



RARE JAW BONE TUMOR: THE IMPORTANCE OF MULTIDISCIPLINARY MANAGEMENT AND MINIMALLY INVASIVE TREATMENT

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ABSTRACT – Objective: The aim of this study was to present a mini review of oral Cementoblastoma and to report a particular case of this tumor. The Cementoblastoma is a rare benign lesion that represents less than 1-6% of all odontogenic tumors. Cementoblastoma, in the current WHO classification of odontogenic tumors, falls under the category of mesenchymal tumors (WHO 2017) and it is characterized by the proliferation of cementum-like tissue and, in all cases, tends to be associated with an erupting permanent tooth, most often the first molar.

Case Presentation: A 15-year-old female presented a great Cementoblastoma with cortical expansion that affected the left mandibular body, extending from the canine to the first premolar (size 28x24 mm) and involving the mandibular canal. Surgery was performed under general anesthesia with total excision of the lesion, which was then sent for histological analysis. Due to the high risk of nerve injuries and the extreme fragility of the remaining bone after surgery, which required plates and intermaxillary blockage, the surgical site was regenerated with an iliac crest graft together with autologous and synthetic bone.

Results: For the mini-review, 107 articles were found, but only 26 were selected. The patient was monitored for 12 months after surgery; a perfect healing was reported without complications, and she showed no signs of recurrence.

Conclusions: This case report emphasizes the usefulness of clinical choices in a rare case of cementoblastoma of great size in a very young patient. In this condition, finding the most effective eradicated and reconstructive treatment, following the last published protocols, could achieve optimal clinical and psychological patient outcomes.

KEYWORDS: Cementoblastoma, Odontogenic Tumor, Histological analysis, Reconstructive surgery.



INTRODUCTION

The Cementoblastoma was first described by Dewey in 1927 as an odontogenic tumor of mesenchymal origin¹⁻⁴. This lesion is considered to be the only true neoplasm of cementum origin⁵. Cementoblastoma represents less than 1% of all odontogenic tumors; more than 75% occur in the mandible, most often in the molar or premolar region. Cementoblastoma is most common in children and young adults, with 50% occurring before age 20 and 75% occurring before age 30⁶. It is characterized by a large mass of cementum or cementum-like tissue that is attached to the roots of an erupted permanent vital tooth and very rarely to the primary tooth. The symptoms may be totally absent, and when they occur, they usually involve pain and swelling. The lesion can be diagnosed by clinical and radiographic examination, but the final diagnosis is made through histopathological examination⁴. The gold standard treatment remains surgical enucleation of the mass and the extraction of the involved tooth to reduce the risk of recurrence. The aim of this work is to present a mini review on this rare tumor and to document the diagnosis and multidisciplinary treatment of a very large benign cementoblastoma, attached to the roots of the mandibular canine and first premolar, with great expansion of the bone cortex, involving the mandibular canal.

MATERIALS AND METHODS

The purpose of this review was to focus on the main clinical and therapeutic strategies and assess whether the treatment of cementoblastoma should be surgical, conservative, or radical. An extensive search in the electronic databases of PubMed and MEDLINE was performed. The specific keywords searched were “oral” and “cementoblastoma” in various combinations. The inclusion criteria were English articles with available full text; human studies, such as case reports, case series, epidemiological studies, and retrospective studies. The exclusion criteria were non-English articles and animal studies.

RESULTS

A total of 638 articles were found but only 107 articles were found to be relevant and matched the inclusion criteria; finally, a total of 26 were selected for this review after reading the abstract (Figure 1 with flow chart). The main clinical aspects and treatments are summarized in Table 1. Sixteen cases involved males; the mean age was 23.4 years old; 21 cases were in the mandible; 24 cases were treated with surgical removal of the mass and/or tooth extraction; 2 cases have had a recurrence^{4,5,7-30}.

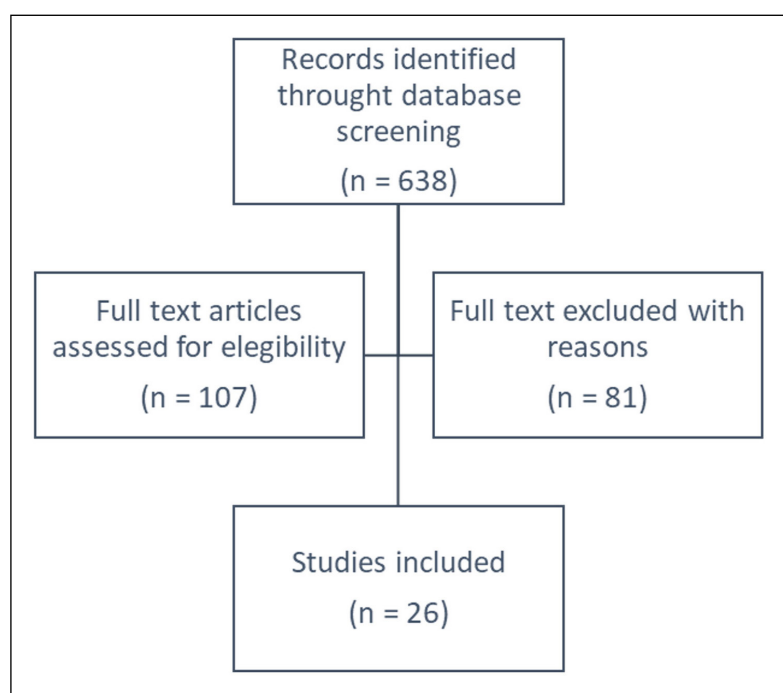


Figure 1. Flow Chart of the selection of the articles for mini-review.

Table 1. Main clinical and treatment aspects of the Oral Cementoblastoma, that synthesize the results obtained.

Authors (Year)	Localization	Sex	Age	Treatment	Recurrence
Huber and Folk ⁷ (2009)	Mandible	Male	18	Mass excision and tooth extraction	No
Sankari and Ramakrishnan ⁸ (2011)	Mandible	Female	42	Tooth extraction	No follow up
Kumar et al ⁹ (2011)	Mandible	Female	55	Mass excision and tooth extraction	No
Feli et al ¹⁰ (2022)	Mandible	Male	32	Root canal therapy, enucleation, curettage, apicectomy	No
Suhasini et al ¹¹ (2020)	Mandible	Male	6	Tooth extraction with mass	No
Yoon et al ¹² (2021)	Mandible	Male	16	Mass excision and teeth extraction	Yes
Nuvvula et al ¹³ (2016)	Mandible	Female	7	Mass excision and tooth extraction	No
Iannaci et al ¹⁴ (2013)	Mandible Bilateral	Male	60	Mass excision and tooth extraction	No
Subramani et al ⁴ (2017)	Mandible	Male	19	Mass excision and teeth extraction	No
Hiremath et al ⁵ (2020)	Mandible	Female	11	Mass excision and tooth extraction	No
Garg et al ¹⁵ (2019)	Mandible	Male	10	Mass excision and tooth extraction	No
Javed et al ¹⁶ (2017)	Upper maxilla	Female	10	Mass excision, teeth extraction and cauterization with carnoye's solution	No
Dadhich and Nilesh ¹⁷ (2015)	Upper maxilla	Female	23	Mass excision and teeth extraction (oral-antral communication)	No
Çalışkan et al ¹⁸ (2016)	Mandible	Female	31	Mass excision and teeth extraction	No
Mohammadi et al ¹⁹ (2018)	Mandible	Male	4,5 (recurrence at 5,5 and 8 years old)	Mass excision and teeth extraction	Yes
Cavalcante et al ²⁰ (2018)	Upper maxilla	Male	22	Mass excision and tooth extraction	No (oral-antral communication)
Pathak et al ²¹ (2019)	Mandible	Male	8	Mass excision and tooth extraction	No
Sumer et al ²² (2006)	Mandible	Male	46	Tooth extraction with mass	No
Pynn et al ²³ (2001)	Mandible	Female	23	Mass excision and tooth extraction	No
Sharma ²⁴ (2014)	Mandible (edentulous area)	Male	16	Mass excision	No
Harada et al ²⁵ (2011)	Upper maxilla	Male	8	Partial maxillectomy	No
Nagvekar et al ²⁶ (2017)	Upper maxilla	Male	12	Mass excision and tooth extraction	No
Qureshi et al ²⁷ (2021)	Mandible	Female	37	Mass excision and teeth extraction	No
Aiyer and Rajagopal ²⁸ (2000)	Mandible	Male	19	Mass excision and teeth extraction	No
Costa et al ²⁹ (2016)	Mandible	Male	18	Root canal therapy, enucleation, curettage, apicectomy	No
Wu et al ³⁰ (2019)	Mandible	Female	55	Mass excision and tooth extraction	No

CASE REPORT

A 15-year-old female was referred to the Maxillofacial Surgery Department of the Santissima Trinità Hospital in Cagliari (Sardinia, Italy), for an asymptomatic lesion in the mandible that was noticed incidentally on an orthopantomographic x-ray (OPT) during a dental examination performed by her dentist (Figure 2).

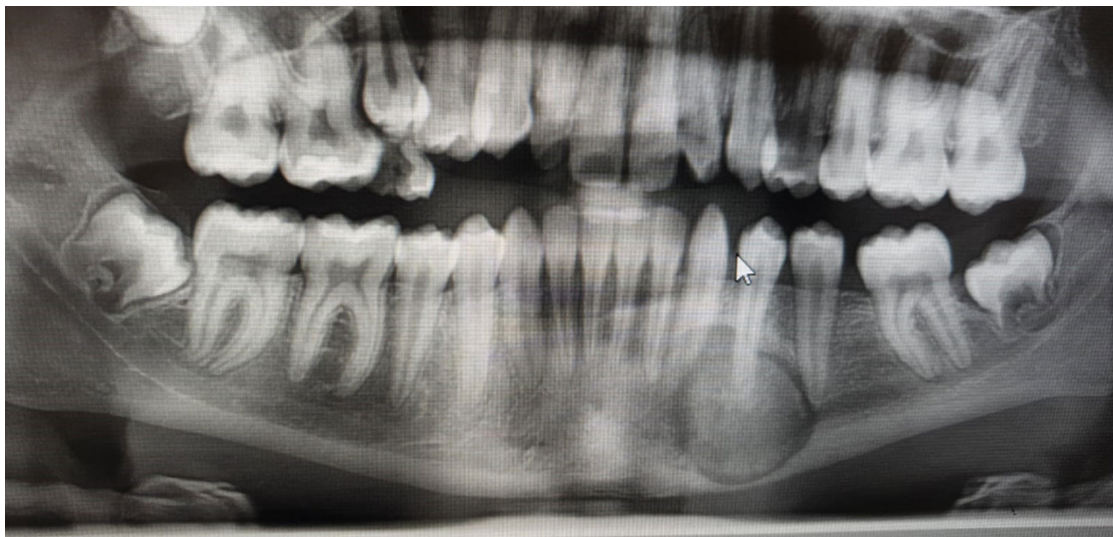


Figure 2. Preoperative panoramic radiograph.

The patient's medical history was unremarkable, and there was no reported history of maxillofacial trauma. Extraoral examination showed asymmetry of the lower third of the face; lymph nodes were not detectable. The clinical examination revealed a blue, non-tender, hard area of swelling on the buccal side of the left mandibular canine area. The overlying mucosa appeared healthy. All the teeth in the left mandibular segment showed a proper response after electric pulp testing. A radiopaque mass that had a close relationship with the root apex of the impacted mandibular left first premolar was detected on the OPT. The mass was surrounded by a thin, radiolucent rim. Cone-beam computed tomography (CBCT) was prescribed for further radiological examination of the mass. A homogenous hyperdense deposit with a size of 28×24 mm was seen in the mentioned area of the CBCT images (Figure 3).



Figure 3. CBCT detail showing the expansion of the lingual and vestibular cortex.

There was also a slight expansion on the mandibular buccal and lingual sides. Significant thinning on the adjacent buccal and lingual cortical bones was detected, but there was no cortical perforation. Involvement of the mandibular canal was suspected. The oblique sagittal images showed that the deposit was continuous with the root apex of the impacted mandibular left first premolar, and there was a well-defined hypodense border between the deposit and surrounding bone. Cementoblastoma, osteoblastoma, odontoma, ameloblastoma, and osteoma were considered in the differential diagnosis. Based on the clinical and radiographic evaluation, a provisional diagnosis of cementoblastoma was made. In the days prior to the surgical operation, impressions and plaster models were taken for making a bi-maxillary fission (IMF) to support the possible fixed containment to be performed with the aim of simplifying bone healing by blocking intermaxillary, in case an iatrogenic fracture occurred during surgery, an aspect to be taken into account given the fragility of the remaining bone structure. A general anesthesia was preferred over a local one. A total Newmann's incision was made whose base was located in the mucogingival line and had two discharge incisions, creating a trapezoidal flap. With the aid of an electrosclerograph, the discharges were performed in a labial direction in the areas of teeth 3.6 and 4.3, with complete exposure of the mandibular buccal bone plate through a full-thickness flap (Figure 4).



Figure 4. Intraoperative photography before excision (Argiolas, L. E. May 2020)

After having identified and safeguarded the mental nerve by positioning a Langenbeck retractor, the mass was sectioned into two parts to facilitate its removal. This section took place with the aid of a piezo-electric device (Piezosurgery®, Mectron, Carasco, Genova, Italy); the two parts had approximately the sizes of 2×2×1.5 cm and 1.9×1.5×1 cm, and the dental elements 3.3 and 3.4 were included. The larger piece was removed together with the resorbed root of tooth 3.4. A part of the mass was subjected to an extemporaneous histological analysis. Intermaxillary fixation (IMF) was performed using six mini-screws, respectively, in positions between 1.2-1.3, 1.1-2.1, 2.4-2.5, 3.5-3.7, 3.1-3.2 and 4.2-4.3.

An autologous iliac crest graft was placed and fixed using two straight miniplates and screws (Medicon®, Budrio, Bologna, Italy). The remaining part of the bone gap was filled with a hemostatic matrix (Flo Seal®, Baxter, IL, USA), as shown in Figure 5. Therefore, a resorbable multifilament polyglactin copolymer that includes an antiseptic agent with bactericidal effect (Vicryl® Plus, Ethicon, Johnson & Johnson, New Brunswick, NJ, USA) A phased removal of the sutures was performed at the start of the seventh day after the intervention, ending the removal after 10–15 days of the surgical operation. During the control appointments, the stitches were removed in the areas of better healing, leaving the points with incomplete healing. The surgical specimen was sent for histopathologic examination in a 10% formalin



Figure 5. Intraoperative photography after excision: autologous iliac crest graft was placed and fixed (Argiolas, L. E. May 2020).

solution; it was sectioned and removed for decalcification. At the radiographical evaluation, the lesion removed appeared as a well-circumscribed, round or ovoid, brownish-colored neoformation around the root of the affected tooth; the texture was hard and chalky⁶. The final diagnosis, although closely related to radiological examinations, always requires a histological examination of the sample. Afterward, serial sections of the entire surgical preparation were taken, previously measured in their maximum dimensions, including both the periphery and the center of the neoformation, and the samples were inserted individually into paraffin blocks. In this case, after mineralization of the lesion, a decalcifying agent had to be used. In observation with an optical microscope, performed in 3 μ m sections stained with haematoxylin eosin, the lesion was characterized by trabeculae of markedly eosinophilic and homogeneous material, similar to cement, with a tendency towards mineralization and concomitant fibrous and vascular tissue (Figure 6).

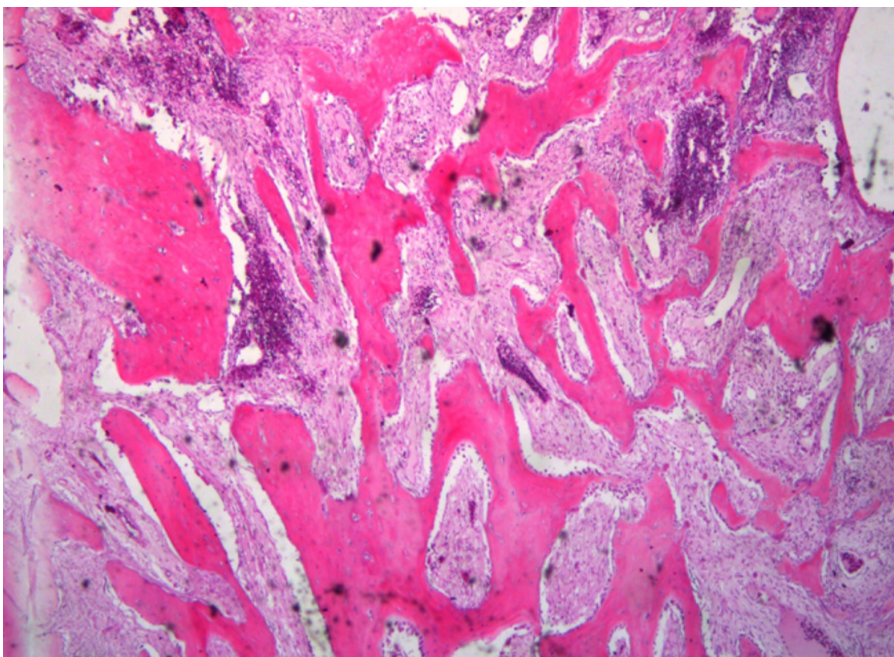


Figure 6. Photo of the histological specimen: hematoxylin-eosin stain, 20x (Gerosa C.)

In the peripheral zones, the trabeculae were arranged in a characteristic radial pattern. Mineralization, well documented by histochemical Goldner staining, was not seen in the younger peripheral areas or the more active growth zones (Figure 7).

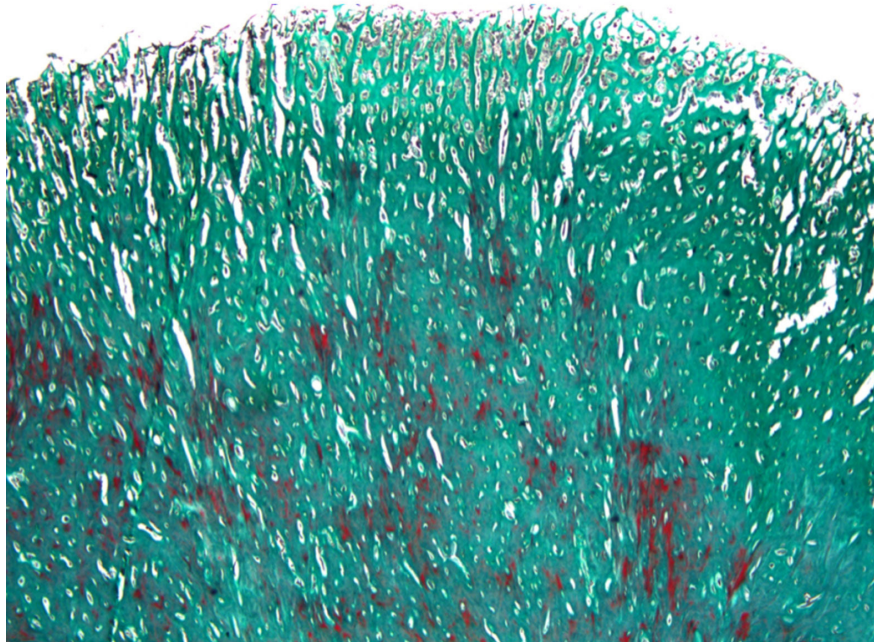


Figure 7. Photo of the histological specimen: Goldner stain, 2.5x (*Gerosa C.*).

The histological diagnosis confirmed the presence of a cementoblastoma. The interposed fibrous tissue contained cementoblasts, isolated or in small groups, distributed in a non-homogeneous way; therefore, it was possible to distinguish hypo-cellular areas or, when more numerous, hyper-cellular areas. Cementoblasts are large and hypertrophic cells, but they are never atypical. The nuclei are round or oval, slightly pleomorphic, with homogeneously distributed chromatin. Occasionally, osteoclastic giant cells may be seen. CD68 immunohistochemical determination highlights giant osteoclastic cells but also a significant number of macrophage cells, not reported in the literature. Mitotic figures are rare, and even if present, in this case in the most active areas, they did not correlate with the prognosis. The KI67 cell proliferation index (MIB1) was low, less than 5% in hot spots, confirming the benign nature of the lesion. Twelve months after surgery, in the follow-up OPT, a complete bone regeneration of the surgical site was observed. It was still necessary to remove one of the osteosynthesis plates, and endodontic treatment was performed on the two teeth adjacent to the area in question, respectively 32 and 35. Informed consent was obtained from the patient to use the data from this observational study for publication.

DISCUSSION

Cementoblastoma is a rare odontogenic tumor, derived from ectomesenchyme^{1,3}, which shows a peak incidence in the second and third decades of life⁶, as demonstrated in the present case in which the patient was 15 years old. There is disagreement in the literature regarding the predominance of gender; some authors reported a predominance of both males and females, while others reported a slight increase in the probability of occurrence in males, which differs from the present case^{3,7,31}. In our mini-review, 16 of the 26 cases are male. Agreement was found between the literature and this case with regard to the most frequent location, in which the mandible (21 cases in our work) is the most affected site compared to less than 25% appearing in the maxilla, particularly in the molar and premolar regions. Teeth generally show vitality^{3,29,31}. Clinically, it is characterized by slow and steady growth. Its dimen-

sions can vary between 0.5 and 5.5 cm in diameter; in the case under examination, a lesion with a diameter of 2.8 cm was found. Due to their slow growth, they are often asymptomatic, as in the reported clinical case^{3,29,31}.

From a radiographic point of view, in agreement with the literature, the following case presented a well-defined area of radiopacity alternating with radiolucent areas circumscribed by a radiolucent halo, closely connected to the root of the tooth³¹. CBCT can be useful to accurately determine the size and extent, location, and relationship of the tumor to the mental foramen and inferior alveolar nerve canal, as well as to assess the degree of root resorption and assist in surgical planning⁶. The characteristics of the CBCT contributed to the diagnosis and planning of the reported case. Compared to the literature, in this case, the lesion was larger and presented marked bone destruction^{2,3,29,31}. Osteoblastoma is the main differential diagnosis of cementoblastoma; however, osteoblastoma and cementoblastoma are essentially identical histologically, and the only distinguishing feature is the attachment of cementoblastoma to the root of a tooth. As it is a rare entity and has similar characteristics to other lesions, it is important to consider other clinical entities, such as hypercementosis condensing complex, complex odontoma, osteoid osteoma, cemento-osseous dysplasia, periapical ossifying fibroma, osteomyelitis, osteoblastoma, florid bone dysplasia, focal sclerosing osteomyelitis, and osteosarcoma³¹.

With regard to treatment, when an early diagnosis is made, it may involve complete excision of the lesion with preservation of the tooth involved, endodontic treatment of the same, and, in some cases, apicectomy³. From the review of the literature performed, it emerged that the gold standard remains the surgical treatment of the lesion with extraction of the element involved; in only two cases, conservative treatment was preferred, involving endodontic treatment, curettage, and apicectomy of the involved tooth^{10,29}. For cases in which a late diagnosis is made and the tumor has already reached important proportions, as in this clinical case, removal of the lesion and associated structures is recommended due to the potential for unlimited growth and eventual recurrence¹. Of the cases analyzed, only two showed recurrence^{12,19}, and one was not monitored⁸.

This type of tumor has an overall recurrence rate of around 12% of cases but exceeds 68% in cases in which the cementoblastoma has caused expansion of the bone cortex, as in the case presented. According to the most recent review on the subject, localization at the mandibular level and the involvement of the dental elements are not factors that predispose to the development of recurrences, a phenomenon that seems more connected with its correct enucleation³¹. In this case, its enucleation was also very complex because it was linked to a high probability of mandibular fracture and nerve damage. The use of minimally invasive surgery through the use of a device such as a piezo scalpel significantly reduces the risk of injury to nervous structures³². However, from our bibliographic research, we have not found any previous work in which the piezoelectric device was used for the enucleation, specifically of a cementoblastoma. However, the use of the Piezo-surgery device for the treatment and removal of maxillary and mandibular lesions has been extensively documented³³⁻³⁵. There are no reports in the literature of cases where the size of the lesion weakened the remaining bone structure to the point of needing the aid of osteosynthesis plates and intermaxillary blocking to prevent iatrogenic fracture of the mandible during removal of the lesion and an autologous graft from the iliac crest to fill in the defect, although the use of iliac crest grafts is well documented in the literature as a treatment for mandibular or maxillary bone deficits after surgery³⁶. Other minimally invasive tools to promote bone growth in the defect are, for example, plasma growth factors^{37,38}.

CONCLUSIONS

A particular case of a great benign cementoblastoma involving the mandibular canal in a 15-year-old female patient was reported. Cementoblastoma is a rare neoplasm; for this reason, it is important to consider similar clinical, radiographic, and microscopic entities as a differential diagnosis. In this clinical case, a lesion with a significant size (2.8 cm in diameter) was found, which caused extensive bone destruction; thus, surgical enucleation of the lesion was the treatment performed in this case, together with the extraction of the teeth involved, in agreement with the literature. The remaining structure may not be enough to remain intact after the removal of the lesion. It was decided, firstly, to fix the bones through osteosynthesis plates and intermaxillary blocking to avoid an iatrogenic fracture of the mandible and then place it in the surgical site with an autologous iliac crest graft removed in the same surgical act, filling the space created with autologous bone and hemostatic matrix. Currently, the patient is being subjected to clinical and radiographic monitoring without a sign of recurrence (Figure 8).



Figure 8. Follow-up panoramic radiograph: twelve months after surgery

AUTHOR'S CONTRIBUTIONS:

A.L.E., V.G. Designed experiments and supervised the research: G.C. Performed the histological analysis: F.D.M., E.S. Analysed data and Formal analysis: C.C., O.G., A.L.E., V.G. Supervised the research and wrote the manuscript. All authors read and approved the final manuscript.

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CONFLICT OF INTEREST:

The authors declare that there is no conflict of interest.

ETHICAL STATEMENT:

In this study, no individual-level personal data was collected. Informed consent was obtained from the patient to use the data from this observational study for publication. In any case, the Helsinki guidelines have been observed and complied with.

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