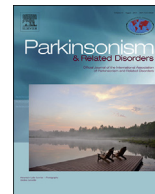




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## Short communication

## Interoceptive sensitivity in patients with cervical dystonia

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## ABSTRACT

**Introduction:** Interoceptive sensitivity (IS) is the ability to perceive sensations arising from the inner body. One method used to measure IS is the heartbeat detection task. The aim of this study was to investigate IS in patients with cervical dystonia (CD) and compare the results with those obtained in healthy controls (HC). We also sought possible correlations between IS and demographic, clinical and emotional features in CD. To evaluate the reliability of IS in a subgroup of CD patients and HC, we retested IS 4–6 months after the first evaluation. We also investigated whether dystonic posture affects IS values in HC.

**Methods:** Twenty CD patients and 20 HC were investigated. The heartbeat detection task was performed according to a standardized protocol. All the participants underwent a clinical, emotional and psychiatric evaluation.

**Results:** IS was lower in CD patients than in HC. The ROC curve analysis showed that an IS value of 0.52 differentiates CD patients from healthy controls. No correlations emerged in CD patients between IS and the demographic, clinical and emotional features. No differences were observed in either CD patients or healthy subjects when IS was retested 4–6 months after the first evaluation. When IS was tested in HC mimicking a dystonic posture, the results were similar to those obtained when they held their heads in a neutral position.

**Conclusions:** The study shows that IS is reduced in CD patients possibly due to an altered functional connection between basal ganglia and limbic circuit, including the insula.

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## 1. Introduction

Interoception is the ability to perceive sensations arising from the inner body and relies on visceral afferent feedback [1]. One technique used to investigate interoception is the heartbeat detection task [2], in which participants are asked to mentally count their heartbeats during rest while their heart rate is measured objectively. The level of concordance between one's heart rate and its subjective perception is considered to yield a stable measurement of interoceptive sensitivity (IS). Studies using functional magnetic resonance imaging on healthy subjects have reported that interoception is, similarly to emotional processes, mediated by the insula and limbic circuits [3]. Several authors have

hypothesized a relationship between IS and emotional processes [3,4] and have found that individuals with higher IS feel their own emotional experiences more intensely. By contrast, IS is lower in patients with depression than in healthy controls [5].

Cervical dystonia (CD) is a focal dystonia characterized by abnormal movements of the head [6], as well as by non-motor disturbances [7] that include depression, anxiety and emotional process abnormalities. IS may be reduced in CD patients owing to the presence of both emotional and psychiatric alterations.

The aim of our study was to investigate IS in patients with CD. In order to do so, we compared IS values in CD patients with those obtained in age-matched healthy controls. To evaluate the reproducibility and reliability of IS, we retested a subgroup of CD patients and healthy subjects 4–6 months after the first evaluation. Lastly, in order to investigate whether dystonic posture of the head per se affects IS values, we compared IS values in healthy subjects assessed in a neutral position with those obtained while they

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mimicked a dystonic posture.

## 2. Materials and methods

### 2.1. Study participants and clinical assessment

Twenty-six CD patients (6 males, mean age  $54.84 \pm 11.4$  years) and 20 healthy controls (HC) (9 males, mean age  $49.6 \pm 15.5$ ) were enrolled from the movement disorder outpatient clinic of the Department of Neurology and Psychiatry, Sapienza University of Rome. The diagnosis of CD was based on standard clinical criteria [6]. We collected information regarding the patients' demographic characteristics and disease duration in a face-to-face interview and by examining clinical records (Table 1). Exclusion criteria were any neurological abnormalities other than head tremor, concomitant known cardiac disorders [8], and clinically significant cognitive deficits as determined by a score  $<26$  on the Montreal Cognitive Assessment (MOCA). We only enrolled CD patients and healthy subjects without psychiatric disorders and excluded any CD patients or HC who were diagnosed with a psychiatric disorder during a full psychiatric evaluation.

The severity of CD was assessed according to the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS). Presence of pain was evaluated using the TWSTRS. The quality of life of CD patients was assessed by means of the Cervical Dystonia Impact Scale (CDIP-58).

Any patients being treated with botulinum toxin were evaluated about 4 months after the last botulinum toxin injection. Only three out of 20 CD patients studied were on clonazepam. All the participants signed a written informed consent form. The experimental procedure was approved by the institutional review board at Sapienza University of Rome and conducted in accordance with the Declaration of Helsinki.

### 2.2. Heartbeat detection task

The Heartbeat Detection Task was performed according to the protocol of Schandry [2], and the heart rate was recorded by means of a Polar wrist-worn, heart-rate monitor (model V 800). Participants were required to place the belt around their chest and to sit quietly and relax for 30 s; the belt was connected via bluetooth to the Polar wrist-worn, heart-rate monitor. To avoid any potential confounding factor due to heart disturbances, the baseline heart rate was recorded for 3 min before the start of the task. Participants were then asked to concentrate on their heartbeats and to count them silently. They were instructed not to take their pulse or attempt any other physical manoeuvre that might help them detect their heart rate. The patients were required to count their heartbeats three times for 25 s, 45 s and 65 s, with each counting phase being separated by a 30-s rest period. The order of the counting phases was randomized for all the participants in each group. An acoustic signal was used to inform the patients when to start and stop counting. After the 'stop' signal at the end of the phase, participants were asked to report how many heartbeats they had counted. Participants were not aware of either the duration of the counting phases or of how they performed. The accuracy of the patients' perception of their heartbeat was calculated as the mean score of three heartbeat perception intervals according to the following transformation:  $1/3 \sum [(1 - \text{recorded heartbeats} - \text{counted heartbeats}) / \text{recorded heartbeats}]$ . Using this formula, the IS score varies between 0 and 1, with higher scores indicating smaller differences between recorded and perceived heartbeats.

IS was measured again between 4 and 6 months after the first assessment in 10 patients with CD and in 10 healthy subjects. Lastly, IS was evaluated in 10 healthy subjects while they mimicked the

**Table 1**

Demographic and clinical data of CD patients studied.

Patients	Sex	Age (y)	Duration (y)	Age onset (y)	Other dystonia
1	F	52	10	42	no
2	F	42	6	36	no
3	F	57	20	37	no
4	F	45	2	43	AD and Cranial Dystonia
5	F	63	38	25	no
6	M	44	20	24	no
7	F	64	10	54	no
8	F	44	2	42	no
9	F	61	15	46	no
10	M	54	6	48	no
11	F	74	20	54	no
12	F	40	5	35	no
13	M	74	7	67	no
14	F	73	4	69	no
15	M	63	7	56	no
15	F	48	5	43	no
17	F	30	19	11	no
18	F	62	2	60	no
19	F	60	10	50	no
20	F	66	16	50	no

CD: cervical dystonia.

dystonic posture that is characteristic of CD patients.

### 2.3. Psychiatric and emotional assessment

All the participants underwent a full psychiatric evaluation that was performed by two experienced psychiatrists using the Structured Clinical Interview (SCID-I) for DSM-IV. The severity of depression was evaluated by means of the Beck Depression Inventory (BDI-II). The severity of anxiety was assessed by means of the Hamilton Anxiety Rating Scale (Ham-A). We used the Toronto Alexithymia Scale (TAS-20) to evaluate alexithymia. The presence and severity of apathy were assessed by means of the Apathy Evaluation Scale (AES). The grade of empathy was assessed by means of the Empathy Quotient (EQ).

### 2.4. Statistical analysis

The Mann-Whitney *U* test was used to evaluate differences between CD patients and healthy subjects. We also performed Mann-Whitney *U* test to assess whether the presence of neck pain might influence IS in CD patients. Spearman's correlation coefficient was used to identify any correlations between IS and the clinical, demographic and emotional assessment in CD patients. We used a ROC curve to identify the IS cut-off value that best discriminates patients from healthy subjects. We used the paired sample *t*-test to investigate any differences between the first and second IS assessments in patients and healthy subjects. The paired sample *t*-test was also used to evaluate any differences in IS performance between the neutral head position and dystonic head position assessments in healthy subjects.  $P < 0.05$  was chosen to indicate statistical significance.

## 3. Results

The psychiatric evaluation showed that 6 of the 26 CD patients enrolled had a psychiatric disorder (1 with adjustment disorder, 1 with generalized anxiety disorder and 4 with major depressive disorder); these six patients were excluded to ensure that psychiatric disturbances did not affect IS. No psychiatric disorders were found in the healthy controls. The 20 CD patients investigated displayed mild disease severity (TWSRS mean:  $15.3 \pm 6.94$ ) and a mild-to-moderate limitation in activities of daily living (CDIP-58

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