Deep Neck Infection Caused by Infected Dentigerous Cyst: A Case Report

Gyeong-Mi Kim, Ji-Su Oh, Jae-Seek You, Seong-Yong Moon, Hae-In Choi

Department of Oral and Maxillofacial Surgery, School of Dentistry, Chosun University, Gwangju, Korea

Received November 11, 2021

Revised December 2, 2021

Accepted December 2, 2021

Correspondence to:

Hae-In Choi
Department of Oral and Maxillofacial
Surgery, School of Dentistry, Chosun
University, 309 Pilmun-daero, Dong-gu,
Gwangju 61452, Korea
Tel: +82-62-220-3810
Fax: +82-062-222-3810
E-mail: beautyluby@hanmail.net
<https://orcid.org/0000-0002-2425-9506>

This study was supported by research fund
from Chosun University Dental Hospital,
2019.

Deep neck infection (DNI) is a potentially serious pathology that can lead to serious complications and high rate of mortality. Since DNI affects the cervical spaces, which can compromise airway, aggressive antibiotic administration and prompt surgical drainage are essential for recovery. Although most causes of DNI are known to be dental infections, developmental cysts such as dentigerous cysts are relatively few causes. In this case, we report a rare patient with severe deep neck space infection caused by infected third molar with dentigerous cyst.

Key Words: Deep neck infection; Dentigerous cyst; Infected third molar

INTRODUCTION

Deep neck infections (DNIs) are life threatening infections in the potential spaces along the fascial planes of the neck [1]. Patients affected with DNIs typically show symptoms such as pain in the neck area, limited mouth opening, laryngeal edema, dysphagia, and odynophagia [2]. DNIs can be caused by infections from various sources, including oral surgical procedures, trauma in the oral cavity and pharynx, and infected congenital cysts in branchial cleft, thyroid glands and salivary glands. Before the era of antibiotics, 70% of DNIs was caused by peritonsillar and tonsillar infections, whereas currently the most common primary source of DNIs is odontogenic (38.8%-49%) [3]. Bacterial cultures collected from DNIs often reveal polymicrobial strains of anaerobic species, including *Streptococcus* species, *Peptostreptococcus* species, and *Staphylococcus aureus*

[4]. Without proper treatment for infection, DNIs can spread rapidly along anatomical structures and eventually to the mediastinum. Such infections can further lead to septic shock, aspiration pneumonia, and airway obstruction by laryngeal edema, resulting in high mortality. Therefore, early surgical interventions with combined efforts from oral and maxillofacial surgeon, otolaryngologists, and thoracic surgeon, as well as adequate antibiotics regimen by infectious physicians and careful observation in the intensive care unit are required. Although the most common cause of DNIs is odontogenic, DNIs from dentigerous cyst, one of the developmental cysts in the oral area, is rather unusual [5]. We report a case of DNI caused by a dentigerous cyst associated with an impacted third molar. This study protocol was approved by the institutional ethics committee of Chosun University Dental Hospital (no. CUDHIRB 2103 001 Q01). Written informed consent was obtained from all of

the patients.

CASE REPORT

An 83-year-old male visited the Chosun University Dental Hospital with severe swelling on both submandibular, submental, sublingual and neck, similar to Ludwig's angina. He was unable to ingest any food except for a small amount of water for 2 days and showed positive signs of

Fig. 1. Computed tomography image taken during the patient's first visit. The abscess that arose from the right dentigerous cyst has invaded the left mylohyoid muscle.

Fig. 2. Computed tomography image taken during the patient's first visit. The abscess at the left lateral parapharyngeal space and air bubbles are seen.

dysphagia, mild dyspnea, trismus, and hoarseness of voice. His general medical history included hypertension, and the laboratory results taken at the time of his first visit revealed acute systemic infection state with white blood cell count of 13,000 cells/mm³, C-reactive protein 42 mg/L, procalcitonin 18.98 ng/mL, Blood urea nitrogen 79.3 mg/dL, creatinine 5.39 mg/dL, and oxygen saturation 93% in arterial blood gas analysis.

Contrast-enhanced computed tomography (CT) images revealed perforation of lingual plate associated with impacted #48 and liquefied pus that was presumably originated in the right submandibular space. The pus pocket with air bubble went across the midline to the left submandibular space that was involving a large part of the mylohyoid muscle and spread even further posterior to the left parapharyngeal space along the left carotid sheath and down to the middle neck level, causing severe trachea deviation (Fig. 1-4). He was diagnosed with DNI.

With combined efforts and supervision from oral and maxillofacial surgeons and otolaryngologists, the patient received emergency surgery under general anesthesia, which included surgical drainage of both submandibular, submental, left parapharyngeal, left buccal, and left thyroidal space. After the surgery, the patient showed hemodynamical instability and low blood pressure, suggesting high risk of septic shock. The patient was, therefore, transferred to the department of infectious disease, and then admitted

Fig. 3. Computed tomography image taken during the patient's first visit. The abscess is moving downward along the carotid sheath.

Fig. 4. Computed tomography image taken during the patient's first visit. The abscess is observed around the carotid sheath at the hyoid bone level, displacing the airway to the right side by soft tissue swelling.

Fig. 5. Panoramic radiograph taken after the patient was discharged. The dentigerous cyst associated with the lower right third molar are seen.

to an intensive care unit with respiratory support attached. Additional sets of incision and drainage were performed as necessary until the patient was weaned off the ventilator 20 days later. The patient showed improved overall general condition and was discharged from the hospital.

Unfortunately, no bacteria could be identified from the antibiotic susceptibility results from the samples collected in the early days of the admission, including the ones taken during the emergency surgery. Therefore, the patient was treated with empirical antibiotics: tazocin (piperacillin/sulbactam) intravenously. Only *Acinetobacter baumannii* could be identified in the pus culture taken two weeks after the admission, which was presumed to be nosocomial infection in the intensive care unit.

Fig. 6. Extraction of the lower right third molar and enucleation of the surrounding cyst are performed under local anesthesia.

Fig. 7. Osteogenesis at the surgical site is observed three months after the surgery.

Two weeks after the discharge, the patient was recalled to the outpatient clinic of the department of oral and maxillofacial surgery for the extraction of #48 and enucleation of the surrounding cyst under local anesthesia (Fig. 5, 6). The cyst was diagnosed as dentigerous cyst by histologic examination. After 3 months, new bone formation at the previous surgical site was observed without any recurrence or abnormal findings on the panoramic view (Fig. 7).

DISCUSSION

DNI is a surgical emergency that can cause serious complications, and its mortality rate is 9% in the field of oral and maxillofacial surgery [6]. The incidence of DNIs is gradually decreasing due to the development of broad-spectrum antibiotics, yet DNI is nevertheless considered life threatening illness especially in elderly and immunocompromised patients. Once it occurs, it progresses rapidly through fascial planes and poses a serious risk of airway obstruction and septic shock within hours or days, which oral and maxillofacial surgeons find hard to manage. Therefore, DNI

requires rapid diagnosis as well as collaboration with medical specialists from other departments for further medical treatment if necessary.

Airway management is a critical yet difficult task in patients with DNIs. For patients suspected of having DNI, airway assessment is essential at the initial physical evaluation. Airway intubation can be very difficult in such patient due to airway obstruction, tissue rigidity, and orifice limitation from swelling. According to a retrospective study by Chen et al. [7], tracheostomy should be considered in patients with DNIs if they are aged 65 years or older, have three or more interstitial spaces infected, or suffer from mediastinal sinusitis.

Contrast-enhanced neck CT is highly sensitive (91%) in the diagnosis of DNI [8] and considered a gold standard that can be utilized to determine the extent of DNIs and to differentiate them from cellulitis. For patients suspected of having DNIs, taking routine contrast-enhanced neck CT is beneficial in evaluating the prognosis and sequelae of the infections [9]. In this case as well, contrast-enhanced neck CT was taken more than 5 times to assess the patient's condition and to determine whether the ventilator should be weaned off from the patient after the edema in the airway subsided.

It is common in DNIs for the abscess to spread posteriorly and downward due to the direction of gravity and the supine position of patient in bed. Naturally, there are some cases, in which drainage is not established well due to poor accessibility of the location, often requiring a few more rounds of incision and drainage [10]. In our case, another incision and drainage was performed under local anesthesia on the 5th postoperative day and additionally under general anesthesia on the 8th postoperative day. There is a report that describes an attempt to reduce the number of such surgeries by connecting a silastic drain to a vacuum assisted closed machine to induce continuous drainage [11].

The pathogenesis of this case is thought to have begun from the infection of the dentigerous cyst associated with the lower right third molar that penetrated through the lingual plate, spread across the mylohyoid muscle, and caused DNI on the left side. Dentigerous cyst is one of the most common odontogenic cysts second only to periapical cyst, and it occurs most frequently in the mandibular third

molars and maxillary canines, which have greater tendency to be impacted [12]. In a retrospective study of infection in the head and neck region, it was reported that 2.1% of patients had a dentigerous cyst [13]. Because dentigerous cysts are essentially asymptomatic and non-inflammatory cysts that originate from reduced enamel epithelium [14], it is speculated that the cyst substance was initially sterile, but the secondary infection took place as the cyst penetrated the lingual plate, eventually leading to DNI. Pus culture and antibiotic sensitivity tests are critical in studying severe infections of non-inflammatory origin such as this kind. Although a standard antibiotics regimen has not been established for head and neck infections, antibiotics are selected by referring to the pus culture and antibiotics sensitivity test. If lab results are not available during the early stage of the infection, empirical antibiotics such as moxifloxacin or combination of β -lactam antibiotics and metronidazole are used. Unfortunately, severe DNIs are usually mixed infection of various anaerobic and aerobic bacteria that are often resistant to β -lactam antibiotics [15]. Furthermore, no specific bacteria were identified in the test results from the samples collected during emergency surgery, which lead to administration of tazocin (piperacillin/sulbactam) as an empirical antibiotic. The absence of cultured bacteria is presumably due to exposure to air in the process of obtaining the specimen, and to prevent such event, specimens should be collected prior to initiation of antimicrobial therapy, ideally by aspiration from undrained abscess.

The standard treatment for dentigerous cyst is to extract the cyst together with the impacted tooth, and decompression before extraction may be preceded to reduce the size of the cyst if it is very large [16]. No major complication such as inferior alveolar nerve damage was observed after the patient was discharged from the hospital and received surgical removal of the right third molar and the dentigerous cyst, and a panoramic radiograph taken 3 months after surgery showed osteogenesis at the surgical site.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

ORCID

Gyeong-Mi Kim

<https://orcid.org/0000-0002-0451-379X>

Ji-Su Oh

<https://orcid.org/0000-0002-8369-5025>

Jae-Seek You

<https://orcid.org/0000-0001-7638-9583>

Seong-Yong Moon

<https://orcid.org/0000-0002-7513-4404>

Hae-In Choi

<https://orcid.org/0000-0002-2425-9506>

REFERENCES

1. Wang LF, Kuo WR, Tsai SM, Huang KJ. Characterizations of life-threatening deep cervical space infections: a review of one hundred ninety-six cases. *Am J Otolaryngol* 2003;24:111-117.
2. Franzese CB, Isaacson JE. Peritonsillar and parapharyngeal space abscess in the older adult. *Am J Otolaryngol* 2003;24:169-173.
3. Pesis M, Bar-Droma E, Ilgiyaev A, Givol N. Deep neck infections are life threatening infections of dental origin: a presentation and management of selected cases. *Isr Med Assoc J* 2019;21:806-811.
4. Huang TT, Liu TC, Chen PR, Tseng FY, Yeh TH, Chen YS. Deep neck infection: analysis of 185 cases. *Head Neck* 2004;26:854-860.
5. Anderson DW, Evans D. Dentigerous cyst of mandible presenting as sepsis. *Am J Emerg Med* 2014;32:1561.e3-e4.
6. Garcia T, Rios M, Paiva JA. Predictors of severity in deep neck infections admitted to the intensive care unit. *Anaesth Intensive Care* 2012;40:832-837.
7. Chen SL, Young CK, Tsai TY, et al. Factors affecting the necessity of tracheostomy in patients with deep neck infection. *Diagnostics (Basel)* 2021;11:1536.
8. Ungkanont K, Yellon RF, Weissman JL, Casselbrant ML, González-Valdepeña H, Bluestone CD. Head and neck space infections in infants and children. *Otolaryngol Head Neck Surg* 1995;112:375-382.
9. Smith JL 2nd, Hsu JM, Chang J. Predicting deep neck space abscess using computed tomography. *Am J Otolaryngol* 2006;27:244-247.
10. Kim YS, Park JH, Chun SS, Han DY, Kim JE, Lee BD. Clinical analysis of deep neck infection. *Korean J Otorhinolaryngol-Head Neck Surg* 2010;53:627-631.
11. Kim B, Kim J, Kim H, Song C, Pai H. Deep neck infection with mediastinal abscess treated by modified vacuum-assisted closure application. *J Acute Care Surg* 2017;7:34-38.
12. Freitas DQ, Tempest LM, Sicoli E, Lopes-Neto FC. Bilateral dentigerous cysts: review of the literature and report of an unusual case. *Dentomaxillofac Radiol* 2006;35:464-468.
13. Smith JL 2nd, Kellman RM. Dentigerous cysts presenting as head and neck infections. *Otolaryngol Head Neck Surg* 2005;133:715-717.
14. Patil AS, Jathar PN, Panse AM, Bahutule SR, Patil RU, Patil M. Infected dentigerous cyst and its conservative management: a report of two cases. *Int J Clin Pediatr Dent* 2019;12:68-72.
15. Mathew GC, Ranganathan LK, Gandhi S, et al. Odontogenic maxillofacial space infections at a tertiary care center in North India: a five-year retrospective study. *Int J Infect Dis* 2012;16:e296-e302.
16. Martínez-Pérez D, Varela-Morales M. Conservative treatment of dentigerous cysts in children: a report of 4 cases. *J Oral Maxillofac Surg* 2001;59:331-333.