CASE REPORT

Transoral robotic resection of a lingual thyroglossal duct cyst

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Abstract Thyroglossal duct cysts (TGDCs) are epithelial rests of the thyroid gland left during embryologic descent from the tongue base. Thyroglossal duct cysts confined to the tongue base alone are even more unusual. We present a case of a 45-year-old woman who presented with complaints of sore throat, globus, and throat clearing for 10 years. On examination she was found to have a 2×1.5 -cm cystic-appearing mass, left of midline in the vallecula. The patient had a transoral robotic resection of this vallecular cyst, which was subsequently found to be a lingual TGDC. We believe this is the first reported case of a TGDC that was successfully excised using a transoral robotic approach.

Keywords Robotic surgery · Thyroglossal duct cyst · Minimally invasive · Transoral

Introduction

During development the thyroid descends along the thyroglossal tract from the foramen cecum to the more caudal location of the thyroid gland over the cervical trachea. Following descent of the thyroid, the thyroglossal tract atrophies; however, up to 7 % of adults have tract remnants [1]. Repeated infections, proliferation of ectopic tissue, and genetics are thought to play a role in the development of a

A. J. Kimple · S. J. Eliades · J. D. Richmon (⊠) Department of Otolaryngology, Head and Neck Surgery, The Johns Hopkins Medical Institutions, 601 N. Caroline Street, 6th floor, Baltimore, MD 21287, USA e-mail: jrichmo7@jhmi.edu thyroglossal duct cyst (TGDC) in a subset of patients with thyroglossal tract remnants [1]. TGDCs are the most common midline mass in the neck [2]. The presenting age for patients with TGDC is bimodal, with nearly an equal number of patients presenting under the age of 10 years and over the age of 30 years [3]. While malignant changes have been described, TGDCs are typically benign; however, they can cause airway obstruction, odynophagia, dysphagia, dysphonia, globus sensation, and oral or cutaneous fistulae [2].

Simple excision of TGDC results in a recurrence rate of approximately 50 % [4]. In the early 1900s, Schlange and Sistrunk developed an extensive open procedure that included removing the midportion of the hyoid bone and a tissue margin around the thyroglossal tract [5]. The "Sistrunk procedure" and modifications thereof have resulted in recurrence rates of <10 % [3].

A variant of the TGDC presents as an isolated mass in the base of tongue, with no evidence of continuity in the neck. These lingual thyroglossal duct cysts (LTGDCs) typically present as a foreign body sensation [6]. Because the LTGDCs are not in continuity with the thyroglossal duct tract, management frequently does not involve a formal Sistrunk procedure [7]. Endoscopic management of LTGDC has become common and resulted in low recurrence rates [8].

The da Vinci robotic system (Intuitive Surgical, Inc., Sunnyvale, CA) is being used with increasing frequency to safely and effectively remove benign and malignant lesions of the oropharynx [9]. As opposed to traditional endoscopic approaches, the robot allows for precise bimanual tissue manipulation with high-resolution three-dimensional visualization. It has been successfully used to remove a lingual thyroid gland [10]. We describe our experience removing a LTGDC with a transoral robotic surgery (TORS) approach.

Case presentation

A 45-year-old woman was referred to our department with a 10-year history of gagging, sore throat, globus, chronic throat clearing, and emesis. Previous conservative management with allergy and anti-reflux medication failed to relieve these symptoms. A computed tomography (CT) scan revealed a vallecular lesion measuring 1.8×1.6 cm. The patient had a history of thyroiditis 3 years previously that spontaneously resolved; there were no other endocrine abnormalities.

Physical examination was unremarkable, with the exception of a fullness that was appreciated at the base of the tongue. Flexible fiberoptic laryngoscopy revealed the presence of a 2×1.5 -cm cystic-appearing lesion in the vallecula that was more prominent on the left side (Fig. 1a). The epiglottis was non-displaced and the remainder of the examination was unremarkable.

Surgical technique

The patient consented to surgical excision of the lesion with the understanding that her symptoms may not improve following removal of the cyst. After successful nasoendotracheal intubation, a maxillary dental guard was fabricated with Aquaplast (WFR/Aquaplast Corp, Wyckoff, NJ), and a 2-0 silk suture was placed through the base of the tongue to allow for extraction anteriorly. A Feyh-Kastenbauer (FK) retractor was positioned with the short base of a tongue blade and suspended on a Louis arm stand.

The da Vinci Si surgical robot was docked with a 30° up-going scope for visualization, a spatula tip Bovie in the right arm, and a Maryland dissector in the left arm.

Electrocautery was used to make a mucosal cut anterior to the cystic lesion in the base of tongue, and dissection proceeded through the deep lingual musculature with identification of the cyst capsule. Dissection proceeded around the cyst capsule, taking a small cuff of muscle around it. At one point, the cyst capsule was entered, and clear mucoid fluid was aspirated. A deeper cuff of tissue was taken in this area to ensure complete removal of the cyst wall. Dissection proceeded along the base of tongue to the vallecula until the nadir of the vallecula was reached, and the lingual surface of the epiglottic mucosa was divided. The mass was removed in its entirety. Hemostasis was obtained, the robot undocked, and the mouth gag removed.

The patient was extubated in the operating room and taken to the postoperative recovery room in stable condition. The patient tolerated the procedure without complications and was discharged on postoperative day one tolerating a liquid diet.

Histological examination demonstrated a cyst lined by ciliated columnar epithelium consistent with a TGDC (Fig. 2). At 2 months after the operation, flexible fiberoptic laryngoscopy demonstrated a remocosalized vallecula with no evidence of recurrence (Figs. 1b).

Discussion

Surgical management of TGDCs has traditionally consisted of a formal Sistrunk procedure with excision of a midportion of the hyoid bone and surrounding normal tissue to reduce the recurrence rate. LTGDC represents a subset (0.67–8.5 %) [8, 11] of TGDCs and are amenable to transoral surgical management because they are not continuous with a thyroglossal tract [8]. Burkart and colleagues

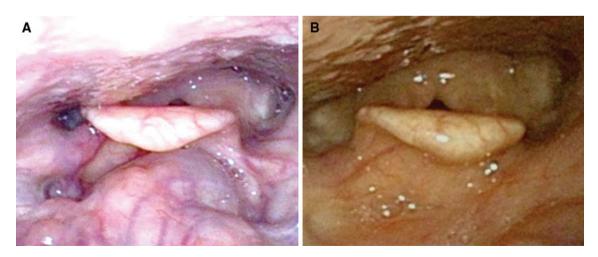


Fig. 1 Flexible fiberoptic laryngoscopy of the base of tongue and epiglottis. **a** Preoperative: a mass can be observed left of midline in the vallecula. **b** Subsequent to the transoral robotic surgery resection

of the lingual thyroglossal duct cyst: normal vallecular contours have returned, and no residual or recurrent mass is visible

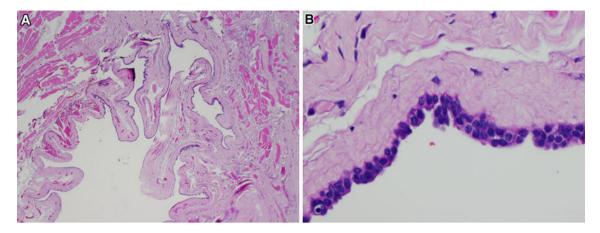


Fig. 2 a Low power microphotograph of cyst wall b High power microphotograph demonstrating cyst wall lined with ciliated columnar epithelium consistent with a thyroglossal duct cyst

reported the largest cohort of 16 LTGDC patients [8]. Half of the patients presented with labored breathing, shortness of breath, and cyanotic spells or obstructive sleep apnea, while the remaining 44 % of patients were identified incidentally on imaging studies due to developmental and speech delay, nystagmus or facial asymmetry [8]. Endoscopic excision, without a formal Sistrunk procedure, was performed in all cases, and with an average follow up of 3.77 years, there were no recurrences.

The low recurrence rate of LTGDC after endoscopic management and increased experience using transoral robotic surgery for management for base of tongue squamous cell carcinomas [9, 12] and lingual thyroids [10] laid the foundation for the removal of a LTGDC via this approach, which allows for two-handed manipulation, three-dimensional visualization, tremor filtration, and increased dexterity over endoscopic instrumentation. Future studies will need to confirm that the recurrence rate is similar to established endoscopic approaches to LTGDC.

Conclusions

We present the first report of a TORS resection of a LTGDC. This rare presentation of a TGDC is well suited for resection using the TORS approach. We believe this is the least invasive method to manage these lesions. However, further studies will need to be conducted to confirm that recurrence rates are similar to endoscopic management of LTGDC.

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Conflict of interest None.

Consent section Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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