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Enigmatic Developmental Cyst: A Case Report of Maxillary Odontogenic Cyst

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ABSTRACT:
Odontogenic keratocyst is a benign intraosseous lesion with high recurrence rate. Due to its peculiar behaviour, unique characteristics of recurrence and disputed treatment modalities, this cyst carries a special attention to maxillofacial clinicians to evaluate its recurrence rate, many treatment modalities do exist with treating for Okc, but recently the topical application with 5 FU has gained attention for maxillofacial clinicians. With the aim of this novel medicament a case report of Okc which is managed with the application of 5FU and a follow up is been illustrated.

Keywords: 5FU (5- Fluorouracil), OKC (Odontogenic Keratocyst).

I. INTRODUCTION:

Odontogenic cyst is the commonest lesions of the oral cavity which are identified on clinical evaluation and routinely or accidentally through radiographic examination. Among the many odontogenic cysts keratocystic lesion represents a unique description because of their aggressive nature and recurrence rate as similar to tumors. Since the first description of Odontogenic keratocyst given by Philipsen in 1956. The name of okc change to kcot and finally back to Okc.[1, 12, 13] Odontogenic keratocyst which seems to originate from dental lamina in mandible and maxilla. The lesion is twice common in mandible than of maxilla the lesions occur before odontogenesis is complete. Maxillary odontogenic keratocyst are rare, mostly maxillary lesions are seen at cuspid region.

The main reason that could be recurrence for okc are:

- Higher level of cell proliferative activity in the epithelium
- Budding in the basal layer of epithelium
- Parakeratinization of the surface layer.
- Supraepithelial split of the epithelial lining
- Sub epithelial split of the epithelial lining.
- Presence of epithelial remnants or cell rests as well as daughter cyst.[1]

A wide range of treatment for treating okc has been proposed in the literature. According to the literature published in 1985 by Eyre and Zakrezewska , the following treatment modalities for okc:

❖ Enucleation
- With primary closure
- With packing
- With chemical fixation
- With cryosurgery.

❖ Marsupilazation
- only
- Followed by enuclation

❖ Resection[2]

The ideal treatment for okc remains controversisal till date there is no general agreement of opinion as to most pertinent treatment for Okc. According to Al.Moraissi et.al,

- Enucleation with cryotherapy.
- Marsupilization followed by residual cystectomy, Reported to have lowest recurrence rate when compared to the enucleation or marsupilization in his 33 metaanalysis studies.[3]
- Enucleation followed by chemical fixation with carnoy's solution of 3 min application onto the nerve was defined by Frerich et.al.[4] However chloroform was vomited out from the carnoy's solution due to its carcinogenic potential, with the application of modified carnoy's solution the recurrence rate of Okc are potential. Due to the recurrence rate a novel approach for treating Okc

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was gained by topical application of 5 FU. The genes that are correlated to OKC are PTCH2 and SUFU. The PTCH2 genes are mapped to chromosome 9q22- q31. The PTCH which is majorly a tumor suppressor gene which is an important molecule for Hedge hog (Hh) signaling pathway. Normally the PTCH forms a receptor complex with the oncogenic SMo(Smoothened) for the SHH(sonic Hedgehog) ligand. The newer trend for the management of okc is based on its molecular pathogenecity as the Okc have similarities with basal cell carcinoma. These SMo genes play a role in development of OkC suggesting of sonic hedge hog antagonism. With the similarities between okc and basal cell carcinoma at molecular level a newer drug 5 FU which is known to induce apoptosis bt inhibiting SHH in hepatocellular carcinoma cell.[1, 6,7,8,9] The present case report presents with topical application of 5 FU for keratocystic lesion.

CASE PRESENTATION:

A 30 yr old female patient reported to our clinic with a chief complaint of swelling over right upper jaw region for the past 2 months .On examination ,patient had no relevant medical history intraorally the swelling was examined measuring of about 3x2 cm which is causing vestibular obliteration. The swelling was firm, soft in consistency. Extraorally the swelling extend from ala of nose to right zygoma region.

INVESTIGATION:

- BLOOD INVESTIGATION
- FNAC
- OPG
- BIOPSY

All the blood investigation was performed and no abnormality detected.

1) FNAC was performed and revealed dirty brown coloured fluid. (Fig.1)

Fig.1 FNAC (Fine Needle Aspiration cytology).

2) OPG revealed a unilocular radiolucent lesion crossing midline extending from 16 to 21 tooth region with resorption of roots from 16 to 21 tooth region.(Fig.2)

Fig.2 Orthopantomogram (OPG) of right maxillary cyst.

BIOPSY

A Small bit of app 1x1cm soft tissue was excised and sent for histopathologically examination. (Fig.3)

Fig.3 Incisional biopsy of soft tissue specimen.

The histopathological reveals:

- Epithelaial lining showing palisading basal cuboidal to collumellar cells.
- Epithelial is detached focally from subepithelail fibroconnective tissue.
- Luminal surface has wavy parakeratinised epithelial cyst.

TREATMENT:

Under all aseptic condition standard drapping and prepping was done. 2% LA with adrenaline administered as right and left buccal vestibular region and at posterior middle and

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anterior superior nerve block, greater palatine nerve block and nasopalatine nerve block were administered. Crevicular incision was given fron 16 to 21 tooth region. Subperiosteal dissection was done. Full thickness mucoperiosteal flap was raised; bony perforation was noticed at 11 to 13 tooth region. Cystic lining with cystic cavity was enucleated (Fig. 4) and extraction of the resorbed tooths were done. The cystic cavity was excised intoto and send for histopathological examination. A 5FU impregnated with gauge ribbon was placed into the cystic cavity and the flap was approximated with 3-0 vicryl. The 5 FU impregnated gauge was taken out from the extraction socket after 24 hrs.
After 2nd, 3rd and 7th day evaluation were done.

Fig:4 Fig:5

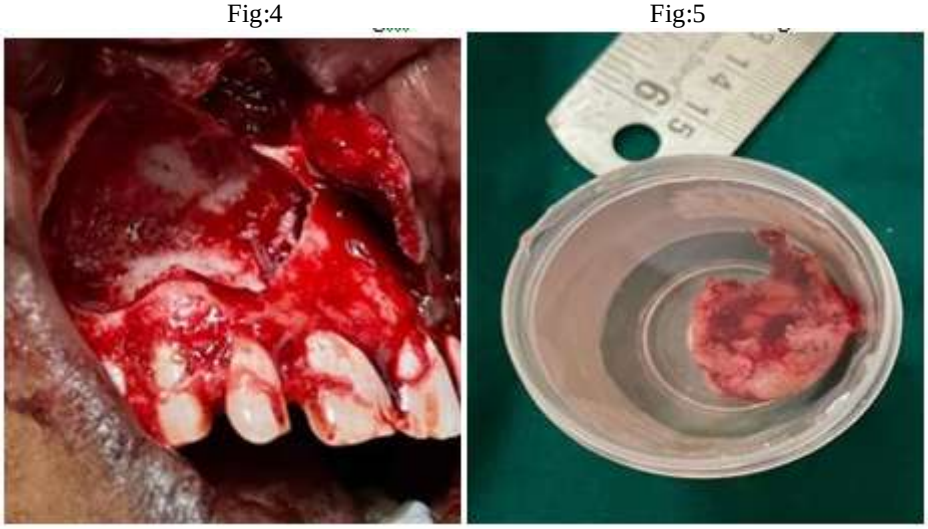

Fig.4: Enucleation of the cyst.
Fig.5: Exicion of the Cystic cavity.

OUTCOME AND FOLLOWUP:

The patient was asymptomatic for 6th months after surgery. As the OKC has high recurrence rate a follow up period of atleast 5 years is necessary.

II. DISCUSSION:

Odontogenic keratocyst is one of the most aggressive odontogenic cyst,common in twice in mandible than the maxilla. The management for OKC showed with the use of carnoys solution the recurrence rate .however the genetic nature of okc was similarto basal cell carcinoma , the newer trend had begun with the topical application of5- FU.

A study by Leddehoff showed that management of OKC with carnoy's solution and 5 FU. He stated that patient treated with modified carnoy's solution showed recurrence rate of 18%IN 26.3+/-1.8 months and there were no recurrence with 5FU and he stated that wound healing has delayed with the use of carnoy's solution.[9] Balamurgun reported 5FU as atrend setter for treatment modality in OKC.[10] Histologically OKC been classified as parakeratotic or orthokeratotic subtype. These subtypes were characterized by the lining and the type of keratin produced. The orthokeratin produced keratin normal to keratin which does not contain nuclei. The parakeratin produce more keratin, no keratolytic granules and cells slough into keratin layer. [11] The lesion reported in this is a parakeratin okc.

III. CONCLUSION:

Odontogenic keratocyst is the most aggressive cyst in oral cavity unidentified it may leads to pathological fracture. Early identification with the availability of newer trends of treatment modality the recurrence rate of okc compared to traditional method of using carnoy's solution had decreased along term follow up is still needed to identify the recurrence rate with the application of 5FU exist. Till date the application of 5FU in management of OKC showing promising results with no evidence of recurrence.

PATIENT CONSENT: Obtained.
CONFLICT OF INTEREST: No conflict of interest.

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