Abstract

**Purpose:** Cutaneous anthrax of the eyelid is a rare condition in clinical practice, but should be considered in the differential diagnosis of preseptal and orbital cellulitis. Here we report a case of palpebral anthrax.

**Case report:** A 5-year-old girl with anthrax infection of the right upper eyelid is presented. Her eyelid showed an edematous, necrotic, and partially bullous lesion. Following complete healing of the eyelid lesion, mild cicatricial ectropion resulted.

**Conclusion:** Anthrax is a rare disease that is not well known by ophthalmologists. Clinicians should be aware of the complications and the management of palpebral anthrax.

**Keywords:** Anthrax, Ectropion, Eyelid

Introduction

Anthrax is an infectious disease of herbivores caused by Bacillus anthracis. Humans become infected when they come into contact with infected animals such as sheep, goat, cattle, and horse or contaminated animal products. There are three main forms of anthrax: inhalation, cutaneous, and oropharyngeal/gastrointestinal.

Cutaneous anthrax most commonly affects the exposed areas of the upper extremities and, to a lesser extent, the head and neck. Cutaneous anthrax accounts for the majority of cases. Cutaneous anthrax develops 2-5 days after skin exposure and penetration of Bacillus anthracis spores. The first sign of cutaneous anthrax is severe and massive edema round the site of infection. The infection begins as a pruritic small papule that enlarges within 24-48 hours to form an ulcer surrounded by vesicles. As the edema subsides, a black, necrotic, central scar then forms, which is characteristic of the anthrax. Two to four weeks later the scar sloughs away, and the granulation tissue heals with scarring.

Palpebral anthrax typically starts at the upper eyelid, and spreads down to the lower eyelid and the cheek. Edema may occur and spread towards the head and neck.
The edema is nonpitting and can spread throughout the body from the lesion. If the edema spreads towards the neck, shortness of breath due to airway obstruction may become evident. Palpebral anthrax can lead to serious complications like cicatrization and ectropion. Longstanding corneal exposure due to ectropion may lead to blinding corneal scar formation.

In this report, we present a patient who developed palpebral anthrax. In this case, a black necrotic scar seen on the right upper eyelid. Irrespective of antibiotic treatment, the patient developed ectropion.

**Case report**

A 5-year-old girl presented with a lesion on the right upper eyelid that developed 3 days before admission to our hospital. On ocular examination her right upper eyelid showed an edematous, necrotic, and partially bullous lesion (Figure 1). The patient was found to have diffuse edema of the right eyelids with inability to open the eye. The right upper eyelid showed a black scar with a plaque. The patient’s father said that the scar had developed 3 days ago and the edema had occurred 2 days ago. Her visual acuity (VA) could not be measured. The anterior segment of the eye was evaluated as normal when the eyelids were opened with an instrument during the ocular examination. There was also a diffuse facial edema that had spread to her neck and upper part of trunk (Figure 1). Her body temperature was 38.5°C. Her family had been living in a village for a long time and kept livestock. She had a history of fall one week ago.

The patient was hospitalized with a diagnosis of eyelid anthrax and intravenous penicillin G (500,000 units/kg/day) was given for 2 weeks. Microscopic examination of the material scraped from beneath the edge of the scar revealed Gram-positive rods. But no microorganism was isolated from the culture. The remaining physical findings and laboratory tests were normal. During the treatment, her facial and trunk edema gradually resolved. In the second week, the edema disappeared completely but the scar was still present (Figure 2).

**Figure 1.** Extensive edema and hyperaemia are present on the head and upper trunk at the first examination.

**Figure 2.** Black necrotic tissue is present in the upper eyelid 2 weeks after presentation.
The patient was discharged, but was asked to return for a follow-up examination after 2 weeks. In the control examination, the scar had fallen off spontaneously, and cicatrisation, and slight ectropion were found in the right upper eyelid (Figure 3). Her father said that the scar had fallen spontaneously two days ago. The patient has been followed for oculoplastic surgery.

**Figure 3.** One month after presentation cicatrisation, lagophthalmos, and slight ectropion are present in the upper eyelid.

**Discussion**

*Bacillus anthracis* is a large, nonmotile, aerobic, spore forming gram-positive bacillus. The spores of *Bacillus anthracis* can remain viable in soil but infective for many years. Cutaneous anthrax accounts for 95% of all anthrax infections. More than 90% of lesions occur in exposed areas, such as the face, neck, arms, or hands.

The eyelid involvement of cutaneous anthrax is rarely seen. After infection via an abrasion, cut, or possible insect bite, a small pimple will develope on the eyelid within 2 to 3 days. After then ring of vesicles develop surround the initial lesion. During the onset period of the disease, the differential diagnosis was difficult because of uncharacteristic symptoms, and negative bacteriological tests due to previous antibiotic treatment. Edema of the involved eyelid regresses, and a black, necrotic scar, which is characteristic of anthrax is formed. This lesion heals and scar falls off within 2 to 4 weeks, commonly leaving sequella. The lymph nodes draining the infected area may be swollen and tender. Although the anthrax ulcer is not painful, adenopathy associated with cutaneous anthrax may be painful. Lymphadenopathy may persist long after disappearance of the scar. The contracture occurs due to cicatrisation, may lead to ectropion. It appears that upper eyelid involvement is more likely to cause ectropion. Succesful medical treatment does not prevent the cicatrisation related to the severity of the lesion. Despite treatment with antibiotic, mild ectropion developed in our case.

Yorston and Foster reported 11 patients with palpebral anthrax, 8 patients had cicatricial ectropion and 3 had corneal scarring as complications. Corneal complications occur due to exposure keratopathy. Our case developed ectropion on the upper eyelid with insignificant exposure.

In the literature, the complications of palpebral anthrax such as exophthalmos, optic atrophy and panophthalmitis have been reported. Palpebral anthrax must be considered in the differential diagnosis of preseptal or orbital cellulitis, dental abscess, and erysipelas.

**Cutaneous** anthrax is mainly diagnosed by the history and rapid development of a painless ulcer surrounded by a zone of edema. The diagnosis is confirmed by bacteriological examination of the lesion or isolation of *Bacillus anthracis* from the lesion or blood by culture. Confirmation of the diagnosis with isolation of bacillus in the culture could not be achieved in all cases. In our case, the culture was negative. The clinical diagnosis was supported with the microscopic examination of scraped material showing gram positive rods.

Early treatment of anthrax is very important. Untreated cases of cutaneous anthrax may be fatal. But the mortality rate can be reduced to less than 1% with proper antibiotic therapy. Penicillin is still the first choice for the antimicrobial treatment of anthrax. However, penicillin resistance has been found rarely in naturally occurring strains. *Bacillus anthracis* is also susceptible to fluoroquinolones, tetracycline, chloramphenicol, aminoglycosides, macrolides, imipenem, rifampicin, and vancomycin.
Reduction in the edema within a few days is the indicator of the effectiveness of the treatment. Effective treatment should be continued to prevent septicemia and meningitis. The mortality rate was 20% in untreated cases due to septicemia and meningitis. Malignant edema may also cause death by obstruction of airway.

Even if the patient is promptly treated with antibiotics, cutaneous lesions progress through scarring. The surgical intervention should be avoided because it may cause spread of the infection. On the other hand, diagnostic surgical biopsy may be used, and surgical treatment may be needed for secondary infections. Surgical interventions for ectropion should be attempted at least 6 months following the arrest of all healing processes.

Although the usefulness of corticosteroids is debatable, they may be necessary to treat malignant edema or may be used as adjuvant treatment for head and neck lesions and for systemic or central nervous system involvement.

**Conclusion**

Cutaneous anthrax on eyelid is rarely seen in clinical practice, but should be considered in the differential diagnosis of preseptal and orbital cellulitis. The patients present with a painless necrotizing ulcer or black scar with surrounding eyelid edema. Cutaneous anthrax on eyelids can lead to cicatriziation and ectropion. Physicians should be aware of the signs and symptoms of anthrax, the importance of detailed medical history and animal contact.

**References**