DOI: https://doi.org/10.2298/SARH200521054G
UDC: 616.716.4-006-079.4

CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Cementoblastoma – an unusual radiographic presentation

Bojan Gačić[1], Branislav Ilić[1], Radojica Dražić[1], Aleksandra Čairović[2], Jelena Sopta[3], Ljubica Simić[3]

[1]University of Belgrade, School of Dental Medicine, Clinic of Oral Surgery, Belgrade, Serbia;
[2]University of Belgrade, School of Dental Medicine, Clinic of Dental Prosthetic, Belgrade, Serbia;
[3]University of Belgrade, Faculty of Medicine, Department of Pathology, Belgrade, Serbia

SUMMARY
Introduction Cementoblastoma is an uncommon tumor of the jaws that originates 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 from 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

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-osseus dysplasia, condensing osteitis, idiopathic osteosclerosis, cementoblastoma, odontoma, osteoblastoma, fibrous dysplasia. In order to get more precise information concerning the lesion, a cone bream 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).

Histopathological examination revealed that the tumor was composed of sheets of dens, irregular lamellated, 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

Received • Примљено: May 21, 2020
Revised • Ревизија: July 10, 2020
Accepted • Прихваћено: July 28, 2020
Online first: September 2, 2020

Correspondence to:
Branislav ILIĆ
Clinic of Oral Surgery
Doktora Subotića 4
11000 Belgrade, Serbia
branislav.ilic@stomf.bg.ac.rs

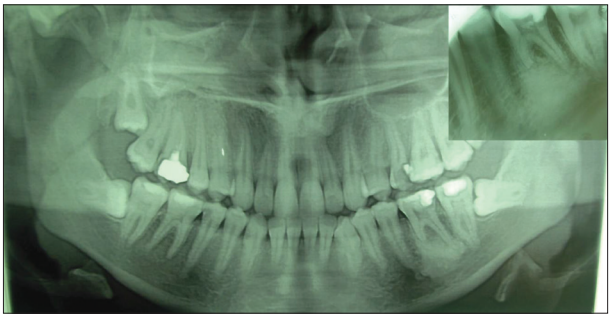

Figure 1. Retroalveolar and panoramic radiography: highly radiopaque mass is attached between the roots of tooth number 36

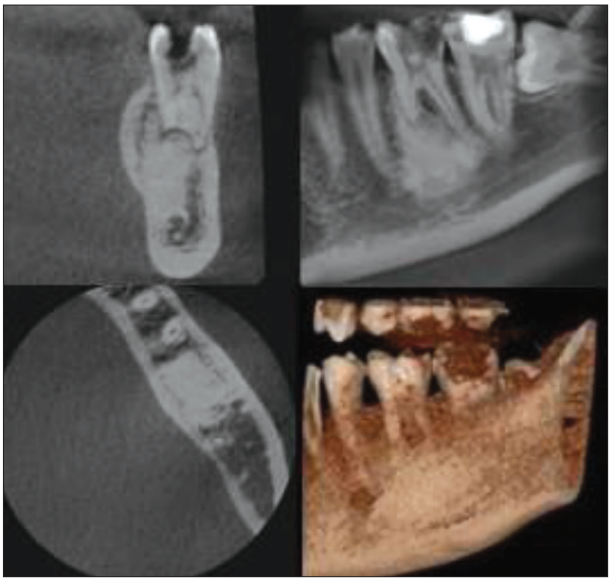

Figure 2. Cone beam computed tomography scans: unclear border of radiopaque mass is pushing down mandibular canal to the base of the lower jaw and causing the resorption of the mesial root

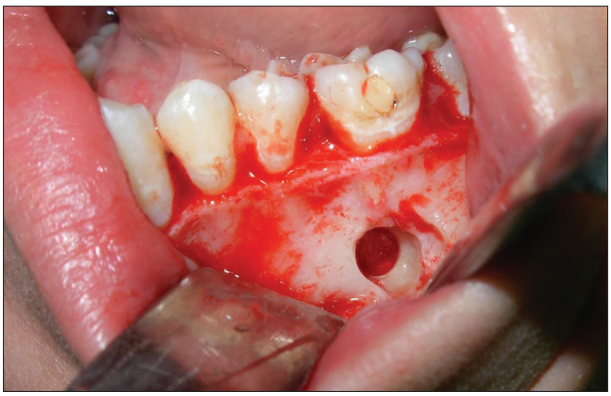

Figure 3. Intraoperative insight in biopsy: It was very difficult to identify tumour and its borders. The biopsy is performed according to preoperative radiography planning

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

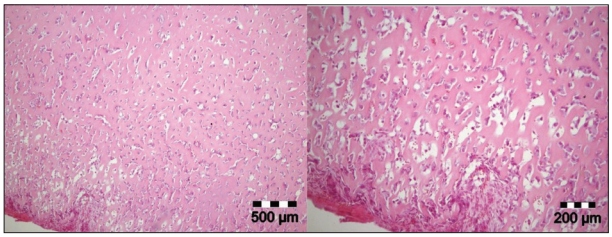

Figure 4a and 4b. Histologic findings: a – tumor consists cementum-like tissue (HE, 10×); b – prominent cementoblasts and trabeculae of uncalcified cemental matrix perpendicular to the surface (HE, 20×)

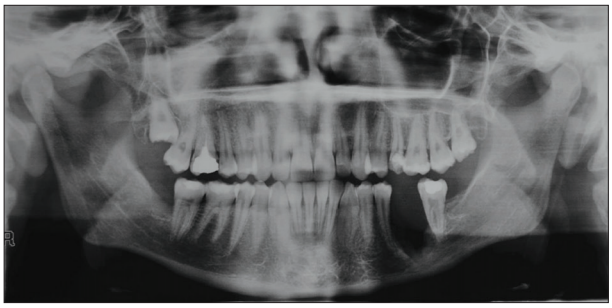

Figure 5. Follow-up radiography: There are no signs of tumor recurrence

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

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

Lesions	Age / Sex	Clinical	Tooth involvement	Radiographic	Histopathology
Hypercementosis	Both / over 40 years old	No symptoms; mandibular premolar area;	Yes (vital, no root resorption)	Well-defined radiopacity with radiolucent halo	Cellular/acellular cementum
Condensing osteitis	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
Idiopathic osteosclerosis	Both / younger population	No symptoms; mandibular jaw;	No	Well-defined radiopacity without radiolucent halo	Thickened trabeculae; reduced marrow fibrovascular spaces
Cementoblastoma	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
Odontoma	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
Osteoblastoma	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
Osteoma	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
Fibrous dysplasia	Female / younger population	Asymptomatic; facial asymmetry, swelling;	No	"Ground-glass" radiographic appearance; loss of lamina dura	Fibroblastic proliferation with irregular shaped trabeculae (Chinese letters)
Osteosarcoma	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 lamellar bone production

and the radiopaque lamina dura are always seen as the outer border of hypercementosis [17].

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

Condensing osteitis is characterized by presence of a low grade, chronical, 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 cemetoblasts and cementoclasts, it is characterized by woven bone production and proliferation of numerous plump activated osteoblasts, many osteoclasts, and fibrovascular 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 to be 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 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 is 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 through 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 did not 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.

REFERENCES

1. Dewey KW. Osteoma of a molar. Dent Cosmos. 1927;69:1143–9.
2. Razek AA. Odontogenic Tumors: Imaging-Based Review of the Fourth Edition of World Health Organization Classification. J Comput Assist Tomogr. 2019;43(5):671–8.
3. Barnes L, Eveson J, Reichart P, Sidransky D, ed. World Health Organization Classification of Tumors. Pathology and Genetics of Head and Neck Tumours, Lyon: IARC Press; 2005. p. 318.
4. El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg P, editors. WHO classification of Head and Neck Tumours. Chapter 8: Odontogenic and maxillofacial bone tumours. 4th edition, IARC: Lyion 2017, p. 205–60.
5. Pathak J, Hosalkar R, Sidana S, Swain N, Patel Sh. Benign Cementoblastoma Involving Left Deciduous First Molar: A Case Report and Review of Literature. J Oral Maxillofac Pathol. 2019;23(3):422–8.
6. Hiremath M, Srinath SK, Srinath S, Ashwathy T. Benign Cementoblastoma Associated With Primary Mandibular Second Molar: A Rare Case Report. J Oral Maxillofac Pathol. 2020;24(Suppl 1):S11–4.
7. Garg B, Chavada R, Pandey R, Gupta A. Cementoblastoma Associated With the Primary Second Molar: An Unusual Case Report. J Oral Maxillofac Pathol. 2019;23(Suppl 1):111–4.
8. Mohammadi F, Aminishakib P, Niknami M, Avarzamani AR, Derakhshan S. Benign cementoblastoma involving deciduous and permanent mandibular molars: A case report. Iran J Med Sci. 2018;43(6):664–7.
9. Cavalcante RC, Petinati MF, de Oliveira ER, Bergamaschi IP, Rebelatto NL, Klüppel L, et al. Benign Cementoblastoma Associated With an Impacted Third Molar Inside Maxillary Sinus. Iran J Med Sci. 2018;43(6):664–7.
10. Chrcanovic BR, Gomez RS. Cementoblastoma: An Updated Analysis of 258 Cases Reported in the Literature. J Craniomaxillofac Surg. 2017;45(10):1759–66.
11. Ghom AG, Meshram V, Diwe A, Kolte V. Benign Cementoblastoma. J Indian Acad Oral Med Rad. 2010;22:42–4.
12. Subramani V, Narasimhan M, Ramalingam S, Anandan S, Ranganathan S. Revisiting Cementoblastoma With a Rare Case Presentation. Case Rep Pathol. 2017;2017:8248691.
13. Borges DC, de Faria PR, Marangon HJ, Pereira LB. Conservative Treatment of a Periapical Cementoblastoma: A Case Report. J Oral Maxillofac Surg. 2019;77(2):272.e1–7.
14. Matteson SR. Benign tumors of the jaws. In: White SC, Pharoah MJ, editors. Oral radiology: principles and interpretation. 4th ed. Toronto: Mosby; 2000; p. 401–2.
15. Huber AR, Folk GS. Cementoblastoma. Head Neck Pathol. 2009;3(2):133–5.
16. Sharma N. Benign Cementoblastoma: A review of literature with a case report. Contemp Clin Dent. 2014;5(1):92–4.
17. Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and maxillofacial pathology (2nd edn.). Philadelphia: W.B. Saunders, 2002; p. 553–71.
18. Salvi A, Patankar S, Desai K, Wankhedkar D. Focal Cemento-Osseous Dysplasia: A Case Report With a Review of Literature. J Oral Maxillofac Pathol. 2020;24(Suppl 1):S15–S18.
19. Ledesma-Montes C, Jiménez-Farfán MD, Hernández-Guerrero JC. Maxillomandibular Giant Osteosclerotic Lesions. J Appl Oral Sci. 2018;26:e20170535.
20. Ledesma-Montes C, Jiménez-Farfán MD, Hernández-Guerrero JC. Idiopathic Osteosclerosis in the Maxillomandibular Area. Radiol Med. 2019;124(1):27–33.
21. Botelho J, Machado V, Gomes JC, Borrecho G, Maia P, Mendes JJ, et al. Multiple Complex Odontomas of the Mandible: A Rare Case Report and Literature Review. Contemp Clin Dent. 2019;10(1):161–5.
22. Kaplan I, Nicolaou Z, Hatuel D, Calderon S. Solitary central osteoma of the jaws: A diagnostic dilemma. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2008;106(13):22–9.
23. Wang HW, Ma CY, Qin XJ, Zhang CP. Management strategy in patient with familial gigantiform cementoma: A case report and analysis of the literature. Medicine (Baltimore). 2017;96(50):e9138.
24. Pereira TDSF, Gomes CC, Brennan PA, Fonseca FP, Gomez RS. Fibrous dysplasia of the jaws: Integrating molecular pathogenesis with clinical, radiological, and histopathological features. J Oral Pathol Med. 2019;48(1):3–9.
25. Barrios BA, Rodriguez LH, Arzate H, Monroy ME, Lopez RS, Diaz DR, et al. Isolation of Cementum Protein in a Cementoblastoma. Oral Surg Oral Med Oral Pathol Oral Radiol. 2013;116:498.
26. Assi R, Kessler H, Ghali G, Yeoh M. Giant cementoblastoma treated with resection. Oral Surg Oral Med Oral Pathol Oral Radiol. 2012;114:66.

Цементобластом – необична радиографска манифестација

Бојан Гачић[1], Бранислав Илић[1], Радојица Дражић[1], Александра Чаировић[2], Јелена Сопта[3], Љубица Симић[3]

[1]Универзитет у Београду, Стоматолошки факултет, Клиника за оралну хирургију, Београд, Србија;
[2]Универзитет у Београду, Стоматолошки факултет, Клиника за стоматолошку протетику, Београд, Србија;
[3]Универзитет у Београду, Медицински факултет, Институт за патологију, Београд, Србија

САЖЕТАК

Увод Цементобластом је тумор виличних костију који води порекло од одонтогеног ектомезенхима, а карактерише га пролиферишуће ткиво налик на цемент.

Приказ болесника У раду је приказан цементобластом доње вилице, атипичне радиографске манифестације: без јасно дефинисане границе и без зоне периферног расветљења. Прегледом доступне литературе евалуирали смо различите туморе/лезије који 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 болесника.

Кључне речи: цементобластом, одонтогени тумори, тумори максилофацијалне регије