Craniocervical necrotizing fasciitis with hepatitis and diabetes mellitus

Shan Chen¹, M.D.; Leihui Chen¹, M.D.; Chongjin Feng¹*, Ph.D.
¹Department of Oral and Maxillofacial Surgery, the First Affiliated Hospital, Sun Yat-sen University, Guangzhou 510080, Peoples R China.
E-mail: chenshan00001@163.com

¹Department of Oral and Maxillofacial Surgery, the First Affiliated Hospital, Sun Yat-sen University, Guangzhou 510080, Peoples R China.
E-mail: happyberry1990.student@sina.com

*Corresponding author: Chongjin Feng, M.D., Ph.D., Department of Oral and Maxillofacial Surgery, the First Affiliated Hospital, Sun Yat-sen University, 58 Zhongshan Rd 2, Guangzhou, Guangdong, 510080, Peoples R China (Tel: 86-20-87755766 ext 8414. Fax: 86-20-87750632.
E-mail: drfcj@aliyun.com

ABSTRACT
Craniocervical necrotizing fasciitis is an uncommon but aggressive infection with high mortality. The aim of this article is to review a case of severe and extensive craniocervical necrotizing fasciitis (CCNF) worsened by hepatitis and diabetes mellitus in a 32-year-old man, with a successful outcome involving early diagnosis by CT scan, broad-spectrum antibiotics, multidisciplinary treatment, multiple surgical debridement, essential intensive medical care and efficient emergency services.

Indexing terms/Keywords
Craniocervical necrotizing fasciitis, mediastinitis, immunocompromised patient, surgical debridement

Academic Discipline And Sub-Disciplines
Oral and Maxillofacial Surgery

SUBJECT CLASSIFICATION : Medicine

TYPE (METHOD/APPROACH) : Case report

INTRODUCTION
1. Introduction
Necrotizing fasciitis (NF) is defined as an infection of potentially fatal infection characterized by rapidly progressive, widespread necrosis of the superficial fascia. The most frequently involved areas are the thorax, limbs, perineum, abdomen, and groin, and more rarely the head and neck (1, 2). When it occurs in the head and neck, it is called craniocervical necrotizing fasciitis (CCNF), and it is very aggressive and life-threatening. CCNF is reported to be rare (3, 4). It is a life-threatening infection originating chiefly from dental or oropharyngeal infection. It may spread to the chest, leading to a descending mediastinitis, which is the most virulent complication of CCNF (5). Immunocompromised patients are more susceptible to developing cervical necrotizing fasciitis, with diabetes mellitus the most common co-morbidity (6). Those who have delayed treatment of a pharyngeal infection and health conditions such as jaundice and hepatitis are also at risk.

We present a case of CCNF of pharyngeal origin with hepatitis and diabetes mellitus managed in our hospital.

Normal or Body Text
2. Case Report
A 32-year-old man presented to Department of Oral and Maxillofacial Surgery, the First Affiliated Hospital of Sun Yat-sen University, because of a 10-day history of left pharyngeal abscess and a 2-day history of pain and swelling at the left side of the neck. 8 days earlier, he had been started on abscess puncture to treat the abscess of the left pharyngeal. He smoked 30 cigarettes a day. His medical history was significant for hepatitis, diabetes mellitus and pneumonia. On admission, physical examination revealed a poor general status with submandibular swelling. Subcutaneous crepitus was palpable in the anterior aspect of the neck. Other major complaints include violent pain, dysphagia, odynophagia, and mild dyspnea. An emergence computed tomography (CT) scan showed a large amount of gas extending subcutaneously from the rear part of the mediastinum up to the anterolateral neck (Fig. 1). An abdominal ultrasound revealed pleural effusion and pelvic cavity effusion.

The patient's temperature was 37.8 °C. The white blood cell count was 11.07×10⁹/L, the platelet was 39×10⁹/L. The blood glucose level was higher than 10 mmol/L. The alanine aminotransferase was 121 U/L, the aspartate aminotransferase was 65 U/L, the gamma glutamyl transpeptidase was 90 U/L, and cholinesterase was 1728 U/L. Prothrombin time (PT) was 17.4 seconds, activated partial thrombin time (APTT) was 53.0 seconds.
The patient was admitted and started on cefuroxime and metronidazole to treat abscess. Entecavir tablets and insulin were prescribed to control hepatitis and diabetes mellitus respectively. On hospital day 1, under local anesthesia, two submandibular incisions and a wide debridement of the soft tissue were performed. Due to coagulation disorders, the patient can't tolerate surgery.

Two days later, the patient did not get better but was intubated in response to increasing respiratory distress. Then, the patient was transferred to the surgical intensive care unit (SICU) with a diagnosis of CCNF from pharyngeal origin, septic shock with acute liver failure, and a previously unknown diabetes mellitus. A new CT-scan and chest radiography were performed, which revealed mediastinitis, pleural effusion, pneumonitis, empyema, and pneumothorax. Then, thoracic surgery was consulted for a right thoracotomy and mediastinal debridement. Intraoperatively, purulent necrotic material was present. Two thoracostomy tubes were placed, allowing drainage. Approximately 1060 ml of pus was drained in the first 24 hours. Successive blood cultures revealed Gram-negative bacilli and Acinetobacter baumannii. The antibiotic regimen was accordingly changed to piperacillin sodium and tazobactam sodium and linezolid tablets. On hospital day 6, CT demonstrated subcutaneous air in the neck and abscess formed bilaterally in supraclavicular fossa and bilateral pleural effusions and pneumothorax and ascites (Fig. 2). The patient required abdominal puncture and further surgical debridement of the neck.

After timely supplement of coagulation factors by frozen plasma to improve blood coagulation function, on day 10, under general anesthesia, surgical debridement was performed aggressively. Three incisions in the skin and platysma muscle and blunt dissection through all cavities were made. Abscess, fluid, and necrotic fatty tissue superficial to the muscles and fascia were present. His blood coagulation dysfunction, entailed blood transfusion support everyday to improve blood coagulation function. Albumin, entecavir tablets and insulin therapy was applied for his severe hepatitis and diabetes mellitus throughout his stay in the SICU. With intravenous antibiotics and general support measures, the patient was improving and then was transferred to Digestive Internal medicine to treat hepatitis and diabetes mellitus on day 20 (Fig. 3). He fully recovered and subsequently was discharged from the hospital on day 33. No complications developed during 13 months of follow-up.

Subsequent Pages

3. Discussion

Cranio-cervical necrotizing fasciitis is uncommon and life-threatening. Most cases of this life-threatening condition result from the extension of infection from a pharyngeal or odontogenic source (710). Our patient's cranio-cervical necrotizing fasciitis was a result of pharyngeal infection. As an immunocompromised patient, and also with diabetes mellitus, the most common co-morbidity, he is more susceptible to developing CCNF (11). The expansion may be due to mechanical or chemical properties or both of the gas produced by the bacteria causing the fasciitis, or may directly indicate virulence of the causative bacteria. If it is produced in large quantities, such gas may dissect the tissues, especially along the fascial planes. While the process presented as an extended deep cervical infection and mediastinitis within a very few days, the development of descending mediastinitis and septic shock worsens the prognosis to this patient.

Early diagnosis of CCNF is the key in treating this infection. The aid of radiographic studies, especially the use of CT, may help in the early diagnosis of CCNF. Diffuse thickening and enhancement of the subcutaneous fat, cervical fascia, and muscles and gas and fluid within the soft tissues are always present on CT images (12). In addition, follow-up CT is useful for tracking the resolution of diseases and for demonstrating any residual fluid collection.

Early detection and adequate emergency treatment are critical in the management of these patients and may reduce mortality and improve survival. Aggressive surgical debridement of the deep cervical spaces and mediastinum, timely supplement of coagulation factors by frozen plasma and intravenous broad-spectrum antibiotic therapy are mandatory. Followed by early and aggressive surgical debridement, a multidisciplinary treatment with critical support and clinical stabilization of patients is critical for the management of this entity.

Besides, multidisciplinary treatment is also very important. As to this patient, his prolonged prothrombin time (PT) and activated partial thrombin time (APTT) was the results of hepatitis. Liver has a regulatory role on the fibrinolytic system. It can rapidly release antiplasmin into the blood and eliminate activator following stimulation of fibrinolytic activity. Therefore, patients with severe liver disease may present with increased abnormal secretion of heparin and heparin material, resulting in abnormal blood coagulation process. With severe liver disease, this patient's PT and APTT were significantly prolonged, which showed that he was in blood coagulation disorder state, with bleeding tendency. Thus, he got medicine to treat hepatitis and required blood transfusion support everyday to improve blood coagulation function.

In conclusion, CCNF with immunocompromised patients and thorax extension spread must be treated aggressively with early diagnosis by CT scan, broad-spectrum antibiotics, multidisciplinary treatment, multiple surgical debridement, essential intensive medical care and efficient emergency services.
FIGURES/CAPTIONS

Fig.1 Computed tomography (CT) scan shows a large amount of gas extending subcutaneously from the rear part of the mediastinum up to the anterolateral neck.

Fig.2 Computed tomography (CT) scan shows subcutaneous air in the neck, abscess formed bilaterally in supraclavicular fossa, bilateral pleural effusions and pneumothorax and ascites.

Fig.3 Computed tomography (CT) scan shows recovery of the oropharynx and mediastinal.
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