

Case Report

Angiectatic nasal polyp diagnostic quandary of sinonasal malignancy

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Received: 08 January 2020

Accepted: 04 March 2020

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ABSTRACT

Angiectatic nasal polyp (ANP) is a rare entity of inflammatory nasal polyp accounts for only 4 to 5% of all nasal polyp. It is often misdiagnosed as a malignant neoplasm due to its aggressive clinical presentation and radiological findings mimicking a neoplastic characteristic. achieving the diagnosis of ANP can be a challenge but crucial as management and prognosis of malignant and benign sinonasal tumour differs significantly. We describe a unique case of an ANP in a 33 year old gentleman presented with left sided nasal obstruction associated with blood stained nasal discharge, facial pain and left eye epiphora. There was proptosis of the left eye and his left nasolabial fold was obliterated due to a vague left cheek swelling. Clinical presentation was highly suspicious of malignancy. CT scan finding was also suggestive of malignant tumour with surrounding bony bowing and erosions, while MRI was reported as haemangioma. Post-surgery of left endoscopic medial maxillectomy, histopathology result was negative for malignancy. After revision of specimen done and further discussions, the diagnosis of ANP was made. Patient was postoperatively well with regression of symptoms along 5 months follow-up. ANPs are relatively uncommon pseudoneoplastic lesion that can cause a significant diagnostic dilemma as illustrated in this case. awareness of distinctive radiological and histopathological findings of ANP is important to establish the diagnosis and managed accordingly. However, little is known to guide the approach in such cases, therefore we find the case report to be noteworthy in this field.

Keywords: Inflammatory nasal polyp, Sinonasal tumour, Pseudoneoplastic

INTRODUCTION

Angiectatic nasal polyp (ANP) is a rare entity of inflammatory nasal polyp accounts for only 4 to 5% of all nasal polyp. It is often misdiagnosed as a malignant neoplasm due to its aggressive clinical presentation and radiological findings mimicking a neoplastic lesion. Detail evaluations of radiological and histological features are very important in order to distinguish ANP from malignancy. However, little is described in literatures suggesting promising methods of

investigations and management of this condition. Hence, we report an interesting case of ANP discussing the nature around the disease, collating with sinonasal malignancy, aiming to contribute in the understanding of the disease.

CASE REPORT

We describe a unique case of an ANP in a 33 year old gentleman who was initially referred to us from the Hospital Emergency and Trauma Unit. His chief

complaint was left nasal blockage, associated with blood stained nasal discharge and occasional left sided headache for 2 months. He also experienced facial pain and numbness over the left maxillary area, with watery left eye for 2 weeks prior to presentation to our clinic. However, no visual disturbance mentioned, smell sensation was still intact and there was no history of frequent sneezing, nasal itchiness, or fever. There was also no history of recent trauma, pulmonary tuberculosis (PTB) contact, asthma, or allergic reaction before.



Figure 1: Proptosis of the left eye and left nasolabial fold obliteration due to a vague cheek swelling.

Upon physical examination, proptosis of the left eye was noted and his left nasolabial fold was seen obliterated due to a vague cheek swelling (Figure 1). Cold spatula test showed diminished frosting over the left side. However, throat and ear examination show no significant abnormality and there was no lymph node palpable in the neck. Rigid nasoendoscopy performed revealed obstructing left nasal mass over the inferior turbinate, limiting the passage of scope into the nasal cavity. However, the right nasal cavity was found to be normal, with no mass or polyp seen, and the fossa of Rosenmuller (FOR) was clear.



Figure 2: (A) CT scan showed heterogeneous mass occupying the left maxillary antrum and bowed out the medial maxillary wall causing compression in the left inferior turbinate, (B) thinning of all of the left maxillary sinus wall with patchy erosion at its cortex.

CT scan showed heterogeneous mass of mixed attenuation value occupying and expanding the left maxillary antrum. It bowed out the medial wall causing compression onto the left inferior turbinate with clear plane of demarcation between those two (Figure 2A). Superiorly, it has pushed up the floor of left orbit without infiltration into the orbital cavity. No intra-lesional calcification depicted. There is thinning of all left maxillary sinus wall with mild patchy erosion parts of its cortex (Figure 2B).

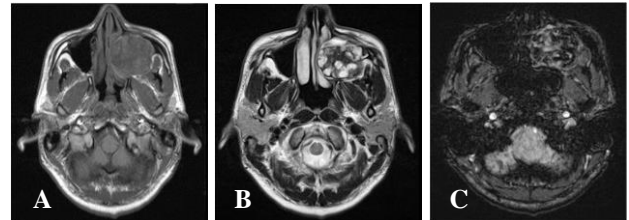


Figure 3: (A) T1 weighted MRI shows heterogeneous mass occupying and expanding the medial wall of left maxillary sinus, (B) MRI shows heterogeneous T2 high signal intensity mass with characteristic hypointense rim suggestive of hemosiderin deposition, (C) susceptibility weighted image sequence MRI showing marked blooming artefact of the left maxillary mass.

MRI showed heterogeneous T2 high signal intensity mass with characteristic hypointense rim suggestive of hemosiderin deposition. It is further confirmed on susceptibility sequence (SWI) which showing marked blooming artefact, suggesting that the mass is hypervascular with previous haemorrhages (Figure 3A-C).

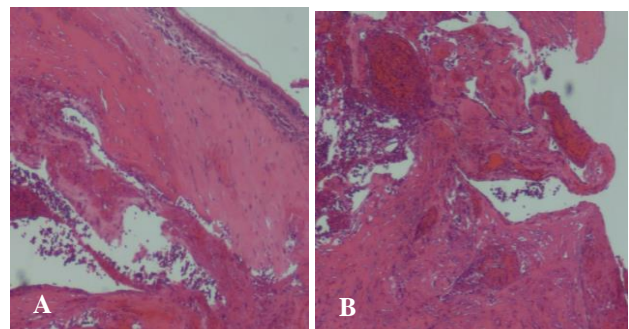


Figure 4: (A) On low power microscope, fragments of highly hyalinised tissue covered by respiratory epithelium seen, (B) microscopic image showing the ectatic vascular formations with extensive areas of stromal haemorrhage.

Microscopically, there are few fragments of highly hyalinised tissue covered by respiratory epithelium (Figure 4A). Ectatic vascular formations with extensive areas of stromal haemorrhage are noted (Figure 4B). No obvious malignant cell is seen. This case was discussed in interdepartmental meeting and diagnosis of ANP was confirmed.

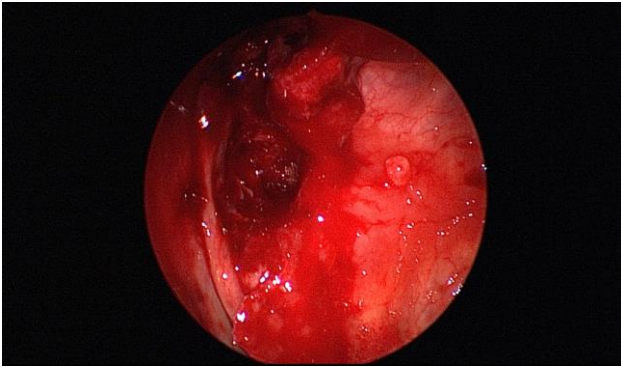


Figure 5: Endoscopic view of the huge, pink, polypoidal mass was found occupying the left maxillary sinus.



Figure 6: Nasoendoscopic finding three months after surgery showing clearance of mass in the left nasal cavity.

This patient underwent left endoscopic medial maxillectomy. Intra-operatively, a huge, pink, polypoidal mass was found occupying the whole left maxillary sinus (Figure 5). It did not appear haemorrhagic neither it bleed upon touch. No necrotic area seen on the mass but the central part was friable and devascularized. Some part of the left orbital wall of maxillary sinus was eroded, consistent with the CT findings. The whole mass was removed with left middle meatal antrostomy, ethmoidectomy, and sphenoidectomy.

DISCUSSION

The ANP, or most commonly called as ANP is relatively rare, and only accounts for about 5 percent of all nasal polyps.⁶ It falls into one of the groups of nasal polyps classified according to types, which includes others such as oedematous, glandular, fibrous, and cystic. ANPs are therefore a benign and pseudoneoplastic lesion, which can be managed just on the same lines as the choanal polyps.² Despite that, due to its aggressive presentation, it is often confused with sinonasal malignant tumour.

The case we described involves a young gentleman in his 30s. With regards to sinonasal malignancy, the gender distribution might be alarming because incidence of

sinonasal malignancy in males is twice that of females, but because he presented at a relatively younger age, diagnosis of malignancy might be less likely. The commonest age for sinonasal malignancy and ANP is 45 to 85 and 12 to 72 years old respectively.⁷

There are some rare industrial exposures which appear to be associated with this malignancy, such as exposure to hard and soft wood dust, leather tanning and rare minerals, for example; nickel, cadmium and chromium. Unlike other head and neck cancers, tobacco smoking is not considered to be etiological factor, but recent study demonstrates a higher incidence of nasal cancers in cigarette smokers. However, none of these risk factors has been related to this patient.

Other than that, ANP can present with symptoms that are identical to the most common presentation of sinonasal malignancy, which is unilateral nasal obstruction, nasal discharge (with or without blood staining), epistaxis, and facial pain.⁷ To further complicate the issue, they also progress rapidly and aggressively as such the malignant lesion would develop. Plus, in a smaller group of patients, invasion to the adjacent structures is often seen as such illustrated in this case, where the left orbit was involved.⁴

Sinonasal angiomatous polyp appears as mass filling up the maxillary sinus or nasal cavity. Unenhanced CT will show a mass which contains a mixed high to low attenuation due to mixture area of haemorrhage, organized thrombi, necrosis and inflammation. Bony changes are always present; mainly expansion which results in erosion, demineralization, resorption and remodelling of the bone.¹ Unlike from its malignant counterpart i.e. sinonasal malignancy, the bony changes are almost always non-destructive. On MRI, the lesion is hypointense on T1WI, heterogeneously hyperintense on T2 with characteristic peripheral hypointense rim indicating hemosiderin deposition due to old haemorrhages.

Dynamic contrast examination is recommended as the lesion will shows gradual enhancement pattern typical of a vascular tumour.² Another imaging differentials that need to be considered in this case is haemangioma. Sinonasal haemangioma is very rare, and usually involves the nasal cavity rather than paranasal sinuses.³ However, absence of intralesional calcification on plain CT study (which represent calcified thrombus in dilated vascular space) in our case made this diagnosis least favourable. Careful imaging analysis is very crucial to come into a correct and most accurate diagnosis. When an isolated nasal or sinus mass is found, particularly with concurrent clinical presentation of epistaxis and in addition to apparently benign bony changes rather than destruction, a haemorrhagic sinonasal mass of vascular origin needs to be entertained first.

Microscopically, ANP usually show similar appearance. In a case series of 13 patients, all cases presented with

irregularly shaped and thin walled blood vessel.⁷ The stroma is hypocellular and expanded by oedema and extravasated eosinophilic amorphous fibrin-like material.⁷ Areas of haemorrhagic necrosis are also seen with thrombus formation and neovascularisation.⁷ The appearance may simulate haemangioma but haemangioma usually has larger vascular lumina.⁷ Even though appear benign morphologically, malignant fibrosarcoma has been reported to show similar tissue changes in superficial biopsy.⁹ Therefore, deep biopsy is warranted to confirm ANP and to exclude malignancy.⁹

According to this case, and most of previous literatures, we also believe that the hypothesis postulated by Batsakis et al, for the pathophysiology of ANP can be accepted where the ANP is thought to derive from an antrochoanal polyp originating from the maxillary sinus and protrudes into the nasal cavity via the sinus ostium.¹⁰ It may also extend posteriorly to the posterior choana or to the nasopharyngeal region. Due to the confining anatomical structure of the ostium, the polyp's vascular supply is vulnerable and compromised. Compression of the feeder vessels leads to vascular dilatation, stasis, oedema, ischemia and subsequently neovascularisation and fibrosis of the polyp. Extensive extravasation of blood components through thin walled vessel results in area of haemorrhage and accumulation of large perivascular pools of amorphous eosinophilic cells, giving rise to the radio pathological features as mentioned above.

Considering the differential diagnosis of squamous cell carcinoma where similarity being the aggressive clinical behaviour and pseudo epitheliomas hyperplasia, however, during surgery it become evident that the ANPs are benign as it relatively easily excisable with minimal bleeding occurrence as demonstrated in this case.² In view of this too, the recurrence rate is very low as revealed by a study done by Dai et al which shows no incidences of recurrence observed during at least in 60 months period follow-up postoperatively.⁵

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: Kashaf NAAFA, Chin AHK, Ibrahim ZM, Rahman IWA, Rahman SKA. Angiectatic nasal polyp diagnostic quandary of sinonasal malignancy. Int J Otorhinolaryngol Head Neck Surg 2020;6:769-72.