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Case Report

CO2 laser supraglottic resurfacing for non-granulomatous chronic supraglottitis in two children

Matthew M. Smith^{a,b}, Johanna Nokso-Koivisto^c, Brittany A. Leader^{a,b}, Lyndy J. Wilcox^d, Michael J. Rutter^{a,b,*}^a Division of Pediatric Otolaryngology, Cincinnati Children's Hospital Medical Center, Cincinnati, OH, USA^b Department of Otolaryngology, University of Cincinnati College of Medicine, Cincinnati, OH, USA^c Department of Otorhinolaryngology, Helsinki University Hospital and University of Helsinki, Helsinki, Finland^d Department of Otolaryngology, Vanderbilt University, Nashville, TN, USA

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ABSTRACT

Chronic epiglottitis and supraglottitis are clinical entities that present with respiratory distress and are primarily associated with autoimmune disorders, gastroesophageal reflux disease, or angioedema. First described in adults with sarcoidosis in 2010, CO2 laser epiglottis resurfacing has been effective in reducing epiglottic edema. We present two cases of adolescent males with non-granulomatous chronic supraglottitis who were successfully treated with CO2 laser supraglottic resurfacing.

1. Introduction

There is a wide range of conditions which have been proposed to cause supraglottitis including gastroesophageal reflux disease [1], infections [2,3], allergies, hereditary angioedema [4], and amyloidosis [5]. A number of rheumatologic diseases can also cause chronic supraglottitis including granulomatosis with polyangiitis [6], systemic lupus erythematosus [7], sarcoidosis [8], and chronic relapsing polychondritis. Chronic non-granulomatous supraglottitis has been defined to be chronic inflammation of the supraglottic larynx without evidence of any specific cause [9]. The occurrence of chronic non-granulomatous supraglottitis is very rare. There is limited literature describing this entity, with only modest systemic improvement on steroid therapy, and the potential for tracheotomy [9,10].

CO2 laser resurfacing of the epiglottis has previously been reported as an effective way to reduce epiglottic edema in patients with laryngeal sarcoidosis [8]. Given the clinical similarities of these two conditions, we present two separate pediatric cases of non-granulomatous chronic supraglottitis from two different institutions and two separate continents, which were successfully managed with a CO2 laser resurfacing technique.

2. Case presentations

2.1. Case 1

A 15-year-old male presented to the emergency department (ED) with a four-month history of odynophagia, dysphagia, increased snoring, dyspnea on exertion, and a ten pound weight loss. He was previously healthy without any significant past medical history. On exam, flexible laryngoscopy revealed an edematous and inflamed epiglottis. His white blood cell count (WBC) and erythrocyte sedimentation (ESR) levels were normal at 11 and 7 respectively. The only abnormal laboratory value was an elevated C reactive protein (CRP) of 0.42. He was stable with no acute breathing difficulties, so he was discharged from the ED on amoxicillin, ranitidine, and dexamethasone, with plan for close follow up. One week later, he underwent a micro-laryngoscopy and bronchoscopy (MLB) (Fig. 1) with biopsies of the epiglottis, which revealed squamous non-keratinizing epithelial mucosa with a lymphocytic component in the lamina propria, without evidence of granulomas. Repeat biopsies two weeks later were consistent with the above findings. Given this non-diagnostic result, he was subsequently evaluated by allergy and rheumatology. Both performed a thorough work up with multiple tests including angiotensin converting enzyme (ACE), complement, mannose binding lectin, lysozyme, immunoglobulin subclasses, lymphocyte subclasses, tuberculosis (TB), and

* Corresponding author. Division of Pediatric Otolaryngology, Head and Neck Surgery Cincinnati Children's Hospital Medical Center, 3333 Burnet Avenue, MLC 2018, Cincinnati, OH, 45229, USA.

E-mail address: mike.rutter@cchmc.org (M.J. Rutter).

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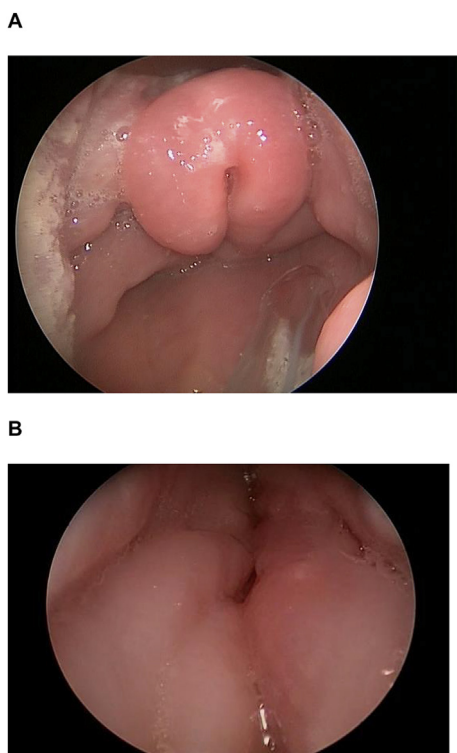


Fig. 1. Case 1 - Initial microlaryngoscopy revealing an edematous epiglottis (A) and arytenoids (B) prior to treatment.

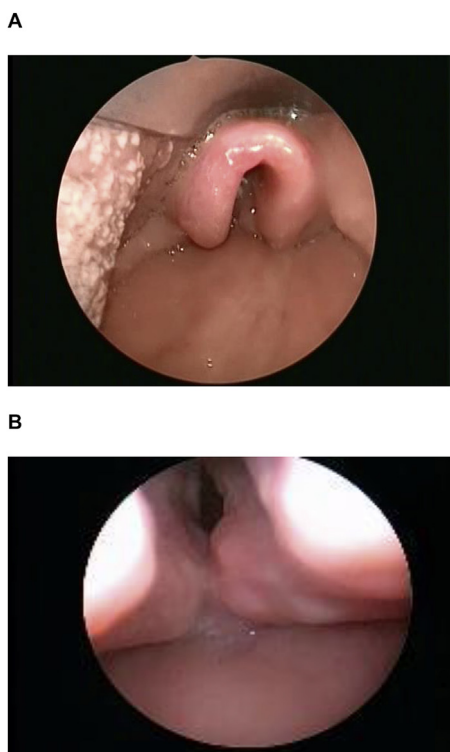


Fig. 2. Case 1 – Final microlaryngoscopy of epiglottis (A) and arytenoids (B) after CO2 laser treatments with pepper pot technique.

anti-streptolysin O, all of which were within normal limits. With no definitive diagnosis, we treated him with a CO2 laser resurfacing (pepper-pot technique) of the epiglottis and arytenoids, similar to previous patients with epiglottic edema secondary to sarcoidosis [8].

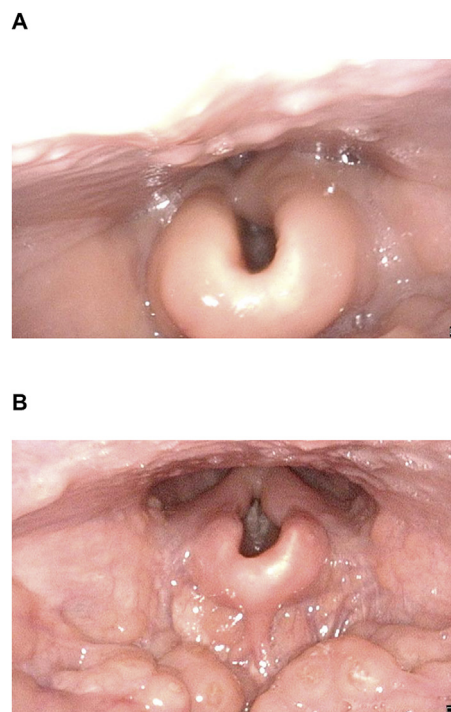


Fig. 3. Case 2 – Initial microlaryngoscopy revealing an edematous epiglottis and arytenoids prior to treatment (A) and microlaryngoscopy after CO2 laser treatments with pepper pot technique (B).

He underwent five separate CO2 laser epiglottis and arytenoid resurfacing procedures every two to three weeks, for a total of ten total procedures. For the fourth and fifth arytenoid treatments, triamcinolone was injected into the arytenoids after CO2 laser resurfacing. Sequential improvement was noted after each procedure (not complete resolution of symptoms) and he was treated with the laser techniques until there was complete resolution of symptoms and edema at the last microlaryngoscopy (Fig. 2). There were no complications with his treatment. He is currently doing well and has returned to all previous activities without shortness of breath.

2.2. Case 2

A 15-year-old presented with 3 months of progressively worsening snoring, inspiratory stridor, dyspnea on exertion, dysphagia and dysphonia. Initial laryngoscopy revealed supraglottic swelling with a thick edematous epiglottis (Fig. 3a). There were no other airway pathologic findings. Magnetic resonance imaging (MRI) demonstrated symmetric supraglottic non-specific edema. Initial blood testing was normal (WBC, ESR, CRP). The patient was admitted and treated with cefuroxime and prednisolone. After five days of observation, he was discharged with stable breathing on high-dose prednisone (40mg daily with slow taper to 7.5mg daily over a 3 week period). He returned six days later for MLB with biopsies of the epiglottis and lingual tonsils performed under local anesthesia which revealed squamous non-keratinizing epithelial mucosa with a lymphocytic component in the lamina propria, without the evidence of granulomas. Extensive evaluations similar to the first case by allergy, rheumatology, and pulmonology did not reveal a diagnosis.

Despite a lack of diagnosis, the patient was treated with a low dose of prednisolone (7.5mg daily slowly tapered to 2.5mg every other day over a four-month period) with resolution of stridor and difficulty breathing at rest. After six months, he had persistent morning dysphagia and nausea, and dyspnea during exercise and underwent an esophagogastroduodenoscopy (EGD) and MLB with biopsies. The findings were consistent with the previous biopsies. Due to continued

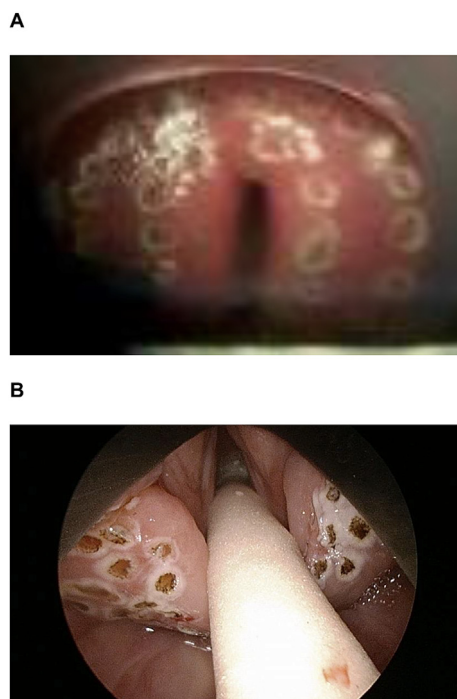


Fig. 4. CO2 laser pepper pot technique of case 1 epiglottis (A) and case 2 arytenoids (B).

dysphagia and dyspnea during exercise, the patient then underwent CO2 laser resurfacing of the epiglottis and arytenoids [8]. After the first treatment there was immediate resolution of the symptoms, but the symptoms gradually returned in 2 months. The patient was treated with a total of three separate procedures, spaced three months apart. After his final treatment there was resolution of the symptoms and the laryngeal edema had diminished (Fig. 3). He is currently doing well and has returned to all previous activities without shortness of breath.

2.3. Surgical technique

The pepper pot technique, which was popularized by Sandhu and colleagues for the treatment of chronic supraglottic edema due to sarcoidosis, was utilized [8]. Using a large Lindholm laryngoscope for exposure, the patient was suspended from the Mayo stand with the Lewy bar system. A CO2 laser attached to a microscope was used (laser settings of 5 W, depth of 1, and a dot shape) with the patient spontaneously ventilating in case 1 and a laser reinforced tube for case 2. For the epiglottis and arytenoids, the laser was used to make small, approximately 1–1.5mm, marks within the surface of the epiglottis which were separated from each other by a small amount of mucosa (1–2mm) in order to prevent scarring between adjacent sites (Fig. 4). The goal depth was to reach the cartilage, in order to allow extrusion of the edema, and cause mucosal scarring to the epiglottic cartilage to eliminate the potential space for fluid to collect. During initial treatments, clear fluid was observed to drain from the laser holes, with this being less evident with subsequent treatments.

3. Discussion

This study is the first to report the successful treatment of CO2 laser supraglottic resurfacing in two pediatric patients with chronic idiopathic supraglottitis. Each of these patients was treated at two different institutions among two different populations from two separate continents. Given the difficulty in managing this condition, and the similarity in appearance with laryngeal sarcoidosis, we utilized the pepper pot technique, described by Sandhu et al., for laryngeal edema in

sarcoidosis [8]. In each patient, CO2 laser supraglottic resurfacing resulted in resolution of supraglottic edema and symptoms.

Chronic supraglottitis and epiglottitis are rare, but when they occur a broad diagnostic work up is often indicated. Given the incidence of gastroesophageal reflux and infectious diseases as underlying risk factors, an antireflux regimen and investigation for viral, bacterial, fungal, tuberculosis, and syphilis as potential causes is warranted [1–3]. In patients where the etiology is not clearly infectious or reflux related, a multidisciplinary approach is beneficial to assist in evaluating for allergies and systemic inflammatory conditions such as sarcoidosis, granulomatosis with polyangiitis, or systemic lupus erythematosus [4–7]. If repeated biopsies and other evaluations are negative, previous literature suggests with continued observation patients will have persistent supraglottic edema, and they intermittently may require steroids, hospital admission, or even tracheotomy [9].

There is only one study to report the treatment of chronic non-granulomatous supraglottitis with local triamcinolone injections with moderate response [9]. Sandhu et al. reported ten patients with supraglottic sarcoidosis that were successfully treated with a median of two CO2 laser resurfacing (range of 1–4 treatments) with simultaneous triamcinolone injections [8]. Using this technique, our patients had resolution of their laryngeal edema without any adverse effects. Due to a lack of treatment algorithm and surgeon preference, one underwent four treatments and one underwent ten.

As this technique was only attempted on two cases of chronic non-granulomatous epiglottitis, it is not possible to report a treatment algorithm or success rate. However, this study demonstrates the safety, efficacy and feasibility of treating pediatric chronic idiopathic supraglottitis. In addition to the surgical treatments, the second case was treated with a longer duration of steroids which could have led to the resolution of symptoms between treatments. Further study is warranted among this patient population to determine if this is effective for other cases of chronic idiopathic supraglottitis.

4. Conclusion

We report two cases of teenage male patients who presented with chronic idiopathic supraglottitis. Each patient was managed similarly and the CO2 laser pepper pot technique, popularized by Sandhu for the treatment of laryngeal sarcoidosis, was effective in treating the edema and eliminating the symptoms.

Conflicts of interest

None of the authors have any pertinent conflicts of interest to disclose.

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References

- [1] D.S. Parsons, R.B. Smith, E.A. Mair, L.J. Dlabal, Unique case presentations of acute epiglottic swelling and a protocol for acute airway compromise, *Laryngoscope* 106 (1996) 1287–1291.
- [2] R. Miller, C.M. Myer III, S. Gray, Otolaryngologic manifestations of chronic granulomatous disease, *Am. J. Otolaryngol.* 9 (1988) 79–82.
- [3] U.M. Musharrafieh, G.F. Araj, N.S. Fuleihan, Viral supraglottitis in an adult: a case presentation and literature update, *J. Infect.* 39 (1999) 157–160.
- [4] D.T. Wong, J.C. Gadsden, Acute upper airway angioedema secondary to acquired C1 esterase inhibitor deficiency: a case report, *Can. J. Anaesth.* 50 (2003) 900–903.
- [5] A.J. Hazenberg, F.G. Dijkers, P.N. Hawkins, J. Bijzet, D. Rowczenio, J. Gilbertson, et al., Laryngeal presentation of systemic apolipoprotein A-I-derived amyloidosis, *Laryngoscope* 119 (2009) 608–615.
- [6] A. Talerman, D. Wright, Laryngeal obstruction due to Wegener's granulomatosis, *Arch. Otolaryngol.* 96 (1972) 376–379.
- [7] D.G. Scarpelli, F.W. McCoy, J.K. Scott, Acute lupus erythematosus with laryngeal

- involvement, *N. Engl. J. Med.* 261 (1959) 691–694.
- [8] C.R. Butler, S.A. Nouraei, A.D. Mace, S. Khalil, S.K. Sandhu, G.S. Sandhu, Endoscopic airway management of laryngeal sarcoidosis, *Arch. Otolaryngol. Head Neck Surg.* 136 (2010) 251–255.
- [9] L.G. Ledgerwood, P.C. Belafsky, Chronic non-granulomatous supraglottitis: a rare and difficult disease, *J. Laryngol. Otol.* 126 (2012) 854–857.
- [10] M. Friedman, D.M. Toriumi, T. Strorigl, Chronic nongranulomatous epiglottitis, *Ear Nose Throat J.* 67 (1988) 524–527.