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RADIOGRAPHIC ASSESSMENT OF KERATOCYST ODONTOGENIC TUMOR IN MAXILLA USING CBCT
(Case Report)

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ABSTRACT

Keratocyst odontogenic tumor (KCOT) is a developmental odontogenic cyst of epithelial origin. This lesion shows features of a cyst and a benign neoplasm, because of its behavior, autonomous growth, and potential for recurrence. The KCOT occurs more significant in the posterior mandible than in maxilla. Occasionally, pain, swelling, and drainage indicate a secondary infection of the cyst. Asymptomatic KCOT usually detected in routine radiograph. The radiographic examination is important to determine KCOT diagnose and treatment planning to prevent recurrence. The aim of this case report is to describe radiographic characteristic of keratocyst odontogenic tumor in maxilla using CBCT. A 20 year-old women patient was referred to the oral maxillofacial radiology department of Padjadjaran University with the chief complaint of swelling, painless in the anterior of the upper jaw. In this presented case, we used cone beam computed tomography to find out the margin of the cortical extension, and diameter of the lesion. The CBCT examination shows radiolucent, well-defined lesion in 12-14 region with displacement of 12. The size of the lesion is about 20x15x19mm extended posterior-superiorly near to nasal cavity and it shows less degree of bone expansion. Based on radiographic and clinical examination, the diagnosis was keratocyst odontogenic tumor. KCOT has some radiographic characteristic distinguishable with another odontogenic lesion. Therefore; cbct examination is recommended for the diagnosis of odontogenic keratocysts and proper surgical planning.

Key word: radiographic characteristic, odontogenic keratocyst, CBCT

ABSTRAK

Keratocyst odontogenik tumor (KCOT) merupakan kista tumbuh kembang yang berasal dari epitel. Lesi ini menunjukkan ciri khas kista dan tumor jinak, karena sifatnya yang memiliki perkembangan otonom dan berpotensi memiliki rekurensi. KCOT terjadi lebih banyak pada posterior mandibula daripada maksila. Gejala klinis seperti nyeri, pembengkakan, dan drainase spontan mengindikasikan adanya infeksi sekunder. KCOT yang tanpa gejala biasanya terdeteksi secara tidak sengaja pada pemeriksaan radiograf rutin. Pemeriksaan radiografi penting untuk menegakkan diagnosis KCOT dan menentukan rencana perawatan untuk mencegah rekurensi. Tujuan dari laporan kasus ini adalah untuk menggambarkan karakteristik radiograf KCOT pada maksila menggunakan *CBCT*.
Pasien wanita 20 tahun dirujuk ke Departemen Radiologi Oral dan Maksilofasial Fakultas Kedokteran Gigi Universitas Padjadjaran dengan keluhan utama pembengkakan, dan tidak sakit pada bagian anterior maksila. Pada kasus ini dilakukan pemeriksaan *CBCT* untuk menentukan batas perluasan tepi kortikal dan luas lesi. Pada pemeriksaan CBCT menunjukkan adanya gambaran lesi radiolusen, berbatas jelas dan tegas pada regio gigi 12-14, disertai adanya *displacement* pada 12. Ukuran lesi sekitar 20x15x19mm meluas ke arah posterior-superior mendekati kavitas nasal dan menunjukkan ekspansi tulang yang kecil. Berdasarkan pemeriksaan radiograf dan klinis, maka ditegakkan diagnosis pada kasus tersebut adalah KCOT. KCOT memiliki suatu karakteristik gambaran radiografi yang dapat digunakan untuk membedakan dengan lesi odontogenik lainnya. Pada kasus ini, pemeriksaan *CBCT* dapat digunakan untuk penegakkan diagnosis KCOT dan perencanaan perawatan bedah selanjutnya.

Kata kunci: karakteristik radiograf, *keratocyst* odontogenik tumor, *CBCT*.

INTRODUCTION

Keratocyst odontogenic tumor (KCOT) is benign lesions of the maxilla-mandibular region which is derived from the remnants of dental lamina and proliferation of cells from the basal layer of oral epithelium into the mandible or maxilla[1]. The term odontogenic keratocyst was first used by Phillipsen in 1956, while Pinnborg and Hansen describe the essential feature of KCOT in 1963[2]. The prevalence of Keratocyst odontogenic tumor is about 22,65% of all odontogenic tumor and predominance of males 55,17%.[3] In a retrospective study of KCOT, 70,5% occured in the mandible, 16.4% occurred in the maxilla, and 13.1% occurred in both.[4] KCOT can occur over a wide age

range, varying from the first to the ninth decade of life. Its peak incidence is during the second and third decades of life.[5]

In 2005, the World Health Organization has reclassified odontogenic keratocyst as a benign tumor, because KCOT shows features of both a cyst and a benign neoplasm.[1,6,7] The KCOT is unique among odontogenic cyst, because of its pathognomonic microscopic features, destructive and invasive behavior, and high recurrence rate (25% to 62.5%).[1,2,8] KCOT often tend to recur after treatment because of the thin, friable wall which is often difficult to enucleate from the bone in one piece and the presence of small satellite cysts within the fibrous wall.[9]

There is no characteristic clinical manifestation of the KCOT, it can be symptomatic or asymptomatic. Asymptomatic KCOT usually detected in routine radiograph. In this case radiographic modalities play a major role to determine the diagnosis, because KCOT has a characteristic in radiographic. The radiograph examination that usually used to determine KCOT diagnosis is conventional radiograph, CT scan, and CBCT. Now days, cone beam computed tomography (CBCT) is a medical imaging technique that popular in treatment planning and diagnosing in oral and maxillofacial diseases[10]. A major advantage of CBCT that has been reported is the three-dimensional geometric accuracy compared with conventional radiographs. Sagittal, coronal and axial view of CBCT images can eliminate the superimposition of anatomical structures. Besides, the advantages of this technique are relatively high isotropic spatial resolution of osseous structures with a reduced radiation dose and low cost compared with conventional computed tomography (CT) scans[10,11]. The aim of this case report is to present cases of maxillary keratocyst odontogenic tumor in 20 years old women with radiographic finding in CBCT.

CASE REPORT

A 20 year-old women patient was referred with the chief complaint of swelling in the front portion of the upper jaw since three weeks. The swelling was bony hard on extra oral examination. Intraoral examination was painless and revealed expansion of buccal cortical bone, with egg-shell crackling on palpation. To visualize the lesion in more detail, the CBCT was performed. Cone beam computed tomography shows a three dimensional image of both jaws (fig. 1). In coronal view shows radiolucent, well-defined, corticated lesion in 12-14 region. The lesion extended mesial-distal around 19,9mm, and superior-inferiorly around 20,1mm. Superior border of the lesion nudge with the inferior border of concha nasal (fig. 2). In axial tomographic view, shows the lesion extended anterior-posterior around 18,3mm (fig. 3). The image showed a multi-locular cavity and scalloped border in the right side of the maxilla (fig.4). Sagittal and axial image (fig. 3,5,6) shows a balloon like expansion, which is indicated a lesion may be a cyst. In sagittal tomographic view, the lesion shows cortical expansion at the vestibular side of the maxilla (fig 5 and 6). The lesion has caused displacement of 12 in distal-palatal. There was no radiographic sign of root resorption of the adjacent teeth.

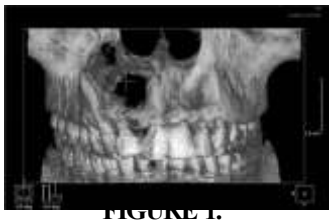

FIGURE 1.
3D image of the lesion in maxilla

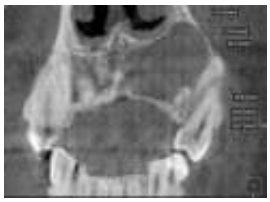

FIGURE 2.
Cbct coronal view, showing radiolucent, well-defined, corticated lesion, extended in 11-14 region

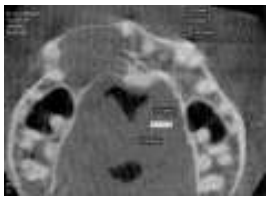

FIGURE 3.
CbCt axial view, showing radiolucent, well-defined, corticated lesion, extended in 11-14 region, the lesion causing an expansion of buccal cortical bone.

FIGURE 4.
CbCT panoramic view show radiolucent, multilocular, well defined, scalloped border in 11-14 region

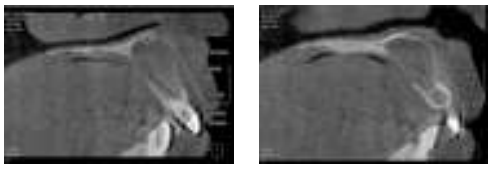

FIGURE 5 and FIGURE 6.
Cbct sagittal view shows a balloon like expansion

DISCUSSION

KCOT arise from derivatives of the dental lamina, an embryonic structure that normally differentiate into tooth buds and enamel producing cells during odontogenesis, whose pathogenesis is not associated with inflammatory stimuli. Some studies report that there is a relation between the alteration tumor suppressor genes occurs on chromosome 2q22.3-q31.36 and the development of KCOT.[12,13,14] The tumor suppressor genes involves in the development of KCOT were p16 (75%), p53 (66%), PTCH (60%), and MCC (60%).[14] The pathogenesis of KCOT involves 2- hit mechanism that associated with allelic loss at 9q22. The 2-hit mechanism refers to the process by which a tumor suppressor gene is inactivated. Inactivation of tumor suppressor genes such as p16, p53, and PCTH cause an inhibition of apoptosis that leads the development of KCOT.[12,13] Besides genetic factors, numerous studies suggest that disregulation of cell cycle and proliferation may be important for KCOT pathogenesis. It is believed that KCOT show increased cell proliferation rates and that such a phenomenon may be related to its aggressive growth. The protein which is involves in cell proliferation of KCOT are PCNA and Ki-67.

Clinically, KCOT doesn't show a special characteristic. Occasionally, swelling, pain, and drainage appear in the late stage, which the KCOT reach a large size or the cyst is infected. An important characteristic of KCOT is its propensity to grow along the internal aspect of the jaws, causing minimal expansion. It was reported that swelling and pain were more frequent in East Asians, whereas KCOT discovered as an incidental finding was more frequent in the Latin American reports15. Some study reported that 37.2% of patients presented with symptoms and 62.8% were asymptomatic with KCOT found incidentally.[16] In the present case, the KCOT causes an unusual clinical sign of buccal cortical expansion despite its small size.

It is also well-known that KCOT frequently recur after enucleation, particularly within first 5 years. Some studies suggested three mechanisms responsible for recurrence: remnants of dental lamina within the jaws not associated with the original KCOT, incomplete removal of the original cystic lining and cortical perforation with adherence to adjacent soft tissue, and cell rests of dental lamina and satellite cysts that remain behind after enucleation[5]. The treatment decisions are based on the age and health of the patient, size, and location of the lesion, involvement of the cortical bone, and presence of important anatomical structures in close proximity to the lesion. To avoid recurrence of the KCOT, we need a radiographic examination.

The radiography examination is important to determine KCOT diagnose and treatment planning. In this case we choose CBCT as imaging modalities. CBCT is a better imaging modality which can be used for the assessment of evaluating cysts or benign tumors. Its offer a real-size data set with multi-planar cross-sectional and 3D reconstructions which are based on a single scan and the internal structures of the pathologic lesions can be investigated by using a low radiation dose.[17] Multi-planar sections (axial, coronal, and sagittal planes) are required when cysts or tumors which are located deep in the tissues. CBCT is also helpful in post- surgical follow-ups of lesions that may have high recurrences, because its offer accuracy in measurement, absence of image distortion, and accuracy the border of the lesion.[17,18] In the present case, CBCTs revealed expansion and thinning of buccal cortical bone, and thinning of nasal cavity floor, contributing to the selection of initial conservative treatment. Moreover, the risk of injury to adjacent anatomic structures, such as teeth, nasal cavity, and the risk of maxilla fracture are clearly identified through CBCT images.

KCOT has characteristic radiographic features such as a radiolucent lesion with corticated and scalloped border, minimal expansion, especially toward medial side, and growth along the internal aspect of the mandibular or maxilla bone. Usually, the lesion causes displacement of the adjacent teeth, resorption of the adjacent teeth roots, and extrusion of erupted teeth. Occasionally, the KCOT present as multiple or bilateral cysts, suggestive of basal cell nevus syndrome. This characteristics radiograph is shown in this case. In the present case, the lesion cause displacement of 12 teeth, but it doesn't cause the resorption of the adjacent teeth roots.

KCOT is often associated with an un-erupted tooth. In 25 to 40% of cases, an un-erupted tooth is seen in association with the KCOT. When associated with un-erupted tooth, a KCOT may be indistinguishable from a dentigerous cyst. The lesion is likely to be a KCOT if the cystic outline is connected to the tooth at a point apical to the cement-enamel junction or if no expansion of the cortical plates has occurred, while the lesion likely to be a dentigerus cyst, if the cystic outline is connected to the tooth at a cement-enamel junction and form around the crown. But in this case, the KCOT doesn't associate with un-erupted teeth. The differential diagnosis of KCOT is also includes ameloblastoma, but usually KCOT tends to grow along the bone without marked expansion, which is characteristic of ameloblastoma. This tumor frequently has internal cystic components, these septa are often remodeled into curved shapes providing honeycomb (numerous small compartments) or soap bubble (larger compartments of variable size) patterns.

CONCLUSION

KCOT shows features of both a cyst and a benign neoplasm and it has some radiographic characteristic distinguishable with another odontogenic lesion. Therefore, a CBCT examination is useful for the diagnosis of keratocyst odontogenic tumor and also for proper surgical planning, because CBCT provided detailed information for evaluation of the lesion borders and the relation of the lesions with adjacent anatomic structures.

REFERENCE

1. Freitas DA, Daniela AV, Alisson LDS, Vinícius AF. Maxillary odontogenic keratocyst: a clinical case report. RGO, Rev Gaúch Odontol, Porto Alegre. 2015 v.63 (4)
2. Chkoura A, Chbiheb S, El wady, W. Keratocyst odontogenic Tumor: a case report and review of the literature. The Internet Journal of Dental science.2008. Vol 6 (2).
3. Ramachandra, S. Poosaria CS, Srinivas P, Krrba KK, Gontu SR, Kantheti LP, Baddam VRR. Prevalence of odontogenicc cyst and tumors : a retrospective clinic pathological study of 204 cases. SRM Journal of Research in dental science. 2014.vol 5.p:170-173.
4. González-Alva P, Tanaka A, Oku Y, Yoshizawa D, Itoh S, Sakashita H, Ide F, Tajima Y, Kusama K. Keratocystic odontogenic tumor: a retrospective study of 183 cases. J Oral Sci. 2008, 50(2):205-12.
5. Suma NK, Pinky C, Venkatesh BNS, Jha S. Odontogenic keratocyst of maxillary premolar region: case report. IJSS casereport & review. 2015. Vol 1 (9).
6. Kargahia N, Kalantari M. Non-Syndromic Multiple Odontogenic Keratocyst: A Case Report. J Dent (Shiraz). 2013. Vol 14(3): 151–154.
7. Rabelo GD, Guimaries H, Jose HM, Silva CJ, Sergio VC, Adriano ML,Antonio FDJ. Non-syndromic Keratocyst Odontogenic Tumor Involving the Maxillary Sinus; case Report. International archives of otorhinolaryngology. 2010. Vol 14 (2).
8. Jardim, ECG, Rossi AC. Faverani LP, Ferreira G R, Ferreira M B, Vicentes LM, Junior IGR. Odontogenic keratocyst tumor: report of two cases. Int. J. Odontostomat. 2013. Vol 7(1):33-38.
9. Kalaskar RR, Kalaskar AR, Chetan AP, Suvarna K G. Keratocystic odontogenic tumor invading the left maxilla: A rare case report. SRM Journal of research Dental Science. 2013. Vol (4) 3: 132-134.
10. Banik S , Samir B , Shaikh MH , Sadat SMA , Mallick PC. Keratocystic odontogenic tumor and its radiological diagnosis by 3 dimensional Cone Beam Computed Tomography (CBCT). Update Dent. Coll. J. 2011. Vol 1(1): 10-13.
11. Berberoðlu HK, Sýrmahan Ç, Amila B, Banu GK, Barýþ AA, Cengizhan K. Three-dimensýonal cone-beam computed tomography for diagnosýs of keratocystic odontogenic tumours; Evaluation of four cases. Med Oral Patol Oral Cir Bucal. 2012. Vol 17(6): 1000–1005.
12. Madras J, Lapointe H. Keratocystic Odontogenic Tumour: Reclassification of the Odontogenic Keratocyst from Cyst to Tumour. JCDA • www.cda-adc.ca/jcda. 2008. Vol. 74 (2)
13. Andriæ M, Brkoviæ B, Jurišiæ V, Jurišiæ M, Milašin J. Keratocystic Odontogenic Tumors – Clinical and Molecular Features. A Textbook of Advanced Oral and Maxillofacial Surgery. 2013.
14. Agaram NP, Collins BM, Barnes L, Lomago D, Aldeeb D, Swalsky P, Finkelstein S, Hunt JL. Molecular Analysis to Demonstrate That Odontogenic Keratocysts Are Neoplastic. Arch Pathol Lab Med. 2004. Vol 128
15. Sumer AP, Sumer M, Celenk P, Danaci M, Gunhan O. Keratocystic odontogenic tumor: case report with CT and ultrasonography findings. Imaging Sci Dent. 2012. Vol 42(1): 61–64.
16. Boffano P, Ruga E, Gallesio C. Keratocystic odontogenic tumor (odontogenic keratocyst): preliminary retrospective review of epidemiologic, clinical, and radiologic features of 261 lesions from University of Turin. J Oral Maxillofac Surg. 2010. Vol 68:2994–2999.
17. Prabhusankar K, Yuvaraj A, Prakash CA, Parthiban J, Praveen B. CBCT Cyst Leasions Diagnosis Imaging Mandible Maxilla. J Clin Diagn Res. 2014. Vol 8(4): ZD03–ZD05.
18. Macdonald-Jankowski DS. Focal cemento-osseous dysplasia: a systematic review. Dentomaxillofac Radiol. 2008. Vol 37:350–60.