Original Article

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The evaluation of swallowing in patients with spinocerebellar ataxia and oropharyngeal dysphagia: A comparison study of videofluoroscopic and sonar doppler

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SUMARRY

Introduction: Spinocerebellar ataxia (SCA) is a degenerative disease that can cause loss of coordination of voluntary muscle movement such as that required for swallowing.
Aims: The purposes of this cross-sectional and comparative case study were: (1) to assess the severity of dysphagia through a videofluoroscopic swallow study, and (2) to compare differences in frequency, intensity, and duration of sound waves produced during swallowing in normal and SCA patients by using sonar Doppler.
Method: During swallow evaluation using videofluoroscopy, a sonar Doppler transducer was placed on the right side of the neck, at the lateral edge of the trachea, just below the cricoid cartilage to capture the sounds of swallowing in 30 SCA patients and 30 controls.
Result: The prevalence in the dynamic evaluation of swallowing videofluoroscopy was by changes in the oral phase of swallowing. The analysis of variance of the averages found in each variable - frequency, intensity and duration of swallowing - shows there was a significant correlation when compared to the healthy individual curve.
Conclusion: The study demonstrates the prevalence of oral dysphagia observed in dynamic videofluoroscopic swallow evaluation. In patients with SCA, the mean initial frequency (IF), initial intensity (II), and final intensity (FI) were higher and the time (T) and peak frequency (PF) were lower, demonstrating a pattern of cricopharyngeal opening very close to that found in normal populations.
Keywords: Deglutition Disorders; Spinocerebellar Ataxias; Doppler Effect.

INTRODUCTION

The stomatognathic function of swallowing is a natural process whose function is to transport the food bolus from the mouth to the stomach while preventing the entry of any substance into the airway.

Food intake, besides being necessary for sustaining life and ensuring adequate nutrition, is one of the greatest human pleasures. However, this pleasure may be interrupted as a result of neurological changes that can alter the normal progression of food through the digestive tract. This symptom, called dysphagia, can occur in children, adults, or the elderly. Heredodegenerative diseases of the central nervous system, including spinocerebellar ataxias (SCAs), are part of the group causing dysphagia and, according to CAMBIER, MASSON, and DEHEN (1999), meet 3 criteria: their genetic determinism, the systematization of the lesions, and the quality of the pathological process that occurs through slow degeneration without necrosis or inflammation.

STEVANIN, DÜRR, and BRICE (2000) report that hereditary cerebellar ataxias are progressive disorders that may begin in childhood or adulthood and are characterized by degeneration of the cerebellum and its afferent and efferent connections.

A study carried out by MATILLA-DUENAS (2012) reports the current identification of at least 43 SCA subtypes whose mutations cause different forms of spinocerebellar ataxia with specific and diverse signs and symptoms. The present study aimed to study the subtypes 2, 3, 6, 7 and 10 of SCAs. These patients present in the common complaints of swallowing disorders, which can cause serious clinical complications, such as dehydration, malnutrition, penetration and tracheal aspiration.

According to Harding (1984) and Teive (2004), ataxia may cause loss of voluntary muscle movement coordination and disturbances in body posture control, eye movements, speech control, word articulation, and swallowing.

However, Logemann (1983b), Groher (1992), Huckabee (1992), and Buchholz (1994) report that the dysphagia may result from weakness in the muscles of the lips, tongue, velum, pharynx, and esophagus owing to cortical/subcortical brain injury and/or brainstem injury. These changes may also cause a lack of movement coordination or lack of sensitivity of the oral and pharyngeal regions that interferes with the voluntary and reflex phases of swallowing and may cause a change of direction of the bolus, leading to penetration and/ or aspiration in the airway.

Investigation of dysphagia is carried out through clinical evaluation and, if necessary, complemented by instrumental examination. Furkim (2001) stresses on the importance of confirming exactly how the dysphagia occurs, so that the therapeutic plan can be formulated. It should be highlighted that technological innovations allow greater objectivity and variability in research, assessment, diagnosis, and monitoring of disorders of the swallowing process that contribute decisively to the definition of sequential conduct.

Among the instrumental methods for evaluation, monitoring, and biofeedback of swallowing are videofluoroscopy(VF) and endoscopic evaluation of swallowing (Wilson, Hoare, & Johnson, 1992; Aviv et al., 1994; Macedo Filho, 1998; Murray, 1999; Hartnick et al., 2000); cervical auscultation with a stethoscope, accelerometer, or microphone (Eicher et al., 1995; McKaig, 1999); surface electromyography (Crary & Baldwin, 1997), swallowing ultrasonography (Brown & Sonies, 1997); and virtual endoscopy with computed tomography (CT) using reconstruction *software* (Burke et al., 2000).

The dynamic study of swallowing using videofluoroscopy is reputed to be the best method for the evaluation of the structures involved and the only one capable of recording radiological images of the process in real time. When preceded by adequate clinical history, this process can characterize the degree of dysfunction and identify the cause of the anomaly with accurately.

Santos (2006) proposes the analysis of swallowing sounds captured by the *sonar Doppler* instrument for therapeutic diagnosis and monitoring of dysphagia.

Hamlet, Nelson, and Patterson (1990) developed a theory about the cause of swallowing sounds while reiterating that the most prominent feature of such acoustic sounds corresponds to the movement of the bolus through the upper esophageal sphincter. They affirmed that a periodic noise, perhaps of laryngeal source, "explodes" with proximity to the closure of the cricopharyngeal muscle and that hyoid, laryngeal, and epiglottal movement can contribute to the acoustic signal when swallowing.

Cichero and Murdoch (1998) describe 3 components that form the swallowing sound: a weak signal associated with the raising of the larynx and the passage of the bolus through the pharynx, a loud sound associated with the opening of the cricopharyngeal sphincter, and a weak signal associated with the descent of the larynx after swallowing.

Santos (2006), in a study based on the aforementioned Cichero and Murdoch's theory, analyzed the time and acoustic sound frequency during performance of swallowing by 50 healthy subjects by tracing the normal pattern of swallowing through the Doppler effect.

Objective

The present study was designed to evaluate the sounds of swallowing function using videofluoroscopy and *sonar Doppler* in patients with spinocerebellar ataxia and complaints of oropharyngeal dysphagia in order to:

1. Evaluate the severity of dysphagia through a dynamic study of swallowing using videofluoroscopy.
2. Compare the differences in frequency, intensity, and duration of the swallowing sound wave in normal and SCA patients by using *sonar Doppler*.

Method

This study was approved by the Ethics Committee in Research (CEP) of the Universidade Federal do Paraná [Federal University of Parana] (UFPR) under Protocol 2072.239/2009-11.

The 30 volunteers participating in this study were diagnosed as being carriers of SCA and complained of oropharyngeal dysphagia. They were referred by the First Aid Post for Movement Disorders at the Hospital de Clinicas (HC), Curitiba. Fifteen (50%) were women and 15 (50%) were men (age range, 28–62 years; average, 44.9 years).

Participants underwent clinical diagnosis for SCA and molecular genetic examination for definitive diagnosis of SCA. Of the 30 subjects, 4 were type 2; 13, type 3; 1, type 6; 4, type 7; and 8, type 10.

The control group consisted of 30 adult volunteers (15 women and 15 men; age range, 28–69 years; average, 46.4 years) with no diagnosis of neurological disease.

The Siemens Axiom model R100 X-ray machine and Siemens monitor model M44-2 were used to carry out the swallowing videofluoroscopy evaluation. The images were digitized on the HP Pavilion tx 2075BR notebook through the TV Capture card USB Sapphire Wonder TV.

The food consistencies used for videofluoroscopic evaluation of swallowing and sound capture by *sonar Doppler* followed the pattern of the American Dietetic Association (ADA, 2002): Liquid—70 mL water and 30 mL of 100% barium sulfate. Pudding—70 mL of water, 30 mL of 100% barium sulfate, and 10 g thickening agent *Resource*® ThickenUp® (Nestlé S.A., Vevey, Switzerland) (above 1751 Cp). Solids: Club Social® biscuits soaked in barium Guedert.

For the videofluoroscopic evaluation of swallowing and sound capture by *Sonar Doppler,* a minimum of 3 swallowing samples were taken for each of the food consistencies: solid, liquid, and pudding. During the examination, data on oral and pharyngeal phases of swallowing were observed, collected, and classified according to the Classification for Severity of Dysphagia to Videofluoroscopy (Ott et al., 1996).

The continuous-wave *Doppler* equipment used in the study to capture the swallowing was a portable ultrasonic detector (model DF-4001 Martec, with a flat disc transducer, single crystal, providing the interface to the *Doppler*. The frequency of *Doppler* ultrasound is 2.5 MHz, with output of 10 mW/cm², and power sound output is 1 W. The equipment was attached to a standard HP Pavilion 2075BR tx notebook computer. The operating system was Windows Vista Home Premium.

The acoustic signals were recorded and later analyzed, using the 2.8h version of VoxMetria software (developed by CTS Computers) and elaborated further using the method described by Behlau and Michaelis (2003).

The *sonar Doppler* transducer was placed on the right side of the neck on the lateral edge of the trachea, just below the cricoid cartilage, characterized by Takahashi, Groher, and Michi (1994) as the best place for cervical auscultation. The beam of ultrasonic energy emitted by the transducer was positioned to form an angle of 30–60°. In order to decrease ultrasound dispersion into the air and increase its body transmission and echo, *contact* gel was used on the area where the transducer was applied.

After registration of the individual (name, age, and address), a sound file was created for analysis. The software's voice analysis function was chosen to record swallowing sounds using the following parameters: audio signal, intensity and fundamental frequency. The *Doppler* device volume was adjusted to number 1 for effective capture of the audio signal by the VoxMetria program and lower external noise interference. A speech therapist with experience in collection and analyses of swallowing sounds using the *sonar Doppler* reviewed a randomly chosen 50% of the samples.

Profile of Swallowing Sound Variables

- Initial frequency (IF) of sound wave: Frequency at the beginning of the acoustic signal measured in Hz
- Peak frequency (PF) of sound wave: Frequency of the highest point of displacement of the acoustic signal measured in Hz
- Initial intensity (II) of sound wave: Intensity at the beginning of the acoustic signal measured in dB.
- Swallowing time (T): Time elapsed from the beginning to the end of the acoustic signal analysis, as measured by the audio signal, in seconds (Figure 1).

The above variables are associated with numbers 1, 2, and 3, which represent solid, liquid, and pudding consistencies, respectively, as IF1, PF1, II1, IP1, and T1 for those variables during swallowing of solid food; IF2, PF2, II2, IP2, and T2 for those variables during swallowing liquid-consistency food; and IF3, PF3, II3, IP3, and T3 and the color blue for those variables during swallowing food of pudding consistency.

To plot individuals' swallowing sounds in terms of frequency, intensity, and duration of swallowing, a sample was selected that was considered to be the best audio and visual representation (graph of frequency and intensity).

The statistical methodology used in this study consisted of descriptive analysis (average) and inferential statistics (significance test).

Figure 1. Sonar Doppler.

Table 1. Prevalence of findings of videofluoroscopic study of swallowing oral and pharyngeal stages in patients with SCA

Oral Phase	Solid		Liquid		Pudding	
	E	I	E	I	E	I
Preparation and chewing	14 (46.67%)	16 (53.33%)	30 (100%)		24 (80%)	6 (20%)
Oral ejection	25 (83.33%)	5 (16.67%)	30 (100%)		25 (83.33%)	5 (16.67%)
Coordination between oral and pharyngeal	25 (83.33%)	5 (16.67%)	25 (83.33%)		25 (83.33%)	5 (16.67%)
Pharyngeal Phase	Solid	Liquid	Pudding			
	A	P	A	P	A	P
Penetration Laryngeal	30 (100%)		30 (100%)		30 (100%)	
Vacuum Laryngeal	30 (100%)		30 (100%)		30 (100%)	
Residues in valleculae, epiglottis, and piriformis	25 (83.33%)	5 (16.67%)	25 (83.33%)	5 (16.67%)	25 (83.33%)	5 (16.67%)

Notes: E = Effective, I = Inefficient, A = Absent, P = Present.

Student's *t*-test was used to determine statistical significance of the acoustic data, with variance equal to 2 samples and a significance level of $p < 0.05$.

The Spearman correlation was calculated using a significance level of 0.05.

Results

Table 1 shows the predominant alteration in the oral phase owing to a reduction in chewing movements and preparation of the bolus.

Application of the swallowing severity rating proposed by Ott et al. (1996) shows that 22 patients (73.33%) had mild dysphagia (Table 2).

During sound recording using Sonar Doppler, the mean baseline frequency (IF) and the initial intensity (II), which represent the beginning of swallowing, were higher in SCA patients than in the control group, showing a slight sound vibration increase for the top of the pharyngeal phase of swallowing. This was not observed in the frequency (FP) and the final intensity (FI), which represents the sound vibrating the opening of cricofaringe, that was inferior in two variables, showing the normal pattern very close to fulfilling this opening. The time (T) was lower in patients with SCA dominant in the control group (Table 3).

Discussion

This cross-sectional and comparative study assessed oropharyngeal swallowing and its related sounds in 30 individuals diagnosed with SCA using videofluoroscopy associated with *sonar Doppler*. This allowed for the analysis

Table 2. Videofluoroscopy-based classification of dysphagia by using the severity scale of Ott et al.

Ott et al classification	Number of patients	%
Normal	6	20%
Mild dysphagia	24	80%
Moderate dysphagia	0	0
Severe dysphagia	0	0

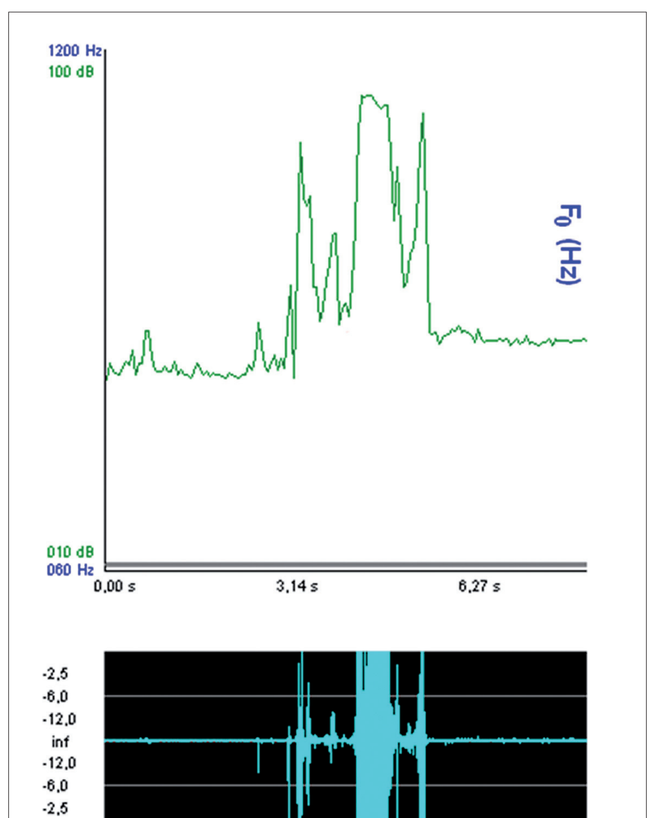

Figure 2. The acoustic signals were recorded and analyzed using VoxMetria software.

Table 3. Comparison of averages between SCA patients and controls using Student's *t*-test

Consistency and variable	SCA		Control		*P* value
	n_1	Mean (SD)	n_2	Mean (SD)	
Solid					
Time	30	1,37	30	1.59	0.0366
Initial Frequency (Hz)	30	675,96	30	558.20	*0.0003
Peak Frequency (Hz)	30	996,22	30	1086.94	*0.0004
Initial Intensity (dB)	30	58.04	30	51.92	*0.0067
Final Intensity (dB)	30	84.14	30	91.08	0.0005
Liquid					
Time	30	1.24	30	1.84	*0.0000
Initial Frequency (Hz)	30	671,75	30	592.79	*0.0000
Peak Frequency (Hz)	30	982,01	30	1092.93	*0.0031
Initial Intensity (dB)	30	58.29	30	50.86	*0.0000
Final Intensity (dB)	30	85. 36	30	91.16	*0.0004
Pudding					
Time	30	1.28	30	1.72	*0.0001
Initial Frequency (Hz)	30	682.16	30	590.97	*0.0003
Peak Frequency (Hz)	30	1004,30	30	1096.08	*0.0003
Initial Intensity (dB)	30	58.28	30	51.60	*0.0002
Final Intensity (dB)	30	84.76	30	92.05	*0.0003

Notes: *Significant at $p < 0.05$.
Values were significantly greater for the SCA group, across all consistencies and variables, than for the control group ($p < 0.05$).

of the swallowing sound signal associated with the positioning of images captured by anatomical structures, enabling more precise pinpointing of the location of each event taking place.

CNS lesions present in SCA patients create alteration of clinical features, including stomatognathic functions. Among these are the swallowing disorders (dysphagias) and dysarthrias (Lemos, 2008). In more severe cases, oropharyngeal dysphagia can bring about clinical complications such as malnutrition, dehydration, and pulmonary problems resulting from tracheal aspiration and impairment of social integration.

In the dynamic assessment of swallowing by means of videofluoroscopy, of the 30 patients studied (100%) the most predominant alteration was the lack of oral coordination in the preparation and chewing of the bolus. The patients showed reduced movements as to chewing and preparation of bolus, indicating decreased mobility. Due to this there was an increased strength in swallowing while decreasing the transit time in the pharynx. The application of the gravity classification for swallowing proposed by Ott et al. (1996) revealed that 24 patients (80%) had mild oral dysphagia (Table 2). In the present study, 24 patients (80%) presented oral dysphagia, which confirms the findings of Costa (2005) when noting that patients with SCA have greater changes in the oral phase of swallowing.

In the pharyngeal phase of swallowing, 5 individuals (16.67%) were observed to have residues in epiglottic valleculae for all consistencies of food offered; however, after multiple swallows, the food waste was cleared, thus avoiding risk of laryngeal penetration and aspiration.

A delay in the swallowing reflex may result in decreased movement of pharyngeal contraction and reduction of glottic efficiency, which, in addition to contributing to the presence of stasis in epiglottic valleculae and piriform recesses, favors the risk of laryngeal penetration and aspiration in these individuals (Logemann et.al., 1983).

It is noteworthy that the patients showed an aspect of dysarthria, which was observed during the exchange of information prior to evaluation of swallowing. Nearly all patients in this study had alterations in the oral and pharyngeal phases of swallowing during videofluoroscopic evaluation, with prevalence in solid consistency (Tables 3 and 4). This confirms the findings of Coutinho (1996), Pontes et al. (2008), and Wolf (2008), which suggest that that dysarthria contributes to the emergence of oropharyngeal dysphagia and jeopardizes the orofacial muscles.

These muscles are responsible for speech disorders and ejection of the bolus from the oral cavity into the esophagus, and weakness of this musculature can cause sporadic gagging, primarily from solid foods, which leads to increased risk of laryngeal penetration and aspiration.

The study identifies and compares the specific characteristics of the sound curves evaluated by means of *sonar Doppler that are* associated with swallowing, as visualized under videofluoroscopy, in patients of SCA dominant types 2, 3, 6, 7, and 10 and healthy adults.

Average IF and II, which represent the beginning of swallowing, they were higher in patients with SCA than in the control group. In other words, we observed increased vibration in the uptake of sound wave to the top of the pharyngeal phase of swallowing. In the final frequency (FP) and final intensity (IF), which represent the opening of the cricopharyngeus, the result was lower vibration than the standard shown in the control group to perform this opening.

The analysis of the variance as to averages of the mean FP presented $p < 0.05$, with significance level. For Hamlet (1992), this signal represents a strong sound associated with the opening of the cricopharyngeal. Data obtained from the initial peak and final peak frequencies come together with the research carried out by Hamlet et al. (1990).

Various studies show differences in acoustic signal time for the swallowing of liquids and pastes (McKaig, 1999; Takahashi et al., 1994; Vice et al., 1990). There was agreement among researchers that the duration of the signal for liquid swallowing is 500 ms. McKaig (1999) points out that the time is specific for each individual, as duration of swallowing may be 1 s for some and 3 s for others without dysphagia.

Swallowing time (T), as described by several authors (Dodds, Stewart and Logeman, 1990, Yoshikawa et al, 2006), was lower in patients with dominant SCA than in the control group (Table 3) as a result of the oral lack of coordination of the swallowing process being much faster. During the analysis of variance between these time averages $p < 0.05$ was presented with significance level. With this it is possible to identify the time of swallowing as being greater in accordance with greater consistency of swallowed material.

Cichero and Murdoch (1998) state that 3 components comprise the swallowing sound: a weak signal associated with laryngeal raising and passage of bolus through the pharynx, a strong sound associated with the opening of the cricopharyngeal sphincter, and a weak signal associated with the descent of the larynx after swallowing. McKaig (1999) often refers to the occurrence of 2 audible clicks followed by an expiratory breath, when the bolus goes into the pharynx, in adults with no change in swallowing. In accordance with these authors, our analysis shows that there is no significant correlation between ataxia and the variables related to different food consistencies. The pickup of the swallowing sound by s*onar Doppler* begins with raising of the larynx and subsequent opening of the cricopharyngeus; as such, it does not demonstrate changes in the oral phase of swallowing, which is allowed in the study analysis only by means of videofluoroscopy.

Conclusions

1. In the dynamic assessment of swallowing by means of videofluoroscopy patients with SCA show changes in the oral phase level of swallowing (mild oropharyngeal dysphagia), due to the reduction in preparatory movements and chewing of diets offered.
2. In the analysis of variance of the averages found in the waveform pattern of swallowing sounds picked up by sonar dippler (in each variable - frequency, intensity and time) of the individual with AEC demonstrates significant correlation exists when compared with the control group.

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