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Carcinoma ex Pleomorphic Adenoma of the Palate with Paranasal Sinuses and Nasal Cavities Extension: A Case Report

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Abstract **Case Report**

Carcinoma ex pleomorphic adenoma (CXPA) of the minor salivary glands is very rare and most cases occur in the palate, tongue, cheek and lip. The authors report herein on a rare case of CXPA occupying the nasal cavities and the hard palate. A 63-year-old man, who presented with a painless swelling in palatal region, nasal obstruction, change in voice and recent onset of epistaxis. Computed tomography revealed an extensive lesion into the nasal cavities. Palatal and nasosinal biopsies were performed. Diagnosis was made on the basis of immunohistochemical study of biopsy samples. Treatment plan by surgical exeresis on an external approach was not accepted by our patient.

Key words: carcinoma ex pleomorphic adenoma, Minor salivary gland, palate, Nasal cavity, paranasal sinuses.

INTRODUCTION

Carcinoma ex pleomorphic adenoma (CXPA) also known as carcinoma ex mixed tumour develops from a primary or recurrent pleomorphic adenoma (PA) and constitutes approximately 12% of all salivary malignancies [1]. The role of Magnetic resonance imaging (MRI) is generally limited to guide diagnosis. Histopathologic with immunomarking should establish the final diagnosis. Treatment is surgical excision with negative margins and subsequent radiation therapy. We report an unusual location of this disease; CXPA of the hard palate with extension into the nasal cavities, and discuss the epidemiology, clinical features and treatment of this disease, in the light of a case report and a review of the literature.

CASE REPORT

A 63-year-old male presented to the otorhinolaryngology department with a history of a painless swelling in palatal region. History revealed that the swelling was detected 20 years ago. The mass was growing slowly. He also complained of nasal obstruction, change in voice and slight epistaxis on forced nose-blowing.

Intraoral examination revealed an oval-shaped, demarcated mass measuring 6cm x 5cm diameter in hard palate region. The mass was asymptomatic, firm, fixed to underlying bone and coating with smooth intact mucosa (Fig. 1). The anterior rhinoscopy found the nasal cavities completely obstructed by the swelling, which could not be penetrated by the rigid endoscope, with apparently healthy covering mucosa. The nasopharynx was healthy and no cervical lymph-adenopathy was detected. Computed tomography of head and neck showed a large, mixed tumour which originated from the palatal bone and extended inferiorly to the oral cavity and superiorly into the nasal cavities and paranasal sinus with bone lysis and total destruction of the nasal septum (Fig. 2). Histopathologic analysis of biopsy samples from the left nasal cavity and hard palate, taken under local anesthesia revealed pleomorphic adenoma with multiple carcinomatous transformation sites. The palatal component was documented as pleomorphic adenoma and the nasal component, as poorly differentiated carcinoma (Fig. 3). We diagnosed the lesion as a carcinoma ex pleomorphic adenoma. Due to the refusal of surgery by the patient, he was referred for chemo-radiotherapy as main therapeutic arm, however the patient has since been lost seen of.

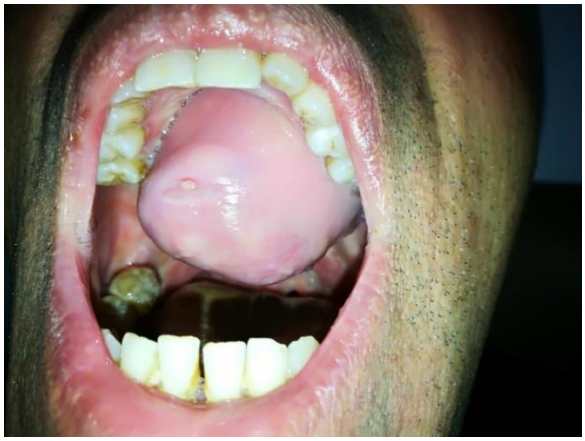

Fig-1: Clinical photograph showing the intraoral swelling on the palate

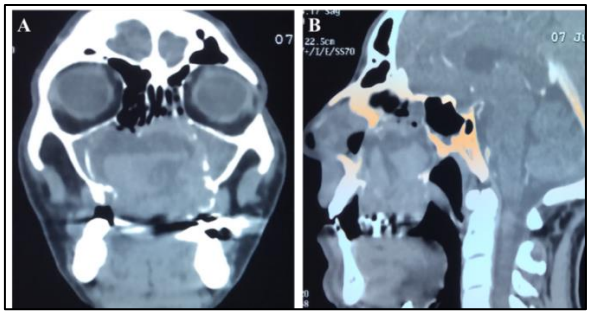

Fig-2: Huge mixed lesion in maxillary and ethmoid sinuses, nasal cavities and entire hard palate in coronal (A) and sagittal (B) slices of computed tomography view

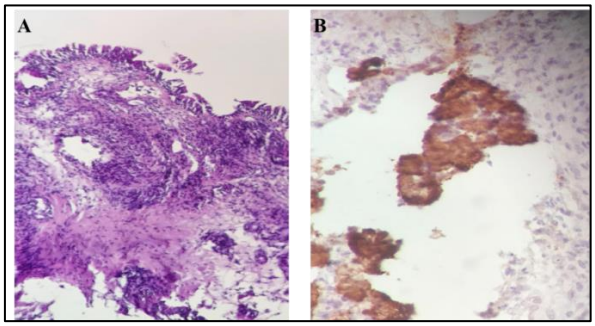

Fig-3: Histology, showing tumoral proliferation with carcinomatous transformation sites in the nasal cavities. HE staining, x40 (A) The tumour was composed of pleomorphic adenoma and Poorly-differentiated epithelial cell carcinoma (B) CK+

DISCUSSION

CXPA, also known as carcinoma ex mixed tumor, malignant mixed tumour, carcinoma ex adenoma is defined as a malignant epithelial component together with remnants of benign pleomorphic adenoma (PA) [2]. These tumours comprise 3.6% of all salivary gland neoplasms, 6.2% of all mixed tumours, and 11.6% of all carcinomas of salivary glands [1]. Most of the malignant changes occur in the major salivary glands, especially the parotid gland, but CXPA may originate from minor salivary glands sites, mainly in the soft and hard palate [2]. Although rare, CXPA cases have been described in nasal cavity. However, extension to the paranal sinuses and nasal cavities of palatal minor salivary gland tumours is extremely rare. There have been few publications [3]. CXPA usually occurs in the 6th-8th decades of life, with a female predominance [4].

The typical clinical presentation of CXPA includes asymptomatic, slow growing firm mass that is very similar to pleomorphic adenomas (PA). Histology usually finds malignant degeneration of a pre-existing pleomorphic adenoma into poorly or nondifferentiated adenocarcinoma after several years' evolution [5]. The imaging data for CXPA are usually nonspecific and demonstrate overlapping features of other malignant and benign salivary gland tumours [6].

MRI is very contributive. It found a tissular aspect, with hyposignal in T1 sequences and hypersignal in T2: this is a classic radiologic feature of PA [5,7]. The presence of y, irregular tumor margin, tumor infiltration into the surrounding tissue, signal heterogeneity and low signal on T2-weighted images, however, rather pointed to carcinomatous aggression, suggestive of malignant degeneration of a pre-existing adenoma. Grossly, CXPA is larger than ordinary benign mixed tumors and contains necrosis and hemorrhage [7].

The diagnosis of CXPA is confirmed by histologic and immunohistochemical analysis. Immunomarking determines the histologic tumor subtype [8].

The recommended treatment is surgical excision with wide margins and post-operative adjuvant radiation therapy [3, 5, 9]. However, this treatment plan was not accepted by our patient.

From the literature, the indication for postoperative radiation therapy depends on the tumour grade and the adequacy of the surgical margins [5, 9]. Recurrence, especially regional and distant metastasis, portends an extremely poor prognosis [10]. The 5-year survival rate of CXPA ranges from approximately 25% to 65% [10].

CONCLUSION

Malignant degeneration of a salivary adenoma with palatal location was very rarely met. Our case is exceptional because the size of tumor in hard palate was huge and extending into the paranasal sinuses and nasal cavities with bone lysis. The recommended treatment is surgical excision with wide margins and subsequent radiation therapy. Prognosis depends on stage of evolution, histologic type and persistence and quality of management.

REFERENCES

1. Gnepp DR. Malignant mixed tumours of the salivary glands: a review. Pathol Annu. 1993;28:279–328.

2. Hellquist H, Skalova A. Carcinoma ex pleomorphic pleomorphic adenoma. In Histopathology of the Salivary Glands, Springer-Verlag Berlin Heidelberg. 2014: 283-296.
3. Goepfert H, Luna MA, Lindberg RD, White AK. Malignant salivary gland tumors of the paranasal sinuses and nasal cavity. Arch Otolaryngol. 1983;109:662–8.
4. Moberger JG, Eneroth CM. Malignant mixed tumors of the major salivary glands. Special reference to the histologic structure in metastases. Cancer. 1968;21:1198–211.
5. Yazibene Y, Ait-Mesbah N, Kalafate S, Mesbah NY, Abdennebi A, Bouzid H, Kalem K. Degenerative pleomorphic adenoma of the nasal cavity. European annals of otorhinolaryngology, head and neck diseases. 2011 Jan 1;128(1):37-40.
6. Kato H, Kanematsu M, Mizuta K, Ito Y, Hirose Y. Carcinoma ex pleomorphic adenoma of the parotid gland: radiologic-pathologic correlation with MR imaging including diffusion-weighted imaging. American journal of neuroradiology. 2008 May 1;29(5):865-7.
7. Fushiki H, Morijiri M, Maruyama M, Motoshima H, Watanabe Y. MRI of intranasal pleomorphic adenoma. Acta oto-laryngologica. 2006 Jan 1;126(8):889-91.
8. Hirai S, Matsumoto T, Suda K. Pleomorphic adenoma in nasal cavity: immunohistochemical study of three cases. Auris Nasus Larynx. 2002 Jul 1;29(3):291-5.
9. Chen AM, Garcia J, Bucci MK, Quivey JM, Eisele DW. The role of postoperative radiation therapy in carcinoma ex pleomorphic adenoma of the parotid gland. International Journal of Radiation Oncology* Biology* Physics. 2007 Jan 1;67(1):138-43.
10. Weiler C, Zengel P, van der Wal JE, Guntinas-Lichius O, Schwarz S, Harrison JD, Kirchner T, Ihrler S. Carcinoma ex pleomorphic adenoma with special reference to the prognostic significance of histological progression: a clinicopathological investigation of 41 cases. Histopathology. 2011 Oct;59(4):741-50.