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Pregnancy Complicated by Mandibular Osteosarcoma: A Case Report and Review of Maternal-Fetal Outcomes

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Abstract

The development of cancer during pregnancy is an uncommon occurrence. While breast and cervical cancers are more commonly reported in pregnant individuals, malignancies affecting the head and neck region—such as osteosarcoma—are particularly rare. Osteosarcoma of the jaw is exceptionally unusual during pregnancy and presents significant diagnostic difficulties. When diagnosed during gestation, this condition can adversely affect both maternal and fetal outcomes. This case report presents a 29-year-old African woman at 28 weeks of gestation who developed osteosarcoma of the jaw. She initially presented with a painful facial swelling that had persisted for approximately three months, following a toothache in the posterior region of the upper left jaw. Despite undergoing multiple surgical interventions, the tumor recurred. She was then treated with chemotherapy, but the response was poor, and she eventually passed away due to disease progression. This report highlights the diagnostic, therapeutic, and prognostic challenges of managing jaw osteosarcoma during pregnancy.

Keywords: Maxilla, Osteosarcoma, Pregnancy, Maternal outcome, Fetal outcome

Introduction

Cancer occurring during pregnancy is highly uncommon [1, 2]. Among the types that do appear, breast and cervical cancers are the most frequently reported, followed by blood-related malignancies [1, 3]. On the other hand, cancers of the head and neck are especially rare during gestation [3, 4]. Within this category, squamous cell carcinoma is the most prevalent subtype [5, 6], but there are a handful of documented instances where pregnant women develop osteosarcoma in the jaw [3, 6, 7]. Typically, osteosarcoma affects long bones near the growth plates of the limbs [7, 8], and its presence in the jaw is extremely rare—making up about 1% of all primary tumors in the head and neck area and roughly 4% to 10% of all sarcomas [9, 10].

When malignancies of the head and neck are identified during pregnancy, both maternal and fetal health may be jeopardized [11, 12]. Despite this, clear treatment protocols are lacking due to the infrequency of such cases [13, 14]. As a result, it becomes essential to document and share these rare cases to enhance clinical knowledge. This report details the presentation, treatment, and outcome of a pregnant woman diagnosed with osteosarcoma in the upper jaw.

Case report

A 29-year-old African woman, 28 weeks into her pregnancy, was referred to the Oral and Maxillofacial

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Surgery Department in July 2022. She had been experiencing a painful swelling on her face for about three months, which initially began as a toothache in the back of her left upper jaw. Within a week, a painful lump developed in the same region and gradually enlarged over time.

She first consulted an oral and maxillofacial surgeon, who initially suspected a central giant cell tumor of the maxilla. A biopsy was performed, and histology indicated it was an odontogenic fibroma. Based on this, the tumor was surgically removed. However, only eight days later, the swelling returned—again painful and progressively increasing in size. As it worsened, she experienced nasal blockage, teeth loosening, and difficulty chewing, prompting her referral to our center for further evaluation.

Upon examination, a large swelling was seen on the left side of her face, stretching from the upper edge of the eye socket down to the lower jaw. The overlying skin appeared reddish, and the mass caused her left eye to shift upward and her nose to deviate to the right. The swelling was warm, firm to hard, immobile, and painful on touch **(Figure 1a)**. Inside the mouth, the mass occupied the left side of her upper jaw and nearly reached the midline of the palate. The lesion was lobulated with areas of keratinization, granulation, and ulceration, and its surface ranged from smooth to rough. The mucosal color was a reddish-purple.

The patient underwent a full diagnostic workup including a histopathology exam, craniofacial CT scan, chest X-ray, complete blood count, and tests for liver, kidney, and electrolyte function. Imaging revealed a large, irregular mass in the upper left jaw invading nearby spaces such as the buccal region, nasal cavity, and sinuses **(Figures 2a–2c)**. Apart from a hemoglobin level of 10.1 g/dL, all other test results were within normal limits. The biopsy confirmed the diagnosis of osteosarcoma **(Figure 3a)**.

A team of maxillofacial surgeons reviewed the case and decided that surgery followed by chemo-radiotherapy would be the best course. The patient and her family were thoroughly informed and counseled on treatment options. A multidisciplinary team was assembled, including anesthesiologists, obstetricians, pediatricians, and ENT specialists. The obstetric team determined that the pregnancy had reached 29 weeks and 5 days. It was decided to perform a cesarean section on the same day as the tumor surgery. Anesthesia evaluation was satisfactory, though a tracheostomy was planned due to potential airway difficulties.

With informed consent, the patient underwent a tracheostomy, cesarean section, and wide tumor resection in a single surgical session. The procedures were completed without complications. A healthy premature baby boy was delivered, and the tumor was removed entirely.

Following surgery, the mother was monitored in the ICU for five days, while the newborn was admitted to the NICU. She then spent an additional week in the hospital for post-operative care **(Figure 1b)**. Final histopathology results confirmed the diagnosis of osteosarcoma **(Figure 3b)**. By the third week post-surgery, the case was presented to a tumor board and the patient was referred to a cancer treatment center for follow-up chemotherapy. Before chemotherapy could effectively begin, the tumor reappeared beneath the left lower eyelid and continued to enlarge despite the initiation of treatment. Over time, the mass not only expanded rapidly but also ulcerated and began to bleed.

Following the third round of chemotherapy, the oncologists referred the patient back to our department for reevaluation and to determine whether further surgical intervention was feasible **(Figure 1c)**. Upon review, the team of maxillofacial surgeons concluded that the tumor was no longer operable. As a result, they recommended palliative care alongside the completion of the remaining three chemotherapy cycles. The palliative approach focused on controlling pain and providing daily wound care.

Tragically, the patient passed away in December 2022 due to disease progression. On a more hopeful note, her newborn son has grown into a healthy and thriving child, recently celebrating his first birthday.

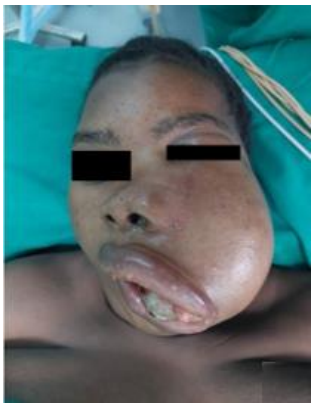

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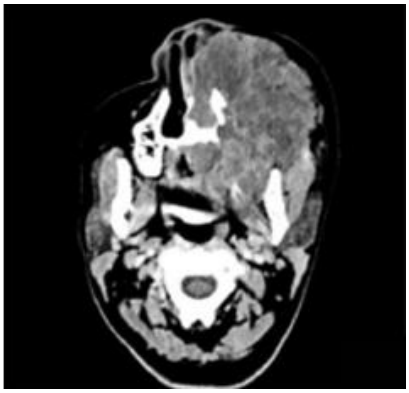
Figure 1. Clinical appearance of the patient; (a) a massive tumor involving the left side of the face, displacing the left eye superiorly and the nose to the right, (b) the post-operative appearance of the patient following wide tumor resection, and (c) clinical presentation of the patient after the tumor recurred; the tumor is shown to have ulcerated and caused significant deformity



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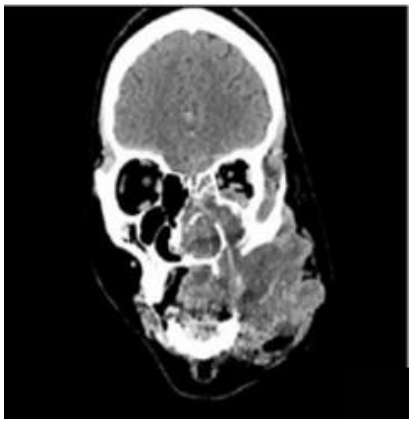

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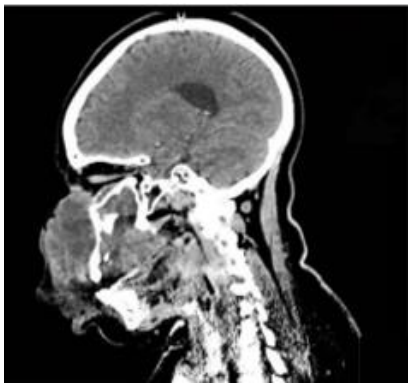
Figure 2. Selected computed tomography (CT) scan of the maxillary lesion (a) sagittal view, (b) axial view, and (c) coronal view; the CT scan demonstrates a heterogeneous soft tissue mass, originating from the left maxilla, involving the left paranasal sinuses and the orbital floor destroying the facial bones.



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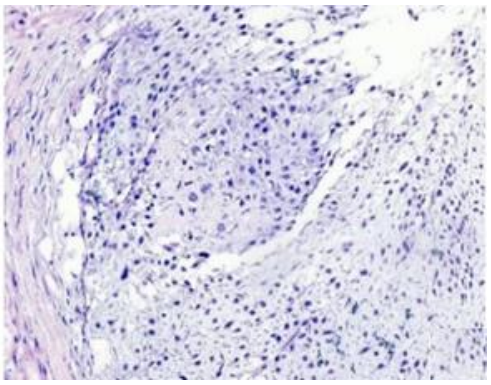

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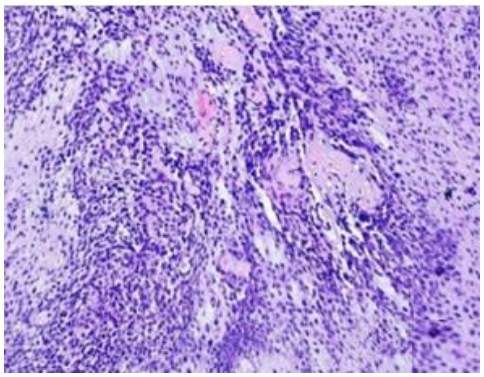

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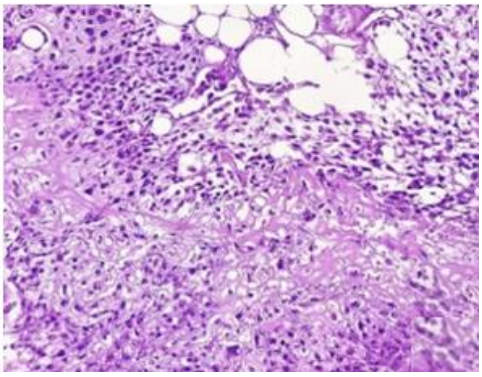

c)

Figure 3. Histopathological images (H&E) of the maxillary lesion: (a) Pre-operative biopsy, showing a poorly circumscribed tumor with pleomorphic spindle cells and irregular osteoid formation (magnification x100), (b) Post-operative, tumor composed of pleomorphic spindled cells with irregular disorganized osteoid formation (magnification x100), and (c) A post-operative, tumor composed of pleomorphic spindled cells, large prominent nuclei, disorganized osteoid formation, and abnormal mitoses (magnification x200).

Results and Discussion

Osteosarcoma tends to affect the mandible more frequently than the maxilla, with a reported ratio of approximately 2:1 [7, 15]. Its development in the jaw during pregnancy is exceptionally rare, with fewer than 20 cases documented in English medical literature [16]. To the best of our knowledge, this particular case stands out due to both its occurrence in a pregnant woman and its unusual location in the maxilla.

As demonstrated in this report, diagnosing osteosarcoma during pregnancy poses considerable challenges—not only because of its rarity but also due to other complicating factors. In the early stages, the tumor's inflammatory features, combined with the limitations of radiographic evaluation during pregnancy, make it difficult to distinguish from more common dental or periodontal infections. This often results in misdiagnosis [3]. Furthermore, the symptoms of jaw osteosarcoma—such as facial swelling, tooth pain, mucosal ulceration, nasal blockage, and displacement of the eye—are common to many other maxillary lesions [15, 17], as seen in the current case, which further complicated early diagnosis.

In this patient, tumor recurrence and rapid regrowth were observed shortly after surgical removal, potentially influenced by the physiological state of pregnancy. Some researchers suggest that the proangiogenic environment of pregnancy, along with hormonal and immune system changes, may accelerate tumor development [18]. For instance, a study by Dohi *et al.* [19] found positive expression of estrogen receptor beta (ER-β) and progesterone receptor (PR) in most osteosarcoma tissue samples. However, this finding remains controversial, as Domínguez-Malagón *et al.* [20] reported negative expression of ER and PR in the majority (95% and 100%, respectively) of jaw osteosarcoma samples they examined.

The primary treatment for osteosarcoma is wide surgical resection, though some experts recommend combining surgery with chemotherapy and radiotherapy [3]. Due to the absence of established protocols for treating osteosarcoma during pregnancy, our multidisciplinary surgical team elected to perform a wide excision of the tumor, followed by adjuvant chemo-radiotherapy. This decision was guided by evidence that pregnant patients can undergo non-obstetric surgeries under general anesthesia without significant risk [21].

Deciding whether to continue the pregnancy to full term depends largely on the prognosis of the tumor and expected maternal survival [18]. In situations where adjuvant therapy is urgently required after surgery, preterm delivery via cesarean section may be considered [21]. In this case, early delivery was medically induced to enable timely cancer treatment. The preterm birth was prompted by the critical timing of the diagnosis, limited knowledge about the safety of standard sarcoma treatments during pregnancy [22], and the elevated risk associated with chemotherapy and radiation. Chemotherapy in the second or third trimester has been linked to complications such as growth restriction, infertility, and premature labor [6], while radiation poses

serious threats, including fetal death, developmental delays, cancer, and genetic abnormalities [6].

In contrast to previous reports that documented favorable outcomes for both mother and child after treatment of jaw osteosarcoma during pregnancy [7, 23, 24], our case unfortunately had a poor outcome. The tumor was unresponsive to chemotherapy, and the patient passed away six months after diagnosis. Nevertheless, the decision to deliver the baby early had a positive impact, as the child continues to grow into a strong and healthy infant.

Conclusion

Osteosarcoma of the jaw during pregnancy presents a serious threat to both maternal and fetal health. Effective management requires a coordinated, multidisciplinary approach centered on the patient's needs. Open communication and detailed counseling with the patient and her family are essential, given the diagnostic and therapeutic challenges. Choosing the most appropriate treatment plan—and whether or not to continue the pregnancy—must be carefully weighed by considering the risks and benefits for both the mother and the unborn child.

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