Infarcted Angiectatic Nasal Polyp With Bone Erosion And Pterygopalatine Fossa Involvement- Simulating Malignancy. Case Report And Review Of Literatures
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Citation

Abstract
Background: Angiectatic nasal polyps, also called angiomatous polyps, are a rare subtype of inflammatory sinonasal polyps, presenting in a variety of ways. Sometimes compromise of their vascular supply may lead to infarction and occasionally erosion with destruction of surrounding bone, stimulating a malignant mass. We present two cases of an infarcted angiectatic nasal polyp, one caused extensive surrounding bony destruction and the other presented with recurrent nasal bleeding, which caused significant clinical and radiological concern. We characterized the histomorphological features of an angiectatic polyp with histochemical stains and reviewed the literature. Angiectatic nasal polyp has distinct histomorphological features, which are poorly documented in literature and awareness of those features would prevent confusion with other vascular or spindle cell lesions of the nasal cavity, the nasopharynx. Correct preoperative radiological diagnosis is important to avoid unnecessary extensive surgery, and histopathological evaluation is mandatory, as they require different treatment since they carry a different prognosis.

INTRODUCTION
The angiectatic polyps (ANP) account for only 4-5% of inflammatory nasal polyps and have characteristic features and growth patterns, which are less often noted, but no less important in establishing a final diagnosis. ANPs are a prominent component of dilated capillary-type blood vessels, as compared to non-angiomatous polyp which have decreased density of blood vessels than normal mucosa. Intraluminal thrombosis is rarely widespread, but necrosis leads to infarction of the polyp and occasionally local aggressiveness, causing extensive bone destruction, mimicking a neoplastic process. Since the infarcted ANP is fragile during excision and is difficult to remove en block, this leads to a large variation in pathologic description and terminology: it is also called “nasal polyp with hemorrhage and necrosis,” due to similarities in CT and MRI features. There are very few studies which addressed the morphologic picture of infarcted ANPs.

PATIENTS AND METHODS
Case 1: A 20-year-old female presented with a one-year history of nasal obstruction, headache, anosmia and swelling above the medial canthus of the left eye. She denied epistaxis, blood stained nasal discharge or visual disturbances. She underwent an operation in an outside hospital for a mass of the nasal cavity, which recurred. No biopsy details were available.

On examination: Anterior rhinoscopy showed a pale white, painless polypoidal mass occupying the whole left nasal cavity, pushing the septum to the right, causing disfiguration of the dorsum of her nose and causing her medial canthus to bulge. A probe could be passed all around the mass except the lateral wall. No bulging noted on probing the mass. A globular mass was noted on posterior rhinoscopy. Both eyes were normal on exam.

CT scan: Nose and paranasal sinus (PNS) revealed a polypoidal mass with peripheral minimal enhancement involving the left nasal fossa and maxillary antrum, bulging anteriorly through the nasal orifice and posteriorly through the nasal choana with destruction of left nasal bone, lamina papyracea pushing the eye laterally and the posterior maxillary antral wall and the pterygo palatine fossa.
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extending to the retroantral fat. This was suggestive of a neoplastic lesion. (Fig. 1)

**Figure 1**
Fig 1- CE CT axial scan of nose & Paranasal sinuses showing mass occupying whole of the nasal cavity on left side, pushing the septum to the contralateral side with cranial extension causing destruction of the ipsilateral ethmoidal air cells, medial wall of the orbit and pushing the eye ball laterally.

Intraoperative small biopsy sent for primary morphological diagnosis by Frozen section examination, which revealed histological picture of benign nasal polyp (Fig. 2).

**Figure 2**
Fig. 2 – Microphotograph biopsy Frozen Section shows fibrous connective tissue with a few vascular spaces in the edematous stroma. (X 100 H. E.).

Then mass was excised almost completely through the transnasal route using an endoscope. The mass was arising from the left maxillary antrum, enlarging the ostia continued superiorly to anterior ethmoidal complex and posteriorly to nasopharynx. Destruction of the maxilla and involvement of the pterygopalatine fossa was a unique feature. Minimal bleeding occurred intraoperatively. Postoperative period was uneventful.

Gross biopsy examination: Multiple irregular grayish-white tissue pieces with two globular brownish polypoidal masses measuring 2x2x1.6 cms and 3x2.5x1.3 cms, respectively, with attached pedicle size showed surface ulceration with multiple hemorrhagic areas and tiny cystic spaces upon cutting.

Histopathological examination: This revealed the structure of a nasal polyp lined by pseudostratified ciliated columnar epithelium displaying focal squamous metaplasia with focal surface ulceration. The stroma showed clusters of dilated congested thin walled capillary type blood vessels, few of ectatic vessels show fibrinoid necrosis of wall and intraluminal fibrin thrombi (Fig. 3), which were positive for Massons trichrome (MT) stain.

**Figure 3**
Fig. 3– Microphotograph permanent section shows luster of thin walled ectatic blood vessels and intraluminal fibrin thrombi with extracellular fibrinoid material in the stroma and positive with MT stain. (Fig. 3- X 100 MT)

Clusters of dilated ectatic blood vessels were well delineated in Reticulin stain and also revealed the intraluminal thrombi (Fig.- 4). The vascular area accompanied by avascular area.
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Figure 4
Fig. 4- Microphotograph show well delineated dilated thin walled blood vessels with intraluminal thrombi (X 200 Reticulin stain).

Deposition of eosinophilic amorphous extracellular material in the surrounding stroma and areas close to the surface of tumor noted, which was congo red negative, (Fig. 5)

Figure 5
Fig. 5- Microphotograph shows extracellular congo red negative pseudoamyloid material in the stroma and cluster of ectatic blood vessels. (X 100 Congo red)

Occasional scattered spindle cells displaying mildly pleomorphic nuclei with stromal edema (Fig. 6) and mild mixed inflammatory cell infiltrate were also noted. The report was signed out as Angiectatic nasal polyp.

Figure 6
Fig. (6) - Microphotograph shows scattered fibroblast- like atypical spindle stromal cells and inflammatory cells.(X 200 )

The patient was discharged after 3 days. No sign of recurrence was observed on regular follow up and the swelling on the medial canthus of Left eye completely disappeared.

Case 2: A 30-year-old male presented to ENT OPD complaining of headache for the last 3 months, pain in the right ear, right nasal obstruction and right nasal bleeding for last 2 months. She had a history of massive nasal bleeding from the right nasal cavity, for which the patient was admitted and three pints of blood were transfused. On examination, the patient was pale, otherwise no significant finding on the face was noted. An examination of the right nasal cavity revealed a polypoidal mass protruding out through right middle meatus. The surface of the mass was slightly bluish, which started bleeding on manipulation. The rest of the nasopharynx could not be visualized because of a deviated nasal septum and syneche. The rest of the examination was insignificant. Clinical finding were suggestive of some vascular lesion, hence the patient was investigated radiologically.

CECT PNS: This showed a polypoidal soft tissue mass showing heterogeneous enhancement on post contrast study, filling the right maxillary antrum extending into the right nasal gallery via widened maxillary hiatus amalgamating with inferior and middle turbinates, causing severe narrowing of the right nasal air passage posteriorly, extending into the right sphenoid sinus and nasopharynx. There is evidence of mild thinning of the roof of the right maxillary sinus with no obvious intra orbital extension,
consistent with a slow growing tumor. No intracranial extension and no definitive bone destruction were seen. Right osteomeatal complex and right fronto-ethmoidal recess are blocked with inspissated secretions seen in the ethmoidal and sphenoidal sinus. (Fig. 7)

**Figure 7**

Fig. 7- CT- Coronal reformat showing right maxillary sinus mass with extension to ipsilateral nasal cavity.

The patient's hemoglobin was 10 gm. The patient was taken for surgery under general anaesthesia. The mass was removed endoscopically via the trans nasal route in pieces sent for histopathological examination. Average bleeding during surgery was noted. Postoperative period was uneventful.

Gross examination: The excised specimen was soft and polypoidal in appearance, having brownish color in most pieces and show whitish shining mucosa in occasional piece.

Histopathological examination revealed part of the tumor showing features of inflammatory nasal polyp with edematous stroma lined by pseudostratified columnar epithelium and large part of the tumor show features similar to first case of infarcted angiectatic nasal polyp with focal ulceration of the epithelium.

**DISCUSSION**

Inflammatory sinonasal polyps are classified into five types on the basis of predominant stromal element seen in histological evaluation: edematous, glandular, fibrous, cystic and angiomatous / angiectatic polyp. These present clinically as soft, gelatinous translucent polypoidal, painless swelling with gradual obstruction of the nasal cavity associated with nasal discharge. ANPs are reported to be a derivative of antrochoanal polyps commonly, but may be a variant of sinonasal polyps of any location and their vascular supply be susceptible to compression at ostial exit site, at the posterior end of inferior turbinate, the posterior choana and at the most dependent part within the nasopharynx. It is hypothesized that vascular compromise causes initial vascular dilatation/ectasia, extravascular edema and possibly infarction followed by reactive and reparative changes with neo-vascularisation, setting the stage for continuing development of polyp, repeated vascular occlusion and further infarction. Resulting hemodynamic condition predisposes the patient to extensive extravasation of blood components (fibrin & platelets) through thin walled capillary - like blood vessels resulting in areas of hemorrhage and accumulation of large perivascular pools of amorphous congo- red negative eosinophilic material. Predominant features of infarcted ANPs are clusters of ectatic blood vessels surrounded by abundant fibrin-like eosinophilic extracellular material and superimposed fibrinoid necrosis, luminal thrombosis of ectatic blood vessels. These polyps can grow rapidly, causing bone erosion that could simulate malignancy preoperatively. Scattered atypical pleomorphic spindle cells (Myofibroblasts) in the stroma are part of reactive secondary changes, seen occasionally in sinonasal polyp, but are quite common in angiectatic polyps - a pseudosarcomatous change. However, other vascularised fibromatous angiomatous polyps do not show deposition of pseudoamyloid-like eosinophilic material, superimposed fibrinoid thrombosis and fibrinoid necrosis of blood vessels wall, or pseudosarcomatous stroma.

Vascular tumors are the most common nonepithelial tumors of the nasal cavity and nasopharynx and it is the prominent vascular component of ANPs, which can pose differential diagnostic problems, mainly with capillary or cavernous hemangioma, sometimes with organized or organizing hematoma and nasopharyngeal angiofibroma. The vascular lumina of cavernous hemangioma are usually larger than those of angiomatous polyp. Predilection for age, sex, site and histomorphology helps in differentiation. Angiofibroma occurs in young males, whereas sinonasal angioma do not show age or sex predilection. Sinonasal angiomas occur more often in the anterior nasal septum, the turbinate and vestibule. Both commonly present clinically with epistaxis, nasal obstruction and bleed significantly on biopsy. Correct diagnosis is necessary, as they require
different treatment and have different prognosis. It is problematic to differentiate angiomatous polyp from juvenile angiofibroma clinically and may need to rely on imaging features and pattern of growth for distinction between these two entities. CT scans show angiomatous polyps, which are non-enhancing or minimally enhancing nasal vault masses without pterygopalatine fossa involvement. Where angiofibroma has typical hypervascular appearance in contrast to angiomatous polyp, the common extension of tumor occurs through the roof of nasopharynx into the sphenoidal sinus and pterygopalatine fossa, so it is difficult to remove surgically. Though invasion of the sphenoidal sinus and ethmoidal sinus by ANPs has been reported. Conventional MRI is a better modality for preoperative diagnosis of the angiomatous nasal polyp, and show characteristic hypointensity on T1 weighted images and internal heterogeneous hyperintensity with a peripheral hypointense rim on T2 weighted images, as well as and strong nodular and patchy enhancement on postcontrast MRIs. Moreover, progressive enhancement on DCE MRI is very important diagnostic clue. Areas of mixed signal intensity on T2 weighted images are supposed to be caused by the extensive areas of organized thrombus and necrosis in that part of polyp and the peripheral hypointense rim on T2 weighted images due to old microhemorrhage with hemosiderin deposition on the surface of the polyp. Post contrast strong enhancement of nasochoanal portion of ANP suggest extensive vascular proliferation and ectasis. 

Angiography may be used for early diagnosis and to differentiate ANP from juvenile angiofibroma. ANPs show hypovascular or avascular appearance on angiography due to their irregular racemose arrangements of diluted capillary-type vessels, in contrast to normal arborizing pattern of vascularity. Specific locations of angiofibroma in pterygopalatine fossa with absent flow voids on MRI can have characteristic histologic features of stellate and staghorn blood vessels set in. Compression by cellular fibroblastic stroma differentiates it from ANPs, which have racemose aggregates of irregularly shaped dilated capillary-like blood vessels. However, correct diagnosis can be based on its anatomic location and association with inflammatory edematous sinonasal polyp. Sometimes pseudothelialomatous hyperplasia of the surface epithelium of ANP may raise suspicion of squamous cell carcinoma which could be easily ruled out in histology. Organized hematoma is usually subepithelial in location and characterized by admixture of fibrin network and hemorrhagic material with surrounding fibrous tissue margin, which prevents reabsorption of hematoma resulting in neovascularization and fibrosis. Most ANPs arise in maxillary sinus and extend towards the choana and into the nasopharynx and the most common symptoms are nasal obstruction and recurrent epistaxis. Gradual enlargement of the lesion may cause erosion, displacement of the adjacent bony structures, cheek swelling, and cause exophthalmos to manifest. Our first case was unique due to the fact that the tumor was destroying the maxilla and the pterygopalatine fossa was involved, which has not been described in literature and was easily removed.

CONCLUSION

Angiectatic polyps present with significant heterogeneity and characterized by extensive deposition of extracellular amorphous eosiophilic pseudoamyloid-like material with clusters of ectatic capillary-type blood vessels, and fibrinoid necrosis with luminal fibrin thrombi in infarcted one. ANPs may behave more aggressively, clinically, than other angiomatous polyps and may simulate malignancy, although entirely benign, hence, awareness of their existence and morphology is of considerable importance.

References
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