

Lingual Thyroid Gland: A Case Report

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Summary: Lingual thyroid (LT) is a developmental disorder due to the failure of the thyroid gland to migrate from its anlage, foramen caecum to its prelaryngeal site. Lingual position represents the most frequent ectopic location accounting up to 90% of ectopic cases. It is found at the junction of the anterior two thirds and the posterior third, between the epiglottis and the circumvallate papillae, often asymptomatic but growing may cause local symptoms as upper airway obstruction, dysphagia and hemorrhage at any time from infancy to adulthood. Thyroid scintigraphy plays the most important role in diagnosing ectopic thyroid gland but neck ultrasound, neck CT- scan, neck MRI contribute as well. Treatment of this anomaly includes exogenous L-thyroxine hormone administration, radioiodine ablation therapy and surgery.

Keywords: lingual thyroid (LT), ectopic thyroid, aberrant embryogenesis, fine needle aspiration cytology (FNAC)

I. Introduction

Lingual thyroid (LT) is a rare developmental disorder due to the aberrant embryogenesis, during the descent of the thyroid gland from the foramen caecum of tongue to its prelaryngeal site in the neck [1- 4]. Ectopic thyroid indicates the presence of the thyroid tissue in other locations than the anterior neck region, which is its normal position [1-4]. In 1869, Hickman reported the first case with lingual thyroid on a newborn baby [5]. Hunt in 1865 reported a similar case of a lingual thyroid in a young woman, although not proved [6]. Haynes in 1912 had removed a proved thyroid tumor of the tongue [5, 6]. Lingual site (at the base of the tongue) represents the most frequent ectopic location accounting for 90% of cases [7] although lower rates (47%) have been reported by others [8, 9]. In 70-75% of cases lingual thyroid is the only thyroid tissue present [10, 11]. It is very uncommon for two ectopic foci to be present simultaneously [11, 12]. The majority of ectopic thyroids are located in the midline along the thyroglossal tract [12], sublingual positions [2, 13], intra-laryngotracheal [15, 16, 17].

Intrathoracic ectopy as mediastinal [15, 16], esophageal [18, 19], and heart [17-19, 20] are described. An overdescent of thyroglossal duct remnants has been suggested as the cause of ectopic thyroid tissue under the diaphragm especially in porta hepatis [22], ovary [23, 24], adrenal glands [26- 28] and gallbladder [28].

It is recently reported that prevalence rates from 1 in 100000 to 1 in 300000 persons, with female to male ratio ranging from 4:1 to 7:1 [1, 2, 15, 16].

Although the pathogenesis of lingual thyroid is unclear, some authors have postulated that maternal antithyroid immunoglobulins may impair gland descend during early fetal life [15, 16].

Clinical features vary from asymptomatic [14] to dysphagia, dysphonia [12, 15, 35], stomatolalia, cough, sleep apnea [10, 11], sensation of foreign body and up to severe upper airway obstruction [36] and hemorrhage [37]. Diagnosis of LT includes local examination of the tongue base associated with the absence of normally located gland, and imaging examinations. Imaging studies consist in neck ultrasound, neck CT-scan, neck-IRM and scintigraphy using Tc 99m, I-131, I-123.

II. Case Report

A 33 years old female patient, Dh.B., was referred to the department of General Surgery, from Endocrinology department with the diagnosis of "lingual thyroid". The patient was hospitalized in this service for a swelling at the base of the tongue, noticed 6 years ago, during the first pregnancy. The mass during these years had been grown, and the patient complained of the sensation of foreign body in the mouth, dysphagia, dispnea, cough and bleeding during the end of pregnancy.

On physical examination it was noticed a well localized lesion measuring 5-6 cm in size, smooth, rubbery and reddish mass at the base of the tongue (Figure 1). No signs of ulceration, bleeding or pus discharge were identified on the surface of the mass.



Figure1. Intraoperative view of lingual thyroid gland preparation

Neck examination revealed no palpable thyroid gland or neck mass but a cervicotomy scar.

Thyroid tests were as follow: TSH 15, 5 uIU/ML, T3 3, 08 pg/ml, T4F 0, 506 ng/dl. The patient was healthy and in her medical history revealed of an operation done during childhood for a subhyoid thyroglossal cyst. She was a nonsmoker. Other laboratory tests were within normal limits. Thyroid ultrasound revealed no thyroid gland in its normal position. Scintigraphy revealed increased isotope uptake in the lingual region and absence of isotope uptake in the neck region. The final diagnosis was ectopic lingual thyroid gland. Treatment began with the administration of L-thyroxine for 3 months and after that with I-131.

The management of the case was discussed fully with the patient and she agreed. Elective surgical resection of the thyroid gland was performed through oral approach under general anesthesia. A nasotracheal intubation was performed. The surgery and postoperative period were uneventful (Figure 2, 3).



Figure 2. The lingual thyroid gland

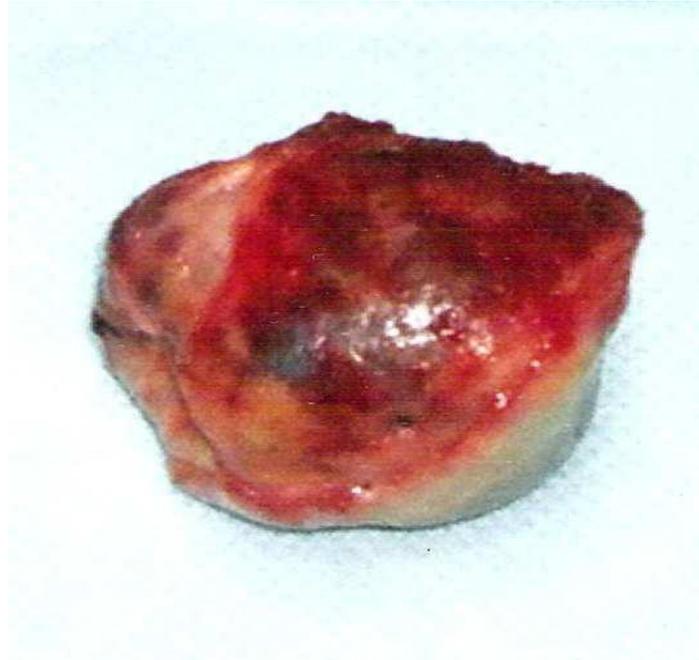


Figure 3. The lingual thyroid gland.

The patient was extubated the third postoperative day in the ICU and was discharged on the 5-th postoperative day, under l-thyroxine supplement treatment as the endocrinologists recommended. The specimen was sent for histopathology analysis and the result was follicular goiter (Figure 4).

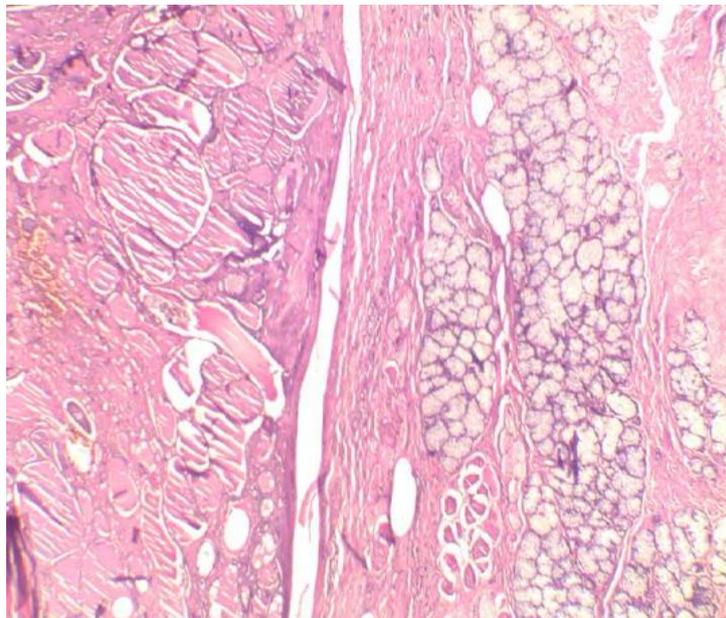


Figure 4. The morphology of removed ectopic thyroid gland, demonstrating the tissue transition from tongue to thyroid gland.

III. Discussion

During embryogenesis, the thyroid tissue descends from the foramen caecum area of the tongue base to the lower part of the neck. This descent process may be impaired anywhere along this path of primitive thyroglossal duct [3, 4]. Maternal antibodies against thyroid antigens are thought to be the cause of the failure in the descending process of thyroid tissue from its origin to its final position [37]. Although it is usually found along the normal path of development it can be found in the mediastinum [15, 16], heart [16-17], oesophagus [16, 17], porta hepatis [28-30], ovary [23], etc. The incidence is reported to be 1:100000-300000 persons, 7 times higher in females than males.

Ectopic lingual thyroid is commonly detected during periods of increased demand for thyroid hormones, for example adolescence and pregnancy [40]. In our case it has become symptomatic during pregnancy. Dysphagia, pain, dyspnea, bleeding and fullness in the throat are the major signs and symptoms [10-12, 32, 33, 35]. Usually they represent a nodular mass in the base of the tongue.

The investigations include thyroid function tests, ultrasonography and scintigraphy. Scintigraphy by using technetium (Tc-99m), I-131, or I-123, is an important diagnosing tool to detect ectopic thyroid tissue and to show the absence or presence of normally positioned thyroid tissue [12, 13, 15]. Fine needle aspiration cytology (FNAC) confirms the diagnosis of ectopic thyroid. It is the only modality to differentiate between benign and malignant lesion [15, 28, 29]

For cases completely asymptomatic and euthyroid regular follow-up is recommended in order to detect mass complications [14, 35, 37]. For mild symptoms and hypothyroidal states, levothyroxine replacement therapy may be effective [14, 35, 37]. Surgical treatment of lingual thyroid depends on size and local symptoms as well as on other parameters such as patient's age, functional thyroid status and complications of the mass [14, 35, 37]. Several surgical approaches have been described such as transoral route, transhyoid route, suprahyoid and lateral pharyngotomy. The transoral route is the preferred approach avoiding deeper structures complications such lingual nerve injury and deep cervical infections [14, 32].

IV. Conclusion

Lingual thyroid as a rare developmental anomaly should be included in the differential diagnosis of tongue masses especially in children and adolescence during periods of increased thyroid hormone demand. Investigations include thyroid function tests, neck ultrasound and neck CT. FNAC is not preferred by some authors because of bleeding possibility. Transoral approach provides good exposure, being less traumatic for the patient with better postoperative recovery.

Figure 1. Intraoperative view of lingual thyroid gland preparation

Figure 2, 3. The lingual thyroid gland

Figure 4. The morphology of removed ectopic thyroid gland, demonstrating the tissue transition from tongue to thyroid gland.

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