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Case Report

CASE REPORT

Lingual Thyroid Presenting as an Asymptomatic Pedunculated Oropharyngeal Mass

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ABSTRACT

Lingual Thyroid is a rare developmental anomaly and constitute 90% of ectopic thyroids, first described by Hickman in 1869. Lingual Thyroid are usually symptomatic and commonly cause dysphagia and dyspnea and are rarely asymptomatic. They are usually seen arising from the tongue base in the midline. We report a very rare case of Lingual Thyroid in a 38-year-female presenting as an asymptomatic pedunculated mass in the oropharynx arising from the tongue base from the left side. The occurrence of Lingual Thyroid as a pedunculated virtually asymptomatic mass is very rare and literature search did not reveal a case of pedunculated Lingual Thyroid in this region. This case report highlights that Lingual Thyroid very rarely can present with pedunculated superficial mass and should be considered in the differential diagnosis of oropharyngeal masses.

Key Words: Ectopic thyroid, Lingual thyroid, Pedunculated mass

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Introduction

Ectopic thyroid is a rare developmental anomaly first documented by Hickman in 1869, which results from fault in descent from the foramen cecum to the pre-tracheal position (in front of 2nd, 3rd and 4th tracheal rings) and the thyroid tissue may be sequestered anywhere along this embryologic path.[1] In case of lingual thyroids (LTs) which constitute 90% of ectopic thyroids, a remnant or whole of the thyroid fails to descent and is found at the base of tongue.[2] Microscopic thyroid tissue is present in 10% population at this location of which 0.01% develop lingual thyroids. Usually LTs are asymptomatic are very rare and incidentally found.[3] We present a very unusual and rare case of Lingual Thyroid, which presented to us without any classic symptoms and as a pedunculated swelling from the tongue base from the left side. The unusual presentation, diagnosis and management is described. Literature search did not reveal any case presenting as an asymptomatic pedunculated LT, however a case of LT placed superficially on tongue which posed a threat to the airway was noted.[4] The rarity is worth documenting this case, and highlights the importance of considering LT in the differential diagnosis of oropharyngeal masses.

Case Report

A 38-year-old diabetic female presented to the Otolaryngology outpatient with a 3 days' history of sore throat. Intraoral examination was consistent with acute pharyngitis and interestingly a post lingual swelling, which was provisionally diagnosed as a vallecular cyst. An endoscopic examination a week later following treatment

of the acute pharyngitis revealed a pedunculated 2 cm diameter mass arising from the tongue base on the left side with smooth surface (figure 1). Other intraoral structures were normal, with palatine and lingual tonsils situated at their normal locations. With a barely palpable thyroid in its normal anatomical location, no other swelling/ palpable lymph nodes was noted in the neck.

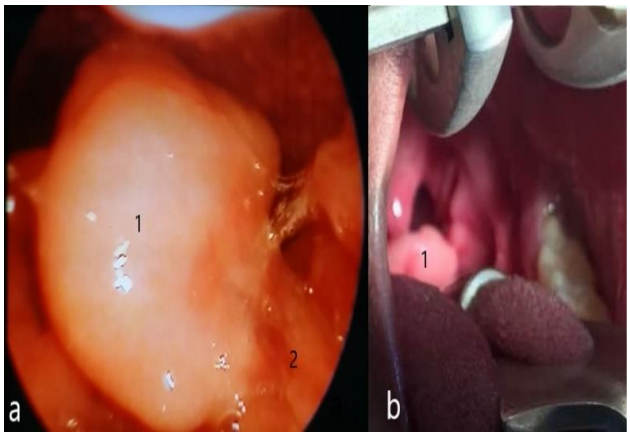

Figure 1: Photograph of LT a) Preoperative endoscopic view showing LT arising from tongue base with a peduncle and b) Peroperative view with Dingman's mouth gag in place and showing LT (1) ready to be removed

Patient's baseline laboratory investigations, including thyroid function tests were within normal limits. Radio nucleotide imaging with Tc 99m-pertechnetate revealed a normal-sized thyroid image with homogenous radiotracer uptake in the pre-tracheal location, however swelling at the base of tongue showed tracer uptake suggestive of LT (figure 2).

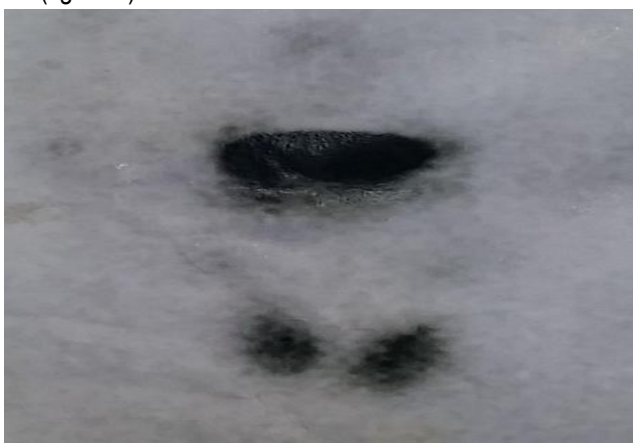

Figure 2: Thyroid Scan with Technetium (Tc 99m) showing isotope uptake at the normal thyroid location in the neck as well as uptake at the tongue base.

Magnetic Resonance Imaging (MRI) scan with contrast (figure 3) revealed a soft tissue density lesion measuring 12 mm TR x 13 mm AP x 16 mm CCL in oropharynx arising from the left side of the base of tongue and projecting within oropharyngeal cavity. It demonstrated intermediate signal on T1W and high signals on T2W sequences with significant post contrast enhancement, consistent with lingual thyroid. Thyroid gland was also seen located in its pre-tracheal location. No cervical lymphadenopathy noted. Keeping in view the posterior placement on tongue base and pedunculated nature of the swelling, FNAC was not considered.

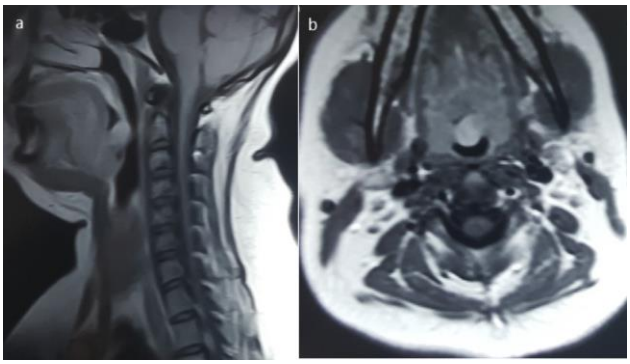

Figure 3: MRI scan (Gadolinium enhanced) with both sagittal view (a) and axial view (b), showing a soft tissue density lesion (LT) measuring 12 in transverse, 13 mm in anterio-posterior and 16 mm in cranio-caudal direction, arising from left side of the tongue base and projecting into the oropharynx.

After taking informed consent, trans-oral surgical removal of the mass was done under general anaesthesia and endotracheal intubation. The mass was carefully approached in tonsillectomy position with Dingman's mouth gag in place followed by removal of the mass with ligation of the pedicle arising from the tongue left side of the tongue base (figure 4).

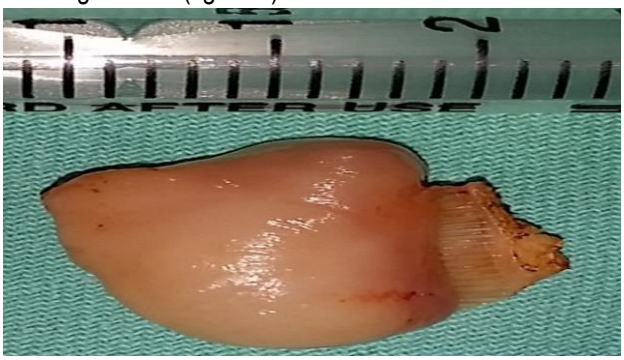

Figure 4: Surgical specimen of LT, measuring approx. 2 cm craniocaudally and transversely with a pedicle.

Post-operative recovery was uneventful and diet restored 6 hours post operatively. The diagnosis was supported by histopathological examination, which revealed normal thyroid tissue (figure 5). No evidence of malignancy noted. One-year follow-up was maintained with no recurrence.

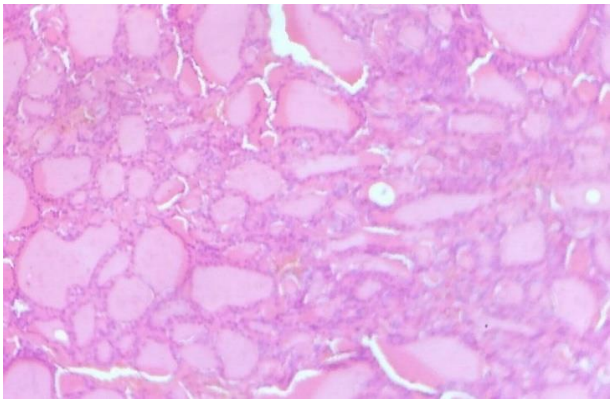

Figure 5: Photomicrograph, showing thyroid follicles which are lined by columnar cells. Follicles are filled with eosinophilic colloid (H&E: 10X100).

Discussion

Ectopic thyroid is a developmental anomaly, commonly occurring at four areas of the aerodigestive tract including lingual, sublingual, thyroglossal and intra-laryngotracheal, with lingual location being the commonest with prevalence between 1:100000 to 1:300000.[5] Patil YS et al., noted a prevalence of lingual thyroid of 1:2400.[1] Ting Gu et al., reported a frequency of LT as 64 %, sublingual 17 %, in 12 % it was found at two locations and, other locations in 7 % cases.[6] A female predominance is noted[6] with a male to female ratio of 1: 5.[1] Usual age of occurrence is between 5 months to 40 years of age[7] and commonly arising at growth spurts like puberty and menstrual age. In 70 -80% cases, ectopic thyroid is the only functional thyroid tissue.[8] Pathogenesis is uncertain.[9] Ting Gu et al., in a study found high levels of marker, TTF-1(Thyroid transcription factor-1) in ectopic tissue and postulated that this could be related to abnormal development of embryo leading to the ectopy.[6] LTs are usually symptomatic. Symptoms include swallow difficulty in swallowing,[1,9,10,] dysphonia,[1,8] pain, cough, airway difficulty,[11] hemorrhage,[12] and patient may also present with symptoms of hypothyroidism [12]. Rashid et al., in a local study have reported a case of snoring and sleep apnea [13], and severe sleep apnea may also result.[14] Very rarely it may be asymptomatic and found incidentally [3], as in our case. Presence of a mass at the foramen cecum should raise suspicion and warrant thorough Head and Neck evaluation, which must include neck palpation for thyroid gland, neck nodes and laryngeal examination. When available endoscopic examination can give important information like status of larynx and hypopharynx, exact site and size of the mass including surface characteristics like ulceration, bleeding etc.[12] In this case endoscopic examination was rewarding and site, size, surface and a peduncle was outlined, making surgical decision easier.

Literature highlights role of preoperative imaging modalities like thyroid scintigraphy and ultrasonography.[1] According to Fiaschetti et al., Radionucleotide imaging with Tc 99m-pertechnetate can be confirmatory for diagnosis of LT. It can detect ectopic thyroid tissue including LT and confirm presence or absence of thyroid in the normal pre-tracheal location [3, 7]. Ultrasonography (US) has contributory role.[7,15] In one study Color Doppler US showed a sensitivity of 90% compared to 70% for Grayscale US and MRI [16]. MRI, and CT are useful modalities in doubtful cases. MRI has been used for the diagnosis of lingual thyroid[18] and displays higher density for thyroid tissue on T1 and T2-weighted images.[15] In our case thyroid scintigraphy revealed the thyroid gland in its normal location as well as the LT and an MRI scan demonstrated the size of the pedunculated LT and contrast enhancement. Fine needle aspiration cytology (FNAC) is helpful in differentiation between benign and malignant lesions.[18] Keeping in view the pedunculated nature of the mass, need of FNAC was not felt rather excision biopsy was preferred.

Differential to be considered with lingual thyroid include other tongue base swellings like Vallecular cyst, lingual tonsils, mucous retention cysts, hemangioma, lymphangioma, thyroglossal cysts and salivary tumors,[19] however in this case, rare conditions like Lingual Osseous Choriostoma, that can present as pedunculated mass were also considered.[20] Rarely carcinoma may also develop in a LT.[2] Surgical removal is the preferred treatment option for symptomatic cases like those having airway compromise, severe dysphagia and hemorrhage.[12] Thyroid hormone suppression therapy for early cases[12,21] and in refractory cases, levothyroxine for suppression and

radioiodine ablation is considered.[1] A number of surgical approaches are advocated, depending upon the size, extent and symptomatology including endoscopic, trans-oral, and trans-cervical and combined approaches.[22] Trans-oral approach is the least traumatic and provides reasonable approach with better recovery,[1] it also proved most suitable for the present case.

The present case is very rare and unusual, with an asymptomatic, pedunculated LT measuring 12 mm TR x 13 mm AP x 16 mm CCL length arising from the left side of the base of tongue and projecting within oropharyngeal cavity along with a normal thyroid in the pre-tracheal location on MRI scan and thyroid scintigraphy in a 38-year-female who visited the otolaryngology outpatients for upper respiratory tract infection, and thus incidentally picked and diagnosed to have LT. Though asymptomatic, with thyroid in normal location and normal hormone analysis, keeping in view the pedunculated nature a trans-oral surgical excision was done without any complication and recurrence.

Conclusion

Lingual thyroid may rarely be encountered as asymptomatic pedunculated mass arising from the tongue base and should be considered in the differential diagnosis of oropharyngeal masses.

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