

## Case Report

# A distinctive case of gastrosplenic fistula: comprehending melioidosis

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## ABSTRACT

This case report delves into an exceedingly rare case of gastrosplenic fistula and concurrent upper GI bleeding in the setting of melioidosis. Although upper GI bleeding is often encountered in the clinical setting, the co-existence of a gastrosplenic fistula adds an unprecedented amount of complexity. Melioidosis is an infectious disease endemic to certain geographic regions of the world, namely that of Southeast Asia and Northern Australia. *Burkholderia pseudomallei*, the culprit organism, is associated with bioterrorism on account of which it has gained notoriety in recent years. This case underscores the array of diagnostic challenges that were combated and the prompt surgical intervention in the form of a splenectomy and partial gastrectomy that culminated in a favorable outcome. Coordination in a temporal fashion and multidisciplinary collaboration between surgeons and physicians was imperative to success. This report emphasizes the significance of recognizing and addressing the atypical manifestations of the disease as it challenges the conventional understanding of melioidosis and offers significant insights into the realm of infectious disease, at large.

**Keywords:** *Burkholderia pseudomallei*, Gastro splenic fistula, Contrast enhanced computer tomography, Oesophago gastro duodenoscopy

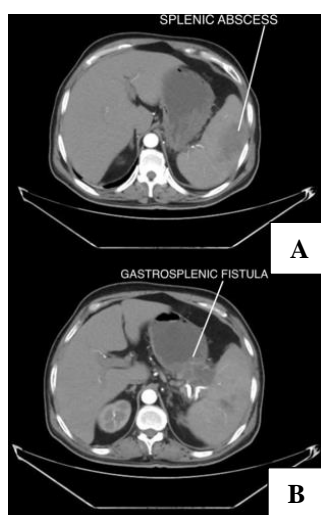
## INTRODUCTION

Melioidosis is an endemic disease in South Asia and is often under-reported in India.<sup>1</sup> It has a wide and complex clinical spectrum, which is often missed or diagnosed late. Splenic abscess related to melioidosis is one of the rare presentations. Moreover, perforating or eroding into the stomach is even rarer.<sup>2,3</sup> This provides the groundwork for the discussion of an intriguing and exceedingly rare case of a gastro-splenic fistula in a patient diagnosed with melioidosis. Understanding this clinical scenario is ever so important, as it poses challenges to the conventional understanding of melioidosis and its possible complications. Emphasis is laid on timely surgical intervention, which proves to be a life-saving measure.

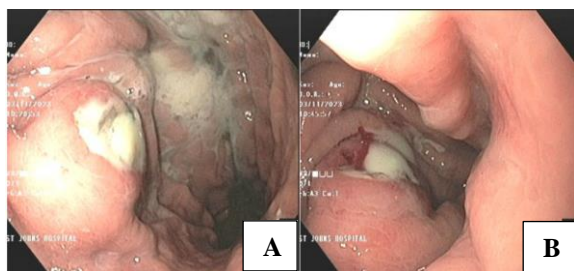
## CASE REPORT

A 54-year-old male who has been a known type II diabetes mellitus for 10 years. Presented to our hospital with 2 months of low-grade, intermittent fever. He had associated non-quantifiable weight loss, generalized weakness, fatigue, loss of appetite, tiredness, and occasional black-colored stools. The patient was evaluated for the next five days, and he also received blood transfusions to optimize his hemoglobin and vitals. Relevant initial investigations are as follows. Hb- 5.5 g/dl, total count- 6640/mm<sup>3</sup>, serum creatinine- 1.67 mg/dl, prothrombin time- 14.2 (12) sec, APTT- 24.4 (30) second, liver function test were normal, HIV (rapid): negative, hepatitis surface Ag: negative, blood culture:

*Burkholderia spp* identified as *Burkholderia pseudomallei*. Ultrasonography of abdomen: spleen of 12.7×6.2 cm with heterogeneous echotexture pole region shows multiple ill-defined confluent hypoechoic areas of volume 20 cc and multiple peripancreatic lymph nodes. CECT showed a splenic intraparenchymal abscess and multiple enlarged lymph nodes at the perigastric and splenic hilar region (Figure 1). These nodes form a necrotic conglomerate having lost the fat planes with the adjacent stomach. upper GI endoscopy revealed grossly oedematous fundal mucosa with a necrotic area discharging copious amounts of pus and associated fresh bleeding, possibly a fistulous communication of the splenic abscess (Figure 2).



**Figure 1 (A and B): Splenic abscess and gastro splenic fistula CECT.**



**Figure 2 (A and B): OGD image showing (A) the GSF; and (B) pus drainage with active bleeding in the GSF site.**

With the above information and a significant Hb drop, it was decided with good rapport and counselling relatives to perform the patient to an emergency exploratory laparotomy with high-risk consent. The patient had an inflammatory phlegmon on the table involving the splenic hilum, the lymph nodes at the hilum, and the stomach, and anticipating the possible splenic abscess communicating with the stomach. It was concluded with performing a partial gastrectomy with splenectomy. Thus removing the whole phlegmon in toto (Figure 3 and 4).

Later, the specimen revealed a communication between the splenic abscess and the stomach. The pus and necrotic lymph nodes sent for culture grew *Burkholderia spp* again. The patient was stable in the post-operative period. He was on injection meropenem 1 g (25 mg/kg) every 8th hourly in view of persistent bacteraemia which was started preoperatively escalated and was discharged on day 7 after surgery with oral eradication therapy of Trimethoprim/Sulfamethoxazole 160/180 mg every 12th hourly for a minimum of 12 weeks postoperatively. However, the patient responded well to surgical as well as medical management.



**Figure 3: On table finding of GSF with pus cavity.**



**Figure 4: On table finding of the fundus of the stomach with the perforated region.**

## DISCUSSION

Melioidosis, also known as Whitmore's disease is an emerging infectious disease in India as it is evident from case reports presented from different parts of India. Melioidosis is endemic across tropical areas, especially in South East Asia and Northern Australia, and is estimated to account for ~89,000 deaths per year worldwide.<sup>2</sup> It is a severe systemic infectious disease caused by the saprophyte *Burkholderia pseudomallei*, gram-negative environmental bacterium commonly found in the soil, groundwater, rice paddies, and ponds throughout endemic regions. The disease is acquired through contact with contaminated soil or water by percutaneous inoculation, aerosol inhalation, or the ingestion of contaminated water or food. The case fatality rate ranges from 19% to 36% in endemic areas.<sup>4</sup> Patients with diabetes mellitus (DM), chronic alcohol use, advanced kidney disease, underwent immunosuppressant therapy and some forms of underlying lung diseases are prone to melioidosis.

The clinical spectrum of melioidosis is complex, with widely-ranging clinical manifestations, varying from asymptomatic infection to localized abscess formation to fulminating disease with multiple organ involvement and even death. Splenic abscess is a rare presentation, even in endemic areas, with only a few cases reported.<sup>3</sup>

The clinical presentation of the disease is quite varied from asymptomatic infection to localized abscess, including pneumonia, ulcerative skin lesions with regional lymphadenopathy, bone and joint involvement, chronic suppurative lesions, and disseminated disease.<sup>5</sup> Pneumonia is the most common primary clinical presentation and splenic abscess. It has variably been reported from one-quarter to as high as three-fourths of patients with melioidosis in different series.<sup>6,7</sup>

Splenic abscess, which was once considered as an uncommon entity, is recently being diagnosed more often due to the widespread use of various imaging modalities in routine practice, and the increasing survival of immunocompromised patients. Splenic abscess is commonly associated with abdominal trauma, intravenous drug abuse and hemoglobinopathies, like sickle cell anaemia. A splenic abscess may also develop following the local spread of infection from the surrounding structures (pancreas, colon, left kidney, pelvic organs), or from distant metastatic sources (like infective endocarditis).

Organisms commonly cultured from these abscesses are aerobic gram-positive (Staphylococci, Streptococci) and gram-negative (Salmonella, *Escherichia coli* and *Klebsiella pneumoniae*). Melioidosis is an important etiology of splenic abscess in endemic areas. The splenic abscess has variably been reported from one-quarter to as high as three-fourths of patients with melioidosis in different series.<sup>6</sup> Melioidosis is an important aetiology of splenic abscess in endemic areas, contributing to about 19% of such cases in some series.<sup>3,6</sup>

Gastrosplenic fistula (GSF) is an unusual complication of benign as well as malignant gastric and splenic pathologies. This pathology acquires an important clinical significance due to its rare association with life-threatening upper gastrointestinal haemorrhage. The commonest causes of GSF in the literature are lymphomas from the spleen and gastric malignancies. Isolated cases of GSF have been reported due to benign aetiologies like peptic ulcers, Crohn's disease, trauma, splenic abscess, and splenic tuberculosis.<sup>8,9</sup> GSF due to benign conditions by itself is very rare less than ten cases have been reported. The pathogenesis mechanism involved progressive infiltration, erosion, and finally penetration of the gastric wall by the infective organism in the spleen.<sup>9</sup>

In our case, we encountered a rare case of upper GI bleeding secondary to GSF due to melioidosis of the spleen. The co-existence of a gastrosplenic fistula adds an unprecedented amount of complexity, particularly in the

setting of melioidosis. Gastro splenic fistula associated with splenic abscess due to melioidosis is extremely uncommon.

GSF is a serious complication that can arise from various gastric and splenic diseases, often requiring prompt embolization or surgical intervention in cases of significant bleeding.<sup>10</sup> Most commonly, GSF occurs as a result of tumour progression in splenic B-cell lymphoma or chemotherapy for advanced-stage disease. However, the development of GSF in patients with benign splenic diseases is a very rare occurrence, typically happening spontaneously. GSF often does not present with severe haemorrhage, and short-term outcomes are generally favourable, with a survival rate of approximately 82%. However, massive bleeding is a severe complication of GSF often associated with significant mortality.<sup>10,11</sup>

The treatment approach should consider the underlying cause of GSF. However, it can be challenging to determine whether GSF is a result of silent gastric perforation with a secondary splenic abscess or a primary splenic abscess leading to the formation of a communication with the stomach. Clinical presentation of gastrosplenic fistula consists mainly of symptoms of the underlying disease. The only symptom that can directly be attributed to the development of a gastrosplenic fistula and also constitutes the most threatening complication is the abrupt development of massive upper GI bleeding.<sup>12</sup> The bleeding is usually the result of the erosion of splenic vessels by gastric juice that might reflux into the splenic pulp. Conservative treatment with percutaneous drainage and antibiotic therapy is often inadequate for GSF patients with large splenic abscesses. In such cases, the most common treatment is splenectomy with partial gastrectomy, aiming to prevent major complications, including massive bleeding.<sup>8</sup> The only other case reported of gastro splenic fistula secondary to splenic abscess due to melioidosis is by Mayatapa et al 'Gastrointestinal: Splenic abscesses-related gastrosplenic fistula: unusual complication of melioidosis'.<sup>11</sup>

## CONCLUSION

The co-existence of melioidosis and gastro splenic fistula pushes the envelope in terms of our surgical and medical grasp of the disease. Through a meticulous examination of the clinical details and review of existing literature, this report sheds light on the management of this exceedingly rare clinical scenario. It emphasizes a multidisciplinary approach and prompt surgical intervention as a pivotal intervention. A thorough understanding of the case and its management promises to enrich the approach towards melioidosis as well as infectious diseases as a whole in the coming years.

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