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Case Report

A rare case of meliodosis presenting as lupus panniculitis with intermittent fever

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Introduction

Melioidosis is a pyogenic infection caused by the bacterium *Burkholderia pseudomallei* and is a soil-associated infection [1]. It is acquired by inoculation or inhalation and is more common in patients with underlying chronic disease. It can cause a range of symptoms including fever, chills, cough, chest pain and skin lesions and can be fatal if left untreated. The infection is typically treated with a combination of antibiotics.

Case report

A 37-year-old, previously apparently well teacher from Kekirawa presented with on and off, high-grade, intermittent fever of two months duration with a history of retropharyngeal abscess one month back. She had undergone abscess drainage and was on oral antibiotics for two weeks. She presented with a painful, non-pruritic skin rash on the left thigh along with fever. She denied any travel history and there was no contact history with soil. She had no contact or past history of tuberculosis.

There was a cattle farm near to her residence. On examination, the patient was afebrile, not icteric or pale with no lymphadenopathy. She had a skin rash over the left side of the thigh which was a papular lesion with panniculitis as shown in Figure 1.

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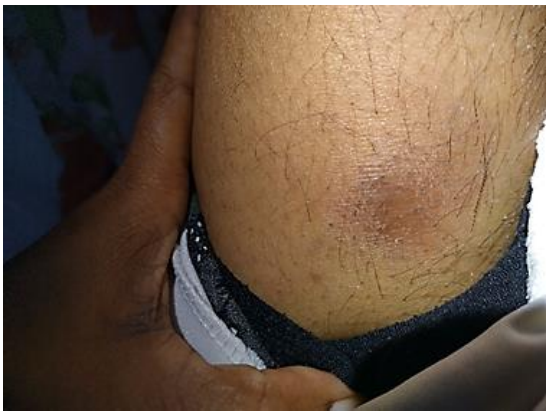

Figure 1: Skin rash over the left side of the thigh

There was a healed ulcer on the left side inner arm as well. Abdominal examination revealed mild splenomegaly. Rest of the systems examination was unremarkable. Her investigations are shown below (Table 1).

Table 1: Investigations

Investigations	**Normal ranges**	**Results (in consecutive days)**		
Full blood count	White blood cells-(4.1-11.2)*10^3	10.2*10^3 /ml		
	Neutrophils–(39.9-73.9)*10*3	62%		
	Hemoglobin-(11.7-15.5)g/dl	9.2g/dl		
	Platelets-(159-388)*10*3	650*10^3/ml		
C-reactive Protein(mg/L)	<10	24	1.6	
ESR(mm/1st hour)	0-25	96	107	53
Serum electrolytes(mEQ/L)	Sodium : 136-145	134		
	Potassium: 3.5-5.5	4.3		
Urine full report		Normal		
Serum creatinine(µmole/L)	53-97	61		
Serum calcium(mmol/ L)	2.1-2.55	2.68	2.35	
Antinuclear antibody		1:80 nuclear pattern		
Complement	C3-(90-207)	C3-196		
	C4-(17-52)	C4-33		
LDH(u/L)	230-460 S	270		
manteaux		Negative		
Sputum TB genexpert		Negative		
Sputum acid fast bacilli		Negative		
Liver biopsy TB genexpert		Negative		
HIV		Negative		
Meliodosis antibody		1:5120 highly positive		
APTT	21-35(seconds)	28		
PT/INR	0.8-1.2	0.93		
Alanine aminotransferase(u/L)	7-40	64		

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Aspartate aminotransferase(u/L)	13-31	71		
Alkaline phosphate(u/L)	53-128	78		
Gamma glutamyl transferase(u/L)	15-30	150		
Total bilirubin (µmole/ L)	(3-21	10.3		
Indirect bilirubin (µmole/ L)	3.4-12	5.7		
Total protein (g/d L)	6.4-8.3	8.1		
Serum albumin (g/dl)	3.2-5.4	3.7		
Serum globulin (g/d L)	2-3.5	4.4		
Random blood sugar (mg/d L)	74-140	112		
Culture reports		Blood- no growth		
		Urine - no growth		
		Sputum - no growth		
		Wound swab – *Acinetobacter* positive		

Her chest X-ray revealed a left sided, middle zone cavitatory lesion (Figure 2). The ultra sound scan of the abdomen showed multiple hypo echoic lesions in the liver and spleen. Her CECT chest, abdomen and pelvis also revealed possible multiple abscesses or metastasis.

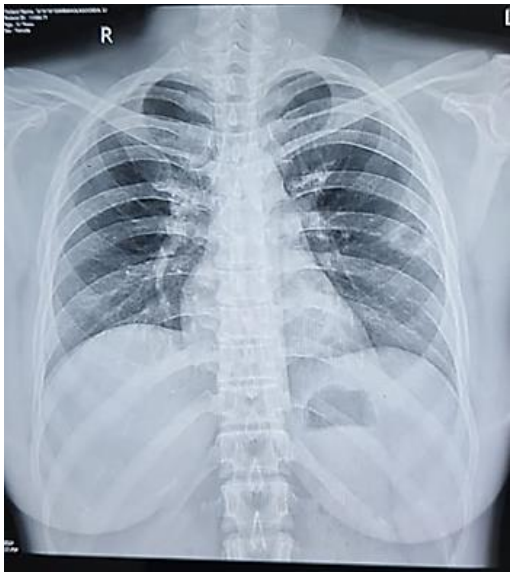

Figure 2: Chest X-ray

We went ahead with a biopsy from the liver for TB PCR and TB culture which came as negative. Liver histology came as nonspecific changes of the liver parenchyma showing moderate chronic inflammation in the portal tracks and lobular disarray which may represent liver parenchyma adjacent to a space occupying lesion such as an abscess or neoplasm. A serum sample for meliodosis antibodies became highly positive with a titre

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of 5120. Her skin lesion biopsy revealed an intact epidermis with acanthosis. Mid and deep derma blood vessels showed a perivascular lymphocytic cell infiltrate. The underlying subcutaneous tissue revealed lobular panniculitis with an infiltrate of lymphocytes. Plasma cells, neutrophils or apoptotic debris were not seen. There were no associated lichenoid changers or vasculitis. The overall features favored the appearances of lupus panniculitis in the correct clinical context. Her ANA titre was 1:80.

However, we started treatment with IV meropenem 1g eight hourly and she responded markedly to treatment. Her ESR gradually reduced to 60, serum calcium to 2.3 and she became fever free and her skin lesion dramatically improved without surgical intervention (Figures 3-6). Her repeat USS abdomen and pelvis also came as normal with no hypoechoic lesions.

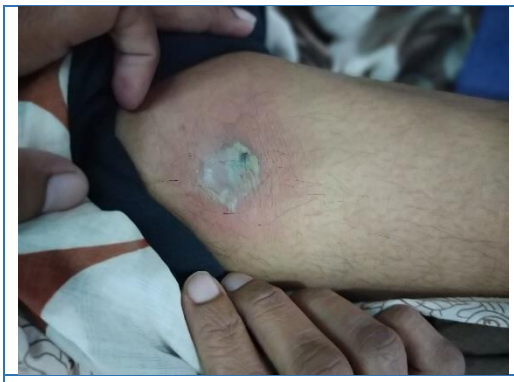
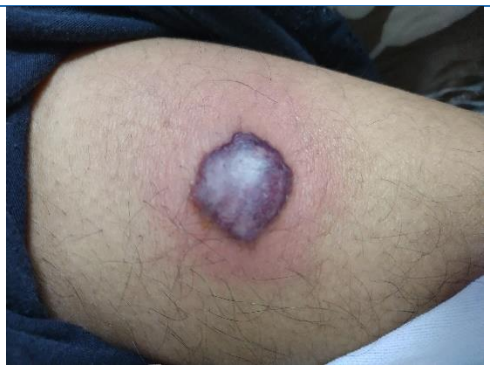


Figure 3:



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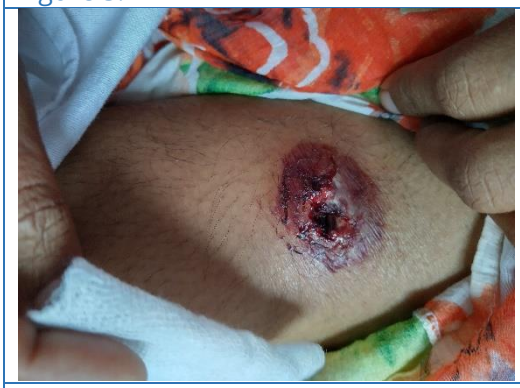

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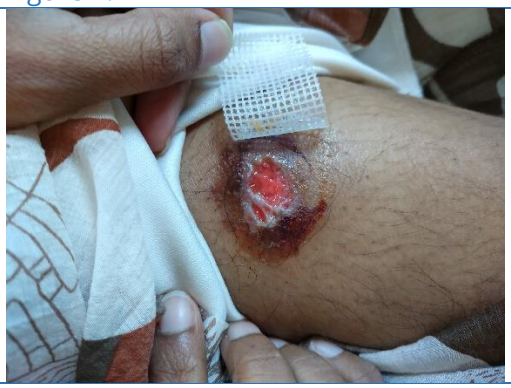

Figure 6:

Figures 3-6: Progressive evolution of the lesion.

Discussion

Although Sri Lanka lies in the endemic belt (south and south east Asia, particularly northern Australia and Thailand), meliodosis was considered as an emerging disease in Sri Lanka until recently, probably due to under-recognition of the disease [2,3].However, after national surveillance was instituted in 2008, melioidosis has become established as an endemic infection throughout Sri Lanka [3,4]. The number of cases have markedly increased recently.

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Meliodosis can present as a spectrum of disease [5]. The acute form of melioidosis typically presents with symptoms such as fever, cough, and chest pain progressing to severe pneumonia. In some cases, it can lead to septicemia which can be fatal if not treated promptly. Patients may present with fever with features of severe pneumonia or with multiple organ involvement manifesting as central nervous system abscesses, epidural abscesses, septic arthritis or localized suppurative infection. The most common form of melioidosis is acute pneumonia, which can range from a mild to severe respiratory illness [6]. The subacute form of melioidosis usually presents with milder symptoms and has a slower progression of the disease. Symptoms can include fever, fatigue, weight loss, and muscle and joint pain. Subacute melioidosis can also manifest as skin and soft tissue infections or as chronic infections such as abscesses, osteomyelitis, or prostatitis. In either form of the disease, prompt diagnosis and treatment with antibiotics is crucial for a favorable outcome. Chronic melioidosis refers to the long-term or persistent form of the disease, which can last for several months to years and may present with recurrent or persistent symptoms. Asymptomatic melioidosis refers to when an individual is infected with *Burkholderia pseudomallei*, but they do not experience any symptoms.

Our case was unique as it initially presented as fever with a skin rash and initial management was directed towards lupus panniculitis until ultrasound scan findings were available. Steroids were not initiated since there was a suspicion of tuberculosis. The patient was found to have a high serum calcium along with hypoechoic lesions on ultrasound scan suggestive of granulomatous diseases such as tuberculosis and sarcoidosis. Her ESR was also suggestive of an underlying chronic inflammatory disease or possible malignancy. Unfortunately, we did not have facilities to perform serum ACE levels or parathyroid hormone levels. We did a skin lesion biopsy which was suggestive of lupus panniculitis. However, she did not have any other features to suggest systemic inflammatory arthritis such as joint pain, any other typical skin lesions or renal or eye involvement. Her haematological investigations were also not suggestive of systemic lupus erythematosus. We had to go ahead with biopsy of the liver lesions to come into a definitive diagnosis after doing CECT chest abdomen and pelvis. Meanwhile we empirically started her on IV meropenem 1g 8 hourly and her clinical symptoms as well as serological investigations markedly improved with the treatment. As the patient was screened for pyrexia of unknown origin, we sent meliodosis serology and it came positive with high titre. However her blood culture was negative, probably because the patient had been treated with antibiotics previously [7]. To our knowledge, there is only one other case of melioidosis with high serum calcium levels reported [8].

Patients with melioidosis can get various types of skin lesions as well as skin abscesses[9]. There are cases reported with Sweet syndrome and erythema nodosum associated with melioidosis [10,11]. However, cases of melioidisis presenting as panniculitis, especially as lupus or lobular panniculitis, are very rare. Patients presenting with melioidosis usually have underlying immunodeficiency or diabetes mellitus. Our patient was an immunocompetent young person, emphasizing the point that we need to consider melioidosis as one of differential diagnoses in young healthy patients as well.

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