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Magnetic Resonance Imaging and Histopathology in a Case of Pleomorphic Adenoma of a Minor Salivary Gland in the Nasal Cavity

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest

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Conflict of interest: None declared

Patient: Male, 56

Final Diagnosis: Pleomorphic adenoma

Symptoms: Epistaxis
Medication: —
Clinical Procedure: —

Specialty: Radiology

Objective: Rare disease

Background: Pleomorphic adenoma of the salivary gland is a common benign tumor of the parotid gland. However, pleomorphic adenoma arising in the minor salivary glands, including the nasopharynx, is uncommon. This report is of a case of nasal pleomorphic adenoma in which the preoperative imaging findings were consistent with the

histological features of pleomorphic adenoma of the minor salivary gland, which differs from that of pleomor-

phic adenoma of the major salivary gland.

Case Report: A 56-year-old Japanese man was referred to the department of otolaryngology following nasal endoscopy per-

formed at a referral center that identified a right nasal mass. The findings of contrast-enhanced magnetic resonance imaging (MRI) were consistent with a pleomorphic adenoma arising in the nasal septum. Needle biopsy and histology confirmed the diagnosis of pleomorphic adenoma. The tumor was resected using endoscopic surgery, and the histology confirmed the diagnosis of pleomorphic adenoma covered by normal nasal mucosa.

Conclusions: This case showed that pleomorphic adenoma arising in the nasal minor salivary gland had typical MRI and his-

tological findings and was a submucosal lesion. Preoperative MRI supported the approach to surgical resection

to ensure complete excision.

MeSH Keywords: Adenoma, Pleomorphic • Magnetic Resonance Imaging • Nasal Cavity • Salivary Glands, Minor

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Background

Pleomorphic adenoma is a benign mixed tumor of the salivary gland that contains both epithelial and mesenchymal tissue. Pleomorphic adenoma of the minor salivary glands is uncommon, and the incidence is between 6–7%, with reported origins that are variable and include the oral cavity, paranasal sinuses, and nasal cavity [1,2]. Pleomorphic adenoma arising in the minor salivary gland of the nasal cavity is less common [3,4], with a reported incidence of 0.4% [5]. The main symptoms caused by nasal tumors are epistaxis and nasal obstruction, but other symptoms may occur, depending on the specific tumor location [1]. Nasal pleomorphic adenomas are often symptomatic and detected and resected in the early stages without detailed evaluations, which may explain the high rate of recurrence. There have been few studies have described and compared the preoperative imaging findings with the pathological features of pleomorphic adenoma arising in the minor salivary glands.

A case is reported of a 56-year-old man with a benign pleomorphic adenoma arising in the minor salivary gland of the nasal cavity, with the diagnosis confirmed on histology and with preoperative magnetic resonance imaging (MRI) that allowed complete surgical resection to be performed.

Case Report

A 56-year-old Japanese man with a five-year history of worsening epistaxis from the right nasal cavity was referred to our otolaryngology department after a nasal mass was identified on the right side by nasal endoscopy performed at the referring medical facility. He was a nonsmoker with a medical history of glaucoma and childhood asthma, both of which were well-controlled without treatment. The patient denied any history of heavy alcohol consumption or substance abuse.

All serum tumor markers were in the normal range, as follows: carcinoembryonic antigen (CEA), 2.7 ng/mL (normal, <5.0 ng/mL); carbohydrate antigen 19-9 (CA19-9), 1.8 U/mL (normal, <37 U/mL); and squamous cell carcinoma antigen (SCCA), 0.9 ng/ml (normal, <1.5 ng/ml).

Nasal endoscopy identified a hemorrhagic mass in the right nasal cavity (Figure 1), with the attachment in the right inferior nasal turbinate or nasal septum. Non-enhanced computed tomography (CT) was performed (Figure 2A, 2B), which showed a mass in the anterior region of the right nasal cavity. There was no calcification in the mass or osteoclastic change in the surrounding bone. Contrast-enhanced magnetic resonance imaging (MRI) was performed that showed a well-defined mass with low signal intensity on T1-weighted imaging

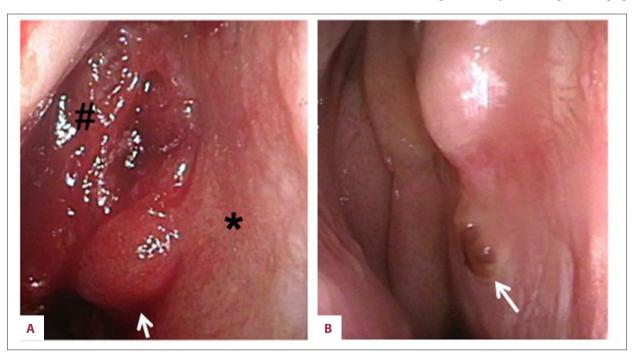


Figure 1. Nasal endoscopy of the right nasal cavity in a 56-year-old man with a nasal pleomorphic adenoma. (A) The preoperative image on endoscopy of a nasal pleomorphic adenoma arising in a minor salivary gland. A hemorrhagic mass (white arrow) is present in the right nasal cavity. The attachment site of the mass is suspected to be in the right inferior nasal turbinate (#) or the nasal septum (*). (B) The postoperative image on endoscopy obtained at three-month follow-up after surgery. The nasal mass was resected, and a scab is present in the nasal septum (white arrow).

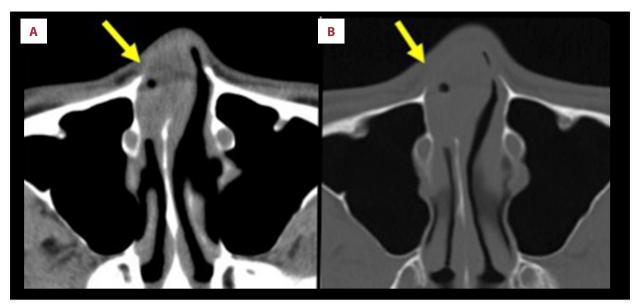


Figure 2. Non-enhanced computed tomography (CT) images of an intranasal pleomorphic adenoma in a 56-year-old man. (A, B) The computed tomography (CT) images show a mass measuring 14×26×25 mm in the anterior part of the right nasal cavity. The CT value of the mass is 46 Hounsfield units (HU). No fat or calcification is identified in the mass, and there are no osteoclastic changes in the surrounding bone.

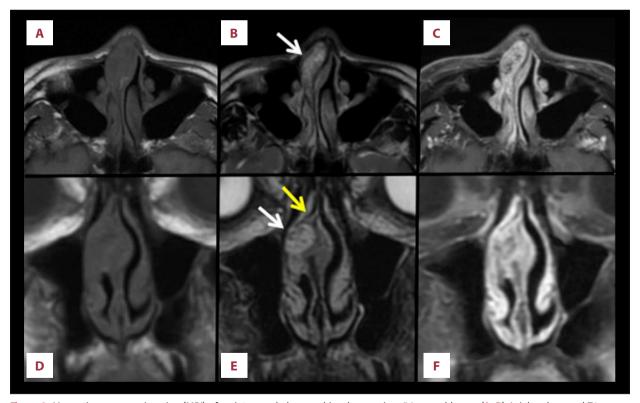


Figure 3. Magnetic resonance imaging (MRI) of an intranasal pleomorphic adenoma in a 56-year-old man. (A, D) Axial and coronal T1-weighted imaging shows a mass with low signal intensity in the right nasal cavity. (B) An axial T2-weighted image shows a tumor with heterogeneous low and slightly high signal intensity. (E) A coronal T2-weighted image shows a structure with low signal intensity that is connected to the nasal septum. The white arrow indicates the tumor, and the yellow arrow indicates the structure with a 'beak sign,' which suggests that it is a submucosal tumor and the attachment site is the nasal septum. (C, F) A contrastenhanced T1-weighted image shows heterogeneous enhancement without tumor infiltration into the surrounding tissues.

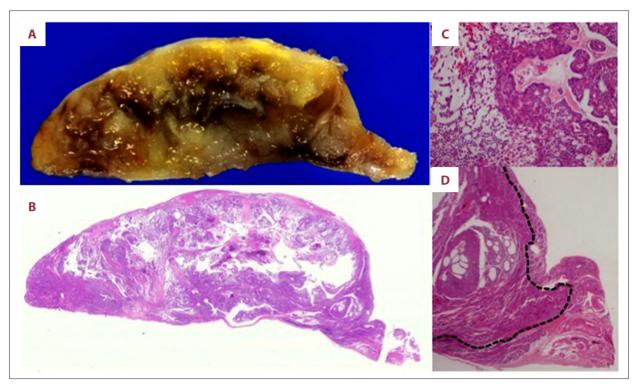


Figure 4. The macroscopic appearance of the resected tumor and photomicrographs of the histopathology of the resected intranasal pleomorphic adenoma from a 56-year-old man. (A) The macroscopic appearance of the resected pleomorphic adenoma. The cut surface of the tumor is light yellow to tan in color, mainly solid, and partly cystic. (B) Photomicrograph of the whole-mount view shows that the tumor is entirely covered by normal nasal mucosa. Hematoxylin and eosin (H&E). (C) Photomicrograph of the histology of the resected tumor shows epithelial components forming structures with lumens and myxoid stromal tissue. There is no atypia and no mitotic figures are present. Hematoxylin and eosin (H&E). (D) The left side of the broken line is a pleomorphic adenoma, and the right side of the broken line is the normal nasal mucosa. The attachment site of the tumor consists of the normal nasal mucosa. Hematoxylin and eosin (H&E).

(T1WI) (Figure 3A, 3D), and heterogeneous low to moderately high signal intensity on T2-weighted imaging (T2WI) (Figure 3B, 3E). Also, there was a structure between the mass and the nasal septum that showed a slightly low signal intensity on T2WI (Figure 3B, 3E), and heterogeneous enhancement in fat-suppressed, contrast-enhanced T1WI (Figure 3C, 3F), and a 'beak sign' that suggested the presence of a submucosal tumor (Figure 3E). There was no tumor infiltration into the surrounding structures.

Needle biopsy and histopathology confirmed the diagnosis of a benign pleomorphic adenoma, and endoscopic surgery was performed. The nasal tumor was identified as a submucosal tumor located at the nasal septum. The tumor was resected with a clear margin, and reconstruction and resection of the cartilage and inferior turbinate were not required (Figure 1A, 1B). Histopathology confirmed the diagnosis of benign pleomorphic adenoma and confirmed that the tumor was completely resected (Figure 4A). The histology showed that the tumor was composed of intermixed myxomatous cells and epithelial cells with no obvious atypia (Figure 4C), covered by normal nasal

mucosa (Figure 4B, 4D), without a pseudocapsule (Figure 4B). Following resection and the confirmed diagnosis of benign nasal pleomorphic adenoma arising in a minor salivary gland, the patient underwent routine three-year follow-up, and no recurrence was found.

Discussion

In this report, the preoperative magnetic resonance imaging (MRI) findings in a case of nasal pleomorphic adenoma were well correlated with the histological features (Table 1). Preoperative magnetic resonance imaging (MRI) identified the area of the tumor and facilitated surgery to ensure adequate surgical resection.

Pleomorphic adenoma of the minor salivary gland is characterized by a moderately high signal on T2-weighted imaging (T2WI) because of the reduced presence of myoepithelial cells, which produce a myxoid stroma [6]. In the present case, these imaging findings were present and supported the

Table 1. Comparison of the features of pleomorpic adenoma of the major salivary gland and minor salivary gland in terms of the histopathology and magnetic resonance imaging (MRI) findings.

	Pleomorphic adenoma of the major salivary gland	Pleomorphic adenoma of the minor salivary gland
Histopathology		
Epithelial cells	Scarce	Abundant
Myoepithelial cells	Abundant	Scarce
Pseudo-capsule	Present	Absent
MRI features		
T2-weighted image	High	Moderately high
Pseudo-capsule	Present	Absent

diagnosis of pleomorphic adenoma (Table 1). Pleomorphic adenoma arising in a minor salivary gland has been previously shown to present as submucosal tumors without a pseudocapsule [7]. In this patient, no pseudocapsule was identified on MRI. In previously reported cases of nasal pleomorphic adenoma with imaging findings [7,8–10], and a previous report published by Ozturk et al. [7] documented that a submucosal lesion was identified by computed tomography (CT) and MRI.

The histopathology of pleomorphic adenoma of the minor salivary gland has previously been reported to show a submucosal tumor with no pseudo-capsule and more epithelial components when compared with pleomorphic adenoma of the major salivary gland [6,11–13]. In the present case, the characteristics of the pleomorphic adenoma of the major salivary gland, such as the high signal intensity on T2WI MRI and a pseudocapsule, were not found.

Because preoperative MRI was able to define the margins of the tumor in this case, it was possible to perform an adequate surgical resection. Most cases of nasal pleomorphic adenoma are benign and are diagnosed and resected in the early stage because they present with symptoms such as epistaxis, as in the present case. Pleomorphic adenoma is classified as a benign tumor. However, between 3–4% of cases of pleomorphic adenoma show malignant transformation [3], and 2.4% of cases of nasal pleomorphic adenoma have been reported to undergo transformation [14]. Postoperative local recurrence has been reported in 7.5–8.8% of cases of nasal pleomorphic adenoma [6,11]. Also, there have been rare cases of the development of low-grade malignancy in nasal pleomorphic adenoma, known as 'carcinoma ex pleomorphic adenoma' [15]

and cases that metastasized [16]. Nasal pleomorphic adenoma has low radiosensitivity and a poor response to chemotherapy and so surgical resection is the first choice of the treatment [17]. Therefore, the margin of surgical resection should be carefully identified. In the present cases, no recurrence was noted at three-year follow-up, which may have been due to the careful identification of the tumor margins by preoperative MRI, and the detailed preoperative evaluation.

Unlike pleomorphic adenoma of the parotid glands, preoperative diagnosis of nasal pleomorphic adenoma remains challenging, even when the tumor is submucosal. The differential diagnosis of submucosal tumors of the nasal cavity includes nonepithelial tumors such as schwannoma, hemangioma, lymphoma, chondroma, chondrosarcoma, monomorphic adenoma, as well as pleomorphic adenoma [18,19]. Therefore, establishing the diagnosis may be difficult in some cases, and the diagnosis may need to be established by imaging studies and histopathology. As this case has shown, preoperative imaging using MRI can identify the extent of the tumor and its margins, which helps to ensure adequate surgical resection and reduce the rate of tumor recurrence.

Conclusions

An uncommon case of pleomorphic adenoma of the minor salivary gland of the nasal cavity is presented that demonstrates the role of preoperative magnetic resonance imaging (MRI) and its correlation with the histological features. MRI confirmed the presence of a submucosal tumor and identified the boundaries of the tumor to ensure adequate surgical resection.

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