

Case Report - Developmental Defect

Lingual thyroid

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ABSTRACT

Lingual thyroid is an abnormal mass of ectopic thyroid tissue seen in base of tongue caused due to embryological aberrancy in development of thyroid gland. Most of the ectopic tissue is seen in the tongue.

Their identification and proper management is essential since they may be the only functioning thyroid tissue occurring in the body. We report a case of lingual thyroid in a 40 year old female patient who was hypothyroid with posterior swelling of tongue. Tc-99 scintigraphy confirmed the clinical diagnosis and surgical excision of entire tissue was done by midline mandibular split osteotomy and patient was placed under lifelong thyroxine replacement. Follow up showed excellent results with minimum patient discomfort.

Keywords: Lingual thyroid, technetium-99, scintigraphy, tongue

INTRODUCTION

Lingual thyroid is a rare clinical entity of the oropharyngeal region caused by failure in descendance of thyroid gland to its normal position during embryogenesis. Although lingual thyroid is an accepted terminology, the term "ectopia" may be used better to describe those general conditions where thyroid tissues are seen in places other than tongue. Site of occurrence of this aberrant tissue may be used as a prefix, as in lingual thyroid.^[1] The exact pathogenesis of this accessory thyroid tissue is not clearly understood, it generally arises from unobliterated thyroglossal duct epithelium.^[2]

The thyroid gland normally descends along the midline with growth of the neck but at times all or part of the gland may fail to migrate along the path from ventral floor of the pharynx to its normal location in front of trachea over thyroid cartilage and it sequesters within the tongue substance giving rise to an embryological defect with location at the base of the tongue, in a zone posterior to the circumvallate papillae.^[3] Female predominance of occurrence is seen seven times higher than male counterpart. They may also occur in more than one ectopic site in the human body. Literature shows that about 70% of patients with ectopic thyroid are hypothyroid.^[4] Histologically, they resemble normal thyroid parenchyma. Clinically lingual

thyroid may manifest with symptoms such as dysphagia, dysphonia, or even bleeding, and upper respiratory tract obstruction. Technetium (Tc-99) scintigraphy is most important diagnostic technique employed in the identification of this ectopic thyroid.^[5]

CASE REPORT

A 40-year-old female patient came to our department with a chief complaint of swelling of the posterior region of the tongue, with a foreign body sensation in the throat and gradual progression of difficulty in swallowing for past 1 year. She also complains of slight difficulty with phonation since 1 month for which she thought the necessity for getting medical attention. Clinical examination did not reveal any extraoral swelling or lymphadenopathy. Intraorally, on indirect laryngoscopic examination showed a well-defined circumscribed swelling on the posterior aspect of the tongue.

Routine blood investigation and Thyroid function tests carried out in the patient revealed an increased thyroid-stimulating hormone (TSH) value with decreased T3 and T4 level, confirmed that the patient was hypothyroid. Ultrasound study revealed the absence of a normal thyroid gland. Diagnosis of a lingual thyroid was made based on the site specific and characteristic nature of lingual thyroid. Surgical excision was



Figure 1: Incision



Figure 2: Oblique bone cuts

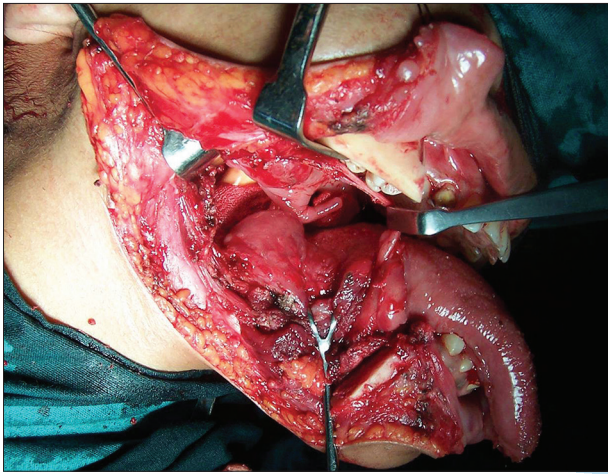


Figure 3: Tumor exposed

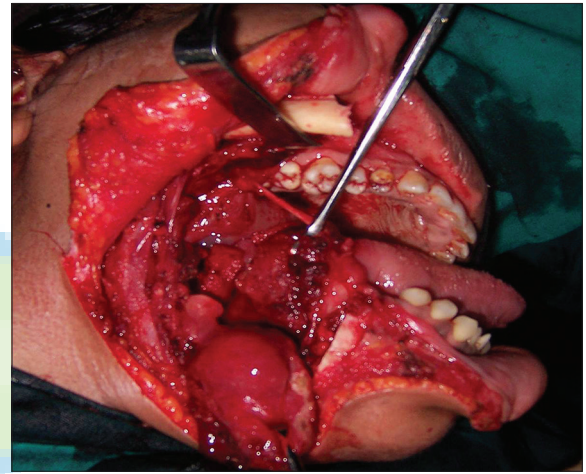


Figure 4: Tumor excised

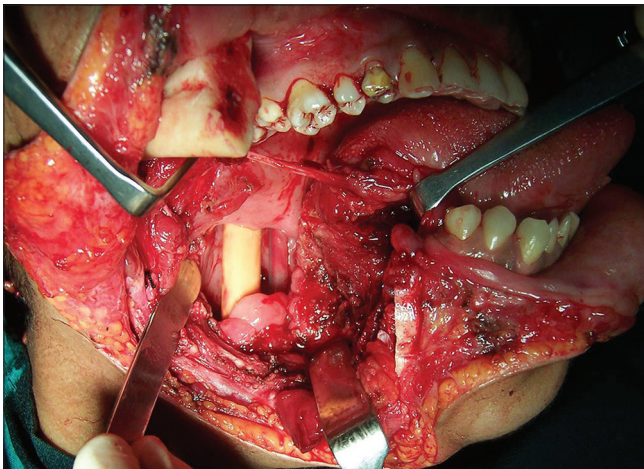


Figure 5: Surgical defect



Figure 6: Mandible repositioned and plated

planned under general anesthesia, we choose lip split [Figure 1] mandibulotomy procedure because this approach would provide a good wide exposure to base of tongue and also suspicion of a vascular tumor was not ruled out, miniplates were adapted and osteotomy cuts were done in the parasymphysis

region in an oblique fashion [Figure 2] mylohyoid muscle was dissected and mandible was swung outward, to reach the base of the tongue, growth was exposed [Figure 3] and removed in toto [Figures 4 and 5]. The osteotomized mandible was repositioned and plated [Figure 6] and primary closure

was achieved [Figure 7]. Excised specimen was sent for histopathology [Figure 8]. Postoperative period was uneventful; speech improved and dysphagia was considerably reduced.



Figure 7: Final closure

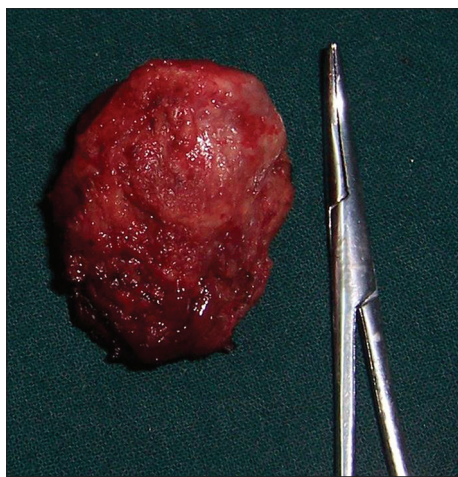


Figure 8: Excised specimen

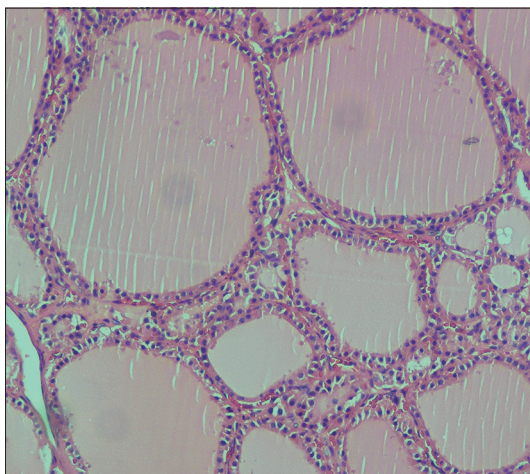


Figure 9: Histopathology shows thyroid follicles lined by columnar cells with eosinophilic colloid inside the follicles

Postoperatively, the patient was placed on replacement therapy with thyroxine. Follow-up showed excellent results with rapid healing of surgical site and return of the normal function. Routine thyroid function tests were within normal limits. Histopathology report confirmed the diagnosis of lingual thyroid [Figure 9].

DISCUSSION

Hickman in 1869 was the first person to report about the presence of lingual ectopia of the thyroid gland. The reported incidence of this condition has been 1 in 100,000 cases which shows the rarity of this ectopic lesion.^[1,2]

The exact pathogenesis regarding the lingual thyroid is not known, but generally it has been accepted by most of the literature that the main cause lies in aberrant embryogenesis. Failure of descendance of medial anlage of thyroid gland from base of tongue to its normal position in front of trachea leads to this aberrant growth referred to as lingual thyroid and usually goes unnoticed until late adolescence or early adulthood.^[6,7]

The literature did not show any age predilection, although Thomas *et al.* found that the peak incidence of occurrence is seen in the third decade of life. Male to female ratio of occurrence is documented as 1:7.^[1,4] Lingual thyroid is mostly seen in females particularly during puberty, pregnancy, or menopause when the plasma TSH level increases in the body leading to hypertrophy of ectopic tissue.^[8]

Other rare sites of occurrence such as submandibular, prelaryngeal, tracheal, laterocervical, esophageal, and substernal have also been reported. The most frequent sites of ectopic thyroid tissue are lingual, sublingual, thyroglossal, laryngotracheal, and lateral cervical. Thyroid tissue can also be found, albeit extremely rarely, in remote structures that were associated with the thyroid anlage during development, including the esophagus, mediastinum, heart, aorta, adrenal, pancreas, gallbladder, and skin. Ectopic occurrence of thyroid tissue usually occurs in a single site although presence of dual ectopic tissue in lingual and subhyoid region has been reported.^[9]

Montgomery in an extensive review done in 1935 have shown that 70% of patients with lingual thyroid have hypothyroidism, as in our case because of absence of normal thyroid tissue.^[10] Conditions such as thyroid adenoma, hyperplasia, inflammation, and carcinoma which may affect the thyroid gland in normal position are also seen in ectopic tissues.^[4]

Clinical symptoms usually seen in patients may range from dysphagia, dysphonia, snoring with a plumy voice, sore throat, occasional bleeding and upper airway obstruction^[11,12] though most of them remain asymptomatic until late adolescence. Rare cases of sleep apnea syndrome due to lingual thyroid have also been reported in the literature.^[13] When acute symptoms develop due to lingual thyroid, these may include sensation of a foreign body or lump in the throat, dysphonia, dysphagia, orthopnea, or dyspnea. The development of acute local symptoms in ectopic thyroid is dependent on the size of the gland and the presence of

other acute processes, such as bleeding. The definitive treatment of acute compressive symptoms from lingual thyroid is surgical excision of the ectopic tissue.^[14]

The surface of the lesion is usually smooth or lobulated, and the color may vary from bluish-red to pure red, with occasional.^[7,15] Histologically, the lingual thyroid is similar to normal thyroid parenchyma. Other histological pattern often seen in this ectopic tissue includes the fetal adenoma type, colloid type, and hyperplastic type.^[16] Our case showed a goitrengous stimulus-induced colloidal changes in thyroid parenchyma also.

The differential diagnosis for lingual thyroid should include vascular tumors, telangiectatic granuloma, teratomas, and benign or malignant processes in the posterior region of the tongue.^[14,16] Lingual thyroid should be investigated based on thyroid function tests and scintigraphy. Increased TSH value with decreased T3 and T4 level are usually seen in thyroid function test.^[17]

In our patient, I-131 thyroid scan showed a nondescended thyroid tissue in the base of the tongue. Radionuclide Tc-99 scintigraphy is a powerful tool in demonstrating any functional thyroid tissue other than lingual thyroid in the body. But this radionuclide scan may give false positive results in case of physiological uptake by salivary glands and nasal mucosa, or pathological uptake as seen in meningioma, dacryo-cystitis, prosthetic eye, sinusitis, dental disease and sialoadenitis.^[18] Radiological examinations are done prior to surgery which may include ultrasound, computed axial tomography in the coronal, sagittal, and axial planes and magnetic resonance imaging to provide useful information regarding other thyroid sources.^[19] Fine-needle aspiration cytology^[20] is also an accurate diagnostic method although thyroid tissue may show more enhancement. Very few studies have found labeled thyroglobulin as a powerful immunohistological marker to confirm the thyroid origin of the tissue.

Treatment modalities employed in lingual thyroid depends on factors such as a general condition of patient, size, and degree of discomfort. Euthyroid patients and asymptomatic patients are followed-up regularly without any treatment. Supplemental thyroxine should be given in symptomatic hypothyroid patients.

Thyroxine suppresses TSH stimulation and minimizes goitrous enlargement.^[7] Failure of medical therapy, obstructive symptoms, and complications such as ulceration, bleeding, cystic degeneration, or malignancy are indications for surgery. Despite providing limited accessibility, transoral approach with cold instruments with mono-polar coagulation and laser CO₂ is most widely employed technique for smaller lesion as they avoid injury to deep neck structures such as lingual nerve. Other methods usually used include lateral pharyngotomy, suprahyoid, and transhyoid techniques. Occasionally, midline mandibular split osteotomy is employed for larger lesions.^[6,8,20]

Ablative radioiodine therapy is an alternative approach recommended in older patients or patients who are unfit for surgery. This treatment should be avoided in children and young adults because of slow response to medication, fibrosis, and dependence on lifelong thyroid hormone replacement and often the systemic doses required have potentially damaging effects on the gonads or other organs.^[18]

CONCLUSION

Malignant transformation of lingual thyroid is rather a rare entity. Lingual thyroid may be the only thyroid producing tissue in the body as in our case. Hence, careful evaluation using adequate diagnostic techniques and appropriate treatment modalities become important as a mere misinterpretation from part of the surgeon may lead to lifelong misery and discomfort to the patient.

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