CASE REPORT

Melioidosis presenting as septicaemia and facial nerve palsy

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SUMMARY

A 43-year-old farmer presented with acute onset pneumonia, septicaemia and peripheral facial nerve palsy (left side). *Burkholderia pseudomallei* was isolated from the blood culture of the patient. The patient was successfully treated with intravenous meropenem and oral cotrimoxazole for 2 weeks followed by maintenance therapy with cotrimoxazole. The case is reported to increase awareness among the clinicians and microbiologists regarding melioidosis.

BACKGROUND

Burkholderia pseudomallei is a soil-dwelling non-fermenting gram-negative bacilli, previously belonging to the family of *Pseudomonas* causing melioidosis. The estimated global incidence of melioidosis is 165 000 cases per year.¹ It is endemic in Southeast Asia and Northern Australia, but the regions of endemicity are expanding with cases being increasingly reported from South Asia, China and Taiwan.² Melioidosis presents with a variety of clinical manifestations usually resembling tuberculosis, malignancy or autoimmune disease and hence rightly named the 'great mimicker'. The usual presentations are pneumonia with or without septic shock, skin ulcers or abscess, septic arthritis or osteomyelitis, abscess in internal organs (spleen, kidney, prostate and liver). Other manifestations commonly seen in endemic areas are suppurative parotitis and neurological manifestations like encephalitis and cranial nerve palsies (seventh nerve being the most common).³ The infection is usually acquired by percutaneous inoculation (by trauma), inhalation, ingestion or aspiration (near drowning).

CASE PRESENTATION

Here we report a 43-year-old man who presented with complaints of fever, acute onset of breathlessness and yellowish discolouration of eyes and urine for a week. The patient was a paddy farmer by occupation; married with two children. There were no comorbidities. Physical examination revealed the patient to be icteric and febrile (temperature 100.4 °F (38 °C)) with tachycardia (pulse rate 135 beats/min). Examinations of the respiratory system, cardiovascular and gastrointestinal systems were unremarkable. The patient also had lower motor neuron facial palsy (Bell's palsy on the left side) of 3 days duration; further central

nervous system (CNS) examination was within normal limits.

INVESTIGATIONS

He was in respiratory distress (respiratory rate 42/min) and arterial blood gas (ABG) analysis showed evidence of a hypoxia (oxygen saturation of 68%) requiring oxygen support. His chest X-ray was normal but CT thorax showed multiple scattered nodular opacities. CT abdomen showed a small renal infarct (probably due to septic emboli), and MRI brain was normal. His blood investigations revealed derangement in haematological and biochemical parameters. Acute phase reactants (serum C-reactive protein 2.4 mg/dL and serum procalcitonin 70.53 ng/mL) were markedly elevated suggestive of sepsis. Total leucocyte count was 7900/mm³ and peripheral smear showed neutrophilia with shift to left. Liver function tests showed mild elevation of total bilirubin 4.59 mg/dL and direct bilirubin 3.05 mg/dL with marginally increased enzymes (serum glutamic-oxaloacetic transaminase 67 U/L, serum glutamic-pyruvic transaminase 43 U/L, alkaline phosphatase 169 U/L); renal function tests were normal.

Screening tests for febrile illnesses, including Widal test, tests for malarial parasite, HIV and autoimmune markers were negative.

Blood sample for automated culture (BACTEC 9120) on the day of admission showed the presence of gram-negative bacilli with bipolar staining (safety pin appearance). On blood agar, small cream coloured colonies with a hazy zone of haemolysis were observed which developed central umbonation on incubation for 72 hours and dry wrinkled colonies on continued incubation for 2 weeks (figure 1). On MacConkey agar, pink coloured colonies were observed due to the uptake of dye from the medium (figure 2).

The organism was found to be motile, oxidase, catalase-positive. It was a non-fermenter which utilized citrate, not pigmented, did not produce H₂S and indole, did not hydrolyse urea and was presumptively identified as *Pseudomonas* sp other than *Pseudomonas aeruginosa*. Since the antibiotic sensitivity pattern by disc diffusion showed resistance to aminoglycosides and polymyxin B, it was further put in VITEK GNI system (bioMérieux, France) for further identification. The organism was identified as *B.pseudomallei*, susceptible to ceftazidime, imipenem, meropenem and cotrimoxazole.

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Figure 1 Colonies on blood agar—medium sized (2–4 mm diameter) cream coloured colonies with a hazy zone of haemolysis, developing central umbonation.

DIFFERENTIAL DIAGNOSIS

Acute pneumonia with facial palsy can be found in *Mycoplasma pneumoniae* infection.

Sarcoidosis can also present with multiple nodules in CT thorax and facial nerve palsy.

TREATMENT

The patient was treated with intravenous meropenem 500 mg three times per day and oral cotrimoxazole (TMP 320 mg/SMX 1600 mg) two times per day for 2 weeks followed by maintenance therapy with cotrimoxazole (TMP 320 mg/SMX 1600 mg) two times per day for 3 months.

OUTCOME AND FOLLOW-UP

Following therapy, there was gradual reduction in the hypoxia. ABG returned to normal within a week. The duration of fever

after commencement of antibiotic was 10 days. The elevated bilirubin and liver enzymes returned to normal within 2 weeks of therapy. The two repeat blood cultures taken on day 5 of treatment showed no growth. The facial palsy showed improvement.

DISCUSSION

Pneumonia with septicaemia is the most common presentation of melioidosis. The mortality rates are high accounting to 90% in the absence of effective intervention and 50% after antibiotic therapy.⁴ The causative agent *Burkholderia pseudomallei* grows on ordinary lab media but can be dismissed as environmental contaminant in non-endemic areas. Confirmation of diagnosis is by culture of the organism from blood, pus samples, wound swabs, synovial fluid, cerebrospinal fluid, urine and sputum, depending on the site involved. Even though culture of the organism from clinical specimens is the 'gold standard'

Figure 2 Colonies on MacConkey agar—pink coloured colonies.

(specificity 100%) for diagnosis, it has low sensitivity (60%). In automated bacterial identification systems, it can be misidentified as *Pseudomonas*, *B. cepacia* or *Chromobacterium violaceum*. Serological testing has even lower sensitivity than culture and is unreliable in endemic areas because of high seroprevalence rates, which affects the specificity. Molecular techniques like PCR in conventional, real time and multiplex formats are available targeting 16SrRNA and TTS (type III secretion system) but the performance is variable and not routinely used in endemic areas.⁵ So our diagnostic approach to melioidosis should take all these factors into consideration.

Radiological investigation plays a very important role in melioidosis as the organism is found to produce abscess in multiple internal organs (prostate, spleen, liver and kidneys). Therefore, CT scan of abdomen and pelvis should be performed routinely in all patients with confirmed or suspected melioidosis. In melioidosis presenting with pneumonia, chest X-ray may show diffuse nodular infiltrates in both lungs, cavitation or upper lobe consolidation.

Infection is common among people with underlying comorbidities such as diabetes mellitus, renal disease, alcoholism, chronic lung disease, thalassaemia, neutropaenia and chronic granulomatous disease.⁶ Our patient suffered from septicaemia with neurological disease though he did not have any risk factors.

Patient presented with facial nerve palsy without symptoms of overt CNS infection. Facial nerve palsy can occur in melioidosis due to direct CNS invasion or the exotoxin-mediated demyelination or nuclear damage.⁷

Current recommendation for drug therapy includes intensive therapy and maintenance therapy. For intensive therapy, ceftazidime or meropenem or imipenem is given for 10–14 days. Any one of the three may be combined with cotrimoxazole for

neurological cutaneous, bone and prostatic melioidosis. Maintenance therapy is given with cotrimoxazole for 3 months.⁸ Our patient was treated the recommended antibiotic schedule—both intensive and maintenance therapy for (2+12) 14 weeks altogether. He has improved dramatically with partial recovery of the seventh cranial nerve palsy. The time taken for the fever to come down after commencement of antibiotic was 10 days, which was similar to the observations made in earlier studies.⁹ Resistance to ceftazidime or amoxycillin among *Burkholderia pseudomallei* is very low (0.6%) and no resistance to carbapenems have been reported.¹⁰ Increased awareness regarding this organism should be present among the clinicians and microbiology laboratory staff to reduce the mortality and morbidity due to melioidosis.

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Learning points

- Acute melioidosis usually presents with fever and pneumonia with septicaemia being the most common presentation.
- Isolation of *Pseudomonas* sp which is sensitive to ceftazidime and carbapenems and resistant to aminoglycosides and polymyxin B should raise the suspicion of *Burkholderia pseudomallei* in people from endemic areas and in travellers who have visited endemic areas.
- The therapeutic response can take over a week, even with the right antibiotic in the right dose.

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