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**RESEARCH ARTICLE**

# ECTOPIC THYROID AT THE BASE OF THE TONGUE: A DIAGNOSTIC AND THERAPEUTIC CHALLENGE

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**ABSTRACT**

Lingual thyroid is a developmental anomaly with thyroid tissue usually seen in the base of the tongue. It arises from the unobliterated thyroglossal duct epithelium. It represents the normal thyroid parenchyma histologically. Clinically it can be mistaken for minor salivary gland tumors and various benign cystic lesions. Fine needle aspiration cytology (FNAC) coupled with radiology aids in the proper diagnosis of lingual thyroid and helps in planning further treatment. We present two cases of lingual thyroid diagnosed on FNAC by an intraoral approach.



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## INTRODUCTION

Ectopic thyroid tissue (ETT) is a rare developmental abnormality originating from aberrant embryogenesis during the development of the thyroid gland. It is four to seven times more common in females. It is rarely seen in children (Santangelo, 2016). Lingual thyroid is associated with absence of thyroid gland in normal position in 70% of the cases. ETT can co-exist with the eutopic gland; there are no vascular or anatomic connections between the two in such cases (Santangelo, 2016). This accessory thyroid tissue's exact pathogenesis is not clearly understood; it generally arises from unobliterated thyroglossal duct epithelium (Noussios, 2011). Clinically lingual thyroid manifests with dysphagia, dysphonia, or even bleeding, and upper respiratory tract obstruction and is likely to be mistaken clinically for various cystic lesions and neoplasms especially minor salivary gland neoplasms. Histologically it resembles normal thyroid parenchyma (Kumar, 2015). Here we are presenting two cases of lingual thyroid diagnosed on fine needle aspiration cytology (FNAC).

## CASE HISTORY

**Case report 1:** A 35 year old female patient, came with a history of swelling since two months. She had difficulty in swallowing and speech. She had no significant past medical history. Her laboratory tests were within normal limits. Thyroid function tests revealed euthyroid levels. On physical examination, there was a firm, non- ulcerated, non-tender mass of size 3*2 cm on the posterior midline of the tongue. Neck examination revealed left level 1 palpable lymph node which was clinically insignificant (Figure 1). CT scan showed a round, well defined hyperdense soft tissue lesion situated at the base of the tongue region which was infiltrative in nature. It was approximately 32* 55 mm anteroposteriorly and in lateral dimension. The soft tissue lesion was showing post contrast homogeneous enhancement (Figure 2. A,B). A provisional clinical diagnosis of lingual thyroid was made. Differential diagnosis thought was a minor salivary gland neoplasm. FNAC was done through intra-oral approach. The smears revealed clusters of follicular cells arranged in micro and macro follicular pattern. Few admixed histiocytes were seen on the background of abundant colloid material (Figure 3. A). So, a cytodiagnosis of lingual thyroid was made. An elective surgical procedure in the form of subtotal excision was carried out under general anesthesia. Grossly, the specimen consisted of subtotal excision of posterior tongue swelling measuring 3.4* 2.5*1.2 cm. The overlying mucosa was unremarkable (Figure 4). On a cut section, it showed brownish soft to firm lesions with gelatinous appearance. Microscopic examination revealed normal mucosa with a submucosal lesion comprised of thyroid follicles of varying sizes lined by cuboidal epithelium. The lumen of the follicles contained eosinophilic colloid material.

**Figure 1. Clinical photograph showing nodular swelling on the posterior midline tongue with intact mucosa**

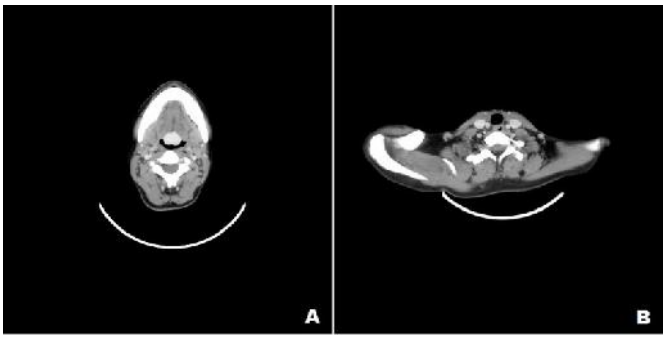


**Figure 2: a) Showing hyperdense soft tissue lesion at the base of the tongue; b)Showing absence of thyroid gland at its usual pre-tracheal location in lower neck**

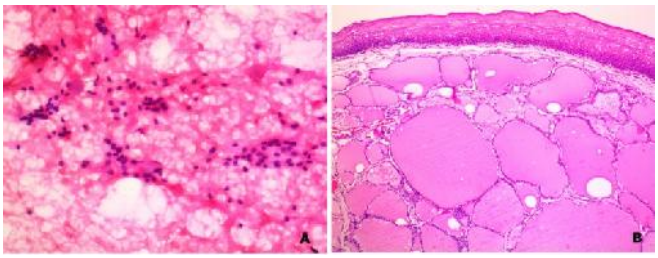

**Figure 3: a) FNAC image (H & E 400X) showing follicular cells arranged in micro and macro follicular pattern with colloid on the background; b) Histopathology (H & E 100X) showing intact squamous epithelium with subepithelial tissue showing follicles of varying sizes lined by low cuboidal epithelium containing abundant colloid**

No features of malignancy were seen. The final diagnosis of lingual thyroid was made (Figure 3. B). Postoperatively, she healed well and was discharged after 5 days. Post operative review CT showed absence of normal thyroid in the neck.

**Case history 2:** A 15 year old female patient came to the clinic with the complaint of pain while swallowing. She had no significant past medical history.

Physical examination revealed firm, nodular, non-tender mass of size 2*2 cm on the posterior midline of the tongue. The overlying mucosa was normal (Figure 5). Clinical diagnosis of lingual thyroid was made with differentials of minor salivary gland neoplasm and lymphoma.

**Figure 4. Gross specimen of excised lingual thyroid**

**Figure 5: Clinical photograph showing nodular swelling on the right posterior tongue with intact mucosa**

FNAC of the lesion was done through intraoral approach. The smears revealed clusters of follicular cells arranged in micro and macro follicular patterns. Few admixed histiocytes were seen in the background of abundant colloid material. No features of malignancy were noted. So, a cytodiagnosis of lingual thyroid was made (Figure 6. A,B).

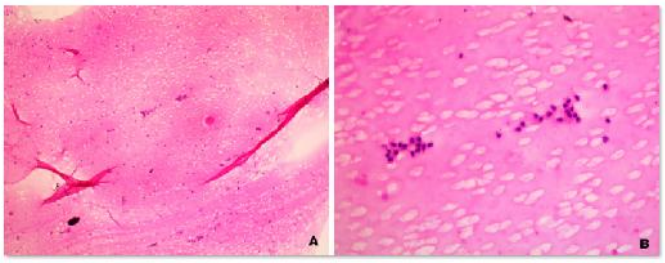

**Figure 6. a)FNAC image (H & E 100X) showing follicular cells clusters on the background of abundant colloid material; b)FNAC image (H & E 400X) showing higher magnification of the same**

Thyroid scan features are suggestive of ectopic sublingual thyroid gland with no functioning thyroid tissue in the thyroid bed. Thyroid function tests revealed euthyroid levels. The patient had refused surgery and is being followed up.

## DISCUSSION

Ectopic thyroid is a rare development of thyroid gland with lingual thyroid being the most common having a prevalence of 1 in 100,000. Lingual thyroid has an incidence of 1 in 3000.(5) Other sites are cervical lymph nodes, submandibular glands, and trachea. ETT has also been found in various other tissues like the cardiac tissue, ascending aorta, esophagus, duodenum, gallbladder, ovary, uterus, fallopian tubes, and vagina.(5) Embryonically, the thyroid gland develops as an endodermal diverticulum between the first and second pharyngeal pouches. At the 7th week of gestation, the thyroid gland is an endodermal pouch in the foramen cecum, which is the remnant of thyroglossal tract. Normally thyroid gland descends along a routine path from foramen cecum in the tongue to the final position in front of trachea (5). The thyroid gland descends typically along the midline with the growth of the neck (3). However, at times, all or part of the gland may fail to migrate along the path from the ventral floor of the pharynx to its usual location in front of the trachea over the thyroid cartilage, and it sequestrates within the tongue substance giving rise to an embryological defect with a site at the base of the tongue, in a zone posterior to the circumvallate papillae (4). ETT is seen at any age but most commonly seen in childhood, adolescence,and around menopause (6). Peak incidence of occurrence is in the 3rd decade of life. The female to male ratio is 4:1. In our cases, both the patients were female and in their third and second decades. About 33-62% of all patients with ectopic thyroid showed hypothyroidism with increased levels of TSH (6). Both our cases were euthyroid. It usually presents as a 2 to 3 cm smooth sessile mass located on the mid-posterior dorsum of the tongue, in the region of the foramen cecum (5). Both masses showed a nodular bulge of 2 cm with normal overlying mucosa. Clinically, lingual thyroid is presented as a mass at the base of the tongue, pink and firm. The most important diagnostic tool in thyroid scan with technetium Tc-99m Sodium; CT and MRI can be used to locate the position of the ETT (5). Another important diagnostic investigation is FNAC done through an intraoral approach. In both our cases the diagnosis was made on FNAC. Differential diagnosis for ETT apart from neoplasms are: Cystic lesions: thyroglossal duct cyst, dermoid cyst, cystic hygroma; benign neoplasms like hemangioma of the tongue, lipoma, lymphangioma. Malignant transformations are very rare (1). Clinical differential diagnosis in our cases were minor salivary gland neoplasm and lymphoma. In approximately 75% of patients the ectopic tissue is the only functioning thyroid tissue in the body. Therefore, it is important to observe that patient at follow-up, being aware of the risk of postoperative hypothyroidism (6). Treatment of a lingual thyroid depends on the age and sex of the patient as well as on the severity of the symptoms and the associated ulceration and hemorrhage. Patients with mild symptoms can be treated successfully by medical suppression by hormonal therapy and radioactive ablation (5,7). Most cases are treated by surgical excision. Surgical treatment includes excision alone or excision with auto transplantation into muscle. Approach for excision is oral, transhyoid, and lateral pharyngotomy (5,7). In the first case, we have used the oral surgical approach for subtotal excision. In the second case, the patient refused surgery and is followed closely. Possibility of ectopic thyroid tissue being the only functioning tissue must be considered and transplantation of the excised ectopic tissue or substitutive hormone treatment is recommended to avoid permanent hypothyroidism. In cases of partial surgical eradication, although transplantation is not necessary, substitutive therapy could be necessary to preserve euthyroid state and to avoid recurrence of the mass. Partial trans-oral ablation is recommended in patients unfit for aggressive surgical approach (6). In our first case, partial thyroid tissue was left back to prevent the hypothyroidism.

### Conclusion

Lingual thyroid is commonly misinterpreted for various other non neoplastic cysts, benign tumors, malignant tumors. High degree of suspicion, radiological correlation and a FNAC through an intraoral approach leads to a correct diagnosis. A preoperative diagnosis helps in relieving anxiety, planning the optimal surgical procedure for the patient.

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