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The relation between clinical and objective gait scores in clubfoot patients with and without a relapse

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ABSTRACT

Background: Objective gait analysis that fully captures the multi-segmental foot movement of a clubfoot may help in early identification of a relapse clubfoot. Unfortunately, this type of objective measure is still lacking in a clinical setting and it is unknown how it relates to clinical assessment.
Research question: The aim of this study was to identify differences in total gait and foot deviations between clubfoot patients with and without a relapse clubfoot and to evaluate their relationship with clinical status.
Methods: In this study, Ponseti-treated idiopathic clubfoot patients were included and divided into clubfoot patients with and without a relapse. Objective gait analysis was done resulting in total gait and foot scores and clinical assessment was performed using the Clubfoot Assessment Protocol (CAP). Additionally, a new clubfoot specific foot score, the clubFoot Deviation Index (cFDI*), was calculated to better capture foot kinematics of clubfoot patients.
Results: Clubfoot patients with a relapse show lower total gait quality (GDI*) and lower clinical status defined by the CAP than clubfoot patients without a relapse. Abnormal cFDI* was found in relapse patients, reflected by differences in corresponding variable scores. Moderate relationships were found for the subdomains of the CAP and total gait and foot quality in all clubfoot patients.
Significance: A new total foot score was introduced in this study, which was more relevant for the clubfoot population. The use of this new foot score (cFDI*) besides the GDI*, is recommended to identify gait and foot motion deviations. Along with clinical assessment, this will give an overview of the overall status of the complex, multi-segmental aspects of a (relapsed) clubfoot. The relationships found in this study suggest that clinical assessment might be indicative of a deviation in total gait and foot pattern, therefore hinting towards personalised screening for better treatment decision making.

1. Introduction

Despite good initial outcome of the Ponseti treatment, a relapse of the clubfoot still causes challenges in clubfoot care. Early identification of a relapse is important in clubfoot care [1]. Relapse clubfeet that are identified in an early stage can often be treated with non-invasive treatment methods [2,3]. Whereas late recognition of a relapse often results in a surgical approach, which might adversely affect foot functionality later in life [4]. Unfortunately, early identification of a relapse can be difficult due to the diverse nature of a relapse [5]. Furthermore, objective measurements that fully capture the multi-planar and multi-joint movement within a clubfoot, are still lacking in a clinical setting. Objective assessment of foot functionality and overall physical ability of the child over time may aid in the early identification of a relapse.

Three-dimensional gait analysis (3DGA) is frequently used as an objective assessment tool to evaluate functional outcomes after clubfoot treatment [6]. 3DGA quantifies the gait pattern by providing objective

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data of numerous gait parameters. Gait indices representing a single score to assess gait quality, such as the gait deviation index (GDI) and gait profile score (GPS), were developed to aid interpretation of these numerous 3DGA parameters [7,8]. In clubfoot patients, deviant GDI and GPS were found when compared to healthy controls. Overall deviations in total gait scores were explained by differences in foot progression, ankle plantarflexion, knee flexion and pelvic tilt [9–12]. In the GDI and GPS, relatively little attention is paid to specific motions of different segments of the foot. Furthermore, they do not include frontal plane kinematics of the foot such as supination/pronation, while this is a typical deviation seen in relapse clubfoot [13–15]. Considering the multi-planar and multi-joint nature of a clubfoot, it is highly important to implement multi-segment foot models in gait analysis. Total gait scores that include the multi-segmental Oxford Foot Model (OFM), such as the foot profile score (FPS), are preferred in the assessment of children with a clubfoot, as it is more clinically meaningful for these patients [14]. Only one study has examined total foot scores of clubfoot patients and found a difference in FPS with healthy age-matched controls, which was explained by variations in forefoot supination and adduction and hindfoot internal rotation [15].

To facilitate interpretation of the FPS score, a transformation of the score (previously used in the GPS and described by McMulkin et al.) could be used. This transformed score, also called the foot deviation index (FDI*), was used to make a classification between normal and abnormal scores, which is not possible using only the raw FPS scores. Besides the existing foot scores (FPS/FDI*), we introduce a new foot score: the clubFoot Deviation Index (cFDI*). This foot score was based on the FDI*, but also includes the angles of the forefoot relative to the tibia which are known to deviate in relapse patients [13,14].

Although the FPS and other gait scores provide a detailed description of clubfoot status, it requires an intensive and time consuming 3DGA which is not always possible in a general clinical setting and therefore not included in the standard care in clubfoot patients. This might be one of the reasons why both 3DGA and total gait scores are still relatively little used in a clinical setting, despite the added value of these objective measurements for clinical decision making [16]. In clinical practice, the Clubfoot Assessment Protocol (CAP) is used as an objective clinical evaluation tool to assess the development of clubfoot patients over time, from which the overall status of the child's functional ability can be derived [10,11,17,18]. Previous literature reported poor to moderate relationships between gait scores and the morphology and quality of movement subdomains of the CAP. This suggests that the clinically observed gait deviations and differences in morphology may be reflected in the total gait scores in clubfoot patients [10,11].

Even though poor to moderate correlations seem to exist between clinical measures and total gait scores in the clubfoot population, information about the relation between clinical measures and detailed foot scores still lacks. To identify a relapse clubfoot, detailed foot scores are clinically relevant considering the multi-segmental characteristics of a (relapse) clubfoot. Hence, the aim of this study was to identify differences between clubfoot patients with and without a relapse clubfoot in total gait and foot deviations, based on total gait and foot scores, and to evaluate the relationship with the clinical status in these patients.

We expected to find lower total gait and foot scores and lower clinical scores in patients with a relapse compared to patients without a relapse. Moreover, we expect positive moderate relationships between total foot scores and CAP subdomains morphology and quality of movement, especially with the item walking [11].

2. Methods

2.1. Study design

A cross-sectional study was performed in a convenience sample of patients with clubfoot, including clinical assessment and objective gait assessment. Objective gait assessment was performed using 3DGA at the Fontys University of Applied sciences. Clinical assessment, using the CAP was performed by the treating orthopaedic surgeon during the regular consultation at the hospital.

This study was approved by the Medical Ethics Committee United (MEC-U) and local review board of the Catharina Hospital and Máxima MC [METC NL53229.100.15/ nWMO 2014-69].

2.2. Study population

Thirty-one uni- or bilateral idiopathic clubfoot patients treated according to the Ponseti method with an age of 4–8 years old were included. Patients were recruited at the Catharina Hospital and the Máxima MC. During consultation, the treating orthopaedic surgeon (AB) assigned the patient to the group of successfully treated clubfoot (the non-relapse group) or relapse clubfoot (the relapse group). A relapse clubfoot was defined as a reoccurrence of one or more of the original deformities of the clubfoot, after initial successful correction and needed additional treatment. Additional treatment comprises of additional casting, bracing, physiotherapy, or surgical treatment (tibialis anterior tendon transfer or anterior distal tibial epiphysiodesis). Unilateral measurements were performed for every patient, which comprised the affected side in unilateral patients and the most affected side in bilateral patients. The most affected side was selected by the treating orthopaedic surgeon, based on clubfoot severity (in the non-relapse group) or planned treatment (in the relapse group). Measurements were performed before relapse treatment. Participants were excluded when they: I) were unable to follow instructions; II) had another disorder of the lower extremity; III) received prior additional surgical treatment for relapse. Renewed-Achilles tenotomy to correct residual equinus early in the bracing period was not considered additional treatment.

2.3. Three-dimensional gait analysis

Gait data capturing and trial selection of the data set were previously reported [14]. Kinematic modelling was performed by using an extended Helen-Hayes model and the OFM. Kinematic gait pattern was evaluated using three total gait scores.

The logarithmic transformed GPS (GDI*) and logarithmic transformed FPS (FDI*) were calculated based on 15 gait parameters [7,19]. Data of 21 typically developing children (mean age 6 $\pm$ 1.57 years, 57 % male) was used as a reference [14]. The GDI* was calculated according to the original description [8,12]. The FDI* was calculated from the FPS according to the same methods as the GDI* was calculated from the GPS. Additionally, we calculated an adapted FDI*-score, the clubFoot Deviation Index (cFDI*), which includes nine angles of the OFM (Supplementary data – calculation cFDI*). Compared to the FDI*, this cFDI* also includes the angles of the forefoot relative to the tibia, which are known to deviate in relapse patients [13,14]. A GDI*, FDI* and cFDI* of 100 with a standard deviation of 10 represents a typical gait pattern and its normal variability, GDI*, FDI* and cFDI* $\leq$ 90 indicate a deviating gait pattern. Furthermore, separate gait variable scores were computed and visualised in a movement analysis profile (MAP) [7].

2.4. Clubfoot Assessment Protocol

The CAP was assessed according to the original protocol [17]. The CAP consists of 19 items, divided over 6 subdomains: passive mobility I and II, muscle function, morphology, and motion quality I and II. Item scores ranged between 0 (severe reduction/no capacity) and 4 (within normal variation). A raw subdomain score was calculated by taking the sum of all item score within the subdomain. This raw subdomain score was transformed to a 0 (extreme deviant) - 100 (normal) scale [17]. The CAP was assessed during a regular visit to the out-patient clinic by the treating orthopaedic surgeon (AB), with a maximum of three months before the 3DGA measurement took place.

Table 1
Demographic characteristics of the clubfoot and relapse group. Presented as mean ± SD or n (%).

	Relapse group (n = 13)		Non-relapse group (n = 18)	
Demographics				
Age [years]	5.46	± 1.51	5.39	± 1.46
Height [m]	1.14	± 0.10	1.14	± 0.09
Weight [kg]	21.19	± 4.25	20.52	± 3.82
Gender [M/F]	8 (62 %)	/5 (38 %)	15 (83 %)	/3 (17 %)
Laterality [uni/bi]	6 (46 %)	/7 (54 %)	9 (50 %)	/9 (50 %)
Walking velocity [/s] *	1.72	± 0.27	1.68	± 0.21
Stride length*	1.45	± 0.14	1.43	± 0.11
Initial treatment				
Achilles tenotomy	13	(100 %)	18	(100 %)
Renewed-Achilles tenotomy**	5	(38 %)	6	(32 %)
Relapse characteristics°				
Equinus/Limited dorsiflexion	8	(62 %)	–	
Hindfoot varus	10	(76 %)	–	
Cavus	1	(8 %)	–	
Forefoot adduction	8	(62 %)	–	
Active supination	10	(76 %)	–	
Relapse treatment				
Additional bracing	5	(38 %)	–	
TATT	5	(38 %)	–	
ADTE	3	(24 %)	–	

TATT – Tibialis anterior tendon transfer; ADTE – anterior distal tibial epiphysiodesis.
* Walking velocity and stride length are normalised for leg length. **Renewed Achilles tenotomy was performed before the age of three. °In 85 % of the cases a combination of relapse characteristics was present. An overview of relapse characteristics for each patient in the relapse group is presented in the Supplementary data (Appendix 1 – characteristics relapse group).

2.5. Statistical analysis

Statistical analyses were performed using SPSS (IBM SPSS Statistics 23). CAP scores, GDI*, FDI* and cFDI*, including gait variable scores, were presented as mean and standard deviation for both the non-relapse and the relapse group. Comparisons between the two groups were analysed using student T-tests, or if applicable, nonparametric alternatives. To assess the relationship between the CAP scores and total gait scores, Spearman's rank correlation was used. Relations were interpreted as poor (≤ 0.4), moderate (0.41–0.6), good (0.61–0.8) and very good (> 0.8)[20]. For the correlation analysis, all patients were taken together to cover a larger range of CAP and gait scores. The item "walking" from the subdomain motion quality was also used for the correlation analysis [11]. After examining the data, the CAP subdomain muscle function and passive mobility II were excluded from the correlation analyses. The majority (muscle function: 74 %, passive mobility II: 94 %) of all patients scored the maximum value. Due to this ceiling effect in the data, the results of the correlation were not deemed informative.

3. Results

3.1. Demographic characteristics

In total 31 patients (13 relapse clubfeet and 18 clubfeet without relapse) were included in this study. In the relapse group, five patients were planned for non-operative treatment and eight patients for operative treatment (Table 1). No differences between groups were found.

3.2. Gait scores

The relapse group showed a significant lower GDI* (88.88 ± 3.88) than the non-relapse group (93.57 ± 5.21) (Table 2). However, no differences were found in separate gait variable scores (Fig. 1A). Although the non-relapse group showed a normal average GDI* above 90, still 33 % of these patients showed an abnormal GDI*. In the relapse group, 62

Table 2
Total gait/foot scores and clinical sub scores for the non-relapse and relapse group, depicted in mean ± standard deviation and percentage of patients with a deviating score.

	Relapse group (n = 13)			Non-relapse group (n = 18)		
	mean	±SD	% deviating	mean	±SD	% deviating
Gait scores						
GDI*	**88.88**	**± 3.88**	**62%**	**93.57**	**± 5.21**	**33%**
FDI*	93.48^	± 5.07	25%	94.83^	± 5.08	9%
cFDI*	87.82^	± 5.53	58%	92.30^	± 4.52	27%
Clinical assessment - CAP						
Passive mobility 1	**48.85**	**± 16.60**	**100%**	**80.97**	**± 14.68**	**40%**
Passive mobility 2	100.00	± 0.00	0%	94.44	± 16.17	2%
Muscle function	89.73	± 14.50	8%	98.14	± 5.40	0%
Morphology	**62.50**	**± 16.93**	**85%**	**94.91**	**± 8.99**	**16%**
Motion quality 1	**68.30**	**± 13.63**	**69%**	**93.77**	**± 6.77**	**5%**
Motion quality 2	**63.54**$^{+}$	**± 14.56**	**85%**	**79.86**	**± 23.93**	**47%**
Walking~	**3.15**	**± 0.80**	**23%**	**4.00**	**± 0.00**	**0%**

Bold text and grey areas indicate a significant difference between the two groups (p<0.05).
^ Non-relapse group n = 11 and relapse group n = 12 due to missing OFM kinematics. $^{+}$relapse group n=12 due to missing value. ~Item was scored on a scale 0-4.

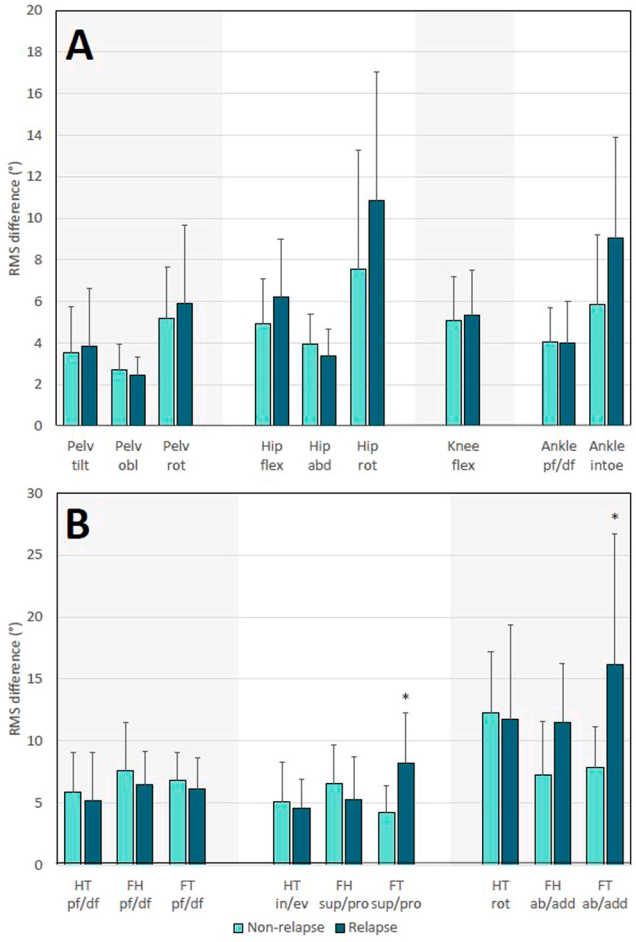

Fig. 1. *Variable scores for the corrected and relapse group. **A**; gait variable scores (relapse n = 13, non-relapse n = 18), **B**: foot variable scores(relapse n = 12, non-relapse n = 11).* * indicates a significant difference between the two groups ($p < 0.05$). Pelv – pelvic, obl – obliquity, rot – rotation, flex – flexion, abd – abduction, pf/df – plantar-/dorsiflexion, HT – hindfoot tibia, FH – forefoot hindfoot, FT – forefoot tibia, in/ev – in-/eversion, sup/pro – supination/pronation.

% of the patients showed an abnormal GDI*.

When looking at the foot scores, no differences were found in FDI* and foot variable scores between the two groups (Table 2, Fig. 1B). However, still 25 % of the relapse group showed an abnormal FDI*. Furthermore, the relapse group showed an abnormal cFDI* score (87.82 ± 5.53), while the non-relapse group showed a normal cFDI* score. Additionally, separate variable scores showed significant more deviation in supination/pronation and abduction/adduction of the forefoot relative to the tibia in the relapse group (Fig. 1B). Furthermore, a deviated cFDI* was found in 58 % and 27 % of the relapse group and non-relapse group, respectively.

3.3. Clinical scores

Significant lower CAP scores were found for the relapse group compared to the non-relapse group. These CAP scores of the relapse group were below the previously established cut-off points indicating good clinical outcome [18], except for the subdomain passive mobility 2 and muscle function (Table 2). Furthermore, relapse patients scored lower on the specific item 'Walking' from the subdomain motion quality.

3.4. Relationships between clinical and foot/gait scores

A moderate positive correlation was found for the CAP subdomain passive mobility 1 and motion quality 1 with the GDI* (Table 3). Also, a moderate positive correlation was found for the individual item 'walking' with the GDI*. No correlations were found for the CAP subdomains and the item 'walking' with the FDI* in all patients. For the cFDI*, however, the CAP subdomains morphology and motion quality 1 showed a moderate positive correlation.

4. Discussion

This study aimed to identify differences in total gait and foot deviations between clubfoot patients with and without a relapse and to evaluate the relationship with clinical function. The relapse group showed lower GDI* and CAP, indicating lower total gait quality and clinical physical functioning. Furthermore, the relapse group showed a deviated score for the newly introduced clubfoot specific foot score, the cFDI*.

The lower total GDI* of the relapse group reflects a more deviating gait pattern when compared to the non-relapse group. These results are in line with previous literature in which common gait deviations associated with a relapse such as forefoot adduction and supination, external hip rotation and limited dorsiflexion were found [13,15,21]. However, there were no statistical differences in separate gait variables that could explain the differences in total gait quality between the relapse and the non-relapse group. When looking in more detail into the variable scores, the largest deviation in the relapse group compared to the non-relapse group was seen in the variable scores for hip rotation and ankle

Table 3
Results correlations between gait/foot scores (GDI*, FDI*, cFDI*) and clinical score (CAP).

	GDI* (n = 31)	FDI* (n = 23)	cFDI* (n=23)
Clinical assessment - CAP			
Passive mobility 1	**0.457‡‡**	0.073	0.315
Morphology	0.312	0.399	**0.587‡‡**
Motion quality 1	**0.422‡**	0.228	**0.504‡**
Motion quality 2^	0.065	0.077	0.221
Motion quality – item Walking	**0.477‡‡**	0.171	0.403

‡/‡‡ and bold style indicating a significant correlation coefficient (‡ p<0.05. ‡‡ p<0.01)).
^ correlation with GDI n=30, FDI* and cFDI* n = 22 due to missing value*

intoeing, both previously described in relapse patients. Intoeing during walking is reported in 20 % of Ponseti-treated clubfoot patients with a relapse [3]. External hip rotation is often seen as a compensation mechanism for intoeing and is reported in 32 % of these patients [3]. The combination of both features might have contributed to the deviation in total gait pattern, which underlines the benefit of using a total gait score that reflects multiple variables into a more global score.

The non-relapse group showed a mean GDI* above 90, reflecting a normal gait pattern. Nevertheless, still 33 % patients in this group showed an abnormal gait pattern which implies variation in their gait pattern. In line with our results, 5 years after treatment approximately 55% of the Ponseti-treated clubfeet without additional treatment needed for a relapse, showed abnormal ankle sagittal plane motion, and 25 % of the feet showed intoeing ($> 5°$) [22]. This indicates a large variability in gait deviations within the non-relapse group, which could be a harbinger of a relapse. Therefore, monitoring patients' scores might be interesting for detecting possible future relapses.

Surprisingly, the FDI* and variable scores of the relapse group did not indicate a deviated gait pattern. Especially in the relapse group we expected an abnormal FDI*, as deviations in detailed foot motion in patients with relapse exist [13,14]. This might indicate that the deviations in detailed foot motion of the clubfoot cannot be fully captured by the original FDI*. One of the major disadvantages of using the FDI* in clubfoot patients is that motion of the forefoot relative to the tibia is neglected, while previous literature stated that most of the deviations in clubfoot patients were found in this angle [13,14]. Based on this, we developed a new foot score which is more relevant for clubfoot patients as it includes motion of the forefoot relative to the tibia: the cFDI*. For this new score, the results were in line with our hypotheses. The relapse group showed an abnormal mean total foot score where the non-relapse group did not. In the separate foot variable scores, statistically significant differences were found for the ab/adduction and pro/supination of the forefoot relative to the tibia, which was in line with foot motion abnormalities that were found in previous studies [13,14]. Subsequently, we recommend the use of the cFDI* when using a total foot score to assess the gait quality of clubfoot patients.

In line with the GDI* and cFDI*, the scores on all subdomains of the CAP were lower for the relapse group, except for the subdomain muscle function where both groups scored high. The ceiling effect for this subdomain was previously reported by Andriesse et al. [23] which suggests that it is a less discriminative subdomain of the CAP. The differences found in the other subdomains were expected, as these subdomains mostly reflect the morphological and specific functional characteristics of the clubfoot and a relapse is a reoccurrence of one (or more) of these characteristics. Not only did the relapse group show lower CAP subdomain scores, these CAP subdomain scores were on average also below the cut-off points of 75 %, indicating abnormal clinical physical functioning [18].

This abnormal clinical functioning in clubfoot patients with a relapse, together with the deviations in total gait and foot movement, suggests that there might be a relationship between 3DGA outcomes and clinical scores. In line with previous research [11], our results showed the a priori hypothesised positive correlation between the GDI* and the CAP subdomain motion quality part one and the CAP item walking. This suggests that a similar construct is measured [11]. The correlation with CAP subdomain passive mobility reflects the high impact joint mobility has on the available functional range of motion during walking. Opposed to previous research of Manousaki et al., our study did not find a correlation between the CAP subdomain morphology and GDI*. However, the small sample size could have resulted in low statistical power, thus, the interpretation of these moderate relationships should be done with caution. Furthermore, we found a moderate positive correlation between the cFDI* and the CAP domain morphology. This could be explained by the items in this domain describing anatomical positions of the different segments, which affect the relative foot motion during gait. Furthermore, the moderate positive relation between the cFDI* and the CAP domain motion quality 1 reflects similarities in objectively measuring detailed foot motion and the overall assessment of the ability of the foot to execute different activities. Additionally, the poor correlations found for the FDI* with the CAP might, again, indicate that the FDI* is not sensitive enough for clubfoot patients and thus measures a different construct than the CAP.

In a clinical setting, clinical assessment and gait analysis are used as complementary assessments to retrieve an overall image of the patients. It could be an advantage to have complementary assessment tools, as one tool may not be enough to capture the complexity of the functioning, clinical characteristics and other aspects that play a role in the complexity of clubfoot. The link between the CAP scores and total gait quality suggests that the CAP could be used as indication at the individual patient level to decide whether additional detailed gait analysis is necessary, in which it is recommended to use the cFDI* and corresponding variable scores. This gait analysis then provides information that fully captures a child's functioning and their deviations, which assists a well-considered treatment decision. In the future, planned treatment could be evaluated based on the results of the 3DGA, since previous literature already showed the added value of detailed 3DGA in clinical decision making [16].

In conclusion, this study showed that clubfoot patients with a relapse have lower clinical status and deviated total gait and foot motion quality. Furthermore, correlations between clinical status and gait/foot motion quality showed that abnormalities in various CAP domains indicate a deviated gait pattern and deviations in specific foot motion. Therefore, in patients with lower scores on these subdomains, detailed gait analysis (using GDI* and cFDI*) is recommended to identify gait and foot motion deviations per individual. This allows more personalised screening and quantification on possible problems which could aid clinical decision making.

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CRediT authorship contribution statement

L. Grin: Investigation, Formal analysis, Writing – original draft. **S.D. N. Wijnands:** Investigation, Formal analysis, Writing – original draft. **A. T. Besselaar:** Conceptualization**,** Resources, Writing – review & editing. **L. van Oorschot:** Investigation, Writing – review & editing. **B.M.P. Vanwanseele:** Conceptualization, Resources, Supervision, Writing – review & editing. **M.C. van der Steen:** Conceptualization, Supervision, Writing – review & editing.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.gaitpost.2022.07.261.

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