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Tall cell variant of papillary carcinoma arising from ectopic thyroid tissue in the trachea

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Abstract
Ectopic thyroid tissue within the submucosa of the trachea is a rare cause of upper airway obstruction. Primary neoplasms arising from such thyroid nests are rare. This report describes a case of tall cell variant of papillary carcinoma arising from ectopic thyroid tissue in the trachea.

Key words: Thyroid neoplasms; Trachea; Ectopic tissue

Introduction

True ectopic thyroid tissue in the trachea is a rare, but well-described abnormality, accounting for six to seven per cent of all primary endotracheal tumours (Fish and Moore, 1963). In 1875, Ziemssen described the first case of intratracheal goitre. The incidence of malignant change in intratracheal thyroid tissue is reported as high as 11 per cent (Dowling *et al.*, 1962). Papillary carcinoma is the most common type of cancer of the thyroid gland. It manifests a histomorphological heterogeneity, so that several variants are recognized. The usual form of papillary carcinoma generally follows a relatively indolent clinical course and has a good prognosis. However, the tall cell variant (TCV) of papillary thyroid carcinoma is associated with more aggressive behaviour and has a poor prognosis (Hawk and Hazard, 1976; Johnson *et al.*, 1988). A patient is presented with a tall cell carcinoma arising from ectopic thyroid tissue in the trachea.

Case report

A 64-year-old man of Australian origin was referred to the ENT clinic with nasal obstruction. Examination showed nasal polyposis and an elective nasal polypectomy was planned. On admission he was found to be in atrial fibrillation. Surgery was postponed and further investigations demonstrated thyrotoxicosis as the cause of fibrillation. Total thyroxin (T_4) level was 210 nmol/L (normal range 70–140 nmol/L) and thyroid-stimulating hormone (TSH) levels were <0.1 mU/L (normal value 0.25–5.8 mU/L). Anti-thyroglobulin antibodies were 1548 IU/L (normal value- less than 360 IU/L) and anti-microsomal antibodies were 1240 IU/L (normal value less than 154 IU/L). He was commenced on carbimazole on a gradually increasing dose and stabilized at a dose of 15 mg three times a day. Hyperthyroidism was controlled and he was readmitted six months later for nasal polypectomy. During this admission he was found to have mild stridor and admitted to recent episodes of haemoptysis. His chest X-ray was normal. Direct laryngoscopy and bronchoscopy were planned along with his nasal surgery. Multiple benign-looking nasal polyps were removed from both nostrils. Direct laryngoscopy was normal and on tracheoscopy a fleshy tumour was noticed arising from the right postero-lateral wall of trachea, which was occupying about 30 per cent of the tracheal lumen. The lesion was situated just above the carina and multiple biopsies were taken. Post-operatively the patient developed severe haemoptysis which necessitated blood transfusion. Bleeding continued and at this stage advice was sought from thoracic surgeons. He was advised to be transferred to a specialist centre for laser bronchoscopy. He was transferred to a chest unit where he underwent repeat bronchoscopy. Further biopsies were taken. Bleeding was controlled by conservative measures and a computed tomograph (CT) scan of neck and thorax was arranged. The scan showed a lesion in the tracheal lumen with no radiological evidence of spread outside the trachea. He had significant mediastinal lymphadenopathy. The biopsies were reported as papillary carcinoma. He underwent radiotherapy for his tracheal tumour. A repeat bronchoscopy was normal a few weeks after completion of radiotherapy. CT scan showed considerable reduction in the size of the mediastinal mass.

One year later the patient presented with a neck node to the Otolaryngology department. The lymph node was located in the mid-jugular region on the right side. His thyroid gland was normal on palpation and ultrasound scan did not reveal any focal lesion. Radio iodine uptake scan was normal. He underwent panendoscopy and excision of the neck node subsequently. Panendoscopy was normal and there was no evidence of recurrence of tumour in the trachea. Histopathological examination of the specimen showed evidence of papillary carcinoma (Figure 1). The original biopsy from the trachea was reviewed again along with the biopsies taken during his admission to the chest unit and was found to be identical to the lesion excised from the neck. Both the lesions showed oxyphil tumour with a striking papillary growth pattern. The tumour cells were tall, had eosinophilic cytoplasm (Figure 2), and showed brisk mitotic activity. Immunocytochemistry showed strong and widespread staining for thyroglobulin. These findings were suggestive of a more aggressive variant of papillary thyroid carcinoma – the tall cell carcinoma. At this stage an occult primary in the

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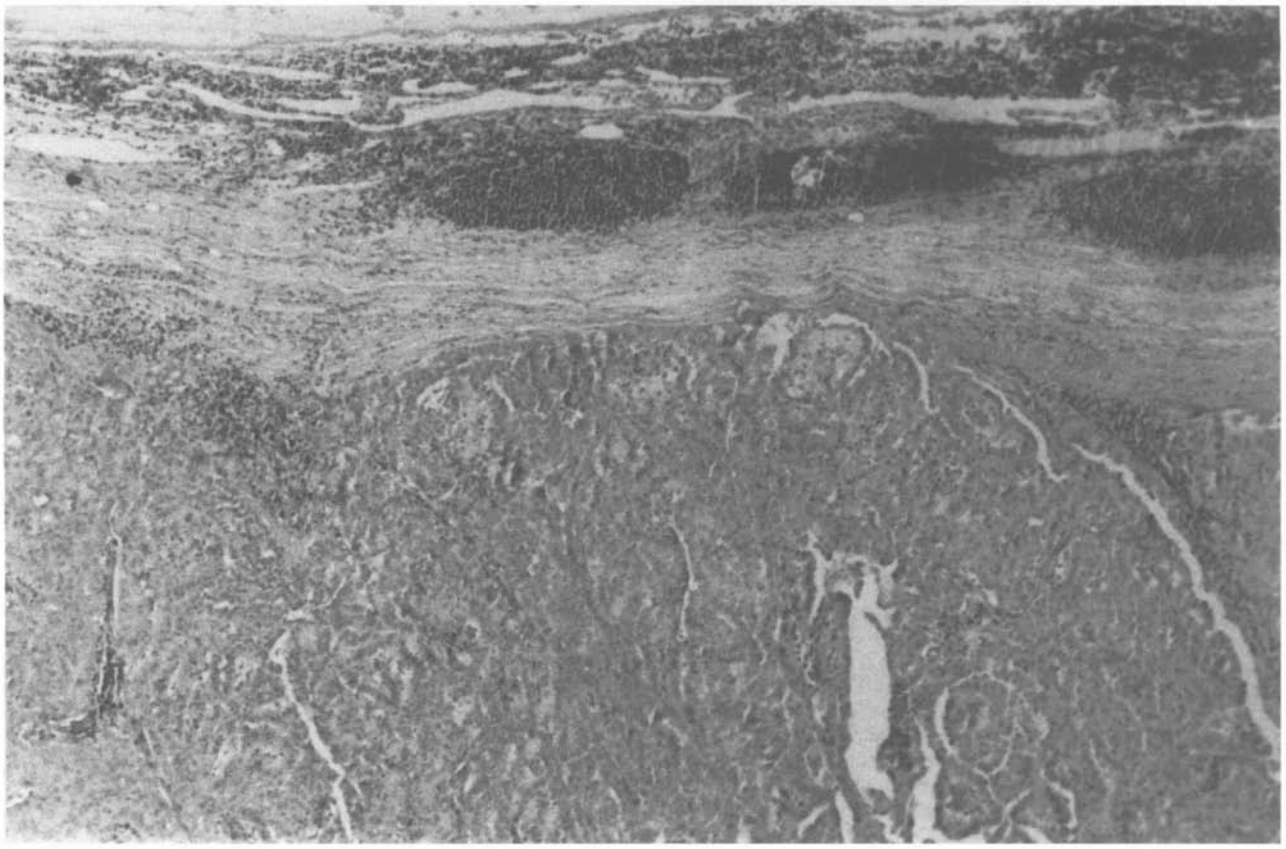

FIG. 1
Photomicrograph of lymph node showing metastatic papillary carcinoma. (H & E; × 100)

thyroid gland was suspected and a total thyroidectomy was performed. At surgery no connection of the thyroid gland to the trachea was identified and no tumour was identified in the paratracheal tissues. Serial sectioning of the thyroidectomy specimen and histopathological examination revealed no focus of occult thyroid carcinoma.

Four months later the patient was readmitted with haemoptysis and breathlessness. Bronchoscopy showed a recurrence of tumour in the trachea. CT scan of the neck and thorax showed that the lesion was extending outside the tracheal lumen and involving the superior mediastinum with multiple secondaries in both lung fields. The patient died of lung metastasis and respiratory failure.

Discussion

The occurrence of ectopic thyroid tissue is due to an abnormal migration of the thyroid during embryonic development. These ectopic masses may be encountered in four locations: the lingual area, sublingual (supra and infrahyoid) area, thyroglossal duct, and the intralaryngotracheal area. Of these sites the intralaryngotracheal area is the most rare area for ectopic thyroid. There is no completely satisfactory explanation for the genesis of intralaryngotracheal thyroid. There are two principal theories for such ectopic localization. One is that the thyroid gland, during embryonic life, is encountered and divided by the later developing tracheal cartilages and thyroid alae; the other is that there is a late foetal or postfoetal ingrowth of thyroid tissue into the tracheal lumen (Dowling *et al.*, 1962; Randolph *et al.*, 1963; Rotenberg *et al.*, 1979). Surgical observations favour the latter theory, since there is a rather consistent location of intralaryngotracheal thyroid masses in the posterior lateral wall of the airway. The posterior wall offers less resistance to the unencapsulated foetal thyroid than does the tighter, fibrous anterior submucosa.

Reports about malignancies in ectopic thyroids show that papillary carcinoma remains the commonest type (Potdar and Desai, 1971). Within the large group of papillary carcinoma, various subtypes have been identified. The variants include follicular, encapsulated, diffuse sclerosing, columnar cell, and tall cell types. An infrequent variant of papillary carcinoma, the tall cell carcinoma is associated with locally aggressive growth and a shortened survival of its host (Hawk and Hazard, 1976). Tall cell carcinoma was first described by Hawk and Hazard in 1976. It is defined as a papillary cancer of the thyroid in which a minimum of 30 per cent of the cells have a height at least twice their width, indicating a more aggressive pattern of growth (Johnson *et al.*, 1988). The incidence of this variant has been reported to be up to nine per cent of all papillary thyroid carcinomas. It tends to occur in older patients and to be large (>5 cm). Johnson *et al.* (1988) found a 75 per cent involvement of cervical lymph nodes, 42 per cent extrathyroidal extension rate, 58 per cent local recurrence, and a 17 per cent incidence of distant metastases. Hawk and Hazard (1976) reported a 22 per cent mortality, often within a few years of diagnosis. The carcinoma was lethal in 25 per cent of the patients reported by Johnson *et al.* (1988).

The reported patient initially presented with a tracheal exophytic lesion, which was thought to be a primary tracheal malignancy. Although this, on biopsy, was reported as a papillary carcinoma, presence of a normal thyroid, and lack of clinical suspicion led to initial treatment with radiotherapy. We feel that all clinicians should be familiar with this anatomical variation so that

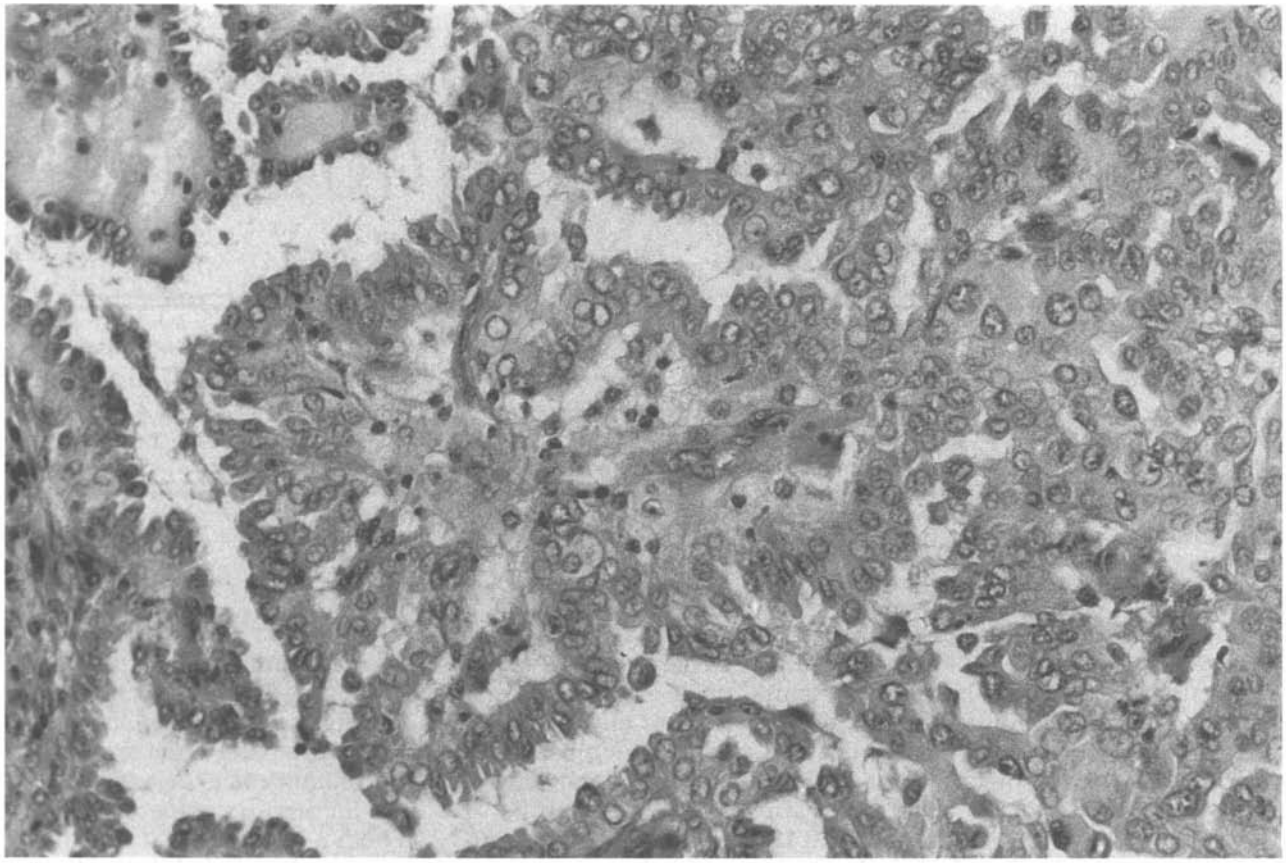

FIG. 2
Photomicrograph of tracheal biopsy showing papillary carcinoma, with tall cells and optically clear nuclei. (H & E; × 200)

undue delay in diagnosis is avoided. Retrospectively, we feel that surgical removal is the recommended form of management for malignant lesions arising from such ectopic thyroid tissue.

Conclusion

A rare combination of an aggressive tall cell carcinoma arising from ectopic thyroid tissue in the trachea is described. Intratracheal thyroid, although uncommon, should be considered in the differential diagnosis of tracheal masses. If there is ulceration and evidence of bleeding, malignancy arising from such ectopic tissue is a possibility. We recommend that if a biopsy is planned the surgeon must be prepared to cope with extensive bleeding and pre-operative consent should be obtained for definitive resection should it be necessary.

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