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Case Report Open Access

Central Giant Cell Granuloma Resistant to Calcitonin Nasal Spray: A Case Report

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Abstract

Purpose: We describe successful treatment of a recurrent central giant cell granuloma (CGCG). This is a rare benign intraosseous lesion that occurs before age 30 and can quickly recur as a painless lesion of the mandible. Because of its aggressive growth and tendency to progress as well as its morphological similarity to a giant cell tumour, it justifies radical surgery.

Patients and methods: A female patient aged 18 years presented with a CGCG lesion in the mandible that recurred despite standard treatment with calcitonin nasal spray after enucleation of the initial lesion.

Results: The recurrence in the mandible required radical surgery and reconstruction with a microvascular iliac bone flap; there was no injury to cranial nerves and facial appearance was normal.

Conclusion: Radical surgical treatment of CGCG recurrence is the best option when calcitonin nasal spray fails to prevent recurrence.

Keywords: Central giant cell granulomas; Calcitonin nasal spray; Radical surgical treatment

Introduction

Central giant cell granulomas (CGCGs) of the jaws are rare benign intraosseous lesions that can quickly recur as indolent lesions in mandible, where they are more commonly found than in the maxilla. Conventional treatment is local curettage, which is associated with a high success rate and low recurrence rate [1]. Recurrences have nonetheless been noted and studies suggest recurrence rates up to 20% with local curettage [1]. A more aggressive curettage or enucleation has been recommended, possibly coupled with adjunctive treatment such as liquid nitrogen cryotherapy [2]. To minimize risk of poor esthetic or functional outcome, many clinicians treat aggressive CGCLs with intralesional triamcinolone, intranasal calcitonin or subcutaneous interferon alpha-2a. Radiation therapy and other chemotherapeutic agents such as methotrexate, doxorubicin, and cyclophosphamide have been employed. Other alternative treatments include intralesional steroid injections at weekly intervals for six weeks [3-6]. A number of alternative nonsurgical therapies have been advocated for the management of CGCG. Calcitonin antagonizes osteoclastic bone resorption and acts directly on other cell types in this lesion, and calcitonin receptors have been identified on giant cells of the lesion [7,8]. Lim and Gibbins reported that immunohistochemistry suggests that stromal cells or fibroblasts are the etiologic basis of CGCG and that the giant cells themselves may be secondary or reactive [9]. Maerki et al. [10] wrote that although it was initially referred to as a "giant cell reparative granuloma", due to the accepted notion of its tendency to repair areas of injury, the term "giant cell granuloma (GCG) is now more frequently used as this lesion has been found in patients without a history of trauma. In addition, several cases of a destructive rather than reparative nature have been observed. Maerki et al. described a case that supports the theory of trauma and inflammation as risk factors for GCG, so that care must be taken when GCG presents in unusual locations as in a case where it was in the middle cranial fossa in a mixed material arts fighter [10]. Because of its aggressive growth and tendency to progress, the central giant cell granuloma has been described as a benign lesion, although its morphological similarity to a giant cell tumour demands a radical surgical approach. This case report describes successful radical surgery after calcitonin nasal spray failed to prevent recurrence of a CGCG lesion.

Patients and Methods

A female patient aged 18 years with a biopsy-proven CGCG was treated with calcitonin nasal spray after enucleation of the mandibular lesion. The indication for additional administration of calcitonin therapy was a clinically and radiographically aggressive variant of CGCG that recurred in the presence of hyperthyroidism. The patient had a biopsy and evaluation for hyperthyroidism (serum phosphate, calcium, parathormone, parathormone related protein) prior to surgical enucleation of the left mandible followed by intranasal salmon calcitonin (100 IU/day) for six months. In spite of the nasal spray, five months after the first operation she developed a recurrent expansive osteolytic process that extended from the first molar to the collum of the left mandible with swelling and pain. The aggressive CGCG lesion grew rapidly and expanded in spite of calcitonin therapy. Calcitonin was stopped and she underwent radical partial resection of the left mandible with a microvascular musculocutaneous iliac bone flap. She was monitored every three months clinically and radiographically; blood samples were taken to assess bone formation and calcium metabolism on the basis of calcium, phosphate, PTH, alkaline phosphatase and osteocalcin. To evaluate bone resorption, 24-h urinary calcium and hydroxyproline were determined every three to six months. She was monitored by PET/CT for regression or recurrence.

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Results

This 18-year-old woman has been followed for three years after first presenting with an aggressive lesion that was enucleated surgically and proved upon biopsy to be a central giant cell granuloma. The criteria for choosing the patient for additional administration of calcitonin were a clinically and radiographically aggressive variant of CGCG and a recurrent CGCG along with a suspicion of hyperparathyroidism. A work-up at our hospital's Division of Endocrinology and Nuclear Medicine failed to confirm this. Our patient was evaluated radiologically and clinically every three months. Orthopantomography after 5 months showed a recurrent lesion more than 4 cm in length and 2 cm in height that was growing rapidly and aggressively in the left mandible (Figure 1), although calcitonin nasal spray had been initiated immediately after the enucleation. Absorption of the nasal spray is known to be erratic and varies between 20% and 100%. The calcintonin spray was discontinued and the patient underwent radical surgery (Figure 2) with a microvascular iliac bone flap (Figure 3). Healing after the second intervention was uneventful and she regained normal facial appearance after resolution of the postoperative edema (Figure 4). The function of the cranial nerves was preserved.

Discussion

Central giant cell granulomas (CGCG) of the jaws are histologically benign lesions characterized by the presence of giant cells in the richly vascularized stroma of the spindle cells. They are morphologically similar to giant cell tumours of the long bones. The giant cell reparative granuloma [11] as it was originally called is not a granuloma in the histologic sense. It is not reparative clinically, apparently demonstrating neoplastic tendencies [12]. Some studies consider giant cell tumors to be a manifestation of the central giant cell granuloma because of the histopathology, whereby age and local factors are responsible for varying clinical characteristics [1,13,14]. The differential diagnosis of CGCL includes the so-called "brown tumor of hyperparathyroidism" (BTOH), peripheral giant cell granuloma, and giant cell tumor of bone. The end result of PTH secretion is osteoclast proliferation, bone resorption, and calcium liberation. Pathologically sustained PTH elevation causes BTOH, which consists of mononuclear osteoclast precursors and multinucleated differentiated osteoclasts. None of these differential diagnoses was confirmed. Intranasal calcitonin spray is the usual postoperative treatment; it can be self- administered and has minimal side effects [15-17]. At present, the nature of the CGCG is unknown, and it is still not known whether the central giant cell granuloma represents an inflammatory, reactive, infective, or neoplastic process. Some authors describe the CGCG as a progressive lesion with potential for aggression. In our case, radical surgery for CGCG with preservation of function and normal facial appearance was effective. In view of the recurrence after the initial enucleation, there was no reason to continue the calcitonin after radical surgery, as it had not prevented recurrence after the first surgery and resistance to the spray had apparently developed.

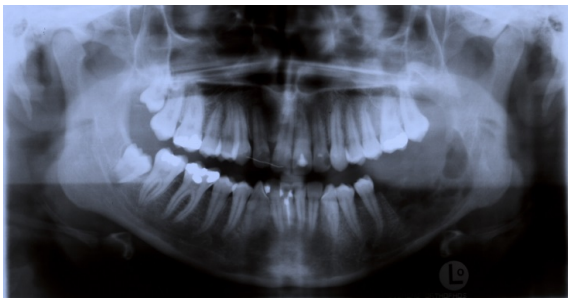

Figure 1: Patient with increased growth of the CGCG.

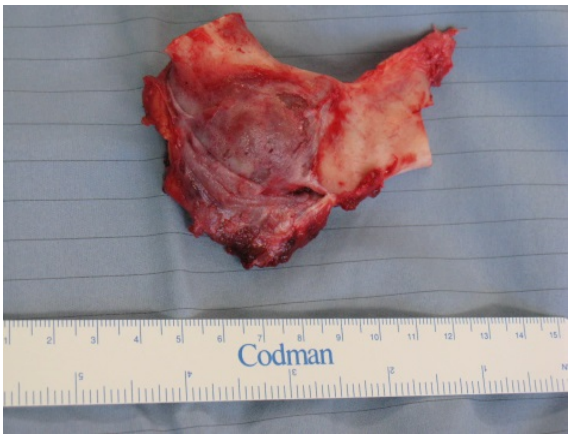

Figure 2: Patient with increased growth of the CGCG.

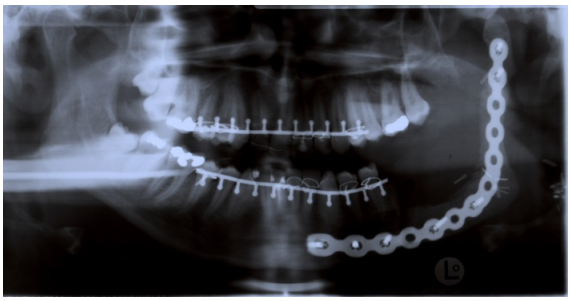

Figure 3: Postoperative x-ray (from Figure 2) after radical partial resection of the mandible.

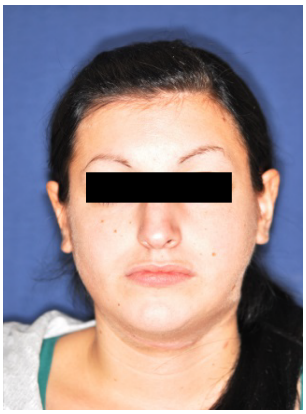

Figure 4: Patient after radical partial resection of the mandible.

Conclusions

We can offer no explanation as to why the patient had no recurrence after radical surgery but suggest that this is the best treatment option when enucleation of a CGCG and calcitonin spray are ineffective.

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