Oropharyngeal Variant of Spasmodic Dysphonia: A Report of Two Cases
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Abstract

Purpose: To discuss a previously undescribed focal oropharyngeal dystonia specific to voice-related tasks

Design: Retrospective chart review of two patients with oropharyngeal variant of spasmodic dysphonia (OVSD)

Results: Both patients were initially misdiagnosed with adductor spasmodic dysphonia and failed standard treatment with botulinum toxin (Botox). A novel treatment was designed, directing Botox injections into the muscles involved in spasmodic valving at the oropharyngeal level. Both patients showed significant improvement in voice post-injection and have experienced only mild dysphagia. These patients have maintained favorable results with repeat injections at 6-12 week intervals.

Conclusions: OVSD is a previously undescribed entity. A novel method of Botox injections into the involved muscles results in improvement in voice without significant dysphagia.

Introduction

Adductor Spasmodic Dysphonia (ASDS) is a task-specific focal laryngeal dystonia that is characterized by hyperadduction of the vocal folds resulting in an effortful, strained-voiced quality. Abductor SD is a more rare form of SD that involves the laryngeal abductor muscle hyperactivity and results in breathy results. Mixed SD has been reported, involving both abductor and adductor muscles of the larynx. Rare variants of these forms of SD have been reported as well, including adductor respiratory dystonia in which the hyperadductory muscle activity occurs during respiration, and singer’s dystonia where the adductor spasms occur exclusively with the singing voice, sparing the speaking voice. We report two cases of a previously undescribed entity, oropharyngeal variant of spasmodic dysphonia (OVSD), as well as a novel treatment option for this condition.

Case #1

KIA is a 56 year-old male with a history of acute onset of dysphonia in August of 2001 after an upper respiratory infection presented 2 months after onset of symptoms. Physical exam revealed a strained-voiced quality, worse with vowels and nasals, but noted across all speech-related tasks. Flexible laryngoscopy revealed hyperadduction of the supraglottal and vocal folds. After voice therapy failed, the patient underwent EMG guided injection of Botox into both thyroarytenoid/lateral cricoarytenoid (TA/LCA) complexes. He developed severe post injection breathy voice with minimal reduction in spasms which returned as soon as the breathiness resolved. Flexible laryngoscopy after Botulin injection revealed persistent spasms involving the oropharyngeal musculature with medial contraction of the pharyngeal walls and posterior displacement of the base of tongue. Further review of the patient’s voice and flexible laryngoscopy from his original presentation showed spasmodic closure at the oropharyngeal level and a cul-de-sac resonance of the voice with the spasms. A diagnosis of Adductor SD with oropharyngeal spasms was made.

Because treatment of oropharyngeal spasms would likely result in severe dysphagia, treatment efforts focused on the larynx. Additional treatment with Botox into the false vocal folds and strap muscle complex in an effort to reduce breathiness resulted in slightly better voice with less breathiness, but the patient was still dissatisfied with voice results. A second opinion from another laryngologist confirmed the diagnosis of Adductor SD with focal dystonia of the tongue base. Consecutive treatments of unilateral injection of 8 units into the TA/LCA complex, as well as 25 units in the submental region resulted in no improvement and the patient returned to our clinic to resume treatment.

We devised an injection strategy of directing Botox superficially at the pharyngoepiglottic fold (PEF), in an attempt to reduce posterior tongue base movement. Anatomically, this approximated the location of the styloglossus, a tongue base retractor that is not an intrinsic muscle of the tongue. The dose was started at 5 units bilaterally and increased in 2.5 unit increments. Significant improvement in the patient’s voice without breathiness was noted as the dose was increased. Eventually, dysphagia for solids was encountered at 17.5 units injected on each side. The dosage was reduced to 15 units superficially in each PEF which maximized vocal benefits and resulted in mild dysphagia. He has continued these treatments every 2-3 months for the past 4.5 years with consistently good results.

Case #2

SC is a 67 year old with an eight-year history of dysphonia characterized by pitch and voice breaks, reduced projection, increased vocal effort and strain, and vocal fatigue. She had been previously diagnosed with adductor spasmodic dysphonia but failed to benefit from prior botulinum injections into the TA/LCA muscles. Voice therapy also failed to improve her voice. Flexible fiberoptic exam revealed spasms involving the oropharyngeal musculature with medial contraction of the pharyngeal walls and posterior displacement of the base of tongue that were most prominent with vowels and nasals; glottic and supraglottic compression were present to a lesser degree. There was no tremor.

Having diagnosed and treated Patient 1 several years earlier, a diagnosis of OVSD was suspected and the patient received a per-oral injection with Botox superficially in the region of the pharyngoepiglottic folds (PEF) (10 units each) as well as 5 units in each false vocal fold. She reported some improvement in her voice, and the dose of the PEF injections was serially increased to 20 units bilaterally, while the FVF injections were withheld. Her voice continued to improve as the PEF injections increased in dosing. Significant dysphagia developed at a dose of 23 units bilaterally, at which time the injection was reduced back down to 20 units bilaterally. She continues to get vocal improvement with PEF injections; FVF injections have been used in 50% of treatments as well, and seem to have added benefit, in contrast to patient #1. She has had a total of 17 injections over a 29-month period (mean interval between injections of 1.7 months).

Her initial VHI-10 was 40, with improvement to 10 after successful treatment. She rates her best voice at 75% of normal, which usually occurs within 5 days of the injection and is maintained for 5 weeks. She returns at 6-week intervals for reinjection. She continues to have dysphagia for solids after injection (food “sticking” at base of tongue), but it is well tolerated with appropriate dietary changes.

Discussion

Since the first report of the use of Botox for Adductor SD in the mid 1980s, variants of SD, as well as alternate treatment strategies have continued to emerge. Botulinum toxin has been injected into the TA, LCA, PCA, cricothyroid, interarytenoid, false vocal folds, and strap muscle complex for a variety of SD types as well as for essential tremor. Refinement of dosages has helped to maximize vocal improvement while also minimizing side effects such as breathy voice, dysphagia and breathing difficulties. Few very descriptions for the use of Botox in the oropharynx have been reported, principally because of the risk of significant dysphagia. Our report demonstrates that oropharyngeal spasmodic activity can be reduced while still maintaining acceptable swallowing function. Although both patients experienced some degree of dysphagia for solids with treatment, the benefits of the voice improvement outweighed the side effects. Both patients were able to manage the dysphagia by dietary modifications and have continued to follow up for several years to receive repeat injections.

Conclusion

OVSD is a previously undescribed entity, involving oropharyngeal spasms during connected speech, resulting in distinctive spasmodic breaks at the oropharyngeal level. A novel treatment involving Botox injections into the muscles involved in spasmodic valving at the oropharyngeal level results in improved voice, with only mild dysphagia.

References