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Automated and objective measures of gait dynamics in camptocormia Parkinson's Disease subthalamic deep brain stimulation

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ABSTRACT

Objective: Axial motor features are common in Parkinson's disease (PD). These include gait impairment and postural abnormalities, such as camptocormia. The response of these symptoms to deep brain stimulation (DBS) is variable and difficult to assess objectively. For the first time, this study analyzes the treatment outcomes of two PD patients with camptocormia that underwent bilateral subthalamic nucleus (STN)-DBS evaluated with disruptive technologies.

Patients and methods: Two patients with PD and camptocormia who underwent STN-DBS were included. Gait parameters were quantitatively assessed before and after surgery by using the NeuroKinect system and the camptocormia angle was measured using the camptoapp.

Results: After surgery, patient 1 improved 29 points in the UPDRS-III. His camptocormia angle was 68° before and 38° after surgery. Arm and knee angular amplitudes (117.32 ± 7.47 vs 134.77 ± 2.70°; 144.51 ± 7.47 vs 169.08 ± 3.27°) and arm swing (3.59 ± 2.66 vs 5.40 ± 1.76 cm) improved when compared with his preoperative measurements. Patient 2 improved 22 points in the UPDRS-III after surgery. Her camptocormia mostly resolved (47° before to 9° after surgery). Gait analysis revealed improvement of stride length (0.29 ± 0.03 vs 0.35 ± 0.03 m), stride width (18.25 ± 1.16 vs 17.9 ± 0.84 cm), step velocity (0.91 ± 0.57 vs 1.33 ± 0.48 m/s), arm swing (4.51 ± 1.01 vs 7.38 ± 2.71 cm) and arm and hip angular amplitudes (131.57 ± 2.45° vs 137.75 ± 3.18; 100.51 ± 1.56 vs 102.18 ± 1.77°) compared with her preoperative results.

Conclusion: The gait parameters and camptocormia of both patients objectively improved after surgery, as assessed by the two quantitative measurement systems. STN-DBS might have a beneficial effect on controlling axial posturing and gait, being a potential surgical treatment for camptocormia in patients with PD. However, further studies are needed to derive adequate selection criteria for this patient population.

1. Introduction

Axial motor features—including gait impairment and postural abnormalities—are common in later stages of Parkinson's disease (PD) with notable clinical importance owing to reduced mobility, loss of independence and recurrent falls [1].

Camptocormia (derived from the Greek words "kamptein" meaning to bend and "kormos" meaning trunk) is an axial postural deformity characterized by involuntary forward-flexed posture of the thoracolumbar spine, appearing in standing position, increasing during walking and abating with supine position [2,3]. Camptocormia occurs in nearly 7–10% of PD patients and is associated with more severe disease, longer disease duration, dementia, and higher levodopa doses [4–6]. It is widely recognized that postural deformities may cause and

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Fig. 1. Pre and postsurgical evaluation of camptocormia angle using the camptoapp.
A. Lateral view photograph before surgery with a calculated angle of 68°.
B. Lateral view photograph 12 months after surgery with a bending angle of 38°.

worsen gait impairments, postural imbalance and functional disability, independently of other motor symptoms [1,7–9].

The response of axial symptoms to deep brain stimulation (DBS) depends on a variety of factors, including patient characteristics, the specific target chosen for DBS, the exact positioning of the electrode within the nucleus, and the stimulation parameters [1]. Chieng LO and colleagues [10] have demonstrated that DBS could be a promising treatment option in patients with PD and camptocormia. However, to date, experience of comparative and objective evaluations of posture and gait performance is limited and our understanding of axial motor symptoms in PD is hindered by the huge variability in methodology across studies. Hence, the literature suggests future focus on more objective assessments, for example, by automated gait and posture analysis.

The aim of this work is precisely to report for the first time an objective assessment approach in PD with camptocormia that underwent bilateral subthalamic nucleus (STN)-DBS with preoperative and postoperative assessment of the Unified Parkinson Disease Rating Scale part III (UPDRS-III), automated measure of gait parameters and bending angle assessment, which we propose as a specific and quantifiable way to assess gait and camptocormia.

2. Materials and methods

2.1. Patient consent

Informed written consent was obtained from all patients. This study was approved by the Ethics Committee of Centro Hospitalar Universitário de São João/Faculdade de Medicina da Universidade do Porto.

2.2. Patient selection

Two patients with PD and camptocormia (defined by forward flexion of 45° or more [4,11], which disappears with recumbent position) who underwent STN-DBS at our Movement Disorders Unit were evaluated by movement disorders specialist, physicist and biomedical engineers. The diagnosis of PD was made according to the UK Parkinson's Disease Society Brain Bank criteria [12].

2.3. Surgical intervention

The target chosen was the STN. To choose the stimulation parameters which best alleviate PD symptoms without side effects, neurologists imposed a passive wrist flexion movement and qualitatively described the perceived decrease in rigidity in a discrete scale (0, 40, 50, 60, 70 or 80% of rigidity improvement). This subjective assessment was compared with the quantitative and objective iHandU® system [13] which is a novel, comfortable and wireless system that classifies rigidity improvement during passive wrist flexion, performing an intra-operative processing providing real-time feedback.

2.4. Pre-operative and post-operative patient assessment

UPDRS-III, bending angle and gait parameters of interest (stride length, stride width, step velocity, double support time, arm swing, knee and hip angular amplitudes) were prospectively collected one day before surgery, during the week after surgery and at 8–12 months after electrode implantation. Gait parameters were assessed using the NeuroKinect® system [14]. Color, depth, infrared, and skeleton data (at a rate of 30 frames per second) of a walking task were collected: 4 m of monitored walking trajectory, repeated 5 times, and three times 5 s of lateral posture. The NeuroKinect system is composed of a single RGB-D camera, such as the Microsoft Kinect, to seamlessly track the 3-D position of 25 body joints, without interfering with the Hospital environment, the clinical routine, not requiring any scenario modification or calibration. The Kinect system has been widely used for the gait assessment, with multiple studies validating its robustness and accuracy [15–20]. An automatic application described by Rodrigues J and colleagues [21] was used to perform gait spatiotemporal feature analysis. We collected photos and video freeze frames to measure total camptocormia angle and posteriorly we followed the recommendations of the consensus presented by Margraf NG and colleagues [22] using the camptoapp.

3. Results

One 61-year-old male (patient 1) and one 70-year-old female (patient 2) were included. The duration of PD was 12 and 9 years, respectively, and the duration of camptocormia was 1.5 and 9 years,

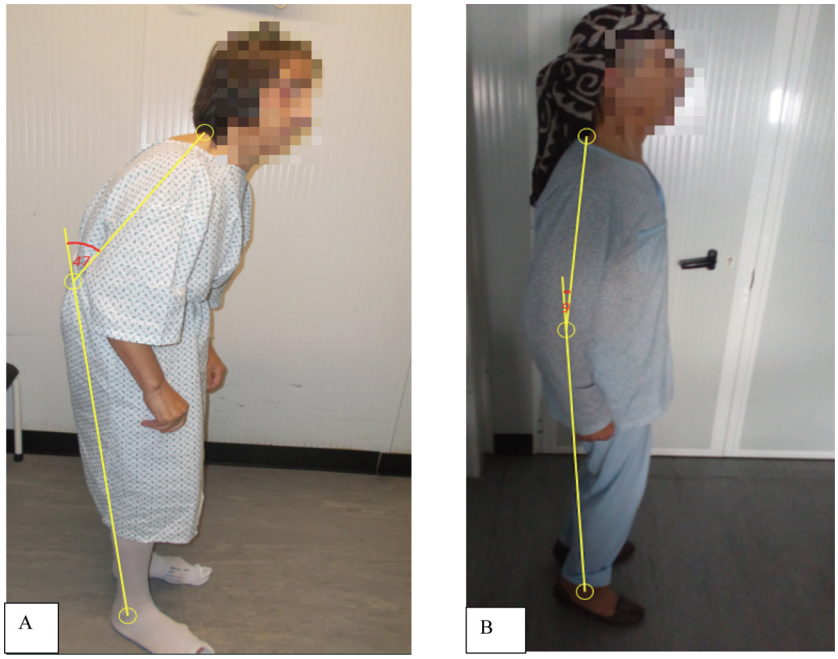

Fig. 2. Pre and postsurgical evaluation of camptocormia forward angle using the camptoapp.
A. Lateral view photograph before surgery with a calculated angle of 47°.
B. Lateral view photograph 8 months after surgery with a calculated angle of 9°.

respectively. Patient 1 had a prior history of dorsal kyphosis and scoliosis with juvenile onset. At the age of 49, he noticed right hand tremor without interference on the activities of daily living (ADL). The tremor resolved with levodopa treatment. Eleven years later, he progressed to an akinetic-rigid form of PD with exuberant camptocormia and lower back pain, despite a fixed dorsal kyphosis and scoliosis (Fig. 1, A). He scored 45 points in UPDRS-III, mainly due to rigidity, bradykinesia and posture, with limited ADL. He was on 1450 mg L-dopa equivalent daily dose and developed motor fluctuations. A L-dopa challenge (800 mg) was performed with an improvement of 47% of UPDRS part III (medication OFF - 51; medication ON – 27) and a slight decrease of camptocormia angle (not shown).

Patient 2 had a history of hypertension and type 2 diabetes. At the age of 61, she started bending forward while developing dysarthria and tremor and rigidity predominantly on the left side. There was a marked improvement when she started levodopa. Five years later, she presented with bradykinesia, mild dysarthria and worsening of her camptocormia angle (Fig. 2, A). Nine years later, dysphagia and occasional falls were prominent symptoms, with a score of 35 points in the UPDRS-III. She was medicated with 1040 mg L-dopa equivalent daily dose without improvement of classical motor symptoms and remained dependent in some ADL. The patient underwent a L-dopa challenge (500 mg) with an improvement of 57% of UPDRS-III (medication OFF - 50; medication ON – 20) and a slight decrease of camptocormia angle (not shown).

Electromyography of bilateral dorsal and lumbar paraspinal muscles did not reveal any myopathic or neurogenic changes on both patients. Our patients fulfilled the inclusion criteria for functional surgery with no contraindication demonstrated in the psychiatric or neuropsychological evaluations. Both patients underwent STN-DBS and there was a good agreement between the medical and the iHandU label.

After surgery, patient 1 had an improvement of 29 points in the UPDRS -III (medication OFF/stimulation ON) compared with the L-dopa challenge (medication OFF). One year later he had an improvement of 26 points in the UPDRS-III (medication ON/stimulation ON) with a decrease of 83% of L-dopa equivalent daily dose (Table 1). An improvement was seen on rigidity and bradykinesia and also on camptocormia angle (68° before surgery to 38° after surgery) (Fig. 1, B). Posteriorly, we also calculated camptocormia angle [22] through pre and postoperative thoracolumbar x-ray (lateral view) of patient 1 and compared with photographs: no significant differences were found in camptocormia angle. There was an improvement of arm and knee angular amplitudes (117.32 ± 7.47 vs 134.77 ± 2.70°; 144.51 ± 7.47 vs 169.08 ± 3.27°) and arm swing (3.59 ± 2.66 vs 5.40 ± 1.76 cm) (Table 1).

Patient 2 had an improvement of 22 points in the UPDRS-III (medication OFF/stimulation ON) compared with the L-dopa challenge (medication OFF) and a decrease of 52% in L-dopa equivalent daily dose. Eight months later, the patient's camptocormia had mostly resolved (47° before surgery to 9° after surgery) (Fig. 2, B) with an improvement of 26 points in the UPDRS-III (medication ON/stimulation ON) (Fig. 2). Gait analysis revealed improvement of stride length (0.29 ± 0.03 vs 0.35 ± 0.03 m), stride width (18.25 ± 1.16 vs 17.9 ± 0.84 cm), step velocity (0.91 ± 0.57 vs 1.33 ± 0.48 m/s), arm swing (4.51 ± 1.01 vs 7.38 ± 2.71 cm) and arm and hip angular amplitudes (131.57 ± 2.45° vs 137.75 ± 3.18; 100.51 ± 1.56 vs 102.18 ± 1.77°) compared with her preoperative results (Table 1).

4. Discussion

Two cases of PD with camptocormia and gait impairment that underwent STN-DBS presented above displayed a clinically improvement of axial motor symptoms, total bending angle and independence in ADL.

Tramonti C. and colleagues [8] have evaluated gait dynamics in patients with PD and severe postural deformities (including 9 camptocormia patients), demonstrating decreased walking velocity, stride and step length in these patients compared to controls, with reduced functional ability; Kinematic data also revealed marked reduction in range of movements and the main differences were pronounced in hip and knee joints in the camptocormia group. In our cases (Table 1), patient 1 had an improvement mainly in arm and in knee angular amplitudes (117.32 ± 7.47 vs 134.77 ± 2.70°; 144.51 ± 7.47 vs 169.08 ± 3.27°), with improvement of 29 points in the UPDRS-III and 30° on camptocormia angle; patient 2 had a global improvement of her gait performance after surgery – stride length, stride width, step velocity, arm swing, and arm and hip angular amplitudes (0.29 ± 0.03 vs

Table 1
Pre and postoperative results regarding UPDRS-III, gait spatiotemporal parameters[a] and total camptocormia angle.

	Patient 1			Patient 2		
	Pre-Op[b]	Post-Op[c]	Follow-Up[d]	Pre-Op[b]	Post-Op[c]	Follow-Up[e]
UPDRS part III	45	16	25	35	28	24
Stride length (m)	0.25 ± 0.03	0.25 ± 0.03	0.29 ± 0.06	0.29 ± 0.03	0.23 ± 0.05	0.35 ± 0.03
Stride width(cm)	13.03 ± 0.25	13.05 ± 0.29	13.88 ± 1.17	18.25 ± 1.16	18.63 ± 0.83	17.9 ± 0.84
Step velocity(m/s)	0.78 ± 0.51	0.91 ± 0.17	0.95 ± 0.31	0.91 ± 0.57	0.52 ± 0.94	1.33 ± 0.48
Double support time (s)	0.67 ± 0.17	0.68 ± 0.20	1.01 ± 0.65	0.79 ± 0.21	0.83 ± 0.23	0.70 ± 0.12
Arm swing (cm)	3.59 ± 2.66	3.39 ± 2.45	5.40 ± 1.76	4.51 ± 1.01	4.49 ± 2.48	7.38 ± 2.71
Arm angular amplitude (°)	117.32 ± 7.47	117.01 ± 1.65	134.77 ± 2.70	131.57 ± 2.45	140.81 ± 2.15	137.75 ± 3.18
Knee angular amplitude (°)	144.51 ± 7.47	144.50 ± 7.64	163.40 ± 2.18	162.89 ± 1.52	160.59 ± 3.78	153.22 ± 2.05
Hip angular amplitude (°)	109.42 ± 3.07	109.46 ± 2.99	99.68 ± 1.98	100.51 ± 1.56	101.95 ± 1.74	102.18 ± 1.77
Camptocormia Angle (°)	68	51.6	38	47	8.6	9

Note: In our center, STN-DBS is programmed within the first 24 h after surgery.
[a] Mean and standard deviation (5–10 steps evaluated).
[b] One day before surgery.
[c] During the week after surgery.
[d] 12 months follow-up.
[e] 8 months follow-up.

0.35 ± 0.03 m; 18.25 ± 1.16 vs 17.9 ± 0.84 cm; 0.91 ± 0.57 vs 1.33 ± 0.48 m/s; 4.51 ± 1.01 vs 7.38 ± 2.71 cm; 131.57 ± 2.45° vs 137.75 ± 3.18; 100.51 ± 1.56 vs 102.18 ± 1.77°, respectively), with improvement of 22 points in the UPDRS-III and camptocormia mostly resolved. Furthermore, patient 2 had better postural and gait outcomes than patient 1 after surgery. We speculate that patient 1's comorbidities (kyphosis and scoliosis), long duration of PD and an onset at young age may contribute to a poor axial motor response [23]. However, both patients referred an improvement in their ADL.

Although the beneficial effects of DBS on appendicular motor symptoms are well recognized, the response of axial disability to this intervention is more difficult to predict and measure quantitatively [1,7,8]. The degree of response to levodopa has been associated with axial motor outcomes (posture, balance and gait) following DBS [1,24]. Umemura and colleagues [25] showed in their study, involving 8 camptocormia patients, four times higher likelihood of improvement of postural changes in patients with a preoperative response to L-dopa. Our patients had a significant improvement of motor symptoms and camptocormia angle after L-dopa challenge, which may suggest that L-dopa response is a preoperative predictive factor of posture improvement in PD-associated camptocormia. The debate whether DBS can alleviate axial disability that is present preoperatively, and whether the response to L-dopa challenge correlates with the response to surgery remains unanswered [1,24–28]. Further larger studies are needed to elucidate the correlation between improvement of camptocormia during L-dopa challenge and the postoperative results using standardized measurements.

This study also highlights the advantages of using quantitative methods to support clinical studies. The NeuroKinect system only relies on a single inexpensive RGB-D camera and a portable computer. The system brings objectivity and provides 3D information from patients, without the need of attaching markers to different body parts, inside the clinical environment, without interfering with the clinical routine. The system is easy to mount and it is super comfortable for the patients. In addition, the system provides an automated report of the gait assessment, which gives neurologists an objective assessment of the gait pattern, surpassing the traditional visual inspection analysis.

However, some important limitations should be pointed out. To reduce the risk of bias, the clinical evaluations should have been videotaped before and after the surgery and given to a blinded rater. On the other hand, patients' camptocormia and gait dynamics have not been objectively quantified before and after L-dopa challenge. These data would be necessary to understand if there is a correlation between the test results and the improvements of axial symptoms after surgery.

5. Conclusion

Our work adds to previous reports of camptocormia improvement after DBS by including a quantitative assessment of motor axial features and gait parameters. Our findings suggest that STN-DBS might have objective beneficial effects on controlling axial posturing and gait. STN-DBS can be a potential surgical means for treating camptocormia in patients with PD. However, further studies need to be performed to confirm this conclusion and to select PD patients who are optimal candidates for STN-DBS.

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