

Glandular Odontogenic Cyst: Report of Two Cases

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Abstract

Introduction: Glandular odontogenic cysts are rare cysts of odontogenic origin affecting the jaw bones. They show unpredictable, aggressive behavior with the potential to grow to a larger size, and recur. Lack of specific clinical and radiographic features makes its diagnosis challenging. They show male predilection and commonly affects mandible as a slow, painless growing mass. Radiographically they may present as unilocular or multilocular lesions with sclerotic borders.

Case Presentation: This article presents two cases of glandular odontogenic cyst treated by surgical enucleation under local anesthesia. Both cases were followed up for 2 years which revealed no evidence of any recurrence.

Conclusion: Histopathological examination of total excised specimen is required for diagnosis. The treatment of choice is usually conservative. However due to its high recurrence rate, resection is also defined in literature as a treatment modality.

Keywords: Glandular Odontogenic Cyst; Enucleation; Mandible

Introduction

Glandular odontogenic cysts are rare cysts of odontogenic origin arising on jaw bones [2]. They are common in males and have predilection for mandible [1]. They show unpredictable, potentially aggressive behaviour with tendency to grow to a larger size and recur [3]. Usually they are asymptomatic, only few cases have presented with pain and paresthesia [4]. Treatment of choice is usually conservative [5]. Enucleation is preferred however because of its high recurrence rate, adjunctive procedures are also used [4]. In this article, we report two cases of glandular odontogenic cyst in mandible treated with surgical enucleation under local anesthesia.

Case Report 1

An otherwise healthy, 60 year-old male patient reported to our department of Oral and maxillofacial surgery with a chief complaint of swelling in lower left front tooth region since 2 years. Initially the swelling was small of a peanut size, which has gradually increased in size to reach the present state. There is no history of any pain, paresthesia, fever or pus discharge.

On intraoral examination, swelling is noted with respect to mandibular left posterior region extending from mesial of mandibular left canine to mesial of mandibular left first molar. Vestibular obliteration is noted. The overlying mucosa appears normal in colour and texture. On palpation, it was firm in consistency, non tender and non fluctuant.

Orthopantomogram reveals a well defined unilocular radiolucency with non scalloped, corticated border seen in left mandibular para-symphysis region extending from distal of mandibular left canine to mesial of mandibular first permanent molar.

FNAC was done. Straw coloured fluid was aspirated.

The lesion was provisionally diagnosed as dentigerous cyst and was treated by surgical enucleation under local anesthesia (Figure 1). Crevicular incision was given extending from mesial of left mandibular lateral incisor (32) to distal of left mandibular first molar (36). A bony window is created and cyst exposed. Following which the cyst is enucleated and removed in toto (Figure 2). Irrigation is done with betadine and saline following which the closure was done using absorbable sutures. Postoperatively the patient was given oral antibiotics and analgesics.

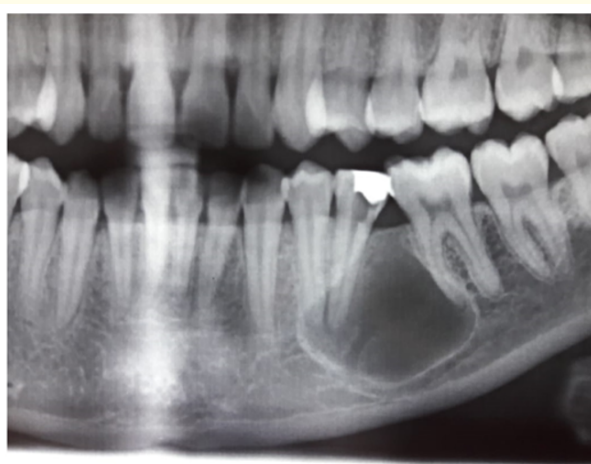


Figure 1: Case 1: OPG.



Figure 2: Case 1: Intraoperative photograph.

Clinical and radiographic follow up was done at 1 week, 1 month, 6 months, 1 year and 2 year. The healing was satisfactory and there was no evidence of any recurrence.

Case Report 2

A 39-year-old male patient reported to our department of Oral and maxillofacial surgery with a chief complaint of swelling in lower right front tooth region since 4 and half years. Initially the swelling was small of a peanut size, which has gradually increased in size to reach the present state. There is no history of any pain, paresthesia, fever or pus discharge. Medical history was not significant.

On intraoral examination, swelling is noted with respect to mandibular right posterior region extending from mesial of mandibular lateral incisor to distal of mandibular right first molar. The overlying mucosa appears normal in colour and texture. On palpation, the inspector findings were confirmed. It was non-tender and hard in consistency.

Orthopantomogram reveals a well defined unilocular radiolucency with non scalloped, corticated border seen in right mandibular parasymphysis region extending from mesial of mandibular right lateral incisor to distal of mandibular right first permanent molar.

FNAC was done. Straw coloured fluid was aspirated.

The lesion was provisionally diagnosed as radicular cyst and was treated by surgical enucleation under local anesthesia (Figure 3). Vestibular incision was given extending from mesial of right mandibular lateral incisor (42) to distal of right mandibular first molar (46). Cyst exposed by making a bony window using hand piece and bur. Following which the cyst is enucleated and removed in toto (Figure 4). Irrigation is done with betadine and saline following which the closure was done using absorbable sutures. Postoperatively the patient was given oral antibiotics and analgesics.

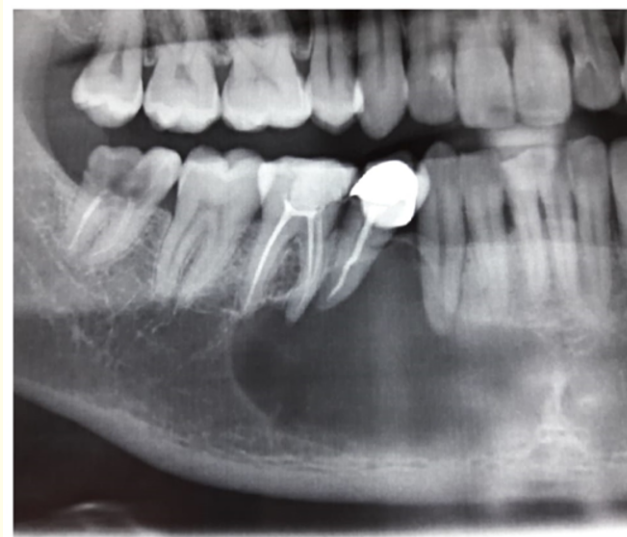


Figure 3: Case 2: OPG.



Figure 4: Case 2: Intraoperative photograph.

Clinical and radiographic follow up was done at 1 week, 1 month, 6 months, 1 year and 2 year. The healing was satisfactory and there was no evidence of any recurrence.

Histopathology report

The H & E stained sections shows non keratinized stratified squamous epithelial lining of variable thickness. The lining epithelium shows superficial cells which are cuboidal to columnar and small cyst like spaces lined by cuboidal cells are seen in focal areas along with clear cells and mucous cells. The underlying connective tissue capsule is densely collagenous with mild chronic inflammatory cell infiltrate and vascularity. Numerous cholesterol clefts and few multinucleated giant cells are also evident. Overall features are suggestive of glandular odontogenic cyst.

Discussion

The first cases of glandular odontogenic cyst were reported by Padayachee and Van Wyk in 1987 which was then known as sialo-odontogenic cyst [1]. The name glandular Odontogenic cyst was proposed by Gardner, *et al.* in 1987 which was further adopted by WHO in 1992 [7].

WHO defined glandular odontogenic cyst as “a cyst arising in the tooth-bearing areas of the jaws characterized by an epithelial lining with cuboidal or columnar cells both at the surface and lining crypts or cyst-like spaces within the thickness of the epithelium” [2].

They are uncommon jaw cysts and the incidence was about 0.2% of all odontogenic cysts [8,9]. They commonly present as slow growing asymptomatic mass in mandibular anterior region [5]. They are common in middle aged group and have male predilection [4].

On radiograph, they may present as a unilocular or multilocular radiolucency, with or without root resorption, the borders may or may not be scalloped and may be associated with an impacted teeth which make the diagnosis more challenging [5]. Also, cortical perforation,

displacement and resorption of root are mentioned in literature [10]. Hence the differential diagnosis include dentigerous cyst, radicular cyst, lateral periodontal cyst, botryoid odontogenic cyst, ameloblastoma and low grade mucoepidermoid carcinoma [2,4]. In most of the cases, they present as unilocular lesion with sclerotic borders often causing cortical perforation [2,5,8].

However, on reviewing the literature most of the cases present like a water pot in OPG. Both our cases are giving similar presentation.

Low viscosity clear fluid on aspiration may be a pathognomonic feature. However secondary infection and repeated surgical procedures may alter this [2].

Definitive diagnosis with small biopsies are difficult because of its similarity to other benign and malignant cyst and tumors of jaw [8,10]. Histopathological examination of complete excised specimen helps in establishing definitive diagnosis [9].

Kaplan, *et al.* suggested that at least the focal presence of each of the major criteria is mandatory for its diagnosis. However, the presence of minor criteria may support the same [8].

Major criteria:

- A lining of Non-keratinized squamous epithelial with a flat interface
- "Spherules"/knobs or "whorls" or focal luminal proliferations
- Surface cuboidal eosinophilic cells or "hob-nail" cells in the epithelial lining
- Mucous cells with or without crypts lined by mucous producing cells with intraepithelial mucous pools
- Intraepithelial glandular microcystic or pseudoglandular duct like structures.

Minor criteria include:

- Papillary proliferation
- Cells with cilia
- Multicystic architecture
- Clear cells in basal or spinous layer.

Establishing an initial provisional diagnosis of glandular odontogenic cyst is very difficult because of its similarity to other odontogenic cysts. Treatment of choice still remains controversial owing to its high recurrence rate (21.6%) and difficulty in diagnosis [2,6]. However, enucleation is the preferred method [4]. Literature have also defined marginal and segmental resections done for treatment of GOC owing to its high recurrence rate and aggressive behaviour [5,6].

Conclusion

Glandular odontogenic cyst can also be included under the category of ABONS (Aggressive benign odontogenic neoplasms). The diagnostic difficulty owing to its clinical, radiographical and histopathological similarity with lateral periodontal cyst, dentigerous cyst, radicular cyst, botryoid odontogenic cyst etc, its aggressive behaviour and high recurrence rate makes glandular odontogenic cyst a plight for the surgeons. Thorough histopathological evaluation of the completely excised specimen is required to establish proper diagnosis, and further patient should be kept on strict follow up to rule out and treat recurrence at an initial stage to prevent severe deformities.

Ethical Approval

Not applicable.

Conflicts of Interest/Competing Interests

Not applicable.

Availability of Data and Material

Not applicable.

Code Availability

Not applicable.

Consent to Participate

Not applicable.

Consent for Publication

Not applicable.

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