

Endotipsitis: Atypical Risk Factor for *Lactobacillus* Bacteremia

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Background

- Transjugular intrahepatic portosystemic shunt (TIPS) is commonly used for decompressing portal venous pressure.
- Endotipsitis is a rare complication of TIPS that is understudied, with only a limited number of cases reported in the literature.
- Endotipsitis may also be underdiagnosed since there is no universally accepted identification or classification.
- Few cases have been reported with *Enterococcus spp*, *Staphylococcus spp*, and *Escherichia coli* as a source.
- *Lactobacillus* bacteremia from endotipsitis is extremely rare.
- This report presents a patient with atypical signs associated with *Lactobacillus* bacteremia, resulting in a diagnosis of endotipsitis.

Case Description

Presentation:

- A 64 y.o. woman with a PMHx of nonalcoholic steatohepatitis, portal hypertension, ascites, esophageal varices with bleeding, s/p TIPS procedure 9 months ago presented with fever, somnolence, and progressive lethargy for 3-4 days.

Physical Examination:

- Remarkable for bilateral scleral icterus, oral temperature of 38.9°C, HR 144, BP 90/70 mm Hg, and RR 18/min.
- No peripheral edema, asterixis, or abnormal heart/lung sounds, and the abdominal exam was benign.

Laboratory Evaluation:

- CBC: anemia (11.9 g/dl), thrombocytopenia (94,000), WBC (5)
- CMP: hyperbilirubinemia (3.1 total, 1.0 direct), AST 120, ALT 62, ammonia 40, ALP 114.

Imaging and Management

Imaging:

- Chest X-ray: no acute cardiopulmonary abnormalities.
- RUQ Ultrasound: a patent TIPS and no evidence of ascites.
- CT abdomen and pelvis: patent TIPS, changes related to chronic liver disease, and no obvious infection source.
- Transthoracic echocardiogram: normal and did not show any vegetations.
- Ruled out obvious source of infection.

Impression/Management:

- Considering sepsis-like syndrome, obtained 2 separate blood cultures → started vancomycin and piperacillin-tazobactam.
- With history of TIPS and no other source of infection, a “probable” diagnosis of endotipsitis was added to differential diagnosis.

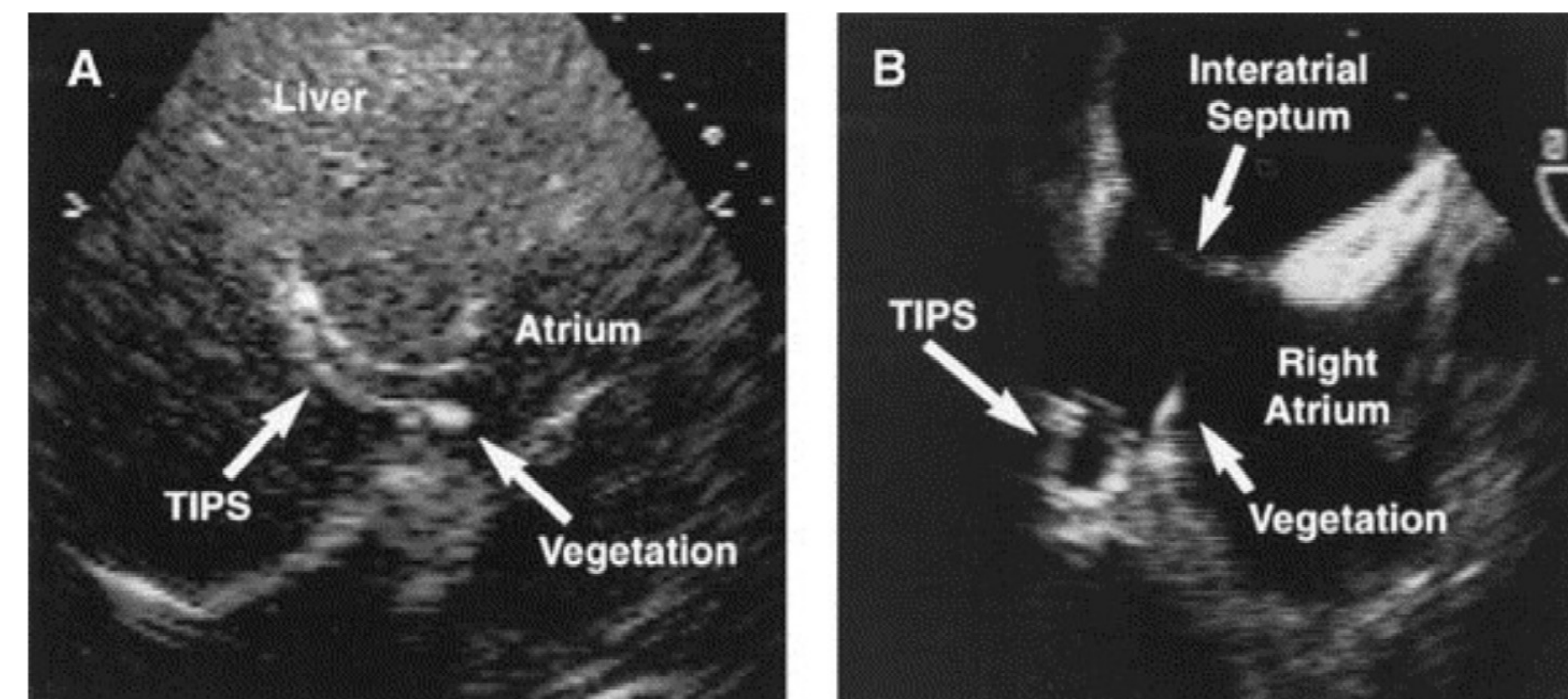


Fig 2 (A) Sonographic evidence of vegetation at the proximal end of a TIPS. (B) The stent extends back into the right atrium observed on transesophageal echocardiography at the atrial end of the stent.

Endotipsitis Definitions

Sanya et al. “Definite” infection: “continuous, clinically significant (positive blood cultures and fever) bacteremia, with vegetations or thrombi inside the TIPS.”

Sanya et al. “Probable” infection: “sustained bacteremia and unremitting fever in a patient with an apparently normal TIPS without an identified site of infection elsewhere in the body.”

Armstrong and MacLeod: “A sustained bacteremia in a patient with a TIPS device, with or without thrombus, plus either no other identifiable infective focus or an identifiable infective focus that is not considered to be the source of the bacteremia after an exhaustive workup.”

Mizrahi: “endotipsitis involves a persistent bacteremia and fever with either shunt occlusion, vegetation, or bacteremia in the presence of a patent shunt, and when other sources of bacteremia have been ruled out.” → our patient

Table 1. Number of cases reported in the literature review for each isolated microorganism causing endotipsitis and the associated outcome.

Etiological agent	Number of cases reported	Outcome death
Enterococcus spp (faecalis, faecium)	14	3
Staphylococcus spp (aureus, epidermidis, MRSA)	9	4
Escherichia coli	7	3
Candida spp (glabrata, albicans)	7	4
Lactobacillus spp (rhamnosus, acidophilus)	4*	2
Streptococcus spp (sanguis, bovis)	2	0
Gemella morbillorum	2	0
Lactobacillus spp (rhamnosus, acidophilus)	3	2
Klebsiella spp (pneumonia, oxytoca)	3	1
Serratia marcescens	1	0
Enterobacter cloacae	1	0
Salmonella typhi	1	0
“Polimicrobial infection”	6	1
Total	56	18 (32.1%)

The most recent literature review from 2017 reported 56 cases of endotipsitis, with 32.1% of those cases resulting in death. This report would result in the fourth case of *Lactobacilli* as an etiologic agent for endotipsitis.

Discussion

- While *Lactobacillus* bacteremia accounts for a mere 0.1% of all positive blood cultures and is often ascribed as contamination, the attributable mortality remains as high as 30%.
- If suspected, workup should start with a color doppler ultrasound to evaluate TIPS patency, with exclusion of all other possible sources of bacteremia.
- A tagged white blood cell scan, Indium-111-labeled leukocyte scan, can also assist by demonstrating increased leukocyte uptake around the TIPS.
- Gold standard diagnosis involves removing the TIPS and culturing it; however, this is not clinically or practically feasible unless there is a liver transplant at the time of TIPS removal.
- Initial treatment is empirical with broad-spectrum antibiotics

Conclusions

- Endotipsitis is a prosthetic infective complication that is an uncommon yet possibly life-threatening condition due to potential evolution to sepsis and death.
- Currently, no uniformly accepted diagnostic criterion.
- Most cases diagnosed from evidence of bacteremia in patients with TIPS and no other identifiable source.
- *Lactobacillus* bacteremia should not always be considered contamination, especially in patients with TIPS.
- Awareness among the clinicians regarding such a condition needs to be enhanced to avert underdiagnosis.
- Future studies are required to standardize diagnostic and treatment algorithms as endotipsitis remains an uncommon infection with significantly high morbidity and mortality.

References

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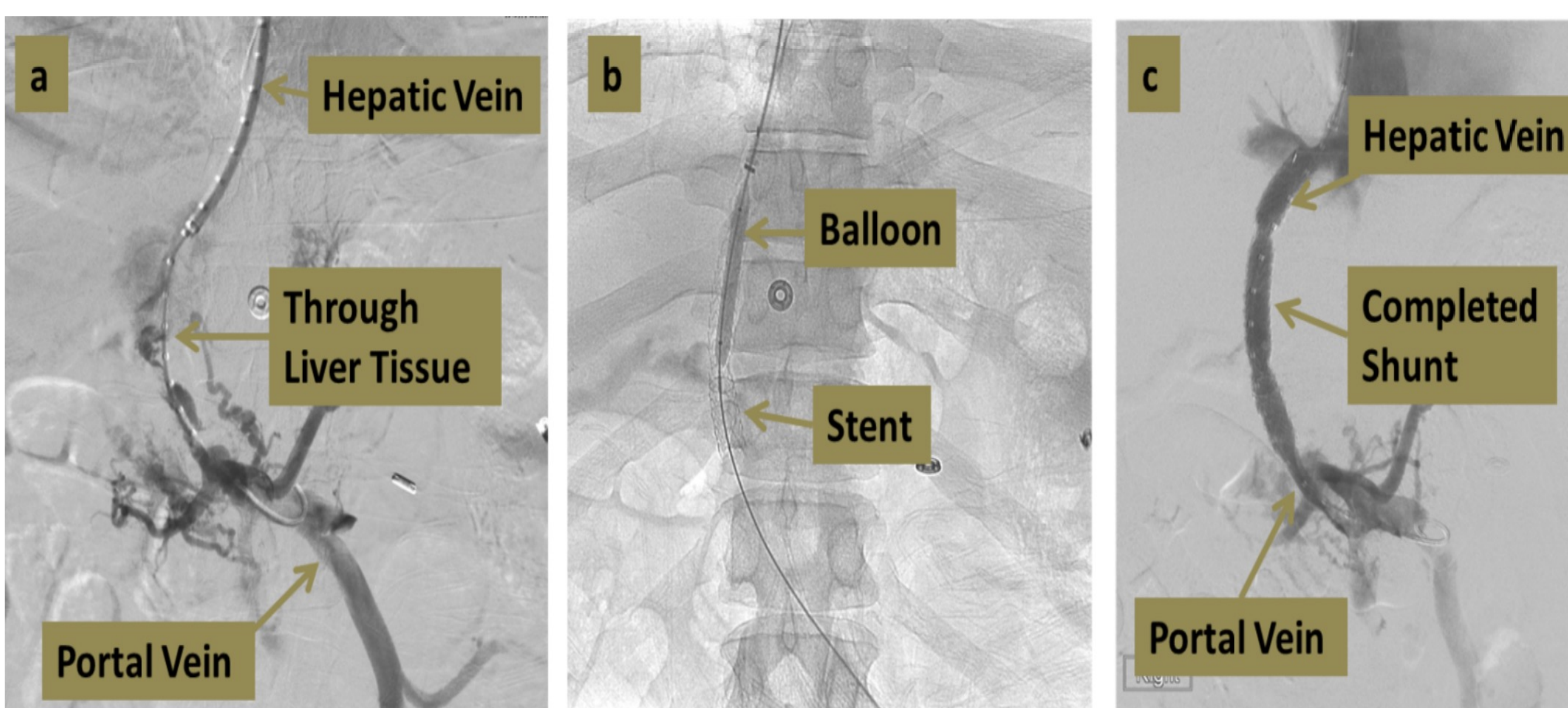


Fig 1 a) Stent extending from the hepatic vein, through liver tissue and into the portal vein. b) Expansion of the stent through the liver tissue by inflation of a balloon. c) Completed shunt allowing blood to flow directly from the portal vein to the hepatic vein.

Clinical Outcome

- Blood cultures grew GM+ bacteria → finalized to be *Lactobacillus* → Abx narrowed to piperacillin-tazobactam.
- Discharged on a 21-day course of IV piperacillin-tazobactam with a plan to repeat blood cultures at that time.
- If repeat blood cultures turned positive, further workup would be pursued, including WBC tagged scan/EGD.
- Completed the antibiotic course without further complications.
- Remained asymptomatic and repeat cultures at day 28 also remained negative.