

Recurrent benign cementoblastoma: A case report and literature review

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ABSTRACT

A 16-year-old male presented with pain in the right posterior mandible on chewing that had lasted for several months. The radiographic features of the lesion included a radiolucent-radiopaque mixed-density mass with a radiolucent rim attached to the root of the mandibular right first molar. The preliminary radiographic diagnosis was benign cementoblastoma, which was confirmed by histopathological examination following surgical excision. The lesion recurred 3 years after treatment; radiographically, it consisted of 3 round foci with mixed radiopacity, each with a radiolucent rim near the root of the mandibular right second premolar and the edentulous postoperative region. The lesion was diagnosed as recurrent benign cementoblastoma and a second surgery was scheduled. This report presented an unusual case of recurrent benign cementoblastoma following surgical excision and extraction of the involved tooth, along with a literature review on reported cases of recurrent benign cementoblastoma with a focus on its clinical features and the best treatment options. (*Imaging Sci Dent 2021; 51: 447-54*)

KEY WORDS: Odontogenic Tumor; Dental Cementum; Recurrence; Cone-Beam Computed Tomography; Radiography, Panoramic

Introduction

The 2005 World Health Organization classification review recognized benign cementoblastoma as a cementoma.¹ In the latest classification, benign cementoblastoma is classified as a benign neoplasm originating from the mesenchyme and/or the odontogenic ectomesenchyme.¹ Although its etiology remains unknown, it is clear that the lesion is derived from mesenchymal tissue.¹ It is a relatively rare

lesion, accounting for 0.69-8% of all odontogenic tumors.²

Benign cementoblastoma tends to be slightly more common in males, with a male-to-female ratio of 1.2 : 1.^{3,4} Approximately 75% of benign cementoblastomas occur in young patients under 30 years of age,⁵ with most cases arising under 20 years.³ Nearly 40% of all benign cementoblastomas are associated with erupted mandibular molars.⁶ A few reported cases are associated with fully or partially impacted teeth.⁵ However, no cases have been reported in association with deciduous dentition.³ Benign cementoblastomas are usually asymptomatic; in symptomatic cases, pain and swelling are the common chief complaints.⁵ Even if a patient has a history of pain, the involved tooth is generally vital and shows no sign of pathosis.⁷ The diagnosis is rarely established until significant bone expansion has occurred.⁷

Because cementoblastoma has unlimited growth potential, treatment includes tumor resection with the extraction of the associated tooth.⁷ The lesions are well-demarcated

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and easily excised from the surrounding bone, often shell- ing out with the tooth attached.³ If the tumor is small at the time of diagnosis, treatment consists of surgical removal with endodontic therapy and retention of the involved tooth.⁷ Patients treated with complete surgical excision show an excellent prognosis with a low risk of recurrence.⁸ Radiographic images are useful for detecting the extent of the lesion and recurrence.⁶ The purpose of this study was to present a case of a benign cementoblastoma in a 16-year old male with unusual recurrence around the site of the previous lesion. The literature on recurrent benign cemen- toblastoma was also reviewed.

Case Report

A 16-year-old male visited the Department of Oral and Maxillofacial Surgery of Kyungpook National University Dental Hospital with pain in the right mandible while

chewing. The patient sensed pain on palpation of the gin- giva near the mandibular right first molar. All the teeth in the affected area were caries-free. The overlying mucosa appeared normal, with no clinical signs of inflammation. The patient reported that the symptoms had begun several months ago. The patient had no significant family or medical history contributing to pain.

An initial panoramic radiograph (Orthopantomograph OP 200 D, Instrumentarium Imaging, Tuusula, Finland) showed a round radiopaque mass with mixed radiodensity surrounded by a radiolucent rim. The lesion was attached to the root of the right first molar and caused displacement of the inferior alveolar canal (Fig. 1). The lesion extended to the periapical area of the mandibular right second molar. Cone-beam computed tomography (CBCT) images (PaX- Flex 3D apparatus, Vatech, Kyungi, Korea) demonstrated a well-circumscribed mass involving the roots of the man- dibular right first molar, causing perforation of lingual cor-

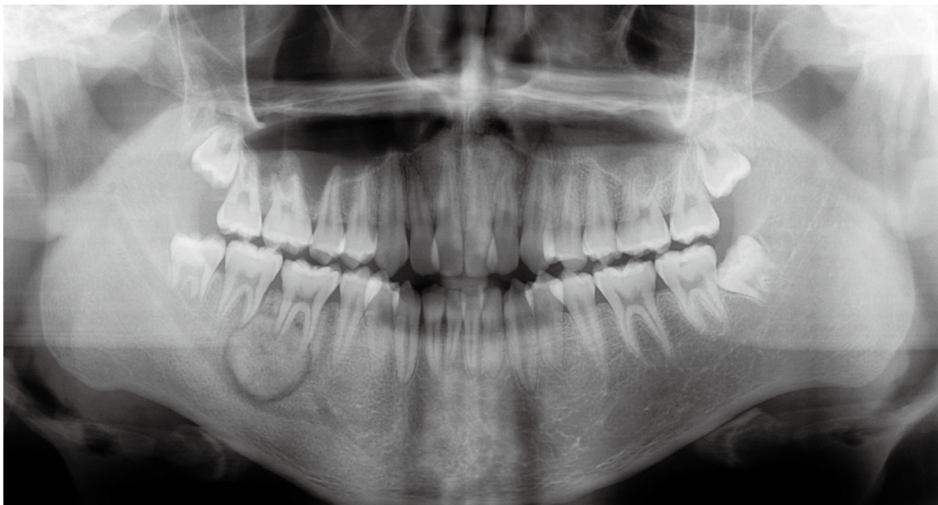


Fig. 1. Panoramic radiograph shows a mixed radiopaque mass with a distinct radiolucent rim attached to the root of the mandibular right first molar.



Fig. 2. Cone-beam computed tomographic images present a mixed-density radiopacity with a radiolucent rim confluent with the root of the mandibular right first molar on axial (A), coronal (B), and panoramic reconstruction (C) images. The lesion perforates the lingual cortical plate with accompanying surrounding sclerosis.

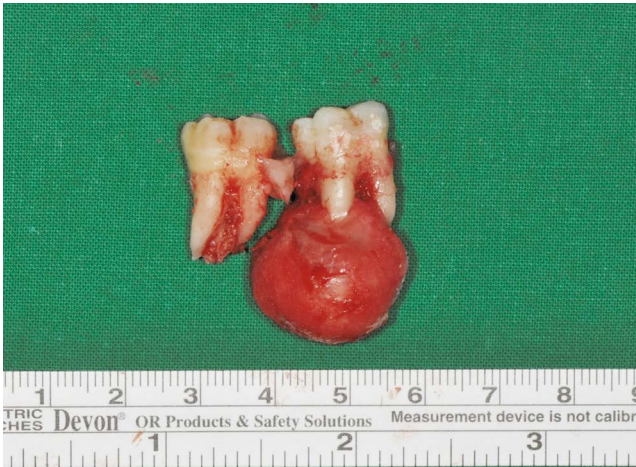


Fig. 3. Surgical specimen along with the affected tooth.

tical bone with sclerosing osteitis around the lesion (Fig. 2). We established a differential diagnosis of benign cementoblastoma with secondary infection based on the clinical and radiographic findings. Under general anesthesia, the patient underwent surgical excision of the lesion with extraction of the mandibular right first and second molars (Fig. 3). During surgery, the mass and the involved tooth were well separated. The defect was immediately filled with bone graft material (Fig. 4) and the specimen was submitted for a histopathological examination. Macroscopically, the surgical specimen consisted of a round hard tissue mass in conjunction with the first molar roots. Hematoxylin and eosin-stained sections showed that the lesion included sheets of cementum-like tissue with basophilic reversal lines surrounded by well-vascularized cellular connective tissue,

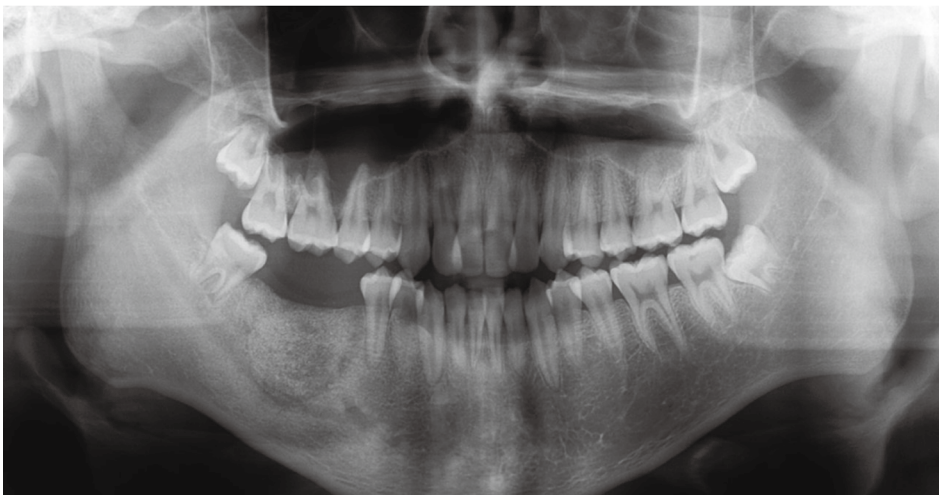


Fig. 4. Postoperative panoramic radiograph reveals the surgical site filled with bone graft material.

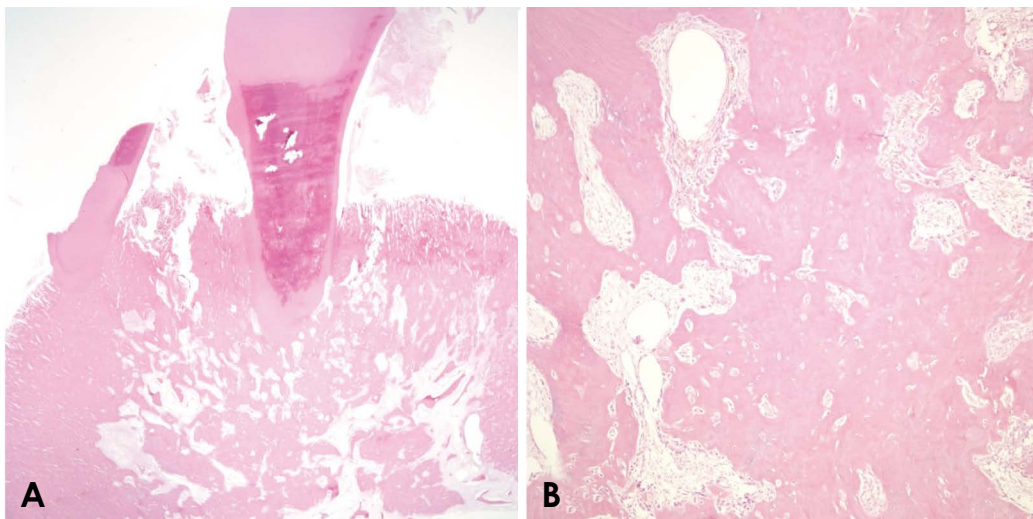


Fig. 5. A. Histopathologic examination demonstrates the margins of the original tumor with radiating cementum-like material attached to the root, exhibiting prominent reversal lines (H&E stain, original magnification $\times 12.5$). B. Sheets of cementum-like material are entrapped within the vascular connective tissue stroma under high-power magnification (H&E stain, original magnification $\times 100$).

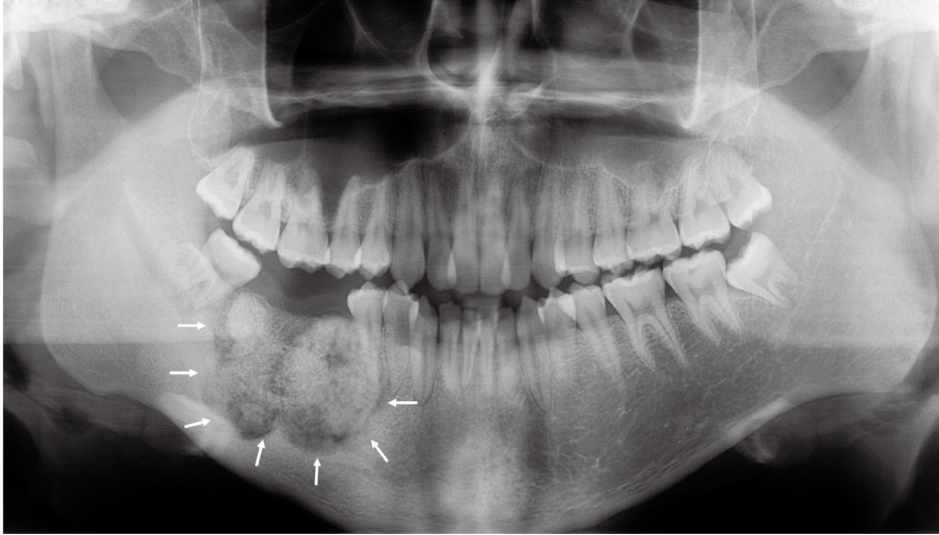


Fig. 6. Panoramic radiograph reveals a mixed-density lesion on the edentulous postoperative region attached to the root of the mandibular right second premolar 3 years later (white arrows).

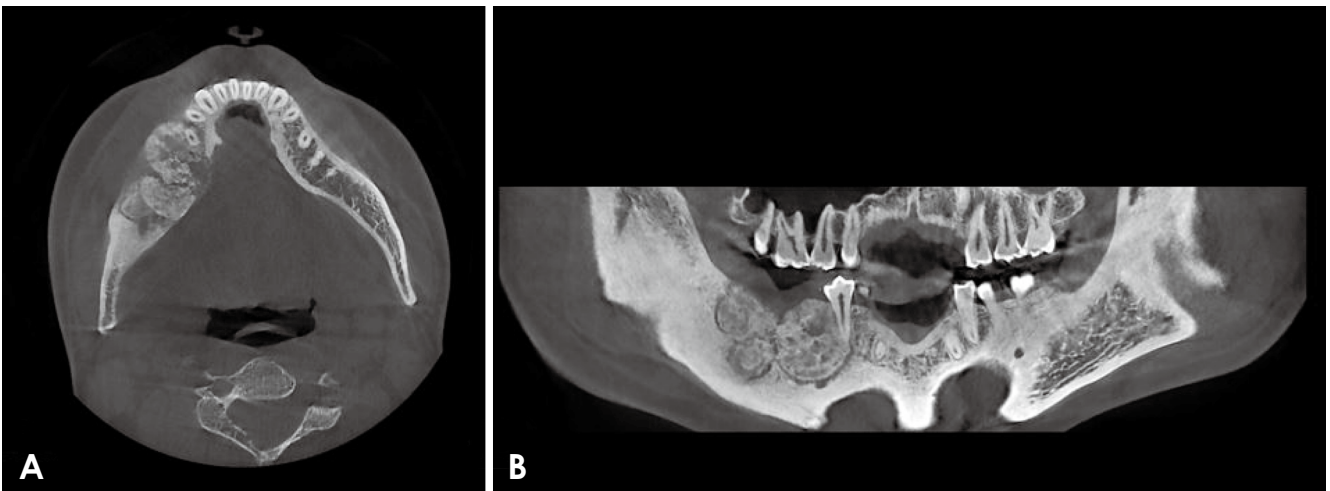


Fig. 7. Axial (B) and panoramic (C) CBCT images show a mass with 3 centers causing expansion and perforation of the lingual and buccal cortical plates.

with large and abundant cementoblasts and irregular lacunae (Fig. 5). The final diagnosis of benign cementoblastoma was established based on the histopathologic examination. Postoperative healing was uneventful.

Routine post-surgical follow-up examinations revealed a new lesion at the age of 20 years on the previous operation site. The clinical symptoms included intermittent pain in the right mandible during physical activity. Three years after the initial surgery, a panoramic radiograph showed masses with heterogeneous radiopacity surrounded by radiolucent rims with sclerotic borders at the previous operation site, suggesting recurrence of the lesion (Fig. 6). CBCT showed perforation and expansion of the bucco-lingual cortical bone (Fig. 7). The patient underwent a re-



Fig. 8. Surgical specimen consists of fragments of the lesion and the extracted mandibular right second premolar.

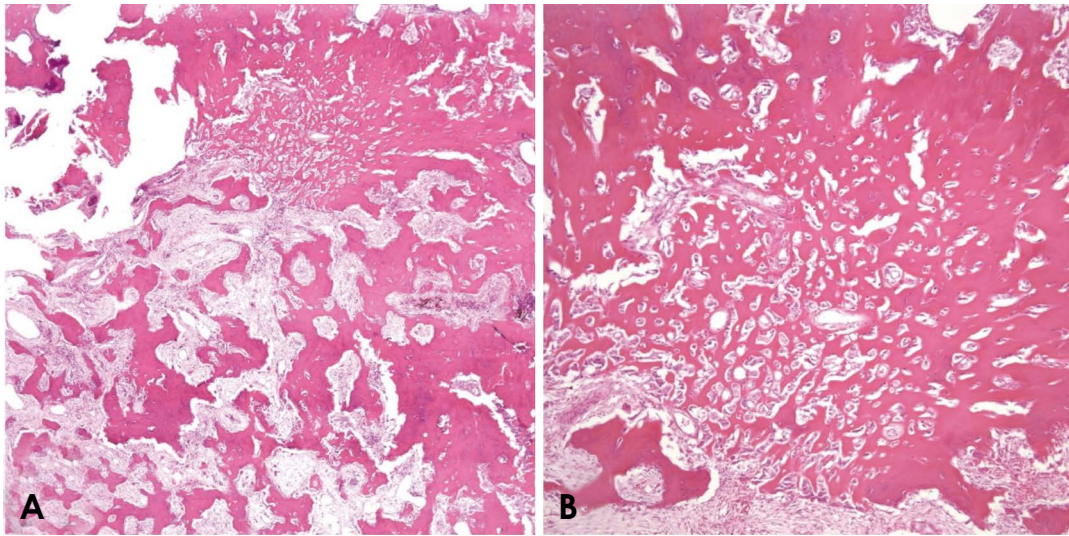


Fig. 9. A. Microscopic examination of the recurrent mass shows sheets of trabeculae of cementum-like material entrapped within a loose vascular connective tissue stroma (H&E stain, original magnification $\times 40$). B. Numerous plump cementoblasts and multinucleated giant cells are seen (H&E stain, original magnification $\times 100$).



Fig. 10. Postoperative panoramic radiograph shows the surgical site with a left iliac bone graft.

operation on the mandibular right posterior region. Under general anesthesia, the patient underwent surgical curettage of the lesion, mandibular right second premolar extraction, and grafting with the left iliac bone. The gross specimen included multiple cemental masses near the operation site (Fig. 8). Areas of cementum-like calcified trabecular tissue with benign cementoblastoma and multinucleated giant cells were present along the loosely arranged vascular connective tissue stroma (Fig. 9). Histopathological examination confirmed the diagnosis of recurrent benign cementoblastoma. Six months after the second surgery, there was no further sign of recurrence at a regular follow-up visit (Fig. 10).

Discussion

Benign cementoblastoma has a low tendency to recur if completely excised. This report describes a case of recurrent benign cementoblastoma after surgical treatment and the results of a literature search for reported cases of recurrent cementoblastoma. After an extensive search, the authors found only 20 reported cases of recurrent benign cementoblastoma involving permanent teeth (Table 1). Brannon et al.³ reported 15 cases of recurrent benign cementoblastoma, and we identified 5 additional case reports (19 case reports in English and 1 in Korean). All case reports met the clinical, radiological, and histological eligibility criteria

for confirming the diagnosis of recurrent benign cementoblastoma. According to the literature review, the age of the patients ranged from 8 to 24 years. The included articles documented 12 affected males and 8 females, yielding a male-to-female ratio of 1.5 : 1. Of the 20 cases with recurrence, 15 were present in the mandible (most often in the posterior area) and 5 in the maxilla. The diameters of the tumors ranged from 1.5 cm to 4.5 cm. Gradually increasing pain in the jaw was the patient's chief complaint in the majority of the reports. The initial treatment for all cases was surgical removal with the extraction of the associated tooth. Clinical and radiographic examination usually showed cortical expansion or perforation of the affected bone, indicating that these features strongly correlate with recurrence of benign cementoblastoma.

Radiographically, benign cementoblastoma is a cemental mass involving 1 or more roots of the permanent tooth. The lesions typically manifest as a well-defined, circumscribed mass fused to the roots, resulting in root resorption, loss of the root outline, and obliteration of the periodontal ligament space.^{3,5} In the case presented herein, the lesion involving the tooth presented as a mass with a distinct radiolucent rim with buccolingual cortical thinning and expansion. The radiopacity showed a thin radiolucent band around the benign cementoblastoma that was more distinctive and uniform than in other lesions such as cemental dysplasia.³ Benign cementoblastoma develops as a circular radiolucent mass at the root of a vital tooth and gradually becomes radiopaque with a radiolucent rim.⁸ As the lesion matures, the differential diagnosis should include odontoma, osteoblastoma, cemental dysplasia, hypercementosis, and sclerosing osteitis, which can be distinguished using the following criteria.^{3,9-11} Odontoma is not attached to the root and typically shows a tooth-like and heterogeneous density with a follicular space.¹⁰ Osteoblastoma does not show tooth-mass continuity and has a more irregular pattern of radiopacity than benign cementoblastoma.³ Due to osteolytic changes, the majority of the patients with osteoblastoma present with persistent pain.³ Benign cementoblastoma has a unique growth pattern in which the overall shape is more uniform and circular than in hypercementosis, which usually shows irregular undulating outlines.¹¹ Hypercementosis is a small lesion surrounded by a thin periodontal membrane space with no signs of root resorption, pain, or jaw expansion.¹¹ Sclerosing osteitis is a well-defined radiopaque lesion that typically appears without any peripheral radiolucent rim, and is therefore easily differentiated from cases of benign cementoblastoma that usually show a distinct rim.⁹ These salient features are crucial for distinguishing benign cemen-

Table 1. Literature review of recurrent benign cementoblastoma

Author	Age (years)/Sex	Involved teeth	Size (cm)	Initial surgical procedure	Clinical and radiographic features	Follow-up (years)
*Brannon et al. ³ (2002)	8-24 (mean: 17.9)/ M:F = 9:6	5 in maxilla, 10 in mandible	1.5-4.5 (mean 2.6)	Surgical excision with extraction (10/15), Surgical curettage with extraction (4/15), Not specified (1/15)	Expansion/swelling (13/15), Cortical perforation (7/15)	0.3-19 (mean: 4.4)
Zaitoun et al. ¹⁶ (2007)	10/F	Right mandibular second molar	2.5	Surgical excision with extraction	Cortical expansion and perforation	0.5
Srikanth et al. ¹⁷ (2009)	22/M	Left mandibular first molar	1	Surgical curettage with extraction	Buccal expansion	1.75
Jung et al. ⁴ (2009)	20/F	Left mandibular first molar	3	Surgical excision with extraction	Buccolingual cortical expansion and perforation	2
Ahmad et al. ¹⁸ (2014)	14/M	Right mandibular first molar	2	Surgical excision with extraction	Lingual cortical expansion	0.6
Mohammad et al. ⁸ (2017)	4.5/M	Right mandibular first molar	1.6	Surgical curettage with extraction	Buccolingual cortical expansion	4

toblastoma from other lesions.

Benign cementoblastoma exhibits slow but unlimited growth potential and is usually asymptomatic.⁷ The associated tooth is vital, and the patient rarely notices the lesion until the appearance of obvious pain, swelling, tenderness, and expansion of the jaw.⁷ Although the lesion is painless in most cases, pain and paresthesia are possible in some cases with substantial bone expansion, where the lesion exerts pressure on the inferior alveolar canal.⁵ In the case presented herein, the patient complained of pain in the right mandible while chewing during the first visit and intermittent pain during exercise upon recurrence of the lesion.

The lesions are encapsulated and easily excised from the surrounding bone, with a low risk of recurrence.⁸ In this case, recurrence was observed within 36 months of the initial surgery, and the patient is under further follow-up. Although the prognosis of benign cementoblastoma after complete surgical excision is excellent, this case is a rare example of rare recurrence, highlighting the importance of follow-up to monitor patients' clinical course. Some authors have advocated for the retention of the affected tooth.^{9,12,13} For small lesions or those where an early diagnosis is made, conservative treatment is achieved by minimal resection and preservation of the affected tooth following apicoectomy and endodontic treatment.^{9,12,13} If a late diagnosis is made, complete removal of the lesion and considerable proportions of the associated structures is necessary.¹³ The majority of the reports have suggested that complete removal of the lesion and the affected tooth is crucial, and an incomplete removal (such as curettage alone) posed a high risk of recurrence for patients.^{3,12,14,15} The decision to remove or maintain the affected tooth remains ambiguous and should be made by the oral surgeon. However, the recommended treatment of benign cementoblastomas consists of surgical removal of the lesion with the tooth/teeth and/or structures affected, followed by a complete curettage of the area or the peripheral osteotomy of the entire region.¹⁵

The recurrence rate of benign cementoblastoma has been reported to be 11.8%, 21.7%, and 37%, indicating considerable variation in the literature.^{3,6} Recurrence usually takes place within 6 months to 1 year after the initial surgery.¹⁴

Cortical expansion and perforation have been found to be related to the recurrence of benign cementoblastoma.^{3,6} The present case and all other recurrent cases in the literature showed cortical expansion and perforation of the adjacent bone. The original lesion in the present case was treated with surgical excision. A previous study found that recurrence only took place in lesions that were treated by excision.⁶ Therefore, surgical curettage could be helpful in pre-

venting recurrence.

In conclusion, this report presents a case of recurrent benign cementoblastoma after surgical excision including the involved tooth. Based on the current case and literature review, recurrence is related to the presence of cortical bone expansion or perforation adjacent to the benign cementoblastoma. Periodic follow-up is recommended in cases of benign cementoblastoma with cortical expansion and perforation.

Conflicts of Interest: None

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