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

Laura Daniela Ruiz Allec a,∗, Xochiquetzal Hernández López a, Juanita Beatriz Arreguín Porras a, Regina Velasco Ramos b, Julia Cristina Pacheco del Valle b, Ángel Israel Pérez García b

a División de Foniatría, Instituto Nacional de Rehabilitación, Mexico City, Mexico
b Departamento de Córnea y Cirugía Refractiva, Fundación Hospital Nuestra Señora de la Luz, Institución de Asistencia Privada, Mexico

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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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∗ Corresponding author.
E-mail address: danyallec@yahoo.com (L.D. Ruiz Allec).

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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.

Alterations in these glands may lead to alterations in the speech mechanism (for example, dysarthria) and swallowing mechanism (dysphagia).

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, and XII), as well as searching for hyposmia.

We carried out nasofibro-laryngeal 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, ventricular 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 5 10 15 20</td>
<td>3 5 10 15 20</td>
<td>3 5 10 15 20</td>
<td>1/4 1/2</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.\(^9,10\) 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 ¼ and ½ 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 (¼ and ½ 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\(^9\) and Núñez et al.,\(^12\) 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)\(^13\) and the percentage of AS in the voice sample,\(^11\) 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
ALTERATIONS IN VOICE, SPEECH AND SWALLOWING IN PATIENTS WITH SJÖGREN'S SYNDROME

Table 2

<table>
<thead>
<tr>
<th>Type of SS</th>
<th>Alteration Symptoms</th>
<th>Physical Exploration</th>
<th>Studies</th>
</tr>
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<tbody>
<tr>
<td></td>
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<td></td>
<td>Alteration Acoustic Analysis</td>
</tr>
<tr>
<td></td>
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<td></td>
<td>Fiberoptic endoscopic evaluation of swallowing: GERD: gastroesophageal reflux disease; NFL: nasal fibro-laryngoscopy; SS: Sjögren's syndrome; SS1: primary; SS2: secondary.</td>
</tr>
<tr>
<td>Gastritis and/or GERD</td>
<td>Voice Problem</td>
<td>Dental Alteration</td>
<td>Altered NFL</td>
</tr>
<tr>
<td>Speech Problem</td>
<td>Swallowing Problem</td>
<td>Lingual Alteration</td>
<td>Hyposmia Altered</td>
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<td></td>
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<td>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.</td>
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<td>+FEES: fiberoptic endoscopic evaluation of swallowing; GERD: gastroesophageal reflux disease; NFL: nasal fibro-laryngoscopy; SS: Sjögren's syndrome; SS1: primary; SS2: secondary.</td>
</tr>
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<td></td>
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<td>See text for reference on what was considered abnormal for each of the sections of this table.</td>
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<td></td>
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<td>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.</td>
</tr>
<tr>
<td></td>
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<td></td>
<td>Fiberoptic Endoscopic Evaluation of Swallowing (Table 3)</td>
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<tr>
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<td></td>
<td></td>
<td>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)</td>
</tr>
</tbody>
</table>
for solid consistency (24/28 patients). According with the data collection system (Table 1), only the bolus control was affected in the evaluation of oral efficacy (n=4); the most affected structure by food residues in the pharyngeal phase was the vallecula (n=24). Oral safety presented involvement only in one case, due to incompetence of glossopalatal closure, while pharyngeal safety was affected in 2 cases by decreased laryngeal pharyngeal sensitivity, accompanied by laryngeal penetration during swallowing in one case and delayed swallowing reflex in the other. Table 3 details the findings according to consistency and volume.

Acoustic Analysis of Voice (Fig. 1 and Table 4)

A total of 28 patients presented abnormalities in one or more parameters of voice analysis and/or rhythm of speech; all of them were classified as Yanagihara Type I. The average phonation intensity was 67 + 10 dB SPL. Up to 15 of the 31 patients (48.38%) presented AS for the vowel /a/, 18/31 (58.06%) for the vowel /e/, 16/31 (51.61%) for the vowel /i/, 6/31 (19.35%) for the vowel /o/ and 4/31 (12.90%) for the vowel /u/. The percentage of AS in the voice sample for these vowels was: /a/ 67.93%, /e/ 74.44%, /i/ 74.35%, /o/ 76.20% and /u/ 62.5%. The AS by frequency and the formant values (and their alterations) for each vowel are described by gender in Table 4.

In the sequences of monosyllables /pa/ta/ka/ and in the trisyllable /pataka/, AS were observed in the accompanying vowel /a/; with 16/31 (51.61%) altered registries for /pa/, 20/31 (64.51%) for /ta/ and 22/31 (70.96%) for /ka/ and 22/31 (70.96%) for /pataka/. The frequencies with AS are shown in Table 4.

The sequence of the trisyllable /pataka/ presented substitutions, omissions and distortions of consonants. In 11/31 patients (35.8%), there was alteration in the rate of articulation, 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 had no statistically significant relationship ($X^2$ [1, n=31], $P=.179$) with alteration of FEES or with alterations in the rhythm of speech articulation ($X^2$ [1, n=31], $P=.2137$).

### Discussion and Conclusions

The characteristics of our population matched those reported by other authors in terms of age, most-affected gender and dominance of SS, 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 that patients might present. The sensation of dry eye and mouth was strengthened as an essential part of the symptoms, given that 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 important frequency of these symptoms.

Dysphagia was established as the most common problem, especially for solid consistencies, as expected for patients with SS. Dysphonia was reported by patients in smaller numbers (n=13) than we identified aurally during examination (n=22). Nevertheless, dysphonia was mild in all cases and did not interfere with speech intelligibility. Pharyngeal-palatal and lingual-palatal diglossia occurred in about half of our patients, logically associated to xerostomia caused by SS. Lingual-palatal diglossia is also reported by other authors as the typical lingual clicking during speech.

Few articles mention dysarthria as a characteristic alteration of SS. In our study, it was the least-common speech disorder (n=3).

With regard to the physical examination, lingual and dental alterations coincided with the literature reviewed. Nasal abnormalities were very common, occurring in over half of our population, coinciding with the report by Freeman et al., who also found nasal alterations in over half of their patients. However, other studies reported these alterations less frequently. Nasosmia (6.45%) was present in a lower proportion than reported 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</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>in female patients</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>
<td></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>F2: 1274 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>F4: 3961+246 Hz</td>
<td>F4: 492+38 Hz</td>
<td>F4: 2346+151 Hz</td>
<td>F4: 2596+207 Hz</td>
<td></td>
</tr>
<tr>
<td>Altered formantsa</td>
<td>F1 - - - - - - 1 patient - - - - - -</td>
<td>F2 - - - - - - - - - 3 patients</td>
<td></td>
<td></td>
<td></td>
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<td></td>
<td>F3 - - - - - - - - - - - - -</td>
<td>F4 - - - - - - - - - - - - -</td>
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<tr>
<td>Frequencies with aperiodic segments</td>
<td>I - - - - - - - - - - - - -</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td></td>
<td>II 11 patients 11 patients 10 patients 5 patients 5 patients</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>III 4 patients 7 patients 6 patients 1 patient 1 patient</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Speech</td>
<td>Pa</td>
<td>Ta</td>
<td>ka</td>
<td>Pataka</td>
<td></td>
</tr>
<tr>
<td>Alterations in articulation rhythm</td>
<td>1 episode 1 patient - - 1 patient 2 patients</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>2 episodes - - - - - - 4 patients</td>
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<tr>
<td></td>
<td>3 episodes - - - - - - 2 patients</td>
<td></td>
<td></td>
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<td></td>
<td>4 episodes - - - - - - - - -</td>
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<tr>
<td></td>
<td>5 episodes - - - - - - 3 patients</td>
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</tr>
<tr>
<td>Frequencies with aperiodic segments</td>
<td>I 2 patients 3 patients 5 patients 5 patients</td>
<td></td>
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<td></td>
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<tr>
<td></td>
<td>II 13 patients 14 patients 15 patients 15 patients</td>
<td></td>
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<td></td>
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</tr>
<tr>
<td></td>
<td>III 16 patients 3 patients 2 patients 2 patients</td>
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</tbody>
</table>

I: 1–4000 Hz; II: 4001–10 000 Hz; III: 10 001–14 000 Hz.

a Refers to formants that have lost their typical morphology (Fig. 1); in no case were they completely replaced by noise.

b All altered formants; none was completely replaced by noise.

neurological involvement in up to 50% of patients. However, there was great variability in the frequency of neurological alterations reported in the medical literature, and some articles referred a presentation of 22%,17,18 Neuropathy in SS has been associated to vasculitis and typically indicates the cranial nerve V as the most affected.14,16 Perhaps the clear phoniatric orientation of our research led us to detect alterations in the XII, X, and VII pairs most often, rather than in V. These alterations force us to consider the role that neurological involvement can play in the occurrence of voice, speech and swallowing alterations. On the one hand, we observed alterations due to the dryness of the oropharyngeal mucosa, such as lingual-palatal diglossia or adhesion of solid food to the pharyngeal walls. On the other, there were also alterations such as tongue deviation, decreased gag reflex, asymmetry of the oral commissure, and so on (not explicable by xerosis), which synergized the involvement of the functions discussed.

Clear chronic irritation was revealed by NFL in 77.41% of patients, a much higher figure than the 20% reported by Freeman et al.4 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 xeroses. We did not find laryngeal lesions associated with autoimmunity and SS, such as bamboo nodules5 and vocal nodules.1 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.5 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 /a/, 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. The rest of the vowels presented F1 and F2, in general within the higher values reported for these formants. 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. and Rosique et al. 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. Nevertheless, for other publications, 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 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 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. Formant frequencies (less than 4 kHz) were not affected in isolated vowels, and were the least altered in association with /p/t/k/, which represents 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 seeking 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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References
