Orbito-Maxillofacial Cutaneous Anthrax

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Abstract

Background: Cutaneous anthrax is a zoonotic disease that spreads to humans primarily through exposure to infected animals, animal products or spore infested soil. Microbiological identification of cutaneous anthrax may be obscured by previous antibiotic use. Thorough history and clinical examination is therefore necessary in making a diagnosis. There is a paucity of reports about these infections in our region. **Methods**: Five cases are presented with a history of contact with animal or animal products and the typical appearance of skin lesions. Two cases were confirmed through microbiologic gram staining. All cases were treated with antibiotics and resolution of the initial cutaneous lesions was noted within three weeks. **Conclusion**: Thorough clinical history, examination and a high index of suspicion are paramount in making a diagnosis. Management with a combination of antibiotics ensures clinical resolution.

Keywords: Anthrax, Orbital-Maxillofacial, Cutaneous

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Background

Anthrax is a zoonotic disease of antiquity mentioned in the bible, and by Homer, Virgil and Hippocrates (1). It is caused by *Bacillus anthracis*, a gram positive, aerobic, non-motile rod, first described by Robert Koch in 1876 (2). In the developed world, the incidence of anthrax has diminished but it still remains of important public health concern in underdeveloped nations where animal husbandry remains a mainstay and veterinary vaccinations have limited penetration (3-5). Human infections occur primarily through direct or indirect contact with infected animals or their products such as hides, wool and inhalation of spores. This makes it a common occupational disease (5,6). Anthrax spores are hardy and can remain dormant in soil for decades even under adverse conditions (5,7). This unique characteristic in addition to high potency and ease of delivery has resulted in its use as a biologic weapon (8). There are three main types of anthrax namely: cutaneous, pulmonary and gastrointestinal. Cutaneous anthrax accounts for 95% of all known cases of anthrax and carries the lowest mortality rate (4). Its hallmark is a painless black eschar that occurs on exposed areas such as face, neck and hands (1,6,9). Pulmonary and gastrointestinal variants are lethal due to delayed diagnosis as well as systemic involvement with release of lethal toxins (3).
Awareness of orbito-facial anthrax in this country as well as the region remains low due to the paucity of cases reported in literature. This paper presents five patients with orbito-facial cutaneous anthrax with emphasis on the associated history, physical and laboratory examination as well as management and outcome.

Cases

All cases presented at the ophthalmology and maxillofacial surgical clinics at a county referral hospital between January 2012 and December 2013 with painless orbito-facial brawny swelling and development of a black eschar were reviewed. Occupational history, falls and/or contact with animal meat was enquired into. Microbiology, culture and sensitivity testing of the tissue fluid was conducted. Informed consents were obtained from the patients for the publication of the case series.

Case 1

A 45 year old male presented with a history of fall from a motorcycle and bruised his left cheek. Two weeks later he presented with a three day old swelling of the left side of the face and extending to the neck (Figure 1A). Bilateral mid facial non-pitting edema and mild clear discharge from the

left lower lid was noticeable from the second day onwards. Culture of fluid isolated Bacillus anthracis highly sensitive to ceftazidime, chloramphenicol, and ciprofloxacin. He was put on intravenous chloramphenicol 1gram (g) four times daily for five days and later oral ciprofloxacin 500 milligrams (mg) twice daily for one week. An eschar developed after 5 days (Figure 1B). He was later discharged on the same dosage of ciprofloxacin for two weeks. On review one week post discharge, an ectropion of the left lower eyelid was developing. He was finally lost to follow-up. It is thought that anthrax spores were in the ground and were inoculated during the fall.

Figure 1A: Initial presentation

Figure 1B: Eschar forming lesion-left eyelid

Case 2

A 45 year old lady who presented with a four day history of left sided facial swelling associated with ulceration of the upper and lower eyelids. She reported that while undertaking her daily farming activities she experienced itchiness of her left facial region followed by rapid swelling within a day. On admission, there was gross left facial non pitting edema with associated discharge from the eyelids. Tissue fluid was taken for culture and patient put on intravenous chloramphenicol (1gm QID for 5 days) and oral ciprofloxacin (500mg BD for 21 days). The culture results isolated *Bacillus anthracis* highly sensitive to doxycycline and ciprofloxacin. A black eschar developed by day 9 with progressive reduction in facial edema (Figure 2). Patient was discharged after 3 weeks after noted marked improvement. It is presumed that while tilling her farm she transferred the spores from the ground to her face while trying to relieve her itch.

Figure 2: Upper and lower eyelid eschar

Case 3

A 36 year old lady presented with left sided facial swelling (Figure 3A) and a history of having slaughtered a cow together with her husband at home. The swelling had been there for 3 days. The husband also presented with an eschar on the right hand at the surgical unit and was managed as an outpatient. There was non-pitting (woody) edema, and discharge from the lower lid. Based on the history and physical evaluation, a diagnosis of anthrax was made. Tissue discharge was taken for culture, and patient put on chloramphenicol and ciprofloxacin. An eschar formed by day 4 (figure 3B) with progressive Improvement noted from day 9. Cultures done from the lesion were negative for *B. anthracis* but treatment was continued based on the clinical diagnosis. She was discharged on day 16 on oral medication. She came for review on day 21 with noted marked improvement but has subsequently been lost to follow-up.

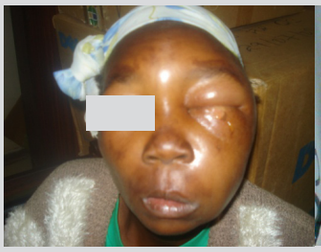


Figure 3A: Initial presentation

Figure 3B: Eschar forming on the left lower eyelid.

Case 4

A 30 year old butcher presented with a week's history of swelling of the lower right lid and discharge (Figure 4A). He was admitted and put on chloramphenicol (5 days) and ciprofloxacin (21 days) while tissue fluid specimen was taken for culture. He developed an eschar two days after admission (Figure 4B). The subsequent results were negative for anthrax. Treatment did not change though, and improvement was noted by day two. By day 5, he had an ectropion, and tetracycline ointment was prescribed to prevent exposure keratopathy. He was discharged on day 13, came for review on day 19 with noted resolution of the disease process but with associated lower lid ectropion and was lost to follow-up subsequently.

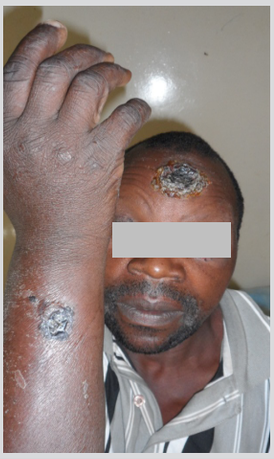
Figure 4A: Patient at initial presentation

Figure 4B: Eschar forming lesion.

Case 5

A 47 year old man reported being bitten by a wasp on the forehead and right forearm five days before admission. He started swelling two days after the bite. He further admitted that a week previously he had slaughtered a sheep at home. Nobody else presented with any problem among the others who handled the meat. His lesions had crusting and serous discharge, with surrounding woody edema (Figure 5). A specimen for culture was taken and no growth was obtained, though microscopy showed endothrix microspores and yeast forms. He was put on chloramphenicol (for 5 days) and ciprofloxacin (for 21 days) on admission, and improvement was noted from day two. He was discharged on day 6 and came for review after 2 weeks where the lesions were noted to be resolving.

Figure 5: Multiple eschar forming lesions - forehead and right forearm.



Discussion

Orbito-facial cutaneous anthrax though rare should be considered a differential diagnosis especially in developing countries where animal husbandry still remains a mainstay economic activity (10). Due to its innocuous development, it is particularly prudent that first contact clinicians be aware of this diagnosis. As noted, some of these patients may present to dentists, clinical officers or nurses in peripheral clinics whom must have a high index of suspicion (10). Cutaneous anthrax carries the lowest mortality rate of the three main forms especially when timely diagnosed and treated (11).

The clinical evolution of cutaneous anthrax is typical with the initial development of minute red macules or papules at the site of inoculation (subcutaneously inoculated anthrax spores). There is subsequent development of gross facial edema (malignant edema) that is typically brawny in nature with minimal fluid or purulent aspirate. Within 48 hours the vesicles rupture to form a moist ulcer that undergoes central coagulation necrosis leading to the development of a black eschar that is characteristic of anthrax (5,10). Orbito-facial anthrax must be approached with caution due to the possibility of involvement of the upper airway leading to asphyxiation.

Diagnosis of cutaneous anthrax involves clinical history and physical evaluation, gram staining and wound and blood culture (11). Dependent on history of antibiotic use as well as hospital facilities available, clinical findings maybe the only way of cutaneous anthrax diagnosis 9,10,12). Karahocagil et al. concluded that though gram staining and simple culture methods before antibiotic therapy may be useful in aiding diagnoses, majority of diagnoses would probably be made on clinical grounds alone (9). Furthermore, from literature, only 60-65% of skin lesion cultures are positive for anthrax (9,13). A case series report by Gelaw et al diagnosed and managed successfully all their cases on clinical grounds (10). As concerns our cases, two were diagnosed on clinical as well as microbiological identification of Bacillus anthracis while the three other cases were diagnosed on clinical grounds as bacterial cultures were negative possibly due to the laboratory procedures or prior antibiotic use. Literature notes that antibiotic therapy renders lesions culture-negative within a few hours (12,13). In the present case series, one patient presented with a positive history of antibiotic use prior to culture of specimens. As such education on the clinical history and evolution of this disease remains paramount.

As concerns therapeutics, ciprofloxacin, doxycline and penicillin are currently recommended for the treatment of cutaneous anthrax (1). The regime used in our case series involved combination therapy of short course intravenous chloramphenicol (5days) and prolonged course of oral ciprofloxacin (21days) as instructed by sensitivity testing (Case 1) and currently prescribed treatment protocols in literature. The use of chloramphenicol was initially instructed by availability viz-a-viz other intravenous antibiotics and latter on by sensitivity testing. All cases responded to treatment with resolution of the initial disease process. However in our case series, surgical management of the sequelae (ectropion) was not possible due to loss to follow-up of the patients.

It is important to note that human to human transfer of anthrax is rare (13). Typical precautions taken during treatment of any infectious disease is adequate when

in contact with anthrax infected patients. Of note is the management of any contaminated personal items such as clothes worn by the patient that may carry the spores (5). Chemoprophylaxis with antibiotics should be instituted only if exposure is confirmed.

Conclusion

Health practitioners should develop a high index of suspicion for anthrax when they encounter orbito-facial lesions with characteristic ulceration and black eschar formation. Occupational history related to animal husbandry should trigger suspicion for anthrax. Management with combination antibiotics will ensure clinical resolution but clinical sequelae like ectropion will need further management.

References

1. Inglesby TV, O'Toole T, Henderson DA, et al. Anthrax as a Biological Weapon: Updated Recommendations for Management. JAMA. 2002;287:2236-52.
2. Brooks GF, Karen CC, Janet SB, et al. Mietzner, Jawetz, Melnick and Adelberg's Medical Microbiology 24th edition. McGraw-Hill. 2007; Chapter 5, 203-212.
3. Holty JE, Bravata DM, Liu H, et al. Systematic Review: A Century of Inhalational Anthrax Cases from 1990 to 2005. Ann Intern Med. 2006;144(4):270-80.
4. Lucey D. Bacillus Anthracis (Anthrax). In Principles and Practice of Infectious Diseases. 6th ed. Churchill Livingstone. 2005.2485-91.
5. Siddiqui MA, Khan AH, Ahmed SS, et al. Recent Outbreak of Cutaneous Anthrax in Bangladesh: Clinic-Demographic Profile and Treatment Outcome of Cases Attended at Rajshahi Medical College Hospital. BMC Research Notes.2012; 5:464.
6. Sternbach G. The History of Anthrax. J Emerg Med. 2002; 24(4):463-7.
7. Richard A, Pherson MC, Mathew R. Henry's Clinical Diagnosis and Management by Laboratory Methods. 21st edition. Saunders;2007;133-6.
8. Centers for Disease Control Update. Investigation of Bioterrorism-Related Anthrax and Interim Guidelines for Exposure Management and Antimicrobial Therapy. MMWR Morb Mortal Wkly Rep. 2001; 50:909-19.
9. Karahocagil MK, Akdeniz N, Akdeniz H, et al. Cutaneous Anthrax in Eastern Turkey: A Review of 85 Cases. Clin Exp Dermat. 2008; 33:406-11.
10. Gelaw Y, Asaminew T. Periocular Cutaneous Anthrax in Jimma Zone, Southwest Ethiopia: A Case Series. BMC Research Notes. 2013; 6:313.
11. Kasper D, Braunwald E, Fauci A et al. Harrison's Principle of Internal Medicine. 16 ed. McGraw-Hill; 2005;1280-1.
12. Oncul O, Ozsoy MF, Gul HC, et al. Cutaneous Anthrax in Turkey: A Review of 32 Cases. Scand J Infect Dis. 2002; 34:414-16.
13. DemirdagK, Ozden M, Saral Y, et al. Cutaneous Anthrax in Adults: A Review of 25 Cases in Eastern Anatolian Region of Turkey. Infection. 2003;31:327-30.