

Odontogenic Keratocyst in Right Maxilla of 15 Year Old Boy: A Case Report of not a Rare Lesion

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ABSTRACT

We present a case of odontogenic keratocyst involving the right maxilla in 15 years old boy. The histochemical of the lesion was analysed by haematoxylin & eosin stain and picosirius red stain. The expansile nature of the lesion was assessed by using picosirius red to categorize the fibrous wall of the cyst. The purpose of this case report is to stress the point that even though the diagnosis of classical odontogenic keratocyst is straight forward with histochemical study alone, it is prudent to profile the biology of each patient using less expensive methods before deciding the appropriate adjunctive surgical procedure, for proper rehabilitation of that particular patient as well as to avoid its recurrence in future.

Key words: Odontogenic keratocyst, picosirius red, polarization microscope.

INTRODUCTION

The odontogenic keratocyst is distinctive among various odontogenic cysts due to various factors such as distinct histological findings, aggressive clinical behaviour and increased recurrence rate. It is mostly seen in the 2nd-3rd decade of life with a slight female predilection.¹ It is seen predominantly in the mandible (around 78%) and mostly in the ramus-third molar area. Clinical presentation varies from being asymptomatic to pain, soft-tissue swelling, bone expansion etc.² Radiologically it is usually unilocular with well-developed sclerotic borders, however it mimics other lesions in most cases like periapical cyst.³ In the treatment respect, there is always a dilemma between conservative surgical excision and the resection of the lesion with adjacent bone, to prevent recurrence.⁴ So many researches have been done on the molecular biology and genetic aspect of odontogenic keratocyst using histochemical,

immunohistochemical, genetic engineering etc, to understand its pathogenesis and substantiate the term called 'keratocystic odontogenic tumour'.⁵

Using eosin and haematoxylin stain, changes in typical characters of the lining epithelium of odontogenic keratocyst are assessed. Special stain, picosirius red is used to stain collagen present in the fibrous wall subsequently to assess the influence of inflammation on the lining epithelium and fibrous wall as well as to categorize the collagen fibres to predict the expansile nature of the cyst.⁶ Hence in this case report we have created a separate profile for the patient by doing immunohistochemical and special stains to decide the subsequent treatment option for this particular patient.

Case report

A 15 year old boy came to our department with the chief complaint of tooth pain on the right side of the upper jaw for past 2 months. Extra oral

examination was unremarkable. On intraoral examination no swelling detected around 14 region. Tooth 14 was vital. Radiographically a well demarcated radiolucency without corticated margin is found periapical to 14. A full thickness flap is elevated and a cystic lesion of 2 cm x 1.5 cm was seen perforating the palatal cortical plate. The lesion was enucleated and submitted for histological examination. (Fig.1) The provisional diagnosis of periapical cyst was considered.



Fig.1: Shows grossing picture of cystic lesion (cut-surface)

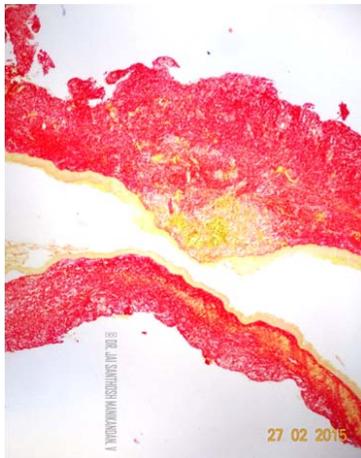


Fig. 3: Shows cystic wall with inflammatory areas in left side and classical cystic lining epithelium of odontogenic keratocyst in the right side of the picture (low magnification- 4 x / Picrosirius red stain)

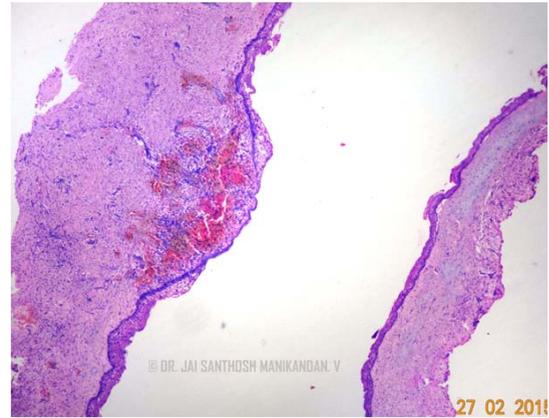


Fig. 2: Shows cyst lining with inflammatory areas (in left side of picture) and classical cystic lining epithelium of odontogenic keratocyst (in the right side of the picture) (low magnification- 4 x / Haematoxylin-Eosin stain)



Fig. 4: Shows fibrous wall with inflammatory areas in left side (seen as greenish- Yellow colour) whereas fibrous wall under classical lining epithelium (seen as yellowish – orange colour) (low magnification- 4 x / Picrosirius red stain seen under polarization microscope)

diagnosed as odontogenic keratocyst. (Fig.2,5) Section stained with picosirius red (Fig.3,6,9) are studied with polarization microscope. (Fig.4,7,10) Most of the areas of fibrous wall present under odontogenic keratocyst are seen as yellowish-orange colour (Fig.4,7) while fibrous wall with inflammatory foci seen as greenish-yellow colour. (Fig.4,10) The patient continues to return for periodic clinical and radiographic follow-up as instructed. Thus far, no evidence of recurrence has been noted during a period of two year, post treatment

DISCUSSION

Odontogenic keratocyst is a distinctive histopathological type of developmental odontogenic cysts.⁷ In 1956, it was first described as odontogenic keratocyst by Philipsen. Finally it was renamed by Philipsen as keratocystic odontogenic tumor and reclassified under the lesions of odontogenic neoplasm according to odontogenic tumours classification given by World Health Organization in 2005.⁸ Remnants of basal lamina and basal cells proliferating from the

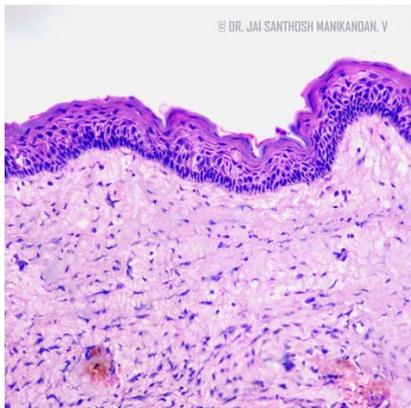


Fig. 5: Shows corrugated parakeratinized stratified epithelium, 6-10 cell thickness, palisaded basal layer with hyperchromatic nuclei, absence of rete process (magnification- 40 x/ Haematoxylin-Eosin stain)

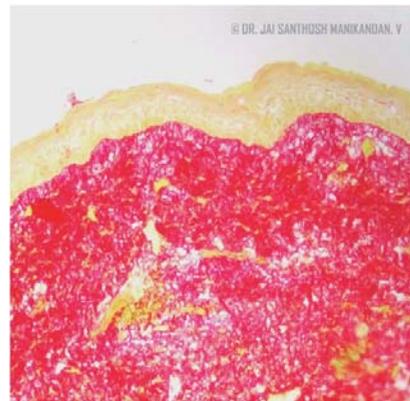


Fig. 6: Show that epithelium was not stained, whereas fibrous wall stained red (magnification- 40 x/ Picosirius red stain)

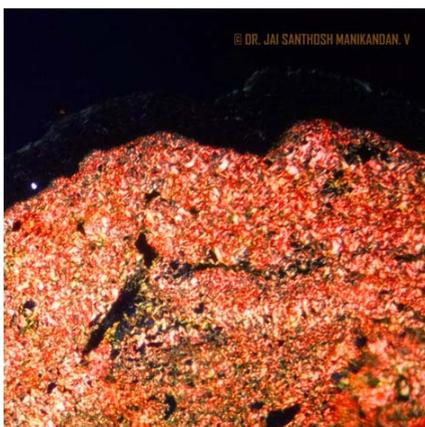


Fig. 7 : Shows fibrous wall under classical epithelium seen as yellowish-orange Colour (magnification- 40 x/ Picosirius red stain seen under polarization microscope)

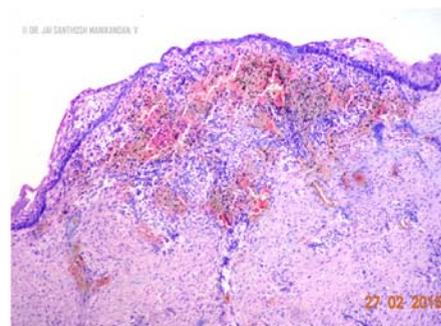


Fig. 8: Non-keratinized stratified lining epithelium. The underlying fibrous wall shows dense infiltration of chronic inflammatory cells. (magnification- 40 x/ Haematoxylin-Eosin stain)

overlying epithelium are two main sources of origin for odontogenic keratocyst.⁹ Important step in pathogenesis of odontogenic keratocyst is the mutation of gene PTCH a tumour suppressor gene and supported its tumour like behavior.¹⁰

Odontogenic keratocyst shows more number of cases are seen between 40-50 age groups.² Odontogenic keratocyst was more frequent in male compared to female.⁹ Mandibular molar-ramus area is the most common site for odontogenic keratocyst.¹¹ In case of maxilla, most frequently involved site is third molar area and next is cuspid region.¹ Odontogenic keratocyst will be asymptomatic in most cases. The lesion are diagnosed later in life and usually attain larger size as they grows mainly through medullary space of the jaw in antero-posterior direction.¹² That usually odontogenic keratocyst which are smaller in size are asymptomatic and discovered only in radiographic examination. Pain and swelling are mostly seen with larger lesions only.² In maxilla, it may extend up to maxillary antrum.¹³

Smaller lesions are usually seen as radiolucent lesion with well-defined lamina dura.¹⁴ That 40% of odontogenic keratocyst are seen as unilocular lesions adjacent to crown of the teeth.¹ Radiographic features of scalloped margin and multilocular appearance seen in odontogenic keratocyst are indicative, but these features are also seen in other odontogenic lesions.¹⁵ Histological features includes 5-8 rows of thin epithelium without rete pegs, predominant

parakeratosis surface layer with wavy appearance, a basal layer made up of columnar or cuboidal cells with palisaded, thin layer of vacuolated cells of stratum spinosum and a fibrous capsule which is thin and devoid of inflammatory cell infiltration.¹⁶ The basal layer is characterized by columnar or cuboidal cells showing palisaded hyperchromatic nuclei showing reversal of polarity. The surface epithelium is usually corrugated. The cystic lumen is filled with desquamated squamous or necrotic material.¹⁷ Fibrous capsule which is usually thin with decreased cellularity often separated by stroma. This stroma is rich in mucopolysaccharide with rare infiltration of inflammatory cells like lymphocyte and monocytes. If there is inflammatory infiltration, the adjacent epithelium thickens and develops rete process. The weak attachment between the epithelium and the connective tissue result in their separation. The cyst wall is mostly seen collapsed and folded.⁹

Described the presence of satellite cyst within the wall, which may be the reason for its higher recurrence rate.¹⁸ Odontogenic keratocyst showed higher recurrence rate ranging from 5% - 62% with most cases encountered in posterior body and ascending ramus area of mandible.² Treatment modalities of odontogenic keratocyst which includes surgical treatment characterized by enucleation with peripheral ostectomy.⁹

Using picosirius red stain, collagen structure can be studied under polarization microscope. under them, collagen takes different

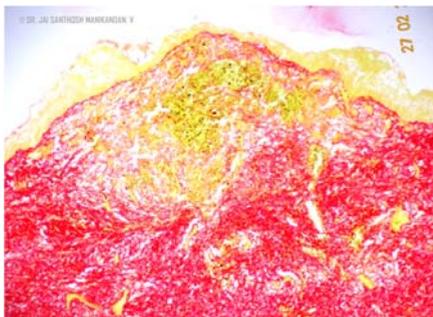


Fig. 9: Fibrous wall shows dense infiltration of chronic inflammatory cells. (magnification- 40 x/ Picosirius red stain)

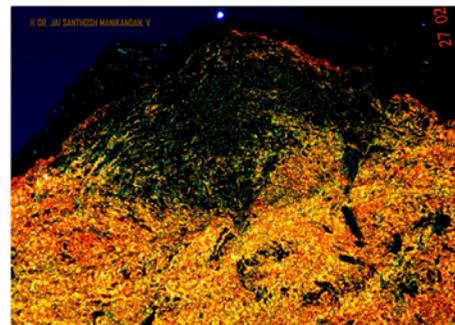


Fig.10: Fibrous wall shows greenish-yellow fibres in area showing infiltration of chronic inflammatory cells.(magnification- 40 x/ Picosirius red stain seen under polarization microscope).

based on fibre thickness, packing and arrangement of collagen fibres.¹⁸ Under polarization microscope, thin normal collagen fibres as well as poorly packed fibres are green to greenish yellow, whereas thick fibres as well as well packed fibres range from yellowish orange through orange to red.^{19,20}

In our case fibrous wall of odontogenic keratocyst showed predominantly yellowish orange fibre, which in correlation with finding found by Aggarwal P *et al.* This indicates that predominantly tight packed fibres are present in the fibrous wall. Similar fibres are also seen in stroma of odontogenic tumours like ameloblastoma.²¹ Hence it can be concluded that aggressive behaviour of odontogenic keratocyst is also derived from its well-organized stroma. But our case result contradicts with the results given by Hirshberg A *et al.*,¹⁸ as he showed odontogenic keratocysts predominantly showed greenish yellow collagen fibres under polarizing microscope.

It also found that areas of fibrous wall corresponding to inflammatory cell infiltration showed greenish yellow fibres, hence it should be made up of predominantly thin and less organized fibres suggestive of procollagen or pathologic collagen. The presence of such fibres may be due to the dense infiltration of inflammatory cells in this cyst wall which releases abundant cytokines, endotoxins and lymphokines into the surrounding connective tissue as well as shows increased collagenase activity in the stroma. All these factors results in the degradation of the extracellular matrix.²² As most of the areas showed tightly packed fibrous wall, this case is considered as aggressive and followed up for recurrence. But in 2 years follow up, it is uneventful. Hence before doing expensive investigation, which consumes valuable time, it is better to do special stain like picrosirius red which is outdated but less technique sensitive. In doing so we are able to additional information regarding in aggressive nature and decide the treatment plan accordingly.

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