International Journal for Multidisciplinary Research (IJFMR)
E-ISSN: 2582-2160 ● Website: www.ijfmr.com ● Email: editor@ijfmr.com

Denovo Carcinosarcoma of Parotid Gland with Three Malignant Components: A Case Report

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Abstract
Carcinosarcoma or true malignant mixed tumor is an extremely rare tumor of the salivary gland. It is a biphasic tumor and is composed of both malignant epithelial and malignant mesenchymal component. We, hereby, report a parotid lesion in a 58-year-old male that contained squamous cell carcinoma as epithelial component and rhabdomyosarcoma and chondrosarcoma as mesenchymal component. We report this unusual case of carcinosarcoma of parotid gland due to its rarity.

Keywords: Adenocarcinoma, Carcinosarcoma, Rhabdomyosarcoma, Chondrosarcoma, Malignant mixed tumor, immunohistochemistry

Introduction

Malignant mixed tumor of the salivary glands is a rare entity, first described in 1951, carcinosarcoma accounts for 0.04% to 0.2% of all salivary gland tumors and is known to be highly aggressive[1]. The mean age at presentation is 62 years with no sex predilection. Salivary gland carcinosarcoma is a rare malignant mixed tumor in which carcinomatous and sarcomatous components coexist[2]. In approximately half of reported cases, carcinosarcoma arises in the background of a preexisting pleomorphic adenoma (carcinosarcoma ex pleomorphic adenoma). Histologically, the neoplasm comprises frankly malignant epithelial and mesenchymal elements. The epithelial element is most often a squamous cell carcinoma or adenocarcinoma, and the most common mesenchymal component is chondrosarcoma, followed by fibrosarcoma, leimyosarcoma, osteosarcoma and rhabdomyosarcoma[3].

Case Report

A patient 58-year-old male came for OPD consultation in the Department of ENT with a mildly painful swelling in right side parotid region for 2 months. He had no history of any parotid lesion in past. It was gradually increasing in size but there were no associated systemic symptoms. On clinical examination, there was a swelling in right parotid region 3 cm × 2 cm mass with extension to cheek region .

The preoperative MRI of the neck showed a multiloculated solid cystic lesion in superficial lobe of right parotid gland measuring 3.7x3.5x4.1cm. It was reported as Pleomorphic Adenoma /Adeno-Cystic carcinoma with locoregional lymphadenopathy.

Gross Examination: We received a grey white soft tissue piece measuring 7x7x2.5 cm, attached skin flap measured 5x3.5cm. On external surface a nodule was present on the skin measuring 2x2x1.5 cm. On serial sectioning a grey white tumor was seen measuring 3.5x3x2.5 cm. External surface was variegated, areas of haemorrhage and necrosis were seen.

Histopathology: Final histopathologic examination of the specimen revealed a biphasic pattern composed of both carcinomatous and sarcomatous elements. The sarcomatous component was a chondrosarcoma and rhabdomyosarcoma and the carcinomatous component was composed of squamous cell carcinoma .There were areas of necrosis and increased mitosis were also seen along with atypical mitotic figures. There was no perineural invasion, lymphovascular invasion, or marginal involvement.(Ref: Figure 2)

Immunohistochemistry: Cytokeratin strong positivity was seen in epithelial component, myogenin strong positivity was seen in rhabdoid areas, S100 was positive in chondrosarcomatous area, ,and desmin was focally positive. (Ref: Figure 3)

Discussion

Malignant mixed tumor of the salivary gland is composed of three distinct clinical entities-carcinoma ex pleomorphic adenoma (the most common type) , metastasizing mixed tumor and carcinosarcoma. The tumor genesis of carcinosarcoma is controversial and two hypotheses are suggested[3,5]. In this case report, patient is 58 year old male. The epithelial component is composed of squamous cell carcinoma whereas mesenchymal component is mainly formed by rhabdomyosarcoma and chondrosarcoma. Histological criteria for diagnosis of that tumor are cell dysplasia, vascular, lymphatic and perineural invasion, infiltrative tumoral growth, necrosis, calcifications. One or two of those listed are sufficient criteria for the diagnosis of malignancy[6,8]. Among mesenchymal components, only chondromyxoid and hyaline material were present and no sarcomatous component was noticed. Carcinoma ex-pleomorphic adenoma and mucoepidermoid carcinoma were excluded on the basis of presence of sarcomatous component. In the literature, recurrence occurs in approximately two thirds of patients and metastases in about half of them. The median period of survival after diagnosis is 10 months in 63%.[3] In our study the patient has been surviving after a follow-up of 4 months following diagnosis.

Conclusion

The reported case is a rare case of carcinosarcoma (true mixed malignant tumor) of the parotid gland arising de novo with squamous cell carcinoma, rhabdomyosarcoma and chondrosarcoma as predominant epithelial and mesenchymal component . Salivary gland carcinosarcoma is rare with poor prognosis. Diagnosis is best confirmed with immunohistochemical staining, with electron microscopy reserved for controversial cases[4,7]. The current treatment of choice is surgery followed by radiotherapy. It has been established that it is aggressive and prognosis of this disease is poor, with local and distant recurrence commonly occurring. However, long-term follow-up with patients who have already undergone treatment is necessary in further elucidating the clinical course

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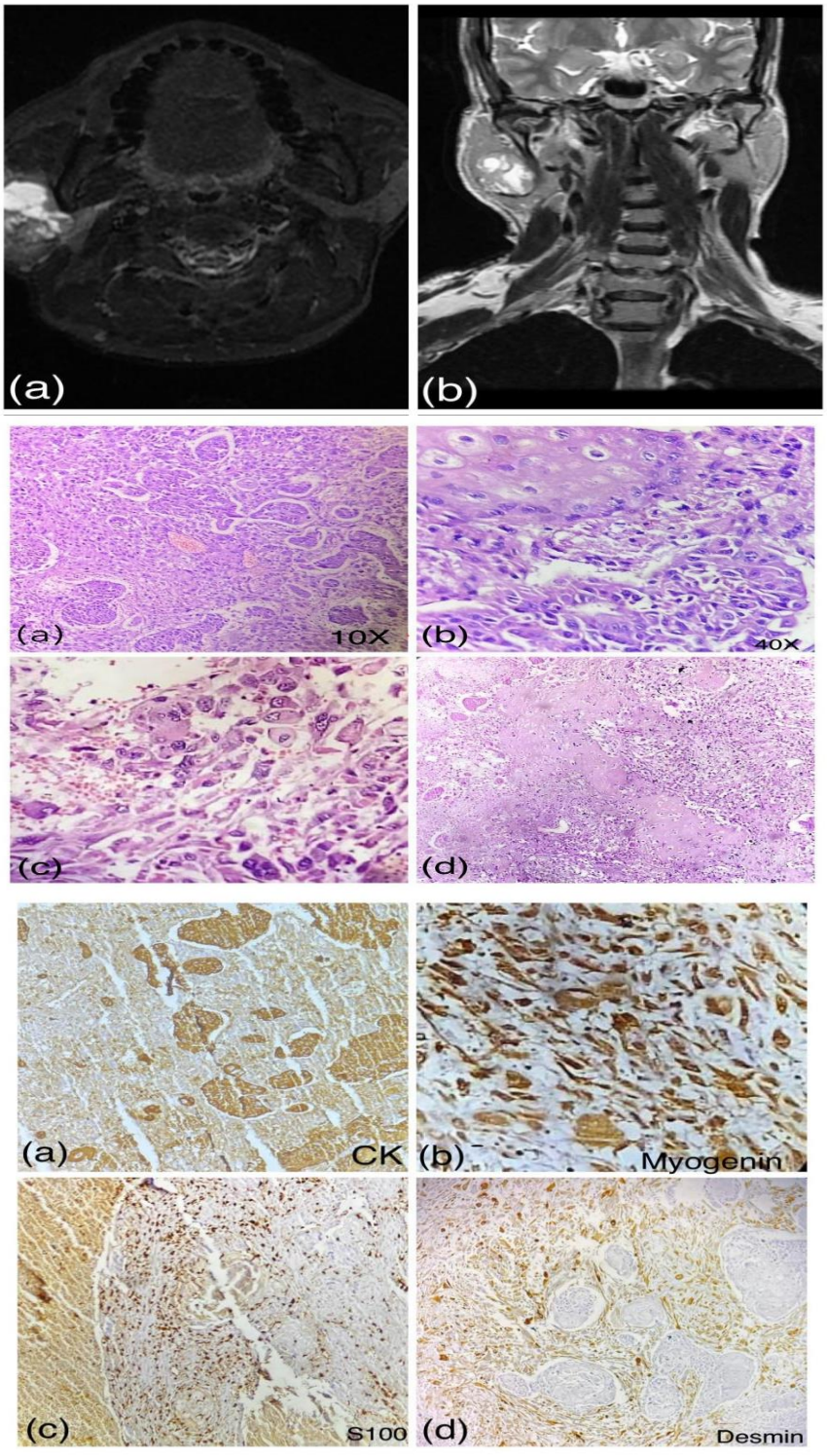
FIGURE LEGENDS

Figure 1: (a) ax stir, (b) cor T2

Figure2: (a)H&E sections at X10, (b) H&E sections at X40, (c) Stromal component: Rhabdoid areas, (d)Stromal elements: Chondroid areas

Figure 3: (a) cytokeratin, (b) myogenin, (c) S 100, (d) desmin


(a)
(b)
(a) 10X
(b) 40X
(c)
(d)
(a) CK
(b) Myogenin
(c) S100
(d) Desmin