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A Case of Lingual Thyroid Managed Using Transoral Endoscopic Surgery (TOES)

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ABSTRACT

Objective: To report a case of lingual thyroid in an adult woman, excised via transoral endoscopic surgery.

Methods:

Design: Case Report
Setting: Tertiary Government Training Hospital
Patient: One

Results: A 57-year-old woman consulted due to dysphagia and was initially treated for laryngopharyngeal reflux for a year. Thorough physical examination revealed a tongue base mass while imaging studies revealed the absence of thyroid in the anterior neck, and thyroid function tests revealed a hypothyroid state. The lingual thyroid was excised via transoral endoscopic surgery, confirmed as ectopic thyroid tissue on histopathology. There were no postoperative complications and the patient was discharged well, expressing high satisfaction with the resolution of her symptoms on follow-up at three months. She remains euthyroid on thyroxine replacement therapy.

Conclusion: The transoral endoscope-guided approach remains a valuable tool in the surgical armamentarium needed for the excision of lingual thyroids. In institutions where robotic surgery is not available, this technique is worth fostering, not only for lingual thyroids but also for lesions of the tongue base.

Keywords: *lingual thyroid; ectopic thyroid; lingual thyroidectomy; transoral endoscopic surgery; tongue base surgery*

Lingual thyroid is a rare anomaly in the oropharynx caused by failure of the thyroid gland to descend to its normal anatomical position during embryogenesis.[1] It is the most common ectopic thyroid anomaly accounting for 90% of cases[2] and has an incidence of 1:100,000 [1,3,4] with a strong female predilection.[3] It commonly presents during adolescence, pregnancy or during the menopausal stage, when there is higher physiological requirement for leading to the hypertrophy of ectopic tissue.[1] In the Philippines, there are only a few documented cases of

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lingual thyroid.[5-7] making its local incidence unknown. We report a case of lingual thyroid in an adult woman, whose diagnosis was delayed due to an incorrect diagnosis of laryngopharyngeal reflux after presenting with persistent dysphagia and globus sensation. Her lingual thyroid was excised via transoral endoscopic surgery.

CASE REPORT

A 57-year-old woman consulted in our institution due to a one year history of dysphagia with associated globus sensation. She had no other symptoms such as odynophagia, hoarseness, cough, dyspnea or heartburn. A private physician diagnosed her as a case of laryngopharyngeal reflux (LPR), and for one year she was repeatedly prescribed proton pump inhibitors (PPI), which only provided minimal relief. One month prior to admission, persistence of dysphagia and globus sensation, now accompanied by snoring and dysphonia prompted consult with a private otorhinolaryngologist. Office indirect laryngoscopy revealed an oropharyngeal mass at the tongue base.

Oral cavity and neck computed tomography (CT) scans with contrast revealed a well-defined, round, hyperdense focus at the base of the tongue measuring approximately 5.5 x 4.5 x 2.5 cm. The lesion was reported to be bulging posteriorly causing narrowing of the oropharyngeal airway. Her thyroid gland was noted to be absent. *(Figure 1)* The patient was then referred to our institution for further evaluation and management.

Further history revealed easy fatigability and weight gain. Flexible nasopharyngolaryngoscopy showed a smooth, pink mass at the base of the tongue. *(Figure 2)* Thyroid function tests revealed a hypothyroid state (thyroid stimulating hormone [TSH]: 4.72 uIU/mL (0.270-4.20 uIU/mL), Free T4 [FT4]: 4.16 pmol/L (12.0-22.0 pmol/L)). Given the history, physical examination, radiologic imaging and thyroid function test results, the patient was diagnosed to have lingual thyroid and started on thyroxine replacement therapy prior to surgery.

A lingual thyroidectomy via transoral endoscopic surgery was performed. Intraoral access was established using a Crowe-Davis retractor and a round silk 2-0 retraction suture (Tudor, Medico (Huaian) Co. Ltd., Jiangsu, China) was placed in the midportion of the oral tongue to provide good exposure of the mass. *(Figure 3)* Under endoscopic guidance using a 30-degree endoscope (Fentex medical GmbH, Neuhausen ob Eck, Germany), Harmonic Focus™ shears (Ethicon Endo-Surgery US, LLC, Cincinnati, OH, USA) were used to incise the mucosa of the lingual thyroid until the plane between the gland and tongue musculature was visualized. Allis clamps were used to grasp the edge of the thyroid to provide counter-traction for easier dissection. After resection, hemostasis using monopolar cautery was done and the surgical bed was approximated using round Vicryl™ 3-0 (polyglactin 910, Ethicon US, LLC, Johnson & Johnson MedTech, Warsaw, IN, USA) simple interrupted sutures. *(Figure 4)* The mass excised measured approximately 4 x 5 x 3 cm mass *(Figure 5)* and histopathology results

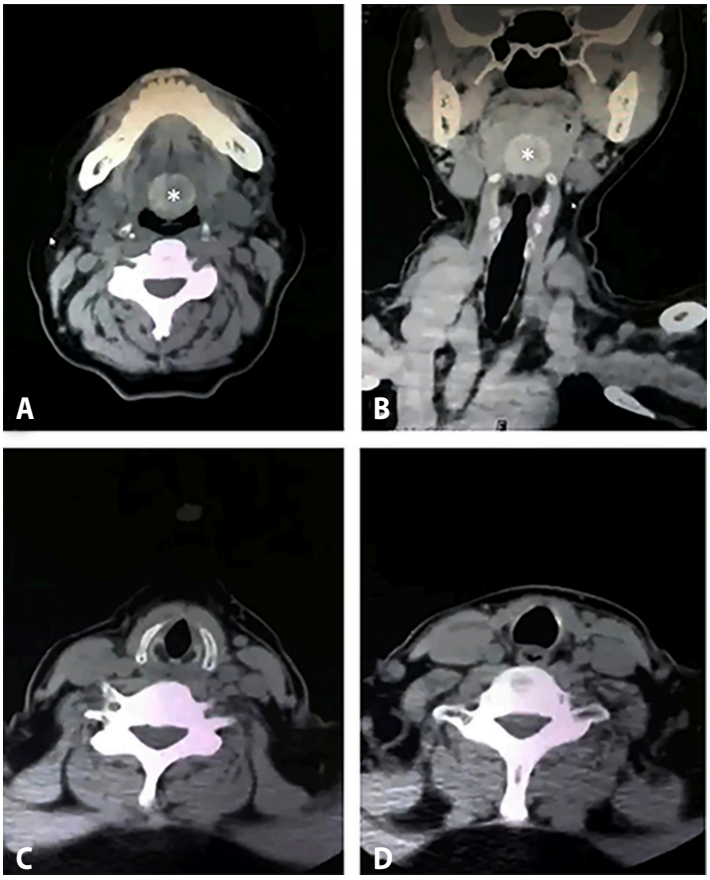

Figure 1. Oral cavity and neck CT scan with contrast showing the hyperdense mass at the base of the tongue and the absence of thyroid gland at the lower anterior neck; **A.** Axial view of the mass at the base of the tongue; **B.** Coronal view of the mass at the base of the tongue; **C; D.** Axial view of the neck, asterisks depicting the typical location of the thyroid gland

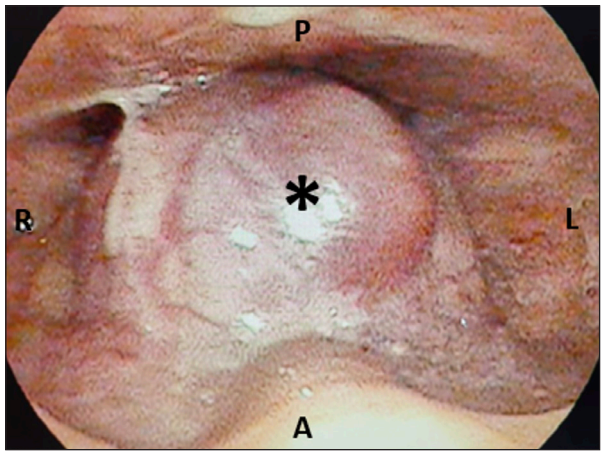

Figure 2. Base of the tongue mass seen on flexible nasopharyngolaryngoscopy (asterisk: lingual mass; A: Anterior side showing the uvula; P: Posterior side showing the posterior pharyngeal wall; R: Right side; L: Left side).

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revealed follicles lined by columnar cells with eosinophilic colloid inside the follicles, indicative of thyroid tissue.

The postoperative course was unremarkable, and our patient was discharged after two days, with home thyroxine replacement therapy. Three months post-surgery, flexible nasopharyngolaryngoscopy revealed excellent healing of the surgical site. *(Figure 6)* The patient was also euthyroid (TSH: 3.77 uIU/mL, FT4: 12.87 pmol/L) on her most recent thyroid function test. One year post-surgery, thyroid scintigraphy revealed no tracer activity in the base of the tongue.

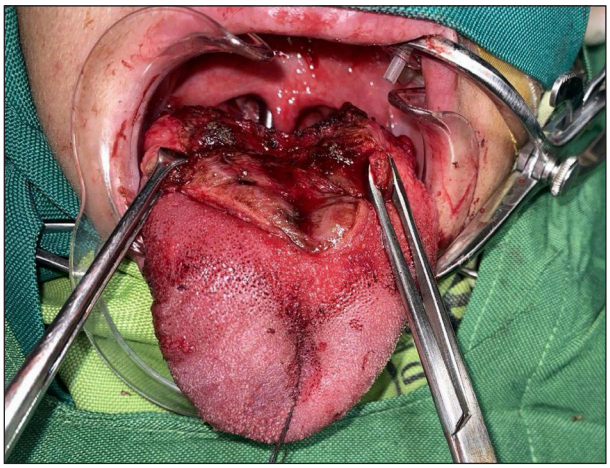

Figure 3. Exposure of the surgical site using mouth gag retractors, retraction suture and forceps

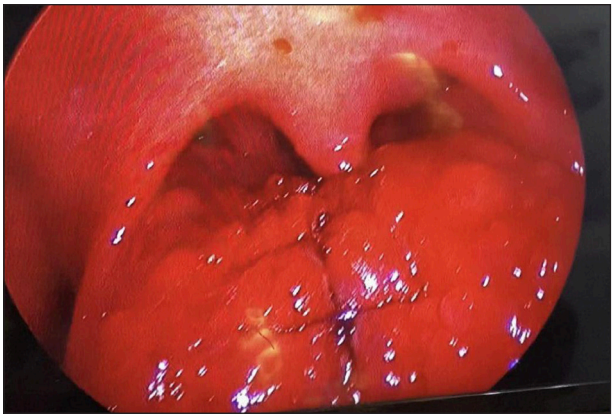

Figure 4. Endoscopic view of the surgical site during suturing of the wound bed

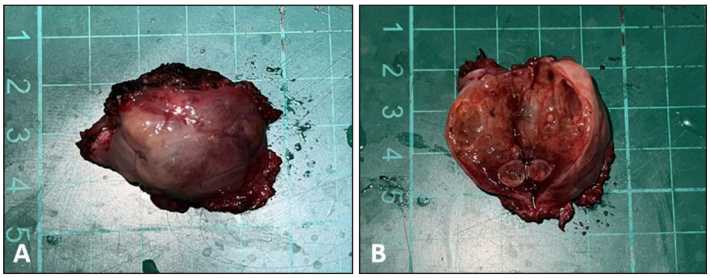

Figure 5. Lingual mass measuring approximately 4 x 5 x 3 cm. (**A:** Anterior view; **B:** Posterior View)

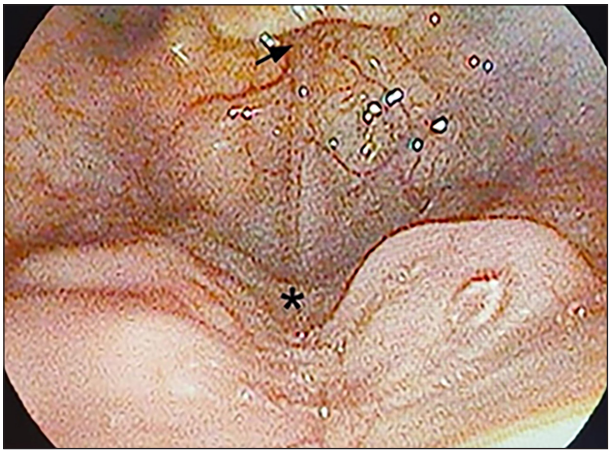

Figure 6. Post-operative site seen on flexible nasopharyngolaryngoscopy after 3 months (asterisk: surgical site; arrow: epiglottis)

DISCUSSION

Lingual thyroid is a rare and elusive clinical entity.[3,8] Recognizing this disease may be challenging, as it requires an integration of history, physical examination, laboratory testing and imaging studies.[8] Patients with lingual thyroid may be asymptomatic and may only present with symptoms once airway obstruction sets in as glandular enlargement occurs.[9] Common symptoms include dysphagia, globus pharyngeus, dysphonia, snoring, sore throat and occasional bleeding.[3,8] Around 70% of cases have hypothyroidism and may present with symptoms such as weight gain, cold intolerance, hair loss, dry skin, lethargy and easy fatigability.[3,10] On physical examination, lingual thyroid may appear as a pinkish, firm and highly vascularized mass on the base of the tongue.[1,3] However, due to its posterior location, visualization of the mass may be difficult and is prone to be overlooked.[11] Thyroid scintigraphy is a reliable tool in diagnosing lingual thyroid.[1,4] Magnetic resonance imaging, computed tomography and ultrasound may also be utilized should thyroid scintigraphy be unavailable.[4] While thyroid function tests are necessary to assess the functional status of the gland,[4] fine needle aspiration biopsy on the other hand, is not usually recommended due to risk of hemorrhage and acute thyrotoxicosis and the low risk of malignant transformation.[12]

The management of lingual thyroid depends on multiple factors such as the patient's symptomatology, size of the gland, thyroid functioning status and cosmesis.[3,13] Depending on such factors, management can range from simple monitoring, to thyroxine suppression therapy, to surgery.[14] Levothyroxine therapy may be used as initial treatment for patients with obstructive symptoms,[14] while surgery is usually indicated in cases where conservative management is futile, as well as during hemorrhage and malignancy.[13] Although there is no standard surgical procedure for lingual thyroid due to is

rarity, surgical approaches can be divided into invasive extraoral or minimally invasive transoral approaches.[9] Extraoral techniques include lip split mandibulotomy[1,12] tongue splitting[12,15] transhyoid approach, suprahyoid approach and lateral pharyngotomy.[16] while minimally invasive approaches include transoral endoscopic excision and robotic lingual thyroidectomy.[9] The advantages of minimally invasive approaches to lingual thyroid include minimized morbidity, better cosmesis, and a shorter hospital stay.[4]

Our case is a clear demonstration of how lingual thyroid can be misdiagnosed given its rather vague and nonspecific presenting symptoms. In our case the patient was managed for a year for laryngopharyngeal reflux disease before proper evaluation by an ENT specialist was made. It should be emphasized that a thorough history and physical examination, including inquiry on possible systemic symptoms, is crucial in arriving at a correct and timely diagnosis. The transoral endoscopic excision performed on our patient was a technique first described by Terris *et al.* in 2010 wherein a 30-degree laparoscope and ultrasonic shears were used to separate the lingual thyroid from the tongue musculature.[17] This approach was again reported by Kamal and El-Fattah in 2012, wherein a 70-degree endoscope was used for visualization.[12]

In conclusion, our case demonstrates that the transoral endoscope-guided approach remains a valuable tool in the surgical armamentarium needed for the excision of lingual thyroids. In institutions where robotic surgery is not available, we believe that proficiency in this technique is worth fostering, not only for lingual thyroids but also for lesions of the tongue base.

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