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Case Report

Melioidosis - A Rare Infectious Disease with Atypical Presentation

Shawhely Mahbub[1], Zaheer Al- Amin[2], Sudhangshu Shekhar Biswas[3], Mohammad Shah Jamal[4], Soma Halder[5]

Abstract

Melioidosis is *an infectious disease caused by a Gram negative bacterium,* Burkholderia Pseudomallei. *It is endemic in parts of Southeast Asia, Taiwan & northern Australia. In Bangladesh it is rare. Laboratory identification of* B. pseudomallei *can be difficult, especially where it is rarely seen. The disease is of public health importance as it is associated with high fatality rate in human being.*

The disease is known as a remarkable imitator due to the wide & variable clinical spectrum of its manifestations. Septicaemia, sepsis, septic arthritis, lung nodules & pneumonia are well recognized manifestations of Melioidosis*. ENT manifestation is rare.*

Here we reported a case of mallioidosis. The patient was diabetic hailing from Gazipur, presented with cellulitis over the neck and septic arthritis of left shoulder and hip with lung nodules. He responded to prolonged treatment with intravenous ceftazidime.

Key words: *Burkholderia pseudomallei, melioidosis.*

Introduction

Burkholderia pseudomallei (also known as *Pseudomonas pseudomallei* or Vietnami time bomb) is a Gram-negative, bipolar, aerobic, motile rod-shaped bacterium found in soil & water[1]. It infects human and animal and causes the disease melioidosis. Melioidosis is endemic in parts of south-east Asia (including Thailand, Laos, southern China, Singapore, Malaysia, Burma and Vietnam), Taiwan and northern Australia[2]. Multiple cases have also been described in Hong Kong, Brunei and India; and sporadic cases in Central and South America, the Middle East, the Pacific and several African countries[3]. The presentation varies from an acute fulminant septicaemia to a chronic, debilitating localized infection & abscess formation. The disease - also known as psuedoglanders or Whitmore's disease is a remarkable imitator due to wide range & variable clinical spectrum of its presentation[4].

1. Senior Medical Officer, Dept. of Otolaryngology & Head-Neck Surgery, BIRDEM General Hospital, Shahbag, Dhaka.
2. Professor & Head, Dept. of Otolaryngology & head-Neck Surgery, BIRDEM General Hospital
3. Assistant Professor, Dept. of Otolaryngology & head-Neck Surgery BIRDEM General Hospital
4. Assistant Professor, Dept. of Gastroenterology, Comilla Medical College & Hospital
5. MBBS, FCPS (Paediatrics)

Correspondence : Dr. Shawhely Mahbub, 1/C North Kamolapur (1st floor) Dhaka- 1217, E Mail: dr.shawhelym@yahoo.com

Case report

A 40 years old, diabetic farmer, hailing from Rashedpur, Borogobindopur, Kaliakoir,

Gazipur got admitted under department of Medicine, BIRDEM on 28th of September, 2013 with the complaints of pain in the neck involving left supraclavicular region and left shoulder, high grade intermittent fever & Mild chest pain for about two months (Fig-I). The pain was gradually increasing day by day and become severe and throbbing in nature and was not responding to usual analgesics. Later on he had developed redness and swelling in the left supraclavicular region with restricted movement of left shoulder. He also complained of high grade intermittent fever for the same duration. The temperature used to rise with chills and rigor and subsided with sweating after taking antipyretics. Highest recorded temperature was 103º F. He had no cough. He was treated with intravenous Meropenem. ENT consultation was taken on 02.10.13 and the patient was advised to be transferred under ENT dept. After 2-3 days the patient left the hospital on request but continued injection Meropenem. He was again got admitted at BIRDEM on 09.10.13, this time, under ENT dept. with exacerbation of his previous complaints along with inability to walk. He also developed severe pain in his left hip joint. He is known diabetic for last 5 years & taking insulin for the last 2 years. His diabetes was uncontrolled for last 2-3 months. He is hypertensive for 1 year and used to take antihypertensive regularly.

On examination his Pulse: 100b/min., Respiratory Rate: 16/min, B.P. 130/80 mm.Hg. Temp. 102º F. Breath sound diminished on left side. Investigations revealed– Hb% was 9.5 g/dl, WBC – 17,080c/cmm, with neutrophilic predominance (Polymorphs– 83.3%, Lymph- 9.4%, Mono-6.9%, Eosino- .4%), Platelet count was-3,09,000/cmm. His fasting & post parandial blood sugar levels were 9.2 & 13.2 mmol/L respectively. X- ray left shoulder (B / V) was done on which was normal. USG of Whole Abdomen was done which was Suggestive of cystitis with bilateral pyelonephritis, bilateral mild pleural effusion, mild hepatomegaly with fatty change of liver. USG of left hip joint was done on which revealed minimal collection in left hip joint, suggestive of minimal joint effusion. CT scan of the neck was done which showed multiple nodules in upper lobes of both lungs; No cervical lymphadenopathy or any neck mass was found (fig- II).repeated blood culture (on: 29.9.13, 10.10.13, and 13.11.13) revealed no growth. Urine culture also revealed No growth. On 10.10.13 a stab incision was made over left supraclavicular region under Local Anaesthesia, very small amount of pus was squeezed out and sent for Culture which revealed Growth of *Burkholderia Pseudomallei,* Colony count were moderate & were Sensitive to– Augmantin, Cefixime, Ceftriaxone, Ceftazidime, Ciprofloxacin and

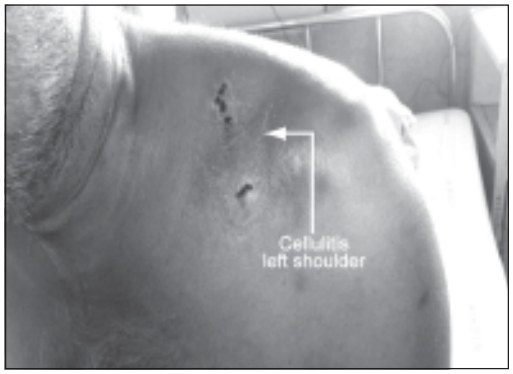

Fig.-1

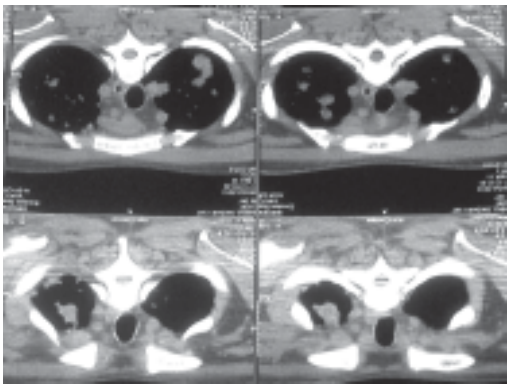

Fig.-2: *CT Neck showing nodules at apical*

many other drugs.. Finally the patient was diagnosed as a case of Melioidosis. From 13.10.13 antibiotic was changed to intravenous Ceftazidime at a dose of 2 gm 8 hourly.

His blood sugar level was controlled with subcutaneous insulin. After a week there was a new cystic lesion developed over left lower neck just posterior to sternomastoid (fig-Iv). Pus was aspirated & sent for C/S which revealed (19.10.13) Growth of *Burkholderia Pseudomallei,* The patient developed septic arthritis of left hip but gradually subsided with ongoing treatment. After about a month of intravenous ceftazidime the patient got improved, became afebrile and could walk alone.

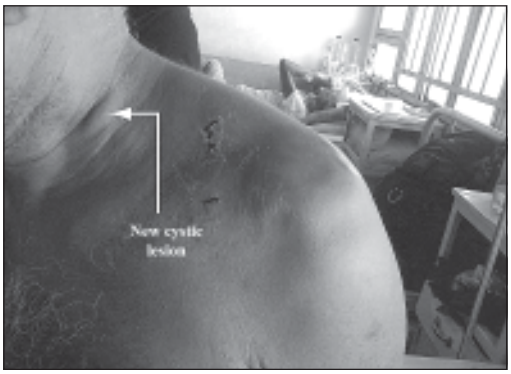

Fig.-3: *New lesion.*

Discussion

Melioidosis can be acquired by inoculation of environmental organisms through penetrating wounds or into existing skin lesions, inhalations or aspiration of contaminated water[5]. The infection is more common in male who indulge in agricultural activities. DM increases the risk by 100 folds[6]. Other predisposing factors are renal disease, alcoholism, cirrhosis, chronic lung disease, thalassaemia, chronic grannulomatous disease, porphyria cutanea tarda, cystic fibrosis, haemosiderosis, splenectomy, aplastic anaemia, febrile neutropenia, mycobacterial disease, malnutrition and immunosuppression[7].

The disease spectrum is quite variable. The infected individual can remain carrier with totally symptom free, remain latent for years and manifest whenever cell mediated immunity is suppressed. Melioidosis can present as a febrile illness, ranging from an acute fulminant septicaemia to a chronic debilitating localized infection and abscess formation. There is usually no obvious infected wound or evidence of recent trauma. It can present as acute septicaemia with high grade fever, lower respiratory infection and hepatosplenomegaly, disseminated cutaneous & visceral abscess and shock. Pneumonia is the most common presentation of melioidosis seen in approximately half of the cases[8]. Acute pulmonary syndrome with pnuemonitis involves upper lobes of the lung & is confused with tuberculosis. Acute or chronic localized suppurative infection can involve skin, brain, lung, myocardium, liver, spleen, bones, joints, lymph nodes and even the eyes[9]. Our first case of melioidosis presented with septicaemia & an abscess over the temporal region later developed orbital abscess, septic shock & subsequently orbital apex syndrome.

Laboratory identification of *B. pseudomallei* can be difficult, especially in Western countries where *B. pseudomalleus is* rarely seen. The large wrinkled colonies look like environmental contaminants and are therefore often discarded as being of no clinical significance. This may be the reason that pus culture from the temporal region of the first case yielded no growth. Non-sterile specimens should therefore be cultured in selective media (e.g., Ashdown's or B. cepacia medium) [10]. Even when the isolate is recognised to be significant, commonly used identification systems may misidentify the organism as *Chromobacterium violaceum* or

other non-fermenting gram-negative bacilli such as *Burkholderia cepacia* or *Pseudomonas aeruginosa*.[11] Blood culture from our first case yielded *Psuedomonas.*

Treatment of melioidosis is divided into two stages- an intravenous high intensity phase and an eradication phase to prevent recurrence[12]. Intravenous ceftazidime is the treatment of choice. Intravenous ceftazidime 2 gm. 8 hourly for 2 to 4 weeks, and are not usually stopped until the patient's temperature has returned to normal & remainafebrile for at least 48 hours[13]. The carbapenem antibiotics (Imipenem & Meopenem), though slightly less effective can be used as an alternative[14]. Even with appropriate antibiotic therapy, fevers often persist for weeks or months, and patients may continue to develop new lesions even while on appropriate treatment, as was seen in our second case. Development of a new lesion is not a reason for changing antibiotic. It usually takes a long time to respond even with appropriate treatment[15]. Our second case took a month and a half to become afebrile.

Trimethorpim (8mg/kg/day) and sulphamethoxazole (40mg/kg/day) in four divided doses is given for 12 to 20 weeks after intravenous therapy to eradicate the organism & to prevent relapse[16]. The alternative option for prolonged treatment is doxycycline or amoxicillin-clavulanate, though less effective[17].

Without access to appropriate antibiotics (principally ceftazidime or meropenem), the septicemic form of melioidosis has a mortality rate that exceeds 90%.[18] With appropriate antibiotics, the mortality rate is about 10% for uncomplicated cases but up to 80% for cases with bacteraemia or severe sepsis[18,19]. It is certain that access to intensive care facilities is also important, and probably at least partially explains why total mortality is 20% in Northern Australia but 40% in Northeast Thailand[20].

Conclusion

Documented reports of Melioidosis from Bangladesh are few. It is an under-diagnosed & under-reported disease in our country & there is an utmost need of creating awareness among the clinicians, microbiologists & other healthcare professionals. The clinical manifestations of Melioidosis are not diagnostic, and such a diagnosis requires high degree of suspicion and experienced microbiologist and laboratory staffs. Prolonged treatment is required to avoid chronic debility and mortality. Multi-disciplinary approaches are often helpful.

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