

Inverted Sinonasal Schneiderian Papilloma With Malignant Transformation

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ABSTRACT

Inverted papilloma is a lesion of the mucosal membrane of the nasal cavity and the paranasal sinuses. It is a unique tumour which is characterized by its tendency to recur after incomplete removal, to destroy the bone and rarely transform to malignancy. It is a rare tumour which comprises approximately 0.5 - 4 % of all the primary nasal tumours. Different names which are used

for this papilloma include villiform cancer, fungiform papilloma, cylindrical / transitional papilloma, schneiderian papilloma, transformation of papilloma and many more. We hereby report a very unusual and challenging case which elicited a range from sinonasal papilloma, with invasion, atypia and foci of transitional cell carcinoma.

Key Words: Inverted sinonasal papilloma, Malignant transformation in papilloma, Sinonasal tumours

INTRODUCTION

Inverted papilloma is a locally aggressive sinonasal tumour that arises from the outlining schneiderian respiratory membrane. It is a benign epithelial tumour that arises within the nasal wall and less commonly in the paranasal sinuses. It is a rare tumour, incidence having 0.5-4 % of all the primary nasal tumours[1]. It has a peak incidence in the 5th and 6th decades of life. It is a controversial topic due to its infrequent occurrence and confused nomenclature. Ward was the first one who described inverted papilloma in 1854. Billroth (1855) was credited for describing the first case of true papilloma of the nasal cavity and he called it 'villiform cancer'[2]. Ringertz, in 1938, was the first one to describe the microscopic appearance and the tendency of the tumour to invert into the connective tissue stroma[3]. He also marked the metaplastic transformation from columnar to squamous epithelium and emphasised its similarity with papilloma and cylindrical cell carcinoma[3]. The age of the patient vary 10 - 87 years, with a majority of the patients in the age group of 50 - 70 years, with a male preponderance (M : F is 3.3 : 1). It is a relatively rare neoplasm with a typical presentation like unilateral nasal polyp. This neoplasm is characterized by its capacity to destroy, its tendency to recur after its removal and its association with malignancy[4]. Synder et al noted a marked atypia and a significant increase in the mucous droplets within the epithelium of recurrent tumours[5]. The aetiology of the tumour still remains unknown. Sulphur, tobacco, infection and other occupational exposures have been considered as the significant aetiological factors. Investigations like fluorescence in situ hybridization (FISH) and polymerase chain reaction (PCR) have been used to determine its link with the human papilloma virus types 6 and 11[6].

CASE REPORT

A 50 - year old male presented with complaints of recurrent left nasal discharge, epistaxis and watering of the left eye since 10 months duration. On local examination, purulent rhinorrhoea and a pink polypoid mass in left nasal cavity were seen, which occluded the left nasal cavity. Radiological evaluation of the paranasal sinuses revealed a soft tissue mass which involved the left sinonasal system. Computed tomography of the nasopharynx

and the paranasal sinuses showed an enhancing mass lesion in the left maxillary sinus, which eroded the superior and the medial and extended into the orbital plate and the ethmoids - which was suggestive of a neoplastic lesion. The systemic examination was within normal limits. Left intranasal polypectomy under general anaesthesia was done and the curettage material was sent for histopathological examination.

PATHOLOGICAL FINDINGS

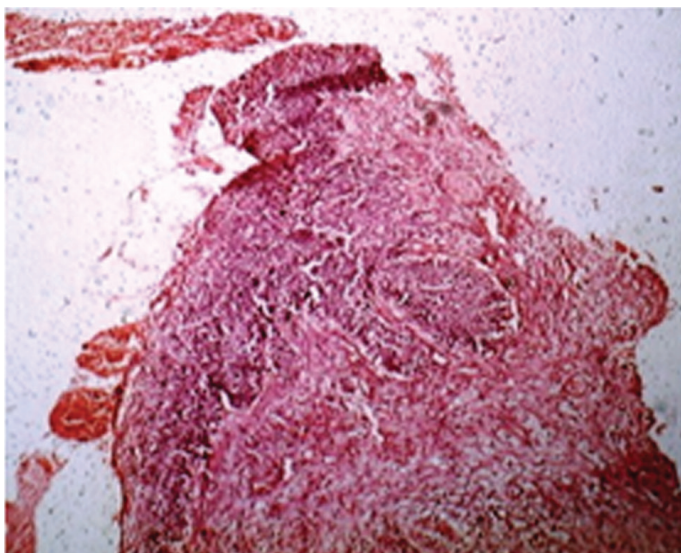
Gross : Received multiple, friable, irregular grey brown to pink, gelatinous tissue bits, totally weighing 20 grams.

Histopathology : Revealed polypoid tissue which was partially covered with columnar ciliated epithelium and areas with neoplastic squamous epithelium which showed inversion into the underlying stroma to form large clefts, ribbons and islands [Table/Fig 1]. The neoplastic epithelium was immature and showed hyperplasia, papillomatosis, extensive cellular and architectural atypia and minimal mitosis [Table/Fig 2]. Foci of transitional cell carcinoma (i.e. nuclear pleomorphism, increased mitotic figures and focal tumour necrosis) were noted in the stroma, with mild mononuclear cell infiltration [Table/Fig 3]. Histopathological diagnosis was given as inverted sinonasal papilloma with malignant transformation.

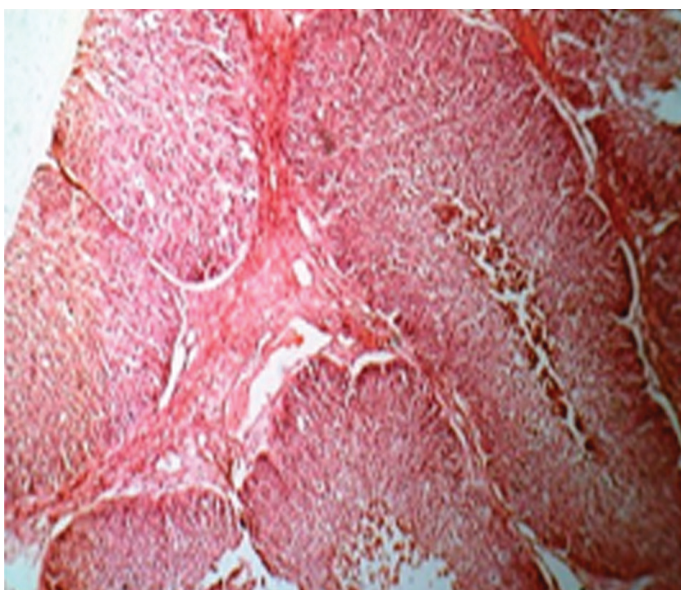
The post operative recovery was uneventful and the patient is on regular follow up with a further plan for radiotherapy.

DISCUSSION

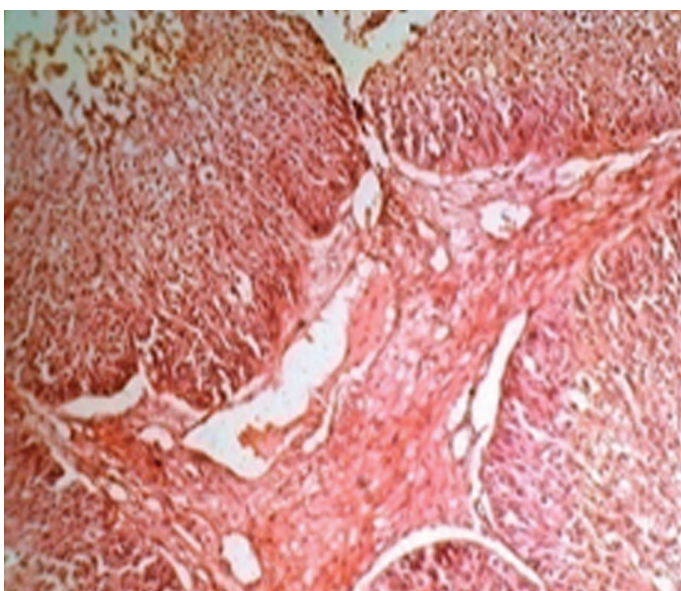
Inverted papillomas which arise from the nose and the paranasal sinuses are distinctive lesions that have been discussed in the literature for over a century. However, because of their infrequent occurrence and confusing nomenclature, they remain a topic of controversy[7]. These have been reported in the literature under a variety of titles. The designation, "inverted schneiderian papilloma" has been recommended as an appropriate title to best convey the tumour qualities of inversion, location and distinctiveness of character[4]. Ward was credited for reporting the first case of inverted papilloma in 1864. However, Ringertz was the first to describe the tendency of the tumour to invert into connective



[Table/Fig-1]: Photomicrograph showing nasal polyp with surface ulceration and lined by transitional epithelium with inverted papilloma. (H&E 10 X)



[Table/Fig-2]: Photomicrograph showing inverted papilloma with atypia. (H&E 40 X).



[Table/Fig-3]: Photomicrograph showing areas of transitional cell carcinoma. (H&E 40 X)

tissue stroma and its metaplastic transformation from columnar to squamous epithelium and he emphasized on the similarity between papilloma and cylindrical cell carcinoma[3]. The neoplasm is characterized by its capacity to invade, its tendency to recur after incomplete removal and its association with malignancy. So, otolaryngologists have given a great deal of attention to this lesion. The association between inverted papilloma and squamous cell carcinoma is well known. The reported incidence of papillomas varies from 1.7 – 7 % [8]. Many reports have documented malignant transformation in recurrent inverted papilloma, as well as inverted papilloma and squamous cell carcinoma co - existing in the initial specimen [9,10].

We report here, an unusual and rare case of inverted, transitional, sinonasal papilloma with architectural and cellular atypia, with the foci of Transitional Cell Carcinoma (TCC) - i.e. nuclear pleomorphism, increased mitotic figures and focal necrosis within the large tumour islands.

Inverted papilloma should be distinguished from TCC – a variant of squamous cell carcinoma. The islands of TCC are larger and more confluent than the discrete island of inverted papilloma (11). This finding was very well appreciated in our case [Table/Fig 3].

The surgical treatment depends upon the location of the tumour. Due to the high recurrence rate, the surgical treatment for this tumour is intranasal polypectomy with medial maxillectomy, or ethmoidectomy with follow up is preferred. The practice of sending the complete tissue which was removed at the time of surgery for histopathological studies, results in an earlier and more accurate diagnosis.

To conclude, inverted papillomas of the nose and the paranasal sinuses with invasion, atypia and malignant transformation is relatively rare. These lesions are given a great deal of attention by otolaryngologists because of their characteristic attributes of high incidence of recurrence and the propensity to be associated with malignant changes.

REFERENCES

- [1] Woodruff WW, Varbae DP . Inverted papilloma of the nasal vault and the paranasal sinuses. *Am J Roentgenol*. 1994;162(2):419-23.
- [2] Brown B. The papillomatous tumours of the nose. *J Laryngol Otol*. 1964;58:889-96.
- [3] Ringertz N. The pathology of the malignant tumours which arise in the nasal and the paranasal cavities and the maxilla. *Acta Otolaryngol*. 1938;27:31-42.
- [4] Vrabec PD. The Inverted Schneiderian Papilloma: A 25-Year Study. *Laryngoscope*. 1994;104:582-604.
- [5] Synder RN, Perzin KH. Papillomatosis of the nasal cavity and the paranasal sinuses. A clinicopathologic study. *Cancer*. 1972;30(3):668-90.
- [6] Buchwald C, Franzmann MB, Jacobson GK, Lindeberg H. The Human Papilloma Virus (HPV) in Sino-nasal Papillomas: A Study of 78 cases by using In-Situ Hybridization and Polymerase Chain Reaction. *Laryngoscope*. 1995;105:66-71.
- [7] Lyngdoh NC, Ibohah TH, Marak IC. A study on the clinical profile and the management of inverted papilloma. *Indian Jr of Otolaryngology and Head and Neck Surgery*. 2006;58(1) :41-6
- [8] Benninger MS, Robert JK, Sibek BA, et al. Inverted papilloma and associated squamous cell carcinoma . *Otolaryngol Head Neck Surgery*. 1990 ;103 :457-61.
- [9] Keshma HK, Kessiss T , Hruban RH. Human papilloma virus in sinonasal papilloma and squamous cell carcinoma. *Laryngoscope*. 1992;102:973-6.
- [10] Futuma Y, Shinohara T, Sano K, et al. Molecular study of the human papilloma virus infection in inverted papilloma and squamous cell carcinoma of the nasal cavities and the paranasal sinuses. *Laryngoscope*. 1991;101:79-85.
- [11] Brandwain-Gasler M. Sinonasal and nasopharyngeal surgical pathology. In *Silverberg's Principles and Practice of Surgical Pathology and Cytopathology*. 4th ed. Elsevier pub. 2006;788-9.

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