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Case Report

Clinical and pathological features in a case of angiomatous nasal polyp

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Abstract: Angiomatous nasal polyp (ANP) is a relatively rare benign lesion, which may be misdiagnosed as a benign or malignant tumor. The characteristic pathological features of ANP are extensive vascular proliferation, accumulation of extracellular amorphous eosinophilic material, and atypical stromal cells. ANP can grow rapidly and exhibit aggressive clinical behavior that could simulate a malignancy preoperatively. We reported a case of ANP of the right maxillary sinus. A 38-year-old man presented with a 2-month history of right-sided nasal obstruction, fulvous rhinorrhea, headaches, right-sided tooth root pain under right face pressure, slight hyposmia and right-sided epiphora. Computed tomography (CT) imaging and Magnetic resonance imaging (MRI) revealed an heterogeneous mass of the right maxillary sinus, which caused extensive bone erosion and extended into the right nasal cavity and the ipsilateral pterygopalatine fossa. The mass was completely resected by endoscopic sinus surgery and had no evidence of recurrence during the follow-up period.

Keywords: Angiomatous nasal polyp (ANP), maxillary sinus, diagnosis, endoscopic sinus surgery

Introduction

Angiomatous nasal polyp (ANP), also known as angioectatic polyp, is a relatively rare benign lesion [1-4]. Based on the predominant elements seen on histological evaluation, inflammatory sinonasal polyps (SNPs) have been classified into five types: edematous, glandular, fibrous, cystic, and angiectatic or angiomatous [2, 5, 6]. Although SNPs are the most common sinonasal lesions examined pathologically, as an uncommon subtype ANP only accounting for 4%-5% of all SNPs [1-3, 5]. Angiomatous nasal polyps (ANPs) are characterized by extensive vascular proliferation and ectasia, with scanty inflammatory infiltrate and abundant extracellular fibrin [2, 4, 5]. In a relatively uncommon presentation, ANPs can grow rapidly and exhibit aggressive clinical behavior such as extensive bone erosion and remodeling or epistaxis which could simulate malignancy preoperatively, and so be a source of diagnostic difficulty [2, 5]. However, to the best of our knowledge, no authors reported ANP of the maxillary sinus caused extensive bone erosion and extended into pterygopalatine fossa in the English-language literature.

In this article, we present an extremely rare case of ANP of the right maxillary sinus that caused extensive bone erosion and extended into the right nasal cavity and the ipsilateral pterygopalatine fossa, and we further describe the clinical, radiological and pathological features of ANP.

Case report

A 38-year-old man presented with an 2-month history of right-sided nasal obstruction, fulvous rhinorrhea, headaches, right-sided tooth root pain under right face pressure, slight hyposmia and right-sided epiphora. He denied postnasal drainage, epistaxis, facial numbness, diplopia, and any impairment in his visual acuity. He was a non-smoker and had no significant past medical history.

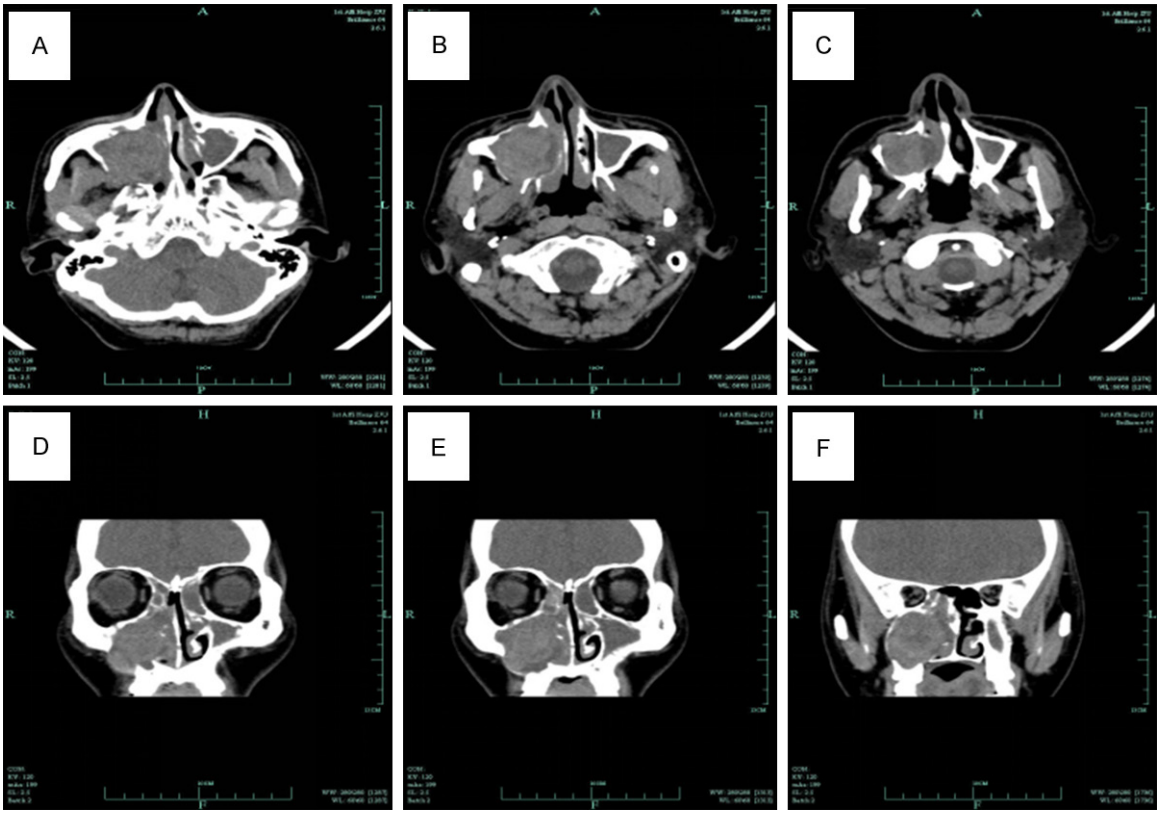

Figure 1. Axial (A-C) and coronal (D-F) CT scan demonstrated an heterogeneous soft-tissue mass occupying the entire right maxillary sinus with erosion of orbital floor, medial, anterior, inferior and posterior walls. The mass extended into the right nasal cavity and the right pterygopalatine fossa.

On otolaryngological examination, we found that the right-sided nasal mucosa were congested and edematou, bilateral inferior turbinate were swelling, while the nasal passage for peering was not clear. On other physical examination, the patient's blood pressure and other vital signs were normal. His visual acuity, eye movement, direct pupillary reflex and indirect pupillary reflexes were normal.

Axial and coronal computed tomographic scan of the paranasal sinuses demonstrated lamellar high-density shadow in both sides of the maxillary sinus, ethmoid sinus, frontal sinus and sphenoid sinus, as well as an heterogeneous soft-tissue mass in the right maxillary sinus and causing sinus wall swelling bony destruction. There was bone destruction involving the orbital floor, medial, anterior, inferior and posterior maxillary walls. The mass extended into the right nasal cavity and the ipsilateral pterygopalatine fossa (**Figure 1**). Magnetic resonance imaging (MRI) of the paranasal sinuses showed the right maxillary sinus component of the mass to have heterogenous hypointensity on T1-weighted images (T1WI) and heterogenous hyperintensity with a peripheral hypointense rim on T2-weighted images (T2WI). T2WI showed the mass was well-defined and the hyperintensity in the ipsilateral maxillary sinus indicated obstructive sinusitis. Diffusion weighted magnetic resonance imaging (DWI) showed the mass of slightly high signal intensity with a peripheral hypointense rim (**Figure 2**). The mass showed heterogeneous nodular and patchy enhancement with the non-enhanced peripheral hypointense rim after gadolinium contrast injection (**Figure 3**).

The patient was taken to the operating room with general anaesthesia and underwent surgical excision of the mass by endoscopic sinus surgery. At surgery, we found that there were yellow soft masses alternating with black necrotic areas occupying the entire right maxillary sinus. A biopsy was performed, the intraoperative frozen section diagnosis was inflammatory infiltrates and necrosis. Based on the

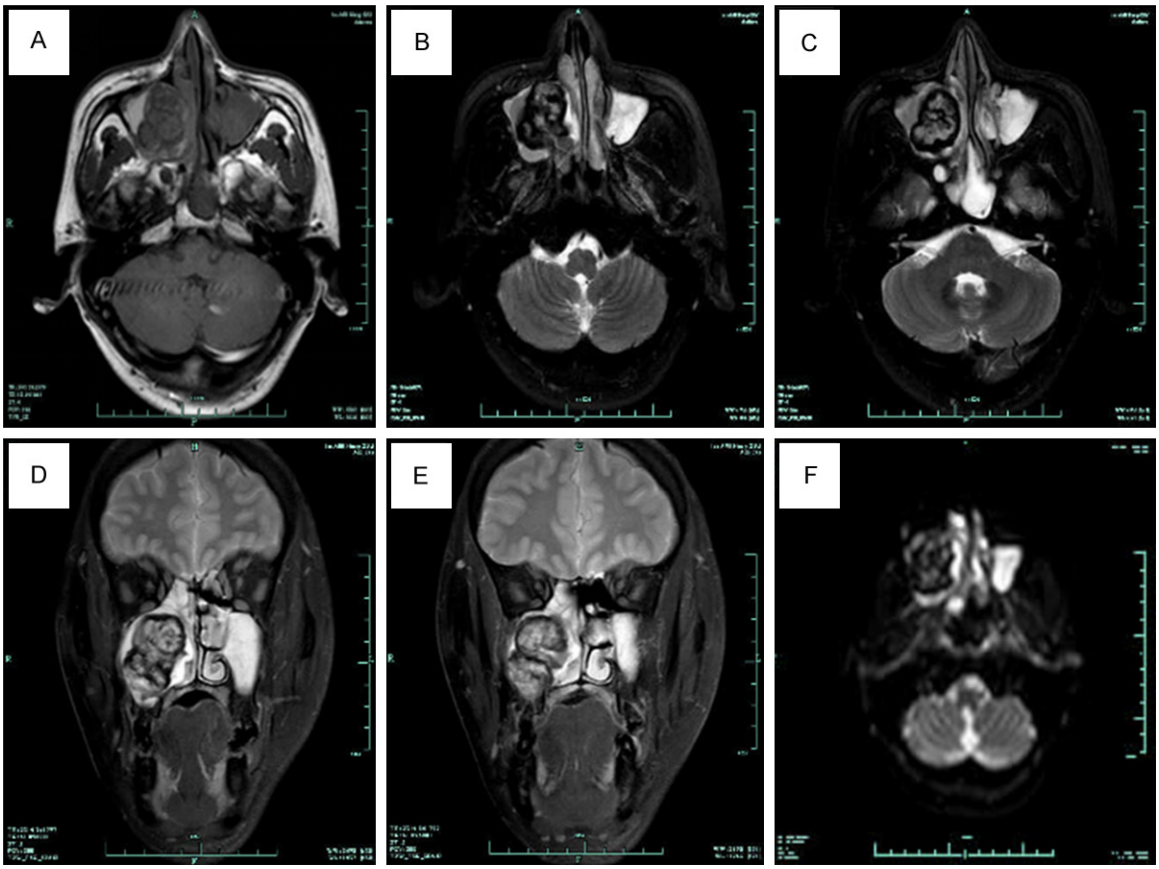

Figure 2. MRI showed the right maxillary sinus component of the mass to have heterogenous hypointensity on T1-weighted images (A) and heterogenous hyperintensity with a peripheral hypointense rim on T2-weighted images (B-E). Diffusion weighted magnetic resonance imaging (F) showed the mass of slightly high signal intensity with a peripheral hypointense rim.

whole findings, the diagnosis of an ANP was made. The ANP was completely excised with no significant hemorrhaging by intranasal endoscopic approach.

Light microscopy was used after staining sections with hematoxylin and eosin. The surface of the mass was covered by pseudostratified ciliated columnar epithelium with areas of squamous metaplasia, edematous subepithelial stroma with infiltration by many inflamed cells, The lesions showed numerous extravasated red cells, fibrin, abundance of irregularly shaped, thin-walled blood vessels, many showing intraluminal thrombus formation, which was associated with wide ares of ischemic necrosis (**Figure 4**). The patient remained asymptomatic and disease-free at follow-up 13 months later.

Discussion

There are many descriptions of ANP in the English literature, names include cavernous hemangioma, pseudotumor, inflammatory granuloma telangiectaticum, pseudoangioma, organized or organizing hematoma, vascular granuloma, hemorrhage necrotic polyp, and angioectatic or angiomatous polyp. In this paper, we use the term angiomatous polyp, because it reflects the fact that the mass is not a real tumor and that the lesion is clinically characterized by extensive vascular proliferation and hemorrhage.

There are many hypotheses of the pathogenesis of ANP have been proposed, however, the definite pathogenesis of ANP is still unclear. One hypothesis is based on the presence of the maxillary sinus and/or nasal cavity polyp, the polyp pedicle is suffer from significant vascular compromise, which resulting in stasis, edema, ischemia, and necrosis of the polyp [1, 2]. Batsakis pointed out four extra-antral sites of vulnerability to vascular compromise for antrochoanal polyp that leaded to formation of ANP:

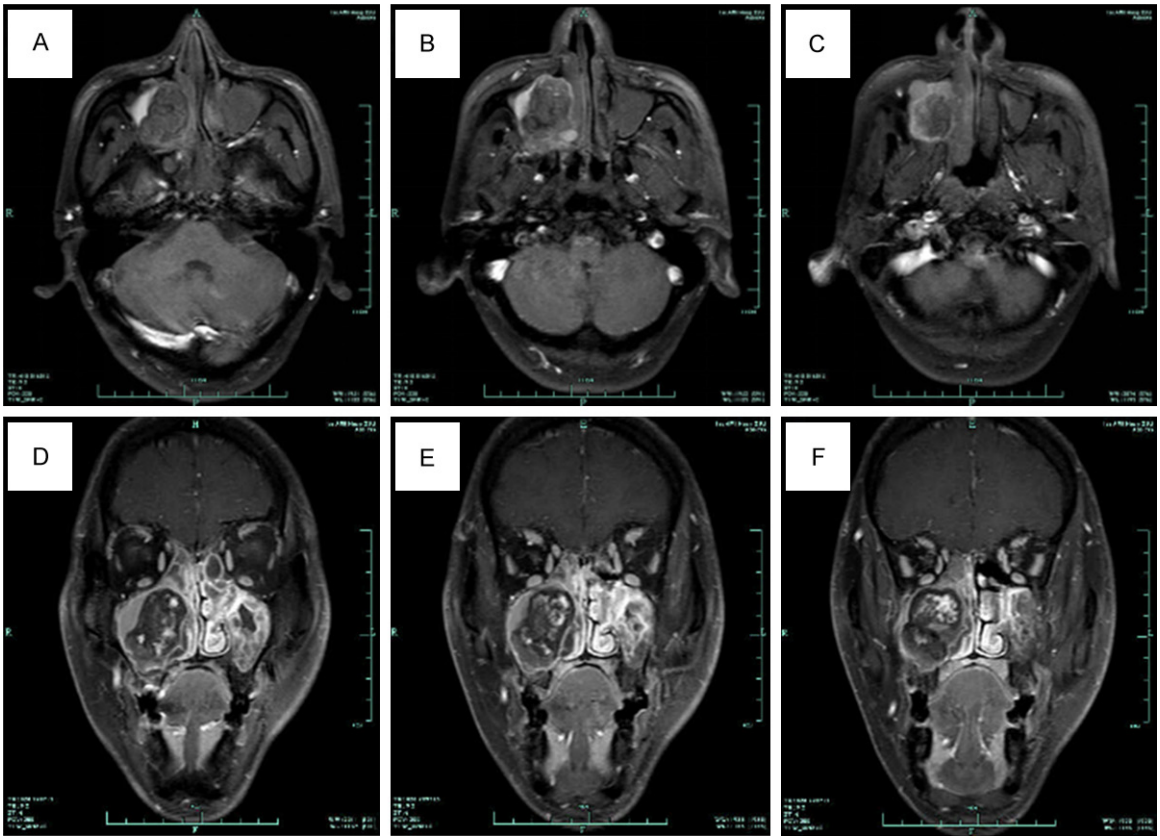

Figure 3. Postcontrast Axial (A-C) and coronal (D-F), T1-weighted images showed heterogeneous nodular and patchy enhancement with the unenhanced peripheral hypointense rim.

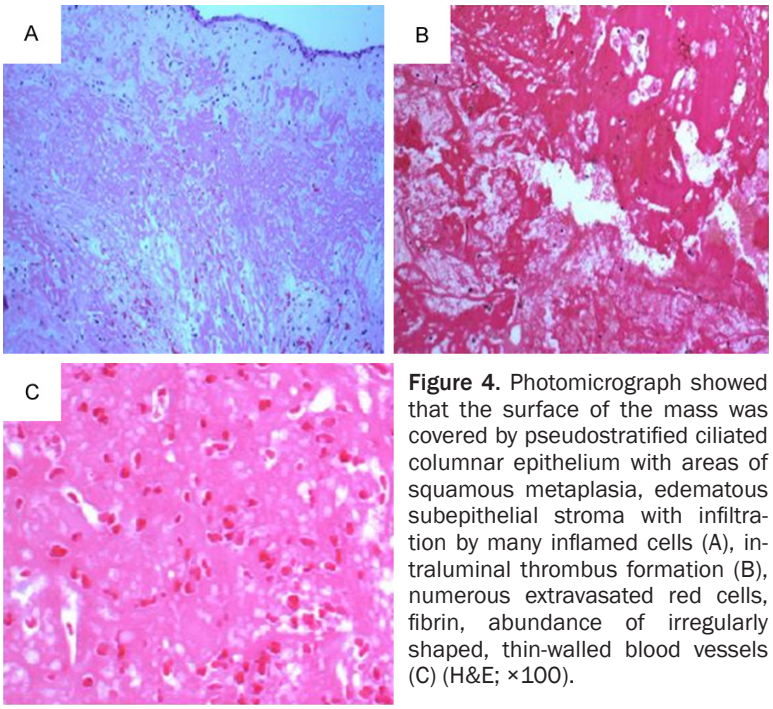

Figure 4. Photomicrograph showed that the surface of the mass was covered by pseudostratified ciliated columnar epithelium with areas of squamous metaplasia, edematous subepithelial stroma with infiltration by many inflamed cells (A), intraluminal thrombus formation (B), numerous extravasated red cells, fibrin, abundance of irregularly shaped, thin-walled blood vessels (C) (H&E; ×100).

the ostial exit site, the posterior end of the inferior turbinate, the posterior choana and the most dependent part within the nasopharynx [7]. Compression of the feeder vessels in these areas is considered to result in initial hemangiectasis and stasis, and edema. This leads to venous infarction followed by neovascularization of the polyp, then setting the stage for repeat vascular occlusion and infarction [7]. Another is that based on the formation of hematoma in the sinus antrum [1]. Many factors such as trauma, surgery, inflammation and/or allergy of the maxillary sinus and nasal cavity, bleeding diatheses, and rup-

tured aneurysm, which resulting in hematoma [1, 2, 8]. Reactive and reparative changes with neovascularization and fibrosis lead to the eventual formation of ANP. To our point of view, the two hypotheses are both reasonable.

The most remarkable pathological features of ANP were the large numbers of dilated blood vessels, many with proof of intraluminal thrombosis, and the sizable extent of necrosis of the lesion, with extravasation of blood components into the surrounding stroma [2-5]. The features of ANP under light microscopy are as follows [5]: (i) racemose aggregates of irregularly shaped blood vessels resembling dilated capillaries, no elastic or muscular layers; (ii) acute and chronic inflammation common; hemosiderin-laden macrophages; (iii) heterogeneity from field to field; patchy areas with features of typical inflammatory polyps; (iv) paucicellular stroma with scattered fibroblasts and myofibroblasts; marked nuclear enlargement; large nucleoli; no mitoses. The features of ANP under electron microscopy are as follows [5]: (i) typical fibroblasts and myofibroblasts with indistinct nuclear fibrous lamina; endothelial cell; (ii) amorphous extracellular matrix (fibrin, plasma, cellular debris). To sum up, the characteristic pathological features of ANP are extensive vascular proliferation, accumulation of extracellular amorphous eosinophilic material, and atypical stromal cells [2-5].

Clinically, the symptoms of ANP were varied and nonspecific, including nasal obstruction, epistaxis, ophthalmoptosis, facial swelling and pain, nasal discharge, snoring, headaches, hyposmia, epiphora, and visual disturbances [1-5, 8-13]. Baumgarten et al [13] reported that the most common symptoms associated with ANP are nasal obstruction and an alteration in olfaction. However, other studies including our previous study found that the most common symptoms associated with ANP are nasal obstruction and epistaxis [1, 2]. In our case, the patient presented with an 2-month history of right-sided nasal obstruction, fulvous rhinorrhea, headaches, right-sided tooth root pain under right face pressure, slight hyposmia and right-sided epiphora.

Radiologic examination is very important in the diagnosis of ANP. Previous reports of ANP clarified that CT findings lacked specificity for ANP identification. The typical appearance of ANP on CT are as follows [1, 2, 4, 7, 12, 14, 15]: (i) an expansile mass filles the paranasal sinus and/or nasal cavities and/or choana/nasopharynx, causing bulge or destruction of the bony wall and heterogeneous isoattenuation on pcontrast CT scans; (ii) on contrast-enhanced CT, the center of the lesions is non-enhanced with peripheral intensification; (iii) the mass on CT shows clear and smooth edges and does not invade the peripheral soft tissue; (iv) does not usually invade the pterygopalatine fossa or sphenoid sinus. However, the imaging features of ANP on conventional MRI are quite characteristic. Due to the high soft-tissue resolution, MRI has remarkable superiority to CT in reflecting the internal structures of ANP and the involved extent. The typical imaging features of ANP on MRI are as follows [1, 3, 4, 12]: (i) an expansive soft tissue mass; (ii) the margin of the mass is well-defined; (iii) blockage of ostium and secondary obstruction; (iv) extension into the nasal cavity, choana and nasopharynx; (v) heterogeneous hypointensity or isointense signal intensity on T1WI; (vi) heterogeneous hyperintense with hypointense linear septum internally and with a peripheral hypointense rim surrounding the lesion on T2WI; (vii) heterogeneous nodular and patchy enhancement with the non-enhanced peripheral hypointense rim on postcontrast MRI. Most prominently, the hypointense peripheral rim of the mass is a very specific contribution to the correct diagnosis of ANP.

In conclusion, ANP of the maxillary sinus invades the pterygopalatine fossa is extremely rare. We believe that CT shows bone changes associate with ANP can provide useful information for the determination of surgical planning, while MRI that shows the typical imaging features in signal intensity can provide an accurate diagnosis of ANP. Therefore, we propose that combined application of CT and MRI is quite necessary for patients with suspicious of ANP. Moreover, endoscopic sinus surgery is considered the best approach for the treatment of ANP.

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Disclosure of conflict of interest

None.

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