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Case Report
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Surgical management of central giant cell granuloma of mandible and prosthetic rehabilitation in a nine year girl: A case report

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Highlights

- Central giant cell granuloma is an idiopathic, osteolytic lesion of jaws.
- A rare case of large destructive CGCG involving anterior region of mandible in a nine-year girl is presented in this case report.
- The case was treated successfully by enucleation and curettage with satisfactory preservation of the continuity of mandible.

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Abstract

Central giant cell granuloma (CGCG) is an uncommon, benign, idiopathic, osteolytic lesion of jaws, histologically characterized by multinucleated giant cells distributed in fibrovascular connective tissue stroma. Accurate diagnosis of the lesion is essential for the successful management and the prognosis of this locally destructive lesion. In this paper, a rare case of large destructive CGCG involving anterior region of mandible, causing expansion of labial cortical plate and mobility of teeth in a nine-year girl is presented. It was treated successfully by enucleation and curettage with satisfactory preservation of the continuity of mandible. Nine months post operatively, the child was rehabilitated with a temporary partial denture to improve esthetics, phonetics and function. One year clinical and radiographic follow up showed new bone formation and no evidence of recurrence.

Keywords: Curettage; Giant cell; Granuloma; Rehabilitation

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INTRODUCTION

Central giant cell granuloma (CGCG) is an uncommon, idiopathic, osteolytic lesion of multinucleated giant cells. It is histologically benign but locally aggressive lesion of bone occurring most commonly in craniofacial skeleton.[1] The lesion was initially described by Jaffe in 1953 as a reparative reaction, later this term was discontinued because it is more destructive than reparative. The World Health Organization (WHO) has defined it as an intra-osseous lesion consisting of cellular fibrous tissue that contains multiple foci of hemorrhage, aggregations of multinucleated giant cells and occasionally trabeculae of woven bone.[2] It occurs most commonly during the first 30 years of life, with nearly 50% occurring in patients younger than 16 years of age. CGCG occurs more often in females than in males, the mandible is involved twice as often as the maxilla.[3]

CGCG is usually asymptomatic and it may present with aggressive growth and expansion of jaws. Radiographically, CGCG may vary from a unilocular radiolucency to a multilocular bone-destructive lesion with displacement of teeth, root resorption and cortical perforation. Sensory disturbance has been rare.[3,4] The lesion should be differentiated with other benign jaw lesions such as Brown's tumor, Ameloblastoma, aneurysmal bone cyst, odontogenic cysts, myxomas, fibrous dysplasia and ossifying fibroma.[5,6,7]

Histologically, the lesion is characterized by loosely arranged fibrous stroma with presence of multinucleated giant cells. These giant cells resemble osteoclasts with 20 or more nuclei.[5] Treatment for CGCG ranges from local enucleation and curettage to enbloc resection which can lead to large defects in the jaws. Recently for management of non-aggressive variants, intralesional steroids, systemic calcitonin, α-interferon and denosumab have been considered.[5,8,9,10] Oral rehabilitation after surgical intervention proves challenging and affected patients generally require prosthetic rehabilitation to facilitate function.[8] The potential for recurrence further complicates this process as recurrence rate[9] ranges from 11-49%. The purpose of reporting this case is to discuss the management of central giant cell granuloma involving mandible in a nine-year old female child followed by prosthetic rehabilitation.

CASE REPORT

A nine-year old girl reported with a chief complaint of swelling in the front region of lower jaw from one month. The swelling increased in size gradually. It was not associated with pain, difficulty in mouth opening and paresthesia. Extra oral examination revealed a solitary, diffuse swelling in symphysis area extending between the corners of the mouth (Figure 1). No abnormalities were reported with mouth opening. Skin over the swelling was normal. Swelling was hard in consistency and non-tender on palpation and it was not associated with lymphadenopathy. Intra-orally diffuse swelling in labial sulcus extending from teeth #31 to #84 region with obliteration of labial vestibule was observed. The mucosa over the swelling was stretched, no sinus opening or any discharge was present (Figure 1). The associated teeth 31,41, 42, 83, 84 showed grade-2 mobility. The swelling was bony hard in consistency and non-tender on palpation.

Panoramic radiograph showed a diffuse radiolucent area in the anterior region of mandible with displacement of incisors (Figure 2). Since the borders and extent of lesion was not clear, Cone beam computed tomography (CBCT) of anterior region of mandible was made. Various sections of CBCT showed unilocular radiolucency with expansion of labial cortical plate and resorption of roots of 31,41 (Figure 2). However, the lingual cortical plate was intact. The lesion measured 3x2cms in maximum dimensions on the orthopantomogram. nose, webbed neck, low posterior hair line, and with competent lips (Figure 2).

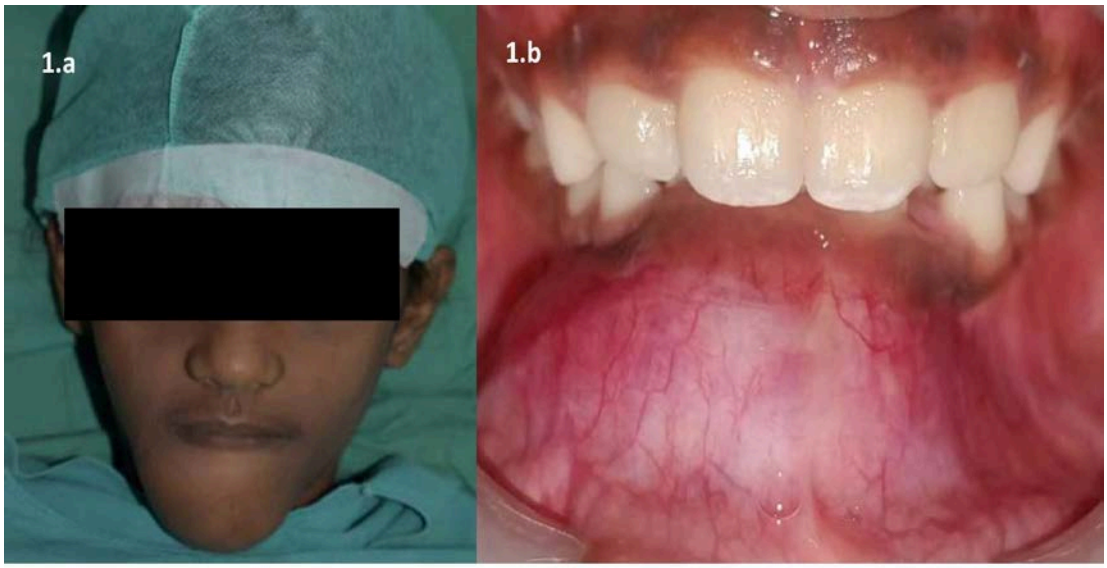

Figure 1. Clinical appearance of the lesion. 1.a: Extra-oral appearance; 1.b: Intra-oral appearance of the lesion showing obliteration of labial vestibule

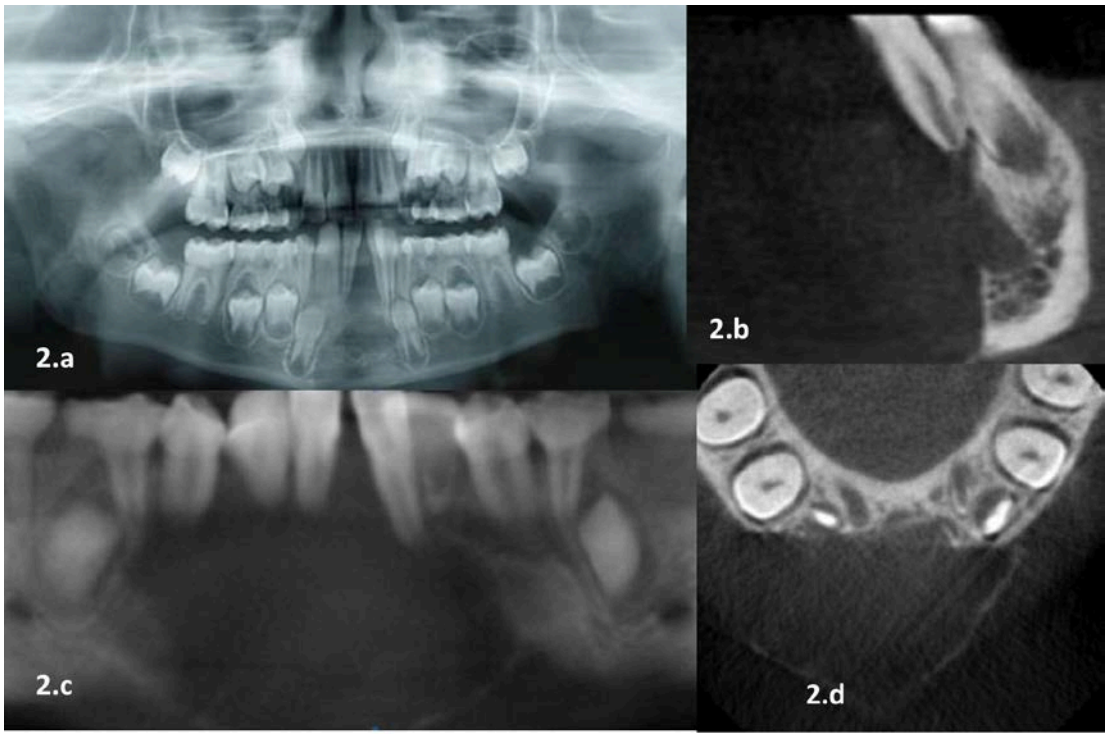

Figure 2. Radiographic appearance of the lesion 2.a: OPG showing diffuse radiolucency in the anterior region of mandible; 2.b, 2.c and 2.d: Lesion in various sections of CBCT

Procedure was explained to the parents and written consent was obtained for the treatment and publication of the case report. A provisional diagnosis of Central giant cell granuloma was made. The differential diagnosis was Odontogenic Cyst, Ameloblastoma, Brown's tumor and Aneurismal bone cyst. Biochemical investigations showed normal serum calcium, phosphorous, alkaline phosphatase and parathormone levels, ruling out the Brown's tumor. Incisional biopsy was performed which showed multinucleated giant cells surrounded by a loose fibrous stroma with areas of hemorrhage and inflammatory cell infiltrates (Figure 3).

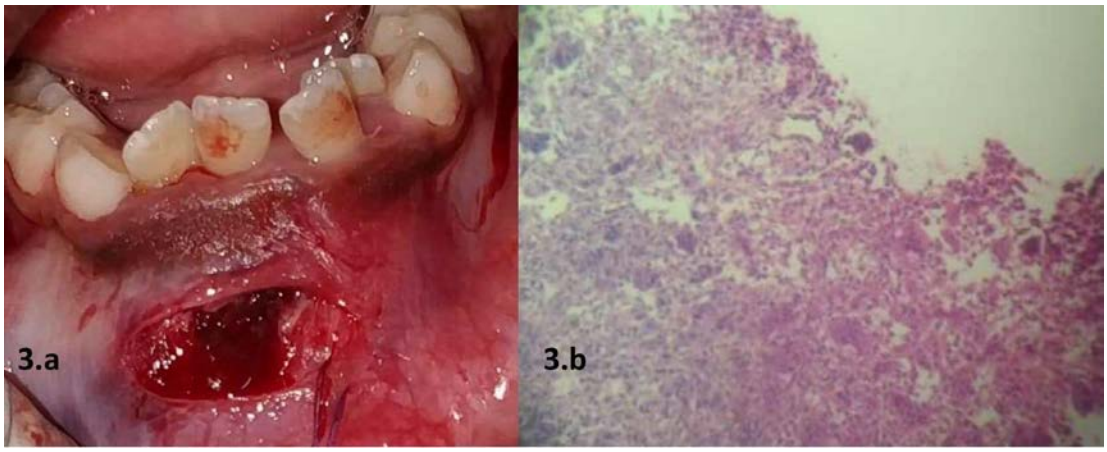

Figure 3. 3.a: Lesion during incisional biopsy; 3.b: Histopathological slide showing multinuclear giant cells in fibrous stroma

This confirmed the diagnosis of central giant cell granuloma. The child was referred to endocrinologist and with his opinion and also by considering the age of the patient, intralesional steroid was the initial chosen treatment modality.[10,11,12] Treatment was started with triamcinolone acetonide 10mg/ml and lignocaine 2% with adrenaline 1:200,000 (50:50), at the dose of 1ml solution for every 1cm of radiolucency, as determined on an orthopantomogram (OPG). The total dose of triamcinolone administered was 60mg over six weeks in increments of 6 injections of 10mg each. Clinical and radiological evaluation one week after the last dose showed no regression in radiolucency (Figure 4). Hence, with the parent's consent surgical enucleation and curettage[13] was planned.

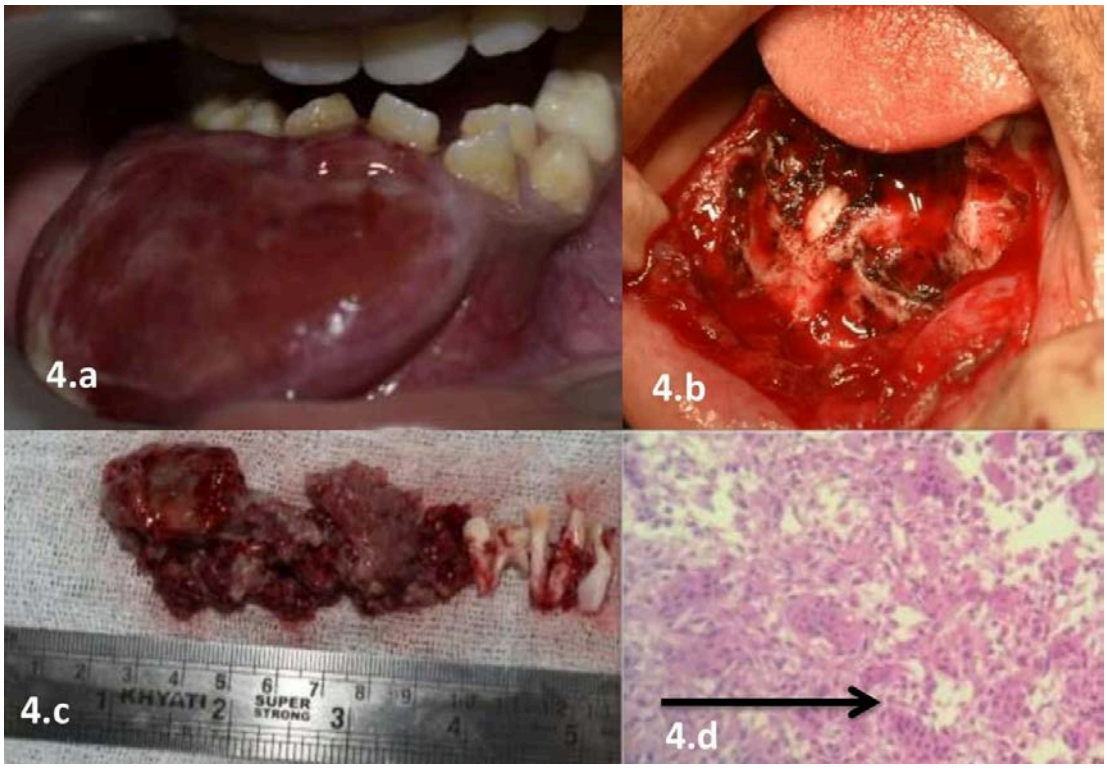

Figure 4. 4.a: Increase in size of the lesion after intralesional steroid therapy; 4.b:Surgical enucleation of lesion; 4.c: Gross appearance of the excised tissue and extracted teeth 31,41,42, 83, 84; 4.d: Histopathological slide showing multinuclear giant cells in fibrous stroma confirming the diagnosis

After taking anesthesiologist's opinion, routine blood investigations were done and the child was scheduled for surgery under general anesthesia. The surgical procedure was performed in association with oral and maxillofacial surgeon. Incisions were made 1cm away from the margins of the bony defect and flap was raised. Since, the patient was a child in the age of growth and development, a radical surgery in the form of bone resection was not considered as the first step. However, complete surgical enucleation[13] of the lesion along with extraction of the mobile teeth 31, 41, 42, 83 and 84 (Figure 4) followed by aggressive curettage was done. This avoids formation of large defects that would compromise esthetics in the young child.[13] Neurovascular bundle was not involved. Tooth buds of 43, 44 were left intact. The bony defect was then irrigated and resorbable sutures were placed. Histopathology of the excised lesion showed multiple giant cells in fibrous stroma (Figure 4). The patient was instructed to use frequent warm saline rinses starting from the day after surgery to keep the wound free of food debris. Child was regularly reviewed until healing was complete. Child was kept under observation and six months post operatively complete healing with new bone formation was seen in the defect area on the panoramic radiograph (Figure 5). At nine months follow up a temporary partial denture (Figure 6) was fabricated with C clasp on 85, Adam's clasp on 75 and it was inserted in the oral cavity.

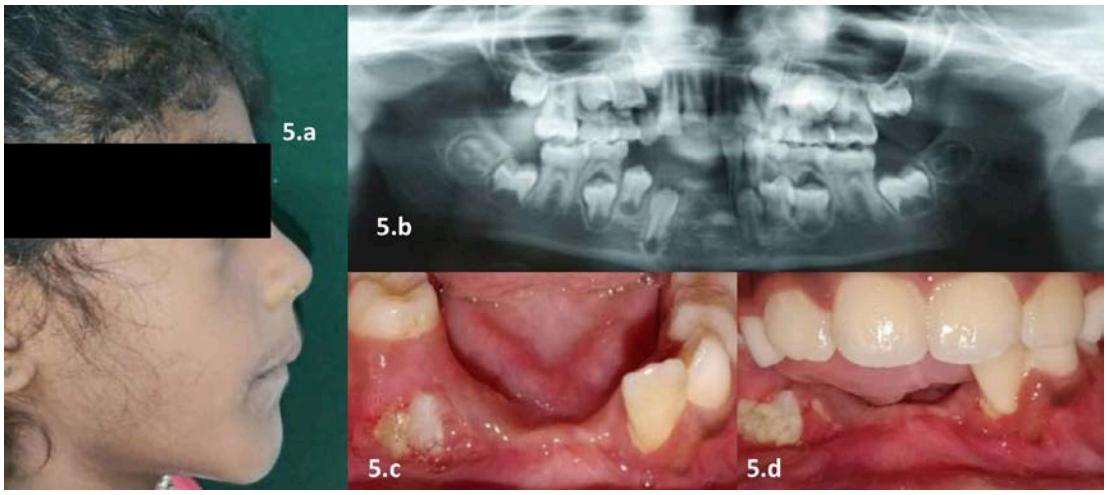

Figure 5. 5.a, 5.c & 5.d: Extra and intra oral image showing 6-months post operative healing, 5.b: 6-month follow up OPG showing satisfactory healing and new bone deposition

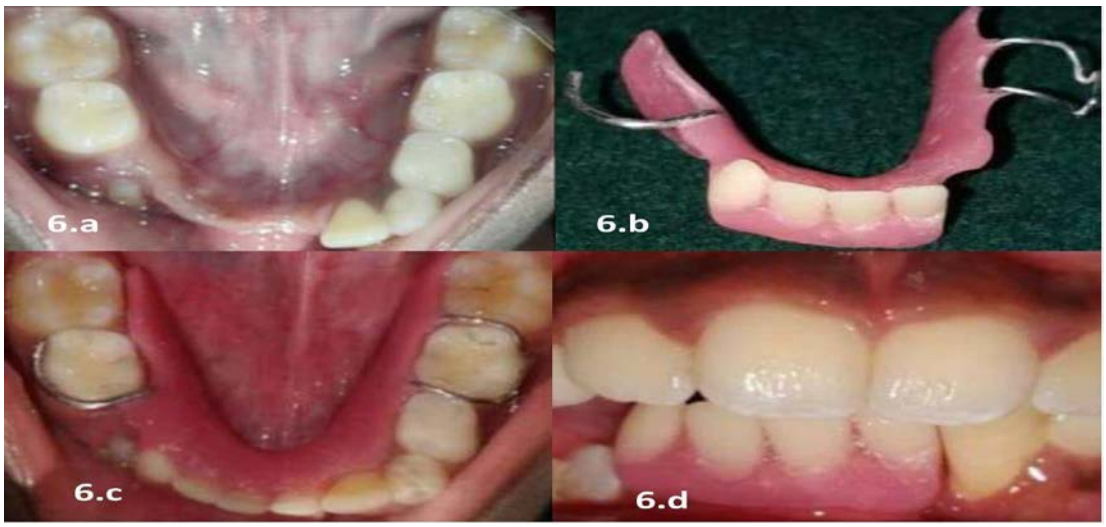

Figure 6. 6.a shows 9-months post-operative healing; 6.b, 6.c & 6.d: Fabrication and insertion of treatment partial denture to improve the esthetics, phonetics and function

Thus, it improved esthetics, phonetics and function in the child patient. One year clinical and radiographic (Figure 7) follow up showed new bone formation and no evidence of recurrence. The child will be followed up for next three years.

DISCUSSION

Central giant cell granuloma is a benign tumor of unknown etiology, it belongs to a group of giant cell tumors.[2] Etiology and pathogenesis of CGCG of jaw bones have not been explicitly defined or recognized clearly. It is believed that it can occur as a response to past injury or as a reactive response to changes in blood supply to the bone.[8] Based on the clinical and radiographic features, it can be classified into two types, non-aggressive and aggressive type. Non-aggressive lesion is slowly expanding and do not show cortical erosion, where as aggressive type is more destructive, grows rapidly with cortical perforation, displacement of teeth and resorption of roots.[5] Correct identification and diagnosis can be quite challenging since clinical and radiographic appearance is not pathognomonic and it is similar to aneurysmal bone cyst, Ameloblastoma, ossifying fibroma and brown's tumor.[4] Final diagnosis can be made by correlating clinical, radiographical findings with histological examination.

In this case, age and gender of the child, location of the lesion and expansile nature of the lesion were indicative of central giant cell granuloma. To rule out Brown's tumor and hyperparathyroidism, biochemical investigations were done which showed normal serum calcium, phosphorous levels, normal alkaline phosphatase levels, normal parathormone levels. Incisional biopsy confirmed the final diagnosis of central giant cell granuloma.

The treatment advocated for CGCG has predominantly been varying extents of surgery, ranging from simple curettage to radical resection.[6] Depending on the size and location of the lesion, surgical intervention will be associated with varying degrees of morbidity. Although radical surgical intervention can be effective, an inevitable loss of teeth and tooth germs will result.[6] Conservative medical regimens have been extensively reported, including systemic calcitonin, α-interferon, denosumab and intralesional corticosteroids.[4] Intralesional corticosteroid use for the treatment of CGCG was first described by Jaco-way et al in 1988. The rationale for its use was that steroids might act by suppressing any angiogenic components of the lesion and inhibit the osteoclast activity. In present case triamcinolone acetonide diluted in an anesthetic solution was infiltrated similar to the protocol recommended by Terry and Jacoway.[10]

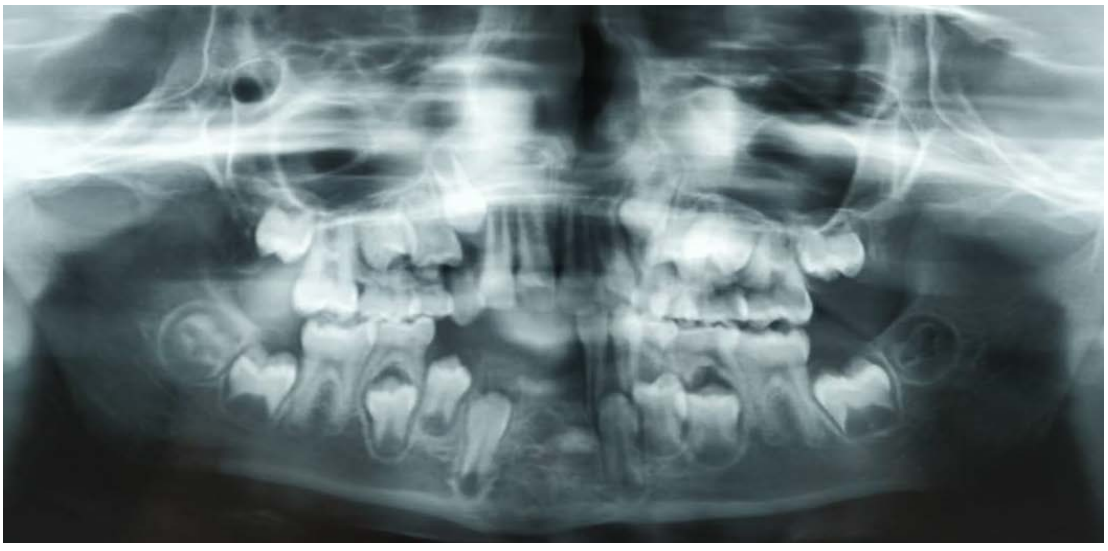

Figure 7. One-year follow up orthopantomogram (OPG) shows new bone formation and no evidence of recurrence.

However at the end of six weeks the lesion did not reduce in size, the reason for this can be attributed to a population of altered osteoclasts that do not have cell membrane receptors to corticosteroids.[4] Since, the patient was a child in the age of growth and development, a radical surgery in the form of bone resection was not considered as the first step. However, complete surgical enucleation of the lesion along with extraction of the mobile teeth 31, 41, 42, 83 and 84 (Figure 4) followed by aggressive curettage was done. This avoids formation of large defects that would compromise esthetics in the young child.[13] The tooth buds of 43, 44 were left intact. Literature search revealed other innovative methods to restore the lost tissues and hasten bone fill in the defect. Use of PRF, native extracellular matrix (ECM) and autologous bone grafts in large defects have been reported. In our case since the lingual cortical plate was intact, adequate healing and bone fill after six months was seen on panoramic radiograph. Hence, at nine months follow up the child patient was rehabilitated with a temporary partial denture to replace the missing teeth.[8] One year clinical and radiographic follow up showed new bone formation and no evidence of recurrence. The child will be followed up periodically.

CONCLUSIONS

Although benign jaw lesions constitute only a small number of pathologic conditions seen by a pediatric dentist, they are of great significance since they have the potential ability to jeopardize the health and longevity of the patient. Hence, early diagnosis and management are very essential.

REFERENCES

1. Jeyaraj P. Management of central giant cell granulomas of the jaws: An unusual case report with critical appraisal of existing literature. Ann Maxillofac Surg 2019;9:37-47

2. Bahbah S, Dghoughi S, Chhoul H, El Wady W. Central giant cell granuloma in a child: case report. Int J odonto stomatol 2016:393-397

3. Dean, JA, David, RA, Ralph, EM. McDonald and Avery's Dentistry for the child and adolescent. 10th ed. St. Louis: Elsevier 2016

4. Ferretti C, Muthray E. Management of central giant cell granuloma of mandible using intralesional corticosteroids: case report and review of literature. J Oral Maxillofac Surg 2011;69:2824-2829

5. Neville BW, Damn DD, Allen CM, Bouqot JE. Text book Of Oral and Maxillofacial Pathology. 2nd ed. Philadelphia: WB Saunders; 2002

6. Schatz JP, Bernard JP, Byloff FK, Samson J. Conservative surgical and orthodontic management of a child with central giant cell granuloma. Int J Paediatr Dent 1995;5:35-41

7. Balaji P, Balaji SM. Central giant cell granuloma – A case report. Indian J Dent Res 2019;30:130-132

8. Masih U, Kour G, Mahay P, Joshi S. Central giant cell granuloma: A rare clinical entity. Int J Pedod Rehabil 2017; 2:41-45

9. Chavis SE, Shrikian TM, Haerian A, Edwards SP, Munz SM. Coordinated pediatric reconstruction and rehabilitation of maxillary central giant cell granuloma: A case report. J Am Dent Assoc 2018;149:1065-1072.

10. Jacoway JR. Central giant cell granuloma –an alternative to surgical therapy. Oral Surg Oral Med Oral Pathol 1988;66:572-575

11. Mohanty S, Jhamb A. Central giant cell lesion of mandible managed by intralesional triamcinolone injections. A report of two cases and literature review. Med Oral Patol Oral Cir Bucal 2009;14:98-102

12. Smriti K, John E, Pentapati KC, Gadicherla S, Bhagania M. Intra-Lesional Medicaments for the Management of Intra-Osseous Lesions of Maxilla and Mandible - Systematic Review. J Int Soc Prev Community Dent 2020:24;10:36-45

13. Altan H, Altan A, Eragca R, Yildirim E, Akbulut N. Surgical treatment of central giant cell granuloma in child and follow-up for 18 months. Pediatr Dent J 2018:28;125-130

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Declarations

Acknowledgements: *Not applicable.*

Conflict of Interest Statement: A*uthors disclose no potential conflicts of interest.*

Ethics Statement: *Procedure was explained to the parents and written consent was obtained for the treatment and publication of the case report.*

Informed Consent: *Informed consent was taken from parents.*

Author contributions: *Conception and design: All Authors; Acquisition of data: All Authors; Interpretation of data: All Authors; Drafting article: MCH, NRK; Revision artice: All Authors; Final approval: All Authors*

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