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Glandular Odontogenic Cyst - A Case Report and Review of Literature

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ABSTRACT

WHO (1992) Classified GOC as a developmental odontogenic cyst. Till now only 200 cases reported in publications and among them less than 30 cases reported in India. This cyst is least known in reference of its clinical behaviours & radiographical findings. Most of the cases reported are in anterior mandible of middle-aged group. Most of the cases include asymptomatic swelling in anterior mandibular region. Radiographic characteristics are non-specific and ranges from unilocular to multilocular hypodense areas. These lesions have characteristic histopathological features which helps in the diagnosis. This paper reports a case of GOC in anterior mandibular region with unusual clinical and radiographical presentation.

Keywords: glandular odontogenic cyst, GOC, developmental cyst

INTRODUCTION

A glandular odontogenic cyst (GOC) is a developmental cyst that is a clinically rare and histopathologically unusual type of odontogenic cyst. It was first discussed at the meeting of the International Association of Oral Pathologists in 1984 but was first documented as a "solo-odontogenic cyst" by Padayachee and VanWyk in 1987.[1] Histologically, in 1988, Gardner et al. found that the epithelial lining of this cyst was odontogenic in nature and gave the term "glandular odontogenic cyst," which was later adopted by the World Health Organisation. A glandular odontogenic cyst is a clinically aggressive lesion with a high recurrence rate, similar to an odontogenic keratocyst. [2]

Clinically, the most common site of occurrence is the mandibular anterior region, presenting as an asymptomatic, slow-growing swelling. GOC occurs mostly in middle age and has a slight male predilection. [3] Radiologically, these cysts may be unilocular or multilocular with a well-defined border. Histologically, GOC is characterized by a cyst wall lining of non-keratinized epithelium with papillary projections, nodular thickenings, and mucous-filled clefts known as ‘mucous lakes '. It also includes cuboidal basal cells, sometimes vacuolated. [4]

We are presenting a case report of GOC in a middle-aged male whose diagnosis was obtained after the complete excision of the lesion.

CASE PRESENTATION

A 50-year-old male patient came to the oral and maxillofacial surgery department of Vyas Dental College and Hospital. Jodhpur, Rajasthan, with a chief complaint of pain and sensitivity in his lower left front teeth region for 1 week. He also complained of a loss of sensation on the left side of his lower lip. There was no prior medical or family history. An extraoral examination showed that the patient had mild swelling in the left lower border (anterior) of the mandible.

On intraoral examination, there was generalized attrition of the lower teeth. No other significant findings were identified (Fig.1). Orthopantomogram (OPG) and cone-beam computed tomography (CBCT) was advised for further evaluation.

Pic.1- Pre operative intraoral photograph

An orthopantomogram revealed a well-defined multilocular radiolucent lesion involving a 35- to 43-tooth region (Fig.2). CBCT results show the extensions as an anteroposterior apical region of 42 up to a disto-apical region of 35. Superior-inferiorly involving the alveolar crest up to the basal bone, measuring 23.6 mm in the greatest dimension in the anterior region Bucco-lingually measuring about 8.2 to 10 mm in its greatest dimension, it has a well-corticated margin. Effects on surrounding structures include the thinning and resorption of buccal and lingual cortices seen with mild expansion. Knife-edge root resorption in 41, 31, 32, 33, 34, and 35 teeth Loss of cortical borders in focal areas of the superior border of the left inferior alveolar canal (Fig. 3). Radiographic findings were suggestive of a radiolucent osteolytic lesion, likely an ameloblastoma, an odontogenic keratocyst, or a central giant cell granuloma.

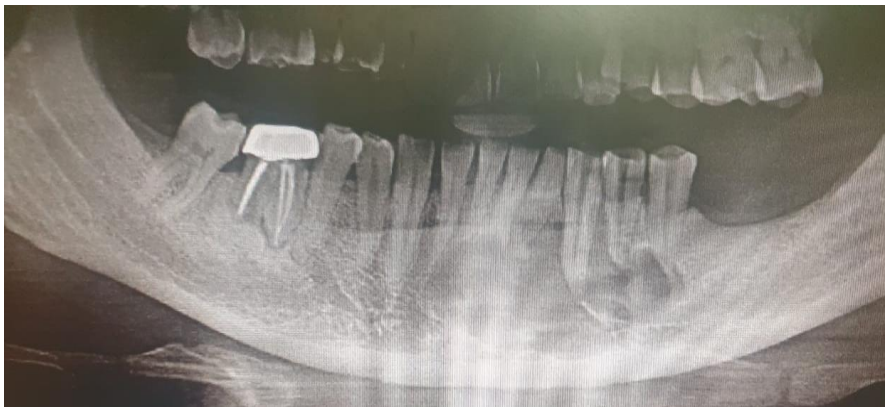

Fig.2- Orthopantomogram revealed a well-defined multi locular radiolucent lesion involving 35 to 43 teeth region.

Fig. 3 CBCT results shows the extensions as antero-posteriorly apical region of 42 up to disto-apical region of 35 region. Superior-inferiorly involving alveolar crest up to the basal bone measuring 23.6 mm in greatest dimension in anterior region Bucco-lingualy measuring about 8.2 to 10 mm in greatest dimension shape well corticated margin. Effect on surrounding structures are thinning and resorption of buccal and lingual cortices seen with mild expansion. Knife edge root resorption in 41, 31 , 32, 33, 34, 35 teeth. Loss of cortical borders in focal areas of superior Border of left inferior alveolar canal.

Complete surgical removal of the lesion under general anesthesia was planned, and consent was obtained from the patient for the same. During the surgery, the lesion seemed to be an odontogenic keratocyst, so enucleation was done, followed by Carnoy solution application as a complementary treatment to give superficial necrosis to reduce recurrence. The samples were sent for biopsies.

Microscopic examination revealed a cystic lumen lined by odontogenic, non-keratinized epithelium with variable thickness. Intra-epithelial micro cysts and clear cells are present. In a few areas, hobnail cells are observed. The connective tissue capsule shows dense infiltration of chronic inflammatory cells and reactive bone trabeculae. Histopathological features are suggestive of a glandular odontogenic cyst (Fig. 4).

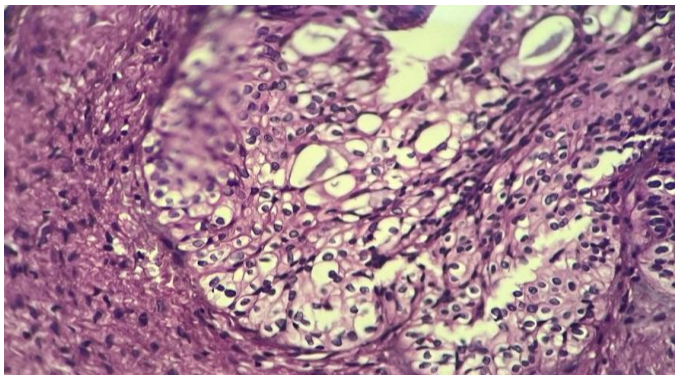

Fig. 4 Microscopic examination revealed cystic lumen lined by odontogenic non keratinized epithelium with variable thickness. Intra-epithelial micro cysts along with clear cells are present. In few areas hobnail cells are observed. Connective tissue capsule shows dense infiltration of chronic inflammatory cells and reactive bone trabeculae. Histopathological features are suggestive of Glandular Odontogenic Cyst.

DISCUSSION

The GOC is a rare but well-established odontogenic developmental cyst and in 1992 it is included in the list of WHO. This cyst is also known as a Sialo-Odontogenic cyst and histopathologically it has a resemblance with Intra-osseous Muco-Epidermoid carcinoma so it is also known as Muco-Epidermoid odontogenic cyst.[2, 5]

The mean age of occurrence is in the fourth to fifth decade of life and few cases are also reported in the second decade of life. This cyst is more common in the male population compared to the female population. Few studies reported a 1.3:1 ratio between male and female populations. The case reported here shows similar findings with compare to reported cases where it mentioned that GOC is more common in the middle-aged male population.[6]

GOC has no specific clinical findings. Literature shows that the most common feature is painless swelling at the affected site. In this case, there is no extraoral as well intraoral swelling identified.[7] The presented case was reported to our institute because of pain and sensitivity at the site of the lesion. In this sense, this lesion has a unique clinical presentation compared to all other reported cases.

This cyst is always associated with the tooth-bearing area of the jaw. Mandible is most commonly affected compared to the maxilla. 80% of all GOC cases are reported in mandible. The involvement of the anterior mandible is twice as compared to the posterior segment.[5, 7] In the present case the lesion is extending from the left mandibular second premolar to the right mandibular lateral incisor. This finding is consistent with published studies wherein it is reported that the anterior mandible has a high probability of a diagnosis of GOC.

There is no unique radiographical presentation of GOC and it is very similar to other osteolytic odontogenic lesions. Radiographical findings of GOC show a well-circumscribed osteolytic lesion with cortical bone expansion and root resorption or displacement of associated teeth. The lesion may be unilocular or multilocular with smooth or irregular margins. Chrcanovic et al. reviewed fifty-eight publications on GOC including 169 cases. Their study identified that 61.5% of cases were unilocular with cortical bone expansion.[2, 8] This present case showed multilocular radiolucency with no noticeable cortical bone expansion. This radiographical finding is unique and very similar to odontogenic Keratocyst where the lesion shows expansion in the anterior-posterior direction rather than the Bucco-lingual direction.

Microscopic features of GOC are variable and sometimes its diagnosis becomes a challenge for the pathologist. Till now less than 200 cases are published with reliable histopathological information. This cyst has a resemblance to a Botryoid Odontogenic cyst, Dentigerous cyst, and Intra-osseous Muco-Epidermoid Carcinoma.[9] Definitive diagnosis of GOC can't be achieved through incisional biopsy as this lesion shows focal variations in architecture. One of the published case reports mentioned that an incisional biopsy of the lesion falsely diagnosed GOC as a Dentigerous cyst and after enucleation the lesion reoccurred because of a conservative surgical approach.[4] Due to the variable thickness of epithelial lining, some areas of the lining may have a resemblance with Reduced Enamel Epithelium and it may mimic Dentigerous Cyst. In another case report the Intra-osseous mucoepidermoid carcinoma is misdiagnosed as GOC in an incisional biopsy sample. Intra-osseous Mucoepidermoid carcinoma is a low-grade malignant tumour that contains multiple cystic spaces and in incisional biopsy samples it can be misdiagnosed as GOC.[10] Kaplan et al. established certain major and minor criteria for microscopic diagnosis to avoid inaccurate diagnosis. Major criteria include non-keratinized stratified squamous epithelial lining with variable thickness, presence of hobnail cells, mucous goblet cells with intraepithelial mucous pool with or without crypts lined by mucous-

producing cells, inter epithelial glandular or duct-like structures. Minor criteria include papillary projections, ciliated cells, and clear or vacuolated cells in basal or supra-basal layers.[6] In the present case microscopic evaluation was done by two independent oral pathologists. Similar findings were recorded by each pathologist which are in accordance with Kaplan's criteria.

Various management protocols are proposed by several researchers for GOC. These protocols vary from conservative to aggressive treatment modalities. Enhanced Ki-67 expression of GOC is linked with the high proliferative nature of the lesion which is associated with chances of recurrence.[6] Most of the studies suggest that aggressive treatment modalities like marginal resection to segmental jaw resection should be carried out to prevent the recurrence of lesion.[4, 6]

CONCLUSION

GOC is a rare odontogenic cyst that commonly affects the anterior region of the mandible. Clinically and radiographically this cyst has no unique characteristic features to differentiate it from other odontogenic cysts and tumours. This cyst may be associated with pain and paraesthesia of the affected site. Incisional biopsy is not a recommended method to diagnose GOC as it may show variable histopathological architectural variations and may be misdiagnosed with other lesions. Prompt and aggressive surgical management is required to prevent the recurrence of the cyst. More publications are required to understand the true nature of lesion.

Declaration by Authors

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