Case Report - Oncology

Carcinosarcoma of parotid gland (malignant mixed tumor)

Access this article online
Website: www.amsjournal.com
DOI: 10.4103/2231-0746.175757
Quick Response Code:

Duan Feng, Nyimi Bushabu Fidele, Mansthumba Milolo Agustin[1], Guan Jian, Sekele Isouradi Bourleyi[1], Lamwe Augustin[2], Ngueji Kakubu Olivier[3]

Department of Oral and Maxillofacial Surgery (Unit I), Second Affiliated Hospital of Jiamusi University, Jiamusi, China, [1]Department of Prosthodontics and Orthodontics, Affiliated Hospital of Kinshasa University, Kinshasa, DR. Congo, [2]Departments of Rehabilitation and Physiotherapy and [3]Pediatrics, the Third Affiliated Hospital of Jiamusi University, Jiamusi, China

Address for correspondence:
Dr. Nyimi Bushabu Fidele, Department of Oral and Maxillofacial Surgery (Unit I), Second Affiliated Hospital of Jiamusi University, No. 522 Hong Qi Road, Jiamusi, Heilongjiang 154002, China.
E-mail: fidelenyimi@yahoo.fr

ABSTRACT

Salivary gland carcinosarcoma is a rare neoplasm; with a poor prognosis. The most common epithelial components are adenocarcinoma or squamous cell carcinoma, whereas the most common mesenchymal components are chondrosarcoma. It should not be confused with the most common carcinoma ex-pleomorphic adenoma, in which the epithelial component alone is malignant. This condition might exhibit with a wide variety of presentation and symptoms along with associated conditions. We present a case of an old patient who presented with a very unusual type clinically with confusing presentation which was eventually diagnosed as carsinosarcoma. In addition, the literature is reviewed, and the possible clinical signs and management of malignant mixed tumor of the salivary gland are briefly discussed.

Keywords: Carcinosarcoma, malignant mixed tumor, parotid gland

INTRODUCTION

Carcinosarcoma (CRS) is a rare salivary gland neoplasm consisting of both carcinomatous and sarcomatous component. The first described mixed carcinoma and sarcoma of the parotid gland in 1951 was Kirklin *et al*., and King OH Jr were the first to use the term true malignant mixed tumor in 1976 (CRS).[1,2] Since then, <73 cases has been reported in published literature till now and the those reported in patients older than 76 years is extremely rare (including our case). It has accounted for 0.16–1.0% of all malignant salivary tumors[3] with 65% in the parotid gland.[4] Although it usually appears isolatedly approximately 33% of patients have either a clinical history or histologic confirmation of a coexisting pleomorphic adenoma. These types of tumors have an aggressive characteristic and are often regarded as high-grade tumors with distant metastasis and occurring in 54% of patients. Patients usually present between 60 and 65 years of age and most series report no sex predominance.[4]

Due to the wide variation of the presentation, this clinical condition remains as an interesting point of study regarding the diagnosis and challenges of management. We present a case of parotid CRS, exhibiting features of salivary ductal adenocarcinoma (carcinomatous element) and fibrosarcoma/osteosarcoma (sarcomatous element).

CASE REPORT

Patient K, 76-year-old female was admitted in the Department of Oral and Maxillofacial Surgery (Unit I), at the Second Affiliated Hospital of Jiamusi University on October 13, 2014 with a painless mass of parotid gland since many years. It had increased

This is an open access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 3.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

Cite this article as: Feng D, Fidele NB, Agustin MM, Jian G, Bourleyi SI, Augustin L, *et al*. Carcinosarcoma of parotid gland (malignant mixed tumor). Ann Maxillofac Surg 2015;5:240-3.

© 2015 Annals of Maxillofacial Surgery | Published by Wolters Kluwer - Medknow

in size over the past 6 months and there were no associated systemic symptoms. On clinical examination, there was deformation of the right parotid gland region by a 10 cm × 8 cm size mass with extension to cheek region [Figure 1]. The palpation revealed 10 cm × 8 cm size tumor, well-circumscribed, hard in consistency, not fixed to deep planes and tender to touch. The superjacent skin was normal and cervical lymph nodes were not palpable.

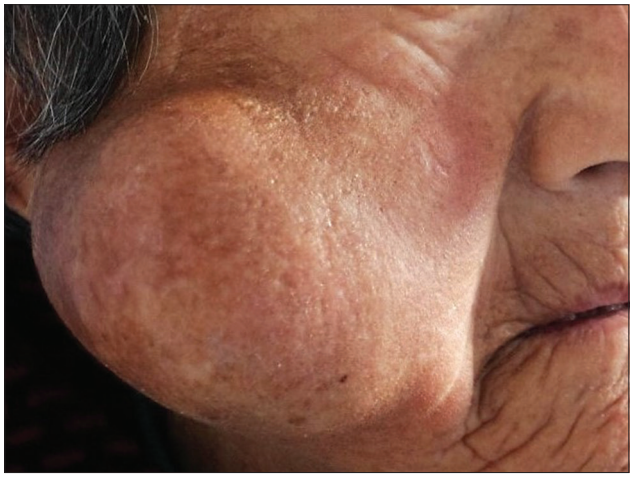

Figure 1: Preoperative

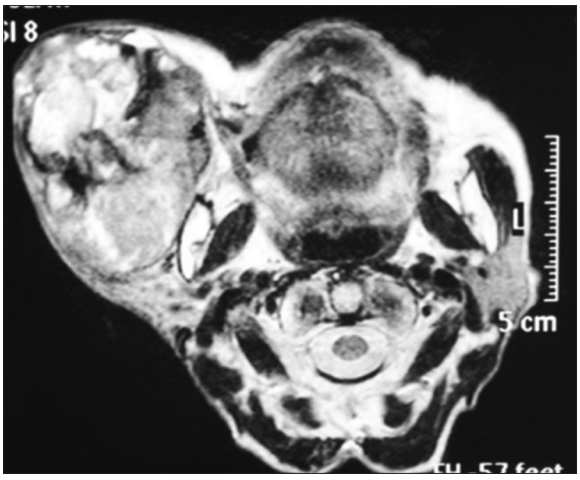

Figure 2: Magnetic resonance imaging revealed a solid heteomogeneous lesion with central, nonvascular areas and sharp borders. A, Axial view

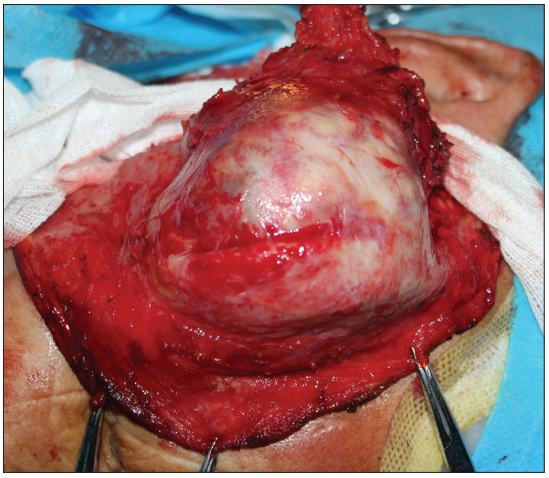

Figure 3a: Intraoperative

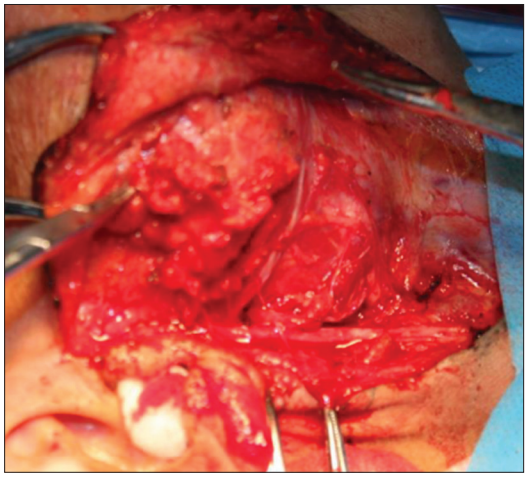

Figure 3b: The excise supercial paratidectomy with conservation of facial nerve

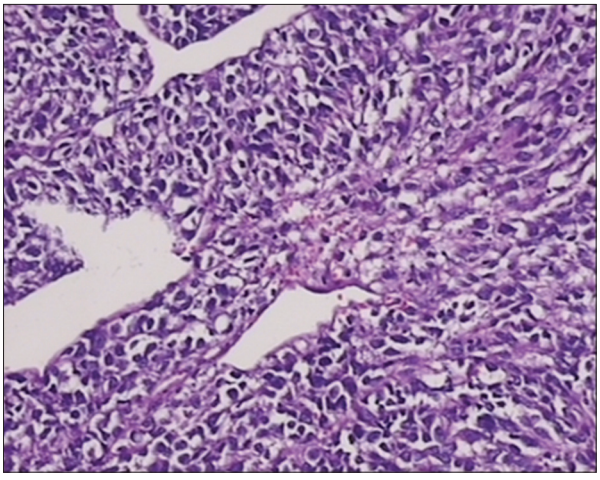

Figure 4a: Histopathology image of the excised parotid gland carsinosarcoma-10x

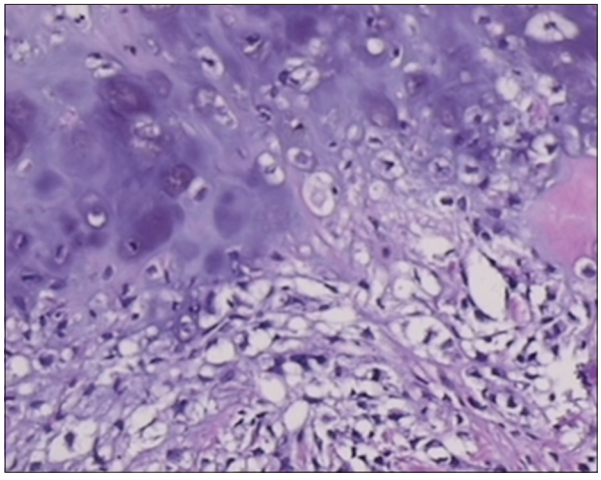

Figure 4b: Histopathology image of the excised parotid gland carcinosarcoma-40x

The preoperative magnetic resonance imaging of the neck showed a heterogeneous lesion, with central nonvascular areas and sharp borders and without any distant metastases to neck lymph nodes [Figure 2]. A presumptive diagnosis of malignant tumor of right parotid gland was made. Based on these data and after consent of patient, superficial parotidectomy had been performed with preservation of facial nerve [Figure 3a and b].

The histopathological exam revealed a solid mass poorly circumscribed that measured 8.5 cm × 7.4 cm × 5.3 cm, apparently totally encapsulated, with moderate consistency, of a gray brown color and calcified. The specimen was fixed in 10% buffered formalin, embedded in paraffin. Final histopathologic examination of the specimen revealed a biphasic pattern composed of both carcinomatous and sarcomatous elements. The sarcomatous part was composed by mainly spindle cell sarcoma, with a small amount of bone tumor and a poorly differentiated cancer nest [Figure 4a and b]. Postoperative evolution, under antibiotic therapy was good, with good healing and without radiotherapy [Figure 5]. After a clinical follow-up of 12 months, no metastases was found.

DISCUSSION

The first described mixed carcinoma and sarcoma of the parotid gland in 1951 was Kirklin *et al*., and King OH Jr. were the first to use the term true malignant mixed tumor in 1976 (CRS).[1,2] Since then, <72 cases has been reported in published literature till now to the best of our knowledge. CRSs of the salivary glands most often arise in the parotid gland (65%), followed by submandibular glands (19%) and minor salivary gland, although the palate is the most common.[5,6] There is no gender predominance. Many cases are seen to arise in a preexisting benign mixed tumor.[5] In addition, the carcinomas have been described in several organs, including uterus, bladder, lung and others.

The origin of this tumor is still controversial.[7] The tumor presents as a mass that may be rapidly enlarging. In some cases, the mass is painful; the pain may be localized, referred, or both. The signs of facial nerve weakness/paralysis are commonly encountered when the parotid gland is involved. Our patient had no sudden painful increase in neck mass size or any signs of facial weakness.

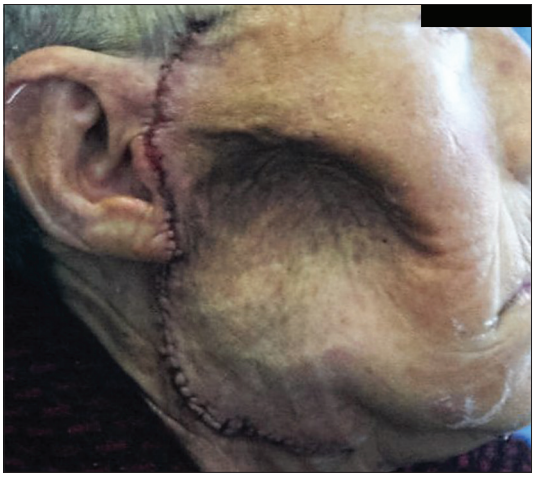

Figure 5: Postoperative

Aspiration cytology represents an uncertain diagnostic method, but fine needle aspiration (FNA) studies can be used for preoperative evaluation.[8] In our case, FNA wasn't done. Correct diagnosis is confirmed only by histopathology in paraffin and sometimes need additional immunohistochemical investigation for a final diagnosis of certainty. The carcinomatous component is most often an adeno-carcinoma, a squamous cell carcinoma, or an undifferentiated carcinoma. The sarcomatous component is usually a chondrosarcoma, but the other types of sarcoma have been also reported include spindle cell sarcoma (not otherwise specified), fibrosarcoma, osteosarcoma, leyomyosarcoma, malignant fibrous histiocytoma and rarely liposarcoma and rhabdomyosarcoma.[9] In our case, histological aspects revealed combination between the characteristics of adenocarcinoma by epithelial components and sarcoma's elements. 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.

The CRS is an aggressive, high-grade malignancy. The treatment is radical surgical resection which should be combined with radiation and chemotherapy.[10] Our case was treated by superficial parotidectomy with conservation of facial nerve without radiotherapy. 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%.[5] In our study the patient has been surviving even after a follow-up of 12 months following diagnosis.

CONCLUSIONS

The case reported in this paper is a CRS, which combines both carcinomatous and sarcomatous components. It is reported because of its rare occurence and lack of malignant symptoms in the parotid gland. Although the number of reported cases is less, the combination of radical surgical excision and radiotherapy not always seems to be the treatment of choice for salivary CRS currently.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

REFERENCES

1. Kirklin JW, McDonald JR, Harrington SW, New GB. Parotid tumors; histopathology, clinical behavior, and end results. Surg Gynecol Obstet 1951;92:721-33.
2. King OH Jr. Carcinosarcoma of accessory salivary gland. First report of a case. Oral Surg Oral Med Oral Pathol. 1967;23:651-9.
3. Kwon MY, Gu M. True malignant mixed tumor (carcinosarcoma) of parotid gland with unusual mesenchymal component: A case report and review of the literature. Arch Pathol Lab Med 2001;125:812-5.
4. Said MS. True Malignant Mixed Tumor (Carcinosarcoma); 2009. Available from: http://www.emedicine.medscape.com/article/1661577-overview. [Last accessed on 2015 Dec 01].
5. Bhalla RK, Jones TM, Taylor W, Roland NJ. Carcinosarcoma (malignant mixed tumor) of the submandibular gland: A case report and review of the literature. J Oral Maxillofac Surg 2002;60:1067-9.

6. Carson HJ, Tojo DP, Chow JM, Hammadeh R, Raslan WF. Carcinosarcoma of salivary glands with unusual stromal components. Report of two cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1995;79:738-46.
7. Thompson L, Chang B, Barsky SH. Monoclonal origins of malignant mixed tumors (carcinosarcomas). Evidence for a divergent histogenesis. Am J Surg Pathol 1996;20:277-85.
8. Granger JK, Houn HY. Malignant mixed tumor (carcinosarcoma) of parotid gland diagnosed by fine-needle aspiration biopsy. Diagn Cytopathol 1991;7:427-32.
9. Fowler MH, Fowler J, Ducatman B, Barnes L, Hunt JL. Malignant mixed tumors of the salivary gland: A study of loss of heterozygosity in tumor suppressor genes. Mod Pathol 2006;19:350-5.
10. Patnayak R, Jena A, Raju G, Uppin S, Satish Rao I, Sundaram C. Carcinosarcoma of the parotid gland: A case report and short review of literature. Internet J Oncol 2007;5:1-7.