Lesser Sac Haematoma-Melioidosis - A Surgical Surprise

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INTRODUCTION

Melioidosis, a potentially fatal disease endemic in South East Asia and Northern Australia is caused by *Burkholderia pseudomallei*, a potential bioterror agent. It is a motile, aerobic non-spore forming gram negative bacillus often characterised by pneumonia and multiple abscesses, but it can also present as septic arthritis, cutaneous ulcer and osteomyelitis. Modes of acquisition are inhalation, inoculation and rarely ingestion from a contaminated environment.[1] General and gastro surgeons rarely come across abdominal melioidosis and rare is a lesser sac haematoma secondary to mycotic aneurysm of splenic artery caused by melioidosis.

Clinical manifestations can vary from asymptomatic infections to localised abscesses to fulminating diseases with multiorgan involvement and eventual death. Due to evolving lifestyle, extensive travel and climate changes the disease which was previously confined to specific countries has crossed its boundaries. Increase in cases of comorbid conditions like diabetes and immunocompromised states have added on to the cause of increasing rates of the disease worldwide. India has seen isolated case reports from few states. Most often *Burkholderia pseudomallei* is misreported as pseudomonas species especially in resource-poor laboratories making the disease potentially fatal due to error in the treatment protocol.[2] Due to its high chance of recurrence, prolonged treatment with combinations of antibiotics is required for complete eradication.

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PRESENTATION OF CASE

Here we present a case of a 40-year-old male patient, with coronary artery disease (CAD) and diabetes mellitus as comorbidities, from Thrissur district of Kerala. The patient is an expatriate who worked as a chef and butcher with frequent travel across Kerala. Primary clinical presentation was fever with evening rise of temperature associated with multiple episodes of vomiting for 2 weeks. Patient was diagnosed with *Burkholderia pseudomallei* osteomyelitis of ileal bone, ischium and acetabulum 2 years back, which was subsequently treated with intravenous (IV) antibiotics for 2 weeks.

Clinical examination revealed Karnofsky performance score of 100 with stable vitals. On examination of abdomen, tenderness was noted over left hypochondrium. Other systems were within normal limits. Relevant biochemical investigations revealed elevated erythrocyte sedimentation rate (ESR) (104 mm / hr.), anaemia (11.6 g / dl) and hypoalbuminaemia (2.8 g) suggestive of chronic inflammatory condition. Primary imaging with ultrasound of abdomen showed hepatomegaly, moderate splenomegaly with two large heterogeneous lesions in splenic parenchyma, possibility of splenic masses (both 5 cm x 8 cm) and hence followed by contrast enhanced computed tomography (CECT), which showed splenomegaly with multiple abscess (3.6 to 6 cm) extending to sub-capsular plane and closely abutting lateral chest wall, similar abscess was observed in right lobe of liver (14 mm x 12 mm & 11 mm x 7 mm), large intramural haematoma involving posterior wall of stomach (8.6 cm x 9.5 cm x 10.2 cm) extending superiorly & reaching the under surface of left hemi diaphragm probably due to mycotic aneurysm leak, few enlarged upper para aortic lymph nodes.

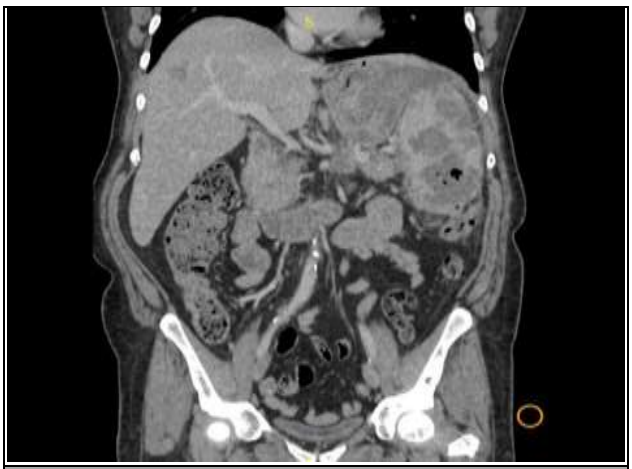

Figure 1. CECT Abdomen Showing Multiple Splenic Abscesses

Figure 2. Lesser Sac Haematoma

Figure 3. Haematoma Removal

Computed tomography guided aspiration of the abscess was done, and pus was sent for culture which confirmed the growth of *Burkholderia pseudomallei*. Blood culture was also sent which came to be sterile. Patient was managed with injection ceftazidime 1 gm intravenous (IV) thrice daily for 2 weeks. Patient became symptomatically better. Review scan showed 41 cc splenic abscess in lower pole and upper pole, a small 5 cc abscess between two, contents in all abscesses partly liquefied and stomach wall lesions were persisting. The patient was discharged with tablet sulphamethoxazole + trimethoprim twice daily for 2 weeks. Stomach wall haematoma was left intact in view of non-expanding nature and benign pathology.

Patient was reviewed 2 weeks later with no fresh complaints and review CECT scan showed no significant changes. Hence, oral antibiotics were continued after discussion with microbiology team.

On his review after 4 weeks, patient complained of generalised tiredness and blood-stained vomiting. On abdominal examination, he had diffuse tenderness with no associated guarding or rigidity. Laboratory investigations showed elevated total count (23,520) and significant drop in haemoglobin (7.1 g / dl). Patient was taken up for exploratory laparotomy on emergency basis in view of probable intragastric rupture of haematoma. Intraoperative findings included large lesser sac haematoma arising from the mycotic aneurysm from splenic artery compressing posterior wall of stomach, causing a small erosion, multiple abscess within splenic parenchyma with large ones located in poles, with dense adhesion to colon, omentum and kidney, liver palpably normal with no ascites / deposits. The haematoma was evacuated, and posterior gastric wall erosion sutured, with adhesiolysis & splenectomy and ligation of splenic artery was done proximal to aneurysm.

Since patient was asymptomatic after surgery, endoscopy was deferred. Postoperatively, he was managed with IV meropenem and was discharged on post-operative day 10 with oral cotrimoxazole. Now patient is on regular follow up since past 12 months and is till now asymptomatic.

CLINICAL DIAGNOSIS

Intra-abdominal abscess.

DIFFERENTIAL DIAGNOSIS

Intra-abdominal abscess due to

- *Mycobacterium tuberculosis*.
- Pseudomonas.
- *Staphylococcus aureus*.
- Enterobacteriaceae.
- *Entamoeba histolytica*.

PATHOLOGICAL DISCUSSION

Melioidosis can present as asymptomatic infection, or with severe clinical manifestations in immunocompromised patients.[3] Infection mainly affects liver, lungs, skin and subcutaneous tissue, but can also rarely affect cardiovascular system, central nervous system and lymph nodes.[4] Mycotic aneurysm is one of the rarest presentations of melioidosis. Systemic involvement can manifest as pneumonia, encephalomyelitis, acute suppurative parotitis and rapidly progressing skin and soft tissue infections.[5]

According to a 20-year prospective study of 540 cases of melioidosis in Northern Australia to understand the various clinical presentations and relation of the disease with various risk factors, 55 % of the patients were bacteraemic, but mycotic aneurysm was only found in 2 of the cases.[6]

Isolation of *B. pseudomallei* from clinical specimens using Ashdown's medium containing crystal violet and gentamicin as selecting agents is still considered as gold standard for diagnosis of melioidosis. Since isolation of the agent is time consuming, preliminary serological tests are performed in endemic regions to initiate treatment.[7]

Pulmonary melioidosis may present as diffuse nodular infiltrates throughout lung fields, with cavitation in acute cases or as mixed nodules or patchy opacities with slower progression as in subacute and chronic cases.[8] Thin walled cavities which rarely contain air fluid level, sparing of lung apex, less occurrence of fibrosis and rare presentation of

mediastinal and hilar lymphadenopathy differentiates it from tuberculosis.

Lesions in the periphery of the prostate with normal or mildly raised prostate specific antigen (PSA) points towards the infection in a patient with disseminated disease with lower urinary tract symptoms when compared to other bacterial prostatic infections with higher levels of PSA is the highlight.[9] *B. pseudomallei* is susceptible to cotrimoxazole and doxycycline and resistant to aminoglycosides and polymyxin B[2].

Treatment of melioidosis comprises of 2 phases. Acute phase, in which parenteral antibiotics like carbapenems and ceftazidime are given for 10 days followed by eradication phase with oral cotrimoxazole to for 20 weeks, which is mandatory due to high chances of recurrence.[2,3] With regard to mycotic aneurysm, surgical replacement with deep vein as a vascular conduit (neoaortoiliac system bypass) has proven to be a definitive management with chance of postoperative complications like wound infections (29 %), post-operative sepsis (18 %), renal impairment (18 %) and vascular graft infection or leakage (18 %).[6]

Surgeons commonly encounter melioidosis from a splenic abscess perspective. Successful treatment depends on correct identification of the organism for which a high index of suspicion is required with an equipped microbiological team to customize the antibiotic regimen for longer duration as incomplete treatment usually results in recurrence.

A lesser sac haematoma secondary to splenic artery aneurysm due to melioidosis is indeed very rarer and hence the description of the case was deemed necessary to the surgeons operating in endemic areas.

FINAL DIAGNOSIS

Multiple splenic abscesses, hepatic abscess, lesser sac haematoma with sepsis secondary to *Burkholderia pseudomallei* infection.

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