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Central Giant Cell Granuloma in an 8-Year-Old Male Child: A Case Report

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ABSTRACT

Central Giant Cell Granuloma (CGCG) is a non-neoplastic, locally aggressive lesion of the jaws, primarily affecting children and young adults. This report details the diagnosis and surgical management of an aggressive, bilateral CGCG in an 8-year-old male child, a presentation that is clinically rare, particularly in the paediatric population. The patient presented with progressive, asymptomatic, firm bilateral mandibular swellings of two years' duration, leading to the premature exfoliation of multiple lower teeth. Computed Tomography (CT) scans revealed extensive, multilocular radiolucencies in both the body and rami of the mandible, exhibiting cortical thinning, root resorption, and cortical perforations, indicative of aggressive biological behaviour. An incisional biopsy confirmed the diagnosis of CGCG. Due to the lesion's extensive size and bilateral involvement, surgical intervention was elected. Management involved bilateral submandibular approach, comprehensive resection, and curettage, followed by intralesional irrigation with triamcinolone acetonide to mitigate recurrence risk. This case underscores the diagnostic challenge and the necessity for aggressive, tailored surgical management in extensive, bilateral CGCG in paediatric patients, while emphasizing the importance of utilizing adjunctive therapies to minimize morbidity and prevent recurrence.

Keywords: Central Giant Cell Granuloma, Mandible, Bilateral, Paediatric, Curettage, Triamcinolone

INTRODUCTION

Central giant cell granuloma (CGCG) is "a non-neoplastic proliferative lesion with unknown etiology which commonly occurs in the mandible" (Nosratzehi et al., 2014). This lesion displays clinical behaviour that ranges from nonaggressive to aggressive variants (de Lange et al., 2007). CGCG was first described by Jaffe in 1953 and was initially not distinguished from giant cell tumour of the extragnathic skeleton, but later it was described by Jaffe as "giant cell reparative granuloma" (Wadde et al., 2020).

LITERATURE REVIEW

CGCG "is found predominantly in children and young adults, with more than 60% of all cases occurring before the age of 30 years" (Chrcanovic et al., 2018; Wadde et al., 2020). The female to male ratio is reported to be "2:1" (Wadde et al., 2020). Lesions are more common in the "anterior region of the jaws, and mandibular lesions frequently extend across the midline" (Wadde et al., 2020). The "etiopathogenesis of CGCG is still not properly known" (de Lange et al., 2007). CGCG can be "difficult to diagnose from other lesion of oral cavity like Hyperparathyroid tumour, Ameloblastoma, Odontogenic myxoma, Haemangioma, Cherubism, Central odontogenic fibroma, Aneurysmal bone cyst, Traumatic bone

cyst" (Wadde et al., 2020). "Histologically and radiographically CGCG resembles giant cell tumour, giant cell lesion of hyperparathyroidism, cherubism, and aneurysmal bone cyst" (Wadde et al., 2020). Radiographically, CGCG "presents as either unilocular or multilocular radiolucent lesions in the maxilla or mandible" (de Lange et al., 2007). The "CT features include unevenly dense expansile mass causing bone destruction and cortical thinning" (Shrestha et al., 2021). On MRI, the lesion shows "low to iso-intensity in T1 weighted and T2 weighted images" (Shrestha et al., 2021). There may be "presence of cystic degeneration, haemorrhage or hemosiderin deposits or osteoid formation, which can cause T1 and T2 signal changes" (Shrestha et al., 2021). On contrast study, "the lesion doesn't enhance but periphery may enhance mildly" (Shrestha et al., 2021).

Clinically, CGCG most commonly occurs in the jawbones as an asymptomatic swelling (de Lange et al., 2007). However, it exhibits a spectrum of behavior ranging from indolent, painless lesions to aggressive variants associated with pain, rapid expansion, and cortical perforation (Kruse-Lösler et al., 2006). CGCG is "diagnosed through histopathological examinations" (Nosratzehi et al., 2014). Histologically, the lesion is characterized by a variable number of multinucleated giant cells embedded in a background of 'ovoid-to-spindle-shaped mononuclear cells' (Kruse-Lösler et al., 2006). Histopathologic examination often reveals a cellular stroma with 'hemorrhagic focuses' and the presence of hemosiderin pigment (Nosratzehi et al., 2014). Imaging modalities, such as CT scan and MRI, play a crucial role in the evaluation of CGCG. "All 7 cases had undergone CT scan, 3 cases had undergone MRI scan" in one study (Shrestha et al., 2021). The CT and MRI findings mentioned earlier provide valuable information for the diagnosis and assessment of CGCG.

The "treatment of CGCG varies from local curettage to wide surgical excision depending upon the extent and progression of the lesion" (de Lange et al., 2007). Numerous treatment modalities are described in literature ranging from non–surgical to surgical (Wadde et al., 2020). Regarding management, the literature notes that 'the common treatment of CGCG is surgical removal,' with interventions ranging from 'simple curettage to en-bloc resection' (Kruse-Lösler et al., 2006). The potential transition from aggressive operative treatments to conservative therapies, which help maintain jaw function, has been highlighted in recent reviews (de Lange et al., 2007). In the past, resection or curettage has been used to treat it. When surgery is linked to higher morbidity or when used as an adjuvant to reduce recurrence, medical therapy is recommended. A RANK ligand inhibitor called denosumab has not been thoroughly studied in relation to the treatment of CGCG, especially in children. In Choe et al. (2021), the discussion revolved around treating six children with CGCG with denosumab. Denosumab appeared to be a useful treatment option with minimally intrusive side effects, as all patients experienced a positive response. In another study, Intralesional infiltration of a solution of Kenacort-A (10 mg/mL, triamcinolone aqueous suspension SQUIBB) and either (1) Lidocaine 2% with epinephrine 1:200,000 Marcaine or (2) Bupivacaine, 50% mixture by volume, was used to treat four cases of central giant cell granuloma. Radiographic and histologic studies were initially used to diagnose these cases in three male Guatemalans (ages 31, 34, and 6), as well as in a female Guatemalan (age 21/2) (Carlos & Sedano, 2002). For adults, the average dosage of the aforementioned solution was 6 mL, or 30 mg of triamcinolone, while for pediatric patients, it was 5 mL, or 25 mg of triamcinolone. An endocrinologist assessed each patient prior to treatment in order to rule out hyperparathyroidism. Prior to beginning treatment, a microscopic examination of the lesion was obtained from each patient through an incisional biopsy.

All of the cases' follow-up radiographs demonstrated the lesions' gradual improvement and eventual resolution (Carlos & Sedano, 2002). Additionally, Harris (1993) found that administering human calcitonin 0.5 mg (100 iu) deep subcutaneously for 1 year can effectively eliminate aggressive central giant cell granulomas. This may eliminate the necessity for invasive surgery or radiotherapy in pediatric patients. Calcitonin therapy seems to be a feasible choice for treating CGCGs. However, due to the lengthy duration of treatment, it is advisable to consider this option for cases involving multiple lesions, recurring lesions, or highly aggressive lesions (Pogrel, 2003). In yet another study Imatinib, a medication that inhibits the activity of tyrosine kinase, was prescribed due to the clinical similarities observed between CGCJ and cherubism. Imatinib has shown effectiveness in treating cherubism. After a period of 2 months, a computed tomographic scan revealed a substantial increase in ossification, which continued to grow over the subsequent 8 months. This case indicates that tyrosine kinase inhibitors could be a viable and low-toxicity treatment option for CGCJ (Tallent et al., 2022).

Literature increasingly emphasizes a shift from radical surgical interventions toward conservative management strategies that prioritize functional preservation (de Lange et al., 2007). In conclusion, CGCG is a non-neoplastic lesion that presents with a wide range of clinical and radiological manifestations, making it a diagnostic challenge. Comprehensive evaluation through imaging, histopathological examination, and clinical features is essential for accurate diagnosis and appropriate management. The treatment options vary from conservative approaches to surgical interventions, depending on the extent and behavior of the lesion.

CASE REPORT

An 8-year-old male child from Sotik County, South Rift Region, Kenya, presented with bilateral mandibular swelling of two years' duration. The swelling had progressively increased in size, leading to the premature exfoliation of several lower teeth. Despite the extensive nature of the lesion, it was asymptomatic; the patient reported no pain, tenderness, or difficulty with mastication. A review of the dental history confirmed that the patient had previously visited a clinic where an incisional biopsy established the diagnosis of giant cell granuloma. No history of surgeries, allergies, admissions, chronic illnesses, or blood transfusions. The child was the last born in a family of 7 children who were alive and well.

On examination, we found a firm body of mandible swellings on both sides of the lower face. There was no scar, skin surface was smooth, and they were also ovoid. The consistency was firm and attached to underlying tissues, not mobile or fluctuant. It was immobile and irreducible. Intraorally, the upper teeth showed no signs of unnatural exfoliation. However, the lower teeth had multiple missing teeth. The following teeth were missing on the lower arch: 84, 85, 74, and 75.

X-ray tests included a CT SCAN, which revealed well-defined multilocular radiolucencies on both sides, as well as multiple strands of abnormally poorly defined trabeculae projecting into the lesion, as well as cortical plate thinning, root resorption, and cortical perforations that are typical of an aggressive lesion. An incisional biopsy confirmed the diagnosis of giant-cell granuloma. Serum calcium to rule out Parathyroid Hyperthyroidism was not done.

Figure 1: 8 year old swelling presenting with bilateral cheek swellings with no pain or tenderness

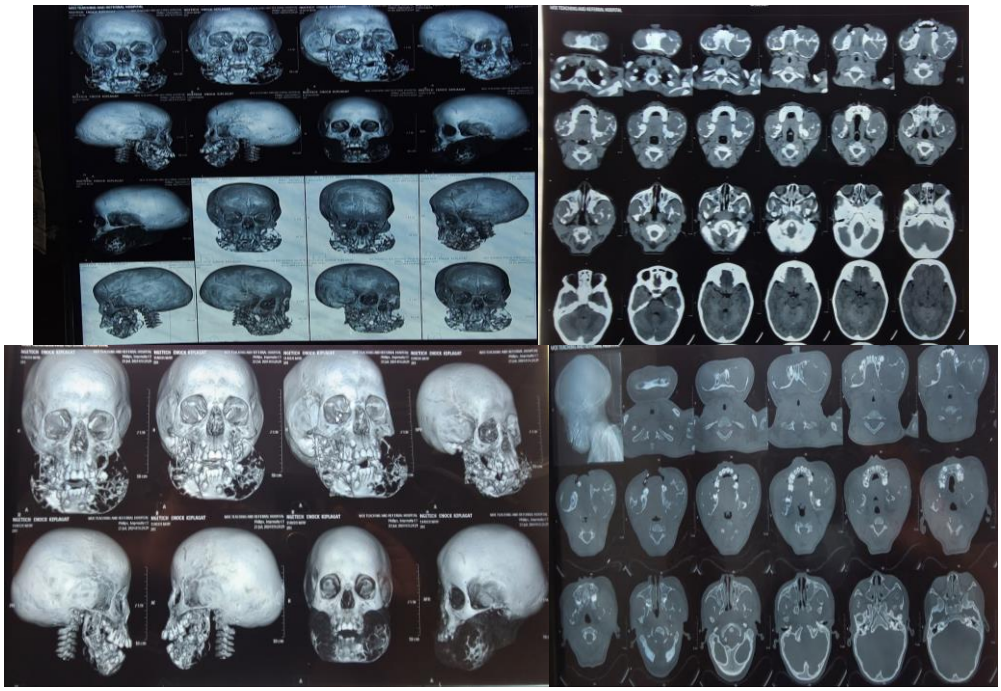

Figure 2: CT scan 3d and Axial views showing a Bilateral Osteolytic Lesion

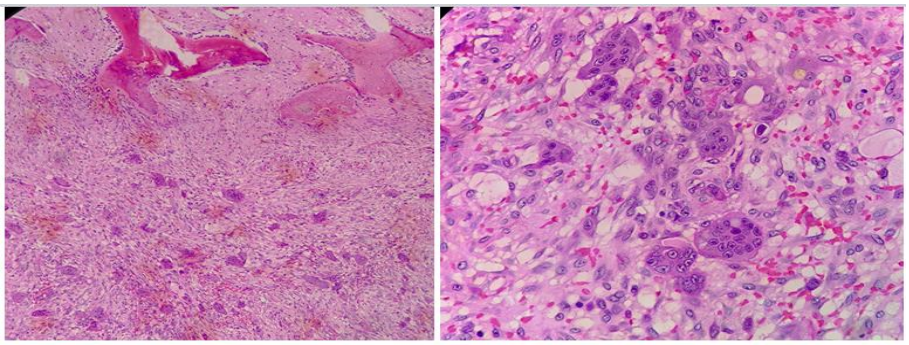

Figure 3: Histological section of Central Giant Cell Granuloma showing multinucleated giant cells scattered within a cellular fibroblastic stroma, with areas of hemorrhage and hemosiderin deposits. Bone trabecular evident at the periphery of the sections.

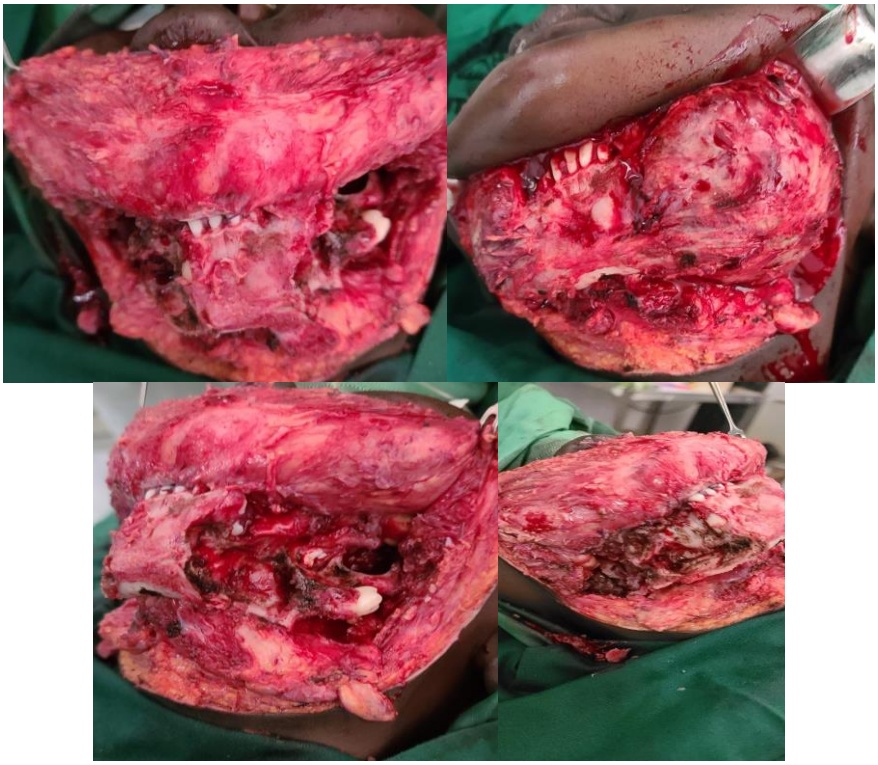

Figure 4: Bilateral Extended submandibular Incision used to raise a flap revealing multiple tumours necessitating multiple enucleations

The decision was to have surgical intervention with resection and plating in mind since the lesion was large in size. The patient was taken to the operating room and intubated nasally for surgical resection under general anesthesia. After cleaning and draping, a bilateral submandibular incision was made, and a flap revealed the body and the rami of the mandible.

Superficially, the lesion was resected as much as possible, leaving the core of the lesion exposed. The completion of the resection was done by curetting the multiple loculations of the lesion in a process of enucleation. Irrigation and continued curettage of all the cortices on both sides achieved an acceptable result. Final irrigation was performed with 160 mg of triamcinolone before closure, which was achieved with 4-0 Vicryl, 2-0 Vicryl, and 4.0 Prolene for the Skin.

DISCUSSION

Central giant cell granuloma (CGCG) is a benign, though potentially aggressive, lesion of the jaws characterized by a wide range of clinical and radiological manifestations. This case report highlights the diagnostic and therapeutic challenges presented by CGCG in an 8-year-old child. The clinical presentation of CGCG can vary significantly, ranging from slow-growing, asymptomatic lesions to more aggressive variants that cause pain, cortical bone destruction, and root resorption (Nosratzehi et al., 2014). In the presented case, the child exhibited bilateral mandibular swellings with missing lower teeth but without pain, which aligns with the typical clinical behavior of CGCG. Radiographic imaging showed multilocular radiolucencies with cortical thinning and root resorption, indicating an aggressive lesion.

Histopathological examination is essential for definitive diagnosis, typically revealing 'cellular fibrous tissue containing multiple foci of hemorrhage [and] aggregations of multinucleated giant cells' (Kruse-Lösler et al., 2006). In this case, an incisional biopsy confirmed the diagnosis, which is consistent with standard diagnostic protocols (Nosratzehi et al., 2014). Treatment of CGCG varies from conservative approaches, such as corticosteroid injections

and calcitonin therapy, to more invasive surgical interventions, depending on the lesion's size and aggressiveness. In this case, surgical resection followed by curettage and irrigation with triamcinolone was chosen due to the lesion's significant size and bilateral involvement. This approach is supported by literature indicating that surgery is often necessary for large or aggressive CGCGs (Chrcanovic et al., 2018; Wadde et al., 2020).

Recent advancements in medical therapy for CGCG include the use of denosumab, a RANK ligand inhibitor, which has shown promising results in pediatric patients (Choe et al., 2021). Although not utilized in this case, denosumab represents a potential alternative for future treatment, particularly in cases where surgery poses significant risks. The outcomes of different treatment modalities underscore the importance of a tailored approach based on the lesion's characteristics and patient factors. Conservative treatments like intralesional corticosteroid injections can be effective for smaller lesions, whereas extensive lesions often require surgical intervention (Carlos & Sedano, 2002; Harris, 1993; Pogrel, 2003). Moreover, emerging therapies such as tyrosine kinase inhibitors (e.g., Imatinib) offer new avenues for treatment, especially for recurrent or refractory cases (Tallent et al., 2022).

In conclusion, the management of CGCG necessitates a multidisciplinary approach involving accurate diagnosis through imaging and histopathology and a treatment strategy that balances the lesion's aggressiveness with the potential morbidity of intervention. Future research should continue to explore and refine both surgical and non-surgical treatments to optimize outcomes for patients with CGCG.

CONCLUSION

Central giant cell granuloma (CGCG) is a complex and varied condition that presents significant diagnostic and therapeutic challenges. This case report of an 8-year-old child with bilateral mandibular swellings highlights the importance of a comprehensive diagnostic approach that includes clinical examination, radiographic imaging, and histopathological analysis. The diverse clinical presentation of CGCG, ranging from asymptomatic lesions to aggressive, painful growths, necessitates a tailored treatment strategy. In this case, surgical intervention involving resection and curettage was necessary due to the lesion's extensive size and bilateral involvement. This approach is consistent with the recommendations in the literature for managing large or aggressive CGCGs. In this case, intralesional corticosteroids were used after resection, which is in line with current conservative treatment methods that aim to lower the risk of recurrence and the health problems that come with more invasive surgeries. Emerging treatments, such as denosumab and tyrosine kinase inhibitors like imatinib, offer promising alternatives, especially for pediatric patients where minimizing surgical intervention is crucial. These therapies represent significant advancements in the management of CGCG and underscore the potential for non-surgical options to play a larger role in the future. Ultimately, the successful management of CGCG requires a multidisciplinary approach that combines accurate diagnosis with a flexible treatment plan tailored to the individual patient's needs. Continued research and clinical studies are essential to further refine these approaches and improve outcomes for patients with CGCG.Conclude your research paper here.

Declaration by Authors

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