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Skull Base Osteomyelitis Secondary to Nasal hamartoma: A Case Report

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ABSTRACT

Nasal masses in the population present with nasal obstruction, epistaxis, and chronic rhinorrhea. We report a 70-year-old female with long-standing nasal obstruction, hyposmia, headache and symptoms of cheek and orbital swelling since 2 years. Nasal endoscopy revealed a polypoid mass with a glandular mucosal surface on the right side of the nasal cavity. Transnasal endoscopically the mass was resected which, on pathologic examination was reported a hamartoma. There was no recurrence at one year follow up. The present case is unique in that the hamartoma arose from the right maxillary sinus and extended up to the cavernous sinus.

Keywords: Nasal Hamartoma, Osteomyelitis, maxillary sinus, FESS.

INTRODUCTION

Seromucinous hamartoma of the nasal cavity is an exceedingly rare diagnosis; however, it should be included in the differential diagnosis of a posterior nasal tumor.However, it is important to distinguish a posterior nasal tumor from hyper vascular lesions, such as angiofibromas, before a biopsy because of the likelihood of severe hemorrhage.[1]We report a rare case of nasal hamartoma extending intracranially through the right superior orbital fissure with the suspicious involvement of right cavernous sinus.

CASE REPORT

70-year-old female presented with right-sided nasal obstruction, hyposmia, swelling of right cheek and right orbit. She underwent otolaryngologic evaluation. Nasal examination revealed a polypoid mass filling the right nasal cavity. The left nasal cavity and sinuses were normal. There were no attachments to the septum, nasal vestibule or nasal floor. There was minimal bleeding on touch. The right maxillary sinus was filled with mucus and the mucosal lining showed polypoid changes.

1. CLINICAL EXAMINATION

On examination of the face, cheek and orbital swelling were present on the right side. Oral cavity examinationrevealed good dental hygiene except some missing teeth's.Ophthalmological examinationrevealed right eye axial proptosis (Rt=22mm,Lt=14mm) on Hertel exophthalmometer and vision was found to be 6/18 bilaterally with dull foveal light reflex. On otoscopic examination bilateral tympanic membranes were found to be intact, no neck lymphadenopathy was present. The mass was excised endoscopically and sent for histopathological examination (Fig. 1).

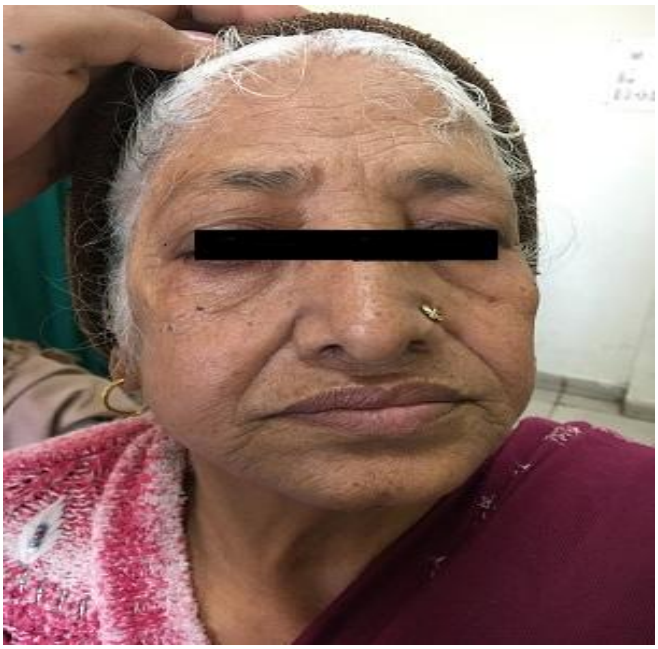

Fig. 1: Right orbital and cheek swelling

2. RADIOLOGICAL FINDINGS.

There was evidence of soft tissue lesion involving the roof of the right maxillary antrum. It was also involving the medial wall of right maxillary sinus extending to the right orbit, causing the destruction of its floor and medial wall. It was also extending intracranially through the right superior orbital fissure with the suspicious involvement of right cavernous sinus. It was causing marked erosion or remodeling of roof /medial wall of right maxillary sinus, right sphenoid sinus, medial pterygoid plate, and medial wall of the right orbit. Mucosal thickening was noted in left maxillary sinus and left orbit appearednormal (Fig. 2).

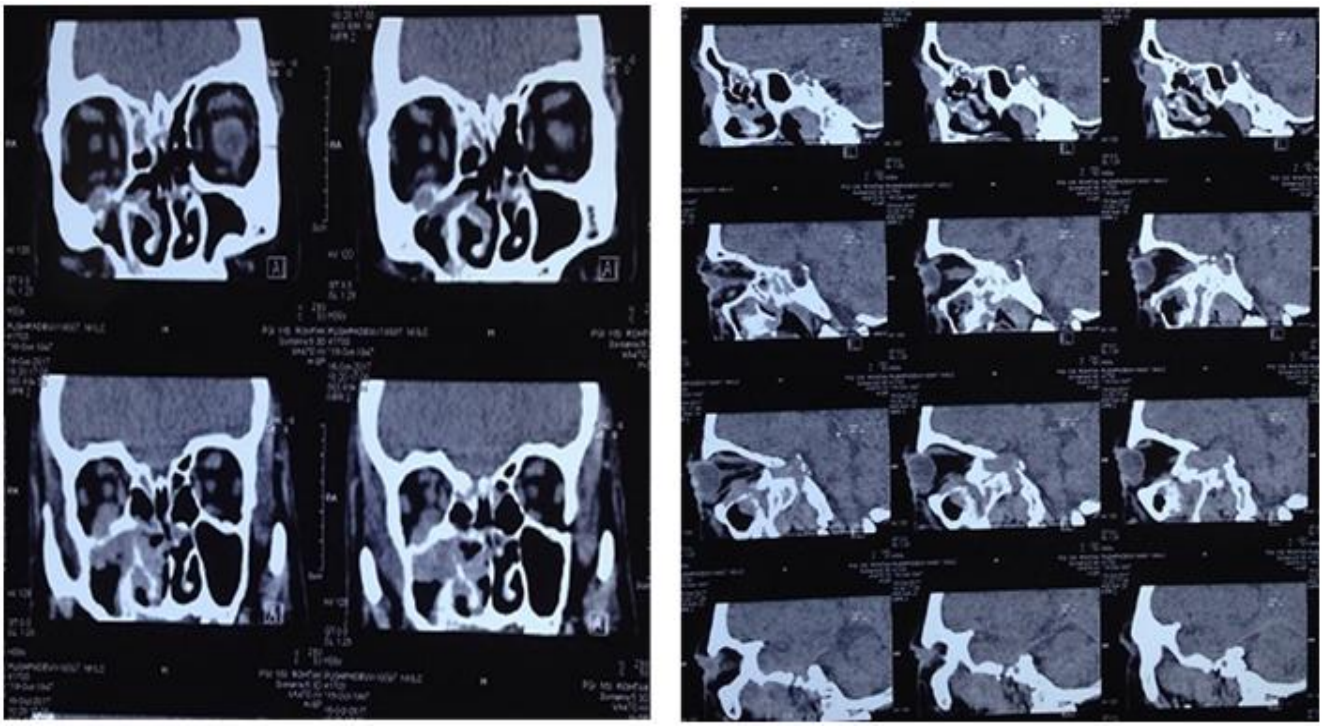

Fig.2. Right maxillary sinus and orbital involvement with the intracranial extension on CT scan.

3. PATHOLOGY EXAMINATION

The cut surface of the specimen showed connective tissue stroma which was intensely infiltrated by chronic inflammatory cells and was LCA positive. These inflammatory cells were arranged in clusters as well as spread into sheets over thick fibrinous and a fibrous background. At places, these inflammatory cells were plump and seen adjacent to the thickened vascular structures which were also CD 31 positive indicating proliferative vessel walls. Few nerve bundles and minor glands were also seen. PAS and Silvermethamine stains were negative for any of the fungus in the tissue.

Sections showed numerous thick-walled vascular structure of varying caliber embedded in fibro-collagenous tissue revealing moderately dense inflammatory reaction, many nerve bundles and lobules of seromucinous glands. Edge of the section shows woven bone formation. The fungus could not be demonstrated. Suggestive of hamartoma with secondary inflammation extending into adjoining bone causing chronic osteomyelitis.

4. DISCUSSION

Hamartomas are non-neoplastic malformations or inborn errors of tissue development. They may be derived from any one of the three germinal layers. They are characterized by an abnormal mixture of tissues indigenous to that area of the body, but with an excess of one or more of the tissue types. Development of hamartomas may involve errors during fetal growth or can develop from disturbances of immature tissues in the postnatal period. Hamartomas are derived from the normally-occurring local tissue. There is no evidence to suggest that hamartomas undergo neoplastic transformation.[2]Eichel and Hallberg differentiated hamartomas from teratomas and dermoids. Teratoma implies a growth derived from pluripotential tissue with elements of all three germ layers. Dermoids are cystic neoplasms originating from, predominantly, ectoderm and, to a lesser extent, mesoderm.[3] Head and neck hamartomas though uncommon are not rare. Fuller in 1963 reported on a pedunculated hamartoma of the esophagus.[4]Eichel and Hallberg resected a hamartomatous growth originating in the Eustachian tube and filling the middle ear.

Seromucinous hamartoma is a rare lesion of the sin nasal tract. The entity was initially described by Baillie and Batsakis in 1974 as a rare benign glandular proliferation of the sin nasal tract and nasopharynx.[5]Grossly, a seromucinous hamartoma is described as apolypoid mass that ranges in size from 0.6 to 6.0 cm, which is actually a type of epithelialhamartoma and has only been clearly described and characterizedin the past few years.The incidence of seromucinous hamartoma in womenis slightly higher than in men. It mostoften affects the middle-aged and elderly (mean age 54). The most common reported symptom was unilateral nasal obstruction; Epistaxis, asymptomatic, nasal discharge, and a chokingsensation have also been reported. Cases have arisen ineither the posterior nasal cavity or the nasopharynx, andmost have arisen from the posterior nasal cavity medialto the middle turbinate rather than lateral to the middleturbinate (15:1), with or without prolapse into the nasopharynx.Transnasal endoscopic excision is the management ofthe firstchoice.[6]Thepatient underwent the excision of the nasal mass under general anesthesia. The clinicalcourse was uneventful and there was no recurrence of themass after 1 year of follow-up.

CONCLUSION

Nasal hamartoma is an unusual entity. However, early and complete excision gives excellent prognosis with recurrence in few cases only as reported in the literature.

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