

A CASE PRIMARY HYPOTHYROIDISM: LINGUAL THYROID

PRİMER HİPOTİROİDİLİ BİR OLGU:LİNGUAL TİROİD

Yasin ÖZTÜRK¹ ,Şenay ARIKAN DURMAZ² , Yunus İlyas KİBAR¹ , Ahmet TAY¹ ,
Mehmet Emin AYHAN¹

¹ Erzurum Region Training and Research Hospital, Department of Internal Medicine

² Erzurum Region Training and Research Hospital, Department of Endocrinology and Metabolism

Yasin Öztürk MD .- yozturk29@gmail.com

Şenay ARIKAN DURMAZ MD. Associate Prof. -sarikan822@gmail.com

Yunus İlyas Kibar MD. -yunusilyas@gmail.com

Ahmet Tay MD. - dr.atay@hotmail.com

Mehmet Emin Ayhan MD. - m.eminayhan@hotmail.com

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Correspondence Address: yozturk29@gmail.com

Phone: +90 0505 673 32 42

ABSTRACT

Lingual thyroid is an abnormal formation appearing as the result of a deficient descent of the thyroid gland to its normal pretracheal location through the thyroglossal duct during embryological development and its a rare embryological aberration. A 19 years old woman was admitted our hospital with the complaints of swallowing difficulty, foreign-body-feeling in her throat, swelling at the back of the tongue, deepening of her voice and constipation. Her physical examination showed a spherical, red, fleshy mass with regular contour located at the back of the tongue While thyroid scan with technetium 99m revealed isotop uptake at the base of the tongue, no uptake was detected at the normal location of the thyroid gland. According to these findings the diagnosis of lingual thyroid was detected. The surgical excision of the mass was not appropriate treatment since the lingual thyroid was the only functioning thyroid tissue and medical treatment was recommended. We aimed at presenting a case of lingual thyroid and hypothyroidism along with laboratory and imaging findings.

Keywords: Lingual thyroid, primary hypothyroidism, thyroid scintigraphy

ÖZET

Lingual tiroid, tiroidin embriyolojik gelişimi esnasında tiroglossal kanalın pretrakeal lokalizasyonuna inmesi sırasında oluşan yetersizlik sonucu görülen, nadir bir embriyolojik anomalidir. Ondokuz yaşında bir bayan yutkunma güçlüğü, boğazda yabancı cisim hissi, dil arkasında şişlik, kabızlık, sesle kalınlaşma şikayetleri ile başvurdu. Fizik muayenesinde dil arkasında küre şeklinde kırmızı düzgün yüzeyli kitle izlendi. Primer hipotiroidi tanısı konulan hastada Technetium 99 m ile tiroid bezi sintigrafik olarak incelendiğinde tiroid bezinin normal lokalizasyonunda izotop tutulumu olmayıp, dil tabanında olduğu görüldü. Bu bulgularla lingual tiroid tanısı konuldu. Hastada tek fonksiyonel tiroid dokusu bu olduğu için cerrahi eksizyon yerine medikal tedavi uygun görüldü. Bu yazımızda hipotiroidizm bulguları olan lingual tiroidli olguyu laboratuvar ve görüntüleme bulguları ile sunmayı amaçladık.

Anahtar kelimeler: Lingual tiroid, primer hipotiroidizm, tiroid sintigrafisi

INTRODUCTION

Thyroid gland develops from a medial embryonic thyroid primordium and a smaller pair of lateral embryonic thyroid primordia. Lateral thyroid primordia do not contribute to the development of the thyroid follicles but give rise to calcitonin producing parafollicular C cells. Medial primordium develops as an endodermal diverticulum in the ventral midline of the pharynx between the primary and secondary pharyngeal pouches and become distinguishable by the end of the third week of pregnancy. This diverticulum lobulates, constitutes two lobules and these two lobules connect to each other through a tube called thyroglossal duct at the point where they arise from pharynx. The pharyngeal connection of the thyroglossal duct is in the ventral pharynx where the tongue formation occurs. Thyroid primordium follows the route of the thyroglossal duct and passes anterior to hyoid bone to descend its normal location. By the end of the seventh week thyroid takes a concentric shape and locates in front of the developing trachea. During this period of time the lumen of the duct closes off and becomes a stalk-like attachment and the pharyngeal attachment point of the duct appears as foramen caecum at the tip of sulcus terminalis on the dorsum tongue (1,2). The abnormalities related to the descent from the foramen caecum are responsible for the ectopic thyroid tissue. This rare embryologic pathology is the lingual thyroid formed in foramen in caecum (3).

In 75 % of the cases of lingual thyroid, the only functioning thyroid tissue in the body is found as this ectopic tissue (4). The first known case of lingual thyroid was presented by Hickmann in 1869 (5). The prevalence of lingual thyroid was reported as 1 per 100 000 people (6). Autopsy studies have determined high prevalence since most of the cases remain asymptomatic (7). Lingual thyroid may enlarge and becomes symptomatic in certain conditions such as pregnancy, thyrotoxicosis and neoplasia (8). We aimed at presenting this rare case of symptomatic lingual thyroid along with radiological and scintigraphic findings and a literature review.

CASE REPORT

When a 19 years old woman was admitted to the otorhinolaryngology outpatient clinic. With the complaints of gradually increasing sensation of a catch in her throat when swallowing, foreign-body-feeling in her throat, swelling at the back of the tongue, deepening of her voice and constipation, hypothyroidism was suspected the patient was referred to the endocrinology outpatient clinic. In her assessment in our outpatient clinic, the patient particularly complained of recently developed constipation, deepening of her voice and gradually increasing sensation of a catch in her throat.

A short stature attracted attention in her physical examination (Height: 152 cm, body weight 69 kg. Body mass index: 29.4). In the examination for the short stature, the height was found below the 25th percentile and -2SD for sex and age. A mental and motor retardation attracted attention. The thyroid gland could not be palpated during the thyroid gland examination. Other systems examination revealed no abnormality and the patient's skin was pale and dry. No hair loss was detected. A spherical, red lesion with regular contour, 2x2x1 cm in dimensions was observed when a more detailed endoscopic examination of the mass was performed (Figure 1-2).

In the laboratory examinations hemoglobin level was found as 12.1 mg/dl, Hematocrit was found as 35.2%, the PLT count was 197 000/ul, serum levels of FT3 was 0.61 pg/mL (N:2,1-5.2). FT4 was 0.15 ng/dl (0,8-1,76), and TSH was >150 mIU/L (0,3-4,9). Both anti-thyroid peroxidase antibody which is one of the thyroid antibodies [16IU/ml, (N: 0-35)] and antithyroglobin antibody [<20 IU/ml (N:0-40)] were found negative and these results were consistent with primary hypothyroidism. Consequently, the ultrasound examination of the thyroid gland was performed. The echo pattern of thyroid parenchyma was not identified at the thyroid location (Figure3).

Ectopic thyroid gland was considered and a thyroid scintigraphy with Tc99m was carried out. Thyroid scintigraphy with Tc99m revealed a homogenous focal activity uptake in a region consistent with tongue base. No activity uptake consistent with thyroid tissue was observed in the thyroid lodge (Figure4). A diagnosis of lingual thyroid was made according to these findings that suggested a hypofunctioning ectopic thyroid tissue at sublingual location. Surgical excision of the ectopic thyroid was not recommended to the patient since although hypofunctioning, it was the only functioning thyroid tissue in the body and it was giving rise to mild symptoms. A thyroid hormone suppression therapy was started to the patient in order

to reduce the mass causing a foreign body sensation in her throat and symptomatic hypothyroidism and outpatient follow up was recommended to the patient.

DISCUSSION

The thyroid gland is the first endocrine gland to develop during the embryonic period (9). Embryonic origin of the thyroid gland is the foramen cecum located at the junction of 2/3 anterior and 1/3 posterior parts of the tongue. During the embryonic development, the thyroid tissue descends to its normal pretracheal location passing through the thyroglossal duct (10). As in our case, the most common ectopic thyroid tissue location is the tongue root just behind the foramen cecum, accounting for 90% of the cases of ectopic thyroid (11). Our other two adolescent cases of lingual thyroid were previously described by Arikan et al (12,13). The first prominent feature that attracted attention in these two cases was the retardation of the development. However, cases of ectopic thyroid with mediastinal, cardiac, esophageal or diaphragmatic locations were also reported (14). Ectopic thyroid tissue may be either functioning or non-functioning and is 7 times more common in females than in males (11).

Ectopic thyroid attracts attention during periods when the increased amount of hormone is required, such as in puberty, pregnancy, menopause (8). Relative to insufficient levels of thyroid hormone lead to increased TSH secretion during these periods. This situation consequently results in the enlargement of the ectopic thyroid gland and may render it symptomatic (15).

Ectopic lingual thyroid may cause symptoms such as foreign body sensation in throat, dysphagia, dysphonia and dyspnea, it may mimic a lingual tumor or may be asymptomatic (16). In our case, swallowing difficulty, foreign body sensation in the throat and deepening of the voice were present for a long period of time. Hypothyroidism has been reported in 33% of the cases of lingual thyroid (17). The hormone levels of our case were also consistent with hypothyroidism.

Lingual thyroid appears as a mass with regular contour and homogeneous structure and is more echogenic compared to the tongue muscles in ultrasound examination. When the ectopic thyroid tissue enlarges or a nodule, cyst or tumor develops within the ectopic thyroid tissue, the homogeneous echogenic structure disappears and findings related to these lesions are detected (18). Due to the different signal characteristics of the colloidal material, the lingual thyroid is observed as a nonhomogeneous structure in Magnetic Resonance Imaging examination. If there is no enlargement of the tissue, nodule or tumor formation within the ectopic thyroid tissue.

it is observed as isointense or mildly hyperintense in comparison to tongue muscles on T1 weighted sequences and hyperintense in comparison to the tongue muscles on T2 weighted sequences (19). Scintigraphic examination reveals similar activities in ectopic thyroid tissue and thyroid tissue at the normal location (19). Mostly no activity is detected at the normal location. In our case, while no activity was detected at the normal pretracheal location, thyroid activity was demonstrated at the tongue root.

The determination of the treatment to cases diagnosed with lingual thyroid depends on several factors such as the dimensions of the mass, the presence of local symptoms, the age of the patient, the state of the thyroid gland, ulceration, hemorrhage, cystic degeneration or malignancy. Suppression therapy may provide prominent regression if the clinical symptoms are not present. The indications for surgical excision generally include severe or recurrent hemorrhage, gland enlargement causing dysphagia, prominent airway stenosis, suspicion of malignancy and dysphonia (20). Radioiodine therapy is reserved for elderly patients who are not suitable for surgery and it is contraindicated in younger patients since it may cause damage to gonads and other organs (21). Surgical removal of lingual thyroid may be performed through transoral, transhyoid routes or through a lateral pharyngotomy approach using external neck incision. Transoral approach is appropriate for smaller lesions. The disadvantages of the transoral approach include limited vision, not always being able to perform a complete bleeding control in spite of the possibility of a life-threatening hemorrhage. However, unlike the aforementioned approaches, the lower risks of damage to vital structures in the neck, fistula formation, deep neck infection and less apparent scar are included to the advantages of this technique (2).

Thyroid hormone suppression therapy was found appropriate in our case since she was young, the only functioning thyroid tissue in the body was the lingual thyroid tissue and other surgical indications were absent. Although lingual thyroid is not a common lesion, we should be kept in mind when a mass at the tongue root is detected in the midline in particular and surgical intervention for the undiagnosed lesions may result in the removal of the only functioning thyroid tissue in the body. We should be used as a first step examination tool and scintigraphic examination should be used to confirm the diagnosis.

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