ORIGINAL ARTICLE

Alterations in Voice, Speech and Swallowing in Patients With Sjögren’s Syndrome

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KEYWORDS
Sjögren’s syndrome; Voice analysis; Fiberoptic endoscopic evaluation of swallowing; Speech; Cranial nerves

Abstract

Objective: To identify and describe voice, speech and swallowing abnormalities in patients with Sjögren’s syndrome (SS).

Materials and methods: This was a prospective cross-sectional descriptive observational study. Patients with SS were interviewed and physically explored. Nasolaryngeal endoscopy, video laryngeal stroboscopy, fiberoptic endoscopic evaluation of swallowing and computerized voice spectrographic analysis (PRAAT® software) of voice and speech were also performed.

Results: We included 31 patients (96.7% women). Average time of evolution was 5 years; mean age was 48.4 years. Of these SS cases, 87% were secondary and 13% primary. Symptomatology: 70.9% dysphagia, 41.9% dysphonia, 35.4% dysglossia, 3.2% dysarthria. We found abnormalities principally in: one or more cranial nerves (V, VII, IX, X, XII) (67.7%), nasopharyngolaryngeal mucosa (77.4%), mucosal wave of vocal cords (90%), swallowing mechanism (90.3%), spectrogram of the vowels /e/ (58.06%) and /i/ (51.61%), and rhythm of the trisyllable “pataka” (35.48%).

Conclusions: Patients with SS have voice, speech and swallowing abnormalities, not only associated to xerosis but perhaps also to neurological abnormalities, probably secondary to the syndrome.

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Palabras clave
Síndrome de Sjögren; Análisis de voz; Evaluación nasoendoscópica de la deglución; Habla; Pares craneales

Introduction

Sjögren’s syndrome (SS) is an autoimmune, inflammatory disorder, chronic and slowly progressive, characterised by infiltration of specialised immune system cells (lymphocytes, monocytes and plasma cells) within the salivary and lacrimal glands. This interferes with the normal function of these exocrine glands, resulting in a significant reduction in the production and secretion of saliva and tears.

It is estimated to affect approximately 1% of the adult population worldwide, although it is also believed to be underdiagnosed. The female: male ratio is approximately 9:1. The common age for diagnosis of SS is between 40 and 60 years, but the syndrome can occur at all ages.

Xerostomia is the main oral manifestation, present in 94% of patients with SS. It causes many problems involving the oral cavity, including difficulty with articulation of words (the tongue can stick to the roof of the mouth), possible loss of clarity of speech or producing "clicks" when speaking, difficulty in chewing and swallowing and adherence of food to dental surfaces.

The main manifestations concerning the topics of this article, reported by Freeman et al., are subjective sensations of dysphagia and pharyngeal dryness, while dysphonia and abnormal laryngeal findings are rare. The findings associated with this syndrome include, among others, bamboo nodules, oscillatory mucosal waves during phonation (usually altered) and vocal nodules, which, as dysphonia, rarely represent the initial manifestation of SS.

Oesophageal dryness due to decreased saliva production is one of the causes of dysphagia. Nervous system alterations may lead to alterations in the speech mechanism (for example, dysarthria) and swallowing mechanism (dysphagia).

There are few published articles noting changes in voice, speech and swallowing, and these will be discussed in the present work.

Materials and Methods

Out of 111 patients with confirmed diagnosis of SS in an ophthalmology hospital, we excluded 78 (those aged over 65, suffering diabetes, hypertension, dyslipidemia or without rheumatological confirmation of the disease and those who could not be located), and removed a further 2 (due to hearing loss and to non-confirmed primary disease). The final number of patients was 31.

We elaborated a clinical history, carried out a physical examination supported by the GRABS phoniatric scale for perceptual analysis of dysphonia and an exploration of the cranial nerves involved in speech and swallowing (V, VII, IX, X, XII), as well as searching for hyposmia.

We carried out nasofibrolaryngeal endoscopy (NFL) and video laryngostroboscopy following the usual techniques using a MediaStroboscope II system Atmos device, a Richard Wolf 90° flexible nasopharyngeal laryngoscope with an Atmos camera, an Atmos L endo-stroboscope and an Optim cold light source by Welch Allyn. We evaluated the anatomy and function of the nasal fossae, velopharyngeal sphincter, pharyngeal walls, posterior third of the tongue, epiglottis, arytenoids, vocal folds, ventriculare bands, pyriform sinuses and vallecula. Using a strobe light, we assessed the vocal folds at rest and in sustained /i/ in medium vocal register, observing fine vibration and glottic closure.
Table 1  Report and Assessment of the Fiberoptic Endoscopic Evaluation of Swallowing.

<table>
<thead>
<tr>
<th>Swallowing Alterations</th>
<th>Nectar, ml</th>
<th>Pudding, ml</th>
<th>Liquid, ml</th>
<th>Solid, biscuit</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>3</td>
<td>5</td>
<td>10</td>
<td>15</td>
</tr>
</tbody>
</table>

**Swallowing efficiency**

*Oral phase*
- Swallowing apraxia
  - Yes
  - No
- Bolus control
  - Normal
  - Insufficient
- Deficit in bolus propulsion
  - Yes
  - No

*Pharyngeal phase*
- Presence of residue in vallecula
  - Yes
  - No
- Presence of residue in pharyngeal walls
  - Yes
  - No
- Presence of residue in retrocricoid space
  - Yes
  - No
- Presence of residue in pyriform sinus
  - Yes
  - No
- Presence of nasopharyngeal reflux
  - Yes
  - No
- Deficit in UES opening
  - Yes
  - No

**Swallowing safety**

*Oral phase*
- Incompetence of glossopalatal closure
  - Yes
  - No
- Presence of preswallowing penetration
  - Yes
  - No
- Presence of preswallowing aspiration
  - Yes
  - No

*Pharyngeal phase*
- Affected laryngeal pharyngeal sensitivity FEES
  - Yes
  - No
- Presence of baseline secretions FEES
  - Yes
  - No
- Delay in swallowing reflex trigger
  - Yes
  - No
- Laryngeal protection
  - Yes
  - No
We also carried out a fiberoptic endoscopic evaluation of swallowing (FEES), which basically consists of an NFL carried out while the patient swallowed boluses of different consistencies and volumes. Endoscopic examination was initially performed without a bolus, to observe the baseline.\textsuperscript{9,10} The positive data were collected in the data collection sheet (Table 1) both for the exploration without bolus and for the exploration with different consistencies. We used Tick-It 2 thickener and prepared consistencies for nectar and pudding according to their own technical specifications. The solid consistency was evaluated with $\frac{1}{4}$ and $\frac{1}{2}$ of 3.8 g wheat biscuit and the liquid with milk. All boluses were stained with vegetable food colouring to distinguish them easily during endoscopy. We started the FEES with the nectar consistency, followed by pudding and liquid at 3, 5, 10, 15 and 20 ml successively, and lastly the solid ($\frac{1}{4}$ and $\frac{1}{2}$ wheat biscuit). The evaluation of each consistency was suspended at the volume in which aspiration appeared (corroborated by a second bolus of the same volume and consistency) and continued with the next safest smaller consistency to the smaller volume. We evaluated the efficiency and safety of swallowing in the oral and pharyngeal phases according to the parameters specified in Table 1.

We used PRAAT software for acoustic analysis of voice and speech, after recording the voice of each patient with a sample window of 44,100 Hz, at a distance of 10 cm between the mouth and the high frequency resolution microphone fitted with an Avance AC 97 audio device. We recorded the emission of vowels /a/e/i/o/u/ for 3 s each, a repetitive sequence of monosyllables /papapa/, /tatata/, /kakaka/ and the trisyllable /pataka/ at the fastest possible speed.

With the digitised signal, using a narrow band and a broad band spectrogram, we determined for each vowel: F1 and F2 (except for the vowel /a/, for which F0 was determined, as well as the first 5 formants [F]); presence of deaf segments in various frequency bands (0–4000 Hz, 4001–10,000 Hz and 10,001–14,000 Hz), and alteration of the morphology of the formants. We classified the vowels spectrographically according to Yanagihara\textsuperscript{11} and Núñez et al.\textsuperscript{12} based on the most affected vowel for each patient: Type I: absence of harmonics in the high frequencies, above 4 kHz and noise component in formant regions; Type II: Type I + absence of harmonics between F1 and F2; Type III: Type II + disappearance of F2; Type IV: Type III + disappearance of F1. We also determined the intensity in SPL decibels for the vowel /a/. For the emission of the monosyllables /pa/ta/ka/ and the trisyllable /pataka/, we analysed the presence of aperiodic segments (AS)\textsuperscript{13} and the percentage of AS in the voice sample,\textsuperscript{11} using the broad band spectrogram, and segments of articulation disruption through oscillograms. We analysed the statistical data with Microsoft Office Excel 2003 and JMP 7.0.

**Results**

We included 31 patients (1 male and 30 females), with a mean age of 48.4 ± 10.30 years. They had no history of risk for phoniatric alterations or vocal pathology that could be distinguished from SS. A total of 48.38% ($n=15$) referred gastritis and/or gastroesophageal reflux.

Four cases (13%) were primary SS (SS1) and 27 (87%) were secondary SS (SS2). Of the latter, 24 were secondary to rheumatoid arthritis, while the remaining were secondary to systemic lupus erythematosus, scleroderma and juvenile RA. The diagnosis of SS had, on average, 5 years and 9 months. The feeling of mouth and eye dryness began before diagnosis in the 31 patients; xerostomia began a mean 6.5 years before and xerophthalmia, 6 years and 1 month. In all, 13/31 patients (41.9%) reported voice problems (10 husky voice, 1 bitonality, 1 hyperrhinophonia and 1 decreased intensity), which became exacerbated with vocal use and dry
environment, and improved with intake of fluids. In total, 12 of the 31 patients (38.7%) reported speech problems (8 with lingual-palatal diglossia [adherence of the tongue to the palate], 1 with pharyngeal-palatal diglossia [adherence of the palate to the pharyngeal walls] and 3 with both diglossia and lingual weakness). Up to 22 of the 31 patients (70.9%) reported dysphagia (17 to solids, 1 to liquids and 4 to both consistencies). The typical sensation was a lack of descent of the bolus (54.83%, n=17) at the level of the pharynx (61.29%, n=19) (Table 2).

Dysphonia, dysphagia and speech problems generally had a late onset. Out of 13 patients with dysphonia, only 31% suffered it before the diagnosis was established, as was also the case with 33% of the 12 patients having speech problems. Finally, only 27% of the 22 patients with dysphagia started with these problems before the diagnosis was established.

Upon physical examination (Table 2), 20 patients (64.51%) presented alterations in the nasal mucosa (hyperaemic and/or dry) and 2 (6.45%) presented hyposmia. We found dental abnormalities (caries and/or edentulism) in 15 patients (48.38%) and dry and/or cracked tongue in 14 (45.16%). Motor impairment and/or sensory impairment, unilateral or bilateral of 1 or more cranial nerve pairs (V, VII, IX, X, and XII) were present in 21 patients (67.7%). The tone of voice in all patients corresponded to their age and gender, and the intensity was normal. The dysphonia of the 31 patients was scored from 0 to 3 (normal, mild, moderate and advanced), using the GRABS system, in the following aspects: G (global grade of severity of dysphonia) 29%=0, 71%=1; R (roughness) 29%=0, 64.5%=1, 6.5%=2; A (asthenia), B (breathiness) and S (strain) 100%=9. None presented rhythm disturbances in their speech during examination.

### Nasofibro-laryngoscopy and Video Laryngostroboscopy

One or more NFL alterations were presented by all patients: 24 patients (77.41%) altered nasopharyngeal laryngeal mucosa (hyperaemia, xerosis and/or thick discharge), 2 patients (6.46%) presented central incompetence (less than 5%) during the issuance of velar phonemes, lingual hyperkeratosis was observed in 6 patients (19.35%), deviated laryngeal pharyngeal axe in 13 (41.93%), oedematous epiglottis in 1 (3.23%), oedematous arytenoids in 18 (58.06%), and ventricular bands with unilateral or bilateral hypertrophy in 5 (16.12%). The pyriform sinuses and vocal folds were normal in all cases. During phonation, we observed glottal closure defect in 15 cases (48.38%) (8 with longitudinal hiatus, 1 with a defect in the anterior third, 1 in the posterior third and 5 in the middle). The stroboscopy showed an altered rippling of the mucosa in 28 cases (90.32%), all of them with decreased amplitude, 26 with reduced vertical movement, and 8 with phase asymmetry.

### Fiberoptic Endoscopic Evaluation of Swallowing (Table 3)

In total, 90.32% (n=28) of patients presented swallowing disorders. The efficiency (28/28 patients) was more altered than the safety (3/28 patients). In swallowing efficiency, the most altered phase was the pharyngeal (27/28 patients).
Dysphonia was reported by patients in smaller numbers. However, our study emphasises, in terms of population, of which 2 had a single interruption, 4 had 2 interruptions, 2 had 3 interruptions and 3 patients had 5 interruptions (Fig. 1).

The neurological disorder in one or more cranial nerves was present in 31 patients (48.38%) and was described as the typical lingual clicking during speech. Dysphagia was established as the most common problem, most-affected gender and dominance of SS2, as well as in the presence of gastritis and/or gastroesophageal reflux as comorbidities of the syndrome.

Our study differed with respect to others in that the time between onset of dry eye and mouth symptoms and the establishment of diagnosis was short. However, these data are unreliable due to the subjectivity of the symptoms, and included all patients presented it.

Many of our patients reported disorders in voice, speech and/or swallowing, also mentioned in other studies as part of SS. However, our study emphasises, in terms of percentage, the total patients presented one or more altered consistencies when assessing the safety of swallowing.

### Table 3
Fiberoptic Endoscopic Evaluation of Swallowing.

<table>
<thead>
<tr>
<th>Swallowing Alterations</th>
<th>Nectar</th>
<th>Pudding</th>
<th>Liquid</th>
<th>Solid</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>3</td>
<td>5</td>
<td>10</td>
<td>15</td>
</tr>
<tr>
<td><strong>Efficacy</strong> (n=28)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Oral phase</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>1</td>
</tr>
<tr>
<td>Pharyngeal phase</td>
<td>3</td>
<td>2</td>
<td>-</td>
<td>3</td>
</tr>
<tr>
<td><strong>Safety</strong> (n=3)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Oral phase</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Pharyngeal phase</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
</tbody>
</table>

Note: each cell indicates the number of patients who had presented alterations in certain consistencies and volumes.

#### Discussion and Conclusions
The characteristics of our population matched those reported by other authors in terms of age, most-affected gender and dominance of SS2, as well as in the presence of gastritis and/or gastroesophageal reflux as comorbidities of the syndrome.

The characteristics reported by other authors in terms of age, most-affected gender and dominance of SS2, as well as in the presence of gastritis and/or gastroesophageal reflux as comorbidities of the syndrome.

The alterations of low cranial nerves were numerically important (67.7%), with these results being similar to those published by Fuentealba, who reported some type of...
Table 4  Acoustic Analysis of Voice and Speech.

<table>
<thead>
<tr>
<th>Voice</th>
<th>a</th>
<th>e</th>
<th>i</th>
<th>o</th>
<th>u</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean value of formants in female patients</td>
<td>Fo: 212±28 Hz</td>
<td>F1: 492±38 Hz</td>
<td>F1: 358±53 Hz</td>
<td>F1: 526±48 Hz</td>
<td>F1: 448±50 Hz</td>
</tr>
<tr>
<td></td>
<td>F1: 788±103 Hz</td>
<td>F2: 2346±151 Hz</td>
<td>F2: 2596±207 Hz</td>
<td>F2: 989±79 Hz</td>
<td>F2: 849±69 Hz</td>
</tr>
<tr>
<td></td>
<td>F2: 1547±141 Hz</td>
<td>F3: 2801±243 Hz</td>
<td>F3: 1274 Hz</td>
<td>F4: 2038 Hz</td>
<td>F4: 358 Hz</td>
</tr>
<tr>
<td></td>
<td>F4: 3961±246 Hz</td>
<td>F5: 3830 Hz</td>
<td>F5: 989±79 Hz</td>
<td>F5: 849±69 Hz</td>
<td>F5: 448±50 Hz</td>
</tr>
<tr>
<td>Value of formants in male patient</td>
<td>Fo: 158 Hz</td>
<td>F1: 457 Hz</td>
<td>F1: 316 Hz</td>
<td>F1: 488 Hz</td>
<td>F1: 464 Hz</td>
</tr>
<tr>
<td></td>
<td>F1: 655 Hz</td>
<td>F2: 2038 Hz</td>
<td>F2: 2311 Hz</td>
<td>F2: 970 Hz</td>
<td>F2: 971 Hz</td>
</tr>
<tr>
<td></td>
<td>F3: 1274 Hz</td>
<td>F4: 2303 Hz</td>
<td>F4: 989 Hz</td>
<td>F4: 849 Hz</td>
<td>F4: 448 Hz</td>
</tr>
<tr>
<td></td>
<td>F5: 3830 Hz</td>
<td></td>
<td>F5: 849 Hz</td>
<td>F5: 448 Hz</td>
<td></td>
</tr>
</tbody>
</table>

Altered formants\a
- F1 1 patient
- F2 1 patient
- F3 1 patient
- F4 1 patient
- T 1 patient

Frequencies with aperiodic segments

I | - | - | - | - | - |
II | 11 patients | 11 patients | 10 patients | 5 patients | 5 patients |
III | 4 patients | 7 patients | 6 patients | 1 patient | 1 patient |

Speech

<table>
<thead>
<tr>
<th>Pa</th>
<th>Ta</th>
<th>ka</th>
<th>Pataka</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 episode</td>
<td>1 patient</td>
<td>-</td>
<td>1 patient</td>
</tr>
<tr>
<td>2 episodes</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>3 episodes</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>4 episodes</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>5 episodes</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
</tbody>
</table>

Frequencies with aperiodic segments

I | 2 patients | 3 patients | 5 patients | 5 patients |
II | 13 patients | 14 patients | 15 patients | 15 patients |
III | 16 patients | 3 patients | 2 patients | 2 patients |

Freeman et al.\d We should also note that the laryngeal pharyngeal axis was found to deviate in 13 patients and that 2 patients presented velopharyngeal incompetence, which also leads us to consider an underlying neurological component, and not only xerosis. We did not find laryngeal lesions associated with autoimmunity and SS, such as bamboo nodules\v and vocal nodules.\w The clear alteration of mucosal wave vibration detected by stroboscopy points to a loss of elasticity of the vocal folds, probably secondary to their dryness.\x However, we cannot discard a possible neurological component (we must bear in mind the asymmetry of the laryngeal pharyngeal axis), which conditions certain alterations such as phase asymmetry).

Evaluation through FEES dramatically reveals the importance of dysphagia in these patients, who, despite not presenting a clear alteration of safety in swallowing, did present, in over 90% of cases, impaired efficiency. This carried a significant risk of malnutrition (not presented clinically by our patients) as well as social limitations.
Figure 1  Spectrogram and oscillogram of patients with alterations in voice and speech. (a) 4 first formants of the vowel /a/, showing irregularity of the morphology with all formants. (b) Broadband spectrogram of the vowel /o/, showing the aperiodic segment. (c) Normal plot in the oscillogram of the trisyllable /pataka/. (d) Abnormal plot in the oscillogram of the trisyllable /pataka/, where the downwards arrows show the segments with rhythm alterations. (e) Broadband spectrogram of the trisyllable /pataka/ analysed in the upper oscillogram, indicating the aperiodic segments.
With regard to voice analysis, the formants of the vowel /a/ corresponded, by gender, to the reports in the medical literature.\textsuperscript{13,19,20} The rest of the vowels presented F1 and F2, in general within the higher values reported for these formants.\textsuperscript{19} The exception was the elevation of F2 for the vowel /u/ by the only male in our study, according to the values reported by Balari et al.\textsuperscript{19} and Rosique et al.\textsuperscript{20} This may be explained by the fact that this formant is directly related to the position of the tongue: the more elevated and anterior it is, the higher the frequency. In the /u/ vowel, the tongue occupies the most posterior position; however, lingual palatal diglossia can establish F2 at higher frequencies.\textsuperscript{21} Nevertheless, for other publications,\textsuperscript{22} the values of this formant are within the established range.

As noise parameters, we studied the presence of AS and their percentage in the voice sample. These segments should not appear in normal voices and are related to a poor production of harmonics in the glottis, either through lack of energy in vocalisation or through anatomical alteration\textsuperscript{12} such as insufficient lubrication of the vocal folds. These AS affected the adjacent F in 7 patients, although they did not predominate over the Fs or replace them in any case, so the classification was Yanagihara Type I for all those who presented any of these AS. The presence of AS in the /a/ vowel increased strikingly after giving this vowel prior reinforcement with the consonants /p/t/k/ (which are aperiodic by definition\textsuperscript{23,24} and therefore increased the amount of irregularities during phonation).

In our study, we focused on observing the changes between 0 and 14,000 Hz. This decision was made because, although spectrographically, the voice concentrates its highest energy between 20 and 4000 Hz, some phonemes have much higher frequencies (for example, fricative sounds, which can reach 10 kHz). However, the loss of this information does not represent a substantial deficit in speech intelligibility. This is because most of the information required for it is below 4 kHz.\textsuperscript{24} Formant frequencies (less than 4 kHz) were not affected in isolated vowels, and were the least altered in association with /p/t/k/, which presents a qualitatively acceptable voice for the majority of patients (coinciding with our assessment using the GRABS scale), bearing in mind that the presence of alterations at higher frequencies represents a morphofunctional glottal alteration.

As for speech, 13 patients presented rhythm alterations during the articulation of the trisyllable /pataka/, which led us to intentionally seek this type of change in the population with SS, as patients did not present it during spontaneous speech (perhaps because they had learned to compensate for their articulation disorders). This alteration also led us to consider how far xerosis and the neurological component intervened, given that the latter could not be associated with statistical significance in our study. This may be due to the small number of patients; however, it could be our basis for pursuing further research. This study will also serve as a basis for further research on voice and swallowing analysis in SS.

We conclude that Sjögren’s syndrome should be conceptualised as a systemic disease that requires multidisciplinary intervention to improve the quality of life of patients.

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