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Recurrent Postoperative Stridor Requiring Tracheostomy in a Patient with Spasmodic Dysphonia

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SPASMODIC dysphonia (SD) is a laryngeal disorder characterized by adductor or abductor dystonic spasms of the vocal cord (VC). It is usually manifested by abnormal phonation but may infrequently be associated with respiratory distress. We describe a patient with SD who developed recurrent severe postoperative stridor that required a tracheostomy.

Case Report

The patient was a 67-yr-old woman with a horseshoe kidney and bilateral uteropelvic junction obstruction who presented for bilateral pyeloplasty. Medical history included long-standing SD for which she had undergone left recurrent laryngeal nerve (RLN) sectioning and Teflon injections (DuPont, Wilmington, DE) of the left VC. She had also been receiving botulinum toxin (Botox; Allergan Pharmaceuticals, Irvine, CA) injections into the thyroaretynoid muscles every 3–4 months for many years. After a diagnostic laryngoscopy several years earlier, she developed postoperative stridor that required tracheal intubation. She had also undergone surgical dilatation of an esophageal stricture. Additional medical history included hypertension, hypercholestrolemia, and peptic ulcer disease. Her medications were simvastatin, indapamide, nifedipine, famotidine, and aspirin. On the day of surgery, she had no shortness of breath or stridor. After placement of routine monitors and sedation with midazolam, an unsuccessful attempt was made to place a thoracic epidural catheter. General anesthesia was induced with thiopental, and mask ventilation was established with ease. Succinylcholine 1.5 mg/kg was given, and the trachea was intubated atraumatically with a 6.5 cuffed endotracheal tube (ETT). There was no resistance to placement of the ETT, and the cuff was inflated with the minimum volume of air required to eliminate the leak. Anesthesia was maintained using air/oxygen mixture, isoflurane, fentanyl, and pancuronium. At the end of the procedure, after complete reversal of neuromuscular blockade with neostigmine and glycopyrrolate, the trachea was extubated. Ventilation was unobstructed, and the patient was transferred to the postanesthesia care unit with supplemental oxygen. Twenty minutes after extubation, soon after arrival in the postanesthesia care unit, the patient developed progressive stridor. She was manually ventilated with 100% oxygen using bag and mask and was treated with nebulized racemic epinephrine with little improvement. After the administration of a hypnotic and succinylcholine, the trachea was reintubated with a 6.0-mm ETT. She was mechanically ventilated, sedated with propofol and fentanyl, and received dexamethasone 4 mg intravenously every 6 h. On the morning of postoperative day 1, the trachea was extubated over a fiberoptic bronchoscope. Examination of the airway as the ETT was being removed revealed limited abduction of the right VC, a thickened immobile left VC, and 50% narrowing of the glottic opening caused by scar formation at the posterior commissure. In addition, there was subglottic stenosis and mild edema of the true and false VC. The ETT was advanced back into the trachea. Because there was a leak around the ETT upon deflation of the cuff, it was judged that an extubation attempt would be appropriate. A tube exchanger was positioned into the trachea, and the ETT was removed. The tube exchanger was left in the trachea to permit rapid reintubation if required. The patient received nebulized racemic epinephrine and inhaled corticosteroids. The patient continued to have mild stridor and hoarseness but maintained adequate gas exchange and arterial oxygen saturation with a respiratory rate of 16–20 breaths/min. The tube exchanger was removed 3 h after extubation. Thirty-six hours after the conclusion of surgery, she was transferred from the postanesthesia care unit to a step-down subacute care unit for vigilant monitoring. On postoperative day 2, the patient again developed severe inspiratory stridor resulting in arterial oxygen desaturation that required immediate reintubation of the trachea. In view of her inability to maintain a patent airway, a flap tracheostomy was

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performed on postoperative day 4. Mechanical ventilation, delivered through a Shiley #4 tracheostomy tube, was required for 48 h because of atelectasis and a right upper lobe pneumonia. After weaning from mechanical ventilation, the Shiley tube was removed, and the patient maintained easy unlabored gas exchange through the tube-free tracheostomy. She was discharged home on postoperative day 10 without respiratory distress. One month later the patient had a strong desire to have the tracheostomy closed. Airway evaluation revealed resolution of edema and a return to baseline airway dimensions. Temporary occlusion of the tracheostomy stoma was well tolerated with no respiratory distress. The tracheostomy was surgically closed, and Botox treatment was reinstituted.

Discussion

Spasmodic dysphonia is a clinical voice syndrome characterized by VC spasm occurring during speech. It may affect the thyroarytenoid muscles (adductor SD), the posterior cricoarytenoid muscles (abductor SD), or both.[1] SD is a type of dystonia affecting the intrinsic laryngeal muscles and may properly be termed focal laryngeal dystonia. Dystonias are manifested by forceful inappropriate action-induced involuntary contractions (spasm) affecting one or more muscle groups. It is believed that SD is a manifestation of defective motor control of speech involving systems of motor neurons in the pallidosubthalamic-supplementary motor area and/or the midbrain. Two thirds of the cases of SD are idiopathic, whereas one third is secondary to other disorders. Stress (physical or psychological) can precipitate or worsen SD symptoms. Associated neurologic abnormalities are present in up to 70% of SD patients and include tremors, weakness, movement abnormalities, and dystonia of other muscle groups.[2,3] Diagnosis requires detailed head and neck and neurologic examinations. Abnormal VC function is identified by fiberoptic or direct laryngoscopy, video laryngoscopy, stroboscopy, and laryngeal electromyography. In one series, 59% of SD patients exhibited narrowing of the laryngeal vestibule caused by epiglottic retraction, cuneiform fronting, and/or false vocal fold adduction. In addition, narrowing of the lower vocal tract may be caused by pharyngeal wall constriction.[4] Treatment requires a multifaceted approach. Speech therapy, psychotherapy, and biofeedback alone do not significantly improve speech in most patients but are useful as adjunctive measures.[5] Botulinum toxin (Botox), which blocks neuromuscular transmission, has been successfully used to treat the dystonic spasms of torticollis, blepharospasm, and SD. Botox acts presynaptically at nerve terminals to prevent calcium-dependent release of acetylcholine, producing, in effect, a chemical denervation.[6] Botox is injected either percutaneously through the cricothyroid membrane under electromyographic guidance or transorally into the affected laryngeal muscle(s).[7,8] Patients may experience mild difficulty in swallowing and slight aspiration transiently after the injection. Other potentially useful treatment modalities include surgical interruption of the RLN, partial laser resection of a VC, and anterior laryngoplasty. Disadvantages of RLN interruption include VC paralysis, pulmonary aspiration, and a 20–40% incidence of recurrence attributed to reinnervation.[9] VC injections (*e.g.*, Teflon) are occasionally used in selected patients with abductor SD or to treat aspiration after RLN sectioning. The VC spasm characteristic of SD typically develops during speech, affecting voice fluency without impeding respiration. There are rare reports of patients with intermittent upper airway distress associated with

Table 1. Causes of Stridor

Lesions of the supraglottic airway
Craniofacial anomalies
Macroglossia
Tumors: large tonsils and adenoids
Foreign bodies
Edema
Infections
Ludwig angina
Vincent's angina
Retropharyngeal abscess or hematoma
Peritonsillar abscess (Quincy)
Trauma
Lesions of the larynx and trachea
Intrinsic obstructive lesions
Tumors, papillomatosis, foreign body, blood
Edema: allergic reactions, angioneurotic edema, inhalation thermal injuries
Infections: supraglottitis, tracheitis, croup
Trauma: laryngeal and tracheal disruptions
Stenosis, strictures, congenital web
Functional obstruction
Dynamic airway collapse
Laryngomalacia
Tracheomalacia
Vocal cord spasm
Perioperative laryngospasm (during induction and emergence from anesthesia)
Hypocalcemia
Neurologic disease
Incomplete recurrent laryngeal nerve injury
Extrinsic compression
Tumors of the neck and mediastinum (*e.g.,* thyroid gland, lymphoma)
Hematomas after head and neck surgeries (*e.g.,* carotid endarterectomy, thyroidectomy or cervical spine surgery), traumatic
Vascular rings
Aortic aneurysms

dystonic involvement of the facial, pharyngeal, and laryngeal muscles.[10]

Our patient had primary SD complicated by laryngeal stenosis at the glottic and subglottic levels. Although her primary symptoms were voice-related, she had also been experiencing recurrent episodes of mild inspiratory stridor for several years. These symptoms are due to glottic and subglottic stenosis and paralysis and thickening of the left VC. The latter had resulted from prior RLN sectioning and subsequent Teflon injection. VC abduction was restricted by fibrosis of the posterior commissure. Stridor did not develop until 20 min after extubation and showed little improvement with racemic epinephrine. We believe that her airway obstruction resulted from VC spasm and laryngeal edema superimposed on the preexisting airway stenosis. Postoperative pain and stress may have contributed to the development of recurrent VC spasm that precipitated the intermittent accentuation of her postoperative stridor. Surgical treatment consisted of a self-sustaining flap tracheostomy that provides a patent airway through a tube-free stoma and preserves voice. The various pathologic conditions that may produce stridor are summarized in table 1.

Spasmodic dysphonia may pose multiple anesthetic challenges in the perioperative period. SD patients who have had multiple airway interventions and those with laryngeal stenosis and VC dysfunction may be at risk of developing airway-related complications. In predisposed patients, airway difficulties could be encountered at the induction of anesthesia, on emergence or postoperatively. Preoperative fiberoptic or direct laryngoscopy may be necessary to define anatomic abnormalities and estimate airway dimensions. Associated neurologic abnormalities and their anesthetic implications should be identified. The presence of laryngeal stenosis may necessitate the use of a smaller-than-usual ETT. The risk of pulmonary aspiration may be increased because of VC dysfunction caused by RLN interruption, a recent Botox injection (within 2 weeks) or other therapeutic and surgical interventions. Vigilant monitoring may be required for 24–72 h, depending on specific patient factors, because respiratory difficulties may develop after the immediate postoperative period.

In summary, we described a patient with SD and airway stenosis who developed recurrent postoperative stridor that required the performance of a tracheostomy. The clinical features and perioperative implications of this disease were discussed.

References

1. Blitzer A, Brin MF, Fahn S, Lovelace RE: Clinical and laboratory characteristics of focal laryngeal dystonia: Study of 110 cases. Laryngoscope 1988; 98:636–40

2. Pool KD, Freeman FJ, Finitzo T, Hayashi MM, Chapman SB, Devous MD Sr, Close LG, Kondraske GV, Mendelsohn D, Schaefer SD: Heterogeneity in spasmodic dysphonia: Neurologic and voice findings. Arch Neurol 1991; 48:305–9

3. Rosenfield DB, Donovan DT, Sulek M, Viswanath NS, Inbody GP, Nudelman HB: Neurologic aspects of spasmodic dysphonia. J Otolaryngol 1990; 19:231–6

4. Painter C, Watanabe H, Tsuzuki T: Laryngeal configuration of 99 dysphonic patients. Eur Arch Otorhinolaryngol 1991; 248:381–5

5. Miller RH, Woodson GE: Treatment options in spasmodic dysphonia. Otolaryngol Clin North Am 1991; 24:1227–37

6. Inagi K, Connor NP, Ford CN, Schultz E, Rodriquez AA, Bless DM, Pasic T, Heisey DM: Physiologic assessment of botulinum toxin effects in the rat larynx. Laryngoscope 1998; 108:1048–54

7. Blitzer A, Brin MF, Stewart CF: Botulinum toxin management of spasmodic dysphonia (laryngeal dystonia): A 12-year experience in more than 900 patients. Laryngoscope 1998; 108:1435–41

8. Schonweiler R, Wohlfarth K, Dengler R, Ptok M: Supraglottal injection of botulinum toxin type A in adductor type spasmodic dysphonia with both intrinsic and extrinsic hyperfunction. Laryngoscope 1998; 108:55–63

9. Weed DT, Jewett BS, Rainey C, Zealear DL, Stone RE, Ossoff RH, Netterville JL: Long-term follow-up of recurrent laryngeal nerve avulsion for the treatment of spastic dysphonia. Ann Otol Rhinol Laryngol 1996; 105:592–601

10. Zwirner P, Dresler D, Kruse E: Spasmodic laryngeal dyspnea: A rare manifestation of laryngeal dystonia. Eur Arch Otorhinolaryngol 1997; 254:242–5