Anthrax: A Rare Cause of Periorbital Cellulitis and Cicatricial Ectropion

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Abstract

Purpose: Among the three forms of anthrax in human (cutaneous, inhalational, and gastrointestinal), cutaneous anthrax is the most common form with prevalence of 95%. The cutaneous anthrax is a rare cause of periorbital (preseptal) cellulitis that may remained without correct diagnosis and appropriate treatment. This may result in severe local complications such as blindness and airway obstruction or dissemination of the disease like as bacteremia and meningitis.

Objective: We report a child with periorbital anthrax to present the clinical manifestation and progression of the disease and remind physicians the interesting contagious cause of periorbital cellulitis.

Case report: A 2.5-year-old boy from a rural area of Guilan province in Iran was seen to have a small erythematous papule in his right lower eyelid 5 days prior to admission. Then, the lesion progressed to a necrotic ulcer with a black central scar and severe surrounding edema and erythema. Severe swelling of the eyelids and edema all over the face occurred as well. Fever was absent. The child had repetitious contacts with domestic herbivores. The CT scan of the orbit and paranasal sinuses showed soft tissue swelling only. Gram-positive rods were seen in the smear of exudate taken from cutaneous lesion but culture result showed negative. Treatment with intravenous ciprofloxacin, penicillin, and clindamycin resulted in clinical improvement but due to cicatricial ectropion formation, oculoplastic intervention was considered to be done.

Conclusion: Physicians must consider a cutaneous lesion to be anthrax if any of the following exists: a history of contact with domestic herbivores and animals, similar lesions in the family members, cutaneous necrotic ulcer with the characteristic black scar and peripheral edema, and observing gram-positive rods in scraped material taken from beneath the scar edge or exudate of cutaneous lesions. Although anthrax of the eyelids is not common, it must be included in the differential diagnosis of periorbital cellulitis.

Keywords: Anthrax, Ectropion, Eyelid, Periorbital Cellulitis


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Introduction

The term "anthrax" is derived from the Greek root "anthrakos" (coal) which refers to the black scar characteristic of cutaneous anthrax. Anthrax is a disease of animals, particularly domestic herbivores, while human is an incidental host. The disease is caused by Bacillus anthracis, a gram-positive, aerobic, non-motile, and spore-forming rod of the Enterobacteriaceae family. Among its clinical forms in human beings (cutaneous, inhalational, and gastrointestinal), cutaneous anthrax is most prevalent (95%) which is mostly observed in the exposed area of the body of livestock breeders or those who are in contact with contaminated animal products such as wool or pelts. Human anthrax is most common in developing countries, especially in regions where the animal-soil-animal cycle of anthrax exists. Heyworth et al. reported that among 431 patients with anthrax, 11% were 2 years old children or younger and 42% were between 3 to 15 years of age. Most lesions in the patients at any age were on the head and neck, few on the trunk, and least on the extremities. Endospores of B. anthracis usually are inoculated through abraded or injured skin. However, anthrax of the eyelids (peri orbital anthrax) is not common, but without correct diagnosis and appropriate treatment, it may result in severe local complications such as blindness and airway obstruction, or dissemination of the disease like as bacteremia and meningitis, or even death. We report a child with periorbital anthrax to present clinical manifestation and progression of the disease and remind physicians an interesting contagious cause of periorbital cellulitis.

Case Report

The patient was a 2.5-year-old boy living in one of the rural areas of Gilan province in north of Iran. Five days before his admittance to 17-Shahrivar Pediatric Hospital in Rasht city, a small papule appeared in his right lower eyelid that become ulcerative and developed into a necrotizing ulcer with a central black necrotic scar and severe surrounding edema and erythema. Obvious swelling of the right side of the face occurred as well. After being admitted to two other hospitals and receiving few doses of vancomycin, penicillin, and clindamycin, the patient was referred to our hospital. He had repeated contacts with domestic herbivores (cows and sheep), yet no previous trauma or other problems were noticed in his history and there was no similar disease in his family members. Moreover, the patient had an appropriate growth rate. He had no fever at the time and during admission. On physical examination, a necrotic ulcer with a central black necrotic scar in his right lower eyelid was observed that accompanied by sero-purulent exudate from the infected eye along with obvious erythema and edema around it and swelling of the right side of the face. It was not possible to open the eyelids for examining the eyeball and cornea due to severe swelling and chemosis (Figure 1).

Figure 1. A necrotic ulcer with a central black scar in the patient's right lower eyelid accompanied by severe surrounding edema and erythema, and swelling of the right side of the face were present at the time of admission.

On the left side of the face, a slighter swelling along with periorbital edema was noted. However, other physical examinations were normal. The white blood cell count was 25,700 (52% neutrophil), and the erythrocyte sedimentation rate (ESR) was 30 mm/h. CSF analysis was normal and there were no other pathological findings. CT scanning of the orbit and paranasal sinuses showed severe swelling of the subcutaneous tissue and muscles of the right temporal, frontal, and preseptal regions and right side of the face,
but the orbit and paranasal sinuses were all normal and only the right maxillary sinus was slightly smaller with thicker mucosa. There was no lesion in the sphenoid sinus, pharynx, and nasal cavities. Microscopic examination of exudate from cutaneous lesion revealed few gram-positive rods. Unfortunately, no bacterial isolation from the lesion and blood culture was seen. The patient was treated as cutaneous anthrax of lower eyelid with intravenous ciprofloxacin, penicillin, clindamycin, and dexamethasone. The ulcer and eyelids were irrigated with sterile normal saline two times a day. One week later, when swelling of the eyelids was reduced, an ophthalmologist examined the patient and observed a clear cornea with red reflex. After three weeks of intravenous antibiotic therapy, swelling of the face had subsided completely, and the lesion remained black and scabby, with some peripheral edema and sharp edges and a size of 2 cm (Figure 2).

The patient was discharged with oral ciprofloxacin and amoxicillin and was asked to return for follow-up examination after a week. The treatment continued for two months. When he was brought to the hospital after 2 months, the scab had fallen off spontaneously, and cicatricial ectropion with restriction of lower eyelid movement were found (Figure 3). The patient were followed for oculoplastic intervention.

**Discussion**

*B. anthracis* can introduce through abraded or injured skin of the eyelids or periorbital area. Rubbing the eyelids with contaminated fingers or possibly an insect vector may lead to cutaneous involvement of the eyelids. Typical periorbital anthrax starts at the upper eyelid; edema spreads down to the lower lid and cheek. Of course, there have been cases in which primary infection of the lower eyelid was observed and our patient was one of such cases. After an incubation period of 2 to 5 days, a small and nontender but frequently pruritic papule develops at the site of inoculation of cutaneous anthrax. The lesion progresses to a serous or serosanguineous vesicle with surrounding severe nonpitting edema within 36 hours. Occasionally, satellite vesicles (pearly wreaths) may be seen. The lesion undergoes central necrosis with a black scar that is a characteristic of cutaneous anthrax. Sometimes, severe edema in head and neck might result in obstruction of the airway for which tracheostomy would be needed. Slight fever and fatigue often exist, yet high fever and severe leukocytosis (20,000-30,000/mm$^3$) are reported in younger children or patients with systemic infections such as bacteremia or meningitis. Definite diagnosis needs to demonstrate *B. anthracis* by direct smear and culture form vesicular fluid or exudate from cutaneous lesions or scraped materials from the area under the scar edge and from pleural fluid, blood, and CSF in systemic infections. Although based on the patient's history regarding his contact with livestock or their products and clinical manifestations and progression of the
disease, physicians can often start the treatment of anthrax even in the absence of positive smear and culture results. Effective and immediate treatment is necessary to prevent the progression and dissemination of the disease. Historically, penicillin has been the treatment of choice for cutaneous anthrax. However, naturally and acquired resistant strains of B. anthracis have been developed. Empirical therapy should be initiated for systemic infections (particularly inhalational anthrax) or cutaneous lesions which are accompanied by fever, toxicity, or severe edema; or are located on the head and neck. In these conditions, ciprofloxacin 10-15 mg/kg every 12 hours (not to exceed 1 g/day) is administered intravenously. Moreover, one or two additional antimicrobials should be administered intravenously until the result of susceptibility testing is available. In order to inhibit protein (toxin) synthesis, it is recommended to add clindamycin to the patient's therapeutic regimen. Local treatment is not much effective but systemic corticosteroid has been recommended in cases of severe edema or meningitis. Debridment or incision of the cutaneous lesion should be prevented because it increases the possibility of bacteremia.

Usually, about 20% of patient with cutaneous anthrax who left untreated will die, but the death occur only in 1% of cases who received on-time and effective treatment. Cutaneous anthrax of the eyelid may recover without any complication or at times it might leave a slight cicatrix or result in cicatricial ectropion, corneal scar or even blindness. Faghihi et al reported a patient with cutaneous anthrax of face and permanent paralysis of the facial nerve. Although, our patient needed an oculoplastic surgery due to cicatricial ectropion, fortunately no corneal scar or visual complications were observed in him.

Conclusion

This case report and other new reports of cutaneous anthrax of the periorbital region and eyelids indicate that despite its low prevalence, anthrax should still be considered in the differential diagnosis of periorbital cellulitis. The physicians must consider a cutaneous lesion to be anthrax if any of the following exists: a history of contact with domestic herbivores and animals, similar lesions in the family members, cutaneous necrotic ulcer with the characteristic black scar and edema of the area around, and observing gram-positive rods in scraped material taken from beneath the scar edge or exudate of cutaneous lesions. Regarding difficult diagnosis of periorbital anthrax in early stages, appropriate treatment should be started soon; otherwise the possibility of local complications or dissemination of the disease will increase.

References