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CLINICAL REPORT

Degeneration of a Huge Pleomorphic Adenoma in the Nasal Cavity Extended to the Hard Palate: A Case Report and Review of the Literature

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Abstract The pleomorphic adenoma is a benign tumor. The patient was 55 years old, with no particular pathological history. For 18 months, she had a progressive and permanent right nasal obstruction associated with intermittent epistaxis, rhinorrhea and chronic unilateral tearing. The nasofibroscopy showed a smooth reddish bleeding tumor of the right nasal cavity, reaching the lower part of the middle cornet. Oropharynx examination revealed on the right side a sessile and bulging tumor of the palate with respect of the mucosa, the tumor goes through the choanae into the oropharynx. The CT scan showed a large aggressive lesion process centered on the right nasal cavity with bone lysis of the posterolateral wall of the maxillary sinus and invasion of the infratemporal fossa. MRI showed a maxillary right naso-sinusal tumor process infiltrating the ethmoidal cells, the right infratemporal fossa with endobuccal extension and filling the nasopharyngeal lumen. The patient was operated through the right external para-latero-nasal, associated to an endonasal abord. Anatomo-pathological with immune-histochemical study was performed to ensure complete removal of the tumor and it showed a high grade cystic adenoid carcinoma.

Keywords Huge pleomorphic adenoma · Degeneration · Nasal cavity · Cystic adenoid carcinoma

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Introduction

The pleomorphic adenoma is a benign tumor, called mixed by BROCA in 1866 to emphasize its heterogeneous aspects composed of both epithelial and myoepithelial cellular contingents. These cluster lesions are with imprecise boundaries, disjointed or scattered within a variable mesenchymal stroma: myxoid, hyaline, chondroid, or osteoid [1, 2]. It primarily affects the main salivary glands (80% in parotid gland and 8% in the sub maxillary gland) [2–4]. The accessory salivary glands are less affected with 6% of cases [2–4]. Pleomorphic adenoma is exceptional in the sero-mucosal glands of the nasal cavity, pharynx, larynx and trachea, or in the lachrymal glands. Malignant degeneration of Pleomorphic adenoma is unusual (1 to 5% of cases). Nasal-sinus localization is exceptional and malignant transformation in this site is unusual and extremely rare [5].

Case Presentation

The patient was 55 years old, with no particular pathological history. For 18 months, she had a progressive and permanent right nasal obstruction associated with intermittent epistaxis, rhinorrhea and chronic unilateral tearing without any other ENT or extra-ENT signs, all evolving in a context of apyrexia and conservation of the general state.

The nasofibroscopy showed a smooth reddish bleeding tumor of the right nasal cavity, reaching the lower part of the middle cornet; in the left nasal cavity, a significant septal deviation blocking the progression of the naso-fibroscope. Oropharynx examination revealed on the right side a sessile and bulging tumor of the palate with respect

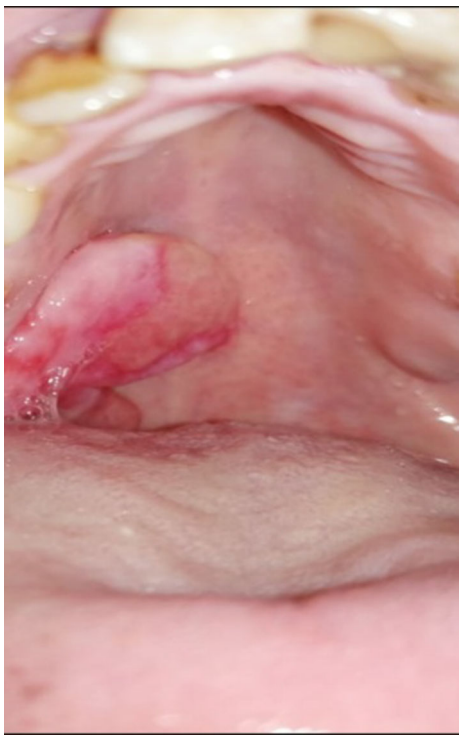

Fig. 1 Sessile and bulging tumor of the palate on the right side

of the mucosa, the tumor goes through the choanae into the oropharynx.

The CT scan showed a large aggressive lesion process centered on the right nasal cavity with bone lysis of the posterolateral wall of the maxillary sinus and invasion of the infratemporal fossa.

MRI showed a maxillary right naso-sinusal tumor process infiltrating the ethmoidal cells, the right infratemporal fossa with endobuccal extension and filling the nasopharyngeal lumen.

A biopsy with anatomopathological and immunohistochemical study was performed which concluded to a Pleomorphic adenoma. The patient was operated through the right external para-latero-nasal, associated to an endonasal abord. The initial exploration by naso-fibroscope discovered a fleshy bleeding tumor filling the entire right nasal cavity. The endonasal component was removed first, followed by a large maxillectomy, Finally, the endobuccal component and the hard palate were removed with curettage and milling around the bony defect of the palate with a passage to a macroscopically safe margins. The immediate and short term postoperative follow-up was simple. Anatomo-pathological with immune-histochemical study was performed to ensure complete removal of the tumor and it showed a high grade cystic adenoid carcinoma.

Discussion

Pleomorphic adenoma is the most frequent tumor of salivary origin (53%) [6, 7]. It is twice as frequent in the main salivary glands, with 65% in the parotid gland. Pleomorphic adenoma is the most common neoplasm of the minor salivary glands [4, 8], but compared to tumors of the major salivary glands, a greater proportion of accessory salivary gland tumors are malignant. This histological type accounts for 21–70% of all tumors arising from the accessory salivary glands, whether benign or malignant. It represents 70–100% of only benign tumors in this location [9–12].

The palate is a frequent site for the pleomorphic adenoma, found in 60% of cases [13]. Other sites are represented by the lips in 15% of cases [14], the cheeks in 12%, the long and the floor of the mouth in 5% each [15]. The oral mucosa, oropharynx and the para-nasal sinuses are less faced [16, 17], the localization of this tumor in the nasal

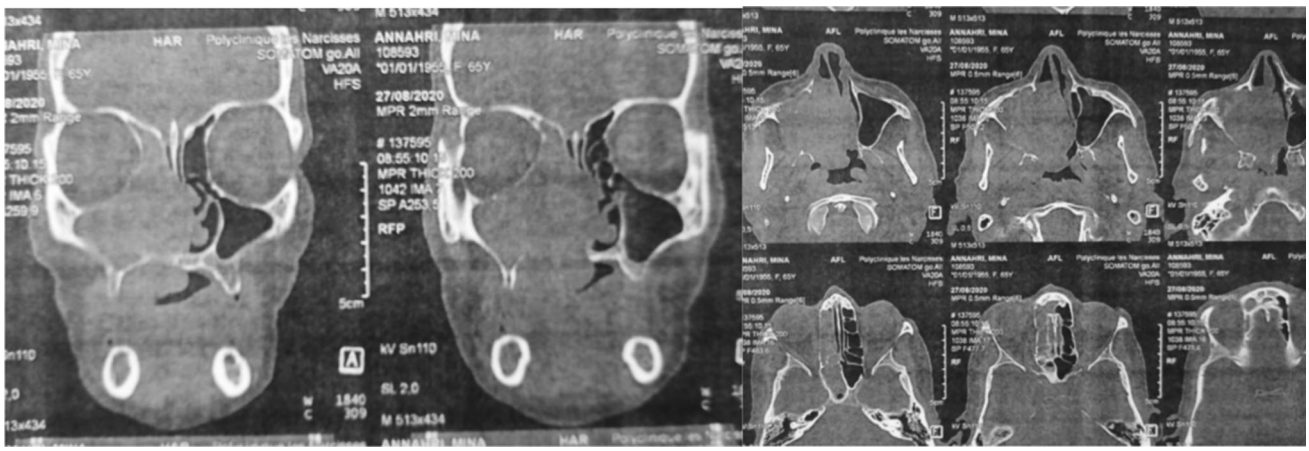

Fig. 2 Facial CT in axial (right) and coronal (left) sections: the right nasal cavity with bone lysis of the posterolateral wall of the maxillary sinus and invasion of the infratemporal fossa

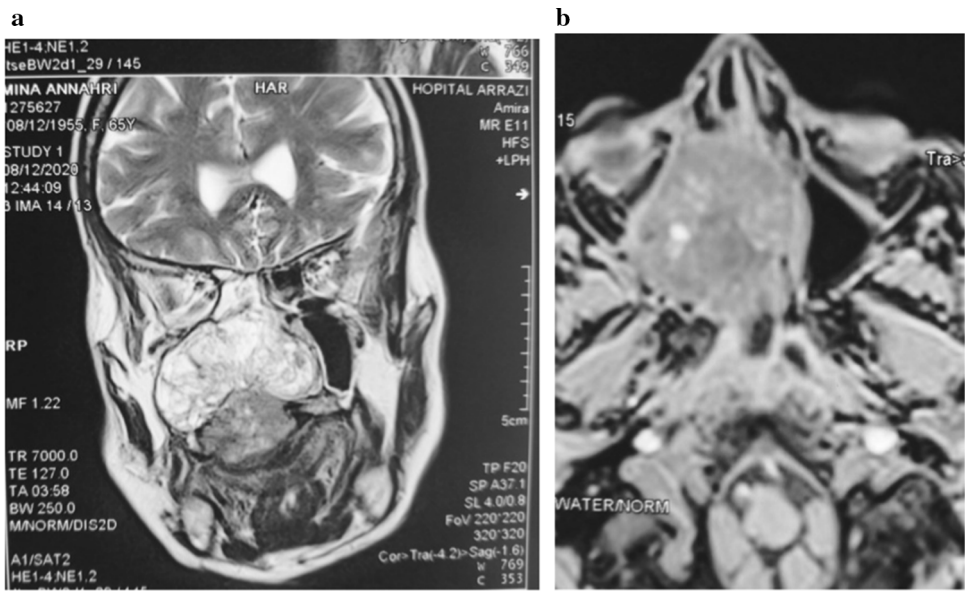

Fig. 3 Facial MRI in coronal section T2 sequence (**a**) and axial T1 after injection of Gadolinium (**b**) showing a right maxillary naso-sinusal tumor process infiltrating the ethmoidal cells and the right infratemporal fossa

cavity is rare [7]. The pleomorphic adenoma is usually solitary, but secondary metachronous or synchronous mixed tumor development within another salivary gland can be seen. In 90%, the tumor lesion is superficial [18]. There are possible associations with other tumors of salivary origin such as muco-epidermoid carcinoma, acinar cell adenocarcinoma, adenoid cystic carcinoma, but the most frequent association is Whartin's tumor (cyst-adenolymphoma) [14].

Computed tomography scan is the best radiological exploration of facial mass tumors in general [xx]. The presence of bone destruction images is a direct sign of malignancy. MRI showed a tissular mass, with T1 hyposignal and T2 hypersignal; these represent a classic radiological feature of pleomorphic adenoma [19]. However, the presence of bone lysis and the extra-sinusal extension are in favor of an aggressive carcinoma, which evoke a malignant degeneration of a pre-existing adenoma. Histopathological examination combined with immunostaining is the only examination that can confirm the diagnosis; it also specifies the histological subtype of the tumor [20].

Treatment is based on surgery, which must be carcinologically valid (wide excision). It may be combined to additional radiotherapy in case of insufficient resection [21–23], while data about chemotherapy are very insufficient because of the rarity of cases. The external surgical approaches are the best way to remove this type of malignant tumors, especially when it is outside the nasosinus cavities. Other surgical approaches, especially endoscopic maneuvers, can only be considered in small localized tumors [5]. The prognosis of these tumors depends on the evolution phase, histological type and persistence of the carcinomatous aggression, either by micro-lesions or by incomplete resection. These factors are the source of loco regional recurrence and distant metastases [5].

Conclusion

The nasal localization of pleomorphic adenoma is a rarely observed, and its malignant degeneration is even rarer. Extra-sinusal extensions with bone lysis are radiological signs that suggest malignancy. The only effective treatment is wide surgical excision. The external approach is preferred because it allows better control of the locoregional extension. The prognosis of these tumors remains poor and depends essentially on the histological type, the stage of evolution and the quality of initial management [5] (Figs. 1, 2, 3).

Declarations

Conflict of interest This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

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