

Case Report

SUBLINGUALE CTOPICTHYROIDGL AND WITH HYPOTHYROIDISM: AN UNUSUAL ASSOCIATION

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ABSTRACT

Ectopic thyroid gland is a rare congenital anomaly which is seen with a frequency of 1:100,000-300,000. Ectopic thyroid tissue is most commonly localized at midline in dorsum of tongue (90%). Generally, ET gland has subclinical course. Symptoms of hypothyroidism are found in 33% of the patients, while ectopic tissue is the only active thyroid gland in 70% of the patients.

A 32 years old woman presented with depression, fatigue and menstrual irregularity. In the laboratory assessment, we identify hypothyroidism secondary to sublingual ectopic thyroid gland. On the thyroid ultrasonography, it was failed to observe thyroid gland at normal localization, and no ectopic tissue was also detected. On the thyroid scan, visual change was seen at the focal uptake area at sublingual region. Thyroid autoantibodies were negative. Here, we report a patient with hypothyroid associated with ectopic sublingual thyroid tissue. The size of the tissue was decreased after the six months thyroxin therapy in comparison to the previous thyroid scan.

Key Words: *Sublingual Thyroid, Ectopic Thyroid Gland, Hypothyroidism.*

INTRODUCTION

Ectopic thyroid (ET) gland is a rare congenital anomaly which is seen with a frequency of 1:100,000-300,000. It is resulted from the descending failure of thyroid gland to normal localization and settled thyroglossal duct during embryological development. It is more prevalent in women than men (Bayram *et al.*, 2004). ET tissue is most commonly localized at midline in dorsum of tongue (90%) (Bayra *et al.*, 2004). Trachea, lateral cervical, submandibular, sublingual and palatine tonsils are the areas in where ET tissue can be localized (De Felice *et al.*, 2004). Generally, ET gland has subclinical course (Ibrahim *et al.*, 2011). Symptoms of hypothyroidism are found in 33% of the patients, while ectopic tissue is the only active thyroid gland in 70% of the patients (Ibrahim *et al.*, 2011). Clinical follow-up is needed for the treatment in terms of hypothyroidism. However, excision is recommended to those with obstructive symptoms (dyspnea, dysphagia etc.) and in malignancy suspicion. In the current study, we aimed to present a patient with ectopic (sublingual) thyroid gland associated with hypothyroidism.

CASES

A 32 years old woman presented to our outpatient clinic with depression, fatigue and menstrual irregularity. The case is suffering from the complaints for 3 years, and has been referred to psychiatry, neurology and gynecology departments. No palpable mass was detected at the neck during physical examination in the family medicine outpatient clinic. In the laboratory assessment, free T3: 2.65 pg/mL (N: 2.5-3.9), free T4: 0.59 ng/dL (N: 0.61-1.12), TSH: 39.3 µIU/mL (range: 0.34-5.60) and thyroid autoantibodies including anti thyroid peroxidase (Anti-TPO) antibody and anti-thyroglobulin antibody (Anti-Tg) were negative. On the thyroid ultrasonography (USG), it was failed to observe thyroid gland at normal localization, and no ectopic tissue was also detected. On the Tc-99m pertechnetate thyroid scintigraphy no uptake was observed at both lobes of thyroid gland in anterior views. A focal uptake was seen at the upper midline region of neck (Figure 1). In the lateral views, the uptake was clearly identified at sublingual region (Figure 2). On the laryngo-pharyngoscopic examination, no mass appearance was

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detected at the tongue root that may affect the function of respiration and swallow. Six months thyroxin replacement treatment was given due to the diagnosis of hypothyroidism secondary to sublingual ET tissue. During the control examination after six months, following findings were recorded: free T3:2.28 pg/ml (N: 2.5-3.9), free T4: 0.88 ng/dl (N:0.61-1.12), TSH: 9.72 μ IU/ml (range: 0.34-5.60). Visual change was seen at the focal uptake area at sublingual region before (25x20 mm) and after (19x19 mm) the thyroxin treatment in the thyroid scans. It is suggested that complaints of patients were improved and the size of ET tissue was decreased after the drug therapy.

DISCUSSION

ET gland is a rare congenital anomaly, and it was firstly reported in a newborn who died due to upper respiratory tract obstruction within the hours after the birth by Hickmann *et al.*, in 1869. It is generally localized at lingual region in the 90% of the cases. Other rare areas for the localization are cervical lymph nodes, esophagus, submandibular region, sublingual region and pericardium (Toso *et al.*, 2009). In the current patient, ET gland was localized at the sublingual area as rarely detected. It is more common in females than males (Bayram *et al.*, 2004). Hormonal changes observed during puberty and pregnancy causes an increase in the volume of ET gland. Our case was a 32-year-old female without history of pregnancy.

In general, cases with ET gland are asymptomatic in terms of hormonal status (Ibrahim *et al.*, 2011). Dysphagia, sensation of stuck in throat, dyspnea and hemoptysis may be seen due to enlargement of ectopic gland. In our patient, there were depression, fatigue and menstrual irregularity without any obstructive symptoms regarding sublingual localization of ectopic gland.

ET tissue remains rudimentary because of developmental defect. Thus, it may not provide sufficient circulating hormone as normal thyroid tissue (Wang *et al.*, 2010). Hormone levels released by ectopic tissue do not cause overt clinical symptoms in the patients. Therefore, there is a subclinical hypothyroidism in 70% of the patients with ET gland (Wang *et al.*, 2010). However, hypothyroidism symptoms may be seen in the terms such as puberty, pregnancy, trauma, excessive psychological stress and infection in where requirement of thyroid hormone is increased, since ectopic gland cannot meet the increased hormone requirement (Willi *et al.*, 1991). In the adult cases, ET gland is associated with hypothyroidism in 33% of cases (Ibrahim *et al.*, 2011). Clinical and laboratory findings were consistent with hypothyroidism as in our case. Ectopic gland is the only functional thyroid tissue in the 70% of the cases with ectopic sublingual thyroid gland (Ibrahim *et al.*, 2011). In the present case, there was no thyroid tissue at normal anatomic localization apart from sublingual ET tissue.

USG, computerized tomography (CT), magnetic resonance imaging (MRI) and scintigraphy can be used in the diagnosis of ET gland (Toso *et al.*, 2009). CT is generally preferred in cases which cannot be detected by USG. MRI is specifically used in lingual thyroid cases in which differentiation between thyroid tissue and tonsillar plica is difficult (Noussios *et al.*, 2011). In our case, no thyroid tissue was detected at midline of neck by USG. On thyroid scan, an ET tissue was detected at sublingual region. As ET gland has an asymptomatic clinical course in 70% of the patients, regular clinical follow-up is sufficient in aspect of hypothyroidism. Thyroxin therapy is effective in case of relieving the symptoms and shrinking the size of ET tissue (Noussios *et al.*, 2011, Chawla *et al.*, 2007). In our case, it was seen that the size of the ET gland was decreased on thyroid scan after 6 months thyroxin therapy in addition to clinical improvement. Surgical treatment is used in cases with obstructive symptoms such as dysphagia or dyspnea (Chawla *et al.*, 2007, Hafidh *et al.*, 2004). Furthermore, radioactive I-131 therapy is one of the alternatives in order to shrink lesion (Kennedy *et al.*, 2007). Rarely, malign transformation may be seen in ET gland (Iglesias *et al.*, 2008). This transformation may occur in different localizations and types. In the literature, there are reports of cases with follicular, papillary and medullar carcinoma (Yetim *et al.*, 2010). Surgical treatment is essential in this setting (Yetim *et al.*, 2010).

Conclusion

To report a woman with hypothyroidism associated with sublingual ectopic thyroid gland. We present a case report including laboratory, radiographic, scintigraphy findings before and after thyroxin therapy in a

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patient with hypothyroidism associated with sublingual ectopic thyroid gland as well as a brief review of the literature. Here, we report a 32 years old female case regarding hypothyroidism associated with sublingual ETgland, which is rarely seen without obstructive symptoms. Also, ET tissue size was decreased after the six months thyroxin therapy in comparison to the previous thyroid scan.

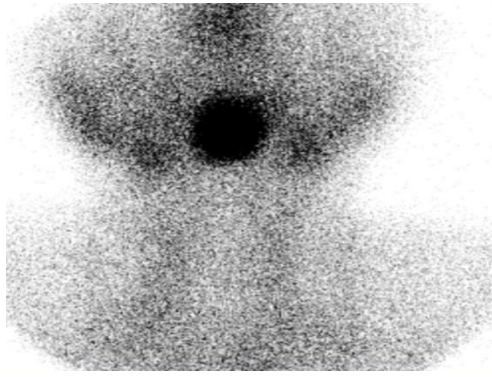


Figure 1: The anterior view of focal activity uptake at superior midline of neck on thyroid scintigraphy.

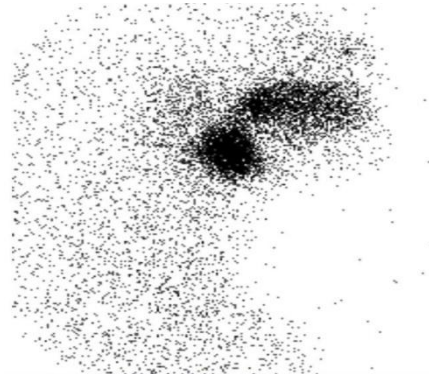


Figure 2: The lateral view of focal activity uptake at sublingual area on thyroid scintigraphy.

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