GLANDULAR ODONTOGENIC CYST - A CONSERVATIVE SURGICAL APPROACH

Dr. Ashutosh Deshpande¹, Dr. Senthil Nathan², Dr. Senthil Murugan P³, Dr. Sneha Krishnan⁴
Department of Oral and Maxillofacial Surgery, Saveetha Dental College SIMATS, Saveetha University
Chennai 77, Tamil nadu, India

ABSTRACT

Glandular odontogenic cyst (GOC) is a rare jaw condition with similarities to botryoid odontogenic cyst and mucoepidermoid tumour. According to WHO’s recent classification, glandular odontogenic cyst is classified as a developmental cyst arising from odontogenic epithelium, despite the fact that its histogenesis is unknown. Histopathologically GOC is characterized by epithelial lining consisting of cuboidal or columnar epithelium with crypts within the epithelium. GOC has a wide range of radiographic appearances that are not characteristic. Curettage, enucleation, and en bloc excision are some of the GOC treatments that have been employed. The recurrence rate of GOC documented in the literature ranges from 21% to 55%, depending on the therapeutic modalities used. We report a case of a 43 years old male presents with glandular odontogenic cyst occurring in the left posterior mandible.

Keywords: Glandular odontogenic cyst; Sialo -odontogenic cyst; odontogenic cyst; cysts of jaw; Surgical enucleation; Conservative surgical treatment

I. INTRODUCTION

Since 1990 many reports have been published identifying association of jaw cysts with number of mucinous glandular components in the lining or the complete lining with glandular elements. In 1987, Padayachee and Van Wyk reported two cases that were similar to Botryoid odontogenic cysts; however, included a glandular component which they described as sialo-odontogenic cysts. Earlier, the odontogenic cysts were reported with clear cell linings and were accepted as glandular prosoplasia from the squamous cell linings which were not characteristic; however, later Gardner et al mentioned that the epithelial lining was odontogenic and described as an unique pathologic entity of the jaws called glandular odontogenic cyst. which was later accepted by the World Health Organization (WHO) [1,2]. These are rare odontogenic cysts that develop in the jaws exhibiting potentially aggressive behaviour and may have the tendency to recur [3]. Glandular odontogenic cysts are more clinically aggressive than radicular and dentigerous cysts, necessitating more radicular surgery and lengthier follow-up; nonetheless, because these cysts frequently return, it is important to get the correct diagnosis. The most common site of occurrence is the mandibular anterior region, which manifests as an asymptomatic, slow-growth swelling[4]. It commonly affects male individuals in the 4th-6th decade of life[5].

On radiographs, the lesion may appear as a single or multiple-locular radiolucency with well-defined boundaries and a scalloped border[6]. The differential diagnosis for these lesions should include ameloblastoma, odontogenic keratocyst, odontogenic myxoma, central giant cell granuloma, aneurysmal bone cyst, radicular cyst, and, low-grade mucoepidermoid carcinoma. The lesions involving posterior mandible should be suspected for low grade mucoepidermoid carcinoma[7].

A non-keratinized epithelial cyst wall lining with papillary projections, nodular thickenings, mucous filled clefts, and "mucous lakes" distinguishes GOC histologically. It also has cuboidal basal cells that are occasionally vacuolated. Although some authors feel marginal resection is a more reliable option due to the cyst's tendency to return after curettage and enucleation[8], the GOC’s can be treated effectively by enucleation and curettage[1]. The presented case was treated conservatively by enucleation and peripheral ostectomy.

CASE REPORT
A 43 years old male patient reported to the department of oral and maxillofacial surgery with a chief complaint of pain and swelling in lower left jaw, since one month. The swelling was intraoral and smaller in size, and it progressively grew to its current size.

On clinical extraoral examination, no gross facial asymmetry was present (fig1); however mild pain was present on palpation. Intraorally, there was obliteration of the vestibule from 47 to the retromolar region with mild inflammation of mucosa (fig.2). No loss of sensation or paresthesia was detected. No pus discharge or any inflammatory exudate was present.

Patient’s blood investigations were within normal limits. Cone beam computed tomography was suggestive of well-defined encapsulated mixed radiopaque-radiolucent lesion in the left angle of the mandible from 46 to mandibular ramus with buccolingual cortical expansion with an impacted molar and involvement of the mandibular canal. (Fig. 3). Incisional biopsy was sent for histopathological examination with differential diagnosis of the lesion included ameloblastoma, Odontogenic Keratocyst, dentigerous cyst, odontogenic myxoma and radicular cyst. Histological examination revealed non keratinized stratified squamous epithelial lining with cuboidal and columnar cells with cilia in few cells along with numerous goblet cells. Few areas showed pseudo glandular structures and pseudo cystic spaces suggesting Glandular odontogenic cyst (fig. 5).

Based on the history, clinical presentation and radiological investigations, the decision was made to treat the lesion conservatively with enucleation followed by peripheral ostectomy under general anesthesia. Using intraoral incision, a mucoperiosteal flap was elevated to expose the lesion. The specimen was enucleated with impacted molar and extraction of 46 followed by peripheral ostectomy. Histopathological examination of the excised specimen showed a lesion measuring 1.5x1x 0.9x 0.7cm (fig.4).

Microscopic description of lesion - Multiple sections show odontogenic epithelial lining and connective tissue wall. The odontogenic epithelial lining is of variable thickness and composed predominantly of non keratinized stratified epithelium and few areas showing stratified squamous epithelium, along with abundant superficial mucous cells. There is evidence of numerous microcysts, basal and parabasal clear cells, epithelial whorls and duct like structures in the odontogenic epithelial lining. The dense connective tissue wall shows moderate chronic inflammatory cell Infiltrate predominantly plasma cells and there is also evidence of loss of characteristic structure of epithelial lining in the areas associated with chronic inflammation(fig 5).

Fig 1: Extraoral photo Showing no gross asymmetry
II. DISCUSSION

Although the GOC is a very uncommon lesion, it should be evaluated when diagnosing unilocular and multilocular radiolucencies of the jaws. GOC has an aggressive potential, a high frequency of cortical perforation, and a reasonably high risk of recurrence, thus a precise diagnosis is critical[9].

The GOC was previously known as sialo-odontogenic cyst, mucoepidermoid odontogenic cyst, and polymorphous odontogenic cyst until the World Health Organization adopted GOC as the preferable name. Although there are several cases of GOC hybrid lesions with other odontogenic malignancies that appear to be odontogenic in origin, the development of GOC is unclear[10].

The glandular odontogenic cyst affects people of all ages. A slight male predilection was reported. The anterior mandible was the most commonly affected area. The lesions progress slowly over time, are painless, and locally destructive. The mandible appears to be involved more frequently than the maxilla (87.2 percent). The average age of occurrence is 49.5 years, with a range of 10–90 years. GOCs range in size from 0.5 to 12 cm in diameter.

Radiographically these lesions appear well defined with a unilocular or multilocular pattern but without specific diagnostic features. Odontogenic keratocyst, unicystic or multicystic ameloblastoma, central mucoepidermoid...
tumour, lateral epithelial cyst (LPC), and central mucoepidermoid tumour all have the same characteristics (a locally aggressive variant of LPC). Ameloblastoma is, without a doubt, the most common differential diagnosis because of its higher prevalence. Furthermore, multilocular types of GOC cannot be differentiated radiologically from ameloblastoma: uneven loculation and the presence of sclerotic boundaries with perforation foci might aid in the diagnosis of GOC. The lesion in the presented case was unilocular with embedded tooth within and showed expansion of the mandible, thinning the cortical plates (fig.3).

Histologically, glandular odontogenic cyst is lined in parts by a nonkeratinized stratified epithelium of varying thickness. The epithelium has a glandular or pseudoglandular structure, with goblet mucous producing cells as well as intraepithelial crypts or microcysts containing mucus. These microcysts may open onto the surface of the epithelium giving a papillary or corrugated appearance. Some cells may also be ciliated. Occasionally the epithelium is thinner, similar to reduced enamel epithelium. Epithelial thickenings or plaques may be present either in this thin epithelium or in the stratified epithelium. Interface between epithelium and connective tissue is flat.

To avoid incorrect diagnosis like odontogenic cyst or ameloblastoma, some microscopic criteria should be strictly applied. The presence of a superficial layer of cuboidal or columnar epithelial cells occasionally with cilia and glandular or pseudoglandular structures and intraepithelial cysts containing mucin are some of the distinguishing features. Thin nonkeratinized stratified squamous epithelium may border the rest of the cyst. According to the literature, to investigate the potential that GOC and mucoepidermoid carcinoma both are reported to be unilocular or multilocular on radiography, they can penetrate and damage bone. A unique characteristic not seen in low mucoepidermoid carcinoma is the presence of epithelial plaques on microscopy (typical of LPC)[11].

Several approaches for the treatment of GOC have been proposed in the literature. Enucleation, marsupialization, curettage with and without peripheral ostectomy, curettage with adjuvant Carnoy solution, or cryotherapy are all treatments that are considered conservative. However, a more severe technique (marginal resection to partial jaw resection) has lately been suggested as the therapy of choice for GOCs by numerous publications [12].

Some large multilocular GOCs appear aggressive enough to require aggressive surgical treatment. According to some experts, GOC behaviour and vulnerability to recurrence are linked to cell dynamics in the lining epithelium. Increased Ki-67 positivity and decreased p53 positivity were found in immunohistochemical testing, indicating a high proliferative index and a low propensity. Some authors state that the high recurrence incidence is attributable to the cyst’s thinness and the existence of microcysts, which interferes with complete removal of cysts. According to Kaplan et al, no recurring instances were recorded in GOCs treated with major surgical operations, but conservative approaches were shown to be often related with recurrent illness[13].

In the presented case we chose to treat the lesion conservatively considering the unilocularity and size of the lesion. the cyst was completely enucleated followed by the peripheral ostectomy. The cavity was packed with chlorhexidine dressing and the wound was closed primarily with a small opening to change the dressings. The chlorhexidine dressings were changed subsequently to allow healing. No recurrence was noted after the follow up of 6 months, the patient is currently under follow up.

III. CONCLUSION

In conclusion, GOC management is exceedingly challenging due to its rarity and aggressive Nature. However, these lesions can be treated conservatively by enucleation followed by adjuvant procedures like peripheral ostectomy, chemical cauterization or cryotherapy. Long term follow up should be carried out to detect potential recurrences.

REFERENCES:


www.turkjphysiotherrehabil.org 16015