Thyroglossal Cyst: An Unusual Presentation

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Abstract To highlight the difference in symptoms, clinical features and management of an intralingual thyroglossal cyst from a classical thyroglossal cyst. We present here the case of a 10 year old boy, who presented to us with the chief complaint of difficulty in speech for 2 years. A marble shaped swelling was seen on the base of the tongue. It was diagnosed as an intralingual thyroglossal cyst. He underwent a Sistrunk operation and the cyst was removed from the base of the tongue. Literature search revealed the rarity of this intralingual thyroglossal cyst, its atypical presentation and difference in way of management. A case report and review of literature regarding this unusual entity is presented. An intralingual thyroglossal cyst is the rarest form of a thyroglossal cyst, and differs from a classical thyroglossal cyst totally in presentation and management.

Keywords Thyroglossal cyst ·
Intralingual thyroglossal cyst · Sistrunk operation

Introduction

Thyroglossal cyst is the commonest cause of midline neck masses in children [1]. Approximately 7% of the population has thyroglossal duct remnants and it accounts for 70% of congenital neck abnormalities [2]. Thyroglossal duct cyst or remnants presents before the age of 6 in 76% of cases [3]. It has been detected in the foetus in utero also [7]. Thyroglossal cyst commonly present as painless midline neck swelling and is commonly found below the hyoid bone (85%) [2]. But they may be situated anywhere between the foramen caecum and the suprasternal notch. Classically the cyst moves upward on protruding the tongue and this sign is virtually pathognomic of it.

Here we report the case of a young boy with an intralingual thyroglossal cyst at the base of the tongue, who presented atypically with a cystic swelling at the base of the tongue interfering with his speech and deglutition. The atypicality in presentation, the differences in its assessment and management and the rarity of the case, encouraged us to present this case.

Case Report

A 10 year old boy presented to our department with the complaint of difficulty in speaking for 2 years. He had no difficulty in respiration or taking food. On examination a smooth marble sized swelling was seen on the base of the tongue, in the midline, near the foramen caecum (Fig. 1). Palpation of the neck revealed no neck swelling or neck glands. Indirect laryngoscopy was normal.

T3, T4, TSH, was normal. Thyroid scan revealed mild thyroid enlargement, mild increased uptake, and no hot or cold areas. Thyroid gland was normal in position.

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Fig. 1 Clinical picture showing marble sized swelling at base of the tongue

The swelling over the tongue did not show any significant uptake of the radiotracer. CT scan showed a well defined hypodense thin walled cystic lesion measuring 2 cm × 1.8 cm × 3.2 cm in size, in relation to the base of the tongue extending up to the hyoid in the midline and projecting into the oropharyngeal air column (Fig. 2). The tongue and thyroid was normal and no lymph nodes were seen.

Pre-operative investigations were done and the patient was admitted for surgery. Under general anaesthesia and orotracheal intubation a classical Sistrunk operation was performed. Through a horizontal incision the hyoid was exposed. The mylohyoid muscle was separated in the

Fig. 3 Histopathological appearance of specimen consistent with thyroglossal cyst (haematoxylin eosin stain, magnification ×80)

midline and the cyst was exposed. The dissection and exposure of the cyst was facilitated by pushing the cystic swelling in the mouth downward by the assistant. The cyst wall was removed totally along with the midportion of the hyoid bone. A drain was placed and the neck closed in layers. Recovery was uneventful. Drain was removed on the third day. Patient was discharged after stitch removal on the seventh day. The surgical specimen was sent for histopathological examination. Report showed a cystic lesion consistent with thyroglossal cyst (Fig. 3).

Discussion

A thyroglossal cyst may appear at any point in the migratory pathway taken by the thyroid gland during its development, including the thyroid gland per se. There are four general locations of the thyroglossal cyst—intralingual (2.1%), suprahyoid (24.1%), thyrohyoid (60.9%), and suprasternal (12.9%) [4]. In our case the cyst was at the base of the tongue, the rarest site for a thyroglossal cyst.

The intralingual cyst differs from a classical thyroglossal cyst in many ways. Firstly it does not present as a neck swelling. The patient presented to us with a soft marble sized swelling on the base of the tongue. His main complaint was difficulty in speech. William et al. [5] reported 2 cases of intralingual thyroglossal cyst in childhood. They observed that thyroglossal cyst of the tongue when encountered may be seen as a lesion which rises when the tongue is protruded. Intralingual thyroglossal cyst should be considered in infants who present with dyspnoea and inspiratory stridor. Lewison and Lim [6] reported 2 cases of infants with recurrent episodes of dyspnoea and chest retraction caused by thyroglossal cyst of the tongue. Lindstorm et al. [7] reported a case of intralingual

Fig. 2 CT scan showing hypodense thin walled cystic lesion in relation to base of tongue

thyroglossal cyst which completely obstructed the oral cavity. Large intralingual thyroglossal cyst may be associated with sudden infant death syndrome also. The common differential diagnosis of such a swelling includes hypertrophied lingual tonsil, dermoid cyst, vallecular cyst, mucus cyst, lingual thyroid, haemangioma, adenoma, lymphangioma, fibroma, lymphoma and lymphosarcoma.

Other than routine investigations we did a T3, T4, TSH estimation, a thyroid scan and a CT scan of the neck in our patient.

We did a T3, T4 and TSH estimation to see whether the patient is euthyroid. Thyroid scan was done to see whether the thyroid gland is in normal position and is functioning normally. Eric et al. [8] stated that there is a high percentage of cases (1–2%) where a thyroglossal cyst is associated with an ectopic thyroid and it is the only thyroid tissue present. Hence preoperative thyroid scan is compulsory to prevent postoperative hypothyroidism.

Computerised tomography scan of the neck showed a cystic lesion in relation to the base of the tongue extending up to the hyoid bone. This is the classical CT appearance of a thyroglossal cyst. CT also shows the presence of a normal thyroid which is of immense clinical importance. Ward et al. [9] found that CT and MRI are useful for thyroglossal cysts in unusual locations, in cases of intralaryngeal involvement, and for recurrent thyroglossal duct remnants. Though not done in this case ultrasound is a cost effective and accurate imaging modality in patients with neck masses suspicious of thyroglossal cyst [1].

Thyroglossal cyst presenting in the neck is managed surgically to prevent infection, for cosmetic reasons, and to prevent the development of carcinoma (in 1% cases) [1]. Surgery for an intralingual cyst is mandatory to cure the symptoms of dysphagia and difficulty in speech. It is life saving in children who present with dyspnea.

Sistrunk operation remains the gold standard for management of thyroglossal cysts of the neck. The definitive treatment of thyroglossal cyst of the tongue is surgical excision or marsupialisation. The lesion may be excised by midline tongue splitting incisions, by lateral pharyngotomy, transoral and transhyoid approaches or via mandibulotomy [5]. William et al. [5] advocated marsupialisation, a simple procedure with a low morbidity specially in children. The lesion is directly approached through the mouth unroofed and the base cauterized. Tracheostomy may be needed prophylactically or postoperatively.

We preferred the sistrunk approach because removal of the hyoid bone reduces the recurrence rate. Rupali et al. [10] also observed that independent of presenting age and symptomatology recurrence of thyroglossal cyst remains the lowest when the Sistrunk procedure is employed.

Conclusion

An intralingual thyroglossal cyst is the rarest site of presentation of a thyroglossal cyst. Instead of presenting as a neck swelling it presents as a swelling at the base of the tongue. The usual complaints are of difficulty in speech, dysphagia and if large, dyspnea. Excluding an ectopic thyroid by thyroid scan or ultrasound is mandatory. CT scan of the neck should be done to assess the site and extent of the lesion and plan the surgical approach. The Sistrunk procedure prevents recurrence. Pushing the cyst downward through the mouth immensely helps in total dissection of the cyst.

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