

Cementoblastoma Solely Involving Maxillary Primary Teeth- A Rare Presentation

Aadithya B Urs*/ Hanspal Singh**/ Garima Rawat***/ Sujata Mohanty****/ Sujoy Ghosh*****

Cementoblastoma is a rare benign neoplasm of odontogenic ectomesenchyme origin, involving the roots of any tooth, which occurs predominantly in second and third decade of life. Very few cases of cementoblastoma associated with a primary tooth or having a maxillary presentation have been reported in the past. Here, a rare case of a ten year old boy who presented to the department with a swelling in maxillary posterior region since one month is being discussed. The radiographic presentation was mimicking an odontoma. The final diagnosis was cementoblastoma. We have advocated the use of polarized microscopy to support the histopathological diagnosis with respect to its cemental origin. Cementoblastoma should be considered in the differential diagnosis of radio-opaque lesions in the transitional dentition.

Key words: Cementoblastoma, maxilla, primary dentition

INTRODUCTION

Cementoblastoma is a rare benign neoplasm of odontogenic ectomesenchyme origin representing only 1–8% of all odontogenic tumors.¹ It is characterized by proliferating cementum-like tissue occurring in relation to the roots of erupted teeth.^{2, 3} It occurs predominantly in second and third decade of life and more often in the posterior region of the mandible.⁴ Very few cases of cementoblastoma associated with a primary tooth or having a maxillary presentation have been reported in the past.^{5, 6} We present an unusual case of cementoblastoma solely involving primary maxillary teeth. To the best of our knowledge, this appears to be the first case being documented in literature.

* Dr. Aadithya B. Urs, M.D.S., Professor & Head, Department of Oral & Maxillofacial Pathology, Maulana Azad Institute of Dental Sciences, New Delhi, India.

**Dr. Hanspal Singh, M.D.S., Senior Resident, Department of Oral & Maxillofacial Pathology, Maulana Azad Institute of Dental Sciences, New Delhi, India.

***Dr. Garima Rawat, B.D.S.(Post Graduate Student) Department of Oral & Maxillofacial Pathology, Maulana Azad Institute of Dental Sciences, New Delhi, India.

****Dr. Sujata Mohanty, M.D.S., Professor & Head, Department of Oral & Maxillofacial Surgery, Maulana Azad Institute of Dental Sciences, New Delhi, India.

***** Dr. Sujoy Ghosh, MDS Associate professor Department of Oral Medicine and Radiology Maulana Azad Institute of Dental Sciences, New Delhi, India.

Send all correspondence to:

Aadithya B Urs, Professor and Head,
Department of Oral & Maxillofacial Pathology,
Maulana Azad Institute of Dental Sciences, New Delhi, India
BSZ Marg, New Delhi-110002
Phone :-+91-9582948857
E-mail : draadithyaburs@gmail.com

Case Report

A ten year old boy reported to the outpatient department with the chief complaint of swelling in the upper back tooth region since one month. The swelling had gradually increased in size since then. The patient was in good general health. On extraoral examination, slight facial asymmetry was observed. The swelling on right side of face was bony hard and tender on palpation. Intra-oral examination revealed a well-defined bony hard swelling in the posterior maxilla which was approximately 2 cm x 1.5 cm in size (Figure 1A). The swelling was present with respect to primary maxillary right first and second molars causing expansion of the buccal cortical plate and obliteration of the buccal vestibule. The overlying mucosa was normal, and all associated teeth were immobile and symptom-free.

Orthopantomogram revealed a well-defined radiopaque lesion measuring 2 cm x 1.5 cm associated with the roots of deciduous maxillary right first and second molar. The radio-opacity was surrounded by a radiolucent line (Figure 1B) and was pushing the tooth buds of developing premolars superiorly. A provisional diagnosis of odontoma was considered. The patient was operated under general anaesthesia and the tumor mass was surgically excised after raising a mucoperiosteal flap starting from 53 with its oblique arm on the buccal side. Bony expansion was present over the roots of 54, 55 in the posterior maxilla. The thin and expanded bone was removed making a window of about 2.5 cm diameter. A greyish white mass of about 2.3 cm diameter was seen attached to the roots of 54, 55 which was hard in consistency. The mass was well delineated from the surrounding healthy bone and could easily be shelled out along with extraction of 54, 55. Primary closure was done and the healing was uneventful.

The received specimen consisted of a bony hard tissue measuring 2.3 cm x 2 cm x 1.8 cm in size with the roots of right maxillary first and second deciduous molar embedded in it (Figure 2). The entire tissue was decalcified as a single specimen. Buccopalatal slices of the decalcified tumor mass disclosed a yellowish white tumor mass located in the apical portion of the above mentioned teeth (Figure 3). Four microns thin haematoxylin and eosin stained sections revealed the tumor mass to be attached to the apical and lateral portion of both the teeth (Figure 4). The lesional tissue composed of sheets of cementum like tissue exhibiting numerous basophilic reversal lines and uncalcified portion at the periphery. Centre of the tumors mass showed cemental agglomerate with cementocytes in lacunae (Figure 5 A, B). Van Gieson staining was performed on the sections which were studied under polarised microscope. The polarized light view revealed quilt like pattern of the tumor mass which is representative of cellular cementum (Figure 6 A, B). This suggested cemental origin of the tumor mass. A final diagnosis of cementoblastoma was made. Patient is on regular follow-up since 2 years and no recurrence is observed till date.

DISCUSSION

Benign cementoblastoma is a rare neoplasm of the jaws. It is classified by 2005 WHO classification as a tumor of ectomesenchyme origin, with or without inclusion of epithelium.⁴ They are defined as true neoplasm of cementum or cementum-like tissue formed on the tooth root by cementoblasts.^{5, 6} Nearly all benign cementoblastomas are associated with the roots of a single erupted permanent tooth and rarely occur in relation to the primary dentition.^{1, 3, 5, 7}

The present case showed an unusual occurrence of the tumor in the maxillary right premolar-molar region of a ten year old boy associated with two primary maxillary teeth (first and second molars). Cementoblastoma of primary tooth origin have usually been associated with mandibular second primary molars.^{4, 8} In the past literature, cementoblastomas involving multiple teeth have been reported to reflect high growth potential.² Table 1 shows previously reported eight cases of cementoblastoma in the primary dentition, out of which only one case was involving the maxilla. This was reported by Ohki *et al* (2003) in a 12 year old patient involving multiple

Fig 1: A) Intraoral photograph showing bony hard tender swelling wrt 54, 55 expanding buccal cortical plate causing obliteration of buccal vestibule. (Inset- Extraoral Photograph showing facial asymmetry)
B) OPG showing well defined radio-opacity surrounded by radiolucent line involving the right maxilla displacing tooth buds of permanent canine, first and second premolar.

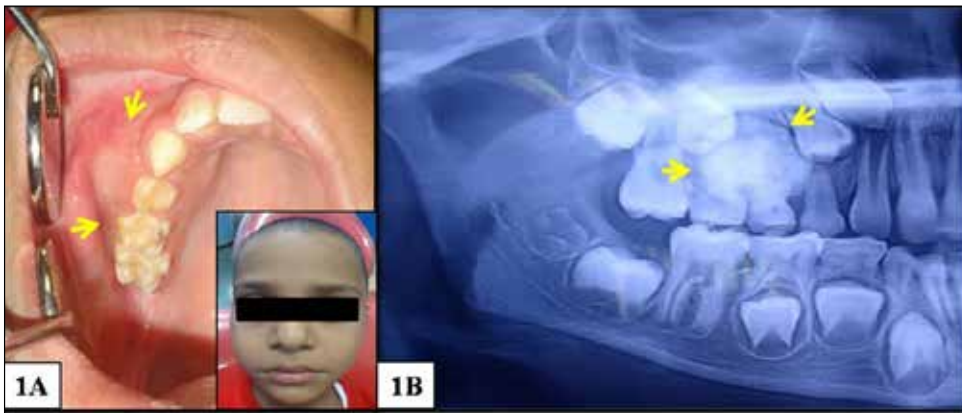


Fig 2: Gross examination showing hard tissue bit composed of teeth and bony hard mass surrounding the teeth.



Fig 3: Decalcified sliced sections showing tumor mass attached to the roots of maxillary first and second deciduous molar.



primary and permanent teeth (right maxillary second primary molar, first and second premolars, and first and second permanent molars).^[2] The present case involved only two primary maxillary teeth (first and second molars). To the best of our knowledge this is the first case of cementoblastoma solely involving maxillary primary teeth.

The radiographic appearance of the current case was that of a radioopaque mass surrounded by a radiolucent line. The differential diagnosis would comprise of odontoma, osteoma and probably an ossifying fibroma of mature type.^{9, 10} Odontoma appeared to be the most probable diagnosis, as these tumors are commonly seen in children with developing dentition. Due to the radio-opaque nature osteoma was also considered though these tumors are seen in the second to third decade.

Benign cementoblastoma is histopathologically characterized by the formation of sheets of cementum-like tissue having many reversal lines, irregular lacunae and cellular fibrovascular stroma. The periphery of the mass or the more-active growth areas are often unmineralized. This tumor may sometimes histologically resemble osteoblastoma, osteoid osteoma, or atypical osteosarcoma and is difficult to distinguish from these tumors.^{5, 7} The only distinguishing feature can be association of cementoblastoma to the root of the tooth, which is not distinctively seen in the other mentioned tumors, but separating the lesions on the basis of only histopathological findings would prove to be difficult if adequate clinical details are not provided.^{7, 8}

Fig 4: Stereomicroscopic view showing tumor mass attached to the tooth.

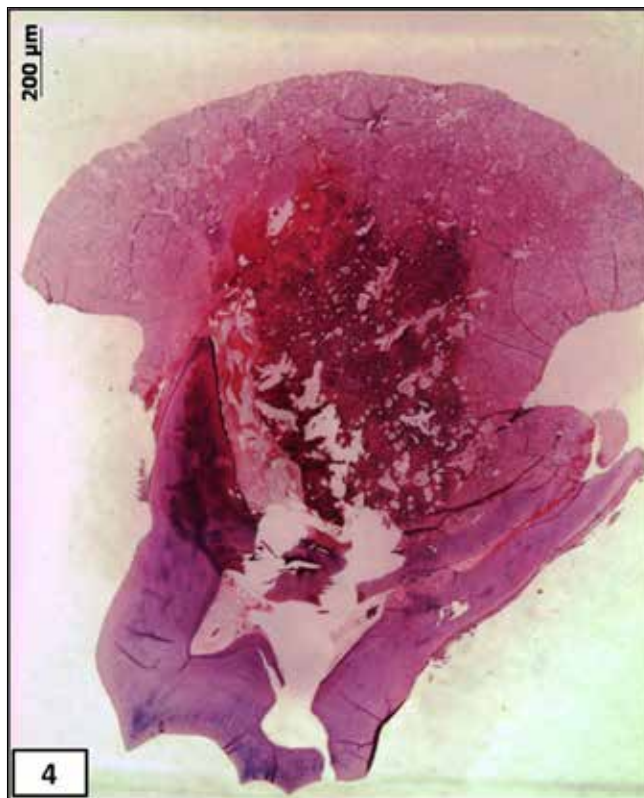


Fig 5: A) Tumor mass attached to the apical portion of tooth (H&E 10X). Arrow heads delineating the junction between the tooth and the cemental mass.

B) Tumor mass composed of cemental agglomerate with cementocytes in lacunae and numerous reversal lines (H&E 40X).

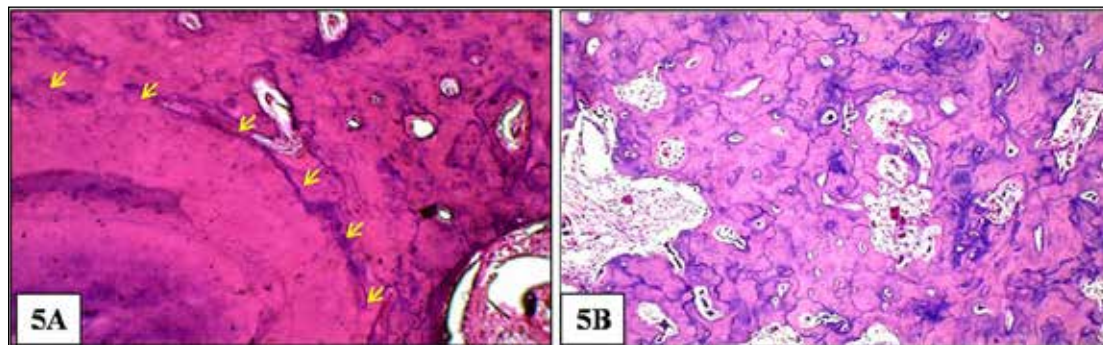
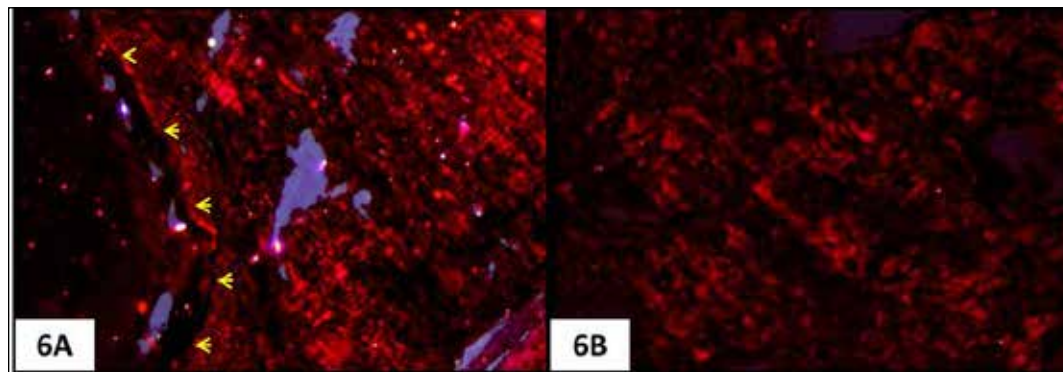


Fig 6: A) Van Gieson stained sections delineating the junction between the tooth and the cemental mass (Polarised light view).

B) Van Gieson stained sections showing quilt like pattern representative of cellular cementum (Polarised light view).



Tissue sections studied under polarized microscope to ascertain the origin can aid in diagnosis. As the pattern and width of collagen fibers in bone and cementum are different, hence, unique patterns will be seen under the polarized microscope. It has been established in the literature, that cementum has an intrinsic collagen bundle width and pattern that are unique when compared to bone. In cellular cementum, three basic patterns can be seen: parallel-fibered, quilt-like and coalescing globule patterns. In acellular cementum, brush border pattern is seen as the extrinsic fibers of periodontal ligament get inserted. Lamellar bone and woven bone show thick parallel- fibered pattern and random arrangement respectively.^{11, 12, 13} In the present case, the decalcified tissue sections stained with Van Gieson stain were studied under polarized microscope. The cementum like tissue revealed quilt like pattern representative of cellular cementum thus, suggesting cemental origin of the tumor. Hence, it would be appropriate to advocate polarised microscopy as an adjunctive aid to diagnosis in radioopaque lesions of odontogenic origin and fibro-osseous lesions of the jaws.

As benign cementoblastomas have unlimited growth potential, preferred treatment is complete surgical excision with extraction of the associated teeth, even though the pulp may be vital. Recurrence is rare if the mass is totally removed. However, incomplete excision of the lesion is usually followed by recurrence.² Cementoblastoma involving multiple teeth show high growth potential, but no recurrences have been reported.

CONCLUSION

Cementoblastoma occurring in association with primary maxillary teeth is exceedingly rare. It should be considered in the differential diagnosis of radio-opaque lesions in the developing mixed dentition. Also, polarized microscopy can be used as an adjunct in arriving at a confirmatory diagnosis.

Table 1

AUTHOR	YEAR	AGE (Y)/SEX	LOCATION	SIZE (CM)	PAIN	RECURRENCE
Chaput and Marc	1965	10/F	Right mandibular first premolar and second deciduous molar	2.1	-	ND
Vilasco et al	1969	8/F	Right mandibular second deciduous molar	ND	+	ND
Zachariades et al	1985	7/F	Right mandibular first and second deciduous molars, and first permanent molar	3.0	-	-
Herzog	1987	7/F	Left mandibular first and second deciduous molars	1.4	-	ND
Papageorge et al	1987	6/M	Mandibular deciduous central incisors	4.5	+	-
Cannell	1991	8/F	Right mandibular second deciduous molar	ND	+	ND
Schafer et al	2001	8/F	Right mandibular second deciduous molar	1.5	+	-
Kousuke Ohki et al	2003	12/M	Right maxillary second deciduous molar, first and second premolars, and first and second permanent molars	3.3	+	-
Present case		10/M	Right maxillary deciduous first and second molars	2.1	-	-

REFERENCES

1. Harada H, Omura K, Mogi S, Okada N. Cementoblastoma Arising in the Maxilla of an 8-Year-Old Boy: A Case Report. *Int J Dent*. doi:10.1155/2011/384578 2011.
2. Ohki K, Kumamoto H, Nitta Y, Nagasaka H, Kawamura H, Ooya K. Benign cementoblastoma involving multiple maxillary teeth: Report of a case with a review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 97(1): 53-8, 2004.
3. Sumer M, Gunduz K, Sumer AP, Gunhan O. Benign cementoblastoma: A case report. *Med Oral Patol Oral Cir Bucal* 11: E483-5, 2006.
4. Souza Laís de Almeida Sena, Cardoso Juliana Andrade, Silva Viviane Palmeira da et al. Cementoblastoma affecting the maxilla of a pediatric patient: A case report. *Rev port estomatol med dent cir maxilofac*. 54(1): 43-47, 2013
5. Vieira AP, Meneses JM Jr, Maia RL. Cementoblastoma related to a primary tooth: a case report. *J Oral Pathol Med* 36: 117-9, 2007.
6. K Lemberg, J Hagstro, J Rihniemi and K Soikkonen. Benign cementoblastoma in a primary lower molar, a rarity. *Dentomaxillofac Radiol*. 36: 364-366, 2007.
7. Slootweg PJ. Cementoblastoma and osteoblastoma: a comparison of histologic features. *J Oral Pathol Med*. 21: 385-9, 1992.
8. Papageorge MB, Cataldo E, Nghiem FT. Cementoblastoma involving multiple deciduous teeth. *Oral Surg Oral Med Oral Pathol* 63: 602-5, 1987.
9. Woldenberg Y, Nash M, Bodner L. Peripheral osteoma of the maxillofacial region. Diagnosis and management: a study of 14 cases. *Med Oral Patol Oral Cir Bucal* 10(2): E139-42, 2005.
10. Chang CC, Hung HY, Chang JY, Yu CH, Wang YP, Liu BY, Chiang CP. Central Ossifying Fibroma: A Clinicopathologic Study of 28 Cases. *J Formos Med Assoc*. 107(4): 288-94, 2008.
11. Giansanti JS. The pattern and width of the collagen bundles in bone and cementum. *Oral Surg Oral Med Oral Pathol*. 30(4): 508-14, 1970.
12. Waldron CA, Giansanti JS. Benign fibro-osseous lesions of the jaws: A clinical- radiologic- histologic review of sixty five cases. *Oral Surg Oral Med Oral Pathol*. 35(2): 190-201, 1973.
13. Hamner JE, Scofield HH, Cornyn J. Benign fibro-osseous jaw lesions of periodontal membrane origin. An analysis of 249 cases. *Cancer*. 22(4): 861-78, 1968.