Validation of a Telephone Screening Tool for Spasmodic Dysphonia and Vocal Fold Tremor

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Summary: Objectives/Hypothesis. The objective of this study was to ascertain whether clinicians can reliably distinguish between spasmodic dysphonia (SD)/vocal tremor and other voice disorders by telephone, despite this modality's limited frequency response.

Study Design. Randomized, single-blinded, and prospective study.

Methods. Voice-disordered patients with (n = 22) and without (n = 17) SD and/or vocal tremor recorded standardized utterances via landline telephone. A laryngologist and two speech-language pathologists blinded to the diagnoses rated each recording as "yes" or "no" to "SD or tremor present?," and if "yes" categorized into adductor, abductor, tremor only, or adductor with tremor subtypes. Twenty-one recordings were presented twice at random so intrarater reliability could be assessed. All ratings were compared with gold standard diagnosis by a second laryngologist who performed a full examination, including videostroboscopy, on each patient.

Results. For the comparison "SD or tremor" yes versus no, sensitivity, specificity, positive predictive value, and negative predictive value are 90%, 95%, 96%, and 89%, respectively. Interrater reliability (Cohen kappa) compared with the gold standard ranged from 0.70 to 0.93 (substantial to almost perfect agreement). Cronbach alpha among three raters was 0.90 for this comparison. Intrarater reliability (number matched/number inspected) was very high, ranging from 0.97 to 1.0. Comparing gold standard and telephone rating of SD/tremor subtypes, kappa ranged from 0.48 to 0.60 (moderate agreement). Cronbach alpha among three raters was 0.88 for this comparison. Intrarater reliability ranged from 0.84 to 0.97.

Conclusions. SD and tremor can be reliably distinguished from other voice disorders over the telephone.

Key Words: Spasmodic dysphonia–Vocal fold tremor–Telephone screening–Abductor spasmodic dysphonia–Adductor spasmodic dysphonia.

INTRODUCTION

Spasmodic dysphonia (SD) is an idiopathic focal dystonia affecting the intrinsic musculature of the larynx.[1] Classified as a rare disease by the National Institutes of Health, SD has a prevalence of roughly 14 per 100 000 and predominantly affects women (2.5:1); peak incidence is from ages 30 to 50.[2] Patients experience vocal symptoms that may range from occasional difficulty with one or two words to sustained inability to phonate, entirely compromising fluent speech, yet other modalities of phonation—including singing—are spared. Nonphonatory functions, such as swallowing, are likewise unaffected.[3]

Little is known about the natural history of SD, making prognostication difficult.[4] There is no definitive treatment of this chronic disease; surgical and chemical denervation of the affected muscles can temporarily palliate symptoms but do not restore baseline function. Although once considered a psychogenic phenomenon, SD's resistance to speech therapy and psychiatric treatment, which can be effective for muscle tension (muscle tension dysphonia [MTD]), true psychogenic dysphonias, and other dysphonias, suggests otherwise. Although some recent inroads have been made, particularly in uncovering potential genetic links, the etiology of many SD cases remains obscure.

Furthermore, diagnosis of SD—like that of many dystonias—requires subjective evaluation by experienced clinicians, often a team comprised of laryngologist, speech-language pathologist (SLP), and neurologist; there is no systematic tool that can reliably distinguish between SD and other speech pathologies. Moreover, even board-certified otolaryngologists may not receive sufficient voice-specific training to make the diagnosis of SD.

Indeed, as recently as the 1970s, patients have been treated in inpatient psychiatric facilities for psychogenic voice disorders. Previous work at the Emory Voice Center has demonstrated that even today there is an increased risk of mood disorders in voice-disordered patients over time.[5] Patients and physicians without access to specialized voice centers may find themselves frustrated by their failure to find a diagnosis or develop a treatment plan. Only a few such centers exist throughout the United States, suggesting that a large proportion of the SD population may be undiagnosed or undertreated.

Most Americans do have access to a telephone, however. Because SD may have very prominent audible characteristics, some centers already offer telephone screening services. However, the utility of this screening modality has not been studied. The standard telephone system was designed to balance intelligibility while minimizing bandwidth use, and to that end, it sacrifices all frequencies above about 3800 Hz and below about 250 Hz. Although this preserves the range containing cardinal vowel formants, some consonants contain much higher

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Coordinator: optionally, practice tasks with patient once. dial xxx-xxx-xxxx from a landline and wait for the prompt. Say "TS-SD patient number _____", filling in the appropriate number; then give the phone to the participant.

Please read the following paragraph aloud:
The rainbow is a division of white light into many beautiful colors. These take the shape of a long round arch with its path high above and its two ends apparently beyond the horizon.

Please read the following sentences aloud:
A dog dug a new bone. We mow our lawn all year. The waves were rolling along.

Please count from eighty (80) to eighty-five (85). Next, hold out an "AHHHH" for five seconds. Then hold out an "EEEEE" for five seconds.

Please read the following sentences aloud:
Keep Tom at the party. Sam usually takes coffee with sugar. Put the cookies in the tin.

Please count from sixty (60) to sixty-five (65).

FIGURE 1. Patient telephone script.

frequencies. The phonemic signs of SD might not be perceptible under such conditions.

Chhetri et al[6] examined the reliability of SD severity assessment comparing telephone-filtered to high-quality digital samples; however, they did not employ control subjects to address the question of distinguishing SD from non-SD. Furthermore, their methodology used the same patient recording for both conditions, resulting in a significant likelihood of bias on the part of their raters. They also found decreased agreement among raters for telephone-filtered samples compared to unfiltered ones, despite claiming that "speech parameters that are important in the perceptual evaluation of ADSD, such as frequency excursions and motor timing of speech, are transmitted well by the telephone" and offering little explanation for the discrepancy.

The aim of this project, therefore, was to elucidate whether SD and the related diagnosis vocal tremor[7] can be distinguished by expert listeners via telephone, despite its inherent limitations.

MATERIALS AND METHODS

This study was carried out under Emory Institutional Review Board (IRB) protocol number IRB00054821, Outcomes in Laryngology Patients.

Participants with SD and/or vocal tremor were recruited from the senior author's weekly in-office laryngeal injection clinic. Inclusion criteria included "gold standard" diagnosis (incorporating full history, vocal characteristics, and stroboscopic laryngeal examination)[8] of abductor (AB) or adductor (AD) SD with or without tremor, or tremor alone, and willingness to participate in the study. Patients were excluded if they carried an additional voice-related diagnosis or if they declined participation. Control patients were recruited from the senior author's regular clinic. Inclusion criteria included diagnosis of a non-SD or tremor voice disorder, including, but not limited to, MTD, unilateral or bilateral vocal fold paralysis, vocal fold polyp or scar, and Reinke's edema. Control patients were excluded if they were unwilling to participate in the study. Verbal informed consent was obtained from each participant before recordings were made.

Recordings were made on one of the landline telephones located in quiet offices in the Emory Voice Center. Participants were seated in a comfortable posture and instructed to look over the patient script (Figure 1). Any questions were addressed, and participants were given the option to practice each task. Each participant was assigned a sequential patient identification number, which was not associated with any personally identifiable information. The coordinator dialed into an electronic voice messaging system that automatically produces an mp3 recording (codec: mpga; channels: mono; sample rate: 16 kHz; bit rate: 16 kB/s) of each message and recorded the introduction as noted in Figure 1.

Participants then recorded a standardized series of stimuli identical to those that would be collected in a standard new patient examination in the Voice Center clinic, to wit: a 34-word excerpt from the Rainbow Passage ("The rainbow … " through "…beyond the horizon"), three sentences phonemically loaded with voiced consonant-to-vowel transitions, the integer sequence from 80 to 85 inclusive, the vowels /a/ and /i/ sustained for 5 seconds each, three sentences phonemically loaded with aspirated/unvoiced consonant-to-vowel transitions, and the integer sequence from 60 to 65 inclusive (Figure 1). Immediately after concluding the recording process for any participant, the senior author recorded the gold standard diagnosis on a separate form (Figure 2).

Patient ID: _____

This patient has some form of SD and/or vocal tremor _____ YES _____ NO

If YES, circle the best diagnosis:
ADductor spasmodic dysphonia
ABductor spasmodic dysphonia
Tremor only
ADductor SD with tremor

FIGURE 2. "Gold standard" diagnosis form.