

PUBLISHED ABSTRACT

Abdominal Lemierre Syndrome – An Odd Presentation of a Rare Entity

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Case description

39-year old hispanic male with no significant past medical history presented to the emergency department with complaints of sudden onset, progressively worsening, severe epigastric and right upper quadrant (RUQ) abdominal pain since 10 days. He also reported subjective fevers, chills, night sweats and unquantified weight loss for 2 weeks. On examination, he had a low-grade fever of 100.4 F, sinus tachycardia (100–110 bpm) and marked epigastric and RUQ tenderness without rebound tenderness, guarding or rigidity. Laboratory studies were remarkable for white blood cell count (WBC) of $16 \times 10^9/L$ with 84% neutrophils, total bilirubin of 2.2 mg/dl, with direct bilirubin of 1.0 mg/dl, mild transaminitis with ALT/AST of 135/98, alkaline phosphatase elevation at 355 and erythrocyte sedimentation rate of 107 mm/h. CT abdomen with contrast revealed right portal vein thrombosis (**Figure 1**). Hepatitis viral serologies, ANA, ASMA, LKM1 antibodies were all negative. He was empirically started on Ceftriaxone and metronidazole for possible pylephlebitis and therapeutic dose of enoxaparin was initiated.

Initial blood and urine cultures showed no growth. He showed marked symptomatic improvement in his abdominal pain, remained afebrile, WBC and liver function tests normalized and he was discharged on day 5 with oral apixaban and hematology follow up. On the day following discharge, blood cultures from admission grew *Fusobacterium nucleatum*. Patient was informed of the results and prescribed another 10 days of metronidazole to complete a 2-week course. During a follow-up hematology visit 1 month later, patient was completely asymptomatic. Thrombophilia workup



Figure 1: CT scan showing portal vein thrombosis.

including coagulation studies, Factor V Leiden mutation, prothrombin gene mutation, protein C and S deficiency, antiphospholipid panel, antithrombin III levels, homocysteine level and testing for JAK 2 mutation were all unremarkable. Apixaban was recommended to be continued for at least 3 months.

Discussion

The *Fusobacterium* species normally reside in the oral cavity, the female genital tract, and the gastrointestinal tract. They are anaerobic, immotile, Gram negative bacilli known for their highly thrombotic nature. *Fusobacterium necrophorum* is a notorious pathogen known to cause Lemierre syndrome, defined as a primary infection of the oropharynx causing secondary thrombophlebitis of the internal jugular vein usually resulting in septic emboli in the pulmonary capillaries [1]. Recently, there have been a few cases of *Fusobacterium nucleatum*, which normally resides in the gut, causing invasion and thrombosis of the portal vein and/or its branches (also referred to as pylephlebitis). There are limited recorded cases as the one presented in this case report. Typically, it is a complication of intra-abdominal or pelvic infections located in the drainage pathway of the portal circulation [2]. Pylephlebitis is a rare but fatal complication that needs immediate attention and rapid initiation of treatment when identified. The usual culprits are *Streptococcus viridans*, *Escherichia coli*, and *Bacteroides fragilis*. *Fusobacterium* is an extremely rare causative pathogen in these cases [1]. It has a unique ability to invade previously healthy people and they do this by the toxins they produce which are unlike those produced by other anaerobic bacilli. They have a lipopolysaccharide endotoxin similar to those in aerobic organisms, which has the lipid A component making them highly biologically active and virulent [3], leading to detrimental outcomes in affected patients.

Treatment is generally with intravenous antibiotics, specifically, a third-generation cephalosporin with metronidazole or monotherapy with a beta lactam/beta lactamase inhibitor or carbapenem for 4 to 6 weeks. Empiric treatment can be given with broad spectrum antibiotics to cover the anaerobes mentioned previously until culture results become available as bacteremia is a common simultaneous occurrence [4]. Management with anticoagulants remains a topic of controversy as there is limited outcomes data and studies on its effectiveness. Overall, there seems to be a benefit to patients if started early in order to reduce the risk of bowel ischemia and infarction [4]. The decision to use anticoagulation must be specific to each patient taking into consideration their individual benefits and risks. Unfortunately, mortality in these cases remains high in spite of advances in diagnostics, sepsis usually being the cause of death [4]. Early recognition and treatment are imperative to patient survival as seen in this case.

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