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An aggressive central giant cell granuloma treated successfully by conventional surgery

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Abstract

Central giant cell granuloma is a rather rare lesion of the jaws aetiology of which is still completely unknown but thought to be of a reactive process to some unknown stimuli. It is an uncommon benign proliferative lesion that may become aggressive leading to the expansion and perforation of cortex, resulting into mobility and displacement of teeth and root resorption, with very high recurrence rate, requiring then a radical surgery. We report the clinical course of aggressive central giant cell granuloma treated successfully by conventional surgery, with fortunately no sign of recurrence during the control period. Authors suggest that the aggressive form of the CGCG should be treated initially by conventional therapy. Regular course of control will allow spotting recurrence signs if any. Radical surgery should be set aside for recurrence cases.

Keywords: Aggressive and non-aggressive form, central giant cell granuloma, conventional surgery, differential diagnosis, maxilla, radical surgery

1. Introduction

The (CGCG) is recognized to be a benign localized proliferation that is osteolytic, consisting of fibrous tissue containing multinucleated giant cells, hemorrhagic areas, and deposits of hemosiderin, and occasionally involving a bone reaction ^[1]. It is described as a rare benign tumor of the jaws with an unknown cause accounting for up to 7% of jaw tumors, ^[2] and by far the most common of giant cell lesions of jaws ^[3]. The peak incidence of CGCG is during the second decade of life, occurs more frequently in males, and the mandible is the most common site ^[3]. Since Chuong's study in 1986, the GCCG are divided into non-aggressive and aggressive forms, with the former being more common ^[4]. The clinical signs and symptoms and the radiographic and histological features are the main factors differentiating the two forms ^[5]. This classification is of a therapeutic purpose since it suggests en bloc resection (radical surgery) for aggressive CGCGs even if local curettage is the conventional treatment. This is due to the recurrence rate following local curettage in aggressive CGCGs which is between 16% and 48%. En bloc resection is associated with the lowest recurrence rate. However, it results in various degrees of deformity and requires complex reconstruction procedures ^[5]. In our report, we share with you a rare case of maxillary aggressive central CGCG treated successfully by conventional surgery, with fortunately no sign of recurrence during the control period.

2. Case report

Mrs. G.S, 32 years old, consulted her dentist for an infectious swelling on the left anterior maxilla involving buccal tissue. Tooth 24, carious, was suspected as causal. It was extracted and antibiotherapy was prescribed. The infection has been then resolved but the swelling still persisted.

One year later, the patient consulted in Center for Consultation and Dental Treatment of Rabat. Physical examination revealed a left maxillary swelling. This manifestation was initially noted 6 months before the first consultation with the treating dentist. There was no facial palsy. No cervical lymph node enlargement was seen.

During the oral examination (fig 1), a lump was found involving the buccal side of alveolar bone crest corresponding to the left premolar region of maxilla, with two nipples on its surface one larger than the other.

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It was firm, slightly depressible, and painful to palpation with a blowhole on the alveolar table. The covering mucosa was bluish. The tooth 23 was non vital and has a degree 2 mobility according to LINDHE classification.



Fig 1: Intra oral examination showing a lump involving the buccal side of alveolar bone crests corresponding to the left premolar region of maxilla, with two nipples (arrows) on its surface, one larger than the other

Pantomographic image (fig 2) showed a wide unilocular osteolytic area on the left premolar region of maxilla, with ill-defined borders, extending from the second permanent molar to the canine. Teeth 24, 25, 26 and 27 were absent. A root resorption was observed on tooth 23. The radiolucency was not related to maxillary sinus and the nasal cavity.



Fig 2: Pantomographic image showing a wide unilocular osteolytic area on the left premolar region of maxilla, with ill-defined borders, extending from the second permanent molar to the canine. A root resorption is observed on tooth 23.

A surgical biopsy under local anesthesia was performed showing a voluminous bone cavity filled with a highly hemorrhagic, red-brown, soft tumor. The provisional diagnosis of giant cell lesion was made.

A phosphocalcic assessment was performed for differential diagnosis of a brown tumor of the jaws secondary to a hyperparathyroidism engendering a similar bone lesion and for witch treatment with calcitonin can be sufficient. The obtained results were normal.

The anato-pathological result of the biopsy suggested a central giant cell granuloma of the maxilla.

Considering results of the orthopantomogram, the biological assessment and the biopsy, a surgical intervention was decided and intraorally performed under local anesthesia.

Once the mucoperiosteal flap elevated, the tumor was immediately exposed. (fig 3 A) The characteristic appearance was found as a "fleshy reddish brown mass, looking like a splenic pulp" (Senthiles and Michaud, 1986) [6] (fig 3 B). During the excision, the tumor was highly hemorrhagic and crisp. A complete curettage of all colonized woven bone trabeculae of "wet sugar" consistency was performed until bone tissue of healthy appearance was obtained (fig 3 C). Due to its seating in the tumor, tooth 23 was extracted.

The excised specimen (multiple fragments measuring between 2.5 and 5 cm and the tooth 23) (fig 3 D) were subjected to an

anato-pathological examination. A coverage antibiotherapy (Amoxycilline, 2g/day for 5 days), a level 2 analgesic (Codeine (20 mg) + paracetamol (400mg) 2cp, 3 times per day) and a mouthwash with 0.12 % chlorhexidine were prescribed. Toothbrushing with a 7/100 post-surgical toothbrush was recommended from the next day. The intra- and postoperative course was uneventful.



Fig 3: (A-B-C) Intraoperative phases. (D) Macroscopic aspect of the lesion excised. It exhibits typical features of giant cell granuloma, "fleshy reddish brown mass, looking like a splenic pulp" (Senthiles and Michaud, 1986). On the right, the canine tooth shows root resorption (arrow).

At 1 month postoperatively, mucosal healing was obtained and no residual pain was indicated. The anato-pathological examination, according to histology and inclusion cytology, showed a gingival mucosa harboring multiple giant multinucleated cells associated with many regular and ovoid mononuclear cells. This cellular infiltrate bathes in much hemorrhagic suffusion. Bone span was also present. Neither cellular atypia nor mitosis were noted. The diagnosis of central giant cell granuloma of the maxilla was confirmed. A radiological follow-up (fig 4) at 3 and 6 month postoperatively showed progressive bony healing and no sign of recurrence was noticed.

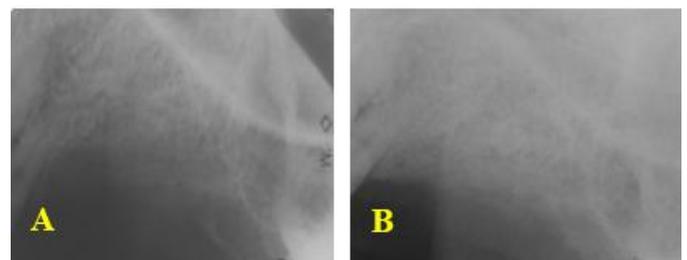


Fig 4: A radiological follow-up at 3 (A) and 6 month (B) postoperatively showing progressive bony healing.

3. Discussion

Central giant cell granuloma (CGCG) was first introduced by Jaffe in 1953 as a "giant cell reparative granuloma" to pass on that it was not a neoplasm [7]. However, despite long standing observations, the real etiology of CGCG is hitherto unknown [8, 9]. It has been suggested that it is the result of an exacerbated reparative process related to previous trauma and intraosseous hemorrhage that triggers the reactive granulomatous process [10]. In addition to trauma, some indications implicate inflammatory foci and a genetic

predisposition [11, 12, 13]. The reparative term has been rejected in recent time because the lesions are typically destructive, never reparative [1] and trauma is not always a mandatory precursor [11].

Since the Chuong study in 1986, [14] several groups of clinicians have proposed that GCCGs may be divided into non-aggressive and aggressive forms, [8, 4] referring to its clinical and radiographic features: [10, 5]

(a) Nonaggressive form, the most common [12], appears as a bone swelling covered with brownish mucosa, functionally silent [8]. The lesion is usually slow-growing and asymptomatic. On the radiologic level, it presents as a well-circumscribed radiolucent lesion [12] with no cortical perforation or root resorption in teeth affected, (which are displaced and sometimes still alive [8]).

(b) Aggressive form is usually found in younger patients. It is more explosive, larger (greater than five centimeters) and grows rapidly [10]. It is usually associated with pain, mucosal ulceration, and sensory disturbances. It usually consists of a large lytic lesion, more often multilocular than unilocular, with cortical expansion, displacement of teeth, and root resorption [8, 12]. Most of the aggressive form features fit with our case which led us to consider it as such.

The radiological appearance of CGCG is not pathognomonic. It varies greatly and may be confused with that of many other lesions of the jaws [10]. Macroscopically the tumor tissue is reddish-brown, lumpy and haemorrhagic. The diagnosis of CGCG is compulsory confirmed by biopsy and according to the WHO definition in 2005, on the histological examination, these lesions include a fibrovascular stroma of reactional form in which we can find, in variable proportion and organization (but without true specificity):

- Areas of dense fibrous tissue;
- Osteoclast-like giant cells irregularly dispersed and often aggregated by spot;
- Hemosiderin deposits;
- Occasionally, spans of osteoid or bone tissue [15].

This histological aspect is common to all giant cell lesions, namely the brown tumors of hyperparathyroidism, cherubism (or familial multilocular cystic disease of the maxilla), the aneurysmal cyst and the giant cell tumor. These lesions must be considered in the differential diagnosis [8]. The brown tumor of hyperparathyroidism is a bone lesion related to primary hyperparathyroidism or secondary to chronic renal insufficiency complicated with osteodystrophy [9]. To help differentiate this lesion, serum calcium and alkaline phosphate levels are increased and serum phosphate levels are low [1]. The cherubism is an autosomal dominant disorder and present different clinical and radiologic features [16]. The lesions are generally bilateral, more mandibular, and affect more boys than girls. Radiologically bone cortices are blown under the effect of multiple multilocular osteolytic plaques [9]. Aneurysmal bone cyst also shows similar histological features and contain multinucleate giant cells with a similar morphology and distribution, but is the only one who present 'cavernous or sinusoidal blood filled spaces' [17]. Above all, CGCG is most difficult to differentiate from GCT without clinical and histological aids [10]. CGCG generally occurs at younger age than GCT [10]. Moreover, in GCT, giant cells are evenly spaced and contain a larger number of nuclei with no fibroblasts, hemorrhage, and deposition of osteoid, [10, 18] all of which are present in our specimen.

The recurrence rate is variable and depends on the biological behavior and the management of the CGCG. Non-aggressive

form seems to have low recurrence rates and is described to be successfully treated by enucleation and curettage alone [15]. However, in case of aggressive form, recurrence rate after conventional therapy ranges from 20% to 70%, and the en bloc resection is recommended for such case [5]. The dilemma is that the en bloc resection results in various degrees of deformity and requires complex reconstruction procedures [5]. This has raised concern and led to a search for other treatment options namely the application of some pharmacological agents. The aim is to prevent or at least minimize the extensive and mutilating surgical procedures characterized by detrimental functional outcomes and preserve vital structures and facial contours [5]. Pharmacological agents that have been used include the application of subcutaneous or nasal calcitonin, intralesional steroids, and subcutaneous injections of alpha interferon [2]. The use of calcitonin was proposed in 1993 by Harris, based on the similarity that exists between CGCG and the tumours of the hyperparathyroidism at histological level [2]. Calcitonin acts to lower calcium levels by inhibiting bone resorption, counteracting parathyroid hormone [4]. Concerning intralesional steroids injections, it was first describe by Jacoway et al. 1988 [2]. Their use is based on the fact that the histopathology of CGCG resembles sarcoidosis, suggesting that because corticosteroid injection is an effective treatment of sarcoidosis, that it would be an effective treatment of CGCG [4]. Interferon is an antiviral and antiangiogenic agent. This treatment is based on the theory that these lesions may be proliferative vascular lesions. The interferon is thought to suppress the angiogenesis resulting in involution of the lesion. It may also induce the differentiation of mesenchymal stems cells into osteoblasts, leading ultimately to bone formation [4]. Concerning the effectiveness of those adjuvant therapies, only one small randomised controlled trial (RCT) of calcitonin for treating central giant cell granuloma (CGCG) of the jaws was conducted and, it was not possible to determine whether calcitonin is beneficially effective or harmful for patients with CGCG of the jaws. The effects of other active compounds are reported in several case reports and series but no RCTs. However the findings are controversial. The management of aggressive variant of CGCG can, thus, include en bloc resection or conventional surgery (aggressive curettage) with or without medical adjunctive treatment, [19, 2]. In the informed consent of the patient, we explained all the therapeutic possibilities and their possible consequences. The patient wanted to try her luck with the conventional surgery because of the aesthetic deficit engendered by the radical surgery. Fortunately we didn't observe any sign of recurrence during the control period (6 months).

4. Conclusion

Central giant cell granuloma (CGCG) is an infrequent lesion in daily odontological practice. The currently available bibliographic data concerning the various possible treatments of the CGCG show that curettage surgery is the conventional treatment generating the least recurrence in case of non-aggressive form. For the aggressive form, there is still no consensus concerning the best therapeutique choices, ranging from conventional surgery with or without adjuvant therapy to radical surgery. Thus, and in the light of our case report, authors suggest that it would be reasonable to start by conventional surgery (enucleation and curettage) and set aside radical surgery for recurrences cases. Finally, the importance of early detection and knowledge of the diagnostic elements of this type of often benign tumors, but whose very

destructive consequences are disabling, should be emphasized.

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