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CLINICAL RECORD

# Ectopic, submandibular thyroid causing hyperthyroidism

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## Abstract

*Background*: Ectopic thyroid tissue in the submandibular region is exceptionally rare. Nevertheless, the treating physician should consider this condition within the differential diagnosis of a submandibular mass.

*Method*: Case report of ectopic thyroid tissue presenting as a submandibular mass in a patient with hyperthyroidism, together with a review of the English-literature concerning ectopic thyroid tissue.

*Conclusion*: To our knowledge, this is the first report of ectopic thyroid tissue presenting as a submandibular mass and causing hyperthyroidism. Removal of the submandibular mass resulted in normalisation of thyroid function, and subsequent reduction in the patient's antithyroid medication dosage. Ectopic thyroid tissue should be suspected in any patient with a submandibular mass. Prior to resection of such a lesion, it is essential to ensure that normal, functioning thyroid tissue is present elsewhere. Ectopic thyroid tissue can also present with pathology similar to that affecting the normal thyroid gland.

**Key words:** Thyroid dysgenesis; Neck; Thyrotoxicosis; Thyroid Neoplasm

## Introduction

The thyroid gland develops in the fourth week of gestation and subsequently descends down the neck to its final position, anterior to the trachea, by the seventh week of gestation. This migration of thyroid cells can give rise to ectopic thyroid tissue. The most common sites of ectopic thyroid tissue occur along this migration path, and include the tongue, larynx, trachea, oesophagus, mediastinum, heart and abdomen.[1,2] Ectopic thyroid tissue has been described at other sites, albeit rarely.

Thyroid tissue in the submandibular region is extremely rare. Nevertheless, it is important that the treating physician consider this rare condition within the differential diagnosis of a submandibular mass.

We present a rare case of ectopic thyroid tissue presenting as a submandibular mass in a patient with hyperthyroidism.

## Case report

A 51-year-old woman presented in January 2010 with a left-sided, submandibular mass. She was otherwise asymptomatic.

On clinical examination, a smooth, soft, $3 \times 2 \times 1$ cm mass was noted in the left submandibular region. The rest of the clinical examination was normal, as was flexible nasal endoscopy.

The patient had been successfully treated for grade one breast cancer in 2001, with wide local excision, radiotherapy and tamoxifen. She was an ex-smoker.

Furthermore, in 1998 she had been diagnosed with Graves' thyrotoxicosis, with raised microsomal antibodies, and had been treated with carbimazole ever since. At that time, she had been noted to have a small, diffuse, palpable goitre together with mild signs of thyroid eye disease, with left-sided lid lag and lid retraction. Although she had remained clinically euthyroid, thyroid function tests had revealed biochemical hyperthyroidism, as follows: thyroid stimulating hormone (TSH) concentration = 0.03 mU/l (normal range 0.27–4.2), free thyroxine (T4) concentration = 23.3 pmol/l (normal range 12–22) and free triiodothyronine (T3) concentration = 7.3 nmol/l (normal range 3.1–6.8). These results were in spite of a fivefold increase in carbimazole dose in the preceding 12 months.

Ultrasonography of the neck revealed a soft tissue mass in the submandibular triangle. The mass was hypoechoic, with chaotic vascularity and a lobulated contour. The remainder of the ultrasound examination demonstrated mild thyroid enlargement only. Fine needle aspiration cytology was performed (Figure 1); the report identified thyroid tissue with no evidence of malignancy.

Following discussion with the patient, open surgical biopsy was performed in March 2010. This revealed a mass in the submandibular triangle measuring $30 \times 20 \times 12$ mm. Histological examination identified a hyperplasic thyroid nodule. There were no features of follicular or papillary carcinoma.

Following surgery, the patient's carbimazole dose was able to be progressively reduced to less than half her pre-surgical dosage, while still maintaining biochemical

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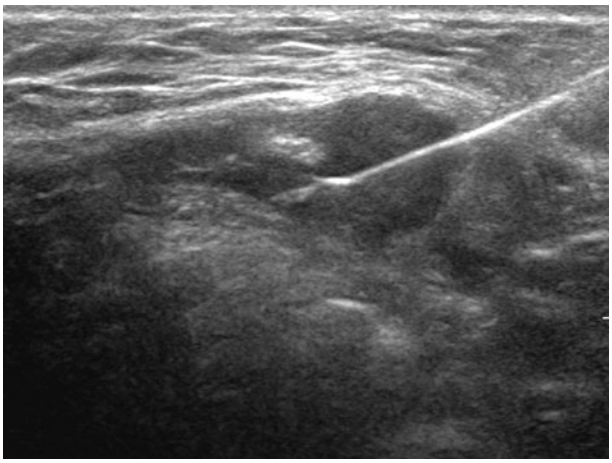


FIG. 1
Ultrasonographic image showing performance of core biopsy of the submandibular mass.

euthyroidism (i.e. TSH = 1.0 mU/l and free T4 = 14.1 pmol/l).

## Discussion

The thyroid gland is the first endocrine gland to develop in the embryo, and begins to form, approximately 24 days after fertilisation, 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 seven 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 ectopic thyroid tissue.

- **This report describes a case of ectopic thyroid tissue presenting as a submandibular mass and contributing to hyperthyroidism**
- **Ectopic thyroid tissue should be included in the differential diagnosis of a submandibular mass**
- **The presence of normally sited, functioning thyroid tissue should be confirmed before ectopic thyroid tissue is resected**

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 actual incidence of ectopic thyroid tissue may be higher than reported, because 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.[3]

In general, the presence of ectopic thyroid tissue in the submandibular region is explained by: (1) displacement during the course of embryonic development, (2) spread of tissue during surgery on a normally located thyroid gland, and (3) metastasis of a highly differentiated papillary thyroid carcinoma.[1]

To our knowledge, since the 1700s there have been 30 reported cases of ectopic thyroid tissue in the submandibular triangle, worldwide. The majority of these cases were benign,[1,3–8] but six had abnormal thyroid pathology[2,9–13] and three had well differentiated thyroid cancer.[11,14,15] The majority of these cases occurred in middle-aged women, although cases in children and the elderly have also been reported. However, there have been no previous reports of patients with functioning, submandibular ectopic thyroid tissue and worsening thyroid status, who despite diagnostic difficulty responded to excision of the submandibular mass.

The presented case illustrates the need to consider ectopic thyroid tissue when evaluating a patient with a submandibular mass and pre-existing thyroid disease, especially when thyroid function deteriorates for no apparent reason.

## Conclusion

Ectopic thyroid tissue should be included in the differential diagnosis of a submandibular mass. Prior to resection of such a lesion, it is essential to ensure that normal, functioning thyroid tissue is present elsewhere. Ectopic thyroid can also present with pathology similar to that affecting the normal thyroid gland.

As demonstrated in this case report, removal of ectopic submandibular thyroid tissue can facilitate the management of hyperthyroidism.

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