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Mandibular Osteosarcoma: Report of Two Cases and Review of Literature

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Abstract **Case Report**

Osteosarcoma is a conjunctive tumor characterized by the elaboration of bony tissue of a malignant aspect. It's a rare malignant primary tumor of the bone. The localization in the jows is different by its clinical and histopathological features. The evolution is characterized by its gravity due to its aggressive potential, late diagnosis and difficulties met to choose the ideal therapeutic project. Clinically, the symptomatology of this tumor is similar to other bone malignant tumors. Radiologically, it is usually responsable of lytic lesion of the bone, giving sometimes a pathognomonic "sunburst" appearance. The histological diagnosis isn't always evident. The treatment is essentially surgical, usually associated to a neoadjuvent and adjuvent chemotherapy. Even though medical progress, the prognosis of these tumors still bad and depends on a wide variety of factors, especially the lateness diagnosis and inappropriate treatment. The main aim of this case report was to focus on the importance of early diagnosis of this tumor based on clinical and radiographic examinations, and confirmation by histopathology. Considering the rarity of the disease type and particularly considering the fast progression and aggressiveness of this neoplasm.

Keywords: Osteosarcoma, Jaw, Mandible, Adults, Chemotherapy, Surgery.

INTRODUCTION

Osteosarcomas (OS), is the most common primary malignant tumor of bone. Osteogenic sarcoma of the jaw is a rare neoplasm, which represents about 7% of all OS and 1% of all malignancies in the head and neck region [1].

Osteosarcoma of the jaw (OSJ) tends to occur in the mandible twice as often as in the maxilla [2]. No gender difference for the incidence has been reported [3], and it occurs in older age and shows good prognosis when compared with that of tumors in the long bones [3].

Osteosarcoma of the jawbone is different by its clinical and histological aspects, late diagnosis and difficulties met to choose the ideal therapeutic project. The treatment is essentially surgical, usually associated to a neoadjuvent and adjuvent chemotherapy [10].

The main aim of this case report was to focus on the importance of early diagnosis of this tumor based on clinical, radiological and histopathological examinations.

OBSERVATION N°1

A 49-year-old women in good general health condition, without any known health problem, reported with facial asymmetry and painful swelling in the mandible and pain in the left mandibular molar region evolving for five months.

On extraoral examination, a diffuse swelling which caused asymmetry of the face (fig.1a). There was paresthaesia of the left side of the lower lip, which was clinically confirmed. The left submandibular lymphnodes were palpable, mobile and slightly tender, measuring 1cm for the largest.

The intra-oral examination revealed an ulcerative and budding lesion, in the lower left molar-premolar region (fig 1b). The size of the lesion was 4x1cm. On palpation, the mass is hard, fixed in relation to the deep plane, filling the mandibular vestibule with difficulty protrusion of the tongue.

Fig. 1a: Extra oral photos showing facial asymmetry by the mandibular mass

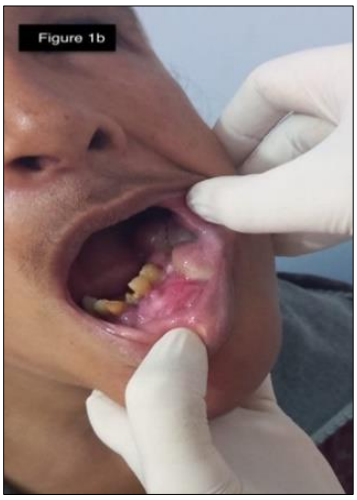

Fig. 1b: intraoral photos showing an ulcerative- boring lesion of the left anterior mandible region.

A panoramic X-ray was taken, showing the presence of a mixed image with poorly defined limits, located in the anterior mandible area, on the left side. (fig.2)

In dentascan maxilla showed a parasymphyseal expansive process to the left side of the mandible, with irregular contours, measuring 29*18mm extending from the region of 31 to 34, slightly hypodense, with bone lysis of the lingual and vestibular cortex and extension to the soft tissues next to it. The lesion infiltrates the lower alveolar arch and encompasses the dental roots of 32,33 and 34 with partial root resorption of 32. It comes into contact with the mental foramen through place without lysis of its bone walls. (fig.3;4)

Fig. 2: Panoramic image showing a mixed aspect area of irregular and undefined edges in the mandibular anterior region

Fig. 3 :Tomographic image detailing the areas affected by injury

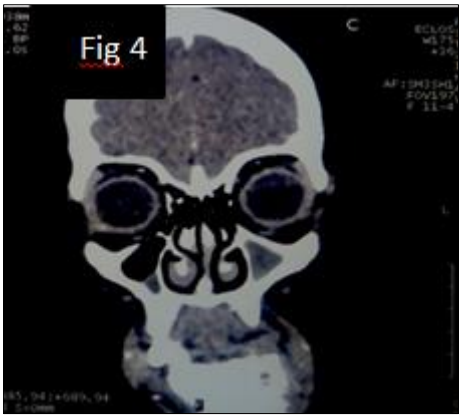

Fig. 4: A coronal CT scan acquired before neoadjuvant chemotherapy showing the lesion on the left mandible

Incisional biopsy was then performed. Histologically, the tumor showed largely myxoid mesenchymal tumor proliferation with tumor osteogenesis compatible with high-grade osteosarcoma (Myxoid).

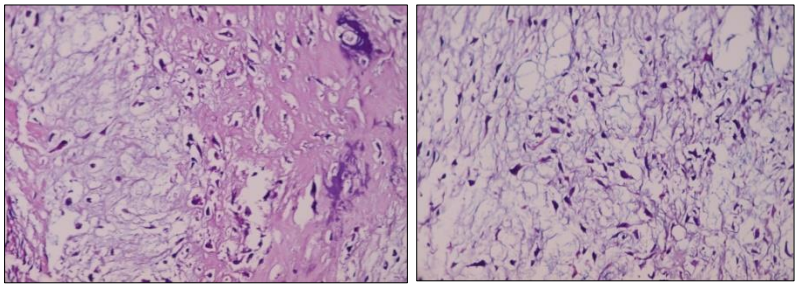

Fig. 5: Photomicrograph showing dysplastic ossifications and lacunae filled with osteoblasts showing hyperchromatic nuclei.

The extension assessment based on thoracic and abdominal CT scanner has not shown any distant metastasis.

Due to the advanced stage, surgical treatment was not conceivable. The patient was treated with neoadjuvant chemotherapy (API-AI) based on Doxorubicin, Cisplatin, Ifosphamide.

The patient received the first course of chemotherapy with good tolerance.After the end of chemotherapy, depending on the clinical response, surgery or radiotherapy is planned.

Observation n°2

Our second case is about a 17 years old boy with no significant past medical history consulted us for the appearance of a painful right gingivo-mandibular swelling, which rapidly increased in volume in June 2020, without dental mobility or dental loss.

His physical examination showed a facial asymmetry, a bulky gingivo-mandibular mass,

occupying the entire oral cavity and externalized by the mouth, associated with a right jugulo-carotid enlarged lymph node of 1.5 cm wide.

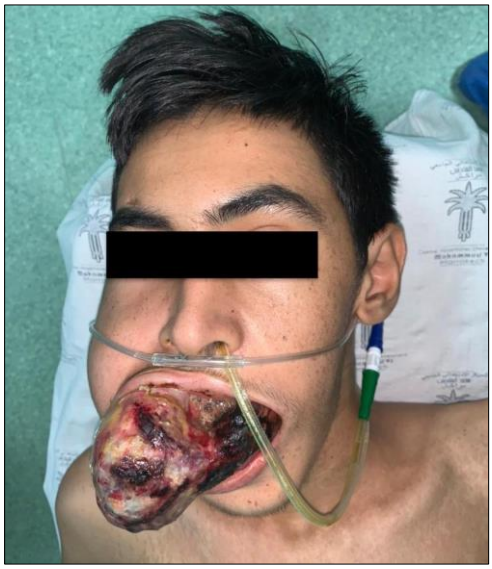

Fig. 6: Bulky gingivo-mandibular tumoral process

Panoramic X-ray showed an osteolytic lesion of the ascending ramus of the right mandible.

Facial CT showed a heterogeneous lytic lesion of the ascending branch of the right mandible with spiculated periosteal reaction and cortical rupture.

Significant infiltration of the soft tissues, particularly the masticatory space, with repression of the parotid.

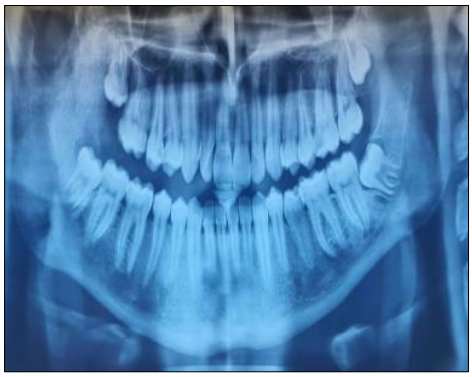

Fig. 7: Panoramic image showing an osteolytic lesion of the ascending ramus of the right mandible.

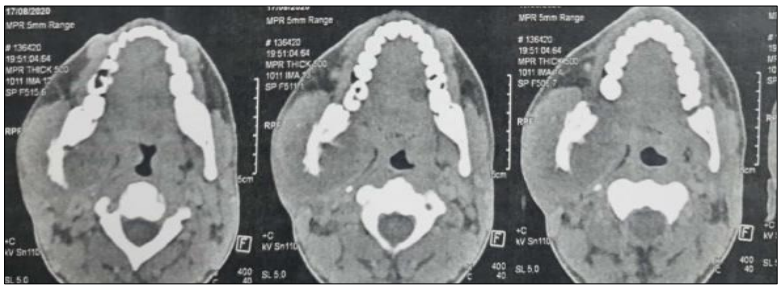

Fig. 8: An axiall CT scan showed a heterogeneous lytic lesion of the ascending branch of the right mandible.

Facial MRI showed a well limited osteolytic mass centered on the ascending branch of the right mandible, measuring 59*78*94 mm, infiltrating the right infratemporal fossa, the right pterygoid, and masseter muscles with intrabuccal extension with bilateral jugulo-

carotid adenomegaly measuring for the largest 12.5mm of the short axis.

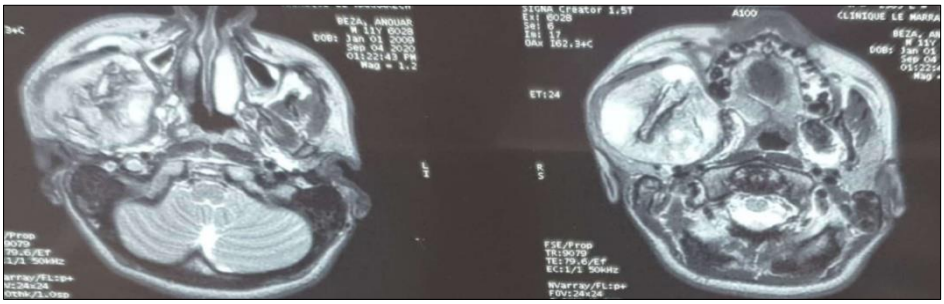

Fig. 9: MRI image showing osteolytic mass centered on the ascending ramus of the right mandible with endobuccal extension

A fine needle biopsy was performed for our patient and found a morphological and immunohistochemical aspect of a conventional osteosarcoma.

The thoraco-abdomino-pelvic CT scanner did not reveal any distant metastasis.

Our patient underwent a surgery that consisted on right hemi-mandibulectomy. The anatomopathological examination showed a 9.5 cm tumor in favor of conventional osteosarcoma, with no vascular or perineural neoplastic invasion and safe surgical margins that remain at least at 1.5 cm from the tumor.

Then the patient received four cycles of API-AI type adjuvant chemotherapy. The evolution was marked by a total regression of the tumoral process. Currently he is at 20 months of supervision

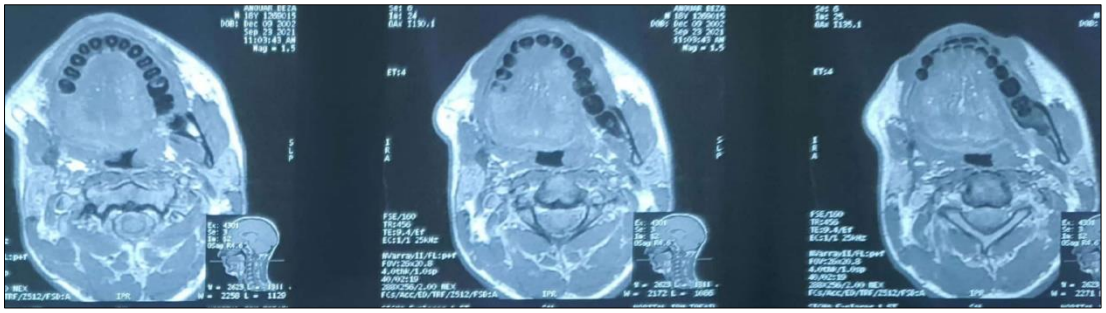

Fig. 10 : MRI image showing total regression of the tumoral process

Fig. 11: image showing clinical result after surgery

DISCUSSION

Osteosarcoma occurs most frequently in the lower long bones, whereas the jaws are unusual primary sites of disease. Maxilla and mandible osteosarcoma represent about 7% of all osteosarcomas [4].

Mandibular OS is often considered as a distinct entity because of its predilection to older patients (mean age: 35 to 40 years). No gender difference for the incidence of OSJ has been reported.

Despite many efforts, the etiology of osteosarcoma remains largely unknown. However, a number of risk factors do exist:[8]

- Rapid bone growth: Increased incidence during the adolescent growth spurt.

- Environmental factors such as radiation. Radiation-induced osteosarcoma is a form of secondary osteosarcoma.
- Genetic predisposition: Bone dysplasias, including Paget disease, fibrous dysplasia, enchondromatosis, and hereditary multiple exostoses and retinoblastoma (germ-line form) are risk factors.

Histopathologically, osteosarcoma is categorized into three subtypes-osteoblastic, chondroblastic and fibroblastic, most series of JOS report predominantly chondroblastic differentiation subtypes, more often myxoid [5].

The duration of symptoms before presentation is typically about 3–6 months. The most common presenting symptoms are swelling at the site of disease, which is almost universally present, and local pain, reported by approximately 70% of the patients. Other complaints are numbness and facial dysesthesia (32%), loosening of the teeth (14%), trismus, limitation of mouth opening, headache and nasal obstruction or bleeding. Patients rarely complain about systemic symptoms like fever, asthenia or weight loss. A few patients have no symptoms at presentation, and their tumors can be discovered incidentally by radiography. Physical examination can demonstrate a painless, firm mass, fixed to the underlying bone covered with normal tissue. Lymph nodes involvement, either cervical, supraclavicular or axillary, is unusual [6].

Radiography is the initial imaging modality in the evaluation of bone tumors.the radiographic appearance may be purely osteolytic, mixed or osteogenic (sun-ray appearance). Periosteal new formation with lifting of the cortex leads to the appearance of a Codman's triangle or attenuation of the lamina dura around the OS are the common radiographic findings [7].

The CT scan allows a better assessment of the bone involvement and extension (better hard tissue definition), whereas the MR imaging aims at defining with considerable accuracy the soft tissue involvement.This is particularly important prior to definitive surgery [15]. As it is the case for other skeletal locations, surgery is a mainstay of osteosarcoma treatment also in the head and neck region. [13].

Obtaining disease-free resection margins is of course imperative, to avoid the risk of local recurrence. Because of the anatomical complexity of the region, tumor resections are occasionally incomplete. Local recurrences and intracranial invasion have long been reported as the major causes of treatment failure due to incomplete neoplasm resection [14].

Chemotherapy and, in particular, postoperative radiotherapy are also complementary therapeutic procedures commonly used in the treatment of head and neck OS [9].

Radiotherapy significantly effects prognosis in skeletal OS, but holds no prognostic significance in OSJs. Associated adjuvant or neoadjuvant chemotherapy has shown promising results both in recurrence incidences and survival, but does not improve poor prognosis of metastatic tumors [3]. Pre-surgical chemotherapy has been a procedure used for large tumors in an attempt to minimize tumor volume and provide a less aggressive surgical approach.

The prognosis of osteosarcoma of the mandible relies on its histological grade and the amount of time elapsing from diagnosis to treatment onset. Osteosarcoma of the jaws has a better prognosis than that of the long bones due to a lower incidence of distant metastases [12]. The survival rate with jaw osteosarcoma is about 50%, compared to a 30% 5-year survival rate with long bone osteosarcoma. Therapeutic failure usually occurs due to local recurrence [11].

CONCLUSION

Osteosarcoma of the jaw is challenging in both diagnosis and management due to the high incidence of faulty biopsy results; rare specific radiological features and the difficulty in proper resection due to proximity to the vital structures.

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