BENIGN CEMENTOBLASTOMA INVOLVING MULTIPLE DECIDUOUS AND PERMANENT TEETH OF THE MAXILLA - A CASE REPORT

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Abstract

Benign Cementoblastoma is a rare tumor of Odontogenic ectomesenchyme origin characterized by proliferation of cementum- like tissue. The neoplasm is closely related to and partly surrounds the roots of teeth. In most cases, this lesion is asymptomatic and was considered as an indolent lesion for several years. Nevertheless, recent studies have demonstrated that some cementoblastoma may exhibit an aggressive biological behavior and can affect several teeth. An unusual case of benign cementoblastoma that was clinically extensive, involving multiple deciduous and permanent teeth is presented. This report also corroborates with some papers that have demonstrated the aggressive behavior of cementoblastomas.
Key words: Cementum- like, aggressive, maxillary teeth, recurrence

Introduction

Cementoblastoma, a rare ectomesenchymal odontogenic tumor, was first described by Dewey as early as in 1927.[1] The first report of cementoblastoma was made in 1930 by Norberg in a dissertation on odontomes.[2] Only 1% of the odontogenic tumors accounted for cementoblastoma.[3, 4] This neoplasm is usually encountered in the second or third decade of life and is more prevalent among males.[5] It is commonly seen in mandibular premolar-molar region.[5-8] Histologically, this lesion is composed of functional cementoblasts forming large masses of cementum or cementum-like tissues in juxtaposition to the involved teeth roots.[5] Although cementoblastomas have been reported in literature several times, very few reports have been published wherein multiple teeth are involved.[6, 9-13]

Here, we present a case of an unusual cementoblastoma involving multiple maxillary teeth in a 7-year old girl, which showed recurrence after 2 years of excision.

Case Report

A seven-year old girl was reported to the department of Oral Medicine with a swelling in the left maxillary region. Seven months back she had developed toothache in the region of the deciduous second molar for which she had consulted a local dentist. Swelling subsided completely after extraction of the offending tooth. However, swelling developed again in the same region a month

and a half ago which gradually increased to the present size. Swelling was insidious in nature and not associated with pain, fever, trauma, bleeding or pus discharge. There were no aggravating or relieving factors for the swelling. Her medical and family history was non-contributory. The physical examination showed that she was a well-developed and well-nourished girl.

Extra-oral examination revealed a diffuse swelling in the middle 3rd of the left side of the face. It extended from infra-orbital rim to 2 cm above the lower border of mandible and mesio-distally from naso-labial fold to 4 cm laterally. The size of swelling was approximately 4cm X 5 cm. On palpation, the swelling was firm to hard, overlying skin was normal and there was no local rise of temperature. Intra-orally, a diffuse large swelling was present on the palatal aspect of deciduous teeth and permanent first molar till midline. It did not cross the midline.

An intraoral periapical radiograph of the tooth showed a radiopaque mass associated with the Maxillary left molars and the permanent first molar (Fig 1). Panoramic radiograph showed a 1.5 cm radio-opaque lesion in the periapical region corresponding to 65,26. Small flecks of radio-opacities were seen superiorly within an unilocular radiolucency (Fig 2). Occlusal view showed a radio-opaque mass in the region of the deciduous first and second molar (Fig 3) Radiograph of specimen revealed a large radio-opaque mass (1.5-2.0 cm) attached to roots (Fig 4). The extracted (by the local dentist) maxillary left deciduous second molar along with the calcified mass attached to its root and which the patient had brought was sent for histopathological evaluation. Surgical excision was carried out and the mass was removed and packed with BIPP (Bismuth Iodoform Paraffin Pack). The permanent first molar was also removed and tissue around the tooth was curetted.

Microscopic examination of the decalcified section of the E with the calcified mass revealed cementum-like tissues attached to the tooth root. The tissue mass had numerous deeply staining hematoxyphilic lines. Many cells entrapped within lacunae were evident throughout the tissue. Thus, the incisional biopsy was suggestive of benign cementoblastoma (Fig 5).

The decalcified section of the permanent first molar with hard tissues attached to the root showed a tumor mass was made up of sheets of mineralized tissues that was arranged in a trabeculae pattern. Irregular lacunae, numerous lines and few blood vessels were present throughout the mineralized matrix (Fig 6). The adjacent stroma resembled that of dental papilla (Fig 7). Hence the histopathological features of excisional biopsy confirmed the diagnosis of benign cementoblastoma.

After 2 years, the patient returned with painful swelling in the same region of 3X 3 cm dimension. A CT scan showed lytic expansion of 3.4 cm X 3.3 cm X 3.0 cm of left maxilla involving anterior wall of left maxillary sinus and inferiorly extending up to median palatal aspect of left maxillary alveolar process with osseous matrix within the lesion (Fig 8).

Enucleation of the tumor under GA was done and the excised mass was sent for histopathological evaluation. The microscopic appearance of the tissue showed interlacing sheets of amorphous basophilic material resembling cementum. This area was lined by focal aggregates of plump cementoblasts (Fig 9) The microscopic features of recurred lesion was suggestive of benign cementoblastoma.

Fig 1: Intraoral Periapical radiograph showing a radioopacity associated with the roots of the maxillary left deciduous and permanent molar teeth

Fig 2: Panoramic radiograph showing a 1.5 cm radio-opaque lesion in the periapical region corresponding to E 6. Small flecks of radio-opacities are present within an unilocular radiolucency.

Fig 3: Occlusal view showing a radio-opaque mass in the region of the deciduous first and second molar.

Fig 4: Intraoral Periapical radiograph of the extracted maxillary deciduous second molar shows a radioopaque mass in continuity with the root.

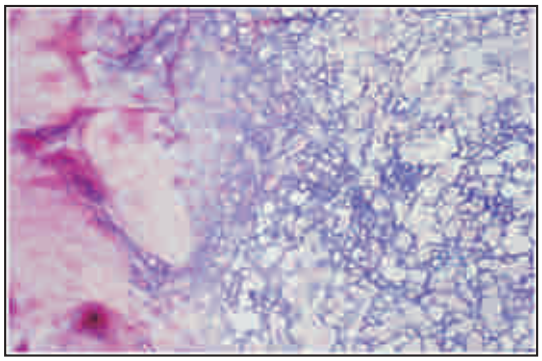

Fig 5: Decalcified section-showing cementum –like tissue attached to the root of the maxillary deciduous second molar with many lacunae containing entrapped cells. (H&E 40X)

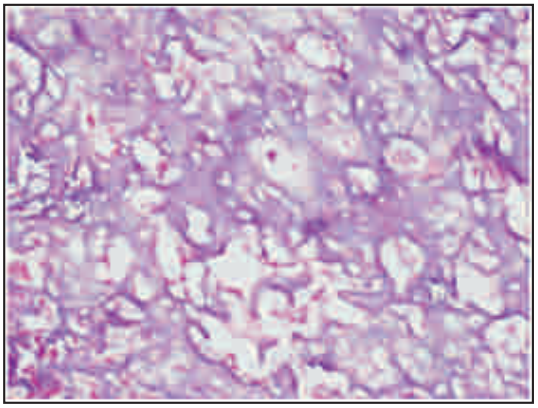

Fig 6: Decalcified sections showing interlacing sheets of amorphous basophilic material that resembled cementum. (H&E 40X)

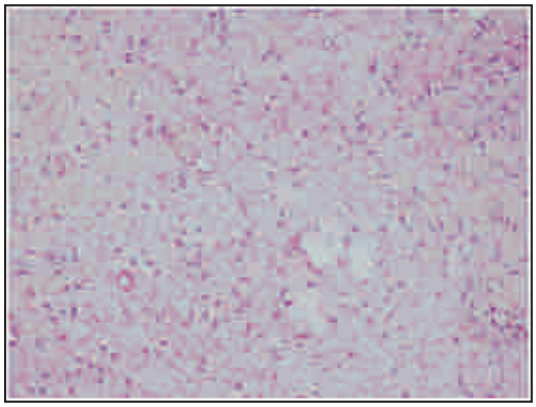

Fig 7: Stroma resembling dental papilla (H&E 40X)

Fig 8: A CT scan showed lytic expansion of 3.4 X 3.3 X 3.0 cm^3 of left maxilla involving anterior wall of left maxillary sinus and inferiorly extending up to median palatal aspect of left maxillary alveolar process

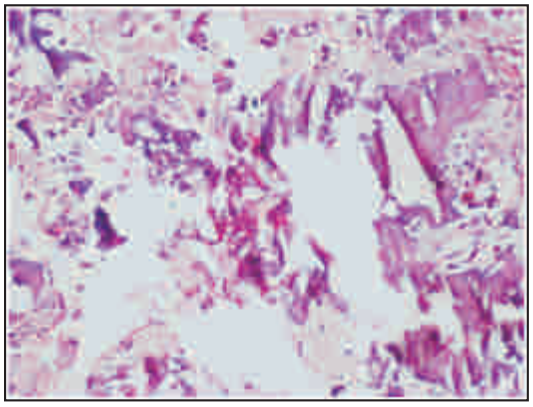

Fig 9: Decalcified section showing interlacing sheets of amorphous basophilic material resembling cementum. (H&E 40X)

Discussion

Benign cementoblastoma is a tumor of the jaws defined as true neoplasm of cementum or cementum-like tissues formed on the tooth root by cementoblasts.[5] This tumor is predominantly seen in young adults with 50% of them occurring below the age of 20 years.[6] Although no significant sex predilection between genders is reported[10], but few authors have found that males are more frequently affected.[5,14] Nearly all benign cementoblastoma usually affect the mandible with permanent first molar being the most commonly affected tooth. [5,14]

In our patient, the neoplasm occurred in the left posterior maxilla involving multiple teeth in a 7-year old girl child. This case is presented because, to our best knowledge, there are only 3 reported cases involving both the dentitions [9,10,13] and the present case is the patient is the youngest among those reported so far.

Clinically, it is considered to be slow growing asymptomatic lesion which may be associated with pain and swelling[5, 7, 15, 16] . However, few cases have shown some signs of local aggressiveness and destruction.[10, 12, 14, 17 &18]

Local aggressiveness in the form of bone expansion with maxillary sinus involvement was noted in the present case. It has been stated that the radiographic features of the lesion is pathognomic and comprises of a radio-opaque mass often fused with roots of involved tooth and is lined by a radiolucent rim.[6]

Microscopically, benign cementoblastoma is characterized by formation of sheets of cementum-like tissues which may have reversal lines and the unmineralized areas at the periphery or in the active growth areas.[5] From the histopathologic view, one of the most persistent confusion is the strong resemblance between cementoblastoma and osteoblastoma. In this case, all the microscopic sections were

reviewed thoroughly and the intimate relationship of the tumor mass with the involved teeth justified its separation from osteoblastoma.

Abrams *et al* recommended that the suffix benign employed with cementoblastoma is redundant because report of malignant benign cementoblastoma or malignant transformation in benign cementoblastoma has not been reported.[18] Later, Langdon described the first case of large benign cementoblastoma which had an aggressive behavior.[19] Many authors do believe that the tumor had an apparently unlimited growth potential with the average growth rate 0.5 cm per year[20,21]. Although, a number of benign cementoblastomas have been documented in literature before 1974 there was lack of a well-defined criteria. The fewer tendencies to recur and short follow-up period also explains the limited documentation regarding its clinical behavior. However, recently Brannon *et al* reviewed and observed a recurrence rate of approximately 37% as compared to the earlier reported rate of just 5 %[8].

Mader and Wadenburg recommended complete excision of the tumor and removal of tooth because of its unlimited growth potential.[22] Brannon *et al* reported recurrence is more likely if curettage without extraction of involved teeth is done.[8] Thus, recommended treatment is complete resection of tumor with enucleation and extraction of associated teeth followed by curettage and peripheral osteotomy. Brannon *et al* also mentioned that the lesions that affected younger individuals were large in size with more signs of aggressiveness noted to be recurred more. The time–interval for the tumor to recur ranges from 4-24 months with a mean interval of 15 months.[8] Even in the present case, the lesion recurred within 2 years and the recurred lesion was removed by subtotal maxillectomy as suggested by Agazzi[23] . This child has been followed-up for more than 2 years now and there had been no sign of recurrence. Therefore, although considered a benign entity, benign cementoblastoma possess a growth potential that that can cause complete destruction of the involved area. Owing to the high possibility of recurrence of this lesion, radical treatment and close patient follow-up is warranted.

Conclusion

This article reports a case of benign cementoblastoma that involved both the deciduous and permanent teeth in a seven-year old girl. Although a benign lesion, it can involve multiple teeth and cause destruction of the surrounding area. As the lesion has a high tendency to recur, radical surgical excision and close follow-up for a long period of time is recommended.

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