



Case Report

Large Thyroglossal Duct Cyst with Atypical Endolaryngeal Presentation: A Rare Entity

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Abstract

Thyroglossal duct cysts are the commonest cystic neck swellings, usually seen in pediatric population. They are typically found in midline, and mostly infra-hyoid. Very rarely thyroglossal duct cysts are found with atypical endolaryngeal presentation. Literature search revealed 15 cases of endolaryngeal thyroglossal duct cysts reported in literature. In this article, we report the 16th case of thyroglossal duct cyst with endolaryngeal extension. This being a very rare case of a large cyst which presented with dysphonia in a 41-years-male who was referred to us for laryngoscopic examination by a speech language pathologist (SLP) His primary complaint was dysphonia and further inquiry revealed a painless, slow growing cystic neck swelling of submandibular triangle. It was diagnosed as a thyroglossal duct cyst with endolaryngeal extension following laryngoscopy and Computed tomography. Cyst was surgically removed with Sistrunks procedure with no recurrence and normal phonation without any therapeutic intervention by SLP post operatively. Literature search revealed that TGDCs with endolaryngeal extension are very rare.

Keywords: Dysphonia, Thyroglossal duct cyst, Laryngeal mass.

Introduction

Thyroglossal duct cysts (TGDC) are the most frequently encountered congenital neck swellings (7%)¹, first reported in the 1915 with hundreds of cases reported since then.² Generally, there is no gender predilection,³ and are commonly seen in pediatric age, but can occur in any age group.⁴ TGDCs are typically found in midline mostly in infra-hyoid location.^{4,5}

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Very rarely TGDCs are found with atypical presentations with endolaryngeal,⁶ prelaryngeal,⁷ and intralingual extensions.⁸ Though TGDCs commonly present with a painful infected palpable neck masses,⁴ however atypical presentations may occur including presentation with symptoms of a laryngeal mass including dysphagia, dysphonia, dyspnea and foreign body sensation.^{6,9}

We report a rare case of a large TGDC with endolaryngeal extension who reported with dysphonia. Literature review revealed 15 cases of TGDCs with endolaryngeal extension,^{6,10} and to the best of our knowledge this is the 16th case of TGDC with endolaryngeal extension and the first reported case from Pakistan.

Case report

A 41-year-male referred by a speech language pathologist presented to ENT outpatients with Dysphonia for four months. Further enquiry revealed a gradually increasing swelling in upper part of neck and foreign body sensation in throat with no other symptom. Videolaryngoscopy revealed a diffuse soft tissue bulge on the right side shifting the epiglottis and laryngeal interoitus towards the left, making it difficult to examine the laryngeal inlet, so much so that the right pyriform sinus and right vocal cord (VC) was not visible, however other laryngeal and hypo pharyngeal structures were normal with mobile left VC. Neck examination revealed a soft cystic swelling which was 5 x 3 cm and was negative for transillumination, involving the upper part of neck including the submandibular triangle. No lymphadenopathy was noted. With suspicion of malignancy, Computed Tomography (CT) (Figure 1), was performed which revealed a cystic lesion centered between hyoid and thyroid cartilage and measuring 54 mm in Anterio-posterior, 35 mm in Transverse and 40 mm in cranio-caudal dimensions. It had a thin wall with some specs of calcification in the posterior wall. The scan showed effacement and deviation of supraglottis towards left and obliteration and compression of the glottis, with lesion splaying the thyroid lamina in the center. To access the functional status of the thyroid, a thyroid scan was performed which revealed a normal functional thyroid at its normal anatomical location and an extrathyroidal swelling at the upper part of the neck (Figure 2). With a provisional diagnosis of TGDC, Sis trunk's procedure was performed and endolaryngeal component of the cyst was dissected off from neighboring structures like epiglottis, thyroid cartilage and excised in along with body of hyoid bone under general anesthesia with endotracheal intubation. Post-operative recovery was uneventful (Figure 3), with normal voice no need of intervention by SLP. 1 year follow up did not reveal any recurrence.

Diagnosis of TGC was confirmed by histopathological examination with characteristic findings. Since the cyst leaked its contents during final delivery of specimen, its Gross examination revealed a bag of tissue measuring 4 x 2.5 x 1 cm, with 0.2 cm thick fibro-muscular wall lined by cuboidal epithelium with lumen showing some colloid with mixed inflammatory infiltrate. The wall also showed moderate infiltration of polymorphs, lymphocytes and foamy histiocytes. The findings were consistent with Thyroglossal Duct Cyst, with no evidence of malignancy.

Discussion

The thyroid gland, following its origin from the foramen caecum at the 3rd week of gestation it descends as a bilobed structure to its final destination in lower neck, maintaining its connection to the foramen caecum, through a narrow canal called the thyroglossal tract. The thyroglossal duct usually gets obliterated and is no more there by the 10th week of gestation but rarely due to unknown etiological reasons the thyroglossal duct fails to involute completely and remains in the form of a tract, duct or develops into a cyst.¹¹ These may be located anywhere from the foramen caecum to the thyroid gland, however they are commonly found below the hyoid in majority of cases, between the foramen caecum and the hyoid being the second commonest location and juxtahyoid and in relation to the thyroid in very few cases.^{4,5} TGDCs with endolaryngeal extension are quite rare with 15 cases reported in literature.^{6,10}

The present case is very rare and unique, being an adult with a TGDC who initially presented with dysphonia to a speech language pathologist and a videolaryngoscopic examination revealed a soft tissue bulge pushing the epiglottis and laryngeal interoitus towards the left. This case is quite similar to a case reported by Asher et al.,⁶ however in the present case the cyst was much larger and presented with dysphonia and initial clinical examination created a suspicion of malignancy, thus mandating a CT Scan.

Though majority of TGDCs are symptomatic, presenting with midline swelling, moving on swallowing and protrusion of tongue, however presentation depends on size and location. Narayana Moorthy S et al., in their study found that 59 % of TGDCs were symptomatic and remaining typically present as painless cysts (41%). Symptoms included pain in 21%; hypothyroidism, dysphagia, breathing difficulty and discharge in 38%.⁴ Atypical presentation is a diagnostic dilemma. The present reported case is rare with unusual presentation since it grew slowly without any complaints, although its location could have caused issues like recurrent infection, cosmetic issues, swallowing difficulty, but it ultimately presented with dysphonia.

TGDCs with endolaryngeal extension may be clinically confused with laryngocoele, saccular cyst, branchial cleft cyst¹² or even malignancy,¹ like in the present case, which gave the clinical impression of a possible malignancy.

Laryngoscopic examination and radiologic investigation like CT scan are usually diagnostic in such cases.¹³ According to Thabet H et al., Magnetic Resonance Imaging (T2 weighted) is more informative and can differentiate typical TGCs from atypical TGCs and other masses.⁸

Gross examination usually reveals either serous or hemorrhagic fluid in majority and rarely colloid material. The wall is usually smooth and histologically lined with respiratory epithelium or squamous epithelium alone or in combination and is rarely without identifiable epithelial lining.¹⁴ Wall may rarely also contain ectopic thyroid tissue,¹⁵ skeletal muscle and adipose tissue and very rarely malignancy.¹⁴ In the current case, having leaked its contents gross examination of cyst delivered revealed a bag of tissue measuring 4 x 2.5 x 1 cm, with 0.2 cm thick fibro-muscular wall lined by cuboidal epithelium. Its lumen showed some colloid with mixed inflammatory infiltrate, with the wall also showing moderate infiltration of polymorphs, lymphocytes and foamy histiocytes and no evidence of malignancy.

Surgical treatment TGDCs include Sistrunk procedure, cystectomy, and / or thyroidectomy,¹⁴ however surgical excision with Sistrunk procedure with excision of the whole tract up to the base of tongue along with central part of the hyoid and central core part of musculature of deep tongue is the mainstay of treatment and has a low recurrence rate.¹⁶ In the present study we were successful in completely resecting the cyst using Sistrunk procedure, without any complication and recurrence with good speech results.

Up to date 15 cases of TGDCs with laryngeal extension have been reported in international literature.^{6,10} This is the first reported case of TGDC with endolaryngeal extension from Pakistan.

Conclusion

Thyroglossal duct cyst with endolaryngeal extension is very rare entity and its current presentation mimicking a laryngeal pathology with dysphonia is very rare. TGDCs should be kept in mind when dealing with laryngeal pathologies.

Author contributions

GS conceived of the study and participated in its design and coordination as well as helped to draft the manuscript; also read and approved the final manuscript. NM participated in its design, manuscript draft, also read and approved the final manuscripts.

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Conflict of interest

All authors declare that they have no conflict of interest.

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