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**Case report / Приказ болесника**

Bojan Gačić<sup>1</sup>, Branislav Ilić<sup>1,†</sup>, Radojica Dražić<sup>1</sup>, Aleksandra Čairović<sup>2</sup>, Jelena Sopta<sup>3</sup>,  
Ljubica Simić<sup>3</sup>

**Cementoblastoma: An unusual radiographic presentation**

**Цементобластом: Необична радиографска манифестација**

<sup>1</sup>University of Belgrade, School of Dental Medicine, Clinic of Oral Surgery, Belgrade, Serbia;

<sup>2</sup>University of Belgrade, School of Dental Medicine, Clinic of Dental Prosthetic, Belgrade, Serbia;

<sup>3</sup>University of Belgrade, Faculty of Medicine, Department of Pathology, Belgrade, Serbia

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**†Correspondence to:**

Branislav ILIĆ

Clinic of Oral Surgery

Doktora Subotića 4, 11000 Belgrade, Serbia

E-mail: [Branislav.ilic@stomf.bg.ac.rs](mailto:Branislav.ilic@stomf.bg.ac.rs)

## Cementoblastoma: An unusual radiographic presentation

### Цементобластом: Необична радиографска манифестација

#### SUMMARY

**Introduction** Cementoblastoma is an uncommon tumor of the jaws that originate from odontogenic ectomesenchyme, characterized by proliferating cementum-like tissue.

**Case Outline** We present the case of a cementoblastoma in the mandible with atypical radiographic image: no well-defined borders and no radiolucent rim. Apart of that, taking into account data from the literature review, different clinicopathological, and radiographic presentations of tumors and lesions that may resemble cementoblastoma are discussed.

**Conclusion** Cementoblastoma must be removed as soon as possible, together with the associated tooth. Recurrence rate is a relevant phenomenon and is estimated to 11.8%, so the long-term follow-up is mandatory.

**Keywords:** cementoblastoma, odontogenic tumours, maxillofacial tumours

#### САЖЕТАК

**Увод** Цементобластом је тумор виличних костију који води порекло од одонтогеног ектомезенхима, а карактерише га пролиферишуће цементу-слично ткиво.

**Приказ болесника** У раду смо приказали цементобластом доње вилице, атипичне радиографске манифестације: без јасно дефинисане границе и без зоне периферног расветљења. Прегледом доступне литературе, евалуирали смо различите туморе/лезије које клиничко-патолошки или радиолошки могу личити на цементобластом.

**Закључак** Цементобластом захтева што ранији хируршки третман при чему је потребно уклонити и захваћени зуб. Рецидиви су релативно чести (око 11.8%) па су због тога неопходне дугорочне контроле пацијента.

**Кључне речи:** Цементобластом, Одонтогени тумори, Тумори максилофацијалне регије

## INTRODUCTION

Cementoblastoma was first documented by Dewey in 1927 [1]. Cementoblastoma is an uncommon tumor of the jaws that originates from the odontogenic ectomesenchyme, characterized by proliferating cementum-like tissue. It represents only 1–6.2% of all odontogenic tumors. The World Health Organization classified benign cementoblastoma and cementifying fibroma as the only true neoplasms [2, 3, 4]. The growth potential of the tumor is unlimited and there are several of the cases reporting the aggressive behavior of the cementoblastoma. Typical radiographic presentation of cementoblastoma is well-defined oval radiopacity with a thin radiolucent periphery.

## CASE REPORT

A 19-year-old female without contributory medical history was complaining about the pain in the mandible molar area. Intraoral examination revealed a large cavity in the distal part of the first lower left molar. The pulp vitality test was negative. The radiographic examination showed a highly radiopaque mass attached between the mesial and distal roots.

The mass was oval (15×20 mm), was positioned toward the base of the lower jaw, and was causing the resorption of the mesial root. Both retroalveolar and panoramic X-rays gave the impression that the mass was fused to the surrounding bone, without clear borders (Figure 1).

Clinical symptoms and findings implied to a chronic pulpal infection. On the other hand, radiological presentations of the lesion suggested to several differentials:

hypercementosis, cemento-osseous dysplasia, condensing osteitis, idiopathic osteosclerosis, cementoblastoma, odontoma, osteoblastoma, fibrous dysplasia. In order to get more precise information concerning the lesion, a cone beam computer tomography was performed. The scans confirmed unclear borders of radiopaque mass that was pushing down the mandibular canal to the base of the lower jaw (Figure 2).

A provisional diagnosis of chronic low-grade infection was made and it was decided to perform a root canal treatment at first. The patient gave her informed consent. Although the endodontic treatment relived the pain, the patient was anxious about the unknown mass inside the bone and the biopsy was scheduled. The bony specimen taken during the biopsy was fixed in 4% buffered formalin and together with the X-rays sent for histopathology (Figure 3).

Histopathology examination revealed that the tumor was composed of the sheets of dens, irregular lamelated and cementum like tissue. Cementum like structures with broad trabeculae were presented as well as sheets of irregularly placed tumor cells within lacunae. Cementoblasts were plump with moderate amount of cytoplasm, hyperchromatic nuclei, but no mitotic activity. Although many authors describe the presence of osteoclast like giant cells, in our case giant cells were not seen. Diagnosis of cementoblastoma was made (Figure 4).

Surgical removal of the tumor, along with the involving tooth and peripheral osteotomy were performed. Preservation of the lower mandibular nerve was obtained.

Postoperative period was uneventful and complete patient recovery was accomplished. Three years follow-up acknowledged the absence of the tumour (Figure 5).

## DISCUSSION

Cementoblastoma, classified as odontogenic ectomesenchymal tumor, arises mostly in the permanent dentition with several incidences reported in primary or unerupted teeth [5–9]. Slow growing mass of cementum or cementum-like tissue is usually located in the posterior area of lower jaw (80%), and associated with permanent first molar. The tumor generally occurs among young population and has equal sex distribution [10, 11, 12]. Associated tooth is usually vital and if the pathological changes of tooth are presented they are coincidental [13]. Cementoblastoma has a pathognomonic radiographic appearance as a well-defined solitary ovoid radiopacity with a thin radiolucent periphery. The tumor is frequently fused to partly resorbed root/roots of the associated tooth [14, 15]. In the case when associated tooth was extracted prior to diagnosis of cementoblastoma, patient pre-extraction X-rays are of great importance.[16] In our case, the resorption of the adjacent root was present, there were no bony expansion and characteristic radiographic appearance was missing. Cone beam computed tomography showed that tumorous mass was more radiopaque than surrounding bone but there were no clear borders and radiolucent rim.

There are several differentials that should be considered: hypercementosis, focal cement osseous dysplasia, condensing osteitis, idiopathic osteosclerosis, odontoma, osteoblastoma, osteoid osteoma and fibrous dysplasia (Table 1).

Hypercementosis is a non-neoplastic condition in which excessive cementum is deposited in continuation with regular radicular cementum. It is widely accepted as an age-related phenomenon involving mostly the older population. Premolars are the most affected teeth, bilateral involvement is not uncommon and is usually presented without clinical

symptoms. Apart from the idiopathic nature of hypercementosis, this condition is associated with several local, more commonly periapical pathosis, or systemic factors. Radiographically, hypercementosis is an occasional finding. The radiolucent shadow of the periodontal membrane and the radiopaque lamina dura are always seen as the outer border of hypercementosis [17].

Cementoosseous dysplasia is reactive or dysplastic process. Clinically is usually asymptomatic and appears in the apical region of vital teeth as frequent coincidental X-ray finding [18].

Condensing osteitis is characterized by presence of a low grade, chronic, dental inflammatory stimulus of the adjacent tooth. Radiographically is seen as localized bony sclerotic area associated to the apex of the tooth but without radiolucent halo [19]. In addition to this, calcifications in condensing osteitis represent necrotic irregularly mineralized bone, contrary to cementum calcifications in cementoblastoma. Therapy is primarily focused to endodontic treatment of the involved tooth.

Idiopathic osteosclerosis is similar to condensing osteitis but without tooth involvement. The cause is unknown, usually affects younger population and the therapy is not required. Radiographical finding is the same as focal sclerosing osteomyelitis but the sclerotic area is not connected to the adjacent teeth [20].

Odontoma is odontogenic tumor composed of various dental tissues. It is slow growing, non-aggressive, true neoplasm found usually in younger population. Usually, odontoma is asymptomatic or can cause delayed teeth eruption. Radiographically is easy to differentiate to cementoblastoma since odontoma is not fused to the adjacent tooth and has tooth shape structure [21].

Osteoblastoma is benign bone forming tumor. It is very similar to cementoblastoma but with few differences. Instead of cementoblasts and cementoclasts, it is characterized by

woven bone production and proliferation of numerous plump activated osteoblasts, many osteoclast and fibro-vascular stroma. Clinically, there is evident night pain that cannot be relieved by salicylate intake. Radiographical finding is the same as cementoblastoma. The degree of opacification on the X-ray correlates to the amount of calcification, but the lesion is not attached to the tooth [22].

Osteoid osteoma is similar to osteoblastoma but with reduced growing potential and sclerotic surrounding bone. Usually, it does not exceed 10 mm in diameter and is not related to the teeth [22].

Fibrous dysplasia is a rare non-neoplastic fibro-osseous lesion of cranial bones. Fibroblastic proliferation with irregular shaped trabeculae, and no osteoblastic rimming are histological criteria for diagnosis. It usually involves younger population and is asymptomatic until causes facial asymmetry, enlargement etc. Radiographical finding shows typical “ground-glass” appearance and the absence of lamina dura [23, 24].

Histologically, cementoblastoma is composed of broad trabeculae of sparsely cellular cementum merged with areas of cemental islands in vascular stroma. The peripheral zone shows radiating columns of cementum running perpendicular to the surface of the lesion [15]. Microscopic specimen of our case had the same characteristics as previously mentioned. Resembling microscopical image can be found in osteoid osteoma, osteoblastoma, and osteosarcoma. Major difference of osteosarcoma is the presence of atypical mesenchymal cells and sharp circumscription with no permeation of surrounding bone [17].

Recent studies involving the expression of cementum protein (CEMP-1) could help better understanding of cementoblastoma. CEMP-1 has been isolated from human cementoblastoma and is considered as a specific marker of cementoblasts, periodontal progenitor cells and mineralization process. The expression of CEMP-1 was positive in

subpopulation of cementoblasts and mineralized tissues. It could help identify and standardize tumoral lesions and it should be considered as a useful diagnostic tool [25].

As seen in our case and from literature data, clinical manifestations of cementoblastoma may vary. In this case, there was not radiolucent rim around tumor, although the aggressive nature of tumor was demonstrated by root resorption. Radiographic aspects of cementoblastoma are correlated with the amount of calcification. Immature lesions are usually radiolucent and with the maturation, radiopacity increases [15].

Histopathologically, cementum is similar to bone and cementoblastoma may be easily misinterpreted as different pathology. That's why the diagnosis cannot be made on examination of the biopsy specimen alone. The pathologist may misdiagnose such lesions if the clinical and radiographic findings are not considered [15].

The treatment of choice is surgical extirpation on tumour. Cementoblastomas must be removed as soon as possible, together with the associated tooth. Recurrence rate is a relevant phenomenon and is estimated to 11.8% [10]. Appropriate treatment should consist of surgical removal of the lesion with the affected tooth, followed by thorough curettage or peripheral osteotomy. Sometimes, en block resection is not sufficient and marginal or even segmental resection of the jaw is required [26]. In our case, tumour was fused to the surrounding bone so additional peripheral osteotomy was necessary. Luckily, the tumour didn't cause bone expansion or cortical bone perforation associated with the higher recurrence rates.[10] Nevertheless, long-term follow-up of the patient is mandatory.

**Conflict of interest:** None declared.

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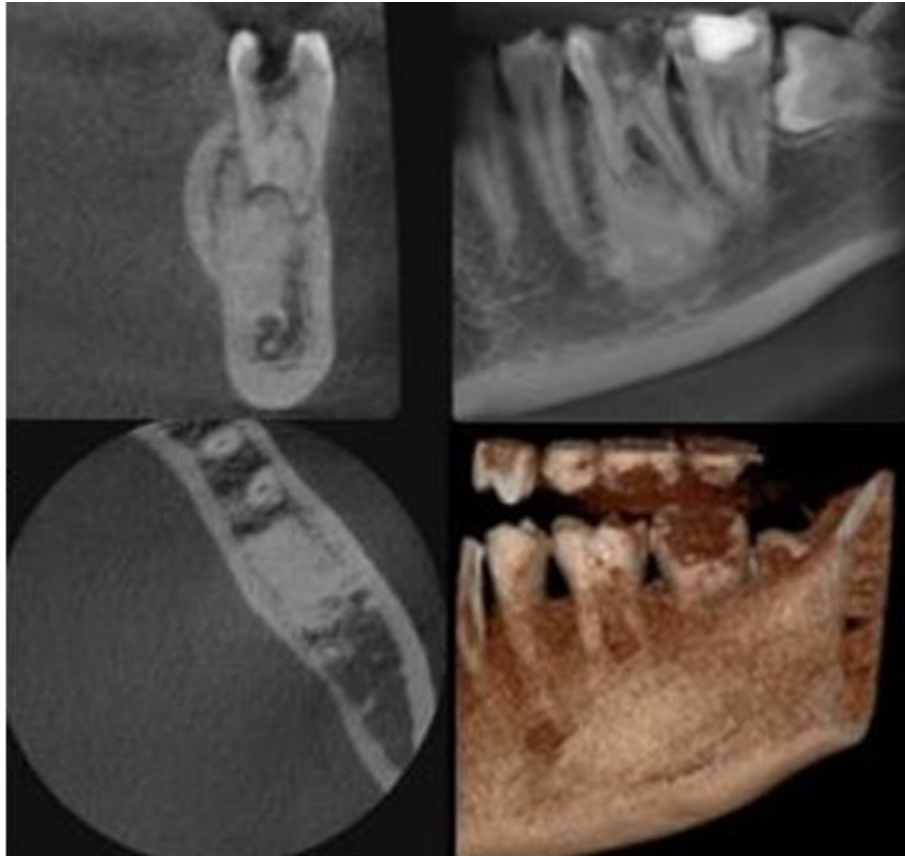
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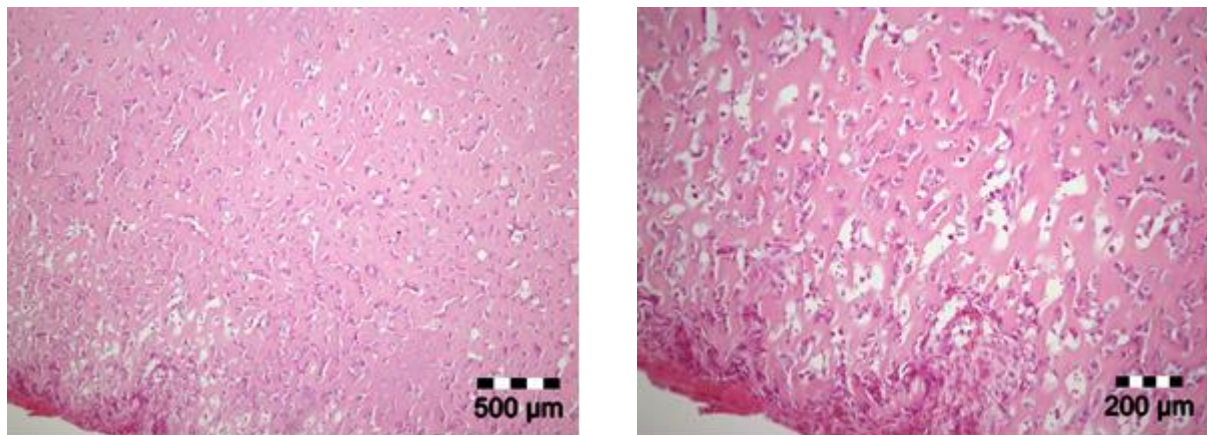
**Figure 1.** Retroalveolar and panoramic radiography: highly radiopaque mass is attached between the roots of tooth number 36



**Figure 2.** Cone beam computed tomography scans: unclear border of radiopaque mass is pushing down mandibular canal to the base of the lower jaw and causing the resorption of the mesial root



**Figure 3.** Intraoperative insight in biopsy: It was very difficult to identify tumour and its borders. The biopsy is performed according to preoperative radiography planning



**Figure 4a and 4b.** Histologic findings: a – tumor consists cementum-like tissue (HE, 10×); b – prominent cementoblasts and trabeculae of uncalcified cemental matrix perpendicular to the surface (HE, 20×)



**Figure 5.** Follow-up radiography: There are no signs of tumor recurrence

**Table 1.** Clinical, radiographic, and histopathological features of radiopaque lesions of the jaws

Lesions	Age / Sex	Clinical	Tooth involvement	Radiographic	Histopathology
<b>Hypercementosis</b>	Both / over 40 years old	No symptoms; mandibular premolar area;	Yes (vital, no root resorption)	Well-defined radiopacity with radiolucent halo	Cellular/acellular cementum
<b>Condensing osteitis</b>	Both / younger population	Discrete or no symptoms; Dental inflammatory stimulus with chronic pulpal involvement; mandibular jaw; no root resorption;	Yes (non-vital, no root resorption)	Well-defined radiopacity without radiolucent halo	Cancellous/compact bone
<b>Idiopathic osteosclerosis</b>	Both / younger population	No symptoms; mandibular jaw;	No	Well-defined radiopacity without radiolucent halo	Thickened trabeculae; reduced marrow fibrovascular spaces
<b>Cementoblastoma</b>	Both / younger population	Discrete or no symptoms; mandibular molar area;	Yes (usually vital; can cause root resorption)	Well-defined radiopacity with radiolucent halo	Cementicles fused to form a mass and fibrovascular stroma
<b>Odontoma</b>	Both / younger population	No symptoms; Frontal parts of maxilla and posterior parts of mandible; Main cause of delayed teeth eruption;	No	Well-defined tooth shape radiopacity with a radiolucent halo	Dental hard tissues; dentin and enamel
<b>Osteoblastoma</b>	Male / younger population	Presence of a mild pain during the night, not relieved with salicylates; Unlimited growth potential; Facial asymmetry, swelling;	No	Well-defined radiopacity correlated with the amount of tissue calcification	Anastomosing trabeculae of woven bone rimmed by single layer of benign activated osteoblasts and numerous osteoclasts
<b>Osteoma</b>	Male / 20–50 years old	Presence of a mild pain during the night, relieved with salicylates; Limited growth potential;	No	Well-defined radiopacity correlated with the amount of tissue calcification	Dense, compact mature bone
<b>Fibrous dysplasia</b>	Female / younger population	Asymptomatic; Facial asymmetry, swelling;	No	“Ground-glass” radiographic appearance; loss of lamina dura	Fibroblastic proliferation with irregular shaped trabeculae (Chinese letters)
<b>Osteosarcoma</b>	Both / no prediction	Symptomatic; Pain; Fast volume increase; Presence of malignant features;	No	May be lytic, sclerotic or both; presence of radiopacity resembling sunrays	Atypical mesenchymal cells with osteoblastic differentiation and new anamelar bone production