A Case of Salivary Gland Carcinosarcoma- Monophasic Sarcomatous Pattern on Tumor Recurrence

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Abstract

Carcinosarcoma is an aggressive, biphasic high grade malignant neoplasm. Carcinosarcoma of salivary gland is an extremely infrequent neoplasm and only less than 80 cases were reported in the literature since the original description by Kirklin et al in 1951. We report a case of Carcinosarcoma ex pleomorphic adenoma in deep lobe of parotid in a 64 year old lady. This case is reported because of the rarity of carcinosarcoma in salivary gland, and to make aware of the unusual monophasic sarcomatous pattern of tumor recurrence, else an erroneous diagnosis of second malignancy may be made.

Keywords: Carcinosarcoma ex pleomorphic adenoma, Malignant mixed tumor, Parotid gland.
swelling in right pre-auricular region which was fixed to mandible. Recurrence of the tumor was suspected, and so right radical parotidectomy, along with right hemimandibulectomy and right submandibulectomy was done to remove the tumor. Peroperatively, the facial and lingual nerves were infiltrated by the tumor. The excision specimen together measured 10x6x6 cm and weighed 200 gm. Ramus of mandible appeared bulged out. Cut section of specimen showed a grey-white glistening neoplasm involving the ramus of mandible and adjacent soft tissue (figure 2). Microscopic study showed an infiltrating neoplasm with lobular architecture, involving ramus of mandible, periosteum and attached gingival soft tissue. Within the lobules, chondromyxoid stroma and small round to oval neoplastic cells were seen, which resembled the myxoid chondrosarcomatous component of the previous biopsy. No evidence of pleomorphic adenoma or malignant epithelial component was seen in the specimen even after thorough sampling. Both resected ends of the bone, submandibular gland and residual parotid gland were free of neoplasm. So, the recurrence of tumor showed only the malignant mesenchymal element in the form of myxoid chondrosarcoma.

![Fig-1](image1)

![Fig-2](image2)
**DISCUSSION**

The term Malignant mixed tumor was first coined by King et al. in 1967[4]. Malignant mixed tumor is a rare disease and comprises four distinct types: 1. Metastasizing mixed tumor is a histologically benign pleomorphic adenoma that inexplicably manifests local or distant metastasis. 2. Carcinoma ex pleomorphic adenoma is a pleomorphic adenoma from which an epithelial malignancy is derived. 3. Carcinosarcoma ex pleomorphic adenoma is a pleomorphic adenoma from which a malignant tumor composed of a mixture of both carcinomatous and sarcomatous elements are derived. 4. Carcinosarcoma or true malignant mixed tumor is a malignant tumor composed of a mixture of both carcinomatous and sarcomatous elements without histological evidence of pre-existing pleomorphic adenoma [1].

Carcinosarcoma of salivary gland is an extremely rare neoplasm. Gnepp et al. in 1993 published a review of 43 cases of carcinosarcoma of salivary glands. Majority were reported in parotid, followed by submandibular and minor salivary glands in palate [5, 6]. Generally these are poorly circumscribed and infiltrative neoplasms, histopathology being the gold standard for diagnosis. In this biphasic tumor, the relative proportion of carcinomatous and sarcomatous component is variable and at times one of the components is evident only on thorough searching. Immunohistochemistry is helpful in confirming the diagnosis. The carcinomatous component may be adenocarcinoma, squamous cell carcinoma, undifferentiated carcinoma, salivary duct carcinoma, mucoepidermoid carcinoma, adenoid cystic carcinoma or epithelial myoepithelial carcinoma. Sarcomatous component is usually chondrosarcoma. However, cases with fibrosarcoma, osteosarcoma, myxoid sarcoma, undifferentiated sarcoma and rarely rhabdomyosarcoma are also reported [6-9]. Destructive local infiltration, angioinvasion and perineural invasion are common in carcinosarcoma. Treatment is wide surgical excision and radiotherapy. However local recurrence and or metastasize to lung, bone or brain occur in 60% of patients within a 30 month period. In our case regular follow up of the patient was available for a period of six months only.

Usually both malignant epithelial and mesenchymal components will be present in tumor when it recurs or metastasizes. In 2016, Mansour et al. reported a recurrent tumor with pure carcinomatous pattern in an old case of parotid carcinosarcoma [10]. In our case, pure sarcomatous pattern was observed in recurrent tumor.

**REFERENCES**