



Case report

Surgical treatment of central giant cell granuloma of the jaws in children

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ABSTRACT

Introduction: There has been much debate in the literature regarding the preferred method of central giant cell granuloma (CGCG) treatment. Non-surgical methods are widely accepted, including intralesional corticosteroid injections, calcitonin therapy and the rarely used interferon therapy but surgical excision is the preferred method of treatment.

Aim: The objective of this study was to describe our experience in the surgical treatment of CGCG in the paediatric population.

Material and methods: A case study of 9 paediatric patients with the average age of 10 years is discussed in this article. Most patients were diagnosed with mandibular CGCG lesions, and multifocal tumours localized in the mandible and the maxilla were noted in 2 cases.

Results and discussion: The surgical procedures performed in our department included enucleation (2 patients), en bloc resection (2 patients) and segmental resection (5 patients). Segmental resections require further reconstruction. A surgically created defect is repaired with the involvement of frozen or autologous bone grafts, distraction osteogenesis and microvascular customized multiple tissue flaps.

Conclusions: Our experience indicates that despite the use of a meticulous surgical technique, patients at risk of tumour recurrence have to remain under strict clinical observation.

1. INTRODUCTION

The term ‘central giant cell granuloma’ (CGCG) was introduced in 1953 by Jaffe.¹ This benign osteolytic lesion of the jaw is also referred to as central giant cell lesion (CGCL).² Central giant cell granuloma occurs relatively rarely and represents 7% of benign jaw lesions.³ It is noted in almost every age group, but its incidence is highest in children and adults younger than 30 years. Research suggests that CGCG is nearly three times more prevalent in women than men.⁴ In the craniofacial region, CGCG is localized mainly in the mandible, and it can cross the midline.³ Multifocal lesions are also observed, often in combination with hyperparathyroidism, Noonan syndrome, neurofibromatosis type 1 and cherubism.²

According to Chuong et al., CGCG can be subdivided into aggressive and non-aggressive types depending on clinical and radiographic characteristics.⁵ The non-aggressive form may be discovered during routine panoramic radiography as a radiolucent mass. This asymptomatic, slow-growing lesion has a low recurrence rate. The aggressive form may demonstrate alarming features upon physical examination, including painful swelling with facial asymmetry and malocclusion. Computed tomography (CT) scans and radiographs reveal cortical bone destruction and, in some cases, root resorption. The aggressive form of CGCG has a higher recurrence rate.^{2–4}

The etiology of CGCG remains unknown.⁴ Morphologically, CGCL reveals aggregations of multinucleated giant cells, multiple foci of haemorrhage and trabeculae of woven bone, and high vascular density. Histologically, CGCG requires differentiation from bone tumours associated with hyperparathyroidism.^{2,6}

Surgical excision is the preferred method of CGCL treatment, and it ranges from curettage with cryotherapy to segmental resection.^{4,7,8} The condition is most prevalent in children, therefore, conservative treatment may be required to reduce secondary deformities. Non-surgical methods include intralesional corticosteroid injections, calcitonin therapy and the rarely used interferon therapy.⁹ According to De Lange et al., surgical treatment carries the lowest risk of recurrence, and the 5-year disease-free survival rate after surgical curettage is 76.1%.¹⁰

2. AIM

The objective of this study was to describe our experience in the surgical treatment of CGCG in the paediatric population.

3. MATERIAL AND METHODS

All of the examined patients were analysed with reference to the anatomical location of the tumour, age, gender, clinical status, radiological features, method of treatment, treat-

ment complications and incidence of recurrence. A total of 9 patients received treatment for CGCG in our department between July 2014 and June 2016, including 2 females (22%) and 7 males (78%). Upon admission, the patients’ age ranged from 2 to 17 years, with the average age of 10 years. Hyperparathyroidism was excluded in all cases by measuring serum calcium, phosphorus and alkaline phosphatase levels. CGCG was confirmed by histopathology.

4. RESULTS

In our study, lesions were most commonly localized in the mandible (5 patients, 56%) and the maxilla (2 patients, 22%). Multifocal CGCG was observed in 2 patients (22%), where a large bilateral lesion between the left and the right mandible ramus with another focus in the left maxilla was noted in the 1st patient (Figure 1), and a lesion in the right angle of the mandible, the mandibular body and the left maxilla near the midline was observed in the 2nd patient.

Six patients had received previous treatment in other institutions, 4 patients had been subjected to intralesional corticosteroid injections with curettage, and 2 patients had undergone lesion enucleation. Six patients were admitted for treatment in our department due to residual lesions or recurrence after the first treatment.

Our approach to CGCG involves complete surgical resection for further reconstruction of the missing tissues. Only 2 (22%) patients were qualified for enucleation. To expand surgical margins, the resected area was additionally devitalized and ablated using argon plasma coagulation. In the remaining 7 cases, the lesions were large, and en block excision was the surgical procedure of choice. Monolateral resection of the ramus and the body of the mandible was performed in 3 patients. In 1 case, bilateral resection of the mandible ramus was required (Figure 1). In this patient, both condyles and a small frontal part of the mandible were left (Figure 2). Mandibular reconstruction with free fibular flap was immediately performed in all 3 patients.

Two patients from this group were also diagnosed with CGCL of the maxilla. These lesions were removed after the resection of mandibular tumours. Two recurrent episodes were noted in one of the patients – 1 in the maxilla and 1 in the mandible. Subsequent widening of surgical margins was performed with good result. In the described cases, non-ossifying fibromas of long bones were observed. One patient was diagnosed with a small asymptomatic lesion near the right knee-joint. In the other patient, a large lesion in the left femoral neck was treated by enucleation and stabilization with the use of LCP hip plates and neck screws.

In the following 3 patients, marginal resection of the anterior part of the mandible was performed without disruption of bone continuity. One of the patients had to be reoperated due to recurrence. Surgical margins were widened, CGCL was not detected, and the mandible was reconstructed by alveolar ridge distraction. In the last patient, the lesion was localized in the left frontal part of the maxilla



Figure 1. 9-Years-old patient with multifocal CGCL tumour.



Figure 2. The same patient as in Figure 1 after tumour resection and reconstruction.

under the nasal cavity. It was resected widely and reconstructed with a free fibular flap 4 months later (Table 1).

5. DISCUSSION

CGCG occurs relatively rarely and represents 7% of benign jaw lesions.³ It is diagnosed in almost every age group, but is most prevalent in children. The diagnosis is made based on physical examination and radiographs, followed by histological confirmation.

In our study, 7 patients were diagnosed with the aggressive form of CGCG, which was associated with rapid lesion growth, facial asymmetry and tooth displacement. CT scans and radiographs revealed large radiolucent masses with cortical bone destruction.

There is mounting evidence to suggest that intralesional corticosteroid injections are effective, especially in aggressive forms of CGCG. Four of our patients were diagnosed with recurring or residual lesions after steroid treatment in

other institutions. We found that unsuccessful injections prolong the time from diagnosis to surgery, which contributes to tumour growth.

Despite the growing number of treatment options, surgery remains the gold standard.^{4,9} The range of the resection is correlated with age, and its extensiveness generally increases in older patients.

In our department, enucleation and curettage involve devitalization and ablation of the post-resection gap by argon plasma coagulation. This surgical technique was applied in 2 patients who remained under observation for 8 months with no signs of recurrence. Segmental resection is always performed with further reconstruction in mind. Reconstruction is performed with frozen or autologous bone grafts or microvascular, customized multiple tissue flaps. Distraction osteogenesis may be performed when sufficient quantities of tissue are available. The reconstruction modality also depends on a child's age. Treatment can begin with a conservative approach, followed by more advanced surgical techniques.

Table 1. Characteristics of study patients

Case no.	Age, y	Gender	Anatomical location	Clinical status	Radiological features	Method of treatment	Previous treatment	Incidence of recurrence in our Department
1	2	M	Mandible, frontal part	Rapidly growing, painless tumour	Radiolucent masses	En bloc resection with ablation of the post-resection gap using argon plasma coagulation	None	None
2	3	M	Mandible, left body	Painless swelling	Radiolucent masses with cortical bone destruction	En bloc resection with ablation of the post-resection gap using argon plasma coagulation	Enucleation in another institution, followed by recurrence	None
3	7	F	Mandible, left body	Rapidly growing, painful tumour	Radiolucent masses with cortical bone destruction	Enucleation with ablation of the post-resection gap using argon plasma coagulation	None	None
4	9	M	Maxilla, hard palate	Painful tumour	Radiolucent masses with cortical bone destruction	Enucleation with ablation of the post-resection gap using argon plasma coagulation	Enucleation and intralesional corticosteroid injections in another institution, followed by recurrence	None
5	9	M	Mandible, bilateral ramus and body, Maxilla, left side	Rapidly growing, soft tissue swelling	Tooth displacement, Radiolucent masses with cortical bone destruction	Segmental resection	Intralesional corticosteroid injections in another institution with no improvement	None
6	12	F	Maxilla, left, frontal part	Painful, soft tissue swelling	Tooth displacement, Radiolucent masses with cortical bone destruction	Segmental resection	Enucleation in another institution, followed by recurrence	None
7	16	M	Mandible, mental part	Tooth displacement, painful tumour	Tooth displacement, Radiolucent masses with cortical bone destruction	Segmental resection	None	One episode of recurrence with subsequent widening of surgical margins
8	16	M	Body of mandible, right side, ramus Maxilla, left, frontal part	Tooth displacement, painful tumour	Tooth displacement, Radiolucent masses with cortical bone destruction	Segmental resection	Intralesional corticosteroid injections in another institution with no improvement	Two episodes of recurrence: one in the maxilla and one in the mandible with subsequent widening of surgical margins
9	17	M	Body of mandible, left side, ramus	Tooth displacement, painful tumour Soft tissue swelling	Tooth displacement, Radiolucent masses with cortical bone destruction	Segmental resection	Intralesional corticosteroid injections in another institution with no improvement	None

Surgery is usually performed during a single-stage procedure, but reconstruction may be delayed when surgical margins are uncertain. CGCG is localized mainly in the mandible, but the observations made in 2 of the studied patients indicate that special attention should be paid to multifocal lesions. The entire skeleton is not routinely scanned for additional lesions. When microvascular reconstruction is needed, the donor site is analysed in a CT scan. In 2 patients, CT revealed non-ossified fibromas in the femur. In 1

of these patients, the lesion was large, therefore, the patient was subjected to enucleation and stabilization with an LCP hip plate and neck screws as well as reconstruction with the use of frozen bone grafts.

Our experience indicates that despite the use of meticulous surgical technique, patients at risk of recurrence have to remain under long-term clinical observation and radiological surveillance.

6. CONCLUSIONS

CGCG occurs relatively rarely and represents 7% of benign jaw lesions.³ It is diagnosed in almost every age group, but is most prevalent in children. The diagnosis is made based on physical examination and radiographs, followed by histological confirmation.

Non-surgical methods of treatment are widely applied, including intralesional corticosteroid injections, calcitonin therapy and the rarely used interferon therapy. Despite the above, surgery remains the gold standard.

The results of this study indicate that wide resection is relatively safe even in children provided that reconstruction is well planned. Surgeons should be able to choose from a variety of techniques to effectively reconstruct the defect.

The patients should remain under clinical observation due to the high risk of GCGC recurrence.

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