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Mucoepidermoid carcinoma in a thyroglossal duct remnant

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ABSTRACT

INTRODUCTION: Thyroglossal duct cysts (TDC) are common midline neck swellings resulting from embryological remnants of the thyroglossal duct. They often contain ectopic thyroid tissue and malignant transformation has been reported, most commonly to papillary thyroid carcinoma. Mucoepidermoid carcinoma (MEC) usually occurs in the salivary glands and only rarely in the thyroid. This is the first case of a MEC occurring within a thyroglossal duct remnant.
PRESENTATION OF A CASE: A 73 year old lady presented with a thyroglossal duct cyst. She declined surgical excision, as she was adamant she wanted to avoid surgery. The neck mass rapidly enlarged at two years following initial diagnosis. Fine needle aspiration cytology was suspicious for carcinoma. She underwent total thyroidectomy and selective central compartment neck dissection with adjuvant radiotherapy. She remains alive and well two years post treatment.
DISCUSSION: Mucoepidermoid carcinoma is the most common malignant neoplasm of salivary glands, although it has rarely been reported in diverse locations including the thyroid, lung and pancreas. To the best of our knowledge, this is the first reported case of mucoepidermoid carcinoma arising from a thyroglossal duct remnant.
CONCLUSION: This case adds weight to the literature favouring surgical excision of thyroglossal duct remnants due to the risk of malignant transformation.

1. Introduction

Thyroglossal duct cysts (TDC) are common midline neck swellings. Usually presenting in childhood, they can be also be found in up to 7% of the adult population [1]. They develop in rests of epithelium sequestered in migration during the formation and disappearance of the thyroglossal duct. Islands of epithelium may persist throughout life as ectopic thyroid tissue but often undergo cystic degeneration to form thyroglossal duct cysts [2]. The commonest site for such cysts is around the body of the hyoid bone. Malignant transformation occurs in 1% of TDC and is usually of the papillary carcinoma type [3]. There are two theories for how this occurs, either the tumour arises from the malignant transformation of ectopic thyroid tissue retained within the duct, or arises de-novo from cells lining the duct [4]. Mucoepidermoid carcinomas are best characterised in salivary glands, rarely occurring in other glandular tissue including lung, lacrimal glands, and thyroid. Of the 40 cases of thyroid MEC in the literature to date the majority are low grade variants, for example, thyroid sclerosing mucoepidermoid carcinoma with stromal eosinophilia. High-grade mucoepidermoid carcinomas are found even less frequently in the thyroid gland. This is the first reported case of a mucoepidermoid carcinoma occurring within a thyroglossal duct cyst.

2. Case report

A 73-year-old lady of Bangladeshi origin initially presented to the endocrine surgeons with a two-year history of a gradually enlarging midline neck lump, which moved on tongue protrusion. There was no lymphadenopathy or abnormality within the thyroid gland itself, and thyroid function tests were normal. Additionally there were no suspicious clinical features; she was a non-smoker, with no history of alcohol consumption. Clinically, the neck lump was felt to be an uncomplicated thyroglossal duct cyst. Ultrasound scan showed a cystic lesion of 27 mm in maximal dimension, superior to and separate from the thyroid gland (Fig. 1), whilst fine needle aspiration cytology (FNAC), revealed features consistent with a thyroglossal duct cyst.

Surgical excision was recommended but the patient declined. She agreed to regular follow-up. The neck mass gradually enlarged over the course of 12 months, but the patient still declined surgery. Two years following her initial presentation, the neck mass rapidly

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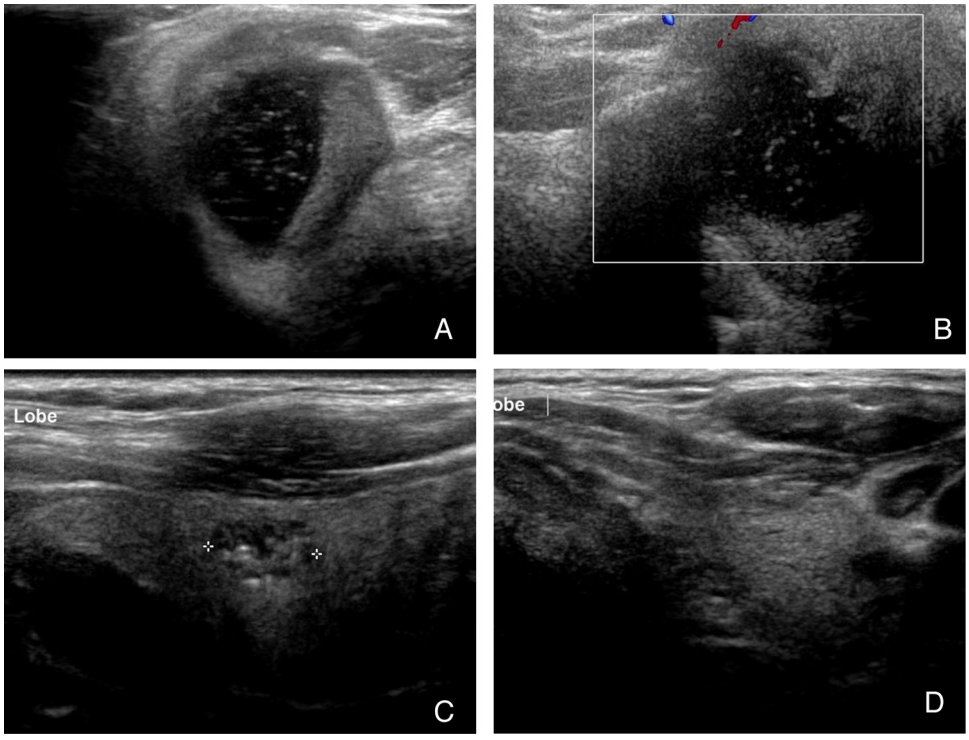

Fig. 1. Ultrasound images from initial clinic presentation.
A: Midline view showing central cystic lesion.
B: Doppler showing no evidence of increased vascularity.
C: Right lobe of the thyroid showing evidence of colloid cyst, otherwise normal appearances.
D: Left lobe of thyroid showing normal appearances of the gland.

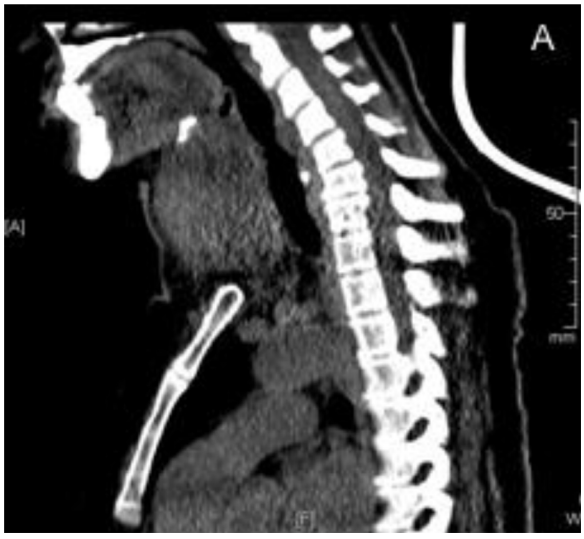

Fig. 2. CT scan of the neck (sagittal view).
A: Sagittal (non-contrast) view of the neck showing anterior midline neck mass extending from the suprasternal notch to the hyoid bone.

expanded in size and the patient was assessed urgently in the head and neck cancer clinic. At this time, a large hard mass with maximal dimension of 70 mm was palpable in the region of the thyroid gland. Clinically, there was no stridor or cervical lymphadenopathy. FNAC was highly suspicious for malignancy. A computed tomography (CT) scan of the neck confirmed the presence of a large anterior midline neck mass measuring, 50 mm × 70 mm × 66 mm, extending from the level of the thyroid to the hyoid bone (Fig. 2). CT scans of chest, abdomen and pelvis, did not show any evidence of distant metastases.

T2 weighted MRI axial (A) and coronal STIR (B) images demonstrating a 68 mm × 47 mm × 93 mm mixed T2 isointense/hyperintense infrahyoid lesion. It indents the superior thyroid gland and displaces the larynx and trachea posteriorly.

A T1/T2 weighted magnetic resonance imaging (MRI) of the face and neck with STIR and diffusion imaging was performed to evaluate the lesion further (Fig. 3). The mass measured 68 mm (long axial dimension) × 47 mm (short axial dimension) × 93 mm (cranio–caudal), extending inferiorly to the suprasternal notch, and superiorly to the level of the hyoid bone. A small volume of thyroid tissue was noted postero-inferior to the mass, and some slightly enlarged level IV lymph nodes (up to 13 mm).

Following discussion at the head and neck cancer multidisciplinary meeting, she underwent total thyroidectomy and level VI neck dissection, during which a partially necrotic tumour mass was excised (Fig. 4). Histological examination revealed a high-grade mucoepidermoid carcinoma above the gland consistent with origin in a thyroglossal duct remnant (Fig. 5). Six central compartment neck nodes were retrieved, with no evidence of metastasis. The carcinoma was investigated for MECT1/MAML2 (CRTC1/MAML2) translocation typical of mucoepidermoid carcinoma by fluorescence in-situ hybridisation using a MAML2 dual colour break apart probe (Zytovision GmBH, Bremerhaven, Germany). The carcinoma cells were negative for the translocation but showed a slightly increased copy number 2–4 throughout.

Apart from a return to theatre for evacuation of neck haematoma at one-week post surgery, she had an uneventful post-operative recovery. She underwent adjuvant radiotherapy (60 Gy in 30 fractions) in view of close surgical margins. She experienced radiotherapy related side effects including mucositis, which were managed conservatively with analgesia and steroids and is alive two years following completion of oncological treatment.