Acoustic evaluation of nasality in cerebellar syndromes

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Abstract

Although previous studies have reported the occurrence of velopharyngeal incompetence connected with ataxic dysarthria, there is a lack of evidence related to nasality assessment in cerebellar disorders. This is partly due to the limited reliability of challenging analyses and partly due to nasality being a less pronounced manifestation of ataxic dysarthria. Therefore, we employed 1/3-octave spectra analysis as an objective measurement of nasality disturbances. We analyzed 20 subjects with multiple system atrophy (MSA), 13 subjects with cerebellar ataxia (CA), 20 subjects with multiple sclerosis (MS) and 20 healthy (HC) speakers. Although we did not detect the presence of hypernasality, our results showed increased nasality fluctuation in 65% of MSA, 43% of CA and 30% of MS subjects compared to 15% of HC speakers, suggesting inconsistent velopharyngeal motor control. Furthermore, we found a statistically significant difference between MSA and HC participants (p<0.001), and significant correlation between the natural history cerebellar subscore and neuroprotection in Parkinson plus syndromes – Parkinson plus scale and nasality fluctuations in MSA (r=0.51, p<0.05). In conclusion, acoustic analysis showed an increased presence of abnormal nasality fluctuations in all ataxic groups and revealed that nasality fluctuation is associated with distortion of cerebellar functions.

Index Terms: Ataxic dysarthria, nasality analysis, velopharyngeal control, cerebellum, multiple system atrophy, spinocerebellar ataxia, multiple sclerosis

1. Introduction

The cerebellum contains 80% of all neurons in the brain and has a crucial influence on motor control of every human [1]. The movement-related functions of the cerebellum consist of coordination, precision and accurate timing [2]. Therefore, damage to the cerebellum results in severe disruption of motor functions manifesting as unsteady gait, loss of balance and loss of fine motor coordination. The primary etiologies associated with cerebellar dysfunction include degenerative diseases such as spinocerebellar ataxia (SCA) and multiple system atrophy (MSA), as well as demyelinating diseases such as multiple sclerosis (MS). These two etiologies are closely related to ataxic dysarthria and together are the primary cause of 54% of all ataxic dysarthria cases reported [3].

Ataxic dysarthria is a speech disruption characterized by the presence of three clusters of deviant speech: articulatory inaccuracy, prosodic excess and phonatory-prosodic insufficiency [3]. Among other distortions, these deviant clusters may lead to inconsistent velopharyngeal closure, which is manifested as a variable level of nasality [3]. A study by Mariën et al. incorporates irregular nasality including both hyponasality and hypernasality between four articulatory manifestations of ataxic dysarthria [4]. In addition, previous studies suggest the presence of velopharyngeal incompetence in multiple sclerosis and ataxic dysarthria of various etiologies [5, 6].

Currently, the gold standard of nasality measurement is perceptual assessment, despite its questionable inter-rater and intra-rater reliability and challenging nature due to the combination of various manifestations in dysarthrias [7–9]. To overcome limited reliability of perceptual assessment, several more objective methods including x-ray tracing, sonography, nasometry, comparison of nasal and oral acoustic outputs and the Horii Oral-Nasal Coupling Index have been developed [10–13]. Some of these methods are invasive or at least put certain restrictions on the patient.

Among objective methods, acoustic measurement is the least invasive and most affordable approach. The two best-known objective measurements of nasality are the voice low tone high tone ratio (VLHR) and the 1/3-octave spectra analysis [14, 15]. The 1/3-octave spectra was originally developed for hypernasality assessment in children with cleft palate and subsequently verified on adult patients after maxillectomy, whereas VLHR was designed for adult speakers [14-16]. Between these two methods, the 1/3-octave spectra analysis has been shown to be more sensitive [17]. Furthermore, the 1/3-octave spectra analysis has been successfully applied to dysarthric speakers [18, 19].

The aim of the present study was to analyze the possible presence of inconsistent velopharyngeal control in participants with MSA, cerebellar ataxia (CA) and MS using the 1/3-octave spectra method, and to examine relationships between disrupted velopharyngeal control and other motor manifestations. We hypothesized that the presence of cerebral damage may lead to an increased presence of the nasality fluctuation as a result of imprecise soft palate coordination.
2. Methods

2.1. Subjects

A total of 74 Czech native speakers were recruited for analysis and were divided into four groups according to diagnosis including CA, MSA, MS and healthy control (HC) speakers. Some participants were recruited for previous studies aimed at investigating speech characteristics in patients with atypical parkinsonian syndromes and cerebellar ataxia [20-22].

The MSA group consisted of 9 men and 11 women with an average age of 60.8 ± 6.5 (range 45–71) years and disease duration 3.9 ± 1.4 years (2–7). Eighteen participants were diagnosed with the parkinsonian subtype whereas the remaining two were diagnosed as the cerebellar type. The MSA participants were rated according to the natural history and neuroprotection in Parkinson plus syndromes – Parkinson’s plus scale (NNIPPS; [23]) and their average overall score reached 72.3 ± 22.8 (35–123). Furthermore, the NNIPPS cerebellar subscore was assessed on 9 patients reaching a non-zero score, and the entire MSA group scored 3.95 ± 6.17 (0–22).

The CA group consisted of 7 men and 6 women with an average age of 53.9 ± 12.1 (34–71) years and a disease duration of 9.9 ± 6.8 (1–21) years. Six of the patients were diagnosed with SCA (one SCA1, four SCA2 and one SCA8) and genetically validated, while the remaining eight patients were diagnosed with idiopathic late-onset cerebellar ataxia. The CA participants were rated according to the scale for the assessment and rating of ataxia (SARA; [24]), and the average overall score reached 13.8 ± 4.4.

The MS group consisted of 7 men and 13 women with an average age of 44.1 ± 9.2 (27–62) years and a disease duration of 17.7 ± 7.9 (6–31) years. The MS participants were rated according to the expanded disability status scale (EDSS; [25]), and their average overall score reached 5.0 ± 1.0 (4–7). All twenty participants manifested deficits in cerebellar functions and were predominantly ataxic (EDSS cerebellar subscore >= 3). The MS group reached an EDSS cerebellar function score of 3.2 ± 0.4 (3–4).

The HC group consisted of 9 men and 11 women with an average age of 60.7 ± 6.6 (45–71) years. None of the HC participants had a history of neurological or speech disorders.

The study was approved by the Ethics Committee of the General University Hospital in Prague, Czech Republic and all participants provided written, informed consent.

2.2. Recording

The recording took place in a quiet room with low ambient noise using a head-mounted condenser microphone (Beyer-dynamic Opus 55; Heilbronn, Germany), positioned approximately 5 cm from the mouth of each subject. The utterances were sampled at 48 kHz with 16-bit quantization. The speech signals were obtained as a part of a larger speech examination conducted by a speech specialist in one session. During the examination, participants were twice asked to perform a sustained phonation of the vowel /i/ at comfortable loudness and pitch, as constant and as long as possible. No time limits were imposed during the recording.

2.3. Acoustic analysis

Acoustic analysis was performed on the sustained vowel /i/, which was shortened by 10% from both sides to avoid the effect of initial vocal fold adjustments. The shortened signal was then resampled to 10 kHz and divided into 60 ms windows with 55 ms overlap. Each window was weighted using a Hamming window function and analyzed using the 1/3-octave spectra method.

The 1/3-octave spectra analysis was based on the multirate filter bank presented by Couvreur [26], and is illustrated in Figure 1. In general, the three highest 1/3-octave frequency bands were obtained using the 3rd order IIR Butterworth passband filters. The highest 1/3-octave bands were centered at 2500 Hz (passband 2245–2828 Hz), 3150 Hz (passband 2828–3564 Hz) and 4000 Hz (passband 564–4490 Hz). After filtering the highest frequency components were removed and the signal was down-sampled by a factor of 2. The decrease of the sampling frequency by a factor of 2 yielded filter characteristics one octave lower; this approach was used iteratively to measure eighteen spectral bands (i.e., from 75 Hz to 4000 Hz).

The energy in each band was estimated using the RMS value and transformed to logarithmic scale according to Eq. 1.

\[
E(i) = 10 \log_{10} \left( \frac{1}{N} \sum_{n=0}^{N-1} E_{\text{filtered}}(i) \right),
\]

where \(E_{\text{filtered}}\) is the energy contained in the single frequency band, and \(E(i)\) is the decibel value of energy contained in the \(i\)-th band.

In accordance with previous research, the energy in the 1000 Hz frequency band was estimated as a marker of increased nasality in each window [19]. The level of nasality in the utterance was then estimated as the average energy (\(E_{\text{NIPPS}}\) mean) across all windows. Furthermore, fluctuation in nasality was estimated as the standard deviation (\(E_{\text{SD}}\) SD) of values measured across all windows.

![Figure 1: Principle of 1/3-octave spectra analysis presented in Couvreur (1998).](image)
2.4. Statistics

The average values of both $E_{Fn}$ mean and $E_{Fn}$ SD measurements were used for all statistical analyses.

Due to the relatively small sample size, the Kruskal-Wallis one-way analysis of variance with posthoc Bonferroni adjustment was employed to estimate group differences. Furthermore, the Spearman correlation was used to estimate possible relationships between acoustic parameters versus non-normally distributed SARA, over-all NNIPPS and EDSS scores, and NNIPPS and EDSS cerebellar subscores.

To estimate the number of participants with abnormal nasality we used the Wald task, which is a non-Bayesian statistical decision-making method based on three classes (i.e., positive, negative and indecisive) [27]. This method was chosen as it enables adjustment of classifier specificity and selectivity, and therefore sets the classifier as more-or-less conservative. This is useful considering that a certain amount of nasality disturbances may occur even in healthy speakers. Comprehensive details on the Wald task have been published previously [28].

3. Results

3.1. Acoustic analysis

As can be seen in the first column of Figure 2, the Wald task showed no clear cut-off threshold between healthy and pathological utterances for the $E_{Fn}$ mean parameter. However, considering nasality variance, the Wald task showed a considerable increase of fluctuation in pathological speech and was able to establish a cut-off threshold between healthy or indecisive versus pathological to values close to 2.5dB. When the 2.5dB threshold was employed, increased nasality fluctuation was present in 65.0% of MSA, 42.9% of CA, 30.0% of RS and 15.0% of HC speakers. The greatest occurrence of nasality fluctuation in MSA compared to CA and MS is further supported by Kruskal-Wallis analysis, which revealed statistically significant differences between MSA and HC groups with a very large Cohen’s effect size ($d = 1.39$).

The results of statistical analyses are shown in Figure 3 and detailed in Table 1.

3.2. Relationship between acoustic and clinical markers

Table 2 summarizes the results of correlation analyses between nasality measurement and over-all clinical scale scores and their cerebellar subscores. A statistically significant correlation was found in MSA (NNIIPPS cerebellar and $E_{Fn}$ SD, $r = 0.51$, $p < 0.05$). No other significant correlation between nasality measures and clinical scores were found.

Table 1: Results of nasality measures including median and median absolute deviation (MAD) values for $E_{Fn}$ mean and $E_{Fn}$ SD parameters across MSA CA, MS and HC groups, as well as results of Kruskal-Wallis including $\chi^2$, $p$, and Cohen’s Effect size.

<table>
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<tr>
<th></th>
<th>MSA</th>
<th>MS</th>
<th>HC</th>
<th>Kruskal-Wallis</th>
<th>Cohen’s Effect Size</th>
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</thead>
<tbody>
<tr>
<td>$E_{Fn}$ mean</td>
<td>$-41.80 \pm 3.84$</td>
<td>$-41.29 \pm 2.34$</td>
<td>$-41.81 \pm 2.79$</td>
<td>$\chi^2(3,70)$</td>
<td>$0.23$ $p = 0.97$ 0.05</td>
</tr>
<tr>
<td>$E_{Fn}$ SD</td>
<td>$2.81 \pm 0.53$</td>
<td>$2.31 \pm 0.50$</td>
<td>$2.15 \pm 0.49$</td>
<td>$18.85$</td>
<td>$p &lt; 0.001$ 1.39</td>
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dysfunction. of nasality may thus represent a novel marker of cerebellar also found. This is in agreement with inconsistent fluctuation and cerebellar subscore of EDS S (fluctuations and SARA for CA (the increased correlation coefficients between nasality Interestingly, even though not reaching level of significance, the increased presence of hypernasality in any of the measured groups. Conversely, significant differences between $E_{na}$ SD for MSA and HC groups and an increased occurrence of nasality fluctuation in certain CA and MS patients suggests abnormal fluctuations in velopharyngeal control connected to ataxic dysarthria. In addition, this is supported by significant correlation between nasality fluctuation and the cerebellar subscore of the NNIPPS in the MSA group ($r = 0.51$). Interestingly, even though not reaching level of significance, the increased correlation coefficients between nasality fluctuations and SARA for CA ($r = 0.40$) and between nasality fluctuation and cerebellar subscore of EDSS ($r = 0.38$) were also found. This is in agreement with inconsistent velopharyngeal closure reported by Duffy [3]. The fluctuation of nasality may thus represent a novel marker of cerebellar dysfunction.

There are some limitations to the present study. First, we did not perform aerodynamic measurements, which would provide ground truth for nasality assessment. However, we used a methodology that was previously validated and successfully used in dysarthria assessment [17–19]. Second, due to differing age of disease onset, groups with different diagnoses could not be age-matched. We therefore matched the HC group to the MSA group including oldest subjects as nasality is expected to remain stable or slightly deteriorate with age [31, 32]. Third, the MSA group included MSA-P patients. As previous work has shown that increased nasality fluctuations may to some extent represent due to hypokinetic-rigid syndrome, the effect of parkinsonism cannot be fully excluded [19]. However, nine of our MSA-P patients has anon-zero cerebellar score. In addition, a study by Rusz et al. [20] reported predominantly ataxic components of dysarthria even in MSA-P and suggested the sensitivity of speech for even minor cerebellar deficits. Furthermore, the moderate correlation with the cerebellar NNIPPS subscore ($r = 0.51$) versus weak correlation with the overall NNIPPS score ($r = 0.27$) also suggests a greater impact of cerebellar dysfunction over hypokinetic-rigid syndrome in our MSA group. Finally, the current approach for nasality assessment is particularly suitable for sustained vowels and cannot be easily generated to continuous speech.

4. Discussion

In the current study, we analyzed nasality in three groups of patients diagnosed with neurodegenerative or demyelinating diseases directly affecting the cerebellum and causing ataxic dysarthria. We employed a 1/3-octave spectra analysis presented by Kataoka et al. [14], which was adapted for the purposes of dysarthria assessment [19]. Using the $E_{na}$ SD parameter, we detected increased nasality fluctuation in 15% of HC, 30% of MS, 43% of CA and 65% of MSA speakers. A significant correlation ($r = 0.51$) was observed between $E_{na}$ SD representing nasality fluctuation and the NNIPPS cerebellar subscore in MSA patients.

Previous studies have reported different results across different etiologies ranging from hyponasality to hypernasality [4, 29]. However, to the best of our knowledge, studies evaluating nasality in ataxic dysarthria are very scarce. Indeed, there are studies that list distorted nasality among other manifestations; nevertheless, nasality assessment was not the primary focus of these studies [4, 5, 30]. This may be due to the fact that nasality assessment is challenging even in speech distortions connected solely to hypernasality such as cleft palate [7]. Considering that dysarthria is a complex combination of different speech distortions and that distorted nasality appears as one of the less-pronounced manifestations (e.g., Hartelius et al. reported 82% of detected nasality distortions as “just noticed” [5]), it is not surprising that nasality assessment provides one of the least reliable results [29].

We therefore employed a 1/3-octave spectra analysis as an objective measure, which has been successfully applied to the assessment of nasality in Parkinson and Huntington disease, and moreover to assess dysarthria in Friedreich Ataxia [18, 19]. When considering the level of nasality, our results did not confirm an increased presence of hypernasality in any of the measured groups. Conversely, significant differences between $E_{na}$ SD for MSA and HC groups and an increased occurrence of nasality fluctuation in certain CA and MS patients suggests abnormal fluctuations in velopharyngeal control connected to ataxic dysarthria. In addition, this is supported by significant correlation between nasality fluctuation and the cerebellar subscore of the NNIPPS in the MSA group ($r = 0.51$). Interestingly, even though not reaching level of significance, the increased correlation coefficients between nasality fluctuations and SARA for CA ($r = 0.40$) and between nasality fluctuation and cerebellar subscore of EDSS ($r = 0.38$) were also found. This is in agreement with inconsistent velopharyngeal closure reported by Duffy [3]. The fluctuation of nasality may thus represent a novel marker of cerebellar dysfunction.

5. Conclusion

Statistically significant differences and correlations were detected only in the MSA group, confirming nasality as a less obvious ataxic dysarthria manifestation. Nevertheless, acoustic analysis presented in the current study showed that the distortion of velopharyngeal control in ataxic dysarthria is not manifested as an increased level of nasality but rather as nasality fluctuation connected with inconsistent velopharyngeal control. This was observed across all examined ataxic groups. Furthermore, our results showed a higher correlation of nasality fluctuation and clinical scores directly connected with cerebellar function deficits suggesting a relation between cerebellar and velopharyngeal dysfunctions.

6. Acknowledgements

This study was supported by the Czech Science Foundation, grant no. 16-03322S, and by the Ministry of Health of the Czech Republic, grant no. 15-28038A.

7. References


