Whitmore's Disease as an Etiology of Necrotising Granulomatous Lymphadenitis- A Case report

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ABSTRACT

Introduction: In India which is located in tuberculosis-endemic zone, all cases of necrotizing granulomatous lymphadenitis are traditionally considered as of tuberculous etiology, unless proved otherwise. However, Melioidosis should be kept in mind as one of the etiological factor of necrotizing granulomatous cervical lymphadenopathy.

Case report: We report a case of Cervical lymphadenitis due to Melioidosis in a farmer who was not treated adequately and developed foot abscess due to lack of awareness about the disease and its management.

Conclusion: Necrotizing granulomatous lymphadenitis due to Melioidosis which if not reported in co-ordination with microbiology department might have been mistreated with anti tuberculous drugs.

Keywords: Necrotising Granulomatous Lymphadenitis, Melioidosis, Eradication Therapy

INTRODUCTION

Melioidosis also called Whitmore's disease [3], is a community acquired infectious disease. It was first described in Rangoon in 1912. The first report of Melioidosis from India was by Raghavan et al from Mumbai in 1991. [4] It is caused by the Gram negative bacterium, Burkholderia pseudomallei found in soil and water.[4] It is endemic in tropical areas of South East Asia (Thailand, Malaysia, Singapore) and Northern Australia.[5] This is an emerging infection in India. Most cases are reported from South India, but has its origin from Eastern and Western coastal belts of India[4].It is transmitted through inhalation, direct contact with contaminated soil or water through penetrating wounds and existing skin abrasions.[4] Pre-existing medical conditions that compromise immunity can predispose to melioidosis e.g. diabetes mellitus, renal disease, chronic alcoholism, immune suppression due to any other cause or occupational exposure to soil and surface water[3]. It occurs in patients aged 40-60 years. Incubation period is 2 days to months or years. The most common presentation is pneumonia. The patient may also present with acute septicemia, prolonged fever, weight loss, localised infections like abscess (liver , spleen, soft tissue etc), lymphadenitis. Some cases are asymptomatic or subclinical. The Gold standard for diagnosis is Culture and sensitivity.[4] Serology and PCR tests are available for detection.[6] Management of melioidosis consists of 2 phases: the intensive phase and the eradication phase. Melioidosis is a preventable disease. Tourists travelling to endemic areas should be cautioned against barefoot walking and recreational activities in water. People with risk factors should use protective footwear and masks during rainy season and while going to paddy fields. Laboratory technicians and research workers should work inside BSC II facility.

CASE REPORT

A 42 year old male patient, farmer by occupation, reported to the Surgery Department with cervical swelling since 4 months with low grade fever. He was given prophylactic Amoxyclav for 10 days without any relief. Fine needle aspiration cytology (FNAC) was advised to rule out Tuberculous lymphadenitis.

On clinical examination, the patient was afebrile. There were non tender left cervical matted lymph nodes. Vitals and systemic examination was normal. On investigating, Hb: 12.5 gm%, TC: 11,500, N: 70, E: 01, M: 02, L: 27 and ESR was raised. Biochemical parameters were normal. HIV and HBsAg tests were Negative. X-ray chest was normal . Tuberculin test was negative.

Fine Needle Aspiration Cytology revealed epithelioid granulomas with caseous necrotic debris with presence of numerous neutrophils (Figure 1). Necrotic pus like material aspirated was subjected to Gram staining, Zeihl Neelson (ZN) staining and Culture and Sensitivity as per Standard Operating Procedure for processing pus samples in the department.

Gram staining revealed numerous pus cells. No organisms were seen. Smear for Acid fast bacilli was negative. Culture initially showed growth of dry and wrinkled colonies on Blood agar and pink, dry and wrinkled colonies on Mac Conkey agar which were assumed as contaminant and not studied further. Repeat FNAC was done for subjecting the sample for CBNAAT (Cartridge based nucleic acid amplification test) testing to rule out tuberculosis. Excess aspirate was subjected to repeat culture and sensitivity testing which again showed growth of similar colonies raising a suspicion of growth of pathogenic organism. Gram stain of the growth showed Gram negative bacilli with irregular bipolar staining (safety pin appearance). The isolate was

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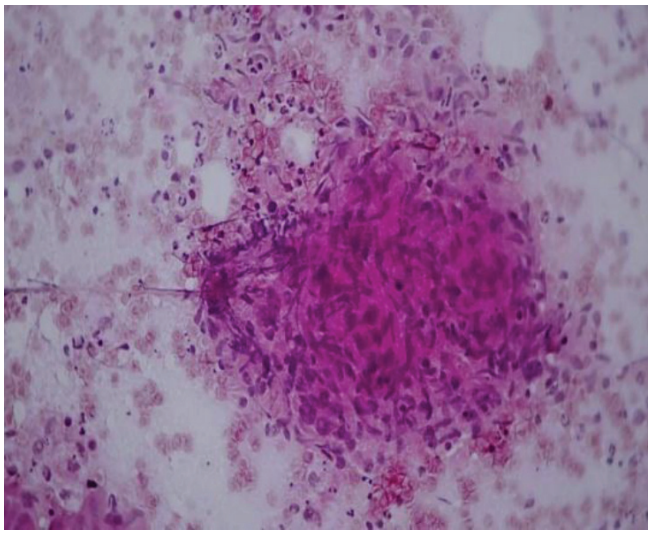

Figure-1:

identified as Burkholderia pseudomallei on Vitek 2 system. The patient was treated with IV Ceftazidime for 10 days and was discharged with disappearance of cervical swelling. The patient came back after 1 month with abscess on the foot. On doing FNAC, pus aspirated from foot, again showed growth of Burkholderia pseudomallei. The recurrence occurred because the patient was not put on eradication therapy due to lack of awareness among the clinicians about melioidosis. He was started on IV Meropenem for 10 days and was put on oral Cotrimoxazole for 3 months. There was complete disappearance of the foot swelling without any surgical intervention in this case.

DISCUSSION

Melioidosis is caused by Burkholderia pseudomallei which is a facultative intracellular gram negative saprophytic bacterium found in soil or contaminated water.[9] The population at risk are those having occupational exposure to wet soil i.e. farming, agriculture, fishing, gardening etc. and in immunocompromised states such as diabetes, alcoholism, chronic renal failure, chronic lung disease and HIV/AIDS.[9]

In our case, cytology alone could not differentiate the cause of necrotizing granulomatous lymphadenitis. The availability of automated bacterial identification system helped in diagnosing the etiological agent responsible for necrotizing granulomatous lymphadenitis.

The differential diagnosis of necrotizing granulomatous lymphadenitis is wide and includes infectious diseases (bacterial, viral, fungal, parasitic), malignant disorders mainly lymphoid malignancies, auto immune disorders like Systemic lupus erythematosus, auto inflammatory diseases and idiopathic causes like Kikuchi's disease and sarcoidosis.[8]

In a tuberculosis endemic country like India necrotising granulomatous lymphadenitis diagnosed on FNAC may be interpreted as tuberculous lymphadenitis and such cases will be mistreated with anti tuberculous drugs. In our case simultaneous AFB staining, CBNAAT testing, Culture and Sensitivity testing helped in identification of the organism. This patient came back with a foot abscess because of lack of awareness about this disease and its management amongst the clinicians.

Management of melioidosis consists of 2 phases: the intensive phase and the eradication phase. The intensive phase consists of 10-14 days of IV antibiotics: IV ceftazidime or IV carbapenems (meropenem/imipenem). If there are fluid collections (including skin abscess/septic arthritis), bone or central nervous system involvement, cotrimoxazole, doxycycline or amoxicillin-clavulanate should be added early during the intensive phase for tissue penetration. Subsequently the eradication phase should be with another 3-6 months of oral co-trimoxazolke alone or in combination with oral doxycycline/ oral amoxicillin-clavulanate.[9]

We present this case to emphasize the importance of having high degree suspicion in making diagnosis of melioidosis with adequate microbiological knowledge and utilising automated bacterial identification system. We also would like to emphasize about creating awareness among the treating clinicians about the treatment of melioidosis which comprises of intensive and eradication phase.

CONCLUSION

The Clinician, Pathologist and Microbiologist should be aware of this infectious disease as it may be misdiagnosed, misreported and mistreated as Tuberculous lymphadenitis. It highlights the importance of doing cytology with simultaneous culture and sensitivity, AFB staining and CBNAAT testing. Utilization of automated bacterial identification and sentisitivity testing systems in Microbiology should be stressed on as in our country most laboratories still rely on conventional culturing methods with low sensitivity leading to under-reporting of these cases. There has to be a good clinico-pathological interaction for creating awareness of Melioidosis among the clinicians as they are unaware of this entity leading to mistreatment of these cases.

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