Two Cases of Gastrointestinal Anthrax with an Unusual Presentation from Kermanshah (Western Iran)

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During 1988 – 1994 a total of 38 cases of human anthrax were admitted to Sina Hospital in Kermanshah (western Iran). There were two cases of gastrointestinal anthrax (5.3%) with culture positive ascitic fluid. Among the many reported gastrointestinal signs and symptoms, unexpectedly one of our patients had only vomiting and ascites whereas the other case had only ascites. Neither had abdominal pain, tenderness, diarrhea, hematemesis, melena, or other expected signs and symptoms of anthrax. Therefore, in contrast to the available reports, these cases presented atypically and despite receiving a sufficient dose of penicillin, the drug of choice at that time, both patients died.

Gastrointestinal anthrax is not as rare as reported but due to an unusual presentation it may be misdiagnosed. Paying attention to gastrointestinal anthrax in the differential diagnosis of ascites with unknown origin and other gastrointestinal presentations in endemic areas may help to diagnose more cases of anthrax. Timely appropriate management in an early stage of the disease, may increase their chances of survival.

Keywords: Anthrax • bacillus anthracis • gastrointestinal anthrax

Introduction

Anthrax is primarily a disease of herbivores, which acquire infection after coming into contact with the soil-borne spores of Bacillus anthracis.1 These spores incidentally infect humans who are exposed to infected animals or their products.2 Intentional and non-humanitarian misuse as a biological weapon is also one of the newer modes of transmission.3 This ancient disease is also described in historical texts.4

Bacillus anthracis is a large, gram-positive, aerobic, spore-forming bacillus5 whose spores germinate when exposed to a nutrient-rich environment, such as the tissues or blood of an animal or human host.6

Infection is initiated with the introduction of the spores through a break in the skin (cutaneous anthrax), entry through the mucosa (gastrointestinal anthrax), or deposition of spores in alveolar spaces by inhalation.7

Human disease manifests itself in different forms such as cutaneous (95%), inhalation, gastrointestinal (GI) (<5%), and rarely oropharyngeal and meningial.8

GI anthrax is extremely rare in industrialized countries but is more frequently encountered in developing countries.9

When the spores are swallowed, they may cause lesions from the oral cavity to the cecum. Ulcerative lesions, usually multiple and superficial, may occur in the stomach and are sometimes associated with similar lesions in the esophagus and jejunum. These ulcerative lesions may bleed. In severe cases, hemorrhage may be massive and fatal. They may also lead to obstruction, perforation, or any combination of these complications. Ascites may complicate GI anthrax. In some patients, the fluid that shifts from the vascular compartment can lead to shock and death.10

Paracentesis may reveal hemorrhagic ascites,
and these cases sometimes simulate an acute or surgical abdomen. Mesenteric lymphadenitis usually occurs with GI anthrax. The primary lesions occur most frequently in the terminal ileum or cecum.11

Case Report

We have admitted 38 cases of anthrax to Sina Hospital in Kermanshah. Of these admissions, two patients (5.3%) were diagnosed with GI anthrax and the remainder (94.7%) had cutaneous anthrax.

The first case

A 13-year-old girl from Kermanshah province, western Iran was admitted to Sina Hospital with complaints of cyanosis, circulatory collapse, and fever. She had a five day history of fever, chills, and repeated vomiting prior to admission.

At the time of admission, the patient was in circulatory collapse and had acrocyanosis. Body temperature (sublingual) was within normal limits. Positive findings upon physical examination were acrocyanosis, undetectable pulses, and abdominal distension without organomegaly but with shifting dullness, suggestive of ascites. Despite the fast progression of the disease and with a probable diagnosis of tuberculous peritonitis, which was prevalent at that time in the province, the ascitic fluid was withdrawn for analysis.

Peripheral blood

WBC 19600 cells/mm$^3$; neutrophils 75%; lymphocytes 18%, stabs 5%; eosinophils 1%; blood urea nitrogen 108 mg%. Creatinine could not be measured due to severe hemolysis and paucity of the serum.

Ascitic fluid

WBC 5950 cells/mm$^3$; neutrophils 93%; lymphocytes 6%; monocytes 1%; RBC 11600 cells/mm$^3$; protein 4g/L; glucose 208 mg/dL (concomitant blood sugar: 340 mg/dL). The smear for acid fast bacilli (AFB) was negative and a gram stain showed presence of gram-positive mycelium-shaped elements.

According to the above presentations and with particular attention to the report of mycelium-shaped filaments; a diagnosis of abdominal actinomycosis was made, although the rapid course of the disease was not suggestive of this diagnosis. In addition to an intravenous fluid infusion and dopamine, crystalline penicillin G was started at a dose of 3 million units QID. Nonetheless the patient died in a state of shock and circulatory collapse less than 12 hours after admission. The probable diagnosis was septic shock.

Two days later, the patient’s ascitic fluid cultured positive for Bacillus anthracis which confirmed a diagnosis of GI anthrax.

Upon confirmation of the diagnosis, the patient’s family members were questioned in order to obtain the source of the infection. Family members, who were farmers and cattlemen, stated that the patient had eaten briefly cooked liver from a sheep that was dying from severe abdominal distention. The sheep had been slaughtered in that condition (before dying) 2 – 3 days before the onset of the patient’s disease. Unfortunately, further evaluation could not be performed because family members rejected a postmortem study, however there were no similar cases of either GI anthrax or cutaneous anthrax amongst other family members.

The second case

A 67-year-old man from Kermanshah province, presented with a three week history of complaints of dyspnea, productive cough, anorexia, weight loss, night sweats, fever, and chills prior to his admission to the infectious ward of Sina Hospital. At the time of admission the patient was afebrile with pulse and respiratory rates of 110/min and 30/min, respectively. He had no complaints except for respiratory symptoms, but during the third day of the admission he developed fever (38.5°C). The fever lasted for two days and upon physical examination of the chest, there were signs suggestive of pleurisy.

He also developed abdominal distention. Abdominal ultrasonography was performed, which revealed ascites with multiple para-aortic adenopathies. Ascitic fluid was sent to laboratory for biochemical analysis, cell count, staining, and culture.

Peripheral blood

WBC 17900 cells/mm$^3$; neutrophils 85%; lymphocytes 14%; large lymphocytes 75%; sodium 134 (meq/lit).

Pleural fluid

WBC 10000 cells/mm$^3$; neutrophils 97%; lymphocytes 3%; protein 4.9 g/L; glucose 135 mg/dL.
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**Ascitic fluid**

WBC 7800 cells/mm³; neutrophils 94%; lymphocytes 6%; RBC 5000 cells/mm³; protein 4.9 g/L; glucose 100 mg/dL (at time blood sugar: 112 mg/dL). An AFB smear was negative, however the gram stain showed the presence of gram-positive bacilli which cultured positive for *Bacillus anthracis*. Urinalysis was also normal. Lymphocytes 6%; RBC 5000 cells/mm³; protein 4.9 g/L; glucose 100 mg/dL (at time blood sugar: 112 mg/dL). An AFB smear was negative, however the gram stain showed the presence of gram-positive bacilli which cultured positive for *Bacillus anthracis*. Urinalysis was also normal.

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Based on the culture results, a diagnosis of atypical GI anthrax was made and treatment with crystalline penicillin, 24 million units per day, was started. Seven days later when the ascites, pleurisy, dyspnea, tachycardia, and tachypnea were still present, the patient died. The source, reservoir, and transmission route were not identified.

**Discussion**

GI anthrax is a rare disease that occurs one to five days after the ingestion of undercooked meat that contains anthrax spores. The signs and symptoms of the classic form include: an initial prodromal phase with fever, malaise, and syncope; a second progressive phase with abdominal pain, nausea, vomiting, abdominal distention, ascites, and severe weakness; and a fulminant third phase with suddenly increasing abdominal girth and expanding ascites, paroxysmal abdominal pain, and shock.

As described above, we reported two cases of GI anthrax that presented with progressive ascites, without the other symptoms such as: abdominal pain, discomfort, diarrhea, hematemesis, and melena. However other reported hospitalized patients had all or several of the mentioned signs and symptoms (Tables 1 and 2). Despite the administration of a high dose of crystalline penicillin G, our two cases died. However, in the other 14 Iranian cases, the mortality rate was 85%. On the other hand, mortality rate of all Iranian cases (87.5%, 14 of 16 cases) in comparison with other sporadic hospitalized cases (22.3%, 2 of 9 cases) was significantly higher (Table 2).

It needs to emphasize that reports concerning the mortality rate are biased towards hospitalized patients with severe cases, in comparison with the epidemic cases. The total mortality rate of sporadic hospitalized patients has been 64% (16 of 25 cases) in comparison with patients admitted during an epidemic, which has been about 5.53% (12 of 217 cases). This could be due to the rapid detection and early management during an outbreak.

Neither of our two patients had significant GI symptoms such as abdominal pain, diarrhea, or hematemesis. In this regard, they were different from classically reported cases. Therefore, especially in the mountainous regions of our country, we must always consider GI anthrax in the differential diagnosis of ascites, whether it is associated with other GI symptoms or not. Because *Bacillus anthracis* can be easily identified in the smears and cultures of discharges; therefore preparing smears and cultures of ascitic fluid samples should be an important priority.

We concluded that the GI anthrax was not as rare as reported, however because of an unusual presentation it may remain undiagnosed. Paying attention to GI anthrax in the differential diagnosis of ascites with an unknown origin and probably other GI presentations in endemic areas may help to diagnose more cases and manage them in the

<table>
<thead>
<tr>
<th>Year Reported</th>
<th>Hospitals and Provinces</th>
<th>No. of patients</th>
<th>Outcome*</th>
<th>GI signs and symptoms (other than ascites)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1954</td>
<td>Firoozabadi Hospital, Tehran, Iran</td>
<td>1</td>
<td>Died</td>
<td>Present</td>
</tr>
<tr>
<td>1970</td>
<td>Khuzestan Province, southern Iran</td>
<td>1</td>
<td>Died</td>
<td>Present</td>
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<tr>
<td>1995</td>
<td>Children’s Medical Center Hospital, Tehran</td>
<td>1</td>
<td>Died</td>
<td>Present</td>
</tr>
<tr>
<td>2002</td>
<td>Imam Hossein Hospital, Tehran, Iran</td>
<td>1</td>
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<td>Present</td>
</tr>
<tr>
<td>2003</td>
<td>Shiraz, Iran</td>
<td>9</td>
<td>8 Died</td>
<td>Present</td>
</tr>
<tr>
<td>2006</td>
<td>Mazanderan Province, Iran</td>
<td>1</td>
<td>Survived</td>
<td>Present</td>
</tr>
<tr>
<td>2006</td>
<td>Kermanshah Province, Iran (Current report)</td>
<td>2</td>
<td>2 Died</td>
<td>Absent</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td></td>
<td><strong>16</strong></td>
<td><strong>14</strong></td>
<td><strong>87.5%</strong></td>
</tr>
</tbody>
</table>

* Mortality rate: 87.5%
early stages of the disease.

The differential diagnoses include: food poisoning, acute abdomen owing to other reasons, and hemorrhagic gastroenteritis caused by other microorganisms, in particular necrotizing enteritis caused by Clostridium perfringens, and dysentery.14

References


Table 2. Published sporadic and epidemic GI anthrax, 1954 – 2006

<table>
<thead>
<tr>
<th>Country</th>
<th>Sporadic cases</th>
<th></th>
<th>Epidemic cases</th>
<th></th>
<th>Total</th>
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<td>—</td>
<td>2</td>
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<tr>
<td><strong>Total</strong></td>
<td><strong>25</strong></td>
<td><strong>16</strong></td>
<td><strong>217</strong></td>
<td><strong>12</strong></td>
<td><strong>242</strong></td>
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