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Case Report

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Cutaneous anthrax associated with facial palsy: A case report

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ABSTRACT

Rationale: Anthrax is a zoonotic disease caused by spores of Gram-positive *Bacillus anthracis*, commonly affects mammals and in rare cases birds. Human infection occurs accidentally through direct or indirect exposure to animal or their products.

Patient concerns: A 63-year-old man was referred to our hospital with flu-like symptoms and severe swelling and redness on the face, the roof of the mouth, and nostrils. He had a history of direct contact with a slaughtered mutton two days ago. He declared controlled diabetes, hypertension, hypertriglyceridemia, and heart failure. Lungs were normal in lung high resolution CT, but multiple lymphadenopathies were seen in the mediastinum. Bilateral axillary lymphadenopathy with a maximum size of 23 mm and pleural effusion on the right side was observed. CT scan of the nose and sinuses showed an increased density of polyps in the left maxillary sinus. Slides were prepared from the patient's lesions and examined under a light microscope. *Bacillus* shape with *Streptococcus* bacteria was seen.

Diagnosis: Anthrax co-infection with herpes systemic virus and *Streptococcus pyogenes*.

Interventions: Multidrug therapy started with appropriate antibiotics.

Outcomes: The symptoms of the patient gradually disappeared. The patient was discharged without any complications.

Lessons: Cutaneous anthrax in endemic areas in patients with skin presentations and a history of contact with infected animals or products should be considered a differential diagnosis. This is more important in mixed infections where the main cause of the problem may be hidden.

KEYWORDS: Case report; Anthrax; Cutaneous; North East; Iran

1. Introduction

Anthrax also known woolsorters' disease, cumberland disease, maladi charbon, malignant pustule, malignant carbuncle, milzbrand, splenic fever, and siberian fever, is a bacterial zoonosis disease of concern[1]. The disease is still endemic in Middle East, central Asian, African, and some Mediterranean countries. Sporadic cases and outbreaks are occurring elsewhere. It has become a re-emerging disease in western countries and a cosmopolitan issue of concern. The disease primarily involves herbivores and all warm-blooded animals are susceptible[2]. Humans acquire disease *via* direct or indirect contact with infected animals or their products. The main clinical symptoms of the disease include cutaneous, respiratory and gastrointestinal anthrax. The most common form is cutaneous anthrax which includes 95% of all cases[3]. Data indicated that 10%-40% of untreated cutaneous anthrax cases might be expected to result in death, while less than 1% of cases are fatal with proper treatment. Although most cutaneous anthrax cases are self-limited, extensive edema and toxic shock can be seen as rare and potentially life-

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threatening complications of cutaneous form. Intravenous therapy with multiple drugs is recommended for cutaneous anthrax with signs of systemic involvement, such as extensive edema or lesions on the head and neck[2,3].

2. Case report

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

A 63-year-old man was referred to the hospital declared he had slaughtered mutton two days ago. Following direct hand contact with mutton and offal, he touched his face (scratched the nose and lips) with the same hand. At first his nose and oral mucosa started itching, consequently partial erythema and swelling in the same area occurred. After one day, he developed flu-like symptoms (runny nose, lethargy, and burning of the nasal mucosa). Within a day after the onset of the symptoms, the patient developed severe swelling and redness of the face, roof of the mouth, and nostrils, as well as sores with a black center in the same area. The patient stated that he had severe shortness of breath and also had a feeling of fullness in his nose, so that he could not breathe through his nose.

At the time of admission to the hospital, the patient was conscious but complained of severe shortness of breath, for which he underwent oxygen therapy with a mask. The patient's vital signs at baseline were as follows: blood pressure was 135/80 mm Hg (reference range: 133/69 mm Hg); heart rate 88 beats/minute (reference range: 60-100 beats/m); temperature 38 °C (36.1-32.7 °C); number of breaths 20/minute (reference range: 12-18/min).

On physical examination, the patient's face had severe diffuse erythema and was completely edematous. Also, the tip of his nose and upper lip were ulcerative and black. On examination of the nasal passages, the mucosa was completely black and swollen, and palate had relatively small erythematous lesions (about 5 mm in diameter) and numerous, but the skin of the upper lip (vermilion) was completely black and ulcerative with scaling. Also, the inner part of the upper lip (mucosal surface) had yellow and black lesions. In addition, the black lesions were not painful (tenderness). Other examinations were normal. The patient had a history of controlled diabetes, hypertension, hypertriglyceridemia, and heart failure. The patient declared that inside of abdomen and liver of butchered animal, was full of blood clots and also the liver was completely abnormal and destroyed. Samples were taken from the patient's lesions as well as blood for laboratory tests. Direct smears from the lesions were prepared and stained. For more investigations, PCR performed on the patient's skin samples. In microscopic examinations of slides, long capsule *Bacillus* and *Streptococcus* were observed (Figure 1). Sections show squamous epithelium with marked spongiosis & necrotic debris. Sub-epidermal blister containing hemorrhage & mixed inflammatory cells. Underlying dermis reveal marked edema & infiltration of lymphoplasm cells & PMNs.

In PCR of patient's samples, *Streptococcus pyogenes* and herpes systemic virus 1&2 were detected (*Streptococcus pyogenes*, sense, 5'-ACAGAGGAAGAAGGTTGATGAAG-3' and antisense, 5'-ACTCATTCGCTGCTTGACTG -3', and HSV-1 and 2, sense 5'-CATCACCGACCCGGAGAGGGAC-3', and antisense, 5'-GGGCCAGGCGCTTGTGGTGTA-3').

CT scan of the nose and sinuses showed an increase in the

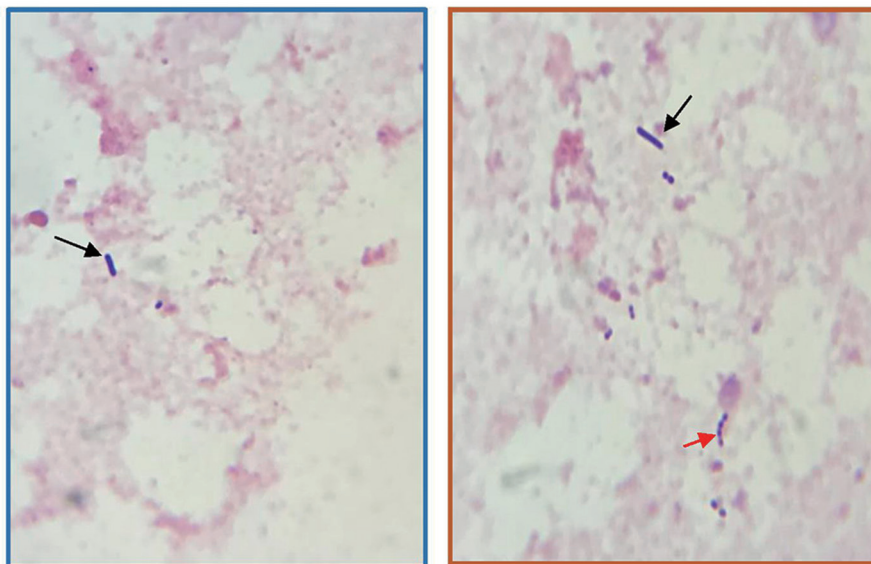


Figure 1. Microscopic examination slides of a 63-year-old patient of cutaneous anthrax associated with facial palsy showing *Bacillus* (black arrows) and *Streptococcus* bacteria (red arrow) infection.

density of polyps or retinal detachment in the left maxillary sinus, but the size and density of the paranasal sinuses were normal and no turbidity was seen. Also, the size and density of both lungs were normal in lung HRCT without injection, but multiple lymphadenopathy was seen in the mediastinum with a maximum sad of 14 mm. Bilateral axillary lymphadenopathy with a maximum sad of 23 mm and pleural effusion on the right side was observed. The umbilicus of both lungs was normal and the airways on the both sides were open. In visible parts of the abdomen, para-aortic lymph nodes with a maximum sadness of 16 mm and a brief fluid around the liver were observed. Following the observation of pleural effusion on CT scan, the patient was taped with pleural fluid under ultrasound guidance, about 25 mL of fluid from the right pleural space. After sampling, the patient was prescribed with following drugs with suspicion of a multi-factorial infection (charbon, group A *Streptococcus*, and herpes simplex infection): vial cefepime (1 gr/8 h), vial vancomycin (1 gr/12 h slow infusion), vial ciprofloxacin (400 mg/12 h), cap doxycycline (100 mg/12 h, orally), ointment tetracycline (per 12 h), amp acyclovir (per 8 h), spray NaCl (2 puff/12 h), amp apotel (1gr at first arrive of patient continued each 12 h). During multidrug therapy, the patient developed gastrointestinal complications such as brief nausea, bloating, and diarrhea, which was self-limiting. During the treatment period, the patient's necrotic lesions gradually became smaller and improved. Also, the patient's respiratory problems were completely eliminated.

3. Discussion

The diagnosis of anthrax is critical. If a patient with signs such as painless papules on skin along with pruritus surrounded with vesicles is referred to the medical care centers, anthrax should be considered in differential diagnosis. The detection of any case is alarming for health systems to enforce preventive measures for this neglected disease. Epidemiologic studies and case reports have shown that most cases of anthrax infection occur in people living in rural areas or in occupations related to animal products. It is suggested that in anthrax-endemic regions, any suspicious lesions or none pitting edema, with or without necrotic lesions, should be examined for anthrax. The most common form of anthrax infection in humans is cutaneous which is diagnosed by a topical skin lesion with central scar and marked non-pitting edema. Cutaneous lesions in anthrax mostly occur on the arms and hands, followed by face and neck. Infection initially appears as an itchy papule, like an insect bite. The papule enlarges during 1 to 2 days and produces a sore, which may be surrounded by vesicles. The lesions are round and regular and 1 to 3 cm in diameter. The production of toxins

by bacteria causes the sore to develop a black scar along with edema. However, the lesions and edema are painless. The lesion dries up after 1 to 2 weeks and the scar being lost in a short time. In contrast, the prevalence of respiratory anthrax is very low. It usually presents with acute respiratory symptoms such as dyspnea, hypoxia, and hypotension 1-3 days after the onset of disease that may results to death within a short time. Reports of cutaneous and gastrointestinal anthrax are available from different parts of Iran. A report of 28 human cases of anthrax from Esfarayen village in North Khorasan province during 2009 in Iran was published. The cases were occurred after an outbreak of anthrax among sheep in the village and consumption of their meat by the villagers. Animal examination of all areas revealed that 16 of them were infected with anthrax[4]. In another report from North Khorasan province in 2015, Hashemi et al. described a case of fatal gastrointestinal anthrax. The patient was a 24 years old man with a history of eating goat meat. The case did not respond to antibiotic therapy and expired 9 hours after hospitalization[5]. Similar study from Kermanshah province (Western Iran) by Hatami *et al.* reported two cases of gastrointestinal anthrax which passed away despite of antibiotic therapy[2]. In a retrospective study carried out in Mazandaran province, 28 cases of anthrax were reported during 2004-2014. Among all, 10 (35%) were rancher and others direct or indirectly were in contact with animals or their uncooked meat[6]. Moreover, a systematic review conducted in 2006 showed that totally 82 cases of respiratory anthrax were reported between the years 1900 to 2005[7]. This reveals that anthrax is a potentially life-threatening disease while rarely reported.

In animals with respiratory anthrax infection, bacteremia is reported in the early phase of the disease before fulminant symptoms. Bacteremia rapidly disappears after antibiotic treatment. Administration of antibiotics before blood culture, which is the most definitive method for diagnosis leads to negative result. Animal vaccination has been suggested to reduce the infection in man[8,9]. *Bacillus anthracis* generates cephalosporinase that inhibits the activity of cephalosporins, such as ceftriaxone. Therefore, cephalosporins should not be prescribed for treatment of anthrax. Early detection and treatment, proper antibiotics, pathogenesis of the bacteria, host sensitivity and immunity or a combination of these factors contribute to the patient survival[1,9].

In addition, many attentions are needed for controlling the disease and prevention of illegal slaughter. Slaughter of animals should be done in the abattoir and under the supervision of a veterinarian and in compliance with hygienic protocols. In cases where the slaughter is carried out in a place other than the slaughterhouse, observance of the hygienic points and awareness of animal diseases is of special importance.

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Conflict of interest statement

The authors declare that they have no conflict of interest.

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Authors' contributions

MG: Concept and design of study and final approval. SMM gathered the data and wrote the manuscript draft. AS critically revised and final approval of the manuscript, submission, and revision.

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