Keratocystic Odontogenic Tumor of the Maxilla—A Serious Entity often misdiagnosed: A Report of Two Cases resembling Dentigerous Cysts

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ABSTRACT

The most common maxillary location for a keratocystic odontogenic tumor (KCOT) is the canine region where they commonly are mistaken for an inflammatory radicular cyst or dentigerous cyst or a lateral periodontal cyst or even a nasopalatine cyst. This misdiagnosis occurs mainly because of the appearance of KCOT as a unilocular radiolucency in the maxilla, particularly if the KCOT is found coincidentally with a nonvital tooth. Additionally, the cyst is frequently infected producing pus that obscures the typical white cheesy material. A misdiagnosis based solely on clinical information can lead to the possibility of the patient being treated with a conservative endodontic therapy or even conservative surgical techniques thereby greatly increasing the chances of progression or recurrence of this aggressive lesion. Here, we report two such cases diagnosed and treated aggressively.

Keywords: Keratocystic odontogenic tumor, Diagnosis, Unilocular radiolucency.

INTRODUCTION

Inflammatory and developmental cysts of the jaw are entities that dentists frequently diagnose in clinics and the possible explanation is the abundance of odontogenic epithelium remnants found in the jaws. The periapical cyst is the most common odontogenic cyst (52.3-70.7% of all odontogenic cysts) followed by the dentigerous cyst (16.6-21.3% of all odontogenic cysts) and keratocystic odontogenic tumor or KCOT (5.4-17.4% of all odontogenic cysts). Keratocystic odontogenic tumor (KCOT) is thought to arise from the dental lamina remnants in the mandible and maxilla. Origin of the KCOT from basal cells of the overlying oral epithelium has also been reported. KCOT generally occurs as a multilocular radiolucency. But a KCOT in the maxilla appears as a unilocular radiolucency, often in a dentigerous relationship. The presence of infection and the coincidental finding of a nonvital tooth can often lead to a misdiagnosis of an inflammatory radicular cyst or dentigerous cyst or a lateral periodontal cyst. Treatment protocol is different for each lesion and the KCOT should be treated aggressively because of the high-risk of recurrence of the lesion (Fig. 1).

CASE REPORT

A 35-year-old female patient of Indian origin came to the department of oral medicine and radiology with a swelling of the middle-third of the face on the right side since 1 month.

Fig. 1: Photomicrograph of biopsy specimen showing the corrugated surface and palisaded appearance of the basal cells. Keratin squames are also seen
The swelling was very small initially and it progressively enlarged to attain the present size. Patient also gives history of pain associated with the swelling since 13 days, which was mild, continuous, localized and dull aching in nature. The patient had consulted a medical practitioner for the same 10 days back and she was prescribed pain killers.

Extraoral examination revealed a solitary diffuse swelling of the middle-third of the right side of the face that extended from the infraorbital margin to the upper lip superioinferiorly measuring about 3 to 3.5 cm (Fig. 2) and from the ala of the nose medially to about 2 cm away from the tragus of the ear laterally measuring about 3.5 to 4 cm. The nasolabial fold was obliterated. On palpation the swelling was tender, soft in consistency and fluctuant. Intraoral examination revealed a solitary diffuse swelling that obliterated the upper right vestibule in the canine, premolar, molar region and it was tender on palpation (Fig. 3). 15 was partially impacted and 13, 14, 15 were non-responsive to electrical pulp testing. Draining sinuses were seen in relation to 13. On radiographic examination a well-defined unilocular radiolucency measuring about 3 cm in diameter surrounding 15 exerting pressure effects on roots of 13, 12, 11 and eroding the floor of the nasal fossa was seen on the panoramic, PNS and maxillary cross-sectional occlusal views (Figs 4 to 6). Aspiration yielded about 1 ml of thick, cheesy material tinged with pus and blood with a dirty whitish yellow color. The aspirate was sent for cytological analysis. A provisional diagnosis of KCOT was given keeping in mind the cheesy consistency of the aspirate but a differential diagnosis of dentigerous cyst could not be ruled out. A final diagnosis of KCOT was given only after the biopsy was histopathologically analyzed.

A similar case of a unilocular radiolucency that had the appearance of a dentigerous cyst in relation to an impacted left canine which revealed to be a KCOT of the maxilla only on histopathological examination of the cystic lining is also...
reported here. A 15-year-old female patient with a swelling of the middle third of the left side of face since 1 month which was small initially and it progressively enlarged to attain the present size. Patient also gives history of pain associated with the swelling since last 5 days, which was mild, continuous, localized and dull aching in nature. The patient also had a swelling of the lower 3rd of the right side of the face which was accompanied by a similar kind of pain. Extraoral examination revealed a gross asymmetry of the face with swelling on both sides. A solitary diffuse swelling of the middle-third of the left side of the face that extended from 1 cm below the infraorbital margin to the upper lip supero-inferiorly measuring about 2 to 2.5 cm and from the ala of the nose medially to the zygoma laterally measuring about 1.5 to 2 cm. The swelling on right side of face measured 4.5 cm mediolaterally and 3 cm supero-inferiorly. On palpation both the swellings were tender, soft in consistency and fluctuant. Intraoral examination revealed a solitary diffuse swelling that obliterated the upper left vestibule in the canine, premolar, molar region and it was tender on palpation. 23 was impacted and there were overretained deciduous 63, 64 and 65. Panoramic radiograph revealed a well-defined unilocular radiolucency measuring about 3 cm in diameter surrounding 23 in close proximity to roots of 63, 64, 65 and eroding the floor of the nasal fossa. A multilocular radiolucency is seen on the right side extending from the body involving the ramus and extending up to the neck of right condyle measuring about 8 × 3 cm. The radiolucency is associated with a radiopacity near the neck of condyle suggesting 48 that has been displaced.

The KCOTs of both the patients were aggressively treated with surgical enucleation and application of Carnoy’s solution. Both patients healed uneventfully and are on regular follow-up, 1 year postsurgically, there has been no recurrence of KCOT.

DISCUSSION

The odontogenic keratocyst was first described by Philipsen in 1956. In 1967 Toller suggested that KCOT may be best regarded as a benign neoplasm rather than a conventional cyst based on its clinical behavior. The odontogenic keratocyst is now designated by the World Health Organization (WHO) as keratocystic odontogenic tumor and is defined as ‘a benign uni or multicystic intraosseous tumor of odontogenic origin with a characteristic lining of parakeratinized stratified squamous epithelium and a potential for aggressive infiltrative behavior’.5

KCOTs are commonly seen in the second and third decades.6 The frequency is higher in males than females.2 The mandible is involved much more frequently than the maxilla in a 2:1 ratio. The most common site is the posterior portion of the body or ramus of the mandible but the anterior portion of the maxilla, the maxillary third molar area and the maxillary antrum also have been involved less frequently.7 The cyst may appear as unilocular or multilocular radiolucency with distinctly corticated, often scalloped border often expansion, especially toward the lingual (medial) side and growth along the length of the mandibular bone with displacement of developing teeth and/or separation or resorption of the roots of erupted teeth and extrusion of erupted teeth. Sometimes a radiolucent lumen and occasionally a cloudy or milky appearance of the lumen on the panoramic radiograph are observed. Occasional presence of multiple or bilateral cysts is evident.8 KCOT has an unusually high recurrence rate that ranges from 5 to 62.5%. Studies have shown that the recurrence rate of keratocysts treated with enucleation was as high as 12%.7 Browne evaluated three different treatment methods, which were marsupialization, enucleation and primary closure and enucleation and packing open and concluded that there was no correlation between treatment method and the rate of recurrence.6 Presence of additional remnants of dental lamina, from which a cyst might develop, presence of satellite cysts and a thin friable epithelium in KCOT all owe to the high recurrence rate and the reason why it develops more aggressively than any other jaw cyst.9 The recurrence rate is almost comparable to that of the ameloblastoma.10 Aspiration from the lesion yields a cheesy white material that has a protein content of less than 4 gm/dl. The histopathology of KCOT shows epithelial layer that lacks rete ridges. In addition, it has a corrugated parakeratinized luminal layer and a prominent basal cell layer. Palisading of the basal cells and a thin friable epithelium are also seen.11 Treatments of OKCs have ranged from marsupialization and enucleation to en bloc resection. Carnoy’s solution and cryosurgery are also advocated as it kills epithelial remnants and dental lamina in the osseous margin. Carnoy’s solution is a tissue fixative that penetrates bone to a depth of 1.54 mm.12 Differential diagnosis of KCOT in the maxillary anterior region includes lateral periodontal cyst, periapical cyst/granuloma, odontogenic cyst, globulomaxillary cyst, dentigerous cyst, adenomatoid odontogenic tumor, ameloblastoma, central giant cell granuloma and calcifying odontogenic cyst.

CONCLUSION

The anterior maxilla is the most common location for the KCOT in the maxilla. KCOTs are commonly mistaken for inflammatory lesions of endodontic origin or for lateral periodontal cysts. These lesions should be treated aggressively and with caution.13
REFERENCES