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A Rare Case of Pleomorphic Adenoma of the Nasal Cavity with Extensive Squamous Metaplasia: A Diagnostic Pitfall

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Abstract

Pleomorphic adenoma (PA) is one of the most common benign tumor of the major salivary glands. Its occurrence in the nasal cavity, particularly arising from minor salivary glands of the nasal septum, is rare. Due to its unusual location and diverse histomorphology, it is often associated with diagnostic challenges. We report a case of a 56-year-old female presenting with unilateral nasal obstruction of 3 months duration on the right side of the nose. Radiological evaluation revealed a well-defined lesion in the right nasal cavity arising from the anterior nasal septum without evidence of bone destruction. Histopathological examination established the diagnosis of PA with extensive squamous metaplasia and keratin pearl formation, imparting a morphology that closely mimics keratinizing lesions of the nasal cavity. In conclusion, PAs in the nasal cavity are uncommon and often show higher epithelial cellularity with reduced stromal components compared to their major salivary gland counterparts, which may lead to misdiagnosis, particularly in limited biopsy specimens. Recognition of such histological variations is essential to avoid overdiagnosis and aggressive treatment.

Keywords: Benign tumor, Histopathology, Nasal cavity, Nasal Tumor, Pleomorphic adenoma

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Introduction

Pleomorphic adenoma (PA) (benign mixed tumor) is the most common salivary gland neoplasm, predominantly arising in major salivary glands. However, similar tumors very rarely also appear in minor salivary glands, which are distributed throughout the body. Rare cases have been documented in the external auditory canal, lacrimal gland, soft and hard palate, and lip,[1-4] although respiratory system cases are incredibly uncommon.[5,6] These tumors most commonly originate from the mucous glands of the nasal septum and may present with non-specific symptoms such as nasal obstruction.

Sinonasal PAs differ histologically from their major salivary gland counterparts by exhibiting increased epithelial cellularity and relatively reduced stromal component, often leading to diagnostic confusion with malignant epithelial tumors.

We report a case of PA of the nasal septum with prominent squamous metaplasia and keratin pearl formation, an uncommon diagnosis at a rare site, posing a significant diagnostic challenge.

Case Presentation

A 56-year-old female presented to the Otorhinolaryngology department of Mata Gujri Memorial Medical College and Lions Seva Kendra Hospital, Kishanganj, Bihar, with complaints of progressive unilateral nasal obstruction for 3 months, on the right side. There was no history of epistaxis, trauma, or significant nasal discharge.

On clinical examination, a mass was noted in the right nasal cavity.

Radiological Findings

Contrast-enhanced computed tomography (CT) scan revealed a well-defined hyperdense lesion (~21 × 17 × 20 mm) arising from the anterior nasal septum, extending into the right nasal cavity. The lesion showed minimal enhancement and caused mild adjacent bony scalloping without evidence of bone destruction [Figure 1a].

Radiological impression suggested a benign nasal cavity lesion with differential diagnoses including: (1) Pyogenic granuloma, (2) Hemangioma, (3) Nasal polyp, (4) Inverted papilloma.

Further excision and histopathological examination was advised and eventually mass was excised under local anesthesia and sent to the Department of Pathology for histopathological examination.

Histopathological Examination

We received a greyish-white piece of firm tissue measuring (2.5 × 2 × 1.5 cm) in size. The cut surface showed shiny white homogenous areas [Figure 1b].

Following standard processing, 3–4 mm thick paraffin slices were cut and histopathological examined using eosin and hematoxylin.

Microscopy

Sections studied show a well-circumscribed neoplastic lesion with overlying respiratory epithelial lining as well as focal metaplastic squamous epithelial lining (Figure 1a). The neoplastic lesion

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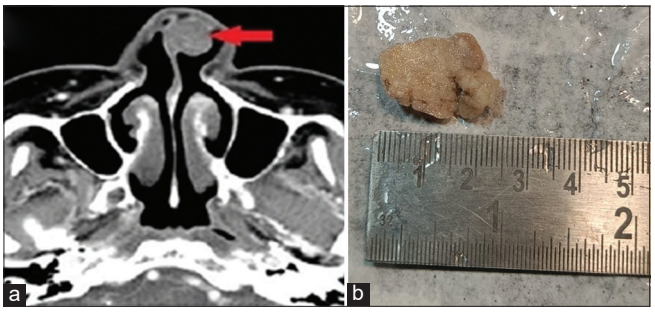

Figure 1: (a) Computed tomography scan showing a soft-tissue margin right nasal cavity arising from the nasal septum. (b) Gross specimen of (2.5 × 2 × 1.5) cm, brown-colored, cut surface showing homogenous brown areas

is arranged in tubules, cords, and sheets, comprising ductal epithelial cells with bland nuclear features (Figure 2b) along with myoepithelial cells of spindle-shaped cells as well as clear cell types (Figure 2c). Surrounding areas show scanty to moderate amount of stroma of chondromyxoid nature (Figure 2d). Few dilated mucinous glands are also noted with intraluminal mucin secretion and presence of mucinophages.

A striking feature is:

Extensive squamous metaplasia (Figure 2a)

No significant cytological atypia, necrosis, or increased mitotic activity is noted.

Final diagnosis

PA of nasal septum with extensive squamous metaplasia.

Discussion

A relatively uncommon benign tumor known as PA of the nasal cavity is sometimes detected in teenagers and is more common in female patients between the ages of 40 and 60.[7,8] It is hypothesized that it results from a viral infection, misplaced ectodermal tissue, or a reminisce of the vomeronasal epithelial duct on the cartilaginous nasal septum. Another requirement is that ectopic embryonic epithelialized cells that move during the migration of nasal buds serve as progenitors for PAs.[9]

PA of the nasal cavity is rare, with most cases arising from the nasal septum due to the presence of seromucinous glands. Clinically, patients present with unilateral nasal obstruction, sometimes accompanied by epistaxis.

A CT scan is therefore required to detect involvement and bone loss. Surgical treatments include excision through endonasal endoscopic surgery or lateral rhinotomy, depending mostly on the size and location of the tumor.

Histologically, sinonasal PAs differ from their salivary gland counterparts by:

- Higher epithelial cellularity
- Reduced stromal component
- Frequent squamous metaplasia

These features may mimic: Squamous cell carcinoma, inverted papilloma, keratinizing lesions.

In the present case, abundant squamous metaplasia with keratin pearls created a strong diagnostic pitfall toward

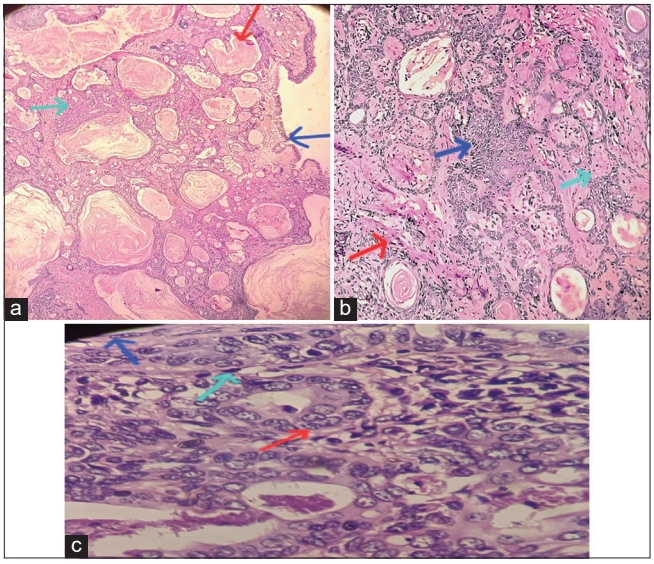

Figure 2: (a) Photo showing a tumor mass lined by pseudostratified ciliated respiratory epithelium (blue arrow), keratin plugging (red arrow), and tumor cells arranged in sheet pattern (cyan arrow) (Hematoxylin and Eosin [H&E], ×40). (b) Tubular (cyan arrow) and sheet-like arrangement (blue arrow) of tumor cells. The surrounding chondromyxoid stroma is relatively scanty (red arrow) (H&E, ×100). (c) Photomicrograph showing ductal epithelial cells forming a tubular pattern(red arrow) surrounded by myoepithelial cells of spindle type(cyan arrow) as well as clear type myoepithelial cells (blue arrow) (H&E, ×400)

malignancy. Occasionally, PAs have little to no stroma and are virtually entirely made up of epithelial cells. This often results in a cancer being incorrectly diagnosed.

After surgical excision, recurrence rates range from 0 to 8%, and recurrent recurrences raise the chance of cancerization.[10] Predominantly myxoid stroma, an uneven or invaded capsule, and multinodularity are risk factors for recurrence.

In the absence of resection, the probability of malignant transformation is expected to be 1.5% within 5 years.[11] The mechanisms underlying malignant transformation, however, are still poorly understood, and there is currently no agreement on the most reliable histology indicators. According to Röijer *et al*., genetic determinants underlying the malignant development of PA may include the amplification and overexpression of HMGIC as well as perhaps MDM2.[12] The most frequent variety of a nasal septal neoplasm, carcinoma ex PA, which has the capacity to spread, is more likely to be malignant than those from other places in the nose. Although bone is the main metastatic location, the liver, local lymph nodes, and the lungs have also been implicated.[13]

Conclusion

PAs in accessory salivary glands are extremely uncommon; they are even more uncommon in the nasal cavity, where they often develop from the nasal septum. When opposed to their major salivary gland counterparts, the ones that develop in the nasal cavity tend to have a higher epithelial and a lower stromal component, and they may be misdiagnosed at an early stage, leading to aggressive therapy. Therefore, if a patient presents with unilateral nasal obstruction or epistaxis caused by a tumor, this diagnosis should be given careful consideration.

Ethical Declaration

Ethical clearance was taken from the competent authority for this work.

Acknowledgment

No additional contributions were made by anyone apart from the authors of this manuscript.

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