

Submitted: 3 Mar 2014

Accepted: 9 Jul 2014

¹ Department of Dermatology and Venereology, Sree Narayana Institute of Medical Sciences, Chalakka, North Kuthiyathodu Ernakulam, 683594, Kerala, India² Department of Surgery, Sree Narayana Institute of Medical Sciences, Chalakka, North Kuthiyathodu Ernakulam, 683594, Kerala, India

Abstract

Ectopic lingual thyroid is a rare developmental anomaly. It is caused by aberrant embryogenesis during the thyroid descent to the neck. It may remain asymptomatic or present with dysphagia, hemoptysis, dyspnoea or dysphonia. Clinically, it presents as a mass lesion on the base of the tongue. The most important diagnostic tool for an ectopic lingual thyroid is the ^{99m}Tc radionuclide scan, but imaging modalities such as computed tomography scan and magnetic resonance imaging may also help to assess its location and extent and to rule out the presence of normal thyroid tissue in the thyroid bed. The management of an ectopic thyroid remains controversial. No treatment is required for asymptomatic patients in the euthyroid state. Patients with hypothyroidism should be treated with thyroid hormone substitution therapy. Malignant transformation is an indication for complete surgical resection. Ablative radioiodine therapy can be considered for older patients and those who are unfit for surgery. In complicated cases, surgical resection is recommended through the transoral, transhyoid or lateral pharyngectomy approach. We report a case of ectopic lingual thyroid in a 35-year-old man who presented with nasal twang and hemoptysis.

Keywords: ectopic, lingual thyroid, hemoptysis, hypothyroidism, radionuclide scan

Introduction

Ectopic lingual thyroid is a rare developmental anomaly. The reported prevalence of the disease is 1 in 100 000 (1). Lingual thyroid is caused by aberrant embryogenesis during the descent of the thyroid gland to the neck (2). Embryonic development starts at 24 days after fertilisation in the primitive hypopharynx. The thyroglossal tube is a narrow tube connecting the thyroid gland to the tongue. Foramen caecum is the opening in the tongue. The thyroglossal duct joins the ultimobronchial body and develops into a functional and mature thyroid gland by the third foetal month. Lingual thyroid was first described by Hickman in 1869. It is the most common location for an ectopic thyroid. Multinodular goiter is rare in the lingual thyroid gland, and only a few cases have been reported in the literature. Ectopic thyroid can occur between the geniohyoid and the mylohyoid (sublingual thyroid) or prelaryngeally. Other rare sites include the mediastinum, precardial sac, heart, breast, pharynx, oesophagus, trachea, lung, duodenum, mesentery of the small intestine, and adrenal gland.

Case report

A 35-year-old male patient presented with complaints of two episodes of hemoptysis in the past one month. He had a history of nasal twang in his voice since childhood. He gave a history of fatigue and constipation for the past one year. He had no history of fever, weight loss or chronic cough. His body mass index was 23. Upon examination of the oral cavity, a swelling of size 5 cm x 4 cm x 2 cm in the posterior aspect of the tongue was noted (Figure 1). It was firm in consistency and not tender. No evidence of palpable thyroid was found in the normal location. Cervical lymphadenopathy was not found. Chest X-ray was normal. Sputum acid-fast bacilli was negative. Thyroid function tests showed a triiodothyronine level of 27 ng/dL (normal range 80 ng/dL–200 ng/dL), thyroxine level of 1.1 µg/dL (normal range 4.5 µg/dL–12.5 µg/dL), and thyroid stimulating hormone level of more than 150 µIU/mL (normal range 0.5 µIU/mL–4.7 µIU/mL), which are suggestive of hypothyroidism. Radionuclide thyroid scan was conducted by the intravenous administration of 5mCi of ^{99m}TcO₄. It showed a large nodular

swelling in the posterior one-third of the tongue with a diffused isotope concentration suggestive of an ectopic lingual thyroid in hypofunction (Figure 2) and no evidence of functioning thyroid tissue in the thyroid bed. Magnetic resonance imaging (MRI) scan indicated a lingual thyroid with a non-visualised thyroid gland in the normal location. Patient was started on thyroid hormones. Patient symptomatically improved in quality of speech and from hypothyroidism. No further episodes of hemoptysis were observed.

Discussion

Ectopic thyroid is a condition characterised by the ectopic location of the thyroid gland, such as lingual, sublingual, prelaryngeal or substernal (3). This condition is common in females, with a female-to-male ratio of 4:1. It usually manifests during childhood, adolescence and menopause, when the demand for thyroid hormone is at



Figure 1: Lingual thyroid (arrow).

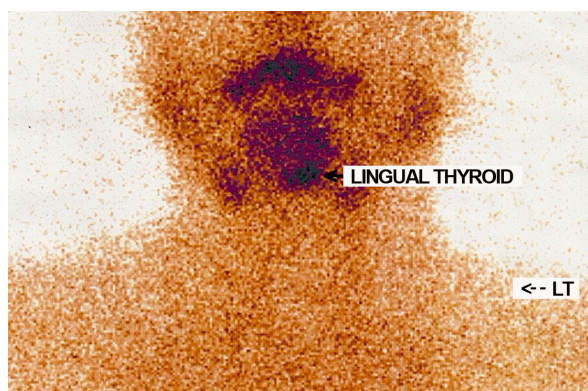


Figure 2: $^{99m}\text{TcO}_4$ radionuclide thyroid scan.

the maximum level. The circulating thyroid-stimulating hormone increases, leading to the enlargement of the ectopic thyroid. Around 33–62% of patients with ectopic thyroid show evidence of hypothyroidism (4).

An ectopic lingual thyroid may remain asymptomatic. It can present with dysphagia, dysphonia, dyspnoea or bleeding (5). Our patient presented with nasal twang and hemoptysis. Upon clinical examination, a lingual thyroid presents as a firm localised mass lesion on the base of the tongue. Various diagnostic modalities are used to confirm the condition, such as Tc 99m pertechnate scan, computed tomography (CT) scan and MRI scan. Thyroid scintigraphy can show the uptake in the base of the tongue in case of a lingual thyroid and helps to rule out the presence of the thyroid gland in its normal location. CT scan can show the high-density mass in the base of the tongue. Imaging modalities help us to determine the location and the extent of the lesion. An interesting case report by Bianco et al. described the role of preoperative carotid arteriography in a lingual thyroid (6). The study showed blood supply from the small branches of the lingual arteries to the lingual thyroid and not from the thyroid arteries. This inference from the arteriography helped the surgeon to predict a lesser chance of bleeding during transoral approach.

Differential diagnoses of a lingual thyroid include lymphangioma, adenoma, fibroma, minor salivary gland tumours, and angioma and midline bronchial cysts (4). A case report described an ectopic lingual thyroid presenting along with a thyroglossal cyst (7). The complications of an ectopic thyroid gland include airway obstruction, dysphagia, cystic degeneration, and rare malignancy.

The management of an ectopic lingual thyroid is controversial. If the patient is asymptomatic with euthyroid status, no treatment is required. In case of hypothyroidism, thyroid hormone substitution therapy should be started. Our patient presented with hypothyroidism and was started on thyroid hormone substitution therapy. In complicated cases or symptomatic patients, surgical procedures may be needed. Radioiodine ablation is an option among patients who are unfit for surgery and among elderly patients. This treatment should be followed by thyroid hormone substitution. However, radioiodine ablation should be avoided in children because of its damaging effects on the gonads (8). Ensuring that the ectopic lingual thyroid is the only thyroid gland available is mandatory. If the ectopic thyroid

is the only thyroid tissue, then transplantation of the thyroid tissue can be conducted (5). The surgical procedure required is total resection of the ectopic thyroid tissue. Various approaches to this procedure are available, such as transoral, transhyoid or lateral pharyngotomy (9). Recent reports have discussed transoral robotic-assisted surgery (TORS) using the Da Vinci surgical system for the transoral approach to the oropharynx (10). One advantage of TORS is a better view of the surgical field with greater magnification. Four-handed surgery, including the robotic arms, gives better manipulation and retraction comparable with open surgery and six degrees of motion. Post-operatively, temporary tracheostomy or the maintenance of the nasotracheal tube is needed for 24 h.

Conclusion

Ectopic lingual thyroid is a rare congenital anomaly. It may present as an asymptomatic lesion. In patients presenting with hypothyroidism, thyroid hormone substitution is needed. In symptomatic patients, surgical resection is recommended. If the ectopic thyroid is the only thyroid lesion, then lifelong thyroid hormone therapy is required.

Acknowledgement

None.

Conflict of Interest

None.

Funds

None.

Authors' Contribution

Conception and design, analysis and interpretation of the data, drafting of the article, final approval of the article: KP, KBA

Critical revision of the article for the important intellectual content, provision of study materials or patient, statistical expertise, obtaining of funding, administrative, technical or logistic support, collection and assembly of data: KP

Correspondence

Dr Krishnan Prasad
MBBS, MS, FMAS, DNB GI Surgery
Department of Dermatology and Venereology
Sree Narayana Institute of Medical Sciences
Chalakkal, North Kuthiyathodu Ernakulam
683594, Kerala
India
Tel: +91-984733 8520
Fax: +91-484-247 8093
Email: dr_kprasad@yahoo.com
drkprasad2000@gmail.com

References

1. Farell M L, Forer M. Lingual Thyroid. *Aust N Z J Surg.* 1994;**64**:135–138.
2. Léger J, Marinovic D, Garel C, Bonaiti Pellie C, Polak M, Czernichow P. Thyroid developmental anomalies in first degree relatives of children with congenital hypothyroidism. *J Clin Endocrinol Metab.* 2002;**87**(2):575–580. doi: <http://dx.doi.org/10.1210/jcem.87.2.8268>.
3. Ulug T, Ulubil SA, Alagol F. Dual ectopic thyroid: report of a case. *J Laryngol Otol.* 2003;**117**(7):574–576.
4. Hazarika P, Siddiqui SA, Pujary K, Shah P, Nayak DR, Balakrishnan R. Dual ectopic thyroid: a report of two cases. *J Laryngol Otol.* 1998;**112**(4):393–395.
5. Gallo A, Leonetti F, Torri E, Manciooco V, Simonelli M, DeVincentiis M. Ectopic lingual thyroid as unusual cause of severe dysphagia. *Dysphagia.* 2001;**16**(3):220–223.
6. Maria Rita Bianco, Alessandro La Boria, Teresa Franco, Pierpaolo Ferrise, and Eugenia Allegra Ectopic thyroid with vascular anomalies. *Int Med Case Rep J.* 2013;**6**:55–58. doi: [10.2147/IMCRJ.S50219](https://doi.org/10.2147/IMCRJ.S50219).
7. Madana J, Kalaiarasi R, Yolmo D, Gopalakrishnan S. Simultaneous occurrence of a thyroglossal duct cyst and a lingual thyroid in the absence of an orthotopic thyroid gland. *J Laryngol Otol.* 2012;**126**(2):217–220. doi: [10.1017/S0022215111002970](https://doi.org/10.1017/S0022215111002970).
8. Alderson DJ, Lannigan FJ. Lingual thyroid presenting after previous thyroglossal cyst excision. *J Laryngol Otol.* 1994;**108**(4):341–343.
9. Mussak EN, Kacker A. Surgical and medical management of midline ectopic thyroid. *Otolaryngol Head Neck Surg.* 2007;**136**(6):870–872. doi: [10.1016/j.otohns.2007.01.008](https://doi.org/10.1016/j.otohns.2007.01.008).
10. Pellini R, Mercante G, Ruscito P, Cristalli G, Spriano G. Ectopic lingual goiter treated by transoral robotic surgery. *Acta Otorhinolaryngol Ital.* 2013;**33**(5):343–346.