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Case Report

Cat Scratch Disease Presenting as Fever of Unknown Origin in a Hemodialysis Patient with Autosomal Dominant Polycystic Kidney Disease

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Introduction

Cat-Scratch Disease (CSD) is an infectious disease characterized by self-limiting fever and localized granulomatous lymphadenopathy, which resolves in immunocompetent hosts in 1-4 month. Typically, a primary lesion appears close to the inoculation site after a cat scratch.

Bartonella hensellae is the etiologic agent involved in most cases of CSD. Immunocompromised hosts are more susceptible to infection from Bartonella, which results in a more severe clinical picture. In these patients, Bartonella can be disseminated and affect multiple organs as gastro-intestinal and respiratory tract, bones, brain and bone marrow [1]. Fever Of Unknown Origin (FUO) is a rare presentation of CSD.

We describe a case of CSD in a hemodialysis patient with Autosomal Dominant Polycystic Kidneys (ADPKD) who presented with abdominal pain and persistent fever.

Case report

A 50 year-old Asian woman with a diagnosis of CKD5 due to ADPKD started hemodialysis one month ago and was admitted to the hospital with a two days history of fever, chills and abdominal pain that started after her last dialysis session. A few days before admission oral Ciprofloxacin was initiated due to suspicion of Urinary Tract Infection (UTI). Physical examination at admission revealed an alert, oriented but ill appearing patient without signs of acute distress. Her temperature was 37.8o C, blood pressure 122/89 mmHg and pulse 84/min. Heart and lung examination revealed no abnormalities and no peripheral lymphadenopathy was palpable.

Abdominal examination revealed enlarged liver and two palpable and slightly painful kidneys. Laboratory findings included leucocytosis-12.000/mm3, C - reactive protein (CRP)-15.3 mg/dl and severe anemia (hemoglobin-5.7 g/dl with MCV 91/fl) which was probably related to recurrent macro hematuria and required a blood transfusion.

Due to persistent fever under Ciprofloxacin and the presence of a central tunneled catheter for hemodialysis, empirical antibiotic treatment with Vancomycin and Amikacin was initiated. Despite of antibiotic therapy, high fever, chills and night sweats persisted with no improvement in abdominal pain. Although the antibiotic therapy was upgraded to intravenous Tazoctam/Piperacillin, peaks of fever persisted.

Multiple blood and urine cultures were negative. Serologic tests of Legionella, Brucella, hepatitis B and C, Q-fever, CMV, EBV, VDRL and HIV were negative. Serology for ANA and ANCA were negative.

Acid fast stain of urine was negative.

Abdominal CT revealed enlarged liver and kidneys with multiple cysts, some of which were hyperdense with probably bloody fluid. Moreover small amount of pleural effusions, hepatic and pelvic fluid were present. Pelvic fluid tests were negative for bacteria, malignant cells and acid fast stain. Transthoracic

echocardiography revealed no vegetations. Bone marrow aspiration was unremarkable.

Since the most likely diagnosis remained cysts infection, a PET CT of the abdomen was performed. No uptake of renal or hepatic cysts was demonstrated, but two 2.6 cm retroperitoneal lymph nodes with pathological uptake were seen (Figure 1).

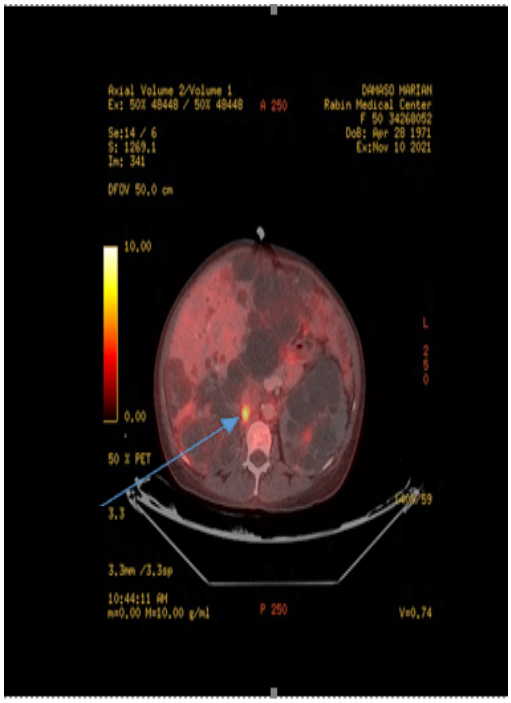

Figure 1: Abdominal PET CT with retroperitoneal lymph node uptake (blue arrow).

Due to significantly enlarged liver and multiple kidney cysts, the removal of a lymph node by a surgical approach was not feasible. Fine needle aspiration of the lymph node was performed using an angiographic approach. Cytological examination of the material revealed no evidence of lymphoma.

Because of the lack of improvement in the patient's condition, antibiotic treatment was continued. Oral therapy with doxycycline was initiated. After starting the drug, there was a gradual decrease in CRP and leukocytosis, and the fever also decreased. The patient was discharged after 24 days of hospitalization. At the same time, IgG antibody to Bartonella hensellae, drawn on the 13th hospital day was found to be positive.

In recurrent anamnesis the patient denied any contact with cats at home and in the environment.

Discussion

In this report we describe a case of *Bartonella hensellae* infection in a hemodialysis patient that presented as a FUO syndrome. Usually, typical CSD is characterized by self-limited regional lymphadenopathy or lymphadenitis, although constitutional symptoms such as fever, malaise and night sweats may occur [2]. The first attempt to characterize CSD-FUO as a unique syndrome was performed in a CSD nationwide CSD surveillance study conducted in Israel in 1991 [3]. The study included immunocompetent population of patients. To date, no similar studies have been conducted with immunocompromised patients and specifically with hemodialysis patients. In contrast to an immunocompetent population, in immunocompromised patients, bartonellosis can be particularly aggressive. Acquired immune deficiency and impaired phagocyte function have been established in maintenance hemodialysis patients. The only case of CSD in a hemodialysis patient reported by Goral et al. [4] presented with 3 week history of fever, chills, nausea and vomiting. Lymphadenopathy and hepatosplenomegaly were found on examination. Biopsy of an inguinal lymph node showed numerous perivascular bacilli organisms. A positive IgM antibody to *Bartonella hensellae* confirmed the diagnosis of CSD. The clinical picture improved rapidly after starting clarithromycin. Most cases of CSD in immunocompromised patients were reported in transplant recipients where the clinical presentation includes prolonged fever with spiking, lymphadenopathy and in more severe cases a disseminated systemic disease [5]. In immunocompetent patients, CSD is a self-limited disease lasting 2-8 weeks and can resolve without antibiotic therapy.

Most immunocompromised patients require antibiotic treatment and the choice between azithromycin, clarithromycin, rifampin, ciprofloxacin and gentamicin have been used with variable efficacy.

The case presented here is the first to describe a patient undergoing dialysis with FUO as the presenting symptom of CSD. It was also unique and diagnostically problematic. Due to enlarged polycystic liver and kidneys, it was very difficult to clarify the diagnosis by performing invasive tests, such as liver and lymph node biopsies. There were minimal clinical and imaging signs, also, the patient denied exposure to cats. Most routine laboratory test were non-specific. Ultimately, the diagnosis was based on a positive serology for Bartonella hensellae. This case highlight the dilemma of fever in ADPKD patients on maintenance hemodialysis. The diagnosis of an infected cyst remains a challenge for clinicians. The differential diagnosis is often problematic including diverticulitis, acute cholangitis or infected urolithiasis. Cyst aspiration can be useful to establish the diagnosis when available. Routine imaging such as ultrasonography and CT have a poor specificity. If available, Fluoro-Deoxy- Glucose (18F-FDG) Positron Emission CT (PET-CT) has been proven to be useful in the diagnosis of infected cyst.

In summary the presence of prolonged fever and disseminated lymphadenopathy in maintenance hemodialysis patient should

raise the suspicion of cat scratch disease.

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