

# Coexistent Double Thyroglossal Duct Cysts: A Very Rare Clinical Entity - Case Report

Plodpai Y, MD<sup>1</sup>

<sup>1</sup> Department of Otolaryngology, Faculty of Medicine, Prince of Songkla University, Hat Yai, Songkhla, Thailand

Reports of double thyroglossal duct cysts at the base of the tongue and infrahyoid region are very rare in the literature. The author reported a 9-year-old boy that presented with a lump in his throat and a midline neck mass. The present case report highlights the physical examination, pre-operative imaging, and surgical management. Combined transoral/transcervical approach should be performed for complete extirpation of the thyroglossal tract and prevention of recurrence.

**Keywords:** Thyroglossal duct cyst, Lingual thyroglossal duct cyst, Sistrunk, Double cyst

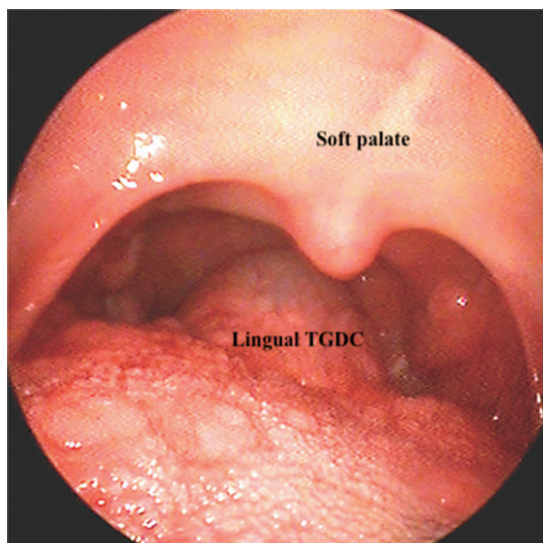
**J Med Assoc Thai 2019;102(7):816-9**

**Website:** <http://www.jmatonline.com>

Received 13 Feb 2019 | Revised 10 Jun 2019 | Accepted 11 Jun 2019

A thyroglossal duct cyst (TGDC) is the most common type of congenital midline neck mass<sup>(1)</sup>. TGDC accounts for 70 percent of congenital neck mass in childhood. During embryonic development, formation of the thyroid gland begins at the foramen caecum at the base of the tongue and descends into the midline of the neck. The thyroglossal duct is the thyroid descent tract. When the thyroid gland reaches its final position in the neck, the thyroglossal duct usually atrophies and disappears. A TGDC is caused by incomplete obliteration of the thyroglossal duct and a cyst can occur anywhere along the tract that arises from the foramen caecum to the location of the thyroid gland. TGDC is most frequently found below the level of the hyoid bone<sup>(2)</sup>. Typically, the clinical presentations are painless fluctuant midline infrahyoid neck mass. Its moves upward on tongue protrusion or moves with swallowing. A TGDC located at the base of the tongue is rare and accounts for only two to three percent of all TGDCs<sup>(3)</sup>.

The present case report describes an unusual case of double TGDC. One cyst involved the infrahyoid region and the other cyst was at the base of the tongue



**Figure 1.** Transoral view of lingual thyroglossal duct cyst before excision.

region (lingual TGDC). The surgical techniques and clinical results were discussed.

## Case Report

A 9-year-old boy presented with a 2-year history of an incidental finding of a lump in his throat. A physical examination revealed a midline cystic-appearing mass at the base of the tongue (Figure 1). It

## Correspondence to:

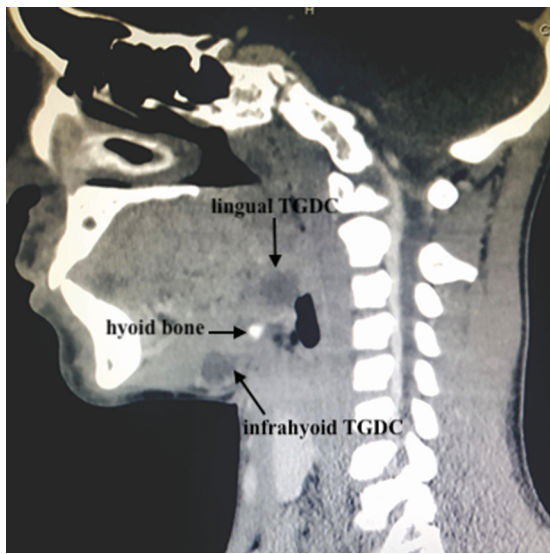
Plodpai Y.

Department of Otolaryngology, Faculty of Medicine, Prince of Songkla University, Hat Yai, Songkhla 90110, Thailand.

**Phone:** +66-74-451381, **Fax:** +66-74-429619

**Email:** yuva078@hotmail.com

**How to cite this article:** Plodpai Y. Coexistent Double Thyroglossal Duct Cysts: A Very Rare Clinical Entity - Case Report. J Med Assoc Thai 2019;102:816-9.

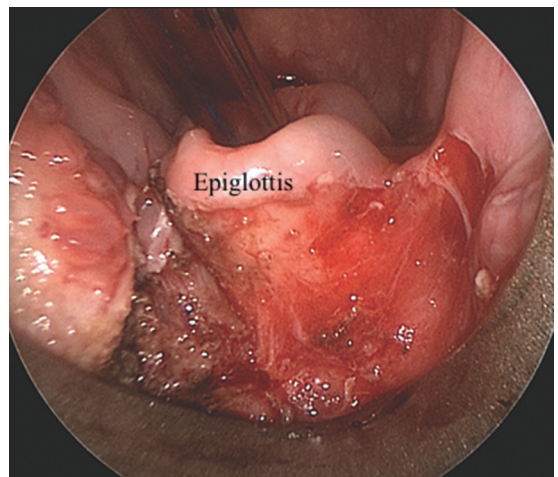


**Figure 2.** Sagittal view of computed tomography of the neck shows 2 cystic masses, one below the hyoid bone and the other located at the base of the tongue which also displaces the epiglottis.

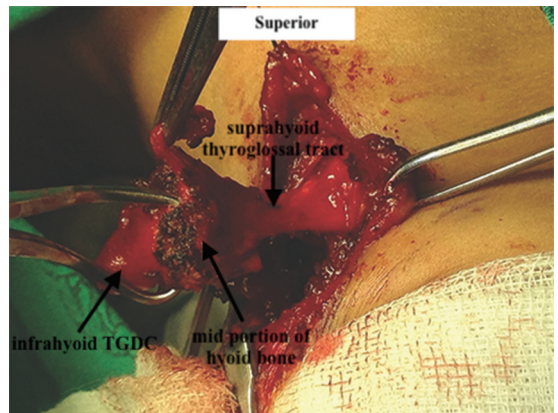
was 2 cm in diameter with a smooth surface and moved on tongue protrusion. The neck examination revealed another midline painless mass at the infrahyoid region that was 1.5 cm in diameter that moved upward with protrusion of the tongue with a palpable thyroid gland. There were no signs of upper airway obstruction. The serum levels of thyroid stimulating hormone, T<sub>3</sub> and T<sub>4</sub>, were normal. Computed tomography (CT) showed a well-defined homogeneous, low-attenuation cystic lesion at the base of the tongue. Its mass effect compressed the epiglottis inferiorly. Another well-defined homogeneous, low-attenuation cystic lesion was found at the inferior aspect of the hyoid bone (Figure 2). The thyroid gland was found to be in the normal position.

Intravenous induction was performed by propofol with spontaneous ventilation technique. Fiberoptic bronchoscope-guided nasotracheal intubation was followed by general anesthesia. A difficult airway cart and emergency tracheostomy set were also prepared in case intubation failed.

The patient was operated on by a combined transoral and transcervical approach. First, the lingual TGDC was removed via the transoral approach. The patient's larynx was suspended with a Benjamin-Lindholm laryngoscope (Karl Storz). Exposure of the entire cyst was developed (Figure 3). At this point, complete resection of the cyst at the base of the tongue was done under microscopic view with electrocautery



**Figure 3.** Transoral laryngoscopic view of lingual thyroglossal duct cyst after excision.



**Figure 4.** Transcervical Sistruck procedure: complete excision of the infrahyoid thyroglossal duct cyst and the hyoid bone together with the thyroglossal duct.

including the capsule of the lesion. The wound was allowed to heal by secondary intention. After removal of the lingual TGDC, the Sistrunk procedure was done to remove the TGDC at the infrahyoid region. A small incision was made over the cyst. The author found the complete entire tract from the foramen caecum to the superior part of the pyramidal lobe of the thyroid gland and performed meticulous en bloc resection of the entire tract and the middle portion of the hyoid bone (Figure 4). The proximal thyroglossal duct and approximately 5 to 10 mm cuff of tissue involving the base of the tongue toward the foramen caecum were removed and the wound was closed in layers. The strap muscle was then repositioned and sutured in the midline. The subcutaneous tissue and skin incision

were sutured with absorbable suture materials after insertion of a suction drain.

The patient was extubated on post-operative day 1. The patient was discharged three days after the operation without complications. In the 2-year course of follow-up, there was no evidence of recurrence.

## Discussion

The occurrence of double TGDC at the base of the tongue and at the infrahyoid region was reported in two cases in the literature. Yorgancilar et al reported a case of two cysts derived from the same thyroglossal duct, one in the base of the tongue region and the other in the hyoid region<sup>(4)</sup>. In another report, Lee et al also described the same findings in an adult and both cysts were completely excised using the Sistrunk operation<sup>(5)</sup>.

Meticulous clinical history taking and physical examination must be performed for the differential diagnosis of lingual thyroid, lingual tonsil, vallecular cyst, salivary gland tumor, hemangioma, lymphangioma, and other congenital midline neck masses<sup>(6)</sup>. Moreover, an untreated TGDC at the base of the tongue may subsequently cause a life-threatening upper airway obstruction.

In general, TGDC is diagnosed clinically and ultrasonography is the preferred imaging modality in children. However, ultrasonography had some limitations in this case. Ultrasound findings of the infrahyoid TGDC showed a unilocular, anechoic, midline cystic lesion closed to the hyoid bone, but it was difficult to show the thyroglossal tract and intraoral lesion.

The benefits of the pre-operative CT scan of the double TGDC included 1) information on the anatomical extent of the cyst in preparation for surgical management, 2) confirmation of the diagnosis, 3) evaluation of the severity of upper airway obstruction or possible synchronous airway anomalies, 4) identification of the thyroid gland, and 5) the classical findings on physical examination were not present<sup>(7)</sup>. However, radiation exposure from a CT scan is a concern in children.

The major problem in TGDC treatment is recurrence. Gioacchini et al reported that the rate of recurrence was 11%<sup>(8)</sup>. The factors associated with recurrences include young age, rupture of the cyst during the procedure, lobulation of the cyst, inflammation, infection, skin involvement, and presentation of fistulas.

The primary goal of treatment for this patient was complete surgical removal of the double TGDC

to prevent a possible recurrence and prevent airway obstruction following induction of anesthesia. The benefit of awake fiberoptic-guided nasotracheal intubation is the anatomical structure of the airway is well preserved and intact muscle tone keeps the airway anatomy separated from each other to enhance visualization of the glottis inlet. However, needle puncture and aspiration of the cyst before intubation is needed in case of difficult intubation or fiberoptic-guided intubation is unavailable.

Although the Sistrunk procedure is the standard treatment for TGDC<sup>(8,9)</sup>, the combined transoral approach is required for removal of a large lingual TGDC<sup>(6)</sup>. In the present case, the large lingual TGDC was completely excised with electrocautery via the transoral approach, which was then followed by the classical Sistrunk procedure using the cervical approach since the imaging indicated that both cysts were possibly adherent to the hyoid. The procedure involved en bloc cystectomy, central thyroidectomy, and removal of the entire tract with the surrounding cone of suprahyoid muscle up to the foramen caecum. During post-operative follow-up of 24 months, no complications or recurrence were seen.

## Conclusion

Double TGDC at the base of the tongue and infrahyoid region is a rare anomaly. The author described successful complete surgical extirpation by a combination of the transoral approach with the formal Sistrunk operation. There was no recurrence or complication.

## What is already known on this topic?

Treatment of TGDC.

## What this study adds?

Successful management of double TGDC in term of anesthetic technique, intubation technique, and surgical technique.

## Conflicts of interest

The author declares no conflict of interest.

## References

1. Allard RH. The thyroglossal cyst. *Head Neck Surg* 1982;5:134-46.
2. Bist SS, Bisht M, Varshney S, Gupta N, Bhatia R. Thyroglossal duct cyst in hyoid bone: Unusual location. *Indian J Otolaryngol Head Neck Surg* 2007;59:366-8.
3. Sameer KS, Mohanty S, Correa MM, Das K. Lingual thyroglossal duct cysts--a review. *Int J Pediatr Otorhinolaryngol* 2012;76:165-8.

4. Yorgancılar E, Yıldırım M, Gün R, Büyükbayram H, Topçu I. Double thyroglossal duct cyst located in the hyoid region and the tongue base: an unusual coexistence. *Kulak Burun Bogaz Ihtis Derg* 2011;21: 106-9.
5. Lee DH, Yoon TM, Lee JK, Lim SC. Double thyroglossal duct cysts in an adult. *J Craniofac Surg* 2017;28:e90-1.
6. Burkart CM, Richter GT, Rutter MJ, Myer CM 3rd. Update on endoscopic management of lingual thyroglossal duct cysts. *Laryngoscope* 2009;119:2055-60.
7. Zander DA, Smoker WR. Imaging of ectopic thyroid tissue and thyroglossal duct cysts. *Radiographics* 2014;34:37-50.
8. Gioacchini FM, Alicandri-Ciufelli M, Kaleci S, Magliulo G, Presutti L, Re M. Clinical presentation and treatment outcomes of thyroglossal duct cysts: a systematic review. *Int J Oral Maxillofac Surg* 2015;44:119-26.
9. Koempel JA. Thyroglossal duct remnant surgery: a reliable, reproducible approach to the suprahyoid region. *Int J Pediatr Otorhinolaryngol* 2014;78:1877-82.