

Case Report

A rare combination of polyp, inverted papilloma and squamous cell carcinoma of sphenoid sinus

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ABSTRACT

Inverted papilloma (IP) is a tumor most commonly originates from lateral wall of nose and benign in nature. Sphenoid sinus is a rare site of origin and involvement. Malignant changes are the rare possibility in the IP. In this case report we reported a rare case of squamous cell carcinoma in back ground of inverted papilloma of sphenoid sinus.

Keywords: Sphenoid sinus, Inverted papilloma, Squamous cell carcinoma

INTRODUCTION

The sphenoid bone contributes to the major part of the middle cranial fosse. Sphenoid sinus is the rare site for the primary malignancy as compare to the other sinuses. It may be secondary site for metastasis from prostate, thyroid, lung, kidney or breast cancer.¹

Inverted papilloma (IP) is a benign growth which most commonly originates from lateral nasal wall, maxillary sinus, ethmoid, frontal sinus and sphenoid sinus, among which sphenoid sinus is extremely rare.²

Etiology of IP is not known but believe to be squally of allergy, chronic sinusitis and viral infection. Any nasal mass originating from the lateral wall of nose and involving any of the para-nasal sinuses and presenting with epistaxis with nasal obstruction and features of chronic sinusitis should arose the suspicious of IP in 5th and 6th decade of life. IP has a potential for bone remodeling at the site of attachment and can be appreciated in CT scan. It may be associated with polypoidal changes as in present case. It's association with squamous cell carcinoma is around 7 to 15%.³ Long standing mass may be associated with malignant changes in the papilloma or incidence of

synchronous or metachronous malignancy. There are 7-15% chances of malignant conversion.

The objective of this study was to report a rare case of sphenoid sinus inverted papilloma along with polyp and features of squamous cell carcinoma due to ignorance of nasal symptoms for long period of time. Wide surgical excision is the treatment of choice. Some cases with intracranial extension may require post-operative radiotherapy.

CASE REPORT

A 71 years old male patient presented to ENT OPD with the chief complaints of bilateral nasal obstruction associated with discharge and recurrent episodes of sneezing from last 8 years.

He had also history of epistaxis from right nasal cavity off and on since last 1 year. He was known case of type 2 diabetes mellitus and hypertension which was under control with medication since last 12 years. On anterior rhinoscopy- polypoidal mass was seen in both nasal cavities. No bleeding was found on probing and was able to pass the probe all around the mass. Mass was showing variable consistency from soft to firm. No neurological or

ophthalmic deficits were present. Rest of the ENT and head neck examinations were within normal limits.

CT and MRI revealed lobulated polypoidal lesions about 4.1×3.2×3.3 cm with heterogeneous intermediate signal on T2 filling the nasal cavity and sphenoid sinus more on left side with erosion of floor of sphenoid sinus, nasal septum and mild extension to the nasopharynx (Figure 1A).

Initial biopsy was reported as inflammatory polyp with single fragment showing features of inverted papilloma. There was no evidence of any malignant changes. Surgery was done with Transnasal endoscopic approach with microdebrider. Wide sphenoidotomy with removal of anterior and inferior wall of sphenoid sinus along with posterior septectomy was done (Figure 2A and 2B).

Post-operative histopathology was suggestive of well to moderately differentiated Squamous cell carcinoma with back ground of inverted papilloma along with some areas showing inflammatory changes. IHC confirmed the diagnosis. Post-operative MRI (1-month follow-up) showed complete removal of tumor with no residual disease (Figure 1B). Patient received post-operative full dose of radiotherapy and doing fine after 6 months on regular follow-ups.

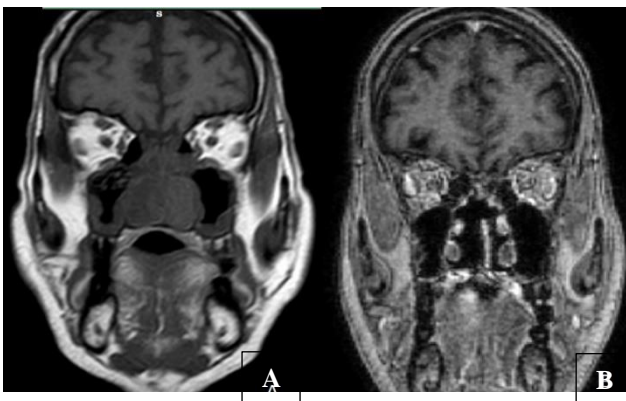


Figure 1: (A) Pre-operative MRI; and (B) post-operative MRI (one-month follow-up).

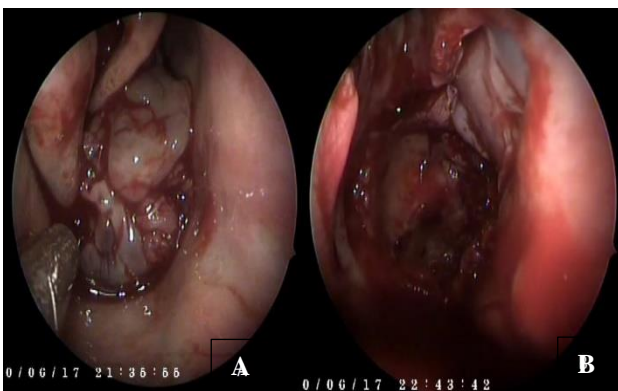


Figure 2: (A) Endoscopic picture showing polyp with firm mass; and (B) endoscopic picture showing cavity after complete surgery.

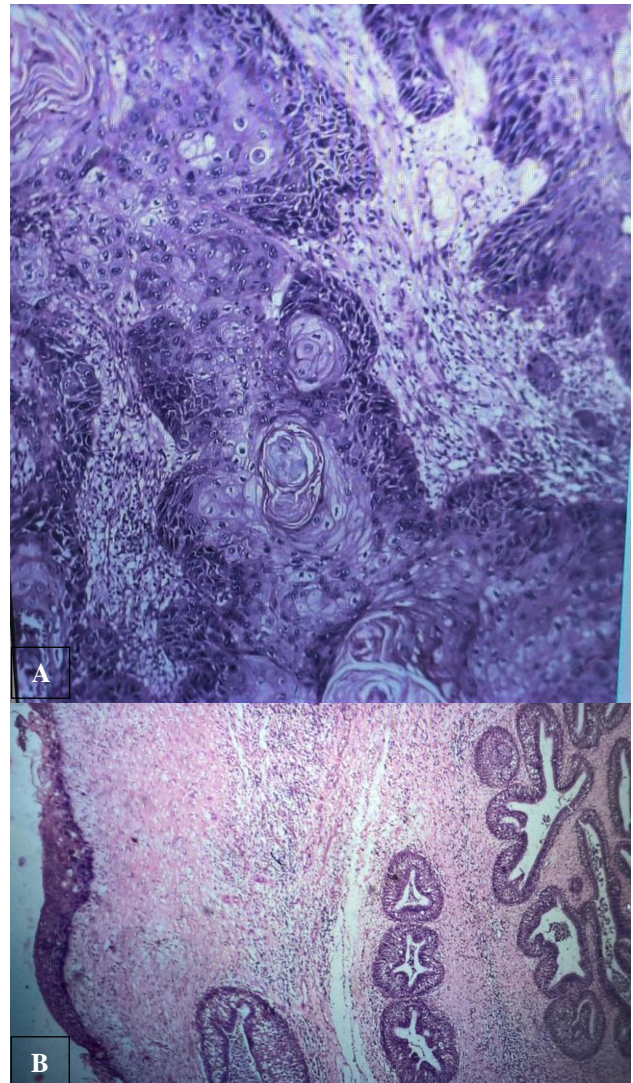


Figure 3: (A) HPR; and (B) post-operative HPR showing features of malignancy in the background of IP.

DISCUSSION

IP is a rare benign tumor most commonly originates from lateral nasal wall of nose as described by Ward in 1854.⁴ Most commonly seen in 5th-6th decade of life.

but it has been reported in all age groups.⁵ Men are affected more as compare to women with a ratio of 3: 1. About 5 to 15 percent of patients are associated with squamous cell carcinoma either subsequently or concurrently.⁶ It accounts around 0.5 to 2% of all tumors of nose. It had an endophytic growth pattern towards the Schneiderian membrane into the stroma hence the name IP.³ Bony remodeling can be seen in 65 to 75% of cases in CT scan. Tendency of its recurrence is around 20 to 47%.⁷ Histological analysis suggests that IP tumor genesis may occur through a stepwise series of discrete events graded according to a four-stage histological grading system (stages I and II, benign IP; stage III, dysplastic IP; stage

IV, carcinoma arising from IP). Viral infection and genetic insults may be required to progress.⁸

Inducing or promoting agents have been suggested in the pathogenesis of this disease. These include human papilloma virus, alterations in tumor suppressor gene p53 and chronic inflammation. Occupational exposure to different smokes, dusts, and aerosols noxious agents may play a possible role in the pathogenesis of IP. Clinical symptoms are very much similar to any nasal mass. They are nasal obstruction mostly unilateral, rhinorhea, epistaxis, facial pain, loss of smell and frontal headache. Patient may be asymptomatic at early stage but later on due to involvement of adjacent structure symptoms may appear. Unilateral epistaxis associated with nasal obstruction or nasal discharge are the warning signs of any neoplastic changes.

The most common symptoms in patients with malignant transformation include facial or dental pain, nasal obstruction, and epistaxis.⁹ On examination IP usually presents as a polypoidal mass until and unless it has involved adjacent structures. If it grows downward it pushes the soft palate anteriorly, medially may involve orbit and may cause restricted eye movements. Hypoesthesia or anesthesia of infraorbital nerve, greater palatine or speno-palatine nerve may be present. Pre-operative radiological evaluations of any nasal mass are mandatory to rule out any malignancy, extent and involvement of any adjacent structures. It also helps us for surgical planning.

There was no specific feature of IP on CT scan. In CT scan it is seen as polypoidal lesion which is usually unilateral occupying lateral wall of nose and sinuses.¹⁰ Bony deformity along with sclerosis is suggestive of slow growing neoplasm such as IP. Unilateral nasal or paranasal mass with lobulated surface is suspicious of IP. MRI study demonstrate a convoluted cerebriform pattern on both T2 and contrast enhanced T1 weighted images as in our case (Figure 1A). This MRI feature can be appreciated in 80% of IP cases. Focal areas of loss of this feature on MRI gives a hint of concomitant malignancy. With review of literature available only 50 cases were found involving sphenoid sinus.

Presenting complains of patient with inverted papilloma is generally unilateral obstruction of nasal cavity, epistaxis, rhinorhea and in the patient having sphenoid sinus involvement is nonspecific and may present with headache. The gold standard treatment of inverted papilloma is endoscopic intra nasal approach with complete removal of the tumor with least co-morbidities as compare to medial maxillectomy and enbloc resection done via lateral rhinotomy or a mid-facial degloving. Endoscopic resection gives a better view, conservative role for nasal mucosa causing less nasal slough, less pain and minimal post-operative bleeding.

No specific treatment protocol or TNM staging was found as occurrence of squamous cell carcinoma in sphenoid sinus. So, removal of tumor along with radiotherapy was the line of treatment. In literature we found radiotherapy is treatment of choice for large and in-operable tumors. Chemo along with radiotherapy can also be given if any recurrence was seen.²

Other rare cases reported for sphenoid malignancies in literature are malignant melanoma, adenocarcinoma, myxofibrosarcoma, esthesioneuroblastoma.¹¹

CONCLUSION

IP of sphenoid sinus is itself a rare tumor and its conversion into squamous cell carcinoma is even rarer. It is rare combination of polypoidal mass with inverted papilloma with malignant changes. Endoscopic resection of tumor along with removal of mucoperiosteum along with the bony site of origin along with the mass. Close follow up after surgery and radiotherapy or combined with chemotherapy is required for inoperable tumors and in cases of recurrence.

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