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 non vital 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 one such case diagnosed and treated aggressively during the primary treatment of the impacted tooth itself.

Keywords: Impacted maxillary canine, Keratocystic odontogenic tumor, Odontogenic keratocyst, Unilocular radiolucency.

INTRODUCTION
Keratocystic odontogenic tumor (KCOT), formerly known as odontogenic keratocyst (OKC) is described as developmental, benign, but locally aggressive lesion characterized by invasive growth into a neighbor structures with high rate of recurrences etiological factors of KCOT occurrence include dental lamina remnants and extension of basal cells of overlying oral epithelium, although DNA mutations are not excluded. The most common clinical symptoms of tumor presence are localized asymptomatic swelling associated with mobility of the teeth, and spontaneous drainage into the oral cavity, which usually occur in mandibular arch, in association with impacted maxillary canine also.1

An impacted maxillary canine is rarely diagnosed during a routine dental examination. Disturbance in the eruption of permanent maxillary canines can cause problems in the dental arch and adjacent teeth, which require special care and attention.

Clinicians have various definitions of ‘impaction’.2–6 Canine impaction can be defined as an unerupted tooth after its root development is complete; or a tooth still unerupted when the corresponding tooth on the other side of the arch has been erupted for at least 6 months and has a complete root formation; or a condition in which a tooth is embedded in the alveolus and is locked in the bone, adjacent teeth, or other obstacles and cannot properly erupt into the oral cavity. This includes teeth in which eruption is significantly delayed and there is no clinical or radiographic evidence that further eruption is likely to happen.

Maxillary canines are among the last teeth to develop and have the longest period of development. They also have the longest and most devious path of eruption from the formation point lateral of the piriform fossa to the final position in the dental arch. Therefore, there is an increased potential for mechanical disturbances resulting in displacement and impaction.7 Genetic factors are largely responsible for this anomaly. Other causes suggested for canine impactions are usually the results of any one or combination of the following factors: Tooth size-arch length discrepancies, prolonged retention or early loss of the deciduous canine, abnormal position of the bud, dilacerations of the root, ankylosis, cystic or neoplastic formation and the absence of the maxillary lateral incisor. This makes the maxillary canine the second most commonly impacted tooth, after third molars.

Permanent maxillary canine impaction has been reported in about 1 to 2% of the population. The presence of the impacted canine may cause some effects such as migration of the neighboring teeth and loss of arch length, internal resorption, dentigerous cyst formation,
external root resorption of itself as well as the neighboring teeth and combinations of the above sequelae. Potential complications emphasize the need for close observation of the development and eruption of these teeth during the examination of the growing child. Research indicates that women are twice as likely as men to have impacted maxillary canines. The prevalence of impacted maxillary canines is between 0.9 and 2%. It has been found that maxillary impacted canines occur palatally 85% of the time while only 15% of impactions occur labially.7

CASE REPORT

A 40-year-old female was reported to OPD of the Department of Oral and Maxillofacial Surgery, IDS, Bareilly, with a chief complaint of swelling over her right palatal region since 2 to 3 months. The patient gave a history of pus discharge from her palate since 1 month for which she went to some private doctor and took medication. She was asymptomatic for some time but the same problem recurred for which she consulted us for further treatment.

On extraoral examination, there was no relevant findings. On intraoral examination, there was a missing tooth in her upper right quadrant along with palatal swelling. The palatal swelling was extending from 11 to 14 region, roughly oval in shape, measuring 1.5 × 1.5 cm in dimension. On palpation swelling was hard, and non tender. Clinically at this stage, palatally impacted canine was suspected as a provisional diagnosis. An occlusal view was advised, in which a well-defined radiolucency was visualized with palatally impacted canine (Fig. 1). On the basis of the above clinical and radiographic examination, a provisional diagnosis of palatally impacted maxillary canine associated with a dentigerous cyst was made. A palatal splint was made and all blood investigations were done before surgical procedure.

Surgical procedure: A crevicular incision was given and palatal flap was raised from first premolar to first premolar crossing the midline (Fig. 2). Full thickness mucoperiosteal flap was raised and cystic lining was removed which was thick and firmly adhered to underlying
Keratocystic Odontogenic Tumor of the Maxilla Associated with Palatally Impacted Maxillary Canine: A Rare Case Report

Journal of Dental Sciences and Oral Rehabilitation, July-September 2015;6(3):141-144

bone, along with impacted canine in toto, followed by closure of surgical site with the help of 3–0 vicryl (Figs 3 to 5). Postoperatively, palatal splint was placed for about 1 week period. The specimen was sent for biopsy. The histopathological report revealed (Figs 6 and 7). Kerato-
cystic odontogenic tumor which is a rare entity in con-
junction with impacted maxillary canine.

DISCUSSION

The OKC 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 OKC is now designated by the World Health Organization (WHO) as KCOT 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 an aggressive infiltrative behavior’. Keratocystic odontogenic tumor is commonly seen in the second and third decades. The frequency is higher in males than females. 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 are involved less frequently. The tumor may appear as unilocular or multilocular radiolucency with distinctly corticated, often scalloped border with 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 or occasionally a cloudy milky appearance of the lumen on the panoramic radiograph are seen. 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.

Occasional presence of multiple or bilateral tumor is evident. Treatment of OKCs have ranged from marsupialization, enucleation to en bloc resection. Carnoy’s solution and cryosurgery are also advocated as it eliminates 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 at the end of 3 minutes of application. 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. Keratocystic odontogenic tumor 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%. Presence of additional remnants of dental lamina, from which a
tumor might develop, presence of satellite cysts and a thin friable epithelium in KCOT all add to the high recurrence rate and the reason why it develops more aggressively than any other jaw tumor. The recurrence rate is almost comparable to that of ameloblastoma.

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.

CONCLUSION

Canine impaction is a relatively frequent clinical presentation in dentistry, with challenges that should be resolved. A good understanding by the clinician of the situation and treatment options can have a significant impact on the treatment outcome. Therefore, clinicians should be competent to perform the proper investigation, provide a correct diagnosis, develop an optimum treatment plan, and render appropriate treatment for each individual patient so each patient realizes the best outcome possible. Any unilocular or multilocular lesions of either jaw irrespective of the internal structure and type of borders should include a differential diagnosis as KCOT as the typical feature of KCOT may not be present.

REFERENCES