Avascular Midline Oropharyngeal Anatomy Allows for Expanded Indications for Trans-Oral Robotic Surgery in Pediatric Patients

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Research Article

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Abstract

Introduction: Transoral robotic surgery (TORS) in children is in its infancy and indications have been primarily limited to lingual tonsillar hypertrophy and superficial mucosal lesions. However, the relatively avascular channel of the midline posterior tongue, vallecula, and posterior hyoid space provides a safe plane of dissection for deep lesions of the tongue and access to structures in the anterior neck. As robotic surgeons gain experience, application of this technology will continue to grow.

Methods: Retrospective case series

Results: We present seven patients who had either a primary (n = 3) or recurrent (n = 4) lingual thyroglossal duct cyst (TGDC) and underwent TORS excision. Four of the seven patients also underwent transoral resection of the central portion of the hyoid bone, while 3 had central hyoid resection during prior surgery. Two minor complications occurred with no evidence of lesion recurrence after mean follow up of 19.7 mo.

Discussion: The midline avascular channel of the tongue allows for relatively bloodless surgical access to pathologies of the midline base of tongue and anterior neck. Lingual thyroglossal duct cysts can safely be removed via TORS approaches with evidence of limited recurrence.

Conclusion: Robotic technology can provide safe and effective surgical alternatives for children with a variety of pathologies, and we aim to promote the widespread adoption of TORS in pediatric head and neck surgery by sharing our knowledge and clinical experience. Further study and publication are needed to establish safety and efficacy.

Introduction

To date, limited publications exist on transoral robotic surgery in the pediatric population. The current literature describes excision of the lingual tonsils, repair of laryngeal clefts, and excision of various mucosal based pathologies. Publications and experience within adult otolaryngology practice have guided a limited number of pediatric otolaryngologists in establishing TORS as a relatively safe and effective means of performing surgery in the oropharynx, hypopharynx, and larynx. Anatomic studies conducted on adult patients on the blood supply to the posterior tongue, as well as cadaveric study on the posterior hyoid space, provide guidance to expand pediatric TORS to encompass novel indications [1] [2] [3] [4]. These anatomic descriptions helped conceptualize the approach of complete transoral removal of lingual thyroglossal duct cysts and the central portion of the hyoid bone [5] .

The thyroid gland develops from an embryonic extension originating in the foramen cecum in the base of tongue/vallecula. The thyroglossal duct should obliterate during embryonic development, however remnants can leave a cyst, tract, or both. Lingual TGDCs represent a small minority of all TGDCs (8%) but can present with significant symptoms and be a challenge to eradicate [6] [7]. Symptoms can range from foreign body sensation to dysphagia and airway obstruction. It is the authors' opinion that many

vallecular cysts are thyroglossal duct remnants. Complete excision of lingual TGDCs via microscopic and endoscopic techniques is not always achievable and marsupialization can result in recurrence due to unrecognized lesion extension into deeper base of tongue musculature [8]. A fine-cut MRI through the tongue base can help identify the extent of lingual TGDCs and tracts in recurrent TGDCs. TORS offers a surgical approach to visualize and eradicate deeper base of tongue lesions with enhanced tissue handling.

We wish to highlight the relevant anatomy as a road map to the deep midline tongue and anterior neck as it relates to TORS excision of primary or recurrent lingual TGDCs. A relatively avascular plane exists in the midline from the vallecula to the hyoid bone. This avenue also includes the midline posterior tongue musculature as it meets along the midline raphe. The main blood supply to the posterior tongue is derived from the bilateral lingual arteries and the subsequent dorsal lingual arteries, which arise from the external carotid system. The hypoglossal nerve travels with the lingual artery at the level of the lateral hyoid bone. Through cadaveric dissection, Lauretano and colleagues described the relationship of this neurovascular bundle, called the HLNVB (hypoglossal lingual neurovascular bundle), for open oncologic approaches to the base of tongue [2]. Based on 10 specimens, they found, on average, that the bundle travels 1.6 cm lateral to the foramen cecum and 0.9 cm superior to the hyoid bone. An additional anatomical study by Gun and Ozer describes the position of these structures for TORS surgeons through cadaveric specimens and live surgical cases. In their research the lingual artery and hypoglossal nerve run from the greater cornu of the hyoid bone anteromedially toward the hyoglossus muscle [1]. These studies confirm that TORS dissection limited to the midline from the vallecula and base of tongue proceeding anteriorly to the level of the hyoglossus muscle should allow the surgeon to avoid hemorrhagic or functional complications.

Surgeons who perform lingual tonsillectomy are aware of the relatively avascular plane that exists between the lingual tonsils and the tongue musculature and minor salivary glands (Figure 1). Son, et al. examined gross surgical cadaveric specimens and histologic specimens and identified few vascular or nervous structures in this plane [4]. Deep to the vallecula lies the hyoepiglottic ligament and the posterior hyoid space. Maddalozzo and colleagues described the posterior hyoid space as it relates to successful trans-cervical excision of TGDCs to prevent recurrence and supported surgical outcomes data with cadaveric study defining this space and showing that it is devoid of significant vasculature [3].

Therefore, whether utilizing the plane through the vallecula, or the plane deep to the lingual tonsils, one can safely dissect anteriorly toward the hyoid bone or anterior neck without the need for an external incision in carefully selected patients.

Methods and Surgical Technique

A retrospective patient review was conducted for all pediatric TORS TGDC surgeries from 2019-2022 at the Ann and Robert H. Lurie Children's Hospital (Chicago, IL, USA). The Lurie Children's Hospital Institutional Review Board deemed this retrospective study exempt from official review. Although no

identifiable patient information is presented and surgical pictures do not identify patients, all patients have consented that their data can be used for publication. Patient demographics, presenting symptoms, operative complications, and postoperative course were analyzed. Patients who presented with a suspected lingual thyroglossal duct cyst or with a history consistent with a recurrent thyroglossal duct cyst underwent magnetic resonance imaging (MRI). Those with suspected residual lingual thyroglossal duct cyst underwent special sequencing with and without contrast with finer cuts (0.8-0.9 mm) through the tongue base to look for a residual TGDC tract, which can be missed with the larger cuts of a traditional MRI algorithm (Figure 2). T2 sequencing in the sagittal plane has provided the best localization and detail. The TORS Sistrunk surgical technique has been detailed in previous publications [5]. A 30-degree endoscope provides visualization for the dissection and central hyoid bone removal (Figures 3a-b). If the hyoid bone remains intact centrally or has not been resected in prior surgical attempts, it is critical for the bedside assistant to place firm pressure on the anterior neck at the level of the hyoid bone to visualize it in the TORS operative field (Figures 4a-b). After cyst and/or tract excision and hyoid bone resection (when indicated), the authors recommend performing an epiglottopexy to approximate the base of the lingual epiglottis to the tongue base musculature (Figure 5). Epiglottopexy helps prevent retroflexion and airway obstruction and promotes closure of the connection of the oropharynx to the soft tissues of the submental neck. Extubation in the operating room and overnight airway monitoring is recommended. A clear liquid diet is advanced to soft diet after 24 hours. Postoperative dysphagia for several days is common, and patients and families should be counseled accordingly.

Case Series

Utilizing the above anatomic concepts, we performed seven surgical procedures for lingual thyroglossal duct cyst (TGDC) excision, which included both primary TGDCs (n = 3) and recurrent TGDCs (n = 4). Four of seven procedures included transoral resection of the central portion of the hyoid bone. Three patients had undergone hyoid resection as part of a prior surgery. Table 1 lists relevant patient information. The three primary surgeries for TGDC were performed for snoring, confirmed obstructive sleep apnea, and dysphagia, respectively. The four revision TGDC procedures were performed for persistent draining fistulas near the hyoid bone in two patients and a submental mass in the other two. The number of prior surgeries in the recurrent group ranged from one to three. Of the seven procedures performed, there were two minor complications. One patient in the recurrent group had a draining oral-cutaneous fistula that presented at 7 days postoperatively and resolved with a short course of oral clindamycin. This complication likely arose because the submental cutaneous fistula tract was excised with an external, transcutaneous approach at the time of the TORS procedure, leaving a surgically dissected plane from the base of tongue to the neck skin. One patient in the primary group developed oral bleeding with blood-tinged secretions (no clots) on postoperative day 8 that was managed conservatively with overnight hospital observation. The site of bleeding was not able to be localized. The average length of postoperative stay was 2.3 days (0-6 days). All patients had resolution of symptoms and have had no recurrence of their lesions at follow up (range 5-36 months, average 19.3 months).

Table 1: Patient Demographics, Operative and Postoperative Data

Age	Primary or Recurrent	Presenting Symptoms and Signs	Transoral Resection of Hyoid	Additional procedure	Complications	Length of Stay (days)	Follow up
8 years	Primary	Snoring, base of tongue mass	Yes		Minor bleeding	3	15 mos
6 years	Recurrent (3 prior procedures)	Draining skin fistula	No		None	1	16 mos
11 years	Recurrent (3 prior procedures)	Draining skin fistula	Yes	External excision of fistula tract	Pharyngo-cutaneous fistula - resolved	3	23 mos
3 years	Primary	Obstructive sleep apnea, submucosal base of tongue mass	Yes		None	2	35 mos
3 years	Primary	Dysphagia, base of tongue mass	Yes		None	6	33 mos
19 years	Revision (1 prior procedure)	Neck mass	No		None	0	12 mos
8 years	Revision (1 prior procedure)	Neck mass	No		None	1	4 mos

Discussion

Previously published studies within the otolaryngology literature can serve as a guide for anatomic concepts that apply to pediatric patients for novel TORS procedures. Anatomic studies on the vasculature of the lingual tonsils, midline base of tongue, and the posterior hyoid space informed the creation of a novel procedure. Our series of 7 patients—who underwent excision of either primary or recurrent lingual TGDCs—demonstrates the feasibility of operating through the midline avascular channel from the tongue base to the hyoid and anterior neck.

Performance of a surgical procedure addressing both the lingual thyroglossal duct cyst and the hyoid bone is done to prevent recurrent disease. This approach is analogous to the modified Sistrunk procedure

utilized for TGDCs presenting as a cervical mass. Simple cystectomy or marsupialization of a lingual thyroglossal duct cyst can result in either persistence or recurrence requiring additional procedures, especially if the lesion has a close relationship to the hyoid bone. Zhang and colleagues reported a series of 7 pediatric patients presenting with tongue base cysts adjacent to the hyoid with recurrent disease after between 1–8 surgeries. The authors definitively managed all 7 with transcervical removal of the central hyoid bone en bloc with the tract leading to the lingual lesion. The lingual component was injected transorally with methylene blue for ease of identification [8]. Others have recommended a transoral cystectomy and transcervical Sistrunk procedure at the same setting for primary lingual TGDCs near the hyoid bone to prevent recurrence (n = 2 patients) [6]. Published data on recurrences after cystectomy or marsupialization of lingual thyroglossal duct cysts are limited, making it difficult to estimate recurrence rates. The authors would, further, argue that TGDC recurrence after a traditional, trans-cervical modified Sistrunk procedure may occur due to an unrecognized base of tongue component. Therefore, the authors believe defining the extent of base of tongue involvement in both primary lingual lesions adjacent the hyoid bone and in recurrent TGDCs is critical. Appropriate MRI sequencing allows for planning an adequate surgical approach to eradicate all lesional tissue.

The present series of 7 patients with either primary or recurrent lingual thyroglossal duct cysts demonstrates how TORS is viable treatment option with the potential of eradicating disease with a single transoral procedure. The resection of the central hyoid bone reduces the chance of recurrence and is a critical step in addressing lingual TGDCs near or attached to the hyoid bone. Of course, any primary or remnant lesion, even a linear midline tongue tract (see Fig. 2) must also be resected. In our series, three patients with recurrent lingual TGDCs had definitive exam and radiographic evidence of absence of the central hyoid bone, so this operative step was not required. Two patients had minor complications that were conservatively managed. After experiencing a pharyngocutaneous fistula in one patient who had the external skin tract remnant excised simultaneous to the base of tongue lesion and central hyoid, we recommend against resecting the skin tract at the same setting. A second patient with multiple previous recurrences showed spontaneous resolution of the skin fistula after the base of tongue lesion was removed (hyoid already removed previously). In a follow up period of 5–36 months, no recurrence has been demonstrated.

TORS in pediatric patients remains a novel domain. Currently, a limited number of specialty centers are gaining experience and attempting to broaden surgical indications. Careful patient selection for novel procedures can help to cautiously expand the boundaries in this emerging field. This expanded case series is limited in patient numbers but provides early results and experience. The authors recommend collaboration with experienced TORS surgeons who practice adult otolaryngology, or pediatric surgeons with extensive head and neck dissection experience to optimize outcomes and assist in careful patient selection.

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Figures

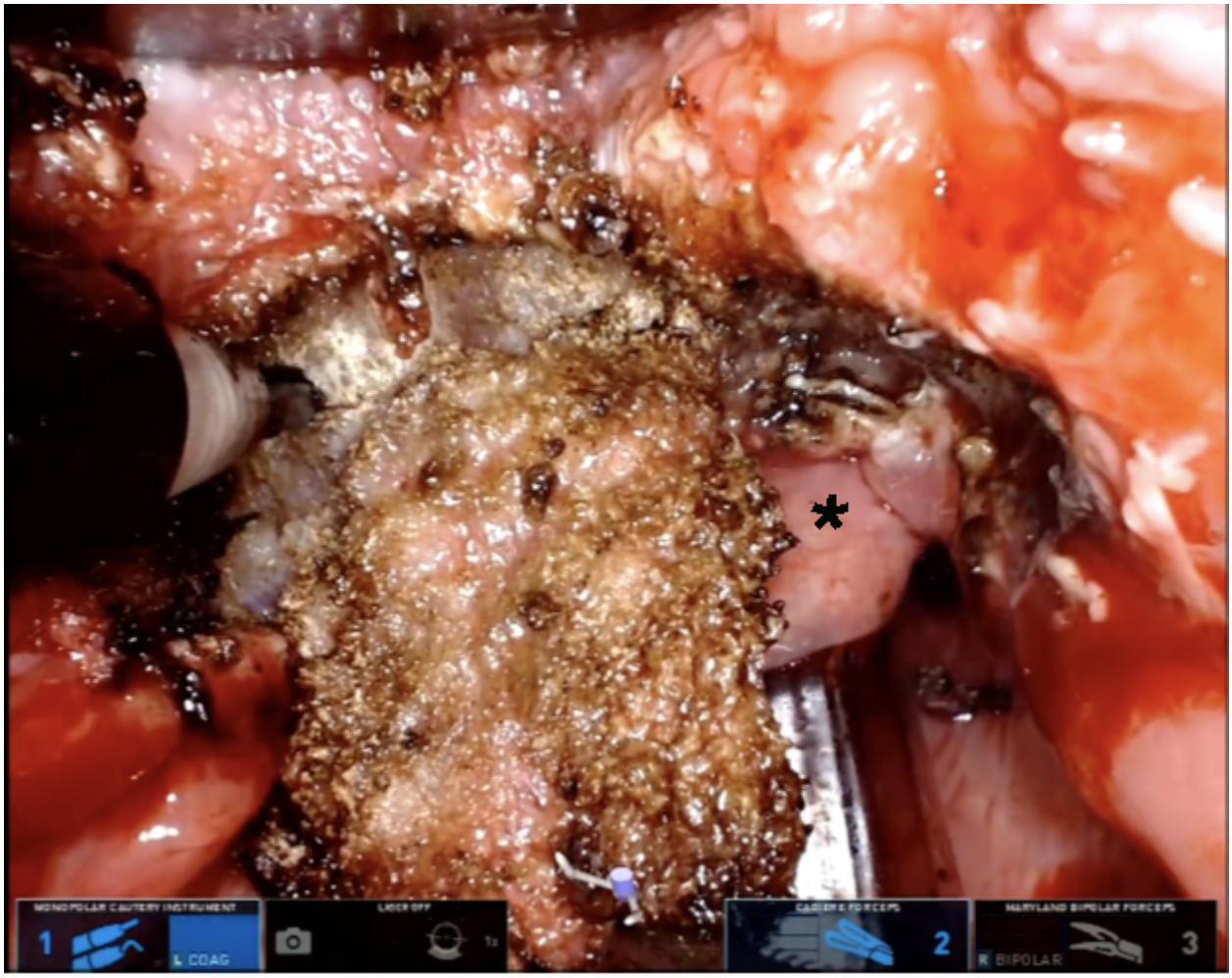

Figure 1

TORS lingual tonsillectomy. Removal of left sided lingual tonsil in avascular plane with monopolar dissection. Right sided dissection already completed. Asterix shows position of epiglottis with endotracheal tube underneath.

Figure 2

T2 weighted sagittal MRI with contrast demonstrating enhancing midline cystic structure (#) with enhancing superior extension into base of tongue and foramen cecum.

Figure 3A

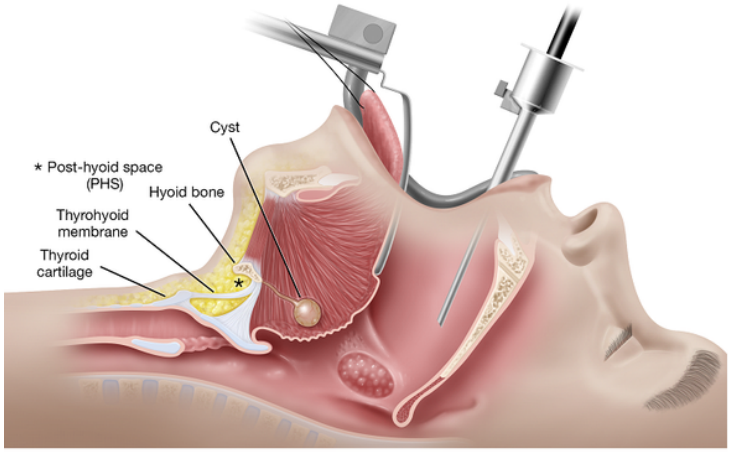

Figure 3C

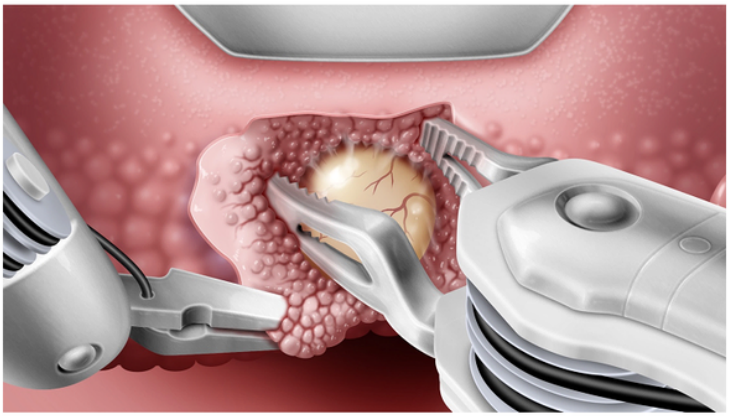

Figure 3B

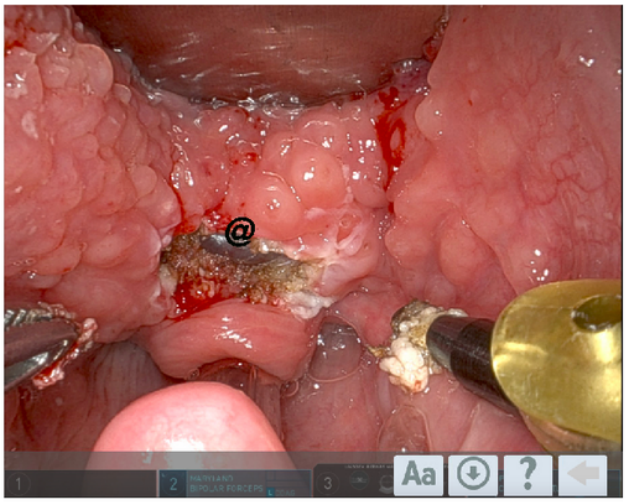

Figure 3

A. Schematic drawing demonstrating surgical set up and view with 30-degree endoscope with visualization of area of dissection anteriorly and inferiorly through posterior hyoid space to hyoid bone. B. TORS view with 30-degree endoscope of base of tongue with exposed cystic structure prior to dissection (&). C. Schematic graphical view of cyst dissection with robotic instrumentation.

Figure 4A

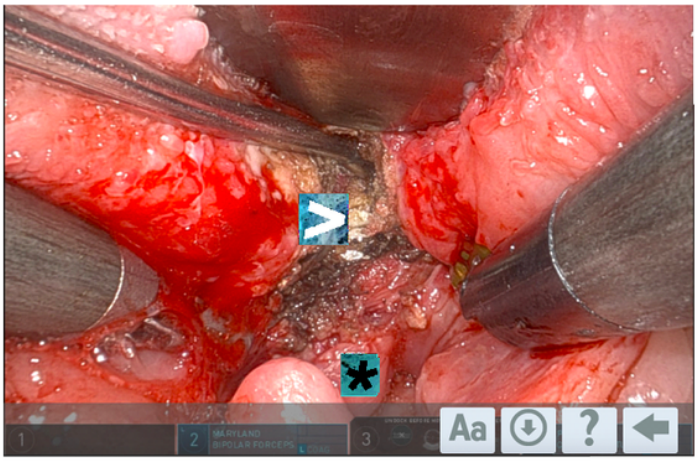

Figure 4B

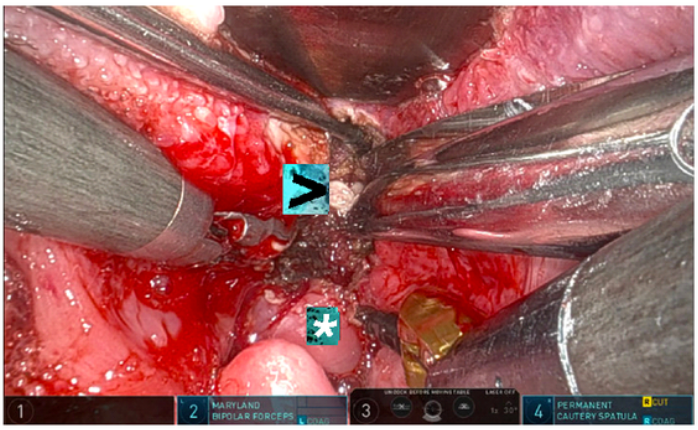

Figure 4C

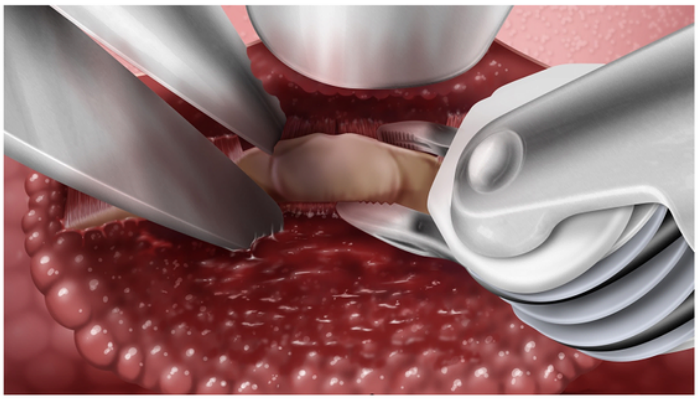

Figure 4

A. TORS view of hyoid bone (>) with anterior neck pressure applied by bedside assistant surgeon. Epiglottis (*). B. TORS view of long heavy Mayo scissors dividing central portion of hyoid bone (>). Epiglottis (*). C. Schematic graphical view of division of hyoid bone with scissors.

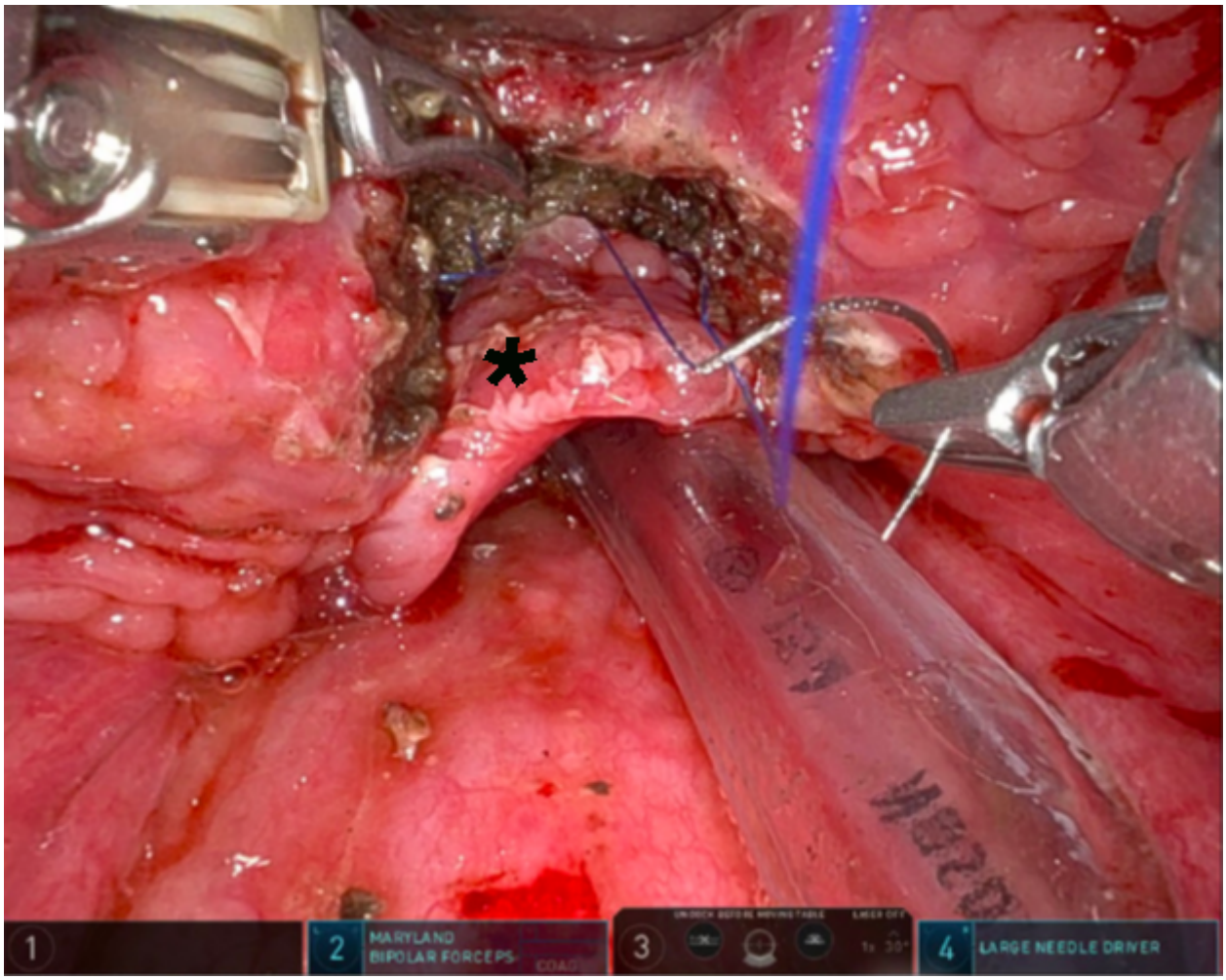

Figure 5

TORS view of epiglottopexy with Prolene suture with PS-2 needle approximating lingual surface of epiglottis (*) to base of tongue musculature.