Transjugular Intrahepatic Portosystemic Shunt for the Treatment of Chylothorax and Chylous Ascites in Cirrhosis: A Case Report and Systematic Review of the Literature

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ABSTRACT

Cirrhosis-related chylothorax and chylous ascites are rare conditions. The pathophysiologic mechanism of cirrhosis-related chylous fluid collections is believed to be excessive lymph flow resulting from portal hypertension. Transjugular intrahepatic portosystemic shunt (TIPS) creation is a well-established method for reducing portal hypertension. The case of a 61-year-old man with cirrhosis-related chylothorax treated successfully with a TIPS is described. A systematic review of the literature revealed nine additional cases of chylothorax or chylous ascites treated successfully with a TIPS. These cases showed that TIPS creation may be effective and safe for the treatment of chylous fluid collections in patients with cirrhosis.

ABBREVIATION

TIPS = transjugular intrahepatic portosystemic shunt

Chylothorax and chylous ascites are uncommon clinical entities characterized by the accumulation of chylous fluid in the pleural and peritoneal spaces (1,2). These conditions are caused by obstruction or disruption of the lymphatic system and can lead to chyle depletion, which results in nutritional, metabolic, and immunologic deficiencies (2,3). The causes of chylous fluid collections can be categorized as traumatic or nontraumatic. Malignancy is the most common cause of nontraumatic chylothorax and chylous ascites, and the remaining cases are attributed to various disorders, such as infection and cardiac disease, or are considered idiopathic (4,5).

Cirrhosis is a rare cause of chylothorax and chylous ascites (6,7). The underlying pathophysiologic mechanism of the formation of chylous fluid collections in cirrhosis is believed to be associated with excessive hepatic and gastrointestinal lymph flow secondary to portal hypertension (2,7). Transjugular intrahepatic portosystemic shunt (TIPS) creation is a well-established method for the reduction of portal hypertension (8). We report a case of cirrhosis-related chylothorax successfully treated with a TIPS. We also performed a systematic review of the literature to evaluate the outcomes of TIPS creation for the treatment of chylothorax and chylous ascites.

CASE REPORT

Institutional review board approval is not required for case reports at our institution. A 61-year-old man with compensated cirrhosis secondary to alcohol abuse was admitted to our center for evaluation of dyspnea. His Model for End-Stage Liver Disease score was 12, and his Child-Pugh score was 6 (class A). He had no history of recent trauma.

Physical examination demonstrated stigmatas of chronic liver disease and decreased breath sounds in the right lung fields. The laboratory results were hemoglobin, 17.9 g/dL; white blood cell count, 10.3 $\times$ 10^3/mm^3 (neutrophils, 73.9%; monocytes, 12.1%; lymphocytes, 9.1%); total bilirubin, 1.9 mg/dL; glucose, 107 mg/dL; total protein, 5.4 g/dL; albumin, 3.1 g/dL; triglyceride, 60 mg/dL; cholesterol,

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196 mg/dL; lactate dehydrogenase, 429 IU/L; international normalized ratio, 1.32; and creatinine, 0.6 mg/dL. A contrast-enhanced computed tomography (CT) scan of the chest and abdomen showed changes consistent with cirrhosis and portal hypertension and a large, right-sided pleural effusion, although with no signs of infection or malignancy (Fig 1a). Echocardiography ruled out cardiac causes for the effusion. Diagnostic thoracentesis revealed a milk-colored fluid with 418 mg/dL of glucose, 0.5 g/dL of total protein, 0.2 g/dL of albumin, 130 mg/dL of triglyceride, 9 mg/dL of cholesterol, and 84 IU/L of lactate dehydrogenase. Microbiologic and cytologic studies were negative for organisms or malignant cells.

A diagnosis of chylothorax was made on the basis of the presence of pleural fluid with a triglyceride level $>$ 110 mg/dL, and the cause was thought to be cirrhosis-related because the patient had no history of recent trauma or evidence of infection, malignancy, or cardiac disease. The patient was placed on diuretics (spironolactone 150 mg/d, furosemide 80 mg/d) and sodium restriction, a low-fat and medium-chain triglyceride diet, and bowel rest and total parenteral nutrition in sequence over a 3-week period and with repeated therapeutic thoracentesis performed as necessary. However, no treatment response was observed except for a reduction in the pleural fluid triglyceride concentration (80 mg/dL), and he was ultimately referred for TIPS creation.

The TIPS procedure was performed with the patient under conscious sedation. Access to the right internal jugular vein was obtained, and the middle hepatic vein was catheterized. The right portal vein branch was punctured with a TIPS set (RTPS-100; Cook, Inc, Bloomington, Indiana) under fluoroscopy, and a 10-mm-diameter, 8-cm-long expanded polytetrafluoroethylene–covered stent graft (Niti-S; Taewoong Medical Co, Ltd, Seoul, Korea) was placed to cover the liver parenchymal tract. An additional 10-mm-diameter, 8-cm-long bare stent (Zilver; Cook, Inc) was placed to cover the hepatic vein segment. A portogram obtained immediately after the TIPS creation showed good passage of contrast medium through the shunt (Fig 1b). The portosystemic pressure gradient was decreased from 26 mm Hg before TIPS creation to 6 mm Hg after TIPS.

The patient had an uneventful recovery after TIPS creation and had complete resolution of chylothorax after 11 days with only diuretics (spironolactone 100 mg/d; furosemide 40 mg/d) and sodium restriction. No further treatment for chylothorax was necessary. He remained well for 2 years with good liver function before developing hepatocellular carcinoma, which eventually led to his death 11 months later. A contrast-enhanced CT scan of the chest and abdomen obtained 1 month before his death showed a patent shunt and no recurrence of pleural effusion (Fig 1c).

SYSTEMATIC REVIEW

Methods

Institutional review board approval is not required for review articles at our institution. A MEDLINE search (PubMed) from January 1990 to August 2015 was conducted to identify eligible studies using the following terms: “Transjugular Intrahepatic Portosystemic Shunt” (all fields) AND “Chylothorax” (all fields) OR “Transjugular Intrahepatic Portosystemic Shunt” (all fields) AND “Chylous Ascites” (all fields). The inclusion criteria for studies were patients with chylothorax or chylous ascites, or both, who underwent TIPS creation. A triglyceride level $>$ 110 mg/dL or the presence of chylomicrons in pleural and ascitic fluid were used to confirm the diagnosis of chylothorax and chylous ascites, respectively (9,10). The exclusion criteria for studies were a language other than English and duplicate data. Two reviewers (J.T., K.C.H.)

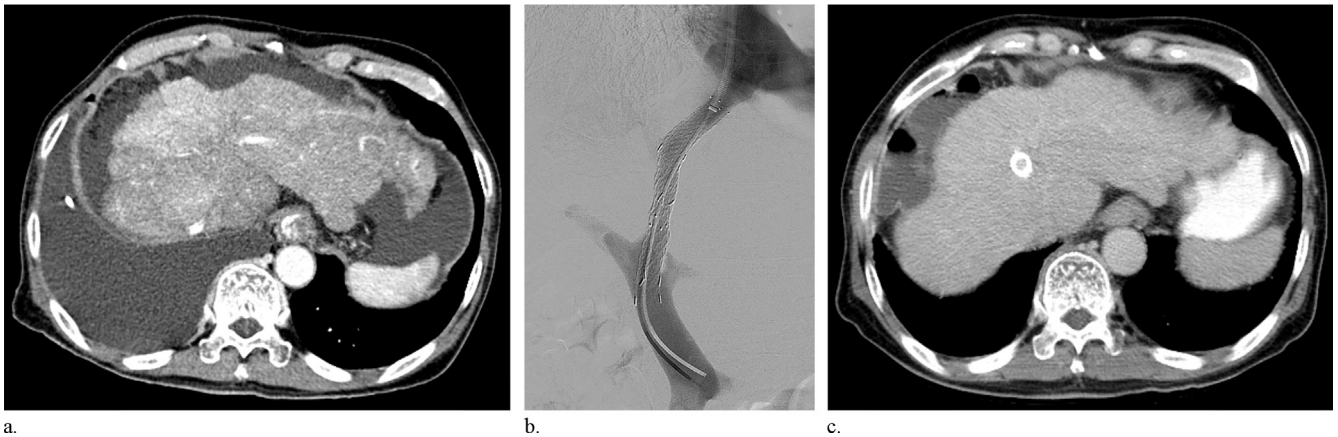

Figure 1. **(a)** Contrast-enhanced CT image of the chest and abdomen obtained on hospital admission shows a large, right-sided pleural effusion. **(b)** Portogram obtained immediately after TIPS creation shows good passage of contrast medium through the shunt. **(c)** Contrast-enhanced CT image of the chest and abdomen obtained 2 years and 10 months after TIPS creation shows no recurrence of pleural effusion.