CASE REPORT

Salivary duct carcinoma of the parotid gland

Mona Mlika, Nadia Kourda, Zidi YSH, Raoudha Aloui, Nadia Zneidi, Soumaya Rammeh, Rachida Zermani, Sarah Ben Jilani

Department of Pathology, Charles Nicolle Hospital, Tunis, Tunisia

Address for correspondence:
Dr. Mlika Mona,
Department of Pathology,
Charles Nicolle Hospital, 2037-Tunis, Tunisia.
E-mail: mlika.zorgati.mona@hotmail.com

ABSTRACT

Salivary duct carcinoma of the parotid gland is an uncommon tumor, highly aggressive. About 200 cases have been reported in the English literature. Pathomorphologically, these tumors showed great similarities to ductal carcinoma of the female breast, which is why they described this tumor as "salivary duct carcinoma." The authors describe a new case of salivary duct carcinoma of the parotid gland. We present the case of a 50-year-old patient with progressive facial paralysis. The MRI examination of the head showed two ill-defined formations. A malignant tumor was strongly suspected, so that a total left parotidectomy with excision of the adjacent facial nerve and left lymph node dissection was performed. Microscopic examination concluded to a salivary duct carcinoma of the left parotid gland negative with Her2/neu antibody with lymph node metastasis. There were no recurrences or metastases within 3 years of follow-up. Salivary duct carcinoma of the parotid gland is a rare tumor with an aggressive behavior. This is due to its propensity to infiltrate distant organs. The diagnosis is based on microscopic examination. Treatment modalities are non-consensual, but some authors advocate the necessity of aggressive approach, especially in tumors negative with Heur2/neu antibody. This is due to the fact that the overexpression of this antigen was reported to be associated with a poor prognosis.

Key words: Parotid gland, salivary duct carcinoma, treatment

INTRODUCTION

Salivary duct carcinoma is a rare tumor accounting for 1% to 3% of all malignant salivary gland tumors.[1,2] Initially, a group of malignant salivary gland tumors characterized by ductal formations and central necrosis was first described by Kleinsasser *et al*. in 1968.[3] Then, a few cases have been reported in the literature resulting in limited data regarding the biologic and the immunohistochemical characteristics of this tumor.

CASE REPORT

We present the case of a 50-year-old patient with progressive facial paralysis. Physical examination showed a mass of the left parotid gland which was painless on palpation, hard and non-compressible. There were no cervical lymphadenopathy and no abnormalities within the oral cavity. The MRI examination of the head showed two ill-defined formations. The first one was in contact with the intraparotid portion of the seventh nerve. The second one was located in the superficial lobe infiltrating the masseter muscle and the homolateral soft tissue. Chest X-ray was normal. A malignant tumor was strongly suspected, so that a total left parotidectomy with excision of the adjacent facial nerve and left neck lymph node dissection was performed. The resultant defect was reconstructed with immediate nerve grafting. On gross examination, we received a 5-cm gland with 2 lesions of 1 cm, ill-defined and not encapsulated, dirty white. Microscopic examination showed ductal lesions comprising pleomorphic and epithelioid tumor cells with a cribriform growth pattern. Solid and papillary areas were also noted [Figure 1]. Lymph node parenchyma was also observed and seems to be infiltrated by the same malignant tumor. Immunohistochemical study was performed using Her2-neu antibody and showed a negativity of tumor cells. The diagnosis retained was a salivary duct carcinoma of the left parotid gland with lymph node metastasis. There were no recurrences or metastases within 3 years of follow-up.

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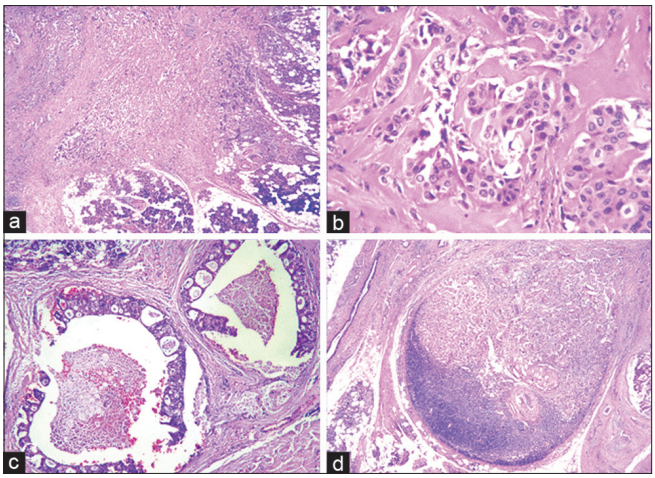

Figure 1: (a) Malignant tumor infiltrating parotid gland parenchyma (HES, ×250). (b) Infiltrative component with trabecular architecture (HES, ×400). (c) Intraductal component with cribriform pattern and intraluminal necrosis (HES, ×400). (d) Lymph node infiltration by the same tumor (HES, ×250)

DISCUSSION

Salivary duct carcinoma of the parotid gland is an uncommon tumor, highly aggressive. About 200 cases have been reported in the English literature. Pathomorphologically, these tumors showed great similarities to ductal carcinoma of the female breast, which is why they described this tumor as "salivary duct carcinoma." It represents a rare tumor with an estimated incidence of 1% to 3% of all malignant salivary gland tumors.[1,2,4] The parotid gland is most commonly involved. Salivary duct carcinoma accounts for 0.9% to 6% of all parotid's tumors.[5] It frequently involves the extracranial portion of the facial nerve and has a propensity to metastasize through the temporal bone via perineural spread.[6] Gingival metastases have also been reported.[7] Rarely, submandibular glands and minor salivary glands are concerned. Salivary duct carcinoma may develop in some cases on the basis of pre-existing pleomorphic adenoma, but it can also occur de novo. Patients are usually elderly men with a mean age ranging between 55 to 61 years.[4] It presents as a rapidly growing mass, which develops aggressively with possibilities of early distant metastases, local recurrence, and high mortality. Facial paralysis is observed in 40% to 60% of cases. Lymphadenopathies are noted in 35% of cases.[4] Imaging findings, especially CT scan and MRI features, are non-specific but they are helpful in the diagnosis of malignancy and in the management. They can indicate the malignant nature of the tumor by showing ill borders or an infiltration of the adjacent tissue. Positive diagnosis is based on histologic examination. The means of diagnosis consist of fine needle aspiration cytology which is useful but not always reliable, fine needle aspiration specimen, and surgical specimen. Gross findings consist in a tumor of variable size, usually firm with a variable cystic component. An infiltration of the adjacent parenchyma is usually obvious. Microscopically, the most peculiar feature is the similarity to ductal carcinoma of the breast. The tumor is composed of an intraductal and invasive components. Intraductal component is cribriform, papillary, solid with comedo-like central necrosis. The infiltrative component is made of glands, cords of cells with desmoplastic reaction. Several variants have been described such as sarcomatoid salivary duct carcinoma, low-grade neoplasm, and mucin-rich variant.[8] Immunohistochemical findings are not useful, but a constant overexpression of keratin, HER/2 neu, CEA, and c-erd-B2 have been described. Frequently, androgen receptors and prostate-specific antigen expression have been reported. In the course of determination of the development of malignant tumors of the salivary glands as well as for the prediction of their clinical course, molecular and biologic research has moved into the center of tumor research. In this context, the cell proliferation rate (Ki-67), the mutation of oncogenes, and the overexpression of growth factor-binding receptors like HER2/neu, p16, and p53 have been identified as important factors in the genesis of salivary gland tumors.[4] Some authors reported that Her-2/neu expression, together with an elevated proliferation rate, is associated with an unfavorable clinical course.[4,9] This observation was also noted in our case. In fact, the negativity of HER-2/neu expression was correlated to a 3-year survival. Differential diagnoses include mucoepidermoid carcinoma, adenocarcinoma NOS, metastatic adenocarcinoma, oncocytic carcinoma, and the most relevant morphologic feature is the presence of an intraductal component which is specific of the diagnosis. Therapeutic approach seems to be non-consensual because of the limited data but many authors recommend, in parotid gland tumors, a total parotidectomy even in T1 tumors because local disease recurrence is often life-threatening.[10] If facial paralysis is present, a radical parotidectomy is mandatory. In patients with involvement of the submandibular or minor salivary glands, tumor resection with wide margins of surrounding tissue is indicated to control local disease.[4] The effects of adjuvant postoperative radiotherapy cannot be evaluated with certainty but many authors suppose that the aggressiveness of this tumor justifies adjuvant measures. Postoperative radiation therapy is indicated in case of extraparotid extension, pathological resection margins, cervical lymph node metastasis, lymphatic embolus, and/or neurologic invasion. Chemotherapy is generally reserved for metastatic forms of the disease.[11] Kuroda *et al*. reported the utility of both anti-androgen therapy and chemotherapy for a patient with advanced salivary duct carcinoma.[12] Salivary duct carcinoma is an aggressive tumor with a worse prognosis. This is due to its metastatic potential. Nearly 50% of the patients die within 4 to 5 years. Prognostic criteria are non-consensual consisting mainly in the young age, a tumor size superior to 3 cm, infiltrative tumor margins, local recurrence, lymphatic and distant metastases, necrosis, percentage of infiltrating, and intraductal components, but there is no real consensus about these prognostic features.[4,8]

REFERENCES

1. Seifert G, Caselitz J. Epithelial salivary gland tumors: Tumor markers. In: Fenoglio-Preiser CM, Wolff M, Rilke F, editors.

Progress in surgical pathology. Vol. 9. New York: Field and Wood; 1989. p. 157-87.
2. Gal R, Strauss M, Zohar Y, Kessler E. Salivary duct carcinoma of the parotid gland. Cytologic and histopathologic study. Acta Cytol 1985;29:454-6.
3. Kleinsasser O, Klein HJ, Hubner G. Salivary duct carcinoma. A group of salivary gland tumors analogous to mammary duct carcinoma. Arch Klin Exp Ohren Nasen Kehlkopfheilkd 1968;192:100-5.
4. Jaehne M, Roeser K, Jaekel T, Schepers JD, Albert N, Loning T. Clinical and immunohistologic typing of salivary duct carcinoma: A report of 50 cases. Cancer 2005;103:2526-30.
5. Etges A, Pinto DS Jr, Kowalski LP, Soares FA, Araujo VC. Salivary duct carcinoma: Immunohistochemical profile of an aggressive salivary gland tumor. J Clin Pathol 2003;56:914-8.
6. Nguyen BD, Roarke MC. Salivary duct carcinoma with perineural spread to facial canal: F-18 FDG PET/CT detection. Clin Nucl Med 2008;33:925-8.
7. Bernabé DG, Veronese LA, Miyahara GI, Conrado-Neto S, Biasoli ER. Gingival metastasis from salivary duct carcinoma of the parotid gland. J Periodontol 2008;79:748-52.
8. Brandwein-gensler, Skalova A, Nagao T. Salivary duct carcinoma. World Health Organization Classification of tumours, Pathology and genetics of head and neck tumours. In: Barnes L, Eveson JW, Sidransky D, editors. Lyon: IARCC Press; 2003. p. 236-8.
9. Kamio N. Coexpression of p53 and c-erbBproteins-2 proteins associated with histological type, tumor stage and cell proliferation in malignant salivary gland tumors. Virchows Arch 1996;428:75-83.
10. De Riu G, Meloni SM, Massarelli O, Tullio A. Management of midcheek masses and tumors of the accessory parotid gland. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;111: e5-11.
11. Pons Y, Alves A, Clément P, Conessa C. Salivary duct carcinoma of the parotid. Eur Ann Otorhinolaryngol Head Neck Dis 2011;128:194-6.
12. Kuroda H, Sakurai T, Yamada M, Uemura N, Ono M, Abe T, *et al*. Effective treatment by both anti-androgen therapy and chemotherapy for a patient with advanced salivary duct carcinoma. Gan To Kagaku Ryoho 2011;38:627-30.

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