Vowel Articulation in Patients with Spinocerebellar Ataxia

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Abstract: *Objective*: Abnormalities in vowel articulation have been reported to be a common feature of dysarthria in spinocerebellar ataxia (SCA); however, findings about the degree and pattern of impaired vowel production are inconsistent. Therefore, the aim of the current study was to characterize the pattern of dysfunctional vowel production in patients with SCA by the means of acoustic analysis.

Methods: 31 patients SCA and 32 healthy subjects were tested. Description of vowel articulation was based upon the frequencies of the first and second formant (F1 and F2) of the German vowels /a/, /i/ and /u/ extracted severalfold from defined words within a given reading passage. The mean as well as the coefficient of variance of the respective F1 and F2 values of each single vowel were taken as measures of distinctiveness and steadiness of vowel articulation.

Results: In the SCA group, F1 and F2 values showed increased variability and a specifically restricted range which was particularly seen in the vowel /i/. Furthermore, the dysfunctional pattern differed between male and female patients with SCA.

Conclusions: Measurement of F1 and F2 revealed dysfunctional vowel articulation in SCA – however, with some genderrelated specifities – that can be explained by imprecision and reduced range of articulatory movements in ataxic speakers. Therefore, objective measurement of vowel formant frequencies provided additional information to the overall perceptual speech score. According to these preliminary findings, acoustic analysis of speech could be a promising tool for diagnosis, monitoring and detection of therapeutic effects in ataxic dysarthria.

Keywords: Spinocerebellar ataxia, ataxic dysarthria, vowel articulation, formant frequency, acoustic analysis of speech.

INTRODUCTION

Spinocerebellar ataxias (SCA) are a group of dominantly inherited progressive neurodegenerative diseases which now are classified according to the underlying mutation into more than 30 subtypes [1, 2]. Within this rare disease entity, the most frequent SCA diagnoses in Germany are SCA 1, 2, 3 and 6. In these genetic subtypes, the mutations consist of an excessive expansion of trinucleotide repeats in different genes. The repeat length acts as one factor responsible for phenotypical heterogeneity and variability in age of onset in patients with SCA1, 2 and 3 [3-5]. The clinical core feature of all SCAs is cerebellar ataxia, typically starting in adulthood with progressive gait unsteadiness and incoordination of the extremities. Although it has long been recognized that SCA patients often manifest with a variant pattern of additional symptoms caused by concomitant affection of non-cerebellar structures [6], e.g. spasticity in SCA1, peripheral neuropathy in SCA2 or dystonia in SCA3 [7-10], the development of speech impairment in the course of the disease has mainly been attributed to dysfunction of cerebellar motor control circuits [11]. In other SCA subtypes, however, the disease process is confined to the cerebellum resulting in a "pure" ataxia phenotype, that in case of SCA6 has a particularly late onset [2, 3].

Since the first systematic description of ataxic speech performed by Darley and colleagues on a sample of 30 patients with etiologically different cerebellar disorders, a wealth of subsequent investigations further refined the features of deviant speech dimensions in ataxic dysarthria which comprise articulatory inaccuracy (represented by imprecise consonants, irregular articulatory breakdowns and vowel distortions), prosodic excess (mainly composed of excess and equal stress, prolonged phonemes, prolonged intervals and slowed speech rate) and phonatory-prosodic insufficiency (mirrored by harshness, monopitch and monoloudness) [12, 13]. Subsequent investigations mainly focussed on the aspect of disturbed temporal motor speech performance and suggest speech slowing, especially in syllable repetition paradigms, and increased variability

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in speech timing as characteristics of ataxic dysarthria [14-17], that contribute to overall speech intelligibility [16]. Descriptions of the patterns of phonatory dysfunction in ataxic dysarthria vary somewhat across studies and include monopitch and monoloudness as well as higher variability in pitch and loudness levels, harshness, breathiness and strain of voice, voice tremor and audible inspiration [12, 13, 18-21]. To date, investigations on the aspect of vowel articulation in ataxic dysarthria are relatively sparse. Though, the imprecision of range and force of articulatory muscles in ataxia might be expected to affect vowel production and thus may compromise speech intelligibility. In fact, perceptual analyses of speakers with mild to moderate ataxic dysarthria revealed higher vocal instability and vowel distortion [22-24]. Several studies dealt with the aspect of coarticulation which reflects temporally staggered activation of coordinative constraints for different phonetic gestures in the interplay of consonant-vowel articulation [25]. However, in ataxic speakers, coarticulation was reported to be widely unimpaired [26] or there were only trends towards increased perseverative coarticulation while anticipatory coarticulation did not differ significantly from healthy speakers [27]. Previous studies on formant frequencies of isolated vowels were inconclusive. While Kent and colleagues found normal formant frequency ranges in samples of conversational speech in ataxic speakers [28, 29], others reported a levelling of formant frequencies, i.e. decrease of normally high formants and increase of normally low formants, which resulted in reduced distinctness of the vowels $/\alpha$, $/\nu$ and $/\nu$ [22]. Summarized, previous results of acoustic analyses do not persuasively confirm the expected inaccuracy and target undershooting suggested upon perceptual impression [30, 31].

The aim of the present study was to further characterize the pattern of vowel articulation in patients with ataxic dysarthria caused by spinocerebellar ataxia. The investigation was based upon an acoustic analysis of formant frequencies of the three German corner vowels $/\alpha$, $/\alpha$ and $/\alpha$ extracted from different words of a standardized reading task. In this way, the results were assumed to mirror a more "natural" speech pattern than non-sense syllable sequences, but at the same time to make group comparisons more valid than extractions from conversational speech. We expected to find a restricted average working space for vowels with a decrease of normally high formant frequencies and an increase of normally low formant frequencies as an indicator of reduced range of motion of the

articulatory muscles. The vowel articulation index (VAI) used here, has been suggested as a useful comprehensive measure of formant frequency centralization in speakers with muscle tension dysarthria [32] and Parkinsonian hypokinetic dysarthria [33]. Therefore, we tested its applicability and usefulness as a surrogate parameter for impaired vowel articulation in ataxic dysarthria as well.

Furthermore, one could assume an increased variability of formant frequencies in repetitive production of the same vowel evoked by the inaccuracy of articulatory movements. Since the position of the tongue mainly accounts for the frequency of the first and second formants of a vowel (F1 and F2), inaccurate articulation should be present particularly in those vowels which require the most extreme position of the tongue related to the degree of hight and frontness. According to this hypothesis, distinctness of articulation should decline from the vowel /i/ over /u/ to the German $/\alpha$ where the tongue position is almost neutral.

We also related the results of acoustic analysis to a perceptual classification of speech intelligibility as well as to global disease severity in SCA patients. Since the sample of speakers comprised several genetic SCA subtypes, a subanalysis of speakers with SCA 6 versus SCA 1, 2 and 3 was performed to compare vowel articulation in "pure cerebellar" dysfunction (SCA 6) with the subgroups of SCA known to feature additional non-cerebellar symptoms.

PATIENTS AND METHODS

Patients

From 2007 to 2008, 31 patients (19 male, 12 female) with genetically confirmed autosomal-dominant spinocerebellar ataxia (SCA 1, 2, 3 and 6) and 32 healthy control persons (17 male, 15 female) were recruited for this study. All participants were native German speakers.

Participants´ age was similar between groups (SCA: mean 54.48 years, SD 14.16, range 32 to 83; healthy controls: mean 57.42 years, SD 11.81, range 40 to 78). In SCA patients, ataxia severity was assessed with the Scale for the Assessment and Rating of Ataxia (SARA) [34] with an average score of 14.55 (SD 8.01, range 2 to 31.5) and the extent of additional non-cerebellar affection was described with the Inventory of Non-Ataxia Symptoms (INAS) with an average score of 5.07 (SD 2.81, range 0 to 12). The SARA score for the

quantitative assessment of ataxic symptoms has been shown to be valid and reliable [34, 35] and consists of 8 subitems (gait, stance, sitting, speech disturbance, finger chase, nose-finger test, fast alternating hand movements, heel-shin slide) with a maximum score of 40 points. The INAS score consists of 30 items which are related to one of the following 16 symptoms: areflexia, hyperreflexia, extensor plantar response, spasticity, paresis, amyotrophy, fasciculations, myoclonus, rigidity, chorea, dystonia, resting tremor, sensory symptoms, brainstem oculomotor signs (horizontal and vertical ophthalmoparesis, slowing of saccades), urinary dysfunction, and cognitive impairment. Only the presence or absence of one of these symptoms was considered [36].

Speech intelligibility was perceptually scored according to the SARA speech item (score 0 to 6 with "6" indicating most severe impairment) and ranged from 0 to 4 points in the SCA group (mean 1.94, SD 1.36). Disease duration was given from reported onset of gait ataxia to the time of examination and ranged from 3 to 22 years (mean 11.87 years, SD 5.44).

Our study was in compliance with the Helsinki Declaration and had been approved by the local Ethics Committees. Written informed consent was obtained from each participant.

Patients´ characteristics are summarized in Table **1**.

Data Acquisition

Each participant had to perform a standardized reading task composed of four complex sentences (see appendix) which has been used by our group in previous research on Parkinsonian dysarthria [33]. In order to exclude difficulties in reading, the participants had to read the text twice; the second sequence was taken for the definite analysis. Speech samples were digitally recorded in a quiet room using a commercial audio software (Steinberg WaveLab[®]; Steinberg, Hamburg, Germany) and a head-set microphone (Plantronics Audio 550 DSP[®]; Plantronics Inc., Santa Cruz, CA, USA) positioned 5 cm from the lips. The data were digitized at a sampling rate of 44.1 kHz. Each of the vowels $/\alpha$, /i/ and /u/ were extracted 10 times from different words within the text (see appendix). The formant frequency values F1 and F2 were measured separately for each vowel for a 30 ms segment at the temporal midpoint, as determined independently by two blinded examiners, using a special speech software (Praat[®]; www.praat.org, Phonetic Sciences, University of Amsterdam, The Netherlands) [37].

Then, the average formant frequency values of F1 and F2 of each vowel were calculated for each speaker based upon the ten separate measurements (mean F1 / α , mean F2 / α , mean F1 /i/, etc.).

The intraindividual coefficient of variance for F1 and F2 values were calculated for each of the three vowels $/\alpha$, /i/ and /u/ as: cov = SD / mean of all formant values x 100 (cov_F1_/ α /, cov_F2_/ α /, cov_F1_/i/, etc.).

As a comprehensive measure of the precision of vowel articulation, the "vowel articulation index / VAI" was calculated based on the formula VAI = $(F2/i / +$ F1/ α) / (F1/i/ + F1/u/ + F2/u/ + F2/ α). VAI was expected to be reduced, if normally high formants (F2/i/ and $F1/\alpha$) decrease or normally low formants ($F1/i$, F1/u/, F2/u/ and F2/ α /) increase as a result of reduced articulatory capacity and has previously been shown to be superior to the traditional measurement of triangular vowel space area at least in speakers with hypokinetic dysarthria [33].

Statistics

All variables were normally distributed (Kolmogorov-Smirnov test). Univariate analysis of variance (ANOVA) with condition (SCA vs. control) and gender (male vs. female) as between-subject factor and post hoc t-test were performed first for all variables with an adjusted significance level according to Bonferroni adjustment set at p<0.004. Since the groups were age-matched, a possible effect of age was not tested. Pearson correlation was used to test for within-group correlations between variables.

According to ANOVA, gender was found to be an independent factor for all of the acoustic measures; therefore, subsequent analyses were performed separately for males and females. No significant differences were seen concerning age, disease duration, SARA sum score, SARA speech score and INAS score between male and female SCA patients.

RESULTS

Numerical data are listed in Table **1**.

Male Group: comparison between SCA Patients and Control Group

In the male SCA subgroup, mean_F1_/i/ values were increased (359.87Hz ± 31.03 vs. 301.96Hz ± 27.10, $p \le 0.001$, whereas mean_F2_/i/ values showed a tendency to be lower than in male controls

gender	male $n = 36$			female $n = 27$		
	SCA (n=19) SCA1: n=4; $SCA2: n=1$; SCA3: n=6; SCA6: n=8	control $(n=17)$	comparison SCA vs. control	$SCA (n=12)$ $SCA1: n=2;$ SCA2: n=3; SCA3: n=5; $SCA6: n=2$	control ($n=15$)	comparison SCA vs. control
	mean /SD	mean /SD		mean /SD	mean /SD	
age (years)	53.68 / 14.45 range 32 - 83	56.18 / 12.13 range 40 - 78	n.s.	55.75 / 14.22 range 32 - 79	58.75 / 11.64 range 43 - 75	n.s.
duration (years)	11.74 / 5.24 range 6 - 22			12.09 / 6.04 range 3 - 22		
SARAsum score	14.13 / 7.64 range $2 - 31.5$			15.27 / 8.95 range $4 - 29.5$		
SARAspeech score	2.00 / 1.50 range $0 - 4$			1.83 / 1.19 range $0 - 4$		
INAS score	4.74/2.94 range $0 - 12$			5.70 / 2.58 range 3 - 11		
	mean /SD	mean /SD		mean /SD	mean /SD	
mean_F1_a	586.17 / 48.85	582.74 / 93.43	n.s.	704.04 / 58.18	697.31 / 86.12	n.s.
mean_F2_a	1248.70 / 147.74	1307.05 / 106.42	n.s.	1433.16 / 145.53	1544.51 / 92.71	$n.s.(p=0.023)$
mean F1 i	359.87 / 31.03	301.96 / 27.10	p<0.0001	416.33 / 71.46	324.50 / 44.73	p=0.0004
mean_F2_i	1889.16 / 174.13	2005.29 / 103.95	$n.s.(p=0.020)$	1980.85 / 250.32	2333.86 / 113.88	p<0.0001
mean_F1_u	400.67 / 43.40	376.67 / 63.99	n.s.	430.94 / 68.75	363.68 / 64.07	$n.s.(p=0.015)$
mean_F2_u	1219.86 / 244.86	1303.15 / 222.20	n.s.	1116.37 / 106.32	1141.30 / 79.30	n.s.
VAI	0.771/0.071	0.791 / 0.065	n.s.	0.792 / 0.074	0.900 / 0.054	p=0.0002
cov_F1_a	12.06 / 5.11	13.66 / 3.57	n.s.	13.35 / 4.57	16.10 / 6.22	n.S.
cov_F2_a	17.46 / 8.56	12.08 / 2.99	$n.s.(p=0.017)$	13.09 / 6.79	11.34 / 2.78	n.s.
cov_F1_i	16.55 / 5.67	12.61 / 3.92	$n.s.(p=0.022)$	21.65 / 6.94	13.09 / 3.97	$p=0.0015$
cov_F2_i	15.84 / 6.63	9.70 / 3.65	p=0.0016	18.21 / 8.71	11.69 / 2.99	$n.s.(p=0.028)$
cov_F1_u	13.11 / 5.42	18.23 / 5.21	$n.s.(p=0.007)$	15.85 / 5.39	16.68 / 5.74	n.s.
cov_F2_u	31.30 / 10.33	24.70 / 6.24	$n.s.(p=0.026)$	30.55 / 11.13	20.70 / 5.07	$n.s.(p=0.013)$

Table 1: Participants´ Characteristics and Results

(1889.16Hz ± 174.13 vs. 2005.29Hz ± 103.95, p < 0.020), but without statistical significance. No betweengroup differences were seen for F1 and F2 values for the vowels $/\alpha$ and $/\alpha$.

VAI was similar in the SCA and the control group.

The coefficient of variance was higher for F2_/i/ $(15.84 \pm 6.63 \text{ vs. } 9.70 \pm 3.65, \text{ p} = 0.0016) \text{ while}$ differences in the same direction for all other F1 and F2 values (except for cov_F1_/ α / and cov_F1_/u/) did not reach the adjusted level of significance.

SARA sum score was positively correlated to mean_F1_/i/ and cov_F1_/i/ (R = 0.530, p = 0.01 and R $= 0.651$, $p = 0.01$), and a similar positive correlation was seen with SARA speech score (R = 0.651 , p = 0.001 and R = 0.530, $p = 0.01$), but not with INAS or disease duration. No correlation of speech parameters and age was seen within the groups (Figures **1-3**).

Female Group: Comparison between SCA Patients the and Control Group

In the female SCA subgroup, the average F1 and F2 values of the vowel /i/ were shifted similarly as in the male SCA group with an elevation of mean_F1_/i/ $(416.33Hz \pm 71.46 \text{ vs. } 324.50Hz \pm 44.73, p = 0.0004)$ and a decrease of mean_F2_/i/ (1980.85Hz \pm 250.32 vs. 2333.86Hz ± 113.88, p < 0.0001).

Figure 1: Mean formant frequency values in male patients with SCA as compared to male controls.

VAI was significantly reduced in the female SCA subgroup compared to female controls (0.792 ± 0.074) vs. 0.900 ± 0.054 , $p = 0.0002$).

Concerning the variability of the formant values, only cov_F1_/i/ was found to be significantly increased $(21.65 \pm 6.95 \text{ vs. } 13.09 \pm 3.97, \text{ p} = 0.0015)$

In contrast to the male SCA group, SARA speech score were not related to the formant measures in female SCA speakers, while F2_/i/ was further reduced $(R = -0.522, p = 0.050)$ and the variance cov $F2_i/i$ increased with higher SARA sum scores ($R = 0.577$, p = 0.032). No correlations were seen between formant values and INAS scores. No correlation of speech parameters and age was seen within the groups.

Male Group: Comparison between Patients with SCA 1, 2 and 3 (ADCA I) and Patients with SCA 6 (ADCA III) [6]

Concerning patients characteristics, age was significantly higher in the 8 male speakers with SCA 6 than in the composed male subgroup with SCA 1 ($n =$ 4), SCA 2 (n = 1) and SCA 3 (n = 6) (64.88 \pm 12.22 years vs. 45.55 ± 9.92 years, $p = 0.0014$). INAS scores were higher in ADCA I than ADCA III (6.18 \pm 2.89 vs. 2.75 ± 1.58 , $p = 0.004$). Disease duration, SARA sum score and SARA speech scores were similar in SCA 6 and SCA 1 and 3.

No significant differences were seen concerning the VAI and the average values and variability of F1 and F2 for the three vowels.

Figure 2: Mean formant frequency values in female patients with SCA as compared to female controls.

Figure 3: comparison of COV of formant frequency values between male and female patients with SCA and controls. *: $p < 0.05$, **: $p < 0.01$.

Female Group: Comparison between Patients with SCA 1, 2 and 3 (ADCA I) and Patients with SCA 6 (ADCA III)

Although the numbers of female patients with ADCA III (SCA 6: $n = 2$) and ADCA I (SCA 1: $n = 2$; SCA 2: n $= 3$; SCA 3: $n = 5$) were too small to formally perform a valuable statistic analysis, age, disease duration, INAS score, SARA sum score and SARA speech scores were similar in ADCA I and ADCA III.

Similarly, no obvious differences were seen concerning the VAI and the average values and variability of F1 and F2 for the three vowels.

DISCUSSION

The present investigation was conducted to characterize and objectively determine the capacity of accurate vowel articulation as one parameter of ataxic dysarthria by the means acoustic analysis. According to our hypothesis, abnormalities of formant frequency precision and steadiness were exspected particularly in the articulation of the German vowel /i/ which requires the maximum range of movement of the articulatory muscles, especially the tongue. Actually, the main finding was an increased variability of formant frequencies of the same vowel throughout the reading task which was preferentially seen in the production of the vowel /i/, but also concerning F2 values of the vowel /u/ and – in male speakers – the vowel / α / which refers to an impresision of frontward/backward tongue movements. The higher variability of the formant frequencies in the ataxic speakers may be intensified by the fact that the same vowels were embedded in different phonemes within the reading task. The

unconstant phonetical neigbourhoud of the single vowels coming along with variable articulatory demands and changing influence of coarticulation might be particularly suitable to unmask deficits in reproducible vowel articulation in ataxic dysarthria [38, 39]. Additionally to the finding of variable formant frequency production, the average F1 and F2 frequencies of the single vowles were found to be less distinctive which again was preferentially relevant for the vowel /i/ which in healthy subjects is characterized by the lowest F1 and the highest F2 values of all vowels. However, patterns of restricted vowel articulation were different in male and femal speakers with SCA: In male patients, only F1 and F2 values of the vowel /i/ were found to be nivellated, but without significant impact on VAI which was only slighly reduced in male SCA patients as compared to male control speakers. In contrast, female SCA patients at least showed an additional tendency to reduced F2 values for $/\alpha$ and elevated F1 values for $/\alpha$ leading to an overall significant restriction of the working space for vowels as mirrored by significantly reduced VAI.

These gender-related differences of the behaviour of formant frequencies in ataxic dysarthria might be due to the sexual dimorphism of the laryngopharyngeal tractus with different size and configuration of the tongue, the three dimensional shape and acoustical properties of vocal cord and the resonatory cavities in male and female. According to previous studies in healthy speakers, gender-related differences of overall speech intelligibility had been attributed to these anatomical factors since fundamental and formant frequncies as well as the resulting working space for vowels have been found to vary significantly between healthy man and women [40-43]. In the present study, correlations between parameters of vowel articulation and global motor performance and speech intelligibility showed differences between the genders as well: In male patients with SCA, higher variability and unphysiological elevation of F1 /i/ was found in speakers with worse overall speech performance, whereas in female ataxic patients, dysfuntional vowel production was related to SARA sum scores instead of the SARA speech item which might be a hint for a differential contribution of vowel articulation to overall intelligibility in male and female speakers with SCA.

Although the findings of the present investigation suggest a specific pattern of dysfunctional vowel articulation in ataxic dysarthria with some particular gender-dependent characteristics, there are some methodical issues concerning the procedure of vowel analysis and the composition of the sample of ataxic speakers: As a typical observation at least in the most severe affected ataxic speakers, there was a reduction of articulatory velocity accompanied by a prologation even of normally unstressed vowels. Therefore, the acquisition of formant frequencies for a 30 ms segment at the temporal midpoint of the vowel, as it has been suggested to be adequate at least in patients with different kinds of dysarthria [32, 33] might be inappropriate in abnormally prolonged vowels. Unsteadiness of formant frequencies within the same vowel might be missed and the measured F1 and F2 variability might actually been underestimated. Furthermore, the SCA group was heterogeneous concerning the perceptually rated degree of global speech impairment. Since 11 out of 31 speakers with SCA featured only a very mild dysarthria (SARA speech score 1) or were completely unimpaired (score 0), the effective pattern of abnormal vowel articulation in ataxic speakers might be diluted. Though, a definite omission of the data of these quasi non-dysarthric speakers would have lessened the statistical power of the entire analysis. However, a pro forma recalculation of the results with focus on the more severe dysarthric speakers (12 male, 8 female) lead to exactly the same pattern of formant frequency abnormalities with the only exception, that the counterintuitive reduction of cov_F1_/u/ in male SCA speakers was not verifiable any more (data not shown). Considering the aspect that the sample of ataxic speakers was also heterogeneous concerning the genetically confirmed subgroups of SCA, an additional comparison between the "pure" cerebellar phenotype (ADCA III / SCA 6) and the clinically compound SCA subgroups (ADCA I / SCA 1, 2 and 3) was conducted without an evidence of different patterns of formant frequency abnormalities, however, this statistical subanalysis was performed only in the male SCA patients since the number of females was too small. In a large study on more than 500 patients with SCA 1, 2, 3, and 6, no differences concerning perceptually rated overall speech performance were found, and it has been suggested that vestibulocerebellar, spinocerebellar and pontocerebellar circuits are functionally impaired to almost the same degree, but at different anatomical levels [11] which seems to somewhat justify the conjoint vowel analysis in the present investigation.

In summary, the present study was able to preliminary define a distinctive pattern of impaired vowel articulation in speakers with SCA which is characterized by an increased variability and reduced range of formant frequency values. These findings seem to mirror the manifestation of ataxia on the articulatory motor system leading to imprecision and restricted range of movements. Comparisons of these results with patients suffering from other kinds of speech impairments as e.g. hypokinetic or spastic dysarthria, should be performed to survey, if this pattern of dysfunctional vowel articulation is specific for SCA or ataxic dysarthria in general and therefore could be further developed as a useful tool in the differential diagnosis of motor speech disorders. Furthermore, subsequent studies with larger samples of genetically homogeneous cerebellar diseases are warranted to disclose possible subtle differences of the type of dysarthria and its development in the course of the disease in the different subgroups of SCA. Since acoustic analsis of speech is an easy applicable, reproducible and non-intrusive research method, the establishment of surrogate parameters and prototypical patterns of deviant speech parameters could be of use for the diagnosis, monitoring and objectification of therapeutic effects in cerebellar diseases and other kinds of movement disorders.

AUTHORS CONTRIBUTION SECTION

TSH, OE, TK, SS and US participated in the conception, design and organization of the research project. SS, TSH and OE were involved in the execution of the research project. SS and TSH contributed to the design and execution of the statistical analysis. TSH and SS contributed to the writing of the manuscript.

All authors have read the manuscript, and the paper has not previously been published and is not under simultaneous consideration by another journal. There has been no ghost writing by anyone not named on the authors list.

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THERE ARE NO CONFLICTS OF INTEREST

There are no special concerns about copyright or federal employment.

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REFERENCES

- [1] Soong BW, Paulson HL. Spinocerebellar ataxias: An update. Curr Opin Neurol 2007; 20: 438-46. http://dx.doi.org/10.1097/WCO.0b013e3281fbd3dd
- [2] Klockgether T. Update on degenerative ataxias. Curr Opin Neurol 2011; 24: 339-45. http://dx.doi.org/10.1097/WCO.0b013e32834875ba
- [3] Paulson HL. The spinocerebellar ataxias. J Neuroophthalmol 2009; 29: 227-37. http://dx.doi.org/10.1097/WNO0b013e3181b416de
- [4] Stevanin G, Dürr A, Brice A. Clinical and molecular advances in autosomal dominant cerebellar ataxias: from genotype to phenotype and pathophysiology. Eur J Hum Gen 2000; 8: 4- 18. http://dx.doi.org/10.1038/sj.ejhg.5200403
- [5] Durr A. Autosomal dominant cerebellar ataxias: polyglutamine expansions and beyond. Lancet Neurol 2010; 9: 885-94. http://dx.doi.org/10.1016/S1474-4422(10)70183-6
- [6] Harding AE. Classification of the hereditary ataxias and
- paraplegias. Lancet 1983; 1: 1151-5. http://dx.doi.org/10.1016/S0140-6736(83)92879-9
- [7] Gomez CM, Subramony SH. Dominantly inherited ataxias. Sem Pediatr Neurol 2003; 10: 210-22. http://dx.doi.org/10.1016/S1071-9091(03)00030-5
- [8] Schöls L, Bauer P, Schmidt T, Schulte T, Riess O. Autosomal dominant cerebellar ataxias: clinical features, genetics, and pathogenesis. Lancet Neurol 2004; 3: 291-304. http://dx.doi.org/10.1016/S1474-4422(04)00737-9
- [9] Whaley NR, Fujioka S, Wszolek ZK. Autosomal dominant cerebellar ataxia type I: a review of the phenotypic and genotypic characteristics. Orphanet J Rare Dis 2011; 6: 33. http://dx.doi.org/10.1186/1750-1172-6-33
- [10] Bettencourt C, Lima M. Machado-Joseph disease: from first descriptions to new perspectives. Orphanet J Rare Dis 2001; 6: 35. http://dx.doi.org/10.1186/1750-1172-6-35

[11] Jacobi H, Hauser TK, Giunti P, *et al*. Spinocerebellar ataxia types 1, 2, 3 and 6: the clinical spectrum of ataxia and morphometric brainstem and cerebellar findings. Cerebellum 2012; 11: 155-66. http://dx.doi.org/10.1007/s12311-011-0292-z

- [12] Duffy JR. Motor Speech Disorders: substrates, differential
- diagnosis and management, ed 2. St. Louis, Missouri, Elsevier Mosby, 2005 [13] Darley FL, Aronson AE, Brown JR. Clusters of deviant
- speech dimensions in the dysarthrias. J Speech Hear Res 1969; 12: 462-96.
- [14] Darley FL, Aronson AE, Brown JR. Differential diagnostic patterns of dysarthria. J Speech Hear Res 1969; 12: 246-69.
- [15] Kent RD, Netsell R, Abbs J. Acoustic characteristics of dysarthria associated with cerebellar disease. J Speech Hear Res 1979; 22: 627-48.
- [16] Ackermann H, Hertrich I. Speech rate and rhythm in cerebellar dysarthria: an acoustic analysis of syllable timing. Folia Phoniatr 1994; 46: 70-8. http://dx.doi.org/10.1159/000266295
- [17] Ziegler W, Wessel K. Speech timing in ataxic disorders. Neurology 1996; 47: 208-14. http://dx.doi.org/10.1212/WNL.47.1.208
- [18] Boutsen FR, Bakker K, Duffy JR. Subgroups in ataxic dysarthria. J Med Speech Lang Pathol 1997; 5: 27-36.
- [19] Joanette Y, Dudley JG. Dysarthric symptomatology of Friedreich´s ataxia. Brain Lang 1980; 10: 39-50. http://dx.doi.org/10.1016/0093-934X(80)90036-X
- [20] Gilman S, Kluin KJ. Speech disorders in cerebellar degeneration studied with positron emission tomography; in Blitzer A, Brin MF, Saaki CT, Fahn S, Harris KS (eds): Neurologic disorders of the larynx. New York, Thieme Medical Publishers, Inc, 1992: 279-85.
- [21] Hertrich I, Spieker S, Ackermann H. Gender-specific phonatory dysfunctions in disorders of the basal ganglia and the cerebellum: acoustic and perceptual characteristics; in: Ziegler W, Deger K (eds): Clinical phonetics and linguistics, London, Whun, 1998: 448-57.
- [22] Hertrich I, Ackermann H. Temporal and spectral aspects of coarticulation in ataxic dysarthria: an acoustic analysis. J Speech Lang Hear Res 1999; 42: 367-81.
- [23] Schalling E, Hartelius L. Acoustic analysis of speech tasks performed by three individuals with spinocerebellar ataxia. Folia Phoniatr Logop 2004; 56: 367-80. http://dx.doi.org/10.1159/000081084
- [24] Schalling E, Hammarberg B, Hartelius L. Perceptual and acoustic analysis of speech in individuals with spinocerebellar ataxia (SCA). Logoped Phoniatr Vocol 2007; 32: 31-46. http://dx.doi.org/10.1080/14015430600789203

[25] Fowler CA, Saltzman E. Coordination and coarticulation in

- speech production. Lang Speech 1993; 36: 171-95. [26] Netsell R, Kent RD. Paroxysmal ataxic dysarthria. J Speech Hear Disord 1976; 41: 93-109.
- [27] Sidtis JJ, Ahn JS, Gomez C, Sidtis D. Speech characteristics associated with three genotypes of ataxia. J Commun Disord 2011; 44: 478-92. http://dx.doi.org/10.1016/j.jcomdis.2011.03.002
- [28] Kent RD, Kent JF, Rosenbek JC, Vorperian HK, Weismer G. A speaking task analysis of the dysarthria in cerebellar diseases. Folia Phoniatr Logop 1997; 49: 63-82. http://dx.doi.org/10.1159/000266440
- [29] Kent RD, Kent JF, Duffy JR, Thomas JE, Weismer G, Stuntebeck S. Ataxic dysarthria. J Speech Lang Hear Res 2000; 43: 1275-89.
- [30] Darley FL, Aronson AE, Brown JR. Motor speech disorders. Philadelphia, Saunders, 1975
- [31] Cannito MP, Marquardt TP. Ataxic dysarthria; in Mc Neik MR (ed): Clinical management of sensorimotor speech disorders. New York, Thieme, 1997: 217-47.
- [32] Roy N, Nissen SL, Dromey C, Sapir S. Articulatory changes in muscle tension dysphonia: evidence for vowel space expansion following manual circumlaryngeal therapy. J Commun Disord 2009; 42: 124-35. http://dx.doi.org/10.1016/j.jcomdis.2008.10.001
- [33] Skodda S, Visser W, Schlegel U. Vowel articulation in Parkinson´s disease. J Voice 2011; 25: 467-72. http://dx.doi.org/10.1016/j.jvoice.2010.01.009
- [34] Schmitz-Hübsch T, du Montcel ST, Baliko L, *et al*. Scale for the assessment and rating of ataxia: development of a new clinical scale. Neurology 2006; 66(11): 1717-20. http://dx.doi.org/10.1212/01.wnl.0000219042.60538.92
- [35] Saute JA, Donis KC, Serrano-Munuera C, *et al.*; Iberoamerican Multidisciplinary Network for the Study of Movement Disorders (RIBERMOV) Study Group. Ataxia rating scales--psychometric profiles, natural history and their application in clinical trials. Cerebellum 2012; 11: 488-504. http://dx.doi.org/10.1007/s12311-011-0316-8
- [36] Schmitz-Hübsch T, Coudert M, Bauer P, *et al*. Spinocerebellar ataxia types 1, 2, 3, and 6: disease severity and nonataxia symptoms. Neurology 2008; 71: 982-9. http://dx.doi.org/10.1212/01.wnl.0000325057.33666.72
- [37] Boersma P, Weenik D. PRAAT: a system for doing phonetics by computer. Report of the Institute of Phonetic Sciences of the University of Amsterdam, 1996, available at: http:// www.fon.humuva.nl/praat

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- [38] Brown A, Docherty GJ. Phonetic variation in dysarthric speech as a function of sampling task. Eur J Disord Commun 1995; 30: 17-35. http://dx.doi.org/10.3109/13682829509031320
- [39] Tjaden K, Wilding G. Effects of speaking task on intelligibility in Parkinson´s disease. Clin Linguist Phon 2011; 25: 155-68. http://dx.doi.org/10.3109/02699206.2010.520185
- [40] Klatt D, Klatt L. Analysis, synthesis, and perception of voice quality variations among female and male talkers. J Acoust Soc Am 1990; 87: 820-57. http://dx.doi.org/10.1121/1.398894
- [41] Byrd D. Relations of sex and dialect to reduction. Speech Commun 1994; 15: 39-54. http://dx.doi.org/10.1016/0167-6393(94)90039-6
- [42] Bunton K, Weismer G. The relationship between perception and acoustics for a high-low vowel contrast produced by speakers with dysarthria. J Speech Lang Hear Res 2001; 44: 1215-28. http://dx.doi.org/10.1044/1092-4388(2001/095)
- [43] Kwon HB. Gender difference in speech intelligibility using speech intelligibility tests and acoustic analyses. J Adv Prosthodont 2010; 2: 71-6. http://dx.doi.org/10.4047/jap.2010.2.3.71