Case Report I

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Glandular Odontogenic Cyst - Dilemma in Management

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Abstract
The glandular odontogenic cyst (GOC) is a rare developmental odontogenic cyst found in the teeth bearing area of the jaws, more often in the anterior mandible. Its’ behaviour is locally aggressive and tends to recur after enucleation. This case report is based on a GOC of a 53 year old male, which responded very well to decompression. However, this method of surgical treatment has not been emphasized in the literature in the treatment of GOC. The prognosis after treatment of this GOC had been good with no clinical and radiological evidence of recurrence for six years.

The finding of this case report highlights the benefits and the importance of this minimally invasive surgical treatment in the management of GOC. In conclusion we propose that this method should be evaluated as a cost effective alternative treatment option in the management of GOC.

Key words: Glandular odontogenic cyst, locally aggressive, anterior mandible, decompression, enucleation.

Introduction
The GOC was first introduced by Padayachee and Van Wyk in 1987 and proposed as “sialo odontogenic cyst”. One year later Gardner introduced the term “glandular odontogenic cyst”. In 1992 World Health Organization accepted glandular odontogenic cyst as a distinct pathological entity and the preferred term GOC since the possible salivary gland origin of the cyst has not been established.

GOC has been reported in 14 - 85 year old individuals and majority of them being above 40 years of age. The occurrence of GOC has also been reported to be common in the 6th decade of life with the mean age of 49 years. There is a slight male preponderance and the reported prevalence of this cyst is 0.017 % to 0.2 % of all odontogenic cysts. The usual presentation is disfigurement, swelling, resent change in alignment of teeth, difficulty in wearing the denture and rarely pain or tenderness.

The histological and immunohistochemical investigations of this cyst have been carried out extensively using paraffin embedded sections stained with hematoxylin and eosin, periodic acid Schiff- Alcian Blue, reacted with a range of monoclonal cytokeratin antibodies according to the alkaline phosphatase-anti-alkaline phosphatase method. The cyst consists of well-defined stratified squamous non keratinized epithelium of varying thickness and a cyst wall. Superficial layers of the epithelium show either cuboidal or columnar cells with cilia. Epithelial
plaque formation is a common feature and there are pseudo-glandular spaces containing periodic acid Schiff positive material and mucicamine positive material. The other possible findings are micro-cysts within the epithelium and goblet cells in the superficial layers of the epithelium. Only a few cases reported with hyaline bodies. The cyst wall contains a few inflammatory cells confirming its developmental origin.

The histopathological features of GOC are described as a combination of findings from a botryoid odontogenic cyst and a mucoepidermoid carcinoma, often causing a diagnostic dilemma for pathologists. The epithelial plaque formation is seen in lateral periodontal cyst and the botryoid cyst. The pseudo-glandular spaces and micro-cysts formation are seen in low grade intra-osseous mucoepidermoid carcinoma. Since these three pathologies share common histological features accurate diagnosis is mandatory in the management of GOC.

The radiological findings are not pathognomonic as the radiolucency can be either unilocular or multilocular. When it is unilocular, it simulates jaw cysts such as dentigerous cyst, radicular cyst and odontogenic keratocyst or tumors with unilocular radiolucency such as some odontogenic tumors, central giant cell lesions, vascular lesions or intra-osseous mucoepidermoid carcinoma. When it appears as a multilocular radiolucency, picture may resemble bone lesions such as ameloblastomas or bone cysts or tumors which give rise to multilocular radiolucency. Almost always the lesions tend to have well-demarcated and sclerotic borders which are scalloped or smooth. When the lesion exceeds the size of 6cm, it often tends to perforate the cortical plate. Tooth displacement and dental root resorption may be a feature. The multilocularity might be a size related phenomenon, and the cortical perforation shows its aggressive behavior.

In the management of GOC the following treatment modalities are being practiced:

1. Enucleation and surface curettage for small unilocular lesions. (+/ thorough extirpation)
2. Marginal, en-bloc or segmental resection for large multilocular lesions.
3. Marsupialization with a second surgery of enucleation and curettage where vital structures to be preserved.

Irrespective of the method used in the treatment of these cysts, it is mandatory to follow up the patients for several years due to reported recurrences of the lesion as long as 7 years after initial treatment.

Due to paucity of published data, there are many questions remaining to be resolved concerning histogenesis, biologic behavior and appropriate treatment of these lesions.

The purpose of this case report is to highlight another treatment option available which was experienced as an alternative approach to management of GOC with minimal morbidity and maximum outcome.

Case report
A 53 year old man reported to oral and maxillofacial surgery unit with the complaint of swelling of the lower jaw as a referral from the OPD dental clinic, Colombo South Teaching Hospital Kalubowila Dehiwala.

He had observed the gradual disfiguring of his chin region for about 1 ½ years and mild changes in the alignment of the lower teeth. There was no pain or tenderness over the affected area or the teeth in the affected region and no numbness or abnormal sensation felt over the affected gingiva or the lower lip.

Significant facial asymmetry was observed due to the swelling of the mental region mainly towards the left side. Distortion of the anterior mandible could be felt and it extended bilaterally from the mental region to parasympathetic regions.
Both jaws were partially edentulous, teeth of the clinically affected area of the mandible did not show discoloration or pathological level of mobility. Swelling was apparent labially and lingually. Labial swelling was extending from right lower canine region to the left lower premolar region. Lingual swelling was extending from the right lower lateral incisor region to the left lower lateral incisor region and appeared in a bluish gray color through the labial alveolar mucosa. It was fluctuant but did not blanch on pressure. Very mild mal-alignment was observed in relation to two lower central incisor teeth and the lower left lateral incisor tooth.

With this clinical picture, radiological investigations were carried out to assess the intra-bony lesion.

The following differential diagnoses were considered:
I. Odontogenic cyst - orthokeratinized odontogenic cyst
II. Odontogenic tumor - unicystic ameloblastoma
III. Non odontogenic cyst- unicameral bone cyst, aneurysmal bone cyst, traumatic bone cyst
IV. Non odontogenic tumor- enchondroma
V. Central giant cell lesion- osteoclastoma, histiocytosis X
VI. Central vascular lesion- hemangioma

The following radiographs were obtained and evaluated for extensions of the lesion, the behavior and features aid in the diagnosis:
A. Intra oral periapical radiographs of lower anterior teeth and right and left premolar regions
B. Lower standard occlusal radiograph
C. Dental panoramic tomography

The radiolucent lesion with ill-demarcated margin was extending from right lower first molar region to the left lower first molar region. (Figure 1) The radiolucency extended in between roots of teeth to varying heights destroying the lamina dura of the alveolar bone with no obvious root resorption. The inferior dental nerve had been displaced inferiorly. As the lesion was extensive and the margins were not demarcated, it was re-evaluated with computed tomography (CT).

An incisional biopsy was performed for histopathological diagnosis. A biopsy specimen of 1.0 x 0.5 x 0.2 cm sent to the Department of Oral Pathology, Faculty of Dental Sciences, Peradeniya. The microscopic findings showed a fibrous cyst wall lined by thin epithelium with two to three cell layers in most areas and focal areas of elongated basal cells with a mild degree of reverse polarity. The cyst wall was not inflamed. Even though those histopathological features were not pathognomonic, the most likely diagnosis was unicystic ameloblastoma.

A second set of periodical radiographs were taken during a waiting time for surgery to assess the root resorption of the affected teeth, revealed the lamina dura formation of pathologically affected alveolar process. It was decided to perform a second incisional biopsy for re-evaluation of the lesion in the absence of pathognomonic features in the histopathological evaluation and radiological finding of lamina dura formation of alveolar process. The second biopsy revealed non-keratinized stratified squamous epithelium with focal epithelial plaque formation, focal pseudo glandular spaces, surface papillary projections and glandular metaplasia. These histopathological features were consistent with those of GOC.

The new pathology report led to evaluate the lesion with a second CT. The comparison of the two CTs indicated a significant bone regeneration of the anterior cortex over the previous bony defect; measurements being 3.2x1.4 cm in the first CT vs. 3.2x0.6cm in the second CT, which has taken place during the course of two months. The IOPA finding of lamina dura formation and CT finding of bone apposition, suggested that surgical sites of two incisional biopsies have served as means of surgical decompression.
With the initial diagnosis of unicystic ameloblastoma, the treatment plan was a marginal resection of the mandible with pathological lesion, reconstruction of the surgical bone defect with a bone graft and mini-plate osteosynthesis under general anesthesia.

However the observed change of behavior of the lesion, led the clinician to follow “wait and see” policy. With the consent of the patient, it was decided to follow up the patient regularly with dental panoramic tomography.

Patient did not have any complaint one year of follow up. A sinus opening in the labial alveolar mucosa was observed in relation to the right lower central incisor tooth. The third CT reporting confirmed the findings of the radiographs. Currently the patient is on regular follow up for the sixth year and there is no asymmetry of the face, no visible or palpable bone distortion over the previously diseased area of the mandible, but only a sinus tract of roughly 0.5 cm in diameter in the labial gingiva in relation to the midline. Last radiograph (March 2014 - Figure 3) showed remnants of a radiolucent lesion in few focal areas, with signs of bone apposition.

**Discussion**

Until 1987, no one knew this cyst as glandular odontogenic cyst and, that does not necessarily mean such cyst did not exist, but could have been misdiagnosed as some other cystic lesion and treated differently.

In 1988 this cyst was introduced as a rare cyst of the jaw\textsuperscript{16}. Almost after 20 years it has remained a relatively rare cystic lesion of the jaw\textsuperscript{17}. As the current microscopic criteria have not been universally accepted; many GOCs have been identified as central mucoepidermoid carcinomas and GOC may not be as rare as it was thought\textsuperscript{11}. Several treatment modalities have been used in

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**Figure 1.** First panoramic tomograph showing the lateral extensions of the radiolucent lesion from right lower first molar region to left lower first molar region and vertical extension in between dental roots and inferior displacement of the inferior dental nerve in left side.

**Figure 2.** Intermediate - dental panoramic radiograph showing reduction of the right lateral and vertical extensions of the radiolucent lesion.

**Figure 3.** Last dental panoramic tomograph showing few irregularly ovoid radiolucent areas in relation to apices of right second premolars to left lower premolars.
the treatment of GOC. These include curettage, enucleation with careful dissection of the margins of bone (Peripheral ostectomy), marginal resection, en-bloc resection and segmental resection of jaw bones. The prognosis of this cyst remains unclear. However, the aggressive nature of GOC has been reported, and at least 25% to 55% recur following curettage making it imperative to follow up the patient carefully for several years after curettage or enucleation. Due to high recurrence rate and aggressive growth potential, some authors believe that radical resection may be more reliable treatment for GOC\textsuperscript{18}. Therefore both conservative and radical methods of treatment for GOC have been suggested in the literature\textsuperscript{19,20}.

The treatment plan for each lesion should be customized, by utilizing the reliable evidence in the literature and patients’ preferences. Those that are treated with conservative approaches must be followed up with regular radiological follow ups.

It may be difficult to define the characteristics and behavior of GOC, leading to dilemma in selecting a proper treatment protocol due to less number of reported cases. The above mentioned case would help to enrich the literature in future treatment planning. This unintentional finding prevented a planned resective and reconstructive surgery and avoided possible surgical morbidity, mortality and the hazards of general anesthesia. The logical and timely taken clinical decision led to a favourable outcome of this clinical scenario. Although the marsupialization is a form of decompression and used in the treatment of GOC, it is for the preservation of vital structures as a preparation for definitive surgical treatment. The other important aspects of this case scenario are the importance of re-evaluation and observing the lesion after interfering with its natural history and re-evaluation of the initial diagnosis in the absence of pathognomonic features.

However the assumption of decompression as a promising treatment for GOC may be wrong in the following circumstances;

1. If the histopathological diagnosis was questionable
2. If the pathology was an unknown sub-type of the diagnosed lesion
3. Individual variation of the pathology
4. Individual variation of the host factors such as subjective immunity
5. Microbiological factors of the individual and
6. Any combination of the above mentioned reasons

The unexpected outcome of this clinical scenario reveals a possible alternative treatment approach which is beneficial to the patient in several ways and rewarding the surgeon and also possibility of expanding it into a treatment protocol which would be useful in reducing the burden on health economics. However, such behavior of GOC has not been reported in literature, this treatment procedure should be further evaluated by; carrying out case control studies to assess the behavior of GOCs following decompression. The findings of this case report contributes to the current understanding of GOC and may pave way forward for the practice of evidence based dentistry and also may help to prevent unnecessary resection of mandibles and maxillae of patients with GOCs.

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