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The feasibility and acceptability of using a novel wrist worn cueing device to self-manage drooling problems in people with Parkinson's disease: A pilot study

Roisin McNaney[1], Nick Miller[2], John Vines[3], Patrick Olivier[4], Karim Ladha[4], Daniel Jackson[4] and Richard Walker[5,6]

Abstract

Introduction: Daytime drooling is experienced by around 50% of Parkinson's patients, who fail to swallow saliva in sufficient volume or regularity, despite normal production. This research explored the feasibility and acceptability of using a cueing device, to improve drooling.

Methods: During a four-week intervention, 28 participants were asked to use a cueing device for 1 h per day. During this time, the device vibrated once-per-minute, reminding the participant to swallow their saliva. A daily diary was used to collect self-report around swallowing severity, frequency, and duration. This was filled out by participants for one week before, four weeks during and for one week immediately after intervention. Diaries were also collected for one week during a follow up, carried out four weeks after intervention finished.

Results: Participants self-reported benefits in drooling severity ($p = 0.031$), frequency ($p \leq 0.001$), and duration ($p = 0.001$) after using the device. Improvements were maintained at follow up. Twenty-two participants explicitly reported a positive benefit to their drooling during exit interview. All felt the intervention and device were acceptable and usable.

Conclusions: Using a cueing device for one month had perceived benefit to drooling severity, frequency and duration in patients with Parkinson's. Participants accepted the device and treatment protocol.

Keywords

Assistive technology, human factors, rehabilitation devices, self-care, therapeutic value

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Introduction

Sialorrhoea, also termed drooling or ptyalism, is reported as a common symptom of Parkinson's. In some studies, drooling is reported to be an issue in up to 70% of participants, especially if one takes into account nocturnal drooling and increasing severity of Parkinson's.[1–4] Saliva is vital for good oral health. Impaired production of, or loss of saliva through drooling exposes individuals to a range of negative effects, from mild annoyance at perceived lack or excess of saliva in the mouth, to major health and psychosocial issues. Saliva helps regulate oral pH and microbiotic homeostasis.[5] The antimicrobial, antiviral, and antifungal properties of saliva aid oral cleansing, protect

[1]School of Computer Science Electrical Engineering and Engineering Maths, University of Bristol, Bristol, UK
[2]Institute for Ageing, Newcastle University, Newcastle upon Tyne, UK
[3]NORTHLab, Northumbria University, Newcastle upon Tyne, UK
[4]Open Lab, School of Computing Science, Newcastle University, Newcastle upon Tyne, UK
[5]Institute of Health and Society, Newcastle University, Newcastle upon Tyne, UK
[6]Northumbria Healthcare NHS Foundation Trust, North Tyneside General Hospital, North Shields, UK

Corresponding author:
Roisin McNaney, SCEEM, University of Bristol, 1 Cathedral Square, Bristol BS1 5DD, UK.
Email: r.mcnaney@bristol.ac.uk

against infection, and support tissue repair. Saliva serves as a buffer against noxious substances. It lubricates the oral cavity, thereby supporting formation and transport of the food bolus to the pharynx for swallowing. It acts as a first stage in digestion and stimulates interaction with chemosensory receptors to aid taste and smell perception. It supports smooth movement of the tongue and lips for speech. If saliva is lost through drooling the person with Parkinson's is at risk of lowered resistance to infection, poor oral health, and added problems with swallowing and speech. Dry mouth—a common consequence of saliva loss—is associated with risks of ulceration, tooth decay, gingivitis, candidiasis, halitosis, and perioral dermatological issues.[6,7] In many societies, the effects of drooling (e.g., odor, stained clothes, constant wiping) are socially frowned upon. In this way, drooling may influence psychosocial health for the pwPD and produce an added burden for the carer (e.g., washing clothes; restricted social life).[8–10]

In Parkinson's drooling is associated not with excess production of saliva, but principally with muscle rigidity and bradykinesia of the facial, tongue, and lip muscles.[11–13] People with Parkinson's Disease (pwPD) who experience drooling fail to swallow saliva in sufficient volume or regularity, despite a normal amount of saliva production.[11,12] This leads to pooling of saliva in the mouth and risk of anterior loss. In addition to the impaired swallow mechanism in Parkinson's, the dysautonomia associated with the condition, as well as changes in sensory perception of food that affect salivation (smell, taste, vision) may complicate the picture.[5,7] Cognitive factors may also play a role. Reynolds et al.,[14] found an association between swallowing frequency and drooling severity, in particular during states of distraction.

Most current pharmaceutical treatments for drooling in Parkinson's aim to decrease saliva production. However, there are potential complications associated with their use. Firstly, as mentioned previously, lack of saliva can cause oral health problems (e.g., gingivitis, tooth destruction, tongue crusting).[15] Secondly, drug treatments such as sublingual atropine can lead to serious cognitive side effects, such as memory impairment and/or hallucinations.[16] Botulinum toxin injection into the salivary glands can be painful and must be repeated every three to six months. In addition, this carries some risk of masseter and pharyngeal muscle weakness that can both impact on chewing and swallowing. Meningaud et al.[15] extensively reviewed the modalities of treatment for drooling problems and maintained that it is important to propose, where feasible, non-invasive treatment options, such as behavioral cueing methods, before drug or surgical therapy is considered. Recent major guidelines underline the importance of this strategy.[17]

Cueing has been employed to successfully improve aspects of impaired activities in Parkinson's, such as gait.[18–21] Cueing generally relies on the implementation of a system of temporal cues, where participants are provided with time-controlled auditory or haptic prompts to instigate or modify a behavior.

The concept of temporal cueing as a treatment for drooling has been built on previous work, which has shown success of cueing, for example in gait training for Parkinson's.[18] The belief is that the training of a metronomic cue brings about the execution of a new motor plan, which facilitates walking and suppresses the impaired motor plan currently inhibiting the intended movement.[19–21] There is a level of automaticity in the complex movements of both walking and swallowing of saliva that link these two symptoms together and allow for cross comparison of motor theory. Both are triggered, patterned responses involving automated neural processes that generally do not require conscious thinking for carrying out the activity. However, in the case of Parkinson's, these automatic movements can become impeded when difficulties with motor initiation arise. In terms of neurophysiology, cueing is believed to suppress pathological basal ganglia activity through activation of corticostriatal pathways.[19] That is to say, the cue causes the initiation of an alternative pathway in the brain, also linked to motor activity, which brings about the initiation of movement that has been halted.

The feasibility of cue provision to improve drooling has been minimally studied. Marks et al.,[22] used a (now) commercially available device, in the form of a brooch, which emitted an auditory cue (a short "beep") at regular intervals to remind the wearer to swallow. They found this yielded positive results for participants ($n = 6$). Although the device was found to be effective for the control of drooling problems, their small sample size did not provide sufficient information around the effectiveness of the intervention on a wider population of people with Parkinson's, nor did the authors discuss the acceptability of the technology they trialed with their participants. A further study by Marron et al.[23] showed that wearers of the same drooling brooch reported several aspects that reduced its acceptability. For example, hearing impaired participants could not use the device, yet the auditory cue was also a source for concern for others in the environment, since the beep attracted attention when worn, drawing into question its social acceptability. The product used also incorporated a switch to turn the device on and off. Some users required assistance to operate this due to their impaired fine motor skills.

In response to these drawbacks, we developed a simple-to-use wrist-worn digital cueing device, the Parkinson's Disease Cueing Device (PDCue)

Figure 1. The cueing device.

(Figure 1). This was iteratively designed with pwPD and their caregivers.[24] This early study established usability and motor and social acceptability of the device we employed; that it was usable even by individuals with marked fine motor and sensory difficulties; and that a vibratory cue was preferable to an auditory cue.[24] The device delivers a silent vibratory cue, once per minute when switched on, to remind the person to swallow. The once per minute setting was decided upon in accordance with previous research which established daytime non-stimulated swallowing frequency in healthy adults of around one swallow per minute.[25] This was also the preferred interval selected by Marron et al.[23] in their study. There is, as yet, no literature relating to the swallowing rate of pwPD.

The purpose of the pilot study presented here was to explore the feasibility and acceptability of using this novel cueing device to help pwPD to self-manage their drooling, and to establish whether there was evidence of an effect on drooling severity and frequency when wearing the PDCue. We also wanted to explore some practicalities relating to recruitment and retention of participants into the study, and how appropriate our outcomes measures were, in order to inform the trial design of any larger scale studies which might arise.

Methods

Experimental design

This study employed quantitative methods to examine for possible effects of the cueing device on perceived drooling severity and frequency. It used qualitative methods (semi-structured exit interview) to establish opinions of participants on the acceptability and feasibility of the intervention program, and experiences of using the PDCue.

Participants and recruitment

The UK based study was approved by the Newcastle and North Tyneside National Research Ethics Service Committee (reference: 11/NE/0257). Informed written consent was obtained from all participants in the study. All study data were collected by employees of Northumbria Healthcare NHS Foundation Trust, who were responsible for the organization of the project. Participants were primarily recruited via the regular Parkinson's clinics at Northumbria Healthcare NHS Foundation Trust, but participants from Participant Identification Centers in Sunderland, Gateshead and Newcastle were also included. Potential participants were identified by clinical staff and then contacted by a researcher via telephone with further information. Written information sheets were then sent to those who expressed sustained interest and, following a one-week period, participants were visited in their homes to obtain informed consent.

Inclusion criteria were (1) anyone with a diagnosis of Idiopathic Parkinson's (stages I–III in Hoehn and Yahr scale[26]), in accordance with the UK Parkinson's Brain Bank criteria,[27] (2) an acknowledged daytime drooling problem, either observed by a clinical professional within a Parkinson's Disease clinic or through patient self-report, and (3) an ability to understand and respond to the instructions given in the study. Exclusion criteria were (1) currently receiving pharmaceutical treatment for drooling and (2) insufficient dexterity with which to use the device.

A full case history was taken from each participant regarding their Parkinson's, drooling, and history of swallowing difficulties. The Mini Mental State Exam[28] and Montreal Cognitive Assessment Test were conducted for screening purposes of cognitive impairment.[29] The Unified Parkinson's Disease Rating Scales (UPDRS) II and III[30] were conducted to gain an indication of overall disease state. All assessments were performed by the same researcher in the participant's own home.

Participants were randomly allocated to either an immediate intervention group ($n = 17$) or a delayed intervention group ($n = 11$). This was to provide preliminary data for comparison of treatment vs. no treatment. The delayed group did not commence intervention until after they had completed a four-week period of no intervention. The randomization protocol was predetermined using an online random number generator (www.randomizer.org/). The numbers were arranged into consecutive order creating a sequence for randomizing individuals (e.g., 1-immediate, 2-immediate, 3-delayed). If a participant left the study, their group assignment (immediate or delayed) was added to the end of this list to be filled by later recruits. We aimed for a 1:1 ratio, with a target recruitment of 30 participants (15 in each group). We fulfilled the capacity of the intervention group, but time restrictions meant that we were unable to complete a delayed

start for the final two participants we had recruited. As such, we entered them into the intervention group leaving final numbers of 17 immediate and 11 delayed participants. This is a limitation of the study and is discussed further in the limitations section.

Measurements

The "saliva" subset of questions from the Radboud Oral Motor Inventory for Parkinson's Disease (ROMP–Saliva),[31] and the UPDRS 2.2 subtest for saliva[30] were conducted with each participant at: one week before commencing use of the cueing intervention (assessment point 1), one week immediately after finishing the intervention (assessment point 2), and four weeks later at a follow up appointment (assessment point 3). For participants in the delayed start group, an additional assessment was collected four weeks prior to the immediately pretreatment assessment (assessment point 0). Figure 2 illustrates the time line.

ROMP-S is a validated tool[31] for use with pwPD. It is derived from the unvalidated Drooling Frequency and Severity Scale (DFSS)[32] originally drawn up for children with cerebral palsy but employed in several other populations. It was slightly modified for ROMP-S, in particular by adding the option to score that one is troubled by (perceived) accumulation of saliva without actually drooling. The nine items, rated on 5-point ordinal scales that describe gradations of drooling activity, cover: day and nighttime frequency and severity of drooling, effects on speech and eating and drinking, how frequently one has to wipe away saliva, limitations on daily activity and social participation, and overall impact.

UPDRS item 2.2 is a 5 point descriptive ordinal scale ranging from 0 to 4; no drooling (0), excess saliva but no loss (1), nighttime but not awake drooling (2), awake drooling but wiping not necessary (3), severe drooling with constant wiping/wet clothes (4).

At each assessment point, participants completed a seven-day drooling severity, frequency, and duration diary. Participants monitored their drooling over the course of one self-selected hour per day when they would typically drool (e.g., after meal times, in the morning). Following Hauser et al.[33] participants completed 100 mm visual analogue scales (VASs). They placed a cross on a 100-mm line (with 0 mm being "no problem" and 100 mm being "as bad as can be") to indicate the number of separate incidents they felt that drooling occurred (frequency), how long in minutes they felt drooling occurred (duration), and how severe they felt drooling was (severity). This method reflects standardized methods of monitoring using paper diaries employed in other medical research (e.g., Montgomery and Reynolds[34] and Stone et al.[35]).

Finally, an exit interview was carried out with each individual to gather qualitative feedback on the participants' experiences. A semi-structured approach was taken to probe; (a) experiences of drooling before taking part in the study; (b) experiences of drooling after taking part in the study; and (c) perceptions around the acceptability, worthwhileness, and effectiveness of the PDCue as a way to self-manage their drooling.

Intervention

All participants were visited at home and received a verbal and practical tutorial on how to use the cueing device. They were asked to use the device for one hour a day, for a total of four weeks, at a time when drooling was an issue for them. Participants were asked to not use the device during the hour that they were self-reporting their drooling on the daily diary.

Data processing and analysis

Quantitative data were analyzed using the IBM SPSS statistical software suite (v.22, IBM Corp, Armonk, NY). For data collected at the ordinal, interval or ratio level, normality of distribution was checked by inspection of histograms and using the Shapiro–Wilk

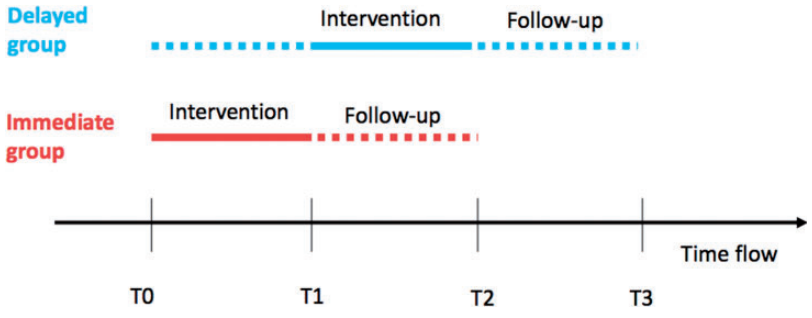

Figure 2. Visualisation of the assessment schedule.

and Kolmogorov–Smirnov tests.[36] Data were summarized using statistics appropriate to the level of the data (e.g., median, interquartile range, frequency). In inferential analysis, the Wilcoxon signed ranks test or Mann–Whitney U test was applied to ordinal, interval or ratio data, and the Chi-squared test to categorical data. For analysis across all 28 participants of change in scores from pre- to post-treatment (assessment points 1 and 2), and from pre-treatment to follow-up (assessment points 1 and 3) for the same variable, the Bonferroni correction was applied, setting significance at 2.5%. For all other inferential tests significance was set at 5%. Two-tailed tests were used throughout. A repeated measures design looked at both between (delayed intervention vs. immediate intervention) and within-group differences (all participants combined to compare baseline pre-treatment, termination of treatment, and follow-up outcomes).

Qualitative data collected during the exit interviews were audio-recorded and transcribed verbatim. Transcriptions were then subjected to an inductive thematic analysis using methods drawn from Braun and Clarke.[37] Data were summarized with short, one or two word codes, at the sentence-to-paragraph level. Codes were then compared to one-another and grouped, which led to the construction of broader themes that captured the core topics and concerns emerging from the data.

Results

Fifty-eight participants were identified for potential inclusion. Twenty of these chose not to join (due to reasons such as not feeling drooling was severe enough; not having time to commit to research). Thirty-eight consented to participate. During the trial, 10 participants left the study. Five stated reasons of ill health, four felt the study was too much for them to manage at the time, and one gave no reason. The data analyzed came from the 28 remaining participants (10 female). Compliance levels for filling out diaries varied. Out of a possible 6699 diary entries, 5069 (76%) were provided. The demographic and case history information can be viewed in Table 1. No significant biases were observed between the two groups with regard to any of the variables investigated.

Intervention vs. no intervention

Results for the first and second assessments with the no-intervention group, and assessments for the immediate intervention group before and at end of intervention, appear in Table 2. There were no statistically significant differences between the immediate and delayed intervention groups at the initial baseline

Table 1. Comparison of demographic data and case histories for delayed and intervention groups.

	Delayed intervention($n = 11$)	Immediate intervention ($n = 17$)	Significance
Demographics			
Median age in years (IQR)	75 (65–79)	72 (65.5–78.5)	$U = 89.0$, $z = 0.212$, $p = 0.832$
Median years since PD diagnosis (IQR)	5 (3–8)	7 (2.5–10)	$U = 78.0$, $z = 0.732$, $p = 0.464$
N. of females (%)	3 (27%)	7 (41%)	$X^2(1) = 0.562$, $p = 0.689$
N. of participants living alone (%)	1 (9%)	5 (29%)	$X^2(1) = 1.638$, $p = 0.355$
Overall Parkinson's severity (SD)	44 (39–70)	53 (35–81)	$U = 76.5$, $z = 0.800$, $p = 0.424$
-Median UPDRS II and III score combined (IQR)	II: 6 (55%) III: 3 (27%)	II: 7 (41%) III: 8 (47%)	$X^2(2) = 1.115$, $p = 0.573$
-Hoehn and Yahr stage	IV: 2 (18%)	IV: 2 (12%)	
Initial perception of drooling severity, self-reported by the participant *N* (%)	Mild = 4 (36%) Moderate = 5 (46%) Severe = 2 (18%)	Mild = 9 (53%) Moderate = 5 (29%) Severe = 3 (18%)	$X^2(2) = 0.878$, $p = 0.778$
Median months since drooling first noticed (IQR)	24 (12–36)	12 (9.5–24)	$U = 75.0$, $z = 0.888$, $p = 0.375$
N. of participants with previous drooling treatment (%)	3 (27%)	5 (29%)	$X^2(1) = 0.015$, $p = 1.000$
N. of participants with reported swallowing problems (%)	7 (64%)	7 (41%)	$X^2(1) = 1.348$, $p = 0.246$

UPDRS: Unified Parkinson's Disease Rating Scales; IQR: Interquartile Range; SD: Standard Deviation.

Table 2. Assessment and diary results for both groups, from assessment 1 to assessment 2.

Assessment	Delayed group ($n = 11$)		Intervention group ($n = 17$)		Significance of between group difference
	Assessment point 1 (T0) Median (IQR)	Assessment point 2 (T1) Median (IQR)	Assessment point 1 (T0) Median (IQR)	Assessment point 2 (T1) Median (IQR)	Mann–Whitney *U* test
ROMP–saliva	20 (17–25)	19 (17–30)	22 (16–23)	22 (17–25.5)	$U = 83.0$, $z = 0.497$, $p = 0.619$
UPDRS 2.2 (saliva and drooling subtest)	3 (1–3)	3 (1–3)	3 (3–4)	3 (2–4)	$U = 69.0$, $z = 1.212$, $p = 0.225$
Drooling diary					
Severity	1 (0–4)	1 (1–5)	3 (1.5–5)	1 (0–2.5)	$U = 39.5$, $z = 2.575$, $p = 0.010$
Duration (No. minutes drooling occurred in 1 h)	1 (0–5)	2 (0.5–10)	5 (1–11)	1 (0–4.5)	$U = 63.0$, $z = 1.440$, $p = 0.150$
Frequency (No. instances in 1 h)	1 (0–4)	3 (1–4)	3 (1–4.5)	1 (0–3)	$U = 54.0$, $z = 1.876$, $p = 0.061$

UPDRS: Unified Parkinson's Disease Rating Scales.
Please note that lower scores indicate a lowered perceived severity in the diary, or lower impact of symptoms in the Parkinson's Disease Questionnaire (PDQ-39) subtests, Radboud Oral Motor Inventory for Parkinson's disease (ROMP)–Saliva and Unified Parkinson's Disease Rating Scale (UPDRS) 2.2.

assessment. There were no significant changes in ROMP-S, UPDRS 2.2, or Diary reports (VAS measurements) for the no-intervention group during the four weeks of no treatment. There were also no statistically significant changes to ROMP-S and UPDRS 2.2 in the intervention group when comparing pre- versus termination of treatment. Patient perceived changes on the VASs did show significant improvement in the intervention group for overall severity, but for frequency of drooling improvement was borderline ($p = 0.06$) and perceived amount of time (duration) of drooling did not alter significantly.

Comparison of pre- and post-treatment scores across all participants

Given that there appeared to be no placebo effect in the delayed intervention group during the no intervention phase (i.e., no significant improvement in any scores), once both groups had completed intervention their scores were combined to provide a larger group ($n = 28$) for comparison of pre- versus post-treatment versus follow-up assessment.

Between treatment termination and four-week follow-up assessment four participants left the study (two moved on to Botox treatment immediately after the intervention ended, one experienced significant health decline and one moved abroad). This left 24 participants available for longer term follow-up assessment comparisons. Outcomes are summarized in Table 3.

ROMP-S ratings saw no significant change comparing scores at pre- versus at termination of treatment. UPDRS 2.2 ratings showed a trend toward significance but were still statistically non-significant. VAS patient perceptions of change in overall severity, duration, and frequency of drooling all evidenced significant improvements.

To examine whether scores returned to baseline status once intervention finished, baseline scores were compared with four weeks post-treatment assessments. ROMP-S demonstrated a move toward significance (but was still not statistically significant), while UPDRS 2.2 now showed a significantly better status. The VAS ratings all showed strongly significant improvements, including after adjustments for multiple testing. The findings suggest maintenance or even improvement of status during the follow-up phase.

Exit interviews

Twenty-seven participants were available for exit interview. One participant had a significant health decline and was thus unavailable. Interviews lasted mean 17:03 min (shortest 6:29-longest 36:37). Following procedures outlined in the Methods section, 26 thematic codes applied to the data. A total of 312 extracts of transcript were assigned these codes (ranging from 1 to 22 extracts per code). A total of four higher level themes were then constructed from this qualitative data analysis, which are summarised below.

The first theme to arise was the impact of drooling on the lives of the participants. By far, the most discussed impact of drooling issues pre-treatment was embarrassment (13/27), with several participants discussing emotional distress *"It really dominated my life . . . it was most distressing, psychologically distressing . . . it clearly ruled my thinking . . . in that I was always clasping this grubby handkerchief just in case"* (P14), and social withdraw, *"At least once a day it would happen. I was out with company and it made me*

Table 3. Assessment and diary results for the entire intervention group ($n = 28$) for pre-, posttreatment, and follow-up results.

Assessment	Entire group ($n = 28$)			Significance of difference	
	Assessment point 1 (T0) Median (IQR)	Assessment point 2 (T1) Median (IQR)	Assessment point 3 (T3) Median (IQR)	Assessment points 1 (T0) to 2 (T1)	Assessment points 1 (T0) to 3 (T2)
ROMP–saliva	20.5 (16–23.75)	20 (16.25–24.75)	17 (15–23.5)	$Z = 0.275, p = 0.783$	$Z = 1.800, p = 0.072$
UPDRS 2.2 (Saliva and drooling subtest)	3 (3–4)	3 (2–4)	2 (1–3)	$Z = 1.801, p = 0.072$	$Z = 2.569, p = 0.010$
Drooling diary					
Severity	3.14 (2.42)	1.18 (1.57)	1.14 (1.51)	$Z = 2.151, p = 0.031$	$Z = 2.809, p = 0.005$
Duration (No. minutes drooling occurred in 1 h)	7.45 (10.64)	4.75 (11.55)	1.86 (2.73)	$Z = 3.362, p = 0.001$	$Z = 2.631, p = 0.009$
Frequency (No. instances in 1 h)	4.18 (5.68)	1.80 (2.32)	1.16 (1.47)	$Z = 3.982, p < 0.001$	$Z = 3.606, p < 0.001$

Please note that lower scores indicate a lowered perceived severity in the diary, or lower impact of symptoms in the Parkinson's Disease Questionnaire (PDQ-39) subtests, Radboud Oral Motor Inventory for Parkinson's disease (ROMP)-Saliva and Unified Parkinson's Disease Rating Scale (UPDRS) 2.2.

feel very embarrassed. I tend to withdraw, avoid going out really. Eat on my own. I am pretty strict about manners, and I thought it looked horrible'' (P12). Several participants (4/27) also discussed physical discomfort that they experienced—constant wetness, changing of handkerchiefs, painful sores around the mouth.

The second theme related to challenges around previous experiences of drooling treatment. Several participants (3/27) had previous experience of Botox; however, for a lot of participants (8/27), Botox was not an option they would have considered. These participants discuss a lack of willingness to take additional medication; *''when I saw the consultant they said I could go and have Botox, an injection. I didn't want to take any more drugs''* (P22). Botox was associated with words such as *''toxic''* (P25) and *''poison''* (P26) and there was a clear preference for avoiding it, and other additional medication, if possible; *''I think if you can have something that avoids taking drugs I think that's great''* (P4). These participants, unsurprisingly, preferred the PDCue as a behavioral treatment option; *''I'd rather have the watch''* (CP7).

Theme 3 related the effect of the PDCue on drooling. Of the 27 interviewed participants, there was a reported positive effect for 22, indicating that the majority of participants successfully engaged with the intervention and found it to be a worthwhile option for supporting the self-management of drooling. Participants also discussed emotional benefits which arose as a result of the PDCue intervention, including improvements to self-esteem, confidence, and feelings of control.

The final theme related to reports of generalization and habituation. There were several cases of participants reporting a generalization effect, wherein they felt an increase in swallowing frequency was being carried over to times when they were not wearing the PDCue (9/27). P4 said *''Even when I wasn't wearing the* [PDCue] *every now and again I think, 'Oh yes, you haven't swallowed. I need to swallow'.''* P3 also noted *''even when I wasn't wearing it I was much more conscious of it.''* Although unexpected, P26 also discussed an improvement to his night time drooling *''I've hardly been drooling at all. No, I haven't. Even during the night I haven't been.''*

There were a small number of participants (3/27) however who reported becoming habituated to the cues, e.g., P10 *''there were occasions when I had the watch on, I seemed to have got so used to it that I didn't get any indication.''* However, these participants reported a positive effect from the intervention, despite this habituation.

Discussion

This pilot study aimed to explore the feasibility, usability, and acceptability of wearing a wrist-worn vibratory cueing device to improve drooling in people with Parkinson's, and to establish whether there was evidence of an effect of the device on drooling severity, frequency, and impact. Twenty-eight people completed a month-long intervention with the device, using it for an hour a day. Altogether they showed significant improvement on severity ($p = 0.031$), frequency ($p \leq 0.001$), and duration ($p = 0.001$) of drooling when comparing results from the self-reported VASs collected pre- and post-intervention. These improvements were also seen to remain at follow up assessment four weeks post-treatment compared to their pre-intervention baseline. A significant improvement was also seen in the UPDRS 2.2 saliva subtest ($p = 0.010$) when comparing the pre-intervention and follow-up assessment time points. Based on this, we conclude

there is some evidence to indicate that the device can be successful in improving saliva control, not only when wearing the PDCue but also when not using it or after intervention has been withdrawn.

Comments from the interviews confirm that people are disturbed by their drooling and that lesser drooling brings benefits for psychosocial well-being. Participants were satisfied with manipulating the device and found it acceptable to wear. They perceived the gains seen made wearing the device worthwhile. This is further reflected in comments participants made in the exit interviews concerning the perceived positive effects of the intervention device. Responses showed that 22/27 participants explicitly reported that they had noticed benefits to their drooling, with several stating that it was a preferable treatment option to other pharmaceutical interventions. In a larger trial, it is unlikely that this interview-based approach could be undertaken at scale. The development of a questionnaire, drawing on the themes outlined from the qualitative data could be an approach to capturing data of this kind.

The lack of positive change in the delayed treatment group during their no-intervention phase suggests that change was not accounted for by a placebo effect from being recruited to the study, being assessed, completing the diary exercise, nor from receiving information about drooling and drooling interventions in general. There was no improvement despite written (in the study information pack) and oral discussion that more frequent swallowing may benefit saliva loss. While the present data suggest placebo effect does not play a significant role here, in a definitive trial a more active comparator condition should be introduced.

Our results showed a measurable change in score in the UPDRS item for drooling, between baseline and four-week follow-up, lending additional weight to the improved VAS responses that participants provided. However, we did not observe any significant differences in the ROMP-S overall score. This may indicate that the types or level of changes experienced over the intervention period were not sufficient for this tool to capture. It could be that employing an overall score across multiple dimensions rather than analyzing each item separately masked significant gains in some areas. For instance, there may have been no change in night time drooling score, or even, after a short period, no shift in overall impact. Nevertheless, frequency of having to wipe the mouth or perceived excessive saliva in the mouth may have altered, but these improvements failed to make a significant difference in overall score against the non-altered variables. A similar factor may be at work in the lesser (compared to VASs) sensitivity to change of the UPDRS saliva item, since this scale combines several features which might actually vary independently (e.g., day vs. night drooling; perceived excess saliva in mouth; frequency vs. severity) in one scale. Significant improvement in one sub-dimension may be missed if the other dimensions do not alter. Further analyses of individual items prior to a definitive trial may aid in separating out which aspects of drooling are more or less susceptible to influence through cueing.

A factor to consider for a larger trial is data completeness for the diaries. We had a 76% completion rate across the study. Although this is not dissimilar to other studies (e.g., Hauser et al.[33]), a larger trial would need to consider the time requirements of participants and the burden of the study to self-report. We make the suggestion that completing the diary throughout the entirety of the study is not required. Only completing the diary for one-week pre- and post-intervention (and again at follow up) would be enough in a follow up trial, as these were the results that we eventually focused on in our analysis.

Self-reported diaries are heavily used in clinical research as a way to monitor the progress of treatment and log patients' activities over time, without the requirement for a researcher to be present, despite long-standing reported issues with compliance (e.g., Montgomery and Reynolds[34] and Stone et al.[35]). Recent research into tools to support self-report in Parkinson's research has provided clear recommendations for improving practice, with Vega et al. finding 99% compliance with their paper-based tool measuring self-reported symptoms over several months with a small number of participants.[38] However, another solution would be to introduce usage logs, collected automatically by the device, which would also provide an indication of participant compliance without the need for the diary.

The body of literature exploring cueing for drooling as a symptom is minimal, with small-scale preliminary work by Marks et al.[22] and Marron et al.[23] being the only examples exploring this space. As such, our work builds upon this nascent body of literature to provide additional evidence that cueing for drooling might be an effective way to manage the symptom, with the qualitative aspects of our study additionally demonstrating reports around increased feelings of control, confidence, and self-esteem post intervention. In addition, we build on our previous work[24] to report that acceptance and usability of our device has been confirmed with a larger and more varied group of participants over a longer period of time. Our cueing approach warrants further exploration in a larger scale trial.

Study limitations

There are several provisos in interpreting the current data. Firstly, one assumes that participants were wearing the devices as requested, for a designated hour each day. However, we did not collect precise usage logs. In future work, there would be benefit in utilizing a

more objective approach, e.g., through digital usage logs collected directly through the device (i.e., using an accelerometer to provide data on when the device is switched on and being used). Secondly, we asked participants to self-select an hour within which to self-monitor their drooling, at times when drooling was a problem. While participants may have selected a self-perceived period of more susceptibility to drooling, it remains unclear how severe their chosen hour might have been. Further laboratory-based work, employing objective measures of physiological drooling (e.g., objective swallow frequency measurement, or saturated gauze weight measurement) would add insight into whether or not orally retained saliva objectively decreased through use of the device. This would also remove, at least in laboratory conditions, the use of self-report diaries that may be open to recall bias. For field-testing, employing devices capable of measuring swallowing events in naturalistic situations (e.g., using an in-ear microphone) would be beneficial.

Finally, although 30 participants were the sample size intended for this first stage feasibility trial, we did not have matched numbers between the delayed and immediate groups. The intention was to have 15 participants in each, but time constraints meant that we were unable to fully recruit to our delayed group (with four participants remaining). We made a decision to include a final two participants in the study as immediate intervention participants. Future studies implementing two treatment strands should not have this problem in future work; however, future researchers should also consider randomisation approaches that allow for equal participant numbers throughout the recruitment process (e.g., even vs. odd participant numbers to each strand). In addition, while the results of our pilot work delivered some positive outcomes, sufficient to suggest the cueing device may be effective, a more definitive answer awaits a trial involving larger numbers in a more highly powered study and with an active intervention comparator.

Conclusions

This study has indicated that our cueing device was acceptable and usable, and that the intervention could be a feasible first step for clinicians, before moving on to pharmaceutical options, which have been shown to have potential complications. While the next step of this research will require a larger multi-center trial to elucidate whether these results are replicable and clearer in a larger population, and to look at the characteristics of responders vs. non-responders to the treatment, the information presented within this paper has provided important, preliminary data around the effect that the cueing intervention could have and issues to address in the development of outcome measures.

Clinical messages

- Providing a regular vibratory cue, through the PDCue device was shown to be an effective treatment for reducing perceived drooling in the great majority of participants.
- Participants accepted PDCue and remained motivated to self-manage their drooling with the device.
- Further studies are needed to confirm the beneficial effects that we observed and for the refinement of outcome measures.

Authors' note

Patrick Olivier is now affiliated with Community, Organisational and Social Informatics, Monash University, Melbourne, Australia.

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Contributorship

RM, PO, RW, and NM were all involved in the conception of the research project and in researching the existing literature. RM, RW, and NM were involved in protocol development, with support from PO, KL, and DJ. Ethical approval and patient recruitment were conducted by RM and RW. Data capture and analysis were conducted by RM, KL, JV, and DJ. RM wrote the first draft of the manuscript. All authors reviewed and edited the manuscript and approved the final version of the manuscript.

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ORCID iD

Roisin McNaney https://orcid.org/0000-0003-3761-296X

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