Case Report

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Malignant Transformation in a Case of Recurrent Sinonasal Inverted Papilloma

Ear, Nose and Throat Section

FARHAT QAMRUDDIN KHAN[1], PRASAD T DESHMUKH[2], SAGAR S GAURKAR[3]

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ABSTRACT

Inverted papilloma or Ringertz tumour is an uncommon benign tumour that arises from the Schneiderian membrane of the ectoderm. It accounts for approximately 70% of all sinonasal papillomas and 0.5-4% of all sinonasal tract neoplasms. It often originates in the middle meatus of the lateral nasal wall and is well-known for its local invasiveness and high recurrence rate. Nasal obstruction, nasal bleeding, and diminished sense of smell are the most common symptoms. Here, the authors report a case of a 58-year-old male complaining of right nasal obstruction since 3 months. The patient had nasal growth for which he underwent nasal endoscopy. History revealed that he had similar growth on the left side, 10 years ago. A nasal endoscopic biopsy was performed and the histopathological examination showed an inverted growth pattern from the sinonasal papilloma. Surgical techniques must be extensively explored and customised, with enhanced radiological assessment of tumour borders, in order to adequately remove the tumour, since incomplete excision increases the risk of recurrence and malignant transformation of the residual disease.

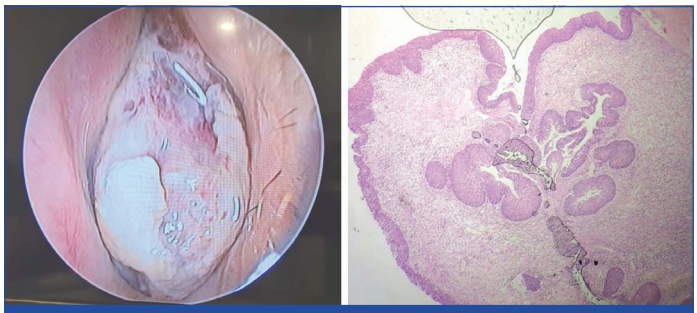
Keywords: Nasal obstruction, Ringertz tumour, Sinonasal malignancy, Sinonasal tumour, Squamous cell carcinoma

CASE REPORT

A 58-year-old male visited the Ear, Nose, and Throat (ENT) Outpatient Department with a 3 months history of nasal obstruction, initially on the left side, which then gradually progressed to the right side. He also complained of facial deformity, nasal discharge, diminution of vision, which was more in the left eye and an associated dull aching continuous type of headache [Table/Fig-1]. Additionally, the patient described a growth in the nasal cavity that had been expanding rapidly over the past 3 months. The patient had a similar history of a growth of left sided nasal mass 10 years before, for which surgery was performed at a private facility outside and the lesion was resected. Histopathology report had confirmed inverted papilloma. On diagnostic nasal endoscopy, a single, pedunculated, polypoidal, firm, reddish pink mass was seen in bilateral nostril [Table/Fig-2]. Probing revealed adherence to the lateral wall of the left nasal cavity and did not bleed on touch.

[Table/Fig-1]: A): Lateral and B): Anterior view of tumour leading to significant facial deformity.

On Computed Tomography (CT) imaging, an aggressive mass was seen in the left nasal cavity, expanding into the adjacent lumens of bilateral ethmoid, frontal, sphenoid, left maxillary sinus and nasopharynx, extending into right nasal cavity, eroding nearby bony structures with left submandibular lymphadenopathy (22×10 mm). A nasal endoscopic biopsy was performed, and the histopathological examination showed an inverted growth pattern from the sinonasal papilloma component with markedly thickened epithelium with ciliated luminal cells growing downward into the underlying stroma, showing areas of severe epithelial dysplasia, suggestive of inverted papilloma associated with squamous cell carcinoma [Table/Fig-3]. In view of recurrence, malignant transformation and extensive local invasion, the patient was referred to the Department of Radiation Oncology for further management where he is undergoing the radiotherapy for the same.



[Table/Fig-2]: Diagnostic nasal endoscopy revealing pinkish mass in the nasal cavity.
[Table/Fig-3]: Histopathological examination suggestive of inverted papilloma associated with squamous cell carcinoma (40X). (Images from left to right)

DISCUSSION

Inverted papilloma, also known as Ringertz tumour, transitional cell papilloma, and sinonasal papilloma, are epithelial neoplasms that arise from the Schneiderian membrane that covers the nasal cavity and paranasal sinuses [1]. It is an uncommon tumour that accounts for between 0.5-4% of all primary nasal lesions [2]. It often originates in the middle meatus of the lateral nasal wall, frequently involving the ethmoid and maxillary sinuses and may extend to the orbit or intracranial cavity. There is a predominance of male gender and more often affects the age group of 40-70 years. Being benign, it is well-known for its local invasiveness. It does not infiltrate the underlying bone; rather, the surface boundary between the inverted papilloma and the bone is pathological, uneven, and riddled with cervices. These bony gaps are replaced with the diseased tissue, resulting in a site for tumour recurrence. The recurrence rate is quite high (13.72%), and it occurs within 2 years following surgical intervention and also has an increased propensity for malignant transformation (7.6%) [3]. Nudell J et al., analysed the behaviour of 740 cases diagnosed in a 10 year period and found a malignant transformation in 20 (prevalence of 1.9%) among the entire patient cohort [4].

Various risk factors, including Human Papillomavirus (HPV) infection, smoking, occupational exposure, and Deoxyribonucleic Acid (DNA)

alterations, such as Epidermal Growth Factor Receptor (EGFR) mutation have been identified as plausible reasons in the malignant transformation of sinonasal papillomas [5]. In a group of 20 patients with inverted papilloma, 70% of the patients had HPV DNA, according to Altavilla G et al., [6]. A p53-positive immunostaining was associated with the presence of HPV DNA, suggesting a link to dysplasia and eventual transformation. Despite the fact that a number of researchers have discussed the potential carcinogenic involvement of HPV in sinonasal papillomas with squamous cell carcinoma, the vast majority of them have been unable to provide statistically significant findings [6].

Nasal obstruction, nasal bleeding, and diminished sense of smell are the most common symptoms. These basic manifestations may be accompanied by headaches, lacrimation, or visual impairment [7]. Owing to its diverse clinical appearance, distinguishing benign inverted papilloma and inverted papilloma associated with malignant transformation presents a diagnostic and therapeutic challenge for the otorhinolaryngologists. The Krouse JH staging approach is used to determine the clinical stage of sinonasal inverted papilloma [8].

Surgical techniques must be extensively explored and customised. Since, the advancement of technology, including the use of CT scans and Magnetic Resonance Imaging (MRI) of the Paranasal Sinuses (PNS), the management of sinonasal papillomas using endoscopic technique has undergone a revolution. Open approaches like lateral rhinotomies and the midfacial degloving surgeries enable better visualisation and a thorough removal of a large tumour, including a maxillectomy, which reduces the probability of recurrence [7]. A long-term follow-up postoperatively is crucial for early detection of recurrence.

In situations involving malignant transformation, the most appropriate treatment is complete excision of the lesion, followed by radiation with or without chemotherapy. Antrochoanal polyps, adenoid cystic carcinoma, maxillary squamous cell carcinoma, fibrous dysplasia, giant cell granuloma, and other neoplasia are the most common differential diagnosis [9].

Squamous cell carcinoma in inverted papilloma has a poor prognosis. Survival rate after five and 10 years are 39.6% and 31.8%, respectively. The prognosis is worse for tumours found in older patients who are poorly differentiated, well-advanced, and have infiltrated the cranial base or the orbital cavity [10].

CONCLUSION(S)

Although inverted papilloma is a benign condition, it is aggressive locally, and the management should include extended and personalised surgical approach with enhanced radiographic assessment of the tumour border, as incomplete excision increases the risk of recurrence and malignant transformation of the remnant tissue. A long-term regular follow-up is recommended for early detection of recurrence and in cases of malignant transformation, radiotherapy with or without chemotherapy should be included in the treatment protocol.

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PARTICULARS OF CONTRIBUTORS:
1. Resident, Department of Ear, Nose and Throat, Datta Meghe Institute of Medical Sciences, Wardha, Maharashtra, India.
2. Professor and Head, Department of Ear, Nose and Throat, Datta Meghe Institute of Medical Sciences, Wardha, Maharashtra, India.
3. Associate Professor, Department of Ear, Nose and Throat, Datta Meghe Institute of Medical Sciences, Wardha, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Farhat Qamruddin Khan,
Resident, Department of Ear, Nose and Throat, Outpatient Department, Jawaharlal Nehru Medical College, Sawangi (M), Wardha, Maharashtra, India.
E-mail: farhatqk@gmail.com

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