

Frontoethmoid Osteoma with Bilateral Nasal Polyposis: A Rare Coexistence

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Abstract

Osteoma and nasal polyposis are common benign conditions seen by ENT practitioner, however it is very rare that both of them present together. Osteoma is the most common neoplasm of the nose and paranasal sinuses. It is commonly asymptomatic, being an incidental finding 1% of plain sinus radiographs and 3% CT scans of sinus. In an extensive review of english literature, we could not find any case reported having large osteoma with bilateral nasal polyposis. We found only two reported cases of osseous metaplasia in cases of nasal polyps but not ivory osteoma hence bilateral nasal polyposis can radiologically have association with osteoma and should be investigated and treated accordingly.

There has been a single case reported in the Spanish literature though. In current case report, we present a case of patient with bilateral nasal polyposis accompanied with unilateral frontoethmoid osteoma.

Keywords: Sinonasal tumors, Nasal polyposis, Osteoma.

INTRODUCTION

Osteoma and nasal polyposis are common benign conditions seen by ENT practitioner, however it is very rare that both of them present together. Osteoma is the most common neoplasm of the nose and paranasal sinuses. It is commonly asymptomatic, being an incidental finding in 1% of plain sinus radiographs and 3% CT scans of sinus. The most frequent site of origin are frontal ethmoid, maxillary, and sphenoid sinuses, in descending order.¹

The symptoms includes headache localized over area of involvement, nasal obstruction, facial pain, rhinorrhea, anosmia and sinusitis or ocular symptoms. Because of cramped nature of ethmoid sinus, symptoms caused by ethmoid osteoma occur earlier than with osteoma of frontal sinus. Embryologic, traumatic, and infective etiologic theories have been proposed for development of osteoma.

Associations of osteomas along with colorectal polyps (Gardner's syndrome)² have been described in literature but not with nasal polyposis. In an extensive review of english literature, we could not find any case reported having large osteoma with bilateral nasal polyposis. There has been a single case reported in the Spanish literature though. In current case report, we present a case of patient with bilateral

nasal polyposis accompanied with unilateral frontoethmoid osteoma.

CASE REPORT

A 35 years old female admitted in our department with a two months history of bilateral nasal obstruction associated with bilateral clear mucoid discharge, headache along with postnasal discharge, sneezing, mouth breathing during sleep and bilateral hyposmia. Her past medical history included no other major diseases except hypertension. No previous history of surgery, trauma or allergy. The patient underwent rigid nasal endoscopic evaluation (4 mm, 0° endoscope) which reveals presence of bilateral greyish nasal polyps. Tympanic membrane of both ears were retracted (Grade-II). Ophthalmological examination was normal.

Computerized tomography scan of the nose and paranasal sinus revealed soft tissue density in bilateral nasal cavities, maxillary and ethmoid sinuses along with hyperdense bony lesion in right frontal and ethmoid sinus suggestive of nasal polyposis along with incidental finding of right frontoethmoid osteoma (Figs 1A to 2B). A standard endoscopic approach was used to remove the polyps from both sides with 0° endoscope, microdebrider and microdrill

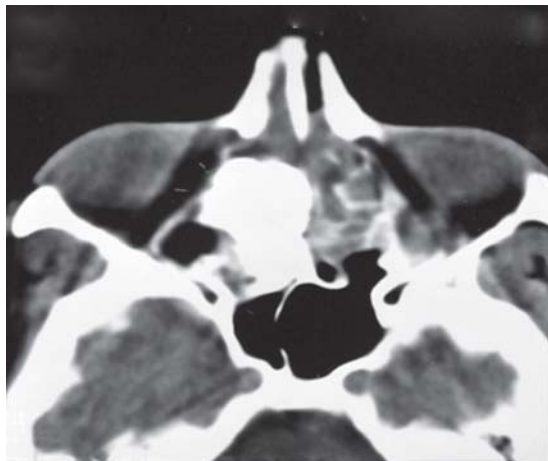


Fig. 1: Homogeneous dense osteoma (right) and soft tissue density of nasal polyp (left)

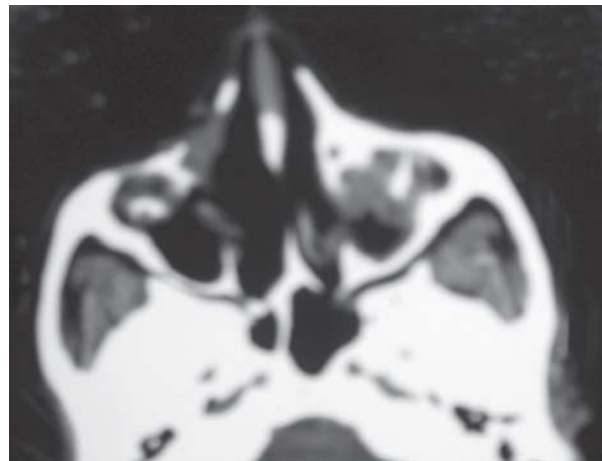


Fig. 4: Axial cut of the patient after 4 months of surgery

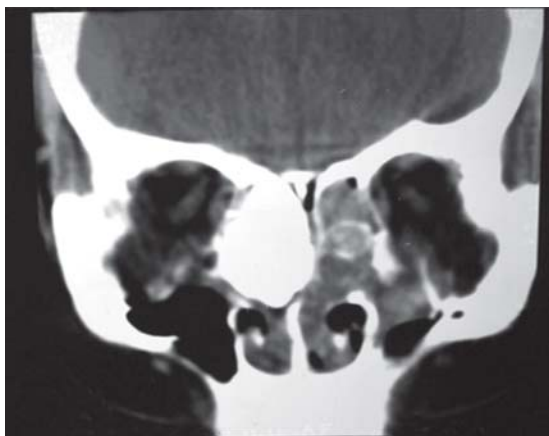


Fig. 2: Osteoma (right) and soft tissue density with ossification in center (left)



Fig. 3: Coronal cut of CT scan of the patient after 4 months of surgery

with burr. Right Lynch Howarth incision was given on right side, subperiosteal flap elevated and ethmoid gallery opened. There was a large 3 × 4 cm bony hard osteoma present in

ethmoid and frontal sinus attached superiorly to lateral lamella and floor of sphenoid sinus with erosion of nasal septum blocking maxillary osteum. It was freed all around with the help of microdrill and removed. Polyps present in maxillary and ethmoid sinuses were removed. The cavity was packed with neosporin soaked ribbon gauze which was removed after 48 hours.

Postoperative period was uneventful. The patient was followed with nasal endoscopy in our OPD weekly in first month and then monthly between 2 and 4 months. CT scan of sinuses were performed 4 months postoperatively and found no residual polyp or osseous tumors (Figs 3 and 4). Histopathology of the specimen was reported as allergic polyp, and ivory osteoma. The patient is asymptomatic on follow-up of two years.

DISCUSSION

Osteomas and nasal polyposis have been medically recognized condition since the time of ancient Egyptians and polyp removal with snare was described by Hippocrates. Osteomas are commonest facial bone tumor with frontal sinus being the most frequently involved sinus. 75% of osteomas arise in frontoethmoid region.³ The true incidence can not be quoted as many osteomas remains asymptomatic but can be estimated as high as 1%.

Osteomas may present at any age and males appear to be affected more often than female, reach peak in those above 40 years. These slow growing neoplasms are of ivory and cancellous kind, the former being the commoner.³ Prevalence of nasal polyposis in normal population is around 1% however it is more commonly associated with allergy, asthma, aspirin, atopy, and cystic fibrosis and can be as high as 80% in AFRS.⁴ Polyps do not seem to arise from all nasal and sinus mucosa, having a predilection for the middle turbinate, middle meatus and ethmoids.

Nasal polyp and osteomas are relatively common conditions but together they are very rare occurrence; we could not find any case report after an extensive search of english literature. Here we report rare coexistence of bilateral nasal polyps with unilateral frontoethmoid osteoma. There is known association between multiple osteomas and intestinal polyps, fibromas, lipomas, neurofibromas, epidermoidcyst, abnormal teeth, pigmented skin lesions (Gardner's syndrome), an autosomal dominant inherited disorder common in Mormons of Utah.² No definite etiology has been identified. The embryonic theory proposed by Cohnheim (quoted by Hallberg and Begley)⁵ holds that the osteomas arise from persistent embryologic cell rests or embryologic cartilaginous cells of the junctional zone around the ethmoid labyrinth. The traumatic theory of Gerber (quoted by Rawlins)⁶ postulates that osteomas are the sequestra of previous head injuries. No single predisposing disease can be implicated in the formation of polyp, though they may be associated non-allergic asthma, aspirin intolerance or sensitivity, atopy or allergy, cystic fibrosis, allergic fungal sinusitis and Kartagener's syndrome. The role infection is thought to be important in genesis of polyp formation. This is based on experimental models in which multiple epithelial disruptions with proliferating granulation tissues have been initiated by the infection *Streptococcus pneumonia*, *Staphylococcus aureus* or *bacteroides fragilis* (all common pathogen in sinusitis) or *pseudomonas aeruginosa*, which is often found in cystic fibrosis.⁷

Osseous metaplasia in mucosal polyps, although rare, is phenomenon that has been reported in the external auditory canal, tongue, intestines and stomach but it is rare phenomenon in nasal polyps. There have been only two case reports of calcification of sinonasal polyposis and nasal osteomas secondary to hypercalcemia.^{8,9} Bone morphogenetic proteins (BMPs) and transforming growth factor beta-1 (TGF beta-1) have been shown to be involved in ectopic bone formation, and therefore may be involved in ossification of mucous polyps. Squamous metaplasia of respiratory epithelium covering nasal polyp is common. DeVries¹⁰ postulated the new bone formation occurring in nasal polyps arose from bony remnants left behind during previous surgery.¹⁰

In our patient the polyp shows no histologically abnormal epithelium overlying the bone formation and it did not reveal

any atypical mesenchymal or enchondral or cartilage formation. This patient had no history of previous nasal surgery and facial trauma. Serum calcium levels were normal. In our case infective pathology can be hold true for genesis of nasal polyp and frontoethmoid osteoma on the ipsilateral side and the cause for polyps on contralateral side can be persistent obstruction due to enlarged osteoma on right side leading to gross deviation of nasal septum towards left side. However, exact etiology in our case is still matter of debate as various pathological processes described above can not hold true for our patient except inflammation.

CONCLUSION

We present a case of unilateral frontoethmoid osteoma associated with bilateral nasal polyposis. This is an extremely rare occurrence and to the best of our knowledge this is the first case report on reviewing english literature. We found only two reported cases of osseous metaplasia in cases of nasal polyps¹¹ but not ivory osteoma hence bilateral nasal polyposis can radiologically have association with osteoma and should be investigated and treated accordingly.

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