Surgical treatment of extensive peripheral osteoma in the mandibular ramus: Case report

Tratamento cirúrgico de extenso osteoma periférico em ramo mandibular: Relato de caso

Tratamiento quirúrgico del osteoma periférico extenso en la rama mandibular: Reporte de caso

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Abstract

Osteomas are benign osteogenic tumors, which clinically manifest as well-defined and asymptomatic lesions. This article aims to report a case of extensive osteoma in the mandibular branch, addressing the main tools used for diagnosis and treatment modalities. Patient L.M.S, 55 years old, male, sought the service of Maxillofacial Surgery, reporting the appearance of a nodule on the face with evolution of 2 years. On physical examination, a significant volumetric increase was observed in the pre-auricular region on the left side, asymptomatic on palpation and non-bleeding. Computed tomography was requested, which showed a hyperdense bone lesion in the mandibular ramus region. The planned surgical proposal was for extra-oral access in the pre-auricular region on the left side, excision of the lesion by osteotomy followed by osteoplasty for the regularization of the remaining bone tissue and suture by planes. Subsequently, the piece was sent for histopathological analysis that identified the lesion as a compact osteoma. The patient was followed up for 1 and a half years, through clinical and radiographic evaluation, through which an excellent bone repair was observed, with no signs of recurrence. Carrying out the clink exam is essential for the diagnosis of osteoma, as it is an asymptomatic pathology. Due to the fact that it has a low probability of recurrence and does not present malignancy, a conservative approach can be performed with the appropriate postoperative follow-up. Therefore, it is concluded that the performance of a thorough clinical examination associated with the imaging and microscopic information are essential for a good clinical management of this pathology.

Keywords: Osteoma; Therapeutic approaches; Pathology; Mandible.

Resumo

Osteomas são tumores osteogênicos benignos, que clinicamente, manifestam-se como lesões bem definidas e assintomáticas. O presente artigo tem o objetivo de relatar um caso de extenso osteoma em ramo mandibular, abordando as principais ferramentas utilizadas para o diagnóstico e modalidades de tratamento. Paciente L.M.S, 55 anos, sexo masculino, procurou o serviço de Cirurgia Bucomaxilofacial, relatando o aparecimento de um nódulo no rosto com evolução de 2 anos. Ao exame físico, observou-se aumento volumétrico significativo em região pré-
aurículo do lado esquerdo, assintomático a palpação e não sangrante. A tomografia computadorizada evidenciou uma lesão óssea hiperdensa em região de ramo mandibular. A proposta cirúrgica planejada foi de acesso extraoral em região pré-auricular do lado esquerdo, exérise da lesão por osteotomia seguida por osteoplastia para a regularização do tecido ósseo remanescente e sutura por planos. Posteriormente, a peça foi enviada para análise histopatológica que identificou a lesão como um osteoma compacto. O paciente seguiu em acompanhamento durante 1 ano e meio, mediante avaliação clínica e radiográfica, por meio da qual se observou excelente reparo, não apresentando sinais de recidiva. A realização do exame clínico é fundamental para o diagnóstico do osteoma, por se tratar de uma patologia assintomática. Devido ao fato de apresentar baixa probabilidade de recorrência e não apresentar malignização, uma abordagem conservadora pode ser realizada com o devido acompanhamento pós-operatório. Diante disso, conclui-se que a realização de um minucioso exame clínico associado às informações imaginológicas e histológicas são imprescindíveis para um bom manejo clínico dessa patologia.

Palavras-chave: Osteoma; Condutas Terapêuticas; Patologia; Mandíbula.

1. Introduction

Osteoma is the term used to designate a benign osteogenic neoplasm, essentially restricted to the craniofacial skeleton, with a rare occurrence in other bones of the human body (Garcia et al., 2007; Starch-Jensen, 2017; Marcondes de Castro Rodrigues et al., 2019). This lesion is characterized by the proliferation of mature or spongy compact bone (Starch-Jensen, 2017; Ata-Ali & Ata-Ali, 2019) and is clinically well defined, and may have a sessile or pedicled base, usually asymptomatic and unitary, and which, depending on its location and size, can generate aesthetic changes associated with the occurrence of facial asymmetry and functional damage to phonation, occlusion and deglutition (Garcia et al., 2007; Starch-Jensen, 2017; Castro et al., 2020).

This pathology can be classified based on the location of origin in endosteal when it originates from the endostean (central region of the bone tissue), periosteal when it is originated on the surface of the bone tissue (periosteum), or extra-skeletal when it develops inside the muscle tissue (Neville et al., 2016; Khandelwal et al., 2016; Marcondes de Castro Rodrigues et al., 2019; Castro et al., 2020). Most osteomas are peripheral, varying from 41.9% to 49%, while those of endosteal origin are less frequent (Autorino et al., 2019). In some situations, the presence of this lesion associated with other signs should be investigated, as they may be indicative of Gardner’s Syndrome, an autosomal dominant disease characterized by the presence of multiple osteomas, intestinal polyps, impacted supernumerary teeth, and cutaneous sebaceous cysts (Neville et al., 2016; Marcondes de Castro Rodrigues et al., 2019; Autorino et al., 2019).

The etiology of osteomas, widely discussed in the literature, is still uncertain; it can be associated with conditions such as inflammatory reactions, embryological and genetic factors, continuous muscle action, osteogenic processes, and terminal stages of traumatic healing (Autorino et al., 2019, Aires, et al., 2020). In the craniofacial region, this lesion mainly
affects the paranasal sinus and can also manifest in the mandible, external auditory canal, orbit, temporal bone, and pterygoid process (Fernandez et al., 2017; Castro et al., 2020). Maxillary manifestation is rare (Caubi, et al., 2013; Castro, et al., 2020). The posterior portion of the body is the most affected region of the mandible, followed by the angle, condyle, branch, coronoid process, anterior region of the body, and mandibular notch (Neville et al., 2016; Aires et al., 2020). In general, osteomas are found in both sexes, with the female gender presenting higher treatment rates. They also do not show a predilection for a specific age group; however, was found a higher occurrence in young adults. (Starch-Jensen, 2017; Marcondes de Castro Rodrigues et al., 2019; Castro et al., 2020). The treatment of osteomas consists of complete surgical excision at the base where a cortical bone is located (Horikawa et al., 2012). The procedure for resection of the lesion is recommended in cases where functional and/or aesthetic impairment is associated with lesions with bigger dimensions. When necessary, surgical removal is quite effective in the treatment, and there are no reported cases of malignancy. Lesion recurrence is rare and may be associated with incomplete tumor removal (Misra et al., 2013; Moura et al., 2016; Marcondes de Castro Rodrigues et al., 2019).

The purpose of this article was to report a case of osteoma in the mandibular branch, showing the importance of a correct diagnosis and choice of the ideal treatment approach.

2. Methodology

This article presents a qualitative and descriptive clinical case study. According to the literature, studies of this nature are characterized by explaining a certain topic and studying it thoroughly, through access to medical records, clinical examination, laboratory and image exams provided, with the researcher being the primary instrument (Pereira et al., 2018). The survey was carried out in databases such as PubMed and LILACS, with an analysis of the articles relevant to the subject of the study. As this is a case report, it was not necessary to submit and approve the Research Ethics Committee, but the patient agreed and authorized the use of images, clinical and radiographic data for educational purposes by signing the Free Consent Form. and Informed Consent Form (TCLE) signed. In addition, the present case was conducted in compliance with the ethical principles of the Declaration of Helsinki.

3. Case Report

LMS patient, male, 55 years old, sought the service of Maxillofacial Surgery at Hospital Universitário Lauro Wanderley- João Pessoa, Paraíba, complaining of the appearance of a nodule on the left side of the face with the evolution of 2 years, without increase or decrease in size since he noticed her presence. In the anamnesis, he denied having systemic comorbidities, smoker or alcoholic, and had no history of previous trauma in the region of the injury.

A considerable volumetric increase was observed in the left preauricular region on extra-oral clinical examination, without ulceration or color changes, asymptomatic on palpation and non-bleeding, resulting in a slight facial asymmetry (Figure 1). Upon intraoral physical examination, no significant changes were observed.
**Figure 1** - **A**: Extra-oral clinical aspect in the patient's frontal view. **B**: Extra-oral clinical aspect in the patient's lower view with a volumetric increase evidenced by the red arrow.

![Image of patient in frontal and lower views](source: personal archive)

In image 1, the initial appearance of the patient in a frontal view is observed in A, with a slight easy asymmetry in the pre-auricular region on the left side. In B, it is possible to view this asymmetry in more detail in a lower view due to the volumetric increase highlighted by the red arrow.

Computed tomography showed the presence of a hyperdense bone lesion with well-defined limits in the region of the left mandibular ramus. With dimensions of approximately 15.20 mm in the lateral-lateral direction, 20.75 mm in the anteroposterior direction, and 29.06 mm in the upper-lower direction (Figure 2).

**Figure 2** - **A**: Axial section of the computed tomography showing the dimension of the lesion in the latero-lateral direction. **B**: Axial section of the computed tomography showing the dimension of the lesion in the anteroposterior direction. **C**: Coronal section of the computed tomography showing the dimension of the lesion in the supero-inferior direction.

![Images of computed tomography sections](source: personal archive)
Image 2 shows the axial and coronal sections in A, B, and C respectively for hard tissues by computed tomography. It is possible to verify the extent of the lesion in question and its intimate contact with the surface of the bone tissue.

The reconstruction allowed a three-dimensional visualization of the area of interest. It evidenced the presence of a well-defined lesion with a benign pedicled appearance in the external cortical layer of the ascending mandible branch (Figure 3).

**Figure 3 - Three-dimensional image showing the dimensions and the pedicled aspect of the lesion.**

In image 3, it is possible to verify in the 3D reconstruction of the computed tomography the positioning and the extent of the lesion in question.

The patient underwent a surgical procedure, under general anesthesia, for the excision of the lesion. Local antisepsis and surgical field preparation were performed. Then, a retromandibular incision was made to access the ascending branch of the left mandible (Figure 4A), in which osteotomies were performed on the osteoma, removing the lesion by fragments (Figure 4B). Subsequently, osteoplasty was performed to regularize the remaining bone tissue (Figure 4C). After copious irrigation of the area with 0.9% saline, the sutures with plans were performed with Vicryl 4-0 for the inner planes and Nylon 5-0 for the skin.
In Image 4 in A, it is possible to observe the surgical access; in B, it is possible to observe the fragments of the lesion after its removal. C shows the aspect of surgical access after removing the lesion and regularization of the remaining bone surface, thus showing the superficial character of the osteoma.

The specimens were sent for histopathological analysis, where dense bone tissue was observed, with osteocytes, vascular spaces, and basophilic inversion lines (Figure 5). Histopathological findings were compatible with the diagnosis of compact osteoma.

**Figure 5** - Histological aspect, showing dense bone tissue with vascular spaces (indicated by red arrows), osteocytes (indicated by blue arrows) and basophilic inversion lines (indicated by black dashes) inside.
In Image 5, it is possible to observe the histopathological aspect of the removed lesion. Microscopic findings such as the presence of osteocytes, blood vessels, and basophilic inversion lines were compatible with the diagnosis of compact osteoma.

The patient was followed up for 1 and a half years through clinical and radiographic evaluation, through which an excellent bone repair was observed, with no signs of recurrence. (Figure 6A e 6B).

**Figure 6 - A:** Postoperative clinical follow-up of the patient. **B:** 3D reconstruction of postoperative computed tomography suggestive of excellent bone repair and absence of recurrence.

Source: personal archive.

In Image 6 A, it is possible to view the patient's clinical follow-up in the postoperative period, showing excellent tissue repair. In B, 3D reconstruction suggestive of effective bone repair is observed, with no signs of recurrence of the lesion, evidencing the success of the recommended treatment.

4. Discussion

Osteomas are benign bone neoplasms resulting from the proliferation of compact or spongy bone that is clinically manifested as a well-defined, rounded, or oval mass, with firm consistency on palpation, usually asymptomatic and of slow growth. Depending on its location and size, it can cause aesthetic and functional damage (Garcia et al., 2007; Neville et al., 2016; Starch-Jensen, 2017; Castro et al., 2020;). In the reported case, the volumetric increase in the region of the left mandibular ramus resulted in the occurrence of facial asymmetry. This aesthetic change caused discomfort to the patient, leading him to seek care.

Although the etiology and pathogenesis of osteomas are still unknown, some authors attribute the occurrence of the neoplastic process in bone tissue to reactive factors, such as trauma. Other authors associate the occurrence of this benign lesion with inflammatory processes or hamartomatous events (Bullot et al., 2010; Caubi et al., 2013; Castro et al., 2020). Conditions associated with the persistence of the embryonic periosteum have also been related to the development of the lesion, especially in cases in which researchers were unable to establish the relationship between the beginning of its
development and the occurrence of trauma or previous inflammatory processes (Bullot, et al., 2010; Aires, et al., 2020). In the present case, the patient was not exposed to any inflammatory or traumatic episode in the mandible region.

As it develops asymptomatically, osteoma is usually discovered in routine radiographic examinations (Bullot et al., 2010). Radiographically, the lesion is characterized as a well-defined, round, or oval radiopaque mass, with a sessile or pediculated base attached to the bone tissue (Misra et al., 2013; Neville et al., 2016; Fernandez et al., 2017). Imaging examinations such as panoramic radiography, ultrasound, or computed tomography contribute significantly to the diagnosis (Bullot et al., 2010; Cardoso et al., 2020). Studies in the literature have shown that computed tomography is the ideal imaging test to determine the actual location and extent of the lesion, as it is a versatile test that produces detailed images of bone tumors, with good sensitivity and great specificity for abnormalities. Cone-beam computed tomographies also reproduce detailed images with high specificity but are obtained with a lower dose of radiation; such a characteristic must be taken into account when choosing the ideal exam (Shintaku et al., 2010; Bullot et al., 2010; Aires et al., 2020). In the case in question, conventional computed tomography was chosen, since in addition to the excellent image reproduction properties, it is an exam available in the Unified Health System, thus not generating additional costs for the patient.

Possible differential diagnoses for osteomas include peripheral ossifying fibroma, torus and bone exostoses, osteosarcomas, osteoblastomas, osteochondromas, osteoid osteoma, among others (De Bartoli et al., 2018; Marcondes de Castro Rodrigues et al., 2019). There is no significant microscopic difference between osteomas and exostoses, therefore, the differential diagnosis is restricted to clinical characteristics (Marcondes de Castro Rodrigues et al., 2019). Exostoses, usually bilateral, are considered developmental hamartomas and tend to stop evolving after the end of puberty, while osteomas may show continuous growth even after the end of this period, indicating the neoplastic character of the lesion (Geron et al., 2017; De Bartoli et al., 2018; Aires et al., 2020). In the case in question, the development of the lesion was detected after puberty; besides, it was a solitary and unilateral mass, showing the classic characteristics of this bone tumor. The diagnosis was confirmed after performing histopathological analysis, in which the findings were compatible with the condition of compact osteoma.

Approaches to the treatment of osteomas are widely discussed in the literature. The slow growth of the lesion allows conservative treatment to be carried out, in which the patient undergoes clinical and radiographic monitoring for a certain period to check for progression. In cases where the lesion does not progress, surgical treatment is not recommended. The decision to surgically remove the lesion should be based on an adequate assessment of the risks of the surgery, including possible damage to critical anatomical structures (Ata-Ali & Ata-Ali, 2019). Surgical resection is indicated for cases that there is a functional and/or aesthetic impairment associated with extensive injuries (Moura et al., 2016; Aires et al., 2020; Torres et al., 2020; Rodríguez-Álvarez et al., 2021). In the case reported, although there was no functional impairment, the volumetric increase in the ascending branch region was associated with an aesthetic change resulting from facial asymmetry, which led to the choice of surgical excision as a therapeutic approach.

The recurrence rate of the lesion is rare and may be associated with incomplete removal of the tumor. For this reason, the patient must be monitored with periodic radiographs during a period to verify whether the procedure performed was effective (et al., 2013; Castro et al., 2020; Aires et al., 2020). In the present case, the patient was followed up for a year and a half using clinical and radiographic evaluation, through which an excellent bone repair was observed, with no signs of recurrence and complications, indicating that the surgical procedure performed was effective.

Therefore, it is necessary to carry out further prospective studies to certify the best therapeutic approach in the case of this type of injury.
5. Final Considerations

It is concluded that the performance of a detailed clinical examination associated with the imaging and microscopic information is essential for a good diagnosis and clinical management of this condition. When faced with osteoma, the professional must perform a thorough investigation since the presence of this lesion may be associated with Gardner's Syndrome.

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