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View from Beneath—Pathology in Focus

Oncocytic papillary cystadenomatosis of the larynx

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Abstract

Symptomatic oncocytic disease of the larynx is rare. Review of the world literature reveals that isolated , symptomatic cases of oncocytoma of the larynx have been previously reported to involve discrete sites usually the laryngeal ventricles and vestibular folds. A unique case of multifocal cystic oncocytic hyperplasia necessitating laryngectomy is reported. CT scan of the larynx suggested destruction of the cartilage. Malignant histological features were not present.

This report raises the question whether previously described cases of oncocytoma of the larynx also had diffuse involvement. If oncocytosis is a diffuse process then it is suggested that patients should be kept under review for recurring lesions.

Introduction

Oncocytes are granular, intensely eosinophilic, swollen cells that replace ductal and acinar epithelium with age. Histochemical staining techniques reveal high levels of mitochondrial oxidative activity. Under electron microscopy the cytoplasm is packed with hypertrophied granular mitochondria with little remaining space for endoplasmic reticulum or secretory apparatus.

They were first described by Schafer in 1897 and later designated the term 'oncocyte' by Hamperl in 1931. MacFarland (1927) first described a tumour entirely composed of oncocytes. Oncocytes are found in many mature tissues including the salivary and seromucinous glands of the upper aerodigestive tract. (Gray *et al.*, 1976). They appear with increasing frequency in ageing individuals and rarely can form the predominant component of cysts and tumours.

Review of the world literature reveals only 142 cases of reported oncocytic lesions of the larynx. A unique further case of extensive, diffuse, multifocal, cystic, oncocytic hyperplasia of the larynx necessitating laryngectomy is described.

Case report

A 74-year-old gentleman presented with a two month history of hoarseness. Indirect laryngoscopy showed paralysis of the left

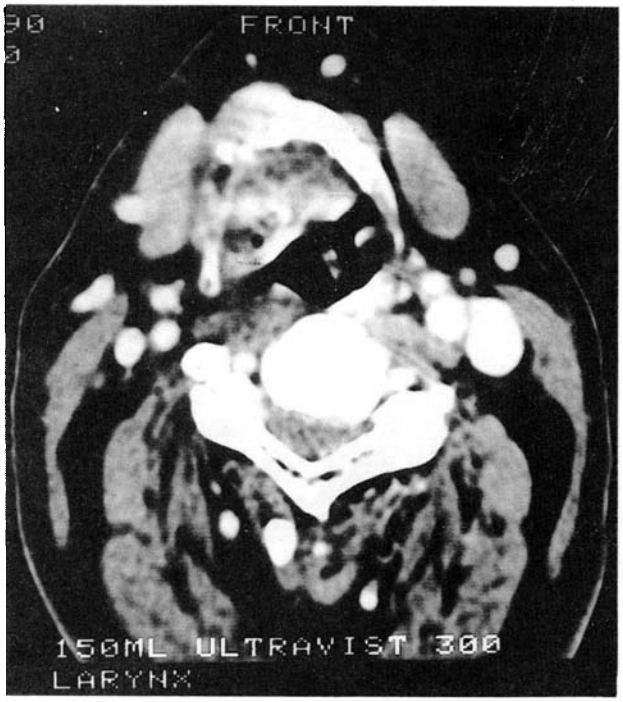

Fig 1(a)
Axial CT scan showing tumour-like mass and destruction of right hyoid bone.

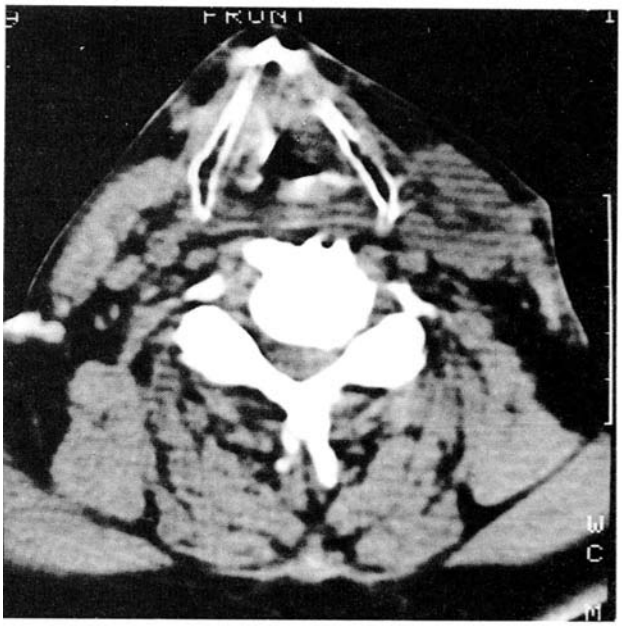

Fig 1(b)
Axial CT scan showing destruction of the arterior left thyroid lamina.

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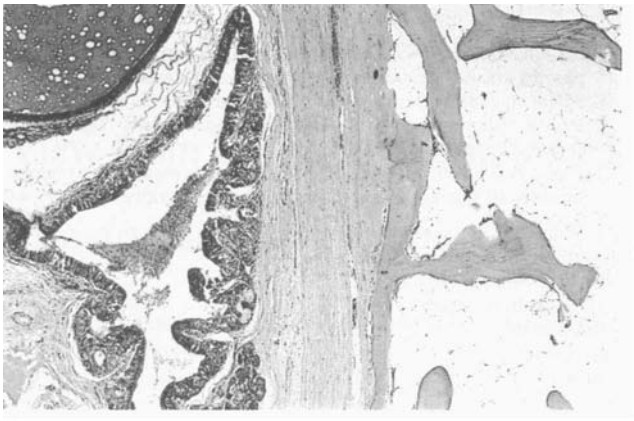

FIG 2(a)
Oncocytic cysts adjacent to ossified thyroid lamina. H & E.

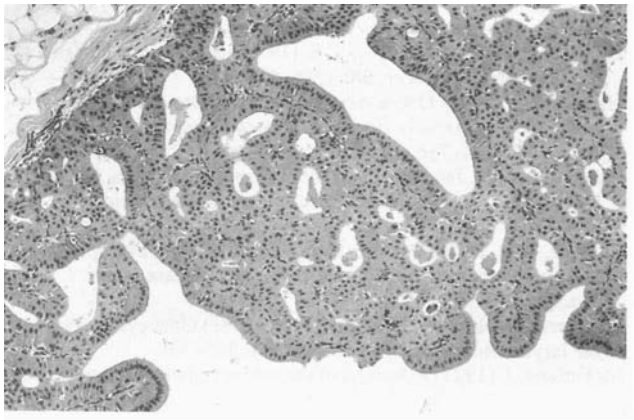

FIG 2(b)
Oncocytic epithelium with cystic spaces, and bland cells showing no malignant cytological features. H & E.

vocal fold and a bulky left vestibular fold. Chest X-ray and laryngeal tomography were normal. Four microlaryngoscopies and biopsies over a period of six months showed no evidence of neoplasia. The patient's symptoms however relentlessly progressed. A CT scan of the larynx (Fig. 1a) showed soft tissue swelling and destruction of the anterior part of the left lamina of the thyroid cartilage. Further laryngoscopy revealed irregularity of the ventral surface of the epiglottis and serous exudation from the anterior left vestibular fold. Multiple deep biopsies revealed a diffuse, oncoytic, cystic metaplasia. The patient was treated conservatively but deteriorated clinically over a period of eight months. Further CT revealed significant progression of the disease (Fig. 1b). In addition to the left-sided pathology, the epiglottis was involved with destruction of the upper part of the right hyoid bone. The right aryepiglottic fold was thickened and the right pyriform sinus partially obliterated. There was now destruction of the right thyroid lamina. Clinically the disease was progressing in a malignant fashion and therefore total laryngectomy was performed. The laryngectomy specimen showed oedematous and nodular aryepiglottic folds, thickened and distorted vocal folds, and replacement of the fatty pad beneath the epiglottis by a spherical mucoid multiloculated mass 2 cm in diameter.

Histology of the mucoid mass showed numerous large, cystic spaces lined by oncocytic epithelium with some papillary infoldings. Similar small oncocytic cysts were present in the stroma of each aryepiglottic fold, within the stroma of each vocal fold, and at the base of the epiglottis. The oncocytic epithelium was composed of bland cells with regular oval nuclei and strikingly eosinophilic cytoplasm. The oncocytic epithelium blended in areas with unremarkable respiratory epithelium of seromucinous glands, and the oncoytes showed no cytological characteristics of malignancy (Figs. 2a & b).

The patient made good recovery and is disease free three years after surgery.

Discussion

The incidence of symptomatic oncoyctic lesions of the larynx has been found to be in the order of 0.5 per cent to 1 per cent. (Busuttil, 1976; Lundgren *et al.*, 1982). However, De Santo *et al.* (1970), reviewed all laryngeal specimens from the Mayo Clinic over a 20 year period and found the incidence of all laryngeal cysts lined with oncocytes to be 11 per cent. This discrepancy might be attributed to the fact that in many cases of oncocytosis of the larynx the disease is clinically inconspicuous. Most research has centred around oncocytosis of the major salivary glands. Several microscopic surveys of the parotid glands have shown that oncocytic foci are almost universal over the age of 70. (Meza-Chavez, 1949).

Oncocytic laryngeal lesions most commonly affect the ventricles and vestibular folds as these are rich in seromucinous glands. Unusually, in our case oncocytic cysts were found in the stroma of the vocal folds which are devoid of any glands. It is likely that these lesions arose in adjacent excretory ducts (Holm-Jenson *et al.*, 1977).

Reports describe the lesions to be macroscopically a discrete single cyst or a small cluster of cysts with solid lesions being extremely rare. (Le Jeune *et al.*, 1980). Only one report (Yamase and Putman, 1979) has described a similar diffuse oncocytic processof the laryngeal mucosa as in our case but as a coincidental finding in a patient who underwent laryngectomy for squamous cell carcinoma.

The CT scan findings of destruction of the laryngeal cartilaginous structures suggested malignancy. This was not born out histologically. Although the oncoytic cysts appeared to compress and distort the normal laryngeal structures no overt malignant invasion was demonstrated. The likely pathogenesis is one of retention cyst formation, degeneration of the epithelial cells into eosinophilic, swollen oncocytes and pressure necrosis. In the larynx no qualified example of a malignant oncocytoma has been reported. (Batsakis, 1979).

Most patients with oncocytic lesions of the larynx present with hoarseness although a small number complain of dyspnoea, stridor, pain or dysphagia. (Lundgren *et al.*, 1982). Most authors have found a female preponderence of approximately 60 per cent to 40 per cent. (Bell *et al.*, 1978; Lundgren *et al.*, 1982). Patients present in the sixth or seventh decade.

The treatment of choice is local surgical excision either by endoscopic or open methods. The unusual extensive diffuse nature of this case warranted laryngectomy.

As there was multifocal involvement of the laryngeal mucosa in this case, the term oncocytic papillary cystadenomatosis is more appropriate than oncocytoma. Previous documents have only centred on discrete lesions that have caused symptoms. It is suggested that in many of these cases a diffuse process is probable and that long-term follow-up for recurrence is essential.

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