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Case report

Intranasal pleomorphic adenoma: An unexpected diagnosis

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Abstract

Pleomorphic adenoma is a heterogeneous benign tumor of the salivary glands, with the parotid gland being the most common site of occurrence. However, its presence in the minor salivary glands is relatively rare. Here, we report the case of a 57-year-old man diagnosed with a nasal pleomorphic adenoma. In this context, we aim to clarify the epidemiological, diagnostic, therapeutic, and prognostic characteristics of nasal pleomorphic adenoma.

Keywords: pleomorphic adenoma, salivary glands, nasal cavity, benign tumor

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1. Introduction

Pleomorphic adenoma (PA), formerly called mixed tumor, is a heterogeneous benign tumor that affects the major salivary glands [1]. The parotid is the most affected salivary gland. However, it is unusual for it to arise in the accessory salivary glands, which are generally found in the oral mucosa and more rarely in the nasal cavity, larynx, or oropharynx [2]. It is worth mentioning that the majority of accessory salivary gland tumors are malignant, and those that are benign are at risk of transforming into malignant tumors [3]. Thus, we present the case of a 57-year-old man diagnosed with a nasal PA.

2. Case report

A 57-year-old male patient, a smoker with a 70 pack-year history who quit 8 years ago, and with a background of moderate obstructive sleep apnea-hypopnea syndrome, consulted for recurrent left epistaxis that had been present for a year, associated with homolateral nasal obstruction. There was no history of fever, weight loss, facial trauma, or facial pain.

Physical examination revealed a febrile, eupneic patient in good general condition. Nasal endoscopy showed a reddish-grey polyploid mass in the left nasal cavity, bleeding upon contact, and attached to an undistorted nasal septum. There were no palpable head and neck lymph nodes, and the cranial nerve examination was normal.

A computed tomography (CT) scan of the facial mass revealed a polypoid formation originating from the nasal septum, measuring 25.3 mm in the long axis. This mass occupied nearly the entire left nasal cavity without osteolysis, and there was focal polypoid thickening of the nasal mucosa (Fig. 1).

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The patient underwent an endoscopic excision of the tumor and the septal cartilaginous implantation base under general anesthesia. The excised material received in our department measured between 10 and 35 mm in the long axis. Histological examination revealed mucosa covered by regular pseudostratified columnar ciliated epithelium and occasional squamous epithelium. The underlying stroma contained a benign tumor with both epithelial and myoepithelial components. The epithelial component consisted of small tubular and glandular structures, lined with regular cubo-columnar cells, some of which contained eosinophilic material. These structures were surrounded by myoepithelial cells that had a plasmacytoid appearance, round nuclei, and granular chromatin, without atypia or mitotic figures. The stroma was fibrous with sparse chondro-myxoid areas (Fig. 2A and 2B). Immunohistochemical staining showed CK 7 positivity in the epithelial component (Fig. 3), while the myoepithelial cells were PS100 positive.

The postoperative recovery was uneventful. After six months of follow-up, the patient showed no signs of recurrence.

3. Discussion

Pleomorphic adenoma (PA) is the most common benign tumor of the salivary glands, primarily affecting the major salivary glands, with 84% of cases in the parotid and 8% in the submandibular glands. Only 4-6% of PAs occur in the minor salivary glands [4]. Among accessory salivary glands, the palate is most commonly involved (42.63%), followed by the upper lip, cheek, retromolar area, and floor of the mouth [2]. However, the nasal cavity is an uncommon site for PA [1-5]. The first report of a nasal PA was made in 1929 by Denker and Kahler, and the first large study series, including 40 cases, was published in 1973 by Spiro et al. [6, 7].

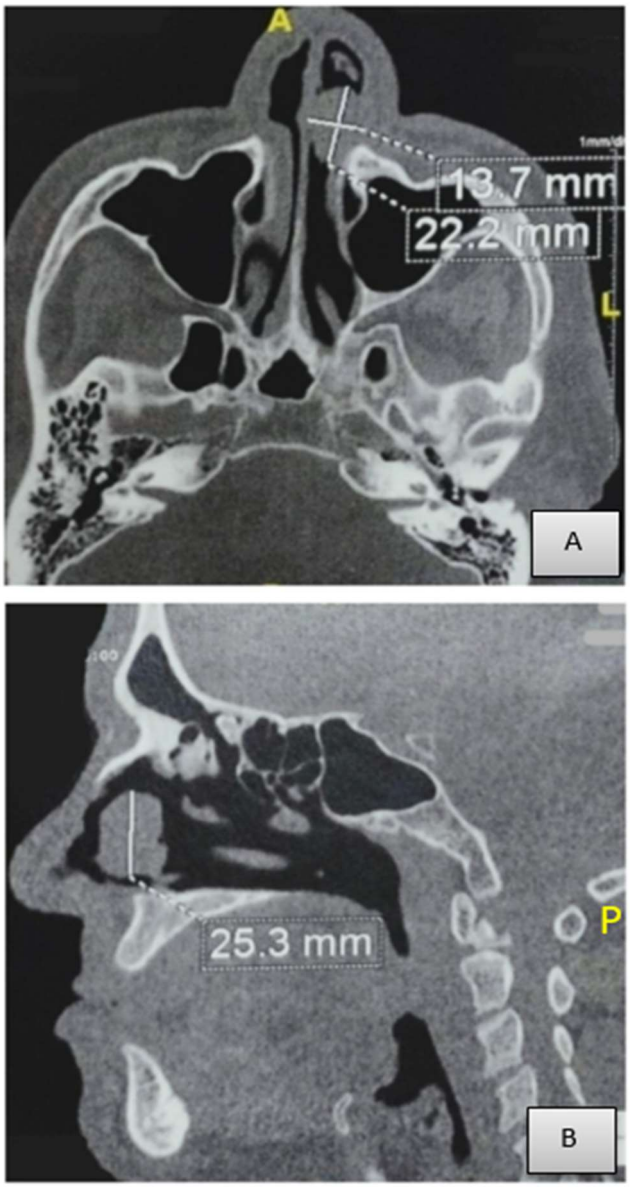

Fig.1. Axial section (A) and sagittal section (B) of a computed tomography scan showing a polypoid formation originating at the nasal septum filling almost the entire left nasal cavity without osteolysis.

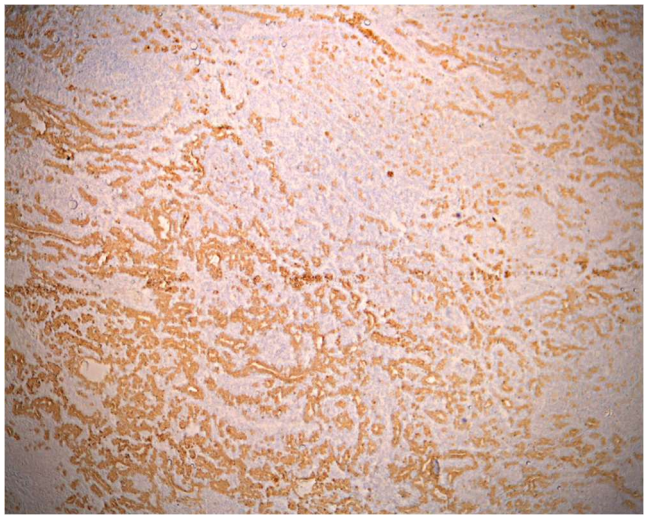

Fig.3. Positive staining for CK 7 highlighting the epithelial component (IHCX40).

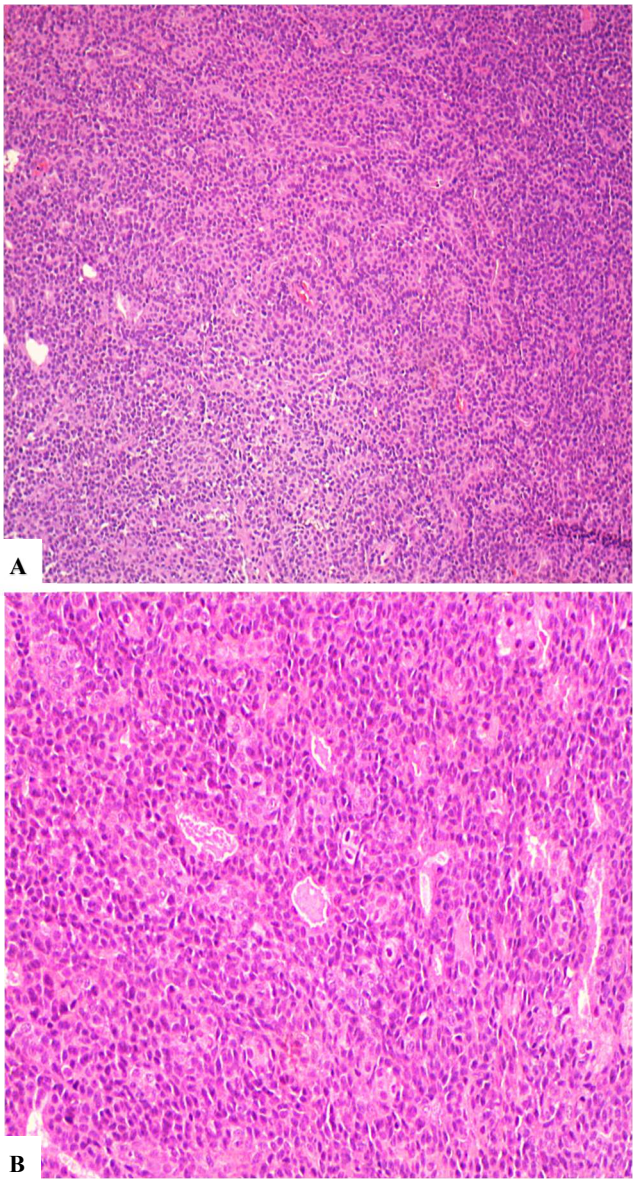

Fig. 2. The tumor is composed of mesenchymal and epithelial components. The epithelial component is made up of glandular and tubular structures of variable sizes (A: HEX100). These structures are filled with eosinophilic material and are surrounded by areas of myoepithelial cells that have plasmacytoid appearance (B: HEX200).

Subsequent series studies have been carried out by Compagno et al. (1977) with 41 cases, Suzuki et al. (1990) with 41 cases, Wakami et al. (1999) with 59 cases, and Vento et al. (2016) with 10 cases [6, 7].

Nasal PA typically presents a female predominance, unlike our patient, and occurs between the fourth and sixth decades of life, as in our case. However, it can affect individuals of any age [1, 5-8]. In our case, the tumor developed in the nasal septum, which accounts for about 80-90% of nasal PAs, while the lateral wall is affected in 20%, despite being rich in mucous and serous glands [5].

Nasal PA often grows slowly, and symptoms appear after a prolonged asymptomatic period [6-8]. Common symptoms include unilateral nasal obstruction, slowly growing nasal mass, nasal deformation, and pain. Intermittent epistaxis may also occur, as in our case. Less commonly, mucopurulent rhinorrhea and external

tumefaction of the nasal pyramid may be observed [6]. On endoscopic examination, nasal PA typically appears as a unilateral, well-circumscribed, pink/reddish, translucent, and soft polyp, with a regular surface. Its size can range from 0.7 cm to 7 cm [5, 6-8].

Nasal PA can present a diagnostic challenge, given the multitude of malignant and benign differential diagnoses, including epidermoid carcinoma, adenocarcinoma, adenoid cystic carcinoma, melanoma, olfactory esthesioneuroblastoma, and lymphomas, as well as benign conditions such as polyps, papillomas, angiofibromas, and osteomas [1-8]. Imaging is primarily done using CT scans and MRI [1]. CT scans are used to rule out an intracranial process and to assess loco-regional extension and pre-therapeutic evaluation [1]. On CT, nasal PA appears as a well-defined soft tissue mass, and it can also detect calcifications within the tumor and explore any bone changes [1, 5-7]. MRI shows nasal PA as low to intermediate signal intensity on T1-weighted images, intermediate to high signal intensity on T2-weighted images, and heterogeneous enhancement on contrast-enhanced images [7]. In our case, MRI was not requested. However, these radiological features alone are insufficient for a definitive diagnosis, and histopathological examination is essential [7].

Some clinicians recommend fine-needle aspiration cytology before surgery, as it provides diagnostic orientation [1]. However, the definitive diagnosis is based on histological examination of the surgical specimen [1, 5-6]. Nasal PA, like other PAs, consists of epithelial, myoepithelial, and mesenchymal components, which is why it is referred to as a "mixed tumor." Plasmacytoid myoepithelial cells are exclusively found in accessory salivary gland tumors [6]. Nasal PA differs from PAs in other locations by its high cellularity, significantly reduced stromal component, and lack of a capsule, which are unusual features in major salivary gland tumors [1,5-8]. As a result, nasal PA may be mistaken for a more aggressive epithelial tumor [1, 5-7]. Therefore, Shama et al. recommend considering PA in the differential diagnosis when a unilateral nasal obstruction is accompanied by epistaxis [6]. An expert in head and neck pathology is recommended, and immunohistochemical markers are necessary for a correct diagnosis. These include cytokeratins, S100 protein (used in our case), glial fibrillary acidic protein (GFAP), alpha-smooth muscle actin, vimentin, and Ki67 proliferation marker [7-9].

The prognosis of PA is generally good, but it may be complicated by recurrence and malignant transformation. Nasal PA tends to recur less frequently than PA of the parotid gland [1]. Recurrence risk factors include multinodularity, irregularity, or invasion of the pseudocapsule [8]. Malignant transformation of nasal PA occurs in 2%-6% of all salivary gland tumors, and the risk is higher with multiple recurrences [1-8]. Moreover, nasal septum-originating PAs have a higher risk of malignancy. Our patient had a benign PA with no signs of malignant transformation. Notably, in 1990, a single case of benign intranasal PA with submandibular lymph node metastasis was reported, despite having a benign microscopic appearance [7].

The treatment for intranasal PA is surgical excision, with wide margins [8]. It can be treated via endonasal endoscopic surgery, with the approach being influenced by the tumor's location and its proximity to the paranasal sinuses [7]. Since incomplete excision can lead to malignant transformation, complete excision with healthy margins and long-term follow-up are mandatory [9].

4. Conclusion

PA of the nasal cavity is rare and should be considered when a fleshy septal formation develops on healthy mucosa. Imaging is crucial to rule out any intracranial involvement, as well as to evaluate the extent of the lesion. The diagnosis relies on a careful histopathological examination, as nasal PAs are characterized by a predominance of the epithelial component compared to those of major salivary glands. This feature often leads to misdiagnosis as an aggressive epithelial tumor. Therefore, it is highly recommended to consult with an expert in head and neck pathology and use appropriate immunohistochemical markers for accurate diagnosis. Treatment involves wide surgical excision to prevent recurrence and malignant transformation. Given the potential for recurrence and malignant change, long-term loco-regional surveillance should be conducted for several years following excision to ensure optimal patient outcomes.

Consent of patient

Written informed consent was obtained from the patient for participation in this study.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Ethical considerations

Ethical approval was obtained from the Ethics Committee of the Taher Maâmouri University Hospital, Nabeul, Tunisia.

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Conflict of interest

None.

Authors' contribution

All authors listed have significantly contributed to the investigation, development and writing of this article. All authors approved the final version. AB : Writing-original draft. SN: Writing-review and editing. AD: Conceptualization. IN: Data collection. GK and EC: Study validation.

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