Case Report / Olgu Sunumu

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Ectopic Thyroid Tissue in the Submandibular Region Presenting with Signs of Thyroiditis

Tiroidit Bulguları ile Prezente Olan Submandibular Yerleşimli Ektopik Tiroid Dokusu

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Abstract / Özet

Thyroid tissue may be localized at various levels in the midline of the neck in the region between the foramen ceacum and the sternal notch. Thyroid tissue can also be found in remote structures that were associated with the thyroid anlage during development. Thyroid tissue is rarely detected in the the lateral neck, such as in the submandibular region. Ectopic thyroid tissue may be subject to the same pathological processes as normal eutopic thyroid tissue, including inflammation, hyperplasia, and tumorigenesis. We present a case of ectopic thyroid tissue located in the left submandibular region in a patient that underwent total thyroidectomy 30 years previously.

Key Words: Ectopic thyroid, neck mass, thyroiditis

Tiroid dokusu boyun orta hatta foramen caecumla sternal çentiğe kadar farklı seviyelerde lokalize ola bilmekte. Tiroid embriyogenez sırasında özofagus, mediasten, kalp, aort, sürrenal, pankreas, mesane ve cilde yerleşerek ektopik hale gelebilmekte. Çok nadir de olsa bez lateral boyun submandibular bölgede gözlene bilmekte. Ektopik tiroid dokusu normal tiroid gibi inflamasyon, hiperplazi ve tümörogenez paterni taşımakta. Klinik şikayetler lokalizasyona bağlı gelişmekte. Bu yüzden bu türden patolojiler submandibular kitlelerin ayırıcı tanısına girmekte. Bu çalışmada 30 sene önce tiroidektomi geçirmiş ve sol submandibular bölgede ektopik tiroid dokusu ile baş vuran hastayı sunmayı planladık.

Anahtar Kelimeler: Ektopik tiroid, boyun kitlesi, tiroidit

Introduction

Thyroid tissue may be localized at all midline neck levels between the foramen ceacum and the sternal notch (1). Thyroid tissue can also be found in remote structures that were associated with the thyroid anlage during development, including the esophagus, mediastinum, heart, aorta, adrenal gland, pancreas, bladder and skin. The localization of thyroid tissue in the lateral neck, such as in the submandibular region, is extremely rare (2). Ectopic thyroid tissue may be subject to inflammation, hyperplasia, and tumorigenesis like normal thyroid tissue (2, 3). It is important to consider this rare condition within the differential diagnosis of a neck mass in the submandibular region.

We present a case of ectopic thyroid tissue in the left submandibular region in a patient that underwent total thyroidectomy 30 years previously.

Case Report

A 60 year-old woman presented with a mass that seldomly caused pain in the left lateral neck region. The patient underwent total thyroidectomy 30 years previously with no subsequent medical treatment. On routine laboratory analysis she had always been euthyroid, despite the detection of high anti-thyroid peroxidase antibodies (anti-TPO) and anti-thyroglobulin (anti-TG) levels. Physical examination revealed a visually apparent, solid, firm, mobile and mildly painful mass of 4x3 cm in size in the left submandibular region on palpation. The mass size was 38x45mm and heterogenic as assessed by ultrasonography. There was no lymphadenopathy in the neck on palpation, which was confirmed by neck ultrasonography. Fine needle aspiration biopsy (FNAB) was performed and reported as suspicous with Hurthle cell metaplasia. The suprasternal region was free of Technetium-99m pertechnetate uptake, apart from the left submandibular mass assessed by the thyroid scan (Figure 1).

The patient was scheduled for excision of the submandibular mass by left-sided neck dissection, and the biopsy was frozen perioperatively. Surgery was conducted under general anesthesia in order to identify and remove the mass. Following a 6 cm neck crease parallel incision over the mass and dissection of the surrounding structures, the mass was identified and removed. The result from the frozen biopsy was reported as a benign-like lesion consisting of thyroid epithelial cells in the background of Hashimoto thyroiditis. The operation was completed with the total excision of the mass. The final histopathology was concordant with the result from the frozen biopsy and was reported as a benign-like lesion consisting of thyroid epithelial cells

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in the background of Hashimoto thyroiditis with sparse Hurthle cell metaplasia (Figure 2). In the first month of post-operative follow up, an increase in Thyroid-stimulating hormone (TSH) with an accompanying decrease in autoantibody levels was noticed. The patient is receiving thyroid hormone replacement therapy 12 months post-operatively.

Discussion

The thyroid gland is the first endocrine gland to develop during the embryonic period and it begins to form approximately 24 days after fertilization from an outgrowth of the pharyngeal endoderm. As the embryo grows, the thyroid gland descends into the neck. For a short time, the gland is connected to the developing tongue by a narrow tube, the thyroglossal duct. At approximately 7 weeks, the gland assumes its definitive shape and reaches its final destination in the neck. By this time, the thyroglossal duct has normally disappeared, although its remnants persist as a small pit, the foramen caecum. Failure of normal descent of the thyroid gland results in the development of ectopic thyroid tissue.

Ectopic thyroid tissue is relatively rare. In the majority of cases, it occurs along the line of descent of the thyroid gland, most commonly in the midline. The prevalence of ectopic thyroid tissue ranges between 7 and 10%. Lingual thyroid tissue is the most common ectopic thyroid tissue site, accounting for 90% of all cases with a prevalence of between 1:100000 and 1:300000 and a clinical incidence of between 1:4000 and 1:10000 (4). Other sites of ectopic thyroid tissue are suprahyoid and infrahyoid, lateral aberrant thyroid, substernal goiters, struma ovarii, and struma cordis. Ectopic thyroid tisue has also been found in the larynx, trachea, esophagus, pericardium, diaphragm, and branchial cysts. Rare cases of ectopic thyroid tissue have been described in the parathyroid, cervical lymph nodes, submandibular gland, duodenal mesentery, adrenals, and the carotid bifurcation. Ectopic thyroid tissue occurs more commonly in females and is usually seen during adolescence and pregnancy when the demand for thyroid hormone increases (3). The actual incidence of ectopic thyroid tissue may be higher than reported, as the diagnosis is made only in the presence of corresponding symptoms or signs. Ectopic thyroids are usually functional, and may become clinically evident with the development of goitres, biochemical hyperthyroidism or malignancy (5).

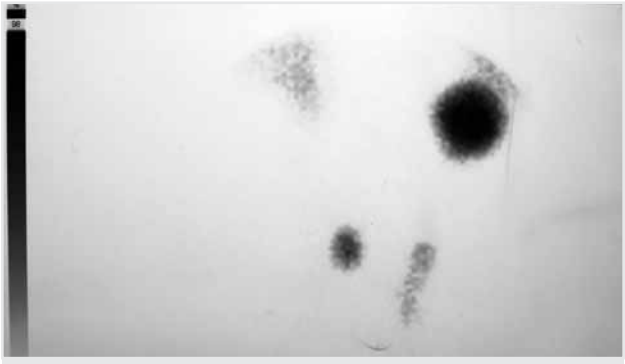

Figure 1. A thyroid scan showing the absence of Technetium-99m pertechnetate uptake in the suprasternal region, apart from in the left submandibular mass

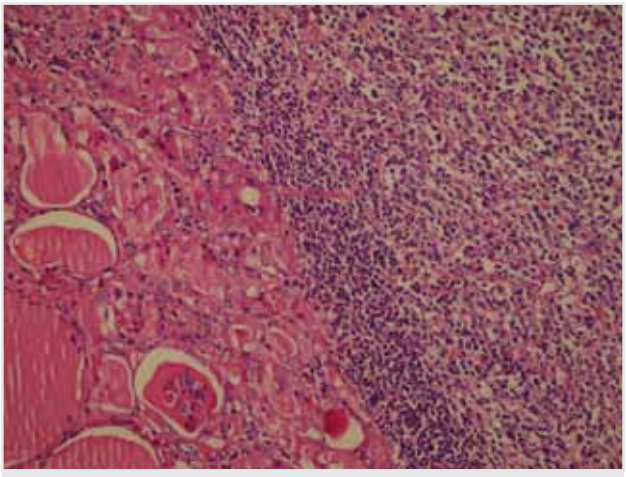

Figure 2. A lymphoid follicle with a prominent germinal center and Hurthle cells (H&E X 300)

The mechanisms leading to the presence of ectopic thyroid tissue in the submandibular region may be due to displacement during the course of embryonic development, the spread of tissue following surgery on a normally located thyroid gland, and metastasis of a highly differentiated papillary thyroid carcinoma. In our case, the management approach was the surgical excision of the submandibular mass because of the suspicious FNAB (5-7). In this case, the history of previous thyroid surgery raises the possibility of inoculation as the mechanism behind the ectopic thyroid tissue development but does not completely exclude embryonic displacement as an additional contributing mechanism.

Conclusion

Ectopic submandibular thyroid tissue is extremely rare and requires case-specific management plans, including surgical excision or follow up with medical treatment. Surgical excision with post-operative hormone replacement therapy is the preferred treatment option in cases of an enlarged symptomatic mass in the neck with the suspicion of malignancy.

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

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References

1. Kousta E, Konstantinidis K, Michalakis C, Vorias M, Sambalis G, Georgiou M et al. Ectopic thyroid tissue in the lower neck with a coexisting normally located multinodular goiter and brief literature review. Hormones (Athens) 2005; 4: 231-4.
2. Klubo-Gwiezdzinska J, Manes RP, Chia SH, Burman KD, Stathatos NA, Deeb ZE, et al. Clinical review: Ectopic cervical thyroid carcinoma-review of the literature with illustrative case series. J Clin Endocrinol Metab 2011; 96: 2684-91. [CrossRef]
3. Kumar R, Sharma S, Marwah A, Moorthy D, Dhanwal D, Malhotra A. Ectopic goiter masquerading as submandibular gland swelling: a case report and review of the literature. Clin Nucl Med 2001; 26: 306-9. [CrossRef]
4. Toso A, Colombani F, Averono G, Aluffi P, Pia F. Lingual thyroid causing dysphagia and dyspnoea. Case reports and review of the literature. Acta Otorhinolaryngol Ital 2009; 29: 213-7.
5. Helidonis E, Dokianakis G, Papazoglou G, Pantazopoulos P, Thomopoulou H. Ectopic thyroid gland in the submandibular region. J Laryngol Otol 1980; 94: 219-24. [CrossRef]
6. Amoodi HA, Makki F, Taylor M, Trites J, Bullock M, Hart RD. Lateral ectopic thyroid goiter with a normally located thyroid. Thyroid 2010; 20: 217-20. [CrossRef]
7. Adotey JM. Papillary adenocarcinoma of thyroid in a patient with right submandibular mass-a rare case of lateral aberrant thyroid. Niger J Clin Pract 2009; 12: 333-4.