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Giant cell reparative granuloma of the cricoid cartilage

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Abstract
Giant cell reparative granuloma (GCRG) is an uncommon benign lesion which has been reported at several sites in the head and neck. We present a case of a GCRG of the cricoid cartilage not previously described in the literature. It must be differentiated from the brown tumour of hyperparathyroidism and true giant cell tumours of bone. These were excluded on clinical, biochemical, radiological and histological grounds. The lesion responded well to surgical debulking and curettage and the patient remained disease-free 15 months after treatment.

Key words: Laryngeal neoplasms; Cricoid cartilage; Granuloma, giant cell

Introduction

Giant cell reparative granuloma (GCRG) is an uncommon benign lesion. The term was first coined by Jaffe (1953) to describe benign reactive intraosseous lesions occurring in the maxilla and mandible.

It has since been described affecting other regions of the head and neck including the paranasal sinuses (Friedberg *et al.*, 1969; Wiatrak *et al.*, 1987), nasal septum (Amin and Samuel, 1990; Govett and Amedee, 1991), temporal bone (Hirschl and Katz, 1974; Tesluk *et al.*, 1989), sphenoid sinus (Rogers *et al.*, 1984; Alappat *et al.*, 1992), skull base and cranial vault (Garza-Mercado *et al.*, 1984; Ciappetta *et al.*, 1990).

Lesions occurring outside the head and neck and jaw bones were first reported by Ackerman and Spjut (1962) who described two cases involving the small bones of the hand. Since then over 100 cases of GCRG have been described in various sites outside the skull and facial bones (Jernstrom and Stark, 1971; D'Alonzo *et al.*, 1972; Lorenzo and Dorfman, 1980; Glass *et al.*, 1983; Ratner and Dorfman, 1990).

Accurate diagnosis depends on the exclusion of similar lesions including the brown tumour of hyperparathyroidism and giant cell tumour of bone.

We present an unusual case of a GCRG affecting the cricoid cartilage, which has not been described previously.

Case report

A 56-year-old female teacher, presented with a two-week history of hoarseness and shortness of breath on exertion. There were no other relevant symptoms: she was a non-smoker, denied excessive alcohol consumption and there were no known allergies. Her past medical history was unremarkable and there was no relevant family history.

Physical examination showed a well nourished female with a hoarse voice and mild biphasic stridor. Head and neck examination was normal with the exception of an oedematous subglottis. There was no cervical lymphadenopathy or thyromegaly and the remainder of the physical examination was normal.

A provisional diagnosis of tracheobronchitis was made and she was commenced on antibiotics and steroids. When reviewed one week later, her symptoms had only slightly improved. Repeat fibreoptic endoscopy showed a smooth circumferential swelling immediately beneath the vocal folds: the overlying mucosa was intact. Computer tomography (CT) confirmed these findings, showing marked subglottic narrowing, with a soft tissue mass predominantly on the right, but also extending posteriorly and to the left. Despite some erosion and widening of the cricoid cartilage it appeared to be an entirely intrinsic lesion (Figure 1).

The patient underwent direct laryngoscopy and biopsy of the lesion. A tracheostomy was performed to protect the airway. Attempted decannulation was unsuccessful and the patient was returned to theatre and the larynx explored via the tracheostomy incision. There was no evidence of any extralaryngeal disease. True-cut biopsies of the lesion were taken.

The patient was referred to St Mary's Hospital, for further management. Laboratory studies including serum calcium, phosphate, alkaline phosphatase and parathyroid hormone assay were normal. A plain chest X-ray and lateral skull showed no abnormal features, radiographs of her hands showed no evidence of hyperparathyroidism or erosive arthropathy.

The lesion was exposed via a laryngofissure approach splitting the thyroid and cricoid cartilages vertically. This revealed normal vocal folds and a circumferential subglottic lesion confined to the cricoid cartilage. The intact mucosa was stripped away to reveal a soft, haemorrhagic, friable mass which was removed piecemeal by curettage. Specimens were sent for histology and microbiology. A laryngeal stent was inserted, extending from above the vocal folds to beneath the excision margin of the lesion. Microbiological examination of the lesion did not show the

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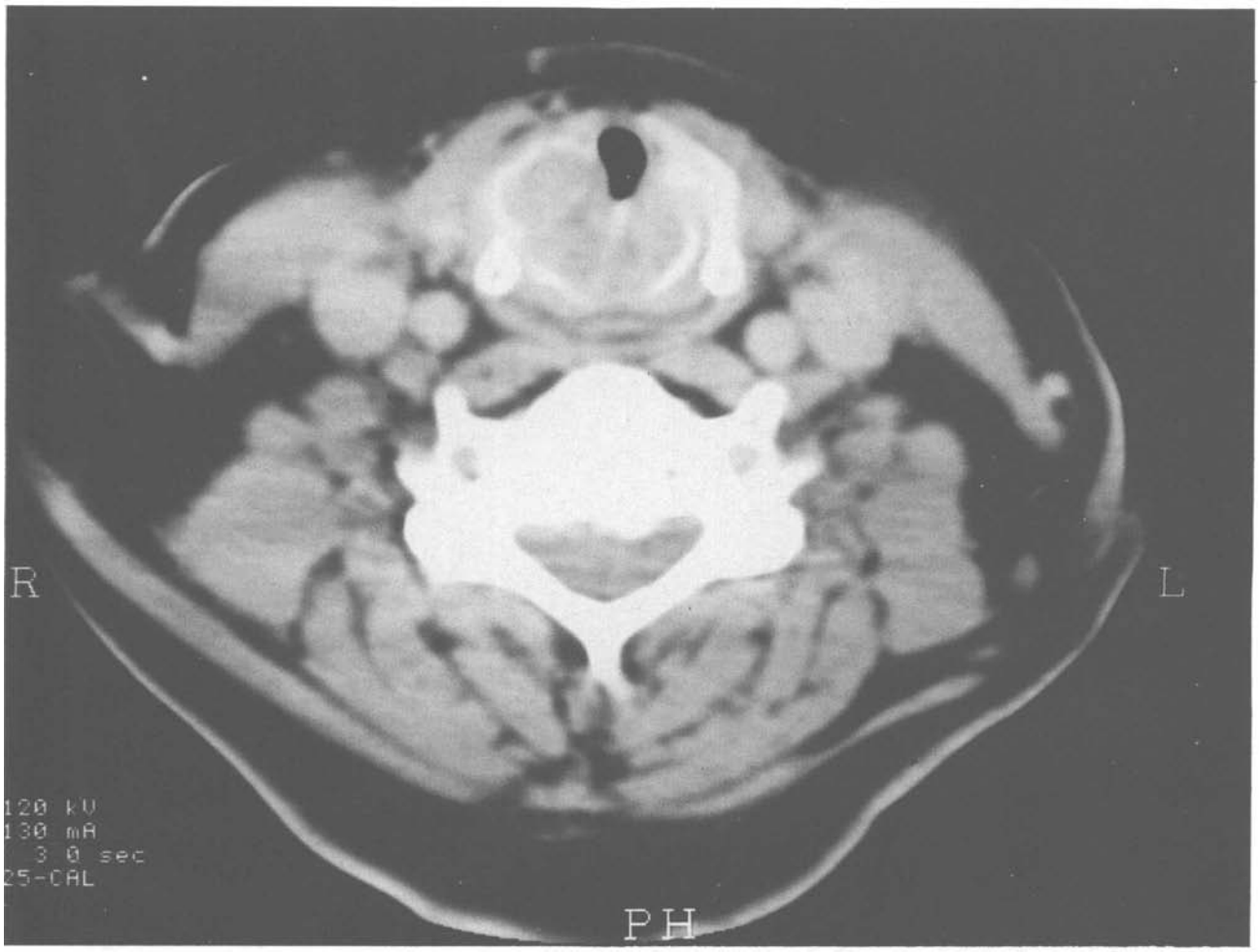

FIG. 1

Axial CT scan at the level of the cricoid cartilage, showing marked subglottic narrowing, with a soft tissue mass predominantly on the right but extending posteriorly and to the left.

presence of any organisms, culture was sterile, acid alcohol-fast bacilli were not grown.

Post-operative recovery was unremarkable, the laryngeal stent was removed on the seventh post-operative day, laryngoscopy showed oedematous vocal folds but a patent subglottis. The tracheostomy was left *in situ*, as the decision to give post-operative radiotherapy had been made.

The patient received external beam radiotherapy (parallel opposed beams) to the larynx and adjacent neck: 6000 cGy were administered in 30 fractions over 43 days, using 6 MV photons. The tracheostomy was removed following the completed course of radiotherapy.

Close follow-up occurred over 15 months: she remained well, the hoarseness had settled and there was no stridor. Repeat fibre-optic endoscopy showed a satisfactory subglottis.

Histopathology

The specimen consisted of multiple fragments of brown tissue, in total measuring 2 × 1 × 1 cm. Histological examination revealed a mass composed of spindle cells of the fibroblastic type. The stroma was vascular and focally there were aneurysm-like formations. Osteoclast-like multinucleated giant cells were irregularly distributed in the lesion. Scattered normal mitotic figures were present in the spindle cell population and there was a mild infiltrate of lymphocytes. There was focal deposition of reactive osteoid (Figure 2).

Although isolated high power fields were indistinguishable from giant cell tumour of bone, the overall appearance with the irregularly distributed giant cells favoured a diagnosis of giant cell reparative granuloma. A review of the histology observed at laryngeal exploration via the tracheostomy showed similar features.

Discussion

The term GCRG was first coined by Jaffe (1953) to describe a benign non-neoplastic lesion mostly affecting the mandible and maxilla. The lesion mainly occurs in children and young adults (Waldron and Shafer, 1966) being commoner in females. In one series 20 per cent of the patients were over the age of 50 years (Waldron and Shafer, 1966). It has now been described in several extragnathic sites confined to the head and neck including the paranasal sinuses (Friedberg *et al.*, 1969; Wiatrak *et al.*, 1987), nasal septum (Amin and Samuel, 1990; Govett and Amedee, 1991), temporal bone (Hirschl and Katz, 1974; Tesluk *et al.*, 1989), sphenoid sinus (Rogers *et al.*, 1984; Alappat *et al.*, 1992), skull base and cranial vault (Garza-Mercado *et al.*, 1984; Ciappetta *et al.*, 1990). There have been several reports of GCRG affecting other parts of the body (Ackerman and Spjut, 1962; Jernstrom and Stark, 1971; D'Alonzo *et al.*, 1972; Lorenzo and Dorfman, 1980; Glass *et al.*, 1983; Ratner and Dorfman, 1990).

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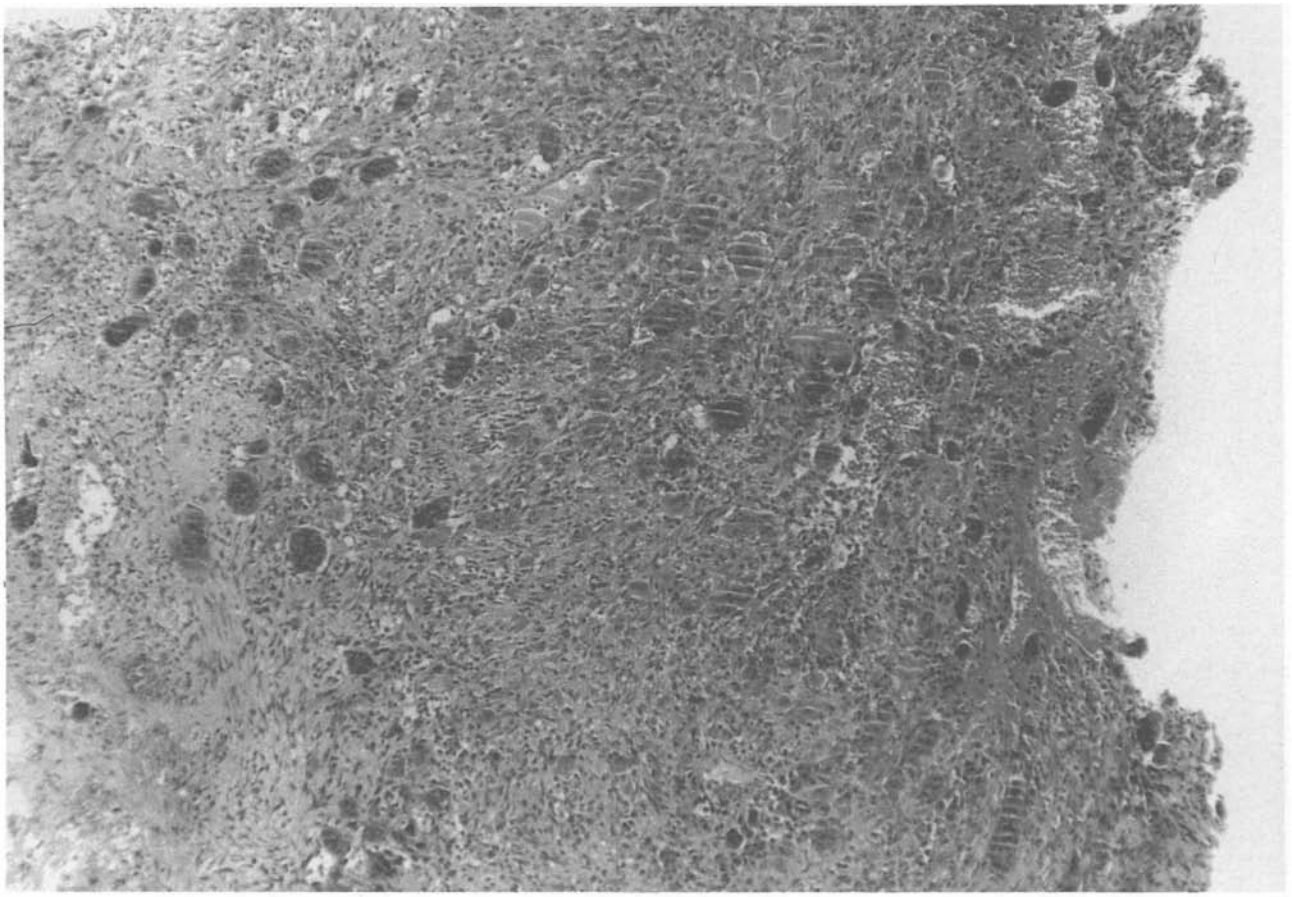

FIG. 2

Giant cell reparative granuloma showing spindle cells and irregularly distributed osteoclast-like multinucleated giant cells. (H & E; × 340).

The aetiology of GCRG is controversial. Jaffe (1953) proposed that it was a hyperplastic-reparative response to injury, a history of trauma in the past can be traced in many cases. Others believe GCRG to be the result of a chronic inflammatory and/or infective process that gives rise to a microhaemorrhage which triggers a reactive process (Katz and Hirschl, 1974).

Histologically the lesion is characterized by a fibrous connective tissue background with scattered multinucleated giant cells and immature fibroblasts. Typically the lesion is vascular and there is focal haemosiderin deposition with osteoid and new bone formation. The histological differential diagnosis includes giant cell tumour (osteoclastoma) and the bone lesion of hyperparathyroidism (brown tumour).

True giant cell tumours rarely occur in the head and neck, though their occurrence is well recognized; the majority have involved the sphenoid and temporal bone (Spjut *et al.*, 1970; Epstein *et al.*, 1982). The lesion is commoner in the third to fourth decades and shows no sexual preponderance. There are no specific biochemical markers but it has a recognized malignant potential and a high rate of recurrence after surgery and radiotherapy (Austin *et al.*, 1959; Hutter *et al.*, 1962; Spjut *et al.*, 1970). An isolated case of a giant cell tumour of the cricoid cartilage has been reported (Badet *et al.*, 1992) and in keeping with the conventional treatment of giant cell tumours the authors recommended wide local excision performing a total laryngectomy. Histologically true giant cell tumours consist of stromal cells and giant cells. The regular distribution of multinucleated giant cells seen throughout the lesion contrasts with the uneven distribution seen in GCRG.

Hyperparathyroidism was excluded on clinical, biochemical and radiological grounds. Repeat estimations of serum calcium, phosphate, alkaline phosphatase and parathormone assay were normal. There was no radiological evidence of bony resorption. The histological appearance of a brown tumour of hyperparathyroidism may closely resemble GCRG.

The treatment of GCRG is based on surgical debulking and curettage (Austin *et al.*, 1959). Due to the unusual site of the lesion and poor surgical access, with a desire to maintain laryngeal function, thorough surgical curettage was followed by external beam radiotherapy. This form of treatment has been used with a considerable degree of success (Katz and Hirschl, 1974; Garza-Mercado *et al.*, 1984). The authors accept that there is a long-term risk of malignant transformation (Quick *et al.*, 1980).

The recognition of GCRG has, therefore, important therapeutic and prognostic implications. The need for a total laryngectomy (the treatment of choice for true giant cell tumours) can be avoided. Local recurrence of GCRG is common in extra-gnathic sites but treatment need not be more aggressive than that of the primary lesion. Its course is almost invariably benign and progressive local spread or metastatic disease has not been reported.

We believe this to be the first reported case of GCRG arising in the cricoid cartilage. The patient remained well and disease-free 15 months after treatment.

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