CASE REPORT

Glandular Odontogenic Cyst Arising from Maxillary Sinus an Unusual Entity: Report of a case and review on Diagnostic Dilemmas

Shrirang Sangle[1], Varsha Sangle[2] , Smita Chaware[3], Dipali Gundre[4] , Dr Yogesh Nagargoje[5], Dr Abhishek Gole[6]

1 Department of Periodontology, MIDSR Dental College, Latur.
2,3,4 Department of Oral Pathology & Microbiology, MIDSR Dental College, Latur.
5,6 Department of Prosthodontics, Crown & Bridge, MIDSR Dental College, Latur.

Abstract: Glandular Odontogenic Cyst (GOC) is a moderately rare cyst of odontogenic origin. It shows glandular features that are thought to indicate the pluripotentiality of odontogenic epithelium. It is seen in middle age group of patients and commonly involves the anterior region of the jaws. It shows non-specific clinico-radiographic findings which may resemble other lesions, but has typical histopathological features which help in its diagnosis. This paper reports a rare case report of GOC arising from maxillary sinus and a review on clinical, radiographic, histopathological, differential diagnosis and Immunohistochemistry (IHC).

Keywords: Glandular odontogenic cyst, sialo-odontogenic cyst, maxillary sinus, epithelial plaques, goblet cells.

Corresponding Author: Dr Shrirang Sangle, Department of Periodontology, MIDSR Dental College, Latur, Email- Sshri58@gmail.com

Case Report

A 40-year-old female patient reported with the chief complaint of a painless swelling in posterior right region of the maxilla since 2 months. History revealed that the swelling was present since 2 months and increased gradually and attained the present size and was associated with pain on pressure since 1 month. Difficulty in breathing through the right nostrils and heaviness on right side of maxillary sinus region. Extra-orally, the swelling was not obvious and did not produce any facial asymmetry. Intra-oral examination revealed a swelling present at palatal aspect with size of 4cm (anterio-posteriorly) x 2.5cm (medio-laterally). The margins of swelling are well defined with smooth surface area. There is no buccal obliteration. On palpation, it was soft, fluctuant, non-tender swelling. A provisional diagnosis of muco-epidermoid carcinoma, retention cyst of maxillary sinus was made.

Radiographic examination revealed a radiolucent area in 16, 17, 18 region (Figure

1). CT Scan showed a well defined 3.5x2.8x3.4 (AP x ML x SI) sized cystic, non enhancing, osteolytic expansile lesion from maxillary alveolar arch involving roots of right 16 & 17 molars evading in the right maxillary sinus and hard palate. Right osteomeatal unit is obliterated with widening and mucosal thickening is seen on right maxillary sinus (Figure 2).

Aspiration yielded 3ml of yellow brown fluid and was submitted for cytological examination which showed predominantly macrophages and few lymphocytes. Impression of a cystic lesion was made. After the surgical enucleation, the specimens were submitted for histopathological examination and for further confirmation of the diagnosis the tissue were subjected to Ki67 IHC markers.

On histopathological examination, the lesional tissue showed cystic lumen lined in parts by nonkeratinized stratified epithelium of varying thickness, few areas showed plaques and epithelial thickenings (Figure 3). Occasionally superficial layer of columnar epithelium with cilia was seen (Figure 4). The epithelium had glandular as well as goblet cell (Figure 5) and at places it resembled reduced enamel epithelium (Figure 4). The interface between the epithelium and connective tissue was flat. The connective tissue capsule was made up bundles of collagen and moderate degree of inflammatory cells. All features were suggestive of glandular odontogenic cyst. High immunoreactivity for Ki 67 further confirms our diagnosis to be Glandular odontogenic cyst (Figure 6).

Discussion

The GOC is an uncommon lesion, comprising only 0.012% to 1.3% of the cystic lesions of the jaw. [1] It is destructive in nature and most commonly occurs in the mandibular symphyseal region, with only a 30% incidence seen in the maxilla. It is extremely rare in maxillary sinus, till date only 4 cases have been reported. GOC usually occurs in middle-aged adults, most often after the fourth decade of life, with a slight male predilection. [5] It is four times more common in the mandible as compared to the maxilla and has a predilection for the anterior region of the jaws The present case differs from the literature in that it was diagnosed in a 40-year old female, and occurred in the maxilla extending antero-posteriorly from maxillary alveolar arch involving roots of right 16 & 17 molars which is not commonly seen. [5, 6]

Clinically, this cyst is not specific, with lesions usually presenting as asymptomatic swellings. The radiographic appearance also varies and is not pathognomonic. It may present as a unilocular or multilocular radiolucency, which typically shows well-defined borders. Occasionally, scalloping of the border may be noted, while root resorption and displacement are not usually seen. [7] Radiographically, GOC may look like lesions like radicular cyst, keratocystic odontogenic tumor, ameloblastoma and central giant cell granuloma. An interesting observation in our literature review of the previous cases of GOC reported in India showed that 7 out of 19 cases were also placed in edentulous or partly edentulous areas (where teeth were extracted previously. This could identify that GOC may possibly grow inside the jaws without any symptoms in the initial stages, and cause swelling and cortical expansion only at a later date. [7]

Histopathologically, GOC shows definite characteristic features that were subdivided by Kaplan et al into major and minor criteria based on the frequency of occurrence of each feature in the formerly reported cases. At least focal presence of the major criteria is mandatory, while the presence of minor criteria further supports the correct diagnosis. [7,8] The major criteria include a lining of squamous epithelium of varying thickness which may show epithelial 'spheres' or 'whorls' with absence

of basal palisading, cuboid-shaped eosinophilic ('hobnail') cells, and intraepithelial mucous / goblet cells or pools, with presence of glandular or duct-like structures. Papillary proliferation of the epithelial lining, multicystic appearance, and presence of ciliated or clear cells in the epithelium were incorporated as minor criteria. The present case showed most of the characteristic histopathological features of GOC described above. [8] The histopathological features of GOC have been create to look like a number of lesions having a wide clinico-pathologic spectrum ranging from other odontogenic cysts like lateral periodontal cyst to malignant neoplasms such as central low-grade mucoepidermoid carcinoma, and therefore suspicious and detailed microscopic examination is essential in arriving at a exact diagnosis. [8, 9] Treatment of choice of this lesion remains controversial, and varies from enucleation and curettage to local block excision depending on the size and aggressiveness. Enucleation is ideal for small unilocular lesions, whereas supervision of large lesions includes enucleation with peripheral ostectomy for unilocular cases and marginal resection or partial jaw. [9]

Conclusion

We report a rare case of Glandular Odontogenic Cyst in maxillary sinus, the purpose of reporting this case was to add to the existing knowledge about this rare cyst which still has an uncertain nature. Till date only 4 cases have been reported in the English literature. Our article describes in detail about glandular odontogenic cyst and its clinical and histopathological differential diagnosis. Advanced techniques like IHC help us in better understating of the origin and pathogenesis of such cases, which creates diagnostic dilemmas.

Figure legends:
Figure 1: Radiograph showing a radiolucent area in 16, 17, 18 region

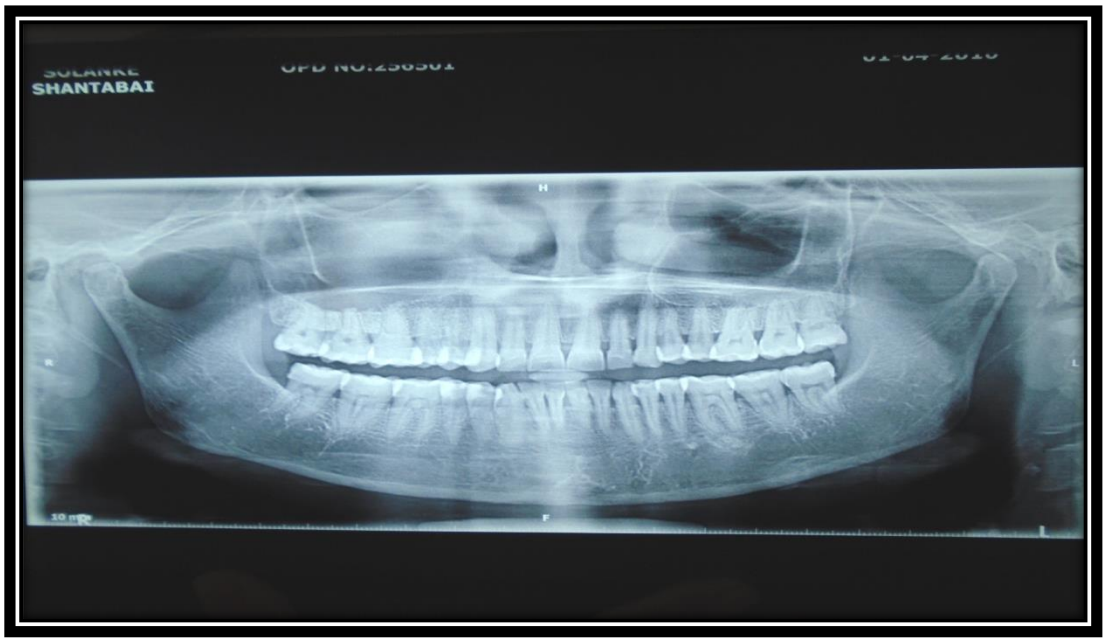


Figure 2: CT scan showing a well defined cystic, non enhancing, osteolytic expansile lesion.

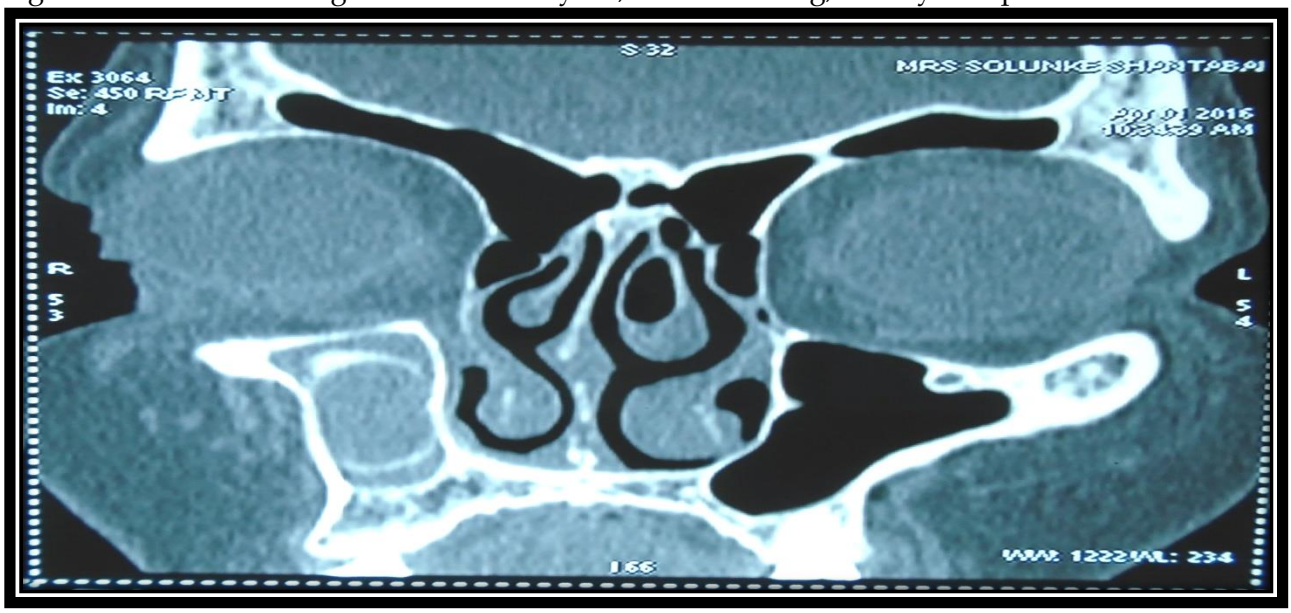


Figure 3: H&E section shows cystic lumen lined by nonkeratinized stratified epithelium with plaques and epithelial thickenings.

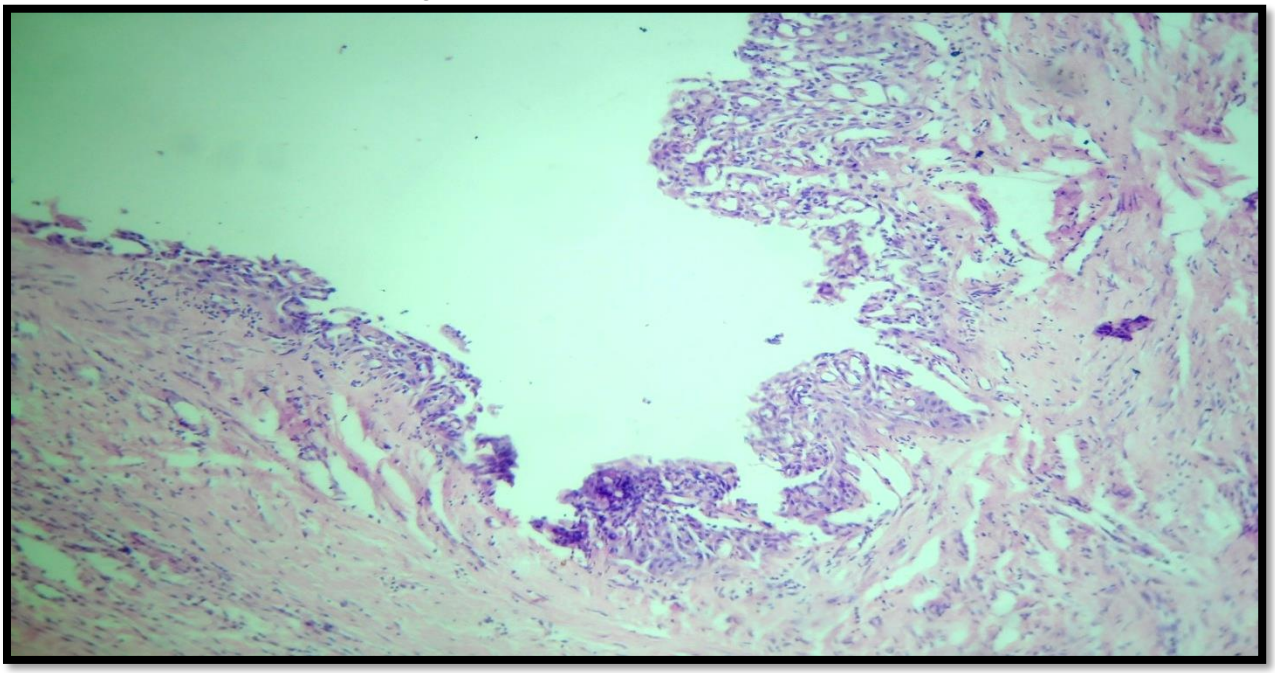


Figure 4: H&E section shows Columnar epithelium with cilia and at places it resembled REE.

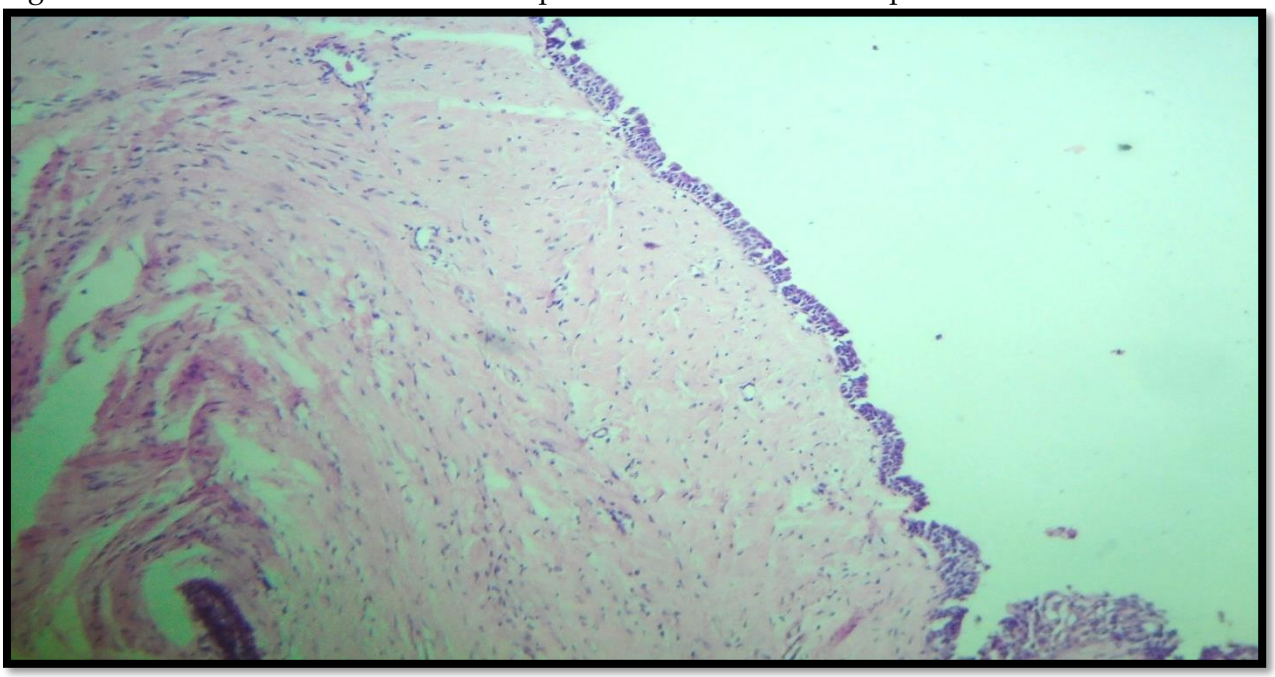


Figure 5: H&E section shows epithelium with glandular as well as goblet cell.

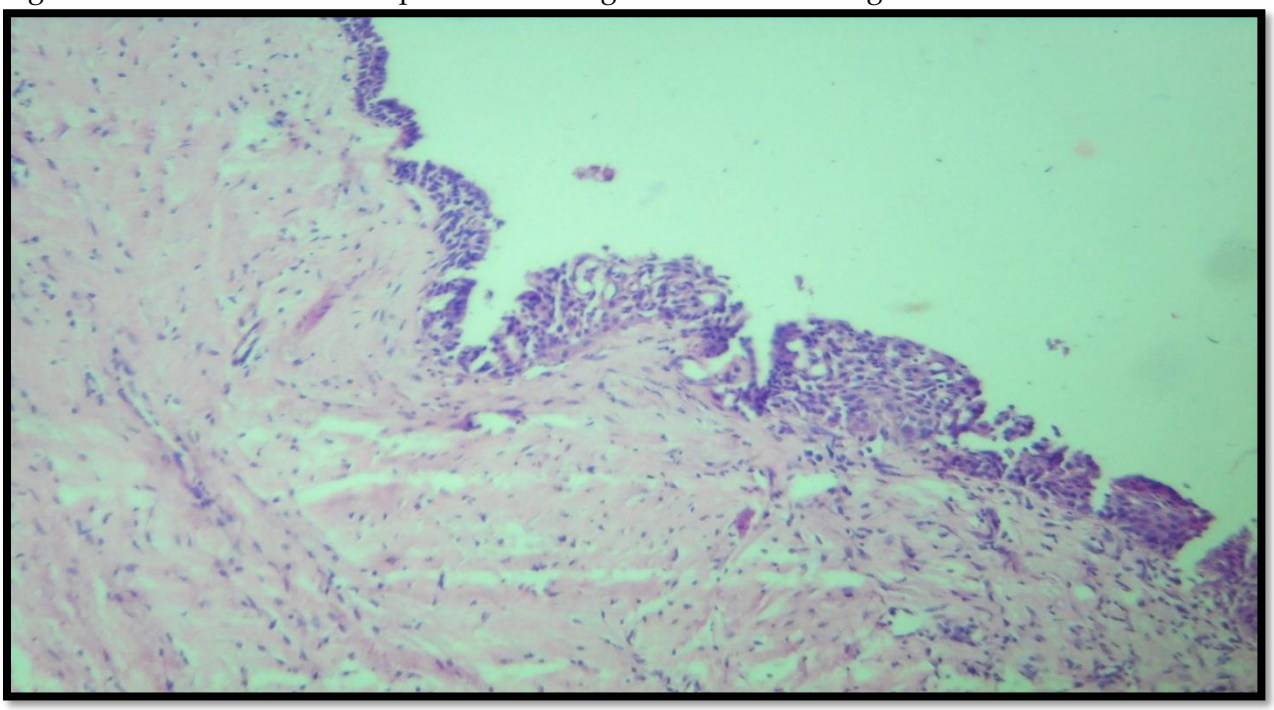
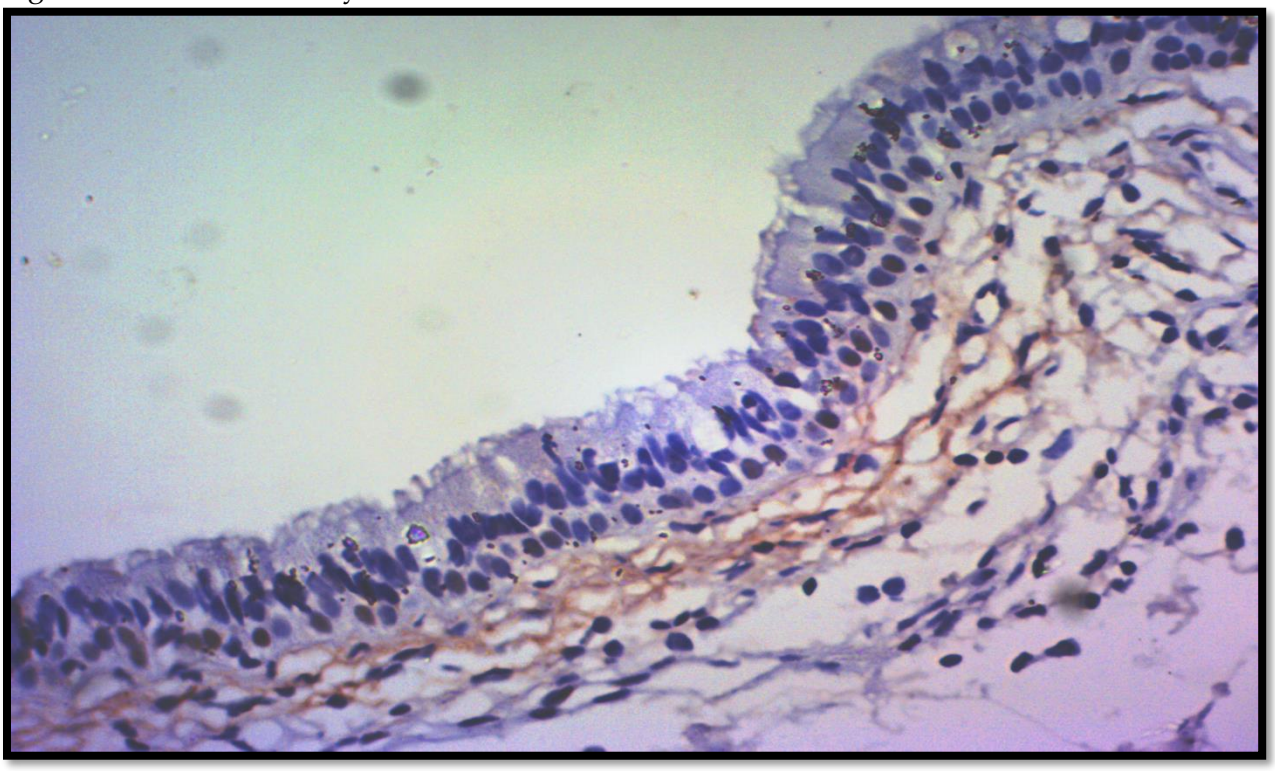


Figure 6: Immunoreactivity for Ki 67



References:

1. Gardner DG, Kessler HP, Morency R, Schaner DL. The glandular odontogenic cyst: An apparent entity. J Oral Pathol. 1988;17:359–66.
2. High AS, Main DM, Khoo SP, Pedlar J, Hume WJ. The polymorphous odontogenic cyst. J Oral Pathol Med. 1996;25:25–31
3. Monali Shah, Hemant Kale, Amena Ranginwala, Govind Patel. Glandular odontogenic cyst: A rare entity. J Oral Maxillofac Pathol. 2014 Jan-Apr; 18(1): 89–92.
4. Shafer'S Textbook Of Oral Pathology9 edited by: R. Rajendran, B.Sivapatha sundharam) 6 th ed.2009. pp254
5. Shear M. Cambridge: University Press; 1992. Cysts of the oral regions; pp. 72–4.
6. Michelle Manzini, Christian Deon, Liliam Dalla Corte,José Carlos Bertotto, Luciana Boff de Abreu, Glandular odontogenic cyst: an uncommon entity Braz J Otorhinolaryngol. 2009;75(2):320.
7. Pornpop Rattana-arpha, Ploi Yongvanijchit, Ekarat Phattarataratip. Glandular Odontogenic Cyst of the Mandible: A Case Report and its Potential Diagnostic Pitfalls. International Journal of Oral & Maxillofacial Pathology; 2013:4(2):34-39.
8. Amir Hosein Jafarian, Amin Rahpeyma, Saeedeh Khajehahmadi. Recurrent Glandular Odontogenic Cyst of Maxilla: a Case Report. Iranian Journal of Pathology ;2015:10 (2), 160 – 164.
9. Nair R G, Varghese I V, Shameena P M, Sudha S. Glandular odontogenic cyst: Report of a case and review of literature. J Oral Maxillofac Pathol 2006;10:20-3