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Original Article

Sinonasal inverted papilloma in eastern part of Nepal

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Abstract
Objective: To evaluate the clinicopathological profile of sinonasal inverted papilloma in a tertiary referral center in eastern part of Nepal.
Methods: A prospective study consisting of total 28 histologically proven cases of sinonasal inverted papilloma was conducted in the ENT department of the center. The study period was from April 2002 to November 2005.
Result: Out of 28 cases studied, 21 were males and 7 were females with male to female ratio of 3:1. The highest incidence was seen in the 5th and 6th decades of life. The duration of symptoms varied from 5 months to 20 years with mean duration of 3.9 years. The lateral nasal wall and nasal cavity involvement was seen in all 28 cases. Maxillary sinus was involved in 27(96.4%) subjects followed by ethmoid (20), sphenoid (10) and the frontal sinus (7). The principle presenting symptom was nasal obstruction seen in 96.4% of the patients. Twenty two cases were treated by lateral rhinotomy with medial maxillectomy while, orbital exenteration was done in two cases. Associated carcinoma was noticed in 10.15% of all the subjects studied.
Conclusion: Sinonasal papilloma mostly presented in fifth to six decades of life mainly affecting the male patients. Majority of the patients were treated by lateral Rhinotomy with medial maxillectomy surgery. Early presentation would have given chances for endoscopic surgery.

Key words: Sinonasal inverted papilloma, lateral Rhinotomy, medial maxillectomy.

Since 1991, three different histopathological types of sinonasal papilloma have been classified by World Health Organization as Exophytic papilloma, Inverted Papilloma, and Columnar cell papilloma[1]. Inverted papilloma (IP) is a true benign epithelial tumour of the sinonasal tract characterized by the invagination of neoplastic epithelium in to the underlying stroma without any sign of local aggression of basement membrane. A variety of names have been given by different authorities like villiform cancer, Ewing's papilloma, papillary sinusitis, transitional cell papilloma, Schneiderian papilloma, epithelial papilloma, squamous papillary epithelioma and papillomatosis[2,3]. The first description of this tumour was given by Ward in 1854 and Billroth in 1855. Ringertz in 1938 described its local invasive nature in details and named it as inverted papilloma[4,5].

Although inverted papilloma is a rare tumour representing from 0.5 to 4 % of all neoplastic growths of the sinonasal tract. It accounts for 70% of all sinonasal papillomas[6,7]. Its exact aetiopathogenesis is still poorly understood. Some proposed aetiological hypothesis include viral infection, smoking, allergy, chronic inflammation and occupational exposure[8].

IP commonly originates from the lateral nasal wall or middle meatus often extending to adjacent paranasal sinuses or other nearby structures such as the orbit or skull base.[2,9] This tumour is rarely multicentric and bilateral and is often associated with malignancy (5to15%). Symptoms depend upon the anatomical site of its occurrence. Unilateral nasal obstruction has been reported as the most frequent presenting symptoms in most of the patients[10,11].

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Because of its characteristic features such as local invasiveness, frequent association with squamous cell carcinoma (SCC), and high incidence of recurrence, IP is considered as an aggressive lesion. It is therefore best treated with a medial maxillectomy by an external approach. However, in the early 1980s, endonasal surgery with a purely endoscopic or a micro-endoscopic approach was introduced, providing the head and neck surgeon with less invasive techniques for the management of IP. Some authors still advocate the superiority of traditional techniques for advanced lesions and cast doubts on the possibility of obtaining adequate exposure of the entire sinonasal complex by endoscopic surgery. Moreover, the need for long-term follow-up to establish the efficacy of a surgical approach has been emphasized for its late recurrences[2-7].

This report analyzes for the first time from Nepal a series of 28 patients with IP treated by external as well as endoscopic approach at BP Koirala Institute of Health Sciences located in Eastern Nepal. We presume that the sample is representation of the population of eastern region of this country because this center is the only tertiary referral center for ENT Head and Neck surgery.

Materials and methods

A total of 28 diagnosed cases of inverted papilloma of nose and paranasal sinus attending the ENT OPD at BP Koirala Institute of Health Sciences, Dharan from April 2002 to November 2005 were included in the study. A detailed history was taken from every patient. A thorough clinical examination including ENT evaluation was performed. Biopsy was taken in all cases and diagnosis confirmed by histopathological examination. All patients underwent CT scan to see extent of the tumour prior to surgical resection under general anaesthesia. Pre and post operative antibiotics were used in all cases. Pre-operative dental and ophthalmic consultations were taken. Dental team was involved to manage post operative deformity and prosthetic implantations. Postoperatively specimens were sent for histopathological examination. Lateral Rhinotomy, medial maxillectomy, total maxillectomy, orbital exentration and endoscopic surgery were performed either separately or in combination.

All patients were followed up subsequently at regular intervals from 6 months to 30 months. First follow up visit was scheduled after 2 weeks, then 2 monthly for 6 months followed by 6 monthly intervals. All the patients underwent detailed clinical examination and endoscopic evaluation to search for any signs of recurrence. Suspicious lesions were biopsied to rule out recurrence. Any other complaints and complications were recorded. Those cases having followed up period of less than 6 months were excluded from the study. The clinical presentation, management and outcome were recorded.

Results

The mean age at onset of symptoms was 46.7 years. The highest number of patients was seen in the 5th and 6th decades of life (Fig. 1). The male-to-female ratio was 3:1 (Fig. 2). Nineteen (67.9%) patients were belonging to Mongoloid race from hilly areas of the country while nine (32.1%) were Indo-Aryans from the plains.

The duration of symptoms in our series varied from 5 months to 20 years with mean duration of 3.9 years. The nasal obstruction was the chief presenting symptom in 27 cases (96.4%) that was unilateral in 12 cases (44.4%) and bilateral in 15 (55.6%). The other presenting symptoms were the nasal mass in 20 cases (71.2%), headache in 15 (57.1%), anosmia in 7 (25%), nasal deformities in 4 (14.3%), proptosis in 4 (14.3%), aural fullness in 4 (14.3%), epistaxis in 3 (10.7%) and palatal swelling in 1 (3.6%) (Fig. 3).

Right side (64.3%) involvement was predominant than the left side (35.7%). None of the patients had bilateral nasal involvement. The lateral wall and nasal cavity were involved in all the 28 cases (100%). The maxillary sinus was the most common sinus involved in 27 (96.4%) followed by ethmoid sinus in 20 (71.2%), sphenoid sinus in 10 (35.7%), and the frontal sinus in 7 (25%) cases (Fig. 4). Orbital exenteration was performed in four patients (14.3%) and of these four subjects, sino nasal malignancy was observed in three patients.

Out of 28 cases studied in present study, lateral rhinotomy with medial maxillectomy was performed in 22 cases (78.6%), while total maxillectomy was done in four cases (14.3%). Out of these four subjects, squamous cell carcinoma was seen in three cases. Two cases (7.14%) with squamous cell carcinoma were treated by orbital exentration while in one; the tumour clearance was performed by medial orbitotomy. In two cases (7.14%), endoscopic sinus surgery was done. Three cases having carcinoma were referred for post operative radiation therapy.

No major surgical complications were seen except for epiphora in 2, ectropion in 3 and prosthesis related problems in 4 cases after total maxillectomy. Two patients developed recurrence of IP in the whole study period.

Fig1: Age distribution of the patients with IP in eastern Nepal

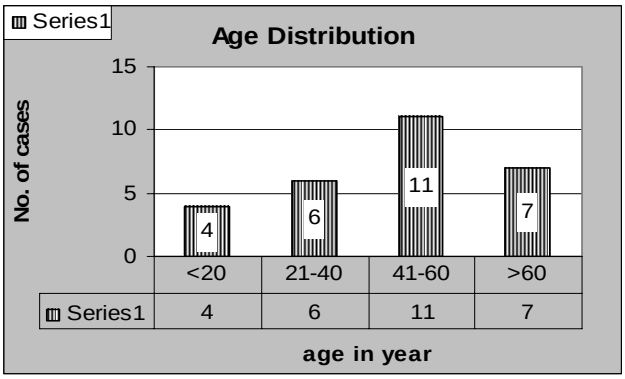


Fig 2: Age distribution of the patients with IP

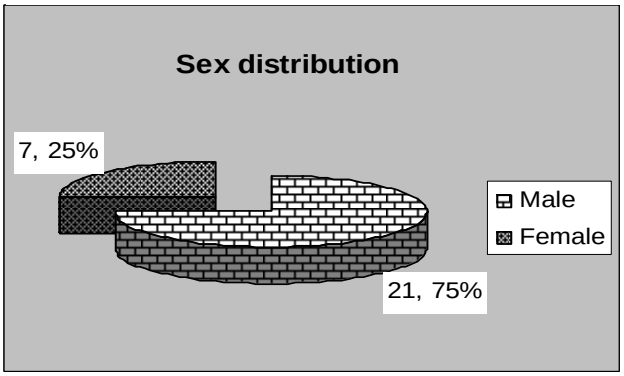


Fig 3: Presenting symptoms of IP

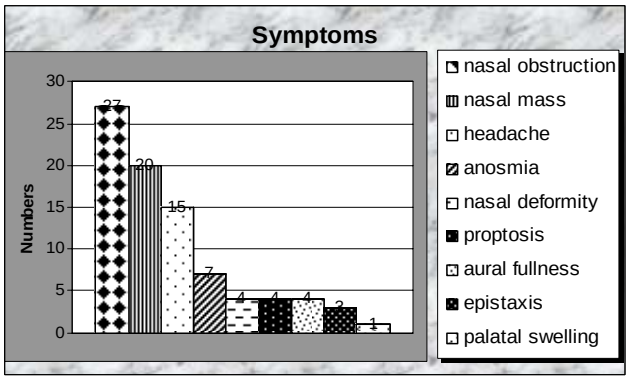
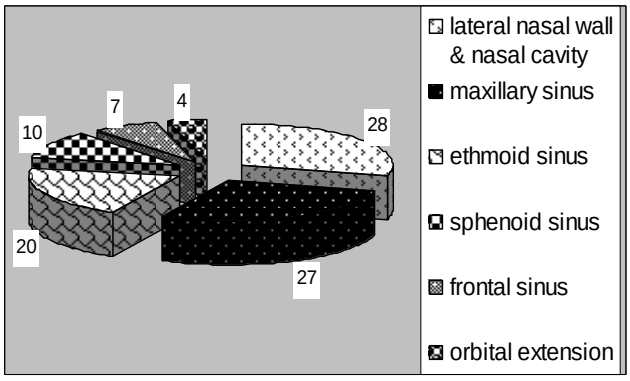


Fig 4: Anatomical location of the IP



Discussion

The clinical presentation of IP has been well studied. It has been reported in all age groups, but is known to have a peak incidence in 5th to 7th decades.[3, 7,8] Although isolated observation in the paediatric age group has been reported in literature, the youngest patient that we got in this study was 17 years old. Age range in our study was from 17 to 73 years (mean 46.7years) with maximum number in the 5th and 6th decades, which is consistent with the study of Tomenzoli et al and Throp MA et al[3,7]. Our results showed a 75% male predominance which is in accordance with most recorded series.[3,12] Interestingly 67.9% of cases were seen among mongoloid races from hills as compared to the 32.1% seen among Indo- Aryan living in the plains. Exact reason for this occurrence is not known. Further study would be helpful to explore the reason for this association.

Although the aetiology of IP is still unknown, recent studies using in situ hybridization and polymerase chain reaction have detected human papilloma virus in up to 86% of IP. In particular, viral subtypes 6, 11, 16, and 18 were most frequently found[7,13]. The other proposed aetiology for the genesis of inverted papilloma would be smoking, allergy, chronic inflammation and occupational exposure. Recently, a number of studies have been done to make genetic predilection for the origin of inverted papilloma. Keles et al revealed reduced level of p27 expression correlated with increased cell proliferation in sinonasal tumours. However, variable p21 and p53 expressions were detected in both benign and malignant tumours of sinonasal epithelium[7,14,15].

The clinical presentation of the IP is variable depending upon the extent of the involvement of the surrounding structures and the presence of other secondary pathology. However, the unilateral nasal obstruction is the commonest presenting symptom reported in most of the series[3,9]. In our patients we got nasal obstruction in 27 cases (96.4%) as a principle symptom that was unilateral in 12 cases and bilateral in 15 cases. Bilateral nasal obstruction may be due to the disease itself causing the pressure and deviation of the nasal septum towards the opposite side that was seen in all the 15 cases. In addition to that, it is due to compensatory hypertrophy of inferior turbinate of healthy side. The other presenting symptoms were the nasal mass, headache, anosmia, epistaxis and proptosis. The proptosis was seen in 4 cases, out of which three had had an associated malignancy. These results are in accordance with the findings in the literature[7,9,10,15].

The duration of symptoms in our series was from 5 months to 20 years with mean duration of 3.9 years. This may be due to the illiteracy, unawareness of health and health services and the poor economic condition. It is a common practice among the general population to choose the cheaper and readily available indigenous methods for treatment, before reporting to the higher center. Thorp et al have reported the mean duration of symptoms of 7.2 months in their series[3]. Vrabec reported the duration of symptoms from 2 weeks to 45 years[16].

In our series, the lateral wall was involved in almost all cases as assessed clinically, from CT scan and by nasal endoscopic examination. The maxillary sinus was the most common sinus involved (96.4%) followed by ethmoid sinus (71.2%), sphenoid sinus (35.7%), and the frontal sinus (25%). Four patients had orbital extension, out of which three were associated with malignancy. In no cases the contralateral nasal cavity was involved. Our study is in accordance with the several other studies in terms of sinus involvement[10,17,18]. However, ethmoid sinus was found to be the commonest sinus involved followed by maxillary, sphenoid and lastly the frontal sinus[2,4,7]. The higher incidence of sinus involvement in our series is because of the delayed presentation. Therefore, most of the cases presented in advanced stage of the disease.

The basic treatment of the tumour is regional resection of the mass leaving free healthy margins. This aim can be achieved more effectively with more aggressive surgery through external approach. Lateral rhinotomy and medial maxillectomy used to be the recommended surgical procedure in majority of the cases of IP[9]. However; more submissive endoscopic surgery is getting momentum now-a-days with controversial results. In spite of that, most authors agreed on the more subdued endoscopic surgery for very limited disease and aggressive external approach for extensive disease. Because we got cases in advanced stage we, did lateral rhinotomy with medial maxillectomy in most of the cases (78.6%), while total maxillectomy was done in four cases. Out of these four three were associated with squamous cell carcinoma and one had extensive erosion of the lateral and posterior wall of maxilla and tumour extension in cheek, infratemporal fossa, pterygopalatine fossa and nasopharynx. Orbital exentration was done in two cases having squamous cell carcinoma. In two cases we did endoscopic approach, where the disease was limited to nose and osteomeatal complex. Our treatment policies are well supported by several other studies[4,7,10,17]. The association of squamous cell carcinoma with IP is well documented in English literature varying from 5-25%. Lawson et al has reported an association in 9% in his series out of the 112 cases studied, while we got an association of carcinoma in 10.7% of cases in our series. The recurrence rate in our series is fairly lesser because we adopted more aggressive surgery and proper tumour clearance with invariably healthy margins in most of the cases[8,9].

Conclusion

The present study indicated that the inverted papilloma is a benign but an aggressive tumour of sinonasal tract. Thus the time of presentation and time of intervention are the two prime factors influence the outcome of the treatment. The patient of IP presented late in Nepal. Majority of the patients are at hand with the sinus involvement. More aggressive surgery is the only option left for the management of this disease however; detection of disease in early stage may provide a chance to do conservative endoscopic surgery.

Larger series and longer period of prospective study and follow up required to determine the histological classification, to determine relation with malignant transformation, to show relation with ethnicity, to compare efficacy of different surgical modalities and to determine the responsible etiological factors.

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