Ectopic submental thyroid tissue, with no evidence of orthotopic thyroid: a case report

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ABSTRACT

Background: Thyroid ectopia refers to a congenital disease with abnormal descending of the thyroid gland. The most common subtype is lingual thyroid ectopia. Sublingual and submental thyroid ectopia are much less common. It may also be found in other neck locations and distant positions .70%–90% of patients with ectopic thyroid do not have eutopic thyroid tissue. It is necessary to distinguish ectopic thyroid from other causes of neck masses.

Case Presentation: A 18-year-old man was referred for the evaluation of a palpable mass in the submental region. On biochemical examination, the thyroid function tests were normal while receiving levothyroxine therapy. Ultrasonography revealed a mass measuring $4.2 \times 2.7 \times 2.6$ cm in the submental region. The thyroid scan did not show any uptake in the thyroid region. A zone of the tracer activity was noted in the central neck, compatible with the palpated mass in the submental region. CT images showed a hyperdense soft tissue in the submental region, too.

Conclusion: Thyroid ectopia is a rare condition and it is often accompanied by hypothyroidism. Ectopia often occurs in lingual and sublingual locations, and submental one is less common. Thyroid scintigraphy is the best imaging modality for the detection of thyroid ectopia. For precise localization, the CT scan correlation may be needed.

Keywords: Thyroid ectopia, submental, thyroid scintigraphy, case report.

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Background

Thyroid ectopia refers to a congenital disease where the abnormal descending of the thyroid gland in the embryogenic stage results in finding thyroid tissue outside of the normal thyroid location, which is anterior to the 2nd–4th tracheal rings [1–5]. It is usually found around the thyroglossal duct [3]. The most common subtype is lingual thyroid ectopia, which accounts for >90% of the reported cases [1,2,5–7]. Sublingual thyroid ectopia is much less common (<10%) [1]. It may also be found in other neck locations and distant positions such as mediastinum, intracardiac, and subdiaphragmatic organs such as the adrenal gland [3,5,6,8].

Until now, there is no known gene responsible for the human ectopic thyroid [2]. The prevalence of the thyroid ectopia is about 1 per 100,000–300,000 population and 1 per 4,000–8,000 patients with the thyroid disease [1,2,6,8]. In postpartum studies, the frequency reaches 10% [1,3,9]. It is more common among females (65%–80% of cases) [1,2,7]. It mostly occurs at younger ages, especially during physiological states, such as puberty, pregnancy, and menopause, where the demand for thyroid hormones increases and the thyroid tissue is susceptible to hypertrophy [1,2,6,8]. Symptoms depend on the location

of ectopic tissue and may present with mass effects, dysphagia, dysphonia, dyspnea, and sensation of chocking, but it is usually asymptomatic with an indolent course for decades [1,6–8]. Hypothyroidism is reported in 70% of patients, concurrently, and it is a predisposing factor for the hypertrophy of the ectopic thyroid tissue [1,2,5,7,8]. Hyperthyroidism state is rare [7]. Furthermore, the ectopic thyroid tissue can potentially develop various functional abnormalities including goiter, thyroiditis, benign, and malignant cancers [1,3,6]. The latter accounts for 1 in 100 of ectopic cases, and the follicular thyroid carcinoma is the most common form, whereas the papillary thyroid carcinoma mostly occurs in eutopic thyroid glands [1].

As 70%–90% of patients with ectopic thyroid do not have eutopic thyroid tissue, it is necessary to distinguish ectopic thyroid from other causes of neck masses, especially thyroglossal duct abnormalities. Because if it is removed, the patient will be at the risk of permanent hypothyroidism [1-7].

Case Presentation

An 18-year-old man was referred to our department for further evaluation of a palpable mass in the submental region (Figure 1). He had recognized a bulging mass in the submental region from the past six years, which has been gradually more prominent in the last two years. He was also suffering from hypothyroidism from 11 years ago and was receiving levothyroxine for two months. He did not have any difficulty in breathing or swallowing. On neck examination, there was a non-tender, soft, 3×3 cm mass below the chin, which moved freely on swallowing and tongue protrusion with no signs of infection.

On biochemical examination, the thyroid function test was normal while receiving levothyroxine therapy. (TSH = 1.81 μ IU/ml, T_3 = 98 ng/dl, T_4 = 4.8 μ g/dl, anti-TPO = 8 IU/ml). Ultrasonography was performed before the scan and revealed a mass measuring 4.2 × 2.7 × 2.6 cm in the submental region near to midline. A hypoplastic thyroid was also detected in the thyroid bed, 0.5 × 0.4 cm and 0.4 × 0.4 cm corresponding to the right and left lobes, respectively. Thyroid scan was performed by dual-head Siemens gamma camera, 15 minutes after IV injection of 185 MBq (3.5 mCi) of Tc-99m-pertechnetate, and static images (anterior, lateral) were obtained in the supine position (Figures 2 and 3) The scan did not show any uptake in the thyroid region. A zone of tracer activity was noted in the submental region.

Computed tomography (CT) scan was performed for the precise localization of the ectopic thyroid tissue. CT images showed a hyperdense soft tissue in the submental region, which was adjacent to the base of tongue



Figure 1. Image of the patient in lateral view, showing a visualized soft tissue mass in submental region.

(Figure 4). There was no visible thyroid tissue in the thyroid bed region.

There was no salivary abnormality or adenopathy in the rest of the neck. Surgery and observation were the two treatment options. He did not have any obstructive symptoms, so the surgical approach was withheld, and the patient was observed for the possible enlargement of the mentioned mass while receiving thyroxine therapy.

Discussion

Thyroid ectopia is a rare condition with an incidence of about 1 in 100,000 populations, and it should be kept in mind during the evaluation of neck masses [1]. There are some clinical pathologies that can imitate thyroid ectopias, such as epidermoid and dermoid cyst, thyroglossal duct cyst, lymphadenopathy, lipoma, lymphangioma, mucous retention cyst, adenoma, hemangioma, subhyoid bursitis, midline bronchial cyst, salivary gland tumors, vascular malformation, and neoplasms [2,10]. Various diagnostic procedures and imaging modalities have been introduced to date including ultrasound, CT scan, nuclear scintigraphy, and MRI [1–2].



Figure 2. Thyroid scan in anterior view showing thyroid tissue in submental region.



Figure 3. Thyroid scan in lateral view, confirming thyroid tissue in submental region.



Figure 4. Ct scan performed from the neck region, showing a hyperdense soft tissue in the sub-mental region, adjacent to the base of tongue.

The main drawback of ultrasound is its low detection rate for thyroid ectopia [11]. If the thyroid tissue is not seen in its normal region, on sonography, a thyroid scan is required to differentiate thyroid agenesis from ectopia [11]. Thyroid scintigraphy is the most important diagnostic modality, as well as the best non-invasive imaging for the evaluation of thyroid ectopia and differentiates it from other causes of neck mass [2,5,6,8,10,11]. It has high sensitivity and specificity and can identify any thyroid tissue throughout the body [5]. FNA has also been used with an accuracy of 95%–97%, especially when there is a suspicion of malignancy [1].

Because the ectopic thyroid tissue is usually the only functional thyroid tissue in these patients, excision will make the patient hypothyroid [1]. For asymptomatic and euthyroid patients, no surgical and medical treatments are needed and follow-up is recommended [2].

Treatment is dependent on thyroid function and the patient symptoms [1]. If the patient has symptoms that are related to mass effects, such as obstructive symptoms, bulk reduction with thyroid hormone replacement is indicated even if the patient is euthyroid [1-2].

Levothyroxine leads to achieve euthyroid status and decreases the size of ectopic tissue [5]. In cases of suspicious or proven malignancies, and patients with mass-related symptoms, such as dyspnea, dysphagia, dysphonia, and hemorrhage, surgery is indicated [1].

List of Abbreviations

CT Computed tomography

MRI Magnetic resonance imaging

Consent for publication

Informed consent was obtained from the patient for the publication of this case report and any accompanying images.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

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References

- Perdoni C, Eustaquio ME. Ectopic thyroid presenting as a submental mass: a case report. Otolaryngol Case Rep. 2017;4:9–11. https://doi.org/10.1016/j. xocr.2017.07.001
- Sarkar S, Panja S, Mandal K, Kumar S. Colloid goiter in submental ectopic thyroid: a rare entity posing a diagnostic and therapeutic challenge. Int Surg J. 2016;3(1):377–81. https://dx.doi.org/10.18203/2349-2902.isj20160263
- Oguz A, Tuzun D, Ozdemir E, Ersoy R, Yazgan AK, Cakir B. Importance of ectopic thyroid tissue detected in the midline of the neck: single center experience. Arch Endocrinol Metab. 2015;60(3):231–5. https://dx.doi. org/10.1590/2359-3997000000073
- Aalaa M, Mohajeri-Tehrani MR. Images in clinical medicine. Ectopic thyroid gland. N Engl J Med. 2012; 366(10):943. https://doi.org/10.1056/NEJMicm1106077
- Sood A, Sood V, Sharma DR, Seam RK, Kumar R. Thyroid scintigraphy in detecting dual ectopic thyroid: a review. Eur J Nucl Med Mol Imag. 2008;35(4):843–6. https://doi.org/10.1007/s00259-007-0672-2
- Kocova M, Zdraveska N, Zdravkovska M, Anastasovska V, Pop Gjorceva D. Submental thyroid ectopy might cause subclinical hypothyroidism in early childhood. SAGE Open Med Case Rep. 2016;4. https://doi. org/10.1177/2050313X16683623
- Jain A, Pathak S. Rare developmental abnormalities of thyroid gland, especially multiple ectopia: a review and our experience. Indian J Nucl Med. 2010;25(4):143–6. https://doi.org/10.4103/0972-3919.78248
- Manohar K, Bhattacharya A, Kashyap R, Kamaleshwaran KK, Mittal BR. Concurrent sublingual thyroid and thyroglossal cyst with functioning thyroid tissue in the absence of an orthotopic thyroid gland. Jpn J Radiol. 2010;28(7): 552–4. https://doi.org/10.1007/s11604-010-0467-4
- Batsakis JG, El-Naggar AK, Luna MA. Thyroid gland ectopias. Ann Otol Rhinol Laryngol. 1996;105(12):996–1000. https://doi.org/10.1177/000348949610501212
- Marković V, Glavina G, Eterović D, Punda A, Brdar D. Dual ectopic thyroid gland: sonography and scintigraphy of lingual and sublingual thyroid. Clin Nucl Med. 2014;39(6):556–8. https://dx.doi.org/10.1097/ RLU.000000000000415
- Karakoc-Aydiner E, Turan S, Akpinar I, Dede F, Isguven P, Adal E, Guran T, et al. Pitfalls in the diagnosis of thyroid dysgenesis by thyroid ultrasonography and scintigraphy. Eur J Endocrinol. 2012;166(1):43–8. https://doi. org/10.1530/EJE-11-0140

Summary of the case

1	Patient (gender, age)	Man-18 y/o
2	Final diagnosis	Thyroid ectopia
3	Symptoms	Submental mass
4	Medications	Leothyroxine therapy
5	Clinical procedure	Thyroid scintigraphy
6	Specialty	Nuclear medicine