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Voluntary control of facial musculature in Parkinson's disease

Michelle Marneweck *, Geoff Hammond

School of Psychology, University of Western Australia, Crawley, WA, 6009, Australia

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ABSTRACT

Aside from being measured in the context of producing facial expressions of emotion, the ability to voluntarily control a range of facial muscles in Parkinson's disease (PD) has not been systematically measured. We used in three enrollment phases an adaptation of the Upper and Lower Face Apraxia test, a measure of the ability to make voluntary movements of the upper and lower face in PD patients and healthy controls. Errors were scored due to (1) pauses prior to movement initiation, (2) loss of individuation, (3) impoverished movement, (4) no movement at all, or (5) content errors (likened to ideational apraxia errors). The results show impaired voluntary control of facial musculature in most but not all with PD (with large effect sizes) which correlated positively and highly with disease severity. Errors by PD patients were predominantly due to impoverished movement and individuation loss whereas those made by controls were predominantly due to individuation loss. Patients committed more errors than controls due to impoverishment and no movement, with negligible differences between groups in other errors. In summary, similarly to spontaneous and voluntary emotional expressions, voluntary non-emotional facial movements are impoverished in PD; impoverishment of all movement types will likely contribute to the mask-like facial appearance that is seen with disease progression. These findings also illustrate the utility of an adapted Face Apraxia test as a practical and sensitive measure of voluntary facial musculature control in PD. The test can be used to supplement clinical observations and as a research tool.

1. Introduction

A "masked" expressionless face is an important clinical sign of Parkinson's disease (PD) [3,13,21]. In a now classic and influential review on facial expression, Rinn [21] proposed that the impression of a mask-like face in PD originates from reduced spontaneous emotional expression, with voluntary emotional expression remaining intact. Following Rinn's proposal, studies found that PD patients were impaired in spontaneous emotional expressions during conversation or in response to emotionally evocative stimuli [14,20,24–26]. These findings have informed our understanding of facial masking in PD as a reduced spontaneity in facial expression of emotion. However, in contrast to Rinn's proposal, PD patients have also been shown to be impaired in voluntarily expressing emotions in response to verbal command (e.g. "look happy") [5,6,12,16,24,25]. In these studies, responses have either been scored by blind raters using Likert scales, the facial action coding system (FACS) [9] which codes observable facial movements as "action units" or action unit combinations, or by digital imaging analyses [6] and kinematic techniques [16]. These findings that were in contrast to earlier propositions [21] spurred the more recent conclusion [6] that facial masking in PD is not limited to spontaneous facial expressions of emotion, but also involves voluntary facial expressions of emotion.

It is reasonable to suspect that non-emotional facial movement is also impaired in PD, thereby contributing with diminished emotional expressions to facial masking. Nevertheless, voluntary non-emotional facial movements remain less systematically explored than voluntary emotional facial movements in PD. Studies on non-emotional facial movement in PD have been limited to specific facial areas, with impairment evidenced in voluntary, spontaneous, and reflex blinking rate and amplitude [1,4,15] and amplitude of jaw and upper lip movement during speech [7]. The group of Simons et al. [24,25] is the only group that has measured, using the FACS, voluntarily imitating a limited set of non-emotional facial movements and found these movements to be impaired in PD. Although the FACS procedure is sensitive to compare action unit patterns with requested patterns, it is not always a practical tool available to clinicians and researchers; FACS certification requires extensive and costly training. There is only one item that assesses facial expressivity on the Movement Disorders Society-sponsored revision of the Unified Parkinson's Disease Rating Scale (MDS-UPDRS) [10]. On this item, PD patients are rated for their facial expressivity for 10 s without moving or speaking. Although this item may pick up some loss of facial musculature control (e.g. reduced control in keeping mouth closed, reduced blinking), the item does not measure the ability to control a range of facial muscles. Therefore, not only is there limited understanding of voluntary facial musculature control in PD, there is

Abbreviations: PD, Parkinson's disease; FACS, Facial action coding system; MDS-UPDRS, Movement Disorders Society-sponsored revision of the Unified Parkinson's Disease Rating Scale; MOCA, Montreal Cognitive Assessment; GDS, Geriatric depression scale; LED, Daily levodopa dose equivalent; CI, Confidence interval.

* Corresponding author at: School of Psychology (M304), University of Western Australia, Crawley, WA, 6009, Australia. Tel.: +61 8 6488 1415.
E-mail address: michelle.marneweck@uwa.edu.au (M. Marneweck).

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no sensitive and practical measure of this ability in PD. As a result, the symptom of facial masking and its understanding remains restricted to impaired emotional expression, and mainly assessed by subjective judgment in clinical contexts.

We measured voluntary facial musculature control in PD in three enrollment phases with an adaptation of the Upper and Lower Face Apraxia Test [2], a test of the ability to make upper and lower facial movements. Initial observations of typical errors committed by both groups guided the process of modification of the scoring structure for a more refined classification of errors committed by PD and control participants. The overarching aim of this study was to explore a range of lower and upper non-emotional facial movements in persons with PD and healthy controls; in doing so, we also aimed to determine the utility of an adaptation of the Face Apraxia Test [2] as a sensitive and practical measure of voluntary facial musculature control in PD.

2. Materials and methods

2.1. Participants

Sixty-six participants (Control $n = 32$, PD $n = 34$), 49 participants (Control $n = 24$, PD $n = 25$), and 40 participants (Control $n = 17$, PD $n = 23$) took part in Enrollment Phases 1, 2, and 3 respectively. There were 22 participants unique to Phase 1 (Control $n = 12$, PD $n = 10$), four unique to Phase 2 (Control $n = 3$, PD $n = 1$), five unique to Phase 3 (Control $n = 2$, PD $n = 3$), and 23 and 26 participants who took part in two or three phases, respectively (two: Control $n = 13$, PD $n = 10$; three: Control $n = 10$, PD $n = 16$). The enrollment phases were approximately 1 year apart. The successive enrollment phases gave the opportunity to establish the reliability of the adapted Face Apraxia Test, by testing some patients in multiple phases; it also gave the opportunity to increase the number of data points in a notoriously variable group of subjects, thereby increasing the reliability of our measures of voluntary facial musculature in PD. PD and control groups were well matched in each phase on demographic and clinical characteristics (Table 1). Patients were diagnosed following clinical evaluation by a neurologist or geriatrician and were under care for PD by a local neurologist or geriatrician at the time of testing. Table 1 also shows PD-related characteristics, MDS-UPDRS-III motor scores, MDS-UPDRS-IV motor complication scores, years since diagnosis, and levodopa dose equivalent (LED) [30]. With the exception of two patients in Phase 1, one patient in Phase 2, and one patient in Phase 3, all patients were on dopamine replacement therapy (see Appendix Table A1 for a list of medications taken by patients for PD management). The local institutional ethics committee approved the study procedures and all participants gave written informed consent.

2.2. Stimuli and procedures

For PD patients, each of the enrollment phases was conducted approximately 1.5 h before their next scheduled dopamine replacement medication intake; we did not have ethical approval to compare patients that were on dopamine replacement therapy with those that were depleted from dopamine replacement therapy; patients were not asked to suspend their dopamine replacement therapy overnight, and thus were not in a practically defined off state. At the start of each phase, patients were assessed for disease severity, using the motor subscale (III) of the MDS-UPDRS [10], depressive symptoms (geriatric depression scale; GDS) [22], and general cognitive functioning (Montreal Cognitive Assessment; MOCA) [18]. The ability to voluntarily control facial musculature was measured using an adaptation of the Upper and Lower Face Apraxia Test [2] in which participants were required to make 9 upper and 29 lower facial movements. In the original test, the examiner gives a verbal instruction and demonstration for each item, and the participant's response follows immediately. For standardization purposes in our study, the instructions and demonstrations of each item were recorded in a video-clip lasting 7 min and watched by all participants on an 11-in. laptop. For scoring purposes, participants' faces were filmed with each face in full frontal view. Participants were instructed to reproduce the intensity and duration of each demonstration as accurately as possible.

Two independent raters, one blind to the study, scored the accuracy of each reproduction as correct or incorrect, with set criteria that determine an item as failed. For scoring purposes, the videorecording of each participant's set of reproductions was viewed alongside the video-clip of instructions and demonstrations. Initial observations revealed that errors by participants did not necessarily fit the error criteria in the original test. Facial movements by patients were often impoverished, with reproductions that were of lower amplitude than the demonstrated movement. Observations of impoverished facial movement are also commonly reported in clinical contexts [28,29]. We also observed loss of individuation of movement in PD and control groups, where the requested movement was temporally coupled with uninstructed movement. Individuation loss has also been reported with manual movements in healthy aging [11,17,23] and in PD [31]. Guided by these typical errors, we scored errors due to impoverished movement and individuation loss separately, allowing for a refined error analysis. We also introduced a category for content errors, which might indicate the presence of ideational apraxia. We observed reproductions by participants at times resembled the demonstrated movement but were incorrect in their content e.g. placing the tongue in the cheek when asked to puff out the cheek. Incorrect items were assigned to one of five error categories: (1) the reproduction was preceded by a pause during which unsolicited movements might have been present; (2) there was a loss of

Table 1
Demographic and clinical characteristics of participant groups in Enrollment Phases 1, 2, and 3.

	Phase 1		Phase 2		Phase 3	
	Control	PD	Control	PD	Control	PD
Age (years)	66 (51–79)	66 (46–80)	67 (55–80)	67 (53–81)	70 (53–80)	68 (58–82)
Males (females)	11 (21)	20 (14)	12 (12)	17 (8)	11 (6)	16 (7)
Education (years)	15 (7–22)	12 (7–20)	16 (8–22)	12 (7–20)	16 (11–21)	13 (9–19)
MOCA	27 (21–30)	28 (22–30)	28 (25–30)	27 (15–30)	27 (22–30)	27 (19–29)
GDS	1 (0–5)	2 (0–13)	1 (0–5)	2 (0–10)	1 (0–4)	1 (0–10)
Years diagnosed	–	5 (1–19)	–	7 (1–20)	–	8 (2–21)
MDS-UPDRS-III	–	38 (10–56)	–	41 (19–56)	–	40 (19–57)
MDS-UPDRS-IV	–	6 (0–14)	–	5 (0–13)	–	6 (0–13)
LED	–	788 (0–2046)	–	916 (0–2312)	–	1057 (0–2662)

Note. Values (except male and female numbers) are expressed as median (range); MOCA scores range from 0 to 30, a score ≥26 reflects normal cognitive functioning; GDS scores range from 0 to 15, a score of ≥6 suggests depression warranting assessment; MDS-UPDRS-III scores range from 0 to 132 (most severe); MDS-UPDRS-IV scores range from 0 to 24 (most severe). LED = daily levodopa dose equivalent [30].