



## Case report

Glanders (*Burkholderia mallei* infection) in an Iranian man: A case reportMaryam Nasiri<sup>a,\*</sup>, Amirali Zarrin<sup>b</sup>, Sina RoshankarRudsari<sup>c</sup>, Javad Khodadadi<sup>d</sup><sup>a</sup> Department of Infectious Diseases, School of Medicine, Iran University of Medical Sciences, Tehran, Iran<sup>b</sup> Student Research Committee, faculty of Medicine, Iran University of Medical Sciences, Tehran, Iran<sup>c</sup> School of Medicine, Islamic Azad University, Tehran, Iran<sup>d</sup> Department of Infectious Diseases, Faculty of Medicine, Qom University of Medical Sciences, Qom, Iran

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## ABSTRACT

**Background:** Glanders is a rare disease that has been eradicated in many countries but may be difficult to diagnose due to its nonspecific symptoms. This disease, which can be highly fatal if left untreated, is caused by a bacterium called *Burkholderia mallei*. Humans can get the disease through contact with infected animals, such as horses. Over time, various treatment strategies have been proposed for this disease, and attempts have even been made to develop a vaccine, but thus far, no effective vaccine has been developed to prevent it.

**Case presentation:** In this article, we describe the case of Glanders disease in KamkarArabnia Hospital in Qom, Iran. The patient was a 22-year-old man with headache, fever, chills, diarrhea, and vomiting of blood, and was admitted to the infectious ward in isolation.

**Conclusion:** The lack of definitive diagnostic symptoms and the rarity of this disease make it difficult to diagnose, and one should be careful in dealing with its symptoms. Also, paying attention to the patient's medical history and travel history to endemic areas, can lead to timely diagnosis and treatment.

## Introduction

Glanders disease was first detected by Greeks in 450–425 BC [1]. The pathogen that is responsible for this disease is called *Burkholderia mallei*, which can affect humans along with animals. Horses, donkeys, and mules are considered as the pathogen's vectors [2].

Glanders is a rare disease, mostly found in Southeast Asia and north Australia [3], but it can also be found in other countries as a result of traveling from and to endemic regions. Most empirical antibiotics have no therapeutic effect on *Burkholderia mallei*; therefore, a lack of administration of the most efficient regimen could be fatal for patients.

If Glanders is left untreated, the estimated mortality rate can be as high as 95% [4,5]. A wide range of clinical manifestations are seen in patients with Glanders, from asymptomatic cases to severe conditions, like septicemic shock [6]. Thus, diagnosing this disease at early stages in order to prevent catastrophic consequences has a crucial role in the management of patients. We report a 22-year-old patient without a significant past medical history.

## Case presentation

A 22-year-old man without any known past medical conditions presented to the emergency department with the following symptoms: fever, chills, cough, runny nose, headache, and diarrhea 5–6 times a day. The symptoms had started a week before his admission and had become more severe over time. He also mentioned two episodes of bloody vomiting and one episode of melena. The patient had recently buried a dead horse, which he claimed had been sick. Although the patient's family had been in constant contact with him, they had no symptoms and did not complain of any particular problems. The patient had no specific underlying disease and was not taking any medication.

Vital signs were stable upon arrival at the emergency department. On physical examination, everything was normal except a 5 mm enlarged lymph node in the posterior left cervical chain of the neck, which was mobile. The consultation of an infectious disease specialist was requested and the patient was admitted to the infectious ward under isolated conditions. Necessary lab tests were requested and CBC showed white blood cells of  $3.9 \times 10^9/L$ , a platelet count of  $55 \times 10^9/L$ , and a hemoglobin level of 13.8 g/dL. C-reactive protein was 56 mg/L (More than 10:Positive). AST and ALT were 108 U/L and 98 U/L, respectively. LDH

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and blood culture were normal. The chest X-ray and abdominal sonography did not show anything significant. Stool exam was also performed because of diarrhea, which was normal.

Due to the patient's contact with the sick horse, empirical treatment with imipenem and doxycycline was started, and after three days of treatment, AST and ALT were reduced (Fig. 1) and platelet counts began to rise after seven days (Fig. 2).

During hospitalization, due to suspicion of Glanders disease, serological tests were requested for *Burkholderia mallei*, and the results were positive after seventeen days. The symptoms resolved and the patient was discharged to continue his therapy with Trimethoprim/Sulfamethoxazole in an outpatient setting. Patient was followed up for three months and he was free from any signs and symptoms.

## Discussion

Glanders is not a common disease and has been eradicated in many countries, including the United States [7]. A bacterium called *B. mallei* is the cause of this disease [8]. This bacterium is gram-negative and can form spores [2,9].

The disease is highly infectious [1]. However, it is rare among humans, except those who are in constant contact with infected solipeds [7]. Untreated Glanders can be up to 95 % fatal, while if treated well, the death rate can be reduced by up to 50 % [7]. The high mortality rate of this disease stresses the importance of its diagnosis and treatment.

Light fever in the afternoon or evening, nausea, fatigue, headache, back pain, lymphadenopathy, and chest pain have been mentioned as the most common symptoms of Glanders [1]. Nevertheless the period of infection can be diverse due to the various forms of exposure [9]. Therefore, the symptoms of this disease should raise suspicion among clinicians in areas with high prevalence.

The differential diagnoses include viral, bacterial, or mycoplasmal pneumonia, malaria, anthrax, CBRNE-plague or smallpox, and typhoid fever [7].

Various treatments have been administered for Glanders, but the small number of patients seems to have made it somewhat difficult to reach a definitive cure. One patient with Glanders was reported to be treated with intravenous imipenem and doxycycline for one month and then azithromycin and oral doxycycline for six months [10]. The duration of intravenous treatment starts from ten days and can be longer [1]. Oral treatment should last at least 3 months and can continue for as long as a year [5]. Surgical treatment may be needed for abscess formation in organs such as the prostate gland [7]. Genetic sequencing of the pathogenesis of this disease may facilitate the development of ways to combat

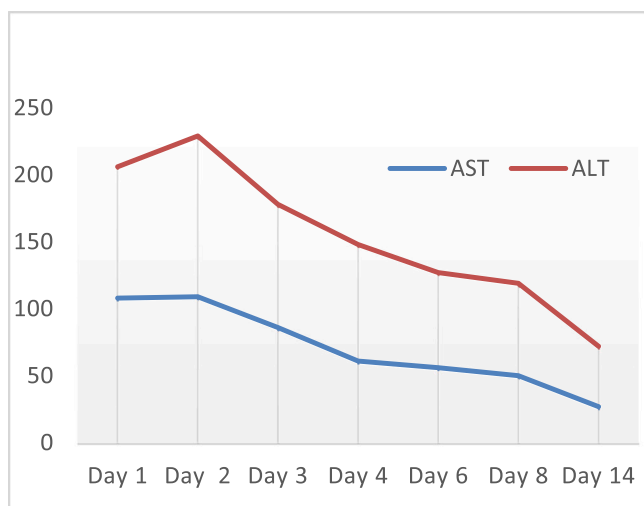


Fig. 1. Evaluation of changes in liver enzymes.

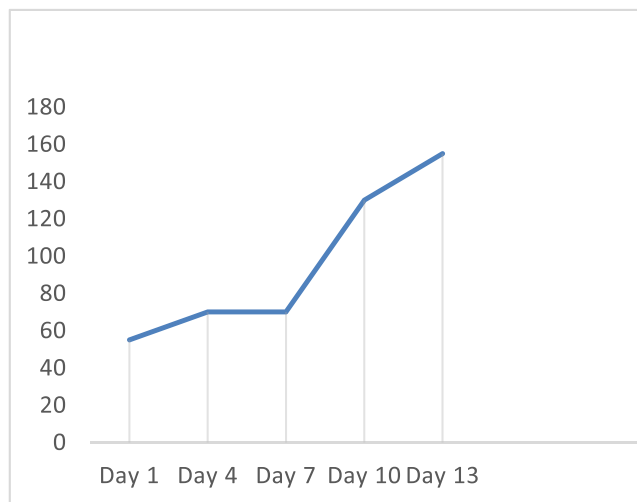


Fig. 2. Evaluation of changes in platelet count.

it [11].

It usually takes 1–3 weeks to incubate the bacteria that cause Glanders, but this time can increase from a few months to several years [7]. Due to the non-specific symptoms of the disease, it is difficult to diagnose it definitively; therefore, the opportunity to treat it in the golden time may be lost.

On the other hand, thus far, no vaccine has been discovered for this disease [11]. Although the causative agent of Glanders disease can be grown in-vitro, the disease-causing bacteria rarely grow in blood cultures [7]. Imaging techniques, such as CT and ultrasound, can be helpful, but they cannot conclusively confirm the disease. Radiography can reveal some lesions [1].

## Conclusion

Glanders is a rare disease in many countries, and the symptoms are commonly observed in many other diseases, which makes it difficult to diagnose. One factor that may play an important role in the diagnosis is the patient's history of exposure to a carrier. It is also necessary to pay attention to the patient's travel history to areas where the disease is highly prevalent. Knowing the pathogenicity of this disease is pivotal in preventing its complications. The necessary protocols to deal with it should be devised by the responsible institutions in each country, and efforts to produce an effective vaccine should be supported.

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## CRediT authorship contribution statement

Please specify the contribution of each author to the paper, e.g. study design, data collections, data analysis, writing, others, who have contributed in other ways should be listed as contributors.

## Ethical approval

Not applicable.

## Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images

## Competing interests

The authors declare that they have no competing interests.

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