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Case Report

Osteosarcoma of the ethmoid sinus: About a case ☆

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ABSTRACT

Osteosarcomas of the craniofacial bones account for less than 10% of all osteosarcomas. Primary osteosarcomas of the nasal cavity and paranasal sinus are rare localization (0.5%-8.1% of osteosarcomas occur in this site). Accordingly, we report a case of osteosarcoma arising de novo from the ethmoid bone in a 46-year-old female. Initially, she presented with headache, bilateral epistaxis, and postnasal drip. Biopsy revealed an osteosarcoma ethmoidal. The patient was treated by a neoadjuvant chemotherapy followed by surgical resection and radiotherapy.

Introduction

Osteosarcoma is a highly malignant tumor, rarely found in the craniofacial region; it represents less than 0.5% of all malignant tumors in this region [1]. The predilected location of this osteosarcoma is in the mandible or maxillarybone, and just few cases have been reported in the other facial bones [2]. Hereby, the ethmoidal sinus is an exceptional primary site of osteosarcoma [3].

Case report

Clinical presentation

A 46-year-old patient with no specific medical history, consulted for headache, bilateral epistaxis, and postnasal drip, evolving for 4 months in a context of preserved general condition.

On clinical examination, we found swelling of the nasal region, with no cervical adenopathy neither other associated signs.

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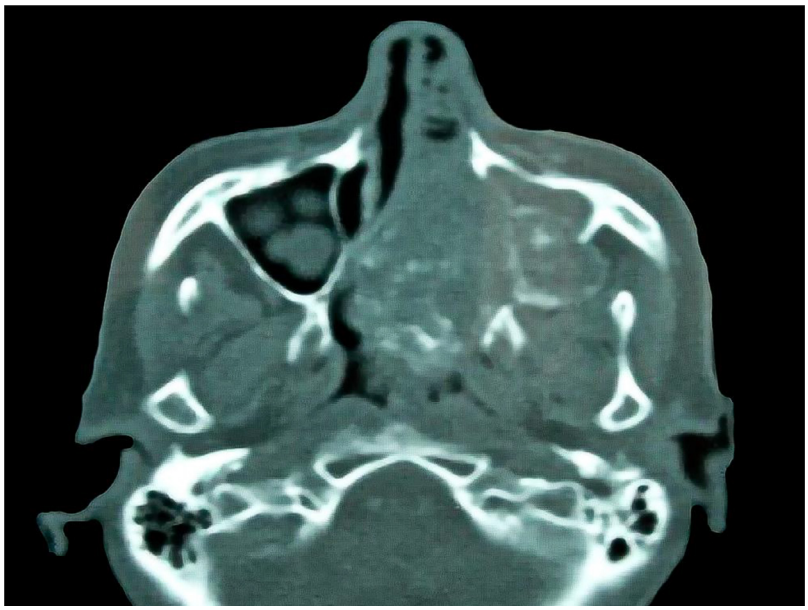

Fig. 1 – Computed tomography revealed a heterogeneous soft tissue mass, centered on the posterior ethmoid and sphenoid body, well-bounded, many calcifications within, measuring 8 x 4 x 4 cm and causing an extensive osteolysis of posterior ethmoidal cells and the sphenoidal bone body.

Diagnosis

Computed tomography (CT) revealed an heterogeneous soft tissue mass, centered on the posterior ethmoid and sphenoid body, well-bounded, many calcifications within, measuring 8 × 4 × 4 cm and causing an extensive osteolysis of posterior ethmoidal cells and the sphenoidal bone body (Fig. 1).

Biopsy showed a tumoral proliferation with many medium-sized cells and granular cytoplasm, poorly limited, and occupied by a hyperchromatic vesicular nucleus. These cells formed dense monomorphic sheets surrounding foci appearing as a characteristic osteoid, with some images of cellular incarceration. These elements suggested the diagnosis of an osteosarcoma (Fig. 2).

Our patient has no distant metastases, verified by a CT examination.

Therapeutic intervention

We first, began with chemotherapy, followed by a tumor resection which was not complete due to the local extension of the tumor, then radiotherapy with a good tolerance.

Follow-up of the patient

After a 12-month follow-up, tumor stability was noted (partial remission without distant metastasis).

Discussion

Osteosarcomas (OS) are the most common primary malignancies of the long bones of the extremities. However, in the head and neck region, they report a prevalence of between 0.5% and 8.1% [1,2]. Primary sites for the OS in the head neck region are bones of the jaw, and the ethmoid sinus is an unusual site for craniofacial [2,3].

These tumors usually occur in the third or fourth decade of life, later than OS occurring in the long bones, and usually arise as secondary tumors after radiation therapy, thorium oxide exposure, chemotherapy, hereditary predispositions to osteosarcoma development, or from pre-existing benign bone disease such as Paget's disease, bone infarcts, osteomyelitis, or trauma [1,2,4]. No gender predominance has been described, but our patient had none of these risk factors and was classified as a primary osteosarcoma.

The exact diagnosis of craniofacial OS depends on the histopathological examination. However, magnetic resonance imaging and CT are very useful diagnostic tools. These imaging modalities can also be used to detect metastatic disease. Bone sclerosis and tumor calcification are the most common imaging features on CT and magnetic resonance imaging, and these findings define tumor extension into adjacent soft tissues, such as intracranial structures or the orbit.

The risk of distant metastasis of long bone osteosarcoma is much higher than with craniofacial OS, but in the literature, metastases to the lungs, other bones, and lymph nodes have been reported for craniofacial OS [5]. However, the main challenge for this tumor is the higher rate of tumor extension to neighboring bones and anatomical structures such as the nasopharynx, orbit, and skull [5,6].

The mainstay of treatment for ethmoidal osteosarcoma is surgical resection of the tumor with negative surgical margins, but neoadjuvant or adjuvant chemotherapy and radiation therapy are the other treatment modalities that can play an important role in locoregional control. The main challenge in the surgical treatment of craniofacial OS is to achieve free

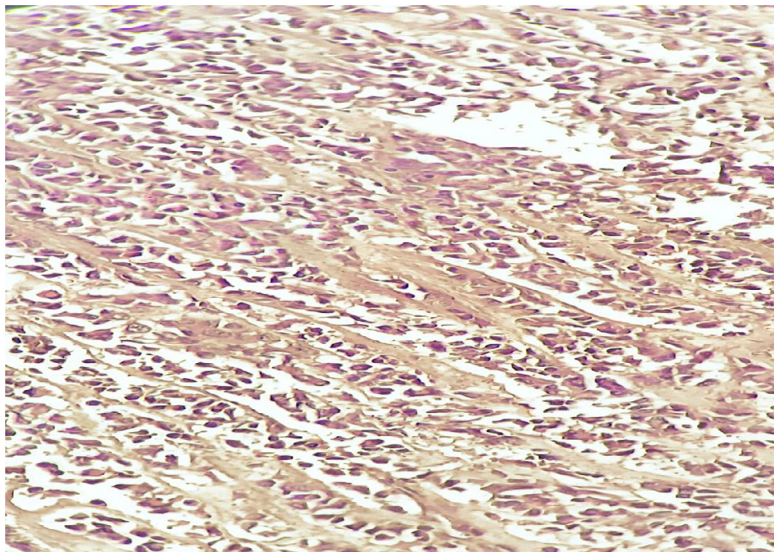

Fig. 2 – Biopsy showed a tumoral proliferation suggested the diagnosis of an osteosarcoma.

surgical margins, which appears to be the only significant predictors of overall and disease-specific survival [6,7].

Chemotherapy is useful in controlling local recurrence and metastasis. The use of adjuvant chemotherapy after primary surgery has been shown to significantly improve 5-year survival rates to approximately 80% [8].

Head and neck OS have a poor prognosis and high rate of postoperative recurrence. However, radiotherapy can potentially improve overall survival. Radiotherapy is more frequently used in the management, especially in cases of partial resections. It can be provided as an adjunctive modality and can be administered before or after surgery, thereby increasing the locoregional control rate and decreasing the rate of metastasis [9,10].

Our patient received chemotherapy followed by tumor resection which was not complete due to the extent of the tumor, followed by radiotherapy with good tolerance and tumor stability (partial remission without distant metastasis after 1 year).

Conclusion

Osteosarcoma of the ethmoidal sinus is an exceptional and aggressive tumor. Multidisciplinary management is very important in all diagnostic, therapeutic and surveillance stages. Surgical excision remains the basic treatment and is a very important factor in the therapeutic success.

Chemotherapy and radiotherapy are also treatments that can improve the prognosis through locoregional and distant control.

Author involvement

All authors were involved in the completion of this work.

Patient consent

The patient was informed of the procedures for publication of this clinical case and gave clear consent.

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