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Pathology in Focus

Post-irradiation carcinosarcoma of the parotid gland

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Abstract
Carcinosarcoma of the salivary glands are rare tumours, often associated with a history of pleomorphic adenoma. A case of carcinosarcoma of the parotid arising following irradiation to the resection site of a pleomorphic adenoma is presented. The clinical and histological features are discussed and the literature reviewed.

Key words: Parotid neoplasms, pleomorphic adenoma; Carcinosarcoma; Radiotherapy

Introduction

Carcinosarcoma (true malignant mixed tumour) is a tumour in which both the stromal and epithelial components fulfil histological criteria for malignancy and which invariably metastasize together. Carcinosarcomas of the salivary glands are rare tumours and are associated with a history of pleomorphic adenoma in one-third of cases (Gnepp and Wenig, 1991). Two previously reported cases occurred following radiotherapy to recurrent pleomorphic adenoma (Hellquist and Michaels, 1986) and one case arose within a carcinoma ex pleomorphic ade-

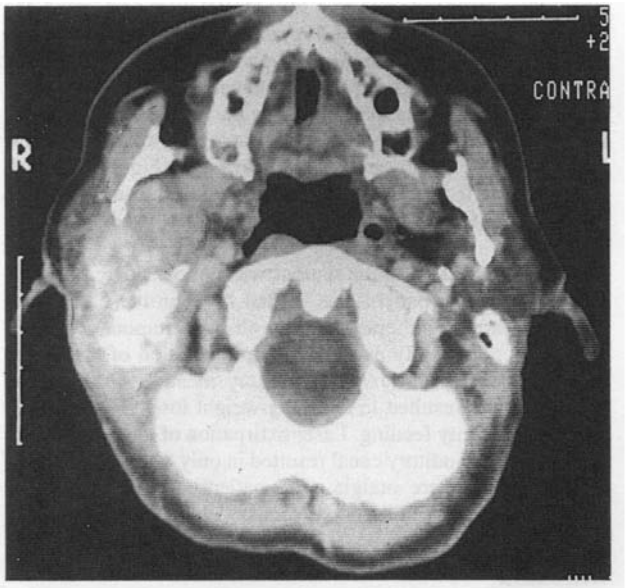

Fig. 1
Axial CT scan with contrast shows extension of the parotid mass into the parapharyngeal space. The tumour surrounds the internal carotid artery and the internal jugular vein and shows central necrosis and calcification.

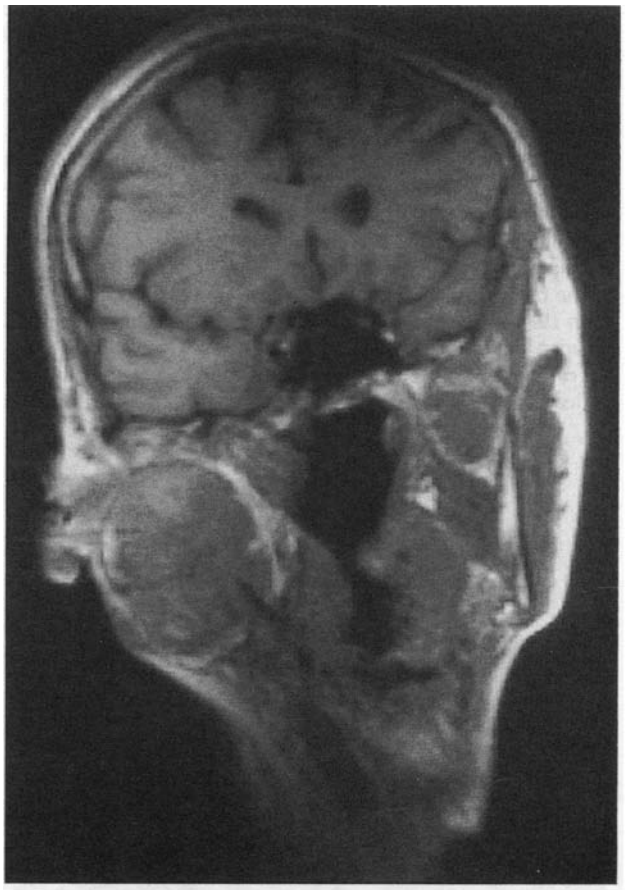

Fig. 2
Coronal T1-weighted MRI scan demonstrating the extent of the mass. Note the infiltration of the mass into the temporal bone.

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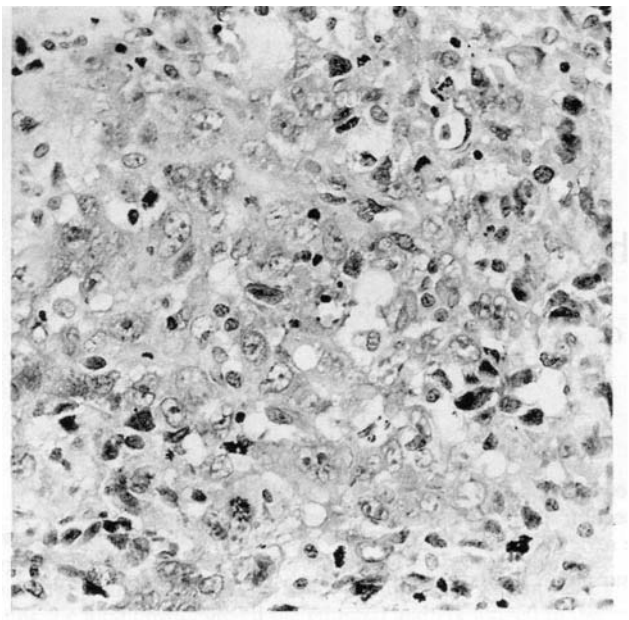

FIG. 3
Sheets of undifferentiated carcinoma (H & E; × 400).

noma after radiation (Suzuki *et al.*, 1990). We report a carcinosarcoma occurring many years after radiation to the resection site of a parotid pleomorphic adenoma.

Case report

A 61-year-old lady had a superficial parotidectomy in 1954 for a pleomorphic adenoma of her right parotid gland. In the subsequent months a keloid scar developed in the cervical and postauricular limbs of the incision for which she received a short course of radiotherapy. The details of the dose, fields and nature of the radiotherapy were not available.

In 1981 she developed a further lump in the right parotid and an open biopsy of the lesion was performed. The histopathological evaluation showed recurrent pleomorphic adenoma and the patient was subsequently advised to have the lump excised. The

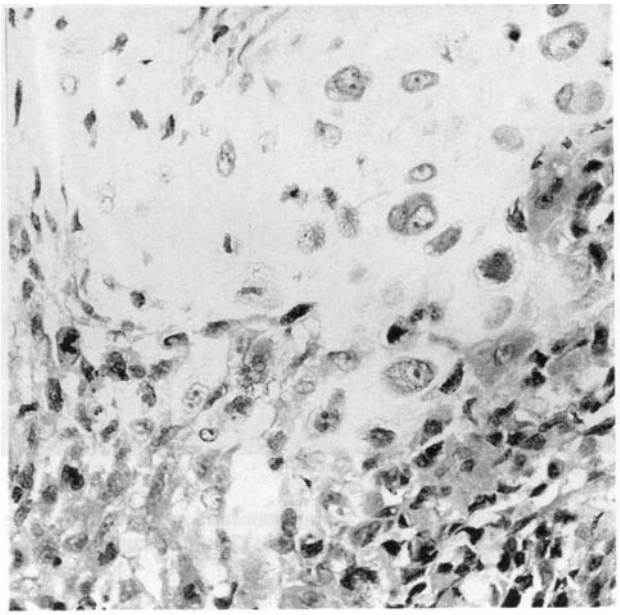

FIG. 4
Showing foci of chondrosarcoma are present. (H & E; × 400).

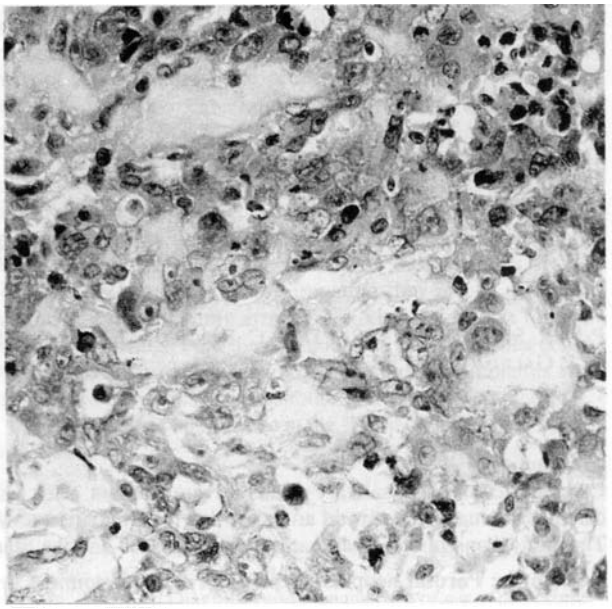

FIG. 5
Osteoid formed directly from malignant cells. (H & E; × 400).

patient however declined further surgery and did not attend follow-up appointments.

The lump remained unchanged for another 10 years until 1991 when there was a dramatic increase in size of the lesion and the patient developed a facial palsy. A fixed indurated mass measuring 5 × 4 cm was found to arise from the preauricular region of the parotid and to extend to the parapharyngeal space and to cause an impression in the lateral wall of the pharynx. The tumour extended into the temporal bone and infiltrated the skin of the external auditory canal where it caused haemorrhagic polyps and granulations. A computed tomography scan (CT) and a magnetic resonance imaging scan (MRI) were performed, the features of which are seen in Figures 1 and 2.

Further open biopsy showed atrophic parotid gland infiltrated by carcinosarcoma. The epithelial element, consisting of sheets of undifferentiated carcinoma (Figure 3), showed positive immunostaining for low molecular weight cytokeratin (CAM 5.2). Foci of chondrosarcoma were present (Figure 4) which were positive for S100 antigen and areas of osteosarcoma were present also (Figure 5). No residual pleomorphic adenoma was seen.

Surgical resection of the tumour was not considered feasible and therefore the patient was treated by a further 55.8 Gy of external beam radiotherapy. Despite this, the tumour continued to enlarge relentlessly and resulted in ulceration of the oropharynx, neck skin and external auditory meatus. Anorexia and severe trismus resulted in alarming weight loss which necessitated gastrostomy feeding. Laser extirpation of the granulations in the external auditory canal resulted in only transitory relief of the patient's severe otalgia. The patient's condition rapidly deteriorated and she died in November 1992.

Discussion

Carcinosarcomas represent approximately one per cent of all malignant salivary gland tumours (Gnepp and Wenig, 1991). The age at presentation ranges from 14–87 years and there is no sex predominance. An enlarging mass or a rapid increase in size of a pre-existing mass is the most common clinical presentation. There may be pain in the face or a facial palsy or symptoms as a result of local or distant metastases.

As in this patient, in the two previously reported cases of carcinosarcoma following irradiation to pleomorphic adenoma the interval between irradiation and subsequent development of car-

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cinosarcoma was many years (36 and 30 years) respectively (Hellquist and Michaels, 1986). Suzuki *et al.* (1990) have however, described a case in which carcinosarcoma was diagnosed within a year of radiation therapy to be a carcinoma ex pleomorphic adenoma.

Typically, the mesenchymal elements, most often chondrosarcoma, dominate the tumour. The epithelial component is usually high grade adenocarcinoma or undifferentiated carcinoma. Hellquist and Michaels (1986) demonstrated co-positivity for cytokeratin and S100 in both mesenchymal and epithelial elements, with no staining for vimentin, leading them to postulate that carcinosarcoma arises from divergent differentiation from a myoepithelial precursor. Our immunohistological findings are in accord with those of Bleiweiss *et al.* (1992) who found keratin and vimentin positivity only in carcinomatous and sarcomatous elements respectively, with no expression of S100 antigen in their case of osteosarcomatous carcinosarcoma. This supports the view that some carcinosarcomas, at least, do not show evidence of myoepithelial cell origin.

Dawson and Orr (1985), in a study of patients treated for pleomorphic adenomas by irradiation, with or without surgery, found a (pure) sarcoma in one out of 193 patients followed-up for 10–15 years, and an adenocarcinoma in three out of 100 patients followed-up for 16–20 years. Conversely, Van Miert *et al.* (1968) had no malignant recurrences in the 62 patients they followed-up for 16–30 years.

Irradiation as an adjunct to surgery for pleomorphic adenoma was common until the 1960s and although it would seem that the risk of carcinosarcoma developing after radiation is small, more large studies with greater than 30 years follow-up are required to define the risk accurately.

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