

A Rare Presentation of Anthrax: Preseptal Cellulitis

Şarbon için Nadir Bir Başvuru Şekli: Preseptal Selülit

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Abstract

Anthrax is a rare zoonotic infectious disease caused by *Bacillus anthracis*. Cutaneous anthrax constitutes most of the cases and occurs in exposed skin areas (arms, fingers, etc.). Here we report a case of periorbital cutaneous anthrax infection. An 8-year-old girl presented to our hospital with a necrotic black lesion on the upper left eyelid and swelling around the eye. She had a history of contact with a dead cow. Her father and brother also had necrotic ulcerative lesions on their forearms. Gram-positive rods were detected in the swab Gram strain taken from her lesion. In addition, *Bacillus anthracis* growth in swab culture taken from her father's lesion. She was diagnosed as "Anthrax" and treated with intravenous penicillin-G and topical oxytetracycline for 10 days. She recovered without functional sequelae. Physicians should consider cutaneous anthrax in the differential diagnosis of ulcerative and necrotizing preseptal and orbital infections, particularly in underdeveloped and developing countries.

Keywords: Cutaneous anthrax, periorbital anthrax, preseptal cellulitis, anthrax

Öz

Şarbon, *Bacillus anthracis*'in neden olduğu nadir görülen zoonotik bir enfeksiyon hastalığıdır. Olguların çoğunu deri şarbonu oluşturur ve açıkta kalan deri bölgelerinde (kollar, parmaklar, vb.) meydana gelir. Burada periorbital kutanöz şarbon enfeksiyonu olan bir olguyu sunuyoruz. Sekiz yaşında kız hasta, sol üst göz kapağında nekrotik siyah lezyon ve göz çevresinde şişlik şikayetiyle hastanemize başvurdu. Ölü bir inekle temas öyküsü mevcuttu. Babasının ve erkek kardeşinin de ön kollarında nekrotik ülseratif lezyonlar vardı. Lezyondan alınan sürüntüde Gram-pozitif çomaklar tespit edildi. Babasının lezyonundan alınan sürüntü kültüründe de *Bacillus anthracis* üremesi oldu. "Şarbon" tanısı konan hastaya 10 gün boyunca intravenöz penisilin-G ve topikal oksitetrasiklin tedavisi uygulandı. Fonksiyonel sekel kalmadan iyileşti. Özellikle az gelişmiş ve gelişmekte olan ülkelerde hekimler ülseratif ve nekrotizan preseptal ve orbital enfeksiyonların ayırıcı tanısında kutanöz şarbonu da göz önünde bulundurmalıdır.

Anahtar Kelimeler: Kutanoz şarbon, periorbital şarbon, preseptal selülit, şarbon

Introduction

Anthrax is a zoonotic infectious disease caused by *Bacillus anthracis*. Essentially, anthrax is a disease of animals, but it can be transmitted from domestic animals such as sheep, goats, and cattle to humans. *B. anthracis* is a Gram-positive, aerobic, and facultative anaerobic, endospore-forming bacillus. These spores can remain in nature for a long time.

It is transmitted to humans by ingesting infected meats, inhaling spores, or by direct contact with the skin, meat, and wool of infected animals⁽¹⁾. Although in developing countries it causes animal and human deaths, in most industrialized countries, anthrax is considered a bioterrorism threat⁽²⁾. There are three clinical types of anthrax, according to the inoculation of spores into the body: Cutaneous, inhalation,



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and gastrointestinal. Cutaneous anthrax constitutes 95% of the cases, and it mostly occurs in exposed skin areas (face, neck, hands, forearms, etc.)^(3,4). Herein, we report a case of periorbital cutaneous anthrax infection in an 8-year-old girl from Turkey with a brief literature review.

Case Report

An 8-year-old girl presented to our hospital with a black lesion on the upper left eyelid and swelling around the eye. The patient's complaints started two days ago. There was no history of trauma or insect bite. The patient's family had been ranching in the village, and their cow had died suddenly a week ago. She, her father, and her brother had contacted the dead cow's meat and skin. Her father and brother also had necrotic ulcerative lesions on their forearms. On physical examination, her body temperature was 38.7 °C, and other vital signs were normal (heart rate: 90 beats per minute, breath rate: 20/min, oxygen saturation: 99% at room air, blood pressure: 110/70 mmHg, Glasgow-Coma score: 15). There was a black necrotic area, swelling, and redness around the left eyelid (Figure 1). In laboratory evaluation, white blood cell count was 9310/mm³ (normal range 4500-9500/mm³) and C-reactive protein was 103 mg/

dL (normal range 0-8 mg/dL). She was also evaluated by an ophthalmologist, and the ophthalmological examination showed normal anterior and posterior segments. Orbital magnetic resonance imaging was performed to exclude orbital cellulite. The findings were consistent with preseptal cellulitis (left frontal, periorbital, zygomatic, and molar subcutaneous edematous signal increases). A swab sample was taken from the necrotic lesion in the eyelid, and Gram staining and culture were performed. Gram-positive rods were observed on microscopic examination. Swab culture was negative. In her father's examination, there was a 2x1 cm black, necrotic, ulcerated lesion on the forearm (Figure 2). *Bacillus anthracis* was isolated from the swab culture taken from the lesion on her father's arm. On microscopic examination, Gram-positive rods were also observed in the swab sample of her brother's necrotic lesion on his forearm. Based on all clinical and laboratory findings, the preseptal cellulite in our patient was considered caused by anthrax. Intravenous Penicillin-G and topical Oxytetracycline treatment were initiated. Other family members were also screened for anthrax. On the third day of treatment, edema and redness of the left eyelid began to regress. Penicillin G treatment was completed in 10 days. The necrotic scar tissue on the eyelid did not regress and was followed up (Figure 3). Three months after treatment, it regressed,



Figure 1. Periorbital cellulitis in the left eye and black necrotic area on the lesion at presentation



Figure 2. Necrotic lesion on the forearm of the patient's father

and no functional or cosmetic loss occurred in the eyelids (Figure 4).

Discussion

Here we report a case of anthrax with a rare presentation, preseptal cellulitis. Anthrax has not been eradicated worldwide, although its frequency has decreased gradually. Because butchers, veterinarians, shepherds, farmers, and farmworkers are at great risk of exposure to infected substances, anthrax can also be considered an occupational disease, especially in underdeveloped countries. It is a common disease, especially in parts of Asia, Africa, Latin America, and Eastern Europe⁽⁵⁾. In our country, it seems more frequent in Eastern and Southeastern Anatolia areas with intense contact with animals⁽⁶⁾. In 2017, 37 cases were reported from 14 provinces in Turkey, but no cases were reported from Sivas. However, in September 2018, anthrax came back on the agenda due to cases in provinces of Turkey, including Sivas. Anthrax reminds us of a serious and fatal disease that can affect both animals and humans. In Sivas, in three patients from the same family, cutaneous anthrax was defined. Here we present a child with preseptal cellulitis in this family.

Anthrax, which can be observed in all animal species, is transmitted mainly from animals such as cattle, sheep, and goats. *B. anthracis* can be transmitted to humans by contact with the wool or meat of infected animals, by inhalation, or by digestion. Although periorbital cellulitis due to anthrax is a rare disease, it should be considered in the differential diagnosis of patients who have close contact with animals or animal products. In our patient's case, the patient's family had been ranching in the village. Their cow had died suddenly, and then her father, brother, and she had contacted the dead cow's meat and skin. Taking this story is important for diagnosis. Symptoms begin to appear 2-7 days after the bacilli enter the body. It starts with swelling and itching on the skin and turns into a blister filled with water within 1-2 days, and then a black wound occurs in the middle. Overall, the most common locations of cutaneous anthrax are the arm and finger. The incidence of lesions around the face and neck region has been reported to be 20%⁽⁷⁻¹⁰⁾. Facial involvement has been observed in five of seven cutaneous anthrax cases diagnosed between 2017 and 2018 in Turkey. Child and adult anthrax cases causing preseptal cellulitis similar to our patients have been reported in our country and other countries. Bayoğlu et al.⁽¹¹⁾ reported



Figure 3. Necrotic scar tissue in the left eye during the first week during discharge



Figure 4. Fully healed image of the patient after 6 months

anthrax on the right eyelid of an 8-year-old patient in 2013, and he healed without complications. In 2016, Gül et al.⁽¹²⁾ reported a 14-year-old patient with a preliminary diagnosis of angioedema who noticed a classic necrotic black lesion in the left eye. Gram-positive bacilli were found in this case, and the patient was treated for anthrax, but scar tissue remained. In a study conducted in India, the causes of preseptal and orbital cellulitis were found to be associated with cutaneous anthrax in 5% of pediatric cases and 21% of adult cases⁽¹³⁾. In a previous study, Gelaw and Asaminew⁽¹⁴⁾ reported a case series of three adult patients with periocular anthrax who were seen in Ethiopia from June 2011 to May 2012. In another study, Munteanu et al.⁽¹⁵⁾ reported a 21-year-old patient with palpebral anthrax.

In the differential diagnosis of cutaneous anthrax, carbuncles, erysipelas, necrotizing cellulitis, cutaneous tuberculosis, tularemia, leishmaniasis, primary syphilis lesion, cat scratch disease, necrotic herpes simplex, and tropical ulcers should be considered^(7,16,17). Anthrax can be diagnosed by detecting bacillus by Gram staining of a swab from the lesions, positive culture, and clinical and laboratory findings. Diagnosis can be rapidly made also by polymerase chain reaction or enzyme-linked immunosorbent assay testing techniques⁽¹⁸⁾.

Delay in treatment or misdiagnosis can lead to varying complications ranging from local complications to death, including ectropion (47%), lagophthalmos (19%), and corneal scars (9.5%) in periorbital anthrax cases⁽¹⁹⁻²³⁾. The biggest anthrax epidemic in the past few years was the Zimbabwe epidemic, which affected 9.711 people in the late 1970s⁽²⁴⁾. Reported cases from Zimbabwe continue to be reported. In 2015, a 3-year-old patient similar to our case was reported to have cutaneous anthrax diagnosed on the left upper eyelid, and the lesion had occurred 2 weeks after eating dead antelope meat⁽²⁵⁾. It was reported that the patient was reconstructed with a skin graft at the end of the treatment and developed mild lagophthalmos. Celebi et al.⁽²⁶⁾ reported that a 4-year-old child developed cicatricial ectropion on the upper eyelid despite anthrax treatment with high-dose penicillin. Similarly, there have been reported pediatric and adult periorbital anthrax cases resulting from ectropion and corneal scars despite appropriate treatments^(20,22,27,28). Unfortunately, the youngest reported case was a 4-month-old baby among these cases⁽²²⁾. Dinc et al.⁽²⁹⁾ reported a 36-year-old farmer patient who was clinically and bacteriologically diagnosed with cutaneous anthrax with a necrotic lesion on the

left eyelid and facial edema. There were no sequelae, except for bilateral pterygium, after 10-day penicillin-G treatment⁽²⁹⁾. In another study, Bozpolat et al.⁽³⁰⁾ reported a 13-year-old female patient with cutaneous anthrax in her left eyelid. After 14 days of penicillin treatment, tissue grafting was performed on the lesion area with tissues taken from the postauricular areas. Devrim et al.⁽⁵⁾ presented a case of a 13-year-old boy who developed eyelid anthrax after contact with a sheep carcass, resulting in eyelid anthrax and cicatricial ectropion.

In underdeveloped and developing countries, physicians should consider cutaneous anthrax in the differential diagnosis of ulcerative and necrotizing preseptal cellulitis and orbital infections, especially in patients with a history of close contact with animals. Recognition of the signs and symptoms by physicians is very important for early and effective treatment. Sequelae development can be prevented in this way. With this article, we also want to highlight the presence and importance of the infectious zoonotic disease "Anthrax" for public health, especially in underdeveloped and developing countries. Therapeutic and preventive measures should be taken.

Ethics

Informed Consent: We obtained written informed consent from the patient's parents for this report.

Authorship Contributions

Surgical and Medical Practices: A.K.T., A.K.A., A.V.Ö., Concept: A.K.T., A.K.A., A.V.Ö., Design: A.K.T., A.K.A., Data Collection or Processing: A.K.T., A.K.A., Analysis or Interpretation: A.K.T., Literature Search: A.K.T., A.K.A., Writing: A.K.T.

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