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Benign ectopic submandibular thyroid with a normotopic multinodular goitre

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Abstract
Ectopic submandibular thyroid tissue is a rare entity and poses difficult diagnostic and management problems. The first case of ectopic submandibular thyroid with a normotopic multinodular goitre is presented

Key words: Thyroid Gland; Ectopic Tissue; Submandibular Gland

Introduction

Ectopic submandibular thyroid is diagnosed rarely and usually presents without a normotopic thyroid gland. Ectopic submandibular thyroid with a normotopic thyroid mass poses difficult diagnostic and management problems. We present such a case.

Case report

A 77-year-old female patient presented with a five-month history of a right-sided neck-lump. The patient was asymptomatic and had no significant past medical history. Clinical examination identified a mass in the right submandibular triangle of her neck measuring approximately 1 × 2 cm. The remainder of the examination including fibre-optic laryngoscopy was normal.

Fine needle aspiration cytology (FNAC) from the submandibular mass identified numerous groups of follicular cells that were arranged in sheets and small groups with some microfollicle formation. The appearances were consistent with follicular thyroid tissue. Cervical ultrasound identified a normotopic thyroid gland with a lesion in the lower pole of the right lobe measuring 5 × 7 × 10 mm. This had a partly solid, partly cystic consistency. The right submandibular mass was noted to contain a small cystic area possibly due to necrosis. Computerized tomography (CT) of the neck confirmed a right submandibular mass, which was separate from the thyroid gland with a small mass in the right lobe. (Figures 1 and 2). A radioisotope thyroid scan identified increased activity in the right lower pole of the gland with no other abnormality. All blood tests were normal including thyroid function, calcitonin and thyroglobulin.

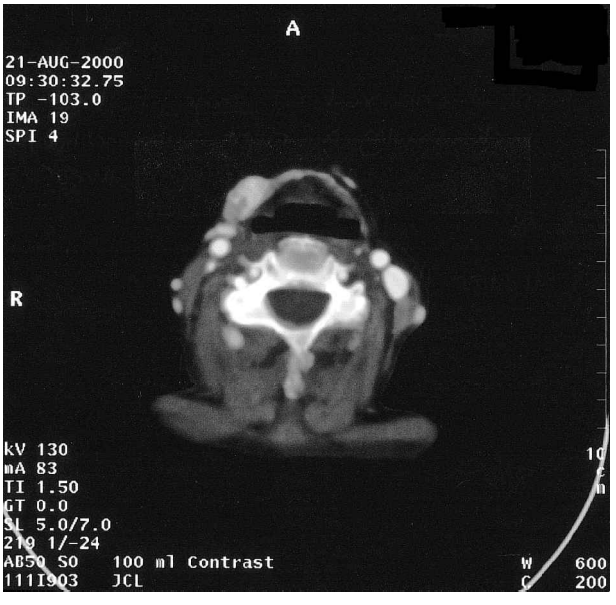

Fig. 1
Transverse CT scan of the neck showing a right submandibular mass.

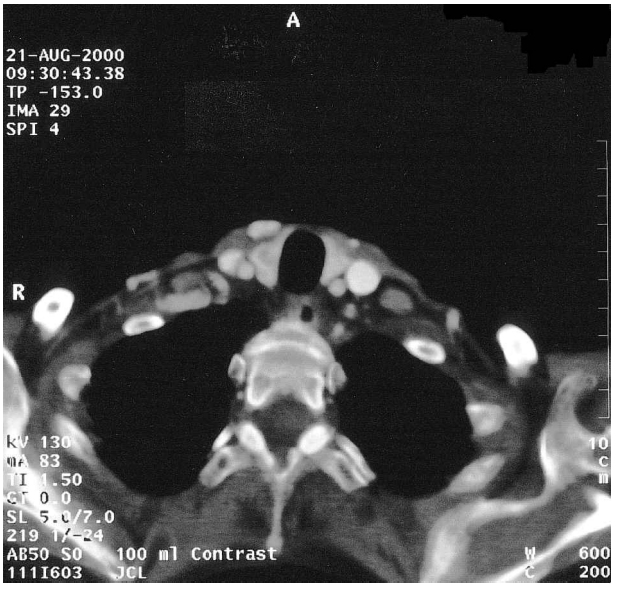

Fig. 2
Transverse CT scan of the neck showing a mass in the right side of the thyroid gland.

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The possible diagnosis of metastatic thyroid carcinoma was discussed with the patient along with the management options. Surgical options were discussed, including a staged procedure, which would be dictated by subsequent pathology results. The patient expressed her strong wish to undergo one definitive operation. A total thyroidectomy and right selective neck dissection were performed. Levels one to four nodes and a tan coloured nodule, lying anterior to the submandibular gland, were excised en bloc.

- **This clinical record reports aberrant thyroid tissue in the region of the submandibular gland**
- **The thyroid gland itself was not ectopic**
- **The authors discuss this finding in the light of what is known about the embryology and development of the normal thyroid gland**
- **The diagnosis presented difficult therapeutic decisions and these are outlined**

Histological examination of the submandibular mass identified thyroid follicular tissue with variably sized follicles, and partly cystic and haemorrhagic degeneration. Findings were consistent with ectopic submandibular thyroid tissue with nodular hyperplasia. The thyroid gland contained multiple nodules of variable size with the largest in the right lower pole. Findings were consistent with a multinodular goitre.

The patient, relieved at the diagnosis, had an uneventful post-operative period.

Discussion

The thyroid gland develops from a median and two lateral primordia. This median anlage forms an endodermal diverticulum in the midline of the ventral pharynx, eventually forming the bulk of the gland. While lobulation occurs it remains connected to the pharyngeal floor by the thyroglossal duct. The lateral anlagen arise as diverticula from the fourth and fifth pharyngeal pouches, eventually fusing with the median portion. Abnormalities in the development result in ectopic thyroid tissue usually found in the midline. This tissue can be functioning or non-functioning, benign or malignant.

Lateral ectopic thyroid tissue is much less common. In the past the term 'lateral aberrant thyroid' has been used and some examples were considered to be metastatic thyroid carcinoma. It is now established that lateral ectopic thyroid tissue can be benign or malignant, and can undergo the same pathological changes as normotopic thyroid gland, including thyrotoxicosis.[1]

There have been seven previous reports of ectopic submandibular thyroid tissue in the English literature.[1–7] All of the cases presented without a normotopic thyroid gland, except Sambola-Cabrer *et al.* who reported a case of ectopic submandibular thyroid with a normotopic atrophic thyroid gland on CT scan. The mass had been present for 23 years and was followed up after repeated benign FNACs without surgical excision.

We report the first case of submandibular ectopic thyroid tissue presenting with a mass in a normotopic thyroid gland. This creates difficult diagnostic and management problems. Ectopic submandibular thyroid should be considered in the clinical differential diagnosis of submandibular masses. The diagnosis is usually suspected from FNAC, but metastatic thyroid carcinoma, in particular papillary carcinoma, cannot be excluded. An ultrasound or CT scan will identify whether a normotopic thyroid gland is present. Surgical excision of ectopic submandibular thyroid tissue without this would result in iatrogenic hypothyroidism. In the case presented, follicular thyroid cells were identified on FNAC and metastatic follicular carcinoma or a follicular variant of papillary carcinoma could not be excluded. Without full histological evidence of malignancy a staged procedure was considered. In accordance with the British Association of Otorhinolaryngologists – Head and Neck Surgeons' Guidelines on thyroid cancer management, the minimum procedure indicated was a diagnostic right thyroid lobectomy and submandibular mass excision. The patient's wishes were taken into account and a difficult decision was made to perform one definitive procedure. The increased morbidity and mortality were taken into account, and balanced against the fact that the final diagnosis of ectopic submandibular thyroid tissue with normotopic multinodular goitre had been unreported previously.

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