Developing an outcome measure for excessive saliva management in MND and an evaluation of saliva burden in Sheffield

Alexander J. McGeachan, Esther V. Hobson, Pamela J. Shaw & Christopher J. McDermott


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Developing an outcome measure for excessive saliva management in MND and an evaluation of saliva burden in Sheffield

ALEXANDER J. MCGEACHAN, ESTHER V. HOBSON, PAMELA J. SHAW & CHRISTOPHER J. MCDERMOTT

Sheffield Institute for Translational Neuroscience (SITraN), University of Sheffield, Sheffield, & Directorate of Neuroscience, Royal Hallamshire Hospital, Sheffield Teaching Hospitals NHS Foundation Trust, Sheffield, UK

Abstract
There are few studies providing evidence to guide the management of oropharyngeal secretion problems in motor neuron disease (MND). There is a lack of a suitable outcome measure for evaluating management strategies. We applied several potential outcome measures for assessing excessive secretions to patients with MND who attended the Sheffield Care and Research Centre for Motor Neurone Disease between 21 November 2012 and 15 May 2013. These measures were the CSS-MND, a symptom rating scale, and the Drool and Wipe quotient, which were designed to semi-objectively measure patients’ drooling. Of the 143 patients seen in clinic during the study period, 58 had symptoms of excessive secretions, and of whom 50 agreed to participate in the study. Semi-objective measures failed to effectively identify patients complaining of secretion problems. The CSS-MND had a relatively low internal consistency (Cronbach’s alpha 0.539; n = 50); however, analysis of the inter-item correlations suggested the appearance of low internal consistency was because the scale was measuring a variety of saliva related symptoms that did not necessarily influence each other. The scale correlated well with patient reported symptom impact ($r = 0.673$, $n = 50$). In conclusion, the CSS-MND would be a useful outcome measure in studies assessing the management of oropharyngeal secretion problems.

Key words: Outcome measures, secretion management, excessive saliva, sialorrhoea, amyotrophic lateral sclerosis, motor neurone disease

Introduction
Motor neuron disease (MND) is a neurodegenerative disorder that variably affects the muscles of the limbs, trunk, bulbar region and respiratory system (1). Half of the patients who develop this disease die within 30 months of their first symptom (2,3). Approximately 50% of MND patients suffer from excessive oropharyngeal secretions of which an estimated 42% are poorly controlled (4).

Evidence from therapeutic trials is limited, due to lack of binding and insufficient sample sizes (5) and a lack of a validated uniformly used, outcome measure (6–10). Consequently, there is little evidence guiding clinicians’ management choices (5).

Several outcome measures have been used to evaluate secretion problems in MND, including patient reported outcome measures (PROMs) or objective outcome measures. Objective approaches often evaluate saliva volume. Such methods include spit collection cups, weighing cotton wool, oral suctioning, counting tissues, drool frequency and radiotracer uptake, which can be time-consuming and hard to administer (5,11,12). They do not reflect the burden caused by secretion problems, which are also influenced by the consistency of secretions, disturbance of sleep, fluctuations during the day and social and physical consequences. In patients with Parkinson’s disease it has been shown that saliva production and problems with excess saliva do not correlate (11).

Patient reported outcome measures (PROMs) may provide a more accurate way of reflecting the distress caused by secretions, but none of the scales used in studies to date has been validated. In a randomized controlled trial (RCT) of salivary gland botulinum toxin injections in MND a ‘global impression of change’ scale was used as the primary outcome (12). While PROMs may reflect the impact that
the treatment had on the patients’ secretion problem, however, there are concerns that these types of scales are too subjective and susceptible to bias (5).

Two short tools, the Oral Secretion Scale and the Saliva Scoring Scale, both developed for Parkinson’s disease, had good inter- and intra-rater reliability when measuring saliva in patients with MND (13), but only reflected the frequency, volume and severity of drooling. There are other factors that may heavily influence the impact a secretion problem has on a patient; in particular, posterior collections of saliva may be an important factor to consider in an outcome measure (14), supporting the idea that a more detailed scale is required.

A further PROM has recently been validated to evaluate excess saliva in Parkinson’s disease. The Sialorrhoea Clinical Scale for Parkinson’s disease (SCS-PD) assesses saliva related discomfort by rating the severity and frequency of a range of symptoms that can be caused by excessive saliva, not just drooling (11).

Alternatively, a semi-objective patient reported measure (such as reporting the number of tissues used to catch saliva) could be adopted to avoid some of the difficulties with subjective measurements (15).

We aimed to evaluate several different measures of sialorrhoea, in order to determine the most appropriate way of evaluating therapies for the problem of oropharyngeal secretions in MND.

Methods and patients

Each patient participating in the study completed four scales: a clinical saliva scale for MND (CSS-MND), Drool Quotient (DQ), Wipe Quotient (WQ), and a Likert scale assessing saliva severity.

The clinical saliva scale for MND (CSS-MND) was adapted for MND from the SCS-PD to evaluate problems caused by excessive saliva in MND (Supplementary Appendix – which is only available in the online version of the journal. Please find this material with the following direct link to the article: http://www.informahealthcare.com/doi/abs/10.3109/21678421.2014.951942) (11). Two items were added to assess the impact of coughing as a result of excessive saliva spilling into the patient’s throat and the impact of excessive saliva on the use of non-invasive ventilation (NIV). This scale was abbreviated to create a modified version to include only the scores from two items, one which assessed drooling frequency and one which assessed drooling severity (Supplementary Appendix – which is only available in the online version of the journal. Please find this material with the following direct link to the article: http://www.informahealthcare.com/doi/abs/10.3109/21678421.2014.951942: Item C + Item E). This gave an extent of drooling scale score, similar to those used in previous studies assessing the management of excessive saliva (7).

The Drool Quotient (DQ) is the number of 15-s intervals in which saliva had spilled onto a patient’s lips or chin during a 10-min period. This method was previously successfully used in a study in patients with cerebral palsy (10). The Wipe Quotient (WQ) is the number of times a patient has to wipe saliva from their lips in a 10-min period.

The CSS-MND was completed by the patient and when necessary the carer or the researcher wrote for the patient. The DQ and WQ were recorded by the researcher. Additionally, the patient’s perspective on the impact of a patient’s saliva problem was evaluated using a modified Likert scale, ranging from ‘mild’ to ‘severe’. Patient reported efficacy of the treatment for these problems was also assessed on a modified Likert scale.

Descriptive statistics were used to identify the incidence and impact of secretion problems. We evaluated the Spearman’s rank correlation between excessive saliva outcome measures and patient reported saliva severity measured on the modified Likert scale (16). The internal consistency of the CSS-MND was assessed with the Cronbach’s alpha coefficient. The inter-item correlations were also evaluated (17,18).

Results

Patient perceptions of the severity of their saliva problem and the efficacy of their current treatment

The study was completed in 21 neuromuscular diseases clinics between the 21 November 2012 and the 15 May 2013; a total of 143 MND patients were seen in these clinics. Fifty-eight (41%) of these patients reported a problem with excessive secretions, 50 of whom agreed to participate in the study. The group included 27 males (54%) and 23 females (46%). The median age of these patients was 65 years, inter-quartile range 57–74 years. The site of onset was limb for 22 (44%) patients, bulbar in 27 (54%), and respiratory in one (2%). Four (8%) of the cases were familial MND.

Of these 50 patients, 31 (62%) reported moderate to severe symptoms (Figure 1). Twenty-four patients were currently receiving treatment for their excessive saliva. Only six (25%) reported their medication to be very effective and five (20%) reported that their medication had no effect on their symptoms (Figure 2).

Figure 1. Impact of excess saliva problem as reported by patients on a modified Likert scale (n = 50).
Variation of saliva related symptoms

The clinical saliva score for MND (CSS-MND) highlighted the variety of symptoms from which patients were suffering. This scale comprised eight items, each item ranging from 0 (no problem) to 3 (severe problems). Variable combinations of problems were experienced. Four patients reported only suffering from excessive saliva during the night, in bed. Twenty-one patients were using NIV, five (24%) of whom had compliance issues as a result of their saliva problem (Table I).

The Clinical Saliva Scale for Motor Neuron Disease (CSS-MND)

Fifty patients with excessive secretions completed the CSS-MND. The time taken to complete was never longer than the 10 min taken to observe for wiping or drooling.

CSS-MND acceptability. Patients were asked to report how acceptable the CSS-MND was to complete on a 5-point scale ranging from not acceptable at all (1) to very acceptable (5). The scale was deemed acceptable, with a mean score of 3.94. The 5-point Likert scale for assessing saliva severity was also deemed acceptable with a mean score of 3.74.

Abbreviated CSS-MND. The items evaluating the effect of excessive saliva on speech and the use of NIV were not applicable to every patient (patients who were anarthric or not using non-invasive ventilation). As a result the analysis for internal consistency was performed on the full scale but also with abbreviated versions of the scale:

- The full CSS-MND, including all items – completed by 17 patients
- Abbreviated CSS-MND 1, which excluded the items relating to both speech and NIV – completed by 50 patients
- Abbreviated CSS-MND 2, which excluded the item relating to speech – completed by 21 patients
- Abbreviated CSS-MND 3, which excluded the item relating to the use of NIV – completed by 42 patients

The Cronbach’s alpha is a coefficient of the correlation between items in a scale and is used to evaluate internal consistency. It is commonly accepted that a Cronbach’s alpha value of between 0.7 and 0.8 indicates sufficient internal consistency (17):

Scale 1, all items: Cronbach’s alpha 0.624 ($n = 17$)
Scale 2, questions about speech and NIV removed: Cronbach’s alpha 0.539 ($n = 50$)
Scale 3, question about speech removed: Cronbach’s alpha 0.649 ($n = 21$)
Scale 4, question about NIV removed: Cronbach’s alpha 0.554 ($n = 42$)

When the CSS-MND was developed from the SCS-PD two items were added to assess coughing (G) and hindrance of the use of NIV (H), in order to evaluate the impact of posterior saliva collection in the mouth. Removing these items increased the Cronbach’s alpha value to 0.681 ($n = 42$).

Additional scales

Extent of drooling. Additionally, the scores of item C (assessing drooling severity) and item E (assessing drooling frequency) were combined to generate a scale that assessed the overall extent of the drooling problem – completed by 50 patients.

<table>
<thead>
<tr>
<th>Item</th>
<th>0</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>Total</th>
<th>Proportion with problem</th>
</tr>
</thead>
<tbody>
<tr>
<td>A</td>
<td>4</td>
<td>15</td>
<td>19</td>
<td>12</td>
<td>50</td>
<td>46 (92%)</td>
</tr>
<tr>
<td>B</td>
<td>22</td>
<td>6</td>
<td>19</td>
<td>3</td>
<td>50</td>
<td>28 (56%)</td>
</tr>
<tr>
<td>C</td>
<td>12</td>
<td>6</td>
<td>6</td>
<td>26</td>
<td>50</td>
<td>38 (76%)</td>
</tr>
<tr>
<td>D</td>
<td>17</td>
<td>16</td>
<td>8</td>
<td>1</td>
<td>42*</td>
<td>25 (60%)</td>
</tr>
<tr>
<td>E</td>
<td>15</td>
<td>14</td>
<td>17</td>
<td>4</td>
<td>50</td>
<td>35 (70%)</td>
</tr>
<tr>
<td>F</td>
<td>20</td>
<td>9</td>
<td>15</td>
<td>6</td>
<td>50</td>
<td>30 (60%)</td>
</tr>
<tr>
<td>G</td>
<td>17</td>
<td>8</td>
<td>12</td>
<td>14</td>
<td>50</td>
<td>33 (66%)</td>
</tr>
<tr>
<td>H</td>
<td>13</td>
<td>3</td>
<td>3</td>
<td>2</td>
<td>21*</td>
<td>8 (38%)</td>
</tr>
</tbody>
</table>

N/A → *Eight of the recruited patients could not speak and 29 were not using NIV. As a result the items D (hindrance of speech) and H (hindrance to the use of non-invasive ventilation) were not applicable in eight and 29 patients, respectively.
Saliva management in MND

Patient reported saliva problem severity, as measured on the 5-point Likert scale, was taken as a measure of saliva impact.

The study aimed to evaluate whether the CSS-MND was a better indication of ‘saliva impact’ (measured on a Likert scale) than the two-item ‘extent of drooling’ scale. To do this, analysis of strength of the correlation of the CSS-MND scale with the saliva impact Likert scale was undertaken. The abbreviated CSS-MND scales and the two-item extent of drooling scale correlated against saliva impact reported on Likert scale, and a saliva impact scale (Likert scale).

Table II. Results of a Spearman’s rank correlation comparing how well the CSS-MND and the Extent of drooling scale correlated with a saliva impact scale (Likert scale).

<table>
<thead>
<tr>
<th>Comparison</th>
<th>Description</th>
<th>Correlation Coefficient (r)</th>
<th>Proportion of Variance (R^2)</th>
<th>Significance (p)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Complete CSS-MND correlated against saliva impact reported on Likert scale</td>
<td>0.465</td>
<td>0.216</td>
<td>0.060</td>
</tr>
<tr>
<td>2</td>
<td>Abbreviated CSS-MND (without NIV or speech items) correlated against saliva impact reported on Likert scale</td>
<td>0.673</td>
<td>0.453</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>3</td>
<td>Abbreviated CSS-MND (without speech item) correlated against saliva impact reported on Likert scale</td>
<td>0.525</td>
<td>0.276</td>
<td>0.015</td>
</tr>
<tr>
<td>4</td>
<td>Abbreviated CSS-MND (without NIV item) correlated against saliva impact reported on Likert scale</td>
<td>0.608</td>
<td>0.370</td>
<td>&lt;0.001</td>
</tr>
</tbody>
</table>

Comparisons 1 and 3 only included the data of 17 and 21 patients, respectively. As a result, the information available for analysis was limited. The full results are summarized in Table II.

The strength of the correlation between each of the items in the CSS-MND (interclass correlations) was analyzed. This highlighted that only two relationships could be deemed strong (r > 0.6). These relationships were frequency of drooling and severity of drooling, and social embarrassment and frequency of drooling. Particularly weak correlations of r = 0.03 were seen between social embarrassment (Item F) and saliva problems at night (Item B) (r = 0.03), and r = 0.01 between coughing on saliva (Item G) and drooling frequency (Item E).

The full inter-item correlation matrix is provided in Tables III–VI.

### Table III. Inter-item correlation matrix, excluding speech and NIV items; n = 50.

<table>
<thead>
<tr>
<th>Item A</th>
<th>Item B</th>
<th>Item C</th>
<th>Item D</th>
<th>Item E</th>
<th>Item F</th>
<th>Item G</th>
</tr>
</thead>
<tbody>
<tr>
<td>Timing of excessive saliva</td>
<td>1.00</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Excess saliva at night</td>
<td>0.23</td>
<td>1.00</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Severity of drooling</td>
<td>0.32</td>
<td>0.11</td>
<td>1.00</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Frequency of drooling</td>
<td>0.08</td>
<td>0.15</td>
<td>0.64</td>
<td>1.00</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Social embarrassment</td>
<td>0.15</td>
<td>0.03</td>
<td>0.42</td>
<td>0.60</td>
<td>1.00</td>
<td></td>
</tr>
<tr>
<td>Coughing/Aspiration</td>
<td>0.07</td>
<td>-0.12</td>
<td>-0.10</td>
<td>0.01</td>
<td>0.11</td>
<td>1.00</td>
</tr>
</tbody>
</table>

### Table IV. Inter-item correlation matrix for speech item; n = 42.

<table>
<thead>
<tr>
<th>Item A</th>
<th>Item B</th>
<th>Item C</th>
<th>Item D</th>
<th>Item E</th>
<th>Item F</th>
<th>Item G</th>
<th>Item H</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hindrance of speech</td>
<td>-0.10</td>
<td>0.20</td>
<td>0.37</td>
<td>1.00</td>
<td>0.52</td>
<td>0.40</td>
<td>0.09</td>
</tr>
</tbody>
</table>

### Table V. Inter-item correlation matrix for NIV item; n = 21.

<table>
<thead>
<tr>
<th>Item A</th>
<th>Item B</th>
<th>Item C</th>
<th>Item D</th>
<th>Item E</th>
<th>Item F</th>
<th>Item G</th>
<th>Item H</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hindering the use of NIV</td>
<td>0.02</td>
<td>0.58</td>
<td>0.24</td>
<td>0.46</td>
<td>0.30</td>
<td>0.38</td>
<td>1.00</td>
</tr>
</tbody>
</table>
Table VI. Inter-item correlation matrix between Speech and NIV items; n = 17.

<table>
<thead>
<tr>
<th>Item D: Hindrance of speech</th>
<th>Item H Hindering the use of NIV</th>
</tr>
</thead>
<tbody>
<tr>
<td>0.54</td>
<td></td>
</tr>
</tbody>
</table>

Objective outcome measures for excessive saliva

Two observational semi-objective outcome measures were used to quantify saliva excess: the Wipe Quotient (WQ) and the Drool Quotient (DQ). Only eight (16%) of the 50 patients reporting a saliva problem scored above 0 on the WQ, and one patient scored above 0 on the DQ.

Discussion

This study demonstrated the CSS-MND to be the most acceptable and reliable PROM in MND to reflect the various difficulties encountered by patients with secretion related symptoms.

The study has highlighted that the incidence of saliva problems among the MND patients attending the clinic was high (41%), yet only 25% of patients receiving treatment for excessive saliva reported that their treatment was very effective, indicating the need for studies to better assess management regimens for excessive saliva. The present study demonstrates that semi-objective tools (the Drool Quotient and Wipe Quotient) were highly insensitive for assessing problems with excessive saliva in the MND population.

When assessing the PROM applicable to all of the participants (comparison 2), the strong correlation between the Likert scale measuring saliva severity and the abbreviated CSS-provides evidence that the CSS-MND is face valid, in that it is a good indicator of the impact that excessive saliva is having on a patient. Moreover, this correlation was stronger than the correlation between patient reported saliva impact and the extent of drooling measure (Item C + Item E), suggesting the CSS-MND is a better reflection of saliva impact than a scale that assesses only drooling frequency and severity, such as those used in previous trials investigating saliva management (7).

When the original scale was evaluated in Parkinson’s disease, the Cronbach’s alpha value was within the acceptable range at 0.78 (11). There were two additional items in the CSS-MND compared to the SCS-PD, which assessed coughing on saliva (G) and saliva hindering the use of NIV (H). Item G correlated weakly with all other items, and item H correlated weakly with five of the seven items. When these two items were removed, the CSS-MND’s internal consistency improved, suggesting that these items were impacting on the internal consistency. This is likely because problems are related to the posterior spillage of saliva and so will be less related to items assessing excessive saliva in the mouth.

While most of the items in the questionnaire poorly correlated with each other, these weak correlations appeared clinically explainable. For example, the item assessing problems with embarrassment and the item assessing saliva problems at night did not correlate ($r = 0.03$), which can be explained by the fact that embarrassment requires social interaction that is likely to only take place during the day. There was correlation between the items that would be expected to correlate, such as drooling severity and drooling frequency ($r = 0.64$). This variable combination of symptoms highlights that the impact of oropharyngeal secretion problems is complex, affects patients in a variety of ways and is not simply a matter of quantity. Low Cronbach’s alpha values for the CSS-MND seem to reflect that this scale is attempting to measure a variety of symptoms associated with excess saliva that are not necessarily closely related.

Assessing multiple saliva related symptoms would provide detailed assessment of saliva related discomfort, and may provide more insight into the relative merits of therapies for saliva control. This promotes the use of the CSS-MND over simple drooling frequency and severity scales.

The CSS-MND provides a good evaluation of saliva related discomfort. However, as with any PROM there are concerns about susceptibility to bias (5). Additionally, due to the lack of recruited patients who were using NIV, insufficient data were collected to properly evaluate the versions of the CSS-MND including this item.

Conclusion

Excessive secretions cause a variety of independent problems in MND patients and each patient can have variable saliva related problems. This study shows that the CSS-MND is an acceptable, easy-to-use tool that assesses multiple independent aspects of oropharyngeal secretion problems.

The CSS-MND is face valid in that it was a good reflection of the impact that saliva problems were having on patients and provided a better reflection of saliva burden than a scale which only assessed the extent of drooling. Moreover, the multi-symptom assessment provided by the CSS-MND would provide a detailed analysis of the relative merits of the various management options.

However, amendments do need to be made to the CSS-MND to ensure that the questions reflect only problems related to excessive saliva. The key areas for the development of this scale are:

- Improving question clarity to ensure patients are only reporting problems caused by excessive saliva
- After ensuring the multi-symptom scale is assessing problems with excessive saliva, assessment of the test re-test reliability is necessary (24)
• Assessment of the scale including the NIV item with sufficient numbers of subjects

It would also be useful to add a subscale for assessing problems with thickened secretions in the mouth or the throat, so that the benefits of excessive saliva treatment can be considered against the detrimental effects of developing uncomfortable thickened secretion problems.

A suggestion for the content of the amended CSS-MND is provided in the Supplementary Appendix – which is only available in the online version of the journal. Please find this material with the following direct link to the article: http://www.informahealthcare.com/doi/abs/10.3109/21678421.2014.951942.

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

References

Supplementary material available online