Lingual Thyroid, A Rare Embryological Aberration of Thyroid Gland and Primary Hypothyroidism

Lingual Tiroid, Tiroid Bezinin Nadir Bir Embriyolojik Atipik Yerleşimi ve Primer Hipotiroidizm

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Abstract
Lingual thyroid is an abnormal formation appearing as the result of a deficient descent during embryological development of the thyroid gland through the thyroglossal duct to its normal pretracheal location, and it is a rare embryological aberration. A 32yr old woman was admitted to endocrinology clinic for hypothyroidism with foreign-body-feeling, swallowing difficulties, dyspnea, hoarseness, constipation, cold intolerance, dry skin, fatigue and lethargy. Oropharyngeal examination showed a spherical, fixed, red fleshy and hard basilingual tumor. The thyroid gland was nonpalpable and patient was diagnosed as primary hypothyroidism. Neck ultrasound showed a heterogeneous, smooth contoured, hypoechogenic solid nodular mass on the base of the tongue. Computerized tomography also confirmed this finding. Thyroid scan with technetium 99m revealed isotope uptake in the base of tongue area and no uptake in the normal thyroid location. With these findings lingual thyroid was diagnosed, and the lingual thyroid mass was resected. Biopsy revealed thyroid tissue with nodular colloidal goiter containing hyperplastic nodule. Levothyroxine treatment was started and there was no complication in the postoperative period. We present here a case of lingual thyroid and hypothyroidism in a 32 year old female patient Turk Jem 2007; 11: 98-100

Key words: Lingual thyroid, hypothyroidism

Özet

Anatol kelimeler: Lingual tiroid; hipotiroidizm

Introduction
Late in the first month of life the anlage of the thyroid gland descends from the posterior dorsal midline of the tongue to its final position in the lower neck. If the embryonic gland does not descend normally, ectopic or residual thyroid tissue may be found between the foramen caecum and the epiglottis (1). It is an ectopic thyroid tissue and most commonly presents itself as a lingual thyroid (2). Lingual thyroid is a rare embryological aberration with incidence of 1:100,000. The lesion consists of a tumor mass of thyroid tissue located at the base of the tongue, in the region of the foramen caecum linguæ (lingual thyroid). In most cases, it is diagnosed in childhood and young adulthood but frequently around menopause. It appears as a mass on the base of the tongue
causing mostly local symptoms often with hypothyroidism, rarely with thrive and mental retardation (1). The majority of patients with thyroid ectopy are asymptomatic, but obstructive symptoms as well as hypothyroidism have been observed up to 70% of patients with lingual thyroid (3). The pathologic findings of lingual thyroid tissue are similar to that of cervical thyroid tissue, including goiter formation. The diagnosis was based on the clinical features, fine needle aspiration biopsy, laboratory tests and radiographic imaging studies (4). Treatment includes surgical removal, transplantation, I-131, and thyroid replacement (2). Lingual thyroid identification is of great significance, since it may constitute the only functional thyroid tissue in the body.

Case

A thirty-two-year-old woman was referred for hypothyroidism to department of endocrinology. She had a three-year history of onset of foreign-body-feeling, swallowing difficulties, dyspnea, articulation disorders, hoarseness, constipation, cold intolerance, dry skin, fatigue, lethargy, headache, anorexia, thinning of hair and muscle cramps. Physical findings revealed bradycardia, diastolic hypertension, slightly diminished cooperation, orientation and decreased attention. She was hypotonic and apathetic, and delayed return of deep tendon reflexes is noted. The thyroid gland was nonpalpable. Oropharyngeal examination with inspection showed a spherical, fixed, red fleshy and hard basilingual tumor approximately 5x4x5 cm in diameter. The surface of the lesion is usually smooth and vascularity can be seen. The patient was diagnosed as primary hypothyroidism with these results (total T3: 1,71 ng/ml (N: 0,846-2,02); total T4: 7,22 ug/dl (N: 5,13-14,06); free T3: 0,43 ng/dl (N: 0,182-0,462); free T4:0,829 ng/dl(N: 0,932-1,710); TSH:12,62uIU/ml (N:0,270-4,2)]. Antithyroid antibodies were negative. Neck ultrasound showed a 50x40 mm, heterogeneous, smooth contoured, hypoechoic solid nodular mass containing cystic and calcific areas on the base of the tongue. Computerized tomography also confirmed this finding (Figure 1). Technetium 99m thyroid scan revealed isotope uptake in the base of tongue area and no uptake in the normal thyroid location (Figure 2). With these findings lingual thyroid was diagnosed, and the lingual thyroid mass was resected. Biopsy revealed thyroid tissue with nodular colloidal goiter containing hyperplastic nodule (Figure 3). Levothyroxine treatment was started and there was no any complication in the postoperative period.

Discussion

Lingual thyroid is a rare embryological aberration with incidence of 1:100,000. More than 400 cases of lingual thyroid have been documented in the literature to date. The development of thyroid tissue can occur in any moment of the migration of the thyroid along the thyroglossal duct from the tongue, resulting in lingual (at tongue base), sublingual (below the tongue), prelaryngeal (in front of the larynx), and substernal (in the mediastinum) ectopy. Thyroglossal duct cyst is the most common type of clinical abnormality related to thyroid ectopy (5). It most commonly presents itself as a lingual thyroid (2). It is 7 times higher in females (6). The lesion consists of a tumor mass of thyroid tissue located at the base of the tongue, in the region of the foramen caecum linguae. The size can vary from a few millimeters to several centimeters in diameter. Lingual

![Figure 1. Thyroid CT Scan showing hypoechoic solid nodular mass containing cystic and calcific areas on the base of the tongue (arrows)](image1)

![Figure 2. Technetium 99m scan showing isotope uptake in the tongue region and absence of isotope uptake in the normal thyroid position. Radionuclide scan, lateral and anterior view of the neck. The activity is in a lingual thyroid (LT: Lingual Thyroid, Parotis Gland, Submandibular Gland)](image2)

![Figure 3. Resected ectopic thyroid biopsy revealed thyroid tissue with nodular colloidal goiter containing hyperplastic nodule (arrows. H&E, original magnification XB2)](image3)
thyroid has been identified in 10% of the tongues examined in some autopsy series (7). The majority of patients with thyroid ectopy are asymptomatic, but obstructive symptoms as well as hypothyroidism have been observed. Such ectopic thyroid glands are probably quantitatively deficient and thyroid function may be low or at a low normal level. Symptoms, if where are any: foreign-body-feeling, swallowing difficulties, dyspnea, articulation disorders, bleeding and hypothyroidism, but in many cases the diagnosis is accidental (7). Up to 70% of patients with lingual thyroid have hypothyroidism and 10% suffer from cretinism (1). It may cause differential diagnostic problems with real malignant tumor. We described a young woman case of lingual thyroid with hypothyroidism in adulthood (3). The diagnosis was based on the clinical features, biopsy, laboratory tests and radiographic imaging studies (4). Thyroid adenoma, goiter, hyperplasia, inflammation, and carcinoma occur in lingual thyroids. In our case, resected ectopic thyroid biopsy revealed thyroid tissue with nodular colloidal goiter containing hyperplastic nodule and CT Scan was confirming this (1). Our aim in presenting this case is distinguishing the lingual thyroid from the masses on the midline of the base of the tongue when the lingual thyroid gland was describeds. The thyroid gland should be researched, on the neck, its localication. Thyroid function tests must be studied. Then, if patient was found as hypothyroid, thyroid hormone should be replacement and elective operation must be planned. Ectopic thyroid mass was resected with transcervical-suprathyroid approach in our patient, and postoperatively, thyroid hormone replacement was started to prevent possible re-enlargement of the lingual thyroid and there was no complication in the postoperative period. We aimed to review the literature about the lingual thyroid and hypothyroidism and to report a case of lingual thyroid in a adult female patient with hypothyroidism.

References