



CASE REPORT

Maxillary sinus squamous cell carcinoma during pregnancy: A new case report

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ABSTRACT

Less than one percent of malignant lesions are malignant tumors of the paranasal sinuses. The majority of maxillary sinus carcinomas are squamous cell carcinomas (SCC). There are few reports of head and neck SCC found during pregnancy; however, in this article, a 23-year-old woman in the fourth month of pregnancy was diagnosed with a maxillary sinus SCC. The histopathologic evaluation of the lesion showed a malignant proliferation of squamous cells with a sheath and cord formation that invaded the surrounding tissue. These findings were compatible with SCC. Hemimaxillectomy, neck dissection, and chemoradiotherapy were conducted; however, despite these therapeutic interventions, the patient died 11 months after the initial visit due to distant metastasis.

INTRODUCTION

Less than one percent of all malignancies are malignant paranasal sinuses. Maxillary sinus carcinomas are only responsible for 3–3.9% of head and neck carcinomas, but they are most prevalent in the paranasal air sinuses. Maxillary sinus carcinomas include: squamous cell carcinomas, sinonasal adenocarcinomas, undifferentiated sinonasal carcinomas, neuroendocrine carcinomas (undifferentiated small cell), and salivary gland adenocarcinomas.¹⁻⁴ Most of the lesions are asymptomatic, and they are typically found when they reach large dimensions and are in an advanced stage.^{1,3,5} Cancer is the second cause of death in reproductive-aged females.⁶ However, there are few reports of SCC in pregnant female patients, but incidence of head and neck carcinoma during pregnancy is increasing. One of the reasons for this increase is increased marriage age.⁷ The treatments for these patients include: surgery, radiotherapy, or a combination of both surgery and radiotherapy. The best treatment is not been identified. The survival rate is poor and is based on the cancer extension with a five-year survival rate of patients with advanced head and neck SCC at just under 40%.^{8,9} Salvage surgery for recurrent lesions has been known to increase this rate.

CASE REPORT

A 23-year-old female in her fourth month of pregnancy with swelling in right maxillary region was treated at the clinic. The swelling began in the first month of the patient's pregnancy, and the woman received a provisional diagnosis of pulpoperiapical lesions. Empirical antibiotic therapy was provided by her dentist. She was referred to the Mashhad Dental School due to an increase in the size of the lesion (Figure 1a).

Right ear otalgia and epistaxis as well as pain at the lesion site were the woman's primary complaints. During the intraoral

examination, a rubbery swelling that extended from the maxillary right first molar to the right side tuberosity was apparent (Figure 1b).

A Panoramic view using a Planmeca ProMax (Planmeca Oy, Helsinki, Finland) showed an ill-defined radiolucent lesion. The lamina dura of the involved teeth had disappeared, and root resorption had occurred in teeth numbers 14, 15, and 16. The involved teeth had lost their bone support and appeared to be "hanging in the air" (Figure 2). Based on the clinical examination and the orthopantomogram findings, the differential diagnoses included: 1) osteogenic sarcoma, 2) Ewing sarcoma, and 3) non-Hodgkin lymphoma.

To assess the extent of the maxillary lesion, a Siemens SOMATOM Sensation 16 multi-slice spiral CT scan was conducted.



Figure 1. Photograph of the patient's facial asymmetry (a). A soft, exophytic, tumoral mass with a palatal expansion was observed in the right maxillary region during the intraoral examination (b)



Figure 2. During the orthopantomograph of the patient before surgery, an ill-defined radiolucency in the right maxillary region with root resorption and a "hanging-in-the-air" appearance were observed

The CT showed that the right maxillary sinus was completely occupied by the lesion. There was significant destruction of the anterior, medial, and posterolateral walls of the maxillary sinus with infiltration into the soft tissue mass and surrounding soft tissues, including the right posterior

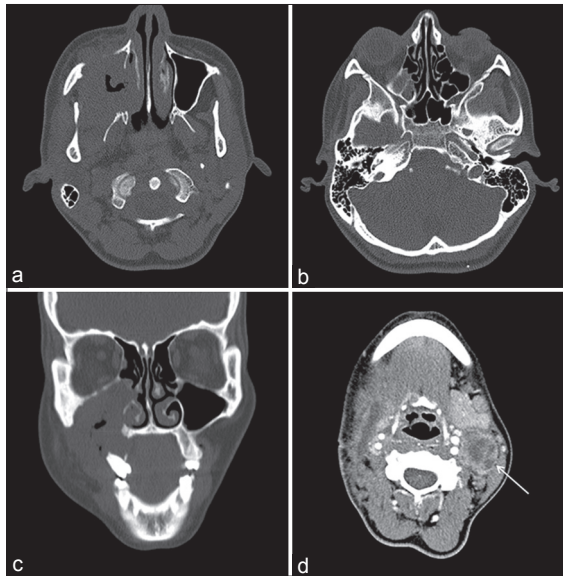


Figure 3. An axial CT section at the odontoid process of the C2 vertebra level showed a destructive soft tissue mass occupying the right maxillary sinus. There is significant destruction of the anterior, medial, and posterolateral walls of the maxillary sinus with infiltration into the soft tissue mass in the surrounding soft tissues, including the right infratemporal and pterygopalatine spaces (a). An axial CT at the lacerum foramina section shows involvement of the right posterior ethmoidal air cell, and the most superior aspect of the right maxillary sinus was observed (b). The coronal CT shows that the mass has eroded a portion of the right orbital floor. The orbital contents appear normal, but the premolar tooth appears to be hanging in the soft tissue mass with no bone surrounding it (c). An axial contrast-enhanced CT at the hyoid bone level shows a metastatic lymph node (white arrow) with central necrosis and peripheral enhancement (d)

ethmoidal air cells and the infratemporal and pterygopalatine spaces (Figure 3a, b). The right middle concha and uncinate processes were also destroyed. Finally, right orbital floor erosion was present (Figure 3c). During the contrast-enhanced CT, a contralateral metastatic lymph node (29.9×26.5 mm) with central necrosis and peripheral enhancement was also found (Figure 3d).

To obtain a definitive diagnosis, an incisional biopsy of the lesion was conducted. Malignant squamous cells with a sheath and cord formation and lesion expansion into the surrounding tissues were apparent. Hyperchromatism, pleomorphism, nuclear atypia, and increased atypical mitosis were present in the squamous cells. Keratin pearls and individual cell keratinisation were present. Cystic degenerative changes were identified in the intraepithelial islands. Connective tissue composed of collagen fibres and fibroblasts with infiltration of lymphoplasmacytes and cross sections of the blood vessels was seen. The surface of the lesion was covered with parakeratinised squamous epithelium. The histopathologic diagnosis was compatible with grade III SCC (Figure 4 a, b).

The patient underwent hemimaxillectomy, and the entire specimen was re-evaluated. The

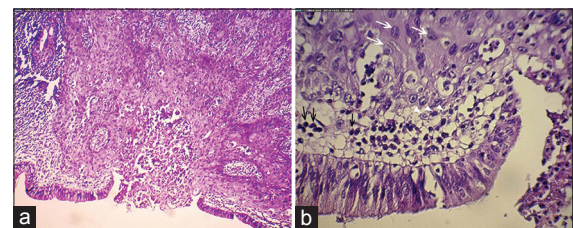


Figure 4. Photomicrograph of malignant squamous cells' invasion into the surrounding tissues (H&E, ×100) (a). Higher magnification of the same site. Hyperchromatism (black arrows), pleomorphism (white arrows), and nuclear atypia (white arrowheads) are present in the squamous cells. (H&E, ×400) (b)

delivery of the child occurred at 37 weeks of pregnancy via a cesarean operation. Despite the bilateral neck dissection that occurred before childbirth and the chemoradiotherapy administered after birth, patient death occurred 11 months after the surgery due to brain metastasis.

DISCUSSION

Paranasal air sinuses neoplasms are rare, and only 0.2% are malignant. The risk of head and neck carcinoma in males is three times the risk for females.⁹⁻¹¹ The mean age of the involvement is 64 years.¹² The occurrence of an SCC during pregnancy is rare, but the number of occurrences is increasing due to increasing marriage age and the increased incidence of SCC in females.² However, the current patient was a 23-year-old female without a history of cigarette smoking or alcohol intake.

Eliassen *et al.*¹³ in a review of 4 SCC cases showed that they were negative for estrogen and progesterone receptors, and human papillomavirus (HPV). Poor survival might be the result of a delay in diagnosis, and p53 overexpression. Also, Smoking and alcohol use were not constant etiologic factors in the development of these tumors. Cudney *et al.*¹⁴ suggested that biomarkers were unremarkable in progression of the benign odontogenic tumor to SCC. However, our patient had no biomarkers expression result and Immunostaining due to low income.

In pregnant patients, the management is complicated. The treatment of various diseases in pregnancy may be difficult due to immunosuppressive, metabolic and hormonal activities. Inappropriate treatment would be hazardous to both mother and fetus. The physician should be alert to “the needs”, “the risks” and “the choices”.¹³

The most prevalent symptoms of maxillary sinus SCC are: pain (59%),

intraoral presentations (40%), facial swelling (38%), nasal blockage (35%), and epistaxis (25%).¹² In the reported case, pain, intraoral swelling, nasal blockage, and spontaneous epistaxis were present.

Osteogenic sarcoma (OS) was considered first in the differential diagnosis list due to pain and swelling. Epistaxis, the age of the patient, the pattern of the bone destruction, and root resorption confirmed this diagnosis, but OS is more prevalent in the mandible and in individuals in their 40s.¹⁵

A secondary diagnosis of Ewing sarcoma (ES) was also considered. This diagnosis was appropriate due to the patient’s age and the radiolucent bone destruction. However, the prevalence of ES is twofold in the mandible than maxilla.^{15,16}

Non-Hodgkin lymphoma (NHL) was the third consideration in the differential diagnosis list. Although the lymph nodes are the most prevalent site for NHL, the jaws can also be affected. The maxilla affects more than the mandible.^{17,18} Symptoms such as pain, ulcer, and tooth mobility are present in lymphomas that involve the maxilla, but the age of the patient did not fit the mean age of NHL patients. Night sweating¹⁹ and pain-free swelling, which are very common in NHL, were also not present in this case.

The image findings of this case were supported by other studies. In the majority of the patients, grade III/IV maxillary SCC was not diagnosed before the adjacent tissues were involved, and in the present case, the destruction of the maxillary sinus walls and the invasion into the nose and floor of the orbit were identified.^{20,21} In addition, metastatic lymph nodes were present in 4–20% of all reported cases,^{12,22} and contralateral neck lymph node involvement was also present in our case.

Hypermobility of the adjacent teeth should encourage the dentist to do a more

precise examination in absence of any signs and symptoms of a periodontal condition. Radiographic examination is the first step to localize the extent of jaw pathological damage. Presence of bone destruction with adjacent soft tissue exophytic mass is highly suggestive of malignancy.¹⁵ The optimal treatment for maxillary sinus SCC was not definitively identified. Treatment modalities for maxillary sinus SCC include: surgery, radiotherapy, and systemic chemotherapy.²³ Maxillary sinus malignancies have a poor prognosis, and the five-year survival rate is 43% and the overall survival is 52 months. High stage, local, and distant metastasis are indicators of a poor prognosis.^{12,24,25} The current case was at the T3N2cM1 stage,²⁶ and despite wide surgical resection and chemoradiotherapy, death occurred.

In conclusion, to increase the survival rate, decreasing morbidity, an early diagnosis of maxillary antral SCC is vital.

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