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CASE REPORT

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Cemento-ossifying fibroma in an 8-year-old patient: a case report

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ABSTRACT

Cemento-ossifying fibroma is a true neoplasm that belongs to the group of benign fibro-osseous lesions. It is characterized by the replacement of bone tissue with fibrous connective tissue that undergoes mineralization, and its diagnosis can be challenging due to its similarity to other lesions. Its origin has been theorized to be odontogenic or from the periodontal ligament, as it can produce cementum-like material within the bone trabeculae. We present the case of an 8-year-old female patient diagnosed with a cemento-ossifying fibroma in the mandible, measuring 50 × 32 mm, who underwent lesion excision along with the dental organs, without recurrence during follow-up. This neoplasm is relatively rare in patients in the third and fourth decades of life and even less common in pediatric patients; therefore, its diagnosis must be carried out comprehensively.

Keywords: cementoma; cement-ossifying fibroma; mandible; case report.

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INTRODUCTION

Cemento-ossifying fibroma is a rare, true benign neoplasm, believed to originate from odontogenic tissue or the periodontal ligament, as it can produce cementum-like material within a fibrous stroma. In some cases, it has been associated with mutations in the HRPT2 tumor suppressor gene, observed in patients with hyperparathyroidism-jaw tumor (HPT-JT) syndrome. This autosomal dominant syndrome is characterized by the presence of adenomas or carcinomas of the parathyroid glands, cemento-ossifying fibromas of the jaws, hamartomas, renal cysts, Wilms tumors, and uterine tumors. It has also been linked to gnathodiaphyseal dysplasia, a condition characterized by fragility of long bones and cortical thickening, caused by mutations in the GDD1 gene (1-3).

Cemento-ossifying fibroma is an uncommon pathology that predominantly affects patients in the third and fourth decades of life, with a higher prevalence in females and a predilection for the mandible, particu-

larly in the premolar–molar region. When located in the maxilla, it most often appears in the canine fossa. This lesion primarily affects individuals of Caucasian descent, followed by those of African descent (1, 2, 4). Clinically, it may cause buccolingual expansion, displacement of the inferior alveolar canal, root displacement, or dental resorption. Radiographically, it appears as a well-defined unilocular radiolucent lesion, which may be mixed or predominantly radiopaque depending on the degree of calcification, with a sclerotic border representing the fibrous interface between the lesion and normal bone (1-3).

Patients are usually asymptomatic, and the lesion is often detected through imaging. Lesion size ranges from 0.2 cm to 15 cm, with larger lesions tending to cause painless swelling and facial asymmetry. Tooth mobility or paresthesia is uncommon (4, 5). Lesions are usually unencapsulated or may present a thin fibrous capsule. Histologically, they consist of cellular fibrous tissue containing mineralized material, including osteoid, bone, or acellular basophilic spherical bodies resembling cementum with striated margins, sometimes surrounded by collagen fibers similar to those of the periodontal ligament. The fibroblastic stroma may exhibit areas of hypercellularity and hyperchromatic nuclei, although mitoses are rare. The presence of osteoblasts at the periphery of the lesion is also uncommon (5-7).

The differential diagnosis includes fibrous dysplasia, which differs in that it presents as a diffuse lesion with a ground-glass appearance. Another diagnosis to consider is Paget's disease, which radiographically manifests as a cotton wool-like lesion with cortical thickening and histologically disorganized bone tissue. Cemento-ossifying fibromas are radiographically indistinguishable from ossifying fibromas, differing only in their histological features (5, 8).

The recommended treatment is enucleation or curettage. Large lesions that cause significant bone destruction may require surgical resection. Radiotherapy is contraindicated because of the lesion's radioresistance and the potential for post-treatment complications. The recurrence rate is approximately 10%, and there is no evidence of malignant transformation (9, 10).

This report presents the case of an 8-year-old female patient diagnosed with cemento-ossifying fibroma—a rare condition in patients under 10 years old— in whom the diagnosis was based on imaging and histopathological studies to rule out other common pediatric pathologies such as fibrous dysplasia or ossifying fibroma. Consequently, a conservative management approach was implemented, considering the patient's age, and no evidence of recurrence was observed. This case report was reviewed and approved by the Ethics Committee of the Mexican Social Security Institute (IMSS, by its acronym in Spanish).

CASE PRESENTATION

An 8-year-old female patient attended the Maxillofacial Surgery Department of the High Specialty Medical Unit, Leon, Guanajuato, Mexico, for the evaluation of a tumor in the mandibular region. The patient's mother reported noticing progressive swelling in the right mandibular region over eight months. Clinically, the patient presented with a firm, non-tender swelling in the right mandibular region, with no changes in the overlying skin. Intraorally, the lesion was in the vestibular region, extending from the midline to the right mandibular first permanent molar; it was firm and slightly erythematous. No dental displacement or mobility was observed.

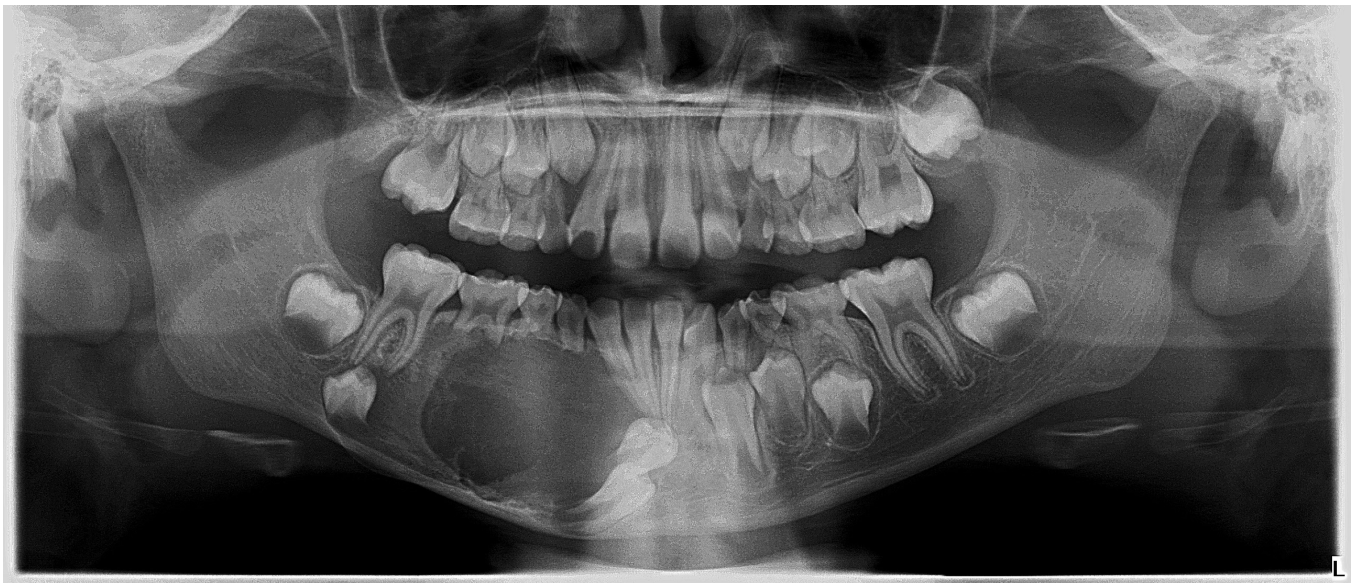

Figure 1. Panoramic radiograph showing a well-defined radiolucent lesion with a sclerotic margin, displaced dental follicles, root resorption of deciduous teeth, and displacement of the roots of permanent teeth.

Radiographically, a well-defined radiolucent lesion measuring approximately 5 x 3 cm was observed, with sclerotic margins and caudal displacement of the dental follicles of the permanent right canine, first and second lower premolars. Additionally, there was evidence of root resorption in deciduous teeth, displacement of lower incisor roots, and thinning of the cortical plates (Figure 1).

Non-contrast computed tomography of the facial skeleton revealed an osteolytic lesion measuring 48 × 32 × 37 mm, with generalized cortical thinning and displacement of the permanent tooth germs toward the basal border (Figure 2).

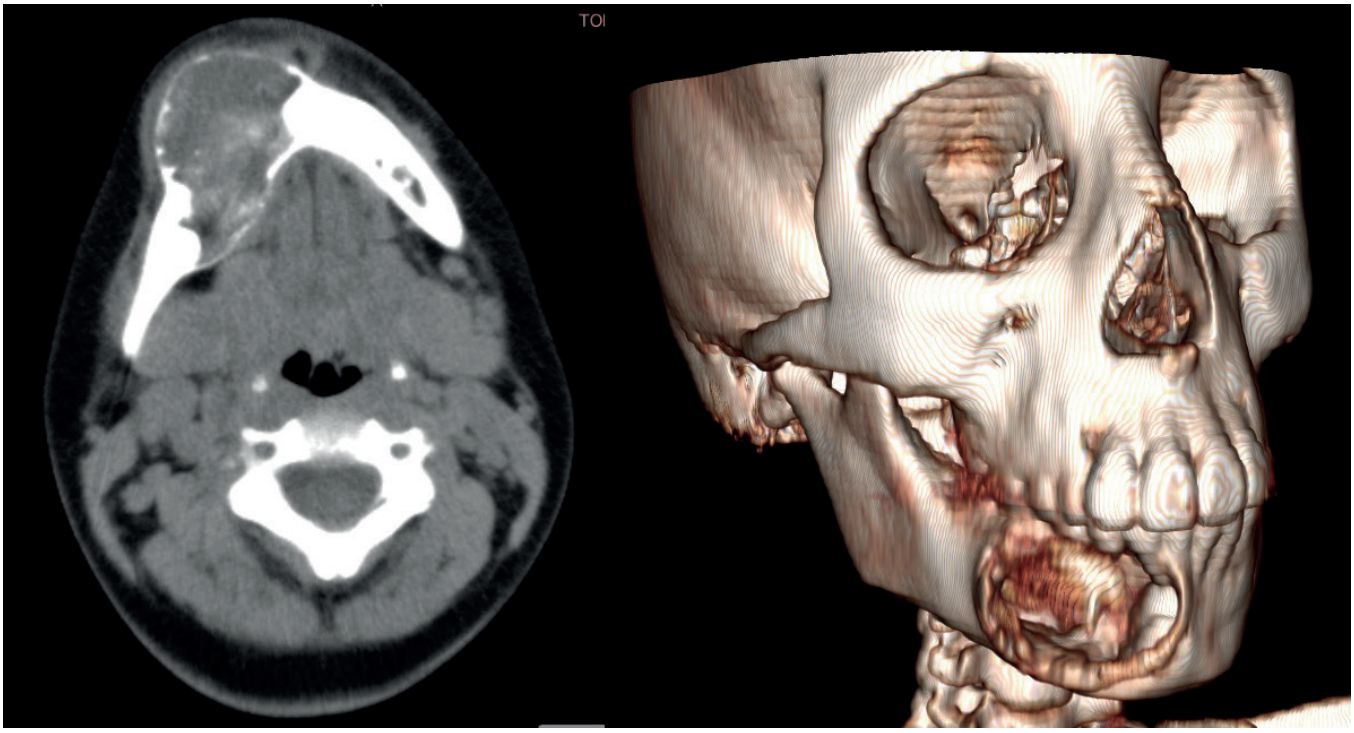

Figure 2. Non-contrast computed tomography of the facial skeleton showing an osteolytic lesion measuring 48 × 32 × 37 mm with cortical thinning.

After obtaining informed consent from the patient's mother, an incisional biopsy was performed under local anesthesia, yielding soft, brown-colored tissue. The sample was sent to the pathology department and stained with hematoxylin and eosin. The study described a benign fibro-osseous lesion characterized by abundant irregularly shaped and interconnected osteoid trabeculae, as well as spherules of acellular cement-like material, findings consistent with cemento-ossifying fibroma (Figure 3).

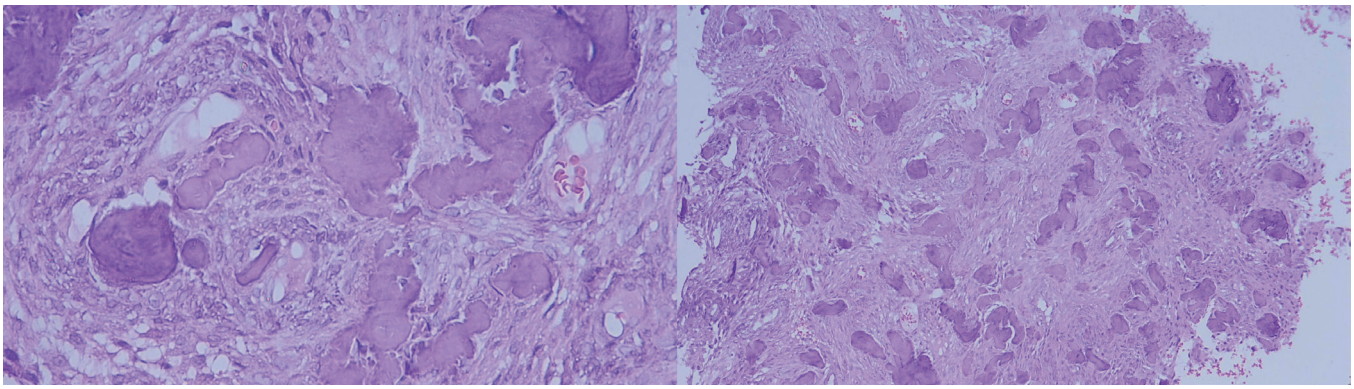

Figure 3. Histological slide showing abundant irregular, interconnected osteoid trabeculae and spherules of acellular cement-like material.

Enucleation of the lesion was performed under balanced general anesthesia, using a scalloped incision extending from the left mandibular incisor to the right first molar. Following the elevation of a mucoperiosteal flap, the well-defined lesion was exposed (Figure 4).

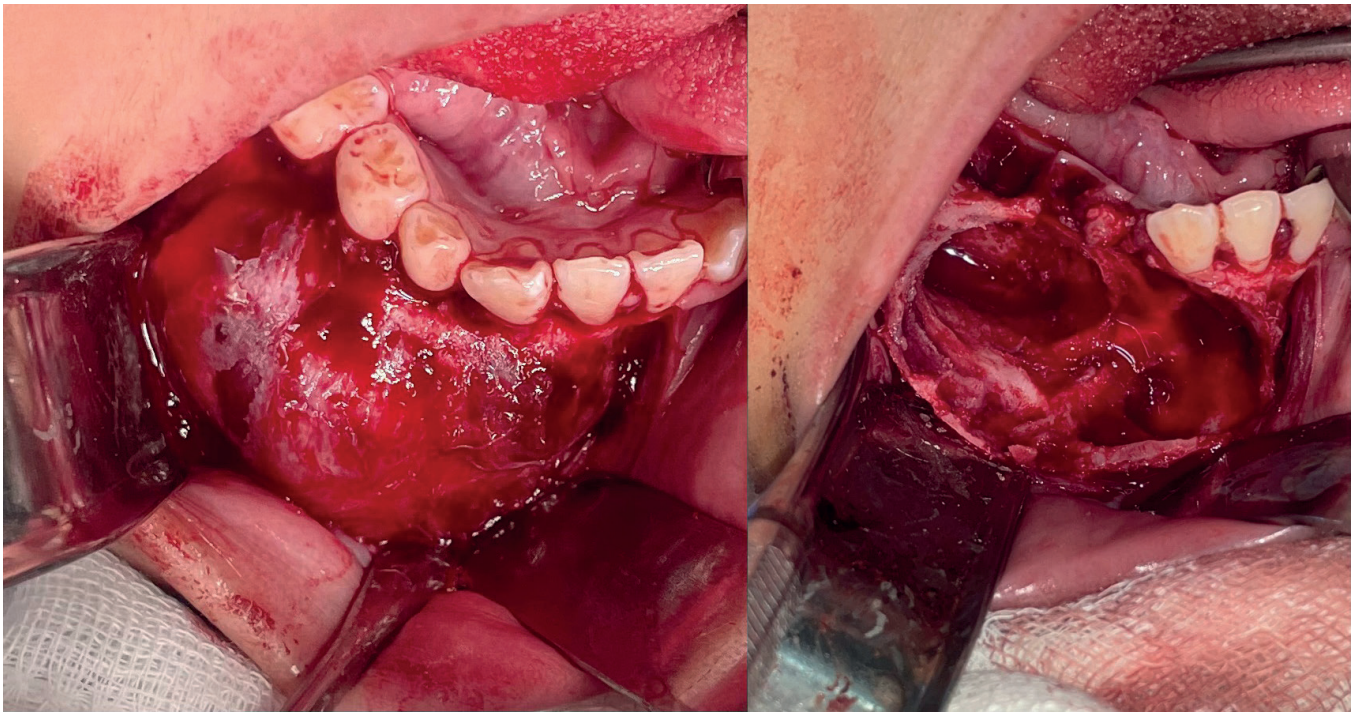

Figure 4. Enucleation of the well-defined lesion.

During enucleation, expansion of the cortical bones, displacement of the inferior alveolar nerve, and downward displacement of the teeth toward the basal edge were observed, without evidence of cortical erosion. The temporary teeth were then extracted, as well as the tooth follicles of the canine, first and second lower premolars. Finally, curettage of the cavity was performed, and the mucosa was closed with 3-0 polyglactin (Figure 5). The specimen was sent for examination, and the diagnosis of cemento-ossifying fibroma was confirmed.

The patient was followed up for eight months with panoramic radiographs, which demonstrated new bone formation at the surgical site and repositioning of the lower incisor roots (Figure 6). A follow-up non-contrast CT scan of the facial skeleton showed no evidence of recurrence.

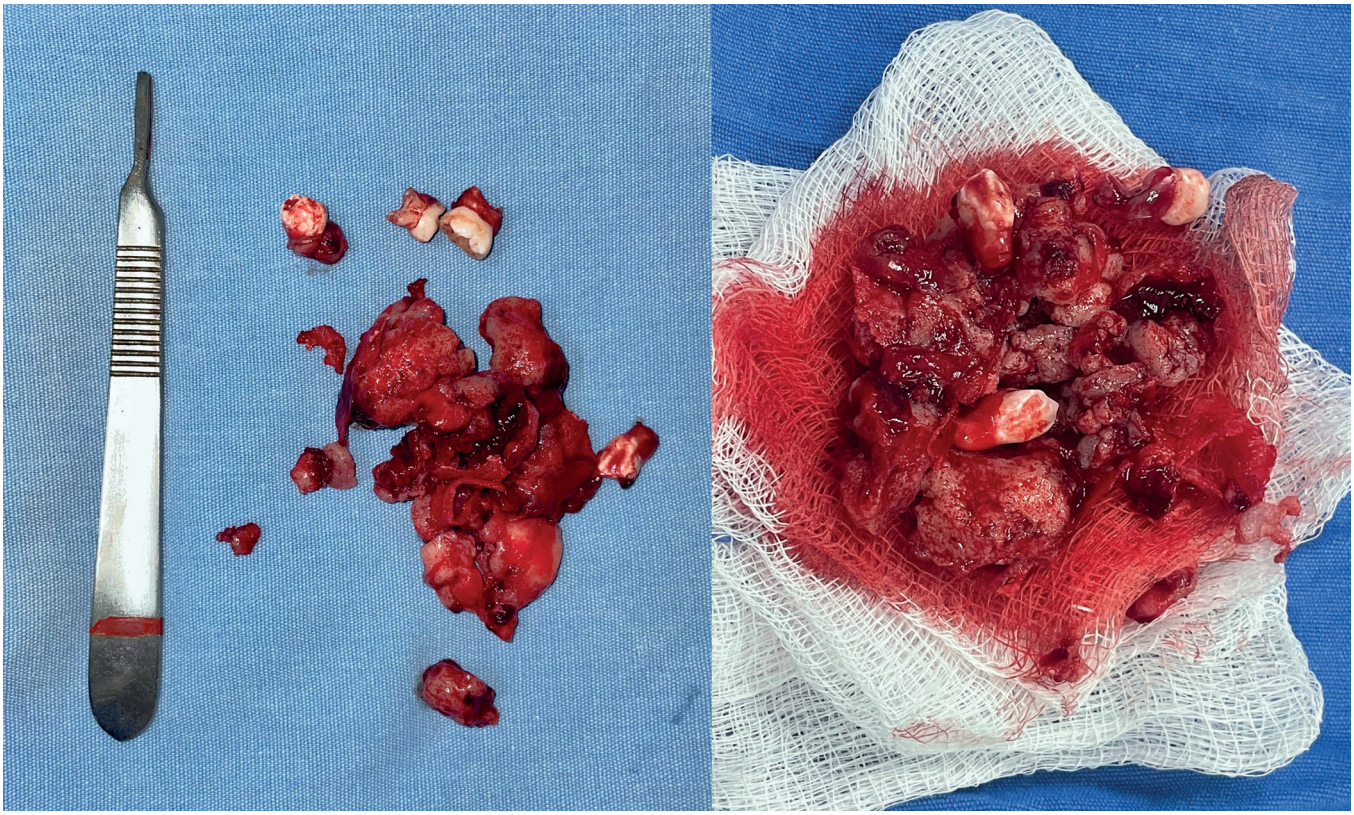

Figure 5. Specimen obtained with follicles and temporary teeth.

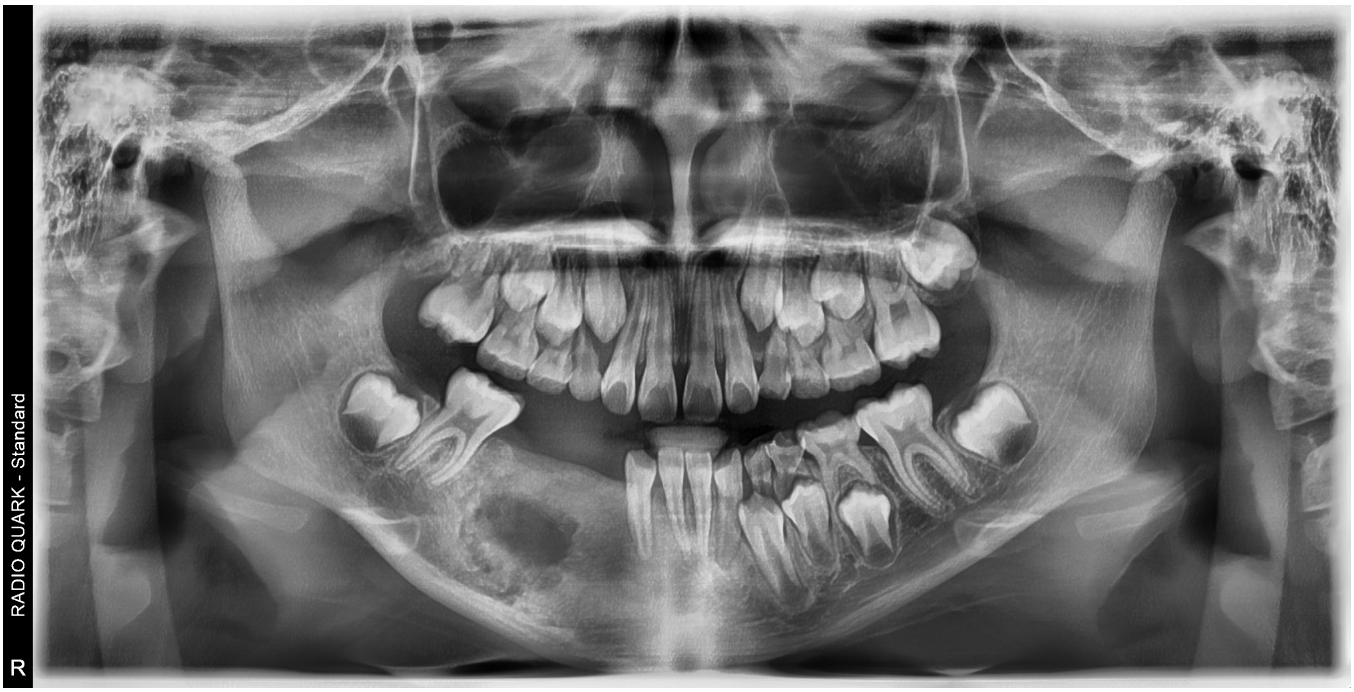

Figure 6. Panoramic radiograph obtained five months after lesion enucleation showing bone regeneration and repositioning of the lower incisor roots.

DISCUSSION

Cemento-ossifying fibroma is a benign neoplasm that typically presents as a slow-growing lesion, most commonly located in the mandibular premolar-molar region. This lesion is rare, accounting for 0.1% of all odontogenic tumors. A more aggressive variant has been reported in patients under 15 years of age, predominantly affecting the maxilla, and is characterized by rapid growth and a higher recurrence rate ranging from 30% to 58% (4, 10, 11).

Although this lesion is typically reported in patients in their third and fourth decades of life, we report a case of an 8-year-old female with a lesion in the mandibular deciduous molar and permanent incisor region, crossing the midline and causing root resorption of deciduous teeth, displacement of permanent teeth, and displacement of the dental follicles of the permanent canine and premolars (11-13).

The diagnosis of this lesion at such an early age can be challenging, as very few cases have been reported in the literature. Instead, psammomatoid, trabecular, and conventional ossifying fibromas are more commonly observed in the 0–10-year age range (9, 14, 15).

According to Contreras-Aedo et al. (3), between 2015 and 2020, no cases of cemento-ossifying fibroma were identified in patients aged 0-20. Two cases were documented in the 21-30-year group, one in the 31-40-year group, and three in the 41-50-year group. Clinically, lesion size ranges from 4.1 to 8 cm in 50% of reported cases, followed by lesions smaller than 4 cm in 33.3% of cases. In the present case, the size of the lesion differed between panoramic radiography and computed tomography, with a radiolucent lesion measuring approximately 5 x 3 cm observed on the two-dimensional radiograph; however, computed tomography revealed a lesion measuring 48 x 32 x 37 mm. This highlights the limitations of two-dimensional radiographs, such as distortion, and underscores the need for three-dimensional radiologic evaluation (3).

Ultimately, the definitive diagnosis of this pathology should be confirmed by histopathologic analysis, which demonstrates cementum-like material within the fibrocellular connective tissue. The recommended treatment for cemento-ossifying fibroma is surgical excision with curettage, with a reported recurrence rate of approximately 10%. Therefore, complete excision of the lesion was performed, including displaced dental structures. During the eight-month postoperative follow-up, radiographic evaluation demonstrated appropriate bone remodeling and repositioning of the displaced permanent incisors, with no evidence of recurrence (16).

The main limitations during the patient's treatment were the lack of institutional resources, which delayed her diagnosis, and the absence of oral and maxillofacial pathology specialists, thereby making it necessary to seek a private facility to analyze the specimen.

CONCLUSIONS

Cemento-ossifying fibroma is a neoplasm capable of producing cementum-like material, leading to buccolingual expansion and root displacement or resorption. Radiographically, it appears as a well-defined unilocular radiolucent, mixed, or radiopaque lesion with a sclerotic border. This lesion accounts for 0.1% of all odontogenic tumors and most commonly occurs in the third and fourth decades of life. Its occurrence in patients under 10 years of age is rare and may be misdiagnosed as other lesions, such as fibrous dysplasia, Paget's disease, or ossifying fibroma.

Conflict of interest:
The authors declare no conflict of interest.

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Ethics approval:
This report was reviewed and approved by the Ethics Committee of the Mexican Social Security Institute (IMSS). In addition, the principles of bioethics were approved by means of informed consent signed by the patient's mother.

Authorship contribution:
KLMM: conceptualization, research, methodology, writing of original draft.
YJTC: research, supervision, validation, writing – review & editing.
JARC: resources, supervision, writing – review & editing.

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