

Ascending Facial Necrotizing Fasciitis in a Patient Taking a Bisphosphonate



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Facial necrotizing fasciitis (NF) is a rare fulminant infection of the soft and connective tissues that spreads along the fascial planes of the face. Its origins most commonly involve odontogenic infection and it is usually associated with a history of dentoalveolar surgery, such as tooth extraction or implant placement. We present a case of ascending facial NF with odontogenic origin in a patient taking a bisphosphonate.

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Necrotizing fasciitis (NF) is defined as rapidly progressive necrosis of subcutaneous fat and fascia. NF is most commonly found in the extremities, trunk, and perineum; NF occurring in the head and neck is relatively rare.¹ Facial NF is associated with a mortality rate approaching 30%, as well as devastating morbidity, including disfigurement and airway compromise.² Usually, facial NF is associated with odontogenic infections, which have a tendency to descend into the neck along the cervical fascia. In the present study, we report an unusual case of ascending facial NF in a patient taking a bisphosphonate.

Case Report

A 61-year-old man who had been a smoker for 42 years presented to our clinic with severe swelling, erythema, and a sensation of heat in his right cheek that had developed 10 days previously. Before his visit, the patient had been treated with second-generation cephalosporin antibiotics (cefaclor) at a local clinic for 5 days under the assumption that he had cellulitis, but his symptoms were aggravated. The patient had a medical history of rheumatoid arthritis and had been treated with corticosteroids and methotrexate for 7 years. In addition, he had been taking alendronate (Fosamax) for 6 months

to treat osteoporosis. Finally, 5 months before the visit to our hospital, he received a dental implant in the right molar area without a drug holiday from alendronate.

On arrival, the patient had severe swelling and erythema in the right cheek and fluctuation in the right temporal area. Also, an approximate 2-cm opening was noted in the right gingivobuccal area, and pus was draining from the opening. The patient's vital signs were as follows: blood pressure, 120/80 mm Hg; heart rate, 112 beats/min; temperature, 38.2°C; and respiratory rate, 20 breaths/min. The initial laboratory values for the patient were as follows: white blood cell count, 24,680/mm³; sodium, 138 mEq/L; hemoglobin, 14.0 g/dL; creatinine, 1.02 mg/dL; and glucose, 127 mg/dL. The Laboratory Risk Indicator for Necrotizing Fasciitis (LRINEC) score³ was 5. In addition, the level of C-reactive protein was 314.20 mg/dL and the erythrocyte sedimentation rate was 68 mm/hour. An initial panoramic view and computed tomography (CT) scan revealed osteonecrosis in the mandible around the site of the dental implant. Also, extensive cellulitis and an abscess was noted in the right masseter muscle and in the right parietotemporal area (Fig 1). With a tentative diagnosis of NF, emergent incision and drainage in the mandible and temporal area was performed; however, the

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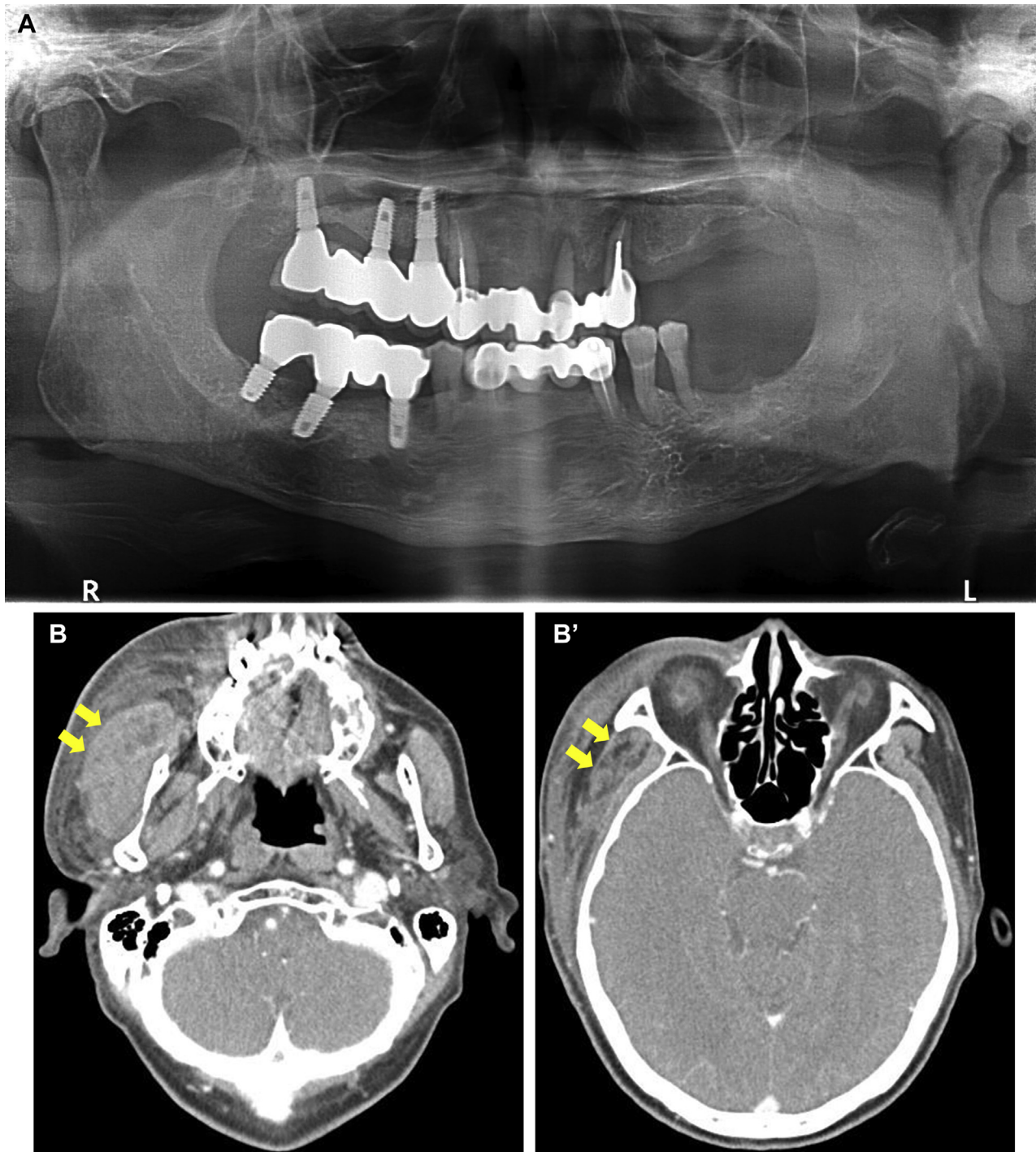


FIGURE 1. A, Preoperative panoramic view showing radiolucency and a bone defect in the right mandible. B, B', Preoperative facial computed tomography scan showing severe swelling in the right cheek and abscesses in the masseter muscle and temporalis muscle (arrows). Kim et al. *Ascending Facial NF and Bisphosphonates*. *J Oral Maxillofac Surg* 2017.

necrosis of the skin of the cheek was aggravated thereafter (Fig 2).

Initially, empirical antibiotics (ceftizoxime, isepamicin, and metronidazole) were administered to cover Gram-positive and Gram-negative organisms and anaerobes.⁴ However, they were later changed to van-

comycin and ceftriaxone in accordance with the results of bacterial culture from the pus (methicillin-resistant *Staphylococcus epidermidis*, *Escherichia coli*, and *Streptococcus anginosus*). After 5 days, definitive surgical debridement of the right cheek and sequestrectomy of the mandible was performed with



FIGURE 2. Preoperative photograph showing severe skin necrosis and eschar formation in the right cheek.

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the patient under general anesthesia. Intraoperatively, a severe abscess in the buccal fat pad and masseter muscle was noted. Also, the superficial lobe of the parotid gland was necrotic, and branches of the facial nerve were exposed (Fig 3). After serial wound irrigation and curettage, the defect was covered with a rotational fasciocutaneous flap and skin graft. Afterward,



FIGURE 3. Intraoperative photograph after debridement. Necrotic tissue in the skin, subcutaneous layer, and superficial lobe of the parotid gland was debrided. Also, the abscess in the masseter muscle was drained. Note the exposed facial nerve.

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serial scar revision was performed. No signs of recurrence developed during 23 months of follow-up (Fig 4).

Discussion

Although the occurrence of NF in the head and neck is rare, when it does occur, the results can be devastating. If it spreads inferiorly to the neck, fatal complications such as mediastinitis can occur, and if it spreads superiorly to the cheek or temples, facial disfigurement can occur.

Most cases of facial NF begin with an infection of odontogenic origin. In our patient, the cause of NF was thought to be highly associated with bisphosphonate-related osteonecrosis of the jaw (BRONJ). Although our patient had been taking bisphosphonates for a relatively short period, his active smoking status and the use of immunosuppressive medication most likely placed the patient at high risk of developing osteonecrosis and NF. In addition, implant insertion could have played an important role in the aggravation of osteonecrosis.⁵

If the diagnostic criteria for BRONJ are strictly applied, our patient's case does not fall into the category of BRONJ, because we cannot demonstrate the exact duration of bone exposure. The patient had undergone dental surgery at a local dentist's office and did not visit our hospital before the development of NF. However, even if the possibility of developing osteonecrosis is low with taking a bisphosphonate for a short duration, we believe it is very risky to perform dental surgery for a patient without drug holiday from bisphosphonates, especially if the patient is taking immunosuppressive drugs.

The International BRONJ Task Force recommends that patients planning to undergo a dental procedure who are at increased risk of developing BRONJ stop taking bisphosphonates at least 3 months before the procedure, especially those with greater cumulative bisphosphonate exposure (>3 years) and those with comorbid risk factors and previous or current usage of steroids or chemotherapeutic agents.^{5,6}

Rapoport et al⁷ postulated that whether an odontogenic infection develops into an acute soft tissue infection or NF is related to the host immunologic state and/or the synergistic effect of the infecting organisms.⁷ Diabetes mellitus is the most commonly associated disease, and other conditions, such as a compromised immune system, malnutrition, chronic renal disease, alcoholism, and peripheral arterial disease, can also be involved. Therefore, patients with such risk factors who are scheduled to undergo dental surgery should be informed of the possibility of developing a soft tissue infection and the importance of early management.

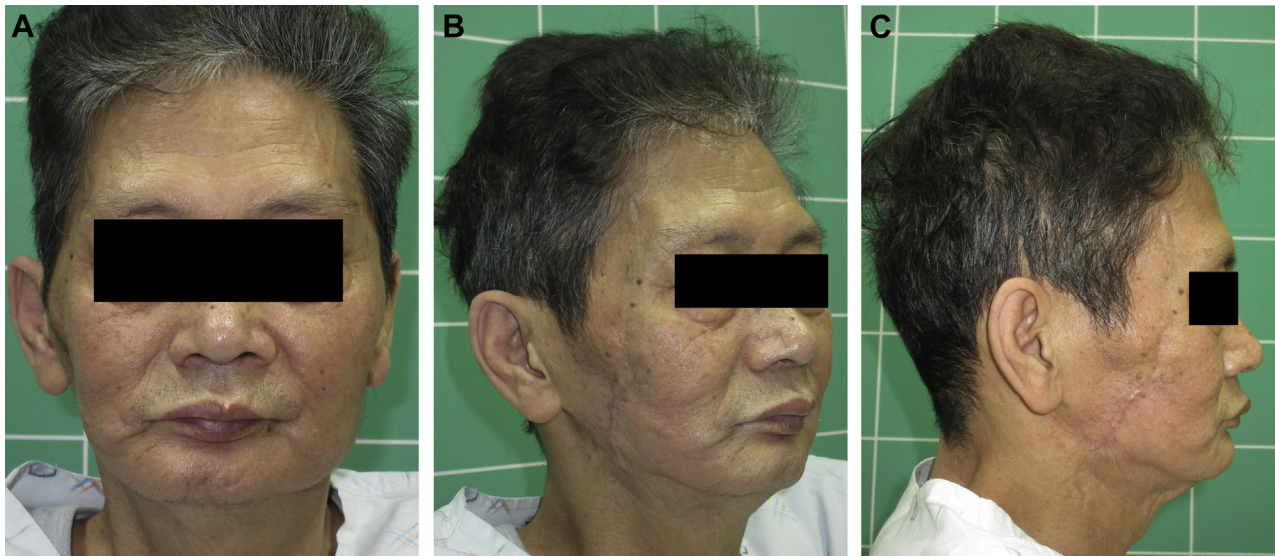


FIGURE 4. A–C, Postoperative photographs taken 23 months after the procedure.

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In our patient, the progression of the disease is another interesting issue. In the vast majority of cases, odontogenic NF spreads inferiorly. It initially involves the submasseteric space, resulting in spasms of the masseter muscle and, thus, trismus. It then migrates inferiorly to the neck, eventually involving the superficial and deep cervical fascia and mediastinal structures in some cases. If mediastinitis develops, the condition can be devastating and is associated with a high mortality rate (39.5%).⁸

The number of reported cases of ascending facial NF are in the single digits.^{9,10} When the condition spreads superiorly, it spreads along the buccal fat pad, temporal fascia, and underlying temporalis muscle. Hence, when a patient is suspected of having facial NF, fluctuation or swelling in the temporal area should be examined because signs of inflammation can be masked by the hair.

The treatment of NF can be challenging. The keys to successful management include early recognition, high doses of appropriate antimicrobial therapy, and early surgical intervention with radical debridement of necrotic tissue.

Many diagnostic tools have been developed and used. The LRINEC has been developed to distinguish NF from cellulitis.³ At a cutoff LRINEC score of 6 or greater, it has a positive predictive value of 92.0% and a negative predictive value of 96.0%. Among the imaging studies, subcutaneous gas on a plain radiograph is a very specific finding. However, it is not very sensitive. CT, magnetic resonance imaging (MRI), and ultrasonography can be used to search for specific findings such as the thickness of fascial layer, with or without enhancement. However, the

limitation is that a comparison must be done with the uninvolved limb, not the non-necrotizing soft tissue.¹¹ Frozen section histopathologic examination has also been recommended.^{12,13} However, a well-experienced pathologist is needed for examination of frozen section specimens. A more reliable method might be to explore the compromised area and identify the macroscopic findings consistent with necrotizing soft tissue infection. These findings include gray necrotic tissue, lack of bleeding, thrombosed vessels, “dishwater” pus, and noncontracting muscle.¹¹

Above all else, a high index of suspicion is most important in diagnosing NF. If clinical findings suggestive of NF (ie, tense edema, skin discoloration, blister or necrosis, crepitus, subcutaneous gas, pain out of proportion) are present with systemic signs of inflammation (eg, tachycardia, hypotension, fever, shock), early surgical exploration and administration of broad-spectrum antibiotics should be performed as soon as possible.

Just as in our patient, NF due to odontogenic infection usually involves a wide variety of microbes, with a combination of aerobic, facultative anaerobic, and obligate anaerobic organisms. Therefore, broad-spectrum antibiotic coverage is initially required, with the antibiotics used changed according to the results of the bacterial, culture, and sensitivity tests.¹⁰ In the surgical management of NF, a thorough knowledge of the anatomy of the fascial spaces of the head and neck is critical in understanding the route of infection. Also, generally, multiple debridement procedures are necessary to drain the loculated pus.¹⁴

In conclusion, odontogenic infection and facial NF can develop secondary to BRONJ. To prevent this

condition, patients with a history of bisphosphonate treatment who need dental procedures should stop taking the bisphosphonates for a sufficient period before the dental procedure. This is especially true for patients in immunocompromised states, because the risk of infection is significantly increased in such cases. If facial NF is suspected, prompt surgical interventions, broad-spectrum antibiotic treatment, and aggressive diagnostic tests such as MRI and CT are essential to minimize complications.

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