

Navigating obstructive lingual thyroid coexisting with sub-clinical hypothyroidism: A case report

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Abstract

Ectopic thyroid tissue results from the embryonic failure of the thyroid gland to descend along the thyroglossal duct, resulting in the presence of thyroid tissue in an abnormal location. One of the most common locations where ectopic thyroid tissue (ETT) is found is at the base of the tongue, referred to as a lingual thyroid, having an estimated prevalence of 1 out of 100,000 to 300,000 in healthy individuals. We present the case of a 15-year-old female with a progressively enlarging mass at the base of her tongue, associated with dysphagia and sleep apnoea. Hormonal tests revealed hypothyroidism. Adopting a transoral approach, total resection was performed via electrocautery. Histopathology report revealed squamous lined tissue exhibiting mucous gland, confirming the diagnosis of ectopic lingual thyroid. The patient was started on thyroxine maintenance therapy and showed significant improvement.

Keywords: Ectopic thyroid, lingual thyroid, hypothyroidism.

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Introduction

The thyroid gland is a butterfly-shaped endocrine gland, consisting of two lobes connected by a narrow band of tissue called the isthmus. Typically found in the peritracheal region of the anterior neck, it is situated inferior to the thyroid cartilage. Its location is generally at the level of the C5 to T1 vertebrae.¹ Ectopic thyroid tissue results from the embryonic failure of the thyroid gland to descend along the thyroglossal duct, resulting in the presence of thyroid tissue in an abnormal location ETT.² An incomplete regression of the thyroglossal duct is called a thyroglossal cyst, which occurs when the duct itself fails to close.

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The most frequent location of ectopic thyroid tissue (ETT) is at the base of the tongue, referred to as a lingual thyroid. The estimated prevalence of ectopic thyroid varies from 1 out of 100,000 to 300,000 in healthy individuals, with a significantly higher prevalence of 1 out of 4,000 to 8,000 cases in patients with existing thyroid disease.

Lingual thyroid is more frequently observed in females, comprising approximately 70-80% of reported cases.³ Thyroid hormone abnormalities, particularly hypothyroidism, is frequently associated with ectopic thyroid tissue.⁴ We present the case of a fifteen-year-old hypothyroid patient with lingual thyroid.

Case Report

In January 2024, a 15-year-old female presented to the ENT outpatient department at Khyber Teaching Hospital, Peshawar with complaints of dysphagia and sleep apnoea accompanied by loud snoring for the past month. The patient's history revealed that she had a mass at the base of her tongue since childhood, which had progressively enlarged after the onset of puberty. She also reported a change in voice.

On oropharyngeal examination, a reddish-blue, smooth and firm midline mass on the posterior one-third of the tongue was identified. Its lower limit was not palpable. The mass was not associated with pain and did not bleed upon palpation. No Cervical lymph node was noted on neck examination.

According to her laboratory investigations, the total leukocyte count was $6.28 \times 10^3/\mu\text{L}$ (normal: $4 - 11 \times 10^3/\mu\text{L}$) and haemoglobin was 11.1 g/dL (normal: 11.5 - 17.5 g/dL). Under the suspicion of ectopic thyroid, the patient was advised to undergo neck ultrasonography and thyroid function tests (TFTs). Ultrasound imaging did not visualise a normally located thyroid gland in the neck. The submandibular and parotid glands appeared normal. However, a thyroid-like mass measuring approximately 3.3 x 3.5 cm was seen at the base of the tongue, strengthening the diagnosis of congenital lingual thyroid. Hormone assessment revealed free T4 to be 14.5 pmol/L (normal: 10 - 28 pmol/L), T3 measuring 1.74 nmol/L (normal: 0.6 - 2.0 nmol/L) and thyroid stimulating hormone (TSH) detected

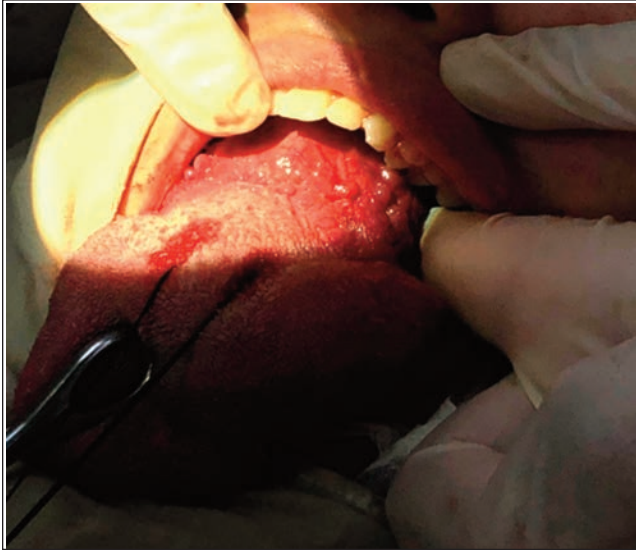


Figure-1: Transoral approach to lingual thyroid at the base of the tongue.



Figure-2: Larger resected mass.

at 11.05 uIU/ml (reference range: 0.3 - 4.2 uIU/ml). These findings indicated primary hypothyroidism, suggesting the lingual thyroid tissue to be non-functional. The patient was prescribed thyroxine 50 mcg, twice daily.

Keeping in view the rapid growth and obstructive nature of the mass, the patient was admitted to the otorhinolaryngology ward for surgical intervention. Under general anaesthesia, the patient underwent endotracheal intubation. Adopting a transoral approach, the tongue was protruded antero-inferiorly to access the lingual thyroid and total resection was performed via electrocautery (Figure 1). Two irregular nodular masses were resected and sent for histopathology. The larger specimen measured 4 x 2.5 x 2 cm (Figure 2) and the smaller mass was 2.5 x 2 cm.

Postoperatively, a soft and cold diet was encouraged.

Histopathology report revealed squamous lined tissue exhibiting mucous glands and thyroid follicles of varying sizes lined by cuboidal cells and filled with colloid. No evidence of malignancy could be determined. A final diagnosis of benign ectopic thyroid was made.

At one-month follow-up, the patient had fully recovered with no active complaints and was started on thyroxine maintenance therapy. Consent for publication of this report was obtained from the patient's guardians.

Discussion

Around the third week of gestation, the thyroid, the body's first endocrine gland to develop, originates between the first and second pharyngeal pouches. Traditionally present at the level of the C5-T1 vertebrae, below the thyroid cartilage, the thyroid tissue sometimes fails to travel along its normal path of descent. This arrested migration gives rise to a rare condition referred to as ectopic thyroid (ET). While the base of the tongue (at the foramen cecum) accounts for the most common site of ET, the tissue can be found anywhere along the thyroglossal duct.¹

There is an increase in size during menstruation, pregnancy, menopause, etc. due to the heightened metabolic requirement for thyroid hormone and associated upsurge in circulating TSH. This accounts for the hypertrophy of the ectopic thyroid tissue.⁵

Clinical manifestations of lingual thyroid can vary from dysphonia to severe airway obstruction along with thyroid hormone abnormalities. Among these, hypothyroidism is seen in approximately thirty-three percent of such patients, as the ETT is the only functioning source of thyroid hormone in roughly 70% of cases.³ In this case, the patient happened to be a subclinical case as her thyroid function tests (TFTs) showed elevated TSH, although, she did not present with any features of hypothyroidism.

Initial evaluation of the patient requires careful palpation and ultrasonography to look for thyroid tissue in its normal position. Additionally, thyroid function tests are necessary to determine hormone levels, and histological analysis may be required, particularly for surgical planning or when there is suspicion of malignancy.⁶

Confirmatory diagnosis of a lingual thyroid requires key imaging techniques like technetium (Tc99m) scintigraphy, iodine 131, iodine 123 and CT scan for precise information on the ETT's location, size, and function.⁷ The utilization of radioiodine therapy was not adopted in the case under study due to its contraindication in children under the age of 15 years. At the same time, it must be noted that while

technetium scintigraphy delivers a lower radiation burden to the body compared to the radioisotopes of iodine making it comparatively safer for use in children, this technique remains inaccessible to the majority of low-resource countries resulting in a significant reliance on ultrasonography as the diagnostic tool of choice. Available treatment options for lingual thyroid include levothyroxine suppression therapy, radioactive iodine ablation, and lingual thyroidectomy.⁸

Conclusion

This report highlights a case of ectopic thyroid tissue in a young female presenting with obstructive symptoms and abnormal thyroid function tests. It underscores the importance of considering lingual thyroid in the differential diagnosis of oropharyngeal masses. Thorough examination and investigation are essential to guide management. In this case, early surgical intervention via transoral resection of the mass effectively relieved the patient's symptoms and confirmed the diagnosis. Lifelong thyroxine replacement remains essential due to the absence of functional orthotopic thyroid tissue.

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Author Contribution:

MI: Concept, design, data collection, clinical assessment and writing.

MH: Data collection, clinical assessment and writing.

UF & WZ: Writing.

IU: Data collection, clinical assessment, critical review, final read and supervision.