

# Lingual thyroid: A case report and literature review

## Clinical Case

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### Abstract

The presence of ectopic thyroid tissue at the base of the tongue is called lingual thyroid. Most cases are asymptomatic, sometimes being diagnosed accidentally, for example, in the context of an upper respiratory infection. In this paper, we present the case of a 44-year-old female patient, with a history of hypothyroidism, who came to the emergency department with fever, odynophagia, dysphagia, and right otalgia. She had edema, purulent exudate and areas of necrosis on the lingual surface of the epiglottis, ventricular and arytenoid bands; a mass was found at the base of the tongue, in the midline, above the epiglottis, with normal-appearing mucosa. Cervical computed tomography confirmed the diagnosis of supraglottitis and showed a hyperdense nodular image with uptake of contrast product at the base of the tongue, consistent with lingual thyroid; in the usual topography of the thyroid, only a small nodular image was identified. The Tc99 scintigram confirmed the diagnosis of lingual thyroid, with a larger nodular image in the oropharynx and a small one in the thyroid topographic area.

Keywords: thyroid gland, lingual thyroid, supraglottitis

### Introduction

The presence of thyroid tissue outside its normal anatomical position is called ectopic thyroid, and when present at the base of the tongue, it is referred to as lingual thyroid. The dorsum of the tongue accounts for 90% of cases of ectopic thyroid. Ectopic thyroid tissue has also been described in the larynx, trachea, mediastinum, lung, heart, and adrenal gland<sup>3</sup>. The presence of thyroid tissue at the lingual level is due to the absence of its migration from the foramen caecum to the pretracheal location, which occurs between the 3rd and 7th week of fetal development<sup>1,2</sup>. It is commonly located between the circumvallate papillae and epiglottis in the region of the foramen caecum<sup>4,5</sup>. Studies report an incidence of 1:1000 to 1:300000<sup>1-3</sup> in the general population and 1:4000 to 1:8000

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in individuals with thyroid pathology<sup>3</sup>. The female-to-male ratio is 4–7:1<sup>12</sup>. Approximately two-thirds of the patients have no pretracheal thyroid tissue<sup>6</sup>, which constitutes the only functioning thyroid tissue in 70–100% of the cases<sup>7</sup>. The potential for malignancy is the same as for the orthotopic tissue<sup>8,9</sup>. The pathogenesis of this entity remains unclear; maternal antithyroid immunoglobulins are assumed to impair embryonic migration and predispose to hypothyroidism<sup>10</sup>. Most of the cases are asymptomatic<sup>11–16</sup> unless there is an increase in the size of the gland resulting in dysphagia, dysphonia, odynophagia, hemorrhage, and dyspnea<sup>17</sup>. A meticulous clinical investigation that includes objective examination, biochemistry, and imaging is necessary for delineating the best therapeutic approach<sup>14–15,18</sup>.

### Clinical Case

The patient was a 44-year-old woman with leukoderma and hypothyroidism. She was being treated with levothyroxine and was a smoker (30 pack-years).

The patient presented to the emergency department of Otorhinolaryngology (ORL) at Pedro Hispano Hospital (Hospital Pedro Hispano - HPH) with fever, odynophagia, dysphagia, and right otalgia for 2 days with no response to deflazacort 30 mg 12/12h, paracetamol 1000 mg 8/8h, and ibuprofen 600 mg 8/8h. The patient had a hot potato voice. Video laryngoscopy showed a mass at the base of the tongue in the midline, above the epiglottis, with normal looking mucosa (Figure 1 - A) along with edema, purulent

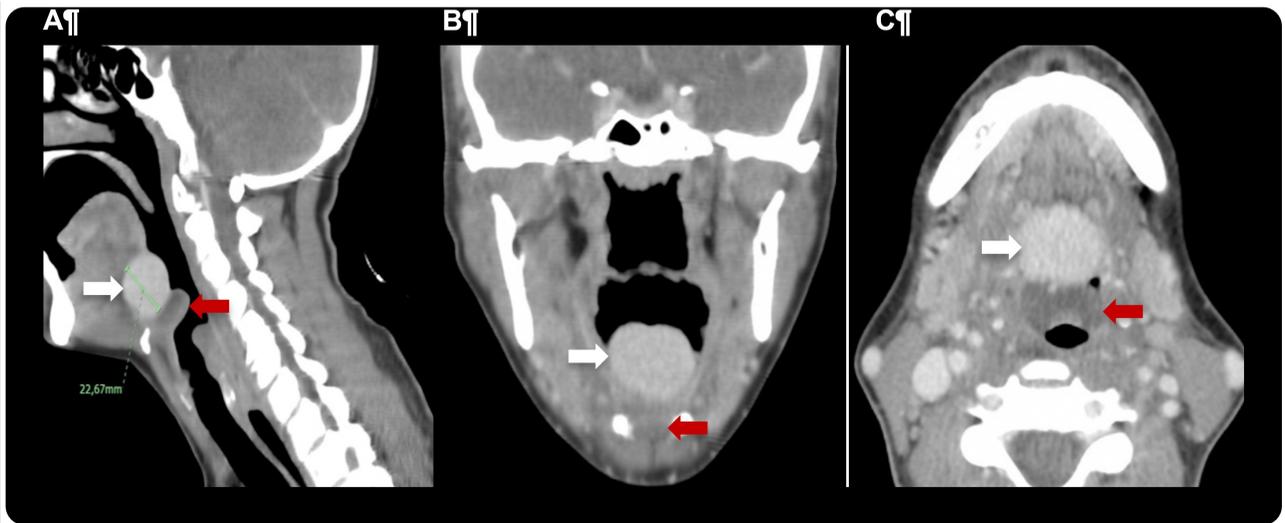
exudate, and areas of necrosis on the lingual surface of the epiglottis, ventricular bands, and arytenoids. Significant salivary stasis was observed (Figure 1 - B and C). The glottic cleft, although reduced, was patent (Figure 1 - D). Blood tests revealed leukocytosis with neutrophilia and a C-reactive protein level of 122.5 mg/dL. The severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) test was negative. Cervical computed tomography (CT) (Figure 2 - A, B, and C) confirmed the diagnosis of supraglottitis, with laryngeal edema most evident in the epiglottis and aryepiglottic folds (red arrow). It also showed a spontaneously hyperdense nodule with contrast uptake at the base of the tongue, measuring approximately 28x26x23 mm, which was compatible with lingual thyroid (white arrow). At the usual thyroid location, there was only a small nodule on the right, 12x4 mm in size, reflecting the thyroid tissue. Blood cultures were collected, and hydrocortisone 200 mg 12/12h and empirical antibiotic therapy with ceftriaxone 1 g 12/12h and clindamycin 600 mg 8/8h were started. After symptomatic improvement and reduction in inflammatory parameters, she was discharged with amoxicillin + clavulanic acid (875 + 125 mg) 12/12h and prednisolone with a weaning scheme. When she was reassessed in an outpatient consultation, she was found to be asymptomatic. On objective examination, the mass was located on the dorsal surface of the tongue, posterior to the circumvallate papillae. The larynx showed no inflammatory signs.

An outpatient Tc99 scintigram confirmed the

**Figure 1**  
Flexible indirect laryngoscopy findings



**Figure 2**  
Computed tomography at admission. A - Sagittal section; B - Coronal section; C - Axial section



**Figure 3**  
Thyroid Tc99 scintigram showing mouth and neck activity



diagnosis of lingual thyroid, showing a larger nodular image with uptake in the oropharynx and a smaller one in the thyroid topographic area on the right (Figure 3).

### Discussion

The lingual thyroid usually presents as a mass at the base of the tongue and can reach dimensions greater than 4 cm. Its surface is usually smooth and vascularized<sup>12</sup>. Most patients are asymptomatic, with lingual thyroid being diagnosed incidentally on routine

observation of the oropharynx or in the context of an upper respiratory infection. However, it can manifest as symptomatic hypothyroidism or, in the case of a larger mass, with dyspnea, dysphonia, dysphagia, and hemorrhage<sup>12</sup>. In the present case, the diagnosis was fortuitous in the context of an upper respiratory infection (supraglottitis). According to the literature, it is mostly diagnosed after the age of 6 years, with most cases being diagnosed during puberty or pregnancy<sup>3,19</sup>. This is assumed to be attributable to the increased physiological need for thyroid hormones during these periods, which, in turn, leads to an increase in the size of the gland. The need for increased thyroid hormone production can also occur in infectious or traumatic conditions. In 70–80 % of the cases, the lingual thyroid tissue is the only thyroid tissue present in the body<sup>1-3</sup>. In the present case, although a portion of the thyroid gland migrated to its cervical location, thyroid tissue remained at the base of the tongue and hypertrophied in response to elevated thyroid stimulating hormone (TSH) secretion. The initial evaluation of patients with suspected lingual thyroid should include a complete objective ORL examination, with a special focus on the oropharynx, as the lingual thyroid often appears as a mass posterior to the lingual “V”<sup>20,21</sup>. Rigid and/or flexible endoscopy allows assessment of the size, position, and characteristics of the

lesion, as well as observation of the larynx and detection of concomitant lesions<sup>1</sup>. Neck palpation is important for assessing the presence or absence of cervical thyroid tissue. Thyroid tests in unsupplemented patients show hypothyroidism in approximately 70% of cases<sup>14,22</sup>. As our patient already had previously documented and treated hypothyroidism, thyroid hormones were not assayed. Lingual thyroid rarely develops hyperthyroidism, and the potential for malignancy is low (1 in 300 cases)<sup>17,22</sup>. The histological types that have been described include papillary (most common), follicular, mixed follicular-papillary, Hurthel cell, and medullary carcinoma<sup>17,23</sup>.

Imaging is essential to confirm the diagnosis. Scintigraphy with Technetium 99m pertechnetate or <sup>131</sup>I usually shows staining at the base of the tongue and no staining at the usual location. In our case, besides lingual labeling, the patient also had cervical labeling. CT is a useful method for determining the gland size<sup>1</sup>. Ultrasound imaging is a widely available and non-invasive test that can be useful for evaluating the thyroid gland and detecting ectopic tissue<sup>13</sup>.

When performed with Doppler, it allows assessment of tissue vascularization. Magnetic resonance imaging (MRI) is the investigation of choice for surgical planning, allowing better evaluation of the sagittal planes than CT. In the present case, MRI was not requested because the lingual thyroid was causing no symptoms, and surgical treatment was not considered.

The differential diagnosis of lingual thyroid includes thyroglossal duct cyst, midline branchial cyst, lymphoma, hemangioma, angioma, lipoma, fibroma, adenoma, malignant lesion, and neoplasms of the minor salivary glands<sup>24</sup>. Fine needle aspiration cytology can help confirm the diagnosis in dubious cases<sup>11,17, 23</sup>. In the present case, it was not necessary.

There is no consensus on treatment due to the rarity of this condition, as only a limited number of cases have been described. Treatment depends on the size, presence or absence of symptoms, and/or complications such as hemorrhage, malignancy, or airway obstruction.

The main goal is to reduce the obstructive symptoms produced by the mass while not putting the patient's life at risk or causing mutilating scars. The basis of medical treatment is suppressive therapy with exogenous thyroid hormones. The goal is to suppress TSH production and thus remove the stimulus that causes a volumetric increase in the ectopic tissue<sup>1,21</sup>. It is also considered to prevent malignant transformation<sup>3</sup>. This is the recommended approach in patients with mild to moderate symptoms (foreign body sensation and odynophagia)<sup>9</sup>. This also seems to be the recommended treatment in asymptomatic euthyroid cases to prevent the appearance of hypothyroidism (which will eventually appear in the majority and inevitably lead to glandular hypertrophy)<sup>9,21</sup>.

However, the rate of size reduction is considerably slow, and a major decrease in the volume is not expected<sup>9</sup>. In the case presented here, the patient was asymptomatic and undergoing pharmacological therapy with levothyroxine. According to the literature<sup>1</sup>, in patients undergoing pharmacological treatment, it is necessary to reassess thyroid hormone levels ideally every three months, paying special attention to periods of increased metabolic stress (such as infections, trauma). Surgical treatment is reserved for patients with severe symptoms or those who experience the worsening of mild to moderate symptoms under pharmacological treatment. It is crucial in cases of recurrent or major bleeding, as well as in patients with dysphagia, dysphonia, and dyspnea<sup>9</sup>. Ablation with radioactive iodine <sup>131</sup>I can be used as an alternative to surgical excision. It is contraindicated in women of childbearing age and children and is most often reserved for elderly patients with contraindications to surgery or who refuse surgery, and lifelong hormone replacement is required after this treatment<sup>9</sup>.

## Conclusion

Lingual thyroid is a rare clinical entity that presents as a mass at the base of the tongue and is often diagnosed incidentally. Diagnostic investigation includes objective ORL examination, thyroid biochemistry, imaging,

and histopathology and is critical for choosing the best treatment approach. Lingual thyroid cases with mild to moderate symptoms can be treated conservatively with levothyroxine. In severe cases or those refractory to medical therapy, surgical or ablative treatment should be considered.

### Conflicts of Interest

The authors declare that there is no conflict of interests regarding the publication of this paper.

### Data Confidentiality

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

### Protection of humans and animals

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the 2013 Helsinki Declaration of the World Medical Association.

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### Availability of scientific data

There are no datasets available, publicly related to this work.

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