

# Northumbria Research Link

Citation: McNaney, Róisín, Miller, Nicholas, Vines, John, Olivier, Patrick, Ladha, Karim, Jackson, Dan and Walker, Richard (2019) 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. *Journal of Rehabilitation and Assistive Technologies Engineering*, 6. pp. 1-11. ISSN 2055-6683

Published by: SAGE

URL: <https://doi.org/10.1177/2055668319852529>  
<<https://doi.org/10.1177/2055668319852529>>

This version was downloaded from Northumbria Research Link:  
<http://nrl.northumbria.ac.uk/id/eprint/39104/>

Northumbria University has developed Northumbria Research Link (NRL) to enable users to access the University's research output. Copyright © and moral rights for items on NRL are retained by the individual author(s) and/or other copyright owners. Single copies of full items can be reproduced, displayed or performed, and given to third parties in any format or medium for personal research or study, educational, or not-for-profit purposes without prior permission or charge, provided the authors, title and full bibliographic details are given, as well as a hyperlink and/or URL to the original metadata page. The content must not be changed in any way. Full items must not be sold commercially in any format or medium without formal permission of the copyright holder. The full policy is available online: <http://nrl.northumbria.ac.uk/policies.html>

This document may differ from the final, published version of the research and has been made available online in accordance with publisher policies. To read and/or cite from the published version of the research, please visit the publisher's website (a subscription may be required.)



**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**

Journal:	<i>RATE Journal</i>
Manuscript ID	RATE-18-0022.R2
Manuscript Type:	Original Manuscript
Date Submitted by the Author:	16-Apr-2019
Complete List of Authors:	McNaney, Roisin; university of Bristol, SCEEM Miller, Prof Nick; Newcastle University, Institute of Health and Society Vines, John; Northumbria University, School of Design Olivier, Patrick; University of Newcastle, School of Computing Science Ladha, Karim; Newcastle University, School of Computing Science Jackson, Daniel; University of Newcastle, School of Computing Science Walker, Richard; University of Newcastle, Institute of Health and Society
Keywords:	Assistive Technology, Human Factors, Rehabilitation Devices, Self Care, Therapeutic Value
Abstract:	<p>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.</p> <p>Methods: During a 4-week intervention, 28 participants were asked to use a cueing device for one hour 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 1 week before, 4 weeks during, and for 1 week immediately after intervention. Diaries were also collected for 1 week during a follow up, carried out 4 weeks after intervention finished.</p> <p>Results: Participants self-reported benefits in drooling severity (<math>p=0.031</math>), frequency (<math>p&lt;0.001</math>), and duration (<math>p=0.001</math>) 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.</p> <p>Conclusions: Using a cueing device for 1 month had perceived benefit to drooling severity, frequency and duration in patients with Parkinson's. Participants accepted the device and treatment protocol.</p>

1  
2  
3  
4  
5  
6  
7  
8  
9  
10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60



# 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

## Introduction

Sialorrhoea, also termed drooling or ptyalism, is reported as a significant 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, anti-viral and anti-fungal properties of saliva aid oral cleansing, protect 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 (pwPD) 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

1  
2  
3 way drooling may influence psycho-social health for the pwPD and produce an added  
4 burden for the carer (e.g. washing clothes; restricted social life) [8-10].  
5  
6

7  
8 In Parkinson's, with over half of all individuals reporting diurnal (daytime) drooling  
9 [1]—a figure which rises even further when nocturnal drooling is taken into account [2].  
10  
11 dDrooling is associated not with excess production of saliva, but principally with muscle  
12 rigidity and bradykinesia of the facial, tongue and lip muscles [11-13]. PwPD who  
13 experience drooling fail to swallow saliva in sufficient volume or regularity, despite a normal  
14 amount of saliva production [11, 12]. This leads to pooling of saliva in the mouth and risk of  
15 anterior loss. In addition to the impaired swallow mechanism in Parkinson's, the  
16 dysautonomia associated with the condition, as well as changes in sensory perception of  
17 food that affect salivation (smell, taste, vision) may complicate the picture [5, 7]. Cognitive  
18 factors may also play a role. in the oral and pharyngeal structures [1] leading to pooling of  
19 saliva in the mouth. People experiencing drooling issues fail to swallow saliva in sufficient  
20 volume or regularity, despite a normal amount of saliva production. Nóbrega et al. [1], using  
21 modified barium swallow with videofluoroscopy and a drooling score, showed changes in  
22 the oral stage of swallowing in 100% of people experiencing drooling problems (n=16), and  
23 in the pharyngeal stage in 94% of patients. They also found a correlation between drooling  
24 severity and swallowing problems (dysphagia). People with the worst dysphagia had the  
25 worst drooling. In Edwards et al [3], self-reported drooling was a problem in 70% of people  
26 with Parkinson's, in both the later stages and early stages of the disease. In addition, a 2017  
27 study by Reynolds, Miller and Walker [14], found an association between swallowing  
28 frequency and drooling severity, in particular during states of distraction.  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56

57 Most current pharmaceutical treatments for drooling in Parkinson's aim to decrease  
58 saliva production. However, there are potential complications associated with their use.  
59  
60

1  
2  
3 Firstly, as mentioned previously, lack of saliva can cause oral health problems (e.g. gingivitis,  
4 tooth destruction, tongue crusting) [155]. Secondly, ~~the use of~~ drug treatments such as  
5 ~~s~~Sublingual ~~a~~Atropine can lead to serious cognitive side effects, such as memory impairment  
6 and/or hallucinations [166]. Botulinum toxin injection into the salivary glands can be painful  
7 and must be repeated every three to six months. In addition, this carries some risk of  
8 masseter and pharyngeal muscle weakness that can both impact on chewing and  
9 swallowing. Meningaud et al. [155] extensively reviewed the modalities of treatment for  
10 drooling problems and maintained that it is important to propose, where feasible, non-  
11 invasive treatment options, such as behavioral cueing methods, before drug or surgical  
12 therapy is considered. Recent major guidelines underline the importance of this strategy  
13 [17].therapy is considered.

14  
15  
16 Cueing has been employed to successfully improve aspects of impaired activities in  
17 Parkinson's, for aspects of Parkinson's such as gait, ~~has been used successfully in the past~~  
18 [18-21]. Cueing generally relies on the implementation of a system of temporal cues,  
19 where participants are provided with time-controlled auditory or haptic prompts to ~~change~~  
20 ~~their~~instigate or modify a behavior.

21  
22  
23 The concept of temporal cueing as a treatment for drooling has been built on  
24 previous work, which has shown success in the domain of cueing, -for example in gait  
25 training for Parkinson's [18]. The belief is that it is built upon observations that the training  
26 of a metronomic cue brings about the execution of a new motor plan, which facilitates  
27 walking and suppresses the impaired motor plan currently inhibiting the intended  
28 movement [19-21]. There is a level of automaticity in the complex movements of both  
29 walking and swallowing of saliva that link these two symptoms together and allow for cross

1  
2  
3 comparison of motor theory. Both are triggered, patterned responses involving automated  
4 neural processes that generally do not require conscious thinking for carrying out the  
5 activity. However, in the case of Parkinson's, these automatic movements can become  
6 impeded when difficulties with motor initiation arise. In terms of neurophysiology, cueing is  
7 believed to suppress pathological basal ganglia activity through activation of corticostriatal  
8 pathways [19]. That is to say, the cue causes the initiation of an alternative pathway in the  
9 brain, also linked to motor activity, which brings about the initiation of movement that has  
10 been halted.

11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24 The feasibility of cue provision to improve drooling has been minimally studied.  
25  
26 Marks et al [2211], used a (now) commercially available device, in the form of a brooch,  
27 which emitted an auditory cue (a short 'beep') at regular intervals to remind the wearer to  
28 swallow. They found this yielded positive results for participants (n=6). Although the device  
29 was found to be effective for the control of drooling problems, their small sample size did  
30 not provide sufficient information around the effectiveness of the intervention on a wider  
31 population of people with Parkinson's, nor did the authors discuss the acceptability of the  
32 technology they trialed with their participants. A further study by Marron et al. [2312]  
33 showed that wearers of the same drooling brooch reported several aspects that reduced its  
34 acceptability. For example, hearing impaired participants could not use the device, yet the  
35 auditory cue was also a source for concern for others due to in the environment, since the  
36 beep attracted attention when worn, drawing into question its social acceptability. The  
37 product used also incorporated a switch to turn the device on and off. S, which some users  
38 required assistance to operate this due to their impaired fine motor skills. degeneration  
39 resulting from Parkinson's.

1  
2  
3  
4  
5  
6  
7  
8  
9  
10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

In response to these drawbacks, we developed a simple-to-use wrist-worn digital cueing device, the PDCue (figure 1). This was iteratively designed with pwPD People with Parkinson's and their caregivers in our previous work [2413]. 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 [2413]. 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 once per minute setting was decided upon in accordance with previous research around swallowing frequency in healthy adults (1.32 swallows per minute) [14] and was~~ the preferred interval selected by Marron et al [2312] in their study. There is, as yet, no literature relating to the swallowing rate of people with Parkinson's pwPD.

The purpose of ~~this the~~ pilot study presented herein this paper was to explore the feasibility and acceptability of using this novel cueing device to help people with Parkinson's 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.

Figure 1 about here



## 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. a repeated measures design and looked at both between and within group differences. It first looked at a 2 (immediate vs delayed intervention groups) x 2 (measure times: first and second assessment point) design; then a 1 (all participants together once all had received intervention) x 3 (measure times: pre, post and 1 month after intervention) design (see fig 2 below). The 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 and Recruitment*

The 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

1  
2  
3 Sunderland, Gateshead and Newcastle were also included. Potential participants were  
4 identified by clinical staff and then contacted by a researcher via telephone with further  
5 information. Written information sheets were then sent to those who expressed sustained  
6 interest and, following a 1-week period, participants were visited in their homes to obtain  
7 informed consent.  
8  
9

10  
11  
12  
13  
14  
15 Inclusion criteria were (1) anyone with a diagnosis of Idiopathic Parkinson's (stages I-  
16 III in Hoehn and Yahr scale [2615]), in accordance with the UK Parkinson's Brain Bank criteria  
17 [2716], (2) an acknowledged daytime drooling problem, either observed by a clinical  
18 professional within a Parkinson's disease clinic or through patient self-report, and (3) an  
19 ability to understand and respond to the instructions given in the study. Exclusion criteria  
20 were (1) currently receiving pharmaceutical treatment for drooling, (2) insufficient dexterity  
21 with which to use the device.  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31

32  
33 A full case history was taken from each participant regarding their Parkinson's,  
34 drooling, and history of swallowing difficulties. The Mini Mental State Exam [28] and  
35 Montreal Cognitive Assessment Test were conducted for screening purposes of cognitive  
36 impairment [2917]. The Unified Parkinson's Disease Rating Scales II and III [3018] were  
37 conducted to gain an indication of overall disease state. All assessments were performed by  
38 the same researcher in the participant's own home.  
39  
40  
41  
42  
43  
44  
45  
46

47  
48 Participants were randomly allocated to either an immediate intervention group  
49 (n=17) or a delayed intervention group (n=11). This was to provide preliminary data for  
50 comparison of treatment vs no-treatment. The delayed group did not commence  
51 intervention until after they had completed a four-week period of no intervention. The  
52 randomization protocol was pre-determined using an online random number generator  
53 (<https://www.randomizer.org/>). The numbers were arranged into consecutive order  
54  
55  
56  
57  
58  
59  
60

1  
2  
3 creating a sequence for randomizing individuals (e.g. 1- immediate, 2-immediate, 3-  
4 delayed). If a participant left the study, their group assignment (immediate or delayed) was  
5 added to the end of this list to be filled by later recruits. We aimed for a 1:1 ratio, with a  
6 target recruitment of 30 participants (15 in each group). We fulfilled the capacity of the  
7 intervention group, but time restrictions meant that we were unable to complete a delayed  
8 start for the final two participants we had recruited. As such, we entered them into the  
9 intervention group leaving final numbers of 17 immediate and 11 delayed participants. This  
10 is a limitation of the study and is discussed further in the limitations section.  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23

## 24 **Measurements**

25  
26 The ~~Parkinson's Disease Questionnaire (PDQ-39) subtests for wellbeing, stigma and~~  
27 ~~communication [19]; the~~ 'saliva' subset of questions from the Radboud Oral Motor  
28 Inventory for Parkinson's Disease (ROMP-Saliva) [3120],; and the Unified Parkinson's  
29 Disease Rating Scale (UPDRS) 2.2. subtest for saliva [3018] were conducted with each  
30 participant at: one week before commencing use of the cueing intervention (assessment  
31 point 1), one week immediately after finishing the intervention (assessment point 2), and  
32 four weeks later at a follow up appointment (assessment point 3). For participants in the  
33 delayed start group, an additional ~~baseline assessment was collected 4 weeks prior to the~~  
34 ~~immediately pre-treatment assessment (assessment point 0). Figure 2 illustrates the time~~  
35 ~~line.~~  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

Figure 2 about here

1  
2  
3 ROMP-S is a validated tool [31] for use with pwPD. It is derived from the  
4 unvalidated Drooling Frequency and Severity Scale (DFSS) [32] originally drawn up for  
5 children with cerebral palsy but employed in several other populations. It was slightly  
6 modified for ROMP-S, in particular by adding the option to score that one is troubled by  
7 (perceived) accumulation of saliva without actually drooling. The nine items, rated on 5-point  
8 ordinal scales that describe gradations of drooling activity, cover day and night-time  
9 frequency and severity of drooling, effects on speech and eating and drinking, how frequently  
10 one has to wipe away saliva, limitations on daily activity and social participation, and overall  
11 impact.

12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24 UPDRS item 2.2. is a 5 point descriptive ordinal scale ranging from 0-4; no drooling  
25 (0), excess saliva but no loss (1), nighttime but not awake drooling (2), awake drooling but  
26 wiping not necessary (3), severe drooling with constant wiping/wet clothes (4). was collected  
27 4 weeks prior to assessment point 1 (assessment point 0) and was immediately followed by a  
28 4-week period of no intervention.

29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

At each assessment point, participants completed a 7-day drooling severity, frequency and duration diary [21]. Participants monitored their drooling over the course of one self-selected hour per day ~~when~~ that they would typically drool (e.g. after meal times, in the morning). Following Hauser et al (2004) [33] participants completed 100 mm visual analogue scales (VASs). They placed a cross on a 100mm line ~~Participants placed a cross on a 100mm line~~ (with 0mm being 'no problem' and 100mm 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. [34, 35, 22, 24]). ~~This same diary was then used to~~

1  
2  
3 collect daily self-report from participants during the 4-week intervention period. A  
4  
5  
6 visualization of this assessment schedule can be viewed in figure 2.  
7

8 Figure 2 about here  
9

10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26 Finally, an exit interview was carried out with each individual to gather qualitative feedback  
27  
28 on the participants' experiences. A semi-structured approach was taken to probe; a)  
29  
30 experiences of drooling before taking part in the study; b) experiences of drooling after  
31  
32 taking part in the study; and c) perceptions around the acceptability, worthwhileness and  
33  
34 effectiveness of the PDCue as a way to self-manage their drooling.  
35  
36  
37  
38

### 39 ***Intervention***

40  
41  
42 All participants were visited at home and received a verbal and practical tutorial on how to  
43  
44 use the cueing device. They were asked to use the device for one hour a day, for a total of  
45  
46 four weeks, at a time when drooling was an issue for them. Participants were asked to not  
47  
48 use the device during the hour that they were self-reporting their drooling on the daily  
49  
50  
51  
52 diary.  
53  
54  
55  
56  
57  
58  
59  
60

## ***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 and Kolmogorov-Smirnov tests [3623]. None of the variables examined were considered normally distributed. Data were summarized using statistics appropriate to the level of the data (e.g. median, inter-quartile 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 versus immediate intervention) and within group differences 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 (2006) [37]. Data was 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 ten 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 twenty-eight remaining participants (ten female). Compliance levels for filling out diaries varied. Out of a possible 6,699 diary entries 5,069 (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.

Table 1 about here

### 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 assessment. There were no significant changes in ROMP-S, UPDRS 2.2 or Diary reports (VAS measurements) for the no-intervention group during the 4 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

1  
2  
3 drooling improvement was borderline ( $p=0.06$ ) and perceived amount of time (duration) of  
4 drooling did not alter significantly.

5  
6  
7  
8 Table 2 presents data for the delayed start (at assessment points 0 and 1) and immediate  
9 start (at assessment points 1 and 2); representing a period of no intervention for the  
10 delayed start group and intervention for the immediate start group. The difference  
11 between the two groups in change from baseline score are compared across the outcomes  
12 investigated. Only the change in drooling severity from the diaries was significantly  
13 different between the two groups; this increased (i.e. got better) for the intervention group  
14 but stayed the same for the no intervention group.  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27

28 Table 2 about here

### 32 **Comparison of pre- and post-treatment scores across all participants**

33  
34  
35 Given that there appeared to be no placebo effect in the delayed intervention group during  
36 the no intervention phase (i.e. no significant improvement in any scores), once both groups  
37 had completed intervention their scores were combined to provide a larger group ( $n=28$ ) for  
38 comparison of pre- versus post treatment versus follow-up assessment  
39  
40  
41  
42  
43

44  
45 Between treatment termination and four week follow-up assessment four  
46 participants left the study (two moved on to Botox treatment immediately after the  
47 intervention ended, one experienced significant health decline and one moved abroad). This  
48 left 24 participants available for longer-term follow-up assessment comparisons. Outcomes  
49 are summarized in table 3.  
50  
51  
52  
53  
54

55  
56  
57 We compared measures before vs after intervention vs at follow up for all 28 participants  
58 (assessment points 1, 2 and 3 respectively). Twenty four participants were available for  
59  
60



1  
2  
3 follow up assessment one month following completion of the intervention. Of the other  
4  
5 four, two moved on to Botox treatment immediately after the intervention ended, one  
6  
7 experienced significant health decline and one moved abroad. Outcomes are summarized in  
8  
9 table 3. There were significant improvements as measured by the Unified Parkinson's  
10  
11 Disease Rating Scale item for drooling and saliva from pre-treatment to follow-up, and from  
12  
13 the self-reported diary for drooling severity (from pre-treatment to follow-up), frequency  
14  
15 (pre-treatment to post treatment, and pre-treatment to follow-up) and duration (pre-  
16  
17 treatment to post treatment, and pre-treatment to follow-up).

22  
23  
24  
25 Table 3 about here

26  
27  
28  
29  
30 ROMP-S ratings saw no significant change comparing scores at pre- versus at  
31  
32 termination of treatment. UPDRS 2.2. ratings showed a trend towards significance but were  
33  
34 still statistically non-significant. VAS patient perceptions of change in overall severity,  
35  
36 duration and frequency of drooling all evidenced significant improvements.

37  
38  
39  
40 To examine whether scores returned to baseline status once intervention finished,  
41  
42 baseline scores were compared with four weeks post treatment assessments. ROMP-S  
43  
44 demonstrated a move towards significance (but was still not statistically significant), whilst  
45  
46 UPDRS 2.2. now showed a significantly better status. The VAS ratings all showed strongly  
47  
48 significant improvements, including after adjustments for multiple testing. The findings  
49  
50 suggest maintenance or even improvement of status during the follow-up phase.

51  
52  
53  
54  
55  
56  
57 **Exit interviews**  
58  
59  
60

1  
2  
3 Twenty-seven participants were available for exit interview. One participant had a  
4  
5 significant health decline and was thus unavailable. Interviews lasted ~~ameann~~ average of  
6  
7 17:03 minutes (shortest 6:29-longest 36:37). Following procedures outlined in the  
8  
9 methodology section, There were a total of 26 thematic codes applied to the data. A total of  
10  
11 312 extracts of transcript were assigned these codes (ranging from 1 to 22 extracts per  
12  
13 code). A total of 45 higher level themes were then constructed from this qualitative data  
14  
15 analysis, which ~~will be~~ are summarised below.  
16  
17  
18  
19

20 The first theme to arise was the impact of drooling on the lives of the participants.  
21  
22 By far, the most discussed impact of drooling issues pre-treatment was embarrassment  
23  
24 (13/27), with several participants discussing emotional distress *“It really dominated my*  
25  
26 *life...it was most distressing, psychologically distressing...it clearly ruled my thinking...in that I*  
27  
28 *was always clasping this grubby handkerchief just in case”* (P14), and social withdraw, *“At*  
29  
30 *least once a day it would happen. I was out with company and it made me feel very*  
31  
32 *embarrassed. I tend to withdraw, avoid going out really. Eat on my own. I am pretty strict*  
33  
34 *about manners, and I thought it looked horrible”* (P12). Several participants (4/27) also  
35  
36 discussed physical discomfort that they experienced—constant wetness, changing of  
37  
38 handkerchiefs, painful sores around the mouth.  
39  
40  
41  
42  
43  
44

45 The second theme related to challenges around previous experiences of drooling  
46  
47 treatment. Several participants (3/27) had previous experience of Botox, however, for a lot  
48  
49 of participants (8/27), Botox was not an option they would have considered. These  
50  
51 participants discuss a lack of willingness to take additional medication; *“when I saw the*  
52  
53 *consultant they said I could go and have Botox, an injection. I didn’t want to take any more*  
54  
55 *drugs”* (P22). Botox was associated with words such as *“toxic”* (P25) and *“poison”* (P26) and  
56  
57 there was a clear preference for avoiding it, and other additional medication, if possible; *“I*  
58  
59  
60

1  
2  
3 *think if you can have something that avoids taking drugs I think that's great"* (P4). These  
4  
5 participants, unsurprisingly, preferred the PDCue as a behavioural treatment option; *"I'd*  
6  
7 *rather have the watch"* (CP7).  
8  
9

10 Theme three related the effect of the PDCue on drooling. Of the 27 interviewed  
11  
12 participants, there was a reported positive effect for 22, indicating that the majority of  
13  
14 participants successfully engaged with the intervention and found it to be a worthwhile  
15  
16 option for supporting the self-management of drooling. **Theme four then related**  
17  
18 **toParticipants also discussed** emotional benefits which arose as a result of the PDCue  
19  
20  
21 intervention, including improvements to self-esteem, confidence and feelings of control.  
22  
23  
24

25 The final theme related to reports of generalization and habituation. There were  
26  
27 several cases of participants reporting a generalization effect, wherein they felt an increase  
28  
29 in swallowing frequency was being carried over to times when they were not wearing the  
30  
31 PDCue (9/27). P4 said *"Even when I wasn't wearing the [PDCue] every now and again I think,*  
32  
33 *"Oh yes, you haven't swallowed. I need to swallow"*. P3 also noted *"even when I wasn't*  
34  
35 *wearing it I was much more conscious of it"*. Although unexpected, P26 also discussed an  
36  
37 improvement to his night time drooling *"I've hardly been drooling at all. No, I haven't. Even*  
38  
39 *during the night I haven't been"*.  
40  
41  
42  
43  
44

45 There were a small number of participants (3/27) however who reported becoming  
46  
47 habituated to the cues, e.g. P10 *"there were occasions when I had the watch on, I seemed to*  
48  
49 *have got so used to it that I didn't get any indication"*. However, these participants reported  
50  
51 a positive effect from the intervention, despite this habituation.  
52  
53

## 54 Discussion

55  
56  
57 This pilot study aimed to explore the feasibility, usability and acceptability of wearing a  
58  
59 wrist-worn **vibratory** cueing device to improve drooling in people with Parkinson's, and to  
60

1  
2  
3 establish whether there was evidence of an effect of the device on drooling severity,  
4 frequency and impact. A total of 28 Twenty-eight people completed a month-long  
5 intervention with the device, using it for an hour a day. These participants- Altogether they  
6 showed significant improvement on severity ( $p=0.031$ ), frequency ( $p=<0.001$ ), and duration  
7 ( $p=0.001$ ) of drooling when comparing results from the self-reported diary VASs collected  
8 pre and post intervention. These improvements were also seen to remain at follow up  
9 assessment 4 weeks post-treatment compared to their pre-intervention baseline. A significant  
10 improvement was also seen in the UPDRS 2.2 saliva subtest ( $p=0.010$ ) when comparing the  
11 pre-intervention and follow up assessment time points. Based on this we conclude there is  
12 some evidence to indicate that the device can be successful in improving saliva control, not  
13 only when wearing the PDCue but also when not using it or after intervention has been  
14 withdrawn.

15  
16  
17 Comments from the interviews confirm that people are disturbed by their drooling  
18 and that lesser drooling brings benefits for psychosocial well-being. Participants were  
19 satisfied with manipulating the device and found it acceptable to wear. They perceived the  
20 gains seen made wearing the device worthwhile. This is further reflected in comments  
21 participants made in the exit interviews concerning the perceived positive effects of the  
22 intervention device. Responses showed that 22/27 participants explicitly reported that they  
23 had noticed benefits to their drooling, with several stating that it was a preferable  
24 treatment option to other pharmaceutical interventions. In a larger trial it is unlikely that  
25 this interview-based approach could be undertaken at scale. The development of a  
26 questionnaire, drawing on the themes outlined from the qualitative data could be an  
27 approach to capturing data of this kind.

28  
29  
30 The lack of positive change in the delayed treatment group during their no-  
31 intervention phase suggests that change was not accounted for by a placebo effect from

1  
2  
3 being recruited to the study, being assessed, completing the diary exercise, nor from  
4 receiving information about drooling and drooling interventions in general. There was no  
5 improvement despite written (in the study information pack) and oral discussion that more  
6 frequent swallowing may benefit saliva loss. Whilst the present data suggest placebo effect  
7 does not play a significant role here, in a definitive trial a more active comparator condition  
8 should be introduced.

9  
10  
11  
12  
13  
14  
15  
16  
17  
18 Our results showed a measurable change in score in the UPDRS item for drooling,  
19 between baseline and 4 week follow-up, lending additional weight to the improved VAS  
20 responses that participants provided. However, we did not observe any significant  
21 differences in the ROMP-S overall score. This may indicate that the types or level of changes  
22 experienced over the intervention period were not sufficient for this tool to capture. It  
23 could be that employing an overall score across multiple dimensions rather than analyzing  
24 each item separately masked significant gains in some areas. For instance, there may have  
25 been no change in night time drooling score, or even, after a short period, no shift in overall  
26 impact. Nevertheless, frequency of having to wipe the mouth or perceived excessive saliva  
27 in the mouth may have altered, but these improvements failed to make a significant  
28 difference in overall score against the non-altered variables. A similar factor may be at work  
29 in the lesser (compared to VASs) sensitivity to change of the UPDRS saliva item, since this  
30 scale combines several features which might actually vary independently (e.g. day vs night  
31 drooling; perceived excess saliva in mouth; frequency vs severity) in one scale which might  
32 actually vary independently. Significant improvement in one sub-dimension may be missed  
33 if the other dimensions do not alter. This would mask changes in one of the sub-dimensions.  
34  
35 Further analyses of individual items prior to a definitive trial may aid in separating out which  
36 aspects of drooling are more or less susceptible to influence through cueing.  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

1  
2  
3 When comparing diary scores at baseline between groups (immediate vs delayed) we  
4 observed no significant differences, except on self-reported severity ( $p=0.010$ ), which  
5 reduced (i.e. got better) in the intervention group but stayed the same in the no  
6 intervention group. This result in itself deserves attention; in the delayed group, the other  
7 perceived measures collected from the diary (duration and frequency) increased (i.e. got  
8 worse), which may indicate that the process of completing the diary increased awareness of  
9 drooling. However, we had considered that this, paired with knowledge that increased  
10 swallowing frequency could reduce drooling (provided in the information sheet and during  
11 the informed consent procedure) might actually lead to an improvement in drooling in the  
12 delayed group. However, this was not the case, again furthering evidence that our results  
13 were not due to placebo effect.  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29

30 Another factor to consider for a larger trial is data completeness for the diaries. We  
31 had a 76% completion rate across the study. Although this is not dissimilar to other studies  
32 [e.g. 33], a larger trial A larger RCT would need to consider the time requirements of  
33 participants and the burden of the study to self-report, ~~although this is not dissimilar to~~  
34 ~~other studies [21].~~ We make the suggestion that completing the diary throughout the  
35 entirety of the study is not required. ~~O~~ only completing the diary for one week pre and  
36 post-intervention (and again at follow up) would be enough in a follow up trial, as these  
37 were the results that we eventually focused on in our analysis.  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48

49 Self-reported diaries are heavily used in clinical research as a way to monitor the progress of  
50 treatment and log patients' activities over time, without the requirement for a researcher to  
51 be present, despite longstanding reported issues with compliance (e.g. [34, 35]). Recent  
52 research into tools to support self-report in Parkinson's research has provided clear  
53 recommendations for improving practice, with Vega et al. finding 99% compliance with their  
54  
55  
56  
57  
58  
59  
60

1  
2  
3 paper-based tool measuring self-reported symptoms over several months with a small  
4 number of participants [38]. However, another solutions would be to introduce Considering  
5  
6 usage logs, collected automatically by the device, which would also provide an indication of  
7  
8 participant compliance without the need for the diary.  
9  
10

11  
12 ~~Self-reported diaries are heavily used in clinical research as a way to monitor the progress of~~  
13 ~~treatment and log patients' activities over time, without the requirement for a researcher to be~~  
14 ~~present, despite longstanding reported issues with compliance (e.g. [24]). Recent research~~  
15 ~~into tools to support self-report in Parkinson's research has provided clear recommendations~~  
16 ~~for improving practice, with Vega et al. finding 99% compliance with their paper-based tool~~  
17 ~~measuring self-reported symptoms over several months with a small number of participants~~  
18 ~~[25].~~  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28

29 Our results showed a measurable change in score in the Unified Parkinson's Disease  
30 Rating Scale subtest for drooling, which adds additional weight to the diary responses that  
31 participants provided. However, we did not observe any significant changes in the  
32 Parkinson's Disease Questionnaire-39 (PDQ-39) subtests for wellbeing, stigma and  
33 communication; nor did we observe any significant differences in the Radboud Oral Motor  
34 Inventory for Parkinson's disease saliva subtest. PDQ-39 is not designed to detect changes in  
35 drooling impact. The lack of significant changes may also indicate that the types or level of  
36 changes experienced over the intervention period were not sufficient for these tools to  
37 capture. Further work, prior to a larger RCT, is required to ensure that the impact of the  
38 PDCue intervention in a larger cohort of participants could be appropriately measured. The  
39 qualitative work that we completed as part of this pilot trial showed that, during exit  
40 interviews, 22 participants explicitly reported that they had noticed benefits to their  
41 drooling, with several stating that it was a preferable treatment option to other  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

1  
2  
3 pharmaceutical interventions. All participants felt the intervention and device were  
4 acceptable and usable. However, in a larger trial it is unlikely that this qualitative approach  
5 could be undertaken at scale. The development of a questionnaire, drawing on the themes  
6 outlined from the qualitative data could be an approach to capturing data of this kind. In  
7 addition, the interviews highlighted that a small number of participants noted habituation  
8 to the device. Whilst we did not set up the study to answer the question about habituation  
9 definitively, future research should look at whether this is a factor in whether the device is  
10 effective or not for an individual. In the present study, in as far it was highly effective for  
11 some, then we can assume that habituation is not an all-pervasive problem. However, in  
12 future we suggest longer follow-up times to look at possible wearing off effects.

13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

The concept of temporal cueing as a treatment for drooling has been built on previous work which has shown success in the domain of cueing for gait training for Parkinson's [7]; built upon observations 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 [8-10]. 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 [8]. 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.



1  
2  
3 The body of literature exploring cueing for drooling as a symptom is minimal, with  
4  
5 only small-scale preliminary work by Marks et al [2211] and Marron et al [2312] being the  
6  
7 only examples exploring this space. As such, our work builds upon this nascent body of  
8  
9 literature to provide additional evidence that cueing for drooling might be an effective way  
10  
11 to manage the symptom, with the qualitative aspects of our study additionally  
12  
13 demonstrating reports around increased feelings of control, confidence and self-esteem  
14  
15 post intervention. In addition, we build on our previous work [2413] to report that  
16  
17 acceptance and usability of our tool has been confirmed with a larger and more varied  
18  
19 group of participants over a longer period of time. Our cueing approach warrants further  
20  
21 exploration in a larger scale trial.  
22  
23  
24  
25  
26  
27  
28

## 29 **Study Limitations**

30  
31  
32 There are several provisos in interpreting the current data. Firstly, one assumes that  
33  
34 participants were wearing the devices as requested, for a designated hour each day.  
35  
36 However, we did not collect precise usage logs. In future work there would be benefit in  
37  
38 utilizing a more objective approach, e.g. through digital usage logs collected directly through  
39  
40 the device (i.e. using an accelerometer to provide data on when the device is switched on  
41  
42 and being used). Secondly, we asked participants to self-select an hour within which to self-  
43  
44 monitor their drooling, at times when drooling was a problem. Whilst participants may have  
45  
46 selected a self-perceived period of more susceptibility to drooling, it remains unclear how  
47  
48 severe their chosen hour might have been. Further laboratory-based work, employing  
49  
50 objective measures of physiological drooling (e.g. objective swallow frequency  
51  
52 measurement, or saturated gauze weight measurement) would add insight into whether or  
53  
54 not orally retained saliva objectively decreased through use of the device. This would also  
55  
56  
57  
58  
59  
60

1  
2  
3 remove, at least in laboratory conditions, the use of self-report diaries that may be open to  
4  
5 recall bias. For field-testing, employing devices capable of measuring swallowing events in  
6  
7 naturalistic situations (e.g. using an in-ear microphone) would be beneficial.  
8  
9

10  
11 Finally, although 30 participants was the sample size intended for this first stage  
12  
13 feasibility trial, we did not have matched numbers between the delayed and immediate  
14  
15 groups. The intentions was to have 15 participants in each, but time constraints meant that  
16  
17 we were unable to fully recruit to our delayed group (with 4 participants remaining). We  
18  
19 made a decision to include a final 2 participants in the study as immediate intervention  
20  
21 participants. Future studies implementing two treatment strands should not have this  
22  
23 problem in future work, however future researchers should also consider randomisation  
24  
25 approaches that allow for equal participant numbers throughout the recruitment process  
26  
27 (e.g. even vs odd participant numbers to each strand). In addition, whilst the results of our  
28  
29 pilot work delivered some positive outcomes, sufficient to suggest the cueing device may be  
30  
31 effective, a more definitive answer awaits a trial involving larger numbers in a more highly  
32  
33 powered study and with an active intervention comparator.  
34  
35  
36  
37  
38  
39  
40  
41

## 42 **Conclusions**

43  
44  
45  
46 This study has indicated that our cueing device was acceptable and usable, and that the  
47  
48 intervention could be a feasible first step for clinicians, before moving on to pharmaceutical  
49  
50 options, which have been shown to have potential complications. Whilst the next step of  
51  
52 this research will require a larger multi-center trial to elucidate whether these results are  
53  
54 replicable and clearer in a larger population, and to look at the characteristics of responders  
55  
56 vs non-responders to the treatment, the information presented within this paper has  
57  
58  
59  
60

1  
2  
3 provided important, preliminary data around the effect that the cueing intervention could  
4  
5 have and issues to address in the development of outcome measures.  
6  
7  
8

## 9 **Clinical messages**

- 12 • Providing a regular vibratory cue, through the PDCue device was shown to be an  
13  
14 effective treatment for reducing perceived drooling in the great majority of participants.
- 15 • Participants accepted PDCue and remained motivated to self-manage their drooling with  
16  
17 the device.
- 18 • Further studies are needed to confirm the beneficial effects that we observed and for  
19  
20 the refinement of outcome measures.  
21  
22  
23  
24  
25  
26

## 27 **Declarations**

28  
29  
30 **Conflicts of interest:** The Authors declare that there is no conflict of interest

31  
32  
33 **Funding:** This research was funded by the National Institute of Health Research, Research  
34  
35 for Patient Benefit program (RfPB PB-PG-0110-21326), and the EPSRC Digital Economy  
36  
37 theme Social Inclusion through the Digital Economy Research Hub (EP/G066019/1)  
38  
39

40 **Guarantor:** RM

41  
42  
43 **Contributorship:** RM, PO, RW and NM were all involved in the conception of the research  
44  
45 project and in researching the existing literature. RM, RW and NM were involved in protocol  
46  
47 development, with support from PO, KL and DJ. Ethical approval and patient recruitment  
48  
49 was conducted by RM and RW. Data capture and analysis was conducted by RM, KL, JV and  
50  
51 DJ. RM wrote the first draft of the manuscript. All authors reviewed and edited the  
52  
53 manuscript and approved the final version of the manuscript  
54  
55  
56  
57  
58  
59  
60

**Acknowledgments:** We would like to acknowledge the time and effort of the staff members who helped to identify participants for the study. We would also like to thank Keith Gray for his help with the statistical analysis of the data.

## References

1. Kalf JG, de Swart BJM, Borm GF, Bloem BR, Munneke M. Prevalence and definition of drooling in Parkinson's disease: a systematic review. *JNeurol.* 2009;256(9):1391-6.
2. Perez-Lloret S, Nègre-Pagès L, Ojero-Senard A, Damier P, Destée A, Tison F, et al. Oro-buccal symptoms (dysphagia, dysarthria, and sialorrhea) in patients with PD. *Eur JNeurology.* 2012;19(1):28-37.
3. Nienstedt JC, Buhmann C, Bihler M, Niessen A, Plaetke R, Gerloff C, et al. Drooling is no early sign of dysphagia in PD. *Neurogastroenterol Motil.* 2018;30(4).
4. Fereshtehnejad SM, Skogar O, Lökk J. Evolution of orofacial symptoms and disease progression in idiopathic PD: longitudinal data from the Jonkoping Parkinson registry. *Parkinsons Disease.* 2017.
5. Pedersen AML, Sørensen CE, Proctor GB, Carpenter GH. Salivary functions in mastication, taste and textural perception, swallowing and initial digestion. *Oral Dis.* 2018;24(8):1399-416.
6. Barbe AG, Ludwar L, Scharfenberg I, Hellmich M, Dano R, Barbe MT, et al. Circadian rhythms and influencing factors of xerostomia among Parkinson's disease patients. *Oral Dis.* 2019;25(1):282-9.
7. Saleh J, Figueiredo MAZ, Cherubini K, Salum FG. Salivary hypofunction: An update on aetiology, diagnosis and therapeutics. *Archives Oral Biology.* 2015;60(2):242-55.

- 1  
2  
3 8. Kalf J, Smit A, Bloem B, Zwarts M, Munneke M. Impact of drooling in PD. *JNeurol.*  
4  
5 2007;254(9):1227-32.  
6  
7
- 8 9. Leibner J, Ramjit A, Sedig L, Dai YF, Wu SS, Jacobson C, et al. The impact of and the  
9  
10 factors associated with drooling in Parkinson's disease. *Parks Rel Dis.*  
11  
12 2010;16(7):475-7.  
13  
14
- 15 10. Rajiah K, Maharajan MK, Yeen SJ, Lew S. Quality of life and caregivers' burden of PD.  
16  
17 *Neuroepidemiol.* 2017;48(3-4):131-7.  
18  
19
- 20 11. Nobrega AC, Rodrigues B, Torres AC, Scarpel RD, Neves CA, Melo A. Is drooling  
21  
22 secondary to a swallowing disorder in patients with PD? *Parks Rel Dis.*  
23  
24 2008;14(3):243-5.  
25  
26
- 27 12. Proulx M, de Courval FP, Wiseman MA, Panisset M. Salivary production in PD. *Mov*  
28  
29 *Disord.* 2005;20(2):204-7.  
30  
31
- 32 13. Kalf JG, Munneke M, van den Engel-Hoek L, de Swart BJ, Borm GF, Bloem BR, et al.  
33  
34 Pathophysiology of diurnal drooling in PD. *Mov Disord.* 2011;26(9):1670-6.  
35  
36
- 37 14. Reynolds H, Miller N, Walker R. Drooling in Parkinson's Disease: Evidence of a Role  
38  
39 for Divided Attention. *Dysphagia.* 2018;33(6):809-17.  
40  
41
- 42 15. Meningaud JP, Pitak-Arnnop P, Chikhani L, Bertrand JC. Drooling of saliva: A review  
43  
44 of the etiology and management options. *Oral Surgery, Oral Medicine, Oral*  
45 *Pathology, Oral Radiology, and Endodontics.* 2006. p. 48–57.  
46  
47
- 48 16. Hyson HC, Johnson AM, Jog MS. Sublingual atropine for sialorrhea secondary to  
49  
50 parkinsonism: A pilot study. *Mov Disord.* 2002;17(6):1318–20.  
51  
52
- 53 17. NICE. Parkinson's disease in adults: *NICE guideline NG71.* London, GB; 2017  
54  
55
- 56 18. Nieuwboer, A., Kwakkel, G., Rochester, L., et al. (2007). Cueing training in the home  
57  
58 improves gait-related mobility in Parkinson's disease: the RESCUE trial. *Journal of*  
59  
60

1  
2  
3 Neurology, Neurosurgery, and Psychiatry, 78(2), 134–40.

4 <http://doi.org/10.1136/jnnp.200X.097923>

5  
6  
7  
8 19. Sarma S V, Cheng ML, Eden U, Williams Z, Brown EN, Eskandar E. The effects of cues

9 on neurons in the basal ganglia in Parkinson's disease. Front Integr Neurosci

10 [Internet]. 2012 Jan [cited 2015 Aug 29];6:40. Available from:

11 [12 \[rez&rendertype=abstract\]\(http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=3405280&tool=pmcent\)](http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=3405280&tool=pmcent</a></u></p></div><div data-bbox=)

13  
14  
15  
16  
17  
18  
19  
20 20. Georgiou N, Iansek R, Bradshaw JL, Phillips JG, Mattingley JB, Bradshaw JA. An

21 evaluation of the role of internal cues in the pathogenesis of parkinsonian

22 hypokinesia. Brain [Internet]. 1993 Dec [cited 2015 Aug 29];116 ( Pt 6:1575–87.

23 Available from: <http://www.ncbi.nlm.nih.gov/pubmed/8293289>

24  
25  
26  
27  
28  
29  
30 21. Bötzel K, Schulze S. Self-initiated versus externally triggered movements. I. An

31 investigation using measurement of regional cerebral blood flow with PET and

32 movement-related potentials in normal and Parkinson's disease subjects. Brain

33 [Internet]. 1996 Jun [cited 2015 Aug 29];119:1045–8. Available from:

34 <http://www.ncbi.nlm.nih.gov/pubmed/8673482>

35  
36  
37  
38  
39  
40  
41  
42 22. Marks L, Turner K, O'Sullivan J, Deighton B, Lees A. Drooling in Parkinson's disease: a

43 novel speech and language therapy intervention. International journal of language &

44 communication disorders / Royal College of Speech & Language Therapists. 2001.

45  
46  
47  
48  
49  
50 23. Marron A, Robinson L, Walker R. Use of a metronome brooch reminder to improve

51 drooling problems in patients with Parkinsonism. Presented at the 9th national

52 conference - Multidisciplinary care in Parkinson's Disease and Parkinsonism from

53 science to practice - Royal College of Physicians,. London; 2004.

- 1  
2  
3  
4  
5  
6  
7  
8  
9  
10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60
24. McNaney R, Lindsay S, Ladha K, Ladha C, Schofield G, Ploetz T, Hammerla N, Jackson D, Walker R, Miller N, Olivier P. Cueing for drooling in Parkinson's disease. InProceedings of the SIGCHI conference on Human Factors in Computing Systems 2011 May 7 (pp. 619-622). ACM.
25. Afkari, S. (2007). Measuring frequency of spontaneous swallowing. Australasian Physical & Engineering Sciences in Medicine / Supported by the Australasian College of Physical Scientists in Medicine and the Australasian Association of Physical Sciences in Medicine, 30(4), 313–7. Retrieved from <http://www.ncbi.nlm.nih.gov/pubmed/18274071>
26. Hoehn MM, Yahr MD. Parkinsonism : onset, progression, and mortality. Parkinsonism: onset, progression, and mortality. Neurology [Internet]. 1967;17427. Available from: <http://www.neurology.org/content/17/5/427.citation>
27. Daniel SE, Lees AJ. Parkinson’s Disease Society Brain Bank, London: overview and research. J Neural Transm Suppl [Internet]. 1993 Jan [cited 2015 Aug 29];39:165–72. Available from: <http://www.ncbi.nlm.nih.gov/pubmed/8360656>
28. Folstein, M.F., Robins, L.N. and Helzer, J.E., 1983. The mini-mental state examination. Archives of general psychiatry, 40(7), pp.812-812.
29. Nasreddine, Z.S., Phillips, N.A., Bédirian, V., Charbonneau, S., Whitehead, V., Collin, I., Cummings, J.L. and Chertkow, H., 2005. The Montreal Cognitive Assessment, MoCA: a brief screening tool for mild cognitive impairment. Journal of the American Geriatrics Society, 53(4), pp.695-699.
30. Goetz CG, Tilley BC, Shaftman SR, Stebbins GT, Fahn S, Martinez-Martin P, Poewe W, Sampaio C, Stern MB, Dodel R, Dubois B. Movement Disorder Society-sponsored revision of the Unified Parkinson's Disease Rating Scale (MDS-UPDRS): Scale

1  
2  
3 presentation and clinimetric testing results. *Movement disorders*. 2008 Nov  
4  
5 15;23(15):2129-70.  
6  
7

8 31. Kalf J, Borm G, De Swart B, Bloem B, Zwarts M, Munneke M. Reproducibility and  
9  
10 validity of patient-rated assessment of speech, swallowing, and saliva control in  
11  
12 parkinson's disease. *Arch Phys Med Rehabil*. 2011;  
13

14  
15 32. Thomas-Stonell N, Greenberg J. Three treatment approaches and clinical factors in  
16  
17 the reduction of drooling. *Dysphagia*. 1988;3(2):73-8.  
18

19  
20 33. Hauser RA, Deckers F, Lehert P. PD home diary: Further validation and implications  
21  
22 for clinical trials. *Mov Disord*. 2004;19(12):1409-13.  
23

24  
25 34. Montgomery GK, Reynolds NC. Compliance, reliability, and validity of self-monitoring  
26  
27 for physical disturbances of Parkinson's disease. *The Parkinson's Symptom Diary. J*  
28  
29 *Nerv Ment Dis*. 1990;178(10):636-41.  
30

31  
32 35. Stone, A. A., Shiffman, S., Schwartz, J. E., Broderick, J. E., & Hufford, M. R. (2003).  
33  
34 Patient compliance with paper and electronic diaries. *Controlled Clinical Trials*.  
35  
36 [http://doi.org/10.1016/S0197-2456\(02\)00320-3](http://doi.org/10.1016/S0197-2456(02)00320-3)  
37

38  
39  
40 36. Shapiro SS, Francia RS. An approximate analysis of variance test for normality.  
41  
42 *Journal of the American Statistical Association*. 1972 Mar 1;67(337):215-6.  
43

44  
45 37. Braun, Virginia, and Victoria Clarke. 2006. Using thematic analysis in  
46  
47 psychology." *Qualitative research in psychology* 3, no. 2 (2006): 77-101. DOI:  
48  
49 <http://doi.org/abs/10.1191/1478088706qp063oa>  
50

51  
52 38. Vega, J., Couth, S., Poliakoff, E., et al. (2018). Back to Analogue: Self-Reporting for  
53  
54 Parkinson's Disease. In *Proceedings of the 2018 CHI Conference on Human Factors in*  
55  
56 *Computing Systems (CHI '18)*. ACM, New York, NY, USA, Paper 74, 13 pages. DOI:  
57  
58 <https://doi.org/10.1145/3173574.3173648>  
59  
60



- 1  
2  
3  
4  
5  
6  
7  
8  
9  
10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60
1. ——— Nóbrega AC, Rodrigues B, Torres AC, Searpel RD, Neves CA, Melo A. Is drooling secondary to a swallowing disorder in patients with Parkinson's disease? *Park Relat Disord*. 2008;14(3):243–5.
2. ——— Kalf J, De Swart B, Borm G, Bloem B, Munneke M. Prevalence and definition of drooling in Parkinson's disease: A systematic review. *Journal of Neurology*. 2009. p. 1391–6.
3. ——— Edwards, L. L., Pfeiffer, R. F., Quigley, E. M., Hofman, R., & Balluff, M. (1991). Gastrointestinal symptoms in Parkinson's disease. *Movement Disorders : Official Journal of the Movement Disorder Society*, 6(2), 151–156.  
<http://doi.org/10.1002/mds.870060211>
4. ——— Reynolds, H., Miller, N., Walker, R. The impact of divided attention in dual task conditions on drooling in Parkinson's Disease (PD): a pilot study [abstract]. *Movement Disorders*. 2017; 32 (suppl 2).  
<http://www.mdsabstracts.org/abstract/the-impact-of-divided-attention-in-dual-task-conditions-on-drooling-in-parkinsons-disease-pd-a-pilot-study/>. Accessed February 15, 2019.
5. ——— Meningaud JP, Pitak-Arnop P, Chikhani L, Bertrand JC. Drooling of saliva: A review of the etiology and management options. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontics*. 2006. p. 48–57.
6. ——— Hyson HC, Johnson AM, Jog MS. Sublingual atropine for sialorrhea secondary to parkinsonism: A pilot study. *Mov Disord*. 2002;17(6):1318–20.
7. ——— Nieuwboer, A., Kwakkel, G., Rochester, L., et al. (2007). Cueing training in the home improves gait-related mobility in Parkinson's disease: the RESCUE trial. *Journal of Neurology, Neurosurgery, and Psychiatry*, 78(2), 134–40.  
<http://doi.org/10.1136/jnnp.200X.097923>

- 1  
2  
3  
4  
5  
6  
7  
8  
9  
10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60
8. ——— Sarma S V, Cheng ML, Eden U, Williams Z, Brown EN, Eskandar E. The effects of cues on neurons in the basal ganglia in Parkinson's disease. *Front Integr Neurosci* [Internet]. 2012 Jan [cited 2015 Aug 29];6:40. Available from: <http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=3405280&tool=pmcentrez&rendertype=abstract>
9. ——— Georgiou N, Ianssek R, Bradshaw JL, Phillips JG, Mattingley JB, Bradshaw JA. An evaluation of the role of internal cues in the pathogenesis of parkinsonian hypokinesia. *Brain* [Internet]. 1993 Dec [cited 2015 Aug 29];116 ( Pt 6:1575–87. Available from: <http://www.ncbi.nlm.nih.gov/pubmed/8293289>
10. ——— Bötzel K, Schulze S. Self-initiated versus externally triggered movements. I. An investigation using measurement of regional cerebral blood flow with PET and movement-related potentials in normal and Parkinson's disease subjects. *Brain* [Internet]. 1996 Jun [cited 2015 Aug 29];119:1045–8. Available from: <http://www.ncbi.nlm.nih.gov/pubmed/8673482>
11. ——— Marks L, Turner K, O'Sullivan J, Deighton B, Lees A. Drooling in Parkinson's disease: a novel speech and language therapy intervention. *International journal of language & communication disorders / Royal College of Speech & Language Therapists*. 2001.
12. ——— Marron A, Robinson L, Walker R. Use of a metronome brooch reminder to improve drooling problems in patients with Parkinsonism. Presented at the 9th national conference—Multidisciplinary care in Parkinson's Disease and Parkinsonism from science to practice—Royal College of Physicians, London; 2004.
13. ——— McNaney R, Lindsay S, Ladha K, Ladha C, Schofield G, Ploetz T, Hammerla N, Jackson D, Walker R, Miller N, Olivier P. Cueing for drooling in Parkinson's disease. In *Proceedings of the SIGCHI conference on Human Factors in Computing*

- 1  
2  
3 Systems 2011 May 7 (pp. 619-622). ACM.  
4  
5  
6 14.—— Afkari, S. (2007). Measuring frequency of spontaneous swallowing.  
7  
8 Australasian Physical & Engineering Sciences in Medicine / Supported by the  
9  
10 Australasian College of Physical Scientists in Medicine and the Australasian  
11  
12 Association of Physical Sciences in Medicine, 30(4), 313–7. Retrieved from  
13  
14 <http://www.ncbi.nlm.nih.gov/pubmed/18274071>  
15  
16  
17 15.—— Hoehn MM, Yahr MD. Parkinsonism : onset, progression, and mortality  
18  
19 Parkinsonism: onset, progression, and mortality. *Neurology* [Internet]. 1967;17427.  
20  
21 Available from: <http://www.neurology.org/content/17/5/427.citation>  
22  
23  
24 16.—— Daniel SE, Lees AJ. Parkinson's Disease Society Brain Bank, London:  
25  
26 overview and research. *J Neural Transm Suppl* [Internet]. 1993 Jan [cited 2015 Aug  
27  
28 29];39:165–72. Available from: <http://www.ncbi.nlm.nih.gov/pubmed/8360656>  
29  
30  
31 17.—— Hoops S, Nazem S, Siderowf AD, Duda JE, Xie SX, Stern MB, et al. Validity  
32  
33 of the MoCA and MMSE in the detection of MCI and dementia in Parkinson disease.  
34  
35 *Neurology*. 2009;  
36  
37  
38 18.—— Goetz CG, Tilley BC, Shaftman SR, Stebbins GT, Fahn S, Martinez-Martin P,  
39  
40 Poewe W, Sampaio C, Stern MB, Dodel R, Dubois B. Movement Disorder  
41  
42 Society-sponsored revision of the Unified Parkinson's Disease Rating Scale  
43  
44 (MDS-UPDRS): Scale presentation and clinimetric testing results. *Movement*  
45  
46 *disorders*. 2008 Nov 15;23(15):2129-70.  
47  
48  
49 19.—— Peto V, Jenkinson C, Fitzpatrick R, Greenhall R. The development and  
50  
51 validation of a short measure of functioning and well being for individuals with  
52  
53 Parkinson's disease. *Qual Life Res*. 1995;  
54  
55  
56 20.—— Kalf J, Borm G, De Swart B, Bloem B, Zwarts M, Munneke M.  
57  
58 Reproducibility and validity of patient-rated assessment of speech, swallowing, and  
59  
60

- 1  
2  
3 saliva control in parkinson's disease. Arch Phys Med Rehabil. 2011;  
4  
5  
6 21.——Hauser, R.A., Deckers, F. and Leheret, P., 2004. Parkinson's disease home  
7  
8 diary: further validation and implications for clinical trials. *Movement*  
9  
10 *Disorders*, 19(12), pp.1409-1413.  
11  
12 22.——Montgomery GK, Reynolds NC. Compliance, reliability, and validity of self-  
13  
14 monitoring for physical disturbances of Parkinson's disease. The Parkinson's  
15  
16 Symptom Diary. J Nerv Ment Dis. 1990;178(10):636-41.  
17  
18 23.——Shapiro SS, Francia RS. An approximate analysis of variance test for  
19  
20 normality. Journal of the American Statistical Association. 1972 Mar 1;67(337):215-  
21  
22 6.  
23  
24 24.——Stone, A. A., Shiffman, S., Schwartz, J. E., Broderick, J. E., & Hufford, M. R.  
25  
26 (2003). Patient compliance with paper and electronic diaries. *Controlled Clinical*  
27  
28 *Trials*. [http://doi.org/10.1016/S0197-2456\(02\)00320-3](http://doi.org/10.1016/S0197-2456(02)00320-3)  
29  
30  
31 25.——Vega, J., Couth, S., Poliakoff, E., et al. (2018). Back to Analogue: Self-  
32  
33 Reporting for Parkinson's Disease. In Proceedings of the 2018 CHI Conference on  
34  
35 Human Factors in Computing Systems (CHI '18). ACM, New York, NY, USA, Paper 74,  
36  
37 13 pages. DOI: <https://doi.org/10.1145/3173574.3173648>  
38  
39  
40  
41 26.——  
42  
43 27.——  
44  
45 28.——  
46  
47 29.——  
48  
49 30.——  
50  
51 31.——  
52  
53 32.——  
54  
55 33.——  
56  
57  
58  
59  
60

1  
2  
3  
4  
5  
6  
7  
8  
9  
10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

34. —

35. —

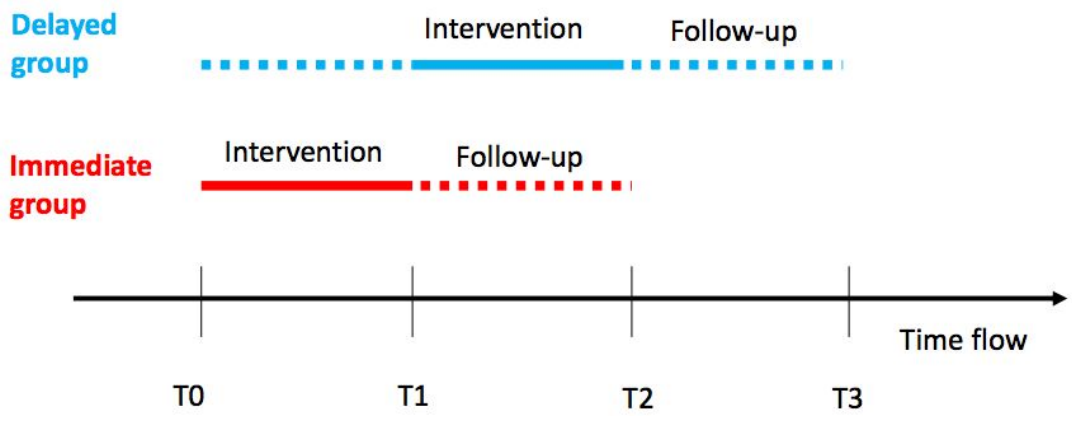
36.

For Peer Review



Figure 1: the cueing device

1  
2  
3  
4  
5  
6  
7  
8  
9  
10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60



For Peer Review

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



Assessment	Delayed group (n=11)		Intervention group (n=17)		Significance of <u>between group</u> difference <u>between the two groups</u> of change from <u>baseline</u>
	Assessment point <u>1 (T0)</u> <sup>0</sup> Median (IQR)	Assessment point <u>2 (T1)</u> <sup>1</sup> Median (IQR)	Assessment point <u>1 (T0)</u> Median (IQR)	Assessment point <u>2 (T1)</u> Median (IQR)	
<b>PDQ-39</b>			-	-	
Wellbeing	5 (4 to 13)	5 (4 to 8)	7 (3.5 to 12)	8 (3 to 10.5)	U = 77.0, z=0.784, p=0.433
Stigma	7 (1 to 9)	4 (1 to 6)	3 (1 to 7)	3 (0 to 6.5)	U = 66.0, z=1.311, p=0.190
Communication	4 (2 to 5)	4 (2 to 9)	4 (2.5 to 6.5)	6 (2 to 7)	U = 92.5, z=0.050, p=0.960
<b>ROMP- Saliva</b>	20 (17 to 25)	19 (17 to 30)	22 (16 to 23)	22 (17 to 25.5)	U = 83.0, z=0.497, p=0.619
<b>UPDRS 2.2</b> (Saliva and drooling subtest)	3 (1 to 3)	3 (1 to 3)	3 (3 to 4)	3 (2 to 4)	U = 69.0, z=1.212, p=0.225
<b>Drooling Diary</b>					
Severity	1 (0 to 4)	1 (1 to 5)	3 (1.5 to 5)	1 (0 to 2.5)	U = 39.5, z=2.575, p=0.010
Duration (No. minutes drooling occurred in one hour)	1 (0 to 5)	2 (0.5 to 10)	5 (1 to 11)	1 (0 to 4.5)	U = 63.0, z=1.440, p=0.150
Frequency (No. instances in one hour)	1 (0 to 4)	3 (1 to 4)	3 (1 to 4.5)	1 (0 to 3)	U = 54.0, z=1.876, p=0.061

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)
<b>PDQ-39</b>	-	-				
Wellbeing	4 (6 to 10)	6 (3.25 to 10)	7 (4.25 to 9.75)	Z = -1.032, p = 0.302	Z = -0.316, p = 0.752	
Stigma	3 (1 to 6.75)	3.5 (0 to 7)	2 (0 to 6)	Z = -0.835, p = 0.404	Z = -0.949, p = 0.343	
Communication	4 (2.25 to 7)	4.5 (2 to 6)	3 (1.25 to 5.75)	Z = -0.523, p = 0.601	Z = -1.150, p = 0.250	
<b>ROMP- Saliva</b>	20.5 (16 to 23.75)	20 (16.25 to 24.75)	17 (15 to 23.5)	Z = 0.275, p = 0.783	Z = 1.800, p = 0.072	
<b>UPDRS 2.2</b> (Saliva and drooling subtest)	3 (3 to 4)	3 (2 to 4)	2 (1 to 3)	Z = 1.801, p = 0.072	Z = 2.569, p = 0.010	
<b>Drooling Diary</b>						
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 one hour)	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 one hour)	4.18 (5.68)	1.80 (2.32)	1.16 (1.47)	Z = 3.982, p < 0.001	Z = 3.606, p < 0.001	