SELECTIVE LARYNGEAL ADDUCTOR DENERVATION-REINNERVATION: A NEW SURGICAL TREATMENT FOR ADDUCTOR SPASMODIC DYSPHONIA

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During the past decade, botulinum toxin (Botox) has emerged as the accepted treatment for adductor spasmodic dysphonia (ASD). This therapy, which produces bilateral weakness of the thyroarytenoid muscle, undoubtedly produces physiologic effects that are beneficial to patients with ASD. However, it also has important limitations, including the need for repeated injections, the unpredictable relationship between dosage and response, and the possibility of short-term swallowing and voice problems. In this study, we will report our preliminary experience with a new surgical treatment for ASD. In this new procedure, the adductor branch of the recurrent laryngeal nerve is selectively denervated bilaterally, and its distal nerve stumps are reinnervated with branches of the ansa cervicalis nerve. Each of the patients was followed for at least 12 months; the median follow-up is 36 months. The outcome of the operation in 21 consecutive patients is reported. Nineteen of the 21 patients were judged to have an overall severity of dysphonia that was "absent to mild" following the procedure. Only 1 patient underwent further treatment with Botox postoperatively. The implications of this new procedure for ASD are discussed.

KEY WORDS — adductor spasmodic dysphonia, laryngeal reinnervation, laryngeal surgery, thyroarytenoid muscle.

INTRODUCTION

Selective paralysis of the thyroarytenoid (TA) muscle produces favorable physiologic changes and dramatic improvement in the voices of patients with adductor spasmodic dysphonia (ASD). This has been demonstrated by the use of botulinum toxin (Botox) injected into the TA muscle as the standard treatment of choice for ASD. This report describes a new surgical treatment that produces TA and lateral cricoarytenoid (LCA) muscle paralysis in patients with ASD by selectively denervating the recurrent laryngeal nerve (RLN) branches to these muscles. In order to prevent unwanted reinnervation by RLN efferents and preserve muscle tone, the TA nerve branch is reinnervated with a branch of the ansa cervicalis. This procedure is a potential long-term surgical solution for patients who desire an alternative to repeated Botox injections.

Surgical treatments for ASD have been rejected because of the lack of long-term success. The first widely accepted treatment for ASD was RLN section, a procedure that was initially embraced by laryngologists.[1] In theory, the creation of a unilaterally paralyzed larynx could prevent vocal fold hyperadduction, at the likely cost of a breathy voice and other typical sequelae of unilateral vocal fold paralysis. Intermediate follow-up on 306 patients[2] showed self-reported recurrent ASD symptoms in only 10% to 15% of patients. Ninety percent of patients considered the operation satisfactory, and would recommend RLN section to others with ASD.

However, another study, by Aronson and DeSanto,[3] indicated that expert listeners judged that only 36% of patients were still improved after 3 years, and few laryngologists continue to treat ASD patients with RLN section. Failure of the procedure to provide long-term relief of dysphonia was attributed to persistent hyperadduction of the TA muscle in the remaining innervated hemilarynx. However, reinnervation by proximal RLN axon regrowth has also been reported on the basis of electromyography (EMG) data.[4]

Blitzer et al[5] introduced Botox as a treatment for ASD. Most laryngologists agree that paralysis of the TA muscle (as provided by Botox) is an effective method of reversing the halting speech and strained voice associated with ASD. Speech of ASD patients is often dysfluent, because the pulmonary expiratory pressure is frequently too low to overcome unpredictable moment-to-moment changes in intralaryngeal stiffness. This results in phonation that is difficult to sustain at normal conversational intensity lev-

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TABLE 1. PATIENTS' REASONS FOR SEEKING SURGICAL INTERVENTION

	No. of Patients
Wanted long-lasting treatment; disliked injections	8
Botox was becoming less effective	6
Botox was ineffective	2
Botox produced excessive breathiness	2
Patient had antibodies to Botox	1
Effect of Botox was too short	1
Anaphylactic reaction to Botox	1

els, and is often interrupted intermittently with voice arrests as the adductory force of the vocal folds overcomes subglottic pressure. Ludlow et al[6] reported that symptom reduction in ASD occurred when there was a reduction in TA muscle activation, and EMG recordings made following Botox administration verified the denervation of the TA muscle.[7] By paralyzing the TA muscle, Botox injection also increases airflow to normal, reduces laryngeal resistance, and produces improvements in acoustic measures, including jitter, shimmer, and signal-to-noise ratio, in ASD patients.[8,9]

The TA muscle has a profound influence on laryngeal biomechanics and voice. The muscle forms much of the bulk of the vocal fold, and therefore, its contraction has a greater impact on airway resistance than that of any other intrinsic laryngeal muscle, particularly during laryngeal adduction. Laryngeal hyperadduction is associated with forceful TA contraction, and studies have linked TA hyperactivity to symptoms in ASD.[10-12] The TA muscle also has a critical role in determining the elastic modulus of the vocal fold, which is directly related to vocal fold stiffness. An abnormally high level of this property allows the development of excessive intraglottic pressures, as seen in ASD.[13,14]

Although Botox injection produces desirable physiologic effects, it has several important limitations. The duration of Botox action is measured in months, and some patients require reinjection several times per year. Voice quality does not improve immediately upon injection, and improvement may be preceded by severe breathiness. Occasionally, patients may experience temporary dysphagia. Finally, the dose-response curve for Botox can vary greatly between individuals.

Currently, there is no ideal, long-term treatment for ASD. For patients who are satisfied with its results, Botox therapy will probably continue to be the treatment of choice in ASD. However, repeated injection is not satisfactory for every patient. For example, several of the patients in this series lived far from a medical center where Botox was available.

The procedure discussed in this report was first studied in a canine model of phonation.[15] It was found that animals did not develop high subglottic pressure with electrical overstimulation of the RLN. After further cadaver dissection, it was decided in 1993 to begin the operation in humans.

The success of previous surgical approaches for ASD has been limited, because dysphonia recurred after long-term follow-up.[3] In this study, patients have been followed longitudinally for up to 5 years to determine whether the present approach provides long-lasting relief from symptoms.

PATIENTS AND METHODS

Patients. Each of the patients had a previous diagnosis of spasmodic dysphonia that was verified by a speech scientist (B.R.G.) and the senior laryngologist (G.S.B.). All but 1 of the patients had undergone at least 1 previous injection with Botox. Patients were excluded from the series if there was less than 1 year of follow-up after the surgery or if they were unavailable for postoperative assessment. Twenty-one patients were selected who fit the criteria and form the basis for this report. Ten of the patients were male, 11 female. The median follow-up period was 36 months; the mean follow-up period was 31.4 months (range 12 to 68 months). The patients' reasons for seeking surgical intervention are listed in Table 1.

Surgical Technique. The patient was intubated with a size 6 or smaller endotracheal tube, and the neck was explored via a low transverse incision. The strap muscles were retracted laterally to expose the thyroid lamina. The inferior pharyngeal constrictor muscle was divided, if necessary, to gain access to the posteroinferior portion of the lamina. The external branch of the superior laryngeal nerve was identified and preserved.

An approximately 14 × 18-mm, inferiorly based window was made in the thyroid lamina as a cartilaginous flap. The superior border was made parallel to the lower border of the thyroid lamina, midway between the upper and lower borders of the thyroid lamina. The anterior edge of the window was immediately anterior to the inferior tubercle of the thyroid lamina. The posterior border was made just anterior to the inferior cornu. The inner perichondrium was incised and the intralaryngeal musculature gently dissected with a hemostat. The distal portion of the anterior branch of the RLN was identified and followed distally. The identity of the terminal branch was verified by stimulating the nerve with a 0.5-mA DC stimulation from a portable nerve stimulator.

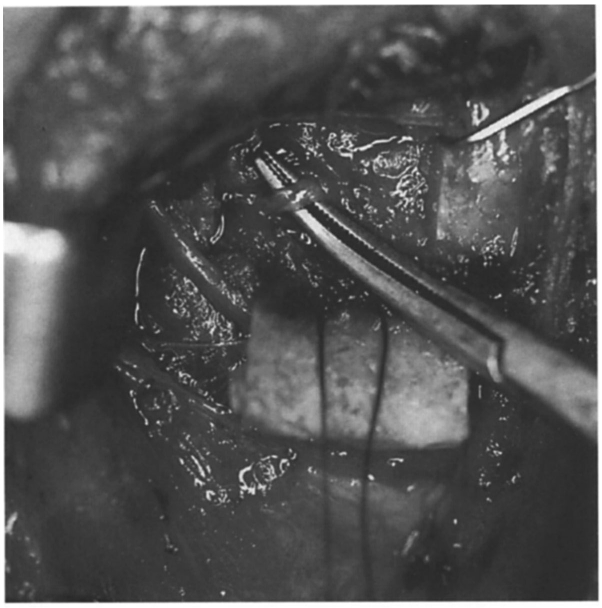

Fig 1. Left thyroarytenoid nerve is exposed through inferiorly based window in thyroid lamina.

Fig 2. Left thyroarytenoid nerve following anastomosis of ansa cervicalis branch to distal branch of left thyroarytenoid nerve.

When available in the later cases in the series, evoked EMG was performed by using an electrode endotracheal tube while stimulating the TA nerve branch. This technique has been described previously by Mermelstein et al.[16]

The TA branch is located beneath the perichondrium (Fig 1). Encased in fat, it typically runs from the posteroinferior portion of the window anterosuperiorly. It occasionally lies in a deeper plane beneath the lateral fibers of the TA muscle. The TA branch of the RLN was ligated 3 mm from its termination and sectioned with a scalpel prior to its arborization into multiple branches within the muscle. The ansa cervicalis nerve was identified and an appropriately sized nerve branch selected. Generally, the sternohyoid branch of the nerve is appropriately sized for TA reinnervation. The ansa was sutured to the TA nerve with a series of interrupted sutures of 10-0 nylon (Fig 2). The proximal stump of the TA nerve branch was ligated with a 2-0 silk suture and sutured outside the cartilage window to the posterior lamina. A 2 × 2-mm (approximately) piece of thyroid cartilage was removed from the window to avoid pressure-induced injury to the ansa cervicalis nerve where it entered into the thyroid lamina window. The window was replaced and sutured to the outer perichondrium with 4-0 nonabsorbable suture.

In the last 6 patients, in addition to TA denervation-reinnervation, nerve fibers destined for the LCA muscle were identified and sectioned to provide maximal reduction of adductory force. The LCA branch of the nerve was identified by following the RLN proximally until the nerve branch directed toward the LCA muscle was identified. Then the LCA branch was sectioned sharply, and there was no attempt to reinnervate the LCA branch.

The strap muscles were reapproximated in the midline. The platysma and skin were closed in layers. A flexible drain was kept in the wound overnight. The patients were observed in the hospital until they could tolerate an oral diet and any airway edema had subsided.

Expert Voice Assessment. A senior speech scientist with at least 10 years of experience evaluating voices rated each patient on the basis of 5 speech characteristics both before and after the surgery. The preoperative assessment was based on the voice findings made before treatment with Botox. For each category, the voice was rated on a scale of 1 (absent to mild), 2 (mild to moderate), or 3 (moderate to severe). The surgeon was excluded from providing the ratings.

Questionnaire. A telephone survey was used to obtain patient opinion about the outcome of the procedure. Each patient was asked a series of questions and asked to respond on a scale from 1 to 5. Patients were asked a series of questions over the telephone. Of the 21 patients in the series, 1 could not be contacted by telephone for completion of the questionnaire, but was included in the series because he was seen frequently postoperatively for more than 3 years.

RESULTS

Patient and Expert Assessment. The results of the telephone survey are listed in Table 2. Each score

TABLE 2. QUESTIONNAIRE GIVEN TO EACH PATIENT IN TELEPHONE INTERVIEW

	Average Score
I would recommend this operation to others with spasmodic dysphonia.	4.58
My speech is more fluent after the surgery.	4.53
My speech is better following surgery than following Botox.	4.50
I am not embarrassed speaking in public.	4.53
My speech is understandable.	4.53
Botox treated my spasmodic dysphonia successfully.	2.94
My speech problems with spasmodic dysphonia have not returned following surgery.	4.05

1 — strongly disagree; 5 — strongly agree.

indicated is the mean response for each question. There was strong agreement with 6 of the 7 statements indicating successful outcome following the procedure. The results of the expert assessment are summarized in Table 3. In general, patients had moderate to severe dysphonia prior to the operation and mild voice symptoms postoperatively.

Surgical Outcome and Further Treatment. No patients experienced airway compromise postoperatively, despite bilateral denervation. All patients experienced severe vocal fold bowing and breathiness in the early postoperative period. This normalized after 3 to 6 months, presumably when the ansa cervicalis reinnervation occurred. Patients were instructed to maintain a dysphagia diet in the week following the surgery.

The surgical morbidity is reported in Table 4. There was 1 serious complication of the procedure, in a patient who developed aspiration pneumonia postoperatively and required hospitalization. The degree of postoperative breathiness is not listed in Table 4, because this finding was consistent with the goal of the procedure.

The additional treatment is listed in Table 5. Only 1 patient has undergone Botox therapy postoperatively. Botox has remained effective in this individual. One patient underwent collagen injection to lessen a vocal fold tremor that was unmasked following TA denervation. Her speech was otherwise without dystonia. One patient underwent successful TA myotomy following aberrant reinnervation with the sectioned RLN. Her voice is slightly harsh but fluent, and the patient is quite satisfied. One patient continues with voice therapy to strengthen her voice.

TABLE 3. SPEECH SCIENTIST RANKINGS OF VOCAL FINDING SEVERITY BEFORE AND AFTER SURGERY

		No. of Patients		
Speech Finding		*Absent to Mild*	*Mild to Moderate*	*Moderate to Severe*
Overall severity	Before	0	8	13
of dysphonia	After	19	2	0
Strained voice	Before	0	13	8
quality	After	18	3	0
Voice breaks	Before	0	5	16
	After	19	2	0
Dysfluency	Before	0	7	14
	After	17	4	0

TABLE 4. SURGICAL MORBIDITY

	No. of Patients
Temporary aspiration (>2 wk)	2
Glottal insufficiency	2
Vocal fold granuloma	1
Wound seroma	1

DISCUSSION

The results presented in this study strongly support the need for further investigation of the role of selective bilateral RLN denervation in ASD treatment. Although the technique must be studied in a larger group of patients with objective measures of phonation, there is room for some optimism for patients seeking an alternative to Botox therapy.

The selective denervation presented in this study appears to have permanent effects similar to the short-term effects of TA Botox injection. This technique is successful, because it prevents the elevated intraglottic (and, secondarily, subglottic) pressures produced in patients with ASD. This prediction is consistent with experience in humans with TA Botox therapy.

Our preliminary results in 21 patients followed for a mean of 3 years indicate that selective bilateral RLN denervation with ansa reinnervation will be less prone to failure than total RLN section. It will take long-term follow-up in a large series of patients to verify this expectation, however. Ansa cervicalis TA reinnervation should prevent unwanted reinnervation from other proximal RLN efferents, a problem that limited the effectiveness of RLN section.[4] Experience with ansa reinnervation of the RLN for laryngeal paralysis indicates that purposeful adduction is not achieved.[17]

TABLE 5. ADDITIONAL TREATMENT

	No. of Patients
Collagen injection	1
Thyroarytenoid myotomy	1
Botulinum toxin treatment	1
Voice therapy	1

The ansa reinnervation nerve provides tone to the TA muscle and lessens the problems with breathiness that plague many Botox patients shortly after injection. Further, the preservation of the interarytenoid muscle allows posterior closure of the glottis to avoid some of the difficulties experienced with whole nerve section of the RLN.

Because of the critical role of the TA muscle in producing the elevated intralaryngeal pressures in ASD, the first 15 patients in the series underwent TA denervation alone. The senior author (G.S.B.) modified the procedure to divide some nerve fibers destined for the LCA muscle to maximize the effect of the operation. Although there is theoretic concern about loss of vocal fold adduction after denervation of the 2 strongest laryngeal adductors, there has not been loss of adduction in any of the 6 patients that have had denervation of both the LCA and TA muscles.

There are several possible problems with the selective denervation procedure proposed in this study. One potential complication is injury to proximal branches of the RLN, particularly the posterior cricoarytenoid (PCA) branch. This is unlikely to occur, because the thyroid cartilage window does not expose the PCA branch in most larynges. Also, because of the direction of the PCA nerve (directed more posteriorly), it is unlikely that a surgeon familiar with normal RLN anatomy would injure the PCA branch. Nonetheless, as the operation is somewhat difficult to perform, it should be first attempted in cadaver larynges. Although changes in vocal quality do occur following the procedure, these effects seem to be less than with Botox treatment. Most patients have normal conversational speech intensity with good inflection and no signs of breathiness. Denervation of the TA muscle reduces fine motor control of the larynx, and the pitch and loudness range of patients is mildly restricted following selective denervation. This occurs because the TA muscle functions in pitch and loudness control, and contracts forcefully during loud phonation and while singing at the higher frequencies. Swallowing problems such as aspiration occurred in 2 patients, but have not been long-lasting, because the closure and sensation of the larynx are not affected. Because the procedure creates a permanent alteration in laryngeal function, it should be performed only in patients with relatively severe symptoms.

In summary, the proposed procedure provides an alternative to Botox with sustained and beneficial effects. Only careful ongoing analysis of results, with adequate follow-up, can determine whether this promise will be fulfilled.

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