

CASE REPORT

Aggressive Central Giant Cell Granuloma of Mandible Transformed to an Enormous Vascular Lesion

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ABSTRACT

Aggressive central giant cell granuloma (ACGCG) is usually found in younger patients and is painful, grows rapidly, larger overall, often causes cortical perforation, root resorption and has a tendency to recur. This article is a case report of ACGCG in mandible. A girl 12 years old had features of typical giant cell reparative granuloma in the anterior mandible. After incision biopsy, she reported after 11 months. The lesion had rapidly increased in size and vascularity to an enormous tumor-like mass with engorged vessels. The lesion was of enormous size and extensive expansion was high in vascularity. The cortices remained but thinned out and perforated despite of extensive expansion. The lesion was curetted resulting in loss of continuity of the mandible. The microvascular surgery with free fibula graft was done to reconstruct the mandible. This is report of rarest case of ACGCG with enormous increase in size and vascularity which was treated successfully and rehabilitated.

Keywords: Aggressive central giant cell granuloma, Vascular lesion, Reconstruction of mandible.

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INTRODUCTION

Central giant cell granuloma (CGCG) is an uncommon benign intraosseous lesion that occurs almost exclusively in jaws, introduced for the first time by Jaffé in 1953.¹ It was hypothesized that the lesion is not a true neoplasm but merely the result of a local reparative reaction. The World Health Organization has defined CGCG as an intraosseous lesion consisting of cellular fibrous tissue containing multiple foci of hemorrhage, aggregations of multinucleated giant cells and, occasionally, trabeculae of woven bone.² The clinical behavior of CGCG varies from a slowly asymptomatic swelling to an aggressive lesion that manifests with pain, cortical perforation and root resorption.³ Giant cell lesion is the most common lesion associated with secondary aneurysmal bone cyst (a vascular lesion) in 39% of cases. Similarly, in 14% of giant cell lesions aneurysmal bone cyst component are seen.⁴ Choung et al⁵ have classified CGCGs into aggressive and nonaggressive lesions based on biological behavior. The case reported here had features of typical giant cell reparative granuloma in the anterior mandible in 11 months, it increased rapidly in size and

vascularity to an enormous tumor-like mass with engorged vessels.

CASE REPORT

A 12-year-old girl reported with painless swelling in the anterior mandible, typically around chin (Fig. 1) since 1 month. The skin over the lesion was stretched but normal. There was expansion of cortices but buccal expansion was more pronounced. The computed tomographic (CT) scan shows multilocular lesion extending from mandibular first molar on one side to that of other side (Fig. 2). There was extensive expansion of buccal cortex. Thus, the lesion was bilateral, multilocular with expansion of buccal cortex and extension from first molar of one side to first molar of other side of mandible. With the clinical diagnosis of CGCG, the biopsy was taken. The biopsy was suggestive of CGCG (Fig. 3). Patient reported after 11 months. The lesion had



Fig. 1: Extraoral diffuse swelling around mandibular symphysis



Fig. 2: Reconstructive CT image of initial lesion

increased very rapidly to an enormous size (Fig. 4). There was enormous, ballooning swelling in the lower part of face bilaterally. The skin over the lesion was stretched and engorged blood vessels could be seen all over. There was huge expansion of both cortices. Intraorally, the mucosa overlying lesion was stretched and reddish; the lingual expansion had raised the tongue. All teeth were mobile and displaced. The CT scan at this stage showed a definite increase in size of the lesion. The bilateral lesion now extended to the half of ramus height on the right side and to the angle of mandible on left side (Fig. 5). The lesion on the CT scan was multilocular, with enormous expansion of buccal as well as lingual cortices, buccal perforation and no bony support to the teeth (Fig. 6). The extension of the swelling was beyond the mandible and was covering the entire neck.

The aggressiveness and the vascularity of the lesion were clinically and radiologically evident. The previous biopsy report was reviewed. It was concluded that the basic lesion

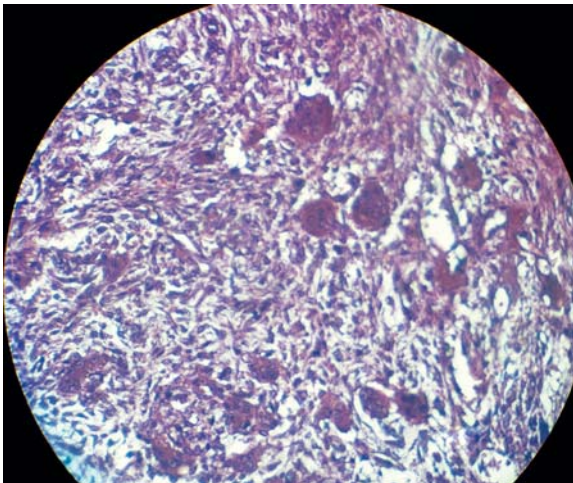


Fig. 3: Histopathology of biopsy from initial lesion suggestive of CGCG



Fig. 4: Delayed clinical presentation

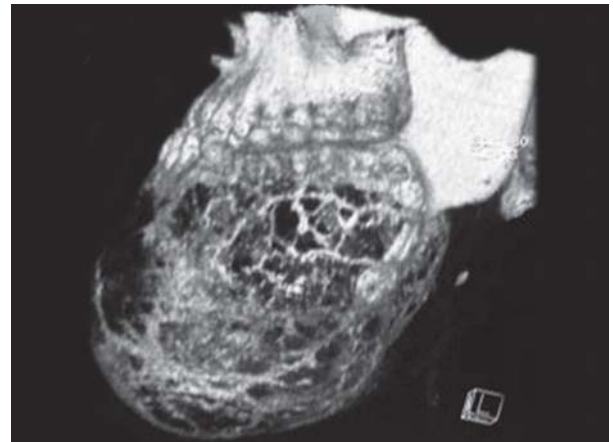


Fig. 5: Reconstructive CT image of delayed lesion sagittal section, left side



Fig. 6: Axial section of CT showing huge expansion of the lesion

is ACGCG with increased vascularity and extensions. The surgery was immediately planned.

The angiography identified bilateral facial and lingual arteries as feeders of the lesion. The embolization of the facial and lingual artery was done beforehand to reduce bleeding. On skin reflection, the blood was oozing from inside the lesion as if the lesion was full of blood—a sponge soaked in blood. The lesion had enormous expansion but the thin, perforated buccal as well as lingual cortices were present which gave a surgical plane of dissection below the periosteum. The lesion could be separated from the bone at angle of mandible on left side and ramus of mandible on right side. No osteotomy cut was taken, but the thinness and perforation of cortices lead to the discontinuation of mandible.

Thus, after curettage, there was loss of continuity of mandible from angle of mandible left side to the ramus on the other side with irregular ends remaining. The peripheral ostectomy was done to smooth the end edges. The excised lesion is of size around 15 cm horizontally and 11 cm buccolingually (Fig. 7). The free fibula grafting was done by the plastic surgeon to maintain the continuity of

mandible. The prosthodontist prepared a spring loaded prosthesis based on maxilla. Figure 8 shows the 1 year postoperative panoramic radiograph, it shows the well accepted fibula graft forming the continuity of the mandible. Also some wire parts of the prosthesis can be seen. Figure 9 shows 1 year postoperative extraoral image. Histopathologically, there is evidence of vascular spaces with fibrous stroma (Fig. 10).

DISCUSSION

CGCG usually occurs in patients younger than 30 years, is more common in females than in males, and is more common in the mandible than in the maxilla. The lesion has frequently been reported to be confined to the tooth-bearing areas of the jaws and is more common in the anterior portion of the mandible, often crossing the midline.⁶ Although CGCGs are benign osseous lesions, some authors separate CGCG into two types, referring to its clinical and radiographic features: (A) Nonaggressive lesion is usually slow growing and asymptomatic, does not show cortical perforation or

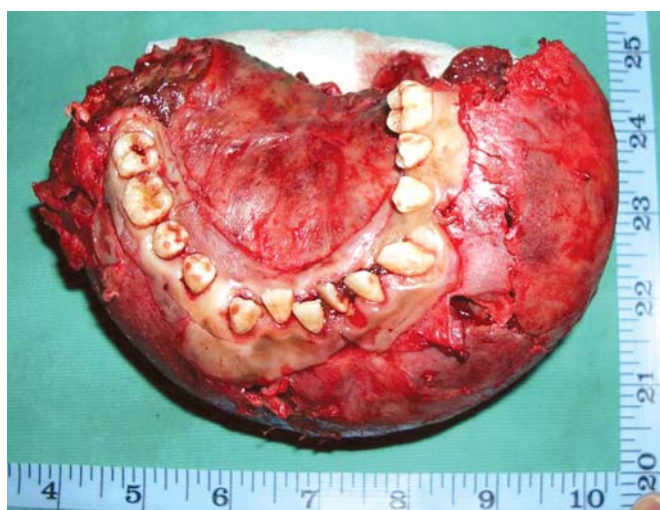


Fig. 7: Excised specimen is of size around 15 cm horizontally and 11 cm buccolingually

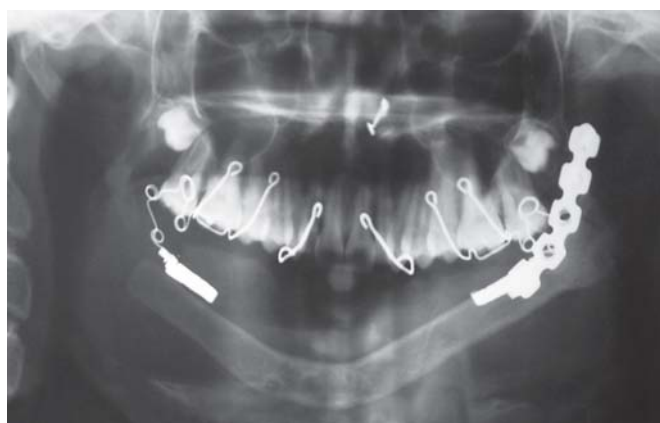


Fig. 8: The 1 year postoperative panoramic radiograph shows the well accepted fibula graft forming the continuity of the mandible



Fig. 9: Extraoral postoperative image photograph

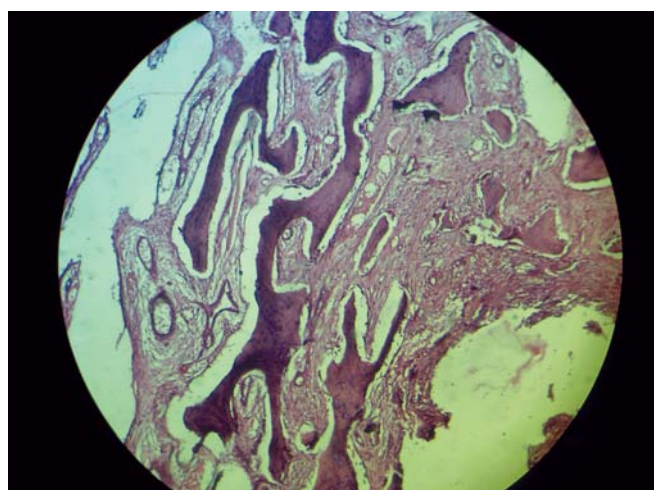


Fig. 10: Histopathological image of excised specimen

root resorption in teeth affected, and it is significantly less likely to recur than the aggressive type;⁷ and (B) aggressive lesions is usually found in younger patients and is painful, grows rapidly, larger overall, often causes cortical perforation and root resorption and has a tendency to recur.⁵

Histological and cytometric differences were analyzed, and immunoprofiles and giant cell DNA quantification were performed in an attempt to identify further features as predictors of aggressiveness for individual lesions. Nevertheless, none of these studies could establish parameters for the prediction of the clinical course of the disease. In comparison, the clinical criteria used by Chuong et al have been more reliable in distinguishing aggressive CGCGs from nonaggressive lesions.⁵ CGCG should also be distinguished from giant cell tumor of long bones. The latter is locally aggressive with a high recurrence rate, and a potential for malignant transformation.⁸ Some CGCGs of the jaws, despite an innocent histologic appearance, show an aggressive behavior and a tendency to recur. Ficarra et al considered that these lesions should be defined as 'aggressive giant cell granulomas' of the jaws, rather than

giant cell tumor.⁹ In cases with aggressive behavior with the lesions, their maximum diameter at the time of the diagnosis was 56 mm, compared to 33 mm in clinically nonaggressive CGCGs.¹⁰ The lesion reported here had size of 35 mm at the time of initial reporting which increased to 15 cm horizontally and 11 cm buccolingually. The surgical techniques used for ACGCG are curettage, curettage with peripheral ostectomy and bone resection. Bone resection is reserved for recurrences. Eisenbud et al⁷ used the technique of curettage or curettage plus peripheral ostectomy. Becelli et al¹¹ described a case treated by means of excision of a mandibular CGCG, reconstruction using autogenous iliac crest graft, dental implants and overdenture prosthesis. The nonsurgical management mentioned in the literature are administration of intralesional corticoids¹² or systemic calcitonins that inhibit the osteoclastic activity¹³ and interferon-alpha or bisphosphonates.¹⁴ Conservative treatments with varying degrees of success have reduced the necessity for reconstructive surgery. In the reported case, due to rapid growth, aggressiveness of lesion and previous history of noncooperation from patient, the surgical treatment was performed immediately, and other treatment modalities were not used. The curettage and peripheral ostectomy eliminated the lesion, and the continuity of mandible was reconstructed with free fibula graft. Aggressive CGCG has a tendency to recur, if inadequately removed, and high recurrence rates have been reported. It has been shown that recurrence usually happens, when the lesion perforates the cortical plates to involve the surrounding soft tissue. Malignant transformation of the CGCG is a rare phenomenon. The reported patient was followed for 2 years. There was no recurrence.

CONCLUSION

The aggressive, extensive and highly vascular CGCG was successfully treated by embolization of bilateral facial and lingual arteries, curettage and peripheral ostectomy. The reconstruction with free fibula graft and spring-loaded prosthesis rehabilitated the patient. This is report of rarest case of ACGCG with enormous increase in size and vascularity which was treated successfully and rehabilitated. To my knowledge, this is the first case with such a huge

size 15 × 11 cm. For the lack of histopathological evidence, the case is not labeled as aneurysmal bone cyst.

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