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CASE REPORT

Collision Lesion of Mandible—Coexistence of Keratocystic Odontogenic Tumor with Central Giant Cell Granuloma: A Rare Case Report

Spoorthi B Ravi, Prashanthi C, Vinayak Karun, Mahesh Melkundi, Sanjay Nyamati, Annapoorna HB

ABSTRACT

Aim and background: An odontogenic keratocyst (OKC) or keratocystic odontogenic tumor (KCOT) and giant cell granuloma (GCG) in the jaws are common lesions which have been studied extensively in detail over the years. However, a lesion showing features of both is exceptionally rare and is reported only twice in the literature till date.

Case description: A rare case of OKC in mandible showing foci of GCG like areas is reported in a 29 years old male patient.

Conclusion: It seems to be a collision lesion, though the possibility of KCOT showing a reactive response to form giant cells or it being a rare variant cannot be totally ruled out.

Clinical significance: This entity requires aggressive treatment since biological behavior of this unique lesion is difficult to predict unless more of such lesions are reported and followed up in future.

Keywords: Odontogenic keratocyst, Giant cell granuloma, Collision lesion, Giant cells, Osteoclasts.

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INTRODUCTION

Odontogenic keratocyst (OKC) is considered to be the most distinct cyst among all the odontogenic cysts and it demands extra attention both clinically and histopathologically because of its growth pattern, clinical behavior and its rate of recurrence. Various studies have proved beyond doubt about the proliferative nature of the lining epithelium. World Health Organization (WHO) reclassified it as keratocystic odontogenic tumor (KCOT) considering the growth potential of its epithelial lining.[1]

Central giant cell lesions as defined by WHO are intraosseous lesions consisting of cellular fibrous tissue containing multiple foci of hemorrhage, aggregation of multinucleated giant cells, and occasionally trabeculae of woven bone. It mainly occurs in children or young adults with a predilection for females. It is more common in mandible than in maxilla. The radiologic features range from unilocular radiolucency to a multilocular radiolucency with varying degrees of expansion of the cortical plates.[2]

Giant cell granuloma (GCG) is a relatively common jaw lesion of young adults that has an unpredictable behavior. Microscopic diagnosis is relatively straightforward; however, this lesion continues to be somewhat controversial because of its disputed classification (reactive *vs* neoplastic) and because of its management (surgical *vs* medical). Its relationship to giant cell tumor of long bone remains undetermined. It is a lesion chiefly consisting of proliferating fibroblasts with macrophages and numerous multinucleated giant cells. These are admixed with collagen, hemorrhagic areas with dense vasculature.

Both KCOT and GCG even when occur singly are clinically known destructive lesions, whose treatment needs are of paramount importance because of KCOT's peculiar tendency to recur following surgical treatment and presence of numerous osteoclasts like giant cells in GCG, both pointing toward more destructive behavior. Conventionally both KCOT and GCG are managed after thorough assessment of the clinical situation with great care before treatment modality is chosen instead of just enucleation. These lesions are relatively common in their own terms, but a lesion showing features of both is found to be reported only twice in the literature.[3,4] We report third such case of KCOT showing focal areas of GCG like lesion.

CASE REPORT

A 29-year-old male patient visited a private dental clinic in Indore, Madhya Pradesh, India, with a chief complaint of swelling on the left lower jaw region since 6 months, which was gradually increasing in size. Patients past medical, traumatic and family histories were noncontributory. On examination a diffuse solitary swelling was observed on the left posterior mandible extending from 1.5 cm posterior to angle of the mouth to mandibular angle region posteriorly (Fig. 1). The swelling was nontender, skin over the swelling was normal. An obvious facial asymmetry was evident. On intraoral examination a diffuse solitary swelling obliterating the buccal vestibule was noted (Fig. 1). The swelling was extending from 34 to retromolar area. On palpation the swelling was firm to hard in consistency, nontender. Associated teeth were appearing normal showing no signs of any pathology but with 35, 36 and 37 showing grade II mobility. Given these findings, a provisional diagnosis of a dentigerous cyst, OKC or ameloblastoma was made.

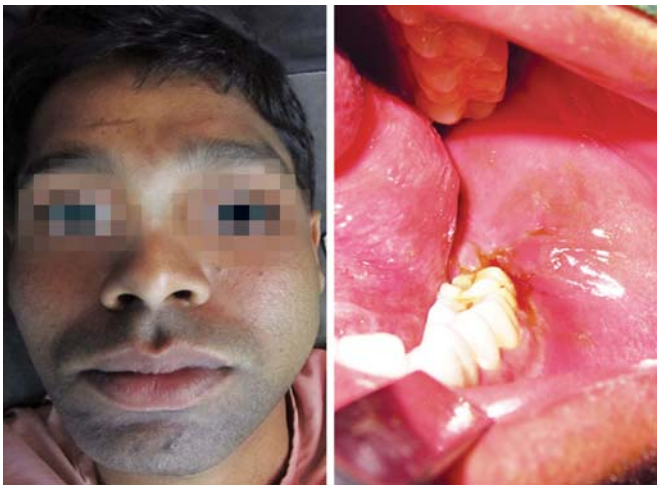

Fig. 1: Extraoral facial asymmetry and intraoral picture showing vestibular obliteration

Orthopantomographic (OPG) examination revealed a well-defined unilocular radiolucency around impacted third molar. The roots of 35, 36 and 37 appeared to be resorbed to about their half length (Fig. 2) computed tomographic (CT) scan revealed a buccal cortical plate expansion (Fig. 2). The lesion was surgically excised and was submitted to Department of Oral pathology, College of Dental Science and Hospital, Indore, for histopathological examination. The specimen was processed routinely and 4 µm sections were cut and stained with hematoxylin and eosin (H&E).

The microscopic sections in majority of areas revealed a typical KCOT lining epithelium of uniform 6 to 8 cell thickness of stratified squamous parakeratinized epithelium with palisaded basal cells and hyperchromatic nuclei with surface corrugations (Fig. 3). Focal areas of lining epithelium also showed plaque or swirl like arrangement with clear cells. The connective tissue epithelial interface was flat with no rete pegs. The cystic capsule showed diffuse chronic inflammatory infiltrate. In many focal areas of the sections, the connective tissue was more cellular, more vascular along with numerous multinucleated giant cells resembling a GCG (Fig. 4). Numerous trabeculae of woven bone were also present within the hypercellular areas.

DISCUSSION

A rare case of KCOT showing focal areas of CGG features is presented here. The most striking feature here is the presence of numerous multinucleated giant cells in the connective tissue capsule. After its first report by Yoon et al in 2004,[3] only single attempt has been made to explain the pathogenesis of multinucleated giant cell component in KCOT.[4] The paucity of such reported cases only adds to the confusion regarding the presence of such giant cells in KCOT. Even though many odontogenic tumors like ameloblastoma, odontogenic fibroma are known to occur

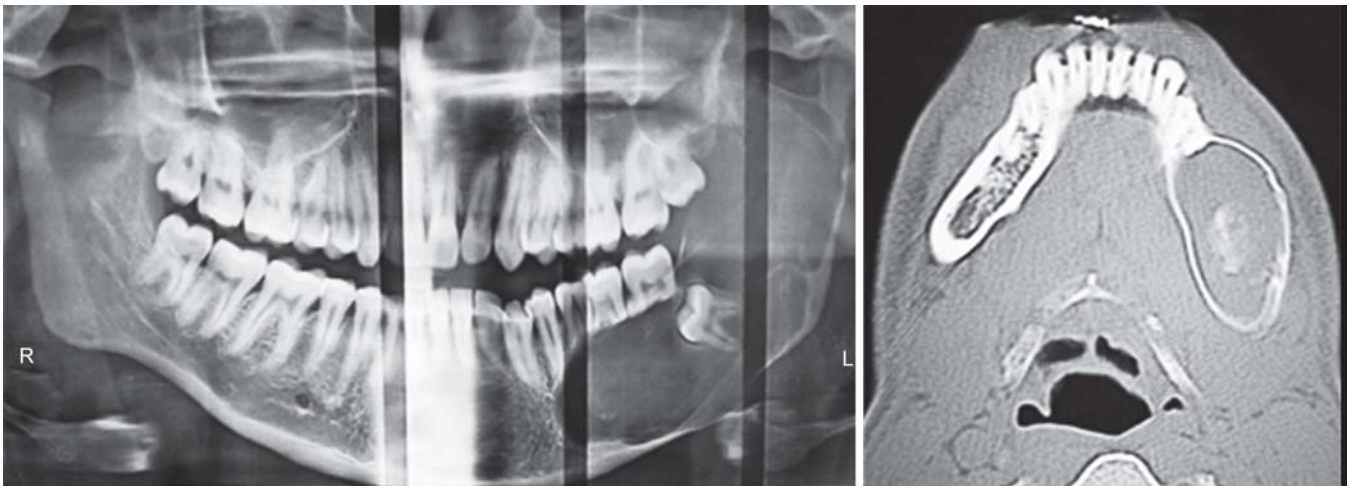

Fig. 2: OPG revealing a unilocular radiolucency with impacted 38 with the root resorption irt 35, 36, 37.
Note: The CT scan revealing buccal cortical plate expansion

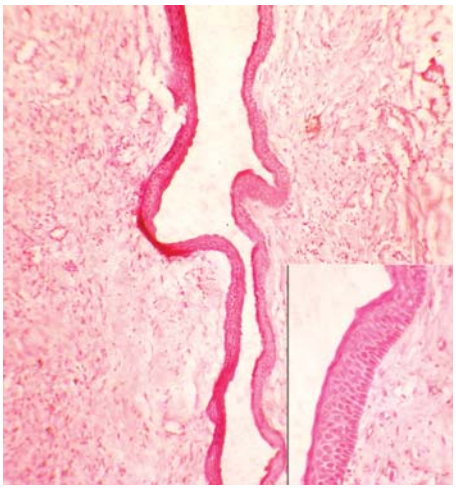

Fig. 3: Section showing typical KCOT features. H&E stain; 5× magnification (inset: 10× magnification)

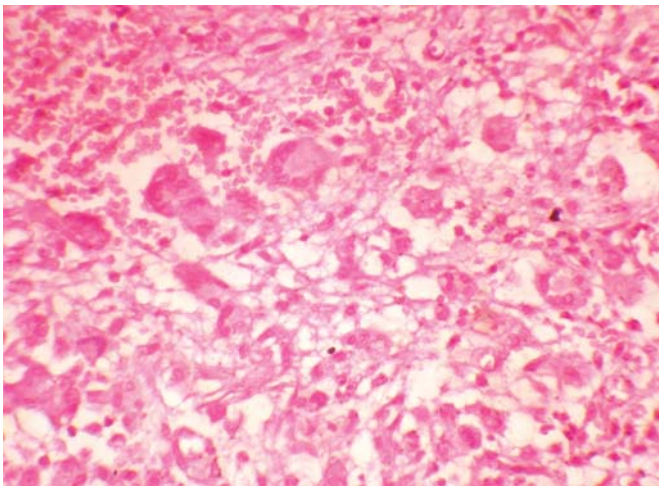

Fig. 4: Section showing numerous multinucleated giant cells with horse shoe pattern arrangement of nuclei (H&E stain; 10× magnification)

along with a giant cell reactive response[5] with an unclear initiating stimulus, the same with odontogenic cysts is a rare phenomenon.

Although there are several case reports of KCOT associated with traumatic bone cyst,[6] ameloblastoma,[7] and KCOT containing cartilage or KCOT associated with intramandibular chondroma,[8] there are only two cases of KCOT with GCG features reported till date in literature,[3,4] likewise GCG is reported to occur with aneurysmal bone cyst, ossifying fibroma, osteoblastoma, chondroblastoma and odontogenic fibroma,[9] but not very commonly with KCOT. Hence, our report stands distinct from others.

Microscopically, the present lesion was unusual by demonstrating bimorphic giant cells. One population of giant cells showed their nuclei arranged in horse shoe pattern as in Langhans giant cells (Fig. 4) and other population with a central arrangement (Fig. 5). The presence of Langhans type of giant cells can be attributed to local production of tumor necrosis factor (TNF) alpha,[10] interleukin (IL)-3[11] which can lead to production of these type of giant cells, and the second type of giant cells seems to be of osteoclast type which appeared larger than the Langhans type giant cells and had more number of nuclei than the other type. These giant cells amidst highly cellular and vascular areas can be attributed to be derived from macrophages.[12]

Surprisingly, the lining epithelium apart from exhibiting typical KCOT features in majority of sections, also demonstrated few clear appearing cells (Fig. 6) with focal areas demonstrating plaque like epithelial thickenings as in a lateral periodontal cyst (LPC) (Fig. 7). Because of the scantiness of such areas, the significance of it is difficult to explain. However, it has been previously suggested that epithelial inclusions in the region of the mandibular third molars might represent pluripotentiality.[13] In view of the multipotentiality of the odontogenic epithelium around the mandibular third molars; it is possible that it would have

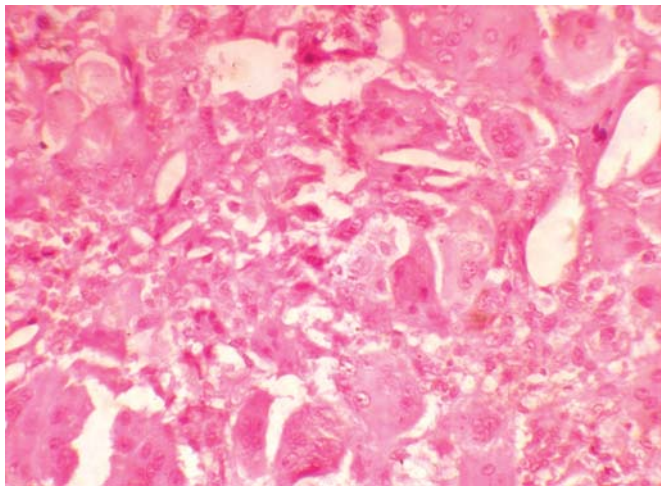

Fig. 5: Section showing many multinucleated giant cells with central arrangement of nuclei (H&E stain; 10× magnification)

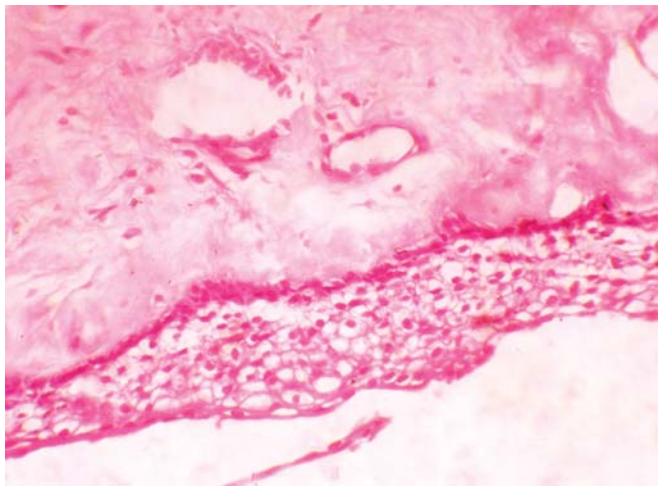

Fig. 6: Section showing numerous clear appearing cells in the lining (H&E stain; 10× magnification)

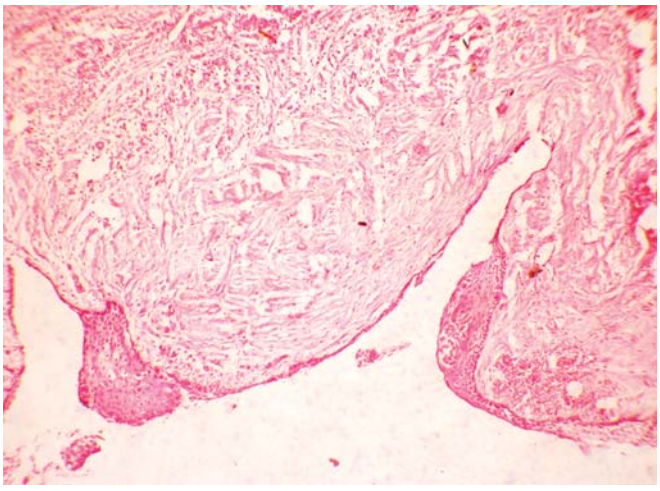

Fig. 7: Section showing few epithelial plaque-like or swirl-like lining, resembling LPC lining (H&E stain; 4× magnification)

the capacity of inducing the formation of cysts with both the KCOT and LPC like lining epithelia. However, according to Shear, if a cyst demonstrates both features of KCOT and LPC, the diagnosis of KCOT should be considered.[14] Based upon the histological findings this peculiar case was regarded as KCOT with GCG. However, the giant cell component was found more in the capsule of LPC areas than the KCOT areas.

Therefore, the pivotal point now is why giant cells, excess cellularity and vascularity in the capsule? Is it a rare variant of KCOT or a case of two separate lesions colliding? (Fig. 8).

Ronnel et al[15] in their explanation of giant cells in odontogenic tumors have put forth three hypotheses.

- Collision tumor: Two lesions are present in a synchronous way, mixed/not mixed/both or just one of them is extremely small
- GCG occur as a reactive response due to the presence of inactive odontogenic epithelium

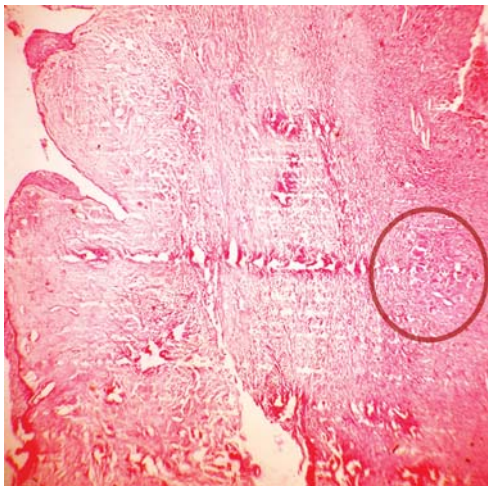

Fig. 8: A section showing collision of cystic lesion with giant cell granuloma. A circle inside represents collection of giant cells with adjoining hemorrhagic areas (H&E stain; 4× magnification)

- Growth factors are produced by a primary GCG lesion that produces the proliferation of odontogenic epithelium.

The present case seems to be due to collision, since the GCG component is not very small or not seen in very small focal areas to represent a reactive response, also it is a known fact that epithelial component of KCOT is not inactive for GCG to occur as a reactive response. Also, root resorption is unusual in KCOT, though it is reported to occur in only 4.4% in Browne's study, 5% cases in Haring's study, 7.5% in Khan MT's study and 11% cases in Maxine partridge's study.[16] However, in the present case, OPG showed severe root resorption of 34, 35, 36 and 37 to about half of their root length. But GCG is reported to be aggressive with root resorption being a common feature.[17] So based on histological, radiological, intramedullary growth pattern with cortical expansion observed, it can be concluded that it is a case of collision lesion, though a reactive response of KCOT to form giant cells or it being a rare variant of KCOT cannot be totally ruled out.

CONCLUSION

The clinical behavior of this collision lesion is a daunting task to predict, as both KCOT and GCG are aggressive clinically and hence demands aggressive treatment, in addition only two such cases are reported in the literature to satisfactorily know their nature. What is clear is that multimodal surgical approach is required to address such cases. So pathologists should be aware of this possibility of collision and report more, because there is still a lot to know about the clinical course and treatment outcome of this unique, rare collision lesion.

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ABOUT THE AUTHORS

Spoorthi B Ravi (Corresponding Author)

Reader, Department of Oral Pathology, MS Ramaiah Dental College and Hospital, Bengaluru, Karnataka, India, Phone: 9742980076 e-mail: drspoorti@gmail.com

Prashanthi C

Senior Lecturer, Department of Oral Medicine and Radiology, MS Ramaiah Dental College and Hospital, Bengaluru, Karnataka, India

Vinayak Karun

Reader, Department of Oral Surgery, RKDF Dental College, Bhopal Madhya Pradesh, India

Mahesh Melkundi

Senior Lecturer, Department of Oral Pathology, College of Dental Science and Hospital, Indore, Madhya Pradesh, India

Sanjay Nyamati

Professor, Department of Oral Medicine, Triveni Institute of Dental Sciences, Bilaspur, Chhattisgarh, India

Annapoorna HB

Reader, Department of Community Dentistry, Triveni Institute of Dental Sciences, Bilaspur, Chhattisgarh, India