Glandular Odontogenic Cyst of Mandible: A Case Report

R Krishna Kumar¹, Rafiya Mushtaq², Sana Fatema³, Chirag Mukhi², Farhana Ashraf⁴
¹Professor & HOD, Department of Oral Medicine & Radiology, M.A. Rangoonwala Dental College, Pune, Maharashtra, India, ²PG Student, Department of Oral Medicine & Radiology, M.A. Rangoonwala Dental College, Pune, Maharashtra, India, ³Senior Lecturer, Department of Oral Medicine & Radiology, M.A. Rangoonwala Dental College, Pune, Maharashtra, India, ⁴PG Student, Department of Oral Pathology and Microbiology, M.A. Rangoonwala Dental College, Pune, Maharashtra, India

Glandular odontogenic cysts (GOCs) are a very rare developmental cyst of the jaws. GOCs are intrabony solitary or multiloculated cyst of odontogenic origin. The importance of GOCs lies in fact that they exhibit a high propensity for recurrence. Thus, the oral physicians, radiologist, and oral pathologist play a major role in the definitive diagnosis of GOCs based on distinctive cases; though they are rare. In large part, this is due to the GOCs complex and frequently non-specific histopathology. This report describes a case of GOC occurrence in posterior mandibular angle region in a 43-year-old Irani Male, which was a rare site for its occurrence.

Keywords: Glandular odontogenic cyst, Jaw cyst, Odontogenic cyst

INTRODUCTION

Glandular odontogenic cyst (GOC) is a rare developmental cyst of the jaws. It is also considered to be a variant of the lateral periodontal cyst. This cyst has been included in the World Health Organization histological typing of odontogenic tumors under the terms GOC or sialo-odontogenic cyst. This cyst was described as an entity by Gardner et al. in 1988.¹ Only 50 cases have been reported in the literature until date. Overall incidence of the cyst has been estimated as 0.012% of the total cyst.²

Although it is almost rare, this cyst is a well-known clinical entity and is important to recognize and diagnose due to its aggressive behavior and tendency of recurrence. This cyst reports a slightly high male predilection with a male: female ratio of 28:19,³ and commonly affected site is anterior mandible. Middle aged people are more often affected, average age of occurrence is 46.7 years in males and 50.0 years in females. Mandible is most commonly affected (87.2%) than maxilla.⁴

Radiographic examination features are the well-defined cyst which may be unilocular or multilocular in appearance, often with scalloped margins and sclerotic borders. GOCs usually seen apical to the teeth and can project into the interdental bone. In addition, there may be root resorption and tooth displacement with cortical perforation, leading to extension of cyst into adjacent soft tissues.⁵

Histopathologically, GOCs peculiar features show that the morphology of the epithelium strongly suggesting an origin from remains of dental lamina. Besides, this presence of intraepithelial mucous containing cystic spaces are also appreciated. Characteristic feature of GOCs is cystic wall lining of non-keratinized epithelium with papillary projections, nodular thickening, and mucous filled lakes.⁶,⁷ In addition to, these features it also includes cuboidal basal cells which are sometimes vacuolated.

GOCs treatment mostly includes enucleation and curettage, although some authors say local block excision or marginal resection to be more reliable modality due to high tendency of cyst to recur.⁷

The objective to present this case report is to present a case of GOC in an Irani male in the posterior mandibular region which was very unusual and rare.

CASE REPORT

A 43-year-old Irani man reported to the Department of Oral Medicine and Radiology with a complaint of mobility
in his lower anterior teeth. His past medical history was non-contributory.

An intraoral examination revealed generalized moderate attrition of the teeth. Grade II mobility was noted in 31 and 41 regions. Prosthesis was noted in the lower left region of the jaw. No extraoral or intraoral swelling was seen. Overlying gingiva and oral mucosa were normal. On palpation, no bony expansion was noted anywhere. There was no discharge of pus and sinuses. No significant extraoral and intraoral findings were noted in the left mandibular posterior region on the examination were noted (Figures 1a and b).

An intraoral periapical radiograph was advised in the region of 32-42, which revealed severe horizontal bone loss from 32 to 42 regions. The patient was referred to the Department of Periodontology to address mobility in lower anterior teeth (Figure 3).

The patient was again referred to the Department of Maxillofacial Radiology for an Orthopantomogram (OPG) to assess periodontal condition, and a panoramic radiograph was obtained using Carestream CS9300 machine which revealed an accidental finding in the lower right region of the mandible.

The panoramic radiograph revealed 38 impaction associated with a unilocular well-defined radiolucency in the left mandibular angle area which extended 2 cm below apices of 38 and just above inferior border of mandible involving the inferior alveolar canal extending up to the superior border of mandible and antero posteriorly from the distal root of 37 up to ramus of mandible. The inferior border of the mandible was well intact; approximate size was 2 cm × 2 cm with the involvement of neurovascular bundle. The impacted tooth was the epicenter of the radiolucency. There was no expansion of posterior or inferior border of the angle of mandible (Figure 4).

Crown of the malposed, unerupted third molar 38 was displaced to the lower border of the mandible. Under local anesthesia fine-needle aspiration cytology (FNAC) was done, which yielded a colorless, low viscosity fluid. FNAC report revealed desquamated epithelial cells and an inflammatory component.

Incisional biopsy was done which revealed a cystic lumen lined by epithelial lining and connective tissue capsule. The epithelial lining was 2-3 layers in thickness resembling reduced enamel epithelium. The connective tissue wall comprised of fibroblasts, collagen fibers, few blood vessels which were suggestive of a dentigerous cyst.

Based on clinical and radiographic findings, a provisional diagnosis of the dentigerous cyst was given due to

Figure 1: (a and b) Frontal and lateral profile view of patient no significant evidence of extraoral swelling

Figure 2: Intraoral picture showing no much evidence of intraoral swelling in lower left back tooth region

Figure 3: Intraoral periapical revealing poorly endodontically treated 37 with prosthesis and pericoronal radiolucency with impacted 38

Figure 4: Orthopantomogram showing unilocular radiolucency associated with impacted 38
its association with impacted teeth. The differential diagnosis given was OKC, lateral periodontal cyst, cystic ameloblastoma.

Following these investigations, surgical enucleation and curettage of the cystic cavity along with the removal of impacted third molar was done under general anesthesia in the Department of Oral and Maxillofacial Surgery and the specimen was sent for final histopathological confirmation.

It showed the presence of cystic lumen, epithelial lining, and connective tissue capsule. The epithelial lining was non-keratinized stratified squamous in nature and exhibited variable thickness. At few places, it was thinner (2-3 cell thick) resembling reduced enamel epithelium. The surface cell layer at most of the places was cuboidal in nature showing the presence of “hobnail cells.” Few areas showed epithelial proliferation in the form of plaques. Occasionally micro cysts filled with eosinophilic material was noted. Confirming overall findings were suggestive of “GOC” (Figures 5 and 6).

**DISCUSSION**

A case of GOCs is very rare and unusual of odontogenic developmental origin which is presented herein. As stated by most of the authors our case also demonstrated significantly mandibular involvement. The histopathology features were also suggestive of a cystic cavity lined with pseudo-stratified, ciliated columnar epithelial lining, and fibrous vascular connective tissue. The literature showed anterior mandible as most commonly affected site, whereas in our present case posterior mandible and angle region was involved.

GOCs recognition based on clinical and radiological examination is practically impossible and misleading, as noted in our case where our provisional diagnosis given was dentigerous cyst due to its association with impacted tooth. Only histopathological examination of the specimen in toto proved the diagnosis of GOC.

The GOCs typically does not show any peculiar characteristics on radiographs and resemble quite similar to the dentigerous cyst, OKC, radicular cyst, and botryoid cyst if radiolucency is unilocular; multilocular radioluencies are suggestive of ameloblastoma, myxoma, and CGCG. Histopathologically, it shows characteristic appearance similar to muco-epidermoid carcinoma and thus should be distinguished.

Our case reported was considered to be GOC because it fulfilled criteria specified by Gardner et al., and unlike muco-epidermoid carcinoma, cellular atypia, and solid and microcystic epithelial proliferation were not seen.

Most authors prefer marginal and segmental resection as the modality to treat GOC due to its high rate of recurrence while some cases have been reported to be managed with conservative approach (enucleation, marsupialization, curettage with peripheral osteotomy, and curettage with adjuvant carney’s solution to destroy satellite cysts or cryotherapy).

As our provisional diagnosis based on OPG, FNAC and incisional biopsy moved toward simple dentigerous cyst which was later found to be GOC only on histopathology confirmation on complete excision. So, clinically and radiographically diagnosis of GOC is always ambiguous. Hence, strict histopathology test has to be made to conclude as GOC.

**CONCLUSION**

GOC is rare and aggressive lesion with a high recurrence rate. Careful clinical and radiological evaluation must be
carried out. Computed tomography (CT) or cone beam CT scans are recommended (which were not carried in our case due to unavailability of resources) as they provide accurate information about locularity of the lesion, cortical integrity, expansion of lesion, and involvement of contiguous soft tissue. Besides, this precise histopathological evaluation also should be carried out.

REFERENCES


How to cite this article: Kumar RK, Mushtaq R, Fatema S, Mukhi C, Ashraf F. Glandular Odontogenic Cyst of Mandible: A Case Report. IJSS Case Reports & Reviews 2015;2(8):24-27.

Source of Support: Nil, Conflict of Interest: None declared.