Verrucous carcinoma of the maxillary antrum

R Indudharan, P K Das, T Thida

ABSTRACT
Verrucous carcinoma of the paranasal sinuses is a rare malignant tumour. It occurs most frequently in the oral cavity and larynx. We present a case of extensive verrucous carcinoma of the maxillary sinus. This paper discusses the clinical and histological features and modes of treatment. All previous reports of verrucous carcinoma cases of paranasal sinuses are reviewed.

Keywords: verrucous carcinoma, paranasal sinus, radiotherapy

INTRODUCTION
Verrucous carcinoma has always been a subject of controversy regarding clinical presentation, diagnosis and treatment ever since Ackerman described it in 1948. The commonest sites of involvement in the head and neck regions have been oral cavity and larynx where it represents 2%-9% and 2% respectively. So far only 4 cases of verrucous carcinoma have been reported from the maxillary sinus. Maxillary sinus tumours are often diagnosed late, but such an advanced case of verrucous carcinoma as is described here has not been reported before. Despite its highly malignant clinical appearance, the tumour is deceptively benign histologically and cytologically.

CASE REPORT
A 35-year-old male Malay labourer presented with a large fungating ulcerated lesion on the right side of the face with trismus. He was apparently well till 14 months ago when he had right upper tooth ache for which he underwent a tooth extraction from right upper quadrant by a dentist. The tooth socket never healed and remained painful. He consulted a general practitioner and received conservative treatment. Six months later, he noticed a gradually increasing swelling on his right cheek for which he visited several general practitioners who treated him for abscess in the cheek, secondary to infected tooth socket. The patient also sought help from traditional healers; the nature of treatment offered by them is not clear. However, over a period of 5 months, the swelling ulcerated with purulent discharge. The patient continued with local GP management. Three months before admission, he noted difficulty in opening his mouth and one month before presentation, he started to have severe right-sided headache, which resulted in frequent insomnia.

On clinical examination, there was a fungating infected growth of approximately 10 x 10 cm involving the entire right cheek and right lateral orbital margin (Fig 1). The growth was protruding through a small dehisence in the right gingivolabial sulcus and through the tooth socket of the second molar of the right upper quadrant. The hard palate appeared infiltrated and had crossed the midline. There was trismus and posterior rhinostompy revealed right choana filled with mucopus. Movement of the eyes and vision were normal. A firm submandibular lymph node of 2 x 2 cm was palpable on the right side. Examinations of all other systems of the body were normal.

Culture of pus from the mass grew mixed gram negative organisms. X-ray of the paranasal sinuses showed an opaque right maxillary antrum with expansion of sinus and destruction of anterolateral wall. CT scan confirmed the X-ray findings and in addition, revealed destruction of floor and lateral margin of the right orbit and infiltration into the right superior nasal meatus, nasopharynx, infratemporal fossa, hard palate and right pterygopalatine space (Fig 2).

Histopathological examination of biopsy from the growth on the cheek and right gingivolabial sulcus showed a proliferative growth made up of stratified squamous epithelium with infiltration as cords of rete pegs containing keratin plugs into deeper tissue. There was minimum pleomorphism and mitosis with large number of chronic inflammatory cells under the surface layer (Fig 3). The histological diagnosis was verrucous carcinoma.

The tumour was staged as T4 N1 M0. Due to the extensive nature of the tumour, a total extended maxilloctomy with possible orbital exenteration and post-operative radiotherapy was suggested. The patient was not willing to go for surgery since a cure could not be assured. Hence a palliative radiotherapy of 3,500 centigray in 12 sittings extended over a period of 2 weeks, which...
Fig 2 - CT scan showing growth involving the floor and lateral orbital margin, superior nasal meatus, nasopharynx, infratemporal fossa, hard palate and right pterygopalatine fossa.

followed by chemotherapy with 4 cycles of methotrexate 80 mg, every 2 weeks and 6 cycles of bleomycin 15 mg every 2 weeks was given. The response of the tumour to the treatment was dramatic. The growth reduced in size completely and the cheek healed. The submandibular lymph node disappeared and there was relief of headache and trismus. Though further surgery was suggested, the patient was not willing. The patient remains free from recurrence at the time of writing this report, which is twenty-eight months after chemotherapy (Fig 4).

DISCUSSION

Verrucous carcinoma is a highly differentiated, slow growing, locally aggressive tumour, occurring most commonly in the oral cavity and larynx in the head and neck regions. In paranasal sinuses, this tumour has been reported only four times (Table I) from the maxillary antrum (6-9) and once from the frontal sinus (10). This is the first time that such a case is reported from South East Asia. The present case reports rapid progression of a generally slow growing tumour. Unless there is close co-operation between the pathologist and the surgeon, and a high degree of suspicion is maintained, the lesion may be reported as benign in spite of the aggressive and destructive clinical picture (29). Ferlito et al. (30) emphasised on the following classic description for diagnosis of verrucous carcinoma:

1. Fungating warty tumour.
2. Thickened club shaped, papillomatous projections which push rather than infiltrate into the underlying tissue.
3. Deeply projecting cleft-like spaces with degenerating keratin and later cystic degeneration of central portion of the filiform projections.
4. High degree of cellular differentiation with absence of features of malignancy.
5. Considerable inflammatory response in invaded tissues.
6. Rare regional lymph node and distant metastasis.

The main histopathological differential diagnosis of verrucous carcinoma is from leukoplakia, papilloma, pseudoepitheliomatosus hyperplasia, verrucous hyperplasia and highly differentiated squamous cell carcinoma (grade one) (32).

Treatment of verrucous carcinoma remains controversial. Krauss (33) et al. reported that radiotherapy is ineffective for this tumour. Radiotherapy is reported to cause anaplastic change in the tumour within 6 months, making it more aggressive with

Fig 3 – Nests of well-differentiated squamous carcinoma having a verrucous profile typical of verrucous carcinoma (H&E x100).

Fig 4 – Post-radio and chemotherapy response of the patient.
chance of distant metastasis\(^{12,13}\). However, Burns et al\(^{14}\) did not have a single example of anaplastic transformation following radiotherapy in 30 cases of oral verrucous carcinomas. Batsakis et al\(^{15}\) suggested that the selection of surgery over radiotherapy should be based on the recorded effectiveness of each modality and not on the phenomenon of anaplastic transformation reported to follow radiotherapy. Bar et al\(^{16}\) reported a case of this tumour in the larynx that responded to radical radiotherapy, though 10 weeks following radiotherapy macroscopic residual tumour was evident and total laryngectomy was considered. At the same time, Nair et al\(^{17}\) reported radiotherapy results of 52 cases of oral verrucous carcinoma to be comparable with that of surgical treatment of these tumours.

In the present case, the extensive nature of the tumour and the lack of patient consent prevented radical mutilating surgery that may appear to be the recourse for a slow growing, limited tumour. Radical surgery as the first choice of treatment for verrucous carcinoma has to be reviewed based on all the reports of clinical response to radiotherapy of this tumour. This case is reported to highlight the possible role of radiotherapy in verrucous carcinomas of the head and neck region and to bring to light the importance of the high degree of suspicion to be maintained in dental and general practice for diagnosing paranasal sinus tumours.

**REFERENCES**


Table I – Verrucous carcinoma of paranasal sinuses (reported cases)

<table>
<thead>
<tr>
<th>Reported by</th>
<th>Presentation</th>
<th>Treatment</th>
<th>Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Elliott et al(^{14})</td>
<td>Dislodgement of denture with growth in hard palate</td>
<td>Infraorbital maxillectomy followed by radium mould with 4000 rads in antrum of 0.5 cm.</td>
<td>No recurrence in 5 years</td>
</tr>
<tr>
<td>Bacon et al(^{15})</td>
<td>Polyp right nasal cavity for 10 years.</td>
<td>Right hemimaxillectomy and ethmoidectomy.</td>
<td>Not reported</td>
</tr>
<tr>
<td>Argawal et al(^{17})</td>
<td>Left-sided nasal discharge, nose block, post-nasal drip, pain over left cheek - few months.</td>
<td>Total left maxillectomy</td>
<td>Not reported</td>
</tr>
<tr>
<td>Present case</td>
<td>Ulcerated lesion right cheek with trismus and right-sided headache.</td>
<td>Radiotherapy 3500 rads followed by chemotherapy. Methotrexate 80 mg/week x 4 cycles and Bleomycin 15 mg/week x 6 cycles</td>
<td>No recurrence in 28 months.</td>
</tr>
</tbody>
</table>