



Parkinsonism and Related Disorders

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

Validity of the wall goniometer as a screening tool to detect postural abnormalities in Parkinson's disease



Parkins

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ARTICLE INFO

Keywords: Movement disorders Parkinsonism Bent spine syndrome Angle measurement Posture

ABSTRACT

Introduction: Software-based measurements of postural abnormalities in Parkinson's disease (PD) are the gold standard but may be time-consuming and not always feasible in clinical practice. Wall goniometer (WG) is an easier, quicker, and inexpensive instrument for screening patients with postural abnormalities, but no studies have investigated its validity so far. The aim of this study was to investigate the validity of the WG to measure postural abnormalities.

Methods: A total of 283 consecutive PD outpatients with \geq 5° forward trunk, lateral trunk or forward neck bending (FTB, LTB, FNB, respectively) were recruited from seven centers for movement disorders. Postural abnormalities were measured in lateral and posterior view using a freeware program (gold standard) and the WG. Both angles were expressed in degrees (°). Sensitivity and specificity for the diagnosis of camptocormia, Pisa syndrome, and anterocollis were assessed.

Results: WG showed good to excellent agreement (intraclass correlation coefficient from 0.80 to 0.98) compared to the gold standard. Bland-Altman plots showed a mean difference between the methods from -7.4° to 0.4° with limits of agreements from -17.7° to 9.5° . Sensitivity was 100% for the diagnosis of Pisa syndrome, 95.74% for anterocollis, 76.67% for upper camptocormia, and 63.64% for lower camptocormia. Specificity was 59.57% for Pisa syndrome, 71.43% for anterocollis, 89.80% for upper camptocormia, and 100% for lower camptocormia. Overall, the WG underestimated measurements, especially in lower camptocormia with an average of -8.7° (90% of cases).

Conclusion: WG is a valid tool for screening Pisa syndrome and anterocollis, but approximately 10° more should be added for camptocormia.

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https://doi.org/10.1016/j.parkreldis.2019.10.024

Received 26 June 2019; Received in revised form 21 October 2019; Accepted 23 October 2019 1353-8020/ © 2019 Elsevier Ltd. All rights reserved.

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1. Introduction

Postural abnormalities are disabling motor complications affecting patients with Parkinson's disease (PD) or atypical parkinsonism [1–3]. The most common are camptocormia, defined as forward trunk bending (FTB) $\geq 30^{\circ}$ at the lumbar fulcrum (lower) or $\geq 45^{\circ}$ at the thoracic fulcrum (upper) [4,5], Pisa syndrome, defined as $\geq 10^{\circ}$ of lateral trunk bending (LTB) [1,5], and anterocollis, $\geq 45^{\circ}$ of forward neck bending (FNB) [1,5]. They may occur isolated or combined at different stages of disease [5]. Postural abnormalities have a multifactorial pathophysiology [1–3] and are associated with higher disability because they increase the risk of falls [5,6], cause back pain and reduce mobility [1–8].

Previous studies investigating their prevalence yielded variable results for three main differences: 1) cohort, as postural abnormalities may be observed in early and late PD stages; 2) diagnostic criteria to define postural abnormalities; and, most importantly, 3) the methods to measure postural abnormality angles [3,9].

Recently, a consensus group suggested two software-based methods to measure camptocormia on photographs of patients: the total camptocormia angle and the upper camptocormia angle [9]. Moreover, most studies on Pisa syndrome and anterocollis used software-based picture analyses to ensure reproducibility and reliability of measurements [10,11]. These methods are reliable and valuable in the research setting [9] but might be less feasible in clinical practice because time-consuming due to the need to take pictures, which are subsequently transferred into a computer for degrees calculation.

Lack of consensus on a validated method to assess postural abnormalities has led to heterogeneity in the methods used among studies and difficulties to compare studies and translate the results of research into clinical practice [9]. Early recognition and accurate measurement of postural abnormalities are crucial to better manage these complications and avoid their progression and associated disability because they prompt an adjustment of pharmacological therapy (e.g. discontinuation of drugs associated with these abnormalities), increase monitoring and physiotherapy prescription [2,11,12].

In particular, since postural abnormalities are currently also diagnosed on the basis of diagnostic threshold angles [4], a valid instrument to visually estimate the flexion angle is needed to rapidly screen patients during routine follow-ups. The wall goniometer (WG) is a graduated laminated paper that can be applied on any vertical surface that allows an esteem of posture abnormalities at a glance [2,4–6]; however, its validity has not been investigated to date. The aim of this study was to analyze the validity of WG for measuring postural abnormalities in patients with PD. We hypothesized that the WG may be a valid instrument to detect postural alterations in PD, both in research and clinical settings.

2. Methods

This validity study compared software-based measurements (gold standard) [9] with the WG [2,4–6] to measure camptocormia, Pisa syndrome, and anterocollis in patients with PD.

2.1. Participants

A total of 283 consecutive outpatients with PD according to the clinical diagnostic criteria of the International Parkinson and Movement Disorders Society [13] were screened at seven Italian (north and central Italy) tertiary centers for movement disorders between March 2018 and November 2018 [5]. Patients with one or more FTB, LTB, and FNB of at least 5°, as measured by the WG, were evaluated. Exclusion criteria were: 1) concomitant neurological diseases affecting posture; 2) previous major spinal surgery, skeletal and/or muscle disease (e.g., vertebral fractures, spondylodiscitis, inflammatory myopathy); 3) diagnosis of atypical parkinsonism (i.e., multiple system

atrophy, progressive supranuclear palsy and corticobasal syndrome) [14]; (4) treatment with medications possibly causing postural abnormalities (neuroleptics other than clozapine or quetiapine and antiemetics, except for domperidone) in the 6 months prior to enrolment.

At each center, patients were assessed on their usual drug treatment, during the ON phase, in a single session by the same rater designated before study initiation. We recorded the following clinical and demographic variables: age, gender, body-mass index, age at PD onset, disease duration (since diagnosis of PD), modified Hoehn & Yahr scale to assess disease stage [15], and Unified Parkinson's Disease Rating Scale parts I-IV to assess disease severity (UPDRS) [16], PD subtype [17], laterality of PD symptoms onset, and Levodopa equivalent daily dose (LEDD) [18]. The institutional review boards of the participating centers reviewed and approved the study protocol. All patients were informed about the nature of the study and gave their written consent to participate. Authorization for disclosure (consent-to-disclose) of any recognizable persons in photographs has been obtained. The study is registered at http://clinicaltrials.gov(NCT03573232).

2.2. Devices and procedures

All study procedures were standardized across the participating centers. Standard anatomical landmarks for the trunk and lower limbs were reviewed and agreed among the examiners. Two instruments were used to measure FTB, LTB, and FNB.

The one instrument entailed the use of software-based measurement on photographs of the undressed patient (with underwear). At each center and for each patient sagittal and coronal views of the patients were taken with the camera lens at approximately waist level and at a distance of about 1.5 m. An experienced rater (CG) was trained in the use of Kinovea[®] [19] and independently drew the lines on all photographs in order to measure the angles.

FTB in the photographs was determined using two routine measurement methods [9]: 1) the upper camptocormia method, defined as the external angle between the two lines of fulcrum-L5 and fulcrum-C7, and 2) the total camptocormia angle using the malleolus method, by drawing a line from the L5 end of the L5/C7 line to the lateral malleolus of the foot, the external angle between these lines was considered. Patients were categorized as having lower FTB when a lumbar fulcrum (L1-sacrum, hip flexion) was evident, and upper FTB when a thoracic fulcrum (C7 to T12-L1) was present [4]. The LTB and FNB angles in the photographs were measured using the perpendicular method [10,11]. For the LTB angle, the external angle between the line drawn from the fulcrum to C7 and the line perpendicular to the ground was measured [9]. For the FNB angle, the external angle between the line drawn from C7 to the tragus of the ear and the line perpendicular to the ground was measured [20]. Photo acquisition and angle calculation took about 30 min to complete per patient.

The other instrument was the WG, which was used to visually estimate the degree of bending during standing or sitting (only for FNB) [4–6]. At each center, a rater was trained in the use of the WG and independently measured all angles. The zero of the WG was positioned at the same level as the fulcrum of trunk bending.

Camptocormia was captured in the sagittal view [4] and Pisa syndrome in the coronal view (showing the back) both with the patient in stance. The external angles between the line perpendicular to the ground and an imaginary line drawn from the fulcrum of bending through the C7 spinal process was considered. Anterocollis was captured in the sagittal view with the patient in a sitting position without support. The external angle between the line perpendicular to the ground and an imaginary line drawn from the C7 spinal process and the tragus of the ear was measured [4–6]. This assessment took place for about 5 min for each patient. The minimal detectable change in the WG measurement was 5°(Fig. 1). The WG measurements and the photographs were obtained by a trained rater at each center at the same time for each patient.

3. Statistical analyses

Descriptive statistics included frequency tables and calculation of means and standard deviation. Non-normality of continuous variables was checked by both visual inspection of distribution and Kolmogorov-Smirnov test.

To test the validity of the WG measurements, we compared them with the gold-standard software-based measurements. Two softwarebased measurements (gold standards) were used to compare the FTB measurements [9]: the upper camptocormia method and the malleolus method. For the LTB and the FNB measurement, we considered the software-based perpendicular method as the gold standard [10,11].

To assess the validity of the WG, we performed the following analyses: 1) Bland-Altman mean differences and 95% limits of agreement [21]; 2) intraclass correlation coefficient (ICC) and standard error of measurement [22]; and 3) Cohen's kappa.

Bland & Altman plots were used to investigate the existence of any systematic difference between WG measurements and the softwarebased gold standards and to compute 95% limits of agreement for each comparison. The 95% limits of agreement were calculated as [mean of the differences \pm (1.96*SD)], in which SD is the standard deviation of mean of the differences [21]. Mean differences are the average difference between the gold standard and WG, while the limits of agreement are the random error or variation between instruments.

A one-way random-effects model ICC(1,1) with 95% confidence interval was calculated to investigate agreement between pairs of observations (WG and gold standards) [22].

The standard error of measurement was calculated for each measurement modality. This was calculated as described by Atkinson and Nevill [23] as standard error of measurement = $SD\sqrt{(1-ICC)}$, in which SD is the standard deviation. The resulting value of standard error of measurement is expressed in degrees (the highest the worst).

From the dataset, we calculated the average and frequency of underestimation, overestimation, and perfect estimation of WG measures compared to the gold standards. We calculated Cohen's kappa [24], the true positive, true negative, false positive, false negative, sensitivity, specificity, positive and negative predictive value [25] for diagnosis of camptocormia, Pisa syndrome, and anterocollis using the softwarebased measurements as gold standard [25].

3.1. Interobserver reliability

A two-way random effect model (2,k) ICC was used to analyze interobserver reliability between the two raters (CAA, MZ) of the continuous variables (degrees) calculated for the FTB, LTB, and FNB using the two measurement methods (WG and software-based) in a sample of 10 patients [22]. Cohen's kappa was used to calculate the interobserver reliability between the two raters (CAA, MZ) of dichotomous variables for the diagnosis of camptocormia, Pisa syndrome, and anterocollis using the two measurement methods (WG and software-based) in a sample of 10 patients. Statistical analysis was carried out using the SPSS package (version 20, IBM-SPSS, Armonk, NY, USA).



Fig. 1. Upper panel. Software-based methods (gold standards): the upper method, the malleolus method, and the perpendicular methods (from left to right). Lower panel. Measurement with the WG for upper and lower FTB, LTB, and FNB. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

E.	Patients with one	e or more PA		Total of	Isolated PA			Combined PA			
	FTB	LTB	FNB	PA	FTB/LTB/FNB			FTB + LTB	FTB + FNB	LTF + FNB	FTB + LTB + FNB
No. of patients 2 Age, mean (SD), y 7	215 73.50 (8.1)	88 73.23 (7)	61 70.97 (7.9)	283	146 73.7 (8.1)	36 72.67 (7.07)	25 69.92 (5.93)	40 74.1 (7.1)	24 71.58 (9.96)	7 71.3 (8.1)	5 72.8 (5.9)
Gender, no. (%), Male 1	134 (62.3)	51 (58)	43 (70.5)		88 (60.3)	19 (52.8)	14 (56)	22 (55)	19 (79.2)	5 (71.4)	5 (100)
Female 8	81 (37.7)	37 (42)	18 (29.5)		58 (39.7)	17 (47.2)	11 (44)	18 (45)	5 (20.8)	2 (28.6)	0 (0)
Body-mass index, mean (SD)	25.41 (3.6)	25.81 (4.3)	26.3 (3.5)		25.2 (3.6)	26.1 (4.33)	26.2 (2.9)	25.4 (4)	26.4 (2.8)	26.5 (6.7)	26.5 (3.6)
Age at PD onset, mean (SD), y 6	55.05 (9.8)	62.69 (10.5)	61.3 (10.3)		66.2 (9.3)	63.7 (9.5)	60.9 (10.5)	62.9 (11.3)	62.8 (9.9)	57.8 (13.1)	60 (7.4)
Disease Duration, mean (SD), y 8	8.02 (5.9)	10.22 (7.1)	9.5 (7.6)		6.9 (4.7)	8.17 (5.2)	8.6 (8.8)	11.2(8.1)	8.7 (5.7)	13.4 (8.1)	12.8 (7.9)
Modified H&Y stage, mean (SD) 2 Modified H&Y stage, no (%)	2.62 (0.8)	3.09 (0.9)	2.5 (0.9)		2.4 (0.7)	2.9 (0.9)	2.4 (0.9)	3.2 (0.8)	2.3 (0.9)	3.4 (0.8)	3 (1)
1 3	10 (4.7)	(1.1)	5 (8.1)		7 (4.7)	1 (2.8)	2 (8)	0 (0)	3 (12.4)	0 (0)	0 (0)
1.5 1	10 (4.7)	1 (1.1)	1 (1.6)		8 (5.5)	0 (0)	0 (0)	1 (2.5)	1 (4.2)	0 (0)	0 (0)
2	76 (35.3)	25 (28.4)	30 (49.2)		54 (37)	14 (38.9)	15 (60)	8 (20)	12 (50)	1(14.3)	2 (40)
2.5	21 (9.8)	2 (2.3)	2 (3.3)		20 (13.7)	1 (2.8)	2 (8)	1 (2.5)	0 (0)	0 (0)	0 (0)
3	60 (27.9)	21 (23.9)	9 (14.8)		43 (29.5)	7 (19.4)	1 (4)	11 (27.5)	5 (21)	2 (28.6)	1(20)
4-5 3	38 (17.6)	38 (43.2)	14 (23.0)		14 (9.6)	13 (36.1)	5 (20)	19 (47.5)	3 (12.4)	4 (57.1)	2 (40)
UPDRS Total score on state, mean (SD) 5	56.9 (24.6)	58.02 (27.7)	62.8 (29.1)		50.5 (20.7)	43.8 (20.2)	49.8 (22)	67 (23.4)	72.5 (24.3)	58.8 (27.2)	86.8 (54.2)
I 7	7.7 (6.4)	7.33 (6.5)	8.8 (6.7)		6.3 (5.8)	4.5 (3.9)	6.3 (6.2)	9.4 (6.9)	11.4 (5.6)	4.1 (3)	15.2(8.8)
П 1	15.3 (7.9)	17.7 (8.9)	15.6 (9.3)		13.3 (6.6)	13.3 (7.9)	11.6 (6.8)	20.7 (7.2)	17.3 (8.8)	18.8 (9.7)	22.8 (15.3)
III 3	31.8 (13.4)	30.3 (14)	36 (15.7)		29.2 (11.8)	23.2 (8.9)	30.4 (12.2)	34.2 (12.7)	40.5 (15.1)	33 (15.2)	46.4(26.5)
IV 2	2 (2.93)	2.7 (3.7)	2.1 (2.8)		1.7 (2.6)	2.8 (3.8)	1.4 (2)	2.6 (3.6)	2.5 (2.7)	2.8 (3.2)	2.4 (5.4)
Dominant Phenotype, no (%)											
Bradykinetic/Rigid type 1	106 (49.3)	47 (53.4)	26 (42.6)		68 (46.6)	15 (41.7)	8 (32.0)	24 (60)	10 (41.7)	4 (57.1)	4 (80)
Tremor type 3	33 (15.4)	20 (22.7)	18 (29.5)		21 (14.4)	11 (30.5)	11 (44.0)	7 (17.5)	5 (20.8)	2 (28.6)	0 (0)
Mixed Type 7	76 (35.3)	21 (23.9)	17 (27.9)		57 (39.0)	10 (27.8)	6 (24.0)	9 (22.5)	9 (37.5)	1(14.3)	1(20)
Laterality of PD Symptoms onset, n (%)											
Right 1	107 (49.7)	46 (52.3)	31 (50.8)		72 (49.3)	20 (55.6)	15 (60.0)	20 (50)	10 (41.7)	1 (14.3)	5 (100)
Left 7	78 (36.3)	30(34.1)	20 (32.8)		55 (37.7)	12 (33.3)	7 (28.0)	14 (35)	9 (37.5)	4 (57.1)	0 (0)
Bilateral 3	30 (14)	12 (13.6)	10 (16.4)		19 (13.0)	4 (11.1)	3 (12.0)	6 (15)	5 (20.8)	2 (28.6)	0 (0)
1-dopa equivalent daily dose, mg, mean 6 (SD)	632.27 (349.54)	666.43 (339.76)	581.13 (330.23)		601.07 (317.96)	590.09 (246.67)	481.36 (267.82)	723.97 (417.29)) 623.81 (394.85)	598.64 (176.53)	850.60 (312.66)

Abbreviations: SD = standard deviation; PD = Parkinson's Disease; PA = Postural Abnormalities including forward trunk bending (FTB), lateral trunk bending (LTB), and forward neck bending (FNB); Isolated PA = patients with only FTB or LTB or FNB; Combined PA = patients with one or more PA; y = years; H&Y = Hoehn and Yahr; UPDRS = Unified Parkinson's Disease Rating Scale; The total number of PA is 283, i.e., the sum of isolated and combined PA. l

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4. Results

4.1. Clinical features of PD patients

A total of 283 patients with PD met the eligibility criteria and entered the study; 207 had isolated postural abnormalities and 76 had combined postural abnormalities (Table 1).

In the sample of patients with one or more postural abnormalities, 215 (75.9%) presented FTB (175 upper type, 27 lower type, and 13 both types), 88 (31.1%) presented LTB, and 61 (21.5%) presented FNB. Upper FTB ranged from 21.3° to 72.3° (WG mean and SD of 41.9 \pm 8.9°; upper method 44.8 \pm 8.6°). Lower FTB ranged from 5° to 60° (WG 25.2 \pm 12.4°; malleolus method 28.9 \pm 10.6°). Coexisting upper/lower subtype ranged from 20° to 60°(WG 45.8 \pm 11.7° upper method 40.7 \pm 10.1°; WG 28.1 \pm 8.8° malleolus method 31.7 \pm 9.2°). LTB ranged from 4° to 45° (WG 11.7 \pm 7.4°; perpendicular method 11.3 \pm 7.4°). FNB ranged from 15° to 106.2° (WG 58.7 \pm 20.4°; perpendicular method 62.5 \pm 20°).

4.2. Discrepancy of the estimated measures between the software-based methods and the wall goniometer

Based on ICC, we found good agreement between the WG and the gold standards for upper and lower FTB. Bland-Altman plots showed a bias between methods, with a mean difference of -2.4° for the upper and of -7.4° for the lower type (Table 2). The limits of agreement were from 9.5° to -14.3° for the upper type and from 2.8° to -17.7° for the lower type (Supplementary Figs. 1A and B).

We found excellent agreement between the WG and the gold standards for LTB (ICC 0.96) and FNB (ICC 0.98). Bland-Altman plots showed a clinically acceptable bias between methods, with a mean difference from 0.4° to -3.9° . The limits of agreement ranged from 6.3° to -5.4° for the LTB and from 5.6° to -13.3° for the FNB, both relatively small and at the limit of clinical importance (Table 2 and Supplementary Figs. 1C and D). The standard error of measurement ranged from 0.6° to 2.3° and was the highest (worst) for the WG compared to the software-based upper and lower method.

When we compared with the gold standard for upper FTB, the WG overestimated the measurement by 5.1° (n = 57, 30.3% of cases), underestimated it by -6.1° (n = 121, 64.4% of cases), and estimated it perfectly in 10 cases (5.3%). For lower FTB, the WG overestimated the measurement by 3.5° (n = 4, 10% of cases) and underestimated it by -8.7° (n = 36, 90% of cases). For LTB, the WG overestimated the measurement by 2.9° (n = 40, 45.4% of cases), underestimated it by -2.5° (n = 31, 35.2% of cases), and estimated it perfectly in 17 (19.4%) cases. For FNB, the WG overestimated the measurement by 2.5° (n = 9, 14.7% of cases), underestimated it by -6.2° (n = 44, 72.1% of cases), and estimated it perfectly in 8 (13.2%) cases (Fig. 1).

4.3. Interobserver reliability

Interobserver reliability between the two raters in the use of WG and software-based measurement to measure FTB, LTB, and FNB was

Table 3

Interobserver reliability	for wall goniom	eter and software-l	based measurement
of FTB, LTB, and FNB.			

Measurement ^a	ICC	95% CI
Wall goniometer		
Upper FTB	0.80	0.18-0.95
Lower FTB	0.86	0.45-0.97
LTB	0.91	0.65-0.98
FNB	0.95	0.80-0.99
Software-based measurements		
Upper FTB	0.91	0.64-0.98
Lower FTB	0.99	0.98-0.99
LTB	0.96	0.84-0.99
FNB	0.99	0.99–1

Abbreviations: ICC, Intraclass correlation coefficient; CI, confidence interval with lower and upper bound); FTB, Forward Trunk Bending; LTB, Lateral Trunk Bending; FNB, Forward Neck Bending.

^a The measurements refer to continuous variables (degree of bending).

good to excellent (ICC 0.80 to 0.99) (Table 3). Interobserver reliability between the two raters in the use of the WG and software-based measurement for the diagnosis of camptocormia, Pisa syndrome, and anterocollis was fair to very good (Cohen's kappa from 0.40 to 1).

4.4. Discrepancy in the diagnosis of postural abnormalities between the software-based method and the wall goniometer

The WG showed moderate to good agreement (Cohen's k) with the software-based methods in the diagnosis of postural abnormalities. The agreement between the WG measurement and the perpendicular method was highest for FNB. Sensitivity was highest for detecting Pisa syndrome (100%) and anterocollis (95.74%) and lower for detecting upper (76.67%) and lower camptocormia (63.64%). Specificity was highest for detecting upper (89.80%) and lower (100%) camptocormia but lower for detecting Pisa syndrome (59.57%) and anterocollis (71.43%) (Table 4).

5. Discussion

The aim of this study was to investigate the validity of the WG to measure postural alterations in patients with PD. Software-based measurements represent the gold standard to quantify the degree of postural abnormalities [9] but may be time-consuming and not always feasible in clinical practice. Taking pictures, transferring them to a laptop, and measuring angles take time away from patient examination, education, and illness management. These drawbacks could be overcome with use of the WG, a simple and quick instrument for the clinical screening of postural abnormalities in PD patients [4–6]. An easy, quick, and validated method to assess postural abnormalities can improve both the clinical assessments and the translation of research results into practice.

Our results showed good agreement between methods when measuring upper and lower FTB. Although the standard errors of

Table 2

Comparison of wall goniometer measurements and gold standards (software-based) measurement in PD patients: ICC, Bland-Altman 95% LOA, and SEM.

Measurement	No. of patients	ICC	95% CI	Mean difference \pm 95% LOA (°)	SEM (°)
Comparisons between the wall goniometer and gold standards ^a Wall goniometer upper FTB vs. software-based upper FTB ^a	188	0.85	(0.80; 0.89)	$-2.4^{\circ} \pm 11.9^{\circ}$	2.3°
Wall goniometer lower FTB vs. software-based malleolus FTB ^a	40	0.80	(0.63; 0.89)	$-7.4^{\circ} \pm 10.3^{\circ}$	2.3°
Wall goniometer LTB vs. software-based perpendicular LTB ^a	88	0.96	(0.93; 0.97)	$0.4^{\circ} \pm 5.8^{\circ}$	0.6°
Wall goniometer FNB vs. software-based perpendicular \ensuremath{FNB}^a	61	0.98	(0.96; 0.98)	$-3.9^{\circ} \pm 9.4^{\circ}$	0.7°

Abbreviations: ICC, Intraclass correlation coefficient; CI, confidence interval with lower and upper bound); LOA, limits of agreement of Bland-Altman; SEM, standard error of measurement.

^a Gold Standards: are the upper, malleolus, and perpendicular method; FTB, forward trunk bending; LTB, lateral trunk bending; FNB, forward neck bending.

Table 4

Contingency table of frequencies for PA determination using software-based (gold standard) and wall goniometer.

Comparisons between the wall goniometer and software-based gold standards			Totals	Cohen's kappa	Sensitivity (%), CI 95%	Specificity (%), CI 95%	Positive predictive value	Negative predictive value	
		Upper FTB Me	thod	_					
		CC Yes $\geq 45^{\circ}$	CC No $< 45^{\circ}$	_					
FTB upper wall	CC Yes $\geq 45^{\circ}$	69	10	79	0.668	76.67 (66.57; 84.94)	89.80 (82.03; 95.00)	87.34	80.73
goniometer	CC No $< 45^{\circ}$	21	88	109					
	Total	90	98	188					
		Malleolus met	hod						
		CC Yes $\geq 30^{\circ}$	CC No $< 30^{\circ}$						
FTB lower wall	CC Yes $\geq 30^{\circ}$	14	0	14	0.612	63.64 (40.66; 82.80)	100 (81.47; 100)	100	69.23
goniometer	CC No $< 30^{\circ}$	8	18	26					
	Total	22	18	40					
		Perpendicular	method						
		PS Yes $\geq 10^{\circ}$	PS No $< 10^{\circ}$						
LTB wall goniometer	PS Yes $\geq 10^{\circ}$	41	19	60	0.579	100 (91.40; 100)	59.57 (44.27; 73.63)	68.33	100
	PS No $< 10^{\circ}$	0	28	28					
	Total	41	47	88					
	Perpendicular method								
		AC Yes $\geq 45^{\circ}$	AC No $< 45^{\circ}$						
FNB wall goniometer	AC Yes $\geq 45^{\circ}$	45	4	49	0.707	95.74 (85.46; 99.48)	71.43 (41.90; 91.61)	91.84	83.33
	AC No $< 45^{\circ}$	2	10	12					
	Total	47	14	61					

Abbreviations: PA = Postural Abnormalities including forward trunk bending (FTB), lateral trunk bending (LTB), and forward neck bending (FNB). CC, camptocormia; PS, Pisa syndrome; AC, anterocollis according to diagnostic criteria. Upper FTB in patients with bending at the thoracic fulcrum (C7 to T12-L1); lower FTB in patients with bending at the lumbar fulcrum (L1-sacrum, hip flexion).

measurements were similar, we found the greatest (worst) mean difference (-7.4°) when comparing the WG to the software-based malleolus FTB measure. This discrepancy could have resulted from the use of different bone landmarks to draw the angle of FTB (Fig. 1). These differences are markedly reduced when the WG and the software-based perpendicular method are compared. The perpendicular method takes as zero reference the vertical axis that intersects with the fulcrum of bending, which is the same as that for visual measurement performed with the WG (Fig. 1). Indeed, we found excellent agreement between the WG and the software-based perpendicular method to measure LTB and FNB, with relatively small mean differences and at the limit of clinical importance, i.e., below 10° [26].

Cohen's kappa indicated moderate to good agreement between the WG and software-based measurements for the diagnosis of camptocormia, Pisa syndrome, and anterocollis. Sensitivity was high, but specificity was low, for the diagnosis of anterocollis and Pisa syndrome. In contrast, sensitivity was lower but specificity was higher for the diagnosis of upper and lower camptocormia. The lowest sensitivity was around 63.64%, which may be considered sufficient to detect lower type camptocormia, accounting for 14.13% of patients in our sample. These results suggest that the WG may be appropriate for screening Pisa syndrome and anterocollis but less suitable to detect patients with camptocormia.

In our sample, the WG tended to underestimate upper and lower FTB and FNB (by -6.1° , -8.7° , and -6.2° , respectively) but overestimated LTB (by 2.9°, on average).

The under/overestimation may stem from lack of precision of the raters in taking measurement, the use of different bone landmarks to measure postural abnormalities, and the anatomical irregularities of the patients' back. For instance, the upper method may display a narrower angle than the WG when there is a combined form of camptocormia (upper + lower type) or when spinal kyphosis under the fulcrum is increased; however, the upper method may also display a wider angle than the WG when spinal lordosis under the fulcrum is increased (Supplementary Fig. 2).

Underestimation of the WG assessment is likely due to the use of different bone landmarks during the visual assessment of the patient when compared with the angle drawn on the photograph. The latter measures more degrees since it takes into account also the knee flexion (malleolus method) [9].

Overall, the results showed that 1) the WG is valid tool to detect postural abnormalities in PD patients; 2) the lower agreement between WG and software-based measurements when assessing the FTB reflects in a low sensibility in detecting camptocormia; and 3) Pisa syndrome and anterocollis can be diagnosed by the WG, while a correction of about 10° should be added when evaluating camptocormia.

Several important aspects and limitations need to be considered when evaluating postural abnormalities with the WG: 1) biases are possible, especially the risk of underestimating postural abnormality measures in patients with camptocormia. These biases could not be attributed to inter-rater variability, as our analysis indicated good to excellent interobserver reliability (although the confidence interval was quite large for the upper FTB due to the small sample); 2) WG does not consider possible lower limb compensations (i.e., hip and knee bending) which may further worsen postural abnormalities; 3) software-based gold standards have been identified for camptocormia [9] but not for Pisa syndrome and anterocollis, for which we reasonably considered the software-based perpendicular method [9–11] as the gold standard; 4) patients should be appropriately exposed (without clothes) to more accurately detect minimal deviations of the trunk/neck; 5) because postural abnormality severity may change over time, even within a short period, patients should be evaluated while standing and walking and during other dynamic conditions. Finally, 6) the definition of each postural abnormality should be always contemplated [1], i.e. camptocormia resolves in supine position or when leaning against a wall, and the WG has only an additive value to quantify the degree of bending.

In conclusion, the WG is an inexpensive, quick, and valid instrument to detect trunk deformities, as well as an aid in supporting management decision to adjust pharmacological therapy and physiotherapy [11,12]. Future studies are desirable to evaluate changes in postural abnormality severity in prospective interventional trials or historical cohort studies.

Funding

This work was supported by the Brain Research Foundation Verona O.N.L.U.S (Grant number 2017).

Author roles

MT and CG: drafting and revising the manuscript, study concept and design, acquisition, analysis and interpretation of data, and study execution; MG, CAA, RL, EZ, RC, AF: drafting/revising the manuscript, study concept and design, interpretation of data, study execution. MC, EA, MGC, LB, MO, RT, CB, PP, PM, SM, SG, LV, FS, MC, MZ, LL: drafting/revising the manuscript, acquisition and interpretation of data, study execution.

Declaration of competing interest

None.

Acknowledgments

We wish to thank Salvatore Bonvegna (University of Messina, Messina, Italy) for his assistance with data collection.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.parkreldis.2019.10.024.

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