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Sublingual thyroglossal duct cyst (SLTGDC): An unusual location

Sailesh Mukul*, Amit Kumar, Ejaz Mokhtar

All India Institute of Medical Sciences (AIIMS) Patna, India

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ABSTRACT

Thyroglossal duct cyst is the most common cause of midline congenital swelling in the neck. Classically, it presents as an asymptomatic midline swelling below the hyoid bone that moves with deglutination and protrusion of the tongue. Sometimes thyroglossal duct cyst presents atypical posing a diagnostic challenge. A sublingual location of thyroglossal duct cyst is rare, and differs quite remarkably in presentation from the classical thyroglossal duct cyst. We describe here the case of a young boy who presented with episodes of postural dyspnea due to elevation of the tongue which was secondary to huge sublingual swelling. Surgical decompression of the lesion was planned on an emergency basis with feasible preoperative workup. The aim of this case report is to highlight the unique presentation and a varied approach in the management of an unusually located sublingual thyroglossal duct cyst. Sublingual/intralingual thyroglossal duct cyst needs to be analyzed in a larger study population for establishing definitive management protocols.

Thyroglossal duct cyst is the commonest cause of a midline neck masses in children [1]. Approximately 7% of the population has thyroglossal duct remnants which account for 70% of congenital neck abnormalities [2]. Thyroglossal duct cyst presents before the age of 6-years in 76% of cases [3]. It has also been detected in the fetus in utero [4]. In 85% of cases the thyroglossal duct cyst is found below the hyoid bone [5]. However, it may be situated anywhere between the foramen cecum and the suprasternal notch.

The sublingual location of thyroglossal duct cyst with its inferior boundary located around mylohyoid region was a diagnostic challenge until it was histopathologically established. Thyroglossal duct cyst (TGDC) is usually diagnosed clinically. Classically the cyst moves upward on protrusion of the tongue which is path gnomic. Ultrasonography is the preferred imaging technique in children. Here we report the case of a young boy with an atypical presentation of sublingual thyroglossal cyst. He presented with a huge midline sub lingual cystic swelling, interfering with respiration and causing progressive postural (supine position) dyspnea. Its atypical presentation, the varied approach to its management and the rarity of the case, encouraged us to present it.

1. Case report

A 6-year old male child from the low socio economic status, resident of rural area presented to the department of Oral & Maxillofacial Surgery with the chief complain of swelling under the tongue for 6 months and difficult in breathing during sleep for 1 month. He also had difficulty in speech and swallowing. There was a history of two previous surgeries for the same problem 3-years and 6 months back, the records of which were not available. Although by history the procedures seemed to be carried trans-orally. Oral examination revealed a marked midline swelling of the floor of the mouth extending bilaterally, with normal overlying mucosa (Fig. 1). On examination no neck scars were evident which could suggest any trans-cervical approach or sistrunk procedure. There was midline sublingual scar present which could have been due to incision and drainage or incomplete enucleation of the lesion.

Examination of the neck was normal. In particular no obvious sub mental swelling could be appreciated. An Ultrasound of the neck was performed which showed a large cystic lesion located deep to the geniohyoid and mylohyoid muscles ({arrows}Fig. 2). No calcifications or mural nodules were noted in the cyst. Color Doppler did not show any vascularity in the wall or the septum. Pre and post ultrasound of the neck revealed normal thyroid gland.

Advanced imaging of the lesion like MRI or CT was not possible due to associated postural dyspnea and lack of an intubation facility at the center. Relevant preoperative investigations were normal. The intubation was anticipated to be difficult and appropriate

* Corresponding author.
E-mail address: dr.saileshmukul@yahoo.co.in (S. Mukul).

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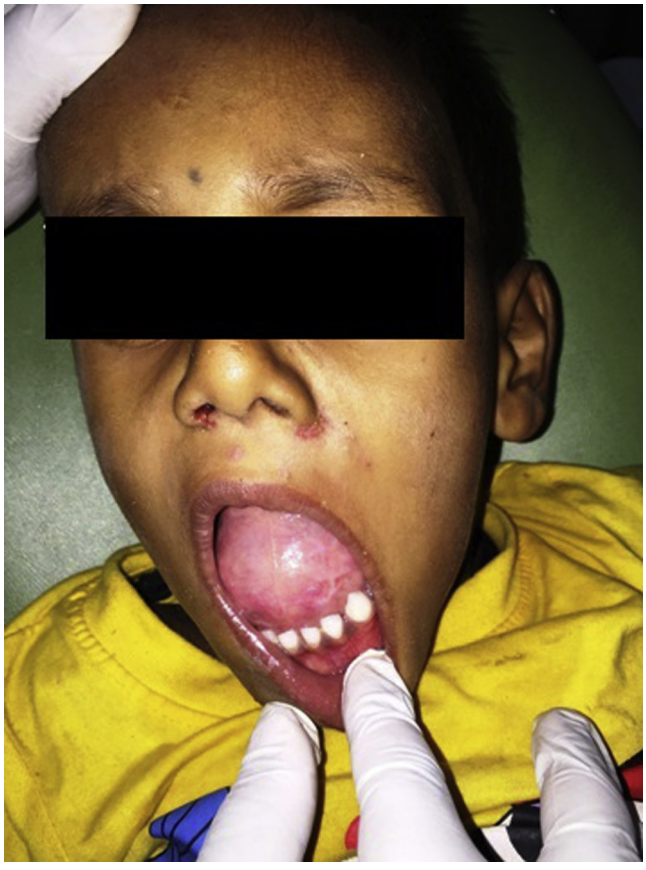

Fig. 1. Marked midline swelling of the floor of mouth extending bilaterally, with normal overlying mucosa.

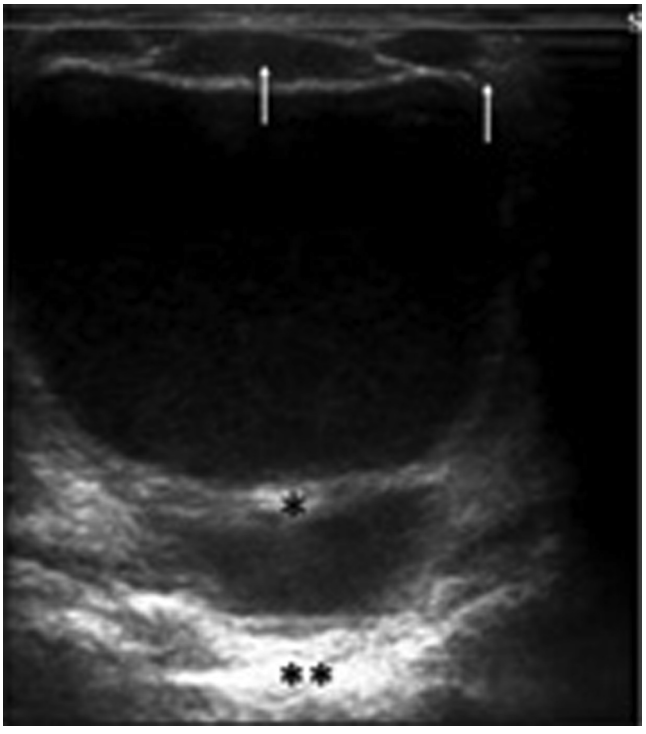

Fig. 2. High resolution ultrasonography image of the floor of mouth showing a large cystic lesion located deep to the geniohyoid and mylohyoid muscles (arrows). The cyst had a single internal septum (*) and was in midline reaching toward the posterior aspect of the tongue (**). It was casting bright posterior acoustic shadowing. There were low level internal echoes within the cyst contents. No calcification or mural nodule were noted in the cyst. Colour Doppler did not show any vascularity in the wall or septum.

provisions were made by the anesthetic team accordingly. The initial surgical plan was decompression of the swelling along with possible exploration to relieve the postural dyspnea. Under general anesthesia a midline sublingual incision was given and layer wise dissection was done to reach the lining of the cyst (Fig. 3). The inferior extent of the lesion was found to be extending up to the mylohyoid. The lining was dissected away to reach superiorly and posteriorly towards the intrinsic muscles of the tongue. The sac narrowed towards the superior aspect in the form of a cul -de- sac to terminate over the dorsal surface of the posterior 2/3rd of the tongue. There was no identifiable tract and the sac plunged into the mylohyoid muscle. The intraoperative findings and the yellowish color of the cystic fluid provisionally established it to be a thyroglossal duct cyst. The cyst was removed in totality from the foramen cecum to floor of the mouth. The surgical specimen (Fig. 4) was sent for histopathological examination, which showed it to be a cystic structure lined by pseudo-stratified to cuboidal epithelium. At places squamous met aplastic changes were also evident (Fig. 5). These histopathological findings confirmed the diagnosis of thyroglossal cyst established during the surgery. Three months postoperative USG finding are shown in (Fig. 6).

2. Discussion

Thyroglossal duct cyst (TGDC) is usually considered to be a benign embryonic malformation where the thyroglossal duct fails to obliterate after the descent of the thyroid gland [6]. The thyroid gland is originally located in the floor of the pharynx, between the tuberculum impar and the copula, during the 4th week of fetal life [7]. During development, the thyroid gland reaches its final position in front of the trachea. It leaves the thyroglossal duct, a narrow canal with an epithelial lining along the descending route of the thyroid gland. Normally, the thyroglossal duct completely disappears before the 10th week [7,8]. However, if the thyroglossal duct is not obliterated, the secretory epithelium of the thyroglossal duct may result in a TGDC.

TGDC is the second most common pediatric neck mass after lymphadenopathy [9]. Thyroglossal duct remnants occur in approximately 7% of the population, although only a minority of these becomes symptomatic [9]. TGDCs may be observed at any age, but most commonly they are noted during childhood, usually by 5-years of age. TGDCs are present at birth in approximately 25% of the cases; one third become apparent after the age of 30 [7]. Unlike most thyroid disorders which are found more frequently in females [7]. TGDCs are found with equal frequency in both sexes. Thyroglossal cyst occurs in 6 different locations [10] viz. infrahyoid cysts (26–65% of TGDCs), suprahyoid cysts (20–25% of TGDC), juxtahyoid cysts (15% of TGDCs), Intralingual cysts (2% of TGDCs), Suprasternal cysts (fewer than 10%), intralaryngeal cysts (very rare) [10]. This case had a location of TGDC which is also very rare.

Infection and abscess formation are frequent complications due to a communication between the cyst and the mouth which leads to contamination by the oral flora. This is the most common presentation in adults [11]. One fourth of the patients present with a draining sinus that results from either spontaneous drainage or surgical drainage of an abscess [12]. These lesions can also fluctuate in size, Other rare presentations can be severe respiratory distress or sudden infant death syndrome due to lesions at the base of the tongue, a lateral cystic neck mass, an anterior tongue fistula or coexistence with branchial anomalies [12]. The two most common complications of TDC are infection and malignancy. The latter occurs in 1–4% of cases [5,13].