Casting a Wider Net: Rare Presentation of Biliary Cast Syndrome in a Non-Liver Transplant Patient

Shil Punatar, DO[1], Ahamed Khalyfa, DO[1],Rida Khan, BS, Zarek Khan, BS[1]
Faizan Khan, MD[1,2], Fares Hamad, DO[2]
Department of Gastroenterology, Franciscan Health Olympia Fields IL [1] Department of Gastroenterology, AMITA St. Joseph, IL[2]

INTRODUCTION

- Originally described in 1975, biliary cast syndrome (BCS) pertains to pigmented dark material molding into intra and extra hepatic bile ducts.
- Primarily described in literature in post-liver transplant patients, BCS reportedly occurs in 2.5 to 18% of patients with varying degrees of severity and patient outcomes (Koo)
- Rarely described and published are cases of BCS in non-transplant patients. Here, we shed light on a case of BCS in a non-transplant patient presenting with a primary complaint of infected decubitus ulcers with isolated alkaline phosphatase elevation.
- While adding to medical literature a non-transplant BCS case, we shed light on the pathogenesis, pathotyping, and treatment of BCS in an effort to recognize and reduce potential patient morbidity and mortality.

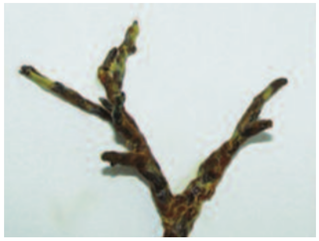
CASE PRESENTATION

The patient is a 57-year old male with past medical history of diabetes mellitus, hypertension, hyperlipidemia, prior venous thromboembolism on warfarin who presented with infected decubitus ulcers and sepsis.

His initial laboratory evaluation was significant for an incidental finding of elevated alkaline phosphatase level of 713 but total bilirubin level of 0.7 and AST/ALT levels of 17 and 11 respectively.

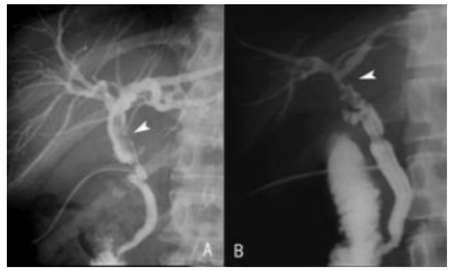
He had a subsequent CT abdomen and pelvis as part of his infectious workup which revealed an incidental finding of a 1.4cm x 1.1 cm lesion of the pancreatic head that was described as a likely pseudocyst. The patient reported no abdominal pain and his sepsis was treated with IV antibiotics and fluids only, as he remained with a stable hemodynamic profile.

ENDOSCOPIC RETRIEVAL AND IMAGING



Top Right: Biliary cast noted on gross imaging to be brown in color with antler shape
Top Left: Yellow brown biliary cast on retrieval
Center: Described as leafless biliary cast with tree shaping

Below: Sparse biliary cast indirect imaging noted within common hepatic duct, with smooth bile duct. In right image, filling defect noted in left hepatic duct with smooth bile duct contour.



Due to institutional restrictions, images are derived from cited studies.
Images utilized cited as Zhu et al

REFERENCES & ACKNOWLEDGEMENTS

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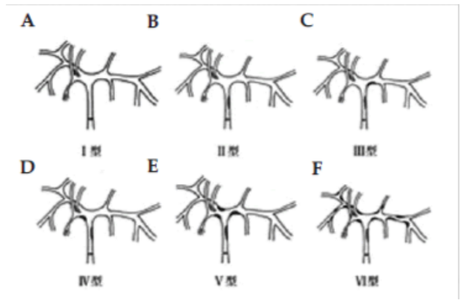
2. Koo JW, Jang NE, Lee HJ, et al. A Case of Biliary Cast Developed in a Patient with Long-Standing Biliary Sludge. *Clin Endosc*. 2013;46(1):98-101. doi:10.5946/ce.2013.46.1.98

Special thanks to Franciscan Health Division of Research for the opportunity and funding for research and presentation endeavors as well as the providers involved in the care of this interesting patient.

MANAGEMENT & OUTCOME

- The patient was evaluated by gastroenterology and underwent EGD with EUS and FNA which revealed no cytological evidence of malignancy but did reveal a lesion in the common bile duct suspicious for polyp.
- A subsequent ERCP was performed which revealed a blockage in the bifurcation of the right and left hepatic ducts.
- At this time, there was concern for biliary parasite infestation, however final pathology reports demonstrated paucicellular proteinaceous fluid containing degenerated pancreatic tissue and chronic inflammatory cells suggestive of biliary casts.
- According to the limited data, most of the non-transplant patients underwent ERCP for evaluation and removal of biliary casts, considering this to be a reasonable step prior to consideration of laparotomy.
- This significantly varies in transplant patients as only 25 percent of BCS patients with liver transplant had effective endoscopic evaluation with many having need for further retransplantation (Koo)

DISCUSSION



Type 1: intact bile duct epithelium, pure BC
Type 2: necrotic epithelium in common hepatic duct
Type 3: necrosis of common/left hepatic duct
Type 4: necrosis of common/right hepatic duct
Type 5: necrosis of common and bilateral ducts
Type 6: necrosis of common, bilateral, and other sources