A Case Report

Conservative Management of Odontogenic Keratocyst in Posterior Maxilla of a Paediatric Patient – A Case Report

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Abstract

Odontogenic Keratocysts (OKCs) are the developmental odontogenic cysts of epithelial origin, which are rare in occurrence, benign in nature but locally aggressive more common in third decade of life, and contributes to around 19% of all jaw cysts, microscopically characterized by palisaded basal nuclei arrangement & para keratin keratinization. Due to its aggressive nature it needs to be enucleated along with removal of associated tooth and other adjunctive procedures. Here we report a case of OKC in a 11-year-old boy associated with unerupted 17,18 teeth extending up to right tuberosity area. Our treatment plan aimed to preserve shape, form and function of maxilla, along with normal growth of patient. Outcome was uneventful and lesion was conservatively managed. Also, a regular follow-up is advised to avoid any recurrence.

Keywords: Odontogenic Keratocyst, Keratocystic odontogenic tumor, FNAC, enucleation, conservative management

Introduction

Odontogenic keratocyst (OKC) is developmental cyst which is odontogenic in origin, benign in nature and presents with locally aggressive clinical behaviour including a high recurrence rate. The odontogenic keratocyst (OKC) was first coined by Philipsen in year (1956), which was later renamed by the World Health Organization (WHO) as a keratocystic odontogenic tumour (KCOT) due to its aggressive behaviour and high recurrence. PTCH (Protein patched homolog) gene mutation and association with Gorlin Goltz syndrome has been seen in cases of multiple OKC. In 2017 WHO has reclassified OKC as a cyst.[1] [2] Neville 2019 edition defines OKC as "a common developmental odontogenic cyst which accounts for 10% to 14% of all jaw cysts.[3]

Radiographically, odontogenic keratocyst presents as a unilocular radiolucency with well-developed sclerotic borders mostly. It may have multi-locular variant which is more common in association with syndromic cases.[4] Rarely any resorption or displacement of the associated tooth is seen. Large cystic lesion may cause expansion of the medullary cavity in bone.Histo-pathological examination helps in confirming the final diagnosis and differentiating it from other lesions like dentigerous cyst, unilocular ameloblastoma, and lateral periodontal cyst. Its characteristic microscopic features are basilar nuclear palisading pattern and keratin production (majorly Para keratin).[5]

OKC has aggressive behaviour and high recurrence tendency due to presence of satellite cyst, it was first pointed out by Pindborg and Hansen in (1963).[6] Recurrence is more common in posterior mandible particularly in posterior body and ascending ramus. Around 18% reduction in the recurrence potential can be noted, when enucleation is accompanied with other adjunctive measures.[7] A regular follow up and re-assessment using radiographic aids helps in identifying early re-occurring lesion.

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In the presented case, an OKC in a 11-year-old boy with pain and purulent discharge in association with the unerupted tooth i.r.t 17,18 and extending up to right maxillary tuberosity area has been reported. Here the provisional diagnosis, radiographic interpretation, report of the histopathological examination and conservative management of the cyst with minimal invasion has been discussed, thereby preserving the anatomical structures associated, without altering the normal growth of maxilla and areas associated.

Case History

In present case an 11-year-old boy reported with gingival swelling, pain and purulent discharge in upper right back tooth region which was sudden in onset with negative history of trauma, food impaction. Patient gave history of purulent discharge since last 1 month which was a greasy fluid, pale in colour with foul odour and no fever was associated. No nasal discharge or obstruction was seen. Medical history was negative for any systemic illness and any congenital disorder in family.

The extra-oral examination of patient revealed no swelling and facial asymmetry. On intra-oral examination gingival swelling was found in upper right back tooth region extending from distal aspect of 16 tooth region to right maxillary tuberosity area buccally, which was tender, non-fluctuant, hard in consistency and presence of purulent greasy pale coloured discharge was seen with no associated food impaction and caries of 16 tooth. Surrounding mucosa was red in colour. The swelling was approx. around 3 cm in size. [Figure 1 A, B, C, D, E]

Patient was advised for OPG which revealed unilocular radiolucency in relation to coronal portion of unerupted teeth 17, 18 extending posteriorly along the right maxillary tuberosity area with well-defined borders without any bony expansion along the medullary cavity, no root resorption or displacement was seen of associated tooth. A radiographic diagnosis of odontogenic keratocyst, was made.[Figure2]

Figure 1 A-E: Pre-operative intra oral photographs

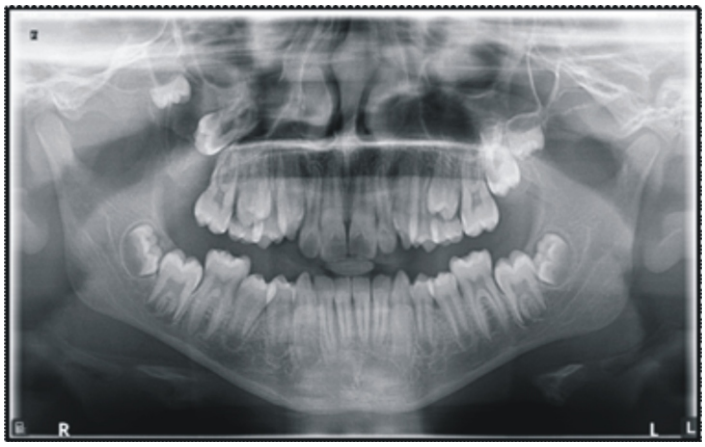


Figure 2: OPG showing unilocular radiolucency i.r.t 17,18

Further an FNAC (fine needle aspiration cytology) was performed; on aspiration a greasy pale straw-coloured fluid was found. With the above clinical findings, a provisional diagnosis of OKC was made.

As per treatment plan extraction of associated teeth i.e., 17, 18 were performed along with enucleation of the cyst under local anaesthesia. [Figure 3,4]

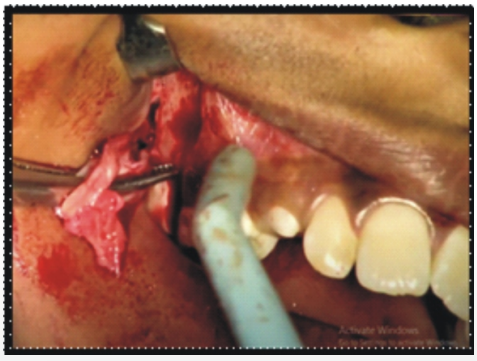


Figure3: Separation of cystic lining

Figure4:Extraction i.r.t 17

Curettage of the underlying area was performed to remove any remnants ofthe cystic lesion. [Figure 5]

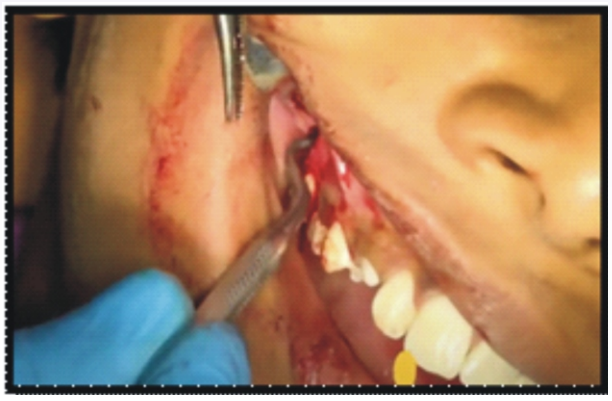


Figure 5: Curettage

Wound closure was performed after irrigation with normal saline with help of 3-0 silk suture. [Figure6]

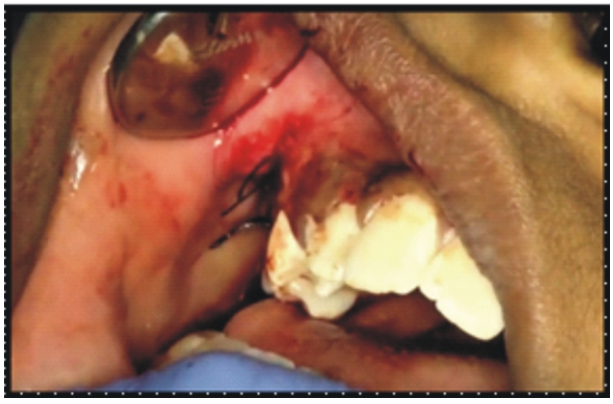
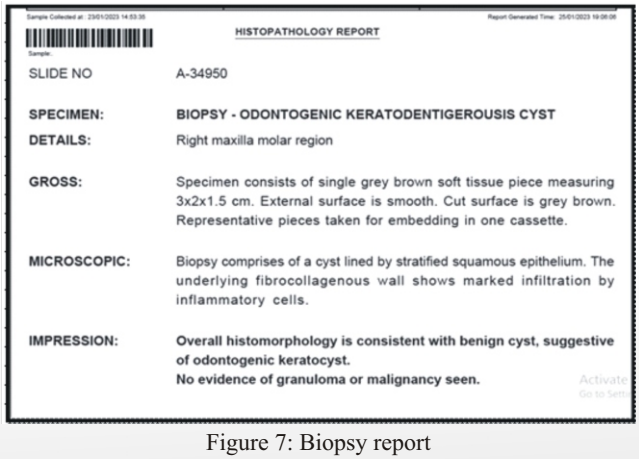


Figure 6: Sutures placed

The enucleated cyst along with its content was sent for biopsy. The biopsy revealed a cyst lined by stratified squamous epithelium with a dense fibro-collagenous tissue underneath infiltrated by inflammatory cells markedly. Overall histomorphology was consistent with benign cyst, suggestive of OKC without any evidence of granuloma or malignancy. [Figure7]



HISTOPATHOLOGY REPORT

SLIDE NO	A-34950
SPECIMEN:	BIOPSY - ODONTOGENIC KERATODENTIGEROUSIS CYST
DETAILS:	Right maxilla molar region
GROSS:	Specimen consists of single grey brown soft tissue piece measuring 3x2x1.5 cm. External surface is smooth. Cut surface is grey brown. Representative pieces taken for embedding in one cassette.
MICROSCOPIC:	Biopsy comprises of a cyst lined by stratified squamous epithelium. The underlying fibrocollagenous wall shows marked infiltration by inflammatory cells.
IMPRESSION:	**Overall histomorphology is consistent with benign cyst, suggestive of odontogenic keratocyst. No evidence of granuloma or malignancy seen.**

Figure 7: Biopsy report

Therefore, final diagnosis of OKC was made based on clinical findings, radiographic interpretation and histo-pathological examination. Patient was recalled for follow up and suture removal after 1 week. On follow-up, good healing with approximation of the tissues was seen along with wound closure and no associated pain and discomfort. Patient has been recalled for regular follow up every 3 months to assess any re-occurrences [Figure 8 A, B, C, D, E]

Figure 8[A –E]:Post-operative follow up intra-oral pictures

Discussion

An OKC is one of the common developmental odontogenic cysts with prevalent clinical and histologic lesion, benign in nature and locally aggressive. It originates from the dental lamina, or from the basal cell component. [8] Most common site of occurrence is mandible around 70%, especially in the third molar, angleand ramus areas followed by the maxillary third molar, mandibular premolar and maxillary canine area.[9] There are few factors which lead to aggressive behaviour and high reoccurrence of OKC thereby reclassifying it as KCOT.[10] It is more common in third decade of life and greater male predictability.[11]

Unilocular radiolucency with well-developed sclerotic borders without any displacement, resorption of adjacent tooth and expansion of medullary cavity is also seen less frequently. It may also present as a multilocular radiolucentlesion with a ratio of unilocular to multilocular varying from 3:1.12 to 1:1.3. Multiple OKC are frequently associated with Gorlin syndrome.[12]

Mandibular OKC recurrence is more common in posterior body and ascending ramus.[13]

OKC has been reported in present case in a young boy, affecting posterior of maxilla, presenting as a unilocular radiolucency i.r.t unerupted 17 and 18. Mostly it affects males in 3rd decade of life but here in the current case report OKC is found in an 11-year-old boy. On FNAC greasy whitish pale-yellow aspirate was found.

Conservative treatment plan was done with extraction of involved teeth 17,18 followed with enucleation and curettage of associated area. Lesion was managed with copious irrigation of normal saline, placement of dressing with final approximation of tissues by suture placement. Conservative treatment plan helps in maintaining the normal anatomical form and function of area.

Biopsy of the involved cystic lesion revealed cyst lined by stratified squamous epithelium and underlying fibro collagenous wall showed marked infiltration by inflammatory cells giving final impression of OKC. There was good healing and approximation of the tissues, resolution of swelling and no pain was present. Patient has been advised for follow up and re- evaluation every 3 months. Here in this case no reoccurrence of lesion was observed.

Conclusion

As OKC is aggressive developmental odontogenic cyst with high reoccurrence rate, OPG is useful in interpreting the extent, position of cyst, expansion of the medullary space nearby. Histopathological examination and FNAC are useful aids in confirmation and final diagnosis of cyst. Due to aggressive behaviour of OKC, enucleation of cyst along with extraction of associated unerupted tooth followed by curettage was performed to prevent any recurrence in future. A close followup and re-evaluation helps to evaluate any recurrence. The result of the presented case suggests that minimal intervention using conservative approach under local anaesthesia without osteotomy and re-sections yields faster healing and allows normal growth of the tissue in pre-adolescent child. Timely diagnosis and a close regular follow up help in preventing any further loss by monitoring any recurrences.

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