

## CASE REPORT

# Imaging of the Lingual Thyroid: A Case Report and Management

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## ABSTRACT

Lingual thyroid is a rare developmental abnormality characterized by the failure of the thyroid gland, or remnants, to descend from anywhere along its embryologic path of origin at the foramen cecum to its normal eutopic pretracheal position. Awareness of the anatomic course of the embryologic thyroid primordium, typical and atypical locations of ectopic thyroid tissue may aid in diagnosis. The reported incidence of lingual thyroid is 1 in 100,000, and it is more common in females, with a female to male ratio ranging between 3:1 and 7:1. Lingual thyroid located at the base of tongue often present with hypothyroidism, usually asymptomatic but may cause dysphonia, dysphagia, upper airway obstruction, and hemorrhage. In the current case report, we present the imaging characteristics of lingual thyroid occurring in a 12-year-old female patient. Partial endoscopic removal of lingual thyroid was performed, followed by substitutive exogenous thyroid hormone therapy. The purpose of this study is to discuss the radiological characteristics and therapeutic issues in the management of lingual thyroid.

**Keywords:** Dysphagia, Dyspnea, Imaging, Lingual thyroid.

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## INTRODUCTION

Lingual thyroid is a rare congenital anomaly, originating from aberrant embryogenesis, and characterized by the failure of thyroid gland, or remnants, to descend from the foramen cecum to its usual prelaryngeal site. Lingual thyroid is relatively uncommon, and carcinoma of the lingual thyroid is extremely rare, with less than 40 cases reported. Lingual thyroid (at tongue base) is the most frequent ectopic location of the thyroid gland, although the reported incidence of lingual thyroid is 1 in 100,000 to 300,000 in the general population<sup>1,2</sup> with a female predominance. Ectopic thyroid tissue can also occur above

the hyoid bone (prelaryngeal thyroid), between the geniohyoid and mylohyoid muscles (sublingual thyroid), and in other rare sites, such as the pharynx, trachea, esophagus, lung, breast, mediastinum, precardial sac, heart, adrenal gland, duodenum, and mesentery of the small intestine.<sup>3-7</sup> Radiologic examination of the usual thyroid bed location is clinically important, because in majority of cases, when a lingual thyroid is found, it is the only functioning thyroid tissue with complete absence of an orthotopic thyroid gland in 70 to 80% of the cases.<sup>2</sup> Any subsequent partial or complete resection of the ectopic thyroid tissue will cause severe permanent hypothyroidism or render the patient athyroid.<sup>3,8</sup>

## CASE REPORT

A 12-year-old female presented with a mass at the dorsal aspect of posterior tongue, which had been present for many years with the sensation of a foreign body. The patient also referred to having dysphagia and dyspnea, especially at night. She was normotensive and nondiabetic. She was a known case of hypothyroidism. Upon examination, the patient presented a solid, nontender, sessile, hyperemic pink, spherical mass measuring 4 cm in diameter, covered with intact mucosa, located at the base of the tongue, occupying the oropharynx and obstructing the visualization of the larynx. Examination of the neck revealed no cervical lymphadenopathy and no palpable thyroid gland in the expected cervical location. The serum thyroid profile of the patient showed normal FT3 and FT4 (0.84 ng/mL and 6.56 ng/mL respectively) and elevated thyroid stimulating hormone (TSH) levels (90 ug/mL). Cervical ultrasonography and computed tomography (CT) examination suggested a well-defined and heterogeneous/hypoechoic mass, measuring 21 × 19 × 23 mm in size with distinct margins restricted to the base of the tongue and absence of the thyroid gland in the normal eutopic pretracheal position. In Figures 1 and 2, CT examination revealed aberrant thyroid tissue as a high density mass in the region of the tongue base, and CT scan with intravenous contrast showed homogenous contrast enhancement of the aberrant mass. A thyroid scan with technetium Tc-99m sodium was performed showing a marked midline focal area of isotope uptake in the region of tongue base, thus representing a lingual thyroid. There was no thyroid uptake in the neck.

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**Fig. 1:** Lingual thyroid in a 12-year-old female child with a history of dysphagia and dyspnea. Axial contrast-enhanced CT examination of neck suggests well-defined, sharply enhancing, homogenous, high density mass in the region of tongue. The posterior median sublingual mass measures  $21 \times 19 \times 23$  mm with absence of the thyroid gland in the normal eutopic pretracheal position



**Fig. 2:** Sagittal contrast-enhanced CT image of the same 12-year-old female child shows aberrant thyroid tissue in the base of the tongue that obscures the epiglottic vallecula and displaces the epiglottis, causing narrowing of oropharynx and mild stenosis of larynx

Partial endoscopic removal of lingual thyroid was performed using standard micro-laryngoscopic surgical approach using a bivalve Weerda-type laryngoscope and a Zeiss microscope. The removal of prominent part of thyroid gland achieved sufficient airway space. Postoperative histopathological examination further confirmed ectopic thyroid tissue. Surgery and postoperative recovery was uneventful. By the 6th postoperative day, soft oral feeding was started and the patient was discharged. Substitutive hormone therapy was commenced in order to maintain the euthyroid state. Since at a 6-month follow-up control, postoperative imaging findings showed no evidence of disease with normal lingual mucosa, regular follow-up was advised.

## DISCUSSION

Ectopic thyroid gland is a rare embryological aberration resulting from incomplete descent of the thyroid gland from the foramen cecum to its original position in pretracheal region in the lower neck.<sup>1</sup> Lingual thyroid is defined as the presence of thyroid tissue in the midline at the base of the tongue anywhere between the circumvallate papillae and the epiglottis. Previous studies reported that 33 to 62% of all patients with ectopic thyroid showed hypothyroidism with increased levels of TSH.<sup>3,8</sup> Imaging techniques, such as thyroid scan with Tc-99m or iodine-123 or iodine-131, CT and magnetic resonance imaging are frequently used for the exploration of a lingual thyroid, topographic diagnosis and confirm the presence or

absence of orthotopic thyroid gland. Due to the rarity of this anatomical and clinical entity, there is no consensus about the optimal therapeutic strategy. Most lingual thyroids are asymptomatic and euthyroid and requires no therapy, except for the regular follow-up. Patients with mild symptoms and hypothyroidism, can be treated successfully by levothyroxine replacement therapy (LT4), leading to partial involution of lingual thyroid volume.<sup>9</sup> Ablative radioiodine therapy (I-131) is an alternative approach recommended in geriatric patients or those who are deemed unfit for surgery. This treatment should be avoided in children and young adults as thyroid tissue is often hypoactive and the systemic doses of radioiodine required are generally high. As in our case, the high doses required have potentially deleterious effects on the gonads or other organs.<sup>10</sup> Literature search showed surgical treatment of lingual thyroid in the neck depends on the parameters, such as patient's age, size of the lesion, severity of local symptoms (oropharyngeal obstruction, dysphonia, and dysphagia), associated complications (hemorrhage, ulceration, malignancy, or cystic degeneration), and functional thyroid status.<sup>9,11,12</sup> Several surgical approaches for lingual thyroid have been discussed in the literature, including surgical resection of the mass with external approaches, such as transoral ablation of the mass and the transhyoid, suprahyoid and lateral pharyngotomy. The former is usually preferred for small masses and successful outcomes may be achieved by using laser diiodine, monopolar coagulation, or the CO<sub>2</sub> laser. Soft diet is usually tolerated sooner than in other

external approaches. Also, it avoids injury to deep neck structures; thus, possible complications, such as lingual nerve injury, deep cervical infections, fistula formation, and visible scar, are avoided.<sup>13</sup> The latter approaches with temporary preoperative tracheotomy can probably provide better control of bleeding and are recommended for larger masses.<sup>11,12</sup> In case of large lesions, younger patients or lesions deeply located in the caudal part of the base of the tongue, total thyroidectomy is recommended because of risk of malignant transformation.<sup>14</sup> However, in our opinion, transplantation of the thyroid tissue is not necessary, substitutive hormone replacement therapy with regular monitoring could be the most appropriate choice.

## CONCLUSION

Findings in this case emphasize the rarity of the lesion and the significance of detailed clinical, radiological, and therapeutic analysis. Our child was managed with partial endoscopic removal of lingual thyroid, followed by substitutive exogenous thyroid hormone therapy instead of radioiodine ablation. The clinician should take into account the potential of this rare entity in any child's hypothyroidism and differentiate it from other masses in the neck and distant sites.

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