Osteoblastoma is a benign neoplasia and is uncommon in the jaws. In some cases, this lesion presents extremely aggressive local characteristics and is termed aggressive osteoblastoma. Because the clinical, radiographic and histopathologic characteristics are similar to those of a variety of benign and malignant tumors, it poses a diagnostic dilemma. This report presents a case of an aggressive osteoblastoma in the mandible and discusses the differential diagnosis of this lesion. A 13-year-old white male sought the Stomatology Clinic at the State University of Paraíba, Campina Grande, PB, Brazil, complaining of asymptomatic swelling on the left side of his face. Cone-beam computerized tomography showed a multilocular, hypodense bone lesion, located in the body of the left mandible and lower third of the ascending ramus. The initial diagnostic hypothesis was juvenile ossifying fibroma or osteosarcoma. After histopathologic examination, the final diagnosis was aggressive osteoblastoma. Surgical resection with a safety margin was performed. There was no evidence of recurrence after a follow-up period of 4 years.

Aggressive Multilocular Osteoblastoma in the Mandible: A Rare and Difficult Case to Diagnose

Maria Luiza Anjos Pontual1, Andréa Anjos Pontual1, Rafael Grotta Grempe1, Leonardo Rocha Mendes Campos1, Antonio de Lisboa Lopes Costa2, Gustavo Pina Godoy2

Osteoblastoma is a benign neoplasia and is uncommon in the jaws. In some cases, this lesion presents extremely aggressive local characteristics and is termed aggressive osteoblastoma. Because the clinical, radiographic and histopathologic characteristics are similar to those of a variety of benign and malignant tumors, it poses a diagnostic dilemma. This report presents a case of an aggressive osteoblastoma in the mandible and discusses the differential diagnosis of this lesion. A 13-year-old white male sought the Stomatology Clinic at the State University of Paraíba, Campina Grande, PB, Brazil, complaining of asymptomatic swelling on the left side of his face. Cone-beam computerized tomography showed a multilocular, hypodense bone lesion, located in the body of the left mandible and lower third of the ascending ramus. The initial diagnostic hypothesis was juvenile ossifying fibroma or osteosarcoma. After histopathologic examination, the final diagnosis was aggressive osteoblastoma. Surgical resection with a safety margin was performed. There was no evidence of recurrence after a follow-up period of 4 years.

Introduction

Osteoblastoma is a solitary, benign bone neoplasm that is rare in the jaws (1). It corresponds to 1% of all primary bone tumors (1,2) and 3% of all benign bone neoplasias (3). This lesion is a neoplastic bone growth characterized by the proliferation of numerous plump osteoblasts forming osteoid and trabeculated bone scattered in a stroma of richly vascularized fibrous conjunctive tissue (2). This histopathologic aspect is similar to the one in the niche of an osteoid osteoma, which is then called a giant osteoid osteoma due to its unlimited growth potential, resulting in differentiation by its greater diameter (>1.5 cm) (3).

Generally, this lesion affects patients in the first three decades of life, with a male/female sex ratio of 2:1, (3,4). It occurs more frequently in the spinal column (2,3,5–7) and long bones (4–8). Other less common sites are the bones of the skull cap, extremities and face (3). The bones of the face are involved in around 10% (5) to 15% (2,8) of the cases, and of these, the mandible is most frequently affected (2,5,8).

Alvares Capeloza et al. (4) performed a systematic review of the English literature involving 66 cases and reported another new case of osteoblastoma. In 2006, Jones et al. (2) added other 24 new cases to the literature and pointed out the need for reports of new cases of osteoblastoma in the jaws to be published in order to elucidate this uncommon neoplasia. Lypka et al. (8) reported a case of aggressive osteoblastoma in the mandible and Wozniak et al. (9) reported a clinical case of malign transformation of an osteoblastoma in the mandible. In the following year, Harrington et al. (5) reported a case of aggressive osteoblastoma in the maxilla, and more recently, reports of a clinical case of osteoblastoma in the maxilla (10) and another in the mandible were published (11). In 2013, three more cases of osteoblastoma in the jaws were reported, two of which occurred in the jaw (6,12) and one involved the maxilla (7). Therefore, to date, a total of 98 cases of osteoblastoma in the jaws have been reported.

Although osteoblastomas are considered as benign lesions, in some cases, aggressive behavior may occur in the affected region with atypical histopathologic characteristics (8). Aggressive osteoblastoma is an uncommon presentation and has a greater tendency to recur. Moreover, this lesion presents imaging and histopathologic characteristics similar to those of other lesions, which has led to much discussion about its differential diagnosis.

The aim of this article was to relate a clinical case of aggressive osteoblastoma in the region of the mandible and discuss its differential diagnosis and the up-to-date findings in the literature.

Case Report

The patient, a 13-year-old white male, sought the Stomatology Clinic at the State University of Paraíba, Campina Grande, PB, Brazil, complaining of asymptomatic swelling on the left side of his face. The patient’s mother
said that the region in his face has been swelling for 10 months. In the extraoral physical examination, consistent swelling was found on palpation on the left side of the mandible (Fig. 1). In the intraoral physical examination, a swollen region was observed in the lingual and vestibular regions and retromolar to the left side of the mandible (Fig. 2). Additionally, were found residual roots of the mandibular left first molar and displacement of both mandibular left premolars and the mandibular left second molar.

Cone-beam computerized tomography showed a multilocular, hypodense bone lesion, with largest diameter of 5.7 x 4.7 cm, located in the body of the left mandible, extending from the apical region of the mandibular left lateral incisor to the mandibular foramen (Figs. 3-5). Displacement of the mandibular left second premolar, the mandibular left first and second molars and of the germ the mandibular left third molar (Fig. 3) was observed, in addition to inferior displacement of the mandibular canal. The cortical area showed expansion, thinning, and destruction in some regions (Figs. 3 and 4). The initial

Figure 1. Facial asymmetry characterized by swelling in the posterior region of the middle and inferior thirds of the face.

Figure 2. On intraoral examination, tumefaction of the body and retromolar regions were found on the left side of the mandible.

Figure 3. Axial reconstructions (A, B, C) and three-dimensional reconstruction (D) exhibiting horizontal inclusion of the mandibular left second molar, extension of the lesion and effects on the buccal cortical and lingual cortical and teeth.
diagnostic hypothesis was juvenile ossifying fibroma or osteosarcoma.

With the aid of imaging examinations, an incisional biopsy was performed, which was then sent for histopathologic examination. A stroma of dense conjunctive tissue with randomly disposed collagen fibres was verified with intense vascularization and numerous multinucleated giant cells. The trabeculated bone varied from immature (osteoid) to mature bone tissue. The bony trabeculae exhibited osteoblastic pavementation, with plump osteoblasts and abundant hyperchromatic cytoplasm disposed in layers. Rare figures of mitosis were observed. Numerous vascular spaces were observed, with an absence of endothelial cell linings. Intense hemorrhagic extravasation completed the microscopic examination of the histology (Fig. 5). The final diagnosis was aggressive osteoblastoma.

Surgical resection and reconstruction with placement of a titanium plate were performed (Fig. 6). There has been no recurrence over a follow-up period of 4 years (Fig. 7).

Discussion

From our research on the English literature, the present clinical case corresponds to the 99th report of osteoblastoma in the jaws. However, in a PubMed search using the keywords “aggressive osteoblastoma” and “jaws”, only six articles were listed, of which only four actually dealt with cases of aggressive osteoblastoma in the jaws. In the case review studies (1,2,4), there was no classification of the osteoblastomas with regard to the aggressive form. Aggressive osteoblastoma is a lesion on the threshold between a conventional osteoblastoma and osteosarcoma,
with a greater tendency to recur despite not metastasizing, and it is characterized microscopically by the presence of plump osteoblasts (5,11,13).

The patient's age in the present case is within the age range of greatest occurrence of conventional osteoblastomas; most cases affect patients aged from 5 to 24 years (2). Furthermore, the patient was in the second decade of life, which is the most prevalent age range (2,4,9). On the other hand, aggressive osteoblastoma is more prevalent in older patients (11). In addition, the patient in the present case is male, the sex most affected in other reports (around 59% (1) and 70.7% (14) of cases). However, in the sample of 24 cases of osteoblastoma studied by Jones et al. (2), osteoblastomas were more common in female patients (83.3% of cases).

Clinically, osteoblastoma may cause edema, bone expansion, and continuous and punctual pain, which may gradually increase (2,3). Except for the pain, the lesion in the present case had these characteristics, similar to the previously reported cases (1,2,4,15,16). Although pain is considered a frequent characteristic of this lesion (1,4,14), the absence of pain symptomatology at the time of diagnosis may occur in 7.2% of cases of osteoblastoma (4). Other studies have verified a higher frequency of the absence of symptomatology in 25% (1) and 50% (2) of cases. In the present case, the patient did not complain until the lesion was diagnosed, similar to what was found in the previously reported cases (4,15). Thus, many cases of osteoblastomas are discovered by routine radiographic examination, or as a result of investigating the facial swelling caused by the lesion, as seen in the reports by Utumi et al. (17) and in the present case.

Evaluating the cases published in the literature, the mandible was the most involved region of the maxillofacial complex (1,2,4,6,7). The lesion in the present case occurred in the posterior region on the left side of the mandible, the

Figure 6. A: Resection of the mandible was performed with a safety margin and a titanium plate inserted. B: Gross specimen.

Figure 7. A: Panoramic coronal reconstruction 3 years after treatment. B: Coronal reconstruction 3 years after treatment.
Agressive osteoblastoma in the mandible

O osteoblastoma é uma neoplasia benigna e incomum nos maxilares. Em alguns casos esta lesão apresenta características locais extremamente agressivas, sendo denominada osteoblastoma agressivo. Devido às características clínicas, radiográficas e histopatológicas serem similares a uma variedade de tumores benignos e malignos, o seu diagnóstico é um dilema. Este relato apresenta o caso de um osteoblastoma agressivo na mandíbula e discute o diagnóstico diferencial desta lesão. Paciente, branco, 13 anos de idade, foi atendido na Clínica de Estomatologia da Universidade Estadual da Paraíba, Campina Grande, PB, Brasil, queixando-se de aumento...
de volume assintomático do lado esquerdo de sua face. A tomografia computadorizada de feixe cônico revelou uma lesão óssea hipodensa multilocular, localizada no corpo do lado esquerdo da mandíbula e no terço inferior do ramo ascendente. A hipótese diagnóstica foi de fibroma ossificante juvenil e osteosarcoma. Após exame histopatológico, o diagnóstico final foi de osteoblastoma agressivo. Foi realizada resecção cirúrgica com margem de segurança. Não houve sinais de recorrência após 4 anos de acompanhamento.

References

Received September 9, 2014
Accepted September 11, 2014