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Positive melioidosis serology in a patient with adult onset Still's disease: a case report of a diagnostic dilemma

Harsha Anuruddhika Dissanayake[1*], Gayani Premawansa[2], Enoka Corea[3] and Inoshi Atukorale[4]

Abstract

Background: Autoimmune disorders are known to produce false positives in serological tests for infections. Aetiological association between infections and autoimmunity, increased susceptibility to infectious and autoimmune disorders with immune dysregulation and non-specific polyclonal expansion of B cells with autoimmunity may cause confusion in diagnosis and patient management. We report a patient with Adult Onset Still's Disease (AOSD) presenting with rising melioidosis antibody titres that caused diagnostic confusion.

Case presentation: A forty-nine-year-old female presented with prolonged fever, sore-throat, large joint arthritis, lymphadenopathy, hepatomegaly and transient rash. She had elevated inflammatory markers and a rising melioidosis antibody titre. The patient responded poorly to prolonged course of appropriate antimicrobials but showed rapid and sustained improvement with glucocorticoids.

Conclusion: Positive melioidosis serology could have been due to a co-infection or false positive antibody reaction due to non-specific B cell expansion or an indicator of true infection that triggered the immune dysregulation to develop AOSD.

Keywords: Adult onset Still's disease, Melioidosis, False positive antibodies

Background

Melioidosis is an emerging infection in the tropics caused by *Burkholderia pseudomallei*. Diagnosis of melioidosis is a challenge [1]. Although isolation of the organism in culture is the gold standard, it has a low sensitivity and negative predictive value [2]. Detection of antibodies against melioidosis in serum provides valuable supportive evidence. However, serology alone is not reliable enough for confirmation. The major disadvantage of serological tests is the false positive results. This is may occur due to the presence of antibodies in people living in endemic areas, cross reactivity with antibodies to different organisms or in association with autoimmune disorders. We report a patient with adult onset Still's disease (AOSD) who developed antibodies against *B. pseudomallei,* a phenomenon not described previously.

Case presentation

A forty-nine-year-old female from a suburban community in Sri Lanka presented with insidious high grade intermittent fever with chills and rigors for 2 months. She experienced one to two febrile episodes daily with complete defervescence in between. She also had anorexia, weight loss, sore-throat and symmetrical large joint arthritis without morning stiffness. Small joints and axial skeleton were spared. She also noticed an itchy desquamating erythematous rash over back of the trunk and proximal limbs. Erythematous patches were transient and recurring but did not temporally correspond to febrile peaks. The patient did not have any symptoms referable to a focus of infection and did not report photosensitivity, Raynaud phenomenon, past history of tuberculosis, or high risk sexual behaviours.

The patient was averagely built (BMI: 23.1 kg/m^2), febrile (39.9 °C), ill and pale. A firm 1.5 cm lymph node in the right posterior cervical group was noted. Throat was non-inflamed. Erythematous macules noted over the trunk and proximal limbs were transient. Symmetric arthritis affected elbow, wrist and knee joints. A smooth

* Correspondence: dissanayakeha@gmail.com
[1]University Medical Unit, National Hospital of Sri Lanka, Colombo, Sri Lanka
Full list of author information is available at the end of the article