

## Ectopic Thyroid Tissue in Submandibular and Infrahyoid Region

### *Submandibular ve Infrahyoid Bölgede Lokalize Ektopik Tiroid Dokusu*

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#### Abstract

The thyroid is the first endocrine gland to form during embryogenesis. At this stage, incomplete or anomalous migration of thyroid tissue causes ectopic localization of the gland. Submandibular ectopic thyroid tissue with a coexisting normally located thyroid gland is extremely rare. In this case aimed to present the findings of the 65-years-old female patient who is bilateral subtotal thyroidectomy operation performed for multinodular goiter of 12 years ago. Case, painless mass in the right submandibular and infrahyoid region for 6 months was admitted to our clinic with complaints. Result of contrast-enhanced neck computed tomography, ultrasound-guided fine-needle aspiration biopsy and thyroid scintigraphy were found of functional residual thyroid tissue in the normal localization as well as 2x3 cm mass in the submandibular area and 1x2 cm mass lesion in the infrahyoid region. The patient referred to excisional biopsy. Normal thyroid follicles and no evidence of malignancy were found in specimen pathologically. Postoperative follow-up of thyroid function tests were normal.

**Key Words:** Ectopic Thyroid, submandibular and infrahyoid space, surgery

#### Özet

Tiroid bezi embriyogenez esnasında ilk oluşan endokrin bezdir. Bu gelişim sürecinde bezin migrasyonunu tamamlayamaması ya da anormal bir şekilde tamamlaması bezin ektopik yerleşimine neden olur. Submandibular ektopik tiroid dokusu ve bu olgularda normal lokalizasyondaki tiroid bezinin varlığı çok az rastlanan bir durumdur. Bu olgu sunumunda 12 yıl önce multinodüler guatr nedeniyle bilateral subtotal tiroidektomi operasyonu uygulanan 65 yaşındaki bir kadın olguya ait bulguları sunmayı amaçladık. Olgu 6 aydır sağ submandibular ve infrahyoid bölgede ağrısız kitle yakınmasıyla kliniğimize başvurdu. Kontrastlı boyun tomografisi, tiroid sintigrafisi ve ultrasonografi eşliğinde yapılan ince iğne aspirasyon biopsisi sonucunda normal lokalizasyonda fonksiyonel rezidü tiroid dokusunun yanısıra submandibular alanda 2x3 cm'lik, infrahyoid bölgede 1x2 cm'lik kitlesel lezyon saptandı. Eksizyonel biyopsi sonrası patolojik incelemede normal tiroid follikül hücreleri gözlemlendi ve malignite lehine bulgu saptanmadı. Postoperatif takiplerinde tiroid fonksiyon testleri normal olarak belirlendi.

**Anahtar Kelimeler:** Ektopik tiroid, submandibular ve infrahyoid bölge, cerrahi

#### Introduction

Ectopic thyroid tissue is a rare congenital anomaly and is generally asymptomatic. The thyroid gland begins its embryological development as an epithelial proliferation in the foramen cecum macula [1]. The thyroid gland embryologically consists of a median and a mass of two lateral cells [2-5]. While a major part of the thyroid parenchyma consists of the median part, 1-30 percent of the total thyroid consists of the lateral part [2]. Moving from the top to the bottom, the thyroid reaches its final position in front of the trachea in the 7th week of fetal life [1]. As a result of an arrest in the ectopic position of the thyroid tissue during its downward migration, the process of embryonic development is deficient or it affects midline development [2, 4, 6]. It is stated that lateral cervical ectopic thyroid formation is occurred in the 7<sup>th</sup> week of embryonic life as a result of failure in

formation of lateral buds of 4th branchial pouch [2, 4]. And it is thought that submandibular ectopic thyroid tissue had developed that way [2, 4, 5, 7]. 90 percent of ectopic thyroid is seen as lingual thyroid, whereas 10 percent of it is identified with other anatomic localizations (infrahyoid, submandibular, prelaryngeal, mediastinum, esophagus, heart, diaphragm, parapharyngeal) [8].

In this study, right submandibular and infrahyoid located ectopic thyroid case, seen as a rare incident in a patient, who previously had subtotal thyroidectomy due to nodular goiter, was presented and discussed accompanied by literature.

#### Case Report

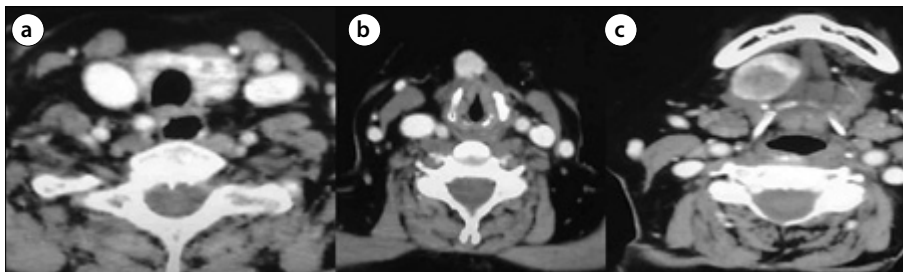
A 65-year-old female patient consulted us with the complaints of a lump, which she had it in upper moiety of right neck and front and upper of the neck for nearly six months.

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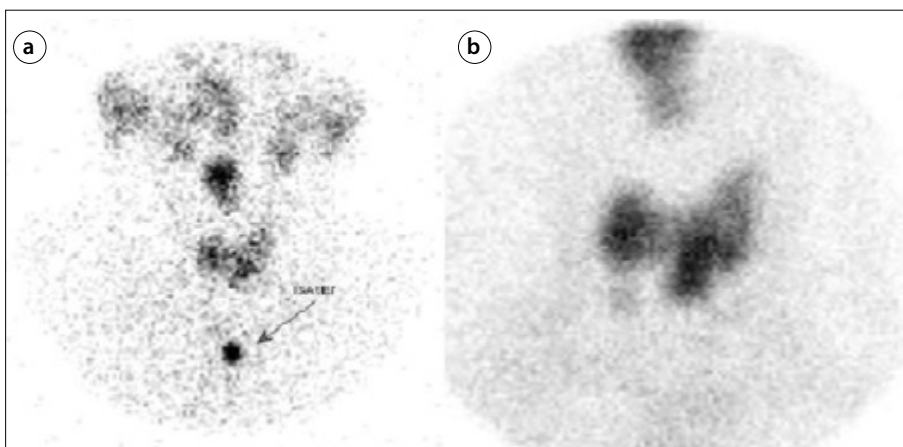
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**Figure 1. a-c.** Contrast-enhanced computed tomography images of the neck, Increase in the left lobe of the thyroid nodular density (a), 17 mm in size with dense nodular contrasting area in the front side of the midline of the neck (b), 30x24 mm in size with dense nodular contrasting area in the right submandibular region was observed (c).



**Figure 2. a, b.** Taken as a result of thyroid scintigraphy with Tc-99m pertechnetate in the thyroid space, midline and submental area is compatible with thyroid tissue activity uptake is observed (a, b).

Patient had a history of subtotal thyroidectomy operation due to nodular goiter 12 years ago and she did not have a history of cigarette smoking or alcohol use.

A 3x2 cm painless lump in right submandibular tract, 2x1 cm painless lump in hyoid bone corpus inferior and almost 5 cm horizontal incision scar in collum anteroinferior and in thyroid localization were determined in the physical examination. The lumps were well-circumscribed, mobile and soft.

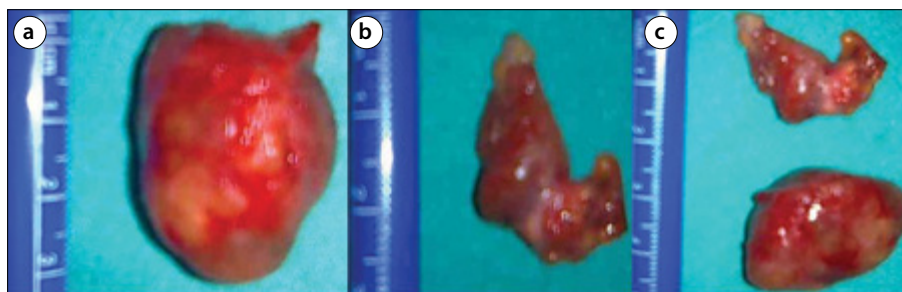
Contrast-enhanced neck tomography, fine-needle aspiration biopsy accompanied with ultrasonography and thyroid scintigraphy were applied to the patient, who was diagnosed with a dermoid cyst or malignancy metastasis at an early stage. 30x24 mm intense contrasted nodular tracts in right submandibular tract by computed tomography and almost 17 mm intense contrasted nodular tracts were monitored in upper front midline, and nodular density rise in the left lobe was monitored (Figure 1). As a result of the fine-needle aspiration biopsy accompanied by ultrasonography, cytology compatible with separate normal thyroid cells were determined in

three pieces taken from the infrahyoid and left thyroid lobe. As a result of thyroid scintigraphy performed by a Tc -99m pertechnetate, activity involvement compatible with thyroid tissue was monitored in thyroid loj, midline and in submental tract (Figure 2).

Preoperative thyroid hormones and biochemical rates were normal. As a result of the reasons such as functional thyroid tissue in thyroid region and malignancy development risk in ectopic thyroid tissue; ectopic thyroid tissues after obtaining consent of the patient were removed totally under general anesthesia with surgical operation by being cut from surrounding tissues.

Pathology results of the patient, who did not have any postoperative complication, was reported as 3x2,5x2 cm in submandibular tract, 2x1x1 cm in infrahyoid tract benign ectopic thyroid tissue (Figure 3).

As postoperative thyroid function test results (FT3) (FT4) were in the normal range in following 6 months, no hormone replacement treatment was started.



**Figure 3. a-c.** Postoperative images from the extracted masses, 3x2,5x2 cm mass extracted from the submandibular region (a), 2x1x1 cm mass extracted from the infrahyoid region (b), With the view of the masses (c).

## Discussion

Thyroid tissue's being in another anatomic localization apart from normal localization in front of trachea was stated as thyroid tissue [2, 4] and this was the most frequent thyroid development disorder (48-61%) [6].

As many patients were asymptomatic, histological ectopic thyroid tissue incidence was determined as 1/100000 in literature and as 10% in cadaver studies as well as unknown real incidence [8].

Ectopic thyroid was generally monitored in midline between foramen caecum and mediastinum [2, 7] and 90% of these cases is seen in the form of lingual thyroid [2, 4, 6]. While lateral located ectopic thyroid was seen rarely [2], submandibular ectopic thyroid was seen extremely rare [2, 4, 5, 7]. We presented a submandibular ectopic thyroid case.

Although lumps seen in submandibular tract were seen in distinctive diagnosis, ectopic thyroid tissue should also be considered [2, 5-7].

Radiological methods; such as ultrasonography, computed tomography, magnetic resonance monitoring and thyroid scintigraphy were used in ectopic thyroid diagnosis [9]. Regional structure with monitoring methods can be evaluated [6] and ectopic thyroid tissue can be determined by hyperdense appearance and contrast involvement [9]. Scintigraphy was helpful in supporting the case and presenting the existence of functional thyroid tissue [9]. And it gave information about the existence of thyroid tissue in normal localization [6].

Lateral cervical ectopic thyroid was seen rarely in normal localization along with thyroid tissue [5]. As only ectopic thyroid tissue was functional in 70% of the cases [7], both diagnosis and treatment became difficult in this case [5]. By performing thyroid function tests and scintigraphy surveys, and as a result of these findings, iatrogenic hypothyroidism risk can be decreased by planned surgery [3, 5, 7].

Ablation treatment and iodine 131 can be performed in the cases in which general anesthesia was risky or surgery

was not preferred. However, the radioactive iodine treatment can be resulted in hypothyroidism and it carries risk in late period connected with radiation [10].

All diseases affecting thyroid tissues in normal position also affected ectopic thyroid tissue [3-5].

Clinical examination, laboratory tests and monitoring methods may not give information the present lump [3]. While ectopic thyroid was diagnosed by performing the fine-needle aspiration biopsy, in some cases, divisions between the benign and malign can be performed only by right histological evaluation [3, 5, 6].

However it was rare, neck lumps suspected to be ectopic thyroid tissue but later became thyroid carcinoma metastases can be confirmed [2-4], and even neoplasm cases, which are primary of ectopic thyroid tissue, can be seen [2, 3].

The treatment of ectopic thyroid was related to some factors, such as greatness of lump, local symptoms, age of the patient, functional position of thyroid gland and related complications (ulceration, hemorrhage, malignancy) [6].

In our case, right submandibular, infrahyoid placement ectopic thyroid tissues and subtotal thyroidectomy secondary residual thyroid tissue previously experienced in normal localization were determined together using contrast neck tomography. It was monitored that the residual thyroid tissue in normal localization by Tc-99m and ectopic thyroid tissue in infrahyoid were functional. As a result of ultrasound-guided fine-needle aspiration biopsy, cytology accompanied by benign thyroid tissue in 3 localizations was determined. By evaluating datum such as age of the patient, malignancy risk, functional thyroid tissue in normal localization, present ectopic thyroid tissues were cut during surgical approach. Both specimens were reported as benign ectopic thyroid tissue according to the pathological results. Postop thyroid function test results were in normal range following 6 months.

Consequently, although ectopic thyroid is seen rarely in distinctive diagnosis of lumps in neck lumps especially submandibular tract, it has to be considered. While monitoring methods are used in ectopic thyroid diagnosis, especially thyroid scintigraphy is certainly necessary when indicating both

diagnosis and functional thyroid tissues. Considering the risk of malignancy metastasis and development of primary malignancy, if there is functional thyroid tissue in normal localizations especially as happened in our case, we think that a surgical intervention is needed.

**Informed Consent:** Written informed consent was obtained from the patient who participated in this case.

**Peer-review:** Externally peer-reviewed.

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

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## References

1. Chawla M, Kumar R, Malhotra A. Dual ectopic thyroid: case series and review of the literature. *Clinical Nuclear Medicine* 2007; 32: 1-5. [\[CrossRef\]](#)
2. Choi JY, Kim JH. A case of an ectopic thyroid gland at the lateral neck masquerading as a metastatic papillary thyroid carcinoma. *J Korean Med Sci* 2008; 23: 548-50. [\[CrossRef\]](#)
3. Fumarola A, Trimboli P, Cavaliere R, et al. Thyroid papillary carcinoma arising in ectopic thyroid tissue within a neck branchial cyst. *World J Surg Oncol* 2006; 4: 24. [\[CrossRef\]](#)
4. Huang TS, Chen HY. Dual thyroid ectopia with a normally located pretracheal thyroid gland: case report and literature review. *Head Neck* 2007; 29: 885-8. [\[CrossRef\]](#)
5. Mace AT, McLaughlin I, Gibson IW, et al. Benign ectopic submandibular thyroid with a normotopic multinodular goitre. *J Laryngol Otol* 2003; 117: 739-40. [\[CrossRef\]](#)
6. Nasiru Akanmu I, Mobolaji Adewale O. Lateral cervical ectopic thyroid masses with eutopic multinodular goiter: an unusual presentation. *Hormones (Athens)* 2009; 8: 150-3. [\[CrossRef\]](#)
7. Babazade F, Mortazavi H, Jalalian H, et al. Thyroid tissue as a submandibular mass: a case report. *J Oral Sci* 2009; 51: 655-7. [\[CrossRef\]](#)
8. Wertz ML. Management of undescend lingual subhyoid thyroid glands. *Laryngoscope* 1974; 84: 507-21. [\[CrossRef\]](#)
9. Erdoğan N, Dirim-Vidinli B, Uluç E, et al. Lingual thyroid multinodular types: Different findings in the two cases. *Ear, Nose and Throat Clinics* 2003; 5: 89-94.
10. Thomas G, Hoilat R, Daniels JS, et al. Ectopic lingual thyroid: A case report. *Int J Oral Maxillofac Surg* 2003; 32: 219-21. [\[CrossRef\]](#)