Case report

Glandular odontogenic cyst coexisting with actinomycosis: A rare case report

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ABSTRACT

Jaw lesions such as radicular cyst, odontogenic keratocyst, osteomyelitis associated with Actinomyces have been reported by many authors in the literature. Actinomycotic colonization in developmental cysts like GOC is extremely rare. So far in the literature search, the previous reports have not described actinomyces co-existing with GOC to our knowledge. Herein, we report a rare case report of GOC coexisting with actinomycotic colonies and pathogenesis of actinomycosis associated with odontogenic cyst.

Keywords: Glandular odontogenic cyst; odontogenic cysts; developmental cysts; actinomycosis.

INTRODUCTION

Glandular odontogenic cyst (GOC), also known as a 'sialo-odontogenic cyst', was initially described by Padayachee and Van Wyk in 1987. Both histopathologically and clinically, it is a rare and unique kind of developmental odontogenic cyst (1). The name 'glandular odontogenic cyst' was suggested by Gardner *et al.,* since the epithelial lining of this cyst was odontogenic in origin (2). They also described the histological characteristics and biological behavior of GOC. Due to its aggressive development pattern, the name 'polymorphous odontogenic cyst' was coined by High *et al.,* (3). Correct diagnosis is very important in this lesion, as it shows frequent recurrence. Hence, it needs more radical surgery and longer follow-up.

Actinomyces isralie, an anaerobic, gram-positive, filamentous bacterium, is the source of the chronic bacterial disease actinomycosis. Actinomyces is a common part of the oral microbial flora with a low pathogenicity, but when it invades the subcutaneous tissue, it becomes pathogenic. Actinomyces-related jaw infections have previously been documented in the past, including osteomyelitis and radicular cysts. However, actinomycotic colonies are discovered to be uncommon in developing cysts (4).

We are emphasizing a unique and uncommon example of a glandular odontogenic cyst that also had actinomycosis in this case report.

Case report

Swelling on the right side of the face was the main complaint of an 84-year-old female patient visiting a dental office.

Upon extra-oral inspection, the area around the top lip showed some slight facial asymmetry. The swelling was soft in consistency. The right upper labial vestibule had a diffuse obliterating edema, according to an intraoral examination. Right maxillary lateral incisor tooth number 12 had an accompanying root stump that was not essential. A right anterior maxilla lesion that was clearly radiolucent was seen on an intra-oral periapical radiograph (Fig.1). Periapical cyst was provided as the tentative diagnosis following both the clinical and radiographic investigation. The patient was further advised for biopsy. An excisional biopsy was performed with respect to the upper right front tooth region (12) and the tissue was sent to the laboratory for histopathological examination.

Fig. 1: Periapical radiograph showing radiolucent lesion associated with 12.

A cystic cavity was found to be bordered with non-keratinized stratified squamous epithelium of varied thickness after routine tissue processing and histopathological examination. In Fig. 2, the epithelium exhibits glandular organization and goblet cells that produce mucus. Some areas of lining epithelium are thin and few areas exhibit plaque like thickening. The cystic epithelium also shows hob-nailing of the nucleus (Fig. 3). Few characteristic basophilic clumps of filamentous actinomycotic colonies, exhibiting sun-ray patterns, were found in the lumen of the cyst (Fig. 4). Eosinophilic clubbing is also evident. The bacterial colonies are surrounded by acute and chronic inflammatory cells, predominantly lymphocytes and plasma cells. PAS staining was done

to confirm the colonies and the clear cells in the epithelium (Fig. 5). Considering the clinical, radiographical and histopathology, definitive diagnosis was given as glandular odontogenic cyst with actinomycosis.

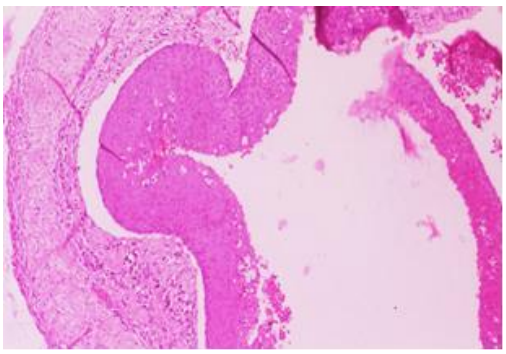

Fig. 2: H&E-stained non-keratinised epithelium exhibiting glandular and goblet cells.

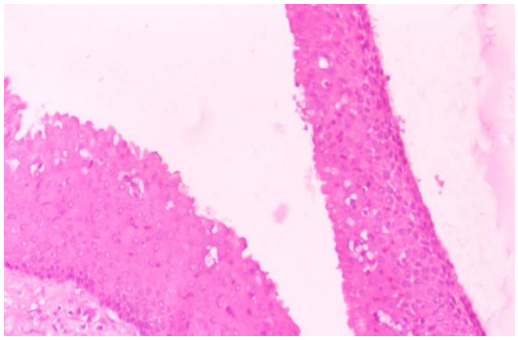

Fig. 3: H & E- stained section showing hob-nailing of the epithelium.

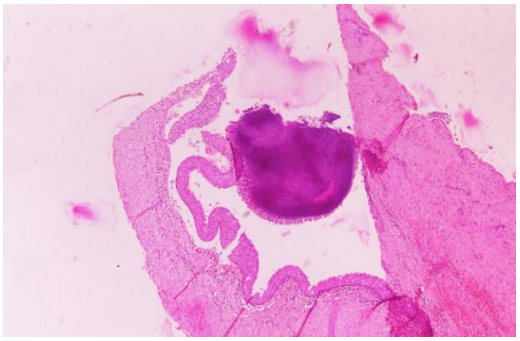

Fig. 4: H&E-stained section showing actinomycotic colony in the lume

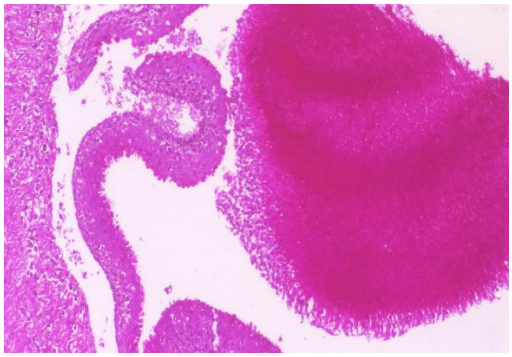

Fig. 5: PAS-stained section showing actinomycotic colony and PAS positive clear cells

DISCUSSION

A rare kind of developmental odontogenic cyst that emerges in the jaws is the glandular odontogenic cyst. Magnusson *et al.*, (5) found that the histological requirements of GOC were met by just 0.012% of the cysts visible under a microscope in the oral cavity. According to a thorough examination of the available literature, the differential diagnosis of GOC may resemble a broad clinicopathological spectrum, ranging from a lateral periodontal cyst to a lethal malignant tumour like central mucoepidermoid carcinoma. All other inflammatory cysts were ruled out in this case, and glandular odontogenic cyst was diagnosed since it met the histological criteria established by Kaplan *et al.*, (6).

It is unusual for actinomycosis to coexist with intra-bony developmental odontogenic cysts. Actinomyces and the relationship between odontogenic cysts can be characterized in two distinct ways: (i) Actinomyces invasion causes an apical inflammatory response that induces odontogenic epithelium and cyst development. (ii) The growth of actinomyces in the haemorrhagic material that fills the cystic cavity (7). Actinomyces-related periapical lesions either had a history of soft tissue damage or prior endodontic treatment. Actinomyces infection was seen in periapical lesions at a frequency of 0.4% to 1.8% (8). Kaplan *et al.*,'s study indicated that the incidence of actinomyces-associated periapical lesions such radicular cyst and periapical granuloma was 12.3% and in dentigerous cyst was 5.7% (9).

Actinomycosis is a persistent and aggressive bacterial disease with the capacity to invade and destroy bone. This filamentous bacterium forms a radiating, basophilic centre with an eosinophilic periphery. The colonies of actinomyces are encircled by neutrophils, polymorphonuclear leukocytes and granulation tissue (10). The commonly used special stains to demonstrate the organisms are Periodic Acid Shift (PAS), Gram, and methenamine silver stains. In the present case, we can see an inflammatory reaction found around the bacterial colonies suggesting a true infection (9). If an inflammatory reaction is not found around the colonies, then it is considered inert floater bacteria (11).

CONCLUSION

In conclusion, colonization of actinomyces may be seen in association with recurrent pathologic lesions of the jaws such as developmental and inflammatory odontogenic cysts. However, the connection between actinomycosis and a glandular odontogenic cyst has not yet been documented in literature.

CONFLICT OF INTEREST

The authors have no conflicts of interest to declare.

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