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ECTOPIC LINGUAL THYROID AS UNUSUAL CAUSE OF PROGRESSIVE DYSPHAGIA

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Abstract

Lingual thyroid gland is a rare clinical entity that is due to abnormal migration of the thyroid tissue during embryonic development. We present a case of lingual thyroid with hemi-agenesis of right lobe and colloid nodule in the left lobe presenting with difficulty in breathing and swallowing with choking episodes. Thorough clinical examination and investigations were carried out. The lingual mass was removed and sent for histopathological examination, which ultimately confirmed the diagnosis of lingual thyroid tissue. We are presenting the case for its rarity in general practice and because of its unusual presentation. The literature is reviewed regarding the incidence and diagnosis of lingual thyroid with possible treatment options discussed. The clinicians and the radiologists must be aware of this entity to avoid mistaking it for evidence of invasion by a malignant neoplasm.

Key words: Dysphagia, Lingual Thyroid, Ectopic

Introduction

Lingual thyroid is the term applied to a mass of ectopic thyroid tissue located at the base of the tongue in the mid line. This uncommon developmental anomaly may be found anywhere between the circumvallate papillae and the epiglottis. It is caused by the faulty descend of the thyroid gland through the thyroglossal duct to its normal pre-tracheal position. The presence of ectopic thyroid tissue has also been reported at other mid-line location of neck near the hyoid bone, larynx, trachea, mediastinum and oesophagus.[1] In majority of cases, there is no functional thyroid tissue in the normal cervical position. The diagnosis is usually made by the discovery of an incidental mass on the back of tongue that may enlarge and cause dysphagia, dysphonia, dyspnoea or a sensation of choking. Hypothyroidism is often present and may cause mass to enlarge and become symptomatic.[2] The choice of treatment of patients depends on various factors including size of lesion, the presence of local symptoms, the age of patients, status of thyroid gland and presence of any complication.3 Here we present a case of 17 year-old girl with mass at the base of tongue presenting with difficulty in breathing, which was excised surgically.

Case History

A 17 year-old adolescent girl presented with mass at the base of tongue with difficulty in breathing since ten years and difficulty in swallowing with occasional choking episodes (during sleep) since last one month. Her father initially noted the mass incidentally when she was seven years of age. The mass gradually increased in size but was asymptomatic until one month back when the patient started having choking spells during sleep.

The oro-pharyngeal examination revealed a hemi-spherical mass at the base of tongue. The mass was moving with deglutition. Initially a diagnosis of vallecular cyst was made. On palpation, the mass was non-tender, non-fragile and smooth. Indirect laryngoscopy was not possible because the mass was obstructing the passage. No lymphadenopathy was found in the neck. Plain radio-graph soft tissue neck lateral view showed the soft tissue shadow at the

level of base of tongue just above the epiglottis. CT scan showed the well defined hyper-dense soft tissue mass of size 2.6 x 2.4 x 2.4 cm arising in the right side of vallecula seen in the mid line at the base of tongue. Rest of oro-pharyngeal and naso-pharyngeal airway was normal. Ultra sonography of thyroid showed small left lobe with single colloid nodule in it. Right lobe of thyroid was not visualised. Radio-isotope scanning for thyroid was not performed due to economic limitation. Thyroid profile was with in normal range. The patient underwent surgery and tissue sent for histo-pathological examination.

Gross: We received mucosa covered soft tissue piece measuring 2.5 x 2.5 cm. Cut surface was brown, translucent.

Microscopy: Sections show numerous variable size thyroid follicles lined by flattened and cuboidal epithelium and filled with colloid. These follicles are present beneath the lingual mucosa and lingual lymphoid follicles (figure 1-5)

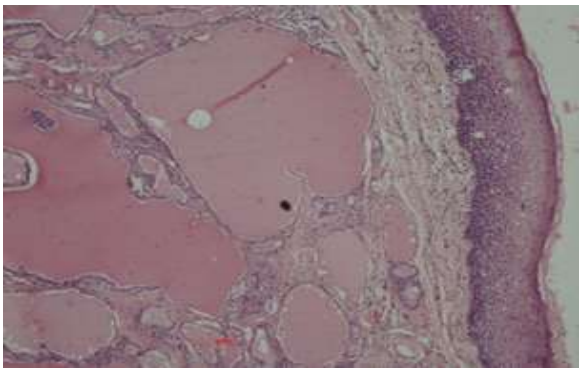


Figure 1 Haematoxyl in and eosin stained sections showing variable sized thyroid follicles beneath the lingual mucosa (Low power view)

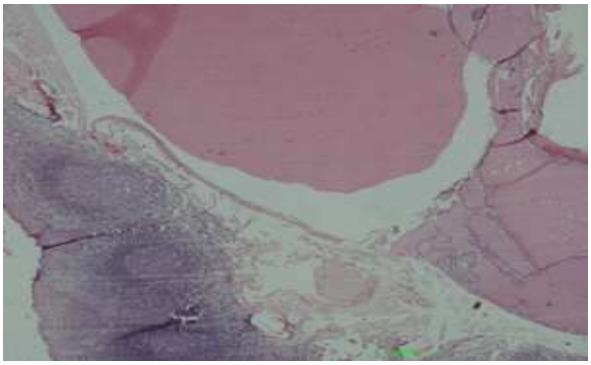


Figure 2 Haematoxyl in and eosin stained sections showing thyroid follicles beneath lingual lymphoid follicles (Low power view)

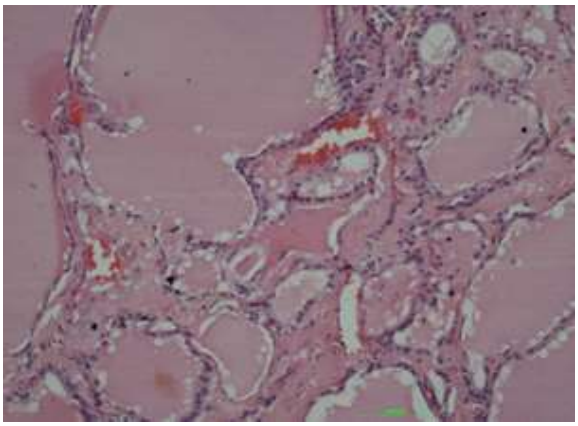


Figure 3 Haematoxyl in and eosin stained sections showing thyroid follicles lined by actively secreting epithelium and filled with colloid (High power view)

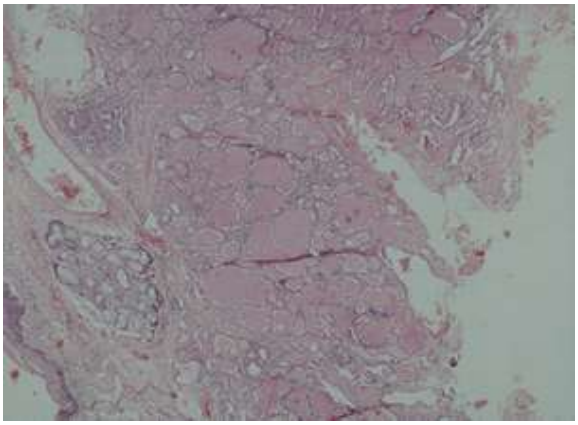


Figure 4 Haematoxyl in and eosin stained sections showing thyroid follicles and sublingual glands (Low power view)

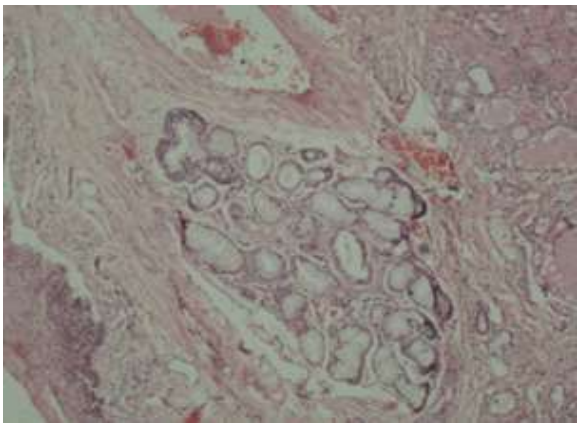


Figure 5 Haematoxyl in and eosin stained sections showing thyroid follicles and sublingual glands (High power view)

Post operative thyroid profile performed after 3 weeks of surgery showed hypothyroid hormone status.

Discussion

The thyroid primordial develops in a 3-4 mm embryo as an endodermal bud from the anterior floor of the pharynx and descends down on either side of the trachea to its pre-tracheal site where it fuses with the caudal prolongation of the fourth pharyngeal pouch to form the thyroid gland. Failure to this process results in ectopic thyroid. Arrest in descend can occur just below the foramen caecum (lingual), between the genio-hyoid and mylo-hyoid muscles(sub-lingual), or just above or below the level of hyoid bone.[4] Other rare sites are larynx, trachea, oesophagus, mediastinum and the heart.[5] Of all ectopic thyroid, 90% are found on lingual dorsum.

Lingual thyroid is a rare developmental anomaly characterised by an aggregate of thyroid tissue in the midline of the base of tongue, between the circumvallate papillae and the epiglottis.[6] The first case of lingual thyroid was reported by Hickmann, who described a 16-hour-old female infant who died of suffocation caused by a mass obstructing the oro-pharynx.[7] The incidence of lingual thyroid varies between 1:3000 and 1:100,000[8] and affected individual have no other thyroid tissue in 70% of cases. Lingual thyroid is generally found in two main age related groups: one consist of children who often suffer from developmental anomalies and mental retardation, the second group presents with the onset of symptoms of dysphagia and oro-pharyngeal obstruction before or during puberty.[9]

The pathogenesis of this condition remains unclear. It has been postulated that maternal anti-thyroid immunoglobulin may arrest the descent of the gland and predispose the patient to poor thyroid function later in the life.

The age at presentation ranged from 6 to 74 years[10] with marked preference towards females, the ratio ranging from 4:1 to 7:1.[11] The clinical evidence oh hypothyroidism is found in up to 33% of the patient.

Even though most of the lingual thyroid glands contain histologically normal, or adenomatous tissue, there are reports of carcinoma, foetal or micro-follicular adenoma arising with in the lingual thyroid.[2] Only one-third of the patients with lingual thyroid have thyroid tissue in the neck, as in the present case.

Most of the times, it is asymptomatic with small size (less than 1cm)[12] but sometimes, as in the present case, it can attain larger dimensions and cause symptoms pertaining to airway obstruction. This makes the present case extremely rare because of the associated neck thyroid mass and normal pre-operative thyroid status.

The clinical management of lingual thyroid remains somewhat controversial because of paucity of data in the literature. The best initial guide to treatment is the presence or absence of symptoms. The use of suppressive therapy with exogenous thyroid hormone is the mainstay of medical treatment. The goal of therapy is to suppress TSH and thereby remove the stimulus for gland enlargement. Surgical therapy is appropriate for patients with clinical signs of upper airway obstruction and severe dysphagia or when malignant degeneration is suspected. Ablative radiotherapy is reserved for older patients when surgical therapy is not appropriate.[1]

Conclusion

We describe the occurrence of an ectopic lingual thyroid. Although not uncommon, this possible location is worth bearing in mind as a possible developmental anomaly. Diagnosis is based on history, physical, radiographic examination, thyroid function test and finally on histo-pathological study. Management of these lesions varies in different patients. The available options in management are surgical excision or radio-iodine therapy.

REFERENCES :

1. Kumar V, Nagendhar Y, Prakash B, Chattopadhyay A, Vepakomma D. Lingual thyroid gland : clinical evaluation and management. Indian journal of paediatrics 71(12):62-64, 2004
2. Tuli BS, Arora S, Soni D, Thapar T. Lingual thyroid with coexisting normal thyroid (one lobe) in neck. Journal of oral and maxillo facial pathology 12(1);23-25,2008
3. William JD, Sclafani AP, Slupchinskij O, Douge C. Evaluation and management of the lingual thyroid. Ann otol rhinol laryngol 105:312-316, 1996
4. Hazarika P, Siddiqui SA, Pujary K, Shah P, Nayak DR, Balakrishnan R. Dual ectopic thyroid: a report of two cases. J Laryngol Otol. 1998 Apr;112(4):393-5
5. Giovagnorio F, Cordier A, Romeo R. Lingual thyroid: value of integrated imaging. Eur Radiol. 1996;6(1):105-7
6. Baldwin RL, Copeland SK. Lingual thyroid and associated epiglottitis. South Med J. 1988 Dec;81(12):1538-41
7. Hickman W. Congenital tumor of the base of the tongue, pressing down the epiglottis on the larynx and causing death by suffocation. Trans Pathol Soc Lond 1869;20:160-1
8. Gallo A, Leonetti F, Torri E, Manciocco V, Simonelli M, DeVincentiis M. Ectopic lingual thyroid as unusual cause of severe dysphagia. Dysphagia. 2001 Summer;16(3):220-3
9. Kalam A, Tariq M. Lingual thyroid gland: Clinical evaluation and comprehensive management. Ear, Nose & Throat J. 1999; 78(5): 340-9
10. Bukachevsky RP, Casler JD, Oliver J, Conley J. Squamous cell carcinoma and lingual thyroid. Ear Nose Throat J. 1991 Aug;70(8):505-7
11. Alderson DJ, Lannigan FJ. Lingual thyroid presenting after previous thyroglossal cyst excision. J Laryngol Otol. 1994 Apr;108(4):341-3
12. Bayram F, Külahli I, Yüce I, Gökçe C, Cagli S, Deniz K. Functional lingual thyroid as unusual cause of progressive Dysphagia. Thyroid. 2004 Apr;14(4):321-4