

Necrotizing fasciitis of the temporalis muscle caused by ascending infection of an orthokeratinizing odontogenic cyst

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ABSTRACT

Introduction: Necrotizing fasciitis is rare in the head and neck region, especially associated with the temporalis muscle, and it is almost always caused by spread of infection secondary to dental extraction. This report presents an extremely rare case of necrotizing fasciitis as a result of ascending orthokeratinizing odontogenic cyst infection involving the temporalis muscle.

Case Report: A healthy 26-year-old man visited our hospital complaining of pain and swelling in the right mandibular third molar region. A panoramic radiograph demonstrated a well-defined unilocular radiolucency at the right side of the mandible. The infection had spread to the temporal region, and a computed tomography (CT) image showed the gas formation. The oral and intravenous administration of antibiotics, incisional drainage, and removal of necrotic tissue and impacted wisdom tooth led to remission of the disease.

Conclusion: The present report was an extremely rare case of necrotizing fasciitis as a result of ascending orthokeratinizing odontogenic cyst infection in mandible involving the temporalis muscle.

Keywords: Computed tomography (CT), Necrotizing fasciitis, Orthokeratinizing odontogenic cyst, Temporalis muscle

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INTRODUCTION

Necrotizing fasciitis of the head and neck is an uncommon [1, 2], rapidly spreading soft tissue infection of polymicrobial origin characterized by extensive necrosis and gas formation in the subcutaneous tissue and superficial fascia [3]. Among cases of necrotizing fasciitis of the head and neck region, those involving the temporal space are rare and infrequently reported. More commonly, temporal space infections are associated with the extraction of both infected and noninfected molar teeth [4]. However, it is extremely rare that infection of the temporal space is caused by a cystic lesion in the mandible. In this report, we present a case involving necrotizing fasciitis of the temporalis muscle caused by infection of an orthokeratinizing odontogenic cyst in the mandible.

CASE REPORT

A 26-year-old Japanese man who had first seen a dentist complaining of pain and swelling in the right mandibular third molar region a week earlier was referred to the outpatient clinic of the Oral and Maxillofacial Surgery Department of Nihon University Dental Hospital. Upon initial examination, he was febrile (37.5 °C), with right facial swelling involving the buccal and submandibular spaces, and trismus with an interincisal opening of 5 mm. The incision and drain were observed on the buccal aspect of the right molar region. However, the previous dentist reported that no pus was identified. A panoramic radiograph demonstrated a well-defined unilocular radiolucency with an impacted third molar on the right side of the mandible (Figure 1). A computed tomography (CT) scan of the head and neck was performed, which demonstrated gaseous content in the lingual region and swelling in the parapharyngeal space (Figure 2). A blood test revealed a leukocyte count of 14,380/ μ L, erythrocyte sedimentation rate of 24 mm/h, and C-reactive protein level of 15.31 mg/dL. Therapy was initiated with daily divided doses of oral amoxicillin hydrate 750 mg and intravenous ampicillin sodium 2 g as an outpatient for 4 days. However, the patient continued to experience severe pain and difficult eating and was hospitalized.

Upon admission, the patient's vital signs were as follows: temperature 38.0 °C, pulse 106 bpm, and blood pressure 136/73 mmHg. A blood test revealed a leukocyte count of 16,230/ μ L, erythrocyte sedimentation rate of 72 mm/h, and C-reactive protein level of 25.66 mg/dL. Computed tomography (CT) findings revealed right facial soft tissue swelling and gas formation in the temporal and facial tissue (Figure 3). Ampicillin sodium was discontinued and administration of ceftriaxone sodium hydrate was initiated and continued for four days. The puncture was performed on the buccal aspect of the right molar region and in the temporal region. Only a small volume of pus could be identified from right molar buccal site and temporal region. After another three days, CT findings revealed increased gas formation (Figure 4). The color of skin became weakly red over the area of fasciitis; therefore, a 30-mm incision and drainage was carried out in the temporal and occipital regions under local anesthesia and sedation. A large collection of pus and necrotic tissue were found. Removal of necrotic tissue could be easily performed through the incision in the temporal and occipital regions. The wound was packed and daily irrigation with normal saline solutions was performed. Microbiological examination revealed *Peptostreptococcus*, *Streptococcus*, and *Prevotella/ Porphyromonas* species, which were sensitive to minomycin and clindamycin. Therefore, antibiotics regime was changed to clindamycin hydrochloride from ceftriaxone sodium hydrate. Swelling and drainage diminished rapidly over the next several days. The patient was discharged 12 days after admission with an

oral opening 20 mm. Forty-two days after discharge, the cyst was surgically enucleated under general anesthesia along with removal of the impacted third molar tooth. The lesion was diagnosed histopathologically as an orthokeratinized odontogenic cyst with inflammatory changes. After two years of follow-up (Figure 5), bone formation was observed, and there was no recurrence of the lesion.



Figure 1: Panoramic radiograph at the initial visit showing well-defined unilocular radiolucency with impacted third molar at the right mandibular.

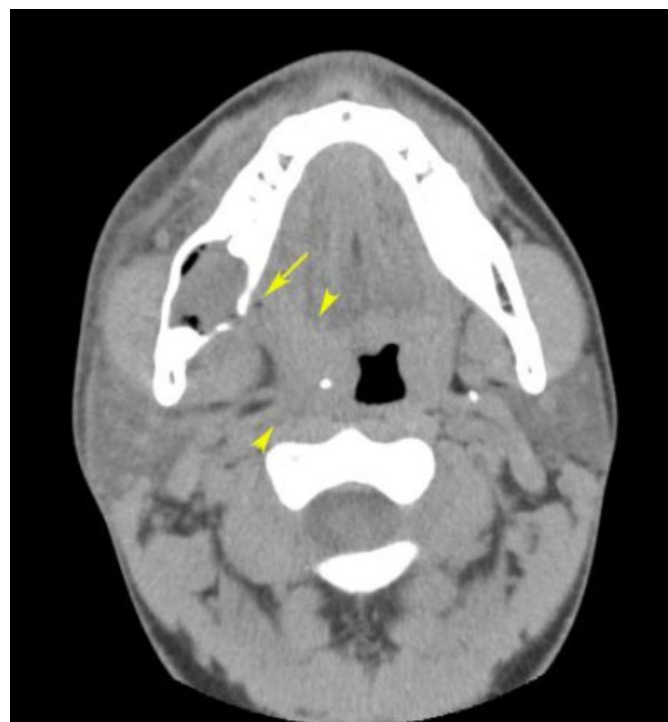


Figure 2: CT image at the initial visit showing gaseous content in the lingual region (arrow) and swelling of parapharyngeal space (arrowheads).

DISCUSSION

Necrotizing fasciitis is rare in the head and neck region, especially when it is associated with temporalis muscle. This paper reports an extremely rare case of necrotizing fasciitis as a result of ascending orthokeratinizing odontogenic cyst infection in mandible involving the temporalis muscle.



Figure 3: CT image after four day from initial visit showing gas bubbles at the fascia of temporal muscle.



Figure 4: CT image after seven day from initial visit showing larger gas formation.



Figure 5: Postoperative two year, panoramic radiograph showing bone formation at the right mandibular.

Previous reports indicate that necrotizing fasciitis in the head and neck region is almost always caused by spread of oral infection, and *Clostridium perfringens* is usually not implicated [1, 2]. Additionally, Nakamura et al. reported that most gas-producing infections in the head and neck region are nonclostridial and proceed along fascia, whereas clostridial infections progress in muscular layers and are often lethal [3]. It has been reported that *Peptostreptococcus* and *Bacteroides*, anaerobic and indigenous nonclostridium bacteria in oral and pharynx are involved in the necrotizing fasciitis in head and neck. Although anaerobic bacteria alone have low pathological significance, high pathogenicity can occur if there is concomitant infection with aerobic bacteria. In the present case, concomitant infection with *Peptostreptococcus* and *Staphylococcus* may have contributed to the development of necrotizing fasciitis in the temporalis muscle.

When necrotizing fasciitis occurs in the head and neck region secondary to odontogenic infection [5–7], the parapharyngeal space is closely related to both the ascending and descending pathways of the spread of infection [8]. The ascending infection tracks the masticator space and parotid gland from the parapharyngeal space, and the descending infection tracks the submandibular region and neck. In the present case, the CT images showed an ascending spread of infection to the temporal space via the pterygoid muscle from the parapharyngeal space. Computed tomography images are highly useful for confirmation of gas formation and estimation of the spread of infection, although it is difficult to distinguish necrotizing fasciitis from inflammation in the head and neck in its early stage [9, 10]. Additionally, clostridial infection mainly spread to muscle tissue and nonclostridial infection spreads primarily to fascia. Therefore, it is useful to confirm whether gas formation is present in muscle tissue or fascia by CT imaging to differentiate clostridial and nonclostridial infections [11]. In the present case, CT imaging showed that the gas formation was present in the fascia, we considered that the causative bacterium was likely to be nonclostridial.

It has been reported that necrotizing fasciitis in the head and neck is mainly caused by molar tooth extraction and infection [6, 7]. Furthermore, almost all cases of temporal abscess have been reported to occur secondary to molar tooth extraction and the presence of pus [4]. To our knowledge, one case exists of a temporal abscess occurring secondary to an infected recurrent keratocystic odontogenic tumor [12]. Furthermore, all cases of necrotizing fasciitis of the temporal region were caused by tooth [13–15]. The histopathology of the present case showed orthokeratinized odontogenic cyst. Therefore, the present report is extremely rare case of necrotizing fasciitis of the temporal muscle caused by infection of an orthokeratinizing odontogenic cyst in mandible. Considering the two cases of necrotizing fasciitis associated with a keratocystic lesion, it could be

hypothesized that the keratinizing epithelium may trigger the inflammatory process.

Successful treatment of necrotizing fasciitis is based on the early recognition, intensive antibiotic therapy, and aggressive surgical intervention with drainage of pus and debridement of necrotic tissue [6, 7]. In the present case, the patient was young and healthy, and necrotizing fasciitis occurred in superficial in the temporal region; therefore, oral and intravenous administration of antibiotics, incisional drainage, and removal of the necrotic tissue led to remission of the disease. All obviously necrotic tissue should be removed because the nonviable tissue serves as a source of further infection. The importance of surgical drainage for the treatment of necrotizing fasciitis is well illustrated in our case. The tract of infection to the temporal region was taken by the deep fascia of temporal muscle.

CONCLUSION

The present report is a case involving necrotizing fasciitis of the temporalis muscle caused by infection of an orthokeratinizing odontogenic cyst in the mandible. The infection had spread to the temporal region, and a CT image showed the gas formation. The oral and intravenous administration of antibiotics, incisional drainage, and removal of necrotic tissue led to remission of the disease.

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Author Contributions

Osamu Shimizu – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Hiroshi Shiratsuchi – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Guarantor of Submission

The corresponding author is the guarantor of submission.

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Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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