Case Report

Benign cementoblastoma involving left deciduous first molar: A case report and review of literature

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Abstract

Cementoblastoma, a benign mesenchymal odontogenic neoplasm is derived from ectomesenchymal cells of the periodontium. Cementoblastomas associated with primary teeth are extremely rare as permanent mandibular first molars are mostly affected. Only 17 cases of those associated with deciduous dentition have been reported so far. The present case report describes a true cementoblastoma of an 8-year-old male child in relation to the left first primary mandibular molar along with emphasis on differential diagnosis.

Keywords: Cementoblastoma, deciduous dentition, differential diagnosis, odontogenic neoplasm

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INTRODUCTION

Cementoblastoma is a slow-growing, benign odontogenic neoplasm of mesenchymal origin, with unlimited growth potential and is derived from ectomesenchymal cells of the periodontium including cementoblasts.[1] Cementoblastoma was first described by Dewey in 1927[2] and was recognized first by Noeberg[1] in 1930. They are commonly seen in children and young adults; males are more frequently affected than females, with more occurrences in mandible than maxilla. Radiographically, benign cementoblastoma appears as a well-defined radio-opacity with a radiolucent peripheral zone. The growth rate for cementoblastoma is estimated to be 0.5 cm/year.[3] The histological features of cementoblastoma include cementum-like tissue with numerous reversal lines, and between these mineralized and trabecular hard tissues, fibrovascular tissue with cementoblast-like cells is present along with multinucleated giant cells.[4] The treatment of choice is complete removal of the lesion with extraction of associated tooth, followed by thorough curettage and peripheral ostectomy. The recurrence rate is 21.7%–37.1%.[3] It is a rare tumor with <300 cases ever reported in literature.[5] Cementoblastoma is more commonly associated with permanent mandibular first molars with deciduous teeth being rarely involved.[6] So far, only 17 cases[6-22] involving deciduous dentition have been reported [Table 1]. The present case report describes a true cementoblastoma in relation to the left first primary mandibular molar in an 8-year-old child along with emphasis on differential diagnosis.

CASE REPORT

An 8-year-old healthy male child reported to the Department of Oral and Maxillofacial Pathology of our

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Table 1: Demographic factors of cementoblastoma cases reported from 1965 to 2018

Author and years	Age	Gender	Site	Teeth affected	Treatment	Recurrence	Follow-up
Chaput and Marc 1965[6]	10	Female	Lower posterior mandible	Right mandibular first premolar and second deciduous molar	ND	ND	ND
Vilasco *et al*., 1969[7]	8	Female	Lower posterior mandible	Right mandibular second deciduous molar	ND	ND	ND
Zachariades *et al*., 1985[8]	7	Female	Lower posterior mandible	Deciduous first and second molars and first permanent molar	Enucleation	Absent	LTF
Herzog 1987[9]	7	Female	Posterior mandible	Deciduous first and second molars	ND	ND	ND
Cannell 1991[10]	8	Female	Posterior mandible	Deciduous second molar	ND	ND	ND
Schafer *et al*., 2001[11]	8	Female	Posterior mandible	Deciduous second molar	Excision	ND	LTF
Ohki *et al*., 2004[12]	12	Male	Posterior mandible	Right maxillary second deciduous molar, first premolar and the first and second permanent molars Both the crown and the root of the un-erupted second premolar	Excision	Absent	FOD
Lemberg *et al*., 2007[13]	10	Female	Posterior mandible	Deciduous second molar	Excision	Absent	FOD
Vieira *et al*., 2007[14]	7	NM	Posterior mandible	Deciduous second molar	Excision	Absent	ND
de Noronha Santos Netto *et al*., 2012[15]	4	Female	Posterior mandible	Deciduous first molar	Excision	Absent	FOD
Solomon *et al*., 2012[16]	7	Female	Posterior maxilla	Deciduous second molar	Sub totalmaxillectomy	Present	FOD
Monti *et al*., 2013[17]	11	Female	Posterior mandible	Deciduous second molar	En bloc resection	ND	ND
Urs *et al*., 2016[18]	10	Male	Posterior maxilla	Deciduous first and second molar	Excision	Absent	FOD
Nuvvula *et al*., 2016[19]	7	Female	Posterior mandible	Deciduous second molar	Excision	Absent	FOD
Javed and Hussain Shah 2017[20]	10	Female	Anterior and posterior maxilla	Deciduous canine to second molar with permanent first molar	Ostectomy with chemical cauterization	Absent	FOD
Nagvekar *et al*., 2017[21]	12	Male	Posterior maxilla	Deciduous second molar	Excision	Absent	FOD
Mohammadi *et al*., 2018[22]	4.5	Male	Posterior mandible	Deciduous second molar and first permanent molar	Excision	Present	FOD
Present case	8	Male	Posterior mandible	Deciduous second molar	Excision	Absent	FOD

ND: No data, LTF: Lost to follow up, FOD: Free of disease, NM: Not mentioned

institute with a chief complaint of pain and mild swelling in the left body of the mandible which had been increasing in size for the past 2 months. On clinical examination, no extraoral swelling was present in lower one-third of the face. Mild intraoral swelling with obliteration of the vestibular space was associated with deciduous mandibular left first molar. The swelling was diffuse and hard in consistency, with expansion of buccal cortex. Tenderness on palpation was noticed. Overlying mucosa was normal with no ulceration or purulent discharge. No carious teeth were seen in the region.

Radiological examination using cone beam computed tomography revealed a localized mixed radio-opaque–radiolucent lesion in the buccal aspect extending from the distal aspect of 32 to the mesial aspect of developing 34 [Figure 1a and 1b]. The lesion was surrounded by a thin, uniform radiolucent line [Figure 2]. It was involving the periapices of 74 and was in continuity with the roots of the same [Figure 3]. It extended inferiorly up to the middle third level of the coronal portion of the developing 33. The approximate maximum dimensions of the lesion were 11.9 mm × 13.8 mm × 16 mm [Figure 4]. Considering the

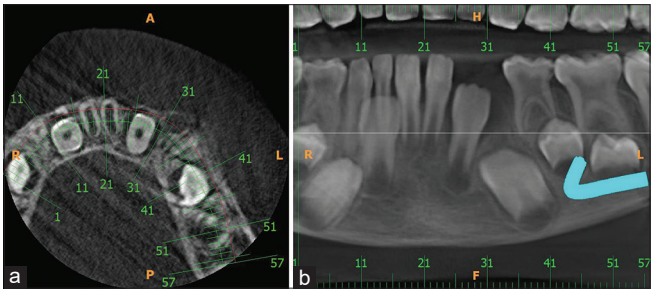

Figure 1: Cone beam computed tomography showing localized mixed radio-opaque–radiolucent lesion in the buccal aspect extending from the distal aspect of 32 to the mesial aspect of developing 34. (a) Axial view (b) Panoramic view

clinical and radiographical findings, differential diagnosis of the lesion included odontogenic tumor, fibro-osseous lesion or hypercementosis. An excisional biopsy was performed for final diagnosis.

The gross specimen included multiple bits of hard tissues and deciduous mandibular first molar. Hematoxylin and eosin stained sections showed sheets of cementum-like tissue with prominent reversal lines [Figure 5]. Areas

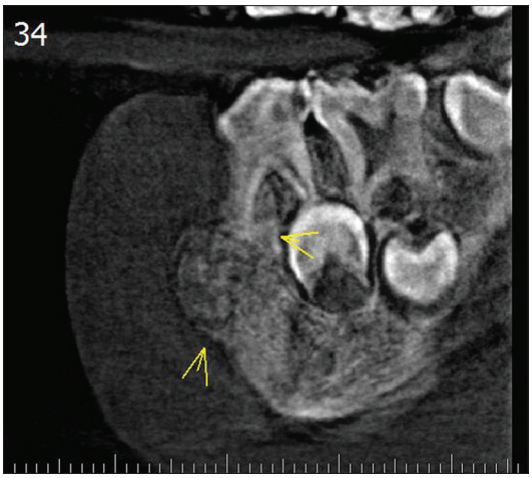

Figure 2: Cone beam computed tomography showing lesion surrounded by a peripheral radiolucency (yellow arrows)

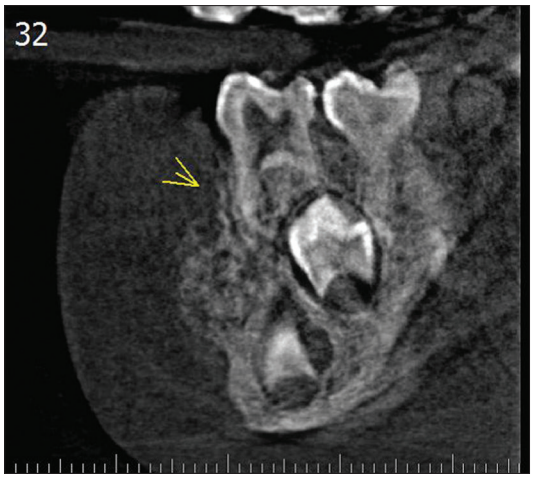

Figure 3: Cone beam computed tomography radio-opacity involving the periapices of 74 and is in continuity with the roots of the same (yellow arrows)

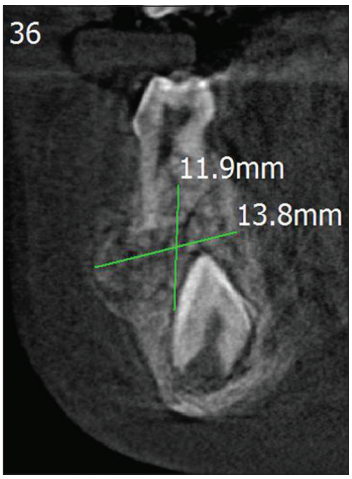

Figure 4: Cone beam computed tomography approximate maximum dimensions of the lesion (11.9 mm × 13.8 mm)

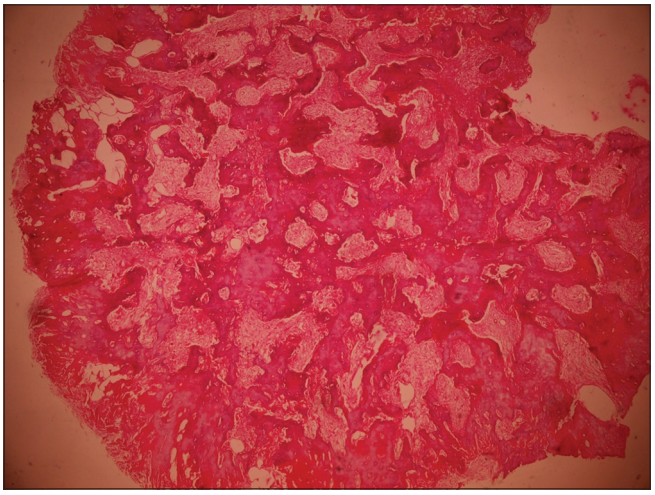

Figure 5: Photomicrograph of hematoxylin and eosin stained decalcified sections show sheets of cementum like tissue with prominent reversal lines (H&E stain, ×4)

of fibrovascular connective tissue interspersed between cementum like masses [Figure 6]. At the periphery of the lesion, radiating columns of cellular unmineralized cementoid tissue was evident [Figure 7]. Multinucleated giant cells and plump cementoblasts were also seen [Figure 8]. Prominent and numerous basophilic reversal lines were appreciable [Figure 9]. On basis of clinical, radiological and histopathological correlation, a diagnosis of cementoblastoma was given. The patient is on follow up since last 6 months and is free of disease.

DISCUSSION

Cementoblastoma is a rare lesion that represents <1% of the odontogenic tumors.[6] Their prevalence among all odontogenic tumors has been reported to vary from 0.69% to 8%.[18-23] It is more common in young patients, with about 50% of them arising under the age of 20 years. Females (78.5%) are more commonly affected than males (21.5%). Most cementoblastomas are closely allied to and partly surround a root or roots of a single erupted permanent tooth.[24] It most commonly occurs in the mandibular molar–premolar region.[25] Primary teeth are very rarely affected. Mandibular arch (93%) is more commonly involved than the maxillary arch (7%). Cementoblastoma was commonly seen on the right side (71.5%) of mandibular arch, followed by the left side of the mandibular arch (21.5%) and the right side of the maxillary molar region (7%), the most common tooth affected being right mandibular second molar (71%).[19] The present case is in accordance with the literature and is only the 18th case report so far, associated with the primary molar.

Painful swelling at the buccal and lingual/palatal aspect of the alveolar ridges is the most common symptom

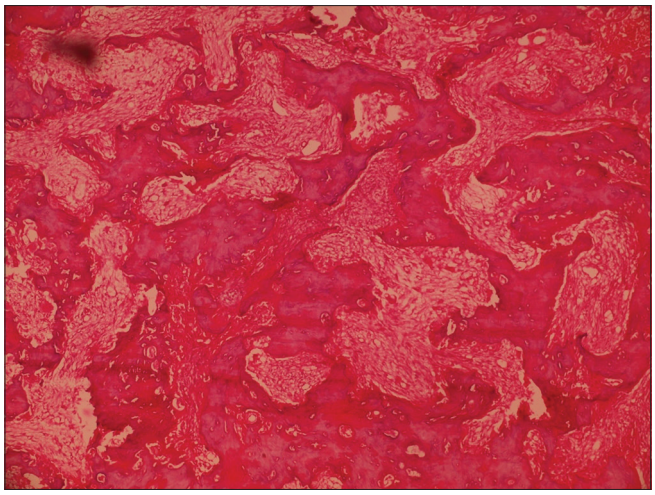

Figure 6: Photomicrograph of hematoxylin and eosin stained decalcified sections show areas of fibrovascular connective tissue interspersed between cementum like masses (H&E stain, ×10)

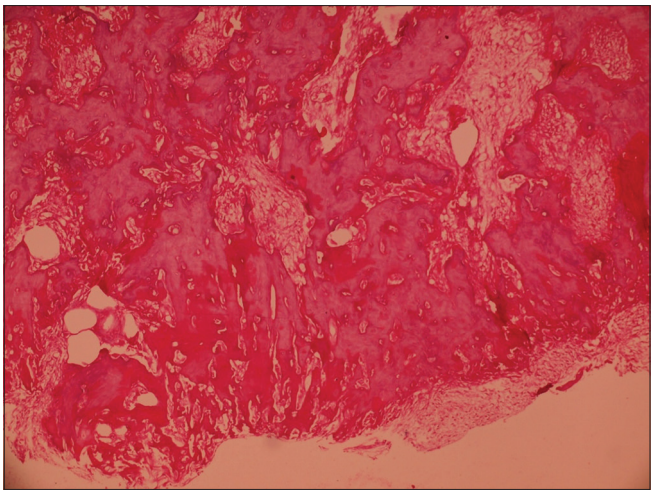

Figure 7: Photomicrograph of hematoxylin and eosin stained decalcified sections show radiating columns of cellular unmineralized cementoid tissue at the periphery of the lesion (H&E stain, ×10)

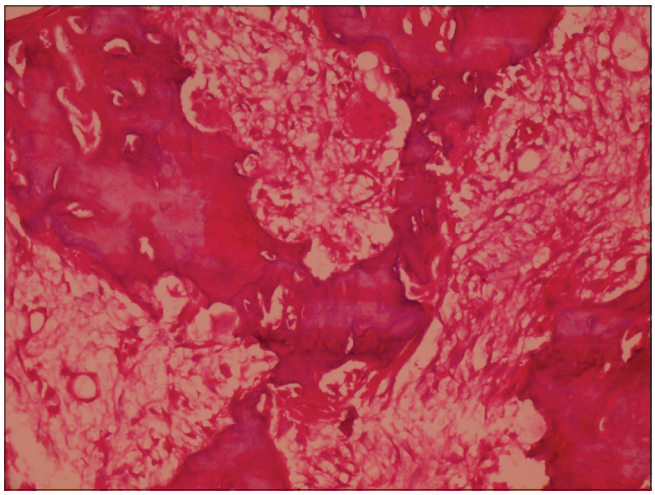

Figure 8: Photomicrograph of hematoxylin and eosin stained decalcified sections show multinucleated giant cells and plump cementoblasts (H&E stain, ×40)

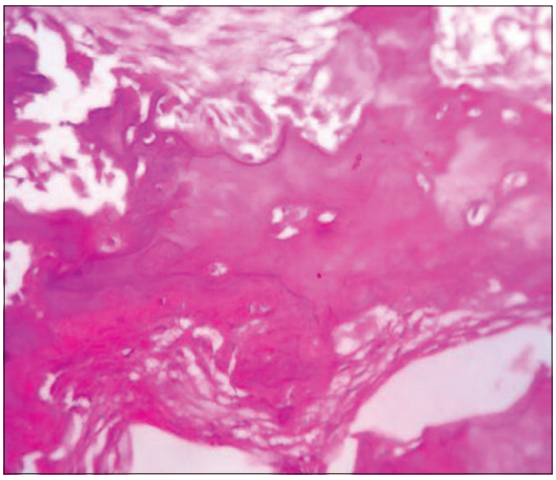

Figure 9: Photomicrograph of hematoxylin and eosin stained decalcified sections show prominent and numerous basophilic reversal lines (H&E stain, ×40)

associated with cementoblastoma. Occasionally, it may be asymptomatic. The involved tooth remains vital. Cortical expansion and facial asymmetry are also common findings. Lower lip paresthesia or a pathologic fracture of the mandible is rarely reported.[26] In the present case, the patient complained of pain and mild swelling in the lower left posterior region.

Cementoblastoma is derived from the functional cementoblasts of odontogenic ectomesenchyme that lay down cementum on the tooth root. Cementoblastoma is continuous with the cemental layer of the apical third of the tooth root and remains separated from bone by a continuation of the periodontal ligament (PDL), all of which supports an odontogenic origin.[27] Pathogenesis of cementoblastoma progresses in three stages with first stage being periapical osteolysis followed by cementoblastic stage and finally with calcification and maturation.[28] Radiographically, it appears as a well-defined radio-opacity surrounded by a radiolucent periphery and is continuous with the apical one-third of the root and PDL. The histopathological features of cementoblastoma include sheets of cementum-like material continuous with the tooth root. The proliferating cementum is lined by numerous plump cementoblasts. Cementoblasts are also present along with prominent reversal lines. Some of the cemental material maybe noncalcified, particularly at the periphery of the mass the tumor and often arranged in struts perpendicular to the capsule. The fibrous stroma is highly vascular.[29] The present case meets the radiological and histopathological criteria of a benign cementoblastoma.

Table 2: Compare and contrast benign cementoblastoma with other lesions

Common features	Osteoblastoma	Cementoblastoma
Clinical features Arises in young adults Slowly progressive Shows bony expansion	Clinical features Not associated with tooth roots Arises in the medullary cavity	Clinical features Intimately associated with the tooth roots Arises from the cementoblasts in the PDL
	Radiographic features Absence of peripheral radiolucent rim	Radiographic features Radiopaque masses attached to teeth and surrounded by a radiolucent periphery
Histopathological features Cellular and vascular stroma with sheets of bone/cemental tissue and multinucleated giant cells	Histopathological features Reversal lines absent	Histopathological features Reversal lines present
Common features	**Odontomes**	**Cementoblastoma**
Radiographic features Both are usually sharply marginated, and sclerotic, with a low-attenuation halo	Radiographic features They are usually pericoronal	Radiographic features Appear periapically directly fusing to the root of the tooth
Histopathological features Cemental tissue with reversal lines	Histopathological features Presence of dentin and pulp also	Histopathological features Presence of only cemental tissue
Common features	**FCOD**	**Cementoblastoma**
Clinical features Both cementoblastoma and FCOD are periapical, sclerotic, sharply marginated lesions with a low-attenuation halo	Clinical features Reactive lesion usually asymptomatic More common in the 4th or 5th decade of life Does not fuse to tooth roots Radiographic features Mature stage is radio-opaque with poorly defined margins Histopathological features Bone and cementum like tissue	Clinical features Neoplastic lesion that maybe associated with pain and bony expansion More common in children and young adults Fuses with the tooth roots Radiographic features Lesion is radio-opaque with well-defined radiolucent margins Histopathological features Only cemental tissue
Common features	**Condensing Osteitis**	**Cementoblastoma**
Clinical features Both occur in younger age group Both are usually seen in premolar molar region Both are sclerotic lesions	Radiographic features Periapical, poorly marginated, nonexpansile, sclerotic lesion associated with a carious nonvital tooth, and it may be unifocal or multifocal It does not show a peripheral radiolucent rim The adjacent tooth usually has a thickened PDL space or periapical inflammatory lesion (e.g., granuloma, cyst or abscess)	Radiographic features Periapical, sharply marginated, expansile and sclerotic lesion Shows a peripheral radiolucent rim No thickening of the PDL space Tooth is vital
Common features	**Hypercementosis**	**Cementoblastoma**
Clinical features Both appear as periapical radiopacities	Clinical features No clinical signs or symptoms	Clinical features Painful swelling at the buccal and lingual/palatal aspect of the alveolar ridges; occasionally, it may be asymptomatic
	Radiographic features The radiolucent shadow of the periodontal membrane and the radiopaque lamina dura are always seen on the outer border of hypercementosis, enveloping it as seen in normal cementum	Radiographic features The calcified mass is attached to the tooth root, with loss of root contour due to root resorption and fusion with the tumor
Histopathological features Disproportional acellular cementum deposit attached to the root of the tooth, associated with a thin connective tissue	Histopathological features Absence of active cementoblasts	Histopathological features Presence of active cementoblasts

FCOD: Focal cemento-osseous dysplasia, PDL: Periodontal ligament

Osteoblastoma, odontoma, focal cement osseous dysplasia (FCOD), condensing osteitis and hypercementosis could be considered in differential diagnosis of benign cementoblastoma.[30-32] An attempt has been made to compare and contrast aforementioned lesions with benign cementoblastoma on clinical, radiological and histopathological features [Table 2].

The treatment includes removal of the tumor en masse with the affected tooth. If the tumor is incompletely removed,

the recurrence rate is as high as 37.1%.[4] The prognosis of the tumor is excellent if it is removed in toto as then there are very minimal chances of recurrence.[33] In the present case also, the lesion was excised along with the extraction of primary first molar and a 6-month follow-up showed no recurrence. There are no reported cases of malignant transformation in benign cementoblastoma till date.

CONCLUSION

The occurrence of cementoblastoma in association with primary teeth is extremely rare (seriously rare). However, it is important to include these lesions in the differential diagnosis of bony lesions in association with tooth roots. Although there are no reported cases of malignant transformation of benign cementoblastoma (rarely serious), there are reported cases in literature exhibiting signs of local aggressiveness and destruction.[34] Also, there seems to be difficulty many a times in differentiating bone from cementum and hence distinguishing cementum-related lesions (benign cementoblastoma, hypercementosis and cement ossifying fibroma) from those related to bone (FCOD, condensing osteitis, cement ossifying fibroma and osteoblastoma). Thus, previous study showing modified Gallego Stain in distinguishing cementum (brilliant red) from bone (green) maybe valuable in concluding the diagnosis where dilemma exists so as to render appropriate treatment and have better patient compliance.[35]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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