



## Submandibular Abscess due to an Infected Keratocystic Odontogenic Tumor associated with Simultaneous Occurrence of a Traumatic Bone Cyst: A Rare Case Report

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### ABSTRACT

**Aim:** The aim of this report is to introduce a rare case in which an infected keratocystic odontogenic tumor (KCOT) was initially diagnosed and treated as a dentoalveolar abscess.

**Background:** Keratocystic odontogenic tumor (KCOT) is a benign neoplasm that can be secondarily infected. However, cervical soft tissue abscess formation as a result of an infected odontogenic cyst or tumor is a rare condition few of which have only been described in the existing literature. Also, there has been a single report regarding the coincidence of a traumatic bone cyst and a keratocystic odontogenic tumor to date.

**Case report:** The patient was a 29-year-old male, complaining of fever, pain and swelling in the left submandibular region. The panoramic radiography showed a well-defined and partially corticated radiolucency between the roots of the second and third left mandibular molars. In addition, a well-corticated radiolucent lesion was incidentally found on the right side of the mandible, which, following surgical exploration, was diagnosed as a traumatic bone cyst.

**Conclusion:** In the present report, an infected KCOT manifested as a cervical abscess, coincided with a traumatic bone cyst.

**Clinical significance:** From the clinical point of view, it is of paramount significance to prevent misdiagnosis of similar presentations as pulp and periapical lesions, which may lead to mistreatment and thus complications.

**Keywords:** Keratocystic odontogenic tumor, Infected odontogenic cyst, Traumatic bone cyst, Case report.

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### BACKGROUND

Keratocystic odontogenic tumor (KCOT) is a benign neoplasm with a keratinized epithelial lining as well as a high recurrence rate.<sup>1,2</sup> KCOT can be secondarily infected, however; cervical soft tissue abscess formation due to an infected odontogenic cyst or tumor is a rare condition few of which have only been described in the existing literature.<sup>3</sup> Also, there has been a single report regarding the coincidence of a traumatic bone cyst and a keratocystic odontogenic tumor to date.<sup>4</sup> In the present paper, we report a case of cervical soft tissue abscess, arising from an infected keratocystic odontogenic tumor and concurrent with a traumatic bone cyst in a 29-year-old male.

### CASE REPORT

The patient was referred to the Oral Medicine Department of Hamedan Faculty of Dentistry, (Hamedan, Iran) in October 2010, for further investigation of a painful swelling in the left submandibular, with forward extension to the submental area. The lesion was fluctuant and tender on palpation. In addition, a linear surgical scar was seen in the submental region (Fig. 1). On intraoral examination, we found a spontaneous drainage into the oral cavity through a fistula on the mandibular lingual aspect, adjacent to the first molar apex. There was no evidence of buccal and lingual plate expansion, nor of tooth mobility and displacement. There was an amalgam filling in the left first molar. The patient had been complaining of pain, fever and swelling since 2 months before. He subsequently underwent surgical drainage coupled with antibiotic regiment, carried out and prescribed by an otolaryngologist surgeon, following which

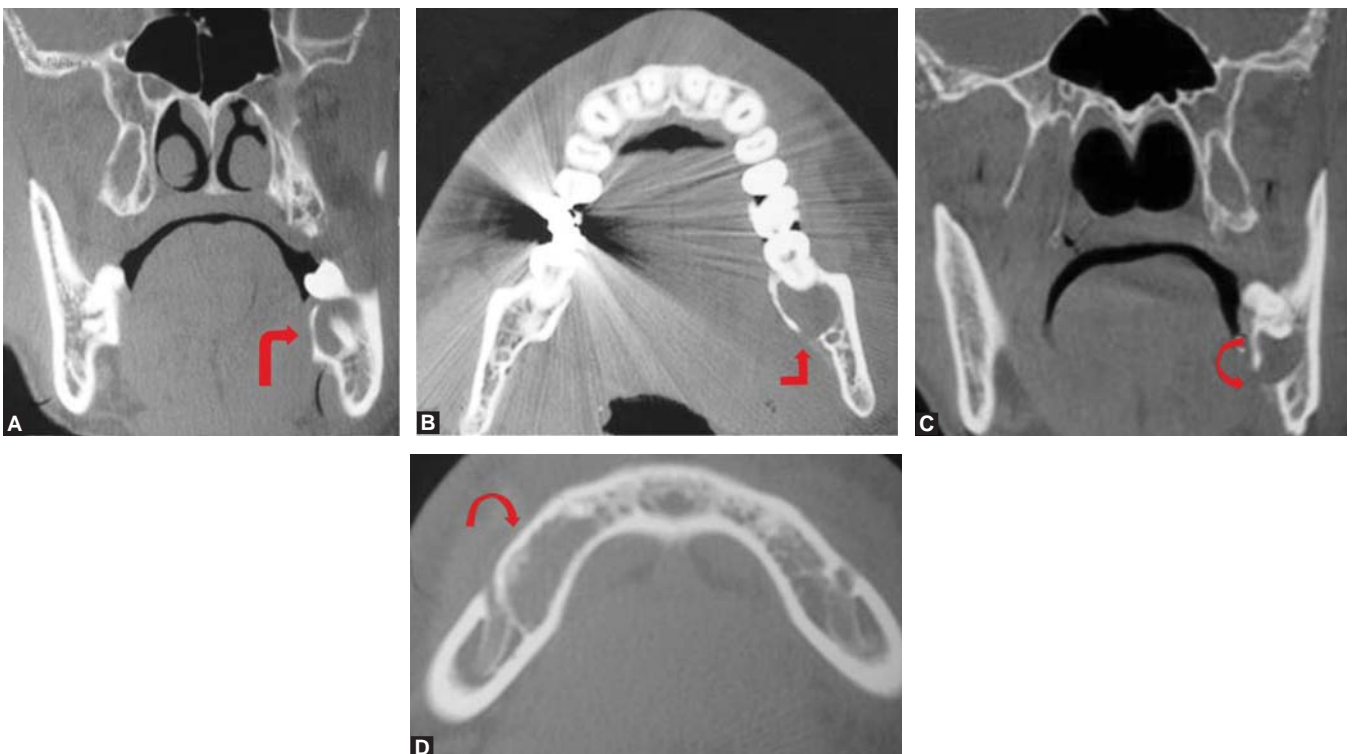


Fig. 1: Swelling in the mandibular left side



Fig. 2: Dental panoramic radiograph indicating two radiolucent mandibular lesions

symptoms subsided. With a differential diagnosis of a dental abscess on the first left molar in mind, the surgeon referred the patient to a dentist afterwards. With the patient failing to do so, symptoms recurred in less than 2 months. Panoramic radiograph revealed a well-defined and partially corticated radiolucent lesion between the roots second and third molars on the left mandible (Fig. 2). Lamina dura was intact in the first molar, but destroyed in the distal root of the second molar and mesial root of the third without any root resorption. Molars on the left mandible were all vital on the vitality test on the pulp tester. In addition, a well-defined and corticated radiolucency, was incidentally found in the right side of the mandibular body. There was neither expansion nor tooth displacement while lamina dura of the involved teeth were intact. Multislice spiral CT scan without contrast enhancement depicted a unilocular cystic lesion in association with the perforation in the medial cortex of the left mandibular molars (Figs 3A to C). These radiological signs were suggestive of the differential diagnoses of a keratocystic odontogenic tumor or a cystic ameloblastoma. Biopsy sections showed a cystic lesion lined with parakeratinized stratified squamous epithelium together with palisading and hyperchromatic basal cell layer and interfacing flat underlying epithelium. Covering epithelium in some areas had an arch shape appearance with exocytosis. Cyst wall connective tissue included dense chronic inflammatory cells infiltration. Axial view showed a well-



Figs 3A to D: (A) Coronal bone window indicating a mild expansion in the medial cortex of the left mandible, (B and C) axial and coronal bone window shows perforation of the mandibular medial cortex, (D) axial bone window shows a non-expansile lesion in the right mandibular body

defined and non-expansile lesion on the opposite side of the mandible (Fig. 3D). On surgical observation an empty cavity without epithelial lining was exposed, proved to be a traumatic bone cyst. Complete surgical removal was performed, including the extraction of the second and third left molars. No clinical signs or symptoms were found in our 6-month follow-up while panoramic radiograph revealed good osseous fill within both lesions (Fig. 4).

## DISCUSSION

The KCOT is one of the benign tumors which are of particular attention due to its high recurrence and aggressive growth. There is no symptom in approximately 50% of cases. Nevertheless, pain, swelling, expansion and drainage have been reported in a number of articles, including ours.<sup>5,6</sup> In many cases, it can be found around an unerupted tooth and thus can easily be misdiagnosed as a dentigerous cyst in clinical investigation. Despite lam and et al having shown that 78% of KCOTs had correctly been diagnosed in clinical examination,<sup>7</sup> our clinical findings namely fever, pain, and a hot and rubbery swelling in superior cervical soft tissues, turned out to be misleading, evoking a diagnosis of dentoalveolar abscess. Although, KCOT can get secondarily infected, cervical soft tissue abscess formation due to an infected odontogenic cyst is a rare condition, with only few articles reported, this complication.<sup>8,9</sup>

Radiographically, KCOTs usually show evidence of a cortical border unless they have become secondarily infected.<sup>10</sup> The cyst may have a smooth round or oval shape or a scalloped outline.<sup>11</sup> The internal structure is most commonly radiolucent.<sup>12</sup> These cysts can manifest as a pericoronal or periapical radiolucency. In pericoronal location, a KCOT may be indistinguishable from a dentigerous cyst.<sup>10</sup>

Periapical radiolucencies often suggest the presence of odontogenic pathosis, usually inflammatory granulomas or

cysts. Some authors have noted that the diagnosis of KCOTs based on radiographic features alone is unlikely to be accurate as it can appear as unilocular radiolucency,<sup>6,13</sup> such as a radicular cyst or periapical granuloma adjacent to a non-vital tooth.<sup>5</sup> However, the epicenter of a radicular cyst, located around at the apex of a nonvital tooth,<sup>10</sup> could be a distinguishing due. In this case, the lesion between the roots of the vital teeth, extending close to the alveolar crest in addition to the radiographic and CT signs evoked the diagnosis of a keratocystic odontogenic tumor or a cystic ameloblastoma. Therefore, periapical radiolucency should not be certainly diagnosed as inflammatory granuloma or abscess, which is normally and subsequently followed by opening and root canal therapy by dentists.

Occasionally, the expansion of large keratocysts may exceed periosteum new bone formation perforating the bone outer cortex.<sup>10</sup> In this case, secondary infection resulted in the disappearance of cortical border in some areas and despite the small size of the lesion, medial cortex perforation ensued. This attests to the aggressive behavior of this lesion. Computed tomography (CT) studies could help to determine the extent of these lesions and detect cortical perforation, which can be helpful in surgical treatment plan.

Our radiological findings, both panoramic radiography and CT scans, enjoyed great precision, underlining the efficacy of the radiological findings in accurate diagnostic of such lesions. Hence, every dental practitioner is advised to update her knowledge of available diagnostic tests namely radiological and differential diagnoses for apical lesions.

However, if routine traditional treatments performed based on initial diagnosis are not effective, biopsy should be considered as confirmatory and possible modification of the treatment plan. Complete surgical removal is the treatment of choice for KCOT. The high recurrence rate requires particular attention to any radiolucent lesion on jaw, necessitating further histological investigation.<sup>6</sup> Authors planned periodic follow-up every 6 months within first 5 years and then annually within 10 years, with regular radiographic examinations to monitor the patient for any signs of recurrence.

To date, there has only been a single report of a concurrent traumatic bone cyst along with KCOT. In our case, an asymptomatic radiolucency was accidentally found on the right side of the mandible, which on surgical exploration proved to be a traumatic bone cyst.

Traumatic bone cyst, recently known as simple bone cyst, is an empty or fluid containing cavity in bones minus epithelial covering. Its pathogenesis is not completely understood. Radiographically, it appears as a lucent lesion



**Fig. 4:** Follow-up panoramic X-ray. Note good osseous filling within both lesions

with a well-circumscribed margin and often scallops between the roots of the teeth, almost always diagnostic.<sup>10,14</sup> It usually reveals no expansion neither tooth movement; these features were reported in few articles though.<sup>15</sup> Surgical exploration was proved not only essential in making the right diagnosis but also curative from a treatment plan perspective.<sup>15</sup> Almost the entire lesions present normal radiographic features after 6 months,<sup>14</sup> as shown in this case.

## CONCLUSION

The present article reported the coincidence of an infected KCOT manifested as a cervical abscess, with a traumatic bone cyst. This report emphasizes the importance of making a firm diagnosis prior to treatment.

## CLINICAL SIGNIFICANCE

As was shown in this patient, KCOT could be misdiagnosed as dentoalveolar abscess, regarding its potential to cause infection and simultaneous drainage. From the clinical point of view, it is important to carry out appropriate para-clinical measures, such as radiography to make a precise diagnosis. Although it is rare, the possible coexistence of traumatic bone cyst with KCOT should be ruled out when multiple radiolucent lesions occur.

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