

Case Report

Submental Ectopic Thyroid in a Patient with an Orthotopic Thyroid Gland

CME
Credits

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Abstract

Ectopic thyroid is a rare clinical disease, with the majority of the patient presented with hypo-functional thyroid tissue and absence of orthotopic thyroid gland. During embryogenesis, the thyroid gland descends from the foramen cecum to its normal position located below the larynx and hyoid bone, anterior to the 2nd, 3rd, and 4th tracheal cartilaginous rings. Any defect that occurs during this period will result in ectopic thyroid. Hence, ectopic thyroid is usually found along the course of the thyroglossal duct, laterally in submandibular glands and even in distant places such as mediastinum or very rarely in sub-diaphragmatic organs. However, the presence of an orthotopic thyroid may lead some difficulties in the diagnosis of this disease; hence, fine-needle aspiration is recommended to avoid unnecessary operation provided the patient is asymptomatic.

Keywords: Ectopic thyroid, nodular goiter, submental

INTRODUCTION

Ectopic thyroid is a rare clinical entity, resulting from abnormal embryonic development and migration of the thyroid gland. Normally, thyroid tissue is found anterior to the 2nd to 4th tracheal rings; however, in ectopic thyroid, the thyroid gland is found outside its typical position. Anatomically, ectopic thyroid can be divided into two subtypes: (a) lingual subtype which consists almost 90% of the cases is found between the circumvallate papillae and epiglottis, along the midline of the tongue base and (b) sublingual subtype, making up <10% of all ectopic thyroids, found along the course of the thyroglossal duct cyst at either supra or infra-hyoid locations, outside the tongue parenchyma, located between the geniohyoid and mylohyoid muscles.^[1-3]

CASE REPORT

A 59-year-old female was referred to our hospital with a 10-year history of a painless, slowly enlarging submental swelling [Figures 1 and 2]. Physical examination showed that the submental swelling was nontender, smooth, and movable measuring about 3 cm × 3 cm × 2 cm in size. She denied any difficulty in swallowing and breathing or any history of fever

or local trauma. Laboratory studies showed mild elevation of TSH 6.7 mU/L (normal: 0.5–5.5), and free T4 level was 0.8 ng/dL (normal: 0.7–1.48 ng/dL)

Neck sonography showed enlarged sizes of both thyroids, multiple heterogeneous hyperechoic and hypoechoic nodules were seen in both thyroid suggesting multinodular goiter [Figure 3]. Bilateral submandibular glands were unremarkable. A well-defined lobulated hyperchoic nodule measuring 2.9 cm × 1.6 cm × 2.7 cm having higher echogenicity than the orthotopic thyroid gland was seen at the submental region [Figure 4], intranodular vascularization was detected with color Doppler sonography [Figure 5], and no calcification was seen. The tissue of origin was not definitely identified. As the nodule was suspicious for neoplasm, fine-needle aspiration of this nodule was performed.

Cytology showed that this was a macrofollicular lesion composed of moderate follicular cells arranged in honeycomb flat sheets, abundant macrophages, and some colloid materials [Figure 6]. Cell block section reveals variable-sized

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Figure 1: Anteroposterior view of the neck showed submental swelling (arrow)



Figure 2: Oblique view of the neck showed submental swelling (arrow)

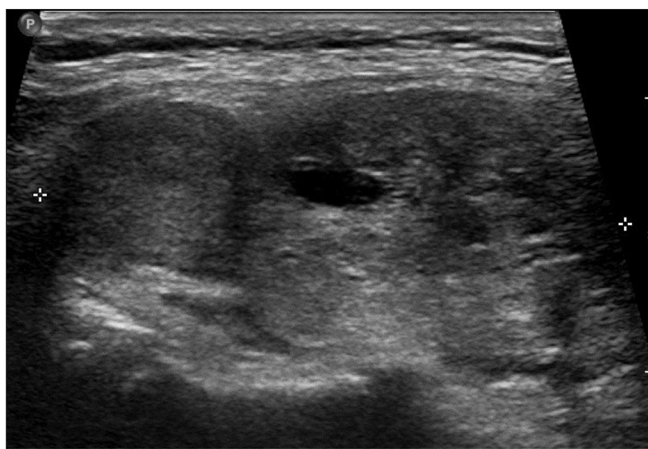


Figure 3: Sonography of the orthotopic thyroid showed enlarged size of the thyroid containing heterogeneous mixed solid and cystic nodule



Figure 4: A well-defined lobulated isoechoic nodule was seen at the submental region

follicular glands filled with colloid [Figure 7]. These findings were suggestive of nodular goiter.

Both observation and surgical intervention were suggested as the treatment options to the patient. Given that the patient was asymptomatic and only TSH was mildly elevated, the patient decided to be followed up regularly.

DISCUSSION

Ectopic thyroid is a congenital condition due to abnormal migration of thyroid gland in embryonic stage, which usually manifests later in life, especially during increased physiological demand of thyroid hormones such as puberty, pregnancy, and menopause.^[4] The prevalence of this entity is 1/100,000–300,000 people and 1 in 4000–8000 patients with thyroid diseases and male to female ratio is 1/4. Interestingly, in autopsy studies, up to 7%–10% of population may have ectopic thyroid tissue along the thyroglossal duct.^[5]

Ectopic thyroid can manifest clinically in any age group, particularly in Asian females over 40 years of age.^[5] Patients

may be asymptomatic or present with symptoms depending on size, location, and associated endocrine dysfunction of ectopic glands. These include dysphagia and dyspnea which are related to mass effect caused by the ectopic thyroid. About 70% of patients develop hypothyroidism, a phenomenon thought to be caused by disrupted vascular supply secondary to abnormal migration of the thyroid.^[6]

Similar to a normal thyroid, the ectopic thyroid is susceptible to the same pathology that affects a normal thyroid, including goiters, Hashimoto's thyroiditis, benign adenomas, and malignancies.^[7] The incidence of carcinoma in ectopic thyroid is reported to be about 1%. In contrast to the malignancies found in the orthotopic thyroid gland, follicular carcinoma is the most common subtype in ectopic thyroid, instead of the more common papillary subtype.^[8]

As 70%–90% of patients do not have normal thyroid tissue outside the ectopic location,^[4] hypothyroidism is common, whereas hyperthyroidism is very rare.^[9] Therefore, we must keep in mind that when surgical excision is considered, these

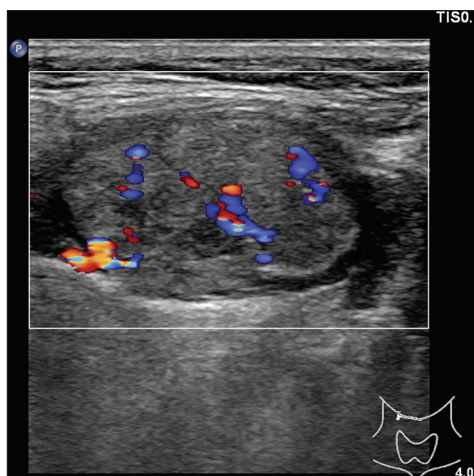


Figure 5: Color Doppler sonography showed presence of intranodular hypervascular flow

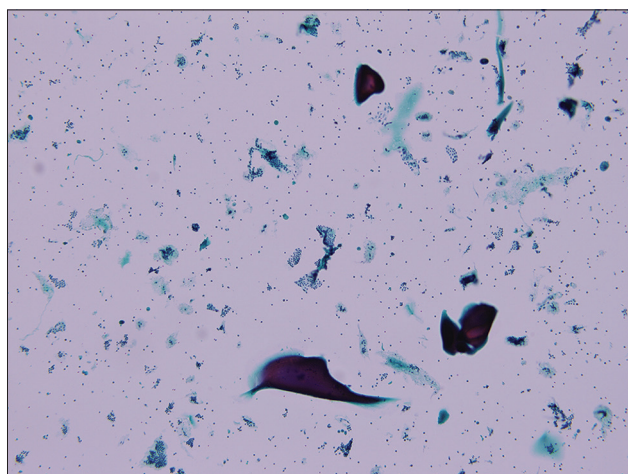


Figure 6: The cytologic smear showed benign macrofollicular fragments and some colloid substances (Pap, $\times 100$)

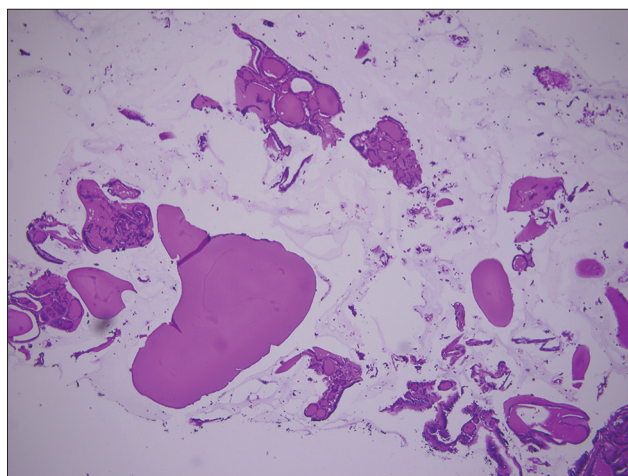


Figure 7: Cell block section reveals variable-sized follicular glands filled with colloid

patients are at risk of developing permanent hypothyroidism postoperatively.

The differential diagnosis of a submental mass should include thyroglossal duct cysts, midline branchial cysts, lymphadenopathy, lipomas, epidermoid cysts, and neoplasm. In our patient, the mass is solid; hence, thyroglossal duct cyst and branchial cyst are excluded. The presence of vascular flow and absence of hyperechoic hilum in the nodule help us to exclude the possibility of lipomas, epidermoid cysts, and lymphadenopathy.

If ectopic thyroid or neoplasia is suspected, particularly in the presence of an orthotopic thyroid gland, fine-needle aspiration is suggested because it can identify thyroid tissue with a 95%–97% accuracy rate and is reliable for excluding malignancy.^[4] Multiple image modalities are available for the diagnosis of ectopic thyroid. Ultrasound, CT, and MRI may show an ectopic thyroid as well as the presence or absence of orthotopic thyroid. However, for ultrasound, when a mass is detected in the neck region, we must practice in a routine manner to screen the normal position of the thyroid gland, so that a correct diagnosis can be achieved. Scintigraphy with technetium-99 m remains as the gold standard for ectopic thyroid diagnosis, not only confirming the location and extension of the ectopic thyroid and the presence or absence of the orthotopic thyroid but can also provide information about the shape and overall activity of the thyroid, which is often unobtainable with other imaging modalities.

The treatment options of ectopic thyroid depend on the patient's clinical symptoms and thyroid functions. If the patient is asymptomatic or minimally symptomatic such as in our case, observation is recommended and no other intervention procedure is necessary. In contrary, bulk reduction with thyroxine replacement therapy are suggested for hypothyroid patients with lingual thyroids and those having symptoms attributable to mass effect caused by compensatory gland hypertrophy.^[10] Surgical intervention is reserved for patients having ectopic sublingual thyroids or lingual thyroids which are refractory to bulk reduction or in symptomatic patients having dyspnea, dysphagia, or hemorrhage regardless of thyroid status.^[3,4]

CONCLUSION

Ectopic thyroid although rare should be kept in clinician's mind in the differential diagnosis of midline neck swelling; sonography and fine-needle aspiration cytology is important in diagnosing the disease, particularly in the detection of the presence/absence of orthotopic thyroid, so that unnecessary operation or postoperative hypothyroidism can be avoided. Another important thing is that as 70%–90% of the patients with ectopic thyroid do not have a functional orthotopic thyroid,^[3] it is necessary to inform patients about the possibility of permanent hypothyroidism if surgical resection is performed.

Declaration of patient consent

The author certifies that he has obtained all appropriate patient consent forms. In the form the patient has given her consent

for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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