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Excision of a Large Lingual Thyroglossal Duct Cyst via Median Labiomandibular Glossotomy (Trotter Procedure) and Hyoid Osteotomy: A Case Report

ABSTRACT

Objective: To report a case of lingual thyroglossal duct cyst in an elderly man, excised via combined Trotter procedure and hyoid osteotomy.

Methods:

Design: Case Report
Setting: Tertiary Government Training Hospital
Participant: An elderly retired male office worker with dysphagia

Results: The cyst was completely excised via median labiomandibular glossotomy, with hyoid osteotomy providing additional exposure. Our patient was discharged in two weeks after decannulation and remains asymptomatic.

Conclusion: The Trotter procedure with hyoid osteotomy may be a viable alternative to the Sistrunk procedure for large lingual TGDCs.

Keywords: *thyroglossal duct cyst, lingual; thyroglossal duct remnants; median labiomandibular glossotomy; Trotter procedure; hyoid osteotomy*

Thyroglossal duct cysts (TGDC) are embryologic remnants of the tract of the thyroid gland as it descends to the neck and are the most common midline congenital neck masses usually diagnosed in the pediatric population.[1] Reports in adults exist, however, only a few cases in elderly patients have been reported.[2,3] Thyroglossal duct cysts can occur anywhere from the foramen cecum down to the neck but are usually associated with the hyoid bone[4] and the tongue base is an uncommon location.[2] We present the case of an elderly man with a rare lingual location of a TGDC surgically managed using an alternative approach other than the classic Sistrunk procedure – a combined Trotter procedure and hyoid osteotomy.

CASE REPORT

A 75-year-old retired male office worker consulted with a chief complaint of dysphagia. Three months before, he noted a sensation of a mass in the throat. No odynophagia, dyspnea or aspiration episodes were noted. Two months prior, the patient noted dysphagia to solids and occasional aspiration episodes. Later, this progressed to dysphagia for both solids and liquids, aspiration episodes and a hot potato voice. No odynophagia and dyspnea were noted. Persistence of symptoms prompted consult and admission. Review of history revealed that the patient is a 30-pack-year history smoker and an occasional alcoholic beverage drinker.

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On physical examination, there was no trismus, the uvula was midline, there were no masses or bulging in the non-hyperemic posterior pharyngeal wall, and tonsils were not enlarged. There were no cervical lymphadenopathies palpated.

Video laryngoscopy revealed an approximately 6 x 5 cm, round, smooth, well-defined, non-ulcerating mass arising from the tongue base and pushing the epiglottis posteriorly with minimal pooling of saliva. *(Figure 1)* Neck Computed Tomography (CT) scan showed a predominantly cystic 5.6 x 3.2 x 4.4 cm mass at the base of the tongue, superior to the hyoid, with no infiltration of surrounding tissues. The thyroid gland was visualized. *(Figure 2)* The initial impression was a vallecular cyst vs. plunging ranula. However, a lingual TGDC was considered after a case conference.

The patient underwent excision of the lingual TGDC via combined median labiomandibular glossotomy and hyoid osteotomy under general inhalational anesthesia via tracheostomy. A vertical lip splitting incision was made with cervical extension below the hyoid. Dissection was carried down through subcutaneous tissue and platysma muscle and bone. The mandible and the hyoid and suprahyoid musculature were exposed and identified. Using a Gigli Saw, the mandible was transected at the symphysis. The floor of the mouth was incised between the orifices of the Wharton's ducts and carried upward through the mucosa of the ventral aspect of the tongue to expose the median raphe and continued to the hyoid bone. The tongue was retracted anteriorly and the mandible was retracted laterally. *(Figure 3)* The incision was extended through the midline of the tongue and towards the tongue base to expose the cyst. Intraoperative findings revealed a 6 x 3 x 4.5 cm well-encapsulated cystic structure at the base of the tongue, closely related to the hyoid with mucoid contents. No attachment nor invasion of the epiglottis or surrounding tissues were noted. *(Figure 4)* Using blunt dissection, the cyst was dissected and removed gently from the tongue musculature.

To provide additional exposure and for complete removal of the cyst, the hyoid was transected in the midline. The cyst was then fully exposed and dissection from the tongue base was completed. The

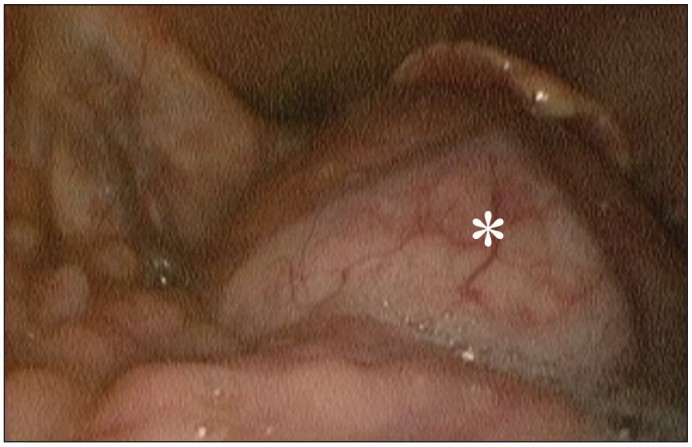


Figure 1. Video laryngoscopy showing a 6 x 5 cm, round, smooth well-defined mass arising from the tongue base anteriorly, and pushing the epiglottis posteriorly (asterisk)

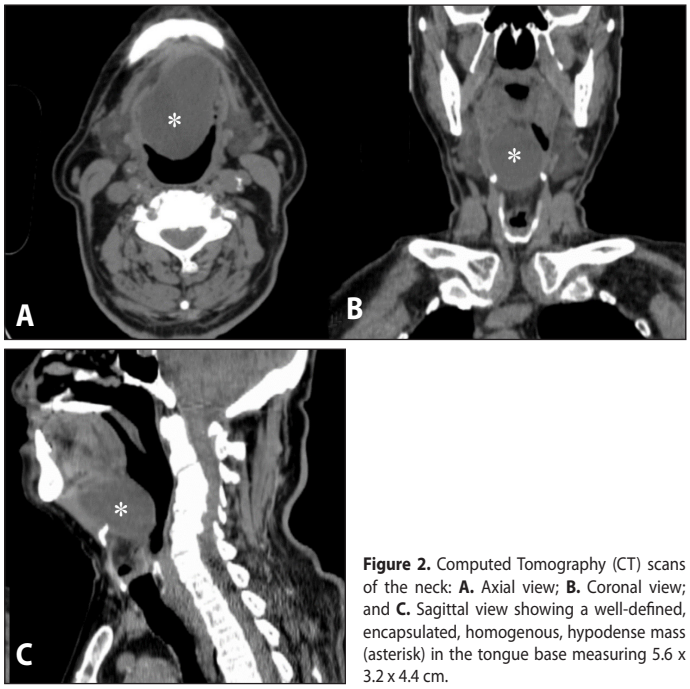


Figure 2. Computed Tomography (CT) scans of the neck: **A.** Axial view; **B.** Coronal view; and **C.** Sagittal view showing a well-defined, encapsulated, homogenous, hypodense mass (asterisk) in the tongue base measuring 5.6 x 3.2 x 4.4 cm.

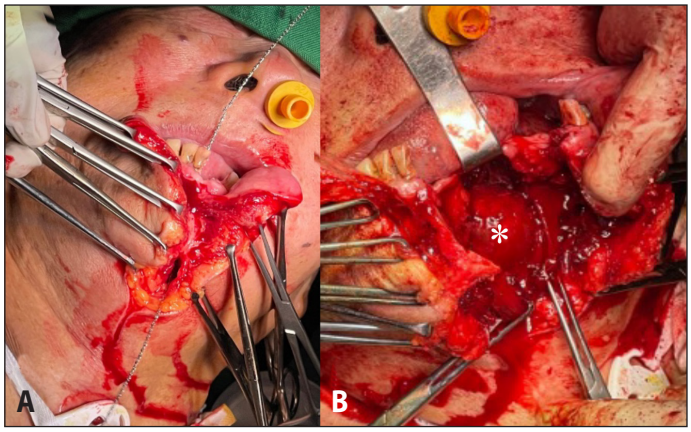


Figure 3. Intraoperative images showing: **A.** the mandible sectioned at the midline using a Gigli saw; and **B.** cystic mass noted at the tongue base (asterisk)

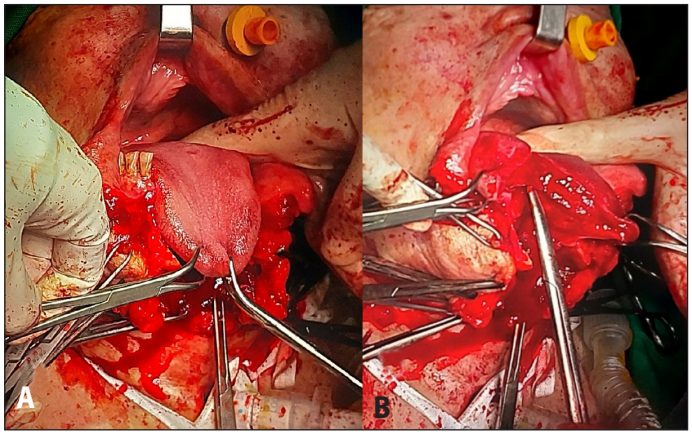
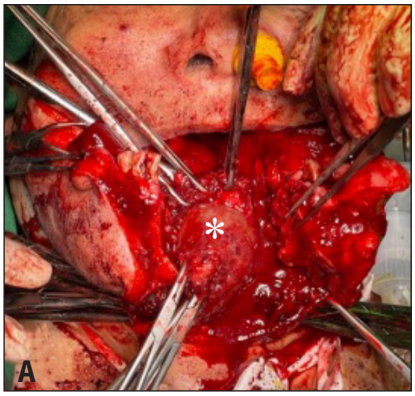


Figure 4. Intraoperative images: **A.** The tongue retracted anteriorly; and **B.** dissection through the median raphe to gain access to the cystic mass at the tongue base (not in view)



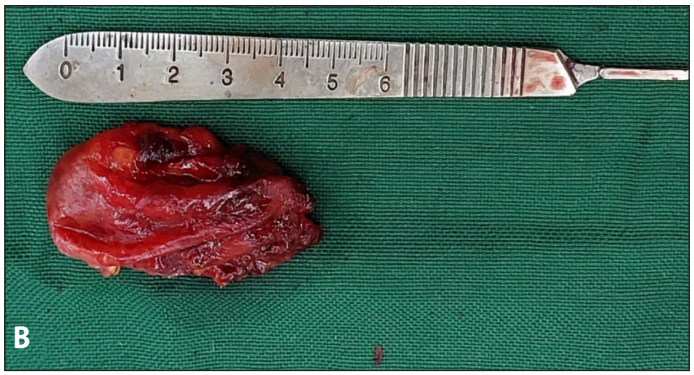


Figure 5. Intraoperative findings: **A.** 6 x 3 x 4.5 cm well-encapsulated cystic structure (asterisk) at the base of the tongue closely related to the hyoid; and **B.** Gross specimen sent for histopathologic analysis

cyst was then removed from its attachment to the hyoid. The lingual incision was closed in three layers and the hyoid and mandible were re-approximated using stainless steel wire. The mucosa of the floor of the mouth and the suprahyoid musculature were approximated. A Jackson-pratt© drain (400ml reservoir with PVC round drain, Cardinal Health, Illinois, USA) and the lip and skin of the submandibular area were closed. *(Figure 5)* Histopathological examination showed a cystic mass lined by flattened cuboidal cells and pseudostratified ciliated columnar epithelium, consistent with a lingual TGDC. *(Figure 6)* The patient was discharged two weeks post-operatively after decannulation.

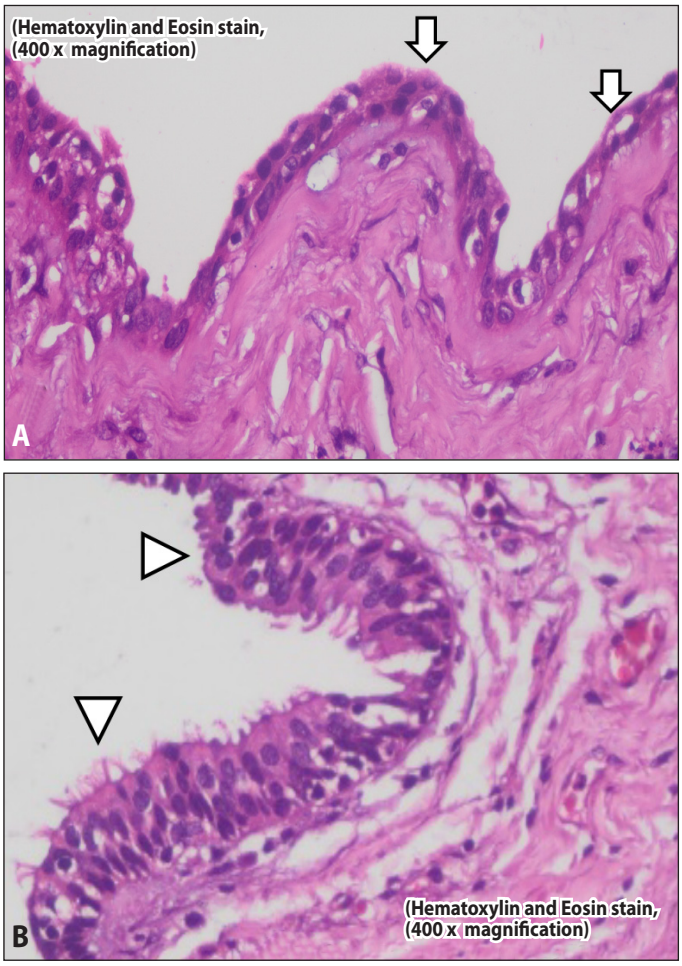


Figure 6. Histopathology slides, hematoxylin-eosin stains on high power objective (400x) showing: **A.** cuboidal (arrows); and **B.** pseudostratified ciliated columnar (arrowheads) epithelial lining, consistent with thyroglossal duct cyst

DISCUSSION

A case of a large lingual TGDC located at the tongue base in an elderly individual was removed via Trotter's procedure and hyoid osteotomy. What makes this case unique was that a congenital cyst which is commonly encountered in a younger age group was found in a septuagenarian, and in an uncommon location at the tongue base which would usually require an extensive surgery such as a lip-split mandibulotomy, glossotomy and trans-hyoid approach to remove the entire cyst.

The diagnosis of TGDC is usually made clinically presenting as cystic anterior midline neck masses that move with tongue protrusion. However, presentation and imaging characteristics are variable.[5] Ultrasound is the preferred first line of imaging especially for children characterized as well-circumscribed, anechoic lesions with thin walls and no internal flow with Doppler imaging.[6] On CT scan, they present as well-circumscribed, hypodense mass with a smooth wall with rim enhancement. While a hypointense mass in T1 and hyperintense in T2 weighted images with the presence of a "tail" closely related to the hyoid bone can be seen on MRI.[5] Diagnosis of lingual TGDC relies on laryngoscopy and CT scan.[7] Laryngoscopy would show a midline, cystic, round, smooth-walled, mass on the tongue base,[8] both were appreciated in our case.

The thyroglossal duct develops from the descent of the thyroid gland from its embryonic location at the foramen cecum and to its final location in the anterior neck. The duct may pass inferiorly, anteriorly, or through the hyoid body.[9] Failure of this tract to involute may fill with mucus from the epithelial lining of the tract and form a cyst.

While TGDCs can persist into adulthood, they are commonly diagnosed in the pediatric population. In adults, the prevalence is lower and the manifestation of new cases tends to be infrequent. The incidence of TGDC over age 50 is 28% and drops to 10% over age 60.[10] TGDC remnants may come in the form of a sinus, fistula or cyst. Infection of the tract may occur, usually after an upper respiratory tract

infection and may cause discharge to the neck or tongue base in the case of sinuses of fistulae, or if an intact capsule is formed, may trigger the formation of a cyst.[5] This may explain the occurrence of TGDCs in adults wherein the persistent duct could have been present since birth but was only triggered later in life.

Notably, cysts situated in the foramen cecum region are uncommon. In a study of 300 treated TGDCs, only two cases (0.67%) were located in the foramen cecum.[10] In another study, the distribution of the TGDC remnants were as follows: lingual (2%), suprahyoid (24%), thyrohyoid (61%), suprasternal (13%) and mediastinal (very rare).[4] The rarity of lingual TGDCs may be attributed to the involution of the duct superiorly first, where the descent of the thyroid begins. This infrequent occurrence contributes to the overall limited experience with these lesions.[11]

A case of TGDC with intralaryngeal extension was reported in local literature in a 71-year-old man who presented with airway obstruction. The mass was in the right supraglottis with an intralaryngeal component which was managed by Sistrunk procedure.[12] Only a few cases of lingual TGDC with intralaryngeal extension have been reported and this underscores the importance of including TGDC as a differential diagnosis in an elderly patient who present with dyspnea and anterior neck mass.

In our case, considering the clinical presentation (aspiration episodes, dysphagia, dysphonia), video laryngoscopy and CT scan findings, the huge size and posterior lingual location of the cyst posed a significant risk of airway compromise, should the cyst become infected and enlarge. However, the patient did not manifest with dyspnea nor stridor at the time of admission.

Most authors prefer complete excision vs. marsupialization and simple aspiration of TGDCs. Recurrence rates can be as high as 30% when inadequately excised.[13] Hence, transcervical and transoral approaches were considered in our case.

The Trotter procedure or median labiomandibular glossotomy is a transoral approach to the tongue base, pharyngeal wall and cervical spine which was described by William Trotter in 1920.[2] This provides direct and adequate access for large benign or low-grade malignant tongue base lesions. It has the advantage of minimal cosmetic and functional deformity and does not violate the neck for future neck surgery. This procedure was utilized in a case of lingual TGDC in an 88-year-old man, however, glossotomy was not done.[2] In our case, however, glossotomy was deemed necessary since the cyst lay within the tongue musculature and a direct approach was achieved by dividing the tongue at the midline to facilitate excision.

In our case, intraoperatively, the cyst was located more inferiorly and posterior to the hyoid bone which made it difficult to completely remove the mass from the surrounding tissue. Hence, the hyoid bone was transected at the midline to facilitate en bloc removal of the cyst. On inspection, no tracts were traced from the TGDC to the hyoid or foramen cecum.

A study of 21 patients who had excision of TGDCs without hyoid bone resection showed no recurrence and with minimal complications. The study concluded that if the tracts have been fully removed along with the cyst, the hyoid bone can be preserved.[16] Recurrence rates after Sistrunk or modified Sistrunk can be as high as 14%, and removal of hyoid bone did not affect recurrence.[17] Factors that contributed to increased recurrence rates include inaccurate initial diagnosis, infection, unusual presentation, presence of multiple tracts, and inadequate excision of the tongue base musculature.[16]

In conclusion, lingual TGDC can occur at the tongue base and in elderly patients, not just along the midline of the neck in younger individuals. While the Sistrunk procedure is the standard technique for TGDCs, it may not be suitable for large cysts confined in the posterior tongue base. Hence, the Trotter procedure with hyoid osteotomy may be a viable alternative. Long-term follow-up is advised to monitor recurrence and complications.

REFERENCES

1. Bell EB, Nugent A, El-Deiry MW. Differential diagnosis of neck masses. In: Flint PW, Francis HW, Haughey BH, Lesperance MM, Lund VJ, Robbins KT, et al., editors. Cummings Otolaryngology: Head and Neck Surgery. 7th ed. Philadelphia: CV Mosby Co.; 2020 p. 1734.
2. Arden RL, Miller LK, Application of Trotter approach for large intralingual thyroglossal duct cyst in an 88-year-old patient. *J Oral Maxillofac Surg.* 2017 Sep;75(9):1921-1925. DOI: 10.1016/j.joms.2017.02.007; PubMed PMID: 28284788.
3. Baisakhiya N. Giant thyroglossal cyst in an elderly patient. *Indian J Otolaryngol Head Neck Surg.* 2011 Jul;63(Suppl 1):27-8. DOI: 10.1007/s12070-011-0179-9; PubMed PMID: 22754829; PubMed Central PMCID: PMC3146692.
4. Fagan J. Thyroglossal duct remnants. The Open Access Atlas of Otolaryngology, Head & Neck Operative Surgery. 2017 [cited 2024 February 13]; Available from: https://vula.uct.ac.za/access/content/group/ba5fb1bd-be95-48e5-81be-586fbaeba29d/THYROGLOSSAL%20DUCT%20REMNANTS-1.pdf.
5. Thabet H, Gaafar A, Nour Y. Thyroglossal duct cyst: variable presentations. *Egypt J Ear Nose Throat Allied Sci.* 2011 Mar;12(1):13-20. DOI: 10.1016/j.ejenta.2011.03.001.
6. Corvino A, Pignata S, Campanino MR, Corvino F, Giurazza F, Tafuri D, et al. Thyroglossal duct cysts and site-specific differential diagnoses: imaging findings with emphasis on ultrasound assessment. *J Ultrasound.* 2020 Jun;23(2):139-149. DOI: 10.1007/s40477-020-00433-2; PubMed PMID: 32052384; PubMed Central PMCID: PMC7242578.
7. Fu J, Xue X, Chen L, Fan G, Pan L, Mao J. Lingual thyroglossal duct cyst in newborns: previously misdiagnosed as laryngomalacia. *Int J Pediatr Otorhinolaryngol.* 2008 Mar;72(3):327-32. DOI: 10.1016/j.ijporl.2007.11.001; PubMed PMID: 18082898.
8. Gupta M, Singh S, Gupta M. Lingual thyroglossal duct cyst treated by intraoral marsupialisation. *BMJ Case Rep.* 2011 Feb 8;2011:bcr1120103546. DOI: 10.1136/bcr.11.2010.3546; PubMed PMID: 22715186; PubMed Central PMCID: PMC3062879.
9. Enepekides DJ. Management of congenital anomalies of the neck. *Facial Plast Surg Clin North Am.* 2001 Feb;9(1):131-45. PubMed PMID: 11465000.
10. Solomon JR, Rangecroft L. Thyroglossal-duct lesions in childhood. *J Pediatr Surg.* 1984 Oct;19(5):555-561. DOI: 10.1016/s0022-3468(84)80103-7; PubMed PMID: 6502427.
11. Urao M, Teitelbaum DH, Miyano T. Lingual thyroglossal duct cyst: a unique surgical approach. *J Pediatr Surg.* 1996 Nov;31(11):1574-6. DOI: 10.1016/s0022-3468(96)90183-9; PubMed PMID: 8943128.
12. Contreras G, Lingan M. Airway obstruction from intralaryngeal extension of thyroglossal duct cyst in an elderly man: A case report. *Philipp J Otolaryngol Head Neck Surg.* 2022 Nov;37(2):38-41. DOI: https://doi.org/10.32412/pjohns.v37i2.2033.
13. Jones JE, Healy GB. Transoral surgical management of lesions of the base of the tongue. *Arch Otolaryngol Head Neck Surg.* 1992 Dec;118(12):1350-2. DOI:10.1001/archotol.1992.01880120076014; PubMed PMID: 1449697.
14. Indudharan R, George CK. Suprahyoid pharyngotomy - a safe and complete approach for management of oropharyngeal, hypopharyngeal and supraglottic benign lesions. *Acta Scientific Otolaryngology.* 2022 Jan:4(1):43-47. [Cited 2024 February 19]; Available from: https://www.researchgate.net/publication/358523879.
15. Choby G, Myers E. Transcervical Pharyngotomy. In: Myers EN, Snyderman CH, editors. Operative Otolaryngology: Head and Neck Surgery. 3rd Edition. China: Elsevier Ltd; 2017 p. 270-278.
16. El-Anwar MW, Nofal AA. Thyroglossal duct cyst excision with hyoid bone preservation. *Eur Arch Otorhinolaryngol.* 2016 Jun;273(6):1521-6. DOI: 10.1007/s00405-015-3624-7; PubMed PMID: 25859938.
17. Ross J, Manteghi A, Rethy K, Ding J, Chennupati SK. Thyroglossal duct cyst surgery: A ten-year single institution experience. *Int J Pediatr Otorhinolaryngol.* 2017 Oct;101:132-136. DOI: 10.1016/j.ijporl.2017.07.033; PubMed PMID: 28964283.