A 54 Year-Old Male with Cholangiocarcinoma and Biliary Sepsis

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Case Report

A 54 year-old male with a past medical history of cholangiocarcinoma and portal vein thrombosis was admitted to an outside hospital with right-sided abdominal pain, leukocytosis and hyperbilirubinemia. Prior to admission, he received 3 cycles of gemcitabine however his tumor had increased in size leading to development of obstructive jaundice. At the hospital, his pain was attributed to hepatomegaly and biliary obstruction secondary to tumor size. He was started on Zosyn for presumed diagnosis of pneumonia and leukocytosis. He was then transferred to Thomas Jefferson Hospital for a second opinion regarding his malignancy.

Upon transfer, patient noted right upper quadrant abdominal pain that he described as constant and dull. He also reported fatigue and weight loss over last few months. He denied nausea, vomiting, diarrhea or any change in his bowel movements.

On arrival, his vital signs were stable. Physical exam was significant for jaundice, distended abdomen and right upper quadrant tenderness. He also had significant peripheral edema up to his groin.

Laboratory studies revealed white blood count (WBC) of 24,500/μL, hemoglobin of 8.5 g/dL, and platelets of 416,000/μL. His sodium was 128 mmol/L and creatinine was 0.6 g/dL. His total bilirubin was 21.2 mg/dL and alkaline phosphatase was 320 IU/L. His INR was slightly elevated at 1.31.

The patient underwent endoscopic retrograde pancreatography (ERCP). He was found to have an occlusive common hepatic duct stricture secondary to a tumor. A plastic stent was successfully deployed. Seven days after the initial ERCP, patient became febrile, tachycardic and complained of worsening right upper quadrant pain. Repeat ERCP revealed purulent debris in the stent and a strictured left hepatic duct. Two metal stents were successfully placed leading to decreased in bilirubin. Within 24 hours, the patient's blood cultures speciated *Stenotrophomonas maltophilia*. He was started on intravenous trimethoprim-sulfamethoxazole (TMP-SMX). Once patient's bacterimia cleared, he was discharged home with hospice services.

Discussion

In the last twenty years, *Stenotrophomonas maltophilia* has been increasingly found as an important cause of infection in hospitalized patients. This hydrophilic organism has been known by multiple names. It was originally called *Bacterium bookeri* when first isolated in 1943 and considered to be a member of the genus *Pseudomonas*. It was then classified as part of the *Xanthomonas* genus in 1983 until finally reclassified as *Stenotrophomonas* in 1993. Although not considered to be a highly virulent organism, *S. maltophilia* can cause serious infections in both immunosuppressed and immunocompetent patients. It is most commonly found in aquatic or humid environments including drinking water supplies, soil and on plants. In health care facilities the organism has been isolated from medical devices, vacuum blood collection tubes, disinfectant and sterile water.[1]

Most infections do occur in severely immunocompromised patients particularly those with cancer and prolonged neutropenia. Risk factors for infection with *S. maltophilia* include extended hospitalization in critical care units, prolonged mechanical ventilation, presence of tracheostomy, indwelling devices such as intravascular catheters and endotracheal tubes, and exposure to broad-spectrum antibiotics. Most *S. maltophilia* bacteremias are related to infected in-dwelling catheters. These infections are usually easily treated with removal of the infected catheter and antibiotic therapy. In patients with non-catheter related *S. maltophilia,* bacteremia treatment failure rates and infection associated mortality are high.[2] In patients with cancer, risk factors associated with poor outcome include prolonged neutropenia, bacteremic pneumonia, shock syndrome, thrombocytopenia and inappropriate initial antibiotic choice.[2]

The respiratory tract is the most common site of *S. maltophilia* infection however it has been found to be the cause of skin infections, endocarditis, urinary tract infections and hepatobiliary infections. There have been five reports in the literature of biliary sepsis secondary to *S. maltophilia*. All of these cases occurred in patients with hepatobiliary malignancy complicated by biliary tract obstruction. Each of these patients had undergone biliary tract instrumentation prior to developing *S. maltophilia* bacteremia.[3,4,5] The organism was recovered from blood in three cases and from bile in the remaining two cases. All of the patients were treated with antibiotics, four out of five patients described made a full recovery, the fifth patient died. The four patients who survived underwent interventional procedures in addition to receiving antibiotics.

Choosing appropriate antibiotic therapy for *S. maltophilia* infections can be difficult and initiation of inappropriate antimicrobial therapy has been linked to poor outcome. Although trimethoprim-sulfamethoxazole (TMP-SMX) has the strongest in vitro activity against *S. maltophilia,* increasing resistance rates have been reported in the literature. Despite these reports, TMP-SMX is still considered the drug of choice against this organism.[2] It has even been suggested that patients who have serious *S. maltophilia* infections and are allergic to sulfa drugs should undergo desensitization in order to be treated appropriately.[4] Other classes of antibiotics such as aminoglycosides and quinolones are not as effective against *S. maltophilia.*

S. maltophilia, although not considered an aggressive pathogen, remains an important cause of morbidity and mortality among immunosuppressed patients. Patients with cancer particularly those with prolonged neutropenia continue to be at risk of

infection with this organism. When *S. maltophilia* infection is suspected it is important to obtain sensitivity and susceptibility data and to institute treatment with TMP-SMX when possible. This organism commonly causes respiratory and catheter-related infections, however there are rare reports of urinary and hepatobiliary infections in the literature. When a patient with hepatobiliary malignancy and biliary tract obstruction presents with cholangitis or biliary sepsis infection, *S. maltophilia* should be considered in the differential of possible pathogens.[3]

References

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"Ben Franklin Bridge"
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