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Case Report

Atypical presentation of thyroglossal cyst in pediatric patient: a case report

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ABSTRACT

Thyroglossal duct cysts are the most common cause of congenital neck swelling that may present at any age. Classically, it presents as an anterior midline neck swelling that moves with protrusion of the tongue. The usual treatment of choice is Sistrunk operation. It can have atypical presentation requiring a thorough preoperative assessment before intervention. Here we discussed a case of atypical presentation of thyroglossal duct cyst in a 4 year old child.

Keywords: Thyroglossal duct cyst, Fine needle aspiration cytology, Pediatric

INTRODUCTION

Pediatric neck masses are very commonly seen in clinical practice. These are generally divided into two broad categories-congenital and acquired neck masses or as lateral and midline swellings.

Congenital masses commonly include branchial cleft cysts, thyroglossal duct cysts, and hemangiomas. Most commonly seen congenital swelling is thyroglossal duct cyst (TGDC) with an incidence of 7% in the general population. TGDCs commonly present as painless midline neck swelling, on a line drawn from foramen caecum to suprasternal notch. Occasionally TGDCs can show atypical presentation either clinically or radiologically, which may pose a diagnostic challenge. Due to variable presentation, imaging, fine needle aspiration cytology (FNAC) and in some cases excision, followed by biopsy of excised tissue is required to reach a definitive diagnosis. We presented one such case of atypical presentation of a TGDC.

CASE REPORT

A 4-year-old girl presented to the ENT OPD with a neck swelling since birth. It was initially small in size and gradually increased. No associated history of pain, difficulty swallowing, difficulty breathing was seen in the patient. On examination, swelling was 3×6 cm sized extending from anterior border of left sternocleidomastoid to midline neck, inferiorly 2 cm above left clavicle and superiorly reaching up to the level of hyoid. It was cystic, non-tender, mobile, not attached to overlying skin, moved with deglutition but not with tongue protrusion. There was no cervical lymphadenopathy.

Patient was further evaluated; thyroid function test was normal. On USG, a cystic lesion was seen anterior to trachea in midline more on left side. Left thyroid lobe and isthmus were not visualized. For further confirmation a CECT neck was done in which there was a well circumscribed non enhancing hypodense complex cystic lesion measuring 3.5×2.9×4.9 cm noted in anterior triangle of left infrahyoid neck in paramedian location (Figure 1). The lesion showed well defined enhancing walls (maximum thickness of 3 mm), few enhancing thin septae (2 mm thickness) and fine internal hyper densities. It appeared to be closely related to the left strap muscles, and appeared to arise from the region of left lobe and isthmus of thyroid gland. The lesion was causing mild tracheal displacement to right side. The possibility of a

benign thyroid cyst arising from left lobe of thyroid was suggested. But FNAC was suggestive of a branchial cyst. So, we had three different diagnoses-thyroglossal cyst/thyroid cyst/branchial cyst.

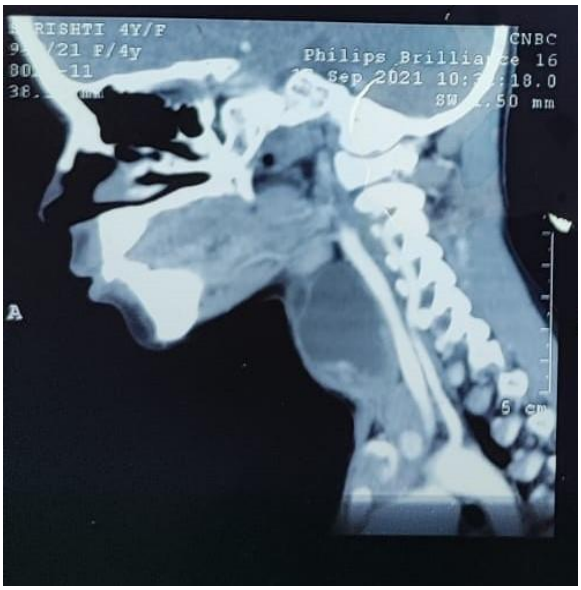

Figure 1: CECT neck s/o hypodense cystic lesion.

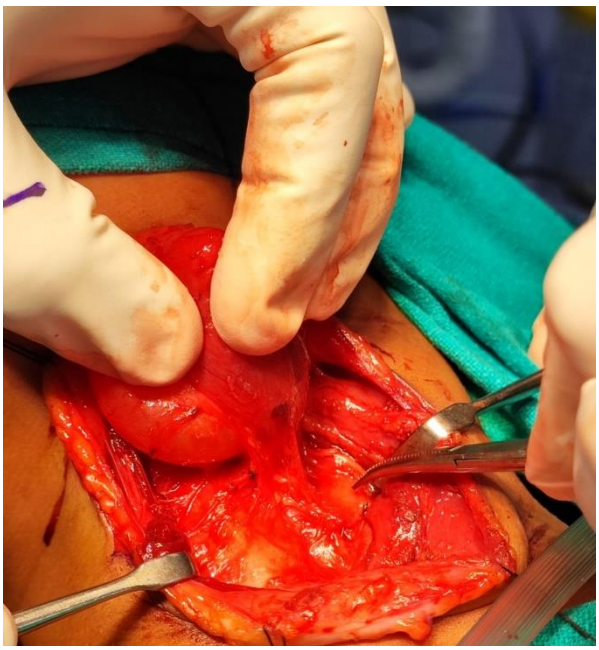

Figure 2: Intraoperative picture.

Thyroid scan was then planned which was suggestive of normal functioning right lobe with presence of a cold nodule in the place of left lobe of thyroid. A probable diagnosis of thyroid cyst arising from left thyroid lobe was made.

Patient was planned for left hemithyroidectomy with cyst excision. Horizontal incision was given over the swelling and subplatysmal flaps raised. A soft cystic swelling approximately 6×6 cm sized was seen on left side of neck which was found attached to a rudimentary left thyroid lobe; isthmus was found to be absent. Swelling was lifted off the carotid artery posteriorly, freed from trachea medially (Figure 2). Superiorly it was attached to the hyoid bone. Strap muscles were found to be splayed over the swelling. The mass was removed in toto along with rudimentary left lobe of thyroid and body of hyoid bone (Figure 3).

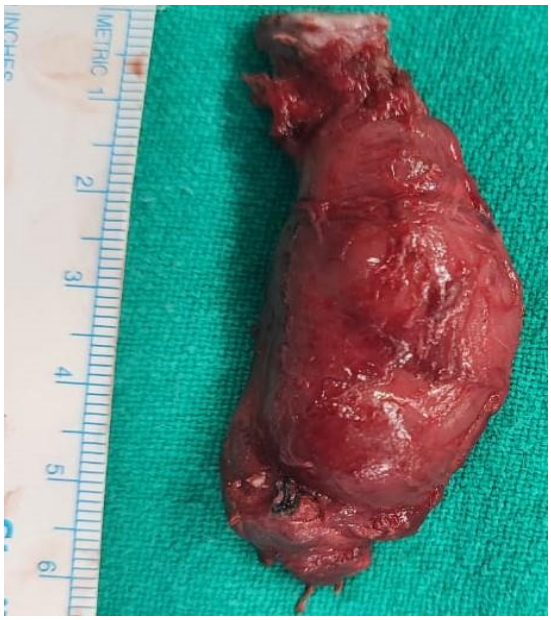

Figure 3: Excised mass.

Histopathology of excised tissue was suggestive of fibro collagenous cyst wall lined by stratified squamous epithelium. At places lining is replaced by acute on chronic inflammatory granulation tissue with mucin secreting glands along with variable sized thyroid follicles suggestive of inflamed and ulcerated thyroglossal cyst.

DISCUSSION

TGDC are the most common congenital neck masses seen in about 70% of cases, followed by branchial cleft anomalies, of which second cleft cysts are the most common. The congenital abnormalities arise out of incomplete closure of thyroglossal duct. In case the duct or any of its part survives gestation period of 10 weeks, the epithelial lining of the duct can lead to secretions, inflammation and eventually cyst formation.

TGDCs can present at any age, with age group ranging between 2 to 50 years. TGDCs present as midline neck swelling in the region of hyoid bone but can occasionally have an atypical presentation. There are few reports in the literature of laterally placed TGDCs along the anterior border of the SCM and included TGDC in the differential diagnosis of branchial cleft cyst. Sometimes it may form a TGDC within the thyroid gland, an intrathyroidal

thyroglossal duct cyst, which is difficult to differentiate from a solitary thyroid nodule. It may also mimic a pre tracheal lymph node by its cystic nature and its upward migration with protrusion of the tongue. Lymphoepithelial cyst of thyroid are uncommonly seen but can be confused with TGDCs clinically and radiologically as it presents as a cold nodule. However, on cytology, intrathyroidal lymphoepithelial cysts are seen to be lined by squamous epithelium predominantly, respiratory type epithelium is typically absent. Presence of lymphoid infiltrate in cyst wall is a feature of lymphoepithelial cyst but not seen in TGDCs.

Intrathyroidal thyroglossal ductal cysts (ITTDCs) are the rarest form of TGDCs, with only 5 reported paediatric cases in literature. ITTDCs are difficult to detect due to their rarity, lack of specific external physical characteristics independent from other thyroglossal ductal cysts. Furthermore, detection becomes even more difficult due to the patients returning normal results in thyroid function tests. Thus, in order to diagnose an ITTDC, histologic evaluation of tissue is a must. Our case mimics the picture of ITTDCs.

Role of fine needle aspiration cytology has also been widely studied in preoperative diagnosis of TGDCs. A study conducted at Johns Hopkins Hospital assessed the role of fine-needle aspiration in preoperative diagnosis of TGDCs in 26 cases in a 15-year period. According to the study, fine-needle aspiration is only moderately sensitive for preoperative evaluation of TGDCs. On cytological examination, TGDCs are lined by squamous epithelium like in branchial cleft cyst but predominantly cuboidal, columnar epithelium is seen in TGDCs which helps in differentiation. In cases of inflammation, fibrous tissue can be seen like in our case. Ultrasonography remains first choice of radiological investigation however CT imaging can be done if clinical presentation is unclear. The treatment of choice of TGDCs is Sistrunk operation, however in a case like ours where the diagnosis was doubtful choice is to be made between hemithyroidectomy and Sistrunk operation.

CONCLUSION

Similar features of TGDCs and other neck swellings can mislead the diagnostic process. These conditions are commonly seen in the paediatric population, present as neck masses, can have thyroid involvement. It is important to differentiate the masses and investigations must be performed to obtain a proper diagnosis. FNAC findings in TGDCs are variable, therefore imaging techniques or FNAC alone may not be adequate for pretreatment assessment in all cases.

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