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Salivary Duct Carcinoma With Rhabdoid Features: A Rare Tumor in Unusual Location?A Case Report

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ABSTRACT

Ductal carcinoma is a rare tumor, which accounts for 1 to 3% of all malignant salivary gland tumors primarily affecting middle-aged men with a high potential for metastasis. In salivary gland neoplasms, rhabdoid cells are not commonly seen. We report the case of a 58-year-old man, who presented a mass of the left cheek evolving for 3 months, who underwent an exofacial parotidectomy, the extemporaneous histopathological analysis of the specimen revealed a ductal carcinoma with rhabdoid features. We completed by a total conservative parotidectomy of the facial nerve. Salivary duct carcinoma with rhabdoid features should be distinguished from malignant lymphoma, malignant melanoma, and myogenic sarcomas. Transformation to ductal carcinoma with rhabdoid features has occurred in metastatic lesions. Treatment was based on extensive carcinological resection with postoperative radiotherapy with regular follow-up.

Introduction

Ductal carcinoma is an aggressive tumor, which may develop de novo or within pleomorphic adenoma accounts less than 1.8% of all major salivary gland tumors and approximately 10% of all malignant salivary gland tumors. This tumor is the rarest in 1% to 3% of all malignant salivary gland tumors affecting mainly middle-aged men with a high potential for metastasis. (1, 2)

It was first described by Kleinsasser, who has used this term because of the tumor resemblance to ductal carcinoma of the breast. Rhabdoid cells have been observed in several neoplasms, including kidney tumors, brain tumors, lung carcinoma, and mammary carcinoma. In salivary gland neoplasms, however, these cells are not seen frequently. (3, 4)

The tumor is clinically characterized by a rapid onset and progression, its recurrence rate is high. Surgery and radiation therapy is the treatment of choice. Despite this aggressive therapy, most patients died from their disease within 2 to 3 years. (5)

In our work, we report the case of a ductal carcinoma with a rhabdoid appearance in a 58-year-old patient.

We report the case of a salivary duct carcinoma with rhabdoid features of the accessory parotid gland, we examined the different clinicopathological and immunohistochemical characteristics, the therapeutic and prognostic aspects of this tumor.

Case report

Our case was a 58-year-old man, treated for viral hepatitis B in 2016, who presented a swelling of a left cheek, gradually increasing in size over the past 3 months. The clinical examination objectified a left cheek mass without inflammatory sign opposite, of hard consistency, painless, making 3cm long axis. A soft tissue ultrasound was performed, which revealed a heterogeneous hypertrophy of the left masseter muscle, probably related to an abscess in its solid phase or a proliferative lesion. The MRI scan shows a 25 x 30x 50 mm heterogeneous and irregular jugal mass, with low intensity on T1 imaging and partially high intensity on T2 imaging, including areas of necrosis and calcifications with low intensity on T1 and T2 imaging, with an infiltration of the masseter muscle and respect of the parotid gland. (Fig.1)

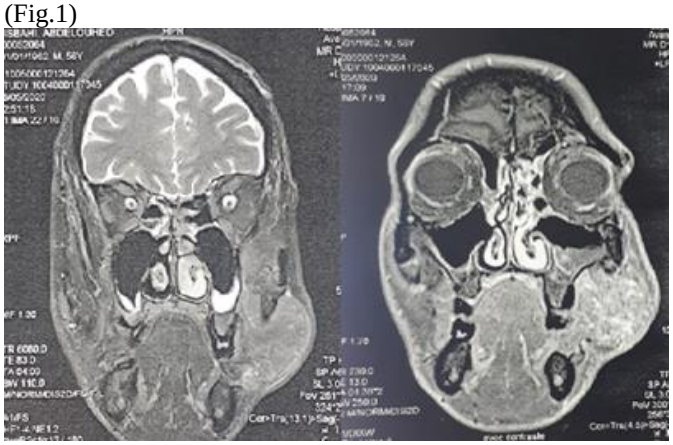

Figure 1. MR images shows an irregular mass in the left cheek region with low intensity on T1 imaging and partially high intensity on T2 imaging, with heterogenous enhancing.

Under general anesthesia, we performed a total parotidectomy, with trans muscular tumor resection. An extemporaneous analysis of a cervical lymphadenopathy was performed which returned in favor of a reactive adenitis. The anatomopathological analysis of the specimen showed a tumor proliferation of large cells with ovoid eosinophilic cytoplasm, eccentric nuclei, marked cellular atypia, with the presence of mitotic activity, the immunohistochemical profile shows positivity for anti AE1 / AE3 antibodies, anti CK7, EMA, HER2, ACE antibodies. This confirmed the diagnosis of a ductal carcinoma with rhabdoid features, then we completed by a radiotherapy (60 Gy). Over a two-year period, no signs of recurrence were detected. (Fig.2)

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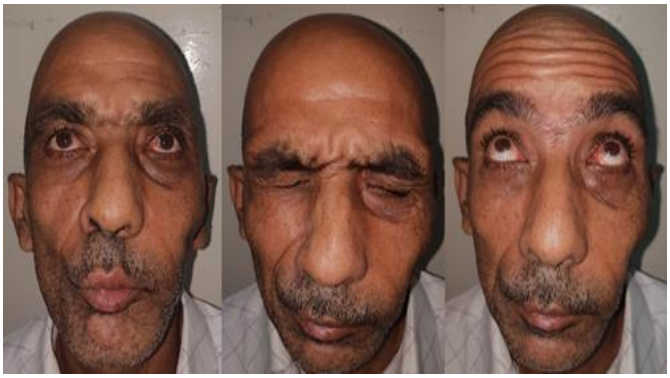

Figure 2. postoperative image of the patient two years after the surgery.

Discussion

Salivary duct carcinoma is a rare, high-grade tumor that histologically resembles high-grade ductal carcinoma of the breast, and includes several variants, among these variants, ductal carcinoma with rhabdoid appearance which is extremely rare. This tumor represents 10%of all malignant tumors of the salivary glands. (6)

Salivary duct carcinoma with rhabdoid features should be distinguished from several other neoplasms. The differential diagnosis includes collision tumors and hybrid carcinomas. (7)

Men are more likely to be diagnosed with ductal carcinoma than women, in the fifth to sixth decade.

Clinically, the tumors are generally characterized by a mass with rapid growth associated with pain, the presence of cervical lymphadenopathy has also been noted. (8). Magnetic resonance imaging can assess the extent of the disease, even in the absence of palpable cervical lymphadenopathy.

Histologically diffuse proliferation of rhabdoid cells, which were weakly adhesive with relatively large eosinophilic cytoplasm, eccentric nuclei, marked cellular atypia, and 1 or 2 large nucleoli (5)

Ductal carcinoma with cells with rhabdoid characteristics was negative for lymphoma markers, such as CD45, CD20, CD5 and / or CD30. They were also negative for melanoma markers, such as S-100 protein, HMB45 or Melan-A, and for skeletal muscle markers such as myogenin, Ia and desmin, they are generally positive for both CK and vimentin (9)

Therefore, salivary duct carcinoma with rhabdoid features should be distinguished from malignant lymphoma, malignant melanoma, and myogenic sarcomas. Kimihide et al (5) have considered that transformation to salivary duct carcinoma with rhabdoid features occurs in metastatic lesions.

In the case of a minor salivary gland carcinoma, tumor resection with wide excision margins is indicated to control the disease. Whatever the primary site of the disease, unilateral and sometimes bilateral lymph node dissection is generally indicated.

The effects of postoperative adjuvant radiotherapy cannot be assessed with certainty. (10) Chemotherapy has only been offered for recurrence. However, the evidence suggests that improvement of the disease could be obtained with adjuvant chemotherapy in combination with post-operative radiotherapy. (8)

Patients with ductal carcinoma younger than 50 years old had a better prognosis than patients older than 50 years. (11)

Conclusion

Ductal carcinoma with rhabdoid appearance is an aggressive and rare tumor with a poor prognosis. Recurrence or metastasis was frequent, involving locoregional and distant sites. The mainstay of treatment is surgery and radiotherapy, but good control remains difficult.

Competing interests

The authors declare no competing interests.

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