Short Report

Surgical presentation of melioidosis in India

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ABSTRACT

Background. Melioidosis, the disease caused by *Burkholderia pseudomallei,* is common in Southeast Asia. It has also been reported from India, where some investigators feel it is under-diagnosed and under-reported. We report our experience with melioidosis presenting as abscesses at unusual sites.

Methods. All consecutive patients with culture proven *B. pseudomallei,* who presented to a single surgical unit between 1995 and 1998, were evaluated.

Results. Three patients presented with splenic abscesses and one with a soft tissue abscess in the neck. One patient developed septicaemia. All patients responded favourably to ceftazidime and/or co-trimoxazole which was started as soon as the diagnosis was confirmed.

Conclusion. Melioidosis is under-diagnosed in India, probably due to a low index of suspicion of this disease among clinicians. It should be considered as a possibility when abscesses are encountered at unusual sites. The pus must then be cultured to identify the causative agent.

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INTRODUCTION

Melioidosis is endemic in Southeast Asia.[1-2] It has been documented to be prevalent in India.[3] *Burkholderia pseudomallei,* the causative agent, is a saprophyte found in the soil and surface water. Human infection occurs when the organism gains entry through broken skin, by inhalation, and perhaps ingestion. Following an incubation period lasting a few days to many years, melioidosis may present as an acute, subacute or chronic illness. Abscess formation is the commonest surgical presentation. This usually occurs in the skin and subcutaneous tissues but it has also been reported at unusual sites such as the liver, spleen, lung, brain, prostate, bone and joints, lymph nodes, muscles and eye.[2]

Despite the fact that melioidosis is prevalent in India, it is seldom diagnosed clinically. The diagnosis is made most often by the microbiologist. We report our experience with the surgical presentations of melioidosis, to alert clinicians to the possibility of this disease when abscesses at unusual sites are encountered.

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PATIENTS AND METHODS

All consecutive patients with culture proven *B. pseudomallei* infection who presented to our surgical unit between 1995 and 1998 were evaluated. In all patients, the organism was isolated from a pus sample and blood culture was done in one patient. In the microbiology laboratory, after Gram staining, the pus sample was cultured on blood agar (BA), chocolate agar, MacConkey agar (MA) and thioglycollate media. *B. pseudomallei* colonies show characteristic haemolysis on BA. On MA, they are non-lactose fermenters, but slowly turn pink on further incubation. They have a characteristic 'earthy' odour. They are Gram-negative bacilli and tend to stain darkly at the ends giving a 'safety pin' appearance. Biochemical tests for differentiating *B. pseudomallei* from other non-fermenting Gram-negative bacilli (NFGNB) were done.[4] A slide agglutination test with homologous antiserum was used to obtain a presumptive result within 24 hours.[5]

RESULTS

The presence of *B. pseudomallei* was confirmed in the abscesses of five patients. A 46-year-old manual labourer from a suburb of Vellore (Tamil Nadu) presented with a swelling of 10 days' duration in the right supraclavicular region, associated with fever and dysphagia. On the day of admission, she developed a bout of cough with expectoration of purulent sputum. This was associated with a decrease in the size of the neck swelling. She was febrile, had fullness in the right supraclavicular region which was tender and had crepitus. A plain X-ray of the neck showed a cavity with an air–fluid level. The abscess was drained and a fistula between the abscess cavity and the trachea was seen and confirmed by bronchoscopy. Therefore, a tracheostomy was performed. Culture of the pus grew *B. pseudomallei* sensitive to tetracycline, co-trimoxazole, chloromycetin and ceftazidime. She was started on ceftazidime, which was given for 2 weeks followed by co-trimoxazole for 6 months. The trachea was decanulated after the wound had healed. At follow up, the patient was well and leading a normal life.

A 20-year-old student from Belgaum (Karnataka) presented with fever and chills of 2 weeks' duration and associated anorexia and weight loss. On evaluation, she was found to have a splenic abscess. Ultrasound-guided aspiration of the abscess was done and the culture revealed *B. pseudomallei.* This was sensitive to cefotaxime, co-trimoxazole, ceftazidime, ciprofloxacin and tetracycline. She was given ceftazidime for 6 weeks and co-trimoxazole for 6 months. At follow up, she had improved clinically, but was found to have a residual splenic abscess. The pus was re-aspirated under ultrasound guidance, and a mixture of gut organisms was isolated in the culture but *B. pseudomallei* was not grown. A splenectomy was performed and the patient was well at follow up a year later.

A 38-year-old farmer from Nagaon (Assam) presented with fever, left upper quadrant abdominal pain, weight loss and a sinus discharging pus in the left flank. He was a known diabetic and was also on treatment for pulmonary tuberculosis. Pus from the sinus was negative for fungus and acid-fast bacilli. *B. pseudomallei* was grown on culture. This was sensitive to tetracycline, co-trimoxazole, chloromycetin and ceftazidime. An ultrasound of the

abdomen revealed multiple splenic abscesses. He was started on co-trimoxazole and the sinuses healed within 2 months of treatment. However, the antibiotic was continued for a year and at follow up, he was well.

A 32-year-old farmer from Midnapur (West Bengal) with homozygous beta-thalassaemia was referred for splenectomy. Preoperative ultrasound examination of the spleen revealed multifocal hypoechoic areas which were thought to be haemangiomas. Following splenectomy the patient developed high-grade fever. The wound was found to be infected and was laid open. Pus and blood were cultured and both grew *B. pseudomallei*. Histopathological examination of the spleen revealed multiple abscesses. The patient was treated with ceftazidime and co-trimoxazole for 2 weeks by which time the fever subsided and granulation tissue started forming in the wound. He has been advised co-trimoxazole for 6 months and is currently well.

A 50-year-old diabetic male with melaena, a chargeman in a steel factory, was referred from Bihar. On investigation, he was found to have a splenic abscess with splenic vein thrombosis and portal hypertension. He had previously been diagnosed to have pulmonary tuberculosis and had completed a nine-month course of antituberculous treatment. Physical examination was unremarkable. Gastroscopy revealed a large fundal varix. Ultrasound revealed splenomegaly and hypoechoic areas in the spleen. There was evidence of splenic vein thrombosis on visceral angiogram. Splenectomy and sleeve resection of the stomach were done. The pus grew *B. pseudomallei* sensitive to tetracycline, co-trimoxazole and ceftazidime. The spleen also showed histopathological evidence of tuberculosis. He was treated with ceftazidime and was recommended six months of co-trimoxazole. The patient was well on discharge but is yet to return for follow up.

DISCUSSION

Melioidosis is still an uncommon disease in India, though it is being increasingly reported. In order to diagnose melioidosis, the clinician needs to have a high index of suspicion. It should be suspected in people who are exposed to contamination by way of their occupation (e.g. farmers). Concurrent medical illness (diabetes mellitus, cirrhosis, AIDS) is a predisposing factor.

Abscesses may occur in various sites (some of them unusual) and may be multiple. These need to be systematically looked for as they may be asymptomatic. Various pathogens cause abscesses at unusual sites. Table I outlines the differential diagnosis and clinical features of some of them. Based on information obtained from our Medical Records Department, approximately 200 patients with abscesses were reported in a single surgical unit during the period of study. Abscesses caused by *B. pseudomallei* constituted 2.5% of the cases. In our microbiology laboratory, *Pseudomonas sp.* was isolated in 7.2% of isolates between 1995 and 1998.

Melioidosis was not suspected at presentation in any of the cases and none of our patients had travelled outside India. All had tested negative for HIV antibodies. Two of the patients were farmers who could have been exposed to the risk of contamination. Ultrasound scan did not reveal abscesses at other sites. In one patient *Burkholderia* bacteraemia occurred following splenectomy. This may have been induced by manipulation of the spleen at surgery. The possibility of inducing septicaemia by handling or puncture of abscesses should always be borne in mind.

In patients with localized disease, monotherapy with amoxycillin/clavulanic acid, tetracycline, chloramphenicol or co-trimoxazole is recommended for 2–6 months.[2] In septicaemic and gravely ill patients, a combination of parenteral ceftazidime and

TABLE I. Unusual causes of abscesses

Organism	Microbiology	Clinical features	Diagnosis	Treatment
Atypical mycobacteria	Rapidly growing colonies at 30–33 °C	Commonly infectious abscess, laparoscopic port-sites, non-healing wounds and surgical scars	Characteristic colonies growing rapidly at 33 °C	Ciprofloxacin, amikacin for 6 weeks
Mycobacterium tuberculosis	Acid-fast bacilli grow in Lowenstein–Jensen medium	Usually bone or lymph node abscesses, can occur anywhere in the body	Acid-fast bacilli on staining, grow in Lowenstein–Jensen medium, caseation and Langhan's giant cells on histology	Isonicotinic acid hyrazide, rifampicin, ethambutol, pyrazinamide, etc.
Coccidiodes immitis	Soil fungus with mycelial forms and arthrospores	Indolent, not so painful subcutaneous abscesses	Microscopic identification of spherules in wet smears or cytopathology	Drainage, ketoconazole, amphotericin B
Actinomycetes	Branching, beaded Gram-positive filaments, weakly acid-fast	Mainly pulmonary or systemic abscesses, chronic or subacute abscesses	By Gram stain, slow-growing orange colonies	Minocycline, amikacin
Nocardia sp.	Gram-positive, non-spore forming anaerobic or microaerophilic rods	Usually abdominal visceral abscesses, may mimic tumour, present long after primary infection	Granules from discharge	Intravenous penicillin
Clostridium perfringens	Gram-positive, encapsulated non-motile bacillus	Severe pain with suppuration and crepitus	By Gram stain, culture in selective media	Penicillin, clindamycin
B. mallei	Aerobic Gram-negative immotile bacillus	Causes glanders, chronic indolent ulcers of skin, inhalation can cause pneumonia	Does not grow on MacConkey agar, oxidase negative	Drainage, sulphadiazine, slaughter of infected horses
B. pseudomallei	Gram-negative bacilli, safety pin appearance	Abscesses at unusual sites, e.g. spleen, bone, muscle	Non-fastidious, non-fermenting Gram-negative bacilli, oxidase-positive	Ceftazidime for 6 weeks, co-trimoxazole for up to 1 year

co-trimoxazole is recommended for the initial 2 weeks followed by co-trimoxazole for 6–12 months. All our patients responded favourably to ceftazidime 1–2 g intravenously thrice a day and/or co-trimoxazole 960 mg twice a day which were instituted as soon as the diagnosis was made. Not all abscesses require surgical drainage, as seen in one of our patients.

The laboratory diagnosis of melioidosis is made by isolation of the organism. *B. pseudomallei* is a non-fastidious organism, which can be easily grown on routinely used culture media. Biochemically, it is a non-fermenter. Microbiologists tend to consider NFGNB to be of no clinical significance and unless they are subjected to additional biochemical tests, *B. pseudomallei* may not be recognized. Since *B. pseudomallei* has been increasingly reported from India, our microbiology laboratory fully characterizes all NFGNB which are positive for oxidase test, motile and resistant to gentamicin.[4] We were thus able to identify *B. pseudomallei* in all our cases.

Kang *et al.*[6] have done a sero-surveillance study in South India and have found that a proportion of the rural population may be exposed to *B. pseudomallei.* Further studies have to be done to determine the usefulness of serological tests in India.

Even though this bacterium was discovered as early as 1912, it was re-speciated only recently.[1] Since very little information was available about melioidosis in India, clinicians and microbiologists in this country did not consider it to be a significant pathogen until the report of a plague-like illness allegedly caused by *B. pseudomallei* in Maharashtra in 1994.[5] Subsequently, many investigators have reported melioidosis, establishing that this disease is widely prevalent in India.[7] Our patients were from Tamil Nadu, Karnataka, Assam and West Bengal suggesting that this infection has a wide geographical distribution in India. Community-based surveys are needed to determine how widely this infection is endemic in India.

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Obituaries

Many doctors in India practise medicine in difficult areas under trying circumstances and resist the attraction of better prospects in western countries and in the Middle East. They die without their contributions to our country being acknowledged.

The National Medical Journal of India wishes to recognize the efforts of these doctors. We invite short accounts of the life and work of a recently deceased colleague by a friend, student or relative. The account in about 500 to 1000 words should describe his or her education and training and highlight the achievements as well as disappointments. A photograph should accompany the obituary.

—Editor