Maxillofac Plast Reconstr Surg 2014;36(2):73-77
http://dx.doi.org/10.14402/jkamprs.2014.36.2.73
ISSN 2288-8101(Print) ISSN 2288-8586(Online)
Case Report

Deep Neck Space Infection Caused by Keratocystic Odontogenic Tumor

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Abstract

Keratocystic odontogenic tumor (KCOT) is a benign cystic intraosseous tumor of odontogenic origin. An infection of a KCOT is not common because KCOT is a benign developmental neoplasm. Moreover, a severe deep neck space infection with compromised airway caused by infected KCOT is rare. This report presents a 60-year-old male patient with a severe deep neck space infection related to an infected KCOT due to cortical bone perforation and rupture of the exudate. Treatment of the deep neck space infection and KCOT are reported.

Key words: Abscess, Complication, Odontogenic cysts, Odontogenic infection

Introduction

Deep neck space infections usually represent the overgrowth of the normal flora of the contiguous mucosal surfaces from which the infection originated[1]. Deep neck infections are serious emergency conditions. They can spread anywhere and are susceptible to serious complications within hours such as mediastinitis, septic shock with disseminated intravascular coagulation, pleural emphysema, pericarditis, necrotizing fasciitis and life-threatening airway compromise[1,2]. Treatment of deep neck space infection has three main components: airway control, medical management, and surgical intervention according to progress[3].

Odontogenic infections are the most common source (50% to 89%) of infections in the head and neck[1,4,5]. However, infections of odontogenic cysts are very rare. In particular, there are few reports about deep neck space infection caused by keratocystic odontogenic tumor (KCOT), a developmental lesion rather than an inflammatory lesion[6,7], although a retrospective study reported that 2.1% of patients presenting with infection in the head and neck had dentigerous cysts[8].

This report presents an unusual case of a severe deep neck space infection caused by an infected KCOT.

Case Report

A 60-year-old male patient with no significant past medical history was referred to the Department of Oral and Maxillofacial Surgery, Chosun University Dental Hospital in September 2011 for painful swelling in both sub-

RECEIVED July 8, 2013, REVISED August 20, 2013, ACCEPTED January 14, 2014

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mandibular and submental areas a few days prior. He presented with mild fever, chill, malaise, dysphagia and odynophagia. The physical examination was non-fluctuant swelling that was firm on both submandibular, buccal and submental areas extending to the supraclavicular region with severe tenderness, and the tongue was elevated posteriorly.

Laboratory examination showed white blood cell count of $40.69\times10^3/\mu L$, erythrocyte sedimentation rate of 60 mm/h, high C-reactive protein concentration of >30 mg/dL. Further lab exams were abnormal due to severe dehydration.

Contrast-enhanced computed tomography (CT) revealed extensive multilocular gas collection and multiple low-density collections in both submandibular, sublingual, parapharyngeal, around the hyoid bone and extending to pretracheal area (Fig. 1). The airway was displaced to the left and narrowed. Panoramic radiograph and CT evaluation showed a large, moderately defined scalloped radiolucent lesion from the anterior mandible to right mandibular body with partial thinning of the cortical border and bony perforation of buccal and lingual cortex (Fig. 2).

Extraoral incision and drainage on both submandibular and submental area and the marsupialization of cyst were performed under general anesthesia. A rupture of the cyst wall was found. Thicker yellowish white creamy exudates seeming like keratin mixed with pus were found in the sublingual and submandibular space. The creamy exudates were removed with blunt dissection of the space as much as possible. The marsupialization with nasopharyngeal tube was performed on the buccal side. The pus culture yielded gram-positive cocci and gram-negative rods. The patient was treated with intravenous antibiotics: ampicillin, metronidazole and isepacin according to antibiotics sensitivity test. The general condition of the patient improved (Table 1) and he was discharged after 25 days without complications.

The histopathological diagnosis was KCOT. Eight months after the marsupialization (Fig. 3), enucleation and bone graft with cancellous iliac bone were performed. Follow-up radiography revealed good osseous fill (Fig. 4). There was neither evidence of recurrence nor complications at 23-month follow-up.

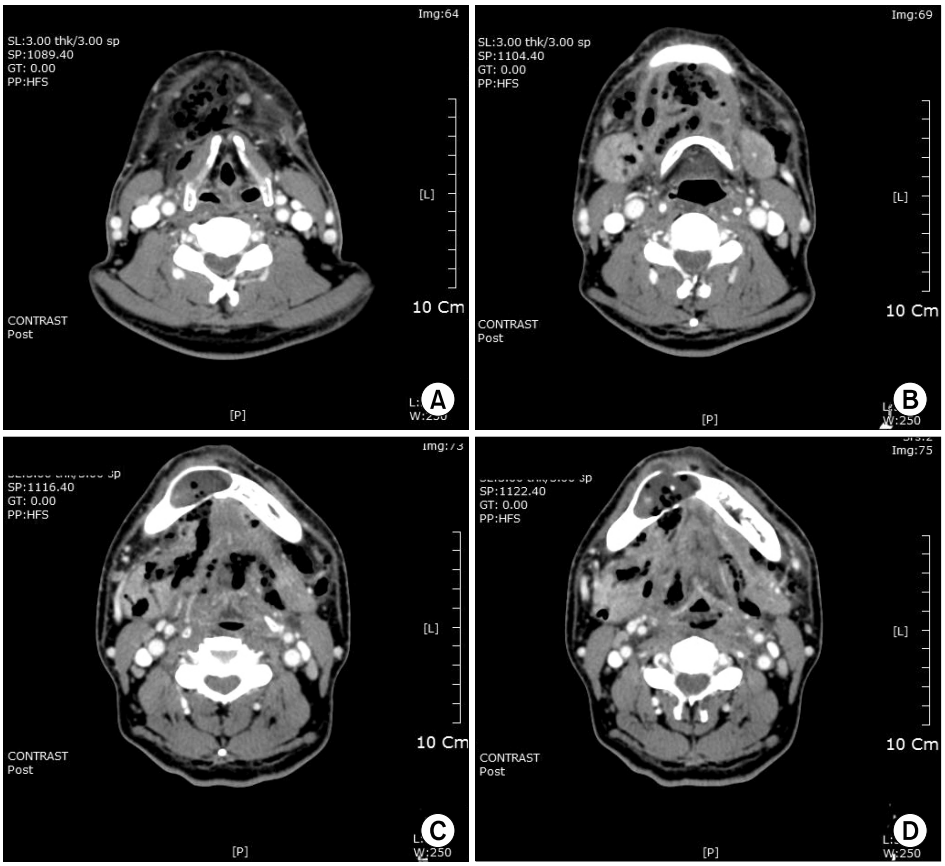

Fig. 1. Contrast-enhanced computed tomography demonstrates multiple gas collections in submandibular, submental, sublingual and lateral pharyngeal space on both sides. The airway is mildly obstructed and deviated.

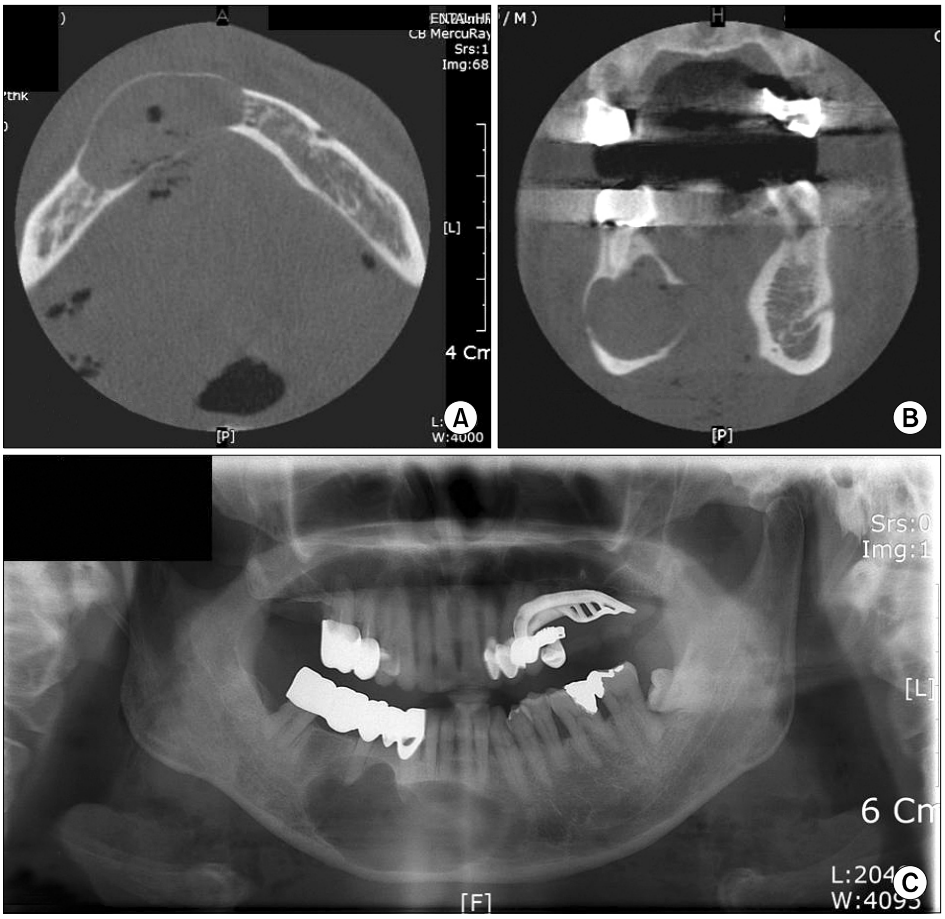

Fig. 2. The computed tomography (A, B) and panoramic radiograph (C) show a large keratocystic odontogenic tumor demonstrating significant expansion and thinning with cortical bone perforation involved mental foramen in the mandible.

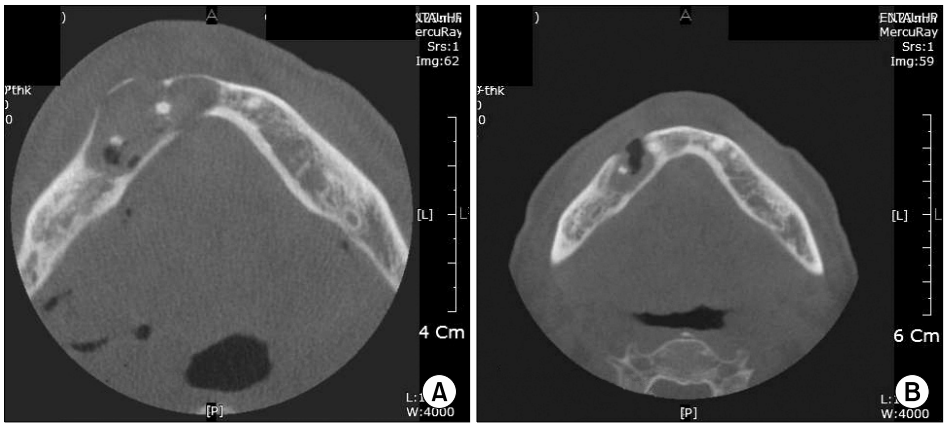

Fig. 3. Bony expansion and radiolucency are decreased and cortical bone remains intact. (A) At the first visit, (B) five months after marsupialization.

Table 1. Laboratory results associated with infection

Day after I&D	WBC ($10^3/\mu L$)	ESR (mm/h)	CRP (mg/dL)
0 day	40.69	60	>30.00
5 days	20.42	52	13.10
10 days	12.78	56	2.72
15 days	11.07	20	1.37

I&D, incision and drainage; WBC, white blood cell; ESR, erythrocyte sedimentation rate; CRP, C-reactive protein.

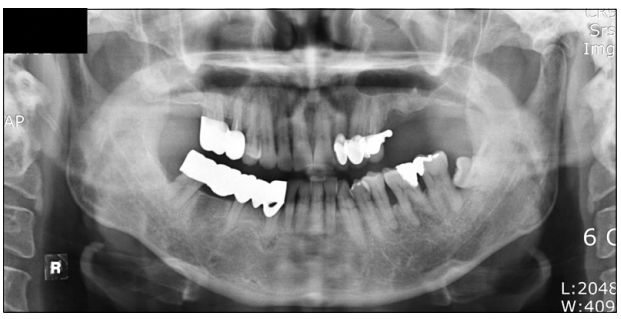

Fig. 4. Follow-up panoramic radiograph show good osseous fill.

Discussion

Deep neck infections are usually polymicrobial infections caused by aerobic and anaerobic bacteria as well as gram positives, and staphylococci and mixed flora predominate in submandibular space abscess[2]. Gram-positive cocci were the predominant bacteria cultured and gram-negative rods were the second most common bacteria isolated in microbiologic study of head and neck space infections of odontogenic origin[9]. Pus culture and antibiotic sensitivity tests are needed in severe infections of non-inflammatory origin. Although there has been no consensus regarding a standard antibiotic protocol, beta-lactamase stable beta-lactam antibiotic and metronidazole are recommended, as in patients with potential airway obstruction, the rates of beta-lactam resistance are very high[5]. In the present case, a mixture of gram-positive cocci and gram-negative rods were isolated, and antibiotic therapy consisted of ampicillin, sulbactam, metronidazole and isepacin.

Infection of the posterior compartment of the lateral pharyngeal space can cause life-threatening vascular complication such as internal carotid artery rupture, internal jugular vein thrombophlebitis[10]. In this case, the abscess involved the carotid artery and internal jugular vein, although vascular complications fortunately did not occur.

KCOTs are non-inflammatory lesions[11] and there is no inflamed supporting fibrous wall in the keratocysts[12]. The cystic fluids are sterile so the presence of microorganisms is due to a secondary infection. It is thought that the cause of the deep neck space abscess in this case was secondary infection following oozed exudates in the space. Aerobic microorganisms are commonly present but anaerobic microorganisms also are present. The KCOT fluid contained numerous anaerobic bacteria identified by polymerase chain reaction, suggesting that oral bacteria can cause symptoms like radicular cyst as an inflammatory response to a chronic irritation[11].

KCOT is an intraosseous benign neoplasm of developmental odontogenic origin with potential for locally aggressive behavior, invading adjacent tissue, with a high recurrence rate[13]. Treatment of KCOTs remains controversial. Simple enucleation has a high recurrence rate, while enucleation combined with decompression or Carnoy's solution has lower recurrence rate[14]. A large, expanding KCOT is best treated with a 2-stage approach. Enucleation and adjunctive measures after marsupialization are recommended to decrease the surgical injury to the patient[15]. Inflammation caused by marsupialization helps change the biologic behavior into a less aggressive form. The existence of greater proliferative activity in the epithelial cells of inflamed odontogenic keratocysts may be associated with the disruption of the typical structure[16,17], and marsupialization changes the cyst epithelium immunohistochemically[18]. However, marsupialization alone has not been recommended because of unpredictable recurrence rates[18].

Stoelinga[19] recommend that when KCOTs penetrate the cortical bone, the overlying attached soft tissues such as periosteum and muscles should be removed to prevent recurrence. In this case, although there was cortical perforation in buccal and lingual cortex, new bone filled the bony defect of the cortex except at the marsupialization site at enucleation, so excision of overlying mucosa was not performed. The recurrences of most KCOT occur within five years but may recur 10 or more years after treatment[20]. Therefore long osbservation is needed although sign of recurrence is not found.

Acknowledgements

This study was supported by a research fund from Chosun University, 2011.

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