CASE REPORTS

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Airway Obstruction from Intralaryngeal Extension of Thyroglossal Duct Cyst in an Elderly Man: A Case Report

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ABSTRACT

Objective: To report an unusual presentation of thyroglossal duct cyst causing airway obstruction in an elderly man.

Methods:

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

Results: A 71-year-old man with an anterior neck mass was brought to the Emergency Room due to progressive difficulty of breathing. A smooth, non-ulcerating right supraglottic mass obstructed the airway. Following an emergency high tracheotomy, contrast computed tomography scan of the neck revealed a hypodense mass with peripheral rim enhancement in the right supraglottis and an extralaryngeal component. Intra-operatively, a dumbbell-shaped cystic mass with a tract connected to the hyoid bone led to a Sistrunk procedure. Final histopathology findings were consistent with thyroglossal duct cyst.

Conclusion: It is possible for an elderly patient with impending upper airway obstruction, dysphonia, and neck mass to still have a benign and congenital thyroglossal duct cyst with intralaryngeal extension.

Keywords: *thyroglossal duct cyst; intralaryngeal extension; saccular cyst; airway obstruction*

Thyroglossal duct cyst is the most common congenital non-odontogenic mass of the neck, accounting for 70% of developmental neck lesions.[1,2] It commonly presents as a painless midline anterior neck mass, and can occur anywhere along the course of thyroid gland development.[3] Most cases (99%) are located in the midline and are below the hyoid (61% between the hyoid and thyroid gland, 13% suprasternal); 24% are suprahyoid and 2% are intralingual.[4] Even though it is anatomically closely related to the larynx, intralaryngeal extension of the cyst is rare, and may be due to massive enlargement over a long period of time and weakness of laryngeal structures.[1,5] With extension to the larynx, there may be hoarseness, dysphagia, and laryngeal obstruction leading to airway compromise.[1,4] We present one such case.

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CASE REPORT

A 71-year-old man presented at our emergency room with a 5-day history of difficulty breathing, a prior 3-year history of hoarseness, and an anterior neck mass that had been present for 35 years, without any prior consults or medication taken. Physical examination revealed a 4 x 4 x 3 cm firm, non-tender mass on the right anterior neck from the level of the hyoid bone superiorly to the thyroid cartilage inferiorly. Videolaryngoscopy showed a right-sided smooth, non-ulcerating, non-fungating, supraglottic mass obliterating the vallecula, pyriform sinus, and displacing the aryepiglottic fold medially, consequently obstructing the airway. *(Figure 1)* Considering a submucosal malignant new growth of the larynx, an emergency tracheotomy was performed to immediately establish the airway. The procedure proved to be difficult yet successful, however it was a high tracheotomy, and its stoma was created at the 1st tracheal ring (the tracheostomy tube was later observed to have assumed an awkward position, with the fenestra outside the tracheal lumen and the distal tip against the posterior tracheal wall). Nevertheless, the patient was relieved of dyspnea.

Contrast computed tomography (CT) scans revealed a well-defined, homogenous, hypodense, non-enhancing mass with peripheral rim enhancement in the right supraglottic area approximately measuring 4.7 x 4.5 x 4.8 cm (AP x T x CC) with an extralaryngeal component and confirmed the awkward position of the tracheostomy tube. *(Figure 2)* Ultrasound-guided fine needle aspiration yielded a greenish-brown turbid aspirate. Cytopathologic findings showed fairly cellular smears consisting of abundant inflammatory cells made of neutrophils and lymphocytes and few red blood cells. A lateral saccular cyst with intralaryngeal and extralaryngeal components was the primary consideration at that time. A primary extralaryngeal pathology was also considered as a differential in this case based on the chronology of the symptoms, wherein the neck mass presented before the hoarseness and dyspnea. Thyroid function tests revealed findings consistent with subclinical hyperthyroidism. A thyroglossal duct cyst with intralaryngeal extension was also considered. Hence we planned an external approach with excision if it was a saccular cyst and a Sistrunk procedure if it was a thyroglossal duct cyst, followed by suspension laryngoscopy to evaluate the larynx.

Intraoperatively, the mass had no connection to the laryngeal saccule and a plane separated the mass from laryngeal mucosa and cartilage. A tract connected the 4 x 6 cm dumbbell-shaped cystic mass to the midline posterior body of the hyoid bone *(Figure 3)* and a Sistrunk procedure was completed. The mass was removed without disrupting the endolaryngeal mucosa and sent for histopathology. The distal tip of the tracheostomy tube was in contact with the posterior tracheal wall. This caused fibrin eschar and granulation tissue formation over the area and needed to be removed. After removal of the mass, supraglottic edema developed and obstructed the airway, and the tracheostomy tube was retained and only changed to a smaller size. While waiting for the final histopathology result, our patient was discharged with tracheostomy tube in place.

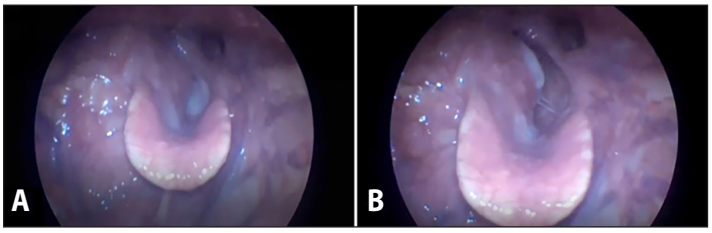

Figure 1. Videolaryngoscopy showing a smooth, non-ulcerating, non-fungating, supraglottic mass, completely obliterating the pyriform sinus, aryepiglottic fold, and vallecula on the right, obstructing the airway; vocal folds cannot be visualized on both **A**. abduction and **B.** adduction.

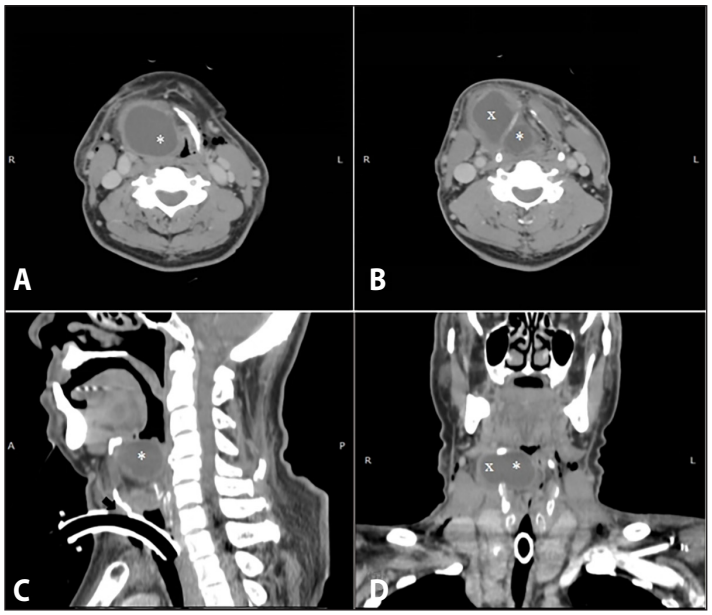

Figure 2. Computed tomography (CT) scans with contrast of the neck: **A.** axial view showing a well-defined, homogenous, hypodense, non-enhancing mass (asterisk), with peripheral rim enhancement in the right supraglottic area measuring 4.7 x 4.5 x 4.8 cm (AP x T x CC); **B.** axial view showing extralaryngeal component (x); **C.** sagittal cut showing the fenestra of tracheostomy tube outside the tracheal lumen *(arrow)*; and **D.** coronal cut showing intralaryngeal (*) and extralaryngeal (x) components.

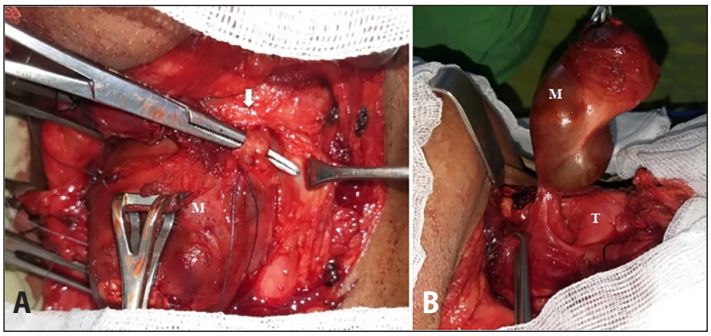

Figure 3. Intraoperative findings: **A.** a tract connecting the 4 x 6 cm dumbbell-shaped cystic mass *(M)* to the midline posterior body of the hyoid bone *(arrow)*; and **B.** plane separating the cystic mass from the laryngeal mucosa *(asterisk)* and thyroid cartilage *(T)*.

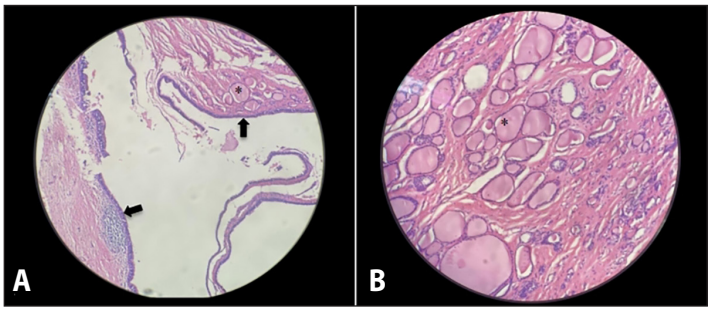

Figure 4 . Post-operative surgical pathology slides, Hematoxylin and Eosin stain interpreted as consistent with thyroglossal duct cyst: **A.** low power view (40 X) shows lining respiratory epithelium *(arrow)* with scattered thyroid follicles *(asterisk)* and with no malignant cells; and **B.** thyroid follicles *(asterisk)* seen on higher magnification (100 X).

Surgical pathology reported the specimen as consistent with a thyroglossal duct cyst, lined with respiratory epithelium with scattered thyroid follicles and with no malignant cells. *(Figure 4)* Follow-up and consults were challenging to most patients during the pandemic; this same reason hindered the timely decannulation of our patient. He was eventually decannulated after supraglottic swelling subsided and suprastomal granulation was excised and remains asymptomatic on latest follow up 4 months after.

DISCUSSION

We reported an unusual presentation of a thyroglossal duct cyst in an elderly man causing airway obstruction and the challenges encountered throughout its clinical course from diagnosis, management, and follow-up, including issues with airway management.

To the best of our knowledge, based on a search of HERDIN Plus, the ASEAN Citation Index (ACI), The WHO Western Pacific Region Index Medicus (WPRIM), Directory of Open Access Journals (DOAJ), MEDLINE (PubMed and PubMed Central), and Google Scholar using the search terms "intralaryngeal extension," [AND] "laryngeal extension," [OR] "thyroglossal duct cyst," we only found 24 previous cases of TGDC with intralaryngeal extension in the English language literature. This includes those already tabulated in previous articles,[1,5,6] making ours the 25th such case."

A thyroglossal duct cyst was not initially considered at the time of presentation since our patient was elderly with an off-midline anterior neck mass (more on the right) with accompanying supraglottic mass, hoarseness, and dyspnea, and had a 75-pack-year smoking history. Hence, a submucosal malignant neoplasm of the larynx was the initial consideration.

Laryngeal carcinoma commonly occurs in older patients, is more common in males, and has a high association with tobacco smoking and alcohol consumption. The most common presentations of laryngeal carcinoma are hoarseness, dyspnea, and neck mass if with nodal involvement or with extralaryngeal extension.[7] It is not uncommon for a thyroglossal duct cyst with intralaryngeal extension to be confused for a neoplasm, because the intralaryngeal extension leads to signs and symptoms that are not usually thought of in regard to thyroglossal duct cyst.[4] Similarly, Bando *et al.* initially diagnosed a laryngeal submucosal tumor based on patient age, 30-year smoking history, and a large submucosal mass in the left aryepiglottic fold, but considered a thyroglossal duct cyst after further examination.[6]

In a patient presenting in the emergency room with difficulty breathing, surgery to establish the airway should not be delayed. In this case, securing the airway was the immediate priority, and CT scan imaging was delayed. The patient was in respiratory distress, with a large neck mass and a short neck, which led to difficult tracheotomy. Moreover, in view of a possible laryngeal malignancy-given a supraglottic and neck mass in an elderly man with a 75-pack year tobacco smoking history, a high tracheotomy was done. High tracheotomy is rarely performed, and only in a dire emergency at the level of 1st or 2nd tracheal rings. It has a very high-risk for tracheal or subglottic stenosis specially if the cricoid cartilage has been damaged. However, it is preferred in patients with dyspnea, where laryngectomy is anticipated, since this leaves the healthy tracheal rings for formation of the tracheostoma after surgery.[8]

Of the 24 reported cases of thyroglossal duct cyst with intralaryngeal extension, only six underwent tracheotomy and these were done at the time of surgical cyst removal, where difficult intubation was noted due to airway narrowing, or wherein post-operative airway edema was anticipated. None reported performing it in an emergency setting due to impending airway obstruction, and further diagnostics such as CT scans could still be obtained.[1,6,9-12] In our case, the impending upper airway obstruction was addressed with tracheotomy before further diagnostics were performed.

On CT scan, a thyroglossal duct cyst appears as low-density mass of 19 Hounsfield units (HU) but can also present as a high-density mass up to 80 HU, especially if with increased protein content. Peripheral rim enhancement and occasional septations can also be appreciated.[2] Even with the aid of a CT scan, diagnosis can still be challenging. Thyroglossal duct cyst with intralaryngeal extension has a similar CT scan and videolaryngoscopy appearance to primary laryngeal lesions such as a lateral saccular cyst. Saccular cysts result from occlusion of the saccular orifice leading to fluid collection in the laryngeal saccule. They appear as submucosal mass on videolaryngoscopy and may also have an extralaryngeal extension to the neck penetrating the thyrohyoid membrane.[6] Hence, a lateral saccular cyst also appears as a cystic mass with both intra- and extralaryngeal components on CT scan. There are some CT scan features that can veer the diagnosis more towards a thyroglossal duct cyst. Booth et al. demonstrated the claw sign, wherein the cystic mass interdigitates with the strap muscles along the inferior hyoid.[13] Bando *et al.* also differentiated it from a saccular cyst based on

proximity to the hyoid bone and laryngeal ventricle; the cyst being closer to the hyoid than to the laryngeal ventricle favored a thyroglossal duct cyst as their diagnosis.[6] However, these distinguishing features were not present in our case; no claw sign was observed, and the cystic mass was closely related to both the hyoid and laryngeal ventricle.

Extension of a thyroglossal duct cyst into the larynx may be due to several factors such as massive enlargement of the cyst over a long period of time and weakness over the laryngeal structures. This exerts pressure and subsequent erosion or herniation through the thyrohyoid membrane or the thyroid cartilage. Intralaryngeal extension can also be due to malignant transformation of thyroid tissue within the cyst.[1,5] The former usually presents as a dumbbell-shaped mass, where the cyst expands on both sides of the hyoid and pushes through an Intact thyrohyoid membrane into the preepiglottic space, leading to its laryngeal symptoms such as dysphagia, hoarseness, and dyspnea.[1,4] This dumbbell-shaped configuration was also seen in our patient, and intralaryngeal extension could be due to massive enlargement over a long period of time given the 35-year history of anterior neck mass.

Fine needle aspiration of the cystic mass is of limited help in diagnosis. While it can determine components of cystic fluid, such as thyroid follicles consistent with a thyroglossal duct cyst as demonstrated in the case of Soliman *et al.*,[4] in our case, the liquid aspirate only yielded inflammatory cells composed of neutrophils and lymphocytes with few red blood cells and with no other cellular elements, insufficient to distinguish the particular cystic mass.

It is important to differentiate a primary laryngeal mass such as lateral saccular cyst with extralaryngeal component, from a neck mass such as a thyroglossal duct cyst with intralaryngeal component since they are managed differently. The latter is managed with a Sistrunk procedure to avoid recurrence, as was done in most of the reported cases. One case by Loh *et al.* was initially managed via endolaryngeal approach and eventually converted to Sistrunk procedure because their initial diagnosis at that time was a saccular cyst.[3] This emphasizes the importance of including thyroglossal duct cyst with intralaryngeal extension as one of the differential diagnoses in an elderly patient with both anterior neck mass and laryngeal mass.

Of the six reported cases where tracheotomy was done, none developed a tracheostomal granulation tissue and were all decannulated with good airway and voice. This may be because these cases were immediately decannulated on the 4th to 5th post-operative day.[6,9-12] However, in our case, immediate decannulation was not done due to the long period of no follow-up brought about by the restrictions and limitations of the ongoing COVID-19 pandemic. The development of suprastomal granulation tissue could be attributed to the disproportionate excision of the anterior cartilage or partial destruction of the cricoid cartilage and friction between the superior aspect of the tracheostomy tube or fenestra and the anterior airway wall due to the awkward position of the tracheostomy tube.[8] This was addressed in our patient by excision of suprastomal granulation tissue with intralesional injection of triamcinolone and subsequent decannulation. Takayama *et al.* demonstrated the effectivity of triamcinolone in eliminating intratracheal granulation tissue in small amounts.[14]

Although the clinical course of our patient was cluttered with several unharmonious events, our management ultimately addressed his needs. When confronted with a diagnostic dilemma (even with available diagnostics such as videolaryngoscopy and CT scan) or management challenges (whether to utilize external or endoscopic surgical approach), the temporal sequence of history of the present illness is helpful in making a sound diagnosis and proper management.

In conclusion, our experience shows that it is possible for an elderly patient with impending upper airway obstruction, dysphonia, and neck mass to still have a benign and congenital thyroglossal duct cyst with intralaryngeal extension.

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