

**Rare Case of Simultaneous Occurrence of Odontogenic Keratocyst in the Anterior Maxilla and Mandible: Our Experience and Review of Literature**

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**Introduction**

Odontogenic keratocysts (OKCs) are epithelial developmental cysts which were first described by Phillipson in 1956. The frequency of OKC has been reported to vary from 3% to 11% of odontogenic cysts. It occurs mainly in second and third decades, with slight male predilection and may occur in any part of jaws with the majority of lesions occurring in the mandible.

Generally, OKCs are solitary lesions unless they are associated with nevoid basal cell carcinoma syndrome. Although odontogenic keratocysts are common in clinical practice, the simultaneous occurrence of multiple cysts in both the maxilla and mandible of a patient is rare. We report case of multiple odontogenic keratocyst in a non syndromic patient of what we believe to be a unique lesion that may represent example of varied clinical presentation

of odontogenic keratocysts that had been described in earlier papers. In addition, cases reported in the past are summarized.

Under general anaesthesia, enucleation procedure was performed, followed by aggressive curettage of bony walls, peripheral ostectomy with bone bur and application of Carnoy solution in both the cystic cavities. The case has not recurred from past 10 years since the procedure was performed.

**Keywords:** Keratocyst, Mandible, Maxilla, Odontogenic cyst,

### Introduction

The odontogenic keratocyst (OKC), was recently designated by the World Health Organization as a keratocystic odontogenic tumor and was defined as “a benign uni- or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behaviour.”<sup>[1]</sup> OKC often occur in the mandibular ramus and angle region but rarely in other mandibular regions or the maxilla.<sup>[2,3]</sup> The OKC is one of the most aggressive odontogenic cysts owing to its relatively high recurrence rate and its tendency to invade adjacent tissue. OKC resembles ameloblastoma in imaging, and, apart from histology, no reliable criteria exist for pretherapeutic differentiation.<sup>[4]</sup> In the literature there is a controversy regarding the treatment of this lesion: some surgeons advocate conservative therapies, whereas others are in favor of an aggressive treatment.<sup>[1]</sup>

The present study aimed at evaluating the variations in which OKC can clinical present in our routine practice along with effectiveness of CS as addition therapy to enucleation in the treatment of OKC in a 10-year retrospective study.

### Clinical Presentation

A 38 year old male presented to our department with a complaint of fluid discharge from right lower back tooth region of mandible from past 1 week. The patient described the fluid to be yellow in colour and salty to taste. On examination, the patient exhibited slight facial asymmetry in the area of left parasymphysial region, with diffuse swelling in chin region. On further query, the patient revealed history of paresthesia in right lower lip and swelling in the chin region from past one month. On intraoral inspection, patient exhibited poor oral hygiene and was partially edentulous. Diffuse swelling in lower buccal vestibule extending from third molar on right side to first premolar on left side of the mandible, measuring about 6 x 4 cms was detected. Expansion of buccal cortex with buccal vestibule obliteration was present in relation to 36 to 48 regions. Bony crepitus was present in buccal cortex in relation to right posterior teeth region. On application of pressure, Yellowish colour fluid was oozing from the crevicular area in relation to 47 and was foul in odour. Lingual cortex and inferior border of mandible was intact. Mobile teeth in relation to lower anteriors were elicited and 13, 33 and 38 were missing since birth and others were missing due to caries and its sequelae. Root stumps were present in relation to 36. On neck examination, Right submandibular lymph nodes were found to be tender and mobile.



Fig 1: Preoperative frontal view showing swelling in left parasymphysial region



Fig 2: Preoperative intraoral photograph showing obliteration of right buccal vestibule in mandible

A Panoramic radiograph revealed a large unilocular radiolucency associated with the impacted right lower canine tooth. The lesion extended from retromolar region on right side to 36 region on left side. It measured about 8cm x 4cm in size, and was unilocular, well circumscribed and was surrounded by thin radio opaque border with scalloped periphery at the superior border. Erosion of mandibular canal as well as thinning of inferior border of mandible could also be appreciated. Simultaneously,

impacted 38 were also noted on the OPG. Another impacted tooth was identified incidentally in the right maxillary anterior region which appeared to be like a canine with a radiolucent lesion surrounding it.

To know more about the extension of cyst we advised an occlusal radiograph of both mandible and maxilla. The maxillary radiograph provided better definition of cyst in relation to impacted 13. It was unilocular, well circumscribed and was surrounded by thin radio opaque border. The periphery of the lesion was smooth and was associated with impacted right maxillary canine. The lesion measured about 2 x 3 cms approximately in size; the point to be noted is that, this lesion was clinically insignificant and totally asymptomatic.

Further the mandibular radiograph revealed the thinning of buccal cortex in relation to 45 and 46 regions and presence of impacted 43.



Fig 3: Preoperative panoramic radiograph showing the impacted right maxillary and mandibular impacted canine with cystic outline



Fig 4: Preoperative occlusal radiograph showing the impacted right mandibular canine with expansion of buccal cortex.

### Differential Diagnosis

The clinical and radiographic features of a radiolucent lesion surrounding an impacted tooth were considered highly suggestive of an odontogenic cyst or neoplasm. The lesions considered in common to both the lesion were based on the following assessment:

#### Differential diagnostic lesions for lower lesion:

Dentigerous cyst is by far, the most common developmental odontogenic cyst. By definition, it arises in association with an impacted tooth and is often first detected on routine radiographic examination as well defined, unilocular, non expansile radiolucency in the posterior mandible of a young adult patient.<sup>[5, 6]</sup>

The possibility of ameloblastoma was also considered, even though these tumors usually present in an older patient population. Unicystic and conventional ameloblastoma was discussed in the differential diagnosis, since these lesions typically present as a circumscribed radiolucency surrounding the crown of an unerupted tooth. However they are more commonly seen in the posterior mandible. Furthermore, while conventional ameloblastoma may present as a unilocular radiolucency,

larger lesions are often multilocular and cause significant expansion of the jaw.<sup>[5, 6]</sup>

Adenomatoid odontogenic tumor typically affects teenage to middle aged adult females as an asymptomatic, pericoronal radiolucency. Involvement of the radicular portion of the impacted tooth as well as the coronal portion is often observed and may serve to distinguish this process from dentigerous cyst. AOT is more frequently observed in the maxilla than the mandible and is usually found in anterior portion of jaw between the apices of the lateral incisor and premolar, intimately associated with an impacted canine.<sup>[5, 6]</sup>

OKC can present with clinical and radiographic features that are identical to dentigerous cyst. Odontogenic keratocyst was also considered because the lesion occurred in relation to an impacted tooth and was connected to the tooth at a point apical to the cemento-enamel junction.<sup>[5, 6]</sup>

#### Additional differential diagnostic lesions for upper lesion

Odontogenic myxoma was considered in the differential diagnosis for upper lesion. In the majority of cases, odontogenic myxomas present as a multilocular radiolucent lesion. When occurring pericoronal to an impacted tooth, they can also present as a cyst-like unilocular lesion.

Consideration was also given to desmoplastic variant of ameloblastoma, since this entity is reported to have a predilection for the anterior maxilla. However this usually presents with a mixed radiolucent-radioopaque pattern resembling a fibro-osseous lesion.

Ameloblastic fibroma can also be included among the diagnostic possibilities since this tumor presents as a symptom free unilocular lesion in the first 2 decades of life. However, 70% of all cases occur in the posterior mandible.

Central giant cell granuloma also merits inclusion in differential diagnosis. It occurs most frequently in the anterior mandible in female patients under the 32 years of age. However, maxillary lesions often arise anterior to the cuspid, and smaller lesions can present as a solitary cyst like unilocular radiolucency<sup>[6]</sup>

The possibility of this lesion representing a malignant tumor was considered highly unlikely because of the painless, slow growing expansile nature of the lesion, the presence of well circumscribed radiographic margins, and an absence of cortical destruction.

Following informed consent and local anaesthesia, aspiration with wide bore needle of lesion in both the jaws was performed. The lower lesion yielded around 2 ml of thick yellowish coloured fluid. The upper lesion yielded around 1 ml of white coloured thick fluid. Following the procedure, the patient was placed on empirical antibiotics. FNAC report revealed features of infected odontogenic keratocyst in relation to 43 and odontogenic keratocyst in relation to 13. Then the patient underwent teeth vitality test which revealed non vital teeth in relation to lower anteriors upto first molar bilaterally.

Nevoid basal cell carcinoma syndrome, an autosomal disorder with a high degree of penetrance and variable expressivity, is characterized by basal cell carcinoma, odontogenic keratocysts, palmar and/or plantar pits and ectopic calcification of falx cerebri.<sup>[7]</sup>

So, we proceeded with our further investigation to rule out any other abnormality or syndromes associated with the lesion. Occipitofrontal circumference and intercanthal distance were within normal limits. General examination and skin examination did not reveal significant findings. Anteroposterior skull radiograph, chest and lumbosacral radiograph showed no abnormality.

## **Management**

After presurgical work up, patient was posted for surgery under general anaesthesia.

An intraoral crevicular incision was made and a mucoperiosteal flap was reflected from the left second premolar to the retromolar area on the right side, exposing the expanded buccal cortical plate. Teeth lying within the area of cyst were extracted, paper thin bone on the buccal side was removed by means of bone rongeur and cystic wall was exposed. During surgery, cystic process was clearly identified beneath the mucosa and pultaceous, cheesy material was oozing from the cyst in relation to 43. The cyst was traced well behind upto the retromolar area on the right side to identify the inferior alveolar nerve which was then transected. Enucleation procedure was performed, followed by aggressive curettage of bony walls. Peripheral osteotomy with bone bur was done and Carnoy solution was applied in the cystic cavity. The surgical site was irrigated with saline and the cystic cavity was packed with iodoform gauze. The mucoperiosteal flap was reapproximated and the surgical site was closed with 3-0 vicryl sutures.

Then we proceeded with the exploration of maxillary cyst. A vestibular incision was given in relation to 13 and the flap was reflected. Bony window was created and enucleation of the lesion was accomplished. The impacted tooth was removed along with the cyst. There was no sinus involvement elicited. It was followed by aggressive curettage of bony walls, peripheral osteotomy with bone bur and application of Carnoy solution in the cystic cavity in relation to 13. Iodoform gauze was placed in situ and the vestibular flap was reapproximated with 3-0 vicryl sutures. Grossly, the specimen was composed of whitish, thick cystic wall with impacted 43 and 13. The surgical specimens were submitted in formalin for histopathologic

review. The biopsy report indicated a diagnosis of OKC in relation to 43 and 13.



Fig. 5: Intraoperative photograph showing bony cavity after enucleation of the cyst in relation to 13.

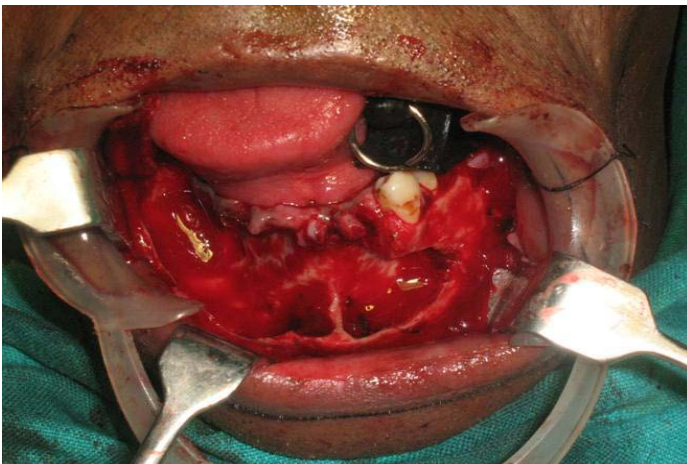


Fig. 6: Intraoperative photograph showing post Enucleation bony cavity in relation to 43.



Fig. 7: Photograph showing the specimen in relation to mandibular impacted canine.



Fig 8: Photograph showing the specimen in relation to maxillary impacted canine

### Microscopic Examination

Microscopic examination revealed epithelial lining of varying thickness showing parakeratinised stratified squamous epithelium. The basal epithelial layer composed of hyperchromatic and palisaded cuboidal to columnar epithelial cells. Epithelial connective tissue interface was relatively flat and separation of epithelium from connective tissue was seen in some areas. In some areas, the epithelial lining was orthokeratinised and prominent keratohyaline granules were present in the superficial epithelial layers. In some areas, the epithelium was non keratinized type and showed hyperplasia with rete ridges formation. The connective tissue wall had daughter cysts along with chronic inflammatory infiltrate and few acute inflammatory cells. The connective tissue also consisted of microcyst in relation to both the cysts. Submitted cystic content showed keratin in relation to both the cysts.

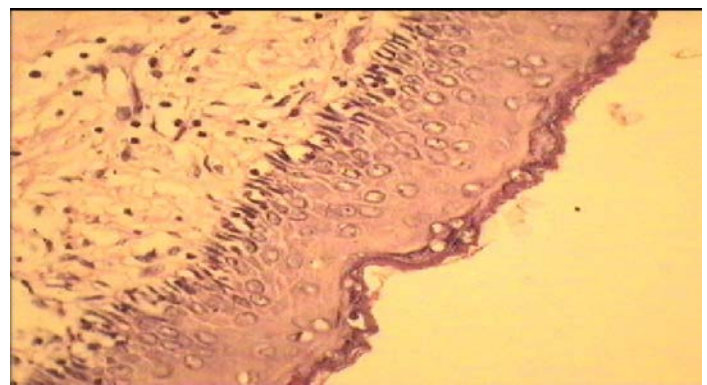


Fig 9: Characteristic OKC lining in relation to 43. (Haematoxylin and eosin stain, original magnification x 400).

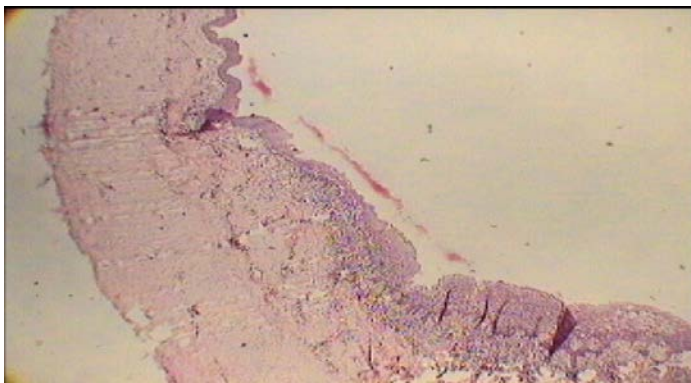


Fig 10 : typical OKC lining in relation to 13 (Haematoxylin and eosin stain, original magnification x 40).

### Final Diagnosis

Finally the lesions were diagnosed to be OKC in relation to 43 and 13

### Clinical Course and Management

After the patient underwent the surgical procedure, he was recalled every third day for irrigation and change of dressing. At each subsequent appointment, the iodoform gauze was shortened and loosely packed until the wound healed completely. Later the patient underwent prosthetic rehabilitation for the replacement of missing teeth. After ten years of follow up there is no evidence of recurrence and radiographs showed that the cystic area had totally recovered. Patient was explained about the scientific importance of his disease and consent was taken regarding publishing the case in scientific literature.

### Discussion

The odontogenic keratocyst (OKC) is classified as a developmental cyst derived from the enamel organ or from the dental lamina. It comprises approximately 12% of all cysts of the jaws. The cysts are most often seen in the mandibular ramus and angle.<sup>[5]</sup>

The histological features of OKCs include a thin epithelial lining, usually consisting of fewer than six cell layers in a corrugated or wavy pattern. The underlying connective tissue is composed of thin, irregular bundles of collagen

and often contains islands of epithelium that may represent daughter cysts. In many cysts there is a tendency for the epithelium to separate from the underlying cyst wall. The most characteristic clinical aspect of OKCs is the high frequency of recurrence.<sup>[8]</sup>

The OKC has been the subject of much debate over the last 50 years with respect to its origin, its growth, clinical presentation and treatment modalities. The clinical appearance of OKC varies from a symptomless incidental radiographic finding to painful cystic lesions. Paresthesia may also be present in some patients as well as secondary fractures when the cyst is large. The maxillary OKC tends to be secondarily infected with greater frequency than the mandibular ones. That may be due to its vicinity to the maxillary sinus. Odontogenic keratocyst is a cyst which is always in controversy as being unilocular or multilocular, parakeratinized or orthokeratinized, benign or malignant and so on. Treatment of OKCs also remains a controversial subject.

Multiple Odontogenic keratocysts have been reported in Nevoid basal cell carcinoma syndrome, but multiple keratocysts in patients with no other syndrome manifestations are rarely found. In the above described scientific case, two extreme clinical presentation of OKC were seen in the same patient simultaneously, with one cyst showing features of full blown cyst whereas the other was symptomless and was identified incidentally on radiograph.

The features that make the above reported case a special one are, in our case though multiple OKC's were present; there were no signs of NBCS. Displacement of teeth rarely occurs in OKC but in the above documented case, 43 was displaced drastically and the cyst was crossing the midline

, as well as bony expansion along with crepitus was present, which is rarely seen as OKC's enlarge at the expense of cancellous bone.

The highlights of various clinical presentation of OKC as respect to review of literature is mentioned in Table.

Table: Variation of clinical presentation of OKC as reported in literature

Sn.	Study	Clinical presentation
1	Neil R. Attenborough, 1974 <sup>[9]</sup>	Recurrence of an odontogenic keratocyst in a bone graft
2	Joseph T. Fay, 1978 <sup>[10]</sup>	Bilocular odontogenic keratocyst
3	Valerie J. Lund, 1985 <sup>[11]</sup>	Odontogenic keratocyst of the maxilla
4	R. I. Macleod, K. B. Fanibunda, J. V. Soames, 1985 <sup>[12]</sup>	A pigmented odontogenic keratocyst
5	Joseph L. Matisse, 22 M. Beto, John E. Fantasia, Allen F. Fielding, 1987 <sup>[13]</sup>	Pathologic fracture of the mandible associated with simultaneous occurrence of an odontogenic keratocyst and traumatic bone cyst
6	Sook-Bin Woo, Leon Eisenbud, Michael Kleiman, Nelson Assael, 1987 <sup>[14]</sup>	Odontogenic keratocysts in the anterior maxilla
7	Gerald A. Cioffi, Geza T. Terezhalmay, Angelo M. Del Balso, 1987 <sup>[15]</sup>	Odontogenic keratocyst of the maxillary sinus
8	R. I. MacLeod, J. V. Soames, 1988 <sup>[16]</sup>	Squamous cell carcinoma arising in an odontogenic keratocyst
9	P.J.W. Stoelinga, 1992 <sup>[17]</sup>	Recurrent odontogenic keratocyst within the temporalis muscle
10	F.S.A. Nohl, K. Gulabivala, 1996 <sup>[18]</sup>	Odontogenic keratocyst as periradicular radiolucency in the anterior mandible
11	H. Reychler, 1997 <sup>[19]</sup>	Penetration of a keratocyst into the skull base
12	Brad W. Neville, Douglas D. Damm, Thomas Brock, 1997 <sup>[20]</sup>	Odontogenic keratocysts of the midline maxillary region
13	Nasser A. H. Said-Al-Naief, Harry Lumerman, Marie Ramer, William Kopp, Gilbert J Kringstein, Floriana Persenchino, Roosevelt Torno, 1997 <sup>[21]</sup>	Keratoameloblastoma of the maxilla
14	Adalberto Mosqueda-Taylor, José Mario de la Piedra-Garza, Frank Troncozo-Vázquez, 1998 <sup>[22]</sup>	Odontogenic keratocyst with chondroid fibrous wall
15	Maria L. Fornatora, Renee F. Reich, Gregory Chotkowski, Paul D. Freedman, 2001 <sup>[23]</sup>	Odontogenic keratocyst with mural cartilaginous metaplasia
16	Kok Han Ng, Chong Huat Siar, 2003 <sup>[24]</sup>	Odontogenic keratocyst with dentinoid formation
17	A. Chi, S. Muller, 2004 <sup>[25]</sup>	Peripheral odontogenic keratocyst



18	J. H. Yoon, S. G. Kim, S. H. Lee, J. Kim, 2004 <sup>[26]</sup>	Simultaneous occurrence of an odontogenic keratocyst and giant cell granuloma-like lesion in the mandible
19	Wei-Yung Yih, John L. Krump, 2005 <sup>[27]</sup>	Odontogenic Keratocyst in the Nasopalatine Duct Associated With Mural Cartilaginous Metaplasia
20	M.K. Nair, J.C. Pettigrew, A.A. Mancuso, 2006 <sup>[28]</sup>	Incidental finding of vascular malformation of internal carotid artery in patient with odontogenic keratocyst
21	Auluck A, Suhas S, Pai KM., 2006 <sup>[29]</sup>	Multiple odontogenic keratocysts
22	Chen AW, Wang JH, Wang KT, Li GJ., 2007 <sup>[30]</sup>	Mandibular keratocyst induced trigeminal neuralgia
23	Ferreira O Jr, Cardoso CL, Capelozza AL, Yaedú RY, da Costa AR, 2008 <sup>[31]</sup>	Odontogenic keratocyst and multiple supernumerary teeth in a patient with Ehlers-Danlos syndrome

The various investigations other than conventional techniques that can be of help in diagnosing OKC are histochemical markers, DNA analysis, electron microscopy, magnetic resonance imaging and CT scan. [12, 32, 33, 34, 35, 36]

The modified Brosch procedure represents a reasonable alternative to resection or marsupialization when treating large OKC that occupy the molar, angle, and ramus regions of the mandible. Extraction of teeth affected by the lesion as well as generous removal of partially eroded bone and overlying soft tissues may contribute to lower recurrence rates. Cryosurgical or mechanical treatment of the bony cavity, which is well visualized using this technique, may also lead to improved long-term results. Treatment must be followed by continued, careful observation of the patient. [37]

The frequency of recurrence after surgical intervention has been reported to vary from 2.5% to 62.5%. Recurrence rates significantly depend on sites of involvement and OKC's in the mandibular molar region have significantly higher recurrence rates than those in other sites. The presence of one or more daughter cysts is significantly related to recurrence. [38]

Follow up of patients with OKC's should be carried out regularly. As most recurrences present within the first five years, yearly follow up during this period is advisable.

Thereafter, follow up at every 2 years seems appropriate but should extend for atleast 25 years, as recurrences may appear after a long time. [39]

### Conclusion

It is one of the most aggressive odontogenic cysts owing to its relatively high recurrence rate and its tendency to invade adjacent tissues. Although OKC is classified as a developmental cyst, there is abundant evidence to support the suggestion that OKC is a benign cystic tumor. [8, 40]

As mentioned in TABLE , OKCs can present as pigmented lesion and can occur in any location in the jaws as well as temporalis muscle and nasopalatine duct. It can manifest as periradicular radiolucency and may be accompanied with squamous cell carcinoma, keratoameloblastoma, mural cartilaginous metaplasia, dentinoid formation, giant cell granuloma and so on. It is also known to induce trigeminal neuralgia and penetrate skull base as well. [11,12,14,15,16,17,18,19,20,21,23,24,26,27,30]

Though OKC is very common, till now it is the most ill understood. This might be due to its diverse manifestation ranging from an asymptomatic cyst to fully blown aggressive cyst with severe signs and symptom. The same applies to the above reported case where in the maxillary lesion was incidentally found out where as the mandibular lesion manifested with pain, paresthesia, extension beyond midline, erosion of the inferior alveolar canal,

expansion of the buccal cortex and so on. To add on, though it is commonly found in mandibular ramus area, in our case, OKC was situated in the anterior jaw region both in maxilla and mandible,

The term multiple cysts refers to the lifetime history of the patient and does not necessarily imply that more than one cyst is present at any one time. [41] Nevroid Basal Cell Carcinoma Syndrome is accompanied with odontogenic keratocysts, often multiple and bilateral. Diagnosis of Nevroid Basal Cell Carcinoma Syndrome may be difficult because of the variability of expressivity and because of different ages of onset for the different traits of this disorder. [7]

In our case though multiple OKCs were present, there was no evidence of any syndrome. Furthermore, though bilateral multiple OKC have been reported in literature, unilateral multiple OKC have never been reported as seen in the above reported case where in both the cysts occurred on the right side of the jaws .

Difficulty in radiographically differentiating between OKC and any other lesion of the jaw such as Dentigerous cyst, Lateral Periodontal cyst and Ameloblastoma is well documented. Furthermore; the diagnosis of OKC was not suspected even at the time of surgery in most cases, as has been reported by Neville et al. [38]

Negligence on part of patient may pave way for extensive manifestation of the cyst leading to facial asymmetry, paresthesia, infection, cortical expansion of jaws, intraoral drainage, mobility of teeth and non vitality as reported in the above case. The sooner the patient reports to the hospital, the better is the prognosis.

The recurrences observed in OKC's may not necessarily be due to the degree of skill of surgeon or the technique used to eradicate the primary cyst, but instead are probably a reflection of the multifocal nature of the pathologic lesion itself. [42] The patient is on regular follow

up from past 10 years and there is no evidence of recurrence till date.

To conclude, multiple OKCs can occur in the absence of Syndromes. Early diagnosis helps in arresting the growth of the cyst at the incipient stage, prevents aggressive manifestation of the cyst and its related morbidity.

Preoperative confirmation of OKC by means of histopathologic investigation or confirmation during surgical procedures by means of frozen section would help clinicians select more suitable treatment modalities. To conclude, not all the cases of odontogenic keratocyst follow the classic teaching lines.,each and individual case is a different entity and one should have a high rate of suspicion while treating cystic cases.

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