture was performed using MX camera's (Vicon Motion Systems, Oxford, UK) at all sites. Marker trajectories reconstructed from raw marker data (Vicon Nexus, version 2.3, Oxford, UK) were used for data analysis.

2.3. Data analysis

For each participant, kinematics for hip, knee and ankle joint, as well as trunk, pelvis and foot segments were calculated using three models. First, HBM was used, as implemented in the Gait Offline Analysis Tool (GOAT; version 3.3; Motek BV, Amsterdam, The Netherlands). Multiple joint constraints and global optimization are used in HBM. Second, PiG was used as implemented in Vicon Nexus (version 2.3). PiG is based on the Newington model, using a minimum marker set mostly placed on bony landmarks. Constraints in PiG are implicit by shared markers and joint centers between adjacent segments, hence restricting segment movements to three degrees of freedom. Third, CAST was used, as implemented in custom-made software package BodyMech (www.bodymech. nl, Matlab 2014a, The Mathworks Inc., USA), based on clusters and virtual markers without constraints. Further biomechanical details of the three models are presented in the Appendix. Outcomes from each model were exported to Matlab for further analyses

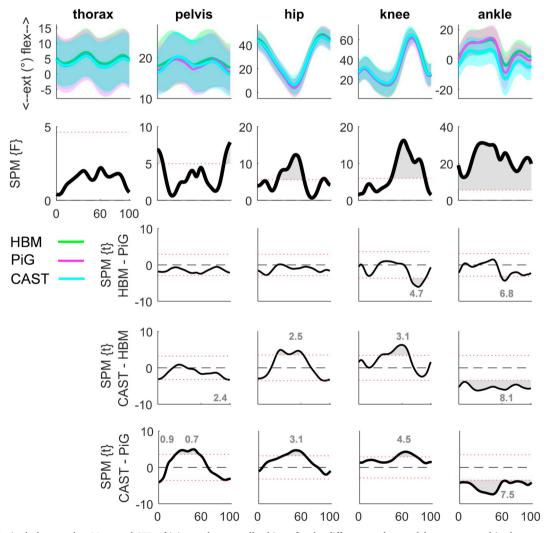


Fig. 2. Sagittal plane angles. Mean and STD of joint angles over all subjects for the different marker models are presented in the top pane. SPM repeated-measures ANOVA F-values are depicted on the second pane. SPM post-hoc paired *t*-test t-values are represented on the bottom three panes. Red dotted lines indicate F/t-threshold values above which curves significantly differ. Grey shaded areas indicate significant differences, with bigger shaded areas indicating lower *p*-values. For each significant region in the *t*-tests, RMSE values are depicted in these graphs in black to quantify the size of a significant difference. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

and statistics.

All joint and segment kinematics were filtered with a bi-directional 6 Hz 2nd order low-pass Butterworth filter, to harmonize bandwidth between methods. Gait cycles were defined from initial contact to next initial contact, with initial contact defined using the method of Zeni, Richards, and Higginson (2008) and gait cycles time-normalized to 0–100%. Three strides of the left leg were analyzed per subject. Next, eighteen clinically relevant outcome parameters, further referred to as kinematic parameters, were calculated (Table 2). The kinematic parameters included parameters as used previously (Schutte et al., 2000), as well as three parameters from frontal and transverse plane following Klejman, Andrysek, Dupuis, and Wright (2010) and two parameters used in kinematic feedback training (Tate & Milner, 2010).

2.4. Statistics

Differences between models were assessed using Statistical Parametric Mapping (SPM; version M.0.4.5) (Friston, Mattout, Trujillo-Barreto, Ashburner, & Penny, 2007). Differences between the two conventional models were also considered relevant, to assess whether differences of HBM with conventional models were similar to differences between conventional models. SPM was chosen as this method allows to detect specific differences in any part of the curve, reducing the necessity of a priori hypotheses. An SPM repeated measures analysis of variance (RM-ANOVA) was conducted over all kinematics with the three models as grouping variable, with post-hoc SPM paired *t*-tests. The Holm procedure (Holm, 1979) was used for all post-hoc tests to maintain probability of a type 2 error at 5%. Root mean square errors (RMSEs) were calculated as an effect size for all significantly different parts of the curves.

Kinematic parameters were tested for normality using Shapiro-Wilk tests and compared between models using RM-ANOVA. For

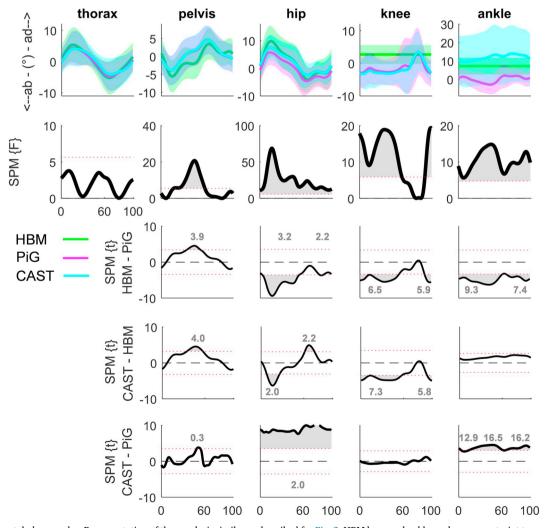


Fig. 3. Frontal plane angles. Representation of the graphs is similar as described for Fig. 2. HBM knee and ankle angles are constraint to zero degrees movement in the frontal plane. The horizontal lines represent the static values for ab —/adduction.

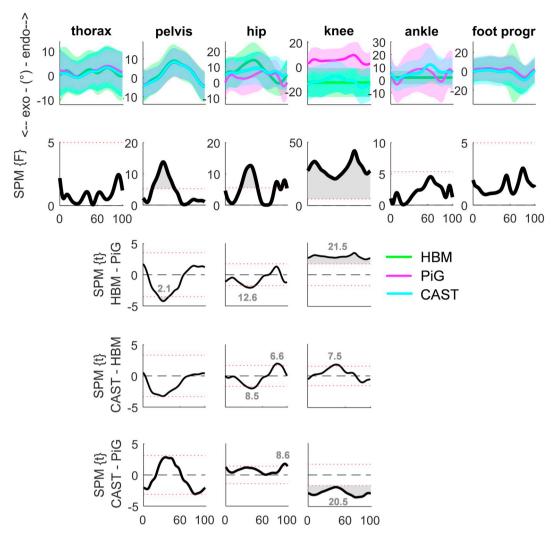


Fig. 4. Transversal plane angles. Representation of the graphs is similar as described for Fig. 2. HBM knee and ankle angles are constraint to zero degrees movement in the transversal plane. The horizontal lines represent the static values for internal/external rotation.

post-hoc analysis, paired-samples t-test with Holm-correction were used. Furthermore, to quantify overall differences between models, RMSE values between the models were calculated over joint kinematic curves for each subject and averaged over all subjects. Additionally, differences in mean kinematics, representing offsets between signals, were calculated to evaluate systematic differences between models. Offset-corrected RMSEs were also calculated to assess differences attributed to the shape of the kinematic curves (Ancillao et al., 2017). The measurement error associated with collecting 3D gait kinematics using the same model, e.g. inter-session and inter-therapist differences in kinematic outcomes, were generally below 5° in previous studies (Ferber, McClay Davis, Williams III, & Laughton, 2002; Ferrari et al., 2008; McGinley, Baker, Wolfe, & Morris, 2009). Systematic differences between models are especially of interest when they exceed those intra-model differences. McGinley et al. (2009) recommended that differences above 5° were considered clinically meaningful. Furthermore, improvements in kinematic parameters after feedback usually exceeded 5° (Booth, Buizer, Harlaar, Steenbrink, & van der Krogt, 2019; Tate & Milner, 2010). Therefore, we considered the cut off of 5° to indicate relevant differences between models.

3. Results

Kinematic curves for the three models and outcomes from SPM can be found in Figs. 2, 3 and 4. Kinematic parameters are shown in Table 2 and RMSE values over the complete strides in Fig. 5.

For thorax, pelvis and hip kinematics, differences between models were not significant or stayed below 5° in all planes, except for transverse plane hip rotation. SPM showed significant differences in hip exorotation, reflected also by hip peak exorotation, showing 5.3° less exorotation for CAST compared with PiG. PiG showed the most amount of exorotation, followed by HBM. RMSE values over complete strides were 11.8° (PiG-HBM), 9.1° (PiG-CAST) and 7.4° (HBM-CAST), with an offset-corrected RMSE of 6.5° between PiG

Table 2

Mean values and standard deviations for clinically relevant outcome parameters from the three models and the three laboratories, including statistical outcomes.

	Mean (°) ± STD			P-values ANOVA & post-hoc			
	PiG	CAST	HBM	ANOVA	HBM-CAST	HBM-PiG	CAST-PiG
Mean pelvic tilt	18.1 ± 6.0	18.1 ± 6.0	18.7 ± 6.1	0.126			
ROM ¹ pelvic tilt	6.1 ± 2.9	6.1 ± 3.0	6.4 ± 3.1	0.699			
Mean pelvic rotation	2.5 ± 6.6	2.5 ± 6.6	2.9 ± 7.5	0.100			
ROM hip flexion	45.8 ± 8.6	42.4 ± 8.6	46.4 ± 9.0	< 0.001	< 0.001	0.214	< 0.001
Minimal hip flexion	2.5 ± 7.8	5.1 ± 7.7	3.0 ± 7.0	< 0.001	< 0.001	0.434	< 0.001
Peak hip abduction swing	-6.6 ± 3.9	-4.5 ± 3.5	-5.9 ± 3.6	< 0.001	< 0.001	0.047	< 0.001
Peak hip exorotation	-4.2 ± 10.2	1.1 ± 9.7	-2.3 ± 8.4	0.021	0.020	0.369	0.015
Peak hip endorotation	11.7 ± 9.9	12.7 ± 9.9	16.3 ± 11.1	0.068			
ROM knee flexion	51.2 ± 12.0	53.4 ± 13.2	54.2 ± 12.6	< 0.001	0.224	< 0.001	< 0.001
Peak knee flexion swing	63.8 ± 6.8	66.8 ± 7.0	66.2 ± 6.0	< 0.001	0.418	< 0.001	0.003
Peak knee extension stance	13.3 ± 12.1	14.1 ± 12.5	12.4 ± 12.6	0.052			
Knee flexion at IC ²	25.7 ± 11.1	26.8 ± 11.0	26.0 ± 11.8	0.247			
Time to peak knee flexion	$74.5^3 \pm 4.2$	$74.2^3 \pm 4.2$	$75.2^3 \pm 4.6$	0.002	0.001	0.035^4	0.298
ROM knee adduction	13.6 ± 3.8	12.1 ± 4.8	0 ± 0^{5}	< 0.001	< 0.001 ⁵	< 0.001 ⁵	0.081
Peak ankle dorsal stance	16.3 ± 7.9	9.0 ± 8.5	14.6 ± 8.7	< 0.001	< 0.001	0.078	< 0.001
Peak ankle dorsal swing	4.6 ± 7.4	-0.1 ± 8.5	6.4 ± 7.3	< 0.001	< 0.001	0.048	0.001
Peak plantar push-off	10.7 ± 10.5	14.4 ± 10.3	5.9 ± 9.0	< 0.001	< 0.001	< 0.001	0.005
Mean foot progression	0.6 ± 13.0	-1.8 ± 15.5	-1.3 ± 15.8	0.204			

Note. Significant values are expressed in bold and differences exceeding 5° clinical significances are underlined. ¹ROM = range of motion. ²IC = initial contact. ³Percentage of gait cycle. ⁴This value is not significant due to Holm-corrections. ⁵ROM is constraint to 0° in HBM; therefore the other two models differ significantly by default.

and HBM.

Knee sagittal kinematics were comparable between models, i.e. differences were below 5°. Since HBM knee angles are constraint to sagittal plane movements only, differences for the frontal and transversal plane were expected. In the frontal plane, knee adduction ROM of PiG and CAST were 13.6° and 12.1°, in contrast to the 0° assumption in HBM. In addition, all frontal and transverse knee RMSE values between the three models exceeded 5°, but offset-corrected RMSE values and offsets for knee adduction remained below 5°. Knee exorotation was over 15° less according to PiG than CAST and HBM over the entire gait cycle and offset-corrected RMSE values were around 5.

Ankle joint kinematics differed significantly between models in sagittal and frontal plane over almost the entire gait cycle, as also reflected in the ankle kinematic parameters showing over 7.3° and 5.6 significantly less dorsiflexion during swing and 5.0° and 6.5° during stance for CAST than for PiG and HBM respectively. Additionally, peak plantarflexion during push-off was 8.5° larger for CAST than for HBM. Furthermore, all RMSE values for the ankle exceeded 5°, with offset values above 5° for comparisons of HBM with CAST in the sagittal plane and with PiG in the frontal plane. None of the offset-corrected values exceeded the 5° threshold.

4. Discussion

This study aimed to assess differences between HBM, a recent marker model using joint constraints, a redundant number of markers and global optimization, versus two commonly used models in gait analysis: i.e. PiG, using a minimum marker set implying some implicit constraints, and CAST, based on cluster markers, calibrated with bony landmarks and no joint constraints, in children with cerebral palsy. Sagittal plane kinematics were found to be quite comparable for the three models, with differences generally below 5°. However, differences of up to 25° were found in the other planes for the hip, knee and ankle joints, between all three models.

HBM provided equivalent outcomes for sagittal plane kinematics, with only ankle kinematic differences with CAST exceeding 5° RMSE. Differences between CAST and HBM/PiG were visible as an offset towards plantar flexion, as was also found in a previous study by Ferrari et al. (2008) between CAST and PiG. This is mainly caused by different markers that define the foot segment (see Appendix A) MT1 and MT5 are used in CAST, which are placed lower on the foot than the MT2 marker used in HBM and PiG. This causes a difference in the line between the heel marker and the MT2 versus the heel marker and the MT1/MT5 plane of approximately 6.0°. This angular difference can be corrected by replacing MT1 and MT5 markers in CAST with the MT2 marker used in the other models. A limitation of the study is that subjects walked on a treadmill in the VUmc and over ground in OPBG and KUL. The effect of treadmill walking versus over ground walking on the different models is not addressed in this study, although the effect is thought to be small based on previous studies. Furthermore, children wore gymnastic shoes on the treadmill and not over ground, which may have affected the foot kinematics and the comparison between models. However, differences between ankle kinematics showed similar results for all three labs, and therefore treadmill walking and shoe movement artefacts are not thought to influence the findings. Overall, our findings suggest that sagittal plane kinematics are generally similar between models.

In contrast, important differences between all three models were found in the frontal and transverse plane. The differences found in this study in children with CP are comparable to differences previously found in healthy adults. For instance, Ferrari et al. (2008)

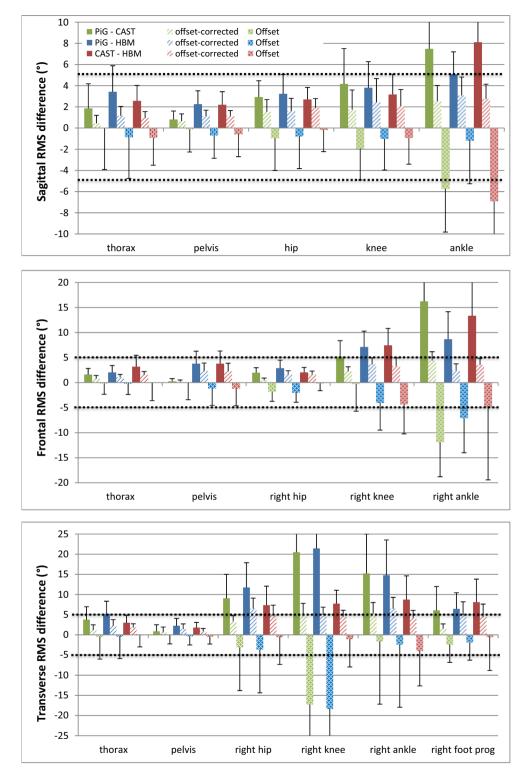


Fig. 5. Differences between the three models, measured as root mean square error (RMSE), offset-corrected RMSE and offset values. Offset values are presented as negative values for visualization purposes. Dotted lines represent the 5° threshold for clinical decision making. Fig. 5a presents sagittal plane values, b presents frontal plane angles and c presents transversal plane angles.

found very low correlation coefficients between PiG and CAST for hip and knee endo-/exorotation. The large differences in frontal and transverse planes are likely due to the differences in constraints applied by all models and partially due to different markers used. In HBM, the knee is modeled as a one degree of freedom hinge joint and the ankle is constrained to two degrees of freedom. Generally constraints will reduce the influence of soft tissue artefacts and marker placement errors (Duprey et al., 2010; Groen et al., 2012). On the other hand, actual rotations in the constrained angles will affect the modeled rotations in other planes and other joints, introducing kinematic errors. True knee or ankle exorotation will for example be measured as hip exorotation, which explains the larger hip exorotation ROM for HBM compared to CAST and PiG. Contrarily, average knee adduction ROM as found by CAST and PiG exceeded 10°, which exceeds the maximum for healthy knees (Lafortune, Cavanagh, Sommer, & Kalenak, 1992). This result is likely due to crosstalk from knee flexion as a consequence of malaligned coordinate system through misplacement of thigh markers (Lafortune et al., 1992). This is supported by findings of Sangeux (2018), who showed that PiG ab/adduction angles can be reduced when correcting the alignment by assuming only one or two degree of freedom movements in the knee. Such a misalignment error leads in turn to errors in calculated hip rotation angles (Sangeux, 2018). Hence it is likely that all models show errors in hip exorotation angles to some extent, albeit of different origin.

Differences in hip kinematics can furthermore be caused by different methods to define the hip joint center. HBM uses the Harrington equation, suggested to be the most accurate regression method according to a recent validation study (Kainz et al. 2016, i.s.o. 2017), in contrast with the Davis equation used in PiG. Leboeuf, Reay, Jones, and Sangeux (2019) showed that supplementing Davis equation with the Harrington equation improved PiG kinematics in the frontal plane, although not in other planes. Kainz et al. (2016), (i.s.o. 2017) advised the use of functional calibration over a regression method. However, experience during performing our measurements showed that most children with CP are not able to perform the star-arc movement with the required range of motion for functional calibration. Therefore, we did not use functional hip calibration in this study. An important note for HBM is that, in absence of medial knee and ankle markers, knee and ankle axes are defined parallel to the line between the ASIS markers in static calibration. Due to femoral and tibial torsion, this is unlikely to be valid for many children with CP and would probably result in errors in hip kinematics. It is clear from PiG outcomes that tibial torsion occurs in the patients in this study: Tibial torsion is accounted for in PiG in ankle angles, but not in knee angles, due to the use of a torsioned tibia. In HBM and CAST, torsions in the tibia are also reflected as an offset in knee exorotation. This results in large differences between PiG knee exorotation and HBM and CAST knee exorotation. Furthermore, the influence of tibial torsion on kinematic outcomes is presented in the Appendix. Due to the large influence of tibial torsion, it is recommended to apply medial markers when using HBM during static calibration when analyzing pathological gait.

Some differences between models may also be due to the use of global optimization, as applied in HBM, versus segment tracking, as used in PiG and CAST. Pelvic kinematics in HBM differed significantly from PiG and CAST, despite identical markers and anatomical frame definitions. Global optimization of the markers used in HBM distributes errors due to marker misplacement and soft tissue artefacts over all segments, but possibly also reduces soft tissue artefacts of the pelvic markers. This is reflected in the differences between models: small differences between HBM and the other two models occur in almost all joints. Despite these differences, overall RMSE differences between HBM and the two conventional models are smaller in magnitude than differences between the conventional models. This suggests that errors are distributed over multiple segments, thereby minimizing overall errors.

The measured differences give guidelines for use in clinical implementation. For all three models, differences exceeded 5°, implying that substituting a marker model with another model introduces a bias that might not be accounted for in clinical decision making. These differences can present themselves as offsets between models, such as in ankle plantarflexion. Such a systematic error is relatively easy to account for. The largest difference in the sagittal plane was present as such an offset. Since those can be corrected for, it can be concluded that kinematic curves in the sagittal plane from different model can be compared for interpretation. This implies that HBM and the conventional models are functionally equivalent for sagittal plane kinematics. However, in general, given that small differences still exist between models, it remains important to collect data from the same subject before and after treatment using the same model. The same holds for a normative dataset used for comparison, which should be based on the same model. Lastly, sagittal plane kinetics can also be input for biofeedback (e.g. Booth et al., 2019), hence validity of kinetics should be assessed before implementation.

Besides offsets, differences between the kinematic patterns are harder to correct for. Such differences in patterns were seen for the frontal and transverse plane kinematics, so for these planes the three models are not equivalent to each other. Constraints in knee and ankle angles in HBM limit the use of HBM in feedback for these angles. However, conventional models are probably also not suitable for providing accurate feedback on these angles, taking into account the large ROM found for knee adduction angles for example. Furthermore, the static angles as calculated by HBM can potentially be used in clinical decision making, for example to determine the amount of tibial rotation. The observed differences between models further emphasize the importance of using a normative database based on the same kinematic model, when comparing patients to healthy subjects. In HBM, global optimization influences all segments, which results in differences with conventional models for all segments, but since most differences remain below 5°, this is not expected to influence clinical decision making. Overall, differences between PiG and CAST were on average larger than between HBM and the two conventional models. Hence these findings do not suggest preferred use of conventional models over HBM. A ground truth would be required to decide on which of the three models is the most accurate. Considering the limited differences and their systematicity, HBM can be useful for providing rehabilitative feedback, and for clinical decision making at least regarding sagittal plane kinematics.

5. Conclusion

Overall, the differences in gait kinematics we found between marker models were not pointing to a specific outlier, i.e. based on agreement there is no preferred use of either PiG, CAST or HBM. For the sagittal plane angles the models were found to be equivalent, i.e. any non-systematic difference was below the minimal clinically significant difference of 5°. However, differences of up to 25°

were found in frontal and transversal planes for hip, knee and ankle joints, between all models. For these planes the three models cannot be used interchangeably. A ground truth would be required to decide on which of the three models is the most accurate.

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Author contribution

We confirm that the manuscript has been read and approved by all named authors, with the exception of P. Cappa, who deceased on 26 August 2016, before the manuscript was finalized. There are no other persons who satisfied the criteria for authorship but are not listed. We further confirm that the order of authors listed in the manuscript has been approved by all of us, with the exception of P. Cappa.

Declaration of Competing Interest

Eline Flux, Marjolein van der Krogt and Jaap Harlaar provided suggestions for improvements to the developers of HBM, without any financial interests. Furthermore, Motek Medical, the company that developed HBM, was included as a partner in the MD-Paedigree project, but did not perform payment for this project. We wish to confirm that there are no further known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome.

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Appendix A. Supplementary data

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References

- Ancillao, A., van der Krogt, M. M., Buizer, A. I., Witbreuk, M. 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.
- van den Bogert, A. J., Geijtenbeek, T., Even-Zohar, O., Steenbrink, F., & Hardin, E. C. (2013). A real-time system for biomechanical analysis of human movement and muscle function. *Medical & Biological Engineering & Computing*, 51(10), 1069–1077. https://doi.org/10.1007/s11517-013-1076-z.
- Booth, A., Steenbrink, F., Buizer, A. I., Harlaar, J., & Van der Krogt, M. M. (2017). O73: Does real-time feedback on ankle power alter dynamic motor control in children with cerebral palsy? *Gait & Posture*, 57, 126–127. https://doi.org/10.1016/J.GAITPOST.2017.06.326.
- Booth, A. T., Buizer, A. I., Harlaar, J., Steenbrink, F., & van der Krogt, M. M. (2019). Immediate effects of immersive biofeedback on gait in children with cerebral palsy. *Archives of Physical Medicine and Rehabilitation*, 100(4), 598–605. https://doi.org/10.1016/J.APMR.2018.10.013.
- Booth, A. T. C., Buizer, A. I., Meyns, P., Oude Lansink, I. L. B., Steenbrink, F., & van der Krogt, M. M. (2018). The efficacy of functional gait training in children and young adults with cerebral palsy: A systematic review and meta-analysis. *Developmental Medicine and Child Neurology, 60*(9), 866–883. https://doi.org/10.1111/dmcn.13708.
- Booth, A. T. C., Steenbrink, F., Buizer, A. I., Harlaar, J., & van der Krogt, M. M. (2016). Is avatar based real-time visual feedback a feasible method to alter gait parameters of interest? *Gait & Posture, 49*, 98. https://doi.org/10.1016/j.gaitpost.2016.07.154.
- 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.
- Davids, J. R., Rowan, F., & Davis, R. B. (2007). Indications for orthoses to improve gait in children with cerebral palsy. JAAOS Journal of the American Academy of Orthopaedic Surgeons, 15(3), Retrieved from https://journals.lww.com/jaaos/Fulltext/2007/03000/Indications_for_Orthoses_to_Improve_Gait_in.8.aspx.
- Davis, R. B., Ounpuu, S., Tyburski, D., & Gage, J. R. (1991). A gait analysis data collection and reduction technique. *Human Movement Science*, 10(5), 575–587. Delp, S. L., Anderson, F. C., Arnold, A. S., Loan, P., Habib, A., John, C. T., ... Thelen, D. G. (2007). OpenSim: Open-source software to create and analyze dynamic
- simulations of movement. *IEEE Transactions on Biomedical Engineering*, 54(11), 1940–1950.

 Duprey, S., Cheze, L., & Dumas, R. (2010). Influence of joint constraints on lower limb kinematics estimation from skin markers using global optimization. *Journal of Biomechanics*, 43(14), 2858–2862.
- Falisse, A., Van Rossom, S., Gijsbers, J., Steenbrink, F., van Basten, B. J. H., Jonkers, I. G., & De, F. (2018). OpenSim versus human body model: A comparison study for the lower limbs during gait. *Journal of Applied Biomechanics*, 34, 496–502.
- Ferber, R., McClay Davis, I., Williams, D. S., III, & Laughton, C. (2002). A comparison of within- and between-day reliability of discrete 3D lower extremity variables in runners. *Journal of Orthopaedic Research*, 20(6), 1139–1145. https://doi.org/10.1016/S0736-0266(02)00077-3.
- Ferrari, A., Benedetti, M. G., Pavan, E., Frigo, C., Bettinelli, D., Rabuffetti, M., ... Leardini, A. (2008). Quantitative comparison of five current protocols in gait analysis. Gait & Posture, 28(2), 207–216. https://doi.org/10.1016/j.gaitpost.2007.11.009.
- Friston, K., Mattout, J., Trujillo-Barreto, N., Ashburner, J., & Penny, W. (2007). Variational free energy and the Laplace approximation. *Neuroimage*, 34(1), 220–234. van Gelder, L., Booth, A. T. C., van de Port, I., Buizer, A. I., Harlaar, J., & van der Krogt, M. M. (2017). Real-time feedback to improve gait in children with cerebral palsy. *Gait* & *Posture*, 52, 76–82. https://doi.org/10.1016/j.gaitpost.2016.11.021.
- Giggins, O. M., Persson, U. M., Caulfield, B., Tate, J., Milner, C., Onate, J., ... Caulfield, B. (2013). Biofeedback in rehabilitation. *Journal of Neuroengineering and Rehabilitation*, 10(1), 1123–1134. https://doi.org/10.1186/1743-0003-10-60.
- Gorton, G. E., Hebert, D. A., & Gannotti, M. E. (2009). Assessment of the kinematic variability among 12 motion analysis laboratories. Gait & Posture, 29(3), 398-402.

- https://doi.org/10.1016/j.gaitpost.2008.10.060.
- Groen, B. E., Geurts, M., Nienhuis, B., & Duysens, J. (2012). Sensitivity of the OLGA and VCM models to erroneous marker placement: Effects on 3D-gait kinematics. Gait & Posture, 35(3), 517–521.
- Harrington, M. E., Zavatsky, A. B., Lawson, S. E. M., Yuan, Z., & Theologis, T. N. (2007). Prediction of the hip joint centre in adults, children, and patients with cerebral palsy based on magnetic resonance imaging. *Journal of Biomechanics*, 40(3), 595–602.
- Holm, S. (1979). A simple sequentially rejective multiple test procedure. Scandinavian Journal of Statistics, 65-70.
- Johnson, A. (2002). Prevalence and characteristics of children with cerebral palsy in Europe. *Developmental Medicine and Child Neurology*, 44(9), 633–640. https://doi.org/10.1017/S0012162201002675.
- Kainz, H., Modenese, L., Lloyd, D. G., Maine, S., Walsh, H. P. J., & Carty, C. P. (2016). Joint kinematic calculation based on clinical direct kinematic versus inverse kinematic gait models. *Journal of Biomechanics*, 49(9), 1658–1669. https://doi.org/10.1016/j.jbiomech.2016.03.052.
- Kerr Graham, H., & Selber, P. (2003). Musculoskeletal aspects of cerebral palsy. Journal of Bone and Joint Surgery (British), 85(2), 157–166. https://doi.org/10.1302/0301-620X.85B2.14066.
- Klejman, S., Andrysek, J., Dupuis, A., & Wright, V. (2010). Test-retest reliability of discrete gait parameters in children with cerebral palsy. Archives of Physical Medicine and Rehabilitation, 91(5), 781–787.
- van der Krogt, M. M., Sloot, L. H., & Harlaar, J. (2014). Overground versus self-paced treadmill walking in a virtual environment in children with cerebral palsy. *Gait & Posture*, 40(4), 587–593.
- Lafortune, M. A., Cavanagh, P. R., Sommer, H. J., & Kalenak, A. (1992). Three-dimensional kinematics of the human knee during walking. *Journal of Biomechanics*, 25(4), 347–357. https://doi.org/10.1016/0021-9290(92)90254-X.
- Leboeuf, F., Reay, J., Jones, R., & Sangeux, M. (2019). The effect on conventional gait model kinematics and kinetics of hip joint Centre equations in adult healthy gait. Journal of Biomechanics, 87, 167–171. https://doi.org/10.1016/j.jbiomech.2019.02.010.
- McGinley, J. L., Baker, R., Wolfe, R., & Morris, M. E. (2009). The reliability of three-dimensional kinematic gait measurements: A systematic review. *Gait & Posture*, 29(3), 360–369. https://doi.org/10.1016/J.GAITPOST.2008.09.003.
- Pfeufer, D., Gililland, J., Böcker, W., Kammerlander, C., Anderson, M., Krähenbühl, N., & Pelt, C. (2018). Training with biofeedback devices improves clinical outcome compared to usual care in patients with unilateral TKA: a systematic review. *Journal of Pediatric Gastroenterology and Nutrition*. https://doi.org/10.1007/s00167-018-5217-7.
- Sangeux, M. (2018). Computation of hip rotation kinematics retrospectively using functional knee calibration during gait. *Gait & Posture*, 63, 171–176. https://doi.org/10.1016/J.GAITPOST.2018.05.011.
- Schutte, L. M., Narayanan, U., Stout, J. L., Selber, P., Gage, J. R., & Schwartz, M. H. (2000). An index for quantifying deviations from normal gait. *Gait & Posture*, 11(1), 25–31.
- Sloot, L. H., van der Krogt, M. M., & Harlaar, J. (2014). Self-paced versus fixed speed treadmill walking. *Gait & Posture*, 39(1), 478–484. https://doi.org/10.1016/J. GAITPOST.2013.08.022.
- Tate, J. J., & Milner, C. E. (2010). Systematic review biofeedback during gait retraining in patients: A real-time kinematic, temporospatial, and kinetic biofeedback during gait retraining in patients: A systematic review. *Physical Therapy*, 90, 1123–1134. https://doi.org/10.2522/ptj.20080281.
- Wu, G., Siegler, S., Allard, P., Kirthley, C., Leardini, A., Rosenbaum, D., ... Stokes, I. (2002). ISB recommendation on definitions of joint coordinate system of various joints for the reporting of human joint motion—Part I: ankle, hip, and spine. *Journal of Biomechanics*, 35(4), 543–548. https://doi.org/10.1016/S0021-9290(01)
- Zeni, J. A., Richards, J. G., & Higginson, J. S. (2008). Two simple methods for determining gait events during treadmill and overground walking using kinematic data. *Gait & Posture*, 27(4), 710–714.