Case Report

Odontogenic Keratocyst with Diverse Differentiation – A Case Report

Imran Mohtesham[1], Sudeendra Prabhu[1], Maji Jose[1], B.H. Sripathi Rao[2]

[1]Department of Oral Pathology and Microbiology, [2]Department of Oral and Maxillofacial Surgery, Yenepoya Dental College, Yenepoya University, Mangalore, Karnataka, India,

Abstract:
Odontogenic keratocyst (OKC) is a developmental odontogenic cyst known for its specific clinical feature and histological architecture. Variation in its epithelial lining has been widely observed and reported. The cystic lumen in OKC is invariably lined by squamous epithelium. Metaplasia is known to be exhibited by the lining epithelium of OKC. Various metaplasia and degeneration observed in the OKC are mucous cells, ciliated cells, para and/or ortho keratinisation and hyaline bodies
Key words: Odontogenic keratocyst, Mucous metaplasia, Keratocystic Odontogenic tumor.

Corresponding author: Dr. Imran Mohtesham, Assistant Professor, Department of oral pathology and microbiology, Yenepoya Dental College, Yenepoya University, Nithyananda nagar, Mangalore-575018, Karnataka, India, Email: himohtesham@yahoo.com

This article may be cited as: Mohtesham I, Prabhu S, Jose M, Sripathi Rao BH. Odontogenic Keratocyst with Diverse Differentiation – A Case Report. J Adv Med Dent Scie Res 2014;2(4):93-96.

Introduction

Odontogenic keratocyst (OKC) is a developmental odontogenic cyst known for its specific clinical feature and histological architecture. Variation in its epithelial lining has been widely observed. The cystic lumen in OKC is invariably lined by squamous epithelium.[1] Metaplasia is known to be exhibited by the lining epithelium of OKC. Various metaplasia and degeneration observed in the OKC are mucous cells, ciliated cells, para and/or ortho keratinisation and hyaline bodies.[2,3] Hitherto very few cases of mucous metaplasia have been reported in the literature. We hereby report a case of OKC with mucous metaplasia in the lining epithelium.

Case Report

A fifty six years old female patient reported to the department of oral and maxillofacial surgery with a swelling of size 10cm x 3cm extending from angle of mandible to the symphysis with a duration of one month. On intra oral examination there were root stumps in relation to the right mandibular canine and premolars. On radiographical examination the lesion showed a well defined unilocular radilucency extending from the angle of the mandible to the other side canine. A clinical diagnosis of odontogenic keratocyst was made. The lesion was excised and was sent for histopathological examination.

On gross examination the lesional tissues was 5cms x 1.5cms with a cystic cavity which was soft in consistency. Routine hematoxylin and eosin staining, PAS and Alcian blue staining was done.

Histopathology

On histopathlogical examination the lesional tissue exhibited 5-7 cell thick parakeratinized odontogenic epithelial lining with surface corrugation (Figure 1A) in one area. But most of the sections with multiple bit

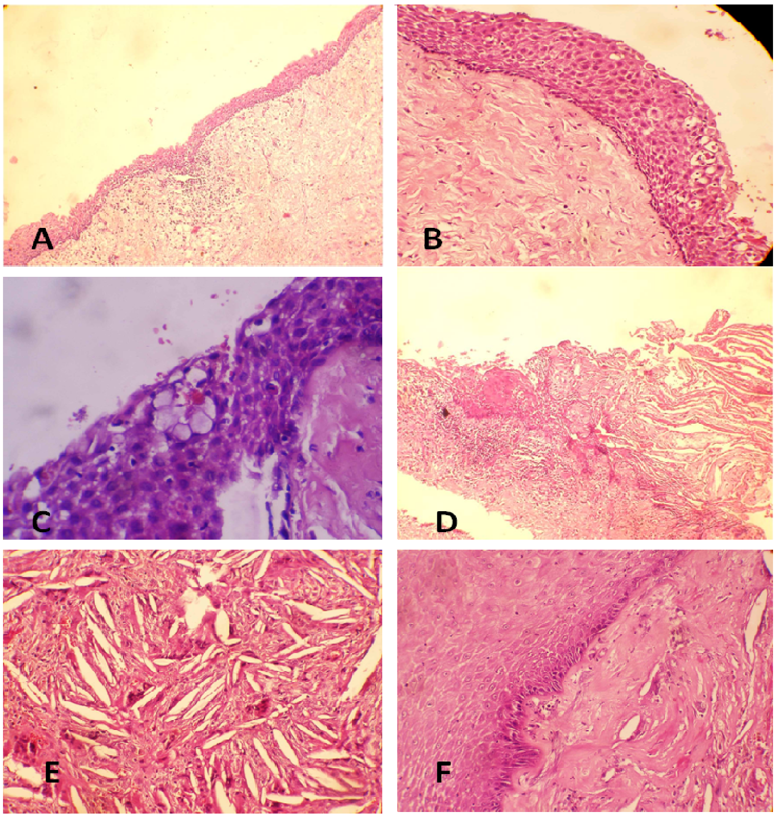

Figure 1 : Cystic lining with diverse histologic features

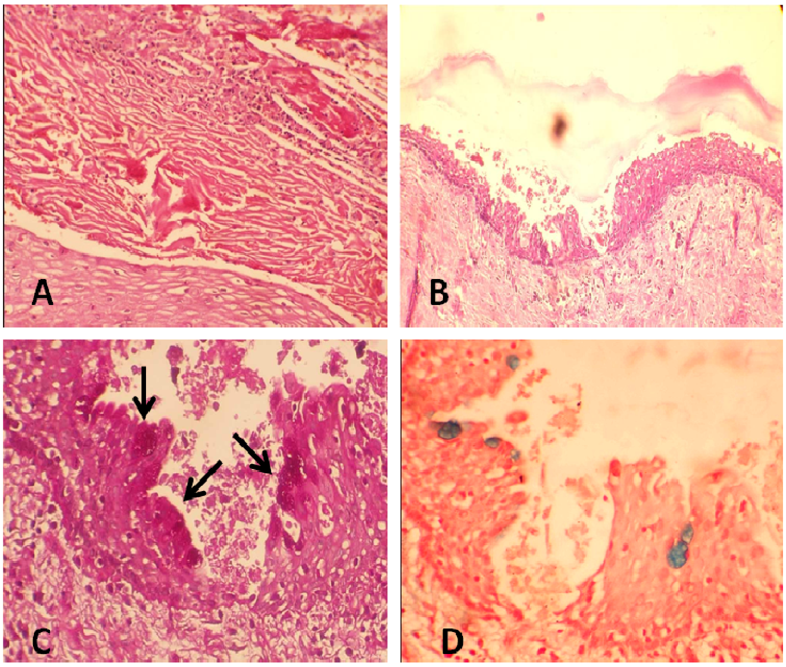

Figure 2: Cystic lining with hyaline bodies, cilia and special stains (PAS and Alcian blue)

evaluation showed proliferating non-keratinized stratified squmous epithelial lining with clear cells (Figure 1B). The Cystic lining showed clusters of mucous cells resembling glandular acinus (Figure 1C). Few areas in the section showed proliferation of cystic lining in arcading pattern, infiltrated with chronic inflammatory cells (Figure 1D) and cholesterol clefts with giant cells (Figure 1E). The section also revealed the proliferation of epithelium with polyhedral cells with abundant eosinophilic cytoplasm and prominent desmosomes resembling squamoid cells. The sub-epithelial haylinization was also evident (Figure 1F) in the section. Presence of hyaline bodies (Figure 2A) and Cilia (Figure 2B) were also evident. Clear cells showed positivity to PAS (Figure 2C) and Alcian blue (Figure 2D)

Discussion

OKC is a developmental odontogenic cyst with unique characteristics. OKC, first described by Philipsen in 1956, differs from other cyst as it shows more aggressive biological behavior and is known for its high recurrence rate owing to its high mitotic count and high epithelial turnover. WHO in 2005 has changed the term OKC to Keratocystic Odontogenic tumor (KCOT) on account of the above mentioned reasons.[4]

OKC has slight male preponderance unlike in our case which was a female patient, and usually seen in second and third decade of life.[5,6] Few reports show its occurrence in fifth and sixth decade.[1,7] However our case occurred in sixth decade. Sixty six percent of OKCs are seen in posterior mandible commonly involving molar region and vertical ramus. The present case showed the presence of lesion extending from the angle of the mandible to the symphysis.

Clinically OKC generally is associated with swelling, pain and discharge with aggressive growth and invading into adjacent tissues. Radiographically OKC usually present with unilocular or multilocular radiolucency with well defined and scalloped borders.[5] The histopathologic examination of an OKC reveals fibrous wall lined by epithelium with a thin layer of stratified squamous epithelium. This epithelium has a basal layer six to eight cells thick and a lining of flattened keratotic epithelial cells. The formed keratin lines the luminal surface of the epithelial cells in a slightly wavy or corrugated pattern.

The present case was in consistent with features mentioned in the literature. In addition our case also showed the presence of mucous cells in the epithelial lining. The presence of mucous cells in the epithelial linings of radicular and dentigerous cysts is well documented [9] but the presence of mucous and ciliated cells in the OKC is rare. It has been previously suggested that epithelial inclusions in the region of the mandibular third molars might represent pluripotentiality.11 In view of the multipotentiality of the odontogenic epithelium around the mandibular third molars; it is possible that it would have the capacity of inducing the formation of cysts with both the squamous and mucous metaplasia. The possible pathogenic mechanism of this case would appear to be a reflection of the pluripotential character of the odontogenic epithelium.[10]

Conclusion

Mucous cells in the epithelial lining of intraosseuos odontogenic cyst could be because of the metaplastic in origin. But its cause and biologic function of this metaplsia is still not known and studies in this direction can open new vistas in the field of research.

References

1. Takeda Y, Oikawa Y, Furuya I, Satoh M and Yamamoto H. Mucous and ciliated cell metaplasia in epithelial linings of odontogenic inflammatory and

developmental cyst. J of Oral Science. 2005; 47(2): 77-81.

2. Kramer IRH, Pindborg JJ , Shear M. Histological typing of odontogenic tumor. 2nd ed. Springe-Verlag: Berlin:1992.
3. Gorlin RJ. Potentialities of oral epithelium manifest by mandibular dentigerous cyst. Oral Surg. 1957;10:271-184.
4. Cawson RA, Odell EW. Essentials of oral pathology and oral medicine. 6th ed. Churchill Livingstone: New York; 1998
5. Gustafson G, Lindhal B and Svensson A. the nevoid basal cell carcinoma. Sweden Dent J. 1989; 13: 131-139.
6. Morgan TA, Burton CC and Qian F. a retrospective review of treatment of OKC. J of oral maxillofac surg. 2005; 63: 635-638.
7. Shear M. Cyst of the oral region. 2nd ed. Wright PSG: Bristol; 1983
8. Brown RM. The OKC. Clinical aspects. Br Dent J. 1970; 128: 225-231.
9. Shear M. Secretory epithelium in the lining of dental cyst. J dent ASS South Afr. 1960; 15: 117-122.
10. Browne RM. The odontogenic keratocyst. Histological features and their correlation with clinical behavior. Br Dent J 1971;131:249-59.

Source of support: Nil
Conflict of interest: None declared