Case Report: Presentation
A 34-year-old active duty Air Force Sergeant presented to the multidisciplinary Laryngology and Sleep Surgery clinics at Wilford Hall Medical Center for treatment of his OSA. His surgical history was significant for a prior uvulopalatopharyngoplasty with tonsillectomy (UPPP/T), followed by a revision UPPP, Repose suspension (THIEMED), and turbinate reduction surgeries. At time of presentation to our clinic, he still complained of excessive daytime sleepiness (EDS) with an Epsilon Wakefulness Scale (ESS) score of 21, concentration difficulty, and morning headaches despite continuous positive airway pressure (CPAP) treatment at 19 cm H2O. His first pre-treatment polysomnogram (PSG) report was unable to be retrieved. However, his most recent PSG done after the above surgeries revealed an apnea-hypopnea index (AHI) of 67. Further background information revealed that he had a left true vocal fold paralysis resulting from a benign left neck mass removed in childhood. He had hoarseness at baseline, and complained of inspiratory stridor with exercise. He denied any dysphagia or aspiration. On physical examination, he had a body mass index (BMI) of 28. Retroglossism was noted and confirmed with cephalometric SNB angle of 74 degrees (reference range of 78-82 degrees) (Figure 1). Notably, his posterior airway space (PAS) measured 6.88 mm (reference range of 10-16 mm). On flexible fiberoptic laryngoscopy, an entire left aryepiglottic fold and arytenoid complex would prolapse into the laryngeal inlet causing obstruction (Figure 2).

Interval sleep study with cuffed tracheostomy tube showed a residual AHI of 7, but there was some noted air leak around his tracheostomy tube which likely resulted in underestimation of his true AHI. This questionable AHI score combined with the presence of persistent EDS symptoms led to the decision to perform maxillomandibular advancement (MMA). Individually the maxilla and mandible were advanced 10 mm. His PAS almost doubled on postoperative cephalometric analysis (6.6 mm to 11.6 mm) (Figure 4). He had normal postoperative occlusion.

Flexible endoscopy revealed significant reduction of his prior arytenoid mucosal swelling and did not show any significant collapse into the airway (Figure 5). He was decannulated, and his EDS symptoms and exercise-induced stridor resolved. He reported being able to perform unrestricted physical training and exercise. PSG was repeated, and his final post-treatment AHI was 1.9.

To treat underlying saddle nose deformity and ptotic tip secondary to lack of maxillary support, he then underwent a reconstructive rhinoplasty with rib cartilage harvest (Figures 6, 7). The patient expressed satisfaction in postoperative appearance and nasal breathing.

Discussion:
Over the course of 10 years, our patient had multi-phase and multi-level surgery in an attempt to resolve his OSA symptoms. All regions of the upper airway (nasal, palatal, retrolingual/hypopharyngeal, and supraglottic) were evaluated and treated. He had a turbinate reduction to address nasal obstruction, UPPP/T to address palatal obstruction, GTA and partial midline glossectomy to address retrolingual obstruction, and supraglottoplasty to address obstructing laryngomalacia. With the advantage of a protected airway in tracheostomy, we were able to expedite his treatment and combine procedures not normally combined (GTA and partial midline glossectomy) with supraglottoplasty. The partial midline glossectomy creates a larger retrolingual airway by removing retrolingual tongue tissue, and can have good response rates (up to 79%) in those who have previously failed UPPP. Controlled studies and level 1 evidence demonstrate improvement in clinical outcomes using radiofrequency ablational tongue reduction as a component of multilevel surgery1-4. With symphyseal osteotomies, the GTA pulls forward the genioglossus muscle to increase retrolingual space. It has been shown to have success rates from 39% to 78% in patients with severe OSA5. Review of the literature did not reveal any publications that combined these procedures in a single operation. However, given a protected airway by tracheostomy, we were able to safely proceed in this aggressive manner.

Our impression at time of surgery was that significant improvement in his posterior airway space had been achieved, however, his symptoms persisted despite this aggressive management. The decision was made to proceed with the MMA. Through Le Fort I maxillary and sagittal-split mandibular osteotomies, the MMA increases the retrolingual and retroglossal airway. Success has been documented from 90-100% of cases that followed the Powell-Riley treatment protocol6-8. In our case, it was clear that despite aggressive soft tissue reduction and advancements, correction of our patient’s skeletal deformities was necessary to achieve success. Cephalometric analysis was used as a guide for surgery, and after MMA his PAS improved from 6.6 mm to 11.6 mm. Literature has documented that narrowing of the PAS less than 7-11 mm is correlated with OSA, and that the MMA can effectively open up the PAS and cure existing OSA9-11.

What is interesting and unique in this case is the presence of laryngomalacia in a non-syndromic adult patient and the effect of supraglottoplasty in conjunction with traditional surgical interventions. There has been significant attention in the literature given to supraglottoplasty for OSA in children with laryngomalacia. Recent case series have demonstrated significant reduction in AHI in children who underwent supraglottoplasty for laryngomalacia and OSA, independent of other sleep surgery procedures12-15. On the other hand, reports of supraglottoplasty treatment for OSA in adults with laryngomalacia are more rare16-18. In one adult case series of 27 patients with laryngomalacia, partial epiglottidectomy alone decreased the overall average RDI from 45 preoperatively to 14 postoperatively. A statistically significant decrease was achieved in 78% of patients16. Though not prevalent, there is support for the role of supraglottoplasty in select cases of adult OSA.

The anatomic phenotype of the epiglottis is not well studied with regards to obstructive sleep apnea (OSA). Studies have clearly demonstrated the importance of targeted surgery for the obstructive sleep apnea (OSA) patient. As a result, multilevel evaluation addressing nasal, palatal, and hypopharyngeal sites of obstruction is common for today’s severe OSA patient. We present a challenging case that required multilevel intervention to include the supraglottis. Such a case demonstrates the need for astute preoperative evaluation and a team approach across disciplines of Otolaryngology for a successful airway reconstruction.

References: