

Obstructive lingual thyroid; suprahyoid intracervical surgical procedure – A case report

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ABSTRACT

Introduction. The treatment of lingual thyroid is controversial and should be individualized. Options include hormonal replacement, surgery in the presence of bleeding and obstruction of the airway and the digestive tract, and radioisotope therapy.

Clinical case. 8-year-old girl presenting with discomfort when swallowing. A pink, well-vascularized mass, not painful or ulcerated, protruding from the base of the tongue and virtually closing the whole oropharynx, was observed. Absence of thyroid tissue in its normal position was reported by the ultrasound department. Cervical computed axial tomography confirmed the diagnosis and the presence of pharyngeal obstruction. Thyroid hormone replacement was established. As a result of dysphagia symptom progression, surgery was indicated. Thyroid removal was performed by means of a cervicotomy, with re-implantation of thyroid tissue laminae. The postoperative course was uneventful and replacement treatment was maintained, with an excellent clinical status four years later.

KEY WORDS: Ectopic; Lingual; Thyroid.

TIROIDES LINGUAL OBSTRUCTIVO, INTERVENCIÓN QUIRÚRGICA VÍA TRANSCERVICAL SUPRAHIOIDEA: REPORTE DE UN CASO

RESUMEN

Introducción. El tratamiento de la tiroides lingual es controvertido y debe individualizarse. Las opciones incluyen el reemplazo hormonal, cirugía en presencia de hemorragia y obstrucción de la vía aérea o digestiva, y la terapia con radioisótopos.

Caso clínico. Niña de 8 años de edad, con molestias a la deglución. Se observa masa rosada, muy vascularizada, no dolorosa ni ulcerada, que protruye desde la base de la lengua y cierra prácticamente toda la orofaringe. Ecografía informa ausencia de tejido tiroideo en su posición normal. Tomografía axial computarizada

cervical comprueba el diagnóstico y la obstrucción faríngea. Se indicó tratamiento sustitutivo de las hormonas tiroideas. Ante la progresión de los síntomas de disfagia, se indicó cirugía. Se describe la exéresis tiroidea por vía cervical, suprahioidea, con implante de láminas de tejido tiroideo. Evolucionó sin complicaciones y se mantiene tratamiento sustitutivo, con excelente estado clínico después de cuatro años.

PALABRAS CLAVE: Tiroides; Lingual; Ectópica.

INTRODUCTION

Ectopic thyroid is an embryological defect in the development of the thyroid gland due to an alteration in the migration from the foramen cecum at the base of the tongue to a pre-tracheal location, between the third and the seventh week of pregnancy⁽¹⁾.

Ectopic thyroid tissue can be found both in the upper aerodigestive tract (lingual, sublingual, thyroglossal, and intralaryngotracheal locations) and in other remote locations (cervical esophagus, superior mediastinum, diaphragm, and heart)⁽²⁾. Maternal antithyroid immunoglobulins have been proposed to be able to stop gland descent and predispose the patient to a poor thyroid function during life.

Clinical incidence ranges from 1:3,000 to 1:10,000. The lingual thyroid is the only functioning thyroid tissue in 70% of cases. The presence of ectopic thyroid tissue is 3 to 4 times more frequent in women than in men, but according to some authors, the proportion could be up to 7:1⁽²⁻⁵⁾.

The most frequent form of ectopic thyroid is lingual thyroid, representing 90% of all cases. Clinical signs depend on patient age, thyroid function, and mass size, and diagnosis is mainly based on imaging tests. Generally, it presents as a submucosal node at the base of the tongue^(4,6). Treatment is controversial and should be individualized in each patient⁽³⁻⁶⁾.

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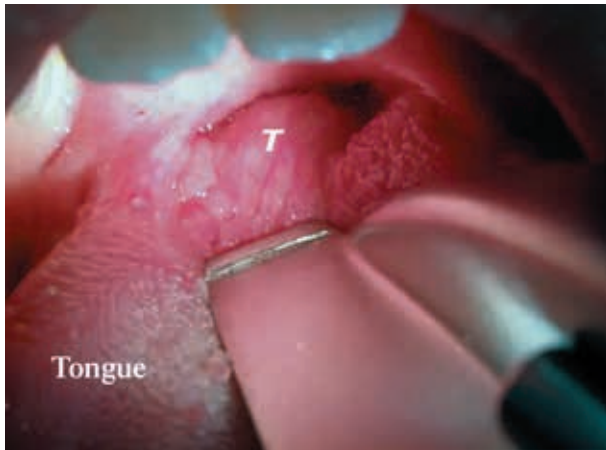


Figure 1. Pink, resistant, well-vascularized mass, not painful or ulcerated, protruding from the base of the tongue and virtually closing the whole oropharynx.

CLINICAL CASE

Eight-year-old female patient presenting with discomfort when swallowing. Physical examination of the oral cavity demonstrated the presence of a pink, resistant, well-vascularized mass, not painful or ulcerated, protruding from the base of the tongue and virtually closing the whole oropharynx (Fig. 1).

Suspected diagnosis of ectopic thyroid was established. Ultrasound examination demonstrated the absence of thyroid tissue in its normal position and its presence at the base of the tongue. A cervical CT-scan was carried out (Fig. 2), which allowed lingual thyroid diagnosis to be confirmed.

Thyroid gland function was studied, with thyroid stimulating hormone (TSH) levels of 6.98 IU/ml (benchmark value: 0.4-4 IU/ml) and thyroglobulin (TG) levels of 109 (benchmark value: 0.1-79).

Thyroid hormone replacement therapy with 3 µg/kg/day levothyroxine was applied over 4 weeks; the mass did not decrease in size, but TSH and TG did return to normal levels.

In light of dysphagia symptom progression, the ectopic gland was surgically removed. Following nasotracheal intubation using fiber endoscopy, a cervical transverse incision was performed at the level of the hyoid bone, the suprahyoid muscle planes were separated in the midline, and the bone's body was resected to facilitate exposure. The mylohyoid muscle was opened and the thyroid gland was identified at the base of the tongue, assisted by an oral digital exploration to improve mass protrusion towards the surgical area (Fig. 3). The gland was completely removed, the oral cavity's muscle floor was repaired, and suprahyoid muscles were put together in the cervical midline. Two small gland laminae were re-implanted underneath the submandibular muscles,

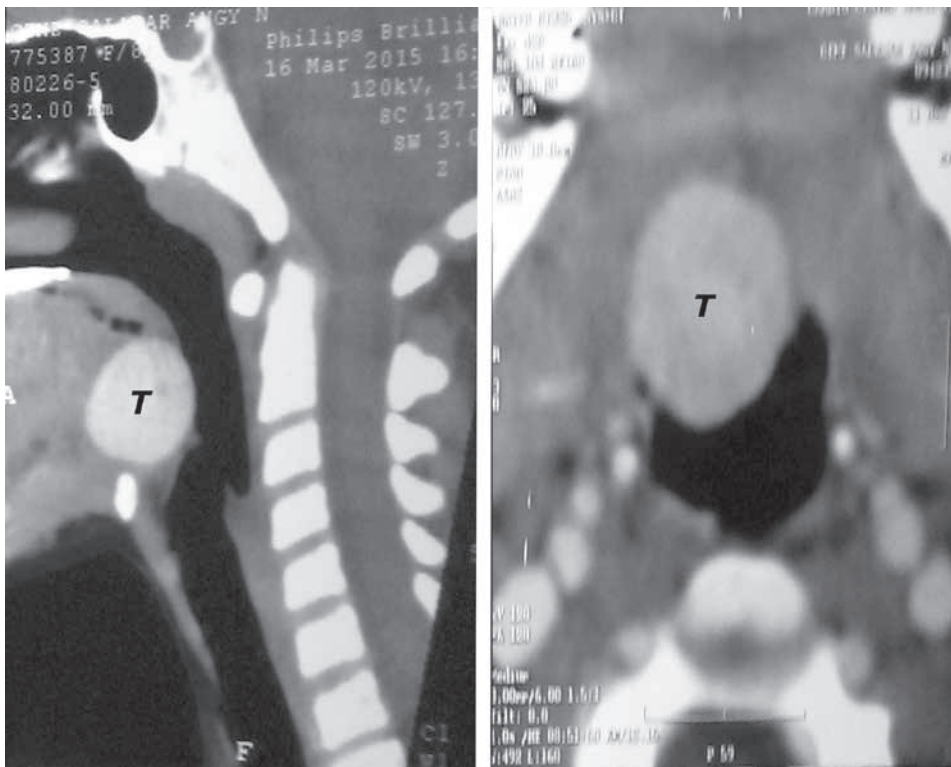


Figure 2. CT-scan demonstrating the presence of the mass at the base of the tongue, partially obstructing the digestive tract without compromising the airway.

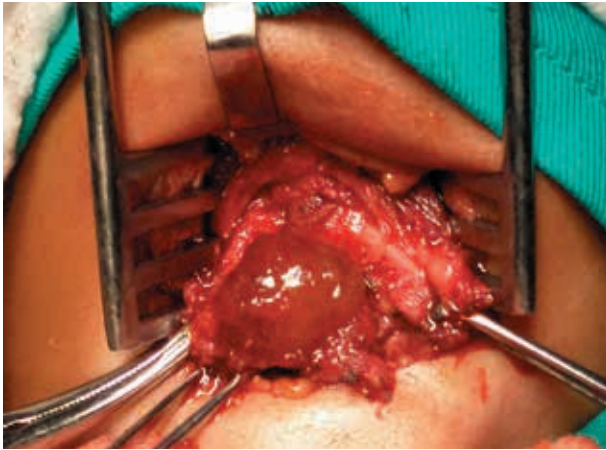


Figure 3. The ectopic thyroid tissue is shown using a cervical approach with hyoid bone body resection and suprahyoid muscle dissection.

and the other ones were submitted for anatomic pathology examination. The permeability of the oropharynx was found to be normal (Fig. 4). Mechanical ventilation was applied for 24 hours. The postoperative course was uneventful. Thyroid hormone replacement treatment was maintained, with an excellent clinical status after a two-year follow-up.

DISCUSSION

Hickman⁽⁷⁾ was the first to describe lingual thyroid in 1869. He did so in a female neonate who died of suffo-

cation 16 hours post-birth. Ectopic thyroid tissue is not associated with a higher malignization risk than the thyroid gland. The presence of carcinoma is extremely rare (1%)^(3,4,6).

Lingual thyroid is not associated with parathyroid glands, usually located on its posterior portion, since they are originated in the dorsal aspect of the third and the fourth pharyngeal pouches – they have a different embryologic origin⁽³⁾.

Most symptoms show up during adolescence, pregnancy, and menstruation. Signs and symptoms include dysphagia, dysphony, dyspnea, foreign body sensation, and occasionally bleeding^(2,4,5).

Intraoral clinical examination usually demonstrates the presence of an irregular, soft, well-vascularized red, pink, or blue mass, variable in diameter, in the midline of the base of the tongue. Upper airway endoscopic assessment determines the size of the gland and to what extent the airway is compromised⁽²⁻⁵⁾.

Thyroid function tests usually demonstrate euthyroidism or hypothyroidism⁽⁷⁾, with normal or low T3 and T4 levels, and high TSH and thyroglobulin levels^(2,3,5,6).

Tc⁹⁹ or I¹³¹ scintigraphy is the most specific study for lingual thyroid diagnosis. It shows activity or radioisotope uptake at the base of the tongue, and no apparent activity in the gland's normal position in the neck. This avoids the need for diagnostic biopsy, which may entail a risk of uncontrollable bleeding or acute thyrotoxicosis, but at the same time avoids the use of ionizing radiation – it has been replaced by more harmless methods, especially in children^(3,5).

CT-scan is useful in determining the size of the gland. However, IRM remains the best non-invasive technique



Figure 4. The postoperative tissue is displayed, with no obstructive mass and with an adequate esthetic result at the incision site.

as it provides with multi-plane images and an excellent definition of the soft tissues^(3,6).

Selective external carotid angiography can detect aberrant vascularization patterns. Surgically speaking, pre-surgery nutrient vessel embolization is a safe method⁽³⁾.

There is no real consensus in the literature regarding the true behavior of lingual thyroid owing to its rare nature and the few and short study series published. However, it seems clear that the basic goals are the complete removal of the obstructive symptoms, which can pose a risk for the patient's life, and the control of the thyroid function^(3,5,6,8).

Treatment depends on various factors: symptom severity, lesion size, patient age and sex, and thyroid function. Euthyroid patients with asymptomatic lingual thyroid should be regularly monitored, but no treatment is required. If lingual thyroid does not cause obstructive symptoms, there should be no need for surgical treatment, and if it presents with hypothyroidism symptoms, it can be treated with thyroid hormone supplements only. The objective is to suppress TSH levels and therefore remove gland growth stimulus^(3,4,8). Treatment duration ranges from 1 to 22 months, with a reported effective response of about 60%⁽⁸⁻¹¹⁾.

Ablation with therapeutic doses of radioactive I¹³¹ is an alternative treatment method, but especially contraindicated in children and fertile women^(2-6,9).

Surgical treatment should be considered in cases where the lesion size causes obstructive or bleeding symptoms, when malignancy is suspected, and in cases where initial symptoms get worse following suppression therapy^(2,3,5,9).

When lingual thyroid is associated with functioning thyroid tissue in its usual location, total removal has healing properties. Otherwise, re-implanting the ectopic thyroid tissue removed has been suggested as the most adequate treatment to prevent the development of a hypothyroid status requiring lifelong thyroid hormone contribution. The tissue is placed as a free graft under the fascia of the anterior rectus muscle of the abdomen or in the submandibular region, after being divided in 4 to 5 mm thick laminae. Thyroxin treatment is not administered in the immediate postoperative course so as to prevent the suppression of the graft. 30% of patients remain euthyroid without thyroxin therapy, which has not been the case in the reported patient, with no evidence of re-implanted tissue growth^(3,6,9,10).

Lingual thyroid surgical removal can be performed intraorally or via a median cervicotomy or a lateral pharyngotomy. Most cases described in the literature have been resolved using an intraoral approach^(3,5,6,10). Various modifications of this approach, such as full thickness incisions in the cheek, median labiotomy, median labiomandibulotomy, and median glossotomy, have been introduced in order to improve exposure. Median glossotomy offers an

adequate esthetic result, with a lower risk of infection and postoperative orocervical fistula. However, the intraoral route provides a worse exposure for large masses and does not allow for an adequate control of potential intraoperative bleeding^(3,4,9,10).

The cervical approach allows for a better global visualization of the lesion, and proves especially useful in large size and posterior location cases such as the reported patient. Intraoperative bleeding is less likely to occur, and when it does, it is easier to control. On the other hand, cervical scar and postoperative orocervical fistula are its main disadvantages. It can be performed via an intra-, supra-, or infrahyoid median pharyngotomy, or else through a lateral pharyngotomy^(2,3,5,6,9,10). Laser and radiofrequency have also been proposed, but they are not widely used for that purpose, so surgery remains the first choice⁽⁸⁻¹⁰⁾. Institutions with robotic surgery devices can use this technology to perform the procedure intraorally⁽¹²⁾.

REFERENCES

1. Sauk JJ. Ectopic lingual thyroid. *J Pathol.* 1970; 102: 239-43.
2. Marín Fernández AB, Fernández Solís J. Tiroides ectópico lingual. Presentación de un caso y revisión del tratamiento. *Rev Mex Cir Bucal Maxilofac.* 2012; 8: 15-7.
3. Zubillaga Rodríguez I, Sánchez Aniceto G, García Recuero I, Montalvo Moreno JJ. Tiroides lingual: un nuevo abordaje quirúrgico. *Rev Esp Cir Oral Maxilofac.* 2004; 26: 316-24.
4. Betancourt AF, Bonnet II, Benedetti-Padrón I, Sáenz JC. Tiroides lingual, revisión del tema. *Rev Colomb Cir.* 2010; 25:231-6.
5. Singh S, Chopra B, Singh A. A rare case of lingual thyroid with hypothyroidism. *Int J Sci Res.* 2015; 4: 7-9.
6. Ghiasi S, Bayazian G, Khansarinia A. Dysphagia and dyspnea by lingual thyroid mass: An appropriate approach. *Egypt J Ear Nose Throat Allied Sci.* 2015; 16: 95-9.
7. Hickman W. Congenital tumour of the base of the tongue, pressing down the epiglottis on the larynx and causing the death of by suffocation sixteen hours after birth. *Trans Pathol Soc. (London)* 1869; 20: 160-3.
8. Aleid H, Alharbi A. Lingual thyroid: a systematic review of hormonal suppression treatment. *J Otolaryngol ENT Res.* 2015; 2: 00026.
9. Haque JA, Rahman HA, Jahan WA. Lingual thyroid diagnosed as a cause of cretinism: A case report. *J Curr Adv Med Res.* 2014; 1: 42-5.
10. Khamassi K, Jaafoura H, Masmoudi F, Lahiani R, Bougacha L, Ben Salah M. Ectopic lingual thyroid. *Case Rep Pediatr.* 2015; 2015: 252357.
11. Amr B, Monib S. Lingual thyroid: A case report. *Int J Surg Case Rep.* 2011; 2: 313-5.
12. Teo EH, Toh ST, Tay HN, Han HJ. Transoral robotic resection of lingual thyroid: case report. *J Laryngol Otol.* 2013; 127: 1034-7.