Life Threatening Delayed Complication of Botulinum Toxin Injection for Treatment of Spasmodic Dysphonia

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Received January 12, 2020; Accepted May 28, 2020.

Key words: Dysphonia – Botulinum toxin – Vocal cords

Abstract: Spasmodic dysphonia is a primary task specific focal dystonia affecting the laryngeal muscles during speech. Most medical and surgical approaches to treatment of spasmodic dysphonia are aimed at the denervation of the laryngeal muscles to block symptom expression in the voice. The standard of care for the adductor form of spasmodic dysphonia is botulinum toxin chemodenervation. The common side effects of treatment with Botox are excessive breathiness and aspiration of fluids. We present the report of a delayed presentation of upper airway obstruction due to a complete vocal cords adduction requiring intubation ten days post Botox injection for the adductor form of spasmodic dysphonia. This presentation may be preceded by a change in voice, productive cough, shortness of breath, or odynophagia. We would recommend supportive treatment in an Intensive Care Unit and close liaison with the otolaryngology team for the management of this complication. Acute upper airway obstruction requiring tracheal intubation is a delayed complication of botulinum toxin administration in the adductor form of spasmodic dysphonia.

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https://doi.org/10.14712/23362936.2020.10
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Introduction
Spasmodic dysphonia (SD) is a primary task specific focal dystonia affecting the laryngeal muscles during speech (Ludlow, 2009). There are three classic types of SD: adductor spasmodic dysphonia (ADSD), abductor spasmodic dysphonia, and mixed spasmodic dysphonia. ADSD is the most common form (Simpson et al, 2016). The diagnosis is based on a careful history and examination of the glottis during a variety of laryngeal tasks (Sulica and Blitzer, 2004).

Most medical and surgical approaches to treatment of SD are aimed at the denervation of the laryngeal muscles to block symptom expression in the voice (Ludlow, 2009). The standard of care for ADSD is botulinum toxin (Botox) chemodenervation (Stachler et al., 2018). The common side effects of treatment with Botox are excessive breathiness and aspiration of fluids (Blitzer et al., 2018).

We describe a case of acute upper airway obstruction due to complete vocal cords adduction requiring intubation ten days post Botox injection for ADSD.

Case report
We present a case of a 64-year-old lady with the background of ADSD, seronegative rheumatoid arthritis, osteoarthritis and hypothyroidism.

She had been treated with triennial Botox injections for ADSD for three years. She was noted to develop mild stridor after previous two injections, but underwent a third injection. On day six, she noticed a change in voice, productive cough, shortness of breath and odynophagia. On day eight, she visited her general practitioner, who prescribed Amoxicillin for presumed respiratory infection.

She presented to the Emergency Department of a District General Hospital (without inpatient otolaryngology services) with acute stridor on day ten post injection. She was initially treated with 10 mg of nebulised adrenaline and 200 mg of intravenous hydrocortisone. Flexible nasendoscopy (FNE) was performed and showed completely adducted vocal cords with very limited motility. She underwent emergency tracheal intubation by an intensive care consultant and was transferred to the Intensive Care Unit (ICU) for ventilation.

On ICU, patient was prescribed her regular medication, proton pump inhibitor, enoxaparin, and subglottic levobupivacaine. She received nasogastric feeding. She required minimal ventilatory support on CPAP ASB with PEEP of 5 and FiO$_2$ 0.3–0.35. She tolerated endotracheal intubation with propofol and remifentanil sedation aiming RASS –1 to 0.

On day five of ICU admission, she was started on Co-Amoxiclav 1.2 g three times a day for ventilator associated pneumonia. A trial extubation on day six was unsuccessful. FNE showed minimal movement and oedema of vocal cords.

A further trial of extubation was attempted on day eight of ICU admission. She was successfully extubated to face mask CPAP.
Following assessment by physiotherapists and swallow and language therapists, patient was deemed medically fit for discharge. She was discharged home on day twelve.

**Discussion**

Different treatment options for SD are described in the literature. The vast majority are aimed at the denervation of laryngeal muscles (Ludlow, 2009).

Dedo described the recurrent laryngeal nerve (RLN) section as the treatment for ADSD in 1976. The subsequent change in voice was treated with speech therapy (Dedo, 1976). Fritzell et al. (1993) reported that this treatment had 72% recurrence rate at ten years follow-up due to the reinnervation of the RLN. Bilateral selective division of the adductor branches of the recurrent laryngeal nerves with immediate reinnervation of the distal nerve trunks with branches of the ansa cervicalis (selective denervation-reinnervation) was reported by Allegretto et al. (2003). Other surgical approaches include RLN avulsion (Nettervillee et al., 1991), laser-assisted thyroarytenoid myomectomy (Woo, 1990), and laryngeal framework surgery (Tucker, 1989).

Despite multiple available options of surgical treatment, the standard of care for ADSD remains botulinum toxin chemodenervation (Sulica, 2004; Stachler et al., 2018). The clinical effects of Botox last approximately 3 to 4 months on average, and reinjection is typically required to maintain clinical benefits. The broad success of Botox may be due to the specificity, repeatability, and reversibility of the chemodenervation (Blitzer et al., 2018). Patients undergoing long-term botulinum toxin treatment report a positive effect of this treatment in their workplace (Meyer et al., 2013).

Unwanted effects of Botox therapy may include loss of muscular volume and breathiness of voice (Simpson et al., 2016). Blitzer et al. (2018) described the following side effects of Botox injection in the ADSD population: mild breathiness on speaking, mild difficulty drinking fluids, and local pain, itching, or rash. The breathiness and dysphagia to liquids were also observed by Novakovic et al. (2011). A unilateral true vocal cord paralysis was reported (Srirompotong et al., 2006). Venkatesan et al. (2010) described bilateral abductor paralysis (BAP) as a complication of Botox administration. Retrospective analysis of 352 patients over the period of 9 years showed that 8 patients developed BAP between 1 week and 1 month post injection. Notably, all affected patients were female over the age of 50 years. The complication was attributed to the diffusion of Botox to the posterior cricoarytenoids. None of the patients were reported to require intubation; however, in one case a temporary tracheostomy was inserted (Venkatesan et al., 2010).

We present the report of a delayed presentation of upper airway obstruction post botulinum toxin administration for ADSD. This presentation may be preceded by a change in voice, productive cough, shortness of breath, or odynophagia. We would
recommend supportive treatment in ICU and close liaison with the ENT team for the management of this complication.

**Conclusion**

Acute upper airway obstruction requiring tracheal intubation is a delayed complication of botulinum toxin administration in ADSD.

**References**


