

CASE REPORT

Suspected cutaneous anthrax in a yak herder in a highland community in Bhutan, 2023: A case report

Ugyen Chopel¹  | Sangay Dorji¹ | Kuenga Sonam²  | Thinley Dorji^{2,3} 

¹Wangdue Hospital, Wangdue Phodrang, Bhutan

²Department of Internal Medicine, Central Regional Referral Hospital, Gelephu, Bhutan

³Faculty of Postgraduate Medicine, Khesar Gyalpo University of Medical Sciences of Bhutan, Thimphu, Bhutan

Correspondence

Ugyen Chopel, Wangdue Hospital, Wangdue Phodrang, Bhutan.
Email: uchopel.96@gmail.com

Key clinical message

This case presents cutaneous anthrax in yak herder from a central highland community in Bhutan. We highlight the clinical presentation, diagnosis and management of the case in a resource-limited setting, and the public health response through the One Health approach.

KEYWORDS

Bacillus anthracis, bacterial zoonoses, disease outbreaks One Health, yak, zoonosis

1 | INTRODUCTION

Anthrax is an ancient zoonosis caused by a soil-borne spore-forming *Bacillus anthracis*, which primarily infects herbivore animals and is lethal in both animals and humans.^{1,2} Humans are secondarily infected through handling of infected carcasses, ingestion of infected meat, and inhalation of anthrax spores from contaminated animal products. Direct human-to-human infection is unknown.³ According to the World Health Organization, globally there has been a decrease in the incidence of human anthrax has decreased from up to 100,000 cases per year in 1958 to 2000 per year in the 1980s.⁴ However, anthrax continues to occur as outbreaks in animals and humans across many countries. In South Asia, it is estimated that 61.6 million cattle, 1.8 million pigs, 72.5 million goats, 18.7 million sheep, 0.33 million buffalo, and 26.6 million humans are at risk of anthrax.⁵

Given that the majority of cases occur in rural areas among the farming population, it is likely that many outbreaks remain unreported. Delays in reporting of human cases, delays in sample collection and administration of antibiotics are some of the factors that do not yield

laboratory confirmation of anthrax as shown in recent experiences from India.⁶ Laboratory confirmation of anthrax is important not only for public health surveillance but has clinical implications in the selection of antibiotic agents and drug dosing.⁷

Based on the type of exposure, disease in humans occurs primarily in three forms. Cutaneous anthrax accounts for 95% of reported cases in the world, followed by respiratory anthrax and gastrointestinal anthrax cases.⁶ Fatal meningial anthrax is rare, but it accounts for 4% which is secondary to other forms of anthrax infection. Diagnosis of the disease is based on a positive history of exposure to the source of infection, patient occupation, clinical syndrome, and laboratory confirmation. The organism grows readily on conventional microbiology media, including blood agar if the specimen has been obtained before the administration of antibiotics.⁸ Other diagnostic modalities include polymerase chain reaction on specimens and serological markers. However, even in the absence of laboratory confirmation, epidemiological response guidelines in resource-limited settings allow for the management of suspected cases of anthrax in the background of disease outbreaks.³

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Bhutan is a country situated in the eastern Himalayas known for its pristine environment with 43.5% of its population employed in the agriculture sector in 2022⁹ and 36.1% of the households owning livestock.¹⁰ Bhutan has three distinct eco-floristic zones with a subtropical climate in the southern belt, a temperate climate in the mid-range, and the alpine mountains. Farmers in the alpine zones are involved in rearing yaks as a source of livelihood. In the 2017 national census, there total yak population reared for livestock in Bhutan was 33,605.¹⁰ While cutaneous anthrax has been reported associated with other livestock, here, we present a case associated with an anthrax outbreak in yaks in one of the central highland districts of Bhutan.

2 | CASE PRESENTATION

2.1 | Case history and examination

A 26-year-old male, educated up to higher secondary education, a yak herder from Baylangdra village, Wangdue Phodrang district in Bhutan, presented to Wangdue Hospital with pain and swelling in the right hand for 3 days. He had handled two yak carcasses with bare hands which had died of an unknown disease with bleeding from orifices and abdominal distension. Two days later, he had pruritus over the dorsal surface of the right hand followed by the formation of small vesicular lesions and progressive swelling of the right hand. Later, the size of the lesion had increased, and produced a thin yellow discharge, but was painless. No other symptoms were reported by the patient, and he had no significant medical or surgical history including diabetes.

On examination, he had a temperature of 98.7F, pulse rate: 56/min, blood pressure: 102/64 mmHg,

respiratory rate: 20/min, and SpO₂: 98% at room air. His right hand had swelling up to the distal forearm with no features of compartment syndrome. A vesicular lesion, a size of 3 × 2 cm was formed on the dorsal surface of the right hand, just over between the second and third metacarpophalangeal joints (Figure 1). The lesion was nontender with serous discharge and had axillary lymphadenopathy (size: 1.5 × 1 cm). Movements of small joints of the right hand were restricted due to the swelling.

2.2 | Investigations

The discharge from the vesicular lesion on Gram stain and culture (specimen collected after 3 days of antibiotics initiation) did not yield any organism. An outbreak response team visited the burial site of the carcass where the index case was handled. Soil samples from the burial site of the index case could not identify *B. anthracis* spores. The findings of the investigations are summarized in Table 1.

2.3 | Differential diagnosis

Our initial provisional diagnosis was cellulitis secondary to an unknown insect bite which was managed with intravenous cloxacillin and co-amoxiclav for 3 days. His hand swelling has improved but the blister lesion remained persistent. A careful review of his history and clinical examinations fulfilled the major and minor criteria for cutaneous anthrax with high-risk exposure to suspected cutaneous anthrax in a yak.³ We notified the disease through the online national reporting system to the Royal Centre of Disease Control for surveillance reporting.

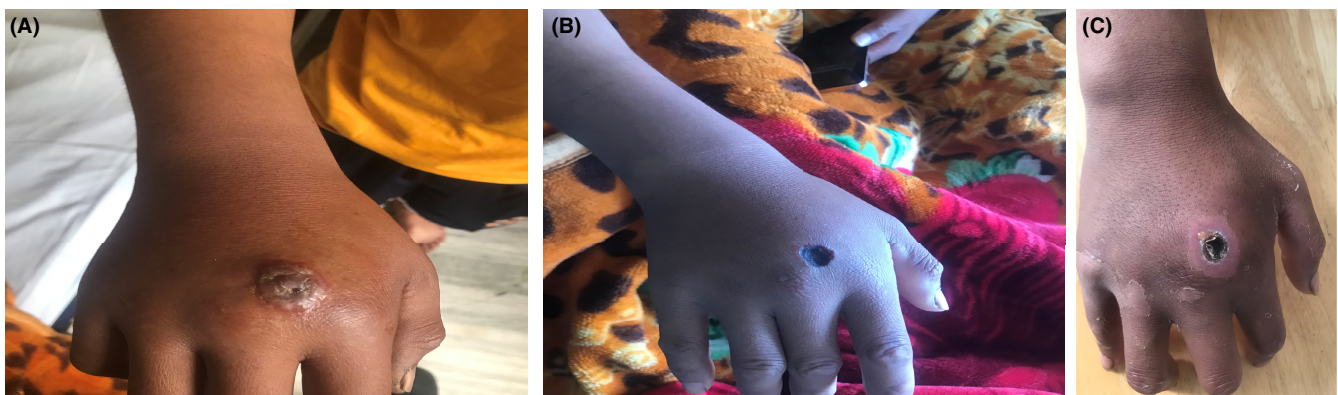


FIGURE 1 Wound over the right dorsum of hand in 26-year-old male treated for suspected cutaneous anthrax in Wangdue Phodrang district, Bhutan, June 2023. (A) Wound during hospital stay on day 3; (B) Wound on day 7; (C) Wound in healing stage during follow-up day 20.

TABLE 1 Summary of investigation findings of a 26-year-old male treated for suspected cutaneous anthrax in a yak herder in a highland community in Bhutan, 2023.

Test parameter	Patient's value	Normal range
White blood cell count (/ μ L)	9.47 \rightarrow 7.99	4–10*10 ³
Neutrophil (%)	77.9 \rightarrow 73.9	40–60%
Haemoglobin (g/dL)	16.5 \rightarrow 15.8	14–18.4
Platelet (/mL)	168 \rightarrow 181	150–450*10 ³
Urea/Creatinine (mg/dL)	18/0.8	15–45/0.7–1.3
Na/K (mEq/L)	142/3.6	133–146/3.8–5.4
Aspartate/Alanine transaminase (IU/L)	107/136	5–40
Bilirubin, total/direct (mg/dL)	2.2/0.39	0.1–1.2/<0.2
Erythrocyte sedimentation rate (mm/h)	12	0–15
Scrub typhus IgM	Negative	
Gram stain (vesicular fluid)	Observed yeast and epithelial cells	
Swab culture (vesicular fluid)	No growth	

2.4 | Treatment, outcome, and follow-up

After we suspected cutaneous anthrax clinically, his antibiotics were revised to intravenous ciprofloxacin 400 mg twice a day. With this, the size of the lesion regressed converting into a plaque with a black necrotic patch while the swelling of the right hand subsided. He received ciprofloxacin and cloxacillin for 14 days. He was discharged after 10 days of admission. On follow-up on the 20th day, the patient was physically fit, and the scar healed well with a blackish patch (Figure 1).

3 | DISCUSSION

This case demonstrates that human cases of anthrax are associated with the handling of infected animals or carcasses without adequate use of personal protective equipment. While sporadic outbreaks of anthrax have been reported with other animals, this case involves possible anthrax infection of yaks that are a source of livelihood for those living in high altitudes in Bhutan, Nepal, and China. This case was managed as a suspected case of cutaneous anthrax as per the national guideline,³ but laboratory confirmation could not be established. It is important to revisit some key aspects of anthrax with a special focus on preventing human and livestock deaths.

Cutaneous anthrax is the most common type of human anthrax that occurs with localized lesions commonly seen on exposed areas like the face, neck, hands, and legs.¹¹ The incubation period is between 2 and 7 days (range 1–19 days).^{1,11,12} The skin lesion begins with initial pruritus over the bacteria-inoculated site, followed by the formation of a small papule (2–3 days), and a ring of vesicles around it (3–4 days). Vesicles may exude, but pus will not be present unless there is a secondary infection.

The lesion itself is painless, although there is painful regional lymphadenopathy (observed in 20%) which is considered a pathognomonic feature.¹² The lesion progresses to localized oedema and ulcerates into a depressed black scar which is characterized by a black necrotic area at the centre called eschar (5–7 days). Around 10 days of infection, eschar begins to resolve and takes 2–6 weeks to resolve regardless of treatment. It may or may not be associated with systemic symptoms like fever, malaise, and headache.^{3,11,12} Complications may include septicaemia, meningitis and depending on the site of the lesion, and depending on the degree of oedema, it may also result in airway obstruction and compartment syndrome.

The diagnosis of anthrax is based on clinical features, history of exposure, and laboratory confirmation. In Bhutan, Gram stain, M'Fayden reaction, rapid antigen detection test, culture and Reverse Transcriptase Polymerase Chain Reaction (RT-PCR) are available at the national reference laboratory.³ Culturing *B. anthracis* from clinical specimens is the gold standard for diagnosing anthrax. The yield of bacterial culture from cutaneous lesions 24–48 h after initiation of any antibiotic is poor.¹³

Antibiotic administration is the mainstay of treatment. In mild or uncomplicated cases, monotherapy with intramuscular penicillin G or oral antibiotics like doxycycline, ciprofloxacin, and amoxicillin are effective. In severe or complicated cases, parenteral antibiotics should be administered. In cases where there is head and neck involvement, adjuvant corticosteroids may be considered as anti-oedema treatment to avoid serious complications like airway obstruction.¹ The duration of antibiotic therapy is debatable but guidelines suggest antimicrobial therapy for 3–5 days for uncomplicated cutaneous anthrax and 10–14 days for systemic anthrax.¹¹ However, antibiotic treatment does not affect the progression of the lesion or toxin-related tissue damage, but prompt treatment will



FIGURE 2 Field investigation at a yak herd camp at Baylangdra village situated at an altitude of 2500–3000 metres above sea level, Wangdue Phodrang district, Bhutan in July 2023. (A) Providing advocacy on the prevention of anthrax to the herders and doing clinical surveillance. (B) Specimen collection and treatment of yaks that were ill. (C) Soil specimen collection from the burial site, where index human cases had handled the dead yak carcass.

limit the size of the lesion.¹¹ Therefore, suspecting cutaneous anthrax in a patient presenting such clinical features with positive exposure and early initiation of antibiotics is essential, especially in contexts where there are no confirmatory laboratory facilities. Mortality from cutaneous anthrax is rare compared to gastrointestinal, oropharyngeal, inhalation, and meningeal anthrax if early antibiotic treatment is instituted. However, the overall case fatality ranges between 5% and 20% in untreated cases.³

Bhutan has reported sporadic outbreaks of anthrax. The last human cases were reported in 2010 from central Bhutan, where nine people had cutaneous anthrax after handling infected animals and one death due to probable inhalation of anthrax.¹⁴ Human deaths were also reported in Trongsa district in 1989 and in Wangdue Phodrang district in 1998 following the consumption of anthrax-infected meat.³

In Bhutan, anthrax is nationally reportable, and suspected or confirmed cases are immediately notified on the National Early Warning, Alert, Response Surveillance, and Information System. Reporting such cases to the national surveillance system is a key step in altering and initiating a public health response. An outbreak response team comprising of medical doctor, livestock officers, and laboratory technician visited the affected herds of yaks in locations at the altitude of 2500–3000 metres above sea level. The herders were provided awareness on clinical recognition of affected animals, proper handling of infected carcasses, and clarifying their traditional beliefs regarding the consumption of infected animal products (Figure 2). During the field investigation, the team conducted clinical surveillance on both herders and animals and disinfected the burial site and its surrounding areas (Figure 2). As shown in this outbreak response, One Health approach is essential in the prevention and outbreak response in such zoonoses with human-livestock interface.

The identification and management of this case had several limitations which reflect key areas that need to

be strengthened. The diagnosis did not have laboratory confirmation which may be due to a delay in specimen collection, inadequate specimen collection, poor technique of specimen collection, or exposure to antibiotics. Because anthrax has not been reported in Bhutan in the last decade, there might have been a decline in the levels of clinical suspicion. We recommend that One Health team members are provided refresher training from time to time especially in the context where there is turnover of all categories of health staff.

4 | CONCLUSION

This was a case of suspected cutaneous anthrax in a yak herder that occurred after handling the carcass of two yaks that died of symptoms suggestive of anthrax. However, the bacillus could not be demonstrated in clinical and environmental sampling. One Health approach was deployed in the field to respond and manage this outbreak. While human anthrax continues to occur as sporadic cases across many countries, there is a need for clinicians, public health practitioners, and national surveillance systems to improve readiness to identify, manage and respond when cases occur.

AUTHOR CONTRIBUTIONS

Ugyen Chopel: Conceptualization; data curation; investigation; writing – original draft; writing – review and editing. **Thinley Dorji:** Conceptualization; data curation; supervision; writing – review and editing. **Kuenga Sonam:** Writing – review and editing. **Sangay Dorji:** Writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

All relevant data sources are cited in the article.

ETHICS STATEMENT

Ethics approval was obtained from the Institutional Review Board, Ministry of Health of Bhutan. Informed written consent was obtained from the patient.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

ORCID

Ugyen Chophel  <https://orcid.org/0009-0002-7333-8072>

Kuenga Sonam  <https://orcid.org/0009-0001-7138-0077>

Thinley Dorji  <https://orcid.org/0000-0003-4932-8704>

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