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Case Report

Patient Presenting with Abscess Unresponsive to Treatment and Progressive to Osteomyelitis: A Rare Cause *Burkholderia mallei*

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Abstract

Glanders is a rare zoonotic disease caused by *Burkholderia mallei* (*B. mallei*). *B. mallei* can cause pneumonia, abscesses, osteomyelitis in severe cases, sepsis, and even death in humans. In this report, we present a 15-year-old male patient living in a rural area who was diagnosed with glanders. The patient, who did not have any previous disease, was followed up with a diagnosis of pneumonia in the hospital, where he was admitted with complaints of cough and abdominal pain and presented to us with pain, redness, and swelling in his leg. Magnetic resonance imaging of the lower extremity revealed osteomyelitis in the fourth and fifth metatarsals of the right foot. *B. mallei* growth was detected in the abscess culture. Meropenem treatment was started. The patient's symptoms regressed with treatment. The patient was discharged with oral ciprofloxacin for *B. mallei* eradication. Glanders are usually transmitted through direct contact with infected animals, especially single-hoofed animals such as horses, or through inhalation of aerosols containing *B. mallei*. It is a rare disease-causing pneumonia and abscesses and can be life-threatening in severe cases. Diagnosis of glanders is difficult because the initial symptoms are non-specific. Isolation of *B. mallei* in culture is the gold standard for diagnosing the disease. There is no clear recommendation for treating glanders and imipenem; meropenem ceftazidime can be used based on antibiotic susceptibility tests.

Keywords: Abscess, *burkholderia mallei*, glanders

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Glanders is a highly contagious and fatal zoonotic disease caused by *Burkholderia mallei* (*B. mallei*), primarily affecting horses.^[1] This disease has been reported to infect mainly horses, ungulates, and, more rarely, humans.^[2]

Case Report

In this case report, we present a 15-year-old patient whose clinic started with pneumonia findings, developed an abscess in the lower extremity, and progressed to osteomyelitis, and *B. mallei* was grown in the abscess culture.

It was reported that a 15-year-old male patient with no previous known disease was admitted to the hospital one month ago with complaints of cough and abdominal pain and was hospitalized with the diagnosis of pneumonia. On the third day of hospitalization, pain, redness, and swelling started to develop in the medial right leg. With a prediagnosis of abscess, the patient was treated with clindamycin and amikacin. Chest X-ray revealed a large consolidated area and infiltration in the left lower lobe, an abdominal ultrasound revealed minimal hepatosplenomegaly and bi-

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lateral 1-1.5 cm inguinal lymphadenopathy, and magnetic resonance imaging (MRI) of the lower extremities revealed soft tissue edema.

After methicillin-resistant *Staphylococcus aureus* (MRSA) growth was detected in the abscess culture, the patient was consulted with us. The patient's general condition was good on physical examination, vital signs were stable, and lung sounds were normal. The patient had 5x3 cm drained abscesses on the medial aspect of the right lower extremity (Fig. 1), ulcerated abscesses around the necrotic periphery, 3x4 cm reddened abscesses with fluctuations on the dorsum of the right foot, and 2x2 cm abscesses with new onset on the medial aspect of the lower extremity. Based on the antibiotic susceptibility test, the patient was treated with piperacillin-tazobactam and teicoplanin. In the patient's follow-up, the right foot's abscess was drained. MRI of the lower extremity was repeated, and findings compatible with osteomyelitis were found in the fourth and fifth metatarsal bones of the right foot (Fig. 2).

B. mallei was grown in the abscess culture. *B. mallei* was also isolated from the abscess material grown in a blood culture tube. According to the antibiotic susceptibility test, piperacillin-tazobactam was discontinued, and meropenem was added to the treatment (Fig. 3). In the medical history, it was learned that the patient lived in a rural area,



Figure 1. Ulcerated irregular wound with a diameter of 5x3 cm on the medial of the right lower extremity.

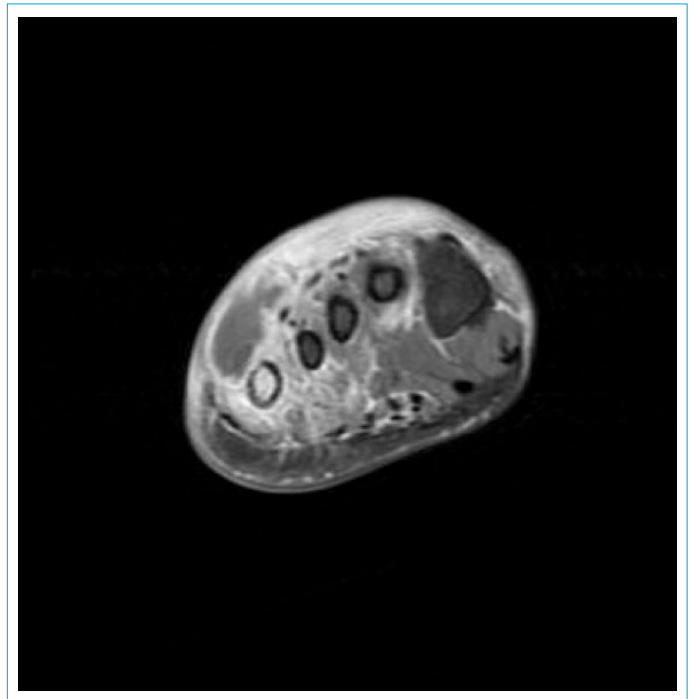


Figure 2. MRI findings consistent with osteomyelitis in the 4th and 5th metatarsals of the right foot.

was engaged in animal husbandry, and was in contact with equine animals such as horses, donkeys, and mules. On the 28th day of meropenem treatment, the findings of osteomyelitis regressed, and the deep ulcerated wound closed completely in the control extremity MRI. The patient was discharged with oral ciprofloxacin for outpatient follow-up. Outpatient follow-up showed that the wound healed completely with oral ciprofloxacin (Fig. 4). Written and verbal consent was obtained from the patient's mother regarding the patient's illness and photo sharing.

Discussion

Data on glanders in pediatric patients are limited. In 1960, one case was reported in Türkiye.^[3] Our case is one of the rare cases reported in Türkiye.^[4]

B. mallei infection can cause different clinical pictures in humans. The disease is usually diagnosed as pneumonia and begins with fever, cough, and respiratory distress. During the disease, bacteremia, pustular skin lesions, and soft tissue abscesses may occur.^[5]

In 2020, a case published in the literature showed *B. mallei* growth in the abscess culture of an 11-year-old male patient who had contact with horses in Brazil. The symptoms of this patient, like our case, started with signs of pneumonia and were characterized by extremity abscess in the follow-up.^[6] Notably, *B. mallei* infection was also found in our patient with extremity abscess and osteomyelitis.

| | | | |
|---|---|-----------------------------|-------------------|
| BACTERIAL IDENTIFICATION AND SENSITIVITY TEST (AUTO) | | Barcode Number: 16727232 | Sample Type: swab |
| *EVALUATION WAS MADE WITH THE ZONE DIAMETERS USED FOR PSEUDOMONAS ACCORDING TO CLSI CRITERIA <small>*Restricted reporting* standards recommended by CLSI/NCCLS are applied in antibiogram result notifications. Our bacteriology laboratory is included in the Central Asian and Eastern European Antimicrobial Resistance Surveillance (CAESAR) network.</small> | | | |
| WOUND CULTURE swab | | Barcode Number: 16727232 | Sample Type: swab |
| REPRODUCING MICROORGANISMS | | | |
| 1. BURKHOLDERIA MALLEI | | Number of Colonies: [DENSE] | |
| Antibiotic Name | - | MIC-1 | MIC-2 MIC-3 |
| AMIKACIN | S | <=1 | |
| AZTREONAM | I | 16 | |
| CEFEPIME | R | >=32 | |
| CEFTAZIDIME | S | - | |
| CIPROFLOXACIN | I | 0.5 | |
| IMPENEM | I | <=0.25 | |
| MEROPENEM (Other) | S | <=0.25 | |
| MEROPENEM (MENINGITIS) | S | <=0.25 | |
| PIPERACILLIN/TAZOBACTAM | I | <=4 | |
| SENSITIVE:S INTERMEDIATE :I RESISTANT:R | | | |
| *EVALUATION WAS MADE WITH THE ZONE DIAMETERS USED FOR PSEUDOMONAS ACCORDING TO CLSI CRITERIA <small>*Restricted reporting* standards recommended by CLSI/NCCLS are applied in antibiogram result notifications. Our bacteriology laboratory is included in the Central Asian and Eastern European Antimicrobial Resistance Surveillance (CAESAR) network.</small> | | | |
| Medical Laboratory Comment: | | | |
| <small>*Restricted reporting* standards recommended by CLSI/NCCLS are applied in antibiogram result notifications. Our bacteriology laboratory is included in the Central Asian and Eastern European Antimicrobial Resistance Surveillance (CAESAR) network.</small> | | | |

Figure 3. Antibiotic susceptibility test.



Figure 4. Patient's wound after treatment.

The diagnosis of glanders is difficult because the initial symptoms are non-specific. Isolation of *B. mallei* in culture is the gold standard in diagnosing glanders, but it is difficult to isolate *B. mallei* even in acute abscess culture.^[7] Although the patient had previously MRSA, *B. mallei* growth was observed in the second abscess culture. In cases not responding to treatment, *B. mallei* should be considered the causative agent, especially in patients with animal contact, even if there is no growth in culture.

Although it is susceptible to many antibiotics in vitro, there is no clear recommendation for treating *B. mallei* infections in humans. In the literature, patients have been reported to be treated with imipenem, meropenem, and ceftazidime. It has been reported that oral trimethoprim-sulfamethoxazole (tmp-smx) or tmp-smx + doxycycline treatment is given for definitive eradication of the disease after long-term intravenous (iv) treatment.^[7,8] In our case, since *B. mallei* was grown in the abscess culture, iv meropenem was added to his treatment, and he received meropenem treatment for 30 days. For definitive eradication, oral ciprofloxacin was given according to the antibiotic susceptibility test. In pul-

monary and bacteremia forms of the disease, the mortality rate is 90-95% without treatment and 40-50% with treatment.^[8] Our patient, who was followed up for a long time, recovered with a cure.

In conclusion, *B. mallei* should be considered the causative agent in unresponsive pneumonia followed by skin soft tissue infections or organ abscesses, especially in cases of contact with single-hoofed animals without culture growth. Rapid and accurate diagnosis of *B. mallei* infection will protect against severe osteomyelitis infection and even sepsis and death. In addition, surveillance for the detection of sick animals and culling of these sick animals is essential for the prevention of glanders. We think that our case, which recovered with early diagnosis and correct treatment, is a guide for rare cases of glanders.

Disclosures

Informed consent: Written informed consent was obtained from the patients' family for the publication of the case report and the accompanying images.

Peer-review: Externally peer-reviewed.

Conflict of Interest: None declared.

Authorship Contributions: Concept – F.T.C., E.B., U.C., D.A., O.O.G., H.G., F.K., B.K.; Design – F.T.C., E.B., U.C., D.A., O.O.G., H.G., F.K., B.K.; Supervision – F.T.C., E.B., U.C., D.A., O.O.G., H.G., F.K., B.K.; Fundings – F.T.C., O.O.G., U.C.; Materials – F.T.C., B.K., F.K., U.C., O.O.G.; Data Collection and/or Processing – H.G., F.K., B.K., F.T.C., O.O.G.; Analysis and/or Interpretation – F.T.C., O.O.G., U.C., E.B., F.K., B.K., H.G., D.A.; Literature Search – F.T.C., B.K., D.A., O.O.G., H.G., F.K.; Writing – F.T.C., O.O.G., U.C.; Critical Reviews – F.T.C., O.O.G., U.C., B.K.

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References

1. Whitlock GC, Estes DM, Torres AG. Glanders: off to the races with *Burkholderia mallei*. *FEMS Microbiol Lett* 2007;277:115–22. [\[CrossRef\]](#)
2. Estes DM, Dow SW, Schweizer HP, Torres AG. Present and future therapeutic strategies for melioidosis and glanders. *Expert Rev Anti Infect Ther* 2010;8:325–38. [\[CrossRef\]](#)
3. Uçar N. Memleketimizde insanlarda tesbit edilen ruam vak'aları. *Etlık Vet Mikrobiyol Derg* 1960;1:65–8.
4. Doğanay M, Yılmaz E, Topluoğlu S, Şahin M, Diker KS, Akçay E, et al. Glanders evaluation report. *Turk Bul Hygiene Experiment Biol* [Article in Turkish] 2020;77 Suppl 1:1–24.
5. Srinivasan A, Kraus CN, DeShazer D, Becker PM, Dick JD, Spacek L, et al. Glanders in a military research microbiologist. *N Engl J Med* 2001;345:256–8. [\[CrossRef\]](#)
6. Santos Júnior ELD, Moura JCR, Protásio BKPF, Parente VAS, Veiga MHND. Clinical repercussions of Glanders (*Burkholderia mallei* infection) in a Brazilian child: a case report. *Rev Soc Bras Med Trop* 2020;53:e20200054. [\[CrossRef\]](#)
7. Lipstiz R, Garges S, Aurigemma R, Baccam P, Blaney DD, Cheng AC, et al. Workshop on treatment of and postexposure prophylaxis for *Burkholderia pseudomallei* and *B. mallei* infection, 2010. *Emerg Infect Dis* 2012;18:e2. [\[CrossRef\]](#)
8. Van Zandt KE, Greer MT, Gelhaus HC. Glanders: an overview of infection in humans. *Orphanet J Rare Dis* 2013;8:131. [\[CrossRef\]](#)