



## Case report

# Disseminated Melioidosis presenting as pneumonia, femoral and sacral osteomyelitis, splenic abscess and high rectal fistula: A case report and review of literature

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## ARTICLE INFO

## Keywords:

Disseminated Melioidosis  
*Burkholderia pseudomallei*  
 Splenic abscess  
 Rectal fistula  
 Osteomyelitis

## ABSTRACT

**Introduction:** Melioidosis is a rare infectious tropical disease caused by *Burkholderia pseudomallei* (*B. pseudomallei*), an environmental saprophyte usually habitating on soils of Southeast Asian fields. Most of the reported cases present with pneumonia and intra-abdominal abscess. Diagnosis is established by culture studies from the blood, sputum or abscess drainage. Management relies on culture-guided antibiotic treatment, with good prognosis. Surgical intervention is required in cases not responsive to medical management.

**Presentation of case:** We are presenting a case of Melioidosis in a 72 year old Filipino who presented with Pneumonia, Femoral and Sacral Osteomyelitis, Splenic Abscess and High Rectal Fistula. He was successfully managed with systemic antibiotic treatment and surgery. The splenic abscess was managed by splenectomy and a transverse loop colostomy was used for fecal diversion to address the rectal fistula.

**Discussion:** Melioidosis varies in its presentation and thus management should be individualized, depending on the organs involved. Our patient presented with multiple foci of infection which rendered the treatment more complicated as compared to those reported previously in published literature. The pneumonia and the osteomyelitis were managed with aggressive systemic antibiotics but the other sites of infection required drainage and surgery.

**Conclusion:** Melioidosis is a rare infection caused by an environmental saprophyte *Burkholderia pseudomallei*. An accurate diagnosis using culture studies is essential to institute appropriate treatment. Antibiotic treatment complemented by surgery for specific organ involvement is essential for cure.

## 1. Introduction

*Burkholderia pseudomallei* is a gram negative facultative intracellular bacteria causing a disease called Melioidosis or Whitmore's Disease. This was first described in 1912 by Alfred Whitmore and C. Krishnaswami, seen mostly in Southeast Asia, particularly in Thailand, Singapore, Malaysia and Northern Australia. Although the disease is mostly distributed in the tropical and subtropical areas, more cases are now being reported in other parts of Asia like India, Hong Kong, South China and outside Asia like Mexico, Puerto Rico and Brazil to name a few. Melioidosis is an environmental saprophyte, inhabiting soil and freshwater fish [1]. The predominant mode of transmission is through percutaneous inoculation of contaminated wet season soil or water through eating freshwater fish. It primarily affects older adults (40–50

years old) but can also affect children. Although healthy individuals may have fulminant Melioidosis, severe disease and mortalities are uncommon without risk factors. Most common risk factor is Diabetes Mellitus, especially for patients with bacteremia and asymptomatic infection. Other risk factors are alcohol use, chronic kidney disease and chronic lung disease [2]. Patients usually present with Pneumonia and bacteremia with either cutaneous or solid organ abscesses, mostly of the spleen, kidney, bone and prostate. Children usually present with cutaneous skin infections. Patients are diagnosed through cultures, either from blood, sputum, abscess fluid or swab from lesion. Imaging tests like Chest XRay, CT scan and MRI can reveal solid organ abscesses. Treatment primarily is with IV antibiotics and management of involved organ (s) accordingly [3].

Locally, the first case in the Philippines was documented in 1948.

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<https://doi.org/10.1016/j.ijscr.2021.106588>

Received 1 October 2021; Received in revised form 6 November 2021; Accepted 9 November 2021

Available online 10 November 2021

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Majority of cases were from Isabela and Metro Manila and presented as either Pulmonary Melioidosis or Disseminated Melioidosis. Among the diagnosed cases of Disseminated Melioidosis, abscesses are usually in the liver, spleen and cutaneous or soft tissue. Most of these patients recovered with administration of intravenous antibiotics, primarily Ceftazidime and Trimethoprim-Sulfamethoxazole. The last reported case of documented Melioidosis in the Philippines was a 44 year old Male in 2016 who presented with hepatic abscess and was successfully treated with Meropenem and Trimethoprim-Sulfamethoxazole. Melioidosis may mimic Tuberculosis and Disseminated Tuberculosis and for this reason may be under-reported in the Philippines [4].

The case being presented in this study is a case of Disseminated Melioidosis presenting with abscess from the spleen, Musculoskeletal (Gluteal, Piriformis muscles; Osteomyelitis) and a High Anal Fistula. The patient was diagnosed by abscess culture and was treated with Ceftazidime, Meropenem, Levofloxacin and Trimethoprim-Sulfamethoxazole. He underwent Splenectomy and Sigmoid Loop Colostomy and was sent home improved.

This case is being presented for the rarity in presentation and also for the complex multidisciplinary management it required. This is being reported in line with the SCARE 2020 guidelines [5].

## 2. Case presentation

The patient is a 72 year old male, diabetic, farmer from Isabela who presented initially with right hip pain treated with pain relievers at home. He was being maintained on maintenance hypoglycemic medications but otherwise had no other co morbidities. The rest of the past medical history, family and social history were unremarkable. Persistence of the pain prompted consult for evaluation and subsequent admission. Work-up revealed WBC count of 22,800 and the Chest x-ray showed Pneumonia. He was given IV Ceftazidime, Meropenem and Co-Trimoxazole and was discharged improved after 1 week. Interim history was unremarkable until he developed high grade fever for which he was readmitted. On his second admission, imaging tests revealed right gluteal, right hip and splenic abscesses (Fig. 1). He was advised surgery, Splenectomy, but deferred decision and sought further treatment in Manila.

On admission in our institution, further work-up included an MRI of the lower abdomen showing Osteomyelitis of the right proximal femur and a recto-gluteal fistula (Fig. 2). Other significant work-up showed a WBC count of 12,000; pus on stool exam and Pneumonia on Chest X-Ray. Further review of the history revealed contamination of a previous non-healing wound of the right foot with soil during farming. He was started on IV Linezolid and Ceftazidime; and underwent CT guided pigtail insertion of Gluteal and Splenic abscesses. Cultures revealed *Burkholderia pseudomallei* and antibiotics were shifted to Meropenem. Recto-gluteal fistula was confirmed through Flexible Sigmoidectomy, noted 10 cm from the anal verge (Fig. 3).

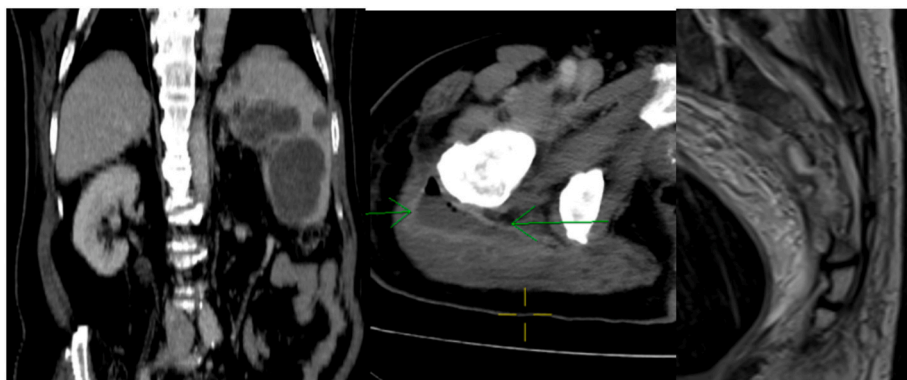
During the course of admission, repeat imaging (Fig. 4) showed almost complete resolution of gluteal abscess but no significant change in the splenic abscess. He then underwent Splenectomy (Fig. 5) and Sigmoid Loop Colostomy and was discharged 2 weeks post operatively. The procedures were performed by a senior attending surgeon. Vaccination for Hib, Pneumococcal, Meningococcal and Flu were given prior to discharge and antibiotics were continued for 6 weeks after discharge.

## 3. Discussion

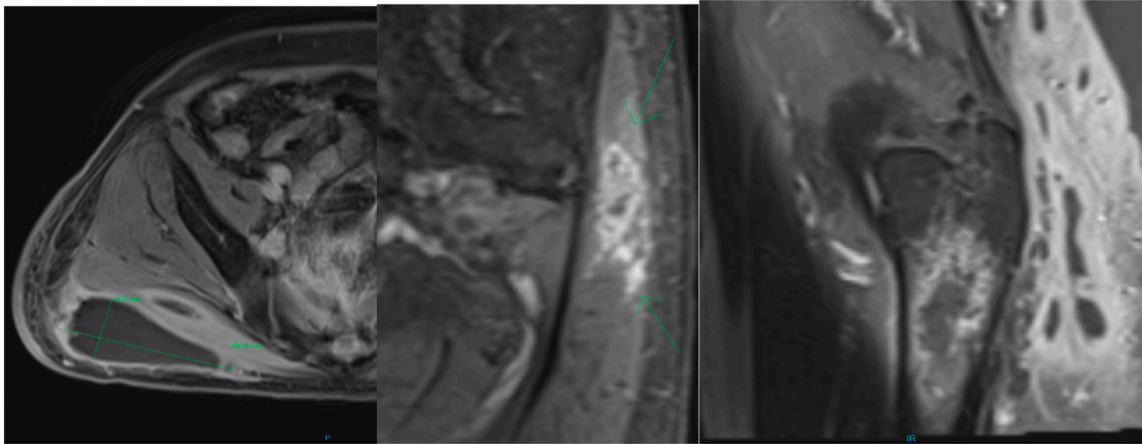
Melioidosis varies in its presentation and thus management should be individualized, depending on the organs involved. Our patient presented with multiple foci of infection which rendered the treatment more complicated as compared to those reported previously in published literature. The pneumonia and the osteomyelitis were managed with aggressive systemic antibiotics but the other sites of infection required drainage and surgery.

Multiple nodular abscesses in the spleen are usually managed with systemic antimicrobial treatment. Splenic abscesses inadequately responding to antibiotics may require a drainage procedure. For cases which fail to resolve with antibiotic therapy and percutaneous drainage, splenectomy may be necessary. Some authors even advocate outright splenectomy for patients with large and multiple splenic abscess collections [3]. Surgical approach may either be laparoscopic or open, depending on the patient's other underlying conditions, functional status and the characteristics of the pathologic spleen. Laparoscopic approach is recommended over the open approach as it has better postoperative pain control, shorter hospital stay and better cosmetic outcome. The only absolute contraindication to the laparoscopic approach is inability to tolerate general anesthesia and insufflation. In this patient, it is of particular concern since he manifested with pneumonia with pleural effusion along with the septicemia from several sources. It was therefore deemed appropriate that an open surgical approach be used to take out the spleen and perform the colonic diversion. Post-splenectomy, patients are required to receive vaccinations from encapsulated bacteria, namely Pneumococcal, Meningococcal and *Haemophilus influenzae* type b vaccines within 1–2 weeks post-operatively to prevent Overwhelming Post-splenectomy Infections (OPSI), which are commonly caused by encapsulated bacteria. Monitoring for infection post-operatively should still be done. It is expected that the patient will have leukocytosis and thrombocytosis, for which anticoagulation is necessary. Leukocytosis more than 20,000 and thrombocytosis more than 1,000,000 count should warrant further investigation. The required immunization protocol was implemented for this patient.

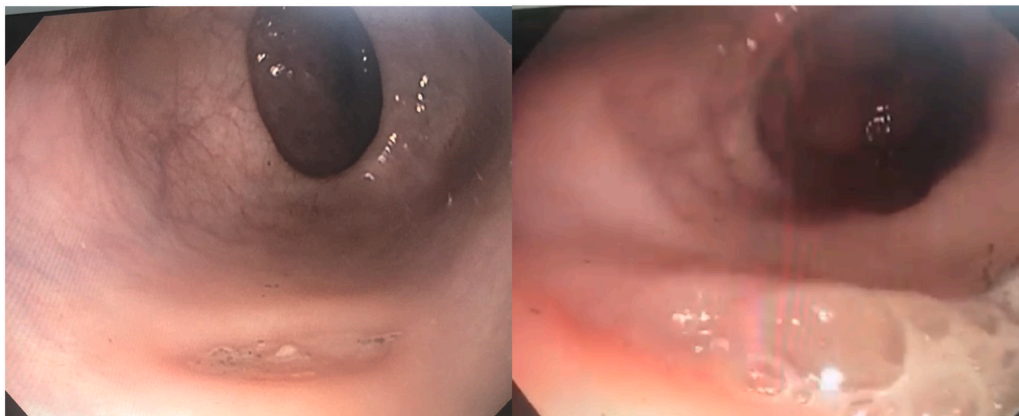
A recto-gluteal fistula was documented in this patient with an MRI of the lower abdomen and directly visualized by flexible sigmoidoscopy. Management of intestinal fistulas depends on patient specific factors. Whether early surgery is required or delayed after 4–8 weeks of maximal medical management likewise depends on patient factors and response



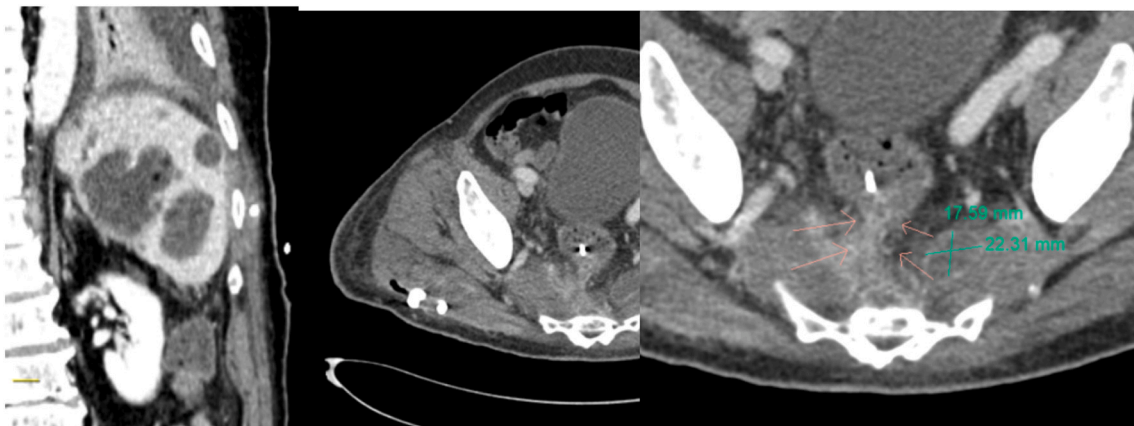
**Fig. 1.** CT scan of the whole abdomen with IV contrast. (a) Enlarged spleen measuring  $146 \times 76 \times 100$  mm, slightly lobulated contour, several varicose non-enhancing hypodense nodular lesions of fluid attenuations with largest measuring  $89 \times 68$  mm. (b) Right gluteal region showing inhomogeneous, hypodense foci with irregular margins, thin enhancing walls and tiny mural air loculations seen infiltrating the muscular soft tissue aspects, measuring approximately  $108 \times 37 \times 75$  mm and in the right hip joint measuring  $62 \times 23 \times 71$  mm.



**Fig. 2.** (a) Peripherally-enhancing, thick-walled fluid collections with restricted diffusion signals are noted in the right gluteal region, the largest component shows few small air locules and measures  $34 \times 105 \times 93$  mm. (b) Smaller multiloculated peripherally-enhancing collection with marrow signal abnormality and cortical destruction involving the sacrum extending to the presacral area and sacral canal associated with surrounding inflammatory change. Intramedullary rim-enhancing collection with surrounding marrow edema demonstrating restricted diffusion in the right proximal femur.

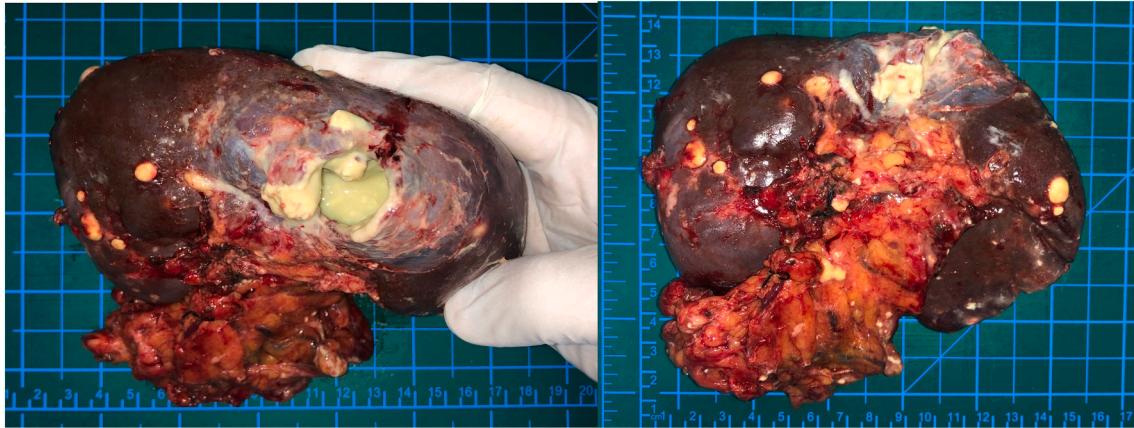


**Fig. 3.** Flexible sigmoidoscopy. 1 cm rectal mucosal opening, 10 cm from the anal verge with bubbling noted after injecting hydrogen peroxide through the gluteal pigtail tube.



**Fig. 4.** Post CT-guided drainage. (a) Multiple rim-enhancing fluid collections in the spleen are noted. Pigtail catheters and small air locules are noted in the two dominant lesions measuring  $60 \text{ mm} \times 75 \text{ mm} \times 63 \text{ mm}$  (APxWxCC) and  $40 \text{ mm} \times 42 \text{ mm} \times 52 \text{ mm}$ . (b) Cortical destruction involving the sacrum and S2-S4 segments with peritoneal inflammatory changes are again noted. (c) Edema in the right gluteus maximus muscles with small fluid collection within. Subcutaneous edema and cutaneous thickening involving the proximal aspect of the right hip and proximal thigh, not significantly change.





**Fig. 5.** Intra-operative findings. Multiple pockets of splenic abscess containing thick non foul smelling purulent material. No spillage of abscess intra-abdominally.

to treatment [6]. Maximal medical management includes drainage of the gluteal abscess and bowel rest to decrease flow to the fistula with the goal of spontaneous closure. In our patient early surgery was deemed warranted to aggressively address the sepsis and in conjunction with the required splenectomy. A diverting Sigmoid Loop Colostomy was performed for fecal diversion with plans for future reversal after resolution of the abscess and the septicemia. A repeat colonoscopy is recommended 6–8 weeks after for surveillance of the rectal fistula.

Osteomyelitis is treated with long term antibiotics, usually for 4–6 months. Our patient with osteomyelitis of the sacrum and proximal femur is classified as Cierny and Mader Class 4Bs (Diffuse, Systemic) [7]. The Orthopedic service deferred the usual surgical management for similarly classified infections, which include debridement and application of antibiotic beads, because of the inaccessible location and other comorbidities. This patient received IV antibiotics during the 1 month admission and was discharged with oral antibiotics to continue for 6 more weeks. Monitoring will include follow-up MRI study to show response to antibiotic regimen.

The patient was discharged subjectively improved. He and his family expressed gratitude for the care he received from the multidisciplinary team.

#### 4. Conclusions and recommendations

Melioidosis is a rare disease caused by *Burkholderia pseudomallei*, which is common in the soils of tropical islands, mostly in Southeast Asia. It's presentation is diverse, management depends on the severity of the spread of bacteria and prognosis is generally good. Culture is the gold standard of treatment and work-up should include investigation for other areas affected. A high index of suspicion is needed to catch the disease early as it may mimic Tuberculosis, especially because Tuberculosis is endemic in the Philippines. Proper documentation and reporting should be done to develop a more in-depth study and to develop management guidelines. Surgical approach to management of Melioidosis is offered when medical management is maximized and exhausted. In this case being presented, the combination of culture guided antibiotic administration and judicious surgical intervention successfully addressed the various complications of Melioidosis affecting several organ systems.

#### Ethical approval

Ethics approval obtained from the Makati Medical Center Ethics Review Board.

#### Funding

No external funding.

#### CRedit authorship contribution statement

Anthony R. Perez, MD: Study concept, writing the paper, final draft.  
Nour Aburayyan, MD: Data collection, review of literature, writing the paper.

Manuel Ramon Sto. Domingo, MD: Data collection, study design, manuscript editing.

Mark Onglao, MD: Data collection, review of literature, final draft.

#### Guarantor

Anthony R. Perez.

#### Registration of research studies

Not applicable.

#### Provenance and peer review

Not commissioned, externally peer-reviewed.

#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Declaration of competing interest

There were no conflicts of interest.

Sources of funding, institutional review board and ethical committee approval were not required.

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