Melioidosis presenting as lymphadenitis: a case report

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Abstract

Background: Melioidosis is an infection caused by the facultative intracellular gram-negative bacterium; Burkholderia pseudomallei. It gives rise to protean clinical manifestations and has a varied prognosis. Although it was rare in Sri Lanka increasing numbers of cases are being reported with high morbidity and mortality. Here we report a case of melioidosis presenting with lymphadenitis which was diagnosed early and treated promptly with a good outcome.

Case presentation: A 53-year-old Sinhalese woman with diabetes presented with fever and left sided painful inguinal lymphadenitis for one month. She had undergone incision and drainage of a thigh abscess three months previously and had been treated with a short course of antibiotics. There was no record that abscess material was tested microbiologically. She had neutrophil leukocytosis and elevated inflammatory markers. Initial pus culture revealed a scanty growth of “Pseudomonas sp.” and Escherichia coli which were sensitive to ceftazidime and resistant to gentamicin. Due to the history of diabetes, recurrent abscess formation and the suggestive sensitivity pattern of the bacterial isolates, we actively investigated for melioidosis. The bacterial isolate was subsequently identified as B. pseudomallei by polymerase chain reaction and antibodies to melioidin antigen were found to be raised at a titre of 1:160. The patient was treated with high dose intravenous ceftazidime for four weeks followed by eradication therapy with cotrimoxazole and doxycycline. As the patient was intolerant to cotrimoxazole, the antibiotics were changed to a combination of co-amoxiclav and doxycycline and continued for 12 weeks. The patient was well after 6 months without any relapse.

Conclusions: Melioidosis is an emerging infection in South Asia. It may present with recurrent abscesses. Therefore it is very important to send pus for culture whenever an abscess is drained. However, it should be noted that the reporting laboratory may be unfamiliar with this bacterium and the isolate may be misidentified as Pseudomonas or even E. coli. Melioidosis should be suspected when an isolate with the typical antibiotic sensitivity pattern of ceftazidime sensitivity and gentamicin resistance is cultured, especially in a patient with diabetes. This will expedite diagnosis and prompt treatment leading to an excellent prognosis.

Keywords: Melioidosis, Lymphadenitis, Burkholderia pseudomallei, Sri Lanka

Background

Melioidosis is an infection caused by the facultative intracellular gram-negative bacterium; Burkholderia pseudomallei. Clinical features of melioidosis are highly variable. They range from asymptomatic disease, localized skin ulcers or abscesses and acute fulminant septicaemia to chronic infection. Predisposing factors for melioidosis include chronic diseases such as diabetes, chronic renal failure, chronic lung disease and alcoholism [1-3]. Infection spreads by percutaneous inoculation, inhalation, or ingestion. Melioidosis is endemic in tropical and subtropical zones of South East Asia and Northern Australia [1,2]. Although it is rare in Sri Lanka, increasing numbers of cases are being reported [4-10] and many of them were fatal [4-6,9]. Timely diagnosis and prompt institution of correct antibiotics will prevent high morbidity and mortality. Here we report a case of melioidosis, presenting as...
lymphadenitis, which was diagnosed reasonably early and
treated aggressively with a very good prognosis.

Case presentation
A 53-year-old Sinhalese woman from Chilaw in the North
Western Province of Sri Lanka, presented with intermit-
tent high fever with chills for one month and a painful left
inguinal mass for two weeks. She had diabetes.

She had undergone an incision and drainage of a left
depth thigh abscess three months previously and had been
-treated with a short course of antibiotics. There was no
record that abscess material was tested microbiologically.

On examination, she was febrile with the temperature of
103°F. She had a tender, fluctuant mass in the left inguinal
region. The rest of the examination was unremarkable.

There was a raised erythrocyte sedimentation rate (ESR)
of 80 mm in the first hour and a very high C-reactive
protein (CRP) level of 208 mg/dl. The white cell count
was 19 × 10^9/L with 75% neutrophils. Liver function and
renal function were normal. Ultrasound scan revealed a
left inguinal abscess. Excision biopsy of the abscess was
done and samples were sent for histology, bacterial culture
and antibiotic sensitivity testing, microscopy for acid fast
bacilli and culture for tuberculosis.

Histology revealed chronic lymphadenitis with perinodal
abscess formation. There were no acid fast bacilli in
the direct smear. Initial culture results showed a scanty
growth of Pseudomonas sp. and E. coli which was sensitive
to ceftazidime and treatment commenced according to
the sensitivity pattern.

However, since the Pseudomonas isolate was resistant
to gentamicin and the patient had a history of recurrent
abscess formation, melioidosis was considered in the
differential diagnosis and the bacterial isolates were sent
to the Department of Microbiology, Faculty of Medicine,
University of Colombo for identification.

Bacterial colonies on blood agar were pin point in size
after overnight incubation developing into 1-2 mm, white,
umbonate colonies after 48 hours. On MacConkey agar
the isolate was salmon pink, which may account for the
erroneous conclusion that the “Pseudomonas” isolate was
mixed with E.coli. The colonies had the characteristic
musty, earthy odour of B. pseudomallei and were slowly
oxidase positive. Gram stain appearance revealed the
typical safety pin appearance. The isolate was resistant
to gentamicin, polymyxin and colisitin and sensitive
to co-amoxycylav. Subcultures of the isolate were cour-
ered to a reference laboratory where they were confirmed
as B. pseudomallei by polymerase chain reaction (PCR).
Serum antibodies to melioidin antigen using an in-house
indirect haemagglutination (IHA) test based on that de-
scribed by Alexander et al. with antigen prepared from
local strains of B.pseudomallei [11] were positive at a titre
of 1:160.

After confirmation of the diagnosis of melioidosis, it
was decided to treat her with intravenous ceftazidime
for four weeks. There was excellent clinical improve-
ment with normalization of markers of inflammation.
Eradication therapy with oral co-trimoxazole and doxy-
cycline was started on the third week, overlapping with
the intravenous ceftazidime for one week. The patient
tolerated the medications well.

The patient was discharged after completion of four
weeks of intravenous ceftazidime with a plan to continue
oral antibiotics for 12 weeks. However she presented to
the outpatient follow up 2 weeks after discharge with an
itchy rash which was presumed to be an adverse effect of
cotrimoxazole. The rash improved after co-trimoxazol
was replaced by co-amoxycylav. The patient was able to
tolerate the co-amoxycylav/doxycycline combination and
completed 12 weeks of eradication therapy. At the time of
this writing she was well without evidence of recurrence
or relapse.

Conclusions
B. pseudomallei infections are known for their protean
manifestations, ranging from septicaemia and pneumonia
to asymptomatic infections, localised ulcers or abscesses.
Diabetes has been found to be the single most common
predisposing factor for melioidosis [2,3]. The disease
occurs mainly in rural areas and is associated with
occupational exposure to soil and surface water. In
the majority of cases it probably goes undiagnosed
and untreated.

Our patient had diabetes but she did not have any
known exposure to soil or water. There was no history
of travelling to a known endemic area suggesting that
she had acquired the disease in Sri Lanka and that the
disease is more prevalent than previously thought.

Ten cases [4-10] of melioidosis have been reported in
Sri Lanka; the first, in an English tea broker in 1927.
Most of them were fatal with only 4 survivors [7-10].
Delay in diagnosis may have been a factor contributing
to the high mortality.

In our patient, we actively suspected melioidosis on
the 4th day after admission, when pus culture revealed a
“pseudomonas species” resistant to gentamicin. The fact
that she was still unwell, three months after surgery and
antibiotics for a deep thigh abscess, strengthened our
suspicion. Rapid initiation of appropriate therapy would
have contributed to her good prognosis.

Isolation of B. pseudomallei in culture is essential to
confirm the initial clinical diagnosis. Our patient may
have been diagnosed three months previously if aspirated
pus had been sent for microbiological examination. It is
also important to note that, if the reporting laboratory
is unfamiliar with this bacterium, B. pseudomallei may be
reported as “pseudomonas species”. Therefore, if melioidosis
is suspected, further bacterial identification should be requested.

Our patient had only intravenous ceftazidime for four weeks in the acute phase. Combination of co-trimoxazole with ceftazidime is not recommended in the acute phase as there is no added benefit [12]. Oral co-trimoxazole and doxycycline was chosen for the eradication phase of antibiotics. They were overlapped with ceftazidime for one week as this is the vulnerable period where reactivation can occur [12].

This case illustrates the fact that a high degree of clinical suspicion is needed to diagnose melioidosis at an early stage. When culture of patient specimens yields a “pseudomonas species” which is resistant to gentamicin, melioidosis should be actively excluded, especially in a diabetic.

Use of a selective medium such as Ashdown’s agar and alerting the microbiological community to the characteristic microscopic and colony morphology and resistance pattern of B. pseudomallei may help to improve the detection rate of melioidosis in the country.

A long course of intravenous antibiotics in the acute phase, overlapped and followed by a prolonged course of a combination of oral antibiotics is needed to improve the prognosis of this potentially fatal, emerging infection.

Consent
Written informed consent was obtained from the patient for publication of this Case Report and any accompanying images. A copy of the written consent is available for review by the Editor—in-Chief of this journal.

Abbreviations
PCR: Polymerase chain reaction; WBC: White blood count; ESR: Erythrocyte sedimentation rate; CRP: C-reactive protein; IHA: Indirect haemagglutination test.

Competing interests
The authors declare that they have no competing interests.

Authors’ contributions
SW made the clinical diagnosis, managed the patient and supervised the manuscript drafting. TP drafted the manuscript, reviewed the literature and was involved in management of the patient. EMC carried out identification of the isolate and performed the IHA and supervised manuscript drafting. JPE carried out the initial microbiological diagnosis and was involved in the early phase of the management of the patient. All authors have read and approved the final manuscript.

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