Widespread metastases from acinic cell carcinoma of parotid gland

Vidyadhara S, Shetty AP, Rajasekaran S

ABSTRACT

Acinic cell carcinoma metastasising to the spine is rare and has been described only once before in the literature. Its metastasis to other organs such as the regional lymph nodes, lungs and orbit have all been described, but the simultaneous occurrence of lymph nodal, pulmonary and spinal spread in a single patient has not been recorded. We believe this 40-year-old man to be the first reported case of incompletely resected acinic cell carcinoma of the parotid gland metastasising simultaneously to regional lymph nodes, upper lobes of both lungs, sphenoid bone and dorsal spine with neurological deficits. This case report stresses the need for postoperative radiotherapy following incomplete resection of the primary tumour. A careful watch for distant metastasis is also of paramount importance at follow-up.

Keywords: acinic cell carcinoma, myelopathy, parotid gland, parotid neoplasm, spinal metastasis

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INTRODUCTION

The acinic cell tumour is a rare neoplasm that accounts for about 1% of all salivary gland neoplasms[1-3]. It was considered benign for a long time, until it was grouped in 1953 among solid parotid tumours with varying degree of malignancy. This tumour is presently regarded as of low-grade malignancy. The term "acinic" refers to the resemblance of the tumour cells to acinar gland cells. Treatment of acinic cell carcinoma includes complete surgical excision. Postoperative radiation therapy may be helpful in cases of questionable margins or residual disease after surgery[4,5].

Unlike other tumours of the parotid or of other salivary glands, skeletal metastasis is rare with acinic cell carcinoma. It is very uncommon for this tumour to spread to the spinal column, compared to other areas[6-10]. The case reported is a young adult with incomplete excision of the acinic cell carcinoma of parotid gland presenting with local recurrence and widespread metastatic deposits in regional lymph nodes, both lungs and also thoracic spine with neurological deficits. To our knowledge, this is the first reported case of acinic cell carcinoma with spinal secondaries producing neurological deficits.

CASE REPORT

A 40-year-old man presented with complaints of sudden-onset severe back pain and girdle pain radiating bilaterally anteriorly up to the nipples. Patient had exacerbation of pain on turning to the sides, attempted sitting and standing. The excruciating pain persisted even at night, disturbing his sleep. The patient had tingling and numbness of both lower limbs as well as the trunk. He had spasticity and grade IV muscle power in both lower limbs. There was no involvement of bowel or bladder. The patient had been operated for acinic cell carcinoma of the right parotid gland four months previously, with incomplete resection of the deep lobe of the parotid owing to severe adhesions.

On examination, there was tenderness over the prominent spinous process of T4 vertebra. Patient had features suggestive of thoracic myelopathy with upper motor neuron type of neurological involvement and sensory blunting below the level of T4 bilaterally for all modalities of sensation. The radiographs were apparently normal but magnetic resonance (MR) imaging revealed a mass lesion of the posterior two-thirds of T4 vertebra, more on the right side. There was evidence of epidural soft tissue component in the spinal canal, producing cord compression (Fig. 1). Computed tomography (CT) revealed local recurrence of the tumour, with dissemination into the deep cervical lymph nodes, upper lobe of both lungs and spinal metastasis to T4 vertebra (Fig. 2). There was increased uptake in the thoracic spine on bone scan.

He underwent posterior decompressive laminectomy of T4 vertebra and pedicle screw instrumentation of T3–T5 vertebrae. Histopathological examination was conclusive of metastatic acinic cell carcinoma (Fig. 3). Postoperatively, the patient had rapid improvement in neurological and instability symptoms. He was mobilised after the third postoperative day with a brace and walker, and was comfortable. He was followed-up by

Department of Spine Surgery, Ganga Hospital, Swarnambika Layout Ramnagar, Coimbatore 641009, Tamil Nadu, India

Vidyadhara S, MS, DNB
Fellow

Shetty AP, MS, DNB
Consultant

Rajasekaran S, MCh, PhD, FRCS
Director and Head

Correspondence to:
Dr S Rajasekaran
Tel: (91) 422 223 5050
Fax: (91) 422 223 2652
Email: sr@gangahospital.com

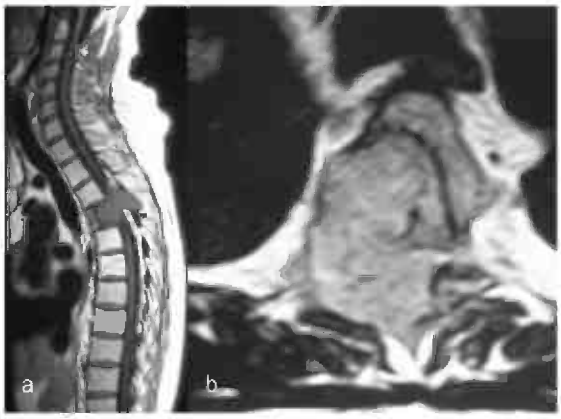

Fig. 1 Preoperative (a) sagittal and (b) axial T1-W MR images shows spinal metastasis to T4 vertebra with cord compression.

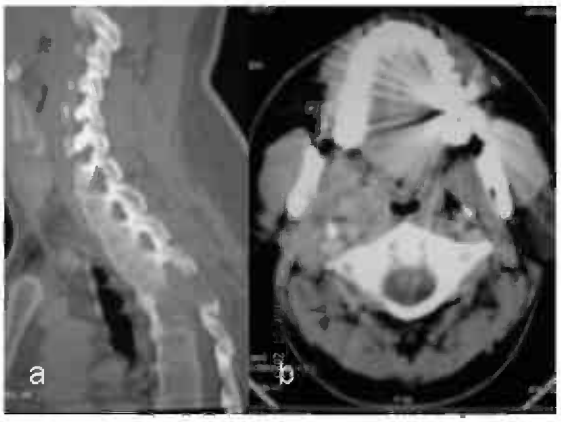

Fig. 2 Preoperative (a) sagittal and (b) axial CT images show local recurrence and widespread metastasis. The axial image taken through the parotids shows local recurrence and regional cervical lymph nodal metastasis.

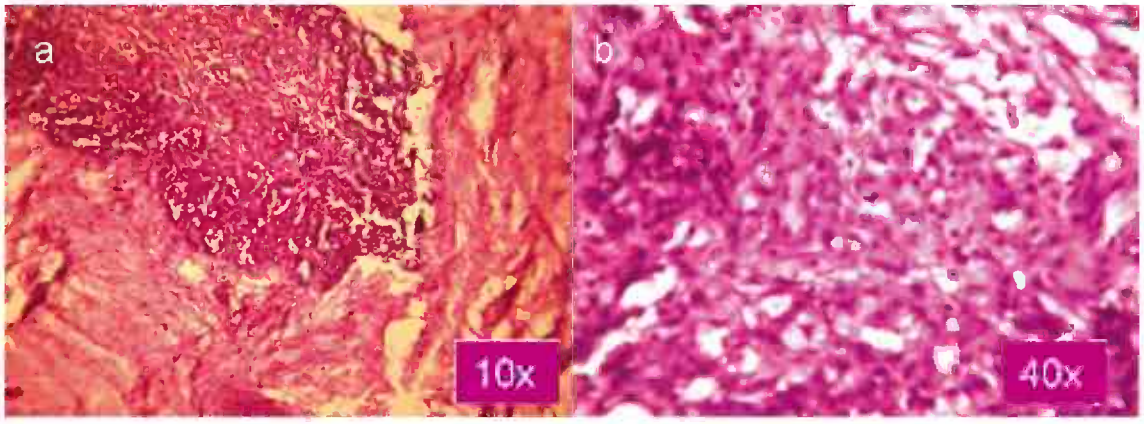

Fig. 3 Photomicrographs of tissue biopsy from T4 vertebra show acinar arrangement of malignant tumour cells (Haematoxylin & eosin, x10 and x40, respectively).

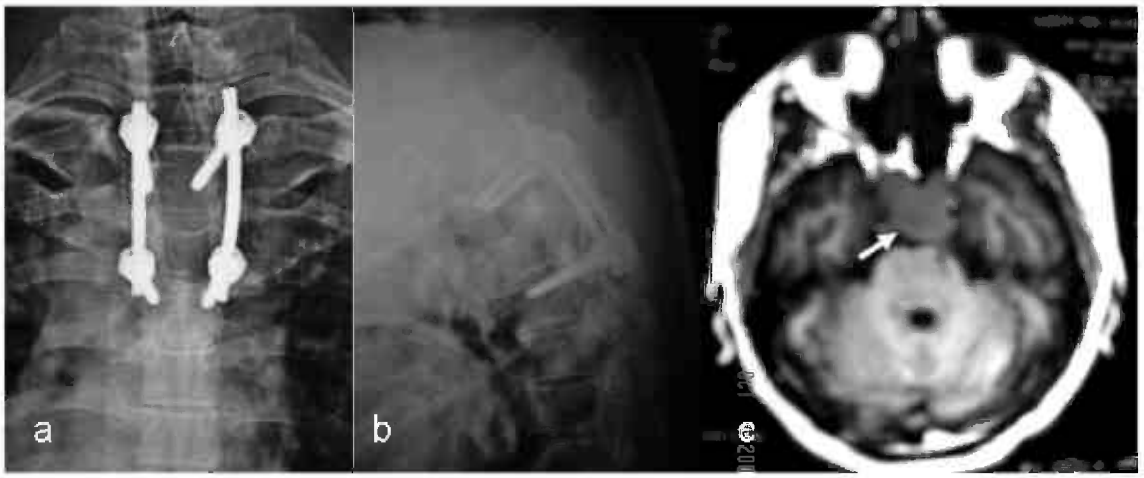

Fig. 4 Sixth month follow-up. (a) Antero-posterior and (b) lateral radiographs show posterior decompression and instrumentation. (c) Axial T1-W MR image shows soft tissue marrow replacement in the body of the sphenoid bone (arrow).

an oncologist for systemic chemotherapy and external radiation. The systemic chemotherapy included nine cycles of Cisplatin, 5-Flourouracil, and Epirubicin, together with steroids. His symptoms recovered within three months of surgery except for mild spasticity preventing him from walking fast. At six-month follow-up, he developed left-sided lagopthalmos. Cranial MR imaging showed an osteolytic lesion with soft tissue marrow replacement in the body of the sphenoid bone. It was eroding posteriorly into the dorsum sella, with pre-pontine soft tissue on the left side (Fig. 4).

DISCUSSION

Acinic cell carcinoma is an uncommon form of primary salivary gland tumour with an incidence of 1% among all salivary neoplasms, and l0%–17% of malignant salivary neoplasms[1-3]. This makes it the fourth commonest type of malignant salivary tumour, after mucoepidermoid carcinoma, adenoid cystic carcinoma and carcinoma ex-pleomorphic adenoma[3]. Although the tumour may occur at any age, there is a peak in the fifth decade of life. Between 8l% and 98% occur in the parotid gland, 11% in the submandibular gland, and 3%–12% in the minor salivary glands, most commonly in the palate.

The surgical treatment of choice is total parotidectomy with preservation of the facial nerve. Facial nerve resection is unavoidable only when the tumour has invaded the perineural structures. Routine excision of the lateral cervical lymph nodes is reserved for patients who have clinically demonstrable lymphadenopathy. While the findings are inconsistent, it is suggested that clinical features are most important with respect to prognosis, especially involvement of its deep lobe[1,8,11], large size[1,8], and infiltrative margins[1,7-10], presumably because these factors influence the completeness of excision.

A variety of morphological and cytological appearances are seen in acinic cell carcinomas. There are four morphological patterns, described as solid, micro cystic, papillary-cystic and follicular, which may appear alone or in combination[11]. The case reported here was of the high grade variety with an aggressive course and widespread metastasis along with local recurrence. In assigning the term "carcinoma" rather than "tumour", the World Health Organisation has recognised this neoplasm's potential to behave in an aggressive manner[6]. Despite being a low-grade neoplasm, death rates due to tumour range from 1.3% to 26%, with local recurrence of 8.3%–45%, regional lymph node involvement of 3.8%–16%, and distant metastasis of 2.6%–14%[1,7-11]. In 82% of cases, secondary lesions appear within five years of treatment.

Metastasis to the cervical lymph nodes, liver, lungs, contralateral orbit, and to other tumours (neurofibroma) has been described in the literature. There is only one such case mentioned in the literature, mentioning the spinal metastasis from acinic cell carcinoma[12-14]. Grage et al reported 11 cases of acinic cell carcinoma with one recurrent tumour having cervical spine metastasis[13]. Patient died of the recurrent disease and there is no mention of his neurological status. Zbaren et al had analysed 98 cases of parotid tumours with 17 being low-grade acinic cell carcinomas. One patient with distant skeletal metastasis was not specified as to the site of metastasis[12].

The reported case had incomplete parotidectomy in view of extensive adhesions. Ideally, the patient should have received postoperative radiotherapy in view of the inadequate removal of the tumour tissue. Within four months of primary surgical tumour resection, the patient developed metastasis in the thoracic vertebra with neurological deficits. This responded favourably to surgical excision and posterior stabilisation of the involved vertebra. It was followed by systemic chemotherapy and external radiation, with resolution of the neurological deficits. Local recurrence, lung metastasis, intracranial secondaries, and pathological fracture of T4 vertebra within four months after the primary surgery proved the aggressive nature of the tumour.

Despite being grouped as a low-grade malignant tumour, acinar cell carcinoma does recur and metastasise. It rarely has an aggressive course of progression. Thoracic spine metastasis with neurological compromise from acinic cell carcinoma of the parotid gland has not been previously reported. The reported case stresses the need for postoperative radiotherapy following incomplete resection of the primary tumour. A careful watch for distant metastasis is also of paramount importance.

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