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A rarely considered diagnosis of unknown fever, disseminated lymphadenopathy and chronic peritonitis in Taiwan: Whipple's disease



Dear Editor,

Whipple's disease is a rare systemic infection cause by *Tropheryma whippelii* and predominantly affects Caucasian men.¹ To our knowledge, it has never been described in Taiwan. We reported an imported patient of Whipple's disease.

A 43-year-old Caucasian gentleman was seen at our hospital for fever, chills, night sweating, joint soreness and weight loss of 5 kg within 2 months. Upon physical examination at our clinic, his conjunctiva was pink and there was no palpable lymphadenopathies in neck, axillary and inguinal area. The breath sound was clear. Abdomen was soft without tenderness, and the bowel sound was normally active. The peripheral joints were not swollen.

Blood examination revealed microcytic anemia (hemoglobin of 10.5 g/dL and MCV of 73.1 fL), leukocytosis (white blood cell count of 13800 cells/ μ L), eosinophilia (absolute eosinophil count of 1159 cells/ μ L) and elevated C-reactive protein (9.2 mg/dL). Septic work-ups including blood cultures, urine culture, acid-fast staining (AFS) of expectorated sputum were negative.

Whole body nuclear scintigraphy after intravenous Gallium-67 citrate revealed lymphadenopathy in the neck, subclavical, mediastinal, para-aortic and mesenteric areas with increased tracer activity. Under the impression of disseminated mycobacterial infection or lymphoid malignancy, repeated biopsy of neck lymph nodes were performed. However, pathological examination was not conclusive. AFS, Periodic acid-Schiff (PAS) and Grocott's Methenamine Silver (GMS) stain as well as tissue cultures all failed to reveal any microorganism.

Six months later, patient's fever persisted and PET scan still demonstrated generalized FDG-avid lymphadenopathies (Fig. 1A). Therefore, we arranged exploratory laparoscopy for sampling of abdominal lymph nodes. During operation, there was adhesion between the liver surface

and peritoneum. Miliary whitish plaques were found on the peritoneum (Fig. 1B) as well as small amount of turbid ascites (Fig. 1C). All of these findings were compatible with chronic peritonitis.

Pathological examination showed interfollicular histiocytic infiltration with occasional fat cyst formation. Foci of neutrophilic and eosinophilic infiltration were also noted. PAS stains disclosed numerous short bacilli (Fig. 1D). This picture was compatible with Whipple's disease. Sequencing of bacterial ribosomal RNA was also identical to *Tropheryma whippelii*.²

The treatment of choice is believed to be intravenous ceftriaxone for 2 weeks, followed by oral maintenance medication for at least a year.^{3,4} However, our patient declined admission. Oral doxycycline and ceftibuten was prescribed for 14 days, followed by trimethoprim-sulfamethoxazole. His clinical symptoms improved promptly after initial therapy. Two months later, he gained 4 kg. His leukocytosis resolved and hemoglobin increased from 8.8 g/dL to 11.6 g/dL. After completing 1-year of maintenance therapy, he remained stable up to the study reported and followed PET scan revealed resolution of lymphadenopathy.

Our report is the first document of laparoscopic finding in Whipple's disease. The findings include abdominal lymphadenopathies, peritoneal adhesion, turbid ascites and peritoneal miliary plaques, all of which are consistent with chronic peritonitis. In Taiwan, this could point to tuberculosis peritonitis.⁵ However, our reports demonstrated that Whipple's disease should be listed as possible differential diagnosis given the suitable clinical context and epidemiological premise.

In conclusion, we report the first case of Whipple's disease ever diagnosed in Taiwan manifested with protracted fever, chronic peritonitis and generalized lymphadenopathies. The diagnosis should be considered in Caucasians or Taiwanese with appropriate clinical manifestation and

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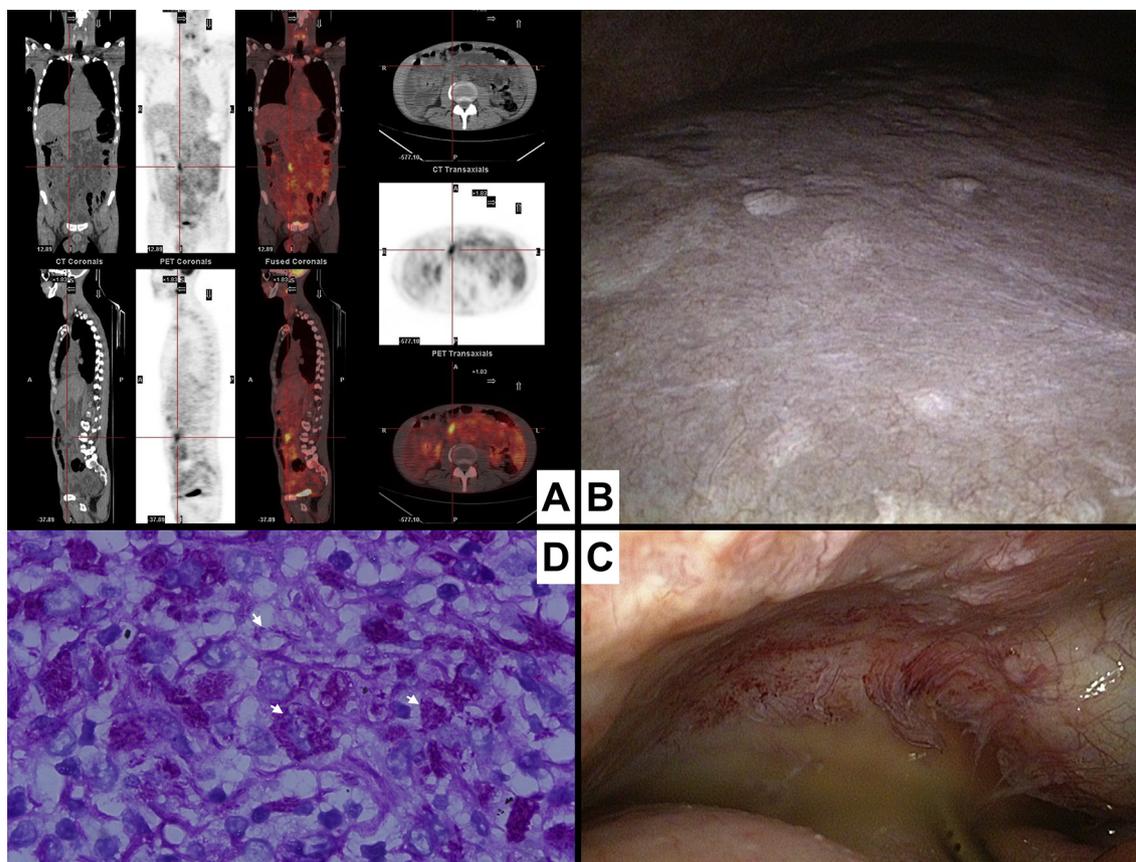


Figure 1. Image and pathological finding of the patient: A) PET scan revealed multiple FDG-avid mesenteric lymphadenopathies; B) Miliary whitish plaques were found on the peritoneum during laparoscopy; C) Inflamed peritoneum and turbid ascites were also found during laparoscopy; D) Numerous PAS-positive short bacilli (arrows) were found in the excised lymph node.

course. Response to antimicrobial therapy is excellent if the diagnosis was made correctly.

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Guan-Jhou Chen
Chien-Yuan Chen
Sung-Ching Pan*

Department of Internal Medicine, National Taiwan University Hospital, Taipei, Taiwan, 100

Ching-Yao Yang
Department of Surgery, National Taiwan University Hospital, Taipei, Taiwan, 100

Po-Ren Hsueh
Department of Internal Medicine, National Taiwan University Hospital, Taipei, Taiwan, 100

Department of Laboratory Medicine, National Taiwan University Hospital, Taipei, Taiwan, 100

Shan-Chwen Chang
Department of Internal Medicine, National Taiwan University Hospital, Taipei, Taiwan, 100
College of Medicine, National Taiwan University, Taipei, Taiwan, 100

*Corresponding author. Department of Internal Medicine, National Taiwan University Hospital No. 7 Chung-Shan South Road, Taipei, Taiwan 100 Fax: +886 2 23707772. E-mail address: scpbpan@gmail.com (S.-C. Pan)

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