Cutaneous Palpebral Anthrax: Case Report

Göz Kapağında Deri Şarbonu

ABSTRACT Anthrax is an infectious disease caused by *Bacillus anthracis*. Cutaneous, respiratory and gastrointestinal involvements can be seen. The face and eyelids are the most commonly involved areas in cutaneous anthrax. If eyelid anthrax is not promptly treated, it may cause severe deformities of eyelids including cicatrization and ectropion. In this study, a 38-year-old male presenting with right eye palpebral cutaneous anthrax was reported. There was periorbital edema and a black eschar on right eyelid leading to closed eye. Gram-positive rods were seen in the microscopic examination of scrapings taken from the lesion. The patient received high dose intravenous penicillin G (16 million units/day) for 10 days. The lesion healed within 10 days, leaving no clinical evidence of any eyelid deformity. No eyelid deformity was observed during one-year follow-up period. Observing this case suggested that early diagnosis and treatment can prevent development of potential anthrax eyelid complications.

Key Words: Eyelids; Bacillus anthracis; outcome assessment

ÖZET Şarbon, *Bacillus anthracis* tarafından oluşan enfeksiyöz bir hastalıktır. Deri, solunum ve gastrointestinal sistem tutulumları görülebilir. Deri şarbonunda yüz ve göz kapakları en çok tutulan alanlardır. Eğer göz kapağı şarbonu hemen tedavi edilmezse göz kapağında kabuklanma ve ektropion gibi komplikasyonlara neden olabilir. Bu çalışmada sağ göz kapağında deri şarbonu olan 38 yaşındaki erkek hasta bildirildi. Sağ göz kapağının kapanmasına neden olan siyah eskar ve periorbital ödem mevcuttu. Lezyondan alınan kazıntıların mikroskopik incelemesinde Gram-pozitif çomaklar görüldü. Hastaya 10 gün boyunca yüksek doz intravenöz penisilin G (16 milyon ünite/gün) verildi. Lezyon herhangi bir göz kapağı şekil bozukluğu bırakmadan 10 gün içinde iyileşti. Bir yıllık takip ve tedavinin Şarbonda potansiyel gözkapağı komplikasyonlarının gelişimini engelleyebileceğini göstermektedir.

Anahtar Kelimeler: Göz kapakları; Bacillus anthracis; sonuç değerlendirmesi

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nthrax is an infectious disease caused by *Bacillus anthracis*. There are three forms of the disease in humans: cutaneous, respiratory and gastrointestinal. The cutaneous disease accounts for the most of the cases in humans. Anthrax is commonly seen among farmers, butchers, veterinarians, shepherds and farm workers. The disease affects primarily herbivores including sheep, cattle, horses and other domestic animals. Humans may rarely be affected. Transmission may result from ingestion of grossly contaminated or undercooked meat or bones of infected animals, contact

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with hair and wool of infected animals, or inhalation of the spores.¹ The first sign of cutaneous anthrax is severe and massive edema around the site of infection. A black, necrotic, central eschar then forms, which is characteristic of the disease.² The face and eyelids are the most commonly involved areas. Early diagnosis and treatment is very important, especially if the eyelid is affected. The diagnosis is confirmed by bacteriological examination of the lesion or isolation of *B. anthracis* from the infected area or blood culture. Inadequately treated eyelid anthrax may cause severe deformities of eyelids including cicatrization and ectropion. High dose parenteral penicillin G is still the recommended first choice treatment.³ We herein present a patient with eyelid anthrax who is diagnosed early, treated properly and resolved without any evelid complications.

CASE REPORT

A 38-year-old male farmer was referred to our clinic with a diagnosis of preseptal cellulitis. He was complaining of pain, itching sensation and swelling of his right upper and lower eyelids which started 4 days ago and a gradual increase of symptoms was reported. There was no prior history of trauma. The patient described a small erythematous papule on the eyelid which then became vesicular and painful, and associated with severe pruritus. There was a black eschar and periorbital edema leading to closure of the right eyelid (Figure 1). The patient's visual acuity was 20/20 and ocular examination was otherwise unremarkable. Microscopic examination of the scraped material obtained beneath the edge of the eschar revealed gram positive rods. Tissue and blood samples were taken for culture. There was no bacterial growth in blood culture of the patient. Routine urinalysis, complete blood count, serum electrolytes, renal and liver function tests showed no abnormalities. No other abnormality was determined by imaging techniques including computerized tomography and ultrasonography. Treatment with systemic penicillin G, 16 million units intravenously per day was initiated and was continued for a period of 10 days. The eyelids began to heal after the fourth day



FIGURE 1: A black eschar and periorbital edema of the right eye. (See for colored form http://tipbilimleri.turkiyeklinikleri.com/)



FIGURE 2: The eyelid began to heal after the fourth day of treatment. (See for colored form http://tipbilimleri.turkiyeklinikleri.com/)

of the treatment (Figure 2). The lesion healed within 10 days leaving no clinical evidence of any eyelid abnormality (Figure 3). No eyelid deformity was developed during one-year follow-up period. Despite the negative culture results for *B. anthracis*, characteristic clinical appearance of the lesion, iso-



FIGURE 3: The lesion healed within 10 days, leaving no clinical evidence of eyelid deformity.

(See for colored form http://tipbilimleri.turkiyeklinikleri.com/)

lation of Gram-positive bacilli from the edge of the eschar, and significant improvement of the lesion after systemic penicillin G treatment helped us to diagnose eyelid anthrax.

Informed consent was obtained from the patient himself for publication of his findings as a case report.

DISCUSSION

Anthrax is an infectious disease caused by Bacillus anthracis. Cutaneous anthrax (CA) is the most common form of the disease and accounts for 95% of cases. CA usually develops on exposed sites such as the hands and face, but eyelid involvement is less commonly observed when compared with the other forms. Periorbital anthrax is primarily an occupational hazard of workers who process animal and agricultural products and should be considered in the differential diagnosis of orbital and eyelid infections, especially in this particular population.^{1,2} Periorbital anthrax primarily involves the upper eyelid. It spreads toward the lower eyelid and the cheek. The first sign of eyelid anthrax is severe and generalized edema. Afterwards, edema begins to reduce in the characteristic necrotic tissue areas, and pustules develop on the lesion. This structure is known as the eschar of anthrax. The granulation tissue, which heals by forming a scar tissue, develops on the eschar within 2 to 4 weeks. Scarring and contraction caused by the lesion on the eyelid can lead to cicatricial ectropion.³ Anthrax is now very rare in Northern America and many countries in Europe, but is a significant problem in Western Africa, Spain, Greece, Turkey, Albania, Romania and Central Asia. Anthrax is still common among domestic farm animals and cutaneous anthrax is an endemic disease among people in the Eastern part of Turkey where traditional animal husbandry is the primary occupation.⁴

Early diagnosis and treatment of anthrax is very important. Fatality may be as many as 10 to 20% of untreated cases, but the death rate can be reduced to less than 1% with appropriate antibiotic therapy. Eyelid anthrax is painless, does not include rash and results in a black eschar. The differential diagnosis of anthrax includes brown recluse spider bite, ecthyma, accidental vaccinia, ulceroglandular tularemia and necrotic herpes simplex. In palpebral lesions, orbital abscess and preceptal cellulitis caused by gram positive cocci such as *Staphylococcus aureus, Staphylocccus epidermidis* and *Streptococcus pneumoniae* should be excluded.⁴

The diagnosis is confirmed by bacteriological examination of the lesion or isolation of Bacillus anthracis from the infected ulcer or from the blood culture. However, the positive culture results do not exceed 65%. A few hours of time after the first dose of antibiotic is sufficient for tissue and blood culture results to become negative. In these cases, the diagnosis is made with the clinical appearance of the lesions, the patient's history, and detection of gram-positive bacilli with microbiological evaluation. There are no known gram positive bacilli other than *B. anthracis* that can cause a similar typical lesion on the eyelids.⁵ In a previous study, Caca et al. similarly made diagnosis of anthrax based on the patient's history, clinical findings, and a Gram staining showing Gram-positive bacilli from the lesion in a patient whose cultures gave no growth.³ Our patient presented with an eyelid abscess and had been previously treated with oral sulbactam + ampicilline antibiotics for two days prior to admis-

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sion to our hospital. Gram staining of the smear showed gram positive bacilli, but the culture was negative. However the clinical features and the patient history was compatible with anthrax. The cause of negative culture results in our patient may be previously given antibiotic treatment.

Penicillin G is still the drug of choice in the therapy of naturally occurring anthrax in many parts of the world. High doses of parenteral penicillin G is the first choice for the treatment of anthrax. General improvement and decreased edema within 2 to 4 days is the most important indicator of the effectiveness of the treatment. Topical treatment is not effective, and the usefulness of corticosteroids is debatable.3 In a series consisting of three patients with eyelid anthrax, all patients responded well to the intravenous administration of penicilin G, and the lesion resolved, leaving scars in two cases.¹ Daboue et al. reported three children with palpebral anthrax; all responded to penicillin G and no complications were noted.⁶ Tetracycline, erythromycin, chloramphenicol, and first-generation cephalosporins can be used in cases of penicillin allergy.⁷ Our patient was treated with penicillin G (16 million units/day) for 10 days.

Another important aspect of cutaneous anthrax is that although antibiotics improve systemic symptoms, they fail to stop the progression of eschar and evelid cicatrization. The main complication of periocular anthrax is cicatricial ectropion. Yorston et al. reported 11 patients with major complications; cicatricial ectropion in 8 cases and corneal scarring in 3 cases.8 In our case, we establised the diagnosis of anthrax based on the patient's clinical presentation, lesion characteristics, and a Gram staining showing Gram-positive bacilli from the lesion, despite negative culture results. We speculated that lack of bacterial growth in cultures of patients may be secondary to previously given antibiotics before their hospital admission. In addition, prevention of eyelid complications may be due to high dose penicillin G treatment.

In conclusion, cutaneous anthrax is still a public health problem in Turkey. Physicians should recognize the signs and symptoms of anthrax, and a possible contact with an infected animal should be considered in the differential diagnosis of preseptal and orbital cellulitis. Early diagnosis and high dose antibiotic treatment can facilitate the treatment and prevents development of eyelid complications including cicatrization and ectropion.

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