Consensus for the measurement of the Camptocormia angle in the standing patient

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Abstract

Introduction: Camptocormia is characterized by a pathological forward flexion of the trunk, which is reversible when lying and worsened by standing and walking. So far there is no consensus on how to measure the angle of flexion, and studies therefore give differing results. Harmonization is needed for both research and clinical practice. Orthopedic measures are not useful for this purpose.

Methods: Two expert raters independently analyzed the photographs of 39 Parkinson patients with camptocormia while standing. They used four different methods to determine the camptocormia angle. The results were compared statistically. An international Consensus Group reviewed the results and drafted recommendations.

Results: The four methods yielded camptocormia angles that differed by up to 50% in the same patient. Inter-rater reliability and test-retest reliability also differed, but were satisfactory to excellent.

Conclusion: This Consensus Group concluded that two of the methods qualified as reliable measures of the trunk angles in standing patients based on their clinimetric properties. They propose that the 'total camptocomia angle' be the angle between the line from the lateral malleolus to the L5 spinous process and the line between the L5 spinous process and the spinous process of C7. They also propose that the 'upper camptocormia angle' be the angle of the lines between the vertebral fulcrum to the spinous processes of L5 and C7, respectively. An app is provided on the web for these measurements (http://www.neurologie.uni-kiel.de/de/axial-posturale-stoerungen/camptoapp).

Keywords: Camptocormia, angle measurement, clinical studies, bent spine syndrome

Introduction

Camptocormia is an involuntary, non-fixed, pathological forward flexion of the trunk frequently associated with Parkinson's disease (PD) [1, 2]. The condition is reversible when the patient is lying. This symptom is often more bothersome for the patient than the cardinal symptoms of PD itself. So far there is no established treatment for camptocormia [3]. Discontinuation of dopamine agonists [4] and chemodenervation with botulinum toxin or lidocaine have been recommended [5-8]. Case series have shown that deep brain stimulation in the subthalamic nucleus or the globus pallidus internus may improve camptocormia at least during the first years after disease onset [9, 10].

Cohort studies have found a prevalence of camptocormia of 5 to 19% [11]. There are many reasons for this broad range, among which are (1) cohort differences, as the condition is mainly observed in late disease stages, (2) differences in the *a priori* threshold angle (between 15° and 45°) used to define the diagnosis of camptocormia, and (3) differences in the methods themselves used to measure the camptocormia angle.

In order for epidemiologic and interventional studies of camptocormia to give meaningful results, there must be a general consensus on the method for determining the patient's flexion angle and on the diagnostic threshold angle. There are currently three commonly used methods for measuring the flexion angle [1, 8, 11]. Methods proposed for orthopedic surgery are not a valid alternative because they mainly address spinal mobility and not posture, and if posture is addressed they rely mainly on radiological findings. [12] This study assesses the advantages and disadvantages of each method as well as those of a new method and concludes with a recommendation.

Patients and methods

Thirty-nine patients with camptocormia and PD according to the UK Brain Bank PD criteria were recruited from the movement disorders outpatient clinic at the Department of Neurology, Kiel University. The diagnosis of camptocormia was based on patient complaints of an involuntary forward flexion of the trunk and confirmation of this condition during a full clinical assessment by a movement disorder neurologist. No *a priori* definitions of the camptocormia angle were used to make the diagnosis.

Lateral view pictures of the patients were taken with the camera lens at approximately waist level were used for the study. The freeware program GIMP 2.8.22 (©Spencer Kimbal, Peter Mattis et al.) was used to draw the respective lines in the photographs and to measure the angles. Two experienced raters (GD, RW) were trained in the use of this program. They employed it to independently determine the camptocormia angle in the photographs using the three currently favored measurement methods. The results were stored for analysis. The measurements were done according to the descriptions in the literature (see Fig. 1).

In the 'fulcrum method' [8] a line is drawn between the spinous processes of vertebrae C7 and L5 (corresponding approximately to the sacro-iliac joint). The inflection point or fulcrum is the most distant point perpendicular to the L5/C7 line. From the fulcrum, a line is drawn

perpendicular to the ground and the outer angle between these two lines is defined as the camptocormia angle (Fig. 1).

In the 'malleolus method' [11] a line is drawn from the L5 end of the L5/C7 line to the lateral malleolus of the foot and the outer angle between these lines defines the camptocormia angle (Fig. 1).

In the 'perpendicular method' [1] a line is drawn from the L5 end of the L5/C7 line perpendicular to the ground and the outer angle between these two lines is defined as the camptocormia angle (Fig. 1).

The fulcrum method was originally developed to assess upper camptocormia and it is used in patients with both lower and upper camptocormia. The angle is affected by the lower flexion because it uses a perpendicular line. Therefore, we propose as fourth method the following new measurement method: the upper camptocormia angle is defined as the outer angle between the two lines fulcrum-L5 and fulcrum-C7 with the fulcrum as defined above (upper camptocormia method) (Fig. 1).

Ten patients were selected for test-retest variability measurements. The data were statistically analyzed with R statistics (https://www.r-project.org) and the irr-package (version 0.84) to calculate the coefficients for inter-rater reliability and agreement. The ethics committee of the medical faculty approved the study. Informed consent was obtained from all participants.

The consensus group was involved in the entire process of defining the optimal measurement. After having agreed on the collaboration, the group first discussed the angles given by the fulcrum, the malleolus and the perpendicular methods. The group was not satisfied with the results of the upper camptocormia angle. The upper camptocormia angle measurement was developed further and the measurements were added. The resulting paper was then discussed a final time, and all members agreed on the interpretation of the findings and the content of the manuscript.

Results

The clinical and demographic data of the cohort are shown in Supplementary Table S1.

Figure 2 near here

The different methods were applied to all patients and the results were used in the analysis. The fulcrum method yielded the largest angles of camptocormia (mean 56.7°) followed by the malleolus (41.6°) and the perpendicular method (33.1°). The upper camptocormia angle measurement gave a mean angle of 45.4°. The differences were statistically significant ($p<0.001$). (Fig. 2, Supplementary Table S2)

Inter-rater reliability between the two raters was lower for the fulcrum method than for the other three methods. The single score agreement measured by intra-class correlation coefficient (ICC) was 0.710 for the fulcrum method, but it was 0.979 for the malleolus method, 0.983 for the perpendicular method, and 0.943 for the upper camptocormia angle. The consistency ICC, which ignores a consistent angle deviation between the raters, was 0.719 for the fulcrum method, 0.985 for the malleolus method, 0.983 for the perpendicular method, 0.942 for the upper camptocormia angle method and 0.943 for the upper camptocormia method. Test-retest reliability measured by the intra-class correlation coefficient was 0.943/0.956 (rater 1/rater 2) for the fulcrum method; 0.990/0.981 for the malleolus method, 0.985/0.984 for the perpendicular method and 0.994/0.947 for the upper camptocormia method.

Discussion

Forward flexion in camptocormia comprises two components: the hip-angle, which represents the lower camptocormia angle and the angle produced by the angulation of the different vertebrae along the spine, which is collectively referred to as upper camptocormia. Together they give the total camptocormia angle, which is the most important measure for the patient. These angles can be determined precisely only by radiography, and the available tools for orthopedic surgery indeed mostly use radiography.[13] Although not validated for the current purpose, inclinometers may possibly be useful [14] but these measures cannot be blindly evaluated (e.g. on photo material) as required for scientific studies. The current study assessed three simple clinical methods from the field of movement disorders used to determine the total camptocormia angle in standing patients and one additional method to measure the upper camptocormia angle. We found that the three methods differed by as much as 50%. The same patient will have two different outcomes when measured by two different methods.

Now that the field of camptocormia research has matured to the stage at which controlled treatment studies can be conducted, a consistent and validated method for measuring the camptocormia angle is needed. Such a measurement must reflect the flexion angle of camptocormia as accurately as possible, it should lend itself for assessment by blinded raters using lateral views of the patient, it should allow this angle to be measured with the patient lightly dressed, and it should be as reliable as possible.

The pathomechanism of upper camptocormia is probably an excessive activity of the external oblique abdominal muscles [15] or a weakness of the dorsal trunk muscles [16] that could cause upper camptocormia, while overactivity of ventral hip muscles or reduced force of dorsal hip muscles could cause lower camptocormia. Future research is required to confirm this hypothesis [15]. In any case, the measurement of the upper and lower angles must be precise and reproducible. The lower camptocormia angle in isolation is almost impossible to identify in the clothed patient because the landmarks, i.e. the femur and the sacrum are difficult to see. This is not such a problem for the upper camptocormia angle. Even though the fulcrum of the vertebral column cannot be unequivocally detected by visual inspection alone in most patients, a line through the top of the inflection of the vertebral column, which is parallel to the line connecting the spinous processes of L5 and C7, can better identify the fulcrum. Therefore, we recommend using the upper camptocormia angle

as a separate measure of flexion of the vertebral column. As shown the proposed method has a high inter-rater reliability.

Regarding the three methods for determining the total camptocormia angle, we found the 'fulcrum method' to have the lowest inter-rater and test-retest reliabilities. The fulcrum method was originally developed to measure upper camptocormia, but essentially encompasses both lower and upper camptocormia. This method has a tendency to overestimate the angle of camptocormia as can be seen in Fig. 1, upper row. The malleolus and perpendicular methods appear to be better suited for determining the total camptocormia angle. Their reliability was equally good but the perpendicular method gave systematically smaller angles than the malleolus method. This is because a forward flexion of the trunk must be compensated by bending the knees and hip in order to maintain stability. On average the angles determined by the two methods differed by 8° or almost 25% of the angle. Therefore, the perpendicular method will give smaller angles and most likely has a lower sensitivity to change for clinical trials. The perpendicular method is also error prone for methodical reasons, because if the camera is tilted while taking the photograph, the perpendicular line will be incorrect and will introduces bias. A strong argument for the malleolus method is that the reference points are anatomical features of the patient's own body and the position of the malleolus is invariant.

A number of important questions are not answered by our study. Bias is possible since the camptocormia angle is not constant but depends on the condition under which the photograph is taken. In some centers, the patient is instructed to walk a few steps before standing still, and the photograph is taken without the patient having straightened up. In other centers, the patients are videotaped while walking 10-20 meters in order to detect the dynamic nature of camptocormia. There is currently no consensus on how to measure and score the dynamic nature of camptocormia, and the present paper does not deal with this aspect. There is no simple solution for this challenge, and more rigid protocols are greatly needed.

Consensus recommendation

We recommend the malleolus method as the standard for future measurements of the total camptocormia angle and recommend measuring the upper camptocormia angle separately, particularly in the context of interventional trials (Fig. 3). We propose to use the term 'total camptocormia angle'. The 'upper camptocormia angle' as defined here is based on earlier work [8, 15] and can serve to identify the specific contribution of the vertebral column to total camptocormia. Photographs should be taken horizontally from a distance of at least 3 to 5 m with the lens approximately at waist height. It is important that the photograph displays the patient in a screen-filling way. We have designed an app, which can be used to conveniently measure these angles (http://www.neurologie.uni-kiel.de/de/axial-posturale-stoerungen/camptoapp).
The consensus group arrived at this conclusion as the currently best approach, which will facilitate scientific communication in the field. We hope that new methods may be developed in the future based on sensor technology. The issue of standards for the best movement sequence, which capture representative flexion angles, needs appropriate future studies. The presented angle assessment methods are not connected to the underlying

etiology or pathophysiology of any camptocormia form. Although not yet formally tested, the two proposed methods should also be applicable for non-parkinsonian camptocormia.

Author roles:

Contributions to the current study (A: study concept and design; B: acquisition of data; C: analysis and interpretation; D: critical revision of the manuscript for important intellectual content)

NM, RW, OG, GD: A, B, C, D

All other authors: A, C, D.

COI:

NGM has received lecture fees from Merz Pharmaceuticals and Bayer Pharmaceuticals and a travel grant from Eisai. He is a government employee in the Department of Neurology, University of Kiel, Germany.

RW, OG and RD have nothing to declare.

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YF has nothing to declare

MH serves as Chair of the Medical Advisory Board for and may receive honoraria and funding for travel from the Neurotoxin Institute. He may accrue revenue on US Patent #6,780,413 B2 (Issued: August 24, 2004): Immunotoxin (MAB-Ricin) for the treatment of focal movement disorders, and US Patent #7,407,478 (Issued: August 5, 2008): Coil for Magnetic Stimulation and methods for using the same (H-coil); in relation to the latter, he has received license fee payments from the NIH (from Brainsway) for licensing of this patent. He is on the Medical Advisory Board of CALA Health and Brainsway. He is on the Editorial Board of approximately 15 journals, and received royalties and/or honoraria from publishing from Cambridge University Press, Oxford University Press, and Elsevier. Dr. Hallett's research

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Figures

Figure 1. Four methods for measuring the camptocormia angle. The upper row shows a very severely affected and the lower row a moderately affected patient. Note the different angle outputs (numbers at the bottom of each photo) from each measurement method.

Figure 2. Results of the camptocormia measurements across the three methods. (A) Mean and SD of the measured angles by two blinded raters in the same patients with these methods (A) and the mean differences and SEM of the measurements between the two raters (B)

Figure 3. The consensus recommendation is to measure the total camptocormia angle as outlined in (A) and the upper camptocormia angle (B). Spinous processes C7 (C7) and L5 (L5); lateral malleolus (LM); fulcrum (FC).

Figure 1
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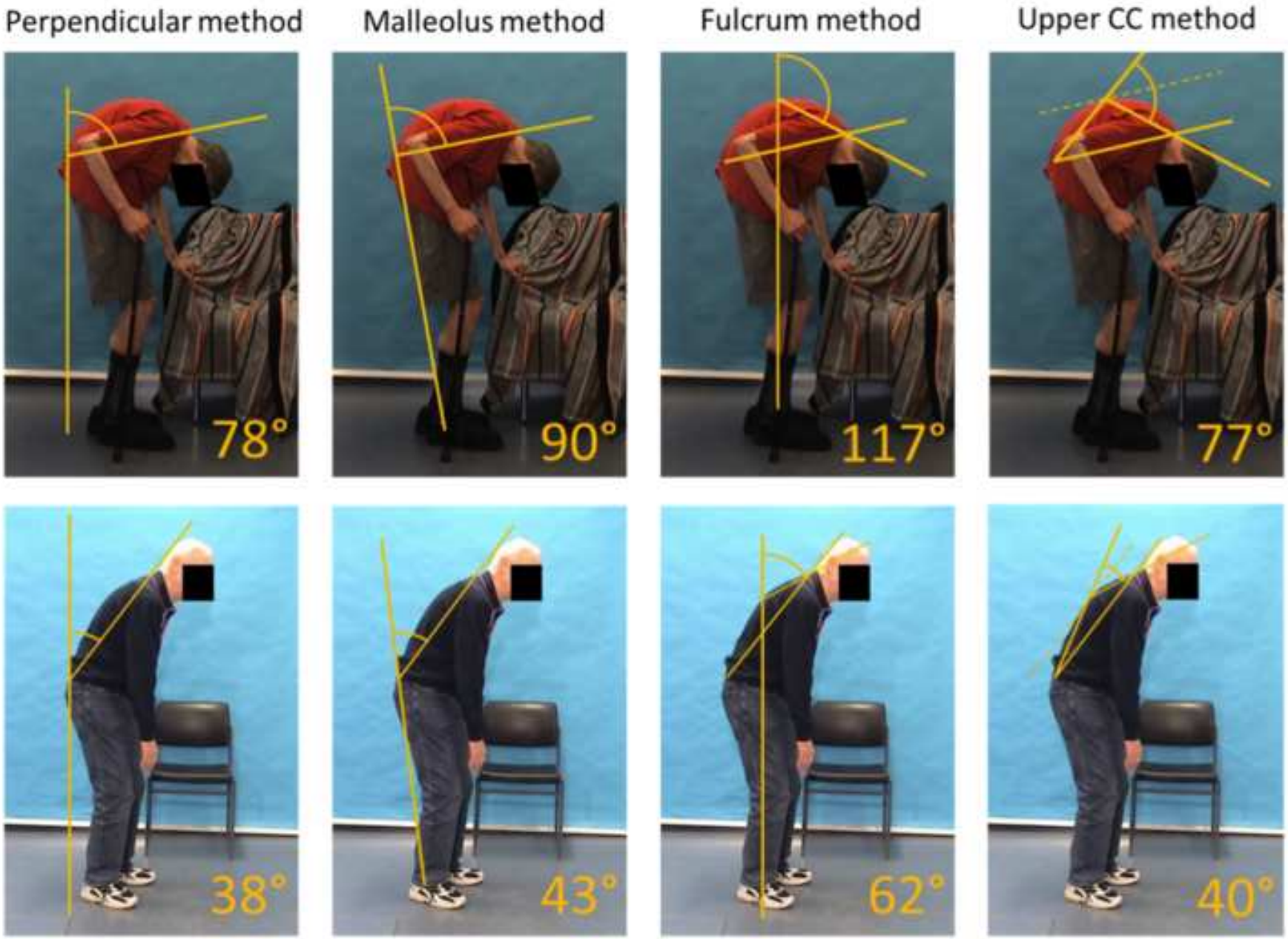

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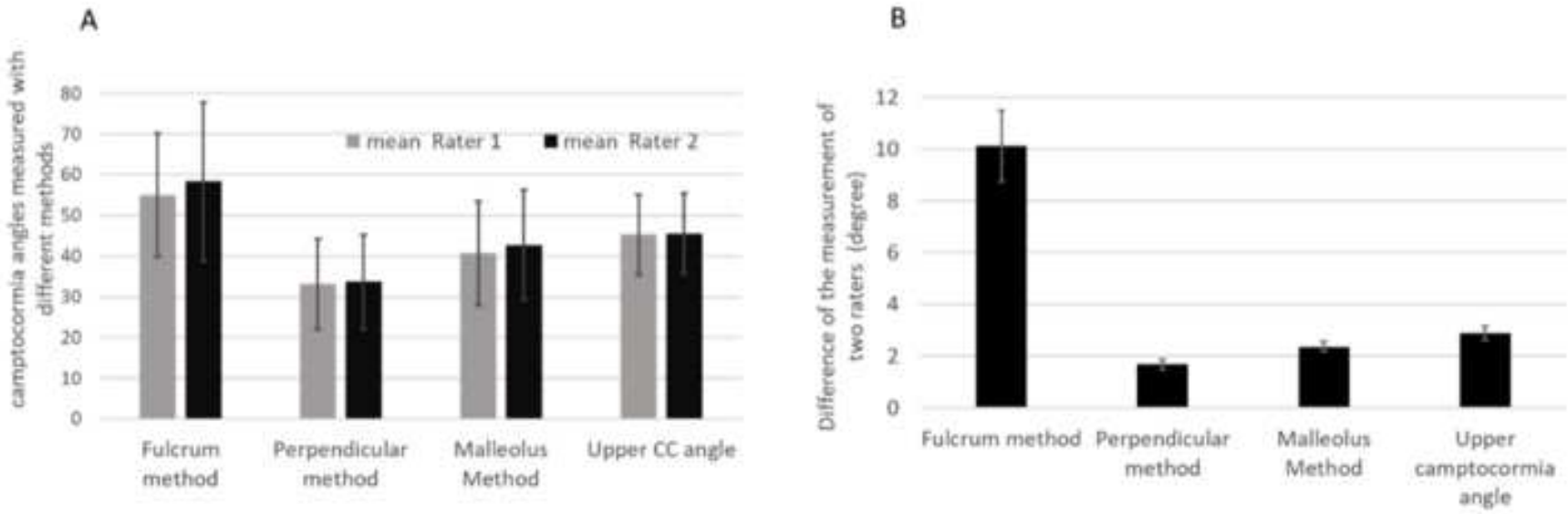
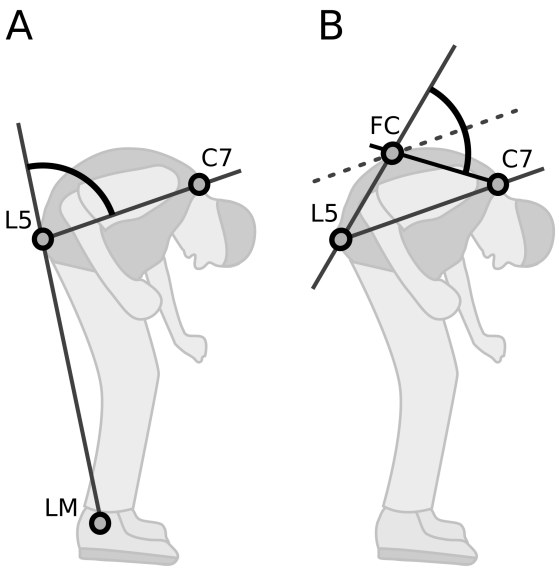


Figure 3



Supplementary Table S1

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Supplementary Table S2

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