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Case Study

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LOCAL RECURRENCE OF PAROTID FIBROSARCOMA: A CASE REPORT AND THE REVIEW OF THE LITTERATURE

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ABSTRACT

Primary sarcomas of the salivary glands occur rarely with few cases having been reported. Fibrosarcoma is a rare type of primary sarcoma with a poor prognosis and low life expectancy. Microscopically, these tumors resemble their counterparts encountered in other areas. They can be easily confused with spindle cell (sarcomatoid) carcinomas, myoepithelial carcinomas or malignant melanomas. We present an unusual case of a primary fibrosarcoma that arose in the left parotid gland of a 44 years male, who presented a swelling in the left parotid compartment. Computed tomography (CT) scan revealed a tumor arising from the left parotid gland. A near total parotidectomy was performed and the histopathology report a poorly differentiated malignant tumor proliferation of very probable carcinomatous origin infiltrating the parotid parenchyma brought back with peri-nervous sheath without vascular emboli or lymph node metastases (14N- / 14). On immunohistochemistry, the tumor was positive for vimentin and was negative for pancytokeratin and S-100, which lead to a firm diagnosis of fibrosarcoma.

KEYWORDS: Fibrosarcoma, Parotid gland, salivary gland, sarcoma.

INTRODUCTION

Parotid malignancies represent 1 to 3% of all head and neck cancers.[1], while sarcomas comprise only 0.3 to 1.5% of all major salivary gland neoplasms.[2] The most frequently described sarcomas of the parotid gland are rhabdomyo-sarcoma and malignant fibrous histiocytoma (MFH), although less commonly a variety of other tumors can occur, including leiomyosarcoma, malignant peripheral nerve sheath tumor (MPNST), angiosarcoma, osteo-sarcoma, fibrosarcoma, liposarcoma, hemangiopericy-toma, Kaposi sarcoma synovial sarcoma, and alveolar soft-part sarcoma.[2] Fibrosarcoma of the head and neck represents 5% of all fibrosarcomas. Microscopically, fibrosarcoma shows an intense proliferation of spindle shaped cells, varying a little in their size and shape and arranged in parallel bands which partly cross each other at acute angles (herring bone pattern).[3] They show a considerable morphological and histopathological overlapping with spindle cell (sarcomatoid) carcinomas, myoepithelial carcinomas and malignant melanomas. Hence, immunohistochemistry is mandatory to differentiate fibrosarcoma from the above-mentioned tumors.

Malignant spindle cell tumors within the salivary gland is a diagnostic challenge for the pathologist. Through this report, the authors are elucidating the existence of fibrosarcoma in the parotid gland and simultaneously emphasizing the need for a high index of suspicion to rule out other lesions with a similar appearance in order to reach the correct diagnosis and form an effective plan of management.

CASE REPORT

44-year-old male, Married and Father of 3 children, Taxi Driver, from a consanguineous marriage, Originally and resident in Ouarzazate, Morocco, Diabetic under oral antidiabetics, operated for a parotidectomy in 2005. The onset of symptoms seems to go back in March 2020 by the gradual installation of a swelling in the painful left parotid compartment without other associated signs, all developing in a context of deterioration of the general condition.The cervico-facial computed tomography (CT) revealed a voluminous tumor process of the poorly limited left parotid compartment measuring 63 * 63 * 53mm infiltrating the deep cervical spaces opposite with bilateral cervical lymph nodes of small sub-centimeter axis (Figu 5,6,7).

Patient underwent almost complete resection of the tumor except for a remainder on the internal carotid artery, the size of which does not exceed 1 cm. Extensive histopathological examination of the tumor, revealed a poorly differentiated malignant tumor proliferation of very probable carcinomatous origin infiltrating the parotid parenchyma brought back with peri-nervous sheath without vascular emboli or lymph node metastases (14N- / 14); to Immunohistochemistry;

A profile in favor of grade II fibrosarcoma according to FNCLCC by the positivity of anti-Vimentin antibodies (clone V9) and anti-ki67 antibodies (clone EP5) estimated at 60%.(figu 1,2,3).

Clinical examination revealed peripheral facial palsy with a positive Charles Bell sign, trismus, the well-healed parotidectomy wound, and exquisite pain points in the temporomandibular joint, malar, and left maxillary sinus. Thoraco-abdominal-pelvic computed tomography did not reveal any anomaly. The patient underwent the dosimetric scan.

After two weeks, the patient presented for a local recurrence of the parotidectomy compartment revealed by an inflammatory and hard mass, fixed to the two 3cm planes without palpable lymphadenopathy. Facial magnetic resonance imaging (MRI) revealed a large tumor process in the left parotid region measuring axially 57 * 52mm and sagittally 70mm responsible for invasion of deep spaces with sheathing of the carotids and involvement of the base of the skull (C1) without endocranial extension(figu 8,9,10).

The patient received adjuvant chemotherapy three courses of AI (Adriamycin 60mg / m2 + Ifosfamide 5g / m2).

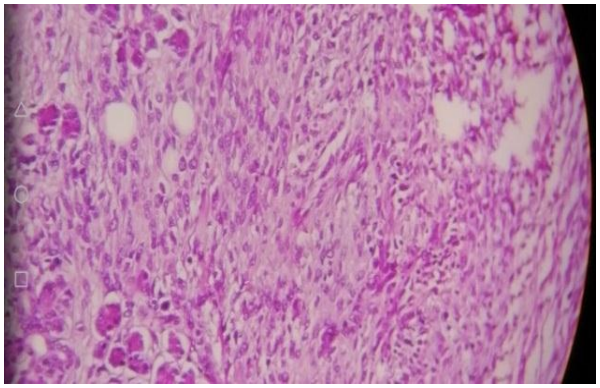

Figure 1: Spindle-shaped cells with a hyperchromatic nucleus, fusiform and lean fibrillar cytoplasm and high mitotic activity (HEx100).

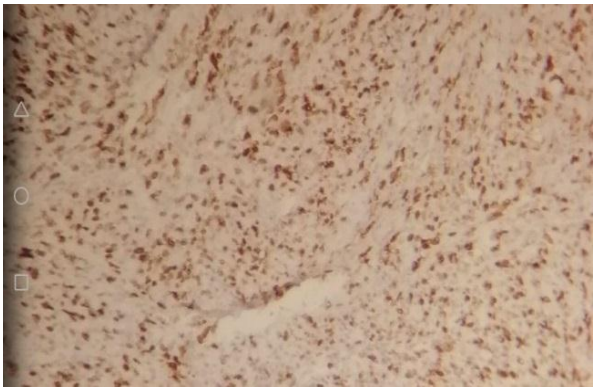

Figure 2: Nuclear labeling of tumor cells estimated at 60% anti-ki67 antibodies (x100).

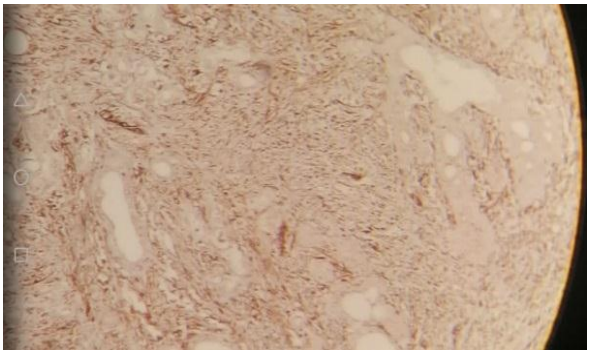

Figure 3: Immunostaining to vimentin showing cytoplasmic positivity in spindle cells (vimentin: 100 ×).

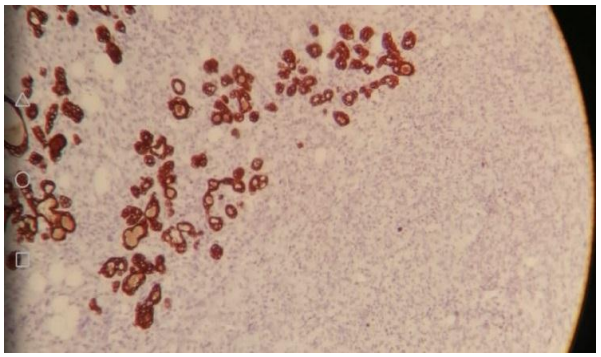

Figure 4: No labeling of tumor cells for anti-EMA antibodies (× 100).

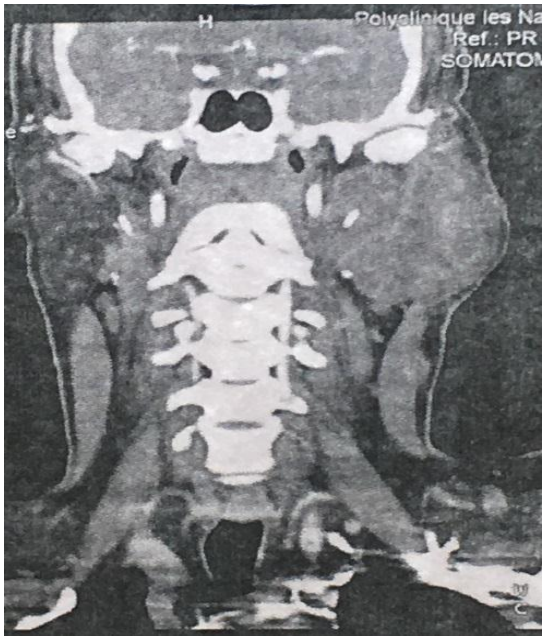

Figure 5: coronal cut showing an ill-defined tumor mass protruding from the left parotid gland.

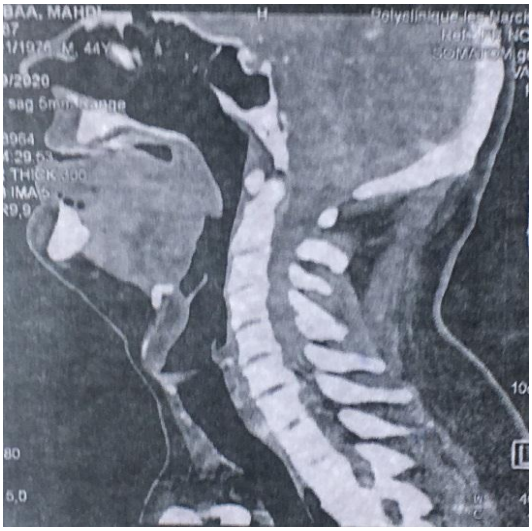

Figure 6: Sagittal section showing the tumor pushing back the deep cervical spaces.

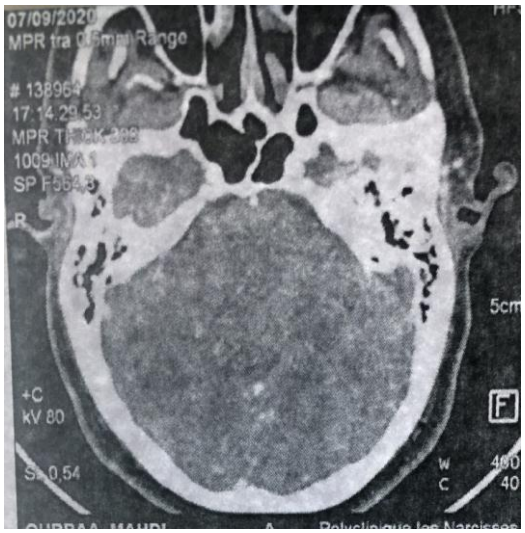

Figure 7: Axial section of a CT scan showing the process Tumor without endocranial extension.

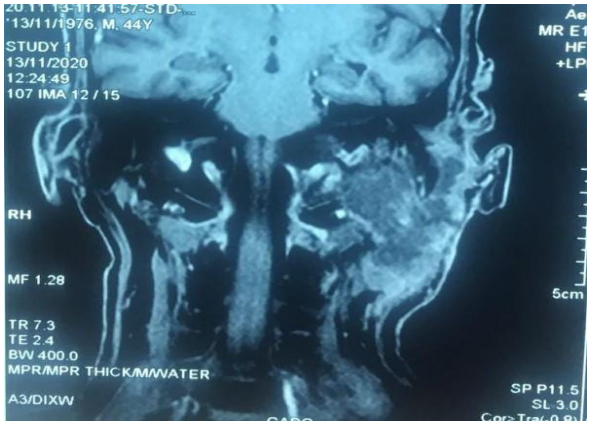

Figure 8: Coronal section of a facial MRI showing the left lateral cervical expansive process responsible for a infiltration of the posterior torn hole, the anterior arch of C1 without endocranial extension.

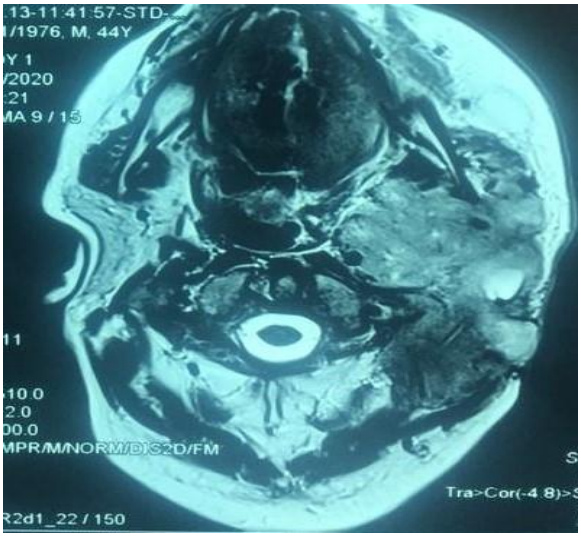

Figure 9: Axial section of a facial MRI showing voluminous tumor process in the left parotid region encompassing the external carotid, infiltrating the masticatory space of the submaxillary gland and intermaxillary commissure.

Figure 10: sagittal section of MRI showing the infiltration of the mastoid, the mastoid insertion of the sterno cleido muscle, the temporo mandibular joint and the mandibular condyle as well as the masseter.

DISCUSSION

Tumors of salivary gland originate a diverse group of neoplasms, and they may present significant challenges in the diagnosis as well as in their management. These tumors are rare with an overall incidence in the Western world of approximately 2.5 to 3.0 cases per 100,000 per year.[5] Malignant salivary gland tumors account for 0.5% of all malignancies and approximately 3 to 5% of all head and neck cancers.[5][6] The most common age of presentation for malignant tumors is in the sixth or seventh decade of life.[7][8] Ionizing radiation has been suggested as a cause of salivary gland cancer but the etiology of most salivary gland cancers is still unknown.[6][7][9][10] Certain

occupations are thought to be associated with an increased risk for salivary gland cancers including rubber products manufacturing, asbestos mining, plumbing, and some types of wood works.[7] The majority of the malignant tumors of salivary glands are epithelial in origin, whereas non epithelial tumors constitute less than 5%, of which nearly 90% are benign.[11]

Primary sarcomas of the parotid are an even rarer group of malignant salivary gland tumors arising from the mesenchymal component of salivary gland and comprises only 0.3 to 1.5% of all salivary gland neoplasms.[2] The two most common type of primary sarcomas of the salivary gland have been reported to be malignant schwannoma and fibrosarcoma.[4] Involvement of the parotid salivary glands are more common than the submandibular glands.[4] The mean age of occurrence has been reported to be 42 years for men and 38 years for women.[4] The most common presenting symptom is swelling followed by pain, tenderness or paralysis.

Fibrosarcoma can be the infantile or new adult type. The infantile type occurs inpatients < 2 years of age with an intermediate grade of prognosis and rare metastasis, whereas adult fibrosarcoma is a diagnosis of exclusion with a profound metastasizing potential and grave prognosis. Grossly, adult fibrosarcoma appears as a well circumscribed mass with firm white to tan cut surface.

Microscopy shows compact spindle cells arranged in short fascicles which split and merge, giving the appearance known as 'herring bone' pattern in a background of variable collagenous stroma. The individual cells are spindle to polygonal in shape with fibrillary eosinophilic cytoplasm and plump hyperchromatic nuclei. Certain conditions should be ruled out before reaching a diagnosis of fibrosarcoma of the parotid. Firstly, in order to rule out the possibility of metastatic sarcoma, four basic criteria should be met are as follows.

1. There should be no present or past history of a sarcoma elsewhere.
2. Metastasis to the gland from a primary in the skin or mucosa of the upper aerodigestive tract should be excluded.
3. The overall appearance should be consistent with originating from within the gland, rather than invasion of the gland by a sarcoma of the adjacent soft tissues
4. Multiple sectioning should be done to rule out a carcinosarcoma (so-called true malignant mixed tumor).[2]

Further, the microscopic appearance of the tumor can be easily confused with spindle cell (sarcomatoid) carcinomas, myoepithelial carcinomas, malignant melanomas and MPNST. But the following characteristics helps in limiting the diagnosis to fibrosarcoma, namely.

- Highly cellular spindle cell lesion
- Herring bone arrangement of cells
- Absence of pleomorphism
- S-100 or CK negativity. A study done by Auclair PL et al, highlighted the fact that 15% of previously diagnosed primary sarcomas of the salivary glands were proven later on to be of epithelial origin.Immunohistochemistry is, therefore, highly recommended to prevent such diagnostic error.

The most successful therapy is either parotidectomy (superficial or total) or a combination of surgery and radiation.[4] Wide surgical excision is the mainstay of treatment, although it is also known to be a chemosensitive tumor and as such adjuvant or neoadjuvant chemotherapy is also employed in its management, especially where a wide excision is difficult to achieve.[12]

The prognosis is poor and most of the patients either experience local recurrences or distant metastases and die of the disease. Mean survival time for those dying from the tumor has been reported to be 2.4 years, and a 5-year survival period remains a significant indicator of cure.[4]

CONCLUSION

Fibrosarcoma of the parotid gland is a very rare neoplasm, and it is a diagnosis of exclusion. In order to reach a correct diagnosis, metastasis from other body parts or adjacent soft tissue should be ruled out. Immunohistochemistry is essential to differentiate these tumors from mixed malignant tumors, melanomas and the tumors myoepithelial origin. The delay in diagnosis and treatment should be avoided in order to limit the progression of tumor stage and grade for a better prognosis.

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