Ectopic Thyroid Tissue in Submandibular Region:
A Case Report

Abstract
Submandibular ectopic thyroid tissue with a coexisting normally located thyroid gland is extremely rare. We present a case of a 31 years old woman. At the age of 27, she has been operated for multinodular goiter and the operation material revealed normal thyroid tissue histologically. Three years after the operation the patient complained about swelling in the submandibular region. Ultrasonographic examination showed normally located residual thyroid tissue and 12x8x6 mm-mass in submandibular space. Tc-99m pertechnetate thyroid scan revealed increased uptake at the same location. The patient referred to excisional biopsy. Normal thyroid follicules and no evidence of malignancy were found in specimen pathologically.

Key Words: Submandibular ectopic thyroid tissues, Tc-99m pertechnetate, thyroid scan

Özet

Anahtar Kelimeler: Submandibular ektopik tiroid dokusu, Tc-99m perteknetat, tiroid sintigrafisi

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t the end of the third embryonal week, the anlage of thyroid gland is formed as an endodermal diverticulum in the median plane at the base of the pharyngeal gut. As the embryo elongates this diverticulum migrates anteriorly and inferiorly to the hyoid bone and laryngeal cartilage, tethered by a slender thyroglossal duct. Thyroglossal duct normally breaks down by the end of the 5th week and the thyroid gland continues its downward migration. By the 7th week of embryonic development, thyroid diverticulum has assumed its adult position. This diverticulum forms a bilobed gland, with the two lobes connected by a small isthmus. Often small amount of thyroid tissue expends superiorly from the isthmus and is known as pyramidal lobe. Aberrant thyroid tissue may be found anywhere along its migratory pathway.

The most frequent location of ectopic thyroid gland is lingual thyroid. Ectopic thyroid may also be found in the larynx or trachea, in pericardium or even in the heart. Lateral aberrant thyroid may be seen as part of enormous development. Various extremely rare sites of ectopic thyroid gland have been reported, including submandibular location. It may then have the same presentation as any other neoplasm of the submandibular space.
Case Report

A 31 year-old woman who had painless swelling in the left submandibular region was referred to our department with the suspicion of submandibular lymphadenopathy. In her clinical story she has been operated for multinodulary goiter at the age of 27 and the operation material revealed normal thyroid tissue histologically. Since then she hadn’t been regularly checked for thyroid hormone levels and was not put on thyroid hormone replacement therapy. Three years after the operation the patient complained swelling in the submandibular region. A 2x1.5 cm mass of an elastic consistency, non-painful on palpation, was noted in the left submandibular region.

The laboratory parameters for thyroid gland indicated hypothyroidism (TSH: 20.4 uIU/ml, range: 0.17-4.05 uIU/ml). Doppler ultrasonography showed normally located residual thyroid tissue (left lobe 11x7x6 mm, right lobe 10x6x7 mm) and 12x9x6 mm-mass between submandibular gland and left upper pole of thyroid cartilage (Figure 1). This lesion had similar parenchymal and degree of vascularity with the residual tissues in thyroid region sonographically. Tc-99m pertechnetate thyroid scan revealed increase uptake in this mass and bilateral residual thyroid tissue in normal location (Figure 2). According to these findings the patient referred to excisional biopsy. Histological examination of this lesion confirmed normal thyroid follicles and no evidence of malignancy were found in the specimen (Figure 3). Postoperative evaluation with ultrasonography showed residual tissue related to submandibular ectopic thyroid tissue. Thyroid function tests indicated hypothyroidism (TSH: 35 uIU/ml) so L-thyroxine therapy has been initiated and patient followed clinically. At the 3rd month of follow up, thyroid function tests were normalized.

Discussion

Any thyroid tissue not located anterior to the upper trachea in the anterior midline of the neck is considered ectopic. Ectopic thyroid may be located anywhere along the thyroglossal duct tract between the foramen cecum and normal location. Intratracheal, eosephageal and even duodenal ectopias have also been described.1 Ectopic submandibular location of thyroid tissue is documented in 16 reports in the literature.2,3,5 Coexistence of normally located thyroid gland with ectopic submandibular
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Thyroid tissue is extremely rare. In our case there had been normally functioned thyroid coexistence with the submandibular ectopic thyroid tissue. However, it is thought that TSH stimulation after subtotal thyroidectomy for multinodular goitre led to stimulation of the ectopic thyroid tissue in the submandibular region.

The most accepted hypothesis for ectopic lateral thyroid in the absence of normally located thyroid gland might represent dysembrioplasia of the lateral anlage, with nonmigration and agenesis of the medial anlage. Nicastri et al reported that benign thyroid tissue may also metastasize to the cervical lymph nodes. But in our case there was no lymphatic structure in the pathologic specimen.

The therapy of ectopic thyroid tissue is surgical removal. However preoperative diagnosis of a tumor mass in the neck region is important in clinical management. Histological examination of the tissue is required because clinical and laboratory findings may not safely exclude the presence of a malign tumor. On that account, standard management (excisional biopsy) of the neck mass can lead to iatrogenic hypothyroidism because in the most cases it is the only functional thyroid tissue in the body. Before the complete removal of ectopic thyroid, it is necessary to ensure that normal, functional thyroid gland is present by thyroid scan. Additionally, post operative thyroid function tests should be performed and patient followed clinically.

REFERENCES