



Submandibular ectopic thyroid with normally located thyroid gland

Normal yerleşimli tiroid bezi ile birlikte görülen submandibüler ektoptik tiroid

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Ectopic thyroid is a rare developmental anomaly of the thyroid gland which is defined as the presence of thyroid tissue at a site other than the pretracheal area. Nearly 1 to 3% of all ectopic thyroids are located in the lateral neck. Simultaneous submandibular ectopic thyroid tissue presenting with a functional orthotopic thyroid gland is extremely rare. In this article, we report a 37-year-old female case admitted to our clinic with a complaint of swollen neck in whom ultrasonography revealed submandibular ectopic thyroid tissue presenting with an orthotopic thyroid gland.

Key Words: Ectopic; submandibular; thyroid gland.

Ektoptik tiroid, pretrakeal alan dışındaki bir bölgede tiroid dokusu varlığı olarak tanımlanan tiroid bezinin nadir görülen gelişimsel anomalisidir. Tüm ektoptik tiroid olgularının yaklaşık %1 ila %3'ü yan boyunda bulunmaktadır. Fonksiyonel bir ortotopik tiroid bezi ile eş zamanlı submandibüler ektoptik tiroid dokusu birlikteliği son derece nadirdir. Bu yazıda, kliniğimize boyunda şişlik yakınması ile başvuran ve ultrasonografisinde submandibüler ektoptik tiroid dokusu ve ortotopik tiroid bezi birlikteliği tespit edilen 37 yaşında bir kadın olgu sunuldu.

Anahtar Sözcükler: Ektoptik; submandibüler; tiroid bezi.

Ectopic thyroid is an uncommon developmental anomaly of the thyroid gland, defined as the presence of thyroid tissue at a site other than the pretracheal area.^[1] In most cases, ectopic thyroid is located along the embryologic descent path of migration as either a lingual thyroid or a thyroglossal duct cyst. Lingual thyroid is the most common presentation of ectopic thyroid.^[2] The presence of ectopic thyroid tissue in the submandibular region is extremely rare.

In 70% of cases of ectopic thyroid, the normal thyroid gland is absent and this ectopic gland is the only functional thyroid tissue.^[3] Simultaneously having submandibular ectopic thyroid tissue and a functional orthotopic thyroid gland is an exceptionally rare event. To our knowledge, only seven cases of simultaneous submandibular ectopic thyroid tissue and a functional orthotopic thyroid gland have been reported in the literature (Table 1).^[4-10] In this article, we report a case of a



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Table 1. Previously reported cases of simultaneous finding of submandibular ectopic thyroid and a functional normally located thyroid gland

Authors	Gender	Histological examination	Treatment
Aköz et al. ^[4]	M	Thyroid tissue and colloidal cyst	Surgical excision
Feller et al. ^[5]	F	Normal thyroid follicles	Surgical excision
Mace et al. ^[6]	F	Thyroid follicular tissue and partly cystic and hemorrhagic degeneration	Surgical excision
Kanaya et al. ^[7]	F	Thyroid tissue with partial adenomatous goiter	Surgical excision
Huang et al. ^[8]	F	Thyroid tissue with goitrous change	Surgical excision
Piantanida et al. ^[9]	F	Thyroid follicular cells with no malignant features	Surgical excision
Amoodi et al. ^[10]	F	Thyroid goiter	Surgical excision

woman presenting with submandibular ectopic thyroid tissue and an orthotopic thyroid gland.

CASE REPORT

A 37-year-old woman was admitted to our outpatient clinic with a cervical mass existing for three years. On physical examination, a painless, uniformly bounded, mobile, soft, nearly 3x4 cm mass was observed in the right submandibular region. A detailed systemic examination did not reveal any abnormality. Ultrasonography (USG) of the neck demonstrated a solid mass of 34x36x26 mm, including cystic areas and showing significant blood flow, localized in the right submandibular region. Her thyroid gland was in a normal location and had normal parenchyma. Thyroid function tests confirmed euthyroidism and she had no anti-thyroperoxidase or anti-thyroglobulin antibodies. Ultrasonography-guided fine needle aspiration biopsy (FNAB) was non-diagnostic. Cervical magnetic resonance imaging (MRI) revealed a 35x41x26 mm lobulated mass showing contrast agent involvement in the right submandibular region and a normal orthotopic thyroid gland (Figure 1). The patient underwent excision of the submandibular mass under general anesthesia (Figure 2). The histopathological examination of the mass revealed thyroid gland tissue with nodular hyperplasia (Figure 3). She was symptom free and euthyroid over the next year and there was no evidence of recurrence.

DISCUSSION

Ectopic thyroid tissue is defined as thyroid tissue that is not localized anterolaterally to the second, third or fourth tracheal rings.^[11] This is the most common thyroid dysgenesis, with

a 48-61% incidence. In the normal population, ectopic thyroid tissue is seen at nearly a 1/100,000-300,000 prevalence and the male/female ratio is 1:4. Ectopic thyroid tissue can be seen from the foramen cecum to mediastinum, which is the migration pathway for the thyroid gland in any situation. Nearly 90% of it is localized in the lingual region, which is the most common region.^[12] Approximately 1 to 3% of all ectopic thyroids are located in the lateral neck.^[2]

Around the fourth week of gestation, the thyroid gland descends through the thyroglossal duct as a bilobed diverticulum from an

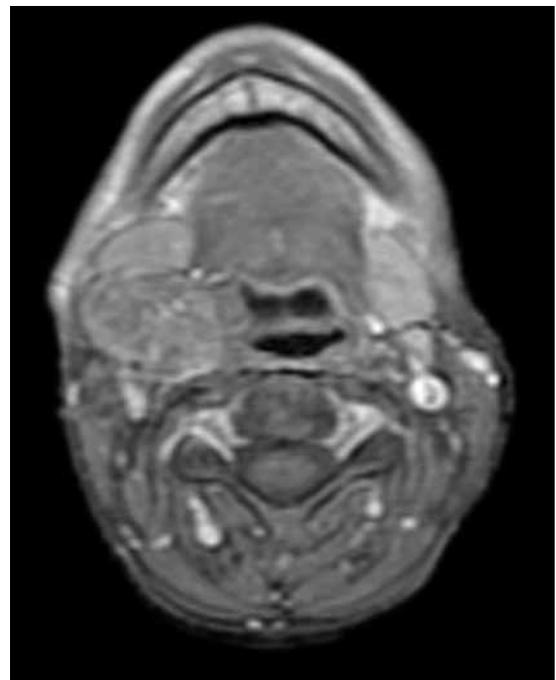


Figure 1. Cervical magnetic resonance imaging shows a lobulated mass in the right submandibular region.

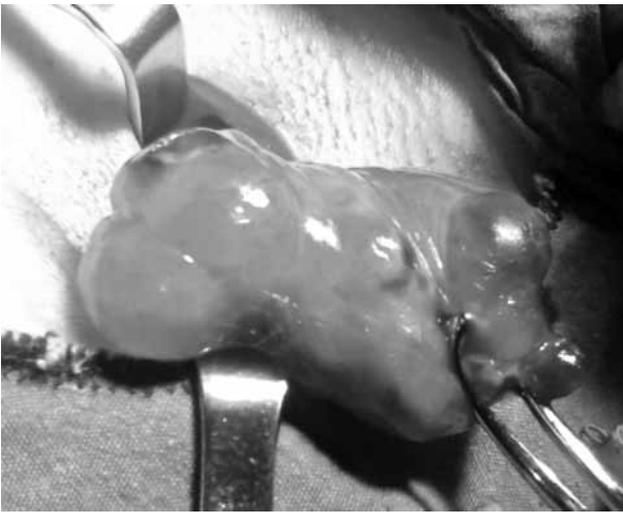


Figure 2. Complete excision of mass.

invagination in the foramen cecum from the base of the tongue to the front of the anterior tracheal wall, where it reaches its final position in the seventh week. Toward the end of the migration, the lateral thyroid primordium dissociates from the caudal base of the fourth pharyngeal pouch and the ultimobranchial body, which contains parafollicular cells, and migrates medially to join the thyroid gland.^[13] A failure to descend or an incomplete migration of the medial anlage of the thyroid during embryologic development leads to midline or near midline ectopias, such as a lingual thyroid or a thyroglossal ectopia.^[13,14] In rare cases, an aberrant migration with cell rests deposited laterally during the development of the gland or a failure of the lateral anlage to fuse with the median anlage can result in the development of true lateral aberrant thyroid tissue. Other possible causes of lateral ectopic thyroid include metastasis of a thyroid carcinoma (lateral aberrant thyroid) or implantation of thyroid tissue during surgery of a normal localized thyroid gland.^[2,13]

In cases of lateral cervical ectopic thyroid, the simultaneous occurrence of orthotopic thyroid is extremely rare. Four of seven reported cases who had submandibular ectopic thyroid and a functional orthotopic thyroid revealed nodular goiter.^[7-10] Furthermore, two of these four cases had a history of previous thyroidectomy for nodular goiter. It may be that implantation of thyroid tissue during previous surgery of an orthotopic thyroid gland is the possible cause of these cases. But in our case there was no

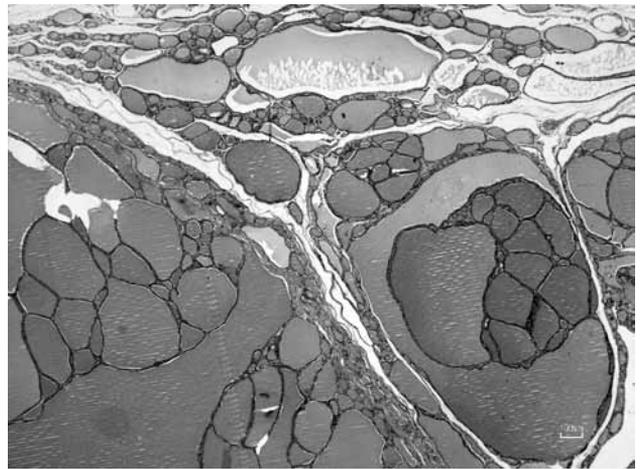


Figure 3. Histopathological examination shows normal thyroid follicles lined by a single layer of flat cuboidal follicular epithelial cells (H-E x 200).

history of previous surgery or trauma in the neck region, and she was euthyroid and had a normal orthotopic thyroid gland.

Since only the ectopic tissue is functional in up to 70% of cases, there are diagnostic and therapeutic difficulties when submandibular ectopic thyroid occurs. To choose the best treatment in these cases, identification of the presence or absence of eutopic thyroid should be done.^[15,16] Otherwise, an inaccurate preoperative diagnosis can result in hypothyroidism. Submandibular ectopic thyroid should first be differentiated from metastatic thyroid cancer. Submandibular tumors such as pleomorphic adenoma or carcinoma, inflammatory lesions, branchial cleft cysts, lymphangiomas, carotid body tumors, and lymphadenopathy of various etiologies should be considered also. With clinical suspicion of submandibular ectopic thyroid, the diagnostic workup should include USG and either computed tomography or MRI, scintigraphy with either technetium-99m or iodine-131, FNAB and thyroid function testing.^[2,16,17] We managed our case as a submandibular mass of unknown etiology. Since orthotopic thyroid gland was detected in radiological evaluation, we did not think this mass was an ectopic thyroid tissue. Therefore, we did not perform scintigraphy with our patient. Prior to surgery, FNAB was performed but its result was non-diagnostic. Hence we decided to excise the mass for diagnosis. Most of the reported cases of lateral ectopic thyroid tissue were diagnosed after surgical excision

for an enlarging mass of unknown etiology, as in our case. The indications for surgery for an ectopic thyroid tissue include risk of malignancy, presence of other thyroid tissue, symptomatic patient and cosmetic deformity.

Conclusion

Ectopic submandibular thyroid tissue is an extremely rare event that poses both diagnostic and management problems. However, physicians should be aware of the possibility that a submandibular mass could be ectopic thyroid tissue. Prior to surgical excision of the ectopic thyroid, it is necessary to ensure that normal, functioning thyroid tissue is present elsewhere.

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