

A Case Report of Cellulitis of the Mouth Floor Presenting as Necrotizing Fasciitis

Luo Huang¹, Hui Qin², Ling Qiu², Kexiong Ouyang¹ and Xiuyi Yang^{2*}

¹Key Laboratory of Oral Medicine, Guangzhou Institute of Oral Disease, Stomatology Hospital of Guangzhou Medical University, Guangzhou, P. R. China

²Department of Otorhinolaryngology Head and Neck Surgery, The People's Hospital of Qian-Nan, Wenfeng Road 9, Duyun City, Qianna Buyi National Minority Miao National Minority Autonomous, P.R. China

Abstract

Cellulitis of the mouth floor can seldom be complicated by necrotizing fasciitis. Cellulitis of the mouth floor that has been identified in association with necrotizing fasciitis tends to arise in cervicofacial region of patients who have predisposing factors. But this article describes a rare case of cellulitis of the mouth floor causing necrotizing fasciitis in a healthy woman. The clinical features, diagnosis, and treatment are discussed in detail.

Keywords: Cellulitis • Mouth floor • Necrotizing fasciitis

Introduction

Cellulitis of the mouth floor is one of the severe oral and maxillofacial space infections, involving the bilateral submandibular space, sublingual space, and submental space [1]. The main cause is odontogenic infection, which is periapical periodontitis of the focal tooth, periodontal abscess, subperiosteal abscess or complications caused by dental treatment. Other common causes include sialadenitis, lymphadenitis, acute tonsillitis, and soft tissues of the floor of the mouth. Injuries and mandibular fractures, etc., [2,3]. Although the incidence of cellulitis at the floor of the mouth has been reduced, it is often accompanied by serious complications. The rapid spread of the infection on the floor of the mouth leads to diffuse swelling of the floor of the mouth, elevated tongue, and compression of the airway. Upper respiratory tract obstruction leads to suffocation and even death. If treatment is delayed or improperly treated, the infection spreads, which can lead to serious complications such as multi-space infection in the deep neck, downward mediastinitis, sepsis and even sepsis, with a high case fatality rate [4-6].

Necrotizing Fasciitis (NF) is a rare soft tissue infection resulting in the death of subcutaneous and fascial tissue. It usually occurs in the extremities, abdomen, and perineum [7]. But cervicofacial NF is rare, and usually occurs as a complication in patients who are immunocompromised. Cellulitis of the mouth floor that have been identified in association with NF tend to arise in cervicofacial region of patients who have predisposing factors including extremes of age, immunosuppression, diabetes mellitus, alcohol, and tobacco smoking [8,9]. Here this article describes a rare case of cellulitis of the mouth floor causing upper limbs and cervicofacial NF in a patient without medical conditions.

Case Report

A 41-year-old woman presented to the emergency room with a seven-

**Address for Correspondence:* Yang X, Department of Otorhinolaryngology Head and Neck Surgery, The People's Hospital of QianNan, Wenfeng Road 9, Duyun City, Qianna Buyi National Minority Miao National Minority Autonomous, P.R. China, E-mail: 66905692@qq.com

Copyright: © 2020 Yang X, et al. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Received 22 December 2020; **Accepted** 07 January 2021; **Published** 14 January 2021

day history of severe, progressive swelling of her neck associated with odynophagia. She reported increased pain that was dull and 7/10 in intensity, localized to her right neck. The patient had no other comorbidities Oral cavity examination was remarkable for trismus with mouth opening limited to two fingers. On physical examination, she was awake, alert, and in mild respiratory distress. There was an extensive tender brawny swelling of right submandibular and submental and cervical regions, in which fluctuation could be touched. On admission, an elevated leukocyte count of 20.89 and Serum High sensitivity C-reactive protein of 337.5 mg/L were recorded. A CT study gave evidences of multiple air pockets with fluid collections got involved in the tissue spaces of right parotid gland and neck at first (Figure 1). Broad-spectrum antimicrobials with cephalosporine and metronidazole were initiated; emergent surgical debridement was performed and fiberoptic support was necessary for a difficult orotracheal intubation. Under general anesthesia multiple incisions and drainage of the involved spaces of both sides were achieved. A multitude of fluid which appeared serous and frankly pus like could be expressed. Multiple through and through drains were placed. Bacterial cultures of intraoperative samples identified *Streptococcus anginosus*. Infectious disease was consulted for further antibiotic management. Her antibiotics were switched to vancomycin and imipenem/cilastatin.

On the afternoon of the fifth day, patient presented diffusing blue-purple ecchymosis over the bilateral inguinal regions. A matter of considerable concern was the observed drop in hemoglobin level and blood platelet count, which worsened to 71 and 50 respectively. The results of coagulation function test at this time showed an ascend except descending thrombin time hence decided to transfuse 2 units of packed cells and human albumin. As her hospitalization progressed, the right side of her chest and left upper limb had draining openings with a necrotic appearance associated with a foul smell (Figure 2). Multiple bullae and small blisters around necrotic area were noted. The clinical picture and CT findings (Figure 3) confirmed the diagnosis of necrotizing fasciitis. The patient underwent aggressive bedside wound debridement of the necrotic tissue exposing the contents and packed open with BIPP dressing. The clinical course was complicated by septic shock, acute respiratory distress syndrome and acute renal failure. Despite aggressive medical and surgical intervention, the patient expired from multisystem organ failure on day twenty.

Discussion

Cellulitis of the mouth floor is an acute diffuse cellulitis involving bilateral submandibular space, sublingual space, and submental space. It is caused by bacterial infection and has the characteristics of rapid onset, rapid spread, and high mortality. Some patients with severe oral cellulitis have complications, which makes the treatment more difficult. When the infection spreads to the mediastinum or forms of purulent pleurisy, despite the use of

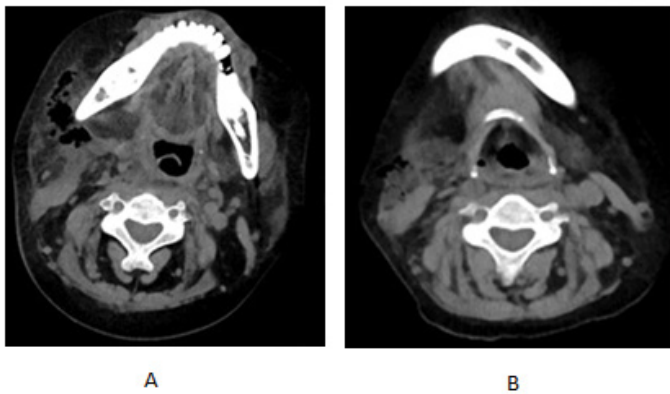


Figure 1. CT showing presence of gas in the tissue spaces of right parotid gland (A) and cervical space (B).

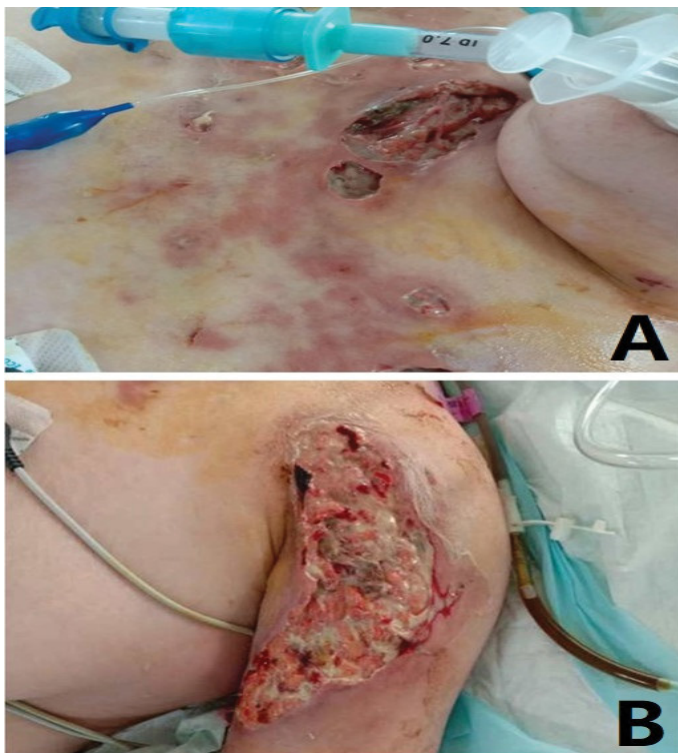


Figure 2. Necrotic fascial layers in the right side of the chest (A) and left upper limb (B).

antibiotics, the mortality rate is still as high as 38% to 60%. When combined with necrotizing fasciitis, the recent mortality rate is reported to be 18% to 22% [8], although cervicofascial necrotizing fasciitis is rarely seen in patients with cellulitis of the mouth floor [9]. Direct and distant spread of necrotizing fasciitis can lead to complications that include intracranial, retropharyngeal, and pulmonary infections. Hematogenous dissemination can also occur, leading to complications such as septic shock, rheumatic disease, and cardiac problems [10]. If left untreated, the rapid dissemination of the infection can be fatal. Cellulitis of the mouth floor and necrotizing fasciitis are the two aggressive manifestations of soft tissue infection to occur in the cervicofacial region. Both of the conditions can be aggressive form of underlying tissue destruction as seen in our case. The natural course of this infection is the rapid extension into contiguous areas and results in supportive fasciitis and cutaneous necrosis of fatty and cellular tissue.

Nevertheless the two conditions represent completely different pathogenic mechanisms; for this reason, the occurrence of both conditions in the same patient is rare and creates diagnostic dilemma at the onset of the disease. All authors agree on the association illness like diabetes mellitus in most reported cases of cervicofacial NF. The immune compromise is the principal deciding factor in the establishment and in the course and prognosis of the disease.

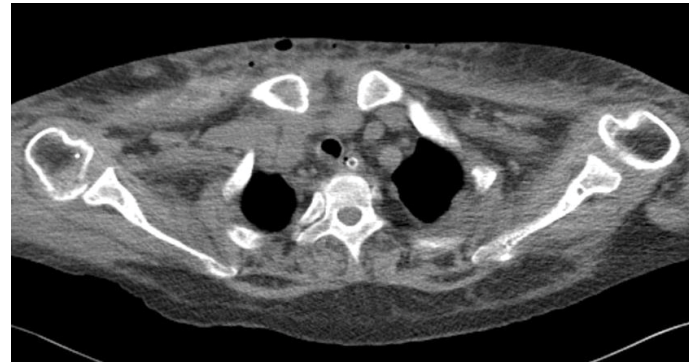


Figure 3. CT showing air dissecting into the anterolateral chest wall.

However, this paper illustrates an interesting scenario in which a middle-age patient with unknown etiology developed a presentation of cellulitis of the mouth floor complicated by necrotizing fasciitis. This demonstrates the two similar clinical entities can occur in the same patient who has no medical condition. Therefore, early differentiation and detection is the key to change the management strategy.

Conclusion

Clinical presentation of cellulitis of the mouth floor can be obvious even in individuals with extensive underlying tissue destruction as seen in our case. It is important to maintain a high index of suspicion for necrotizing fasciitis in the setting of Ludwig's angina. Delay in the diagnosis and treatment is associated with a high mortality and morbidity.

References

1. Scully Cult, Langdon James and Evans J. "Marathon of Eponyms: 12 Ludwig Angina." *J Oral Dis* 16 (2010): 496-497.
2. Marple, Bradley. "Ludwig Angina: A Review of Current Airway Management." *J Arch Otolaryngol Head Neck Surg* 125 (1999): 596-599.
3. Murphy, Sharon. "The Person behind the Eponym: Wilhelm Frederick Von Ludwig (1790-1865)." *J Oral Pathol Med* 25 (1996): 513-515.
4. De-Vicente, Rodriguez. "Maxillofacial Cellulitis." *J Med Oral Patol Oral Cir Bucal* 9 (2004): 126-133.
5. Wasson Jones, Hopkins Cumper and Bowdler David. "Did Ludwig's Angina Kill Ludwig?" *J Laryngol Otol* 120 (2006): 363-365.
6. Roberto Bertolai, Alessandro Acocella, Roberto Sacco and Tommaso Agostini. "Submandibular Cellulitis (Ludwig's Angina) Associated to a Complex Odontoma Erupted into the Oral Cavity: Case Report and Literature Review." *J Minerva Stomatol* 56 (2007): 639-647.
7. Faisal A. Quereshy, Jonathan Baskin, Anca M. Barbu and Marc A. Zechel. "Report of a Case of Cervicothoracic Necrotizing Fasciitis Along with a Current Review of Reported Cases." *J Oral Maxillofac Surg* 67 (2009): 419-423.
8. Kavarodi, Marcos. "Necrotizing Fasciitis in Association with Ludwig's Angina: A Case Report." *Saudi Dent J* 23 (2011): 157-160.
9. Kristelle Chueng, David J. Clinkard, Danny Enepekides and Yousef Peerbaye, et al. "An Unusual Presentation of Ludwig's Angina Complicated by Cervical Necrotizing Fasciitis: A Case Report and Review of the Literature." *Case Rep Otolaryngol* 2012 (2012): 931350.
10. Jiménez Yolanda, Jose V. Bagan, Judith Murillo and Rafael Poveda. "Odontogenic Infections Complications Systemic Manifestations." *Med Oral Patol Oral Cir Bucal* 9 (2004):143-147.

How to cite this article: Luo Huang, Hui Qin, Ling Qiu, Kexiong Ouyang, et al. "A Case Report of Cellulitis of the Mouth Floor Presenting as Necrotizing Fasciitis." *Clin Case Rep* 11 (2021): 1408.