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CASE REPORT

Cutaneous anthrax cases leading compartment syndrome

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ABSTRACT

Bacillus anthracis is the causative agent of anthrax. Anthrax is a zoonotic disease with three clinical forms. Clinical forms are skin, gastrointestinal and inhalational anthrax. Cutaneous anthrax is 95% of the cases. Cutaneous anthrax frequently defines itself. Clinical presentation of anthrax may be severe and complicated in some cases. There may seem complications like meningitis, septic shock and compartment syndrome. Compartment Syndrome is a rare complication of cutaneous anthrax and it is life threatening. Physicians working in the endemic area should be aware of this form. In this study, three cases were shown which developed compartment syndrome following cutaneous anthrax. *J Microbiol Infect Dis 2013;3(4): 214-217*

Key words: anthrax, compartment syndrome, bacterial infection, zoonosis

Kompartman sendromuna neden olan deri şarbonu olguları

ÖZET

Bacillus anthracis şarbon hastalığının etkenidir. Şarbon, üç klinik formu bulunan, zoonozdur. Klinik formlar deri, inhalasyon ve gastrointestinal şarbondur. Olguların % 95'ini deri şarbonudur. Hastalık genellikle kendi kendine sınırlanır. Şarbonunun klinik seyri bazı olgularda ağır ve komplike olabilir. Menenjit, septik şok ve kompartman sendromu gibi komplikasyonlar görülebilir. Kompartman sendromu nadir ve hayatı tehdit eden bir komplikasyondur. Endemik bölgelerde çalışan klinisyenler bu tablonun farkında olmalıdır. Bu çalışmada deri şarbonu sonrası kompartman sendromu gelişen üç olgu incelendi.

Anahtar kelimeler: Şarbon, kompartman sendromu, bakteriyel infeksiyon, zoonoz

INTRODUCTION

Anthrax is a disease caused by a gram-positive bacteria named *Bacillus anthracis*.[1,2] It is transmitted via direct contacts like slaughter and skinning of a sick animal, inhalation of spores and consuming infected meat.[1,3] Due to the route of entry of the agent there may develop cutaneous, gastrointestinal and pulmonary anthrax.[4,5] Known risk factors are slaughter of sick animals, direct contact with contaminated meat, skinning of a slaughtered animal and consuming contaminated meat.[6] All three clinical forms of anthrax might be mortal. Mortality rates are; at meningoencephalitis 100%; at pulmonary anthrax 92%; at gastrointestinal anthrax between 25% and 75%.[7] In case of cutaneous anthrax disease, considering the fact that the treatment administered lately or insufficiently the patient may go into a septic shock and may result even death.[6] Rarely there may develop compartment syndrome due to the excessive edema and vascular pressure. We describe three patients who had cutaneous anthrax on the forearm and arm.

CASE 1

A 50-year-old male farmer slaughtered his sick bovine animal. Ten days after the slaughtering has been noticed black lesion and edema. Thirteen days after the slaughtering a pruritic pustule had occurred on the farmer's body. In a couple of days that pustule had turned into a scar. When the patient had admitted to the clinic, there was a wide edema and erythema on the left arm with 3x2 cm measured crusted hemorrhagic scar. On the left backhand there were hemorrhagic bullae, there were echymotic lesions interdigital and metacarpal areas. Passive movements of corpus were noticed and fingers accompanied by intense pain. Radial pulse was palpable. As for ulnar digital pulse

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monophascic current was available with Doppler. In physical examination, body temperature was 39°C, arterial blood pressure was 120/65 mmHg, pulse was 96 beats/min. Laboratory findinds showed: leukocyt (WBC) 19200/mm³ (ref.4000-10000); erythrocyte sedimentation rate (ESR) 29 mm/h(0-16 mm/h); C- Reactive Protein (CRP) 30 mg/l (0-5 mg/l); Aspartate aminotransaminase (AST) 66 IU/l (0-31); Alanine aminotransaminase (ALT) 93 IU/l (0-37). Gram positive bacillus was seen in the lesion smears. There was no reproduction in blood and swap culture. The patient was diagnosed with cutaneous anthrax and treated with crystalline penicillin G and steroids.

Upper extremity venous Doppler ultrasonography (USG) was normal. In the arterial Doppler USG, ulnar arterial blood current was decreased in comparison to the radial. In the superficial monitoring USG of the patient who had an extensive edema, strictness and pain, abscess was shown. Fasciotomy was diagnosed as compartments and it was operated by orthopedists. No abscess was found in surgery, muscles were fine, and there was no color change.

CASE 2

A 41-year-old male patient who works at animal husbandry had slaughtered five sick bovine animals a week before. On the same night that day, he had a slight diarrhea. One or two days after he had a pruritic, pustule-like lesion. The patient had squeezed that lesion and after that the lesion was cut and excreted by a primary care phsycian the patient had admitted. He had administered with penicillin treatment. A day after the panicula had extended to his shoulder. He had hospitalized for two days at public hospital. As his condition got worse, he was transferred to emergency service of our hospital. In his examination, paresthesia, edema and pain on the left arm were found. On his left carpal volar surface 3x4cm measured hemorrhagic crust, erythema from arm to shoulder, panicula, bullae from place to place, necrotic centers and common edema was observed (Figure 1 a and 1 b).

Figure 1 a. and **1 b.** Edema and necrosis on the anthracis lesion

The procaine penicillin treatment which was administered at public hospital before was carried on. Pulse was not palpable. Urgent fasciotomy was operated at orthopedic department. In the operation fasciotomy was operated parallel to the second and fourth metacarpals on hand dorsal, and carpal tunnel was caved on the wrist volar surface level; fasciotomy was operated on the forearm volar surface downward to the cubital area via that carpal tunnel. Due to the lack of blood current in brachial arteria, fasciotomy was operated till middle arm line, on that level onset brachial blood current was seen. There were some ischemic changes monitored in biceps brachi muscle. The patient was admitted to our clinic a day after operation. In physical examination body temperature was 38°C, arterial blood pressure was 110/65 mmHg, pulse was 80 beats/min. Laboratory findings were; WBC 15,500/mm³, CRP concentration 149 mg/l (0-5 mg/l), ESR 16 mm/h (0-16), AST 42 U/l (0-31), ALT 25 U/l (0-37). There was no reproduction in material derived from vesicular lesion and in blood culture. There was seen no microorganisms in straightforward search. The edema was regressed, there was no fever and the treatment covered 10 days. Urgent fasciotomy was operated by orthopedic surgeons. On tenth day of treatment-as the ninth day of fasciotomy- a covering surgery for fasciotomy was operated by the orthopedist after the last reevaluation.

CASE 3

A 35 year-old male patient who works at husbandry had seen scars and panicula on the left hand and on left forearm. He had hospitalized and consulted by dermatology department at a public hospital. As the patient had cooling on his fingertips, he had been transferred to emergency service. In the initial examination there were seen erythematous lesions; 2 x 2cm measured lesion on the left hand dorsal and two lesions on the left hand forefinger necrotic crusted in the center and edematous on the sides. There was seen a widespread area of edema, disseminated bullae and from place to place cyanosed, crusted lesions. Fingertips were cooled down and pulseless. On the upper extremities there was rash and increased temperature. Pulse could be measured with Doppler only. Left elbow and left wrist movements were limited and dolorous. In physical examination, body temperature was 37°C, arterial blood pressure was 110/70 mmHg, pulse was 85 beats/min. Laboratory findings were; WBC 17800/mm³, CRP concentration 15 mg/l (0-5), ESR 100 mm/h (0-16), AST 96 U/l (0-31), ALT 82 U/l (0-37). In direct search there was seen no microorganisms. There was no reproduction in cultures. We thought about the diagnosis compartment syndrome due to the cutaneous anthrax. As a treatment, crystalline penicillin G for five days, and procaine penicillin for five days were administered. Anti-edema treatment and medical dressings were administered. A week after complaints were regressed and he had no fever. Although not having any surgery, the symptoms of compartment syndrome were gone.

DISCUSSION

Anthrax is a zoonotic bacterial infection transmitted via entrance of *Bacillus anthracis* spores through bruises and chaps of skin, inhalation and digestion. It is transmitted to human via contact of sick animals and veterinary products like fur, skin, meat etc. It remains as a common health issue in developing countries, although there is a decreasing numbers of cases in developed countries.[1,2] Virulence factors are polysaccharide capsule and extracellular toxin. Anthrax toxin is composed of three proteins named edema factor (EF), protective antigen (PA) and lethal factor. These proteins are not toxic alone. Combination of PA and EF leads to decrease in edema and polymorphonuclear neutrophile functions.[2] In all three cases progressive edema was monitored.

Cutaneous anthrax is seen 95% of cases worldwide. The infection limits itself. Lesions can be treated without complications and with scar 80 to 90 percent. Clinical state changes slight lesions to severe forms. Life-threatening important complications are extreme edema and toxic shock. In cutaneous anthrax cases mortality rates were 10 to 40 per cent before immunization and modern therapies whereas it is below rate of 1 per cent at present.[8] Risk groups include butchers, farmers and veterinarians. Lesions are mostly seen at upper extremity, head and cervix.[5] In our cases, the patients were farmers or people who work at animal farms or husbandry. All three had lesions on hand and arm.

Acute extremity compartment syndrome (AECS) is a common threat for extremities leading ischemia in muscles and nerves because of bleeding and edema; as is also a life threatening factor. It occurs due to the increasing pressure in closed fascial area.[9] AECS occurs mostly in upper extremities in result of too many factors. These factors are bone fractures, muscle and soft tissue injuries, compression, ambustion, bug bite, excessive training, bleeding, intravenous drug use and infection. It requires immediate surgery. There might occur high pressure in tissues because of edema which restricts compartment area.[10] In our cases we thought that extreme edema was so severe that it led to pressure increase in compartment. In the case 2, the patient was affected on muscles. The color change on skin was normalized, and the pain was stopped thanks to the fasciotomy. The first and the second case benefited from fasciotomy, besides, in the case 3, anti-edema and antibiotics therapy were effective enough so fasciotomy was not necessary.

Symptoms of AECS are persistent severe pain and paraesthesia. There might be some other symptoms like panicula, being pulseless and colour change due to circulatory disorder. It may be accompanied by sense and motor deficits due to the effected peripheral nerves. In worse cases which are not treated, muscle and nerve ischemia and paralysis can be seen due to the arteriolar compression. Complaints of the patient and examination findings are more valuable data than the other researches.[10] In our cases there was no nerve involvement, aside from some symptoms due to the circulatory disorder.

Infections are rare reasons of AECS.[9] A case of 45 year-old Crimean-Congo Hemorrhagic Fever was reported, in which compartment syndrome was developed secondary to bleeding in the upper extremity.[11] In our cases increase of pressure was related to the edema; in contrast to that case in which pressure increase was related to bleeding.

In cutaneous anthrax cases, incision at early stage is not recommended because of the prob-

ability of progression in infection and septicemia.[5] There were cases reported about anthrax disease progressing with palpebra involvement, and reconstructive surgery was operated to fix scatricial ectropion.[12,13] Cases were reported in which soft tissue necrosis developed on right hand dorsal due to the anthrax, backflow forearm flap was operated.[14] We showed that in case of compartment syndrome surgery can be operated in addition to the medical treatment in proper cases. Deteroriation and/or septicemia were not seen in our cases.

In compartment syndrome, if it is diagnosed and decompressed in 6 hours, full recovery can be achieved; on the contrary, possible results after more than 12 hours might be unsatisfactory.[15] Both of our cases benefited from early operated fasciotomy; recovered with no functional or tissue loss.

Penicillin treatment is a still preferred therapy. Doxycycline and ciprofloxacin are best alternatives accepted. World Health Organization (WHO) recommends procaine penicillin, amoxicilline and penicillin V in uncomplicated minor cases, and intravenous penicillin in major cutaneous anthrax cases with extreme edema.[8] We administered crystalline penicillin G, procaine penicillin or quinolone. WHO recommends 3-7 days of treatment in uncomplicated minor or mild cases. The lesion regresses after 7-10 days from the therapy start.[8] In our cases the lesion was regressed and the pain was relieved in a week time.

Spores are resistant to environmental effects. It can survive in barns or pasturelands for years. Immunization of animals and proper burial of dead animals might prevent emergence of new cases.[4] In all three cases, there was anamnesis of bovine animal slaughtering or contact of skin or meat was reported. In our country, if we prevent direct contact and slaughter of sick animals we might decrease these cases about rate of 90 per cent.[4]

In our country, anthrax has remained an endemic disease yet. Although to a lesser extent, life threatening incidents might be seen in cutaneous anthrax. Early diagnosis, proper treatment and intervention are lifesaving factors. Clinicians work in that region should be aware in case of sick animal and/or animal products contact. Typical lesion of the disease should be identified. Immunization of animals, educating people and prevention of sick animal slaughter should be done. It should not be forgotten that compartment syndrome may develop albeit it's rare. Very early diagnose and effective intervention might help saving extremities and even lives.

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