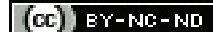


# Exudative Benign Cementoblastoma with Radiographic Rarities- A Case Report with Review of Literature

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## ABSTRACT

Cementoblastoma is a rare benign odontogenic neoplasm of mesenchymal origin. The benign cementoblastoma was previously classified as one of the cementoma neoplasias by the World Health Organisation's (WHO) classification of odontogenic tumours. The benign cementoblastoma in 2005 has been added to the list of odontogenic tumours that involve "mesenchyme and/or odontogenic ectomesenchyme, with or without odontogenic epithelium". It accounts for around 0.69-8% of all odontogenic tumours. The lesion usually is asymptomatic and slow growing causing cortical plate expansion of both buccal and lingual plates. Cementoblastoma has male predilection, commonly seen involving mandibular first molar diagnosed in early age. There are very few incidents of an infected cementoblastoma in the literature. Radiological examinations can reveal a blend of radiolucent and radiopaque mass presenting a wheel spoke pattern. The histopathological results for both cementoblastoma and osteoblastoma are comparable. The relationship of the lesion with the tooth is the distinguishing element. Hereby, authors present a case of infected cementoblastoma in a 51-year-old female patient involving impacted mandibular second molar, clinically aggressive with unique radiologic features. Cementoblastoma is usually an incidental finding while the present case was associated with an impacted tooth and was symptomatic with infection. The uniqueness of the present case was the complete involvement of the root structure and the cyst like expansion of the lesion instead of the usual thin radiolucent rim. Also, the lesion was infected with no dental caries associated with the tooth.

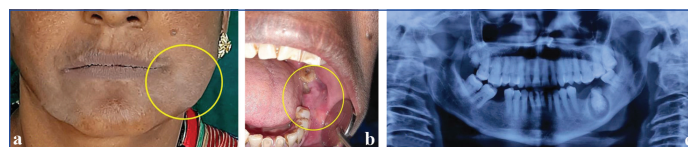
**Keywords:** Dental cementum, Impacted, Odontogenic tumour

## CASE REPORT

A 51-year-old female patient reported to the Dental Hospital with the chief complaint of swelling and pain on the left side of the face which she had noticed two months ago. The pain was moderate in intensity, intermittent in nature with no aggravating factors and relived on taking analgesics. The swelling was initially small which gradually grew to attain the present size. The patient had visited a dental practitioner with the same complaint two months ago where she got her tooth left mandibular first molar (36) extracted which was misdiagnosed as the cause for swelling and pain without prior diagnostic radiographs. The patient reported to our dental hospital since there was no relief from pain and swelling. The patient's medical and family history were unremarkable.

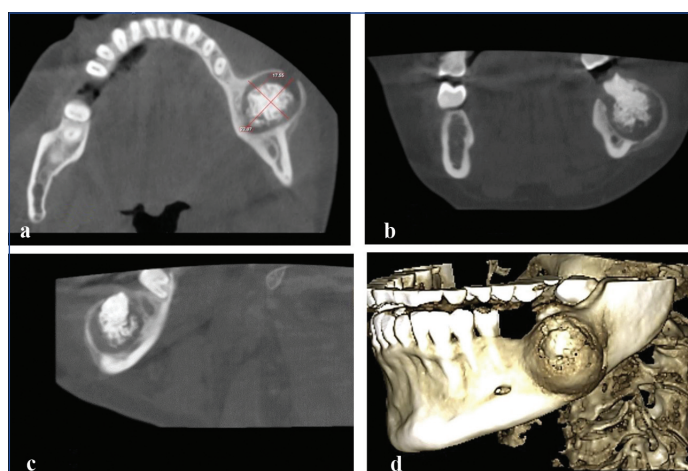
Clinically, patient presented with an extraoral swelling in the left lower third of the face measuring around 5×6 cm in diameter with no remarkable surface changes [Table/Fig-1a]. On palpation, the swelling was well defined and hard in consistency and no signs of paraesthesia noted. Intraoral examination revealed obliteration of the left buccal vestibule in the region of 36 and 37, associated buccal cortical plate expansion with intact lingual cortical plate. Pus discharge was noted in the region of 37 which was clinically missing [Table/Fig-1b].

Considering the above history and examination a provisional diagnosis of dentigerous cyst with respect to impacted 37 was given and differential diagnoses of infected residual cyst (in relation to 36) and odontogenic tumour with impacted teeth were considered. Orthopantomograph (OPG) revealed an impacted 37 with a radiopaque mass involving the entire root. Loss of complete root morphology and well circumscribed wide spacious concentric ring around the root which had fibrous root appearance was evident [Table/Fig-1c] which is rarely seen as compared to common spoke wheel appearance. Cone Beam Computed Tomography (CBCT) images of different sections, showed a well-defined radiolucent band inside the cortical border with a radiopaque mass at the centre accompanied by minor areas of radiolucency [Table/Fig-2a-c].



**[Table/Fig-1a-c]:** a): Extraoral image ; b): Intraoral image showing swelling and pus discharge; c): OPG shows the impacted 37 with radiopaque (fibrous root appearance) mass involving the entire root morphology.

There was no invasion of the lesion to the mandibular canal in all three sections- axial, coronal and sagittal. There was evidence of buccal cortical plate expansion and perforation of buccal cortical plate, giving more evidence of an aggressive matured benign cementoblastoma [Table/Fig-2d].

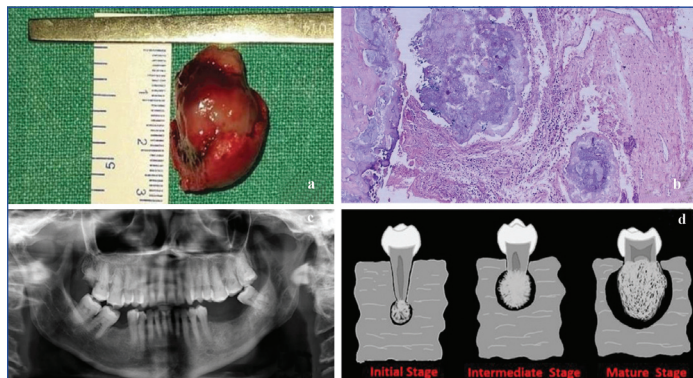


**[Table/Fig-2a-d]:** Cone beam computed tomography images of axial (a) coronal (b) sagittal (c) and (d) 3-dimensional reconstruction showing loss of root morphology, with extension, expansion and perforation of the buccal cortex.

Since, the lesion was infected, the patient was put on broad spectrum antibiotic coverage and routine haematological examination before surgical excision showed normal blood parameters. Excisional biopsy of the lesion along with the extraction of the associated 37 was done

under general anaesthesia. The gross specimen consisted of a tooth along with bony tissue mass measuring 2.5×2×2 cm [Table/Fig-3a]. There was loss of the root morphology while the crown appeared normal. Microscopically, the sections showed basophilic masses with inflammatory infiltrate, fibrovascular connective tissue, areas of fibrosis and bony trabeculae lined by osteoblasts and association of the lesion with the root of the tooth, confirmed the diagnosis of cementoblastoma [Table/Fig-3b].

The patient was advised periodic follow-up upto six months. The patient showed remarkable improvement in terms of healing and pain control in two months. Postoperative follow-up OPG taken at the end of six months showed complete resolution of the lesion with no recurrence [Table/Fig-3c]. Patient was advised for replacement of missing teeth. Schematic representation of the proposed staging of cementoblastoma based on root involvement with varied radiographic features [Table/Fig-3d].



**[Table/Fig-3a-d]:** a) Gross specimen involving the tooth; b) Histopathological examination on H&E stained decalcified section, shows basophilic mass with inflammatory infiltrate, areas of fibrosis lined by osteoblasts and association of the lesion with the root of the involved tooth (H&E, 10X); c) Follow-up OPG shows complete resolution of the lesion with no recurrence; d) Schematic representation of the proposed staging of cementoblastoma based on root involvement with varied radiographic features.

## DISCUSSION

Cementoblastoma is a relatively rare and uncommon benign neoplasm of mesenchymal origin [1]. Cementoblastoma accounts for around 0.69-8% of all odontogenic tumours [2]. It is formed by the haphazard deposition of cementum like material by the proliferating cementoblast cells. The neoplasm usually presents as a slow growing bony hard swelling which is predominantly asymptomatic [3]. Paraesthesia was also observed when the lesion involves the mandibular canal [4]. The lesion has a slight male predilection usually occurring in the 2<sup>nd</sup> and 3<sup>rd</sup> decade of life. While there are cases reported in the literature in patients ranging between 10-72 years of both genders [4,5]. Few literature points to their equal gender predilection [6]. A fully erupted mandibular permanent 1<sup>st</sup> molar is most commonly associated with the lesion. Nevertheless, the incidence of occurrence in the maxilla and primary dentition cannot be denied [7]. The present case under discussion is unique in its presentation which is highlighted in [Table/Fig-4]. Based on the literature survey very few cases have been reported involving impacted mandibular second molar [Table/Fig-5] [1-3, 5-25].

The lesion usually is asymptomatic and slow growing causing cortical plate expansion of both buccal and lingual plates [5]. There are very few incidents of an infected cementoblastoma in the literature [5,8,9]. The clinical examination in the present case revealed pus discharge in relation to unerupted 37 with intraoral sinus opening and 36 was missing. The present case seemed aggressive and more matured (considering the age at diagnosis) with the expansion of the buccal cortical plate and an extraoral swelling which the patient had noticed for the past two months.

Chrcanovic BR and Gomez RS, analysed 258 cases of cementoblastoma deriving the following result i.e., equal gender prevalence and higher prevalence was evident in the 2<sup>nd</sup>/3<sup>rd</sup> decades

Timeline	Features
History given by patient	Swelling and pain on left lower back teeth region for two months.
Clinical features at time of reporting to clinic	Extraorally a well-defined swelling noted on left body of mandible which was hard in consistency with no remarkable surface. Intraorally obliteration of the left buccal vestibule in the region of clinically missing 36 and 37, with buccal cortical plate expansion and intact lingual cortical plate. Pus discharge was noted in the region of 37.
Final diagnosis	Benign cementoblastoma (based on clinical, radiological and histopathological examination).
Management	Antibiotic coverage with routine blood examination. Surgical excision under GA.
Outcome and follow-up	Periodic follow-up upto six months. Follow-up OPG taken after six months showed complete resolution of the lesion with no recurrence.
Uniqueness of case	<ul style="list-style-type: none"> <li>• Age/Gender: 51/F</li> <li>• Location/tooth involved: 37</li> <li>• Eruption status: Infected/Vertically Impacted</li> <li>• Complete root involvement with loss of entire root morphology with fibrous root appearance suggestive of aggressive and more mature lesion.</li> </ul>

**[Table/Fig-4]:** Timeline of events and unique features of the present case report.

of life. The common site of occurrence is mandibular 1<sup>st</sup> molars. Lesions were frequently related with bone expansion (74.9%), clinical symptoms (70.2%), vital teeth (78%), and root resorption (59.8%). Less frequent observations included perforation of cortical bone (16.3%), inferior displacement of the mandibular canal (23.6%) [10].

The results of a radiological examination can present three stages i.e., initial, intermediate and mature stages. In the initial stage, the lesion is usually radiolucent. The intermediate stage manifests a blend of radiolucent and radiopaque mass. While a completely mature lesion displays a dense radiopaque mass surrounded by a radiolucent halo [26]. Going through the radiographic data of case reports, it was observed that almost all the lesions were in the maturing stage and few were found to be in the intermediate stage. Therefore, lesions in their initial stage, seldom show any symptoms. Also, the case reports of cementoblastoma associated with infection or impacted tooth, are only a few [Table/Fig-5] [1-3, 5-25].

In the present case, the radiographic examination via orthopantomogram and CBCT images revealed an impacted 37 with a radiodense mass involving the entire root component from the cervical region to its apex showing a fibrous root appearance which was different from the regular wheel spoke pattern. The density of the cemental mass was obscuring the entire outline of the enveloped root resulting in loss of entire root morphology. The mass had invaded the root canal and had obliterated the periodontal ligament space. The tumour was considered to be in the matured stage since the central radiopaque mass involving the entire root was surrounded by a radiolucent band from the center to the periphery. The lesion was extensive enough to cause the expansion of mandible perforating the buccal cortical plate without any periosteal reaction. In the literature review most of the reported cases involved either apical 3<sup>rd</sup> or extended up to middle third of root which was different in the present case with the involvement of the entire root. Considering the unique radiographic features in the present case, a new classification for staging can be proposed based on the level of root involvement [Table/Fig-3d].

1. Initial stage-Tumour involving apical third.
2. Intermediate stage-Tumour involving middle third of root.
3. Mature stage-Tumour involving entire root.

Cementoblastoma in its initial stage can be treated endodontically. While the intermediate and mature stages require extraction of the involved tooth [27].

Histopathological investigation gives similar findings for both cementoblastoma and osteoblastoma. The differentiating factor is through the association of the lesion to the tooth. In Cementoblastoma,

Author and year	Place of study	Age (years)/ Sex	Teeth affected	Signs and symptoms	Infected/uninfected	Radiographic features including expansion of cortex perforation staging
Chatha AA et al., (2017) [1]	India	15/Female	36 Erupted	Painless extraoral swelling with paraesthesia of lower left jaw	Uninfected	Radiodense lesion Expansion of buccal and lingual cortex No perforation Mature stage
Sirigala L et al., (2015) [2]	India	8/Male	84, 85 Erupted	Pain and swelling	Uninfected	A radiopaque mass Expansion of buccal cortex Perforation of buccal cortex Mature stage
Milani C et al., (2012) [3]	Brazil	19/Male	45, 46, 47 Erupted	Painless hard swelling	Uninfected	Well-defined, round, radiopaque lesion Radiopaque mass Expansion of buccal and lingual cortex No perforation Mature stage
Kumar S et al., (2011) [5]	India	55/Female	48 Erupted	Swelling and pus discharge	Infected	Radiopaque mass surrounded by a radiolucent band Expansion of buccal and lingual cortex No perforation Mature stage
Pellizzaro D et al., (2017) [6]	Brazil	52/Female	27 Erupted	Asymptomatic	Uninfected	Well defined, radiopaque mass No expansion of cortices No perforation Mature stage
Bal Reddy P et al., (2012) [7]	India	28/Male	14 Erupted	Dull ache	Uninfected	Radiopaque mass No expansion of cortices No perforation Mature stage
Seifi S et al., (2018) [8]	India	31/Female	36 Erupted	Pain and pyorrhea	Infected	Radiopaque mass surrounded by a radiolucent rim No expansion of cortices No perforation Mature stage
Dinakar J et al., (2010) [9]	India	41/Female	48 Partially erupted	Pain and swelling	Infected	Radiopaque mass Expansion of buccal cortex No perforation Mature stage
Chrcanovic BR and Gomez RS, 2017 [10] Including analysis by Brannon et al., (Total=258 cases) (2002)	-	Age range: Male: 4-64 years Female: 7-75 years	Maxilla: 57 Mandible: 196 Unknown:5 Eruption status: unknown	Symptomatic: 158 cases Asymptomatic: 67 cases Unknown: 33 cases	Uninfected	Radiopaque mass: 220 cases Radiolucent mass: 4 cases Unknown: 34 cases Expansion of cortices:164cases No expansion of cortices: 55 cases Unknown: 39 cases Cortical bone perforation Yes: 35 cases No: 180 cases Unknown: 43 cases
Cavalcante RC et al., (2018) [11]	Brazil	22/Male	28 Impacted	Asymptomatic	Uninfected	A well-defined hyperdense mass with hypodense centre inside the maxillary sinus No expansion of cortices No perforation Mature stage
Berwick JE et al., (1990) [12]	Canada	15/Female	36 Erupted	Pain and swelling	Uninfected	A radiopaque mass surrounded by a radiolucent halo Expansion of buccal cortex No perforation Mature stage
Mogi K et al., (1996) [13]	Japan	24/Male	36 Erupted	Pain and extraoral swelling	Uninfected	A round radiopaque mass measured 20 mm in diameter Expansion of buccal cortex No perforation Mature stage
Piattelli A et al., (1997) [14]	Italy	35/Female	48 Impacted	Pain and intraoral swelling	Uninfected	A radiopaque lesion surrounded by a radiolucent halo Expansion of buccal cortex No perforation Mature stage
Aiyer RG and Rajagopal S, (2000) [15]	India	19/Male	35,36,37 Erupted	Painless extraoral swelling	Uninfected	Fairly large opacity with surrounding well defined zone of radiolucency No expansion of cortices No perforation Mature stage
Sumer M et al., (2006) [16]	Turkey	46/Male	38 Impacted	Pain, trismus and swelling	Uninfected	A radiopaque lesion surrounded by a radiolucent halo Expansion of buccal cortex No perforation Mature stage
Agrawal M et al., (2013) [17]	India	16/Male	37 Erupted	Pain and swelling	Uninfected	Well-defined mixed radiopaque radiolucent lesion surrounded by a radiolucent rim Expansion of buccal and lingual cortex No perforation Intermediate stage

Dinakar C et al., (2015) [18]	India	33/Male	48 Impacted	Pain and swelling	Uninfected	A radiopaque lesion surrounded by a thin radiolucent rim Expansion of buccal cortex No perforation Mature stage
Revathi S et al., (2016) [19]	India	28/Female	38 Partially Erupted	Pain	Uninfected	A well-defined round to ovoid radiopaque lesion demarcated with thin radiolucent line No expansion of cortices No perforation Mature stage
Subramani V et al., (2017) [20]	India	19/Male	36 Partially Erupted	Asymptomatic extraoral swelling with cortical bone expansion	Uninfected	A well-circumscribed radiolucent mass Expansion of buccal and lingual cortex No perforation Initial stage
Borges DC et al., (2018) [21]	Brazil	33/Female	28 Erupted	Asymptomatic	Uninfected	Well-demarcated lesion circumscribed by a radiolucent halo No expansion of cortices No perforation Mature stage
de Andrade MG et al., (2018) [22]	Brazil	60/Female	37 Erupted	Pain	Uninfected	Hyperdense mass No expansion of cortices No perforation Mature stage
Quadri F et al., (2019) [23]	India	33/Female	45 Erupted	Pain and buccal cortical plate expansion	Uninfected	Dense cemental thickening Expansion of buccal cortex No perforation Mature stage
Hussein WA, (2019) [24]	Egypt	29/Male	47 Erupted	Painless Intra-Oral Swelling	Uninfected	Radiopaque mass with a uniform radiolucent halo No expansion of cortices No perforation Mature stage
Santana SF et al., (2020) [25]	Brazil	24/Female	35,36,37 Erupted	Pain and swelling	Uninfected	Hyperdense lesion with a well-defined and rounded hypodense halo No expansion of cortices No perforation Mature stage
Present case, 2022	India	51/Female	37 Impacted	Pain, swelling and pus discharge	Infected	A radiopaque lesion surrounded by a radiolucent halo Expansion of buccal cortex Perforation of buccal cortex Mature stage

**[Table/Fig-5]:** Demographic and clinical data of reported cases of cementoblastoma with emphasis on radiographic features, eruption and infection status [1-3, 5-25].

the radiopaque mass is attached to the root of the associated tooth while in osteoblastoma the radiopaque mass is usually not in contact with the root of any tooth [13].

The present case of infected cementoblastoma associated with impacted tooth, had a history of extraction of the adjacent tooth. The chances of infection of cementoblastoma associated with an unerupted tooth, is highly unlikely [5]. The lesion may have incurred the infection from an infected periapical lesion of the adjacent tooth or could be idiopathic. The probable cause of infection of cementoblastoma associated with an erupted tooth with no dental caries may be from the per coronal abscess or periodontal infection of the associated tooth or idiopathic.

## CONCLUSION(S)

The present case was a case of cementoblastoma with distinct clinical and radiological manifestations. The present case mandates the inclusion of cementoblastoma in the differential diagnosis of impacted/clinically missing tooth. Also, highlights the importance of radiographic investigation prior to arriving at a specific diagnosis and intervening with any surgical intervention. Radiographic investigation should be considered mandatory by all practitioners to avoid misdiagnosis and inaccurate treatment planning.

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