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CASE REPORT / OLGU SUNUMU

Evaluation of a Central Giant Cell Granuloma and a Sialolithiasis with Cone Beam Computed Tomography and Ultrasonography: A Case Report

Santral Dev Hücreli Granülom ve Tükürük Bezi Taşının Konik Işınlı Bilgisayarlı Tomografi ve Ultasonografi ile Değerlendirilmesi: Olgu Sunumu

Melisa BOZKURT[1], Fatma Nihan AKSAKALLI[2], Nuran BAYRAMOV[3], Baturay ORNAĞ[4], Mehmet Oğuz BORAHAN[5], Ferit BAYRAM[6], Şebnem ERÇALIK YALÇINKAYA[7]

ABSTRACT

Central giant cell granuloma (CGCG) is a benign intraosseous tumor that can cause rapid enlargement, expansion and/or perforation of cortical bone and has no significant etiology. Its differential diagnosis depends on both histological and systemic examination combined with the radiologic findings.

In this case report, a CGCG case located in the anterior maxilla revealing a fibromatous and erythematous painless swelling of a 72 – year old female patient with a sialolithiasis as an incidental finding was aimed to be presented by orthopantomagram (OPG), periapical, Cone Beam CT (CBCT) and ultrasonographic (USG) findings.

Keywords: central giant cell granuloma, sialolithiasis, ultasonography, cone beam computer tomography

ÖZ

Santral dev hücreli granuloma (SDHG) etyolojisi belirgin olmayan benign bir intraosseöz tümör olup hızlı büyümeye, ekspansiyona ve/veya kortikal kemikte perforasyona neden olabilir. Ayırıcı tanıları histolojik ve sistemik bulgulara da bağlı olup, radyolojik bulgular ile beraber değerlendirilmelidir.

Bu olgu sunumunda 72 yaşındaki kadın hastanın maksilla anterior bölgesinde bulunan, fibromatöz ve eritematöz bir şişliğe neden olan ağrısız bir SDHG olgusu sunulacaktır. Ayrıca, tesadüfi bulgu olarak sialolith ile karşılaşılmıştır. SDHG ve siyalolit; ortopantomagram (OPG), periapikal radyografi, Konik Işınlı Bilgisayarlı Tomografi ve ultrasonografi ile değerlendirilmiştir.

Anahtar kelimeler: santral dev hücreli granülom, siyalolit, ultrasonografi, konik ışınlı bilgisayarlı tomografi

Dt. Melisa Bozkurt (✉)
Başıbüyük, Başıbüyük yolu, Marmara Üniversitesi Sağlık Yerleşkesi 9/3 Maltepe/ İstanbul
melisabozkurtt@windowslive.com

Fatma Nihan Aksakallı
Assoc. Prof. Dr. Istanbul University, Institute of Oncology, Oral Pathology, Istanbul, Turkey

Nuran Bayramov
Dr.,Dt. Marmara University, Faculty of Dentistry, Oral and Maxillofacial Radiology, Istanbul, Turkey

Baturay Ornağ
Dt. Marmara University, Faculty of Dentistry, Oral and Maxillofacial Surgery, Istanbul, Turkey

Mehmet Oğuz Borahan
Assoc. Prof. Dr. Marmara University, Faculty of Dentistry, Oral and Maxillofacial Radiology, Istanbul, Turkey

Ferit Bayram
Asst. Prof. Dr. Marmara University, Faculty of Dentistry, Oral and Maxillofacial Surgery , Istanbul, Turkey

Şebnem Erçalık Yalçınkaya
Prof. Dr. Marmara University, Faculty of Dentistry, Oral and Maxillofacial Radiology, Istanbul, Turkeyy

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INTRODUCTION

Central giant cell granuloma (CGCG) is a benign intraosseous tumor that has an unclear etiology (Balaji & Balaji, 2019) which was first described in 1953 by Jaffe as reparative giant cell granuloma (Jaffe, 1953). Since 2017, it is subtyped under the giant cell lesions and bone cysts according to the classification of odontogenic lesions by World Health Organization (Soluk-tekkesin & Wright, 2017).

CGCG is mostly represented in adolescents and young adults and tends to occur in anterior region of the jaw bones. Approximately %70 of the cases occur in mandible (Zhang et al., 2019). Orbita, temporal bone, ethmoid bone and vertebrae are reported as the other locations of CGCG cases except jaw bones. Due to the variety of origin cells of head and neck, the characteristics of these pathologies differ within individuals and even locations in the same individual (Lee & Huang, 2020).

Clinically, enlargement of the lesion is fast and the expansion and /or perforation of cortical bone is not a rare finding. Well-defined or infiltrative borders are also the most frequent feature (Lee & Huang, 2020). The covering mucosa may represent a red/purple layer which may be misdiagnosed as a vascular lesion. The aggressiveness of tumor may vary although the discovery of the lesion depends on the symptoms such as swelling, paresthesia, pain, tooth mobility and displacement (Nilesh et al., 2020).

Radiologic findings of CGCG is not specific. CGCG usually presents a circumscribed and multilocular radiolucent lesion with non-corticated and well-defined borders. Soap bubble appearance is not rare if the lesion when multiloculated (White & Pharoah, 2014).

Histopathology of the tumor is comprised of multiple hemorrhagic foci, multinucleated cells and trabecular bone (Kramer et al., 1991, Bocchialini et al., 2019). Brown tumor of hyperparathyroidism, aneurysmal bone cyst, simple bone cyst, cherubism may also show similar histopathological and radiological findings. The formation of the septa makes differential diagnosis complicated (Bocchialini et al., 2019). Brown tumor has prominent compartments and structure with septa formation which is distinct in compartment of CGCG. The histopathological features of giant cell lesions are variable, thus its diagnosis may be a complicated pathway and consideration of other pathologies is inevitable (Chrcanovic et al., 2018, Candeiro et al., 2020).

In this case report, a CGCG case in maxilla anterior region revealing as fibromatous and erythematous painless swelling of a patient with a sialolithiasis as an incidental finding was presented.

CASE REPORT

A 72 – year old female patient referred to outpatient clinic of Department of Oral & Maxillofacial Radiology. The patient reported a painless swelling which has been presented for the last 3 months. Clinical examination revealed fibrous and erythematous tissue on the edentulous anterior maxilla and panoramic and periapical images showed an unilocular radiolucent lesion (Figure 1 & 2). Panoramic radiograph has also revealed a radiographic mass which was preliminary diagnosed as sialolithiasis (Figure 1).

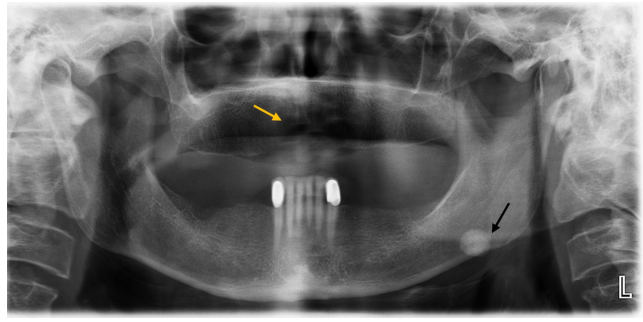

Figure 1. First OPGT of the patient. Osteolytic and unilocular radiolucent lesion (yellow arrow). Note the left unilateral sialolithiasis as an incidental finding (black arrow).

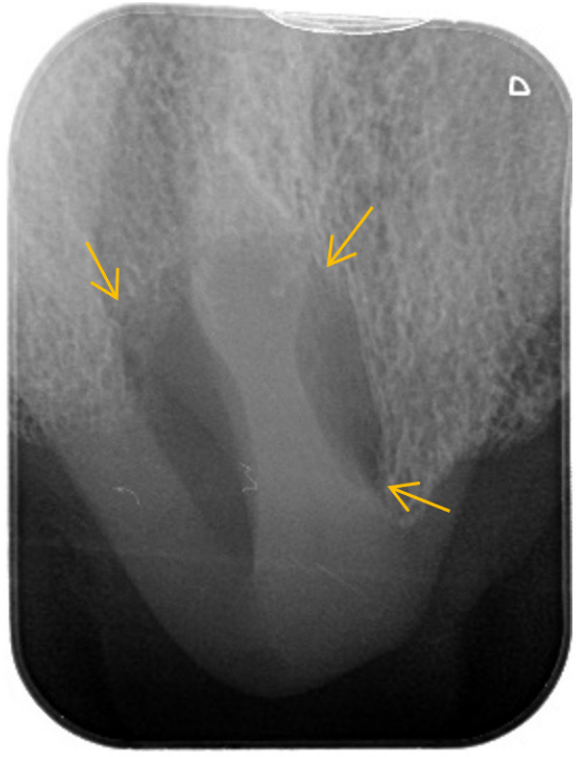

Figure 2. Periapical radiography of the anterior maxilla. Note the non-corticated borders of the radiolucent lesion (yellow arrows)

To further assess the lesions, cone-beam computed tomography (CBCT) was used to examine the anatomic borders and bone destruction. On CBCT; maxillary buccal and palatal cortical bone perforation with the lesion in close relation with the nasopalatine canal cortex was detected (Figure 3 a,b). Additionally, CBCT showed the sialolithiasis (Figure 3 c,d).

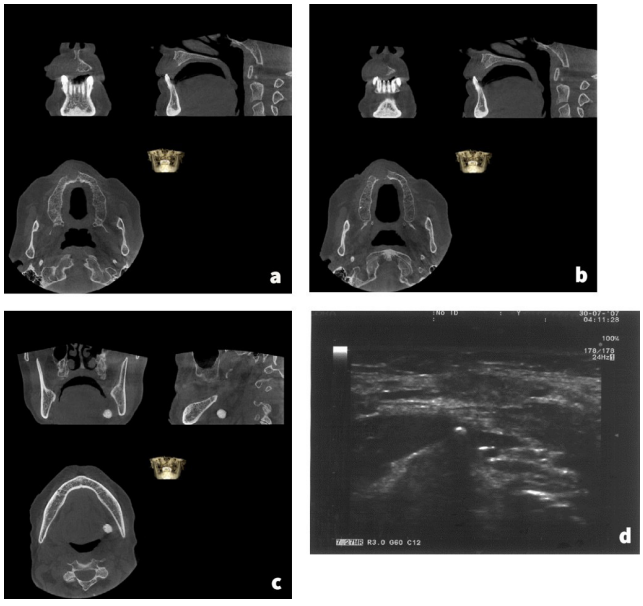

Figure 3. a. CBCT sections of the CGCG. Note the buccal and palatinal bone cortex perforation. Corticated and well-defined borders. **b.** Close relation with the lesion and nasopalatine canal cortex. **c** Unilateral left sialolithiasis on the CBCT sections. **d.** Sialolithiasis on the USG imaging.

To evaluate the vascular features (e.g. hemangioma) of the lesion, ultrasonography (USG) was used. Ultrasonographic findings revealed buccal bone cortex perforation and snowing-like hyperechoic particles inside the lesion. In the inferior line of the lesion, posterior eco has not increased which is an ultrasonographic finding which occurs due to the acoustic competency difference between soft tissue (or liquid) and bone tissue. This lesion was observed as a solid mass instead of a cystic lesion in ultrasonography (Figure 4).

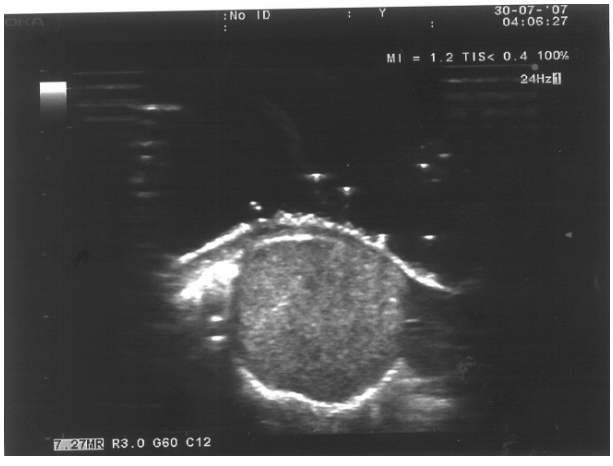

Figure 4. To evaluate the vascular features of the lesion USG was performed. There was no significant vascularity or blood flow. Lesion can be detected with USG imaging as an isoechoic and solid intraosseous mass. Internal structure of the lesion was observed and described as snowing-like.

The lesion was removed surgically at Marmara University, Faculty of Dentistry, Oral & Maxillofacial Surgery Department under local anesthesia. Enucleation and curettage of the lesion was performed with monopolar electrocautery and minimal bleeding was seen in the field. The patient was prescribed anti-inflammatory agents, antibiotics and mouthwash (Figure 5).

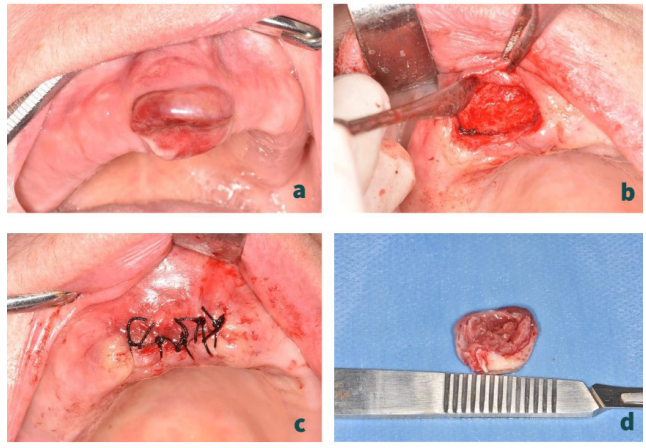

Figure 5. a. Pre-operative intraoral appearance of the lesion. The impression of anterior mandibular tooth as linear impression, in the middle of the tumor. **b.** Intra-operative minimal bleeding in the field. **c.** Post-operative. Bone cavity has filled only with gelatine sponge (absorbable gelatin sponge heamostatic). **d.** The excision biopsy material.

The histopathological findings supported the preliminary diagnosis of CGCG revealing hypertrophic shuttle fibroblasts scattered in the distribution of multinucleated giant cells, clustered around hemorrhagic foci (Figure 6).

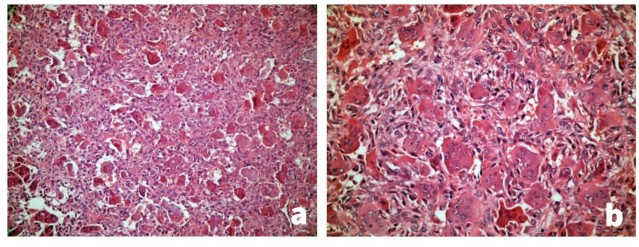

Figure 6. **a.** Histopathological photomicrograph, 200x magnification, 400x magnification, **b.** showing tissue characteristics of central giant cell granuloma.

Since Brown tumor shows the similar histopathological features, blood analysis [serum Ca, ionized Ca, alcalen phosphatase, Vitamin D, kreatinin, thyroid stimulating hormone (TSH), parathyroid hormone (PTH), triiodothyronine (T3) and thyroxine (T4)] has been requested. Results have shown that patient has a normal PTH value and the Brown tumor was eliminated as differential diagnosis.

DISCUSSION

Central giant cell granuloma is a rare intraosseous mass and a non-neoplastic tumor of bones. Location of the mass, age of the patient, systemic condition are clues for referral diagnosis. Histopathologically, genetic disorders such as Cherubism, Noonan syndrome, neurofibromatosis and Brown tumor of hyperparathyroidism have similarity with CGCG which all contain the presence of numerous multinucleated cells in large amounts of loose connective tissue containing innumerable spindle cells, macrophages and blood vessels. Areas of considerable erythrocyte extravasation and hemosiderin deposition are significant features of aforementioned lesions (Nilesh et al., 2020, Kramer et al., 1991, Bocchialini et al., 2019, Chrcanovic et al., 2018, Candeiro et al., 2020).

The presented case was histopathologically diagnosed as CGCG however evaluation for Brown tumor is essential. The %60 of the CGCG cases is usually present before the age of 30 years (Neville et al., 2002). The age of the patient was a rare entity of this case thus systemic conditions should be considered. Ficcara et al. (1987) and Choung et al. (1986) categorized CGCG as aggressive and non-aggressive. Non-aggressive form is more common, grow slowly as a painless swelling; although aggressive form is rare, grow fast and mostly encountered in younger patients. Defining the borders according to this classification, aggressive tumors have ill-defined borders and mostly cause cortical destruction. Radiologic examination of the lesion has significant spots such as non-corticated borders or multilocularity is not invariable as well. Two-Dimensional images are the first-line in the diagnosis of bone-effected lesions. CBCT has advantages as a second-line examination of hard tissues. Ultrasonography was also used in this case due to the vascular features of the tumor, which is a critical benefit for the surgery.

Radiologically, CGCG may also represent a small unilocular radiolucency, which may be confused with periapical granulomas or cysts (Candeiro et al., 2020). In this case report the lesion was radiolucent as expected and unilocular and positioned in edentulous anterior maxilla. Extension of the radiologic examination is necessary if lesion contains both soft tissue and bone. USG and magnetic resonance imaging (MRI) are imaging options for soft tissue maintaining lesions. In this case, the lesion was solid and had well-defined borders with adjacent structures. The USG was preferred as a non-invasive, real-time imaging method to examine soft tissue relations of the lesion. Internal structure of the lesion has been revealed in USG imaging. Vascularization has not been seen and surgeons were informed with both CBCT and USG examination reports (Neville et al., 2002, Ficcara et al., 1987, Choung et al., 1986).

Ultrasonography is an easy-to-use imaging method that does not contain invasive radiation and may be helpful for clinical evaluation of differential diagnosis [Caglayan & Bayraktar, 2018) however USG imaging has limitations such as the size of the lesion (Arslan et al., 2020). Increasing the variety of imaging methods may adjoin multiple findings until the final diagnosis. Using multiple imaging techniques may decrease the risk of misdiagnosing and also the risk of complications during operation. In this case, to measure the size of the lesion, CBCT was used as a highly reliable radiological method.

CONCLUSIONS

USG imaging is an option in order to differentiate cystic lesions from solid lesions in maxillofacial radiology and clinicians should consider using USG for vascular lesions in referral diagnosis and before the surgery. USG is one of the easiest, harmless and low-cost imaging methods to prevent complications. USG may provide important diagnostic information such as the thinning, expansion or perforation of the buccal cortex, convexity or continuousness of the bone cortex and internal structure of the lesions.

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None.

Conflıct of Interest

We have no conflict of interest.

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