EPIDEMIOLOGY OF VOICE AND SWALLOWING DISORDERS IN SJÖGREN'S SYNDROME

by

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ABSTRACT

Individuals with Sjögren's Syndrome (SS) have autoimmune symptoms causing systemic dryness. The disease purportedly affects voice and swallowing function. However, little is known regarding the true prevalence of voice and swallowing problems and symptoms in SS and their effects on quality of life. This preliminary epidemiological investigation aimed to (1) assess the prevalence of voice and swallowing problems in SS, (2) identify risk factors for voice and swallowing problems in SS, and (3) better understand the functional, social, occupational, and emotional effects of voice and swallowing problems in SS. One hundred and one individuals with SS (7 males, 94 females; mean age 59.4, SD = 14.1) were interviewed using an extensive questionnaire. Questions surrounding the individual's medical, psychosocial, occupational, and social/lifestyle history, as well as voice and swallowing symptoms, SS severity, and health-related quality of life, were explored. The results were analyzed using summary statistics, chi-square tests, risk ratios, and confidence intervals ($p < .05$). Of the 101 individuals with SS, 59.4% reported a current voice disorder and 64.4% reported a current swallowing disorder. These disorders began gradually, were chronic, and correlated with SS disease severity. These results indicated that voice and swallowing problems are relatively common in SS and are more frequent as disease severity worsens. Voice symptoms, including frequent throat-clearing, chronic throat soreness, difficulty projecting the voice, and discomfort with voice use, were significantly correlated with

health-related quality of life; chronic throat dryness, a monotone voice, a wobbly or shaky voice, and chronic throat soreness were significantly associated with SS disease severity. Swallowing symptoms, including difficulty swallowing medications, sneezing with eating, wheezing after eating, food sticking in the throat, increased mucous in throat, and taking smaller bites for safety were correlated with health-related quality of life; taking smaller bites, mucous in throat, difficulty placing food in the mouth, and wheezing while eating were significantly associated with SS disease severity. However, only 15.8% with voice disorders and 42% with swallowing disorders sought treatment for these symptoms. These findings have implications for evaluation and treatment paradigms in individuals with SS.

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INTRODUCTION

The capacity for oral communication and swallowing are central to human health and well-being. These behaviors, both of which share a common anatomy may be adversely affected by numerous disease states. Damage to this shared anatomy may result in the loss of two behavioral sets that are central to the human experience, namely voice and swallowing. Autoimmune diseases are associated with the body's inappropriate defense against its own healthy tissue and, consequently, may affect voice and swallowing function. Sjögren's Syndrome (SS) is one such disease. This study aims to examine the epidemiology of voice and swallowing disorders in individuals with this autoimmune disease.

Sjögren's Syndrome

Chambers (2004) and Kassan and Moutsopoulos (2004) have described SS as an autoimmune disease characterized by systemic dryness, also known as sicca. Sicca symptoms occur most notably in the eyes, mouth, throat, skin, and vagina. There are two forms of SS: primary and secondary. In general, primary SS is defined as occurring in the absence of another inflammatory autoimmune disease. Secondary SS occurs with—or secondary to—other autoimmune diseases and is more prevalent than primary SS (roughly 60% of cases). However, the primary versus secondary distinction does not necessarily account for the range in severity of symptoms. That is, individuals with

secondary SS may experience sicca symptoms as severe as those of primary SS. As reported by Vitali et al. (2002), at least four of the following six criteria must be met for a diagnosis of SS: (1) symptoms of dry eye, (2) signs of dry eye, (3) symptoms of dry mouth, (4) positive tests of salivary gland dysfunction, (5) positive salivary gland biopsy, and/or (6) autoantibodies SS-A or SS-B. Additionally, one of these criteria must be biopsy or blood sample positive for containing autoantibodies. It is noteworthy, however, that the diagnostic criteria and testing techniques for SS remain topics of discussion in the rheumatic disease medical community. These diagnostic considerations have the potential to modify criteria for assigning the SS diagnosis, as well as possibly increasing recognition of patients with subclinical or early forms of the disease (Cornec et al., 2013; Goules, Tzioufas, & Moutsopoulos, 2014). SS is slowly progressive, has no cure, and is treated symptomatically with medication, moistening agents as well as inhaled lubricants, and other therapies, such as lifestyle modifications for co-occurring laryngopharyngeal reflux (Belafsky & Postma, 2003; Hilgert, Toleti, Kruger, & Nejedlo, 2006; Kasama et al., 2008; Lee, Fang, & Li, 2002; Rhodus & Bereuter, 2000; Seitsalo et al., 2007; Strietzel et al., 2011; Tanner et al., 2013).

The etiology of SS is unknown, although it may involve genetics, hormones, and/or viral components (Mathews, Kurien, & Scofield, 2008). SS occurs most often in perimenopausal women, but it sometimes occurs in men and younger adults and teenagers (Skalova, Minxova, & Slezak, 2008). It is diagnosed in approximately 1% of the adult population. However, it is likely underdiagnosed as SS symptoms are similar to symptoms of other health conditions, particularly those related to menopause and aging, and true prevalence may be closer to 2% (Kassan & Moutsopoulos, 2004). Sicca

symptoms affect not only skin and bodily orifices but also the function of other connective tissue, internal organs, and digestion (Sjögren's Syndrome Foundation, 1998).

Reported laryngeal manifestations common to SS include bamboo nodules, esophageal reflux, laryngopharyngeal reflux, and cricoarytenoid arthritis. However, a recent study conducted by our group suggests that individuals with primary SS do not experience significant laryngeal structural or mucosal changes—other than dryness—and that in cases of secondary SS these changes may be more related to the primary disease process, such as rheumatoid arthritis or lupus (Heller et al., 2013). Individuals with SS also have auditory-perceptual features of dysphonia characterized by hoarseness, breathiness, and strain, as well as auditory-perceptual and acoustic severity values in the mild to mild-to-moderate range (Doig et al., 1971; Heller et al., 2013; Ogut et al., 2005; Ruiz Allec et al., 2011). Additionally, individuals with SS experience dysphagia due to reduced saliva and esophageal abnormalities (Belafsky & Postma, 2003; Bitter, Volk, Lehmann, Wittekindt, & Guntinas-Lichius, 2011; Caruso, Sonies, Atkinson, & Fox, 1989; Doig et al., 1971; Hilgert et al., 2006; Murano et al., 2001; Ruiz Allec et al., 2011; Seve, Poupart, Bui-Xuan, Charbon, & Broussolle, 2005; Skalova et al., 2008). Other major health issues include Raynaud's phenomenon (Seitsalo et al., 2007), neurologic components (Cheung, Chen, Hsin, Tsai, & Leong, 2010; Michel et al., 2011), chronic fatigue (Ng & Bowman, 2010), non-Hodgkin's lymphoma (Lee et al., 2002; Theander et al., 2011), and various oral manifestations such as dental caries, loss of teeth, and articulatory changes (Heller et al., 2013; Mathews et al., 2008; Ruiz Allec et al., 2011). Besides sicca symptoms, factors contributing to voice and swallowing disorders in SS may include neurological processes, cricoarytenoid joint arthritis or fixation, and

gastroesophageal reflux (Belafsky & Postma, 2003; Cheung et al., 2010; Prytz, 1980), to name a few.

Only a few studies exist that have examined the frequency of voice and swallowing problems in SS, and these indicate somewhat conflicting observations. Table 1 summarizes and critiques studies from the literature reporting epidemiological data for voice and swallowing disorders in SS. (Note. Single case studies/reports are not included in Table 1.) Most of the studies in Table 1 include typical signs and symptoms of voice and swallowing dysfunction. Though each of these studies provides important insight into effects of SS, none include a thorough analysis of the epidemiology of voice and swallowing disorders in SS. Common weaknesses include a small sample size, a lack of specificity regarding disease history and severity, a limited discussion of the specific contributing factors of disorders, a limited discussion of socioemotional effects of voice and swallowing symptoms, and the absence of psychometrically validated voice and swallowing measures.

Perhaps the largest and most relevant study examining voice and swallowing disorders in SS was conducted by Ruiz Allec et al. (2011). This group examined voice, swallowing, and speech dysfunction in SS using subjective and objective measures and discussed prevalence of each of these areas of dysfunction. However, this study included a relatively small group (n = 31), only two of whom had primary SS, and provided only a limited discussion of the risk factors that might contribute to the presence of voice and swallowing problems in this population. Additionally, limited information was provided by the authors regarding the potential adverse effects of these disorders on socioemotional functioning and/or quality of life.

Table 1. Characteristics of studies examining voice and swallowing dysfunction in SS

Study (chronologically)	# of Subjects	Gender Representation	Age	Methods	Findings	Weaknesses/ Limitations
"Otolaryngological aspects of Sjögren's syndrome" Doig et al., 1971	22 pSS 31 SS+RA 21 RA	Not specified	pSS: 30–81 mean 53.3 SS+RA: 31–78 mean 51.4 RA: 34–70 mean 51.7	Oral exam, ear exam, ophthalmological exam, indirect nasopharynx exam, indirect laryngopharynx exam, radiological barium swallow	Dysphagia: pSS 27.3%, SS+RA 32.3%, RA 9.5%; Hoarseness: pSS 27.3%, SS+RA 19.4%, RA 9.5%	Small *n*, gender not specified, limited patient history/symptom review, hoarseness is only voice symptom noted, no discussion of socioemotional effects
"Objective measures of swallowing in patients with primary Sjögren's syndrome" Caruso et al., 1989	34 pSS 34 controls	Not specified	32–72 mean 55	Salivary function via saliva collection, real-time B-scan mechanical sector ultrasound imaging during swallows	pSS: longer dry swallows and 40% had longer water bolus swallows than dry swallows; salivary gland dysfunction alone can cause dysphagia	Small *n*, gender not specified, only measured swallow times and included few other swallow complaints, did not analyze voice dysfunction, no discussion of socioemotional effects
"Quantitative assessment of dysphagia in patients with primary and secondary Sjögren's syndrome" Rhodus, Colby, Moller, & Bereuter, 1995	13 pSS 15 sSS+SLE 14 controls	pSS: 12F 1M sSS+SLE: 15F controls: matched	pSS: 54.5 ± 3.4 sSS+SLE: age 57.4 ± 2.5 controls: 56.3 ± 4.4	Oral exam, oral and written questionnaire re. xerostomia, sialometric evaluation, subjective swallowing difficulty measures, videofluoroscopy	SS and SLE: significant dysphagia sSS+SLE: more severe except in pharyngeal transit time with a water bolus	Small *n*, specific prevalence not reported, no voice symptoms studied, no discussion of socioemotional effects
"Sjögren's syndrome. New diagnostic aspects" Haga et al., 1997 [Article in Norwegian]	96 pSS	Not specified in abstract	Not specified in abstract	Clinical and laboratory diagnostic criteria	71% had hoarse voice	Article in Norwegian, only voice symptom reported was hoarseness, no report of contributing factors, no report of swallowing dysfunction, no discussion of socioemotional effects

Table 1. Continued

Study (chronologically)	# of Subjects	Gender Representation	Age	Methods	Findings	Weaknesses/ Limitations
"Ear, nose, and throat manifestations of Sjögren's syndrome: Retrospective review of a multidisciplinary clinic" Freeman, Sheehan, Thorpe, & Rutka, 2005	107 pSS 25 sSS 53 sicca syndrome	pSS: 82%F sSS: 95%F sicca: 82%F	pSS: 14–79 mean 49 sSS: 30–75 mean 55 sicca: 38–79 mean 55	Otolaryngology history, audiometry, otoscopy, rhinoscopy, indirect laryngoscopy, palpation of salivary glands and neck	Discrepancy between symptoms and abnormal exam: pSS 79%/27%, sSS 17%/5%, sicca 34%/7%, average 60–70%/20%	Not age/sex-matched, only voice symptoms specified were chronic pharyngitis or laryngitis, no discussion of swallowing, no discussion of socioemotional effects
"Laryngeal findings and voice quality in Sjögren's syndrome" Ogut et al., 2005	77 SS 77 controls	SS: 70F 7M Controls: 70F 7M	SS: 23–70 Controls: 22–72	Reflux Symptom Index, Reflux Finding Score, Jitter, Pitch Period Perturbation Quotient, Shimmer, Amplitude Perturbation Quotient, Noise-to-Harmonic Ratio	Findings of reflux in SS was statistically significant, significant association between SS and variety of other laryngeal pathologies	Did not specify if participants were pSS or sSS, did not include swallowing measures, only named reflux as potential cause of pathologies, no discussion of severity or socioemotional effects
"Dysphagia and dysmotility of the pharynx" Mandl, Ekberg, Wollmer, Manthorpe, & Jacobsson, 2007	20 pSS 30 controls	pSS: 18F Controls: 27F	pSS: 31–55.5 mean 47 Controls: 31–54.5 mean 48	15-item pharyngeal and oesophageal symptoms questionnaire, pharyngeal and oesophageal video radiography, autonomic nervous function tests	pSS: 65% dysphagia, 60% GERD, 45% pharyngeal swallowing symptom, 80% oesophageal swallowing symptom, sensory dysfunction may contribute to dysphagia	Small *n*, no pt history gathered to determine potential causes, no discussion of voice, no discussion of socioemotional effects

Table 1. Continued

Study (chronologically)	# of Subjects	Gender Representation	Age	Methods	Findings	Weaknesses/ Limitations
"Alterations in voice, speech, and swallowing in patients with Sjögren's syndrome" Ruiz Allec et al., 2011	31 SS: 4pSS 27sSS	30F 1M	48.4 ± 10.3	Clinical history, GRBAS scale, exploration of speech/swallowing cranial nerves, search for hyposmia, naso-laryngeal endoscopy, video laryngostroboscopy, FEES, acoustic speech/voice analysis	Reported: 41.94% voice problems, 48.38% gastritis and/or GERD, 38.7% speech problems, 70.9% dysphagia Observed: 70.97% voice problems, 3.23% oedematous epiglottis, 58.06% oedematous arytenoids, 16.12% hypertrophic ventricular bands, 48.38% glottal closure defect, 90.32% altered rippling of the VF mucosa, 90.32% swallowing disorders, 90.32% abnormalities in voice/speech analysis, 9.68% dysarthria	Small *n*, limited discussion of causes, limited discussion of severity and socioemotional effects
"Effects of reduced saliva production on swallowing in patients with Sjögren's syndrome" Rogus-Pulia & Logemann, 2011	20 SS: 10p 10s 20 controls	SS: 19F 1M Controls: matched	30–74	12-item questionnaire of perception of dry mouth and swallowing abilities, stimulated saliva production, FEES	SS: generally perceived their swallowing impairment but 97% of swallows were functional and only 3% aspiration	Small *n*, excluded pts w/no complaints of dry mouth, no discussion of voice

Table 1. Continued

Study (chronologically)	# of Subjects	Gender Representation	Age	Methods	Findings	Weaknesses/ Limitations
"The prevalence of dysphonia, its association with immunomediated diseases and correlation with biochemical biomarkers" Sanz et al., 2011	80 autoimmune: 44 RA, 32 SLE, 4 SS 60 controls	Auto: 66F 14M Controls: 32F 28M	Auto: 28–73 mean 58.2 Controls: 22–62 mean 48.3	VHI, TIOS dysphonia scale, autoimmune symptoms questionnaire, QOL questionnaire, biochemical and immunohistochemical measures	Autoimmune: 32–38% dysphonia; voice disorders in SS no significant prevalence; voice disorders 81% worse during flare; steroids, voice rest, and speech therapy are suggested treatments; quality of life reduced due to voice disorders	Small *n* in SS subgroup, unbalanced gender %, gender/age not specified for RA/SLE/SS, swallowing not studied

Note. pSS = primary SS; sSS = secondary SS, SLE = systemic lupus, GRBAS = Grade/Roughness/Breathiness/Asthenia/Strain, FEES = fiberoptic endoscopic evaluation of swallowing, VF = vocal fold

The current and lifetime prevalence of voice disorders in the general adult population is 6.6% and 29.9%, respectively, and voice disorders are strongly associated with sex, age, voice use patterns and demands, esophageal reflux, chemical exposures, and frequent cold/sinus infections (Roy, Merrill, Gray, & Smith, 2005). By comparison, it is reported that the prevalence of current voice disorders in SS ranges dramatically from 12% to 41.9% (Ruiz Allec et al., 2011; Sanz et al., 2011). The prevalence of current swallowing disorders in the general adult population is 12–13% (Groher & Bukatman, 1986). Reports on the prevalence of swallowing disorders in SS range from 32% to 70.9% (Doig et al., 1971; Ruiz Allec et al., 2011). Some assert that these symptoms may be related to sensory deficits (Rogus-Pulia & Logemann, 2011). Based upon these results, it appears that voice and swallowing problems are more common in SS as compared to the general population, but the true prevalence in SS remains undetermined.

While it appears obvious that SS has a significant impact on an individual's health and well-being, little is known regarding (1) the prevalence of voice and swallowing disorders in SS, (2) patterns of voice and swallowing decline in SS, (3) risk factors associated with this decline, and (4) consequences of voice and swallowing disorders on social and emotional functioning in the SS population.

Purpose

Epidemiology is the study of the relationships of various factors determining the frequency and distribution of diseases and disorders in a population. At present, the true prevalence of voice and swallowing disorders in SS is undetermined. This descriptive,

epidemiological investigation interviewed a sample of patients with SS to provide much needed data regarding voice and swallowing disorders associated with these autoimmune conditions. There were three specific aims related to this study: (1) *to provide data regarding the prevalence of voice and swallowing disorders in the SS population*, (2) *to identify risk factors for voice and swallowing disorders in SS, and* (3) *to better understand the functional, social, occupational, and emotional effects of voice and swallowing disorders in SS.*

METHODS

Participants

Participants included 101 adult (age 18+) individuals identified and recruited from the SS population. A 3-year retrospective chart review of patients from the Division of Rheumatology at University Hospital, University of Utah Health Care was undertaken in the fall of 2012 using diagnosis codes associated with this disease. Medical records were reviewed to verify diagnosis. Criteria for SS diagnosis included clinical presentation of sicca symptoms, positive SS-A or SS-B antibodies, and/or positive minor salivary gland findings from lip biopsy (Chambers, 2004; Kassan & Moutsopoulos, 2004).

Consecutive patients with SS diagnosis were identified and invited to participate until at least 100 participants were enrolled. Inclusion criteria were the diagnosis of SS, age 18 or older, English-speaking, adequate hearing for purposes of the telephone interview, and no known or reported cognitive deficits. Primary SS was the target population; however, when 100 participants with primary SS could not be identified, secondary SS was also included. It should be noted that autoimmune diseases often coexist. Because one of the purposes of this study was to sample the range and variety of voice and swallowing symptoms experienced by individuals with SS, individuals were not excluded on the basis of other health conditions, diagnoses, or diseases. The chart review identified 160 eligible participants who were subsequently invited to participate. Of these, 59 potential participants declined; therefore, the final participant group included

101 individuals with SS, or 63% of eligible participants.

Participant recruitment procedures included written correspondence and a subsequent telephone call. Individuals received a letter indicating that they would receive a call regarding possible study participation and that they might opt out of the call by contacting the principal investigator by phone or in writing. The envelope included two copies of the consent form and a postage-paid return envelope. Within a 2-week period, individuals received a telephone call by a co-investigator, providing details about the study and inviting them to participate. If the individual was interested in participating, the consent form was reviewed via telephone, and a signed copy was returned to the investigators. A time for telephone administration of the survey was scheduled, allowing time for return of the consent form. Once the signed consent form was received, the survey was administered via telephone. The participant was instructed that she or he could choose to withdraw from the study and cancel the telephone survey at any time. Interviews were conducted over a 6-month period between fall of 2012 and winter of 2013 by graduate and undergraduate students in the Department of Communication Sciences and Disorders at The University of Utah. This study was approved by the Institutional Review Board at the University of Utah (IRB#00058438).

Data Collection

The survey instrument used in the present investigation included a comprehensive, 45-item survey involving medical, lifestyle, psychosocial, and occupational factors, voice and swallowing symptoms, and individual disease severity scales. For the purpose of this study, we considered a voice disorder to be "any time the

voice does not work, perform, or sound as it normally should, so that it interferes with communication" (Roy et al., 2005, p. 1989). This definition of a voice disorder was previously used in the largest National Institutes of Health epidemiology study of voice disorders in the U.S. (R01 DC02285 Elaine Smith, Principal Investigator, National Institute on Deafness and Other Communication Disorders). Similarly, a swallowing disorder was considered to be "any time an individual experiences difficulty moving food or liquid from mouth to stomach, or experiences choking or throat clearing during or following mealtime" (Roy, Stemple, Merrill, & Thomas, 2007b, p. 859). This is the conventional definition of a swallowing disorder employed in applied research and in contemporary texts dedicated to the topic of dysphagia. Thus, the survey probed the domains of both voice and swallowing history to identify symptoms and signs, practices and patterns, and relate these findings to medical, psychosocial, occupational, and social and lifestyle factors. The survey required approximately 75 minutes to complete. The survey consisted of six primary sections detailed below.

Medical History

Voice production and swallowing function are influenced by a variety of medical conditions. We selected 21 disorders with the potential to influence the structures and/or function of voice and swallowing. We considered the presence or absence of these conditions in the participant's lifetime and, if appropriate, mechanisms of medical treatment. The inclusion of the 21 disorders was based upon the examination of previously employed surveys, a review of current literature (Roy et al., 2005; Roy, Merrill, Thibeault, Gray, et al., 2004; Roy, Merrill, Thibeault, Parsa, et al., 2004), and the

clinical experience of the investigators. This section also included more detailed questions regarding upper respiratory conditions and the use of medications.

Psychosocial History

Psychosocial factors may contribute to and be affected by voice and swallowing disorders (Roy, Stemple, Merrill, & Thomas, 2007a; Roy et al., 2007b). This section of the survey probed two main areas within the psychosocial domain, including psychosocial risk factors, such as gender and activity level, as well as the psychosocial impact of voice and swallowing disorders. Three standardized batteries, the Voice-Related Quality of Life (V-RQOL; Hogikyan & Sethuraman, 1999), the M.D. Anderson Dysphagia Inventory (MDADI; Chen et al., 2001), and the Short Form 36 Health Survey (SF-36; Ware, Gandek, & International Quality of Life Assessment Project Group, 1994) were included to comprehensively examine the impact of voice and swallowing disorders and general health on this population. The MDADI is a psychometrically validated, dysphagia-specific instrument that assesses psychosocial aspects of dysphagia. The V-RQOL is a validated, reliable, 10-item instrument that has been used to determine the impact of voice impairment on quality of life and as an outcome measure in patients with voice disorders. The SF-36 is a psychometrically validated survey. The SF-36 was developed to assess eight different measures of functional health and well-being and psychometrically based physical and mental health; that is, four scales reflect different dimensions of physical health and four scales reflect different dimensions of mental health. The SF-36 is a generic tool in that it does not target a specific age, disease, or treatment group. It has been shown to effectively survey general and specific

populations, comparing the relative burden of eight profiles (physical functioning, bodily pain, role limitations due to physical health problems, role limitations due to emotional problems, emotional well-being, social functioning, energy/fatigue, and general health perceptions). An additional question involving how the participant felt on a daily basis was also included.

Occupational or Employment History

Research has identified a link between occupation and the risk of voice disorders (Roy et al., 2005; Roy, Merrill, Thibeault, Gray, et al., 2004; Roy, Merrill, Thibeault, Parsa, et al., 2004); however, the relationship between occupation and swallowing problems has not been documented. This survey examined aspects of the participant's occupational history in order to determine its potential role in voice symptomology. This section examined work history, job related voice use patterns, and the participant's perception of the impact of job-related voice use on voice production.

Social and Lifestyle History

Lifestyle patterns and choices may influence voice and swallowing (Roy et al., 2005; Roy et al., 2007a, 2007b). Diet may influence these processes via its impact on hydration and on levels of harmful stomach acid refluxing into the larynx and pharynx. The use of tobacco products, alcohol, and recreational drugs may also influence function by altering the hydration and integrity of tissues in the aerodigestive tract. This section probed the participant's dietary history, such as caffeine intake, use of dairy products, or the intake of spicy foods, as well as their history of using tobacco products, alcohol, and

recreational drugs.

Voice and Swallowing Disorder History

Rates of voice and swallowing disorders may be higher among patients with SS as compared with the general population (Doig et al., 1971; Roy et al., 2005; Ruiz Allec et al., 2011; Sanz et al., 2011). However, a comprehensive examination of the etiology, onset, course, presentation, and treatment of these conditions in patients with SS has yet to be undertaken. This survey examined details of the participant's current and/or previous voice and swallowing concerns with the intention of defining correlates of voice and swallowing concerns, patterns of change across the lifespan, typical disease-related features, rates of treatment seeking, and outcomes.

Disease Severity

Numerous items in the survey rated the severity of specific symptoms, general health, and the autoimmune condition. Along with these items included in the main body of the survey, severity appendices were administered specific to the autoimmune diagnoses. Severity scales specific to SS included Sicca Symptoms Inventory-Short Form (SSI; Bowman et al., 2003) and European League Against Rheumatism (EULAR) Sjögren's Syndrome Patient Reported Index (ESSPRI; Seror et al., 2011). The SSI is a 10-item, psychometrically validated, dryness-specific instrument that is found to distinguish patients with SS from other rheumatic diseases. The ESSPRI is a three-item, construct validated instrument designed to measure symptoms of primary SS and is intended for clinical research. Additional severity scales were included to quantify the

potential effects of other comorbid autoimmune conditions in the study population. Severity scales for these comorbid autoimmune conditions included the Rheumatoid Arthritis Pain Scale (RAPS; Anderson, 2001), the Audit of Diabetes-Dependent Quality of Life, version 19 (ADDQOL19; Bradley et al., 1999), the Lupus Quality of Life (Lupus QOL; McElhone et al., 2007), and the World Health Organization Disability Assessment (Ustun et al., 2010). These scores were compared with the voice and swallowing severity scales to determine relationships among symptoms and comorbid conditions.

Thus, the survey, as constructed, facilitated understanding of the prevalence of voice and swallowing disorders in SS. Further, it permitted analysis of interactions among the above domains, including the delineation of risk factors, prognostic indicators, treatment outcomes, and functional implications. Consistency and accuracy of survey instrument administration was ensured using three primary methodologies. First, all research assistants received individual training and instruction on survey administration. They were permitted to ask questions and take notes regarding administration and response to participant questions. Second, each research assistant was required to practice administering the survey to nonparticipants prior to interviewing participants. Third, research assistants were audited periodically via survey instrument data entry and through bimonthly research team meetings to ensure accuracy of survey administration. Interrater reliability of the accuracy of data entry was calculated on 10% of the surveys and revealed 99.997% agreement, which confirmed excellent reliability of data entry.

Statistical Analyses

Data analysis procedures included those used in other epidemiology studies conducted by this group (Roy et al., 2007a, 2007b; Tanner et al., 2011). Medical, familial, environmental, and social history data were examined using contingency tables, summary statistics, chi-square tests (χ^2), and risk ratios (RRs). In the following section, data analysis procedures are described in detail.

The prevalence and severity of voice and swallowing problems in the SS population were evaluated using descriptive statistics. Variables were examined using contingency tables and multiple logistic regression. Bivariate analyses of association were evaluated for statistical significance using the χ^2, the Mantel-Haenszel Chi-square (MH χ^2), and the Cochran-Mantel-Haenszel Statistic (CMH). The χ^2 is used to test for independence between variables, the MH χ^2 is used to test for differences in trends between variables, and the CMH is used to test for independence after adjusting for potential confounding factors. Tests that beta coefficients in the logistic regression models equal zero were based on the Wald test, which follows an approximate χ^2 distribution.

To analyze risk factors for and impact of voice and swallowing disorders in SS, cross-tabulations were used to perform bivariate analyses between selected variables, with statistical significance based on the χ^2. The *t* statistic was used for testing the null hypothesis of equality of means between groups (i.e., those with voice and swallowing disorders vs. those without), with the *t* statistic computed using approximate degrees of freedom from Satterthwaite's approximation when the variances between the two groups are unequal. In addition, unadjusted and adjusted (controlling for the presence of other

variables) RRs were estimated using logistic regression to establish specific risk factors for reporting voice and swallowing disorders. The association between history of a voice or swallowing disorder and presence of selected risk factors (e.g., employment classification, work-related voice use issues, history of gastroesophageal reflux disease, frequent colds or sinus infections, chemical exposures, and many other disease entities) were assessed for homogeneity across the levels of age and sex using the Breslow–Day test for homogeneity. Multiple logistic regression modeling was employed, with interaction terms among significant main effects evaluated in the model.

RRs can range from 0 to infinity. The RR is statistically significantly different than 1.0 at the 0.05 level of significance if the 95% CI does not include 1.0. A RR of less than 1 indicates a negative association, a RR of 1 indicates no association, and a RR of more than 1 indicates a positive association between two variables. If both the lower and upper limits of the CI are less than 1, there is a significant negative association, whereas if both the lower and upper limits of the CI are greater than 1, there is a significant positive association. RRs may be interpreted literally and describe the extent to which the likelihood of possessing a particular risk factor (i.e., a specific health condition) increases the presence of a voice disorder. For example, individuals with SS who have a voice disorder are 1.5 times as likely to possess the specified health condition versus individuals with SS who did not report voice problems.

Finally, whether the relationship between severity of voice or swallowing disorders and selected dichotomous risk factors depends on duration, frequency, sex, and age, was addressed by assessing interaction terms among the significant main effects in multiple regression models. Only variables significant at the 0.2 level, based on

backward stepwise logistic regression, were retained in the model. This conservative value was used to minimize the probability of committing a type I error. Two-sided tests of significance were based on the 0.05 level against a null hypothesis of no association. Analyses were performed using SAS version 9.1 (SAS Institute Inc., Cary, NC, USA, 2003). Procedure statements used in SAS for assessing the data included PROC FREQ, PROC GLM, PROC FACTOR, PROC TTEST, and PROC LOGISTIC.

Results

Demographic Characteristics, Autoimmune Comorbidities, and Voice Disorders

Participants with SS ranged in age from 20.5 to 93.4, with mean age 59.4 (*SD* = 14.1). A description of the 101 participants is shown in Table 2. Most were women, non-Hispanic Whites, had a current household yearly gross income of $60,000 or more, and had at least some college education.

Years with SS ranged from 1 to 54 (*M* = 10.5, *SD* = 9.9). The number of participants currently taking medication for the syndrome was 76 (75.2%), ranging in years from 1 to 53 (*M* = 9.4, *SD* = 9.8). Years with the syndrome was only associated with ethnicity (*M* = 12.4, *SD* = 11.1 for Hispanics vs. *M* = 10.3, *SD* = 9.8 for non-Hispanics, *p* = .005). Use and duration of medication was not significantly associated with age, sex, race, ethnicity, income, or education.

Two severity measures of SS were derived (SSI, with a possible range from 0 [least severe] to 28 [most severe], and ESSPRI, with a possible range of 0 [least severe] to 10 [most severe]). Participant SSI scores ranged from 0 to 26 (*M* = 14.3, *SD* = 5.3) and ESSPRI scores ranged from 1 to 9.7 (*M* = 5.9, *SD* = 2.0). Severity scores did not significantly differ across the levels of age, sex, race, ethnicity, income, or education. Nor did they significantly differ by the years with SS, whether medication was currently

Table 2. Description of participants

	n	%
Age		
20–39	13	12.9
40–59	9	8.9
60–69	24	23.8
70–79	33	32.7
80+	22	21.8
Sex		
Men	7	6.9
Women	94	93.1
Ethnicity/Race		
NH–White	89	88.1
NH–Asian	1	1.0
NH–Native American/Alaska Native	3	3.0
Hispanic	8	7.9
Household Yearly Gross Income		
< $20,000	18	19.8
$20,000–$39,999	19	20.9
$40,000–$59,999	17	18.7
$60,000+	37	40.7
Missing	10	
Education		
< High School	5	5.0
High School	16	15.8
Some College	36	35.6
College Bachelors Degree	26	25.7
Graduate Training/Degree	18	17.8

being used, or the years of medication use.

A new variable was created that is the sum of the frequency of additional comorbid autoimmune conditions including Rheumatoid Arthritis (RA), Type 1 Diabetes, Wegener's Granulomatosis, Scleroderma (Scl), Dermatomyositis, Polymyositis, Mixed Connective Tissue Disease (MCT), and Systemic Lupus (SLE). Among participants, 56 (55.45%) were SS only, and 44 (45.6%) were SS with other comorbid autoimmune conditions (SS+). The most common other combination was SS and RA (26, 25.7%), though several other combinations were present in fewer participants (see Table 3). Those with SS only compared with SS+ were not significantly more likely to have a current voice disorder; that is, RR = 1.12 (95% CI [0.81, 1.57]). SS only versus SS+ did not significantly differ by age, sex, race, ethnicity, education, or whether they live alone.

Table 3. Distribution of autoimmune conditions in the cohort

	n	%
SS only	56	55.45
SS+RA	26	25.74
SS+SLE	5	4.95
SS+MCT	4	3.96
SS+RA+SLE	3	2.97
SS+Scl+SLE	1	.99
SS+RA+Scl	1	.99
SS+RA+MCT	1	.99
SS+Scl	1	.99
SS+SLE+MCT	1	.99
SS+RA+Scl+SLE	1	.99
SS+RA+Scl+MCT	1	.99

However, this variable was significantly associated with yearly gross income and religion. Income was only significantly associated with occurrence of SS versus SS+ in the < $20,000 category ($n$ = 6 in SS vs. n = 12 in SS+, p = .02). Religion was only significant in 2 participants of Protestant faith with SS versus 0 with SS+ (p = .037).

Prevalence of Voice Disorders

The majority of participants (n = 60, 59.4%) classified themselves as having a current voice disorder. The prevalence of current voice disorders did not significantly differ across the levels of age, sex, race/ethnicity, income, or education. In most participants their voice disorder tended to begin gradually (88.3% vs. 11.7%) and lasted for more than 4 weeks (83.3% vs. 16.7%). For those with a voice disorder, 4.0% first noticed the disorder within the past year, 33.3% 1 to 3 years earlier, 31.4% 4 to 9 years earlier, and 31.4% 10 or more years earlier. The severity of SS was significantly associated with having a voice disorder based on mean ESSPRI scores (6.3 [SD = 1.8] vs. 5.4 [2.1], p = .020) and approached significance based upon mean SSI scores (15.1 [SD =

4.8] vs. 13.1 [5.7], $p = .061$).

Voice Symptoms

Levels of selected voice-related symptoms were identified in our participant population (Table 4). Participants were asked to quantify the frequency of selected voice symptoms, where "daily" = 1, "weekly" = 2, "monthly" = 3, "several times a year" = 4, and "yearly or less" = 5. The mean frequency of the selected voice symptoms ranged from 1.3 to 3.0 ($M = 2.4$, $SD = 0.5$). All but two of the voice-related symptoms were significantly associated with the participants who indicated that they had a voice disorder (see Table 4). Regression analysis showed that the SSI severity score was significantly associated with chronic throat dryness (Partial $r^2 = 7.3\%$, $p = .006$), a monotone voice ($r^2 = 5.7\%$, $p = .011$), and a wobbly or shaky voice ($r^2 = 4.3\%$, $p = .031$). The ESSPRI severity score was only simultaneously significantly associated with chronic throat

Table 4. Levels of selected voice-related symptoms and presence of a current voice disorder

	Current	Frequency*	Voice Disorder		χ^2
Symptom	**%**	***M* (*SD*)**	***n***	**%**	***p***
Chronic dryness in your throat	**80.2**	**1.3 (0.8)**	**54**	**66.7**	**.003**
Frequently clear your throat	**60.4**	**1.8 (1.2)**	**42**	**68.8**	**.017**
Hoarseness	**51.5**	**3.0 (1.5)**	**44**	**84.6**	**< .0001**
A loss of singing range	**50.5**	**2.1 (1.2)**	**39**	**76.5**	**.0004**
Voice tires or changes quality after short use	**42.6**	**2.3 (1.3)**	**37**	**86.1**	**< .0001**
Difficulty projecting your voice	**41.6**	**2.3 (1.5)**	**31**	**73.8**	**.013**
An effort to talk	**40.6**	**2.8 (1.5)**	**35**	**85.4**	**< .0001**
Trouble speaking or singing softly	**38.6**	**2.4 (1.4)**	**35**	**89.7**	**< .0001**
Discomfort while using your voice	**35.6**	**2.3 (1.5)**	**31**	**86.1**	**< .0001**
A bitter or acid taste	30.7	2.8 (1.3)	20	64.5	.487
Chronic soreness in your throat	**26.7**	**2.1 (1.1)**	**21**	**77.8**	**.023**
Wobbly or shaky voice	**23.8**	**2.5 (1.3)**	**21**	**87.5**	**.001**
An "airy" or "breathy" voice	**16.8**	**3.0 (1.2)**	**15**	**88.2**	**.008**
A monotone voice (monopitch)	**13.9**	**2.3 (1.3)**	**12**	**92.3**	**.010**
Wet, gurgley voice quality	6.9	3.0 (1.7)	5	71.4	.502

*1 = Daily, 2 = Weekly, 3 = Monthly, 4 = Several times per year, 5 = Yearly or less
Note. Only *p* values significant at the < .05 level are bolded.

soreness (7.1%, $p = .007$).

Voice Use Patterns, General Health, Lifestyle, and Voice Disorders

To assess the impact of participants' voice use patterns, general health, and lifestyle on experiencing a current voice disorder, they were asked questions related to each of these areas. RRs for the significant variables were also determined in order to report how much more likely someone who possesses the respective attribute or condition is to report a voice disorder versus someone who does not have the attribute or condition. Questions regarding voice use patterns were asked to establish the frequency of specific vocal behaviors during the day, vocally demanding jobs, and any job-related voice disorders. Participants were asked general health questions to establish the history of specific medical conditions and coinciding medication use, history of environmental inhalants, bodily tension, and family history of voice and swallowing problems. Lifestyle questions were asked regarding use of tobacco, alcohol, and drugs, as well as exercise patterns and personality descriptors. Table 5 lists each condition addressed, the number of participants with each condition who also reported a current voice disorder, the resulting significance patterns, and RRs. The self-reported factors associated with significantly elevated RRs, from greatest to least, include bronchitis (RR = 1.88), jaw tension (RR = 1.7), abdomen tension (RR = 1.67), current exposure to secondary tobacco smoke (RR = 1.64), vocally demanding occupation (RR = 1.60), stomach or duodenal ulcer (RR = 1.58), job-related voice disorder (RR = 1.58), often/constant neck/throat tension (RR = 1.55), asthma (RR = 1.53), family history of any swallowing problems (RR = 1.50), pneumonia (RR = 1.49), hearing loss (RR = 1.4), and past exposure to

Table 5. Current voice disorder and selected conditions

Medical Condition	Current Voice Disorder *n*	%	χ^2 *p*	RR [95% CI]
Arthritis	35	58.3	.578	
Heart disease	9	81.8	.187	
Hypertension	26	57.8	.765	
Circulatory problems	24	70.6	.169	
Kidney problems	15	75.0	.113	
Thyroid problems	29	64.4	.428	
Stomach or duodenal ulcer	**24**	**80.0**	**.006**	**1.58 [1.18, 2.11]**
Esophageal reflux	40	64.5	.187	
Stroke	8	66.7	.600	
Respiratory allergies	23	63.9	.745	
Pneumonia	**39**	**69.6**	**.041**	**1.49 [1.04, 2.13]**
Emphysema	2	50.0	.696	
COPD	2	100.0	.483	
Hearing loss	**23**	**74.2**	**.044**	**1.40 [1.04, 1.90]**
Bronchitis	**43**	**74.1**	**.002**	**1.88 [1.26, 2.80]**
Asthma	**18**	**81.8**	**.016**	**1.53 [1.16, 2.05]**
Severe neck, back, or head injury	22	66.7	.301	
Chronic pain	43	64.2	.170	
Cancer	15	68.2	.343	
Depression or anxiety	37	61.7	.576	
Sleep disorder	19	73.1	.167	
Stopped menstrual periods	45	60.8	.280	
Swallowing				
Current swallowing problem	41	63.1	.313	
Swallowing problem began with RX reaction	1			
Swallowing problem began with illness	8	50.0	.159	
Swallowing problem began after surgery	5	100.0	.076	
Past swallowing problem	12	52.2	.422	
Family history of any type of swallowing problem	**14**	**82.4**	**.035**	**1.50 [1.12, 2.02]**
Medications for the following conditions				
Bronchitis	0			
Asthma	12	75.0	.176	
Severe neck, back, or head injury	6	75.0	.566	
Chronic pain	31	68.9	.250	
Cancer	2	50.0	.383	
Depression or anxiety	23	60.5	.811	

Table 5. Continued

Medical Condition	Current Voice Disorder *n*	%	χ^2 *p*	RR [95% CI]
Sleep disorder	10	66.7	.390	
Condition				
Colds (3+ vs. < 3 year)	45	60.0	.836	
Sinus infections (3+ vs. < 3 year)	41	56.2	.284	
Sore throat (3+ vs. < 3 year)	38	54.3	.115	
Postnasal drip (Chronically vs. less)	23	60.5	.859	
Head and neck surgery	28	60.9	.870	
Breathing assistance	11	73.3	.234	
Ever used tobacco products	15	65.2	.518	
At any year in your life, drank an average of one or more alcoholic beverages a week.	23	62.2	.622	
Ever used recreational drugs	7	71.4	.502	
Experience tension				
Neck/Throat (often/constantly vs. otherwise)	**35**	**72.9**	**.009**	**1.55 [1.11, 2.16]**
Jaw	**20**	**87.0**	**.002**	**1.70 [1.30, 2.22]**
Shoulders	29	67.4	.157	
Abdomen	**11**	**91.7**	**.016**	**1.67 [1.29, 2.15]**
Family history of any type of voice problem	7	87.5	.092	
Voice activity				
Talk (often/constantly vs. otherwise)	47	58.0	.569	
Talk quietly	30	68.2	.115	
Whisper	3	50.0	.629	
Talk loudly	20	55.6	.558	
Sing	7	53.8	.662	
Shout, yell or cheer	1	25.0	.153	
Clear your throat	26	68.4	.152	
Laugh	35	59.3	.984	
Cough	15	60.0	.944	
Ever in an occupation requiring you to talk a lot on a daily basis	**52**	**64.2**	**.048**	**1.60 [0.92, 2.81]**
Experience voice disorders with this job	**28**	**77.8**	**.023**	**1.58 [1.17, 2.14]**
Exposures				
Excess dust – current	7	41.2	.093	
Excess dust – past	**31**	**70.4**	**.047**	**1.38 [1.01, 1.90]**
Fumes from cleaning products – current	16	55.2	.582	
Fumes from cleaning products – past	28	69.9	.784	
Secondary tobacco smoke – current	**10**	**90.9**	**.024**	**1.64 [1.26, 2.13]**

Table 5. Continued

Medical Condition	Current Voice Disorder *n*	%	χ^2 *p*	RR [95% CI]
Secondary tobacco smoke – past	39	65.0	.166	
Dry air – current	53	62.4	.165	
Dry air – past	55	63.2	.052	
Exercise (yes vs. no)	54	60.0	.728	
Personal Description				
Quiet vs. talkative	23	57.5	.803	
Easy-going vs. worrier	30	55.6	.398	
Active vs. inactive	46	56.1	.160	
Happy vs. sad	54	58.7	.642	

Note. Only RRs significant at the < .05 level are bolded.

excess dust (RR = 1.38).

Voice Disorders and Quality of Life

To assess the functional impact of a voice disorder on the SS population, several quality of life measures were included in the survey. These measures were associated with current voice disorders, current voice symptoms, and SS severity. The VRQOL, a measure of the impact of having a voice disorder on quality of life, ranged from 17 (poorest quality of life) to 100 (best quality of life), with mean 83.2 (*SD* = 18.3). VRQOL was not significantly associated with age, sex, race, ethnicity, income, or education, but was significantly related to whether the participant indicated having a voice disorder (75.7 [*SD* = 19.7] vs. 94.2 [*SD* = 7.8], $p < .0001$). Mean VRQOL was significantly lower for all of the selected voice-related symptoms except the same 2 items identified in Table 4 (see Table 6). Regression analysis showed that the VRQOL measure was significantly associated with the SSI severity score (Partial r^2 = 6.9%, p = .008) and the ESSPRI severity score (7.2%, p = .007).

To assess the relationship between daily, overall health and the potential effects on the voice, SS severity, and quality of life, an additional single-question measure was used. This variable about "how you feel, overall, on a daily basis" was significantly associated with chronic throat soreness and frequent throat clearing. Of those with excellent/good health, 19.7% had chronic throat soreness compared with 37.5% for those with fair/poor health (χ^2 p = .048). Of those with excellent/good health, 50.8% had frequent throat clearing compared with 75.0% for those with fair/poor health (χ^2 p = .015). Along with specific voice symptoms, "how you feel..." was compared with

Table 6. Voice symptoms and relation with VRQOL scores

Symptom	VRQOL Score		
	No Current Symptom *M* (*SD*)	Current Symptom *M* (*SD*)	*t* statistic *p* value
Chronic dryness in your throat	**91.4 (11.9)**	**81.2 (19.1)**	**.004**
Frequently clear your throat	**89.3 (13.8)**	**79.2 (19.9)**	**.003**
Hoarseness	**89.3 (13.8)**	**77.4 (20.2)**	**.001**
A loss of singing range	**89.5 (15.0)**	**77.0 (19.2)**	**.0004**
Voice tires or changes quality after short use	**89.3 (15.6)**	**74.9 (18.7)**	**< .0001**
Difficulty projecting your voice	**91.0 (11.3)**	**72.3 (20.7)**	**< .0001**
An effort to talk	**92.2 (9.1)**	**70.0 (20.5)**	**< .0001**
Trouble speaking or singing softly	**90.2 (11.5)**	**72.1 (21.5)**	**< .0001**
Discomfort while using your voice	**90.1 (12.4)**	**70.7 (20.8)**	**< .0001**
A bitter or acid taste	84.1 (17.7)	81.2 (19.8)	.462
Chronic soreness in your throat	**87.4 (14.4)**	**71.5 (22.8)**	**.002**
Wobbly or shaky voice	**88.6 (12.7)**	**65.9 (22.7)**	**< .0001**
An "airy" or "breathy" voice	**85.9 (16.4)**	**69.9 (22.0)**	**.001**
A monotone voice (monopitch)	**85.8 (16.3)**	**65.5 (22.3)**	**.0001**
Wet, gurgley voice quality	83.1 (18.8)	83.7 (10.9)	.938

Note. Only *p* values significant at the < .05 level are bolded.

reporting a voice disorder, VRQOL, SSI, and ESSPRI. As reported in Table 7, reductions in reported overall health were significantly associated with the presence of a voice disorder, as well as reductions in voice-related quality life, and worsening disease severity.

The general quality of life measure, SF-36, and its subscales were compared with VRQOL, current voice disorder, and current voice symptoms. The two summary measures of the SF-36 are physical health and mental health. Each summary measure is associated with four subscales (physical health: physical function, role-physical, bodily pain, general health; mental health: vitality, social functioning, role-emotional, mental health). The raw scores from the survey are coded into a 0 to 100 scale, with 100 representing the highest state of health. Average scores for each of the scale items across all participants have been reported previously, ranging from the 70s for general health, bodily pain, and energy/fatigue, to the 80s for social functioning, physical functioning,

Table 7. "How you feel on a daily basis" and relation with presence of a current voice disorder, as well as VRQOL, SSI, and ESSPRI scores

		Voice Disorder		VRQOL	SSI	ESSPRI
How feel on a daily basis	***n***	**Yes %**	**No %**	***M* (*SD*)**	***M* (*SD*)**	***M* (*SD*)**
Excellent	10	3.4	19.5	93.5 (7.6)	11.0 (3.3)	2.9 (1.1)
Good	51	53.3	46.3	85.4 (18.9)	13.6 (5.5)	5.5 (1.5)
Fair	33	33.3	31.7	80.7 (16.4)	15.4 (5.0)	7.0 (1.6)
Poor	7	10.0	2.5	64.4 (20.5)	18.0 (4.4)	8.2 (1.2)
			χ^2 *p* = .034	***F p* = .007**	***F p* = .019**	***F p* < .0001**

role limitations – physical health, and emotional well-being, and to the 90s for role limitations – emotional stresses. The highest possible score for physical functioning is obtained by performing all types of physical activity including the most vigorous without limitations due to health; for role-physical is obtained by having no problems with work or other daily activities; for bodily pain is obtained by having no pain or limitations due to pain; for general health is obtained by evaluating personal health as excellent; for energy/fatigue is obtained by feeling full of pep and energy all the time; for social functioning is obtained by performing normal social activities without interference due to physical or emotional problems; for role – emotional is obtained by having no problems with work or other daily activities; and for mental health is obtained by feeling peaceful, happy, and calm all of the time. Both SF-36 summary measures and six of its eight subscales were significantly correlated with the VRQOL (see Table 8). This association seems to disproportionately contribute to reductions in the mental health, rather than the physical health, summary measure. Table 9 identifies the extent that selected voice symptoms influenced specific quality of life domains as assessed by the SF-36. Specific voice symptoms were statistically correlated with subscales, namely frequent throat

Table 8. Mean SF-36 subscales and correlations with VRQOL scores

	M	*SD*	Correlation Coefficient (r) with VRQOL	Correlation Coefficient *p* value
PHYSICAL Health	**49.9**	**23.8**	**.23**	**.018**
Physical Functioning	60.3	29.6	.18	.066
Role-Physical	50.0	43.3	.14	.167
Bodily Pain	**53.6**	**25.4**	**.26**	**.009**
General Health	**37.4**	**21.0**	**.29**	**.003**
MENTAL Health	**66.9**	**18.4**	**.35**	**.0003**
Vitality	**43.8**	**23.8**	**.25**	**.012**
Social Functioning	**70.2**	**30.5**	**.35**	**.0004**
Role-Emotional	**81.8**	**31.1**	**.27**	**.006**
Mental Health	**75.0**	**16.7**	**.24**	**.014**

Note. Only *p* values significant at the < .05 level are bolded.

clearing, chronic throat soreness, and difficulty projecting voice. These symptoms seemed to disproportionately contribute to reductions in the physical health summary measure aspect of quality of life. For the model with summary physical health as the dependent variable, only frequent throat clearing was significant (slope = -16.2, *SE* = 4.6, *p* = .001). For the model with summary mental health as the dependent variable, only discomfort while using voice was significant (slope = -13.7, *SE* = 3.6, *p* = .0002). Reporting a voice disorder was not significantly associated with any of the eight subscales or the physical or mental health summary measures.

Of the 60 participants who reported a current voice disorder, only 16 (15.8%) had ever sought professional help to improve their voice. Of the 16, six saw a singing or acting teacher or coach, five saw a physician, three saw a speech-language pathologist, and two saw both a physician and speech-language pathologist. Almost all indicated that it helped their voice (13/16 = 81.2%). Seeking professional help to improve their voice was not significantly associated with age, sex, race, ethnicity, income, or education.

Table 9. Mean SF-36 subscale scores according to current voice symptoms

		PF		RP		BP		GH		VT		SF		RE		MH	
	n	**Slope**	***SE***	***M***	***SD***	***M***	***SD***	***M***	***SD***	***M***	***SD***	***M***	***SD***	***M***	***SD***	***M***	***SD***
Hoarseness	52																
Voice tires	43																
Trouble speaking	39																
Difficulty projecting	**42**					**-10.2**	**5.0**										
Loss of singing range	51																
Discomfort	**36**			**-18.3**	**8.9**							**-17.3**	**6.1**	**-22.2**	**6.1**	**-12.6**	**3.3**
Monotone	13																
Effort	41																
Chronic dryness	81																
Gurgley voice	7																
Chronic soreness	**27**					**-12.7**	**5.5**	**-9.7**	**4.6**	**-13.5**	**5.2**						
Frequent throat clear	**61**	**-19.9**	**5.7**					**-10.9**	**4.1**								
Bitter or acid taste	31																
Wobbly/shaky	24																
Airy/breathy	17																

Note. Only significant slope estimates appear in the table, $p < .05$.
Physical Health: Physical Function (PF), Role-Physical (RP), Bodily Pain (BP), General Health (GH)
Mental Health: Vitality (VT), social functioning (SF), Role-Emotional (RE), Mental Health (MH)

Discussion

Epidemiologic studies of voice disorders in SS are relatively rare and report varying results. The current investigation reports findings from the largest epidemiology study of voice disorders undertaken in SS. Important results are discussed below.

The majority (59%) of participants with SS reported a current voice disorder. Most of these voice disorders began gradually (88%), were chronic, lasting more than four weeks (83%), and in many cases were longstanding (i.e., 63% persisted 4+ years). The likelihood of reporting a current voice disorder was associated with increasing disease severity of SS, but interestingly was not associated with the age or sex of the participants. Furthermore, additional comorbid autoimmune conditions did not appear to increase the likelihood of reporting voice disorders. These results confirm that chronic, longstanding voice disorders are common in SS and appear to be related to worsening disease severity. Differences in prevalence rates were not observed between primary SS, secondary SS, or when the disease coexisted with other autoimmune conditions.

Patients with SS reported numerous voice symptoms, which often occurred on a daily and/or weekly basis. With the exception of bitter/acid taste and a wet/gurgley voice quality, all of the voice symptoms were significantly associated with reporting a current voice disorder. Four symptoms were reported by more than half of all participants: chronic throat dryness (80%), frequent throat clearing (60%), hoarseness (52%), and loss of singing range (51%). Furthermore, it appears that certain voice symptoms, such as chronic throat dryness and chronic throat soreness, are particularly meaningful for individuals with SS and accounted for a significant amount of variance in overall SS severity scores. This suggests that symptoms especially involving physical sensations

within the laryngopharyngeal region have particular salience for individuals with SS.

This study also examined associated medical conditions and risk factors that potentially contributed to voice disorder reporting. Certain medical conditions and lifestyle or occupational factors increased vulnerability for experiencing a voice disorder. Based upon inspection of the RR data, several interesting factors emerged. Vocally demanding occupations and job-related voice disorders were among the significant risk factors. This association seems obvious since extensive vocal use may logically contribute to voice disorders. Other risk factors were associated with inhaled irritants, such as current exposure to secondary tobacco smoke and past exposure to excess dust. These inhalants expose the laryngeal structures to potential damage and, therefore, may contribute to structural or compensatory voice problems. Several of the risk factors which are associated with decreased breathing efficiency—bronchitis, asthma, and pneumonia—may impact the respiratory pressure required for adequate voicing and may in turn effect ability for efficient voicing. Several other risk factors statistically associated with reporting a voice disorder included jaw tension, neck/throat tension, and hearing loss. These risk factors seem to have anatomical and/or acoustic connections to the voice. Tension in areas in close proximity to laryngeal structures may contribute to, or be a result of, a current voice disorder. Additionally, hearing loss may effect one's perception and judgment of his or her own voice quality and abilities.

Effects of Voice Symptoms and Disorders on Quality of Life in SS

Our cohort reported high rates of undesirable voice symptoms and related voice disorders, and this high frequency of voice-related problems may help to explain some of

the untoward effects on quality of life reported by the participants. Inspection of the VRQOL results revealed that voice disorders associated with SS produced mild to moderate reductions in voice-related quality of life. A mean VRQOL score of 83 for our group was substantially lower than both the highest possible score of 100 and the non-voice patient population average score of 98 (Hogikyan & Sethuraman, 1999). However, this mean score is in the mild to moderate range of voice-related handicap. Lower VRQOL scores were associated with increasing severity of SS. Our finding that severity of SS was not correlated with years with SS, but was correlated with lower VRQOL, is important. This indicates that a person with SS can experience lower voice-related quality of life at any time point along their disease process.

The SF-36 general health quality of life measure also revealed important findings. Participants in the current study reported a lower overall quality of life than the normative data, whether or not they had a voice disorder. Comparisons between the SF-36 physical health and mental health summary measures and the VRQOL were statistically significant ($p = .018$ and $p = .0003$, respectively). Additionally, those who reported a voice disorder also had poorer quality of life scores on all subscales of the SF-36 versus those who did not report a voice disorder. Although statistical significance was not found on the SF-36 subscales for those who reported a voice disorder, some voice symptoms were statistically significant: difficulty projecting voice, discomfort while using voice, chronic throat soreness, and frequent throat clearing. Additionally, the voice symptoms independently associated with reduced SF-36 physical and mental health subscales were frequent throat clearing and discomfort while using voice, respectively. Interestingly, the symptom that seems most obviously related to SS, chronic dryness, was not significantly

associated with subscales or summary measures of the SF-36. Dryness was correlated with the SSI, but this is unsurprising given that questions on the SSI specifically address the nature and severity of these symptoms. This discrepancy of the correlations between SF-36 and symptoms versus disorders seems to suggest that specific symptoms could be disproportionately contributing to reduced overall quality of life. Specifically, the symptoms of discomfort and chronic soreness are linked to more quality of life parameters on the SF-36. Similar to other studies in non-SS populations (Merrill et al., 2011; Merrill, Roy, & Lowe, 2013), symptoms associated with effort, discomfort, and chronic sensations in the throat seem to decrease quality of life. The negative impact of voice disorders on physical and emotional functioning seen in the current study also reflects similar observations made by Baylor, Burns, Eadie, Britton, and Yorkston (2011), who reported negative effects of communication disorders in a variety of non-SS populations, although studies by their group utilized instruments other than the SF-36 (Baylor et al., 2011; Baylor et al., 2013; Baylor, Yorkston, Eadie, Miller, & Amtmann, 2009).

An additional quality of life survey question, "how do you feel, overall, on a daily basis," was significantly correlated with reporting a voice disorder, VRQOL, and SS severity. Similar to the VRQOL and the SF-36, this question was significantly associated with specific voice symptoms, including chronic throat soreness and frequent throat clearing.

The one symptom that was significantly associated with all three quality of life measures was frequent throat clearing. It has been reported that chronic throat clearing is associated with voice disorders and may be a reaction to triggers such as the perception

of dysphonia, foreign body sensation, and increased viscosity of mucous (Bonilha, Gerlach, Sutton, Dawson, & Nietert, 2012; Cobeta, Pacheco, & Mora, 2013). Chronic throat dryness may manifest in thicker laryngeal secretions, and it may be the increased viscosity of mucous associated with SS that might instigate this frequent symptom in our participants.

Although 59% of participants reported a voice disorder, only 16% sought professional help to improve their voices. Importantly, 87% of those who received treatment indicated that their voices improved. This finding is particularly interesting given that both SS disease severity and (expectedly) VRQOL scores were worse in those who reported a voice disorder. Also interesting is the fact that help-seeking behavior was not significantly associated with age, sex, race, ethnicity, income, or education. One possible explanation for this low rate of seeking treatment may be limited patient education regarding treatment options for voice disorders. It is important to note that, while the reported severity of voice disorders in this group was generally mild, the SS severity was moderate. Additionally, SS severity predicted the presence of voice disorders. Therefore it is possible that the overall SS severity and associated symptoms eclipsed the voice symptoms, perhaps evidenced in their treatment-seeking behaviors. Regardless of the potential causes of these treatment-seeking behaviors, the results from this study have important implications for symptom identification and referral practices in this population.

Further research surrounding voice disorders in SS is warranted based on these quality of life findings. Having a voice disorder statistically impacted VRQOL and "how you feel on a daily basis." VRQOL and "how you feel…" were also correlated with the

SF-36. Multiple voice symptoms were statistically associated with all three measures. These associations demonstrate the negative impact SS and related voice disorders have upon those with this autoimmune condition. Additional research in this area to further analyze the correlations between voice disorders, symptoms, and quality of life may demonstrate significance between voice disorders and the SF-36. Perhaps more importantly, future research should investigate novel treatment options for voice disorders in this population.

Comparisons with the Extant Literature

Direct comparison of our results from the current study with the studies of SS described and critiqued in Table 1 is difficult due to differences in sample sizes, patient populations (i.e, numbers of primary SS in the rheumatic groups studied), methodologies, and definitions used to determine the presence of a voice disorder. Therefore, only general comparisons can be made. Our prevalence rate for voice disorders in SS (59%) is clearly at odds with absence of significant voice problems reported by Sanz et al. (2011) and the 42% reported by Ruiz Allec et al. (2011). It is important to keep in mind, however, that the study by Ruiz Allec et al. (2011) included only 31 SS participants, four of whom had primary SS. The study by Sanz et al. (2011) included only four total SS participants of their rheumatic diseases cohort. Methodologies in both studies were distinct in that they defined voice disorders differently. Similar to findings in the current study, Heller et al. (2014) found that dysphonia in SS typically ranges from mild to moderate. This report was based on objective and quality of life measures. Other studies focused on voice symptoms, and hoarseness seemed to be the symptom highlighted most

frequently, with a prevalence rate of 19–71% (Doig et al., 1971; Haga et al., 1997). Reports of hoarseness from the current study fall in between that range at 52%; however, among the 14 other symptoms analyzed in the current study, two other symptoms were more prevalent: chronic throat dryness (80%) and frequent throat clearing (60%). It is important to note that the SS diagnostic criteria used by Doig et al. (1971) differs greatly from that used in the current study, which followed more recent guidelines. In a recent study, Tanner et al. (2013) reported number of years with SS was associated with voice symptoms: vocal effort and throat dryness. The current study did not reveal any associations with years with SS, but with SS severity. However, the Tanner et al. (2013) study focused on hydration voice treatment, rather than epidemiology. Some studies reported a large difference between voice symptoms and signs of voice dysfunction observed during instrumentation procedures. Freeman et al. (2005) reported the difference between symptoms and observations was 60–70% versus 20%. In that study, however, only two symptoms were analyzed versus the 15 analyzed in the current study. Ruiz Allec et al. (2011) found opposite results in that the prevalence of observed voice problems was greater than reported problems (90% vs. 42% in pSS and sSS). Though the current study did not use similar objective measures, we did find a higher rate of both voice symptoms and disorders than either of these studies. Importantly, according to Sanz et al. (2011), there seems to be an 81% worsening of voice disorders during rheumatic flares. It is also important to keep in mind that symptoms of a disorder may impact quality of life more than signs of dysfunction observed objectively. When comparing risk factors presented in the extant literature, reflux seemed to be the one highlighted in other studies (Ogut et al., 2005; Ruiz Allec et al., 2011). These reports

differ from that in the current study, which did not find reflux to be a significant risk factor. The final comparison to be made is in the area of quality of life differences resulting from voice disorders in SS. Sanz et al. (2011) was the only study in Table 1 to address socioemotional effects of these disorders. They reported that voice disorders negatively impact physical and daily living parameters and reduce quality of life. This is similar to findings in the current study; however, the Sanz et al. (2011) study did not find statistical significance for voice disorders in SS, likely due to the small number of SS participants in that study.

Comparisons between the current study and several non-SS studies were made to review interesting risk factors in the current study that were not independently associated with reporting a current voice disorder. These factors include frequent colds and sinus infections and reflux, which have previously been associated with reporting a voice disorder in non-SS populations (Merrill, Anderson, & Sloan, 2011; Roy et al., 2005). Increased age has also been associated with increased prevalence of voice disorders in a non-SS population compared with the general population (29% in age 65+ vs. 6.6%, respectively; Roy et al., 2005; Roy et al., 2007a). Although the current study included an uneven distribution of participants across each age stratum, the majority of participants (78%) were age 60 or older. The geriatric population was well represented in this study, and statistical analyses did not find a correlation between increased age and increased prevalence of voice disorders. This, again, seems to show that SS severity is the true risk factor for reporting a voice disorder, irrespective of age. The finding that comorbid autoimmune conditions did not seem to be associated with voice problems is also intriguing. In a concurrent study (Wright, 2014), RA was found to have a higher

incidence of voice disorders than the general population (35% vs. 6.6%), but not as high as the incidence found in SS or SS with RA in the current study. This, again, seems to imply that SS alone may represent the greatest risk factor for voice disorders and that comorbid autoimmune conditions do not intensify these problems.

Results

Prevalence of Swallowing Disorders

The majority of participants (n = 65, 64.4%) classified themselves as having a current swallowing disorder wherein they experienced difficulty moving food or liquid from their mouth to their stomach or anytime they experienced choking or frequent throat clearing during or following mealtime. In most participants (n = 55, 84.6%) their swallowing disorder began gradually and lasted for more than 4 weeks (n = 59, 90.8%). For those with a swallowing disorder, 15% first noticed the disorder within the past year, 18.3% 1–3 years earlier, 30% 4–9 years earlier, and 36.7% 10 or more years earlier. The prevalence of a current swallowing disorder did not significantly differ across the levels of age, race/ethnicity, income, or education. However, significant difference was found for sex, in that male participants were more likely to report a swallowing disorder (100% of males vs. 61.7% of females, p = .041). Having SS+MCT was significantly associated with reporting a current swallowing disorder (χ^2, p = .041). Other comorbid autoimmune conditions showed no significant correlation. SS severity was significantly associated with having a swallowing disorder based on mean SSI scores (15.8 [SD = 5.2] vs. 11.5 [SD = 4.2], p < .0001), approximated significance based upon mean ESSPRI scores (6.2 [SD = 1.9] vs. 5.5 [SD = 2], p = .085), but was not significantly associated with the overall disease severity question (p = .111). Length of time with SS was not

significantly associated with reporting a swallowing disorder.

Swallowing Symptoms

Levels of selected swallowing-related symptoms were identified in our patient population (Table 10). Nearly all of the 101 participants reported at least one swallowing symptom (*n* = 99, 98%). Of the 19 swallowing symptoms, five were reported by more than 50% of participants: dry mouth (96%), difficulty swallowing solids (64%), food sticking in the throat (61%), need for excessive chewing (55%), and taking smaller bites in order to swallow safely (55%). According to CMH statistical analyses, 13 of the 19 swallowing symptoms were significantly associated with the participants who indicated they had a swallowing disorder (see Table 10). RRs reported in Table 10 reveal how much more likely someone is to report a swallowing disorder if they also have a specific symptom. The swallowing symptoms associated with significantly elevated RRs, from greatest to least, included food sticking in throat (RR = 3.1), difficulty swallowing solids (RR = 3.0), take smaller bites of food (RR = 2.4), take a longer time to eat (RR = 2,3), pain/pressure in the throat/chest while swallowing (RR = 2.3), need for excessive chewing (RR = 2.2), coughing/throat clearing/choking while eating (RR = 1.8), forcibly regurgitate stuck foods (RR = 1.8), avoid eating certain foods (RR = 1.8), difficulty swallowing liquids (RR = 1.7), inability to control liquid/food/saliva in the mouth (RR = 1.5), increased mucous/phlegm while eating (RR = 1.5), and difficulty swallowing medication (RR = 1.3). The length of time of having SS was significantly associated with only one swallowing symptom: sneezing while eating; those with this symptom had SS longer (*M* = 1995.1, *SD* = 15.7 vs. *M* = 2003.5, *SD* = 8.4, *p* = .022).

Table 10. Levels of selected swallowing-related symptoms and presence of a current swallowing disorder

Symptom	Current %	Frequency* Most reported	Swallow Problem *n* (%)	χ^2 *p* Value	RR [95% CI]
Take longer time to eat because of swallowing problem (*n* = 48)	**47.5**	**1**	**44 (91.67)**	**< .0001**	**2.3 [1.6, 3.3]**
Difficulty swallowing liquids (*n* = 21)	**20.8**	**2**	**20 (95.24)**	**.001**	**1.7 [1.4, 2.1]**
Difficulty swallowing solids (*n* = 65)	**64.4**	**1**	**55 (84.62)**	**< .0001**	**3.0 [1.8. 5.2]**
Difficulty swallowing medications (*n* = 47)	**46.5**	**1**	**35 (74.47)**	**.049**	**1.3 [1.0, 1.8]**
Gurgley or wet voice during or after eating (*n* = 5)	5.0	2–3	4 (80)	.456	
Coughing, throat clearing, or choking before, during or after eating (*n* = 46)	**45.5**	**1**	**39 (84.78)**	**< .0001**	**1.8 [1.3, 2.4]**
Inability to control food, liquid, or saliva in the mouth (*n* = 28)	**27.7**	**1**	**24 (85.71)**	**.006**	**1.5 [1.2, 2]**
Sneezing during or after a meal (*n* = 12)	11.9	2	9 (75)	.415	
Pain or pressure in the throat or chest during swallowing (*n* = 40)	**39.6**	**2**	**39 (97.5)**	**< .0001**	**2.3 [1.7, 3.1]**
Wheezing after eating (*n* = 6)	5.9	1	6 (100)	.061	
Food comes out of the nose while eating (*n* = 3)	3.0	4	3 (100)	.193	
Need to chew excessively in order to swallow safely (*n* = 55)	**54.5**	**1**	**47 (85.45)**	**< .0001**	**2.2 [1.5, 3.2]**
Dry mouth (*n* = 97)	96.0	1	63 (64.95)	.543	
Difficulty placing food in mouth (*n* = 3)	3.0	2	3 (100)	.193	
Sensation of food sticking in throat (*n* = 62)	**61.4**	**2**	**54 (87.1)**	**< .0001**	**3.1 [1.9, 5.1]**
Forcibly regurgitate food stuck in throat (*n* = 18)	**17.8**	**3**	**18 (100)**	**.001**	**1.8 [1.5, 2.1]**
Avoid eating certain foods because of swallowing disorder (*n* = 41)	**40.6**	**1**	**36 (87.8)**	**< .0001**	**1.8 [1.4, 2.4]**
Increased mucous or phlegm in throat before, during, or after eating (*n* = 30)	**29.7**	**1**	**25 (83.33)**	**.010**	**1.5 [1.4, 1.9]**
Take smaller bites of food to swallow safely (*n* = 55)	**54.5**	**1**	**48 (87.27)**	**< .0001**	**2.4 [1.6, 3.5]**

*1 = Daily, 2 = Weekly, 3 = Monthly, 4 = Several times a year, 5 = Yearly or less

Note. Only *p* values significant at the < .05 level are bolded.

Regression analysis showed that the SSI severity score was simultaneously significantly associated with taking smaller bites (partial r^2 = .161, p < .0001), mucous/phlegm while eating (partial r^2 = .068, p = .004), and difficulty placing food in the mouth (partial r^2 = .050, p = .011). The ESSPRI severity score was simultaneously significantly associated with mucous/phlegm while eating (partial r^2 = .088, p = .003) and wheezing while eating (partial r^2 = .056, p = .013). The overall disease severity question was only significantly associated with one symptom: taking a longer time to eat. Of those with no problem, 33.3% took a longer time to eat compared with 70.8% for those with a severe problem (MH χ^2 p = .010). Participants were asked to quantify the frequency of selected swallowing symptoms, where "daily" = 1, "weekly" = 2, "monthly" = 3, "several times a year" = 4, and "yearly or less" = 5 (Table 10). The majority of the selected swallowing symptoms occurred daily in most participants (11 of 19 [57.9%] daily, 6 [31.6%] weekly, 2 [10.5%] monthly, 1 [5.3%] several times a year). Increased frequency of one symptom was significantly associated with length of time of having SS: pain in the throat/chest while swallowing (p = .044). SSI severity scores were significantly associated with increased frequency of difficulty swallowing solids (p = .011) and taking smaller bites in order to swallow safely (p = .004). ESSPRI severity scores were significantly associated with increased frequency of coughing while eating (p = .031). The question regarding overall disease severity was significantly associated with increased frequency of food getting stuck in the throat (p = .004) and taking smaller bites in order to swallow safely (p = 0.002).

Voice Use Patterns, General Health, Lifestyle, and Swallowing Disorders/Symptoms

The potential influence of possible risk factors including participants' voice use patterns, general health, and lifestyle on reporting a current swallowing disorder was examined. RRs for the significant variables were also determined in order to report how much more likely someone who possesses a particular attribute/condition is to report a swallowing disorder, versus someone who does not have the attribute/condition. Questions regarding voice use patterns were considered for analyses to determine whether these voice patterns impacted swallowing. Table 11 lists each condition addressed, the number of participants with each condition who also reported a current swallowing disorder, the resulting correlations, RRs, and CIs. The self-reported factors associated with significantly elevated RRs, from greatest to least, include voice problem longer than 4 weeks (RR = 2.0), esophageal reflux (RR = 1.92), current exposure to secondary tobacco smoke (RR = 1.67), often/constant neck/throat tension (RR = 1.66), frequent throat clearing (RR = 1.42), chronic postnasal drip (RR = 1.4), and stomach or duodenal ulcer (RR = 1.38).

Age, race/ethnicity, income, and education were not significantly associated with swallowing symptoms. Stepwise regression showed that swallowing symptoms were significantly associated with many risk factors, and polytomous logistic and stepwise regression showed symptom frequencies were also significantly associated. In the end, 60 (51%) of the 118 risk factors analyzed, along with the mentioned demographic questions, were significantly associated with symptoms and increased frequency of symptoms of swallowing problems. Risk factors most frequently associated with

Table 11. Current swallowing disorder and selected conditions

	Current Swallowing Disorder		χ^2	RR [95% CI]
Medical Condition	***n***	**%**	***p* Value**	
Arthritis	38	63.33	.795	
Rheumatoid arthritis	20	58.82	.408	
Heart disease	9	81.82	.200	
Hypertension	29	64.44	.987	
Circulatory problems	25	73.53	.170	
Kidney problems	15	75	.267	
Thyroid problems	30	66.67	.664	
Stomach or duodenal ulcer	**24**	**80**	**.033**	**1.38 [1.06, 1.81]**
Esophageal reflux	**49**	**79.03**	**.0001**	**1.93 [1.29, 2.87]**
Stroke	10	83.33	.144	
Respiratory allergies	25	69.44	.427	
Pneumonia	40	71.43	.097	
Emphysema	3	75	.650	
COPD	1	50	.669	
Hearing loss	23	74.19	.170	
Bronchitis	38	65.52	.777	
Asthma	15	68.18	.672	
Severe neck, back, or head injury	19	57.58	.322	
Chronic pain	44	65.67	.699	
Cancer	15	68.18	.672	
Depression or anxiety	43	71.67	.064	
Sleep disorder	20	76.92	.121	
Stopped menstrual periods	43	58.11	.132	
Voice				
Current voice problem	41	68.33	.313	
Did the problem begin suddenly?	5	71.43	.851	
Did the problem last more than 4 weeks?	**38**	**76**	**.004**	**2.00 [1.18, 3.38]**
Past voice problem	19	73.08	.281	
Family history of any type of voice problem	5	62.5	.910	
Medications for the following conditions				
Bronchitis	12	75	.262	
Asthma	32	71.11	.180	
Severe neck, back, or head injury	3	75	.746	
Chronic pain	26	68.42	.463	
Cancer	10	66.67	.147	

Table 11. Continued

Medical Condition	Current Swallowing Disorder *n*	Current Swallowing Disorder %	χ^2 *p* Value	RR [95% CI] Current Swallowing Disorder
Depression or anxiety	12	75	.262	
Sleep disorder	32	71.11	.180	
Condition				
Colds (3+ vs. < 3 year)	48	64	.899	
Sinus infections (3+ vs. < 3 year)	47	64.38	.993	
Sore throat (3+ vs. < 3 year)	42	60	.170	
Postnasal drip (Chronically vs. less)	**28**	**53.85**	**.023**	**1.40 [1.04, 1.89]**
Head and neck surgery	32	69.57	.318	
Breathing assistance	11	73.33	.432	
Ever used tobacco products	15	65.22	.922	
At any year in your life, drank an average of one or more alcoholic beverages a week	22	59.46	.435	
Ever used recreational drugs	5	71.43	.686	
Experience tension				
Neck/Throat (often/constantly vs. otherwise)	**26**	**49.06**	**.001**	**1.66 [1.22, 2.25]**
Jaw	46	59.74	.083	
Shoulders	35	60.34	.328	
Abdomen	56	62.92	.412	
Family history of any type of swallowing problem	12	70.59	.556	
Voice activity				
Talk (often/constantly vs. otherwise)	13	65	.947	
Talk quietly	37	64.91	.894	
Whisper	60	63.16	.317	
Talk loudly	40	61.54	.427	
Sing	56	63.64	.694	
Shout, yell or cheer	61	62.89	.129	
Clear your throat	**35**	**55.56**	**.017**	**1.42 [1.08, 1.87]**
Laugh	27	64.29	.99	
Cough	45	59.21	.060	
Ever in an occupation requiring you to talk a lot on a daily basis	52	64.20	.947	
Experience voice disorders with this job	25	69.44	.378	
Exposures				
Excess dust – current	9	52.94	.281	
Excess dust – past	31	70.45	.261	
Fumes from cleaning products – current	19	65.52	.877	

Table 11. Continued

Medical Condition	Current Swallowing Disorder *n*	χ^2 %	RR [95% CI] *p* Value	Current Swallowing Disorder
Fumes from cleaning products – past	31	67.39	.560	
Secondary tobacco smoke – current	**11**	**100**	**.009**	**1.67 [1.41, 1.97]**
Secondary tobacco smoke – past	42	70	.152	
Dry air – current	54	63.53	.689	
Dry air – past	56	64.37	.995	
Exercise (yes vs. no)	56	62.22	.200	
Description				
Quiet vs. talkative	25	62.5	.752	
Easy-going vs. worrier	35	64.81	.918	
Active vs. inactive	53	64.63	.904	
Happy vs. sad	58	63.04	.378	

Note. Only the RRs significant at the < .05 level are reported.

Note. Only *p* values significant at the < .05 level are bolded.

swallowing symptoms or frequency of symptoms, along with how many symptoms with which they were associated, were esophageal reflux (7), frequent coughing (5), stomach or duodenal ulcer (4), frequent sinus infections (4), current choral singing (4), being male (4), kidney problems (3), bronchitis (3), chronic pain (3), sleep disorder (3), abdomen tension (3), past church singing (3), past volunteer activities (3), acidic foods (3), and past exposure to secondary tobacco smoke (3). Other risk factors were significantly associated with one or two symptoms or symptom frequency. Four of the 19 swallowing symptoms were not found to have associated risk factors: wet/gurgley voice while eating, sneezing while eating, dry mouth, and avoiding certain foods.

Swallowing Disorders and Quality of Life

To assess the functional impact of a swallowing disorder on the SS population, several quality of life measures were included in the survey and used in analyses. These measures were associated with current swallowing disorders, current swallowing symptoms, and SS severity.

Scores on the MDADI, a measure of swallowing-related quality of life, can range from 0 (poorest quality of life) to 100 (best quality of life). Participants' scores ranged from 30 to 100, and scores were significantly lower for those who indicated they had a current swallowing disorder (M = 75.9, SD = 15.4 vs. 90.1, SD = 11.5, $p < .0001$). MDADI was not significantly associated with age, sex, race, ethnicity, income, or education. Mean MDADI score was significantly lower for 15 of the 19 selected swallowing-related symptoms (see Table 12). Regression analysis showed that the MDADI measure was significantly associated with SSI severity scores ($p = .001$),

Table 12. Swallowing symptoms and relation with MDADI scores

Symptom	MDADI Score		
	No Current Symptom *M* (*SD*)	Current Symptom *M* (*SD*)	*t* statistic *p* value
Take longer time to eat because of swallowing problem (*n* = 48)	**88.2 (11.4)**	**73.0 (15.9)**	**< .0001**
Difficulty swallowing liquids (*n* = 21)	82.4 (15.3)	75.8 (16.0)	.086
Difficulty swallowing solids (*n* = 65)	**87.8 (11.9)**	**77.2 (16.2)**	**.001**
Difficulty swallowing medications (*n* = 47)	**84.7 (14.0)**	**76.7 (16.5)**	**.010**
Gurgley or wet voice during or after eating (*n* = 5)	**81.8 (15.2)**	**64.8 (17.3)**	**.017**
Coughing, throat clearing, or choking before, during, or after eating (*n* = 46)	**86.3 (12.6)**	**74.6 (16.6)**	**.0001**
Inability to control food, liquid, or saliva in the mouth (*n* = 28)	**83.3 (14.2)**	**75.0 (17.8)**	**.017**
Sneezing during or after a meal (*n* = 12)	81.8 (14.5)	74.8 (22.1)	.142
Pain or pressure in the throat or chest during swallowing (*n* = 40)	**85.9 (14.0)**	**73.5 (15.1)**	**< .0001**
Wheezing after eating (*n* = 6)	**82.4 (14.2)**	**58.0 (20.6)**	**.0001**
Food comes out of nose while eating (*n* = 3)	**81.6 (15.2)**	**60.0 (17.8)**	**.018**
A need to chew excessively in order to swallow safely (*n* = 55)	**90.0 (10.3)**	**73.5 (15.4)**	**< .0001**
Dry mouth (*n* = 97)	78.5 (24.6)	81.1 (15.3)	.748
Difficulty placing food in mouth (*n* = 3)	81.2 (15.8)	73.3 (7.4)	.393
Sensation of food sticking in throat (*n* = 62)	**88.7 (12.8)**	**76.1 (15.4)**	**< .0001**
Forcibly regurgitate food stuck in throat (*n* = 18)	**83.9 (13.4)**	**67.7 (18.6)**	**< .0001**
Avoid eating certain foods because of swallowing disorder (*n* = 41)	**87.8 (11.6)**	**71.0 (15.5)**	**< .0001**
Increased mucous or phlegm in throat before, during, or after eating (*n* = 30)	**83.8 (13.8)**	**74.3 (17.8)**	**.005**
Take smaller bites of food to swallow safely (*n* = 55)	**90.5 (9.7)**	**73.1 (15.3)**	**< .0001**

Note. Only *p* values significant at the < .05 level are bolded.

ESSPRI severity scores (p = .004), and the question regarding overall SS severity (p = .032).

To assess the relationship between daily, overall health and the potential effects on swallowing, SS severity, and quality of life, an additional single-question measure was used. The variable about "how you feel, overall, on a daily basis" was independently significantly associated with two swallowing symptoms: food getting stuck in the throat and having mucous/phlegm while eating. Of those who reported food stuck in throat, 95.2% with good/fair/poor health compared with 4.8% with excellent health (MH χ^2 p = .025). Of those who reported mucous/phlegm while eating, 100% had good/fair/poor health compared with 0% with excellent health (p = .021). Along with specific swallowing symptoms, "how you feel..." was compared with reporting a swallowing disorder and the MDADI. "How you feel..." was statistically associated with the MDADI (p = .024) but not with reporting a current swallowing disorder (p = .255).

The SF-36 general quality of life measure was compared with current swallowing disorders and current swallowing symptoms. The two summary measures of the SF-36 are physical health and mental health. Each summary measure is associated with four subscales (physical health: physical function, role-physical, bodily pain, general health; mental health: vitality, social functioning, role-emotional, mental health). The raw scores from the survey are coded into a 0–100 scale, with 100 representing the highest state of health. Average scores for each of the scale items across all participants have been reported previously, ranging from the 70s for general health, bodily pain, and energy/fatigue, to the 80s for social functioning, physical functioning, role limitations – physical health, and emotional well-being to the 90s for role limitations – emotional

stresses. The highest possible score for physical functioning is obtained by performing all types of physical activity including the most vigorous without limitations due to health; for role-physical is obtained by having no problems with work or other daily activities; for bodily pain is obtained by having no pain or limitations due to pain; for general health is obtained by evaluating personal health as excellent; for energy/fatigue is obtained by feeling full of pep and energy all the time; for social functioning is obtained by performing normal social activities without interference due to physical or emotional problems; for role – emotional is obtained by having no problems with work or other daily activities; and for mental health is obtained by feeling peaceful, happy, and calm all of the time. Of the eight subscales, reporting a swallowing disorder was significantly associated with two: mental health (M = 72.5, SD = 17.9, p = .042) and general health (M = 33.9, SD = 20.1, p = .023). The relationship between reporting a swallowing disorder and the physical (p = .060) and mental health (p = .085) summary measures of the SF-36 approached significance. As detailed in Table 13, a logistics model showed that six swallowing symptoms were independently associated with the SF-36 subscales. These symptoms seem to disproportionately contribute to reductions in specific aspects of quality of life including bodily pain and vitality. For the model with summary physical health as the dependent variable, only food sticking in throat (slope = -9.4, SE = 4.8, p = .009) was independently associated. For the model with summary mental health as the dependent variable, difficulty swallowing medication (slope = -7.2, SE = 3.5, p = .046), wheezing while eating (slope = -31.2, SE = 7.5, p = .001), and mucous/phlegm while eating were independently associated (slope = -8.6, SE = 3.9, p = .038).

Of the 65 who reported a current swallowing disorder, only 27 (42%) had ever

Table 13. Mean SF-36 subscale scores according to current swallowing symptoms

Symptom (n)	PF Slope (SE)	p	RP Slope (SE)	p	BP Slope (SE)	p	GH Slope (SE)	p	VT Slope (SE)	p	SF Slope (SE)	p	RE Slope (SE)	p	MH Slope (SE)	p
Longer time to eat (48)																
Difficulty swallowing liquids (21)																
Difficulty swallowing solids (65)																
Difficulty swallowing meds (47)									**-10.5 (4.6)**	**.023**						
Gurgley/wet voice with eating (5)																
Cough/ throat clear/ choke with eating (46)																
Unable to control food/ liquid/ saliva in mouth (28)																
Sneezing with eating (12)					**-9.8 (7.4)**	**.049**										
Throat/chest pain/ pressure during swallowing (40)																
Wheezing after eating (6)									**-25.0 (9.6)**	**.018**	**-36.1 (12.9)**	**.006**	**-37.5 (12.4)**	**.002**	**-21.6 (6.7)**	**.002**
Food comes out nose while eating (3)																
Excessive chewing to swallow safely (55)																
Dry mouth (97)																
Difficulty placing food in mouth (3)																
Food sticking in throat (62)	**-13.2 (5.9)**	**.029**					**-14.6 (4.1)**	**.001**								
Need to forcibly regurgitate food stuck in throat (18)																

Table 13. Continued

Symptom (n)	PF Slope (SE)	p	RP Slope (SE)	p	BP Slope (SE)	p	GH Slope (SE)	p	VT Slope (SE)	p	SF Slope (SE)	p	RE Slope (SE)	p	MH Slope (SE)	p
Avoid eating certain foods due to poor swallowing (41)																
Increased mucous/ phlegm in throat with eating (30)					**-15.2 (5.1)**	**.011**										
Smaller bites of food to swallow safely (55)					**-7.5 (5.3)**	**.002**										

Note. Only significant slope estimates appear in the table, $p < .05$.
Physical Health: Physical Function (PF), Role-Physical (RP), Bodily Pain (BP), General Health (GH)
Mental Health: Vitality (VT), Social Functioning (SF), Role-Emotional (RE), Mental Health (MH)

sought professional help to treat their swallowing problem. Of those who sought help, 21 saw a physician, five saw a speech-language pathologist, three saw a dietician, and three saw another provider. Fifty-six percent indicated that it helped their swallowing (15/27), and half received a diagnosis (14/27 = 52%). Seeking professional help to treat their swallowing problem was significantly associated with a history of ulcer or chest/thoracic surgery.

Combined Voice and Swallowing Disorders

To assess the comorbidity of voice and swallowing disorders in SS, frequency counts were undertaken. Table 14 outlines the number of participants who reported both voice and swallowing disorders, one of these, or neither. Overall, 41 (40.6%) reported both current voice and current swallowing disorders. Of the 60 participants who reported a current voice disorder, 41 (68.3%) also reported a current swallowing disorder. Of the 65 participants who reported a current swallowing disorder, 41 (63.1%) also reported a current voice disorder. These results suggest an association between voice and swallowing disorders in the SS population.

Table 14. Prevalence of combined current voice and swallowing disorders

Current Voice Disorder	**Current Swallowing Disorder**	***n* (%)**
Yes	Yes	41 (40.6)
Yes	No	19 (18.8)
No	Yes	24 (23.8)
No	No	17 (16.8)

Discussion

The current study describes self-reported data from an epidemiologic study of swallowing disorders in SS. Prevalence, potential risk factors, and effects on quality of life were analyzed. The majority (64%) of participants with SS reported a current swallowing disorder. Most of these swallowing disorders began gradually (85%), were chronic, lasting 4 weeks or more (91%), and in many cases were longstanding (i.e., 85% persisted for 1+ years). The likelihood of reporting a current swallowing disorder was associated with increasing disease severity of SS, but interestingly was not associated with the age of the participants. One demographic variable, being male, was significantly associated with reporting a swallowing disorder. One other autoimmune condition, MCT, was also associated with reporting a swallowing disorder. These results confirm that chronic, longstanding swallowing disorders are common in SS, and appear to be related to worsening disease severity. Differences in prevalence rates were not observed between primary SS and when the disease coexisted with RA, which was the most frequently co-occurring autoimmune condition.

Patients with SS reported a high prevalence of swallowing symptoms (98% reported at least one), which often occurred on a daily and/or weekly basis. Increased likelihood of reporting a current swallowing disorder was associated with 13 of the 19 swallowing symptoms studied. The five most reported symptoms were dry mouth (reported by 96%), difficulty swallowing solids (64%), food sticking in the throat (61%), need for excessive chewing to swallow safely (55%), and taking smaller bites to swallow safely (55%). Only one symptom, sneezing while eating, was significantly associated with having SS longer. Reporting symptoms and increased frequency of symptoms were

significantly associated with SS severity, suggesting that worsening of SS severity is associated with increased prevalence of swallowing symptoms and disorders. Specifically, SSI was associated with four symptoms, ESSPRI was associated with three, and the question regarding overall disease severity was associated with three.

This study also examined associated medical conditions and potential risk factors that potentially contributed to swallowing disorder reporting. Certain medical conditions and lifestyle or occupational factors increased vulnerability for experiencing a swallowing disorder. Based upon inspection of the RR data, several interesting factors emerged. Some risk factors were laryngeal irritants, such as current exposure to secondary tobacco smoke, postnasal drip, and esophageal reflux. These irritants expose the laryngeal structures to potential inflammation within the upper aerodigestive tract and, therefore, may contribute to swallowing dysfunction. These irritants may also contribute to another risk factor, frequent throat-clearing, which may be a way the body attempts to clear these irritants or a way to compensate for dryness or laryngeal dysfunction associated with SS. Several other risk factors that were statistically associated with reporting a swallowing disorder included neck/throat tension and voice problems lasting longer than 4 weeks. These risk factors seem to have anatomical connections to structures involved in swallowing. The voice-related risk factor noted here demonstrates the potential effects of voicing on swallow function, again possibly due to related anatomy, and the comorbidity of voice and swallowing disorders in this group.

Of the medical, occupational, and lifestyle risk factors considered, 51% of them were associated with at least one, and up to seven, swallowing symptoms. Such a

diversity of risk factors may indicate that SS may be the true risk factor for swallowing symptoms, with increased prevalence of symptoms with additional medical and lifestyle components. However, several interesting factors were associated with having more, or more frequent, symptoms. As discussed above regarding risk factors for reporting swallowing disorders, some were potentially damaging laryngeal irritants, including esophageal reflux, acidic foods that may be related to reflux, frequent sinus infections, and past exposure to secondary tobacco smoke. These irritants may also contribute to another risk factor, frequent coughing, which is the body's defense mechanism against irritants. Some voice-related risk factors–current choral singing, past church singing, and past volunteer activities–may again reflect the frequent comorbidity of voice and swallowing disorders in this population, and the shared anatomy involved in both voice and swallowing function. Four symptoms were not found to have risk factors of those examined, and three of these four were some of the same symptoms not associated with reporting a swallowing disorder: wet/gurgley voice while eating, sneezing while eating, and dry mouth. Since dry mouth is a hallmark of SS, the finding that it is not significantly associated with risk factors or reporting a swallowing disorder is interesting. This finding may be related to the high rate (75%) at which the participants were medicated for SS and the description of the participant group being of moderate SS severity.

Effects of Swallowing Symptoms and Disorders on Quality of Life in SS

Our cohort reported high rates of undesirable swallowing disorders and related swallowing symptoms, and this high frequency of swallowing-related problems may help to explain some of the detrimental effects on quality of life reported by the participants. Inspection of the MDADI results revealed that swallowing disorders associated with SS produced mild to moderate reductions in swallowing-related quality of life. Lower MDADI scores were associated with increasing severity of SS. Our finding that SS severity was not correlated with years with SS, but was correlated with lower MDADI scores, is important. As with the voice results, this indicates that a person with SS can experience lower swallowing-related quality of life at any time point along their disease process.

The SF-36 general health quality of life measure also revealed important findings. Those who report a swallowing disorder also have poorer quality of life scores on two of the SF-36 subscales versus those who do not report a swallowing disorder. Together, these two associated subscales represent both the physical and mental health summary measures. Although statistical significance was not found between reporting a swallowing disorder and all SF-36 subscales, some swallowing symptoms were statistically significant: difficulty swallowing medications, sneezing while eating, wheezing while eating, food stuck in the throat, mucous/phlegm while eating, and need to take smaller bites. Of these symptoms, those independently associated with reduced SF-36 physical and mental health summary measures were difficulty swallowing medications, wheezing with eating, food stuck in the throat, and mucous/phlegm while

eating. Interestingly, the symptom that seems most related to SS, dry mouth, was not significantly associated with subscales or summary measures of the SF-36. As mentioned above, this was also the case when analyzing symptoms significant for reporting a swallowing disorder.

The additional quality of life variable, "how do you feel, overall, on a daily basis," was not significantly correlated with reporting a swallowing disorder. However, it was associated with the MDADI. This question was significantly associated with two of the same symptoms associated with both the MDADI and the SF-36: food stuck in the throat and mucous/phlegm while eating. These two symptoms may be related to the high rate of reflux that is associated with swallowing problems in this population.

Although 64% of participants reported a swallowing disorder and MDADI scores for these participants were lower, only 27% had ever sought professional help to treat their swallowing disorder. Seeking professional help was associated with a history of ulcers and chest/thoracic surgery. One possible explanation for this low rate of seeking treatment may be limited education regarding prevalence of and treatment for swallowing disorders in this population, as well as limited treatment options. Although the average swallowing problem in this population appears to be moderately severe, additional health concerns associated with SS may consume more health-seeking resources. However, regression analysis suggested that as SS severity worsens, so do swallowing disorders. The participants in this study reported moderate average SS severity. Perhaps swallowing problems and quality of life ratings would be negatively impacted in an individual whose SS is more severe. This finding is important for health professionals to consider for treatment paradigms. Importantly, 56% of treatment-seeking participants indicated

improvement following treatment. Improved treatment options for this population would likely improve success rate of treatment.

Further research surrounding swallowing disorders in SS is warranted due to these quality of life findings. Swallowing disorders statistically impacted the MDADI and portions of the SF-36. SS severity was statistically associated with reporting a swallowing disorder and subsequently impacted MDADI. Various swallowing symptoms were also statistically correlated with reporting a swallowing disorder, along with all disease severity and quality of life measures. Additional research in this area to further analyze the correlations between swallowing disorders, symptoms, and quality of life may demonstrate additional correlations with the SF-36. As with voice disorders research in SS, there is a need for continued research investigating novel treatment options for swallowing disorders in this population.

Comparisons with the Extant Literature

In reviewing the extant literature in the area of swallowing disorders in SS, the studies critiqued in Table 1 could not be directly compared to the current study due to low numbers of SS in the rheumatic groups studied, methodology differences, and/or differences in definition of a swallowing disorder. Therefore, loose comparisons were made. The high prevalence for swallowing disorders in SS found in the current study (64%) is similar to the range of 65 to 71% previously reported by participants in other studies (Mandl et al., 2007; Ruiz Allec et al., 2011). Prevalence was similar even though these two studies included only 20 and 31 SS participants, respectively, and they defined swallowing disorders differently. Though the current study did not use objective

measures as in other studies, a common report among most of the swallowing-related studies in Table 1 is that dysphagia is significant in SS (Mandl et al., 2007; Rhodus et al., 1995; Ruiz Allec et al., 2011). The prevalence of swallowing symptoms has been reported at 80–100% (Doig et al., 1971; Mandl et al., 2007; Rogus-Pulia & Logemann, 2011). Similarly, the current study found at least one symptom of dysphagia in 98% of participants. Dry mouth was reported by 96% of participants in the current study; Doig et al. (1971) and Mandl et al. (2007) did not include this symptom, and their prevalence of symptoms was 81 and 80%, respectively. It is also important to note that the SS diagnostic criteria used by Doig et al. (1971) differs greatly from that used in the current study, which followed more recent guidelines. Additionally, both the Doig et al. (1971) and the Mandl et al. (2007) studies included a smaller number of participants than the current study (53 SS and 20 SS, respectively, of 101). Some studies reported a large difference between perception of swallowing problems and signs of swallowing dysfunction as observed using objective measures of swallowing function. Rogus-Pulia and Logemann (2011) reported their SS participants rated their swallowing as worse than the 3% found on objective measures. Conversely, Ruiz Allec et al. (2011) found that swallowing problems were objectively worse than were reported (90% vs. 71%). Both studies had a relatively small cohort and used a much smaller swallowing symptom questionnaire than the current study, asking only 12 and one swallowing-related question(s), respectively. When comparing risk factors for swallowing disorders presented in the extant literature, reflux seemed to be the one suggested in other studies (Mandl et al., 2007; Ruiz Allec et al., 2011). These reports support findings in the current study that found reflux to be a significant risk factor for swallowing disorders.

The final comparison to be made is in the area of quality of life resulting from swallowing disorders in SS. No studies in Table 1 addressed socioemotional effects of these disorders.

Comparisons between the current study and several non-SS studies were made to review interesting risk factors in the current study that were not independently associated with reporting a current swallowing disorder. These factors include stroke, COPD, and chronic pain, which have previously been associated with reporting a swallowing disorder in a geriatric population (Roy et al., 2007b), although similar associations were not found in the current study. Other factors not associated with reporting a current swallowing disorder include years with SS and current use of medication. Although RA was the most common comorbid autoimmune condition, MCT was the only one significantly associated with reporting a swallowing disorder. This finding may be important as SS awareness increases with potential diagnosis of comorbid MCT, and future research may examine swallowing disorders in a larger cohort with this autoimmune condition. In a concurrent study (Wright, 2014), RA was found to have a higher incidence of swallowing disorders than the general population (41% vs.12–13%; Groher & Bukatman, 1986), but not as high as the incidence found in SS with or without comborbid RA in the current study. Increased age has also been associated with increased prevalence of swallowing disorders compared with the general population (33% in age 65+ vs. 12–13%, respectively; Groher & Bukatman, 1986; Roy et al., 2007b). The majority of participants in the current study (78%) were aged 60+, and statistical analyses did not result in a correlation between increased age and increased prevalence of swallowing disorders. SS seems to be associated with a higher prevalence

of swallowing disorders than both the general population and the geriatric population. This seems to show that SS severity is the true risk factor for reporting a swallowing disorder, regardless of age or the majority of comorbid autoimmune conditions.

CONCLUSIONS

This study, based on self-report data from a large SS cohort, revealed a high rate of voice and swallowing disorders and symptoms, and their possible negative influence on quality of life. Various medical, lifestyle, and personality risk factors were associated with reporting these disorders and symptoms. Correlations between voice and swallowing disorders and SS severity were also found. It appears that SS severity contributes to reductions in voice- and swallowing-related quality of life. Our results contribute to and expand those from previous studies and suggest a greater need for education and awareness of voice and swallowing disorders in the SS population. These results, combined with a low rate of seeking treatment, demonstrate the need for awareness of voice and swallowing disorders at any stage of this disease. Additionally, research in this area should continue to better understand the nature and relationship between comorbid voice and swallowing disorders, symptoms, and resulting quality of life.

APPENDIX

EPIDEMIOLOGY OF VOICE AND SWALLOWING DISORDERS IN AUTOIMMUNE DISEASES: A PILOT STUDY

Subject #: _____

Questionnaire

Date: _____ START TIME __ __ : __ __ am pm Survey administrator: _____

To start, I have a few questions about you.

1. What is your date of birth?

   ___ / ___ / ___
   Mo. Day Yr.

2. How many years of schooling have you completed including any college, vocational, or technical training?

   _____ **[ACTUAL # OF YEARS]**

   12 = High School (includes GED)
   14 = Associate Degree
   16 = B.A., B.S.
   18 = M.A., M.S.
   20 = Ph.D., M.D., J.D., etc.

3. a. What is your race or ethnicity? Are you …(read list)
   1 = White,
   2 = Black or African American,
   3 = Native Hawaiian or Pacific Islander,
   4 = Asian,
   5 = Native American or Alaska Native, or
   6 = Hispanic
   7 = Something else? (Specify: _____)

b. Are you Hispanic or of Spanish origin?

1 = Yes
0 = No

4. Which of the following categories best describes your current household yearly gross income (before taxes for everyone who lives with you)? …

1 = Under $20,000,
2 = From $20,000 to $40,000,
3 = From $40,000 to $60,000, or
4 = More than $60,000?
5 = DON'T KNOW
6 = PREFER TO NOT ANSWER

Subject #: _____

This first section of the interview relates to your health and medical history during your entire life.

5. a. During your entire life, have you ever had…

1 = Yes **(IF YES, CONTINUE 5b-5e)**
0 = No **(GO TO NEXT MEDICAL CONDITION)**
7 = Don't Know
9 = Refused to answer

b. What year did you first have (this condition)?

__ __ __ __

c. Are you currently taking medications for this condition?

1 = Yes
0 = No

d. In what year did you first start taking this medication?

Medical Condition	**a. Ever had**				**b. First Time**	**c. Currently taking**		**d. Year started**
	Yes	No	DK	Ref	Year	Yes	No	
Arthritis?	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Sjogren's Syndrome? Rheumatoid Arthritis? Diabetes Type 1? Wegener's Granulomatosis? Scleroderma? Dermatomyositis?	1	0	7	9	__ __ __ __	1	0	__ __ __ __

Polymysitis? Systemic Lupus? Mixed connective tissue? Other disease?								
Medical Condition	**a. Ever had**				**b. First Time**	**c. Currently taking**		**d. Year started**
Heart disease	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Hypertension / high blood pressure	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Circulatory problems	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Kidney problems	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Thyroid problems If YES Did you have… hyperthyroid (overactive) hypothyroid (underactive)	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Stomach or duodenal ulcer	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Esophageal reflux, such as burning, burping acid taste, or acid indigestion?	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Stroke	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Respiratory allergies?	1	0	7	9	__ __ __ __	1	0	__ __ __ __

(IF YES) Are they… 1 = seasonal 2 = non-seasonal								
Pneumonia	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Emphysema	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Chronic Obstructive Pulmonary Disease or COPD?	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Medical Condition	**a. Ever had**				**b. First Time**	**c. Currently taking**		**d. Year Started**
Bronchitis	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Asthma	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Severe neck, back, or head injury	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Chronic pain	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Cancer	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Depression or anxiety	1	0	7	9	__ __ __ __	1	0	__ __ __ __
Sleep disorder	1	0	7	9	__ __ __ __	1	0	__ __ __ __

6. On average, how many times a year do you get **(CONDITION)**?

Would you say never, less than once a year, once or twice a year, 3 to 5 times, 6 to 8 times, or more than 8 times a year?

Condition	**Never**	**<1**	**1-2**	**3-5**	**6-8**	**>8**
a. colds?	0	1	2	3	4	5
b. sinus infections?	0	1	2	3	4	5

c. sore throats?	0	1	2	3	4	5

7. Do you have post-nasal drip …

   1 = Chronically,
   2 = Seasonally,
   3 = Occasionally with colds, or
   4 = Not at all?

8. Have you ever had surgery?

   1 = YES
   0 = NO (if no, skip to question 9)

Surgery	**Yes**	**No**
a. Head and neck surgery	1	0
b. Chest / Thoracic surgery	1	0
c. Abdominal surgery	1	0
d. Other. Specify_____ _____	1	0

9. Did any of your surgeries or hospitalizations require you to be on a respirator / ventilator / breathing machine or trach?

   1 = YES
   0 = NO

10. **CODE SEX.** ASK IF UNSURE: Are you…

0 = Male or **(GO TO Q12)**
1 = Female?

11. [FOR WOMEN ONLY]

a. Have you stopped having menstrual periods? **(If NO, go to Q 12)**

b. How many years ago did you stop menstruating?

1-2 years = 0
3-5 years = 1
6-10 years = 2
10+ years = 3

c. Are you taking estrogen replacement therapy?

1 = YES
0 = NO **(IF NO, GO TO 12)**

d. How long have you been taking replacement therapy?

1-2 years = 0
3-5 years = 1
6-10 years = 2
10+ years = 3

12. Now we have a few questions about some of your personal habits including cigarette smoking, other tobacco use, and the use of alcohol.

a. Have you ever used any tobacco products for a year or longer?

1 = Yes
0 = No **(GO TO Q 13)**

b. Have you ever (smoked/used) **[TYPE]** for a year or longer?

	Yes	No
a. Cigarettes	1	0
b. Cigars	1	0
c. Pipes	1	0
d. Chewing tobacco	1	0

c. What year did you first start (smoking/chewing)?

— — — —

d. Do you still (smoke/chew)?

1 = Yes **(GO TO f)**
0 = No

e. In what year did you stop (smoking/chewing)?

— — — —

f. On average, how many (cigarettes/cigars/pipefuls/pinches) (do/did) you (smoke/use) a day?

_____

13. a. At any year in your life, did you drink an average of one or more alcoholic beverages a week?

1 = Yes
0 = No **(GO TO Q14)**

b. How old were you when you first started drinking alcoholic beverages regularly?

__ __ years

c. Do you still drink? **[THIS MEANS ANY AMOUNT OF ALCOHOL]**

1 = Yes **(GO TO e)**
0 = No

d. What year did you stop drinking?

__ __ __ __

e. On average, how many drinks do/did you drink in a week?
_____ # drinks

14. a. Have you ever used recreational drugs?

1 = Yes
0 = No (IF NO, GO TO 15)

b. Have you ever used (TYPE) for a year or longer?

a. Marijuana
b. Heroine
c. Cocaine
d. LSD

e. Other. Specify _____

c. How old were you when you first started using recreational drugs regularly?

__ __ years

d. Do you still use recreational drugs?

1 = Yes **(GO TO f)**
0 = No

e. What year did you stop using recreational drugs?

__ __ __ __

f. On average, how many times do / did you use recreational drugs in a week?

_____ # of times

15. How often do you use / partake of the following items? (read "constantly, often…never")

Activity	Constantly	Often	Occasionally	Rarely	Never
a. Coffee	5	4	3	2	1
b. Tea	5	4	3	2	1
c. Colas	5	4	3	2	1
d. Chocolate	5	4	3	2	1
e. Dairy Products	5	4	3	2	1

f. Mint Products	5	4	3	2	1
g. Acidic Foods	5	4	3	2	1
h. Spicy Foods	5	4	3	2	1
i. Water	5	4	3	2	1

16. How often do you experience tension in the following areas? (read "constantly, often…never")

Activity	Constantly	Often	Occasionally	Rarely	Never
a. Neck / Throat	5	4	3	2	1
b. Jaw	5	4	3	2	1
c. Shoulders	5	4	3	2	1
d. Abdomen	5	4	3	2	1

17. Now I would like to ask you some questions about your voice. For the purpose of this study, we consider a voice problem to be any time your voice does not work, perform, or sound as you feel it normally should, so that it interferes with communication.

a. Do you currently have a voice problem like this?

1 = Yes (continue to 17b below)
0 = No (skip to 18)

b. Did the problem begin suddenly or gradually?

1 = Suddenly

0 = Gradually

c. Has this problem lasted for more than 4 weeks?

1 = Yes
0 = No (IF NO – GO TO 18)

d. When did you first notice the problem?

1 = 1-6 months ago
2 = 7-12 months ago
3 = 1-3 years ago
4 = 4-9 years ago
5 = 10+ years ago

18. a. Have you ever had a voice problem like this in the past?

1 = Yes
0 = No **(GO TO Q 19a)**

b. Approximately what year did you first notice that you had a voice problem?

— — — —

c. Did this problem last for 4 weeks or more?

1 = Yes **(CHRONIC)**
0 = No **(ACUTE)**

d. Did the problem begin gradually or suddenly?

1 = Gradually
2 = Suddenly

e. Have you had any voice problems since that time?

1 = Yes
0 = No **(GO TO Q 19)**

f. Have your voice problems been …

1 = continual, or
0 = off and on?

g. Have you ever seen a doctor or speech pathologist about any type of voice problem?

1 = Yes
0 = No **(GO TO Q 19)**

h. What year did you first see a doctor or speech pathologist about a voice problem?

— — — —

i. What was the problem?

_____

19. Do you have a family history of any type of voice problem?

1 = Yes
0 = No

Subject #: _____

20. Now, I will read a list of voice symptoms. For each one, please tell me if you have ever had that symptom.

a. Have you ever had (SYMPTOM)?

1 = Yes
0 = No **(GO TO NEXT SYMPTOM)**

b. Do you have this symptom currently?

1 = Yes
0 = No

c. How often do you have this (symptom)? Would you say …

1 = daily,
2 = weekly,
3 = monthly,
4 = several times a year, or
5 = yearly or less?

d. Do you think this (symptom) is the result of your job?

1 = Yes
0 = No

SYMPTOM	**a. Past**		**b. Current**		**c. Frequency**					**d. Job**	
	Yes	No	Yes	No	Dy	Wk	Mn	Sev	Yr	Yes	No
aa. Hoarseness?	1	0	1	0	1	2	3	4	5	1	0

Subject #: _____

SYMPTOM	a. Past		b. Current		c. Frequency					d. Job	
	Yes	No	Yes	No	Dy	Wk	Mn	Sev	Yr	Yes	No
ab. Your voice tire or change quality after using it for even a short time?	1	0	1	0	1	2	3	4	5	1	0
ac. Trouble speaking or singing softly?	1	0	1	0	1	2	3	4	5	1	0
ad. Difficulty projecting your voice?	1	0	1	0	1	2	3	4	5	1	0
ae. A loss of singing range?	1	0	1	0	1	2	3	4	5	1	0
af. Discomfort while using your voice?	1	0	1	0	1	2	3	4	5	1	0
ag. A monotone voice (monopitch)?	1	0	1	0	1	2	3	4	5	1	0
ah. To make an effort to talk?	1	0	1	0	1	2	3	4	5	1	0
ai. Chronic dryness in your throat?	1	0	1	0	1	2	3	4	5	1	0
aj. Wet, gurgley voice quality	1	0	1	0	1	2	3	4	5	1	0
ak. Chronic soreness in your throat?	1	0	1	0	1	2	3	4	5	1	0
al. To frequently clear your throat?	1	0	1	0	1	2	3	4	5	1	0
am. A bitter or acid taste?	1	0	1	0	1	2	3	4	5	1	0

an. Wobbly or shaky voice?	1	0	1	0	1	2	3	4	5	1	0
ao. An "airy" or "breathy" voice?	1	0	1	0	1	2	3	4	5	1	0

21. Next, I will read a list of activities. For each one, please tell me whether you do this constantly, often, occasionally, rarely or never during an average day.

How frequently do you (**ACTIVITY)** during an average day? Would you say constantly, often, occasionally, rarely or never?

Activity	**Constantly**	**Often**	**Occasionally**	**Rarely**	**Never**
a. Talk	5	4	3	2	1
b. Talk quietly	5	4	3	2	1
c. Whisper	5	4	3	2	1
d. Talk loudly	5	4	3	2	1
e. Sing	5	4	3	2	1
f. Shout, yell, or cheer	5	4	3	2	1
g. Clear your throat	5	4	3	2	1
h. Laugh	5	4	3	2	1
i. Cough	5	4	3	2	1

Subject #: _____

22. Some people use / have used their voices for other activities.

a. Do you currently participate in (ACTIVITY)?

1 = YES
0 = NO

b. In the past, have you participated in (ACTIVITY)?

1 = YES
0 = NO

Activity	Current		Past	
	Yes	No	Yes	No
aa. choral singing	1	0	1	0
ab. singing at church	1	0	1	0
ac. solo singing	1	0	1	0
ad. stage acting	1	0	1	0
ae. public speaking	1	0	1	0
af. volunteer activities	1	0	1	0

23. Now we would like some information about your work history. Some occupations require extensive voice use such as sales, teaching, broadcasting, clergy, telephone operator, or receptionist.

a. Are / were you ever employed in a job that required you to talk a lot on a daily basis?

1 = Yes
0 = No (IF NO, GO TO 24)

Subject #: _____

b. How many years have you been / were you employed in this type of job?

1 = 0 – 9
2 = 10 – 19
3 = 20 – 29
4 = 30 – 39
5 = 40+

c. What is / was your job title? _____

d. What are / were your activities on this job?_____

_____

e.. Do / Did you experience any voice problems with this job?

1 = YES
0 = NO (IF NO, GO TO h)

f.. How long did the problem last? Would you say…

1 = A few days
2 = A few weeks
3 = A few months, or
4 = A year or more

g. Would you say…

1 = your voice **limits / limited** your ability to do certain tasks in this job?

2 = your voice **makes / made you unable** to do certain tasks
3 = your voice **does not / did not affect** your ability to do various tasks

h. Did you ever change (IF EMPLOYED or feel you need to change) your occupation or job because of your voice?

1 = YES
0 = NO

i. Did you have to retire or take disability due to your voice problem?

1 = YES
0 = NO

24. a. Have you ever sought professional help to improve your voice in any way?

1 = Yes
0 = No **(GO TO Q 25)**

b. Did you see … **[CIRCLE ALL THAT APPLY]**

1 = a speech or language pathologist,
2 = a physician,
3 = a singing or acting teacher or coach, or
4 = someone else? Specify_____

c. Did it help you?

1 = Yes
0 = No

Subject #: _____

25. We are interested in whether you have ever been exposed to any of the following?

a. Are you currently exposed to (ITEM)?

1 = YES
0 = NO

b. Have you ever been exposed to (ITEM)?

1 = YES
0 = NO

Exposure	Current		Past	
	Yes	No	Yes	No
aa. Excess Dust	1	0	1	0
ab. Fumes from cleaning products	1	0	1	0
ac. Secondary tobacco smoke	1	0	1	0
ad. Dry air	1	0	1	0
ae. Other. Specify_____ _____	1	0	1	0

Subject #: _____

26. I will now ask you 10 questions about problems you may be having with your voice. Each question will ask you to rate the severity of the problem for you. To determine how "bad" it is, think of both how often the problem occurs and how severe it is when it happens. Rate each question on a 5-point scale below, with "5" being the worst possible. Your answers should be based on your average voice quality over the past 2 weeks or so. (Read the 5-point scale when completing this section.)

1 = None, not a problem
2 = A small amount
3 = A moderate (medium) amount
4 = Frequently
5 = As bad as it can be

Read "Because of your voice" prior to each question; (give options "none, small amount…as bad as can be" for each question)

	None	Small Amt.	Mod. Amt.	Freq.	As bad as can be
a. Do you have trouble speaking loudly or being heard in noisy situations?	1	2	3	4	5
b. Do you run out of air and need to take frequent breaths when talking?	1	2	3	4	5
c. You do not know what will come out when you begin speaking?	1	2	3	4	5
d. Do you get anxious or frustrated?	1	2	3	4	5
e. Do you get depressed?	1	2	3	4	5
f. Do you have trouble using the telephone?	1	2	3	4	5
g. Do you have trouble doing your job or practicing your profession? (Indicate if not applicable ____NA)	1	2	3	4	5
h. Do you avoid going out socially?	1	2	3	4	5
i. Do you have to repeat yourself to be understood?	1	2	3	4	5
j. Have you become less outgoing?	1	2	3	4	5

Subject #: _____

Now, I would like to ask you some questions about your ability to swallow. For the purpose of this study we consider a swallowing problem to be anytime when you experience difficulty moving food or liquid from your mouth to your stomach or anytime you experience choking or frequent throat clearing during or following mealtime.

27a. Do you currently have a swallowing problem like this?

1 = Yes
0 = No (Skip to 28a)

b. Did the swallowing problem begin suddenly or gradually?

1 = suddenly
0 = gradually

c. Has this swallowing problem lasted for more than 4 weeks?

1 = Yes
0 = No (IF NO, GO TO 28)

d. When did you first notice that you have this swallowing problem?

1 = 1-6 months
2 = 7-12 months
3 = 1-3 years
4 = 4-9 years
5 = 10+ years

e. Did your swallowing problem begin with a reaction to medicine:

1 = Yes
0 = No

f. Did your swallowing problem begin following an illness?

1 = Yes
0 = No

g. Did your swallowing problem begin following surgery?

1 = Yes
0 = No

28a. Have you ever had a swallowing problem like this in the past?

1 = Yes
0 = No (Skip to 29)

28b. Approximately what year did you notice that you had this problem?

— — — —

29. Have you ever had?

a. A feeding tube

1 = Yes
0 = No

b. A need to use nutritional supplements (e.g. Ensure, Boost) to make sure that you are taking in enough calories each day?

1 = Yes

0 = NO

30. Now, I will read a list of swallowing symptoms. For each one, please tell me if you have ever had that symptom.
    a. Have you ever had (SYMPTOM)?

        1 = Yes
        0 = No **(GO TO NEXT SYMPTOM)**

    b. Do you have (SYMPTOM) currently?

        1 = Yes
        0 = No

    c. How often do you have (SYMPTOM)? Would you say…

        1 = daily,
        2 = weekly,
        3 = monthly,
        4 = several times a year, or
        5 = yearly or less?

SYMPTOM	a. **Past**		b. **Current**		c. **Frequency**				
	Yes	No	Yes	No	Dy	Wk	Mn	Sev	Yr
aa. To take a longer time to eat because of your swallowing problem?	1	0	1	0	1	2	3	4	5
ab. Difficulty swallowing	1	0	1	0	1	2	3	4	5

Subject #: _____

liquids?									
ac. Difficulty swallowing solids?	1	0	1	0	1	2	3	4	5
ad. Difficulty swallowing medications?	1	0	1	0	1	2	3	4	5
ae. A gurgley or wet voice during or after eating?	1	0	1	0	1	2	3	4	5
af. Coughing, throat clearing, or choking before, during or after eating?	1	0	1	0	1	2	3	4	5
ag. An inability to control food, liquid, or saliva in the mouth?	1	0	1	0	1	2	3	4	5
ah. Any sneezing during or after a meal?	1	0	1	0	1	2	3	4	5
ai. Pain or pressure in the throat or chest during swallowing?	1	0	1	0	1	2	3	4	5
aj. Wheezing after eating?	1	0	1	0	1	2	3	4	5
ak. Food come out of your nose while eating?	1	0	1	0	1	2	3	4	5
al. A need to chew excessively in order to swallow safely?	1	0	1	0	1	2	3	4	5
am. Dry mouth?	1	0	1	0	1	2	3	4	5
an. Difficulty placing food in mouth?	1	0	1	0	1	2	3	4	5

ao. A sensation of food sticking in your throat?	1	0	1	0	1	2	3	4	5
ap. To forcibly regurgitate food that is stuck in your throat?	1	0	1	0	1	2	3	4	5
aq. To avoid eating certain foods because of your swallowing disorder?	1	0	1	0	1	2	3	4	5
ar. Increased mucous or phlegm in your throat before, during, or after you eat?	1	0	1	0	1	2	3	4	5
as. To take smaller bites of food in order to swallow safely.	1	0	1	0	1	2	3	4	5

31. The next few questions ask about your views of your swallowing ability. This information will help us understand how you feel about swallowing. The following statements have been made by people who have problems with their swallowing. Some of the statements may apply to you. Please listen to each statement and indicate the response which best reflects your experience in the past week. You may refer to this key as you consider your answer. (READ KEY OF 5 POSSIBLE RESPONSES)

	Strongly Agree	Agree	No Opinion	Disagree	Strongly Disagree
a. My swallowing ability limits my day-to-day activities	1	2	3	4	5
b. I am embarrassed by my eating habits.	1	2	3	4	5
c. People have difficulty cooking for me.	1	2	3	4	5

Subject #: _____

d. Swallowing is more difficult at the end of the day.	1	2	3	4	5
e. I do not feel self-conscious when I eat.	5	4	3	2	1
f. I am upset by my swallowing problem.	1	2	3	4	5
g. Swallowing takes great effort.	1	2	3	4	5
h. I do not go out because of my swallowing problem.	1	2	3	4	5
i. My swallowing difficulty has caused me to lose income?	1	2	3	4	5
j. It takes me longer to eat because of my swallowing problem.	1	2	3	4	5
k. People ask me, “Why can’t you eat that?”	1	2	3	4	5
l. Other people are irritated by my eating problem.	1	2	3	4	5
m. I cough when I try to drink liquids.	1	2	3	4	5
n. My swallowing problems limit my social and personal life.	1	2	3	4	5
o. I feel free to go out to eat with my friends, neighbors, and relatives.	5	4	3	2	1
p. I limit my food intake because of my swallowing difficulty.	1	2	3	4	5
q. I cannot maintain my weight because of my swallowing	1	2	3	4	5

problem.					
r. I have low self-esteem because of my swallowing problem.	1	2	3	4	5
s. I feel that I am swallowing a huge amount of food.	1	2	3	4	5
t. I feel excluded because of my eating habits.	1	2	3	4	5

32. Do you have a family history of any type of swallowing problem?

1 = Yes
0 = No
DK= Don't know

33. a. Have you ever sought professional help to assess or treat your swallowing problem in any way?

1 = Yes
0 = No **(GO TO Q 34)**

b. Did you see … **[CIRCLE ALL THAT APPLY]**

1 = a speech or language pathologist,
2 = a physician,
3 = a dietician, or
4 = someone else? Specify_____

c. Did it help you?

1 = Yes

0 = No

d. Did you ever receive a diagnosis related to your swallowing?

1 = Yes
0 = No **(Go To Q 34)**

e. What was the diagnosis you received? _____

34. a. Do you live alone?

1 = Yes (IF YES, GO TO 35)
0 = No

b. How many people (including you) live in your home? ____ (insert #)

35. a. Do you have a hearing loss?

1 = Yes
0 = No (IF NO, GO TO 36)

b. Do you wear hearing aids on a regular basis?

1 = Yes
0 = No

36. a. Do you exercise?

1 = Yes

0 = No (GO TO 37)

b. How often do you exercise?

1 = 1-2 X week
2 = 3-4 X week
3 = 5+ X week

37. Overall, how do you feel on a daily basis?

1 = excellent
2 = good
3 = fair
4 = poor

38. Would you describe yourself as …

a. Quiet or talkative

1 = Quiet
0 = Talkative

b. Easy-going or a worrier

1 = Easy-going
0 = Worrier

c. Active or inactive

1 = Active
0 = Inactive

d. Happy or sad

1 = Happy
0 = Sad

39. What is your religious preference?

1 = Protestant
2 = Catholic
3 = LDS/Mormon
4 = Jewish
5 = Other
6 = No Religion
7 = Prefer not to answer

40. Now I would like to ask you some questions about your speech. For the purpose of this study, we consider a speech problem to be any time when you experience difficulty with speech production (including slurred speech or imprecise sound production/articulation) so that it interferes with communication.

a. Do you currently have a speech problem like this?

1 = Yes (continue to 40b below)
0 = No (skip to 41)

b. Did the problem begin suddenly or gradually?

1 = Suddenly
0 = Gradually

c. Has this problem lasted for more than 4 weeks?

1 = Yes
0 = No (IF NO – GO TO 41)

d. When did you first notice the problem?

1 = 1-6 months ago
2 = 7-12 months ago
3 = 1-3 years ago
4 = 4-9 years ago
5 = 10+ years ago

41. a. Have you ever had a speech problem like this in the past?

1 = Yes
0 = No **(GO TO Q 42)**

b. Approximately what year did you first notice that you had a speech problem?

— — — —

c. Did this problem last for 4 weeks or more?

1 = Yes **(CHRONIC)**

0 = No **(ACUTE)**

d. Did the problem begin gradually or suddenly?

1 = Gradually
2 = Suddenly

e. Have you had any speech problems since that time?

1 = Yes
0 = No **(GO TO Q 42)**

f. Have your speech problems been …

1 = continual, or
0 = off and on?

g. Have you ever seen a doctor or speech pathologist about any type of speech problem?

1 = Yes
0 = No **(GO TO Q 42)**

h. What year did you first see a doctor or speech pathologist about a speech problem?

— — — —

i. What was the problem?

_____

Subject #: _____

42. Do you have a family history of any type of speech problem?

1 = Yes
0 = No

43. *I would like to ask you a few more questions about your general health.* (READ NUMERIC CHOICES)

(Medical Outcomes Study: 36-Item Short Form Survey Instrument (RAND 36-Item Health Survey 1.0 Questionnaire Items)

1. In general, would you say your health is:	
Excellent	1
Very good	2
Good	3
Fair	4
Poor	5

2. **Compared to one year ago**, how would your rate your health in general **now**?	
Much better now than one year ago	1
Somewhat better now than one year ago	2

About the same	3
Somewhat worse now than one year ago	4
Much worse now than one year ago	5

The following items are about activities you might do during a typical day. Does **your health now limit you** in these activities? If so, how much?

(Circle One Number on Each Line)

	Yes, Limited a Lot	Yes, Limited a Little	No, Not limited at All
3. **Vigorous activities**, such as running, lifting heavy objects, participating in strenuous sports	[1]	[2]	[3]
4. **Moderate activities**, such as moving a table, pushing a vacuum cleaner, bowling, or playing golf	[1]	[2]	[3]
5. Lifting or carrying groceries	[1]	[2]	[3]

6. Climbing **several** flights of stairs	[1]	[2]	[3]
7. Climbing **one** flight of stairs	[1]	[2]	[3]
8. Bending, kneeling, or stooping	[1]	[2]	[3]
9. Walking **more than a mile**	[1]	[2]	[3]
10. Walking **several blocks**	[1]	[2]	[3]
11. Walking **one block**	[1]	[2]	[3]
12. Bathing or dressing yourself	[1]	[2]	[3]

During the **past 4 weeks**, have you had any of the following problems with your work or other regular daily activities **as a result of your physical health**?

(Circle One Number on Each Line)

	Yes	No
13. Cut down the amount of time you spent on work or other activities	1	2
14. **Accomplished less** than you would like	1	2

Subject #: _____

15. Were limited in the **kind** of work or other activities	1	2
16. Had **difficulty** performing the work or other activities (for example, it took extra effort)	1	2

During the **past 4 weeks,** have you had any of the following problems with your work or other regular daily activities **as a result of any emotional problems** (such as feeling depressed or anxious)?

(Circle One Number on Each Line)

	Yes	No
17. Cut down the **amount of time** you spent on work or other activities	1	2
18. **Accomplished less** than you would like	1	2
19. Didn't do work or other activities as **carefully** as usual	1	2

20. During the **past 4 weeks,** to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbors, or groups?

(Circle One Number)

Not at all = 1

Slightly = 2

Moderately = 3

Quite a bit = 4

Extremely = 5

21. How much **bodily** pain have you had during the **past 4 weeks**?

(Circle One Number)

None = 1

Very mild = 2

Mild = 3

Moderate = 4

Severe = 5

Very severe = 6

22. During the **past 4 weeks,** how much did **pain** interfere with your normal work (including both work outside the home and housework)?

(Circle One Number)

Not at all = 1

Subject #: _____

A little bit = 2

Moderately = 3

Quite a bit = 4

Extremely = 5

These questions are about how you feel and how things have been with you **during the past 4 weeks**. For each question, please give the one answer that comes closest to the way you have been feeling.

How much of the time during the **past 4 weeks** . . .

(Circle One Number on Each Line)

	All of the Time	Most of the Time	A Good Bit of the Time	Some of the Time	A Little of the Time	None of the Time
23. Did you feel full of pep?	1	2	3	4	5	6
24. Have you been a very nervous person?	1	2	3	4	5	6
25. Have you felt so down in the dumps that nothing could cheer you up?	1	2	3	4	5	6
26. Have you felt calm and peaceful?	1	2	3	4	5	6

Subject #: _____

27. Did you have a lot of energy?	1	2	3	4	5	6
28. Have you felt downhearted and blue?	1	2	3	4	5	6
29. Did you feel worn out?	1	2	3	4	5	6
30. Have you been a happy person?	1	2	3	4	5	6
31. Did you feel tired?	1	2	3	4	5	6

32. During the **past 4 weeks,** how much of the time has your **physical health or emotional problems** interfered with your social activities (like visiting with friends, relatives, etc.)?

(Circle One Number)

All of the time = 1

Most of the time = 2

Some of the time = 3

A little of the time = 4

None of the time = 5

Subject #: _____

How TRUE or FALSE is each of the following statements for you.

(Circle One Number on Each Line)

	Definitely True	Mostly True	Don't Know	Mostly False	Definitely False
33. I seem to get sick a little easier than other people	1	2	3	4	5
34. I am as healthy as anybody I know	1	2	3	4	5
35. I expect my health to get worse	1	2	3	4	5
36. My health is excellent	1	2	3	4	5

44. Lastly, we would like to ask you a few questions about your autoimmune condition. How would you rate the overall severity of your autoimmune condition (Sjogren's Syndrome, Rheumatoid Arthritis, Diabetes Type 1, Wegener's Granulomatosis, Scleroderma, Dermatomyositis, Polymyositis, Systemic Lupus)?

1=no problem
2=mild problem
3=moderate problem
4=severe problem

Subject #: _____

45. (Examiner, **circle** the diagnosis below and **administer** the appropriate Disease Severity Scale included in the **Appendix**).

1=Sjogren's Syndrome
2=Rheumatoid Arthritis
3=Diabetes Type 1

4=Wegener's Granulomatosis
5=Scleroderma
6=Dermatomyositis
7=Polymysitis
8=Systemic Lupus
9=Mixed connective tissue disease

CLOSING:
That is all the information we need. Do you have any questions or comments about the study?

_____

_____

_____

The University of (Utah) thanks you for your time (today/this evening).

END TIME __ __ : __ __ am
pm

Subject #: _____

APPENDIX: Sjogren's Syndrome/Wegener's Granulomatosis

Sjogren's Questionnaire 1

Please consider the past 2 weeks when rating the following:

Sore Eyes

0 1 2 3 4 5 6 7

0=No problem at all 7=As bad as imaginable

Eye Irritation

0 1 2 3 4 5 6 7

0=No problem at all 7=As bad as imaginable

Poor Vision

0 1 2 3 4 5 6 7

0=No problem at all 7=As bad as imaginable

Difficulty Eating

0 1 2 3 4 5 6 7

0=No problem at all 7=As bad as imaginable

Dry Throat

0 1 2 3 4 5 6 7

0=No problem at all 7=As bad as imaginable

Bad Breath

Subject #: _____

0 1 2 3 4 5 6 7
0=No problem at all 7=As bad as imaginable

Wetting Mouth (carried fluid during day/night)
0 1 2 3 4 5 6 7
0=No problem at all 7=As bad as imaginable

Oral Problems
0 1 2 3 4 5 6 7
0=No problem at all 7=As bad as imaginable

Vaginal Dryness
0 1 2 3 4 5 6 7
0=No problem at all 7=As bad as imaginable

Skin Dryness
0 1 2 3 4 5 6 7
0=No problem at all 7=As bad as imaginable

(SSI Sicca Symptoms Inventory—Short Form, Bowman et al., 2003)

Sjogren's Questionnaire 2
Dryness
0 1 2 3 4 5 6 7 8 9 10

Subject #: _____

Limb Pain

0 1 2 3 4 5 6 7 8 9 10

Fatigue

0 1 2 3 4 5 6 7 8 9 10

(ESSPRI, Seror et al., 2011)

APPENDIX: Rheumatoid Arthritis

Rheumatoid Arthritis Pain Scale (RAPS) DIRECTIONS: The following items relate to pain and arthritis. For each item, choose one number from 0 (never) to 6 (always) to describe how you have felt in the last week.

	0 (Never)	1	2	3	4	5	6 (Always)
1. I would describe my pain as gnawing.	0	1	2	3	4	5	6
2. I would describe my pain as aching.	0	1	2	3	4	5	6
3. I would use the word exhausting to describe my pain.	0	1	2	3	4	5	6
4. I would describe my pain as annoying.	0	1	2	3	4	5	6
5. I am in constant pain.	0	1	2	3	4	5	6
6. I would describe my pain as rhythmic.	0	1	2	3	4	5	6
7. I have swelling of at least one joint.	0	1	2	3	4	5	6
8. I have morning stiffness of one hour or more.	0	1	2	3	4	5	6
9. I have pain on motion of at least one joint.	0	1	2	3	4	5	6
10. I cannot perform all the everyday tasks I normally would because of pain.	0	1	2	3	4	5	6

11. Pain interferes with my sleep.	0	1	2	3	4	5	6
12. I cannot decrease my pain by using methods other than taking extra medication.	0	1	2	3	4	5	6
13. I would describe my pain as burning.	0	1	2	3	4	5	6
14. I find that I guard my joints to reduce pain.	0	1	2	3	4	5	6
15. I brace myself because of the pain.	0	1	2	3	4	5	6
16. My pain is throbbing in nature.	0	1	2	3	4	5	6
17. I would describe my pain as sharp.	0	1	2	3	4	5	6
18. I would say my pain is severe.	0	1	2	3	4	5	6
19. I feel stiffness in my joints after rest.	0	1	2	3	4	5	6
20. My joints feel hot.	0	1	2	3	4	5	6
21. I feel anxious because of pain.	0	1	2	3	4	5	6
22. I would describe my pain as tingling.	0	1	2	3	4	5	6
23. I feel my pain is uncontrollable.	0	1	2	3	4	5	6
24. I feel helpless to control my pain.	0	1	2	3	4	5	6

When looking at the scale below, overall I would rate my pain as:

0 1 2 3 4 5 6 7 8 9 10

NONE SEVERE

APPENDIX: Diabetes Type 1

ADDQoL 19

If I did not have diabetes…

Subject #: _____

…I would enjoy my leisure activities

very much more much more a little more the same less

…My working life would be

very much better much better a little better the same worse

…Local or long distance journeys would be

Very much easier much easier a little easier the same more difficult

…My holidays would be

very much better much better a little better the same worse

…Physically I could do

very much more much more a little more the same less

…My family life would be

very much better much better a little better the same worse

…My friendships and social life would be

very much better much better a little better the same worse

…My closest personal relationship would be

very much better much better a little better the same worse

…My sex life would be

very much better much better a little better the same worse

…My physical appearance would be

very much better much better a little better the same worse

…My self confidence would be

very much greater much greater a little greater the same less

…My motivation would be

very much greater much greater a little greater the same less

…The way people in general react to me would be

very much better much better a little better the same worse

…My feelings about the future (e.g. worries, hopes) would be

very much better much better a little better the same worse

…My financial situation would be

very much better much better a little better the same worse

Subject #: _____

…My living conditions would be

very much better much better a little better the same worse

…I would have to depend on others when I do not want to

Very much less much less a little less the same more

…My freedom to eat as I wish would be

very much greater much greater a little greater the same less

…My freedom to drink as I wish (e.g. fruit juice, alcohol, sweetened hot and cold drinks) would be

very much greater much greater a little greater the same less

APPENDIX: Systemic Lupus

LupusQoL Questionnaire The following questionnaire is designed to find out how SLE affects your life. **Read** each statement and then circle theresponse, which is **closest to how you feel**. Please try to answer all the questions as honestly as you can.

How often over the last 4 weeks

1. Because of my Lupus I need help to do heavy physical jobs such as digging the garden, painting and/or decorating, moving furniture

All of the time most of the time a good bit of the time occasionally never

2. Because of my Lupus I need help to do moderate physical jobs such as vacuuming, ironing, shopping, cleaning the bathroom

All of the time most of the time a good bit of the time occasionally never

3. Because of my Lupus I need help to do light physical jobs such as cooking/preparing meals, opening jars, dusting, combing my hair or attending to personal hygiene

All of the time most of the time a good bit of the time occasionally never

4. Because of my Lupus I am unable to perform everyday tasks such as my job, childcare, housework as well as I would like to

All of the time most of the time a good bit of the time occasionally never

5. Because of my Lupus I have difficulty climbing stairs

All of the time most of the time a good bit of the time occasionally never

Subject #: _____

6. Because of my Lupus I have lost some independence and am reliant on others
All of the time most of the time a good bit of the time occasionally never
7. I have to do things at a slower pace because of my Lupus
All of the time most of the time a good bit of the time occasionally never
8. Because of my Lupus my sleep pattern is disturbed
All of the time most of the time a good bit of the time occasionally never

How often over the last 4 weeks
9. I am prevented from performing activities the way I would like to because of pain due to Lupus
All of the time most of the time a good bit of the time occasionally never
10. Because of my Lupus, the pain I experience interferes with the quality of my sleep
All of the time most of the time a good bit of the time occasionally never
11. The pain due to my Lupus is so severe that it limits my mobility
All of the time most of the time a good bit of the time occasionally never
12. Because of my Lupus I avoid planning to attend events in the future
All of the time most of the time a good bit of the time occasionally never
13. Because of the unpredictability of my Lupus I am unable to organise my life efficiently
All of the time most of the time a good bit of the time occasionally never
14. My Lupus varies from day to day which makes it difficult for me to commit myself to social arrangements
All of the time most of the time a good bit of the time occasionally never
15. Because of the pain I experience due to Lupus I am less interested in a sexual relationship
All of the time most of the time a good bit of the time occasionally never not applicable
16. Because of my Lupus I am not interested in sex
All of the time most of the time a good bit of the time occasionally never not applicable
17. I am concerned that my Lupus is stressful for those who are close to me
All of the time most of the time a good bit of the time occasionally never
18. Because of my Lupus I am concerned that I cause worry to those who are close to me
All of the time most of the time a good bit of the time occasionally never
19. Because of my Lupus I feel that I am a burden to my friends and/or family
All of the time most of the time a good bit of the time occasionally never

Subject #: _____

Over the past 4 weeks I have found my Lupus makes me

20. Resentful

All of the time most of the time a good bit of the time occasionally never

21. So fed up nothing can cheer me up

All of the time most of the time a good bit of the time occasionally never

22. Sad

All of the time most of the time a good bit of the time occasionally never

23. Anxious

All of the time most of the time a good bit of the time occasionally never

24. Worried

All of the time most of the time a good bit of the time occasionally never

25. Lacking in self-confidence

All of the time most of the time a good bit of the time occasionally never

How often over the past 4 weeks

26. My physical appearance due to Lupus interferes with my enjoyment of life

All of the time most of the time a good bit of the time occasionally never

27. Because of my Lupus, my appearance (e.g. rash, weight gain/loss) makes me avoid social situations

All of the time most of the time a good bit of the time occasionally never

28. Lupus related skin rashes make me feel less attractive

All of the time most of the time a good bit of the time occasionally never

How often over the past 4 weeks

29. The hair loss I have experienced because of my Lupus makes me feel less attractive

All of the time most of the time a good bit of the time occasionally never not applicable

30. The weight gain I have experienced because of my Lupus treatment makes me feel less attractive

All of the time most of the time a good bit of the time occasionally never not applicable

31. Because of my Lupus I cannot concentrate for long periods of time

All of the time most of the time a good bit of the time occasionally never

32. Because of my Lupus I feel worn out and sluggish

All of the time most of the time a good bit of the time occasionally never

33. Because of my Lupus I need to have early nights

All of the time most of the time a good bit of the time occasionally never

34. Because of my Lupus I am often exhausted in the morning

All of the time most of the time a good bit of the time occasionally never

APPENDIX: Scleroderma, Dermatomyositis, Polymyositis, Wegener's Granulomatosis

World Health Organization Disability Assessment Schedule II Phase 2 Field Trials – Health Services Research
36-Item Self-Administered Version

H1 How do you rate your overall health in the past 30 days?

Very good Good Moderate Bad Very Bad

This questionnaire asks about difficulties due to health conditions. Health conditions include diseases or illnesses, other health problems that may be short or long lasting, injuries, mental or emotional problems, and problems with alcohol or drugs.Think back over the last 30 days and answer these questions thinking about how much difficulty you had doing the following activities. For each question, please circle only one response.

In the last 30 days, how much difficulty did you have in:

Understanding and communicating

D1.1 Concentrating on doing something for ten minutes?

None Mild Moderate Severe Extreme/Cannot Do

D1.2 Remembering to do important things?

None Mild Moderate Severe Extreme/Cannot Do

D1.3 Analyzing and finding solutions to problems in day to day life?
None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D1.4 Learning a new task, for example, learning how to get to a new place?
None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D1.5 Generally understanding what people say?
None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D1.6 Starting and maintaining a conversation?
None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

Getting Around
D2.1 Standing for long periods such as 30 minutes?
None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D2.2 Standing up from sitting down?
None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D2.3 Moving around inside your home?
None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D2.4 Getting out of your home?
None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D2.5 Walking a long distance such as a kilometer (or equivalent)?
None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

Subject #: _____

In the last 30 days, how much difficulty did you have in:

Self Care

D3.1 Washing your whole body?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D3.2 Getting dressed?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D3.3 Eating?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D3.4 Staying by yourself for a few days?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

Getting along with people

D4.1 Dealing with people you do not know?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D4.2 Maintaining a friendship?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D4.3 Getting along with people who are close to you?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D4.4 Making new friends?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D4.5 Sexual activities?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

Subject #: _____

Life activities

D5.1 Taking care of your household responsibilities?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D5.2 Doing most important household tasks well?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D5.3 Getting all the household work done that you needed to do?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D5.4 Getting your household work done as quickly as needed?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

IF YOU WORK (PAID, NON-PAID, SELF EMPLOYED) OR GO TO SCHOOL, COMPLETE QUESTIONS D5.5-D5.8 BELOW. OTHERWISE, SKIP TO D6.1 AT THE TOP OF THE NEXT PAGE.

In the last 30 days, how much difficulty did you have in:

D5.5 Your day to day work/school?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D5.6 Doing your most important work/school tasks well?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D5.7 Getting all the work done that you need to do?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D5.8 Getting your work done as quickly as needed?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

Subject #: _____

In the last 30 days:

Participation in Society

D6.1 How much of a problem did you have in joining in community activities (for example, festivities, religious or other activities) in the same way as anyone else can?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D6.2 How much of a problem did you have because of barriers or hindrances in the world around you?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D6.3 How much of a problem did you have living with dignity because of the attitudes and actions of others?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D6.4 How much time did you spend on your health condition, or its consequences?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D6.5 How much have you been emotionally affected by your health condition?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D6.6 How much has your health been a drain on the financial resources of you or your family?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D6.7 How much of a problem did your family have because of your health problems?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

D6.8 How much of a problem did you have in doing things by yourself for relaxation or pleasure?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

H2 Overall, how much did these difficulties interfere with your life?

None **Mild** **Moderate** **Severe** **Extreme/Cannot Do**

H3 Overall, in the past 30 days, how many days were these difficulties present? RECORD NUMBER OF DAYS ___/___

H4 In the past 30 days, for how many days were you totally unable to carry out your usual activities or work because of any health condition? RECORD NUMBER OF DAYS ___/___

H5: In the past 30 days, not counting the days that you were totally unable, for how many days did you cut back or reduce your usual activities or work because of any health condition? RECORD NUMBER OF DAYS___/___

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