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RESEARCH ARTICLE

CONSERVATIVE MANAGEMENT OF A LARGE GLANDULAR ODONTOGENIC CYST IN THE ANTERIOR MANDIBLE: A RARE CASE REPORT WITH TWO YEARS FOLLOW-UP

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ABSTRACT

Gardener *et al*. in 1987 described Glandular odontogenic cyst as an uncommon developmental odontogenic cyst. It may manifest as an intra-bony solitary or multilocular cyst of odontogenic origin. It has a high rate of recurrence similar to that of the keratocystic odontogenic tumor. The increased recurrence rate shall be due to its multilocular nature, intrinsic behaviour and incomplete removal of the lining. We report a case of large glandular odontogenic cyst in the anterior mandible treated by enucleation followed by chemical cauterisation with carnoy's solution. The patient was followed for two years, no recurrence noted, and gradual increase in new bone formation was seen in the follow-up orthopantomograms.

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INTRODUCTION

Gardener *et al* first described glandular odontogenic cyst in 1988 as a distinct clinicopathologic entity and a cyst of odontogenic origin with a rare occurrence. Padayachee and Van Wyk1 as Sialo odontogenic cyst described it, as it was similar to that of the botryoid odontogenic cyst with a glandular element in it. Sites of predilection are the anterior mandible and maxillary tuberosity (Gardener and Morency, 1993). Middle age groups are more commonly affected with a slight predilection for men. Only 111 cases of Glandular odontogenic cyst (GOC) have been reported in the literature (Kaplan *et al*., 2008). Magnusson *et al*. (1997) observed that only 0.012% of the cysts seen in the oral cavity have fulfilled the criteria of GOC microscopically. It appears as a well-defined unilocular or multilocular radiolucency with scalloped and sclerotic margins. Very rarely, there may be root resorption and tooth displacement with cortical perforation leading to an extension of the cyst into adjacent soft tissues (Junior *et al*., 2004; Tran *et al*., 2004; Noffke and Raubenheimer, 2002). Histopathological appearance of GOC shows a cyst wall lined by non-keratinised epithelium with papillary projections, nodular thickening, mucus filled clefts, and mucous lakes. Sometimes vacuolated cuboidal basal cells can be present. The aim of this report is to present a case of large GOC treated conservatively with a follow-up of 2 years without recurrence.

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CASE REPORT

A 49-year-old female patient reported to our institute with the chief complaints of painful swelling in the lower front teeth region for past 2 years. History revealed that it started 2 years back as a small swelling and gradually increased in size. Extra orally, the diffuse swelling was obvious and produced facial asymmetry, approximately 3x3 cm extending from right parasymphysis region to left parasymphysis region, inferiorly towards lower border of the mandible (Fig 1). On intraoral examination, a well-defined single diffuse swelling was present in the lower anterior region of size 5x5 cm approximately from the distal aspect of 35 to distal aspect of 45. On palpation, grade 3 mobility was found from 43 to 33. 46 were missing (Fig 2). An Orthopantomogram (OPG) demonstrated a well defined unicystic corticated radiolucency present in the lower anterior region of size 5x8 cm approximately which is extending from distal of 45 to mesial of 36 with thinning of the cortical plates showed displacement of teeth but no root resorption (Fig 3). Computed tomography imaging revealed a large well-defined lytic and expansile lesion involving the mentum and a portion of the body of the mandible, causing thinning of the cortical bone along its outer aspect (Fig 4).

Figure 1. Extraoral picture showing swelling in chin

Figure 2. Intraoral photograph showing labial mandibular diffuse swelling from 35 to 45

Figure 3. The panoramic radiograph shows a well defined, radiolucent, unilocular lesion from distal of 45 to mesial of 36, with smooth borders and tooth displacement

Figure 4: The Axial CT showing a large well defined lytic and expansile lesion involving the mentum and portion of body of the mandible, causing thinning of the cortical plate along its outer aspect where as lingual cortex is intact

Occlusal view showed unilocular radiolucency in the anterior mandible with buccal expansion and thinning of cortical bone. The lingual cortical plate was intact. On histological examination, the tissue appeared to be part of a cyst, lined by epithelium and connective tissues. The lining epithelial cells are of cuboidal in shape with a centrally placed nucleus and are in three to four layers in thickness and resembling the reduced enamel epithelium. In certain areas, epithelium is pseudostratified ciliated epithelium. Apart from the epithelial cells, mucous cells and microcystic areas filled with mucin are also noticed (Fig 5).

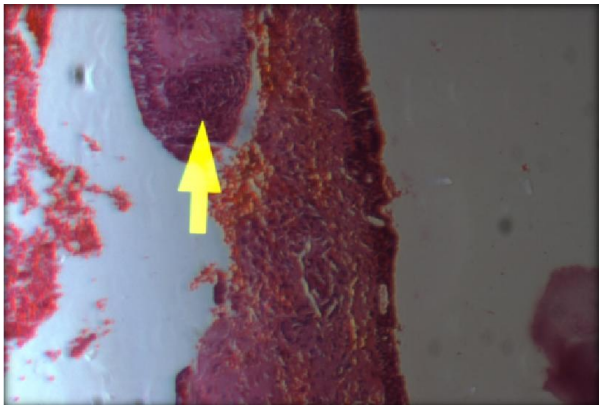

Figure 5. Photomicrograph showing cystic lining exhibiting focal thickening of epithelium. Mucous cell and microcystic area filled with mucin are also noticed

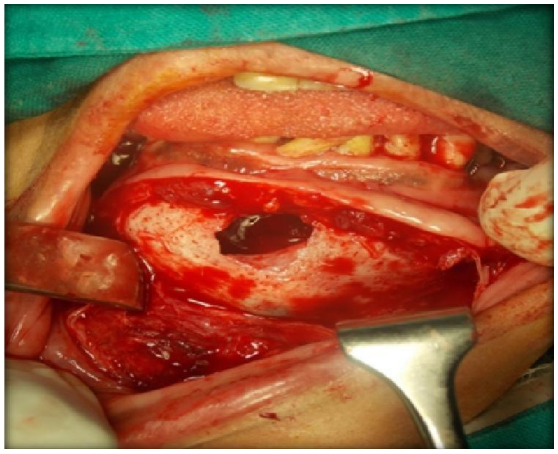

Figure 6. Intraoperative photograph showing the lesion which is present in the anterior region of the mandible

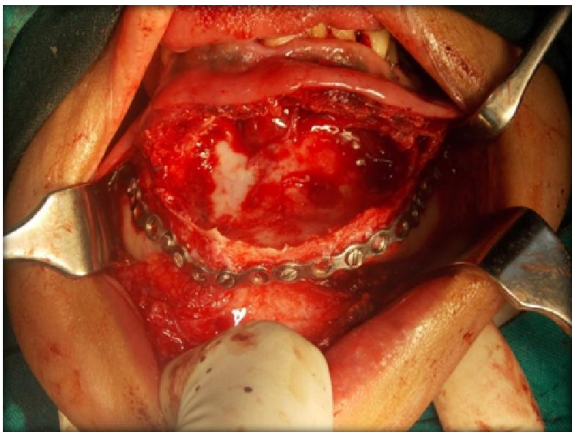

Fig. 7. Intraoperative photograph showing cystic cavity after thorough enucleationreinforced with 2mm stainless steel reconstruction plate

Fig. 8. Third month post op OPG

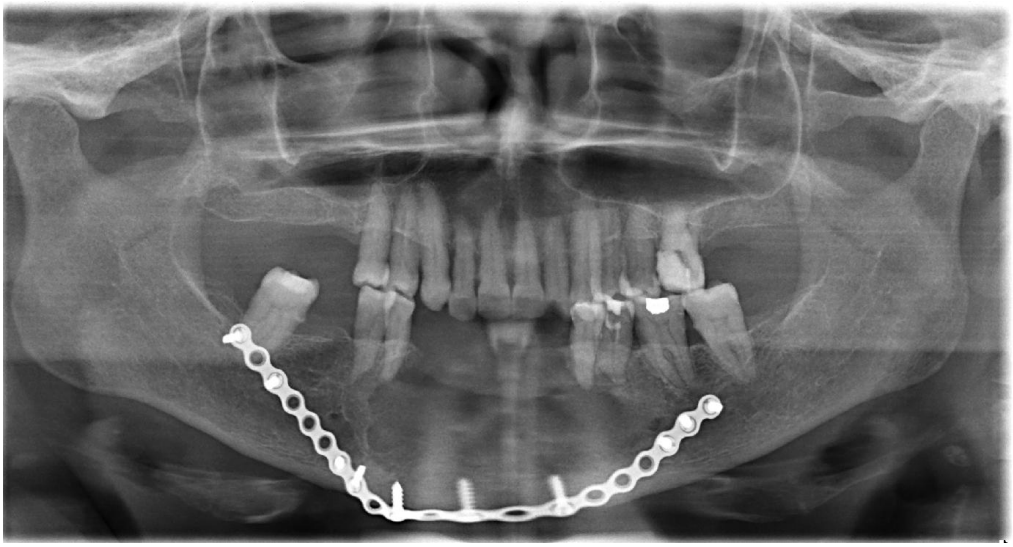

Fig. 9. sixth month post op OPG shows healing of lesion

Fig. 10. Ninth month post op OPG

Fig. 11. One-year post op OPG

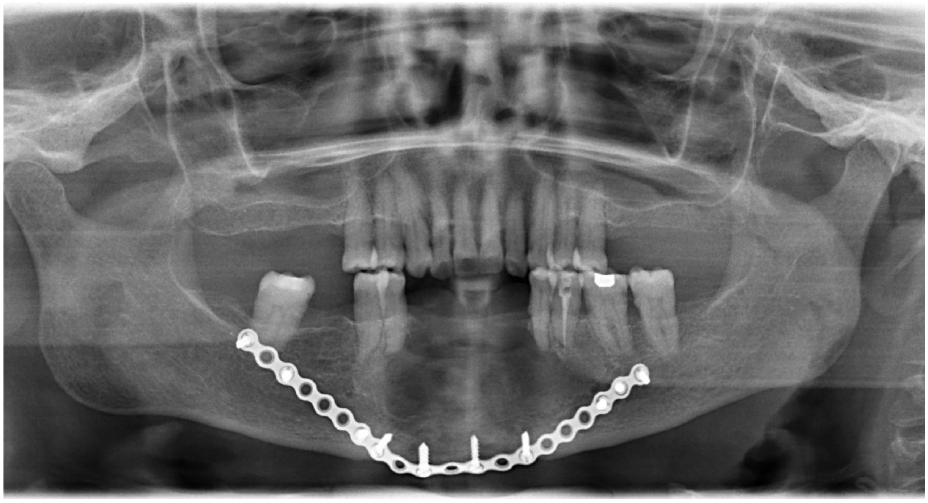

Fig. 12. Two-year post op OPG shows new bone formation and healing of lesion

The underlying connective tissue exhibits fibrous tissue running in parallel bundles resembling capsule of the cyst with wavy and active fibroblast. Deeper areas exhibit dense fibrous connective tissue with extravasated blood. Based on the clinical, radiographic, and histopathologic features the diagnosis was given as glandular odontogenic cyst. Under general anaesthesia, the cyst was thoroughly enucleated (Fig 6) and chemical cauterisation did with carnoy's solution. A long 2mm miniplate was fixed to reinforce and thereby pathologic fracture can be avoided as the labial cortex was completely removed due to thinning of the cortex (Fig.7). The patient was followed for two years on a basis of 3 months, 9 months, 6 months, 1 year and 2 years. (Fig 8 - 12). No evidence of recurrence noted. The evidence of new bone formation in the enucleated bone cavity was reasonably increasing in the series of postoperative radiographs taken.

DISCUSSION

Although histogenesis was thought to be from salivary gland, the present histopathological features of GOC strongly suggest the odontogenic origin (Oliveria *et al*., 2009). Motals *et al* (2012) studied the cytokeratin expression in GOC and Mucoepidermoid carcinoma. They found GOC to be positive for CK 19 were as low-grade Mucoepidermoid carcinoma. Thus, they concluded that CK 18 and 19 could be useful markers in differential diagnosis of GOC and confirmed the odontogenic origin of GOC. Ide *et al* reported a case of glandular odontogenic cyst presenting as a dentigerous cyst and containing hyaline bodies, which have been considered exclusive to odontogenic cysts (Ide *et al*., 1996). These findings also support the evidence that glandular odontogenic cyst is odontogenic in nature. Treatment options for GOC in literature are varying and not evidence based as the cyst is very rare to occur. It includes simple curettage to enucleation for small and unilocular lesions and peripheral osteotomy, marginal resection and segmental resection for large and multilocular lesions (Hussain *et al*., 1995). The recurrence rate in literature ranges from 21% to 55% and is attributed to the size of the lesion. Only 14.4% of the small lesions recur in contrast to 85.6% of the large lesions (Kaplan *et al*., 2005). Similarly, resorption of roots, cortical bone perforation, and multilocular lesions are associated with high rate of recurrence (Kaplan *et al*., 2005). Hence, it is better to have a follow-up of at least 3 years. The cyst in our case was enucleated and chemical cauterisation did with carnoy's solution. We had a follow-up of 12 months, there is no clinical and radiological evidence of recurrence, and new bone formation was evident in subsequent X rays. We conclude a diagnosis of glandular odontogenic cyst is only possible with histopathological examination.

As there is difficulty in predicting the biology of the disease with a diagnosis of glandular odontogenic cyst from the literature, patients with GOC should have a still a longer follow-up because of the possibility of recurrence even after several years of treatment.

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