

# Giant cell granuloma of the maxilla: case report

## *Granuloma a cellule giganti del mascellare superiore: descrizione di un caso clinico*

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### Key words

Maxilla • Benign lesions • Giant cell granuloma • Surgical treatment • Case report

### Parole chiave

Mascellare superiore • Lesioni benigne • Granuloma a cellule giganti • Terapia chirurgica • Caso clinico

### Summary

Giant cell granuloma is an uncommon bony lesion in the head and neck region, most commonly affecting the maxilla and mandible. Although it is a benign disease process, it can also be locally destructive. Surgery is the traditional and still the most accepted treatment for giant cell granuloma. The case described here involved the maxilla which was treated with surgical excision, followed by local injection of steroids.

### Riassunto

*Il granuloma a cellule giganti è una patologia di raro riscontro nella regione cervico-facciale, per lo più localizzato in corrispondenza della mandibola e del mascellare superiore. Sebbene si tratti di una neoplasia benigna, può risultare molto aggressiva localmente. Il trattamento chirurgico è quello tradizionale ed è ancora quello più in uso, sebbene altri tipi di approccio (es. terapia con calcitonina) siano stati indicati in letteratura. Gli Autori presentano un caso di granuloma a cellule giganti localizzato nel mascellare superiore, trattato con chirurgia e successive iniezioni locali di steroidi.*

## Introduction

Giant cell granuloma (GCG) was first described by Jaffè in 1953<sup>1</sup>. This lesion is characterised by proliferation of fibroblasts and multinucleated giant cells, in a densely packed stroma<sup>1</sup>. Central GCG (CGCG) (e.g., GCG of the mandible and maxilla bones) is less common than peripheral GCG of the extremities<sup>2</sup>.

Jaffè<sup>1</sup> recommended curettage for these lesions, since they have the potential for extensive bone destruction. Others have advised complete en-bloc excision<sup>2</sup>.

Although defined, histologically, as benign lesions, GCGs have the capacity of local destruction; thus, a treatment modality that would arrest growth is mandatory<sup>2</sup>. Surgery is the traditional and still most accepted treatment for GCGs<sup>3</sup>.

A case of GCG of the maxilla is described which was managed by surgery, followed by local injection of steroids.

## Case Report

A 34-year-old female was referred to the Otolaryngology Service at the University of Perugia. She was first admitted in December 1998.

Symptoms noted a few weeks before admission included painful swelling of the upper cheek and right gum and acute right maxillary sinus infection. She had initially been treated with antibiotics, which gradually led to an improvement in the sinus infection. However, the swelling remained stable, and was initially thought to be due to a dental infection. Since the symptoms persisted, a computed tomography (CT) scan was taken. It revealed a lytic lesion, apparently of dental origin, which caused bone destruction in the inferior portion of the maxilla. Surgery with curettage of the lesion was then performed. Histological examination revealed giant cells in a fibroblastic stroma, with large areas of haemorrhage, which the pathologist then interpreted as a giant cell granuloma.

After surgery, the patient was monitored at regular (bi-monthly) follow-up. No change in the lesion was observed upon physical examination.

The swelling recurred 20 months after surgery. The patient was again referred to our service for evaluation and treatment. The intra-oral examination revealed a firm submucosal mass, 2 cm in diameter, adhering to the anterior wall of the maxilla.

The feasible therapeutic options were discussed with the patient. Partial maxillectomy was suggested, with the use of a prosthesis to correct the resulting defect.

The patient decided to undergo repeat curettage of the lesion, in order to avoid possible problems related to further major surgery.

Histological findings showed giant cells in a fibroblastic stroma, with large areas of haemorrhage. There was no evidence of mitosis or of proliferations of other mononucleated cells. Considering the localization and the relatively small size of the giant cells (with few nuclei), the specimens were interpreted, by the pathologist, as a "Reparative Giant Cell Granuloma".

The patient underwent clinical examination every 2 months. No signs of recurrence were detected over the next 17 months of follow-up, when the swelling recurred (Fig. 1).



Fig. 1. The mass involves the anterior wall of the maxilla, causing asymmetry of the right hard palate. The patient presented loosening of both the incisive teeth on the right maxillary side.

Intra-oral examination revealed loosening of both the incisive teeth on the right maxillary side. The mass was adhered to the anterior wall of the maxilla, resulting in asymmetry of the right hard palate. Moreover, the floor of the right nasal cavity was deformed. A new CT scan was performed which revealed a lytic mass involving the anterior part of the maxilla and the alveolar ridge (Fig. 2).

All the therapeutic options, including partial maxillectomy, were again discussed with the patient who was still determined to avoid major surgery. We then decided to carry out surgical treatment of the lesion followed by local injection of corticosteroids, in an attempt to avoid further recurrence.

Using an upper right gingival labial sulcus approach, the entire tumoural mass was removed, under general anaesthesia (Fig. 3). Careful curettage of the residual bone cavity was performed. Histological examination of the surgical specimens again revealed giant cells in a fibroblastic stroma, with large areas of haemorrhage



Fig. 2. CT scan. The mass, a lytic lesion, involves the anterior part of the maxilla and the alveolar ridge.

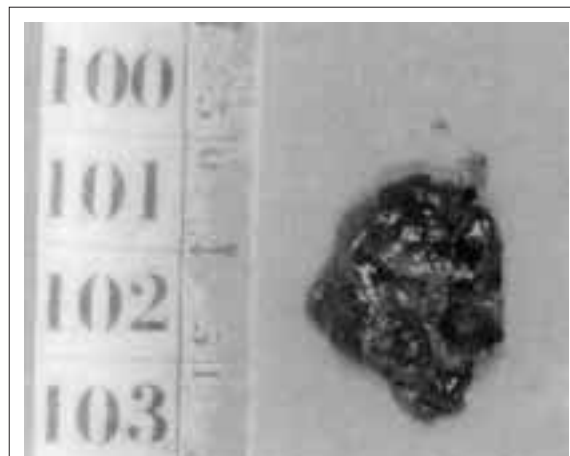


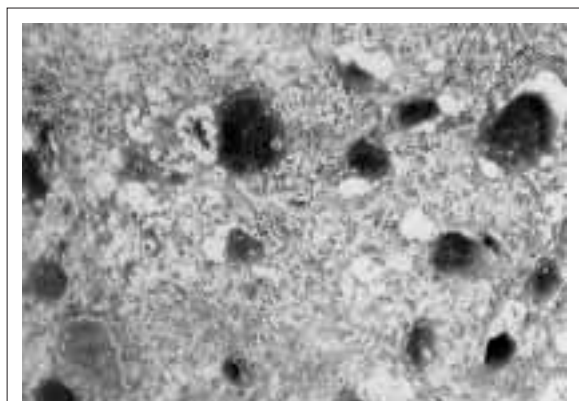
Fig. 3. The tumoural mass removed.

which were interpreted, by the pathologist, as "Recurrent Reparative Giant Cell Granuloma" (Fig. 4). Surgical treatment was followed by consecutive weekly local injection of corticosteroids, for a period of 6 weeks. Except for mild discomfort during the first injection, the injections were well tolerated by the patient.

At present, 18 months after this last treatment, the patient is still monitored at follow-up, and shows no signs of recurrence.

## Discussion

Central GCG is a rare disease<sup>2</sup>. GCG, or RGCG is an uncommon benign bony lesion located in the mandible and maxilla<sup>1</sup>.



**Fig. 4.** Histologic examination shows large areas of haemorrhage, multinucleated giant cells, in a cellular vascular stroma and new bone formation.

It has been defined by the World Health Organization as an intra-osseous lesion consisting of cellular fibrous tissue that contains multiple foci of haemorrhage, aggregations of multinucleated giant cells, and, occasionally, trabeculae of woven bone<sup>4</sup>.

GCG accounts for <7% of all benign lesions of the mandible and maxilla in tooth-bearing areas. The mandible, anterior to the first molar teeth, is the most commonly affected site<sup>3</sup>. Central GCG (e.g., GCG of the mandible and maxilla) is less common than peripheral GCG of the extremities<sup>2</sup>.

GCG affects children and adults<sup>5</sup>, and may occur at any age, but is most commonly seen in the first 3 decades<sup>6</sup>. It is more frequent in females than in males<sup>5</sup>. The aetiology of the GCG still remains to be defined. It has been reported that the origin of this lesion could be triggered by trauma or inflammation<sup>5</sup>.

The common effects of GCG are: painless swelling that remains undetected until facial asymmetry, impaired nasal breathing, and loosening or displacement of teeth. Localized swelling is an important clinical feature. The swelling is smooth, and palpation can reveal a rubbery, elastic sensation where the bone has thinned. Although the lesion is expansile and invasive, it does not usually involve the perineural sheets. For this reason, paresthesia is not usually observed in these patients<sup>5</sup>.

The clinical behaviour of GCG ranges from a slowly growing asymptomatic swelling to an aggressive lesion<sup>3</sup>. When GCG is a slow-growing lesion, it can be asymptomatic and discovered on a routine X-ray, while the rapidly expanding, aggressive variety is characterized by pain and facial swelling. These fast-growing lesions also have a high rate of recurrence<sup>7,8</sup>. It has been reported that recurrence is usually found when the lesion perforates the cortical plates to involve the surrounding soft tissue<sup>9</sup>.

There have been reports of recurrence as early as 3 years and as late as 22 years<sup>10</sup>.

Radiologic features vary from undefined destructive lesions to a well-defined, multilocular appearance. Teeth or roots displacement are the most consistent features which are clearer on CT than on plain film<sup>11</sup>. However, none of these radiological features is specific for GCG<sup>12</sup>. It is important to bear in mind that the X-ray appearance of the lesion is not pathognomonic and may be confused with many other lesions of the maxilla and the mandible<sup>3</sup>.

Histologically, multinucleated giant cells, in a cellular vascular stroma, and often new bone formations are detected. The osteoclast-like giant cells have a patchy distribution and are usually associated with areas of haemorrhage. Ultrastructurally, the proliferating cells include spindle-shaped fibroblasts, myofibroblasts, and inflammatory mononuclear cells<sup>13</sup>. GCG is a vascularized lesion that shares many features with the aneurysmal bone cyst. Differentiation from the latter can also be difficult histologically. However, the aneurysmal bone cyst is characteristically composed of honeycomb blood-filled spaces with a lining of flat non-endothelial cells<sup>14</sup>. Brown tumour of hyperparathyroidism is histologically very similar to GCG. Thus, all patients with suspected GCG, should have serum calcium, phosphate and alkaline phosphatase levels evaluated, to exclude the possibility of hyperparathyroidism<sup>5</sup>. The non-ossifying fibroma is histologically very similar to the GCG which can, however, be differentiated from the latter since the granulo-ma-like aggregate of giant cells and the fibrous stroma found in GCG, are not typical features of non-ossifying fibroma<sup>5</sup>.

Surgery is the traditional and most accepted form of treatment for GCGs. However, the extent of tissue removal ranges from simple curettage to en bloc resection<sup>3</sup>.

The incidence of recurrence, after surgery, is 4-20%, whereas local aggressive giant cell lesions have a higher recurrence rate<sup>15,16</sup>.

Several surgical techniques have been proposed for the removal of more aggressive GCG lesions, and, indeed, for aggressive lesions presenting with pain, rapid growth, facial swelling, Choung et al. advise en bloc resection, with immediate reconstruction of the affected area, as the most appropriate treatment<sup>8</sup>.

To avoid the disadvantages of surgical treatment, such as recurrence (see above), severe facial mutilation and loss of teeth and tooth germs, GCGs have also been treated by non-surgical methods which include radiotherapy, daily systemic doses of calcitonin, and intra-lesion injection with corticosteroids<sup>3</sup>.

Some GCG may also be sterilized thermally, using laser or cryoprobe. Much controversy still exists, in the literature, concerning radiation treatment and has been reported as a potential risk for malignant trans-

formation of the lesion<sup>17</sup>.

Treatment of GCG of the maxilla and mandible with calcitonin has also been reported<sup>18</sup>. GCGs have been successfully treated with calcitonin, in various concentrations, for at least 1 year, and complete remission of GCG has been observed, without signs of recurrence<sup>18,19</sup>. However, calcitonin therapy can be complicated by side-effects, such as hypocalcaemia and secondary hyperparathyroidism<sup>18</sup>. Furthermore, the relatively long duration of treatment can be more intolerable, for some patients, especially children<sup>3</sup>.

Another form of treatment for GCG consists in weekly intra-lesional injections with corticosteroids, successful results having been reported in the literature<sup>20</sup>. Steroid treatment was first suggested by Body et al., in 1981<sup>21</sup>. Corticosteroid treatment is, however, relatively contraindicated in certain medical conditions, such as diabetes mellitus, peptic ulcer, and generalized immuno-compromised states.

Non-surgical therapies for GCG are probably good treatment options for small slow-growing lesions, while successful treatment of painful, large, and rapidly growing lesions is still more likely to be achieved by surgical removal<sup>3</sup>.

## Conclusions

Giant cell granuloma is a rare disease of the head and neck region.

Surgery is the traditional and still most accepted treatment for GCGs, but it is important to bear in mind that, today, modern surgery can be performed in association with new approaches, in an attempt to avoid recurrence.

Indeed, the present case, in which we used a combined approach with local injection of corticosteroids, after surgery, supports this opinion.

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