CASE REPORT

GIANT OSTEOMA IN THE ASCENDING RAMUS OF THE MANDIBLE – RADIOGRAPHIC AND TOMOGRAPHIC ASPECTS OF A CASE

OSTEOMA GIGANTE EM RAMO ASCENDENTE DE MANDÍBULA – ASPECTOS RADIOGRÁFICOS E TOMOGRÁFICOS DE UM CASO

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ABSTRACT

Osteoma is an unusual benign tumor of bone origin characterized by proliferation of compact or medullary bone. It is most common in adults between the third and fifth decades of life, with no gender predilection and can be categorized as central, peripheral or extra-skeletal. This study aims to report the case of a patient who was seen at the Odontoclínica Central da Marinha with a final diagnosis of Osteoma with approximately 10 years of evolution. A 58-year-old black woman was referred complaining of pain when chewing on the left side and displacement of the jaw to the right side. On physical examination, facial asymmetry was observed with a well-defined swelling of firm consistency in the parotid region on the left side, in addition to a swollen palate. Radiographically, a rounded and well-defined radiopaque image was observed, affecting the ascending ramus of the mandible, extending to the condyle and coronoid process on the left side. On cone beam computed tomography, a hyperdense, multilobular, well-defined and corticalized image was noted, located on the left side of the mandible, involving the ascending ramus, breaking the lingual cortex and invading the cranial region of soft tissues. The patient underwent an incisional biopsy with a histopathological report compatible with osteoma. Giant osteomas are unusual, mainly in the jaw region. The dentist must be aware of this pathology due to the risk of involvement with syndromes, also because it affects the aesthetics and function of affected patients.

Keywords: Osteoma; Bone tumors; Bone pathology; Diagnosis; Cone beam computed tomography.

RESUMO

O osteoma é um tumor benigno incomum de origem óssea, caracterizado por proliferação de osso compacto ou medular. É mais comum em adultos entre trinta e cinquenta anos, sem predileção por gênero e pode ser categorizado como central, periférico ou extra-esquelético. Este estudo tem por objetivo relatar o caso de uma paciente que foi atendida na Odontoclínica Central da Marinha com diagnóstico final de Osteoma com cerca de 10 anos de evolução. Paciente mulher, 58 anos, melanoderma, encaminhada por queixa de dor à mastigação do lado esquerdo e desvio de mandíbula para o lado direito. Ao exame físico, observou-se assimetria facial com aumento de volume bem delimitado de consistência firme em região parotídea do lado esquerdo, além de abaulamento em palato. Radiograficamente, observou-se imagem radiopaca de formato arredondado e bem definida, acometendo ramo ascendente da mandíbula, estendendo-se para côndilo e processo coronoide do lado esquerdo. Na tomografia computadorizada de feixe cônico, notou-se imagem hiperdensa, multilobular, bem definida e corticalizada, localizada do lado esquerdo da mandíbula, envolvendo o ramo ascendente, rompendo a cortical lingual e invadindo a região craniana dos tecidos moles. A paciente foi submetida à biópsia incisional com laudo histopatológico compatível com osteoma. Osteomas gigantes são incomuns, principalmente, em região de maxilares. O cirurgião-dentista deve estar atento a essa patologia devido ao risco de envolvimento com síndromes, também por afetar estética e função dos pacientes afetados.

Palavras-chave: Osteoma; Tumores ósseos; Patologia óssea; Diagnóstico; Tomografia Computadorizada de Feixe Cônico.

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INTRODUCTION

The craniofacial osteoma is a rare benign tumor of bone origin, characterized by proliferation of compact or medullary bone (1,2). Although the literature presents a wide age range of individuals affected by this type of neoplasia, it is more common in adults between the third and fifth decades of life, with no sex predilection (2,3).

This pathology can be categorized as central, affecting the medullary region of the affected bone, peripheral, presenting as a pedunculated volume increase at the margin of the craniofacial bone, or extra-skeletal, involving its manifestation in soft tissues, such as muscles (3,4).

The craniofacial skeleton is the preferred site for the development of osteoma, being more common in paranasal sinuses (frontal, ethmoidal, and maxillary) and frontal bone (2,4). In jawbones, these lesions are rare and, when they occur, are more frequent in the posterior mandibular body and less frequent in the maxilla and mandibular condyle (2,4). Even though most cases are asymptomatic, there may be aesthetic complaints due to facial asymmetry, discomfort during chewing, difficulty using dental prostheses, opening the mouth, among others (2,5).

The diagnosis of osteoma is made through the correlation of clinical, imaging, and histopathological examination. Clinically, these lesions present as well-defined and hardened swellings. Regarding imaging exams, it is possible to initially use panoramic radiography, but computed tomography is considered essential for assessing the size, precise location, and anatomical relationship of the lesion with adjacent structures (1). In the exam, these lesions present as dense radiopacities or well-defined and corticated hyperdensities (3).

Osteoid osteoma, cementoblastoma, complex odontoma, ossifying fibroma, osteoblastoma, chondroma, osteosarcoma, Paget's disease, and even idiopathic osteosclerosis represent the main differential diagnoses for this tumor (1,6). It is also important to differentiate osteomas from exostoses and torus, as they can be very similar clinically, since they present as a hardened swelling in the maxillo-mandibular region (1).

Another relevant factor in the identification of osteomas in the craniofacial region is their relationship with Gardner's Syndrome, a genetic disease with a high predisposition to the development of malignant colorectal tumors (1,7). All cases should be investigated, especially if there is more than one bone lesion, as this may be the first warning sign for the diagnosis of this syndrome (4,7).

Histopathologically, there is a proliferation of compact or cancellous bone with a normal appearance, with bony trabeculae that may be permeated by fibroadipose marrow (2,3). Osteoblasts and osteoclasts are generally imperceptible, but some may contain areas similar to osteoblastoma, suggesting an active remodeling process, although this does not characterize a sign of tumor aggressiveness (3). According to this analysis, osteomas can also be classified as central or peripheral (3).

The treatment of osteoma is usually surgical and with rare recurrence (3). In more extensive cases, although it is a benign tumor, bone plasty may be indicated for aesthetic reasons. In this way, the aim of this study was to report the case of a large osteoma in the region of the ascending ramus of the mandible, detailing its main radiographic and tomographic findings.

CASE REPORT

This is a descriptive, retrospective case report study. The present study was approved by the responsible Research Ethics Committee (CAAE: 81171024.0.0000.5256, No. 6,939,705). The patient has read and signed the informed consent form.

A 58-year-old black woman, hypertensive and diabetic, with no habits of alcohol or tobacco consumption, attended the Odontoclínica Central da Marinha (OCM), in Brazil, referred in October 2022, by a primary care dentist with a complaint of pain during chewing on the left side of the palate and jaw deviation to the right side, with a reported evolution of approximately 10 years. She was referred to the Stomatology Clinic and, upon physical examination, facial asymmetry was observed with a well-defined firm consistency mass in the left parotid region, as well as swelling in the palate, extending to the retromolar region up to the tonsillar pillar on the same side (Figure 1). It was found that the upper prosthesis was in contact with the swollen palate, which justified the discomfort during chewing. The patient had a mandibular deviation to the right side and limitation in mouth opening movement.

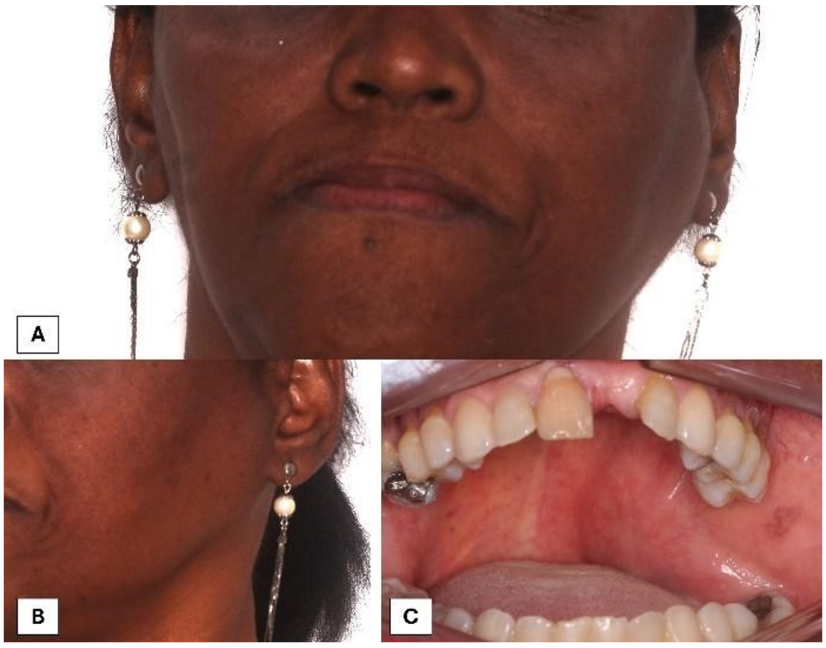

Figure 1: Clinical aspect demonstrating extra-oral swelling in the parotid and intra-oral region.. **A** and **B**- front and side views, respectively, showing lump in the left parotid region; **C**- Swelling in the soft palate and left retromolar region.

When evaluating the previous medical record, it was found that an examination from 2013, almost 10 years earlier, already showed the lesion, although no treatment was proposed at that time (Figure 2A). A new panoramic radiograph was then requested, in which an image of radiopaque density with a rounded and well-defined shape was observed, affecting the ascending ramus of the mandible, extending to the mandibular condyle and coronoid process on the left side. An extension of the lesion beyond the posterior bone cortex of the mandible was also noted (Figure 2 B). At this moment, salivary gland tumor, such as pleomorphic adenoma due to its location, or bone pathology, including osteoma and benign fibro-osseous lesions, were suggested as diagnostic hypotheses. A cone beam computed tomography (CBCT) was requested to better clarify the extent of the lesion and the structures involved.

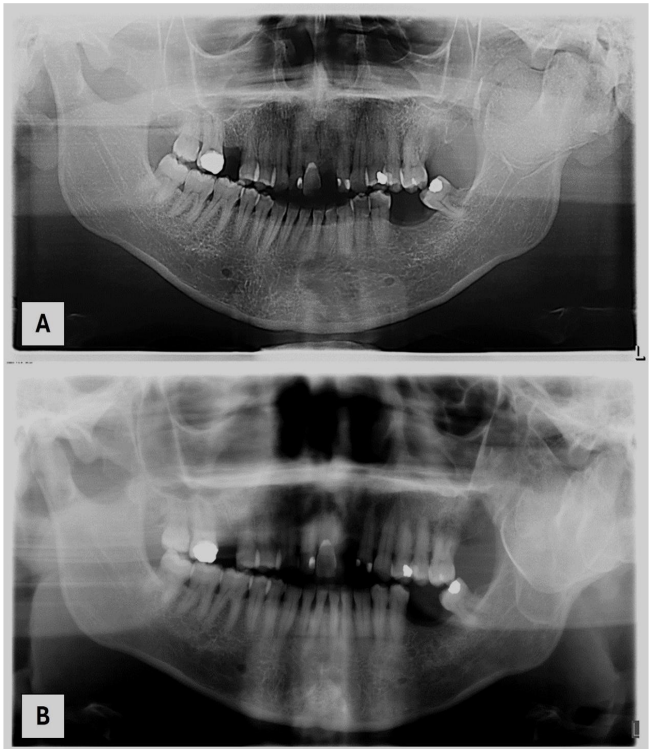

Figure 2: Panoramic radiographs showing the evolution of the lesion over approximately 10 years (**A** – 2013; **B** – 2022), with a multilobulated and corticated radiopaque image in the region of the ascending ramus of the mandible and the condyle.

Volumetric acquisition was performed using a cone beam X-ray on the i-CAT tomograph (Imaging Sciences 23 International, Hatfield, PA, USA), with a voxel of 0.2mm and subsequent multiplanar axial, sagittal, and coronal reconstruction with a thickness of 1mm by the software OnDemand3D. In the evaluation of the CT scan, a hyperdense, multilobular, well-defined, and corticalized image was observed, located on the left side of the mandible, involving the ascending ramus, breaking the lingual cortex and invading the cranial region of the soft tissues. In the craniocaudal direction, the lesion extended from the maxillary tuberosity to the ascending ramus of the mandible, while in the anteroposterior direction, the lesion affected the region from the maxillary tuberosity to the mastoid process. The measurements obtained were 54mm in the craniocaudal direction, 62.4mm in the antero-posterior direction, and 42.8mm in the vestibular-lingual direction. Interestingly, in a region of the ascending ramus of the mandible, in the upper third, vestibular cortex, the image did not show corticalization, becoming homogeneous with the bone trabeculae. The resorption of the mandibular edge, involvement and inferior displacement of the mandibular canal, rupture of the lingual cortex of the condyle, as well as resorption of the maxillary tuberosity and constriction of the left maxillary sinus were also evidenced (Figures 3-7). The observed images suggested a pattern of benign growth, tending towards fibro-osseous lesion as ossifying fibroma, or benign bone tumor.

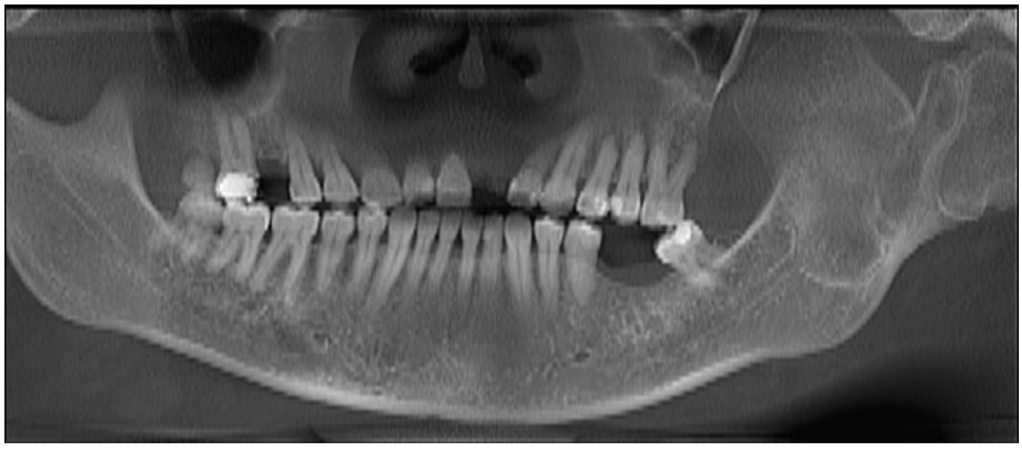

Figure 3: Panoramic reconstruction. Extensive lesion is observed in the region of the ascending ramus of the mandible and the condyle on the left side.

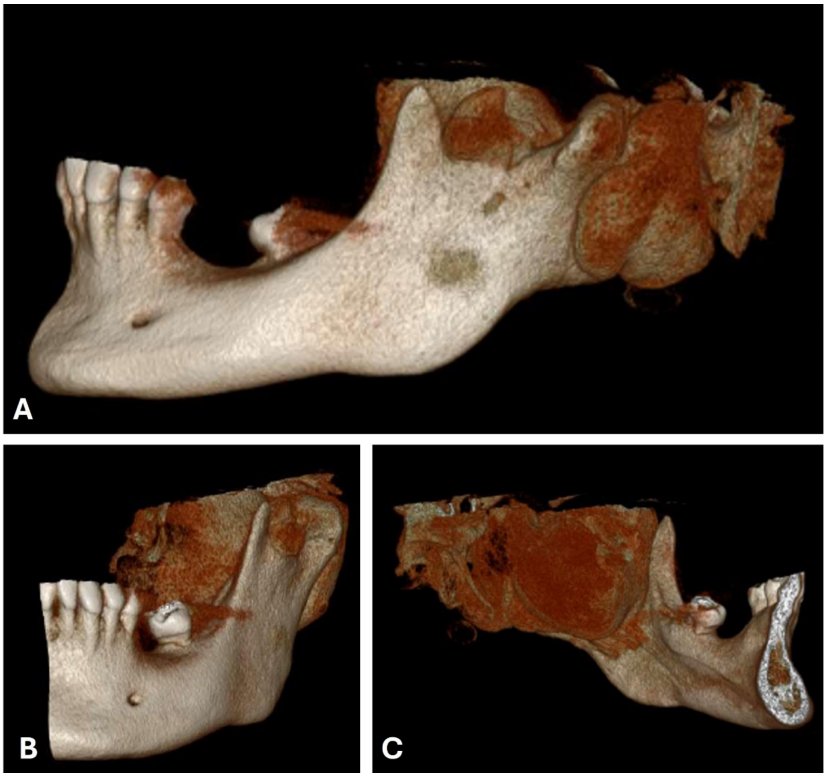

Figure 4: Three-dimensional volumetric reconstructionss (**A** – Lateral view; **B** – Front view; **C** – Lingual view).

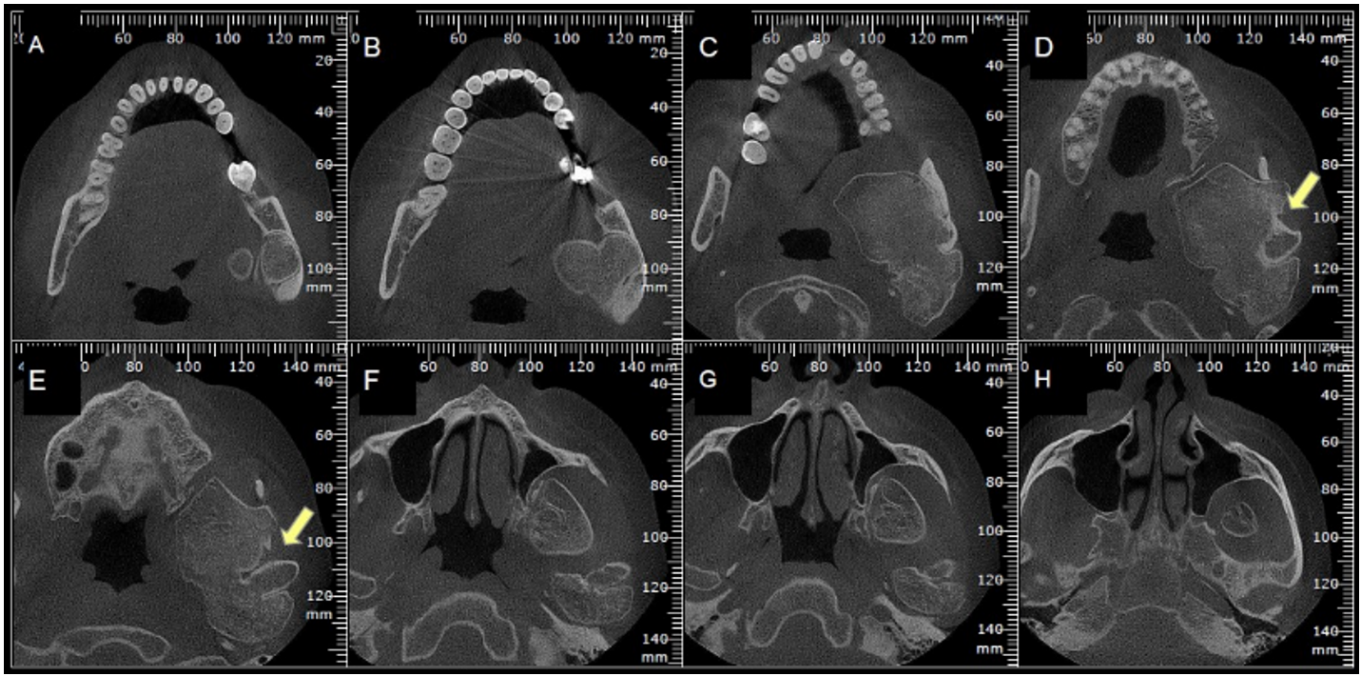

Figure 5: Axial reconstructions (craniocaudal direction) demonstrating the extension of the lesion from the body of the mandible to the cranial region of soft tissues. Note the mandibular condyle involved by the lesion (yellow arrow).

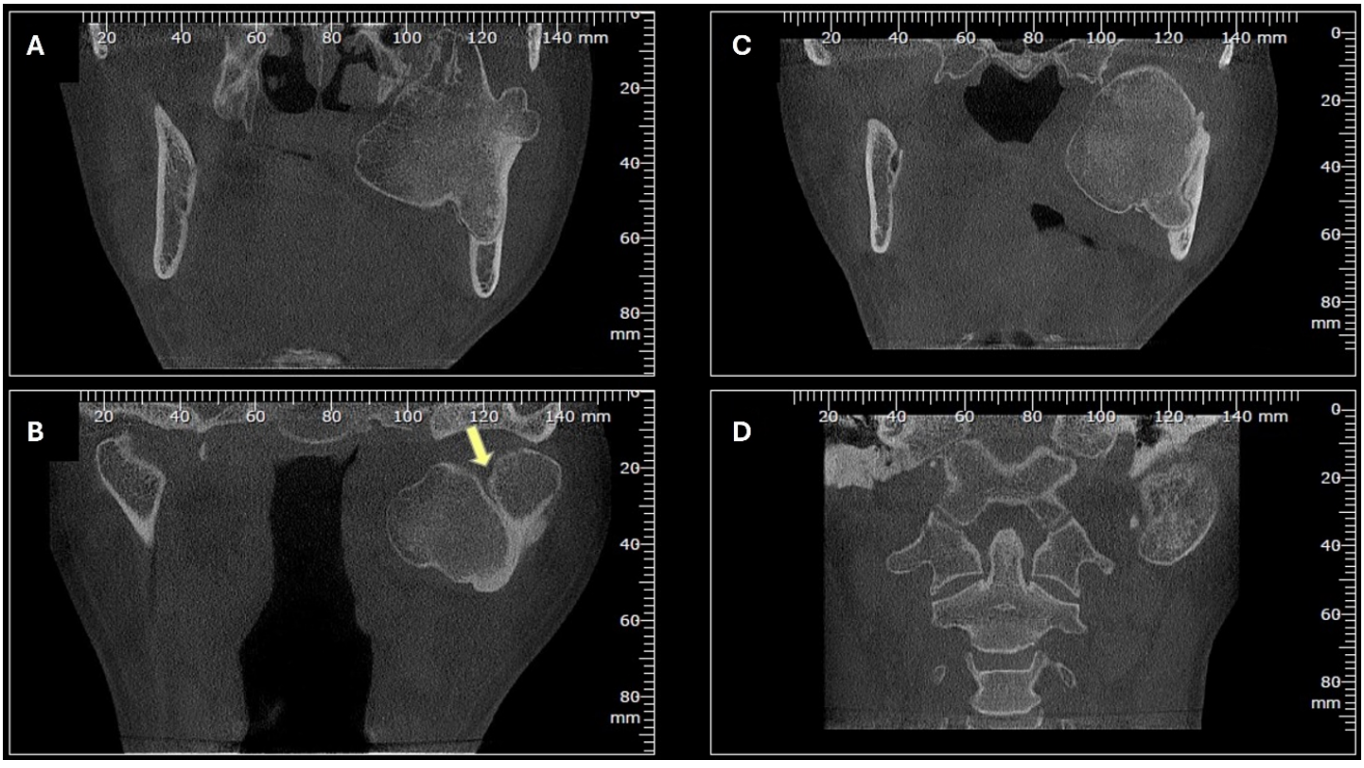

Figure 6: Coronal reconstructions (antero-posterior direction) showing the extent of the lesion, with thinning of the mandibular cortex and proximity to the mandibular condyle (yellow arrow).

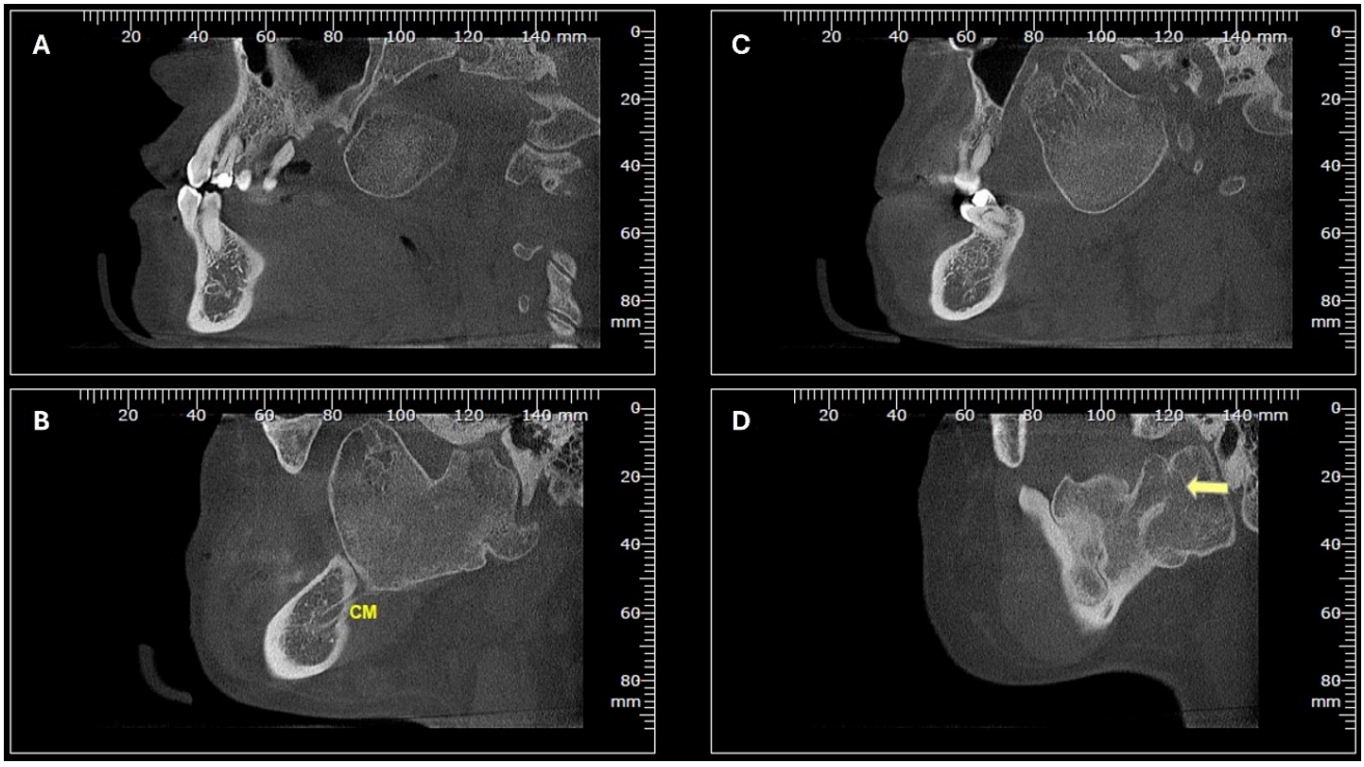

Figure 7: Sagittal reconstructions showing lesion invasion into soft tissues, constriction of the mandibular canal (CM), and rupture of the bone cortex of the mandibular condyle (yellow arrow).

The patient underwent an incisional biopsy for diagnostic elucidation and subsequent therapeutic planning. The histopathological examination showed cortical and medullary bone tissue with fibroadipose marrow, compatible with osteoma. The patient was referred to the Oral and Maxillofacial Surgery clinic at the Hospital Naval Marcílio Dias, in Brazil, for surgical intervention. Until the time of this case report, the team was discussing the case, as the lesion is close to critical areas and requires a delicate approach, possibly needing multidisciplinary intervention. Currently, the patient has no pain complaints, but has aesthetic discomfort due to facial asymmetry.

DISCUSSION

Osteomas are rare tumors, mainly in the jaw region, as their preferred location is the paranasal sinuses (3). A study conducted in Turkey demonstrated the radiographic aspects of rare non-odontogenic lesions in the jaws, and after analyzing more than 8,000 computed tomography and magnetic resonance imaging scans, the authors found 19 cases of non-odontogenic tumors, among them only one osteoma, accounting for less than 0.01% of the sample (8). Another study, with a literature review of 69 cases of peripheral osteoma, showed only 7 occurrences in the ascending ramus of the mandible and 18 in the condyle (2). Considering the giant osteomas, Hasan conducted a literature review with 30 cases in the mandible, with only five in the ascending ramus of the mandible (9), corroborating the rarity of the lesion presented in this case report.

In terms of pathogenesis, there is still controversy in the literature. Some authors advocate the theory that trauma in the jaw region or even dental extractions could trigger the development of this type of lesion, although it does not justify all cases (5,10). Other possibilities include the development of true neoplasia, developmental anomalies, and endocrine changes (2). Many authors prefer, however, the mechanism that suggests that the trauma associated with muscle traction could cause small periosteal hemorrhages, elevating the periosteum and thus generating an osteogenic reaction, especially in cases of peripheral osteomas (2,4,10). The present case did not show evidence of trauma, the patient did not report any symptoms, and the lesion had existed for about 10 years, according to previous radiographic examinations, with slow and gradual progression.

Osteomas can affect patients across a wide age range, with reports from 16 to 74 years, but they are more common between 20 and 50 years of age,

which is consistent with the case presented here, because although the patient was 58 years old at the time of diagnosis, the lesion had already been present in radiographic findings for almost 10 years. Often, these cases may not be properly diagnosed because the lesion is asymptomatic and with a slow growing pattern. Gawande *et al*. reported a similar case, in which the patient was a 45-year-old woman with facial asymmetry, significant volume increase in the jaw, asymptomatic and with no prior history of trauma (2).

It is important to note that this bone neoplasm may be related to some syndromes, such as Gardner's syndrome, Haberland syndrome, Opitz G/BBB syndrome, and acromegaly (4). Gardner's syndrome is most frequently associated with osteoma, especially in cases of multiple bone tumors. This syndrome presents a mutation in the APC gene and predisposes the development of colorectal cancer in more than 90% of patients, in addition to the possibility of the emergence of other occasional tumors (11). The diagnosis of osteoma can occur in younger patients and in some cases assist in the early diagnosis of the syndrome, favoring the patient's prognosis, which draws particular attention to the proper understanding of this pathology (11). In the case presented here, the patient did not have other tumors or a history of colon cancer, which would rule out the possibility of Gardner's syndrome.

Clinically and radiographically, the osteoma can suggest several differential diagnoses, especially due to its highly variable presentation in shape and size. Among them, benign fibro-osseous lesions, complex odontoma, osteoblastoma, cementoblastoma, exostoses, idiopathic osteosclerosis, osteoid osteoma, among others (1,4,6). For this reason, clinical correlation, imaging, with radiographs and CT scans, with histopathological analysis becomes essential for the correct definition of diagnosis and treatment. The present case had as its main differential diagnosis the ossifying fibroma, due to its size and its relationship with the mandibular bone, which suggested the appearance of a tumor detachable from the affected bone, however, the histopathological analysis revealed an aspect compatible with the diagnosis of osteoma.

The osteoid osteoma, for example, is an entity that can cause diagnostic confusion with central osteomas. Nonetheless, this other tumor presents some particularities that aid in diagnosis, as it generally causes constant pain, with reports of worsening at night, and improves with the oral administration of acetylsalicylic acid (6,12). Osteoid osteoma presents radiographically as a well-defined radiopaque image with a radiolucent halo and is histologically differentiated from osteoma by having a highly vascularized substrate and osteogenic connective tissue associated with newly formed bone trabeculae (6). Kammoun *et al*. reported a case of osteoid osteoma where the lesion was osteolytic at the base of the mandible and was masked on the panoramic view by an overlap of the hyoid bone, due to poor patient positioning during the radiographic technique. After the extraction of a tooth and without resolution of the patient's complaint, a CBCT was requested which showed the lesion at the base of the mandible, being fundamental for the diagnosis and management of the case (12).

In terms of imaging diagnosis, panoramic radiography is the first examination requested and, often, can reveal osteomas as accidental radiographic findings. However, the X-ray is a two-dimensional exam and limits the determination of the actual dimensions of the lesion, in addition to presenting many areas of overlapping structures that can confuse the diagnosis (12). The case presented in this report had already shown a radiopaque image in the region of the ascending ramus of the mandible and condyle, but the patient did not undergo a diagnosis, as she had no complaints in the region. After the report of discomfort, a new panoramic X-ray and completion with CBCT were requested, which effectively directed the diagnosis.

CTCB is an imaging modality that ensures three-dimensional visualization of the maxillo-mandibular complex, being increasingly used in dentistry for various purposes (13,14). In the diagnosis of bone lesions, such as osteoma, tomography is the best choice for determining the size, location, and anatomical relationship of the lesion with adjacent structures (1). The present case evidenced, through CT scan, a large bone lesion with thinning of the cortices, invasion of soft tissue, and displacement of the mandibular canal. This type of detail provides crucial information for surgical planning and ensures that the treatment is carried out more safely and with a better prognosis for the patient in terms of recovery and recurrence of the lesion (14).

CONCLUSION

Giant osteomas are uncommon, especially in the jaw region. The dentist must be aware of this pathology due to the risk of involvement with syndromes, as well as affecting the aesthetics and function of affected patients. In this sense, the clinical, histopathological, and imaging diagnosis of this pathology is of great relevance.

The authors declare that there is no conflict of interest.

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