Hypertrophic Lingual Thyroid Causing Sleep Apnea

Case Report

Harun Soyalıç[1], Erkan Gökçe[2], Battal Tahsin Somuk[1], Can Koçak[1], Ahmet Eyibilen[1]

[1]Department of Otolaryngology Gaziosmanpaşa University Faculty of Medicine, Tokat, Turkey

[2]Department of Radiology Gaziosmanpaşa University Faculty of Medicine, Tokat, Turkey

Abstract

Ectopic lingual thyroid disorder, developing as a consequence of thyroid gland migration during early embryogenesis, is a rarely seen congenital anomaly. In this paper, we report a 39-year-old male patient with snoring and moderate-level obstructive sleep apnea who received the diagnosis of lingual thyroid at our clinic. We treated the patient with thyroxin suppression therapy. During our clinical follow-ups, the amount of lingual thyroid tissue significantly decreased, and the symptoms of obstructive sleep apnea disappeared.

Key Words: Lingual thyroid, obstructive sleep apnea, thyroxin suppression therapy

Introduction

Lingual thyroid is a rare developmental anomaly of the thyroid gland. It is characterized by a complete or partial thyroid tissue descending from the foramen cecum to the normal pre-tracheal thyroid location in the tongue. Ectopic lingual thyroid is often asymptomatic, but it can lead to dysphonia, dyspnea, dysphagia, foreign body sensation in the throat, and hemoptysis. Obstructive sleep apnea syndrome (OSAS) is a disease characterized by respiratory arrest or periodic airflow reduction as a result of partial or complete collapse of the upper respiratory tract while asleep. Lingual thyroid is a rarely seen cause of OSAS. In this paper, a case of hypertrophic lingual thyroid causing OSAS and snoring is presented, and the literature is reviewed.

Case Presentation

A 39-year-old male patient applied to our clinic with complaints of recently developed morning and daytime fatigue, a foreign body sensation in the throat, snoring, and disruption of night sleep due to shortness of breath. The mid-line of the soft palate and nasal septum appeared normal upon flexible fiberoptic examination of the patient. On the base of the tongue, a vascularized mass (approximately 3x2 cm) that was pink/purple in color and smooth-surfaced was settled slightly on the left side of the centerline (Figure 1). An REM-dependent moderate-level OSAS was identified; apnea-hypopnea index (AHI) was 15.3 (AHI-REM 56.8, AHI-non REM 5.5) via polysomnography (PSG).

During neck tomography, a mass lesion that was settled at the base of the tongue with a smooth lobulated contour indenting into the oropharyngeal air passage and left vallecula was observed. In addition, ectopic lingual thyroid tissue showing intense contrast enhancement inside of a slightly heterogeneous internal structure was also observed. Density belonging to the thyroid gland was not seen in the normal location of the thyroid gland (Figure 2). No thyroid tissue-compatible activity involvement was observed by 5 cmC Tec-99 m pertechnetate thyroid scintigraphy. Thyroid tissue-compatible focal uptake focus was observed at the base of the tongue. TSH and FT4 were identified as follows: TSH=48.95 (normal range: 0.27-5.6 μIU/mL), free T4 = 0.57 (normal range: 0.93-1.7 ng/dL). As a result, the patient received thyroxin suppression therapy. In the third month of the patient's treatment, the thyroid function tests returned to normal, and a significant reduction in the dimensions of the lingual thyroid on the base of the tongue was observed. Surgical treatment was not deemed necessary. A PSG was not repeated, since the symptoms of the patient's sleep apnea disappeared. A slight mucosal swelling that was observed during his 2-month fiberoptic control examination is shown in Figure 3.

Discussion

Lingual thyroid was first documented by Godart in 1760 and was reported by Hickman in the medical literature in 1869 (1, 2). There are more than 400 published lingual thyroid cases (1). The prevalence of lingual thyroid is 1/100,000-300,000. Its clinical incidence has been reported to be 1/4000 to 1/10,000 (3-6). It occurs at a rate 4-7 times higher in women than in men (4, 6).

The ectopic thyroid tissue settles at the base of the tongue at a rate of 90%. In addition, ectopic

Address for Correspondence:
Harun Soyalıç, Department of Otolaryngology Gaziosmanpaşa University Faculty of Medicine, Tokat, Turkey
Phone: +90 505 560 40 26
E-mail: harun_soyalic@hotmail.com

Received Date: 30.10.2013
Accepted Date: 26.02.2014

www.turkarchotolaryngol.net
DOI:10.5152/tao.2014.186

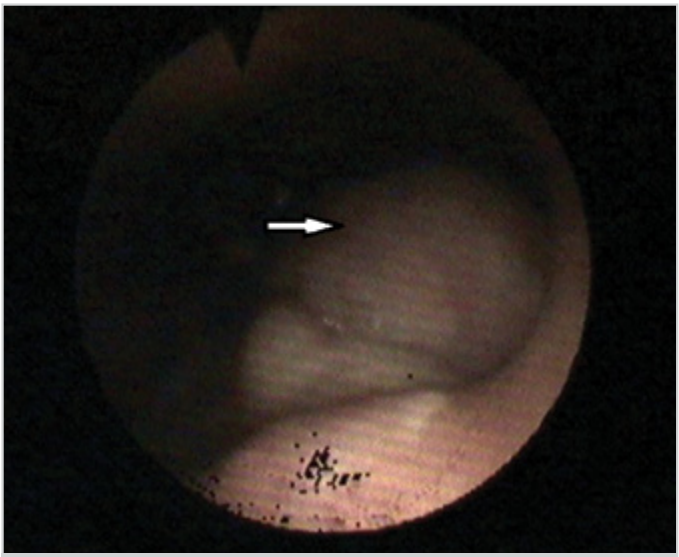

Figure 1. Pre-treatment clinical image of the lingual thyroid (white arrow)

Figure 2. Ectopic lingual thyroid under contrast-enhanced computed tomography (white arrow)

thyroid with sublingual, submandibular, prelaryngeal, tracheal, esophageal, and mediastinal locations may be observed. Ectopic thyroid tissue located in the mesentery of the mediastinum, heart, porta hepatis, diaphragm, lung, duodenum, adrenal gland, and the small intestine has also been reported (3, 4). Adenoma, hyperplasia, inflammation, and malignancy seen in the normal thyroid gland can also be observed in the ectopic thyroid tissue (7). Differential diagnosis of lingual thyroid includes hemangioma, lymphangioma, fibroma, adenoma, lipoma, angioma mucous retention cyst, dermoid cyst, salivary gland tumor, tongue base tumor, and lingual thyroid cancer (3, 8).

A small number of oropharyngeal masses causing OSAS have been reported. While lipomas are the most frequently observed masses, other lipomas that have been reported are lymphoma, plasmacytoma, hemangioma, retention cysts, lingual tonsil hypertrophy, lingual cysts, and ectopic lingual thyroid (9). To date, six ectopic lingual thyroid cases causing OSAS have been reported (4, 10). Different treatment options, including hormone suppression, radioactive iodine ablation, or excision using various surgical approaches, have been applied to these reported cases.

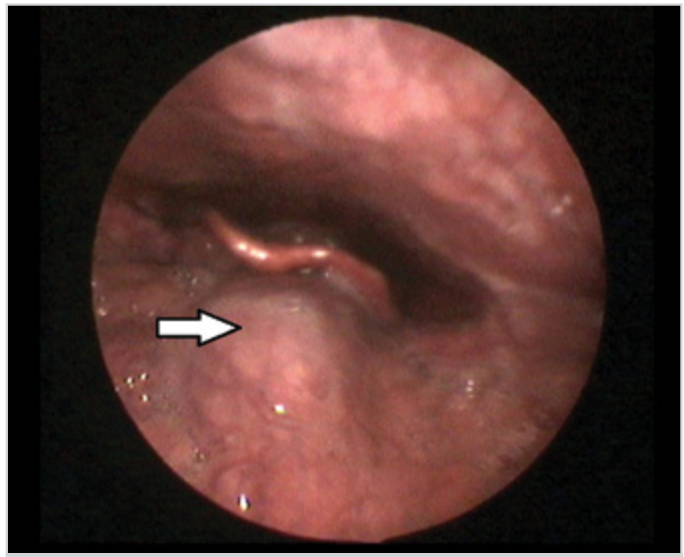

Figure 3. Post-treatment clinical image of the lingual thyroid (white arrow)

The ectopic lingual thyroid grows and produces more symptoms when endocrine changes take place, particularly during the periods of puberty, pregnancy, and menstruation. In the case of an overgrown lingual thyroid, snoring and obstructive sleep apnea can be observed (4, 10, 11). There is hypothyroidism and increased TSH levels in 33-62% of the cases with ectopic lingual thyroid (3). In addition, our patient received thyroxin treatment for 10 years after being diagnosed with hypothyroidism. However, the increase in TSH due to the patient's discontinuation of the drug caused the growth of the lingual thyroid. The disappearance of snoring and OSAS after the reduction of the lingual thyroid through the use of suppression therapy has suggested to us that the cause of the sleep apnea was related not only to hypothyroidism but also to the growth in the ectopic thyroid tissue on the base of the tongue. Lingual thyroid treatment involves either surgical or medical procedures, depending on the dimensions of the mass and the symptoms of the patient. Surgical therapy is indicated when there is growth, recurrent severe bleeding, and suspicion of dyspnea, dysphagia, malignancy, and uncontrolled hyperthyroidism in the lingual thyroid or in cases where the patient is not responding to suppression therapy (3, 5, 6). Our case responded to the thyroxin suppression therapy very well. There was a significant decrease in the lingual thyroid, and therefore, we did not need to pursue other treatment options. Radioactive iodine therapy may be an alternative approach for patients who refuse surgery or are not suitable for anesthesia (12). The other current treatments are radio-frequency ablation, excision with a CO_2 laser by transoral approach, or excision of lingual thyroid with a Coblator (11, 13, 14).

Conclusion

Endoscopic examination of the base of the tongue should absolutely be performed on thin patients who apply with OSAS clinical symptoms but do not have pathology of the nose or soft palate. It should be noted that sizable masses that have settled on the base of the tongue may result in sleep apnea. Whenever a mass is seen along the midline of the tongue base, an ectopic lingual thyroid should be considered in the differential diagnosis. As hypothyroidism may be seen in patients with ectopic lingual thyroid, these patients should be explicitly treated.

Informed Consent: Written informed consent was obtained from patients who participated in this case.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - H.S., E.G.; Design - H.S., BT.S.; Supervision - H.S., A.E.; Funding -H.S., C.K.; Materials - H.S., C.K.; Data Collection and/or Processing - H.S., E.G.; Analysis and/or Interpretation - H.S., BT.S.; Literature Review - H.S., A.E.; Writing - H.S., E.G.; Critical Review - H.S., A.E.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this case has received no financial support.

References

1. Kennedy TL, Riefkohl WL. Lingual thyroid carcinoma with nodal metastasis. Laryngoscope 2007; 117: 1969-73. [CrossRef]
2. Falvo L, Berni A, Catania A, D'Andrea V, Palermo S, Giustiniani C, et al. Sclerosing papillary carcinoma arising in a lingual thyroid: report of a case. Surg Today 2005; 35: 304-8. [CrossRef]
3. Toso A, Colombani F, Averono G, Aluffi P, Pia F. Lingual thyroid causing dysphagia and dyspnea. Case reports and review of the literature. Acta Otorhinolaryngol Ital 2009; 29: 213-7.
4. Peters P, Stark P, Essig G Jr, Lorincz B, Bowman J, Tran K, et al. Lingual thyroid: an unusual and surgically curable cause of sleep apnoea in a male. Sleep Breath 2010; 14: 377-80. [CrossRef]
5. Singhal P, Sharma KR, Singhal A. Lingual thyroid in children. J Indian Soc Pedod Prev Dent 2011; 29: 270-2. [CrossRef]
6. Rahbar R, Yoon MJ, Connolly LP, Robson CD, Vargas SO, McGill TJ, et al. Lingual thyroid in children: a rare clinical entity. Laryngoscope 2008; 118: 1174-9. [CrossRef]
7. Thomas G, Hoilat R, Daniels JS, Kalagie W. Ectopic lingual thyroid: a case report. Int J Oral Maxillofac Surg 2003; 32: 219-21. [CrossRef]
8. Cunningham CL, Vilela RJ, Roy S. Radiofrequency ablation as a novel treatment for lingual thyroid. Int J Pediatr Otorhinolaryngol 2011; 75: 137-9. [CrossRef]
9. Guerrissi JO. Follicular variant of papillary carcinoma in submandibular ectopic thyroid with no orthotopic thyroid gland. J Craniofac Surg 2012; 23: 138-9. [CrossRef]
10. Yaday S, Singh I, Singh J, Aggarwal N. Medullary carcinoma in a lingual thyroid. Singapore Med J 2008; 49: 251-3.
11. Barnes TW, Olsen KD, Morgenthaler TI. Obstructive lingual thyroid causing sleep apnea: a case report and review of the literature. Sleep Med 2004; 5: 605-7. [CrossRef]
12. Babademez MA, Günbey E, Acar B, Günbey HP. A rare cause of obstructive sleep apnea syndrome: lingual thyroid. Sleep Breath 2012; 16: 305-8. [CrossRef]
13. Lin WN, Lee LA, Wang CC, Li HY. Obstructive sleep apnea syndrome in an adolescent girl with hypertrophic lingual thyroid. Pediatr Pulmonol 2009; 44: 93-5. [CrossRef]
14. Patel Z, Johnson L. Iodine 131 ablation of an obstructive lingual thyroid. J Radiol Case Rep 2009; 3: 3-6.