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Benign Cementoblastoma- Repeated Recurrence: A case report

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ABSTRACT: A 16 year old male presented with pain and swelling in the right posterior mandible that lasted for several months. The radiographic features of the lesion included a radiopaque lesion attached to the root of right mandibular first molar. The histopathological report confirmed the diagnosis of benign cementoblastoma followed by surgical excision of the same along with the tooth involved. The lesion recurred after 1 year of treatment. Radiographically there was diffuse swelling on the right angle of the mandible which was soft, tender and fluctuant. The tooth 46 was missing. Buccal sulcus was obliterated and buccal expansion was noted. The lesion was diagnosed as recurrence of the cementoblastoma lesion. Wide excision of the lesion along with marginal mandibulectomy was done on the right side. The same patient again reported to the clinic with a swelling on the same side after 6 months. Histopathological report suggested the recurrence of cementoblastoma.

KEYWORDS: Odontogenic tumour, Dental Cementum, Benign Cementoblastoma, Recurrence, Cone Beam Computed Tomography

I. INTRODUCTION

The benign cementoblastoma or true cementoma was first described by Norberg in 1930. It is a rare benign odontogenic tumor of ectomesenchymal origin. It is considered to be the only true neoplasm of cemental origin and is characterized by the proliferation of cellular cementum. The benign cementoblastoma is probably a true neoplasm of functional cementoblasts which form a large mass of cementum or cementum-like tissue on the tooth root.[1] The lesion is frequently microscopically indistinguishable from the benign osteoblastoma or giant osteoid osteoma. [2] In the past, this tumor has been considered to exhibit a very low recurrence rate[3]. In this case report we are presenting a rare case of cementoblastoma with recurrence.

II. CASE REPORT

This is a case report of 16-year-old male patient who reported to the Department of Oral Pathology, Govt. Dental College, Trivandrum with the chief complaint of pain and swelling on the right lower side of the face for more than two weeks(Figure 1). Patient noticed a swelling in the right back tooth region (46) which was gradually increasing in size and went to private clinic for the treatment of the same. After taking radiograph it was diagnosed as cementoblastoma and the tooth 46 was extracted along with the lesion, followed by placement of bone graft in the same site(Figure 2 & 3).

FIGURE 1 : Extra oral swelling of right side

FIGURE 2: Biopsy specimen with tooth 46

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FIGURE 3: Pre-extraction radiograph showing radiopaque lesion attached to roots

After the procedure, pain subsided but the swelling remained the same. After six months the patient was referred to the department of Oral and Maxillofacial Surgery, Govt. Dental College, Trivandrum. On examination, there was a diffuse swelling on the right angle of the mandible which was soft, tender and fluctuant. The tooth 46 was missing. Buccal sulcus was obliterated and buccal expansion was noted. Right submandibular node was palpable which was soft and tender. Radiological examination revealed expansion of hemi mandible with areas of cortical breach, well defined central sclerotic area involving the body of right hemi-mandible adjacent to the alveolar margins between the apex of second premolar and molar teeth. From the radiological findings of expansion and thinning of right hemi-mandible with well-defined central sclerotic area it was suggested as tumor recurrence(Figure 4,5,6). Biopsy was taken from the right lower molar region in relation to the extracted socket and reported as benign cementoblastoma.

Patient underwent wide excision of the lesion with marginal mandibulectomy of the right mandible for the same and patient was under regular follow up. After 6 months patient again reported with pain on the surgical site. Further investigations revealed the recurrence of cementoblastoma.

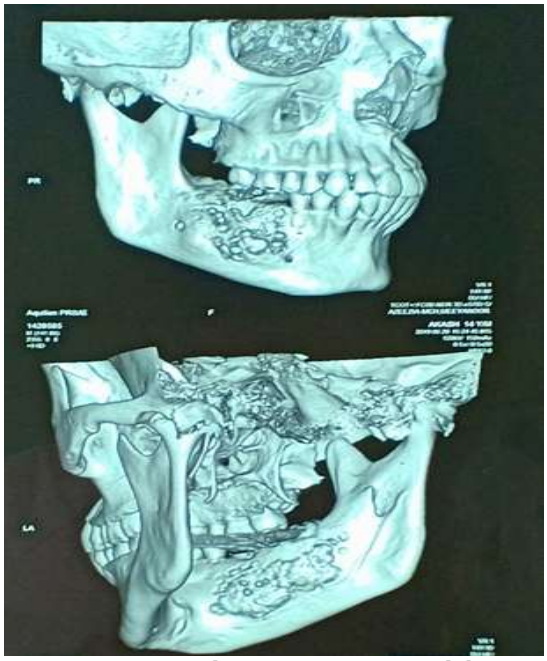

FIGURE 4: CBCT showing expansion and thinning of right hemi mandible with well defined central sclerotic area and areas of cortical breach

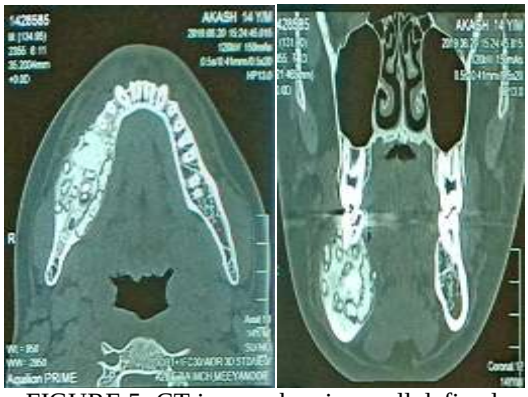

FIGURE 5: CT image showing well defined radiopaque lesion with intervening radiopaque flecks surrounded by radiolucent zone in the body of mandible

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Figure 6: OPG showing extensive lesion in the body of mandible right side with central sclerotic area

Histopathological examination of the present case revealed sheet like mineralized material resembling cementum with irregularly placed lacunae containing cells and prominent reversal lines with intervening fibro vascular stroma. The recurrent cases also presented with the same histopathologic features(Figure 7,8,9).

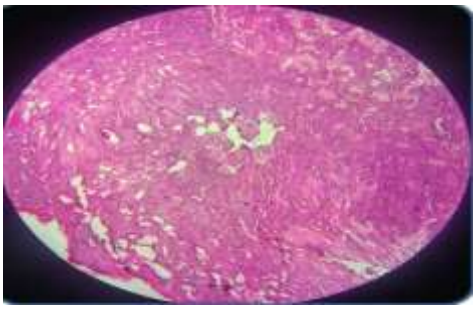

Figure 7

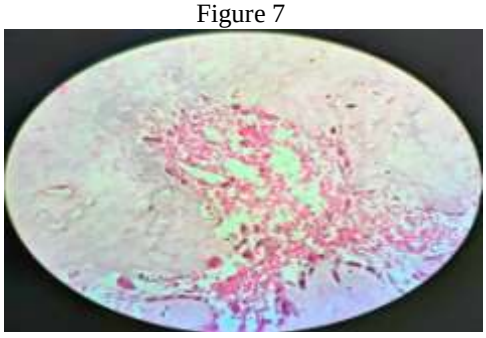

Figure 8

FIGURE 7,8: Histopathology showing sheet like mineralized material with irregularly placed lacunae containing cells and prominent reversal lines with intervening fibro vascular stroma.

III. DISCUSSION

Cementoblastomas are rare neoplasms, representing less than 1% of all odontogenic tumors.[3] It was first described by Dewy in the year 1927[4] and was described by Norberg in 1930 as a slow growing, benign odontogenic tumor arising from cementoblast.[5] Cementoblastomas in the current World Health Organization classification of odontogenic tumor, is in the category of tumors of mesenchymal origin.[6] It is considered to be the only true neoplasm of cemental origin and is characterized by the proliferation of cellular cementum.[1] The benign Cementoblastoma occurs most frequently under the age of 25 years, with no significant gender predilection. More than half of the tumors have occurred in persons under 20 years of age, although the range has been 10–72 years. Mandible is affected three times more frequently than the maxilla and the mandibular first permanent molar is the most frequently affected tooth[2] and few cases were reported to involving the deciduous dentition.[7] A vitality test of teeth involved in the process is generally positive.[1]

The present case was reported in a 16-year-old male patient in relation to extracted 46. Involvement of this mandibular first molar has accounted for approximately 50% of all reported cases. Pain and swelling are present in approximately two thirds of reported patients[2] and in the present case also patient presented with pain and swelling of the right lower back tooth region of 2 weeks duration. Clinical examination of the case revealed a tender, diffuse, soft and fluctuant swelling on the right angle of the mandible and 46 was missing with a history of extraction of the same 6 months back. Obliteration of the buccal sulcus and expansion of the buccal cortical plate noted. Right submandibular node was palpable which was soft and tender. Although most investigators consider the Cementoblastoma to represent a rather innocuous neoplasm, signs of locally aggressive behaviour may be observed, including displacement of adjacent teeth, envelopment of multiple adjacent teeth, maxillary sinus involvement, infiltration into the pulp chamber and root canals, cortical erosion and bony expansion[3] as in the present case. Little is known about the pathogenesis of this lesion.

They are odontogenic tumors, derived from ectomesenchymal cells of the periodontium, including cementoblasts. It is thought to evolve in three stages. The first stage is characterized by periapical osteolysis, followed by a cementoblastic stage, and then an inactive stage of maturation and calcification and is considered a neoplasm with unlimited growth potential. Its etiology is unknown, and trauma does not seem to play a role.[1] Radiographically, tumor mass is attached to the tooth root and appears as a well-circumscribed dense radiopaque mass often surrounded by a thin, uniform radiolucent line. The outline of the affected root is generally obliterated because of

resorption of the root and fusion of the mass to the tooth.[3,2] Radiological examination of the current lesion revealed expansion of hemi mandible with areas of cortical breach, well defined central sclerotic area involving the body of right hemi-mandible adjacent to the alveolar margins between the apex of second premolar and molar teeth. Expansion and thinning of right hemi-mandible with well-defined central sclerotic area suggested it as tumor recurrence.

Histopathologically the main bulk of the tumor mass is composed of sheets of cementum-like tissue, sometimes resembling secondary cellular cementum but other times being deposited in a globular pattern resembling giant cementicles. Reversal lines scattered throughout this calcified tissue are often quite prevalent. There is a variable soft-tissue component consisting of fibrillar, vascular and cellular elements. Many of the cemental trabeculae in area of activity are bordered by layers of cementoblasts. Away from these trabecular surfaces, cementoclasts may be evident. In such active areas, the lesion is frequently microscopically indistinguishable from the benign osteoblastoma or giant osteoid osteoma. In fact, some areas are so cellularly active that they bear strong resemblance to osteosarcoma. Multinucleated giant cells often are present. This calcified mass will be found united to the tooth root through obliteration of the periodontal ligament, resorption of portion of the root and replacement by the tumor tissue. The periphery of the tumor generally shows a soft tissue cellular layer resembling a capsule, corresponding to the radiolucent zone seen on the radiograph[1,2,3]. Histopathological examination of the present case revealed sheet like mineralized material with irregularly placed lacunae containing cells and prominent reversal lines with intervening fibro vascular stroma.

In 2002 Brannon et al, clinicopathologically analyzed 44 cases of cementoblastoma with a mean follow-up interval of 5.5 years and recurrence was documented in 13 cases with a recurrence rate of 37.1 %[8]. In the past, this tumor has been considered to exhibit a very low recurrence rate, although a recently reported series of a large number of cases suggests recurrences may be more common than previously thought, with an overall recurrence rate as high as 22%. Completeness of removal is most closely related to recurrence. Total removal of the mass and the associated tooth minimizes but does not completely eliminate the chance of recurrence. Progressive growth of the tumor after extraction of the involved tooth and incomplete removal of the mass has been documented[3]. The present case was recurred within one year, even after marginal mandibulectomy of the affected side.

IV. CONCLUSION

Although cementoblastoma has a benign nature, local aggressiveness and recurrence may occur. Hence, despite its rarity, it is very important to improve the awareness of this type of lesion among practitioners. Through this article we aim to provide an insight to dental practitioners regarding the clinical features, radiographic findings, histologic findings and treatment options of an aggressive benign cementoblastoma.

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