



UNIVERSIDADE FEDERAL DE UBERLÂNDIA
FACULDADE DE ODONTOLOGIA



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**UNUSUAL CASE OF PERIPHERAL
OSTEOMA IN YOUNG PATIENT**

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OSTEOMA IN YOUNG PATIENT**

Trabalho de conclusão de curso
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Odontologia da UFU, como requisito
parcial para obtenção do título de
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Orientadora: Prof^ª. Dr^ª. Gabriella Lopes
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 TRABALHO DE CONCLUSÃO DE CURSO

ATA DA COMISSÃO JULGADORA DA DEFESA DE TRABALHO DE CONCLUSÃO DE CURSO DO (A) DISCENTE **Anaísa Oliveira Palhares** DA FACULDADE DE ODONTOLOGIA DA UNIVERSIDADE FEDERAL DE UBERLÂNDIA.

No dia **08 de novembro de 2018**, reuniu-se a Comissão Julgadora aprovada pelo Colegiado de Graduação da Faculdade de Odontologia da Universidade Federal de Uberlândia, para o julgamento do Trabalho de Conclusão de Curso apresentado pelo(a) aluno(a) **Anaísa Oliveira Palhares**, COM O TÍTULO: **“UNUSUAL CASE OF PERIPHERAL OSTEOMA IN YOUNG PATIENT”**. O julgamento do trabalho foi realizado em sessão pública compreendendo a exposição, seguida de arguição pelos examinadores. Encerrada a arguição, cada examinador, em sessão secreta, exarou o seu parecer. A Comissão Julgadora, após análise do Trabalho, verificou que o mesmo se encontra em condições de ser incorporado ao banco de Trabalhos de Conclusão de Curso desta Faculdade. O competente diploma será expedido após cumprimento dos demais requisitos, conforme as normas da Graduação, legislação e regulamentação da UFU. Nada mais havendo a tratar foram encerrados os trabalhos e lavrada a presente ata, que após lida e achada conforme, foi assinada pela Banca Examinadora.

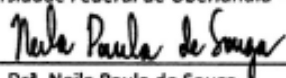
Uberlândia, 08 de novembro de 2018.


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*“São as nossas escolhas, Harry,
que revelam o que realmente somos,
muito mais do que as nossas
qualidades.”*

(Albus Dumbledore)

SUMMARY

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ABSTRACT

Osteoma is a benign slow-growing osteogenic neoplasm that commonly occurs in the craniofacial region and is characterized by the proliferation of compact and/or cancellous bone. Osteomas may be peripheral, central or extraskeletal. The exact etiology and pathogenesis of peripheral osteoma are still unknown. Recurrence of peripheral osteoma after surgical removal is extremely rare and there are no reports of malignant transformation. The aim of this paper was to describe an unusual case of an 11-year-old male patient presenting a solitary peripheral osteoma in the right retromolar trigone region, a site that was not previously reported. The patient was initially asymptomatic and after one month, the lesion started to cause facial asymmetry and discomfort while chewing. This paper also emphasizes its clinical, radiographic, surgical and histopathological features, as well as the differential diagnosis.

Keywords: Osteoma, Oral Diagnosis, Diagnostic Imaging

RESUMO

Osteoma é uma neoplasia osteogênica benigna de crescimento lento e comumente ocorre na região craniofacial e é caracterizada pela proliferação de osso compacto e/ou esponjoso. Os osteomas podem ser periosteais, centrais ou extraesqueléticos. A etiologia exata e a patogênese do osteoma periosteal ainda são desconhecidas. A recorrência de osteoma periosteal após a remoção cirúrgica é extremamente rara e não há relatos de transformação maligna. O objetivo deste trabalho foi descrever um caso incomum de um paciente masculino de 11 anos apresentando um osteoma periosteal solitário na região do triângulo retromolar direito, um local que não foi relatado anteriormente. Inicialmente, o paciente estava assintomático e, após um mês, a lesão começou a causar assimetria facial e desconforto durante a mastigação. Este trabalho também enfatiza suas características clínicas, radiográficas, cirúrgicas e histopatológicas, bem como o diagnóstico diferencial.

Palavras-chave: Osteoma, Diagnóstico Bucal, Diagnóstico por Imagem

INTRODUCTION

Osteoma is a benign slow growing neoplasm characterized by the proliferation of compact or cancellous bone, and one of the most common tumors of the nose and paranasal sinuses. It was first recognized as a tumor by Jaffe in 1935.¹ Multiple osteomas are mainly associated with Gardner's syndrome while solitary osteomas can be further classified based on the location as: (1) the central osteoma arising from the endosteum; (2) the peripheral osteoma deriving from the periosteum and (3) the extraskeletal soft tissue osteoma, which usually develops within the muscles.² Solitary peripheral osteomas affecting the maxillomandibular region are reasonably rare. These lesions are more frequent in the mandible than in the maxilla. Sayan *et al.*, (2002) reported that most cases of osteoma in their sample affected the frontal bone (28.57%) while 22.85% were found in the mandible and 14.28% in the maxilla.³

Also, Kaplan *et al.* (2008) reported that the mandible was affected more frequently in cases affecting jaw bones.⁴ The peripheral variant is the most common and it occurs most frequently in the frontal, ethmoidal, and maxillary paranasal sinuses. The exact etiology and pathogenesis of peripheral osteoma are unknown, three theories are more accepted: developmental defect, neoplastic nature, and reactive lesion due to trauma or local infection.

Although not completely elucidated, the etiopathogenesis of peripheral osteomas has been associated with the reactional theory, relating its onset with the previous history of local trauma, being some cases named of traumatic peripheral osteoma.⁵ This lesion is commonly unilateral with mushroom appearance and there is no predilection for age or sex and it may develop from 4.8 months to 50 years of age. Clinically, peripheral osteomas are usually asymptomatic, but depending on the location and size of the lesion, it may cause swelling, pain, esthetic disfigurement and functional impairment.⁶

Regarding its radiographic features, the peripheral osteoma often appears as a unique well-defined radiopaque lesion with rounded or oval shape, attached to the cortex by a broad base or a pedicle.^{2,7} Histologically, they can be compact osteoma, cancellous osteoma, and mixed osteoma. The slow growth of osteoma justifies a conservative approach toward an asymptomatic lesion, whereas surgical removal is indicated when the lesion is symptomatic, actively growing, or causing esthetic disfigurement and functional impairment.⁷

In this sense, this article reports an unusual case of osteoma regarding its location, radiographic appearance and patient's age: a case of an 11-year-old male patient presenting a peripheral osteoma on the right retromolar trigone region with unusual radiographic appearance.

CASE REPORT

An 11-year-old male patient was referred to the Diagnosis Clinic of the School of Dentistry - Federal University of Uberlândia for evaluation of a “nodule on the right cheek”. The patient and his mother reported a 1-month evolution of the lesion and pain while chewing during the initial interview. It was observed in the extra-oral examination swelling on the right side of the face causing asymmetry (Figure 1A). During palpation and intraoral inspection, it was noticed a single lesion of hardened consistency, apparently originated from the posterior mandibular region, distally to the lower first molar on the right side. The overlying mucosa was normal in color and appearance but showed occlusion marks of the adjacent teeth (Figure 1B). The patient had no history of previous facial trauma, and his medical history was not contributory.



Figure 1 – Facial asymmetry noticed on the right side of the patient in extra oral examination (A) and intraoral aspect of the lesion presenting occlusion marks (B).

A panoramic radiography was performed for an initial evaluation of the extension and internal characteristics of the lesion. It showed a well-circumscribed and radiopaque mass on the posterior right side of the mandible, superiorly to the lower second molar (Figure 2).

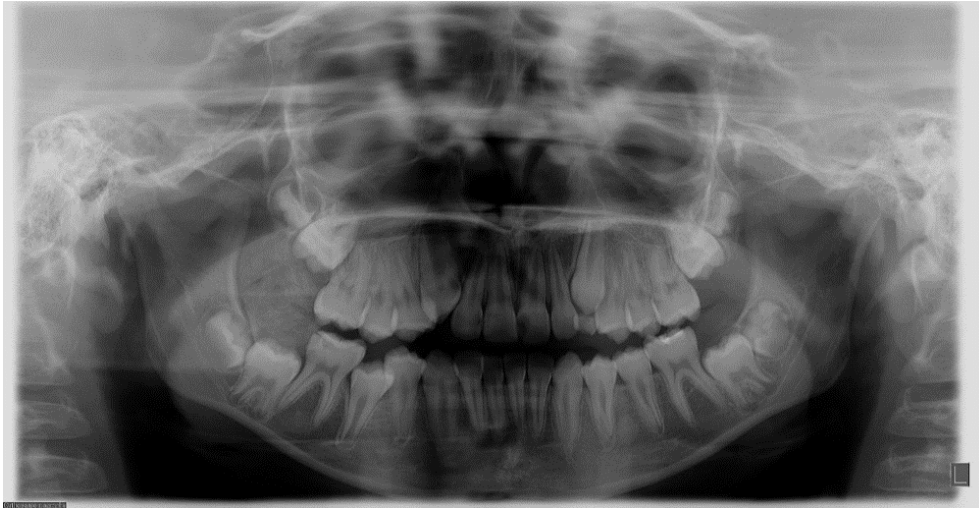


Figure 2 - Panoramic radiography showing radiopaque lesion on the posterior right side of the mandible, superiorly to the lower second molar.

Thus, to have a more detailed and three-dimensional vision of the region, a cone beam computed tomography (CBCT) was acquired. CBCT exam revealed a mixed unilocular lesion with well-defined margins, measuring approximately 30x19x22mm, originated from the retromolar trigone of the right mandibular side, giving it a pedunculated appearance (Figure 3).

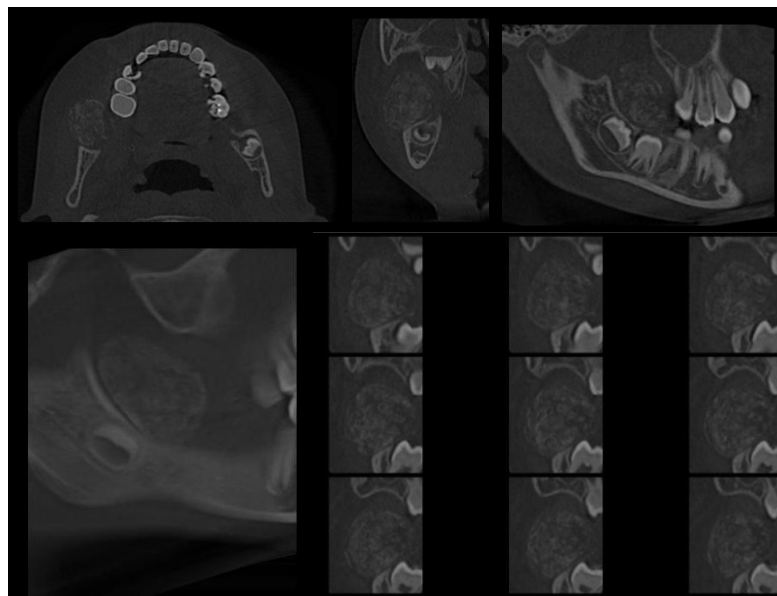


Figure 3 – CBCT showing mixed lesion with pedunculated appearance in the right retromolar trigone region.

As a result of the collected data, peripheral osteoma, peripheral ossifying fibroma and parosteal osteosarcoma were the raised hypotheses. Subsequently, an incisional biopsy was performed, and the collected material was sent to histopathological analysis. It was prescribed Amoxicillin (500mg), Ibuprofen (600mg), Dipyron (500mg) and chlorhexidine digluconate 0.12% and the patient was advised about postoperative care

regarding feeding and cleansing of the surgery area. Previously to biopsy, 1.5mg of Alprazolam was administered.

Microscopic examination of the sample showed that the fragments were predominantly of disordered dense connective tissue, moderately cellular and vascularized, permeated by globular or trabecular areas of mature bone tissue with moderate infiltrate of mononuclear cells (Figure 4). The histopathological aspect observed in the fragments obtained by incisional biopsy could be found both in peripheral osteoma and in peripheral ossifying fibroma.

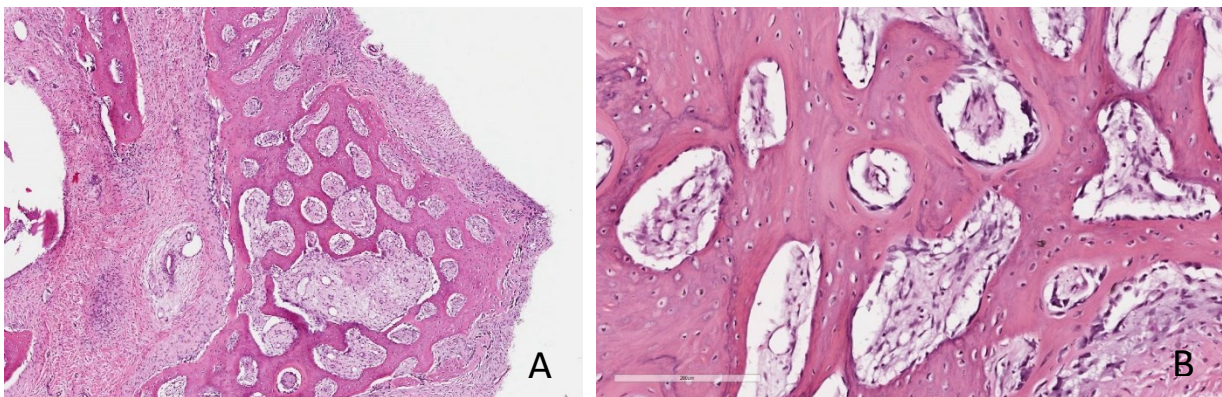


Figure 4 - Microscopic features showing disordered dense connective tissue permeated by trabecular areas of mature bone tissue (haematoxylin-eosin stain, magnification x20 in A and x40 in B).

Since the histopathological analysis revealed benign characteristics of the lesion, it was surgically removed in operating room under general anesthesia and nasotracheal intubation. It was performed intra and extra-oral antiseptics with povidone-iodine 1%. Subsequently, it was infiltrated bupivacaine 0,5 % with adrenaline 1:200.000, diluted in 1:3 proportion with saline solution 0,9%.

For access to the lesion, a linear incision was performed in the central area of the occlusal aspect of the lesion. Then, a blunt dissection led to the apical aspect of the lesion and the exeresis was completed (Figures 5 and 6). A fissure bur was used for bone plasty. The wound was cleansed with saline solution 0,9% and sutured with resorbable mattress (monocryl 4.0). The collected material was stored in 10% formaldehyde and sent to histopathological analysis.

The patient underwent endovenous antibiotic (cefazolin 1000 mg), non-steroidal anti-inflammatory (tenoxicam 20 mg) and analgesic (dipyrone 1000 mg) therapy, all of them based on the patient's weight, while inpatient. Post-operative care was advised and oral rinsing with clorexidine digluconate 0,12% prescribed, such as amoxicillin 500 mg, nimesulide 100 mg and dipyrone 500 mg after hospital discharge, based on the patient weight. The follow-up was every 2 days, for the first week, until complete healing.

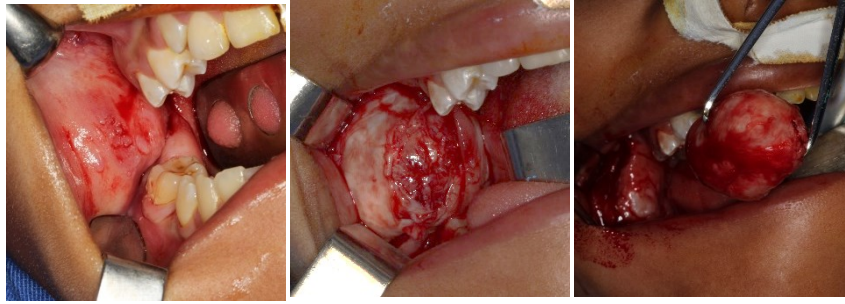


Figure 5 – Surgical access and exeresis of the lesion.

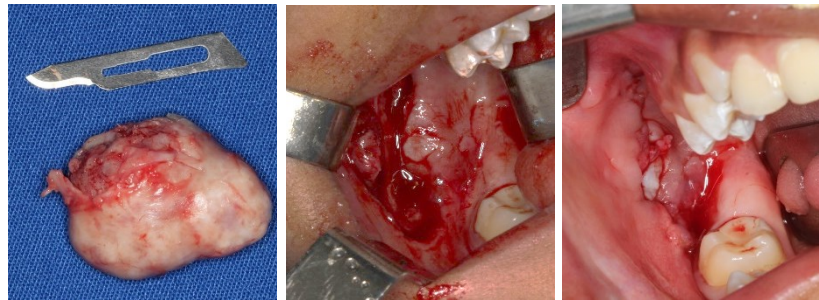


Figure 6 – Excised specimen and final surgical aspect.

After complete surgical resection of the lesion and histopathological analysis of the surgical specimen, it was verified that it presented a clear appearance of normal bone, with spongy architecture and large spaces of fibro-fatty marrow and, to a lesser extent, hematopoietic (Figure 7). There were foci of chronic inflammatory infiltrate, however confined to the surface of the lesion and probably due to trauma during masticatory function, thus discarding the suggestion of a reactive lesion (peripheral ossifying fibroma).

Finally, considering that the lesion was not a simple bone protuberance and that it did not accompanied by the development of the mandible, it was concluded that the pathological condition was compatible with the clinical hypothesis of periosteal osteoma.

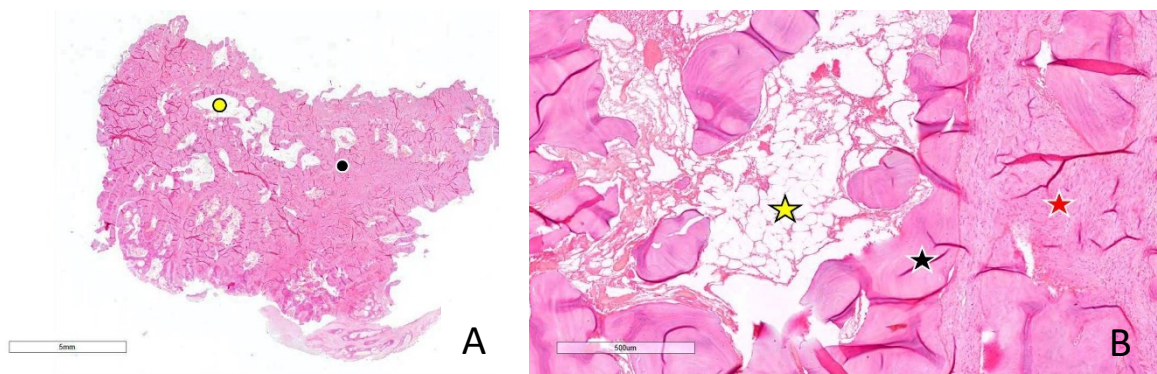


Figure 7 – Photomicrography of the sample obtained by surgical excision showing fragment composed by compact bone tissue, with small spaces of fibro-fatty marrow in A and showing the presence of mature/lamellar bone (black star), fatty marrow (yellow star) and fibrous (red star) in B (Haematoxylin-eosin stain).

The patient remains under strict clinical and radiographic assessments and after 9-month follow up no recurrence or alteration was observed at the surgical site (Figure 8).

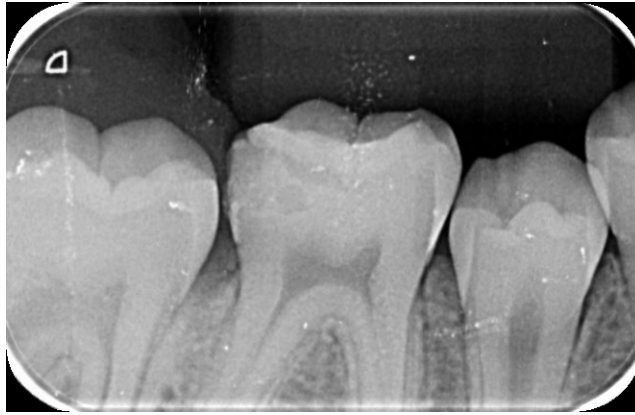


Figure 8 - Periapical radiography 8 months after excision of the lesion

DISCUSSION

According to Chung *et al.* (2015), osteoma is a benign tumor that occurs from the proliferation of cancellous bone, compact bone, or their combination.⁸ Osteomas are slow-growing and asymptomatic lesions; however, when they occur in the paranasal sinuses or lesions of exacerbated growth, symptoms such as facial asymmetry, trismus, buccal opening deviation, malocclusion and swallowing disturbances can be reported.¹

We present a case report of an 11-year-old male patient with a peripheral osteoma in the right retromolar trigone region. According to Chattopadhyay *et al.* (2012), osteomas can be observed at any age, but children are almost never affected unless they have Gardner's syndrome.⁹ Conversely, in the present case, the patient had no systemic alterations or previous facial trauma.

Khandelwal *et al.* (2016) confirm that although a solitary peripheral osteoma of the jaw bones is very rare, the mandible is involved more frequently than the maxilla. When it involves the mandible, the angle and the inferior border of the body of the mandible are more commonly involved.¹⁰ However, the present case reports a solitary peripheral osteoma in the right retromolar trigone region. A region that was not previously reported as site of this lesion in the available literature in English.

The authors, Khandelwal *et al.* (2016) and Agrawal *et al.* (2015), further claim that the lesion grows slowly, and usually appears as an asymptomatic increase in volume and is often discovered by routine radiographic examinations, which differs from the present report since the swelling was noticed after one month when it started to cause facial asymmetry and discomfort while chewing.^{7,10}

According to Agrawal *et al.* (2015), the peripheral osteoma is described radiographically as well-circumscribed radiopaque mass that appears round or ovoid in shape.⁷ These radiographic characteristics were partially observed in the present case, since it also has well defined margins and ovoid appearance. However, the density differs from the literature, instead of a homogenous radiopaque appearance, we present a mixed lesion. The internal density could only be accurately observed in the CBCT exam and lead us to a hypothesis that the lesion was not mature to justify the radiolucent-radiopaque internal aspect.

Initially, it was considered the possibility of case of a peripheral ossifying fibroma, which is one of the differential diagnosis for peripheral osteoma and one of the most common reactive inflammatory hyperplastic lesions of the oral cavity.¹¹ Peripheral ossifying fibromas are benign neoplasms of the jaws, usually slow growing, asymptomatic and present a proliferation of fibrous cell tissue, with varying amounts of bone, cementum or a combination of these. The most common clinical aspect is focal

growth, presenting as elevated ovoid lesions with smooth or ulcerated surface, usually with normal staining of the adjacent mucosa with sessile or pedunculated base and firm consistency.¹² These clinical characteristics are also observed in this case report, one of the reasons for being considered differential diagnosis. Radiographic examination shows that the edges of the lesion are usually well defined, with a thin radiolucent line representing a fibrous capsule. The internal structure has a radiolucent-radiopaque mixed density, with a pattern depending on the shape and quantity of the calcified material present. In tomographic images, a hyperdense mass is found enveloped by a hypodense halo, without invading the cortical bone, differentiating this tumor from the peripheral osteoma and from the present case.¹³

The definitive diagnosis is based on histological exams, where the presence of a proliferation of multinucleated giant cells permeated by cells of the connective tissue, with possible areas of reactional bone or dystrophic calcifications. Nevertheless, the histopathological exam revealed, in the present case, mature bone throughout the examined piece, eliminating the possibility of the diagnosis of peripheral ossifying fibroma.

Another differential diagnosis was the parosteal osteosarcoma, a lobulated nodule attached to the cortical bone which was included kept in mind due to its lack of continuity with the underlying bone. It is reported that parosteal osteosarcomas present thin periosteal radiolucency, known as "string sign", that separates the tumor from the subadjacent cortex in about 30% of the cases.¹³ However, a malignant lesion is composed by poorly differentiated malignant cartilage lobes that may exhibit central calcification, which was not found on the present case, excluding this differential diagnosis due to the eminently benign cellular aspect of the lesion.¹⁴

The treatment chosen in the present case corroborates the options described in the literature. In most cases, invasive treatments are chosen to relieve pressure on neighboring structures, and in our case the surgical excision was performed due to the local discomfort and facial asymmetry. In the literature, surgical intervention is indicated in symptomatic cases and rapidly growing osteomas, two characteristics of the case (Larrea-Oyarbide et al., 2008).¹⁵ The surgical management was also justified once the lesion was in the eruption path of the lower second molar. The patient is being followed up for 9 months without complaints and did not present clinical and radiographic signs of recurrence.

CONCLUSION

Peripheral osteoma is a slow-growing osteogenic neoplasm and its diagnosis can be achieved with radiographic and histological exams.⁴ Even though it has well-known appearance, our report brings awareness that its radiographic characteristics can differ and may appear as mixed lesions and in different regions other than paranasal sinuses and inferior cortex of the mandible.

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