

Imaging Characteristic of 11 Lesions of Odontogenic Keratocyst in the Indian Subpopulation: A Cone-Beam Computed Tomography Experience

Abstract

Background: The clinical feature and radiographic appearance of orthokeratinized odontogenic keratocyst (OKC) are not characteristic, which may lead to the misdiagnosis. The radiographic appearance of OKC may range from a small unilocular radiolucency to a large multilocular radiolucency, resembling other odontogenic cysts and tumors. **Aim:** The aim was to illustrate the characteristic feature of OKC presented on the digital panoramic radiograph and cone-beam computed tomography (CBCT), which may provide great value for the differential diagnosis and the treatment planning and also to compare the various radiographic features of OKC in CBCT and digital panoramic radiograph. **Materials and Methods:** Digital panoramic and CBCT records of seven cases, with 11 lesions of OKC were analyzed retrospectively from the patients' database from our institute (2014 to 2016), which was histopathologically diagnosed as OKC. **Results:** The mean age of the patients was 24.1 years, and the female-to-male ratio was 3: 4. Six of 11 lesions were localized within the mandible, and five lesions were in the maxilla. **Conclusion:** The presurgical assessment with radiological information is extremely important for treatment planning, and CBCT provides us with an accurate and faster three-dimensional representation of a lesion at a lower dose and cost, but the role of panoramic radiograph cannot be refuted.

Keywords: Cone-beam computed tomography, keratocystic odontogenic tumor, multilocular radiolucency, panoramic radiography, unilocular radiolucency

Introduction

First described by Philipsen in 1956,^[1] the odontogenic keratocyst (OKC) arises from the remnants of the dental lamina and basal cells of the oral epithelium.^[2,3] OKC was designated by the World Health Organization (WHO) as a keratocystic odontogenic tumor (KCOT) reclassifying this into developmental odontogenic neoplasia in 2005, reflecting its aggressive clinical behavior, high recurrence rate, association with nevoid basal cell carcinoma syndrome and mutation in the Protein Patched Homolog 1 (PTCH) tumor suppressor gene. Histologically, the two variants, orthokeratinized and parakeratinized were considered as different entities. The parakeratotic variant of OKC was designated as KCOT by the WHO, whereas orthokeratotic variant was excluded from the definition of KCOT and called as orthokeratinized odontogenic cyst (OOC).^[3]

In 2017 January, 4th edition of the WHO reverted KCOT from neoplastic category

back into the cyst category as OKC, based on extended genetic and molecular data. This is because many studies showed the presence of PCTH gene mutation even in nonneoplastic lesions including dentigerous other cysts. Moreover, the resolution of cyst after marsupialization may occur, while the neoplastic process does not respond. Histologically, OKC is characterized by palisaded and hyperchromatic basal cells with layer with a corrugated surface layer of parakeratin. OOC, which was referred to as a type of OKC originally, later excluded from KCOT in 2005 WHO classification, now accepted as a separate entity for the first time in 2017 WHO classification. OOCs differ from OKCs as the former are not associated with any syndromes, nonaggressive, do not have high recurrence rate, and are predominantly lined by orthokeratinized stratified epithelium.^[4]

Imaging plays an important role in the diagnosis of maxillofacial jaw lesions because any cyst or neoplasm can achieve dimensions so substantial that imaging

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helps to understand the extent of the lesion.^[5] Significant number of OKCs have no clinical signs and symptoms until they cause bone displacement, cortical disruption, or pain and are found incidentally during routine radiographic examination. Intraoral periapical, occlusal, and panoramic radiographs provide an initial assessment of the position and size of OKCs.^[6] However, cone-beam computed tomography (CBCT) provides high spatial resolution, accurate three-dimensional representation of jaw lesion, borders of large lesions, bone expansion, and cortical perforation with a relatively low dose of radiation.^[7] The objectives of this article were to illustrate the characteristic feature of OKCs presented on the digital panoramic radiograph and CBCT, which may provide great value for the differential diagnosis and treatment planning.

Materials and Methods

Seven cases with 11 lesions were analyzed retrospectively from the patient's database of the Department of Oral Medicine and Radiology, Vydehi Institute of Dental Sciences and Research Center, Bangalore, from 2014 to 2016. The selected cases were histopathologically diagnosed as OKC with both panoramic and CBCT records. Age, gender, history, clinical features including the location of the lesion, associated tooth, cortical plate expansion, and other symptoms were assessed along with consideration for the provisional

diagnosis. The digital panoramic radiographs and CBCT were performed with Kodak Carestream CS9300 (Carestream Health, Rochester, NY, USA) with 60 kV, 8.0 mA for panoramic radiographs, and 90 kV at a time of 12–20 s. In orthopantomography (OPG) and CBCT, we assessed for location (maxilla or mandible – anterior, premolar, and molar); size and shape; periphery (well defined or partially diffuse); internal structure (radiolucent – unilocular or multilocular, scalloping); and the influence of the OKC on adjacent structures, such as teeth, buccal and lingual cortices, lower border of the mandible, inferior alveolar canal (IAC), and the maxillary antrum, was noted.

Results

Clinical features

Seven cases of histopathologically diagnosed as OKC were analyzed. The mean age of the patients was 24.1 years and ranged from 8 to 38 years. The female-to-male ratio was 3:4. There were 11 OKC lesions in seven patients; two patients had multiple OKCs (Case 4 had two OKCs: one in the anterior maxilla and one in the posterior mandible; Case 7 had four OKCs, each in the maxillary and mandibular posterior region). Five of 11 lesions were localized in the maxilla. Six lesions were localized within the mandible [Figure 1]. The recorded duration of the lesion in the form of swelling as

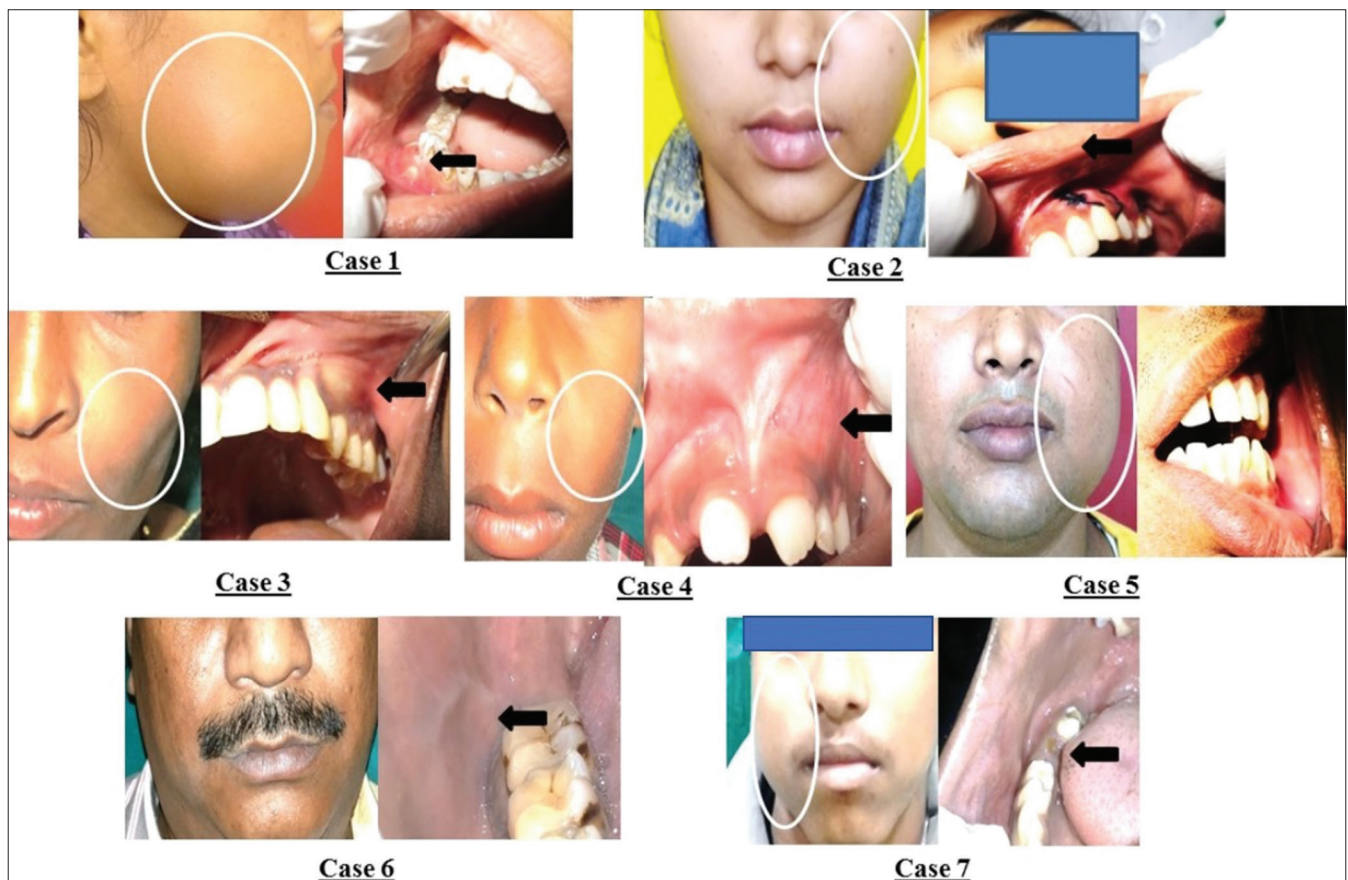


Figure 1: Clinical features of seven cases of odontogenic keratocysts

recognized by the patients ranged from 1 to 6 months. Six of the 11 lesions presented with pain and swelling, whereas one lesion presented with pain as its sole complaint, while four lesions were asymptomatic without pain or swelling. On clinical examination, four lesions did not show any clinical signs of swelling, tenderness, cortical plate expansion, or perforation; one showed the presence of sinus tract with pus discharge; vestibular obliteration was present in four lesions; buccal or labial cortical plates were expanded in five lesions; lingual or palatal cortical plate was expanded in three lesions; except for one lesion where there was perforation clinically, no other cases showed clinical perforation. Four lesions were provisionally diagnosed as dentigerous cyst, one each as radicular cyst, globulomaxillary cyst, and localized periodontitis [Table 1].

Radiological findings

Location

Six lesions were localized within the mandible, and five lesions were in the maxilla. All six lesions that were

localized in the mandible were located in the posterior retromolar ramus region of the jaws. Of the five lesions occurring in the maxilla, two were located in the anterior region and three in the posterior region. Only one among the 11 lesions occurring in the mandible was observed to cross the midline [Figure 2]. One of the 11 lesions showed incidental findings in the panoramic radiograph [Figure 2 and Case 6]. Apart from a chief complaint, two cases [Case 4 and Case 7] showed the presence of additional lesions in the other quadrants, which were first diagnosed incidentally in panoramic radiographs [Figure 3 and Table 2].

Size

The anterior–posterior dimension of the lesions ranged from 12.3 to 61.3 mm, with latter being the only lesion to be extensive as to even cross the midline [Table 2]. All the lesions were well defined.

Internal structure

In panoramic radiograph, three lesions showed unilocular appearance, two lesions showed multilocular, three lesions

Table 1: Clinical features and demographic data

Case number	Age/sex	Pain	Swelling	Location	Clinical features	Provisional diagnosis
1	25/female	2 months	4 months	4 th quadrant	Febrile, sinus tract in 43 and 46 region, missing 45, Buccal and lingual expansion	Infected dentigerous cyst with impacted 45
2	18/female	1 year	6 months	2 nd quadrant	Missing 28, vestibular obliteration from 22 to 27, tender to palpate	Dentigerous cyst with impacted 28
3	30/female	10 days	1 month	2 nd quadrant anterior	Vestibular obliteration from 23 to 25, labial and palatal expansion	Globulomaxillary cyst with 22, 23
4	8/male	1 month	3 months	2 nd quadrant	Vestibular obliteration from 21, 62, 63, minimal labial and palatal expansion 21, 62, 63, 64	Dentigerous cyst with impacted 23
5	24/male	2 months	4 months	3 rd quadrant	Febrile, vestibular obliteration from 36, buccal cortical plate expansion	Infected dentigerous cyst with impacted 38
6	38/male	4-5 months	Nil	4 th quadrant	48 missing distal periodontal pocket with 47	CGG with LP
7	14/male	1-2 months	2 months	4 th quadrant	Vestibular obliteration from 46, 47, 48 region, Buccal cortical expansion, grossly decayed 46, 47	Radicular cyst with 46, 47

CGG: chronic generalized gingivitis; LP: localized periodontitis

Table 2: Radiological features - location, size, and internal structure

Case number	Location	Internal structure (panoramic radiograph)	Size (mm) (CBCT)	Internal structure (CBCT)
1	4 th quadrant	Unilocular, opacities in superior aspect of 32, 31, 41, 42, 43, 45, 46, 47	61.3×43.7×35.1	Unilocular anteriorly Multilocular in posterior Scalloped Bony remnants in the superior aspect
2	2 nd quadrant	Cannot comment	34.9×28.4×23	Unilocular, sclerotic border, discontinuous anterolaterally
3	2 nd quadrant	Unilocular	23.6×28×21.1	Unilocular scalloping near 24 region
4	2 nd quadrant 4 th quadrant	Unilocular, borders are not visible	24.7×24.1×23.5	Unilocular Well defined
5	3 rd quadrant	Multilocular scalloped	30.2×10.5×25	Unilocular
6	4 th quadrant	Multilocular scalloped	32.4×28.5×9.4	Multilocular scalloped
7	4 th quadrant 1 st , 2 nd , 3 rd quadrant	Unilocular, envelopmental	44.1×56.6×24.9	Unilocular Follicular space is intact

CBCT: Cone-beam computed tomography

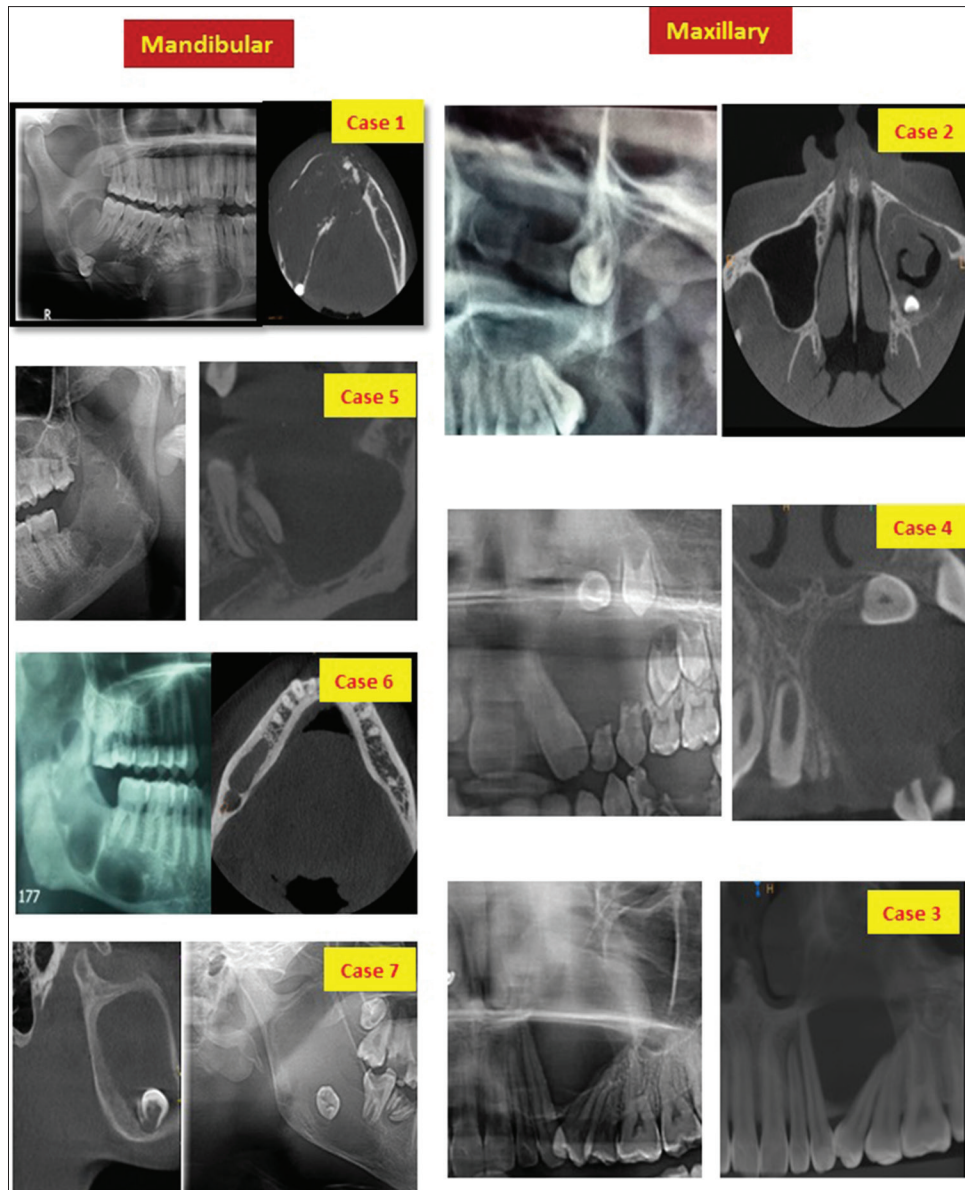


Figure 2: Radiological features – location, size, and internal structure: Case 1: Unilocular radiolucency extending from right ramus, crossing midline; thinning and perforation of buccal and lingual cortical plates; Case 2: Lesion located in left maxilla, with impacted 28; Case 3: Unilocular lesion located in the left maxilla in 23, 24 regions; Case 4: Unilocular lesion located in in the left maxilla in 21,22, 23, 24 and 25 regions; Case 5: Lesion present in the left mandible in ramus body region; Case 6: Multilocular scalloped lesion present in the right mandible in ramus body region; Case 7: Unilocular lesion in right mandible, in ramus region

with unilocular pericoronal, and two lesions were unilocular envelopmental type of radiolucency. Panoramic radiograph was not conclusive about the internal structure in one of the lesions. In CBCT, six showed unilocular appearance, one with multilocular appearance, and four lesions were unilocular pericoronal radiolucencies with intact follicular spaces [Figure 3 and Table 2].

Effect on adjacent structure

Three of the 11 lesions were not associated with impacted tooth, and eight lesions were associated with impacted teeth. The impacted teeth were third molars in five lesions; in one lesion second premolar; in one

more lesion second molar were involved; in another lesion, two teeth that is lateral incisor and canine was involved. There were no impacted supernumerary teeth in any of the lesions. One out of 11 lesions, panoramic radiograph showed resorption in one, no resorption in nine lesions. In one lesion, panoramic radiograph failed to show the presence or absence of resorption. In CBCT, four lesions clearly showed the presence of resorption, and seven lesions showed no resorption associated with adjacent teeth [Figure 4 and Table 3]. When we compared the duration of symptoms with the presence or absence of root resorption in CBCT, the maximum duration of symptoms (pain or swelling) reported in patients with

Table 3: Radiological features - impacted tooth and root resorption

Case number	Adjacent tooth (panoramic radiograph)	Root (panoramic radiograph)	Adjacent tooth (CBCT)	Root (CBCT)
1	Retained 85, impacted 45 displaced inferior, toward lower border of mandible	Resorption	Retained 85, impacted 45 displaced inferior, and lingually 45 is close to and buccal to IAC	Resorption knife edge In relation to 33, 32, 31, 41, 42, 43, 44
2	Impacted 28 displaced posterior and superior	NAD	Impacted 28 displaced posterior and superior	Root of 27 is displaced mesially
3	Displaced mesially with 23 and distally 24, 25	Not appreciated	Displaced mesially with 23 and distally 24, 25	No resorption
4	Impacted 22, 23 displaced superiorly toward lower border nasal fossa, displaced root of 21 mesially	Incomplete root formation	22, 23 displaced superior floor of nasal cavity Retained 62, 63, mesially displaced root of 21	Root resorption with 62, 63
5	Associated with 37, 38 missing	NAD	Associated with 37, 38 missing	NAD
6	Missing 48	NAD	Missing 48	Knife edge with distal root of 46, 47
7	46 RS, periapical abscess, 47 DDC	NAD	Impacted 48 displaced buccally, inferiorly, and posteriorly	Root resorption with 47

CBCT: Cone-beam computed tomography; IAC: Inferior alveolar canal; CEJ: cemento enamel Junction; NAD: no abnormality detected; DDC: Deep dental caries

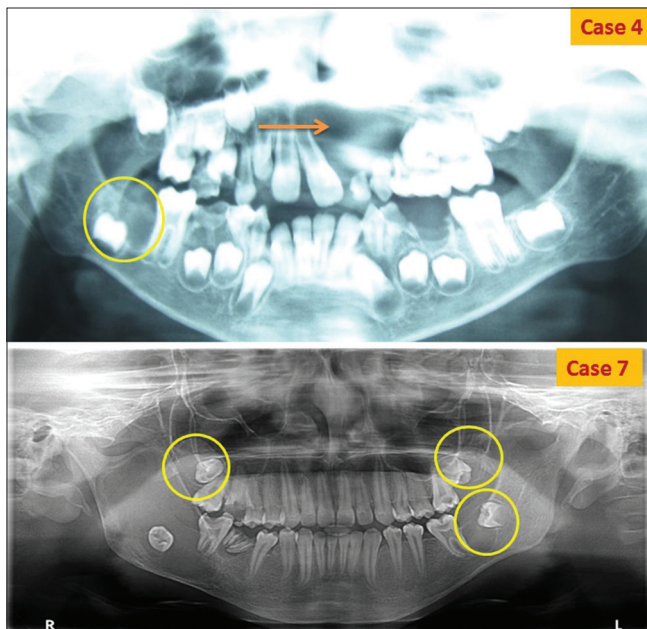


Figure 3: Multiple odontogenic keratocyst; Case 4: In addition to the chief complaint of lesion in left maxilla (yellow arrow), another lesion was incidentally found in right mandible (yellow circle); Case 7: Similarly, apart from chief compliant lesion in right mandible, three more lesions were found in relation to 18, 28, and 38 regions (yellow circle)

root resorption was 4 months; and the patient without root resorption ranged up to 1 year.

Of the six mandibular lesions, five lesions showed the agreement between OPG and CBCT findings, except one where CBCT showed an intact IAC, and OPG showed the inferior displacement of IAC. Moreover, CBCT could give additional information on the buccal or lingual displacement of IAC and any erosion of the cortical boundaries.

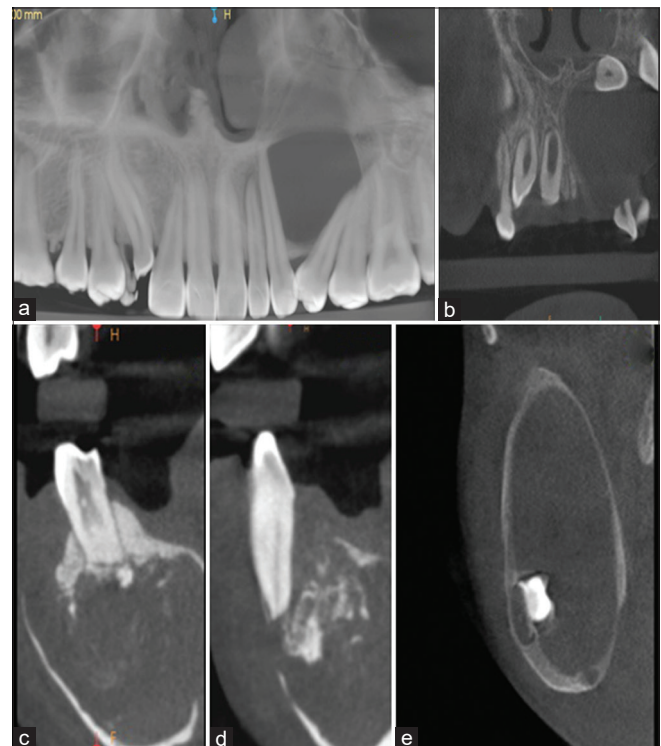


Figure 4: Radiological features-impacted tooth and root resorption. (a) root of 23 displaced mesially and 24, 25 distally; (b) 22, 23 are impacted and displaced superiorly; (c and d) showing root resorption; (e) Impacted 48 displaced buccally, inferiorly and posteriorly

Of the five lesions affecting maxilla, no effects on maxillary sinus or nasal floor in four lesions, and the effect of lesion on maxillary sinus was not clearly defined in one lesion as seen on panoramic radiograph. CBCT clearly showed disruption of one of the walls, perforation, thickening of mucosa in four lesions, and unaffected walls in one lesion [Table 4 and Figure 5].

Table 4: Radiological features - effect on maxillary sinus/nasal floor and inferior alveolar canal

Case number	Maxillary sinus (panoramic radiograph)	Inferior alveolar canal (panoramic radiograph)	Maxillary sinus (CBCT)	Inferior alveolar canal (CBCT)
1	NA	Displaced inferiorly	NA	Not affected
2	Walls are intact, haziness over inferior portion left maxillary sinus	NA	Obliteration of airspace, posterior wall is disrupted	NA
3	Inferior border is not appreciated	NA	Antero-inferior wall perforated Thickening of mucosa	NA
4	No comments	NA	Disruption of floor of nasal fossa	NA
5	NA	intact	NA	Traceable till the distal aspect of 37
6	NA	Displaced inferiorly, posteriorly	NA	Displaced inferiorly, lingually, superior cortex is eroded
7	NA	Displaced posteriorly	NA	Displaced inferiorly and posteriorly

CBCT: Cone-beam computed tomography; NA: Not applicable

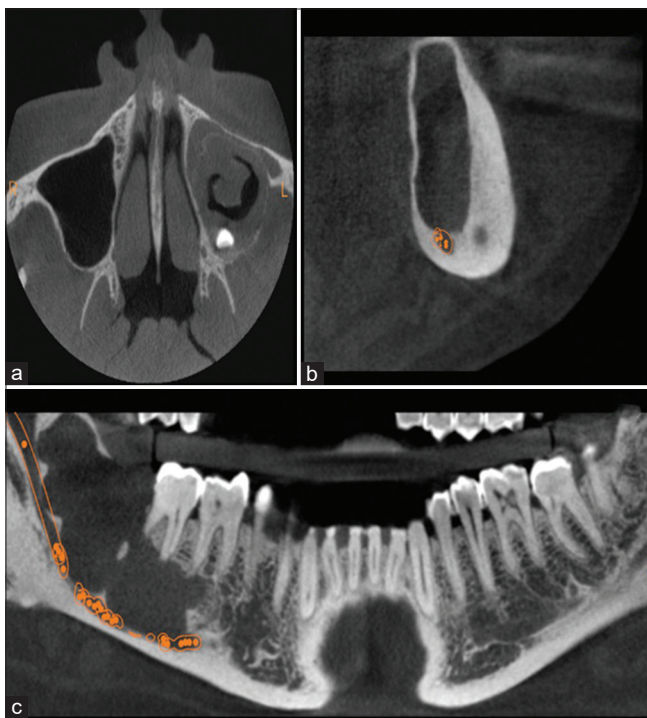


Figure 5: Radiological features-effect on maxillary sinus/nasal floor and inferior alveolar canal. (a) Obliteration of left maxillary sinus; (b and c) Inferior displacement of inferior alveolar canal

In the panoramic radiograph, thinned out lower cortical borders were appreciated in three lesions, no expansion in four lesions, and expansion could not be commented in the remaining four lesions. In CBCT, nine cases showed expansion, and two did not show any expansion. In addition to these details, perforation and thinning of buccal, lingual, and palatal cortices were clearly visible [Figure 6 and Table 5].

Histopathological findings

Histologically, all the cases were characterized by a uniform, usually corrugated parakeratinized epithelium, 8–12 cells thick presenting a flat basal surface lining the fibrous wall. The columnar basal cells showing reversed

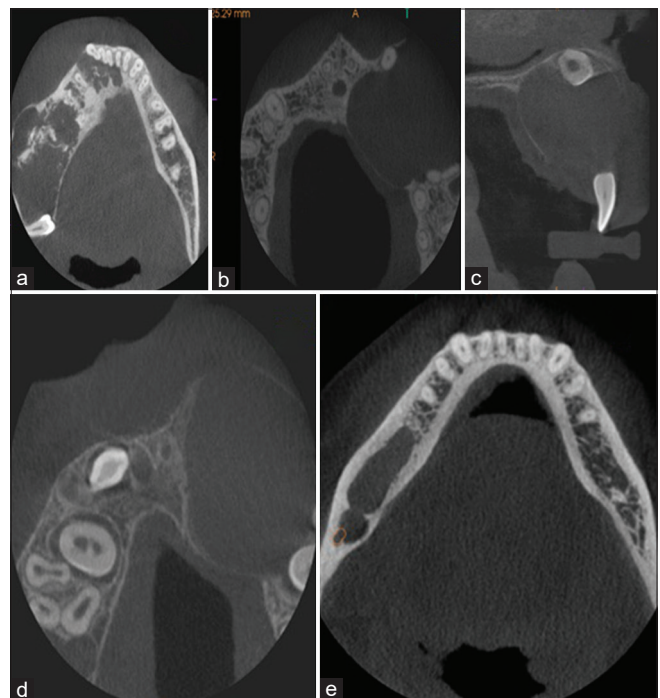


Figure 6: Radiological features-effect on cortical plate. (a) Thinning and expansion of lingual and more of buccal CP in right mandibular lesion; (b) Expansion and perforation of buccal and palatal cortical plate in left maxilla; (c) Expansion and perforation of labial and palatal cortical plates; (d) Significant expansion thinning and perforation of buccal and palatal CP; (e) No CP expansion

nuclear polarity is an important feature, and satellite cysts were found in some lesions.

Discussion

The occurrence of OKC in our institution is 3.6/years. This was more than that of reported in the Hong Kong population, which was 2.1/years.^[8] MacDonald-Jankowski expressed that these patients displayed differences between global groups, patients of East Asian origin may present early symptoms, while in the Western community, it may be found as an incidental finding at a later stage of the disease.^[9]

Table 5: Radiological features - effect on cortical plate

Case number	Cortical plate (panoramic radiograph)	Cortical plate (CBCT)
1	Lower border mandible is perforated	Thinning and expansion of lingual and more of buccal CP Perforation lingual CP (anterior) Sporadic irt buccal CP
2	Cannot comment	Expansion, thinning, and perforation, especially in the posterior aspect of buccal CP
3	Cannot comment	Expansion, thinning, and perforation of buccal and palatal CP
4	Cannot comment	Significant expansion thinning and perforation of buccal and palatal CP
5	Intact	No expansion Thinning perforation of buccal and lingual CP, posteriorly
6	Thinning of inferior border of mandible	Thinning of inferior border of the mandible and buccal and lingual CP
7	Thinning of anterior and posterior ramus, inferior border	Expansion, thinning, and perforation of buccal and lingual CP

CBCT: Cone-beam computed tomography; CP: Cortical Plate

Radiologic diagnosis of a lesion depends on the location, internal structure, extension, cortical involvement, periosteal alterations, and relation to the adjacent structures within the jaw. MacDonald-Jankowski and Li evaluated the clinical and conventional radiological features of 33 cases affecting a Hong Kong Chinese community and concluded that those lesions that presented early were significantly associated with unerupted teeth. They also concluded that multilocular lesions had a significant predilection for older patients, longer in mediobuccal extension, whereas unilocular lesions had a predilection for the maxilla.^[8] The OKCs may begin as unilocular lesions and gradually become multilocular as proposed by Haring and Van Dis in their study.^[10] OKCs show minimal buccolingual expansion as compared to ameloblastoma, assuming a fusiform shape rather than the balloon expansion more of the ameloblastoma. Contrary to this, nine lesions showed expansion in CBCT in our cases. One among the 11 lesions showed extensive mandibular involvement crossing midline although lesion was extensive, the patient reported with the swelling of 4-month duration and pain about 2 months only.

In our case series, two cases [Case 4 and Case 7] had multiple OKCs where the additional lesions were detected incidentally, and patients had no signs and symptoms with respect to additional lesions [Figure 3]. Further investigations were done to evaluate for the presence of other systemic abnormalities to rule out of nevoid basal cell carcinoma syndrome and found that both the cases were solitary (or sporadic) or nonsyndromic variety. According to a systematic review, solitary OKC is the most common accounting nearly 94% of all OKCs, and the global recurrence rate of solitary OKC is 28%.^[11]

Panoramic and intraoral radiographs are the basic imaging methods in the diagnosis of OKC. In our case series, one case of incidental finding and two cases where multiple OKC were first detected in panoramic radiography, thus establishing the importance of scout radiographs. Although panoramic radiography is the primary choice of radiologic

examination for dentists, the superimposition of craniofacial structures generates images with limitations, distortions, and magnifications. Thus, for detailed examinations of the OKC, CBCT can be advised to know the anatomical extensions and dimensions. The risk of injury to adjacent structures such as teeth, mandibular canal, maxillary sinuses, and cortical plates can be evaluated, with reduced radiation dose and cost as compared to multidetector computed tomography (MDCT). The smaller device size and shorter scanning time in CBCT allow it to be a convenient appliance as compared to MDCT. Shweel *et al.* compared the accuracy of CBCT and MDCT in the preoperative radiological assessment of odontogenic cysts and tumors and found that both CBCT and MDCT were identical in detecting location, borders, and internal structure of examined lesions. They observed that the CBCT was more accurate in linear measurements, identification of tooth displacement, and buccal bone defect and concluded that CBCT is an optimal radiological modality for preoperative radiological assessment of odontogenic tumors.^[12]

However, despite all these advantages, CBCT was unable to discern the contents of the lesion. CBCT is insufficient for displaying the contrast within soft tissue, even though, the dynamic range of CBCT has been increased from 8-bit to 14-bit depth. Sometimes, failure to consider the whole image may result in missed neoplasm.^[6] Furthermore, a review by MacDonald revealed that CBCT has little to contribute to the imaging of malignancy and its subsequent management. In such patients, MDCT, magnetic resonance imaging (MRI), and positron emission tomography can be used for appropriate imaging.^[5]

Conclusion

OKC, being an aggressive lesion with varied radiographic appearance, may resemble ameloblastoma, dentigerous cyst, lateral periodontal cyst, and radicular cyst. The presurgical assessment with radiological information is extremely important for shaping the surgeon's approach

for treatment planning, as incomplete removal may lead to recurrence. CBCT provides us with an accurate and faster 3-dimensional representation of a lesion at a lower dose and cost. Nevertheless, the role of 2-dimensional radiographs like panoramic radiograph as well as other advanced imaging such as MRI and MDCT cannot be refuted.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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