Ectopic Lingual Thyroid: A Case Presentation

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Abstract

Background: The lingual thyroid gland is a rare clinical entity that was found to occur due to the failure of the thyroid gland to descend into its normal ectopic pretracheal position during embryogenesis. The reported incidence of lingual thyroid is 1 in 100,000, and it is more common in females, with a female: male ratio of 3:1. When located at the base of the tongue, the ectopic gland is often asymptomatic but may cause local symptoms, such as dysphagia, dysphonia, upper airway obstruction, hemorrhage, and often hypothyroidism. The diagnosis of lingual thyroid is usually made clinically and radionuclide scanning is used to confirm the diagnosis. A case of lingual thyroid is presented for its rarity and differential diagnosis of midline base of the tongue lesions.

Case description: A 35-year-old woman presented with complaints of increasing difficulty in breathing and a constant foreign object sensation in the throat (FOSIT) for the past one year. Flexible video-laryngoscopic examination revealed a well-demarcated midline tongue base lesion measuring 3 × 3 cm. Technetium 99m scan reported it to be ectopic thyroid gland tissue. Transoral excision of the lesion was done along with cautery with bipolar cautery.

Conclusion: Lingual thyroid is a rare developmental anomaly, the treatment of which is still controversial in view of the rarity of the condition. It should be included in the differential diagnosis of midline masses of the base of the tongue, especially in children and adolescents. The approach to treatment should be transdisciplinary and should not only take into consideration the clinical condition of the lesion but also the hormonal aspects of the patient.

Keywords: Thyroid, Thyroid scintigraphy, Tongue base.

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Background

Lingual thyroid is a rare clinical condition that occurs during embryogenesis due to failure of descent of the thyroid gland into its normal pretracheal position. It has a reported incidence of 1 in 100,000, and it is more common in females, with a female:male ratio of 3:1. Lingual thyroid is often asymptomatic but may cause local symptoms such as upper airway obstruction, dysphonia, dysphagia, and often hypothyroidism. Clinical examination is the key to the diagnosis of lingual thyroid and radionuclide scanning is used to confirm the diagnosis. A case of lingual thyroid is presented for its rarity and differential diagnosis of midline base of the tongue lesions.

Case description

A 35-year-old woman is reported here who presented with complaints of difficulty in breathing and foreign object sensation in the throat (FOSIT). Flexible video-laryngoscopic examination revealed a well-demarcated midline tongue base lesion measuring 3 × 3 cm mass with normal mucosal covering. The surface of the swelling was normal without any signs of ulceration, bleeding, or pus. However, numerous anastomosing blood vessels were seen over the mucosa of the swelling (Fig. 1). Provisionally, a diagnosis of lingual thyroid was made based on its location and symptomatology further investigations were carried out. The thyroid profile of the patient was taken which was reported to be within normal limits. Technetium 99m thyroid scan revealed an absence of isotope uptake at the root of the neck, i.e., at the usual location of the thyroid gland and a midline focal area of increased isotope uptake in the lingual region (Fig. 2). The patient was provisionally diagnosed as having lingual thyroid and was advised regarding lifelong thyroxine replacement in-light of the above diagnosis; for which she was very keen to proceed with surgery despite the long-term morbidity. Transoral surgical resection is under general anesthesia with nasotracheal intubation. Surgical steps included the usage of a Boyle–Davis mouth gag and bipolar cautery forceps, a cuff of normal tongue base tissue was also removed to take the swelling in its entirety (Fig. 3). The grossly resected specimen measured 3 × 3 cm and firm in consistency (Fig. 4). Her postoperative recovery was uneventful, voice and diet restoration were immediate. A postoperative video-laryngoscopy was carried out on day 5 revealing a healthy and healing mucosa at the resected site with no signs of the slough, pus, or bleeding (Fig. 5). Postoperatively, our patient was commenced on thyroid replacement treatment and has been reviewed by our endocrine colleagues, as part of a multidisciplinary approach in managing endocrine conditions.

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Literature defines lingual thyroid as the presence of thyroid tissue in the midline at the base of the tongue anywhere between the circumvallate papillae and the epiglottis.\(^1\) It occurs due to aberrant embryogenesis during the descent of the thyroid gland to the neck.\(^2\) In 1869, Hickmann reported the first case of lingual thyroid.\(^3\) It occurs due to embryonic failure of normal thyroid tissue to descend from the foramen caecum area of the tongue base to the lower part of the neck in front of thyroid cartilage.\(^4\) Lingual thyroid is reported to be seven times higher in females and has an incidence of 1:100,000.\(^5\) It is more commonly seen during childhood, adolescence, and during menopause.\(^6\) During early fetal life maternal antithyroid antibodies, authors have postulated that impairment of gland descent is the pathogenesis of lingual thyroid.\(^1\)

Some of the other locations of ectopic thyroid are geniohyoid and mylohyoid muscles (sublingual thyroid), above the hyoid bone, and in rare locations like pharynx, heart, mediastinum, pericardial sac, esophagus, breast, lung, trachea, duodenum, adrenal gland, and mesentery of the small intestine.\(^6\) Usually, lingual thyroid is asymptomatic but it can be associated with hypothyroidism in up to 70% of patients.

Lingual thyroid may also rarely be associated with symptoms such as dyspnea, dysphagia, and the sensation of a lump in the throat. Less common complaints include dysphonia or bleeding.\(^7\) Lingual thyroid presents as a nodular mass in the midline at the base of the tongue. On video-laryngoscopy examination, the
lingual thyroid appears as a smooth surface mucosal lesion with vascularity. An essential part of the examination is palpation of the neck to check the presence or absence of the thyroid gland in a normal anatomical location. Investigations for the diagnosis and treatment plan for lingual thyroid include radionuclide Technetium-99m and iodine-131 thyroid scans in addition to serum thyroid profile (T3, T4, and TSH). Ectopic thyroid presence at the base of the tongue is confirmed with technetium scanning. Our case did not reveal any normal thyroid gland on scintigraphic and radiological examinations. Histologically; on FNAC, the lingual thyroid resembles normal thyroid tissue. The treatment options that are available for lingual thyroid include chemotherapy, surgery, and radioiodine ablation. Although controversial, clinical treatment may be attempted, using suppressive therapy with exogenous thyroid hormone in small oligosymptomatic lingual thyroids. An alternative approach recommended in older patients or patients who are deemed unfit for surgery is ablative radioiodine therapy. Surgical excision can be carried out only after confirmation of adequate thyroid tissue in the neck by iodine-131 radionucleotide scan. The transoral approach offers the best approach among different types of surgical access and provides good exposure, better postoperative recovery, and is less traumatic for the patient.

**CONCLUSION**

Lingual thyroid is a rare developmental anomaly, the treatment of which is still controversial in view of the rarity of the condition. It should be included in the differential diagnosis of midline masses of the base of the tongue, especially in children and adolescents. A recommendation before and after surgery is to check thyroid function tests due to the risk of postoperative hypothyroidism. Treatment options are conservative with substitutive hormone treatment in patients with mild symptoms, while cases with airway obstruction surgery are the preferred line of management.

**ETHICS APPROVAL AND CONSENT TO PARTICIPATE**

All procedures performed in the case report were in accordance with the ethical standards of the institution. Informed consent was obtained from the patient included in the case presentation.

**CONSENT FOR PUBLICATION**

Informed consent was obtained from the patient included in the case presentation.

**AVAILABILITY OF DATA AND MATERIAL**

Not applicable

**AUTHORS CONTRIBUTIONS**

All the authors have contributed to writing up this case presentation. Kanishka S Rao: Drafted the work. Anas Gomati: Revised the drafted work. Vijendra S Shenoy: Substantial contribution to the conception. Panduranga M Kamath: Contribution to conception. All listed authors have approved the submission of this case presentation to this journal.

**REFERENCES**