

CASE REPORT

High-grade mucoepidermoid carcinoma of maxillary sinus

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ABSTRACT

Malignant tumors of the paranasal sinus are uncommon, constituting less than 1% of all malignancies and 3% of all head and neck cancers. Nonsquamous cancers of the maxillary sinus are even rarer as is evident from the limited data available on the clinical characteristics and outcomes. Mucoepidermoid carcinoma accounts for 13% of all malignancies occurring in maxillary sinus. We report a rare case of high-grade mucoepidermoid carcinoma of maxillary sinus in a 45-year-old female along with review of the literature.

Key words: Carcinoma, maxillary sinus, mucoepidermoid, salivary gland

INTRODUCTION

Malignancies of the nasal cavity and paranasal sinuses constitute fewer than 1% of all malignancies and 3% of upper aerodigestive tract malignancies.^[1] The majority of these tumors are in the maxillary sinus, and squamous cell carcinoma is the commonest histological type.^[2] As a subset of maxillary sinus malignancies, non-squamous cell cancers of the maxillary sinus are rare entities.^[3] Kraus *et al.* in a study of 49 patients with nonsquamous tumors of maxillary sinus reported sarcomas, adenoid cystic carcinomas, lymphoma, and adenocarcinoma accounting for most of their cases.^[4] In contrast Bhattacharya^[3] in his series of 188 cases had adenoid cystic carcinoma (34%) as the predominant histological type followed by sarcomas (24%). Sinonasal malignancies occur twice as often in males as in females, and are most often diagnosed in patients 50 to 70 years of age.^[5] Mucoepidermoid carcinoma arising from mucous glands of maxillary sinus are extremely rare and accounts for 13% of all malignancies occurring in maxillary sinus.^[1]

We report a rare case of high-grade mucoepidermoid carcinoma of maxillary sinus in a 45-year-old female.

CASE REPORT

A 45-year-old female came to the department of oral medicine and radiology with the complaint of pain and swelling on the

right side of the face. She gave history of visiting a dentist 2 months back with complaint of pain in the right maxillary posterior teeth region following which extraction of 16 and 17 was done. There was no relief; further she noticed occurrence of a mild swelling on the right side of the face that made her to report to our department. The pain was continuous, radiating, aggravated on mouth opening and on lying down, not getting relieved on medication. The swelling was sudden in onset, noticed few days after extraction of tooth, static and mildly painful.

There was no significant past medical history, family history or personal history. On general examination she was moderately built and nourished, alert, responsive, and cooperative. Vital signs were all within normal range.

Extra orally, a solitary diffuse swelling was noticed unilaterally on the right middle one-third of the face causing gross asymmetry of the face [Figure 1]. Swelling was irregular,



Figure 1: Swelling of the right maxilla causing gross asymmetry of face

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measuring about 3 cm in its greatest extension, located over the malar region, extending from infraorbital rim to the ala of nose. Skin over swelling was normal.

On palpation the swelling was mildly tender, firm, not mobile, not fluctuant, not compressible and not reducible. Skin over the swelling was normal.

Intraorally, a solitary swelling was noted on buccal and palatal alveolus with respect to the right posterior maxilla. Swelling was irregular in shape, extending from alveolar region of 14 to tuberosity of maxilla, partially obliterating the buccal vestibule. Palatally a minimal swelling was noticed extending till midline, the associated palatal mucosa was erythematous. On palpation the swelling was nontender, firm, not fluctuant, not mobile, compressible, and not reducible. An ulcer was also noted on the buccal alveolar swelling in respect to 18. The ulcer was irregular, 5 mm in its greatest extension, the floor was erythematous, margins were slopy, mildly tender and its base was indurated.

Generalized recession of gingiva was noticed. Teeth missing as a result of previous extraction were 11, 16, 17, 21, 23, 31, 32, 36, 37, 41, 42, 46. 18 was mobile and tilted mesially.

Considering a soft tissue alveolar swelling in a middle-aged women, with generalized recession of gingiva, missing and mobile teeth, a provisional diagnosis of aggressive periodontitis was given. Based on the history and clinical findings the differential diagnosis considered were necrotizing sialometaplasia, pleomorphic adenoma, mucoepidermoid carcinoma of salivary gland, adenoid cystic carcinoma, polymorphous low-grade adenocarcinoma, lymphoma and carcinoma of maxillary sinus.

Necrotizing sialometaplasia is a locally destructive inflammatory condition of salivary gland affecting palatal salivary glands most frequently. Although it may occur at any age, it affects adult males more commonly. It affects hard palate more frequently than soft palate. It presents initially as a nonulcerated swelling that converts into craterlike proliferative ulcer mimicking a malignant lesion.

Pleomorphic adenoma is the most common salivary gland tumor. Most pleomorphic adenoma present as a smooth, dome shaped, firm submucosal mass or nodule with growth rate that is usually indolent. It can occur at any age but is seen in middle aged adults with slight female predilection.

Carcinomas are known to occur in middle and elderly individual, causing rapid destruction of jaw bone and present as firm swelling associated with mobile teeth. Mucoepidermoid carcinoma is the most common malignant salivary gland neoplasm. Minor salivary glands of palate is the second-most common site of occurrence after the parotid gland. It usually presents as asymptomatic slow growing swelling that are sometimes fluctuant to firm with a blue or red color.

Adenoid cystic carcinoma occurs most frequently in the palate. It presents as a painful, slow growing mass affecting the surrounding structures in middle-aged adult individuals.

Polymorphous low-grade adenocarcinoma occurs most commonly in palate and usually presents as asymptomatic slow growing swelling. It usually affects older adults and has definite female predilection.

Extra nodal lymphoma may present in the palate as nontender, diffuse swelling with boggy consistency although buccal vestibule is the most common site.

Rarely carcinomas of maxillary sinus (mucoepidermoid carcinoma, adenoid cystic carcinoma, adenocarcinoma) may perforate the hard palate and present intraorally as diffuse swelling or as an ulcerated lesion.

Vitality test suggested that all the teeth were vital. Complete blood hemogram revealed all the values within the normal range.

The intraoral periapical radiograph taken in respect to maxillary right posterior region revealed radiolucent lesion around 18. The borders of the radiolucency was irregular and ill-defined, the trabecular pattern of alveolar bone was completely lost resembling moth eaten appearance with no cortication of bone. The lamina dura along 18 was absent along with widened periodontal ligament space giving a floating tooth appearance.

Panoramic radiograph revealed a massive unilateral osteolytic lesion on the right side completely destroying the posterior floor of maxillary sinus and tuberosity of maxilla, extending anteriorly till 14. The borders of the lesion were ill-defined and appeared invasive [Figure 2]. Waters view (paranasal sinus view) revealed completely obliterated right maxillary sinus. Lateral wall and the floor of the maxillary sinus were lost.

Computed tomography of right maxilla revealed an extensive contrast enhancing lesion in maxillary sinus with a soft tissue density (H.U. 74) and measuring 6 × 5.4 cm. Coronal and axial sections revealed a primarily osteolytic lesion destroying



Figure 2: Panoramic radiograph showing unilateral osteolytic lesion destroying tuberosity and posteroinferior wall of maxillary sinus on the right side

orbital floor superiorly, medial and lateral wall of maxillary sinus (medially and laterally) and hard palate inferiorly. However other sinuses, pterygoid plates and pterygoid muscles appeared normal [Figure 3].

Radiographic findings suggested that the lesion was extensive, aggressive, osteolytic with its epicenter within the maxilla. This led us to give a radiological diagnosis of primary intraosseous carcinoma of maxilla.

Incisional biopsy was performed intraorally [Figure 4]. Two bits of soft tissue measuring about $1.5 \times 1 \times 0.5$ and $1.5 \times 1 \times 1$ cm that were yellowish brown in color and soft to firm in consistency were taken as specimen for histopathological examination.

The routine H and E stained sections revealed highly cellular stroma arranged haphazardly, in sheets and nests at areas. Tumor cells were seen containing eosinophilic cytoplasm and round to oval hyperchromatic pleomorphic nuclei with plenty of mitotic figures. Upon examination in higher magnification,

a mixture of morphologically altered epidermoid cells, intermediate basaloid-like cells and mucous cells are observed of which epidermoid component is predominant [Figure 5]. The proliferation of epidermoid and intermediate components is in the form of sheets with some areas resembling squamous cell carcinoma. The lesional tissue show great variability in the composition of cells and many of them demonstrate extensive degree of cellular atypia. Production of mucin, formation of cyst-like areas is barely minimal. Areas that demonstrate clear appearing cells (vacuolated cytoplasm) and a minimal inflammatory infiltrate also characterize the lesional tissue. Adjacent skeletal muscle and tiny bony spicule was infiltrated by tumor cells [Figure 6]. The microscopic picture was suggestive of a high-grade mucoepidermoid carcinoma.

The radiographic evidence of complete destruction of maxillary sinus along with histological features of high-grade mucoepidermoid carcinoma made us to reach final diagnosis of high-grade mucoepidermoid carcinoma of maxillary sinus.

Considering that lesion was aggressively growing but not yet had involved other sinuses, muscles (pterygoid muscles)

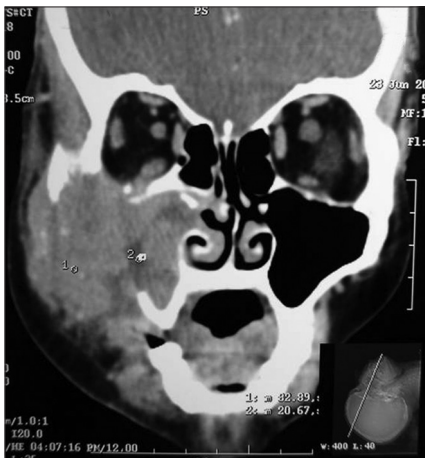


Figure 3: CT scan (coronal section) revealing contrast enhanced lesion in right maxilla involving, superior, medial, lateral, and inferior borders of maxilla

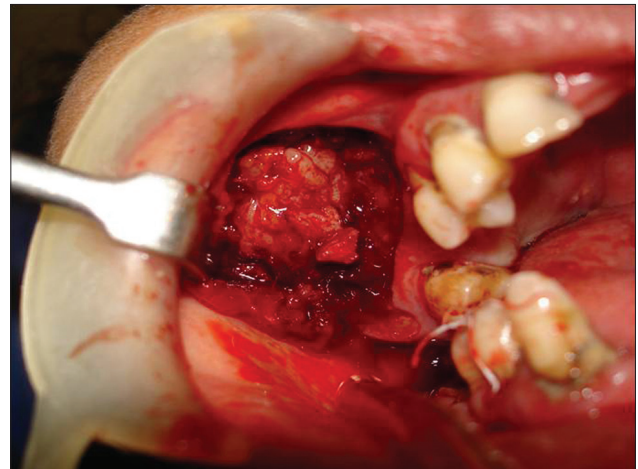


Figure 4: Incisional biopsy through intraoral approach

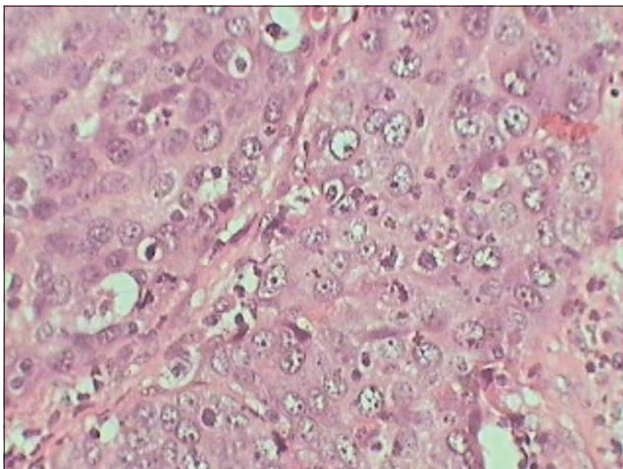


Figure 5: Showing predominantly epidermoid cell component (H and E staining, higher magnification, $\times 40$ view)

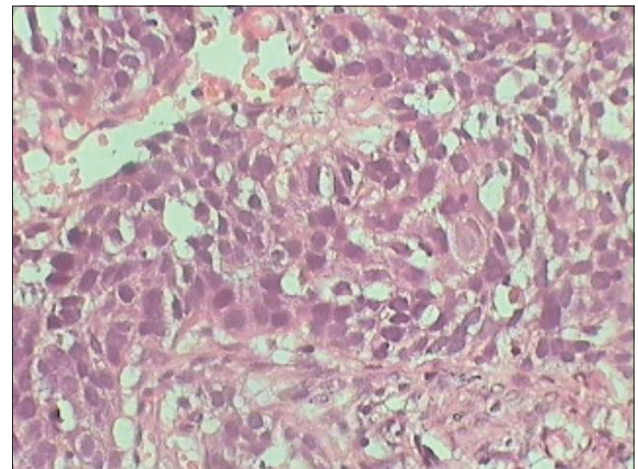


Figure 6: Demonstrating extensive degree of cellular atypia (H and E staining, $\times 40$ view)

and nerves a surgical approach was planned immediately. All the maxillary teeth were extracted. Hemimaxillectomy was performed under general anesthesia. Two weeks postoperatively a temporary obturator was given. After 6 months of follow up when no complication was encountered a permanent obturator with complete denture was given. Patient is under regular follow up. There is no signs and symptoms of recurrence of neoplasm from the past 1 year since the surgery.

DISCUSSION

Mucoepidermoid carcinoma is a malignant epithelial neoplasm composed of both mucus secreting cells and epidermoid-type cells in varying proportions. It was first studied and described as a separate entity by Stewart *et al.* in 1945.^[6] After a systematic review of its histology and degree of differentiation, the WHO classification in 1991 recommended that, the term “mucoepidermoid tumor” be changed to “mucoepidermoid carcinoma”.^[7] It comprises 5–10% of all salivary gland neoplasms and accounts for 13% of all malignancies occurring in maxillary sinus.^[1] Maxillary sinus non-SCCs mainly affect adults, with a male predilection. Although both ACCs and MECS affected adults, ACCs were more common in males and MECs in females.^[8]

MEC arising in maxillofacial region can have its origin from the maxillary sinus lining or central MEC arising from within the bone or from the minor salivary gland. In this case, the histological picture was suggestive of high-grade mucoepidermoid carcinoma but it was not conclusive about the origin of it. Considering the radiographic features of completely destroyed maxillary sinus and the fact that high-grade MEC are more commonly associated with maxillary sinus, we gave the final diagnosis of high-grade MEC of maxillary sinus. More over the central MEC and the one arising from minor salivary glands are usually of low-grade variety.^[9] Also central MEC are usually confined within the cortical plates and are more common in mandible.

Maxillary sinus malignancies are very difficult tumors to treat and traditionally have been associated with a poor prognosis. One reason for these poor outcomes is the close anatomic proximity of the nasal cavity and paranasal sinuses to vital structures such as the skull base, brain, orbit, and carotid artery.^[10] This complex location makes complete surgical resection of sinonasal tumors a challenging and sometimes impossible task. In addition, MEC of maxillary sinus tend to be asymptomatic at early stages, appearing

more frequently at late stages once extensive local invasion has occurred. The unfortunate combination of complex surrounding anatomy with late, advanced stage presentation therefore leads to the frequent local recurrence and subsequent poor outcome associated with sinonasal malignancies. Overall 5-year survival for maxillary sinus MEC is 36%.^[8] The early diagnosis is critical for better prognosis of this tumor. So also it suggests that MEC of maxillary sinus should be considered in the differential diagnosis of swellings in maxilla.

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