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RADIOLOGIC-PATHOLOGIC CORRELATION

Glandular Odontogenic Cyst of Maxilla

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ABSTRACT

Glandular odontogenic cyst (GOC) is a rare developmental lesion that is considered a distinct entity because of its uncommon histopathological characteristics. It has morphological similarities to other lesions, which makes its diagnosis challenging for pathologists. It strikes distinct age groups, with an average patient age being 50 years. This lesion can involve either jaw, but the anterior region of the mandible is the most commonly affected area. It exhibits a tendency toward recurrence when conservative treatment is administered. It is believed that the low prevalence of GOC in the literature is due not only to its rarity, but also to the fact that its main characteristics are also found in other pathological entities. We report here radiologic-pathologic features of GOC of the maxilla in a 17-year-old female patient.

Key words: Glandular odontogenic cyst, maxilla, odontogenic cyst

INTRODUCTION

Literature reports only 50 cases of glandular odontogenic cyst (GOC),[1] a rare pathology also known as sialo-odontogenic cyst. It was first described by Gardner et al.[2] Clinically, the most common site of occurrence is the mandible (85%), especially in the anterior region. When in the maxilla, the lesion seems to affect mainly the tuberosity region. It occurs primarily in middle-aged people.[3] A slight predilection for men can be seen; female to male ratio being 19:28.[3] It is a slow growing, asymptomatic swelling, seen radiographically as a unilocular or multilocular radiolucency, with well-defined borders.[3-5] Histologically, GOC is characterized by a cyst wall lining of non-keratinized epithelium, with papillary projections, mucus-filled clefts and 'mucus lakes'. Cuboidal basal cells are also seen, sometimes vacuolated.[6,7] It is therefore seldom suspected on clinical and radiological examination. Some researchers believe that GOC is often misdiagnosed because of the overlap of its histological features with other odontogenic cysts, such as botryoid or lateral periodontal cysts or central low-grade mucoepidermoid carcinoma.

Our patient was a 17-year-old female with a history of pain and swelling in the right maxillary region for 8 months. On extraoral examination, facial swelling with a size of 4 x 6 x 3 cm was present on the right maxillary sinus region and was nontender with normal overlying mucosa [Figure 1]. Correlating with the history and clinical examination, panoramic radiograph was advised. This revealed a well-defined unilocular radiolucency in the right maxillary sinus with a posteriorly impacted third molar [Figure 2].

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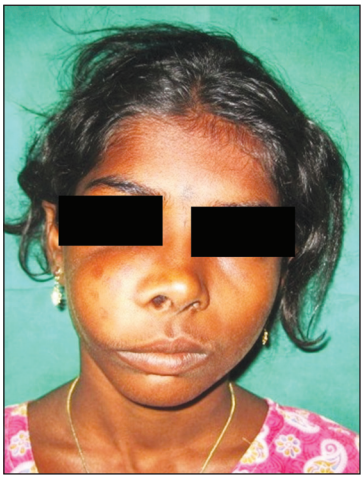

Figure 1: Extraoral photograph showing well-defined swelling on right side of the face causing gross asymmetry.

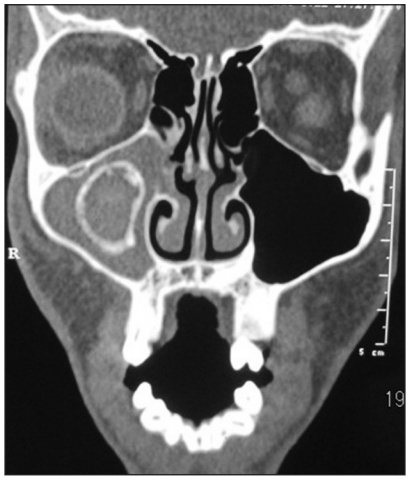

Figure 3: CT coronal section of the skull showing well-defined unilocular lesion in the right maxillary sinus confined within the boundaries of maxillary sinus.

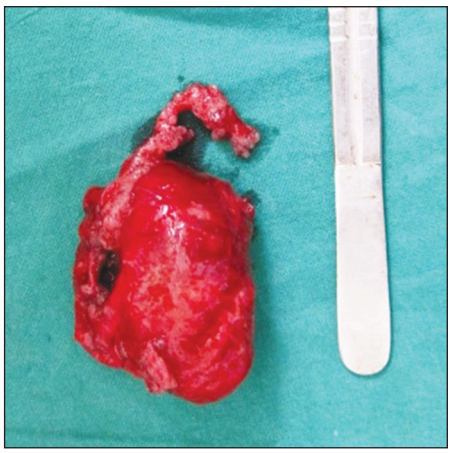

Figure 5: Excised lesion showing the cystic lining.

CT scan of the right maxillary sinus showed well-defined unilocular radiolucent lesion with impacted third molar confined within the boundaries of the maxillary sinus [Figure 3]. Aspiration of the lesion was performed to rule out vascular, cystic or mass lesions. Straw-colored aspirate fluid was obtained. Provisional diagnosis of calcifying epithelial

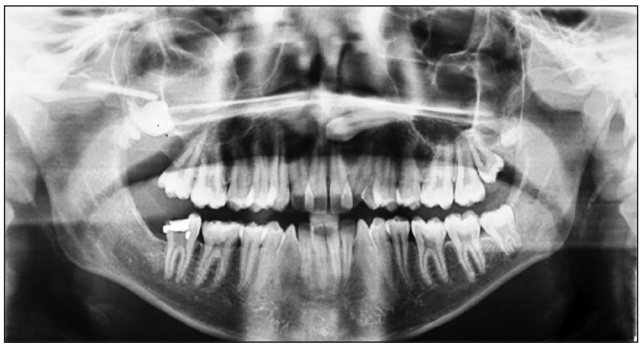

Figure 2: Panoramic radiograph showing well-defined unilocular radiolucent lesion in the right maxillary sinus.

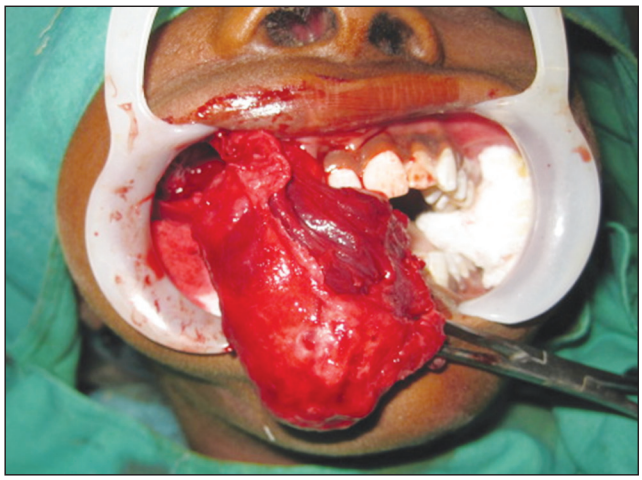

Figure 4: Intraoperative photograph showing the lesion which is still attached to the floor of the maxillary sinus.

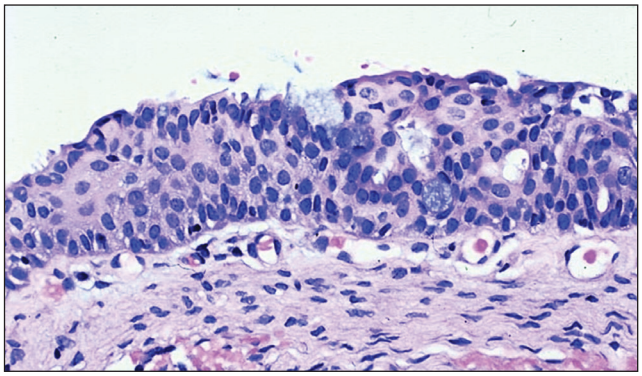

Figure 6: Parakeratinized squamous epithelial lining exhibiting cuboidal and columnar cells with numerous goblet cells and foci of epithelial cells showing eosinophilic material resembling mucin (Hemotoxylin and Eosin Stain, 40x magnification).

odontogenic cyst, odontogenic keratocyst, adenomatoid odontogenic tumor, and dentigerous cyst was made. The patient was sent for surgical exploration under local anesthesia and the lesion was enucleated using Caldwell-luc approach and the entire lesion was excised along with the impacted third molar [Figures 4 and 5]. The cavity was closed primarily after aggressive curettage. The excised tissue was sent for histopathological examination. The report revealed parakeratinized squamous epithelial lining exhibiting cuboidal and columnar cells with numerous

goblet cells and foci of epithelial cells showing eosinophilic material resembling mucin. Underlying connective tissue showed few chronic inflammatory cells infiltrate, predominantly lymphocytes, suggestive of GOC [Figure 6]. Patient was followed up for 1 year with no evidence of recurrence.

RADIOLOGICAL FEATURES

GOC is a rare lesion with a frequency rate of only 0.012-1.3% of all jaw lesions. It does not differ from the other jaw bone cysts in the typical radiological projections and that is why the diagnosis of the dentigerous cyst, odontogenic cyst, radicular and the keratocyst is made on the basis of the radiographical examination and in the case of the multilocular cyst, the picture may incline toward considering ameloblastoma.[7] Most commonly, GOC shows unilocular radiolucency with well-defined sclerotic borders with cortical expansion along with tooth displacement and root resorption. In the absence of extensive bony destruction or displacement or resorption of teeth, the radiographic presentation is similar to the more common odontogenic lesions, so that GOC is often not suspected.[8]

HISTOPATHOLOGICAL FEATURES

Histopathological examination revealed cystic lesion with luminal epithelium and surrounding connective tissue. The cystic space was lined by stratified non-keratinized squamous epithelium. The surface of the epithelium was composed of cuboidal and columnar epithelial cells with areas of the surface showing ciliated cells. In addition to the glandular structures, regions of the lining epithelium were composed of a uniform layer of stratified squamous epithelium with a hyper chromatic, palisaded basal cell layer.[9] The underlying connective tissue consisted of densely fibrous tissue with no inflammatory cells with smooth epithelial to connective interface.[10] Our patient had similar histopathological features suggestive of GOC.

CONCLUSION

In conclusion, GOC being a rare occurrence in maxilla, it is important to consider both radiological and pathological features, for a possibility of such a cyst cannot be ruled out while making differential diagnosis.

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