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Multiple Infected Odontogenic Keratocyst of Jaw: A Case Report

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Abstract

Multiple Odontogenic keratocyst (OKC) of jaws is one of the rarest findings occurring in young teenage patients. Odontogenic keratocyst is a rare and benign tumor of jaws and one of the common odontogenic cyst, having high recurrence rate and unique radiographic pattern. It most commonly occurs because of cell rests of dental lamina which are most commonly seen in 2nd or 3rd decade of life with slightly male predilection. Here is a case report of a young female patient who was diagnosed with Multiple Odontogenic Keratocyst of maxilla and mandible on the basics of clinical , radiographical, and histological findings and was treated surgically and had good surgical outcomes on her follow ups.

Keywords: Multiple Odontogenic keratocyst , High reccurence , Clinical radiographical and histological findings

Introduction

Odontogenic keratocyst was first identified by Wright which was described by Philipsen in 1986 which is a rare and benign aggressive tumor of jaws and most common odontogenic cyst. One of the unique cyst of jaws as it is most aggressive, having high recurrence rate, and having characteristic radiographic and histological appearances.[1]

One of the most common etiological factors of OKC is that it develops from rest cells of dental lamina or from off shoots of the basal cell layer of epithelium. There are 2 characteristic features of OKC: orthokeratocyst and parakeratocyst. The behaviour of the former seems to be less aggressive, whilst the latter exhibits potential for local destruction and extension into adjacent tissues, rapid growth, a higher rate of recurrence and a tendency for multiplicity.

Cyst usually presents as an asymptomatic, solitary, radiolucent lesion located in the posterior mandible. OKC

has a unique feature to grow in an anteroposterior direction within the medullary cavity of the bone without causing cortical bony expansion. Radiographical analysis can be unilocular or multilocular hence it resembles with other tumors like ameloblastoma or cysts like dentigerous cyst, lateral periodontal cyst and radicular cyst.[2]

Case Report

A 19year old female reported with chief complain of swelling in right upper face region since 1 month. Patient was apparently asymptomatic 1 month back as patient had undergone extraction of upper deciduous teeth by a local practioner after which patient had hot fermentation due to which swelling increased in upper right side of face which was incidious in onset, tender on palpation and with pus discharge intra and extra orally with sinus opening present extra orally on right cheek region.(Fig 1,2) Extraoral physical examination showed floating facial edema in the region of the right cheek.(Fig 3) There was local rise in temperature of skin region.

On Examination a 6 X 8 cm swelling extending from right lateral nasal border till right tragus of ear anteroposteriorly and from lower eyelid to lower border of mandible superoinferiorly was present which was firm in consistency.

Intra oral examination showed impacted permanent canine irto 13,23,33, and 43 with marked swelling in the right side upper vestibular region(Fig 4,5)

Radiographical Examination with OPG showed multiple impacted canine in maxilla and mandible with right side of maxillary impacted canine in maxillary sinus. CECT scan of face revealed large expansile lobulated cystic lesion involving right maxillary alveolar process and maxillary crest with extension. There was another expansile septated cystic lesion seen in the body of mandible diagnosing it as odontogenic keratocyst or dentigerous cyst (Fig 6,7,8,9)

Histopathological examination revealed a cystic cavity lined with an orthokeratinized epithelium with an average thickness of 10-12 cells. The basal layer exhibited a cuboidal morphology with no tendancy for hyperchromatism or palisading nucleus. The cyst consisted of dense connective tissue with inflammatory exudate which suggested it a Infected Multiple Orthokeratinized Odontogenic Cyst.

Treatment Plan - Patient was admitted in Oral Surgery ward and was administered IV antibiotics as Inj. Amoxicillin 500mg 8hrly and Inj. Metrogyl 100ml 8hrly and was planned for Surgical Enucleation of the cystic cavity with chemical cauterization with Cornoy's Solution under general anesthesia was done. Patient was under observation and was recalled for 3 months for follow up

On 1st month follow up of the patient the swelling of the patient was reduced and healing was satisfactory. (Fig 10,11)By the end of 3rd month of her follow up patient had no swelling with satisfactory healing occurring intra orally.(Fig 12) Intra orally patient had wound dehiscence in maxillary vestibular region irto 13 region post operatively which was closed by primary closure.(Fig 13) Patient is under observation for further follow up.

Discussion

Odontogenic keratocyst was first described by Mickuliz in 1826. In 1926 it was termed as "cholesteatoma." which meant cystic or "open" mass of keratin with a living "matrix". However in 1956 the cyst was named odontogenic keratocyst by philipsen.[3]

Odontogenic keratocyst being an aggressive nature as said by WHO termed it as Keratocystic Odontogenic Tumor to describe the cyst is derived from the remnants of dental lamina with it's biologic behaviour similar to benign neoplasm.[4] It is named as keratocyst because of the cystic lining that produces keratin. OKC can occur in any age

group but most commonly it occurs during 2nd and 3rd decade of life having male predilection over females.[5]

Presenting symptoms in OKC are mainly pain, swelling, expansion of cortical bones, pus discharge and presthesia.[6]

In our case patient had symptoms of pain, swelling and pus discharge with sinus opening extra orally in the infra orbital region.

Radiographically,OKCs are more commonly unilocular with scalloped margin when presented at the periapical region and can be mistaken for radicular or lateral periodontal cyst. When the cyst is multilocular and located at the molar ramus area it may be mimicking ameloblastoma. The septa present in ameloblastoma are coarse and curved; originate from the normal bone trapped within the tumor. On radiographical examination these septa resembles honeycomb or soap bubble appearance which is not seen in OKC.[7]

In our case patient had multiple radiolucenies with aggressive behaviour at the right side of maxilla with expansion of bucco- palatal expansion and antero posterior expansion in right body of mandible associated with impacted canine.

Various attempts have been made to reduce the high recurrence rate of OKCs by modifications in the operative technique. Depending on various factors such as age , location of the lesion, size , primary or recurrent lesion treatment modalities can vary accordingly.

Main surgical treatment for OKC can be simple marsupilization , or enucleation or resection with adjuvant surgical treatment like peripheral ostectomy , liquid nitrogen or cornoy's solution.[8]Carnoy's solution is composed of 3 ml of chloroform, 6 ml of absolute ethanol, 1 ml of glacial acetic acid, and 1 g of ferric chloride. An alternative to chemical cauterization is the use of cryosurgery after removal of the lesion. A temperature of -20 C is required to devitalize tissues and only liquid nitrogen can deliver this on a consistent basis.

In our case patient was treated with cystic enucleation followed by peripheral ostectomy and chemical cauterization with modified cornoy's solution. Modified Cornoy's solution consist of 1g of ferric chloride , 1ml glacial acetic acid , and absolute alcohol 6ml.

Various post op complications such as paresthesia , wound dehiscence , reinfection or recurrence can occur in OKC. Chances of recurrence are one of the most common complications in OKC. In our case study patient had complication of wound dehiscence but there is no evidence of recurrence till date.

Recurrence rates of Odontogenic Keratocyst are highest with enucleation alone and range from 9% to 62.5%. Enucleation with Carnoy's solution provides the least recurrence (4.8%) from any of the conservative techniques.[9] In a systematic review, the authors reported that resection was found to have the lowest recurrence rate (0%) but the highest morbidity rate.

Conclusion

Odontogenic keratocyst if not diagnosed early with time it has been seen that it may grow and expand to large size before becoming clinically evident unlike other jaw cysts and have greater tendency to recurr after surgical treatment. It is one of the aggressive but asymptomatic cyst which can be well diagnosed with clinically, radiographic, and histopathological correlations for proper patient treatment and follow up. Majority of the treatment protocol used is surgical enucleation , marsupilization or resection with chemical cauterization with cornoy's solution or liquid nitrogen. Hence the correlation of histopathologic findings with clinical and radiographic features is necessary to achieve a definitive diagnosis.

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Legend Figure

Fig 1: Frontal View with swelling on right side of face with sinus opening

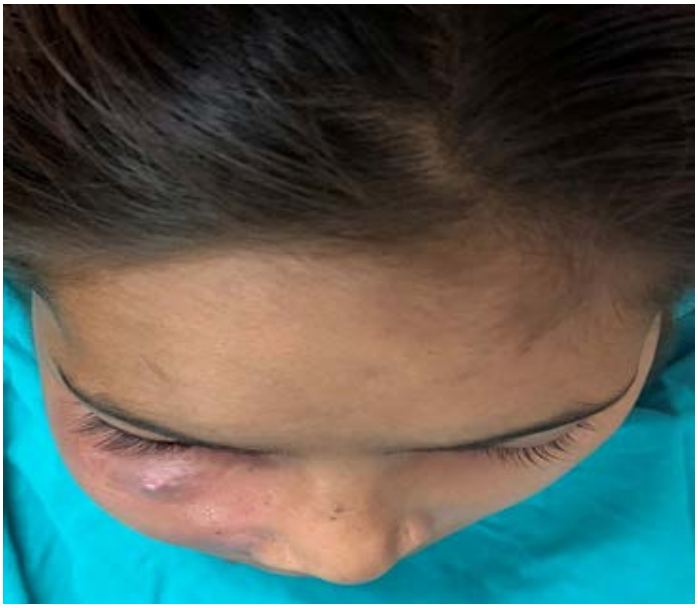

Fig 2: Bird's eye view with extra oral sinus opening

Fig 3: Lateral view measuring 6 X 8 cm swelling extending from right lateral nasal border till right tragus of ear anteroposteriorly and from lower eyelid to lower border of mandible superoinferiorly

Fig 4: Intra oral occlusal view – Multiple impacted canine with sinus opening in right vestibular region irto 13

Fig 5: Occlusal view showing impacted canine and sinus opening in right vestibular region irto 13

Fig 6: OPG of the patient showing multiple impacted canine in maxilla and mandible. Impacted canine in right side of maxillary sinus in maxilla

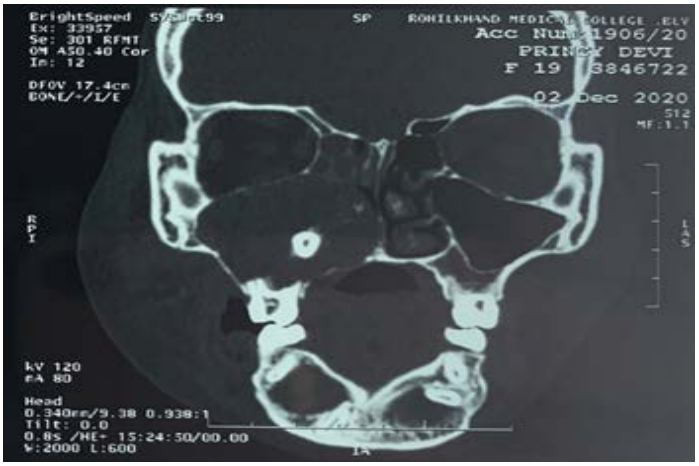

Fig 7: Coronal view of CECT showing impacted canine and atrophy of sinus with excessive destruction of maxilla

Fig 8: Axial section of CECT showing bucco palatal expansion on right maxilla crossing the midline with destruction of maxilla.

Fig 9: Axial section of CECT showing anteroposterior expansion of mandible

Fig 10: 1 month follow up of patient

Fig 11: 3 month follow up

Fig 12: Post-operative healing on right side maxilla

Fig 13: Post operative compilation of wound dehiscence