



Case report

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Acute proptosis: Unusual initial presentation of sinonasal inverted papilloma

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ABSTRACT

Inverted papilloma is an uncommon histopathological finding of maxillary sinus tumor. We reported here a rare case of inverted papilloma of maxillary sinus presented initially with ipsilateral proptosis. A middle-aged lady with history of having allergic rhinitis presented with progressive left eye proptosis. Assessment revealed presence of orbital mass arising from paranasal sinus, in which the histopathological examination was initially inconclusive, and later it turned up to be inverted papilloma, partly with malignant transformation. High index of suspicion is necessary especially in those patients with recurrent non-specific sinonasal symptoms, which may need repeated biopsy and histopathological examination. Ocular manifestation signifies a more advance disease.

1. Introduction

Inverted papilloma is a rare disease in the general population, with only 0.75–1.5/100000 incidence rate per year, and more prevalent in male than female[1]. It arises from an ectodermal respiratory epithelium lining on the lateral nasal wall and paranasal sinuses. It is called the Schneiderian membrane, a specialized mucosa derived from the invaginating olfactory placode in the embryo. Inversion of the neoplastic epithelium into the underlying stroma, rather than its outward proliferation, characterised the growth pattern of this tumor, hence it is termed as inverted papilloma[2].

Ophthalmic involvement is rare. Only 2.7% of inverted papilloma appears to invade orbit. Inverted papilloma with ocular involvement tends to occur in middle-aged men. Of these, lacrimal obstruction, a medial canthal mass, ocular and/or periorbital pain, diplopia, exophthalmos, and decreased visual acuity have been reported.

Aetiology of this tumor is not well understood. Several proposed risk factors include exposure to organic solvents, welding fumes, nickel compound, cigarette smoking and infectious agents. Infection

by human papilloma virus has been considered as a potential causal agent in the pathogenesis of inverted papilloma, which influences the transformation of lesion from benign to malignant[1,3,4].

2. Case report

A 52 year old lady with hypertension and allergic rhinitis, presented with six weeks history of progressive left eye proptosis with reduced vision and left facial pain. It was mild proptosis as she did not noticed any changes over the eye, but she was told as such by family members. No eye redness was otherwise noted. Ocular examination revealed visual acuity of 6/6 on the right eye, and 6/9 on the left eye. Left eye-relative afferent pupillary defect was present. Anterior segment examination showed left eye axial proptosis (Hertel exophthalmometry differs by 5 mm compared to the fellow eye) with mild restriction in depression and levoversion. There was no evidence of left eye lagophthalmos, exposure keratopathy or dry eye. The rest of the anterior segment and posterior segment details of both eyes were unremarkable. On further history, she revealed there were episodes of left nasal block and blood stained nasal discharge which spontaneously resolved after a while during which she did not seek any otorhinolaryngology advice.

Computed tomography of brain, orbit and paranasal sinuses showed aggressive left sinonasal mass (Figure 1). Examination under anesthesia and biopsy was done by otorhinolaryngology team, with histopathological examination of the tissue biopsy

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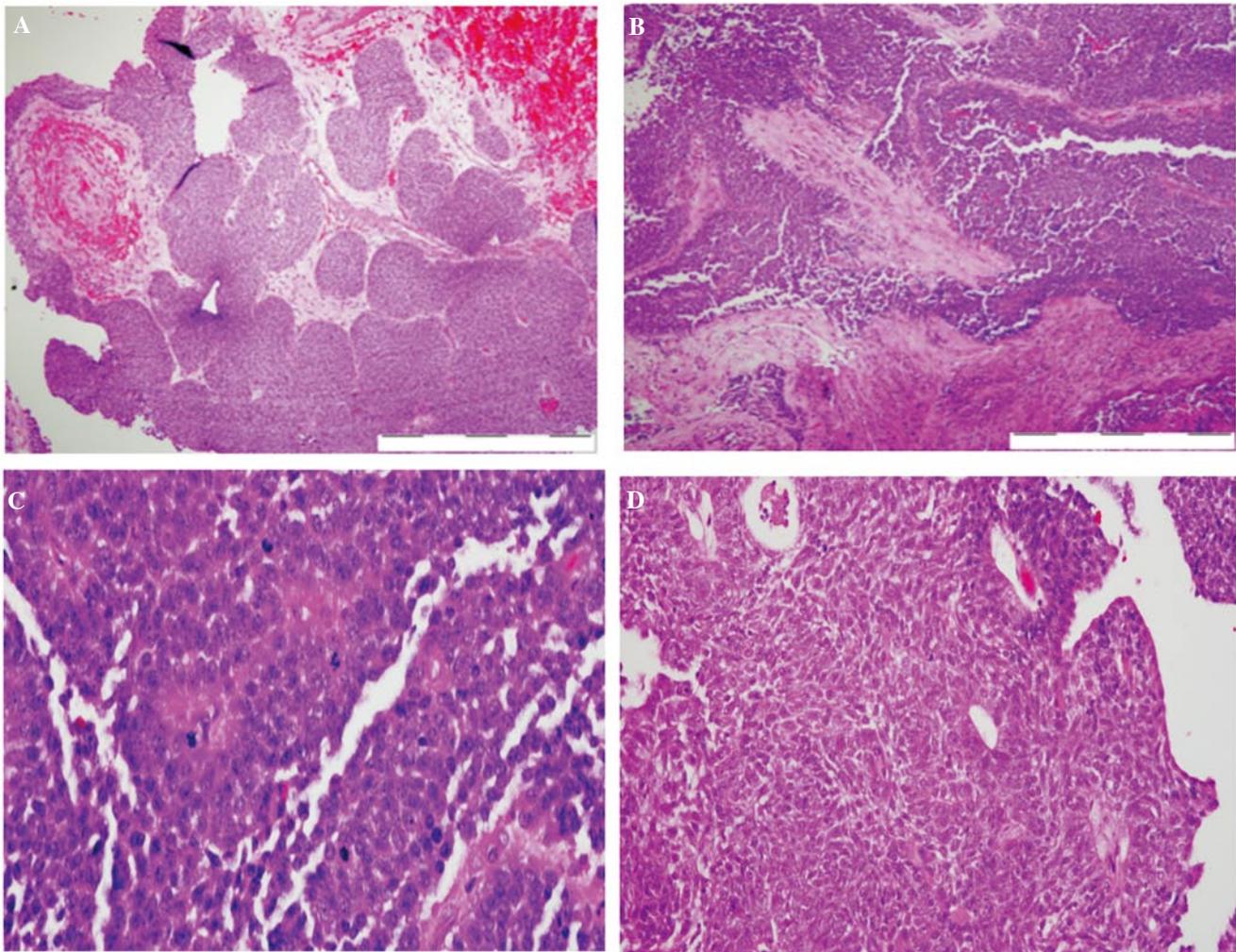


Figure 3. A: Typical histopathological features of sinonasal inverted papilloma exhibiting endophytic growth of transitional-like epithelium with intact basement membrane. (H&E x 100 magnification); B: Aggressive inverted papilloma invading and burrowing into deep stroma. The papillary epithelial cells exhibit a disordered arrangement, the basement membrane is blurred and the surrounding stroma is desmoplastic. (H&E, x 100 magnification); C: Mitotic figures including atypical form; D: Solid sheets of tumor cells showing squamoid differentiation.

reported as inverted papilloma. Further evaluation of the lesion was then done during which she was subjected for magnetic resonance imaging (MRI) of orbit and paranasal sinuses (Figure 2). The MRI revealed large mass occupying left maxillary sinus with local infiltration. Endoscopic medial maxillectomy, debulking of medial and inferior orbital mass partial resection were then performed on her. Intraoperative sample of tissue biopsy showed malignant transformation of inverted papilloma (Figure 3). Following that she was advised for total extended maxillectomy with orbital exenteration which she refused.



Figure 1. Computed Tomography of paranasal sinus and orbit showing heterogenous soft tissue mass occupying the left maxillary sinus, extending to left ethmoid sinus and left nasal cavity medially, and left temporal fossa laterally. Superiorly it invades left orbital cavity and frontal sinus. Left inferior rectus bulky and embedded within the lesion, left inferior oblique not visualized suggesting infiltration. The mass also stretching the left medial rectus. Left eye appeared proptosed out.

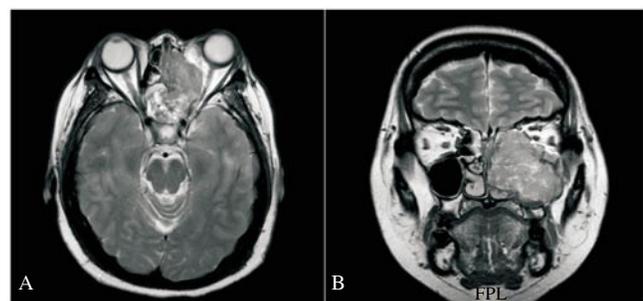


Figure 2. MRI of brain/paranasal sinus/orbit done two months after CT scan in figure 1 (a) and (b) showed large mass occupying left maxillary sinus with multiple foci of hemorrhages within, and multiple well defined proteinaceous content. The mass showed local infiltration with no intracranial extension.

3. Discussion

Paranasal sinus tumor is a rare disease of head and neck region. Cancer of paranasal sinuses accounts for only about 5% of overall head and neck malignancy. The incidence is twice more common in male than female. Early symptoms of paranasal sinus tumor may be missed, hence causing delay in diagnosis. These patients may just present with non-specific sinonasal symptoms like nasal obstruction, nasal discharge, epistaxis and facial pain, which are

similar to those typical inflammatory condition[4,5]. The ocular presentation signifies a more extensive stage of disease though it is not a common symptoms, and at most of time occurs following malignant transformation of the lesion[6]. This scenario is what happened in our patient, leading to delay in diagnosis which posed a challenge towards providing optimal treatment. Apart from ocular involvement, extension of the lesion beyond sinonasal area may cause neurological or dental symptoms.

Inverted papilloma of sinonasal primarily arise from lateral nasal wall. Despite being benign in nature, it tends to recur, and be locally aggressive and has the propensity to be associated with malignancy[7,8]. The association of inverted papilloma with squamous cell carcinoma is well recognized[2,5,6,9]. The most frequent location of this type of tumor was lateral nasal wall, followed by maxillary sinus, ethmoidal labyrinth, nasal septum, frontal sinus and the sphenoid sinus[7]. Inverted papilloma that invades the orbit has a high incidence of malignancy and is locally aggressive tumor. Definitive diagnosis is certainly by biopsy of lesion. However, false negative preoperative biopsies is possible as lesions may coincide with benign polyps[10]. This is consistent with our experience in this patient, in which malignant transformation of lesion only noted in post-operative biopsy.

Clinical staging of inverted papilloma was based on Krouse's staging system, which is widely used in most studies on this disease[7,11]. In this system, the extension and severity of disease was staged from T1 to T4. Variable extension of tumors without concurrent malignancy was staged from T1 to T3. Tumor confined to nasal cavity only was described as stage T1. In T2, the tumor grew in ostiomeatal complex, ethmoid sinuses, and/or the medial portion of the maxillary sinus, but it was not necessarily seen in nasal cavity. Stage T3 involved tumor of the lateral, inferior, superior, anterior or posterior walls of the maxillary sinus, the sphenoid sinus, and/or the frontal sinus, with or without involvement of area described in T2. All tumors with any extranasal/extrasinus extension to involve adjacent, contiguous structures such as the orbit, the intracranial compartment, or the pterygomaxillary space with associated malignant transformation was grouped in T4 stage.

The mainstay of treatment regardless of stage of inverted papilloma is complete removal of tumor bulk, and now mostly accomplished via endonasal endoscopic surgery[7]. Some authors do suggest chemo-radiotherapy following complete surgical resection patients with T4 disease in order to control recurrence at primary tumor site[5,12].

Even though our patient has been experiencing vague symptoms of chronic sinusitis, the possibility that it was tumor-related has never been thought of. Protrusion of ipsilateral eye which developed later triggered her to get a specialist opinion. By this time, assessment already showed evidence of local infiltration which is in stage T4 based on Krouse's staging system. Despite that, she is still having good vision of both eyes. This condition present as a great barrier for optimum management, as full debulking of tumor tissue is impossible without exenteration of the involved eye. In this case the involved eye is still functionally good. Hence patient's decision/consent to remove the eye is difficult to be obtained, even though risk of intracranial extension and distant metastasis from inadequate cancer tissue removal is well aware of. This will remain as a challenge to the treating surgeon.

4. Prognosis

Those with orbital involvement are likely to have a higher rate of tumor recurrence due to extensive tumor growth and incomplete

tumor excision as it is technically difficult[13]. Therefore, full tumor debulking and meticulous follow up of patient are warranted[8,11].

5. Conclusion

Inverted papilloma is an uncommon sinonasal tumor and orbital extension is even rare. Like other sinonasal masses, ocular symptoms rarely present during initial visit as it represents malignant stage of tumor.

Conflict of interest statement

The authors report no conflict of interest.

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