

Lingual Ectopic Thyroid Gland with Subclinical Hypothyroidism: A Case Report

A. Khanam¹ and S.K. Dey²

¹Medical officer, Institute of Nuclear Medicine and Allied Sciences, Faridpur

²Director, Institute of Nuclear Medicine and Allied Sciences, Faridpur

Abstract

Lingual ectopic thyroid gland is a rare developmental disorder of the thyroid gland. A young female patient with soft tissue swelling at the base of the tongue was referred to Institute of Nuclear Medicine and Allied Sciences, (INMAS), Faridpur for evaluation. Using ultrasonography and Tc-99m scintigraphy, the swelling was diagnosed as an ectopic lingual thyroid gland. Thyroid hormones (S. T₃ & S. T₄) as well as thyroid stimulating hormone (TSH) levels were also evaluated, and the patient was managed medically.

Keywords: Ectopic thyroid, ultrasonography, scintigraphy

1. Introduction

Thyroid gland is an endocrine gland normally located at the lower anterior neck in front of 2nd to 5th rings of trachea, which plays a vital role in the metabolic functions of the body by producing the hormone thyroxine. It develops during 3rd to 4th week of gestation, primarily from the endodermal thyroid diverticulum extending downwards from the floor of the primitive foregut along the midline [1, 2]. Ectopic thyroid gland is a rare occurrence, affecting approximately 1 in 100,000 to 300,000 patients with a female preponderance, and is often identified incidentally due to being asymptomatic [3, 4]. Here we present a case of a young woman with ectopic thyroid at the base of tongue, and discuss the approach to diagnosis and management.

2. Case Report

A 22 year old female patient from Madhukhali, Faridpur presented with the complaints of swelling at the back of her tongue for 3 years, with occasional pain and difficulty in swallowing. She gave no history of fever, weight loss, cough, bleeding from the swelling, difficulty in breathing or malodorous breath. She had no known co-morbidities, no history of surgery or regular medication aside from occasional intake of antiulcerants. She had no behavioural or menstrual abnormality. None of her family members had similar illness. Her body built was average; vital signs were within normal limit.

On examination, there was a reddish swelling at the base of the tongue near tonsillar fossa. Palatine and nasopharyngeal tonsils were not enlarged. There was no other abnormality in the oral cavity. No enlarged lymph nodes were palpable in neck.

Ultrasound examination was done using Mindray DC-7 machine and a 7-12 MHz linear array transducer, with the patient in supine position. On ultrasound examination, the thyroid bed was found empty. A well-demarcated soft tissue structure with uniform echotexture was seen in the lingual region measuring 2.7 × 2 cm. The structure was separated from the surrounding muscles and great vessels, but closely adherent to the base of the tongue. On colour Doppler sonography, peripheral vascularity was noted.

Parotid and submandibular glands were normal, and no enlarged lymph nodes were seen in the neck.

The patient underwent Tc-99m thyroid scintigraphy using Symbia S dual head SPECT camera. Acquisition was taken 20 minutes after IV injection of 3 mci Tc-99m pertechnetate. Scan showed no tracer uptake in the usual thyroid bed. Tracer was concentrated in the region of the lingual swelling, indicating ectopic lingual thyroid gland.

Serum T₃ and T₄ were estimated using radioimmunoassay (RIA) and TSH was estimated using immunoradiometric assay (IRMA). Biochemical examination showed normal S.T₃ and S.T₄ levels (S. T₃ 1.43 ng/ml, S. T₄ 96.1 ng/ml) with raised S.TSH (11.44 μIU/ml), which indicated subclinical hypothyroidism.

Following confirmation of diagnosis, the patient was treated with levothyroxine replacement therapy 50 μgm daily orally, and advised for regular follow up.

3. Results and Discussion

Ectopic thyroid gland is caused by varying degrees of abnormal migration of thyroid tissue during development. Commonly the ectopic gland is situated along the course of the thyroid diverticulum, though ectopia at distant sites i.e. mediastinum, gall bladder, adrenal glands, ovaries, gut and mesentery, have also been reported [2, 5]. The ectopic tissue may be present in conjunction with normal orthotopic thyroid gland, but in 70-90% cases the ectopic gland is the only thyroid tissue present [2]. The most common form of ectopia is lingual thyroid, in which the gland develops at the base of the tongue near the site of foramen caecum. In about 70-75% cases the lingual thyroid is not accompanied by an orthotopic thyroid, indicating a failure of the diverticulum to descend along the normal course. Hypothyroidism is found in most patients, but a euthyroid state may be present due to thyroxine produced by the ectopic gland, which raises the probability of delayed diagnosis [2, 4-6].

Aside from the symptoms of hypothyroidism, the patients present with local symptoms due to hypertrophy of the ectopic tissue. This is believed to be caused by stimulation by raised TSH in the circulation in response to the inadequate production of thyroxine by the gland [2, 5].

*Corresponding author: khanamafrida006@gmail.com

Symptoms of lingual thyroid enlargement include dysphonia, respiratory tract obstruction, foreign body sensation, cough, sleep apnoea and dysphagia. In our case, the patient only presented with symptoms caused by the swelling, but had no signs or symptoms relating to clinical hypothyroidism. Generally transoral examination reveals a smooth pink or reddish swelling of variable size at the base of the tongue; fiberoptic laryngoscopy may be required to visualize the swelling in some cases.



Fig. 1: Transoral examination showing a reddish swelling at the base of the tongue

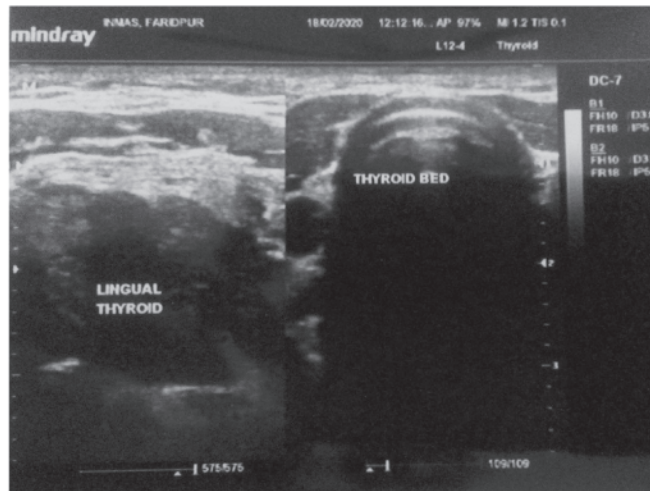


Fig. 2: Ultrasound examination showing empty thyroid bed and ectopic thyroid tissue in lingual region

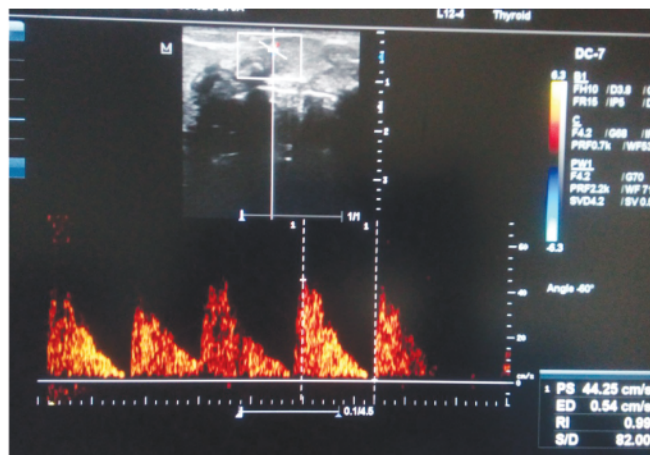


Fig. 3: Peripheral vascularity around ectopic thyroid tissue

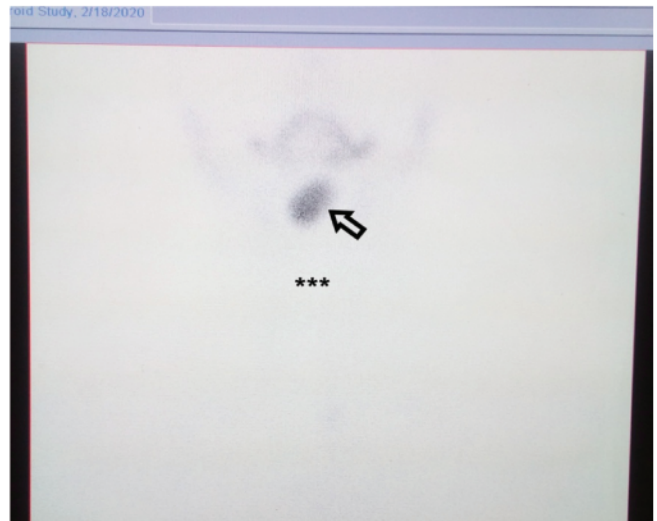


Fig. 4: Tc-99m scan showing no uptake in usual thyroid bed (***) and ectopic thyroid in lingual region with tracer concentration (arrowhead)

When suspected, a series of diagnostic tests are required in order to exclude the possible differentials, which include various adenoma and cysts of the pharynx, tonsillar enlargement and metastatic deposits [2]. Evaluation of the thyroid hormone status not only aids in a complete diagnosis but also is important in planning the management. Ultrasonography aids in diagnosis by evaluating the ectopic tissue as well as the thyroid bed. Grayscale sonography has been found to be equally sensitive in detecting ectopic thyroid tissue as magnetic resonance imaging (MRI), with a sensitivity of 70%. Moreover, the sensitivity is increased to upto 90% with the demonstration of internal or peripheral vascularity using colour Doppler ultrasound [7]. Computed tomography (CT) scans and MRI are valuable tools for acquiring more precise anatomical information such as the location, size and extent of ectopic tissue [2].

Thyroid scintigraphy remains the most reliable noninvasive test for confirming the presence of ectopia, showing tracer uptake in the ectopic tissue instead of the normal thyroid bed. However, in presence of an orthotopic thyroid, the uptake in the ectopic site may be masked, necessitating correlation with CT scan, MRI or cytology [2].

Fine needle aspiration cytology (FNAC) and histopathology can also confirm ectopia with a high accuracy, and are the only modalities to differentiate between benign and malignant swelling. Although primary malignancy in the ectopic thyroid tissue is uncommon, it is found in some cases, often in presence of an orthotopic thyroid as well. Most of these malignancies are of papillary variety. It is, however, difficult to differentiate between primary carcinoma of ectopic thyroid and metastatic deposit from carcinoma involving the orthotopic thyroid [2, 4, 5].

The choice of management of lingual ectopic thyroid is multifactorial, depending on the size, symptoms, thyroid hormone status and possibility of complication. Asymptomatic patients with euthyroid state require no intervention but should undergo regular follow up. For patients with hypothyroidism and mild local symptoms

similar to our patient in this case, it is recommended to start levothyroxin replacement therapy [2]. Generally subclinical cases with S.TSH below 10 μ IU/ml do not require thyroxine replacement therapy, but the risk of overt hypothyroidism increases with this approach, and starting levothyroxine may be useful by halting further growth of the tissue by suppressing TSH [8]. Surgical management is required in cases of malignancy and also in case of benign enlargement causing severe local symptoms i.e. dysphagia or airway obstruction. Complete excision or partial reduction followed by levothyroxin therapy is reportedly effective in cases of benign swelling [4, 9]. Some authors also recommend surgical excision in even smaller benign swellings due to the risk of malignant transformation. Radioiodine ablation using I-131 is reserved for patients who are not suited for surgery, as it requires a higher dose than that necessary for orthotopic thyroid, and carries an increased risk of complication [2, 5]. Keeping in mind the prognosis and complications of an untreated lingual thyroid gland, it is important to evaluate and confirm diagnosis at an early stage, as doing so may provide a scope of medical management, and prevent the necessity for surgical intervention.

References

1. S. Standring, ed. Gray's anatomy: the anatomical basis of clinical practice, 41st ed. (Elsevier health sciences), 470, (2015).
2. G. Noussios, P. Anagnostis, D.G. Goulis, D. Lappas and K. Natsis, Ectopic thyroid tissue: anatomical, clinical, and surgical implications of a rare entity, *Eur J Endocrinol*, **165**(3), 375, (2011).
3. D. Benedetto, Ectopic thyroid gland in the submandibular region simulating a thyroglossal duct cyst: a case report, *J Pediatr Surg*, **32**(12), 1745-6, (1997).
4. G. Thomas, R. Hoilat, J.S. Daniels and W. Kalagie, Ectopic lingual thyroid: a case report, *Int J Oral Maxillofac Surg*, **32**(2), 219-21, (2003).
5. G. Guerra, M. Cinelli, M. Mesolella, D. Tafuri, A. Rocca, B. Amato, S. Rengo and D. Testa, Morphological, diagnostic and surgical features of ectopic thyroid gland: a review of literature, *Int. J Surg*, **12**, S3-S11, (2014).
6. M. Kaplan, R. Kauli, E. Lubin, M. Grunebaum and Z. Laron, Ectopic thyroid gland: a clinical study of 30 children and review, *J Pediatr*, **92**(2), 205-9, (1978).
7. H. Ohnishi, H. Sato, H. Noda, H. Inomata and N. Sasaki, Color Doppler ultrasonography: diagnosis of ectopic thyroid gland in patients with congenital hypothyroidism caused by thyroid dysgenesis, *J Clin Endocrinol Metab*, **88**(11), 5145-9 (2003).
8. A.S. Fauci, E. Braunwald, D.L. Kasper, S.L. Hauser, D.L. Longo, J.L. Jameson and J. Loscalzo, *Harrison's principles of internal medicine*, 17th ed. (New York: McGraw Hill Professional), 2233 (2008).
9. A. Gallo, F. Leonetti, E. Torri, V. Manciooco, M. Simonelli and M.D. Vincentiis, Ectopic lingual thyroid as unusual cause of severe dysphagia, *Dysphagia*, **16**(3), 220-3 (2001).

