

## LATE ONSET DYSPHAGIA AFTER THYROIDECTOMY: THYROID REMNANT OR THYROGLOSSAL HYPERPLASIA?

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### Abstract

Lingual thyroid is a rare developmental disorder, and it is the result of failure of the thyroid gland to descend from the tongue root to its normal site. The ectopic thyroid with a functioning gland is even rarer. In this case, we present a 44-year-old female patient with a complaint of foreign body sensation, progressive dysphagia and dyspnea due to ectopic thyroid tissue. Her complaints were totally resolved after L-thyroxine treatment. Here we report a patient with functioning ectopic thyroid tissue who had had bilateral subtotal thyroidectomy 10 years ago due to multinodular goiter. In conclusion, if progressive dysphagia, dyspnea, and foreign body sensation occurs in a patient who had thyroidectomy and living in an endemic goiter region, lingual thyroid may be underlying disorder.

**Key words:** thyroglossal hyperplasia, dysphagia, dyspnea, lingual thyroid

## INTRODUCTION

Lingual thyroid, which occurs due to the defective descent of thyroid gland to the normal position at the base of the thyroglossal duct, and it is a rare congenital anomaly (1).

Ectopic lingual thyroid tissue may be localized at sublingual, submandibular, esophageal, mediastinal region. The prevalence of lingual thyroid is 1 in 1000000 patients, 4-7 times greater in women compared to men. It is seen more common in the third decade of life (1-2). However, the present case was not associated with endemic reason and could possibly be due to compensatory thyroid remnant hypertrophy of a pyramidal lobe after insufficient removal and insufficient hormone replacement.

Patients with lingual thyroid may be asymptomatic or associated with dyspnea,

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dysphagia, dysphonia, bleeding, foreign body sensation of throat. Also, hypothyroidism may also be seen in lingual thyroid cases. The aim of this report is to add a new case to the literature in terms of the diagnosis and management of functional lingual thyroid tissue, and to discuss these topics (3-5).

## CASE REPORT

A 44 year old woman was admitted to the outpatient clinic due to swelling at the base of tongue, foreign body sensation in the throat, progressive dysphagia and cough for 8 months. Dysphagia was not prominent but the patient claimed that she had difficulty especially while swallowing stiff food. Ten years ago, the patient had bilateral subtotal thyroidectomy due to multinodular goiter. She was living in an endemic goiter region. There was a 3x2 cm mass at the root of the tongue in the patient's oral-pharyngeal examination. The other part of physical examination and biochemical parameters were normal.

The patient had no thyroid hormone replacement therapy for the last six years. The serum free T3:2.61 pg/ml (N:2.5-3.9), free T4: 0.76 pg/ml (N: 0.61-1.12), TSH: 7.6  $\mu$ IU/ml (range: 0.34-5.60) levels revealed subclinical hypothyroidism. Serum anti-thyroidperoxidase antibody levels were normal.

A thyroid scan with Tc<sup>99</sup>-pertechnetate found bilateral thyroid tissue left after subtotal thyroidectomy as well as between these subtotally operated lobes (Fig. 1). Thyroid replacement therapy was started after the diagnosis of hypothyroidism. Two months later, the complaints regarding hypothyroidism and mass effect decreased. She was followed up 11 months and during the last control, the patient

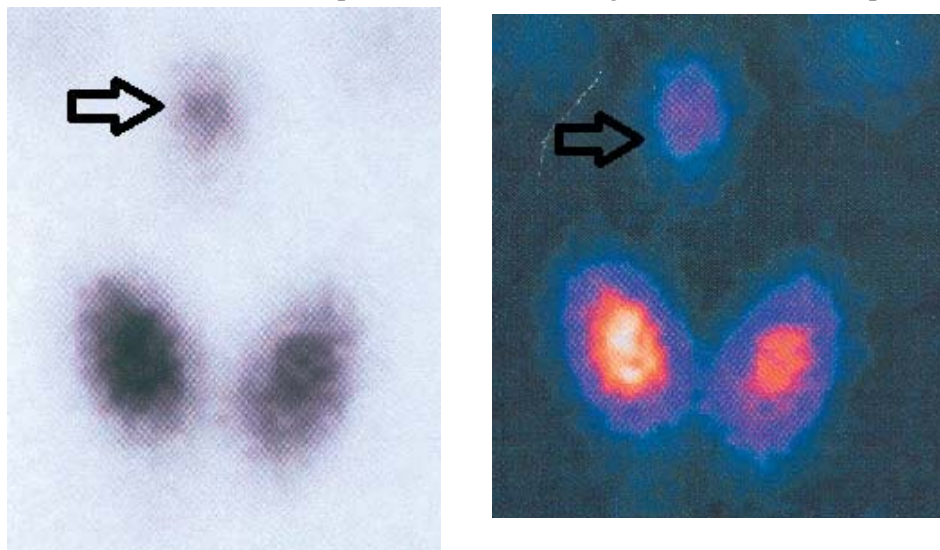


Figure 1. Thyroid scan-showing Tc<sup>99</sup>-pertechnetate uptake along thyroglossal duct and thyroid remnants after thyroidectomy

was completely asymptomatic in terms of hypothyroidism and mass effect. In this case, surgical operation was not planned for lingual thyroid due to the loss of symptoms and signs related to mass effect such as dysphagia and dyspnea.

## **DISCUSSION**

Lingual thyroid is a rare condition localized at the midline of tongue root. Its size may vary from millimeters to centimeters. Symptoms and signs may change according to the size of ectopic gland. The incidence has been reported to be 1 in 100,000. However, the prevalence of ectopic thyroid tissue is 10 % in postmortem reports. The first patient with lingual thyroid was reported by Hickman *et al.* in 1869. The case died after the birth due to respiratory stress (5-7). Lingual thyroid is more common in females in comparison to males. The symptoms and signs of these patients are commonly seen in puberty, pregnancy and menopause due to increased serum TSH levels causing the gland hypertrophy. Also, the need for thyroid hormone-thyroxine as a replacement therapy increases in these periods (8-10).

The patient with lingual thyroid became symptomatic after bilateral subtotal thyroidectomy. The case did not use thyroid hormone replacement therapy for 6 years. During this period, increased TSH levels after surgery caused hypertrophy of the ectopic thyroid tissue. In order to synthesize enough thyroid hormone, the thyroid tissue became hyperplastic and this mass effect resulted in progressive dysphagia. Also, the living in an endemic goiter region has a role in the development of the tissue hypertrophy and mass effect (11-13).

In the diagnosis of lingual thyroid, sonography, computerized tomography, magnetic resonance imaging and thyroid scan can be used after physical examination. The scintigraphy can have a more important role for the functionality of the tissue (13).

In the current patient, the diagnosis was confirmed by clinical findings, radiological studies and thyroid scan. Medical and surgical approaches are used in the management of lingual thyroid. Euthyroid and asymptomatic patients should be followed regularly. Radioactive iodine therapy is an alternative approach in the treatment of this disorder (13-14).

However, hypothyroidism may be seen after this therapy as a complication. In these patients, thyroid hormone replacement therapy has beneficial effects in terms of symptoms and signs due to hypothyroidism and mass effect as in our case (14-16).

There are many surgical methods in the management of lingual thyroid such as total thyroidectomy, transposition and trans-oral laser excision. If recurrent bleeding, progressive dysphagia, airway obstruction occurs in these patients, surgical therapy should be applied (13-14). These patients need regular follow-up visits, in order to monitor the state of thyroid function and the size and shape of the gland.

As in our case living in an endemic goiter region, if there is hypothyroidism, the most important symptoms related to mass effect are progressive dysphagia, cough and dyspnea due to this disorder. Ectopic thyroid tissue can be functional

after hypothyroidism and it causes mass effect as in our current report. Therefore, we should keep in mind that if progressive dysphagia occurs in a patient who had thyroidectomy and living in an endemic goiter region, lingual thyroid may be the underlying disorder (14-17). Though being a rare entity; malignant lingual thyroid, follicular cancer, or exceptionally medullary cancer, may occur in such ectopic thyroids (18).

**In conclusion**, if there is a mass at the root of the tongue, lingual thyroid or should be thought for differential diagnosis.

#### REFERENCES

1. Sauk JJ. Ectopic lingual thyroid. *J Pathol* 1970; 102: 239-243.
2. Farrel ML, Forer M. Lingual thyroid. *Aust N Z L Surg* 1994; 64:135-138.
3. Batsakis JG, El-Naggar AK, Luna MA. Thyroid gland ectopias. *Ann Otol Rhinol Laryngol* 1996; 105: 996-1000.
4. Gallo A, Leonetti F, Torri E, Manciocco V, Simonelli M, Devincetis M. Ectopic lingual thyroid as unusual cause of severe dysphagia. *Dysphagia* 2001; 16: 220-223.
5. Spieker MR. Evaluating dysphagia. *Am Fam Physician* 2000; 61(12):3639-3648.
6. Quarracino M, Aguas S. Lingual thyroid: a clinical case. *Med Oral* 2003; 8: 57-60.
7. Hickman W. Congenital tumor 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 Pathology Soc* 1869; 20: 160-163.
8. Erdogan MF, Agbaht K, Altunsu T, Ozbas S, Yucesan F, Tezel B, Sargin C, Ilbeg I, Artik N, Kose R, Erdogan G. Current iodine status in Turkey. *J Endocrinol Invest* 2009; 32(7):617-622.
9. Oppenheimer R. Lingual thyroid associated with chronic cough. *Otolaryngol Head Neck Surg* 2001; 125: 433-434.
10. Kumar V, Magendhar Y. Lingual thyroid gland: clinical evaluation and management. *Indian J Pediatr* 2004; 71: 1143-1147.
11. Lai YT, Chen HS, Chang YL. Lingual thyroid. *Otolaryngol Head Neck Surg* 2009;140: 944-945.
12. Sethi MJ, Bhatia V. Lingual thyroid: a case report and review of literature. *S D Med* 2009; 62: 13-14.
13. Huang TS, Chen HY. Dual ectopic thyroid: case series and review of the literature. *Head Neck* 2007; 29: 885-888.
14. Cappelli C, Gandossi E, Cumetti D, Castellano M, Pirola I, De Martino E, Agosti B, Micheletti L, Cherubini L, Mattanza C, Rosei E. Ectopic lingual thyroid tissue and acquired hypothyroidism: case report. *Ann Endocrinol* 2006; 67: 245-248.
15. Barthel A, Bornstein SR. Images in clinical medicine. Obstructive lingual thyroid. *N Engl J Med* 2005; 352(1):e1.
16. Kumar V, Nagendhar Y, Prakash B, Chattopadhyay A, Vepakomma D. Lingual thyroid gland: clinical evaluation and management. *Indian J Pediatr* 2004; 71: 62-64.
17. Prasad KC, Bhat V. Surgical management of lingual thyroid: a report of four cases. *J Oral Maxillofac Surg* 2000; 58: 223-227.
18. Hari CK, Kumar M, Abo-Khatwa MM, Adams-Williams J, Zeitoun H. Follicular variant of papillary carcinoma arising from lingual thyroid. *Ear Nose Throat J* 2009; 88(6):E7.