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Central Giant Cell Granuloma in a Dialyzed Patient

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Summary

Background. Central giant cell granuloma (CGCG) is regarded as a benign tumour affecting the jaws. The first description of this rare lesion was introduced by Jaffe in 1953. CGCG usually occurs in patients younger than 30 years, is more common in females than in males, and is more common in the mandible than in the maxilla. Electronmicroscopic and immunohistological analyses showed that CGCG is a process that arises from monohistocyte-like cells. CT is excellent for demonstration of bone thinning or destruction. MRI is the best modality of evaluating the extent of the lesion. Histologically, multinucleated giant cells in a cellular vascular stroma, and often – new bone formation, are demonstrated. Traditional management of CGCG is local excision by curettage. Intralesional corticosteroid injection as an alternative nonsurgical method for managing giant cell lesions was suggested. Calcitonin also has been reported as a treatment for CGCG.

Material and methods. Medical literature review was made and a case history presented. A 16-year-old patient presented with swelling on the right side of the mandible. The patient was on hemodialysis for the end stage renal disease for 5 years.

Results. According to medical literature, histologically there is no difference between brown tumors and CGCGs, and every histologically-confirmed giant cell lesion should be regarded as a potential brown tumor.

Conclusions. General practitioner dentists, oral and maxillofacial surgeons should be aware for the possible presence of brown tumors in the jaws of patients who have been diagnosed hyperparathyroidism.

Keywords: central giant cell granuloma, hemodialysis, hyperparathyroidism, child.

INTRODUCTION

The central giant cell granuloma (CGCG) is a common benign lesion of the jaws of unknown etiology, accounting for approximately 7% of all benign tumors of the jaw. The World Health Organization has defined it as an intraosseous lesion consisting of cellular fibrous tissue that contains multiple foci of hemorrhage, aggregations of multinucleated giant cells and occasionally trabeculae of woven bone [1].

CGCG usually occurs in patients younger than 30 years, is more common in females than in males, and is

more common in the mandible than in the maxilla [2]. CGCG is generally thought to be reactive rather than neoplastic in nature. Electronmicroscopic and immunohistological analyses showed that CGCG is a process that arises from monohistocyte-like cells. In the literature reviewed, a slight female predominance was found for CGCG, which may be explained by recent suggestions of the association between hormonal secretion and the appearance of CGCG in females. In young children, the craniofacial skeleton is actively developing to include osteogenesis, exfoliation and eruption of teeth. These processes cease in

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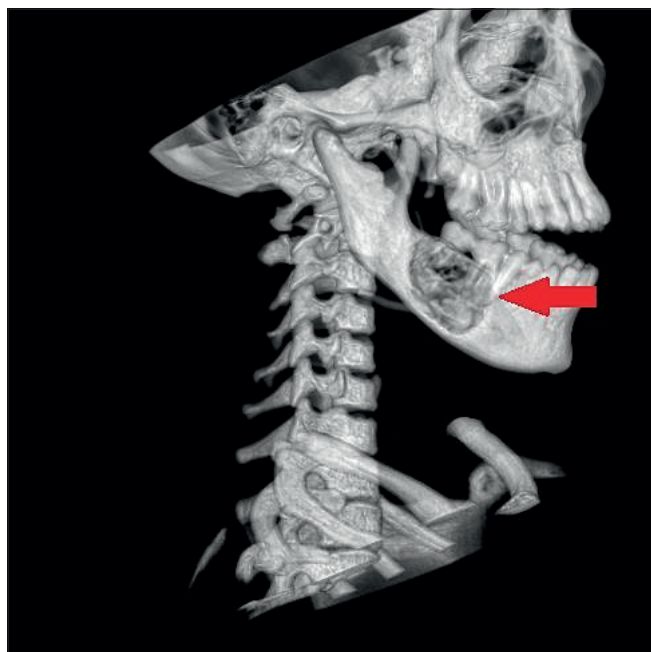


Fig. 1. The tumor in the right of the mandible is seen in 3D computerized tomography

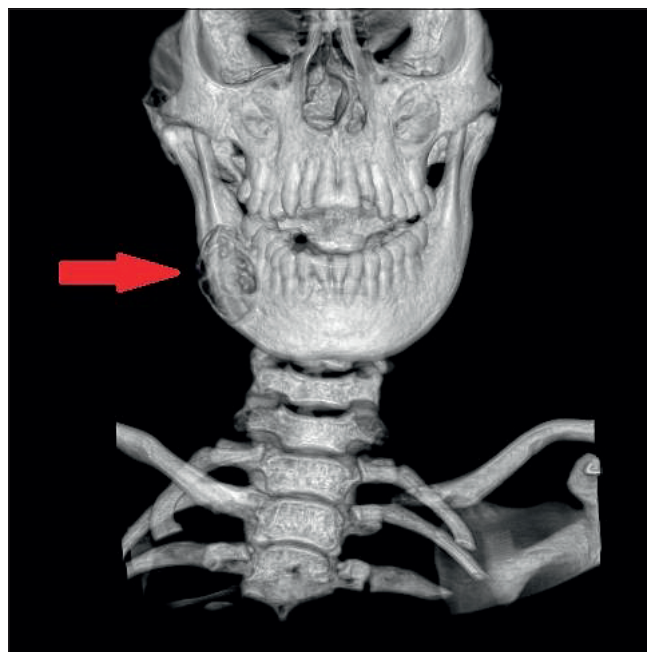


Fig. 2. The tumor in the right of the mandible is seen in 3D computerized tomography

adulthood and may therefore predispose CGCG formation in younger individuals [1].

The radiologic features of CGCG may appear as a unilocular or multilocular radiolucency, with well-defined or ill-defined margins and varying degrees of expansion of the cortical plates. It is important to remember that the radiologic appearance of the lesion is not pathognomonic and may be confused with that of many other lesions of the jaw [2].

CT is excellent for demonstration of bone thinning or destruction. The lesion attenuation is similar to muscle. MRI is the best modality of evaluating the extent of the lesion, as well as evaluating adjacent soft tissue. It has low to intermediate intensity signals on both T1W and T2W images, similar to Giant cell tumor. Mild post-contrast enhancement is evident both on CT and MRI [1].

Histologically, multinucleated giant cells in a cellular vascular stroma, and often new bone formation, are demonstrated. The osteoclast-like giant cells have a patchy distribution and are usually associated with areas of hemorrhage. Ultrastructurally, the proliferating cells include spindle-shaped fibroblasts, myofibroblasts, and inflammatory mononuclear cells [3].

Traditional management of CGCG is local excision by curettage. This may result in a major physical defect in the patient, especially in young patients with developing dentition.

Intralesional corticosteroid injection as an alternative nonsurgical

method for managing giant cell lesions was suggested by Jacoway et al., and this therapy may be very attractive option particularly for the young age group. Resolution of the lesion, or at least a significant reduction in size, may be achieved by using this method. Before beginning with intralesional corticosteroid injections, confirmation of the lesion by biopsy is mandatory. A careful pretreatment evaluation must be carried out in all patients for untoward side effects [4].

Calcitonin also has been reported as a treatment for CGCG. This hormone causes an increased influx of calcium into the bones, and thus, functions antagonistically to parathormone. It has been demonstrated that giant cells express calcitonin receptors. It is thought that giant cells are directly inhibited in their function by calcitonin [5].

A 16-year-old patient presented with swelling on the right side of the mandible. The swelling resulted in facial asymmetry in the right cheek and was painless. He did not have fever. There was no history of trauma or dental problems. Physical examination revealed a fixed mass on the right side of the mandible without tenderness. The range of motion in opening his mouth was not limited. Intraorally the crepitation in the mucofacial fold was palpated due to the thinned and inflated buccal wall of the mandible. The mucofacial fold was not infiltrated. There was no pain in teeth 46/47 but which showed increased mobility (II). Swallowing was not impaired. The tissue in the floor of the mouth showed

no change. Sensation in the teeth and lower lip were otherwise unaffected. In computerized tomography the right side of the mandible showed the bone thickened and filled with content similar in density to the soft tissues of the nasal septum. The lesion's size was 30×35.2×28 mm (Figs. 1 and 2). The cortical layer had thinned at the sides. The nerve canal was beneath the affected bone.

The patient underwent local excision of the mass and teeth 46/47. The postoperative course was uncomplicated.

The histopathological examination revealed an osseous formation composed of fibrous tissue with bundles of fibro/microfibroblasts, various sizes of osteoclast-type cells having multiple nuclei, areas of hemorrhage, hemosiderins, and immature osseous trabeculae with active osteoblasts at the periphery. The histopathologic diagnosis of central giant cell granuloma was made.

The patient was born with multiple anomalies: congenital anomalies of the kidney and urinary tract, congenital heart disease, retarded development. The patient undergone many surgeries for hydroureter, hydro-nephrosis and heart disease problems and was on hemodialysis for the end stage renal disease for 5 years. Laboratory investigations revealed elevated serum parathormone level of 1942 pg/mL (normal range 10–69 pg/mL). During this time he was treated with alfacalcidol daily for secondary hyperparathyroidism because of chronic kidney disease.

RESULTS

This is the first case of central giant cell granuloma in all patients with chronic renal failure we have had in our practice. According to medical literature, histologically there is no difference between brown tumors and CGCGs, and every histologically-confirmed giant cell lesion should be regarded as a potential brown tumor. A final diagnosis can be defined only by evaluating the radiological findings with histopathological, laboratory and clinical data. A brown tumour is considered as pathognomonic of hyperparathyroidism secondary to renal failure, especially in patients on long-term hemodialysis [6]. Normalization of parathormone levels will often cause the brown tumours to regress or sometimes even resolve spontaneously [7]. Since this patient felt discomfort in the mouth the surgery was chosen for the treatment of the tumor. In other hand, it was very difficult to treat secondary hyperparathyroidism in this patient.

CONCLUSIONS

In our opinion, since our patient had a secondary hyperparathyroidism, the final diagnosis is a brown tumour. The basic prevention of brown tumor is early diagnosis and treatment of secondary hyperparathyroidism. General practitioner dentists, oral and maxillofacial surgeons should be aware for the possible presence of brown tumors in the jaws of patients who have been diagnosed hyperparathyroidism. ♦

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Santrauka

CENTRINĖ GIGANTINIŲ LĄSTELIŲ GRANULOMA: ATVEJO PRISTATYMAS

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Darbo tikslas. Centrinė gigantiųjų ląstelių granuloma (CGLG) yra gerybinis žandikaulių navikas. Pirmą kartą šią rečiau ligą aprašė Jaffe 1953 metais. CGLG dažniausiai diagnozuojama jaunesniems nei 30 metų pacientams, dažniau moterims nei vyrams. Dažnesnė jos lokalizacija yra apatinis, o ne viršutinis žandikaulis. Elektroninės mikroskopijos ir imuno-histocheminiai tyrimai rodo, kad CGLG susidaro iš panašių į histiocitus ląstelių. Kompiuterinė tomografija puikiai parodo kaulo suplonėjimą ar destruktiją. Magnetinio rezonanso tomografija yra geriausias būdas pažeidimo mastui įvertinti.

Šio straipsnio tikslas – pateikti 16 metų amžiaus paciento, kuriam atliekamos hemodializės ir diagnozuota centrinė gigantiųjų ląstelių granuloma, klinikinį atvejį, taip pat apžvelgti literatūrą centrinės gigantiųjų ląstelių granulomos etiologijos, patogenezės, diagnostikos ir gydymo klausimais. Histologinių tyrimų duomenimis, kraujagyslių stromoje randamos daugiabrandoelės gigantinės ląstelės ir dažnai – naujo kaulo formavimasis. Tradici-

nis CGLG gydymas yra vietinė ekscizija su kiuretažu. Buvo pasiūlytas alternatyvus nechirurginis gydymas vietinėmis kortikosteroidų injekcijomis. Kalcitoninas taip pat gali būti skiriamas CGLG gydyti.

Tyrimo medžiaga ir metodai. Apžvelgiama medicinos literatūra apie centrinę gigantiųjų ląstelių granulomą ir aprašomas klinikinis atvejis. Šešiolikos metų pacientas skundėsi dešiniojo apatinio žandikaulio patinimu. Pastaruosius penkerius metus jam buvo atliekamos hemodializės dėl galutinės stadijos inkstų funkcijos nepakankamumo.

Tyrimo rezultatai. Medicinos literatūros duomenimis, nėra histologinio skirtumo tarp rudųjų navikų ir CGLG. Kiekvienas histologiškai patvirtintas pažeidimas, radus gigantiųjų ląstelių, turėtų būti laikomas ruduoju naviku.

Išvados. Bendrosios praktikos odontologai, burnos, veido ir žandikaulių chirurgai turėtų žinoti, kad pacientų, kuriems diagnozuotas hiperparatiroidizmas, žandikauliuose gali išsivystyti rudasis navikas.

Reikšminiai žodžiai: centrinė gigantiųjų ląstelių granuloma, hemodializė, hiperparatiroidizmas, vaikas.

REFERENCES

1. Venkateshwarlu M, Geetha P, Radhika B. Central giant cell granuloma. Indian Journal of Dental Advancements 2010; 2(1).
2. Bataineh AB, Al-Khateeh T, Rawashdeh MA. The surgical treatment of central giant cell granuloma of the mandible. J Oral Maxillofac Surg 2002; 60: 756–61.
3. Üstündağ E, Iseri M, Müezzinoğlu B. Central giant cell granuloma. J Pediatr Otorinolaryngol 2002; 65: 143–6.
4. Rajeevan NS, Soumithran CS. Intralesional corticosteroid injection for central giant cell granuloma. Int J Oral Maxillofac Surg 1998; 27: 303–4.
5. De Lange J, Rosenberg AJWP, van den Akker HP, Koole R, Wids JJ, van den Berg H. Treatment of central giant cell granuloma of the jaw with calcitonin. Int J Oral Maxillofac Surg 1999; 28: 372–6.
6. Fineman I, Johnson JP, Di-Patre PL, Sandhu H. Chronic renal failure causing brown tumors and myelopathy: case report and review of pathophysiology and treatment. Journal of Neurosurgery 1999; 90: 242–6.
7. Alvarado F, Waguespack SG, Campbell JH, Williams TP. Expansile intraosseous lesion of the mandible. J Oral Maxillofac Surg 2003; 61: 1318–23.