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Case Report

Intrahepatic lymphatic channel sclerotic embolization for treatment of postoperative lymphatic ascites: a report of 3 cases ☆,☆☆

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ABSTRACT

Postoperative hepatic lymphorrhea is extremely rare and there is no standard treatment for this condition. We report the cases of 3 men, 32-, 56-, and 37-year-old, with postoperative hepatic lymphorrhea, which was refractory to conservative treatment. Transhepatic lymphangiography allowed locating the lymphatic leak and treating it with hepatic lymphatic vessels injection of foam sclerotic agent. This technique seems efficient and safe.

Background

Lymphatic ascites due to postoperative hepatic lymphorrhea is extremely rare, with only few case reports in the literature [1–3] and poses diagnosis difficulties. Indeed, although the biochemistry's profile of peritoneal fluid is typical of lymphorrhea with high protein concentrations and low triglycerides levels, conventional intranodal lymphangiography usually cannot visualize the leakage point. Definitive diagnosis is based on transhepatic lymphangiography that can visualize hepatic lymphatic system and thus reveals the leakage point. Various

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☆☆ Written informed consent was obtained from the three patients describes in this case report.

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treatment are described, including surgery with intermittent abdominal drainage [2], retrograde injection of OK-432 at the leakage cavity [1], and embolization of intrahepatic lymphatic vessels using Onyx [3]. In this study, we report three patients with hepatic lymphatic ascites treated successfully by anterograde injection of 2% lauromacrogol foam into intrahepatic lymphatic vessels.

Case 1

A 32-year-old Vietnamese man had a partial gastrectomy for pylorus adenocarcinoma. Early postoperative period was unremarkable and he returned to a normal diet 7 days after surgery. At day 10, he complained of a swollen abdomen. Abdominal ultrasound found a large amount of ascites, which biochemistry's profile showed low level of triglyceride (0.5 mmol/L) but high concentration of protein (30.6 g/L). Cellblock cytology, microbiology, and tuberculosis tests were negative. A peritoneal drain (pigtail 8F, Bioteq, Taiwan) was inserted, which allowed removing 900-1400 mL of yellow peritoneal fluid per day during 2 months.

The patient was then sent to our hospital for suspicion of lymphatic leakage. A dynamic intranodal magnetic resonance lymphangiography (MRL) with the technique previously described [4] was performed, showing normal cisterna chyli and thoracic duct, without any sign of leakage (Fig. 1A).

A transhepatic lymphangiography was then performed, in order to explore the lymphatic vessels of the hepatic hilum. We punctured into the periportal vein space using a 25G-needle (Chiba, Cook medical, Bloomington, IN 47402-4195 U.S.A.) under ultrasound guidance. The target puncture was the anterior branch of the right portal vein and the left trunk of the portal vein. Then 10 mL of nonionic contrast (Xenetic 350, Guerbet, France) was injected into the needle on the right side and the left side, while retracting the needle until visualization of the lymphatic vessels. The procedure was done under local anesthesia. We succeeded in depicting the intrahepatic lymphatic vessels that go parallel with portal veins (Fig. 1B). Extravasation of contrast material at the hepatic hilum was also visualized when we injected the contrast through the needle on the right liver (Fig. 1B). We decided to embolize the lymphatic vessels on the right hepatic lobe. Six mL of foam sclerosant (the mixture included 2 mL of lauromacrogol - Aetoxisclerol 2%, Kreussler Pharma, France – and 4 mL of air) was injected through a needle.

After sclerotherapy, drainage volume was significantly reduced. Three days after, there was no more liquid and peritoneal pigtail drain was removed at day 5. The patient was discharged from the hospital at day 7. Six months later, on follow-up consultation, there was no recurrence of ascites and his liver function blood markers remained normal. He was also in complete remission of his gastric cancer.

Case 2

A 56-year-old Vietnamese patient was admitted to our hospital for suspicion of lymphatic leakage. He had a 7 months history of ascites, occurring after total gastrectomy for gastric adenocarcinoma. Biochemistry, cytology, and microbiology profiles of the peritoneal fluid ruled out carcinomatous, tuberculous, and other infectious causes of ascites. He also had a peritoneal drain inserted for 7 months, which drained 1000-1500 mL of fluid per day. The protein and glyceride concentration at arrival was 43.9 g/L and 0.7 mmol/L, respectively.

MRL showed no extravasation. The following transhepatic lymphangiography did not demonstrate any leakage point (Fig. 2A) but the abdominal computed tomography scan without contrast performed right after this procedure showed presence of contrast, mainly located in the gall bladder's bed and in the hepatic hilum (Fig. 2B). We decided to inject sclerotic agent into the intrahepatic lymphatic vessels. We first punctured into periportal space by a 25-G needle (Chiba, Cook company) and opacified the lymphatic vessels by nonionic contrast (Xenetic 350). Through the needle, a total of 2 mL of lauromacrogol (Aetoxisclerol 2%, Kreussler Pharma, France) mixed with 4 mL of air was injected into the lymphatic vessels. After intervention, drainage of peritoneal fluid reduced significantly at day 3 (below 100 mL per day). The drain was removed at day 7. There was no changing in liver function at day 7 and 3 months after the procedure.

Case 3

A-37-year-old man had a clear-colored ascites one day after partial gastrectomy for gastric adenocarcinoma. Biochemistry of ascites fluid highly suggested lymphatic fluid and ruled out other causes (carcinoma, tuberculous, parasitic or bacterial infections). Protein and triglyceride levels in paracentesis fluid were 43.4 mg/L and 0.7 mmol/L, respectively. A peritoneal drain was inserted, allowing removal of 1-2 L of ascites per day during one month before the patient was referred to our hospital. Intranodal MRL did not show extravasation of contrast. Transhepatic lymphangiography demonstrated a leak of contrast material at the liver hilum (Fig. 3A). Therefore, sclerotic injection was performed through the lymphangiography needle, with totally 6 mL of sclerotic foam (lauromacrogol mixed with air at the ratio 1:3). Computed tomography scan done just after the procedure showed that air bubble was distributed in lymphatic vessels, hepatic hilum, and retroperitoneal lymphatic vessels. The volume of peritoneal fluid drainage reduced significantly after the procedure but still remained about 500 mL per day for a week. We decided to repeat injection of sclerosant. After the second intervention, the volume of drain reduced and the drainage was withdraw after one week. His liver function remained normal at 3 months of follow-up.

Discussion

Postoperative chylous ascites is well known but hepatic lymphorrhea (not containing chyle), due to damage of the gastroduodenal ligament, is extremely rare. Lymphatic fluid from liver contributes approximately 20%-50% lymphatic volume of thoracic duct [5]. Almost the totality of hepatic lymph is

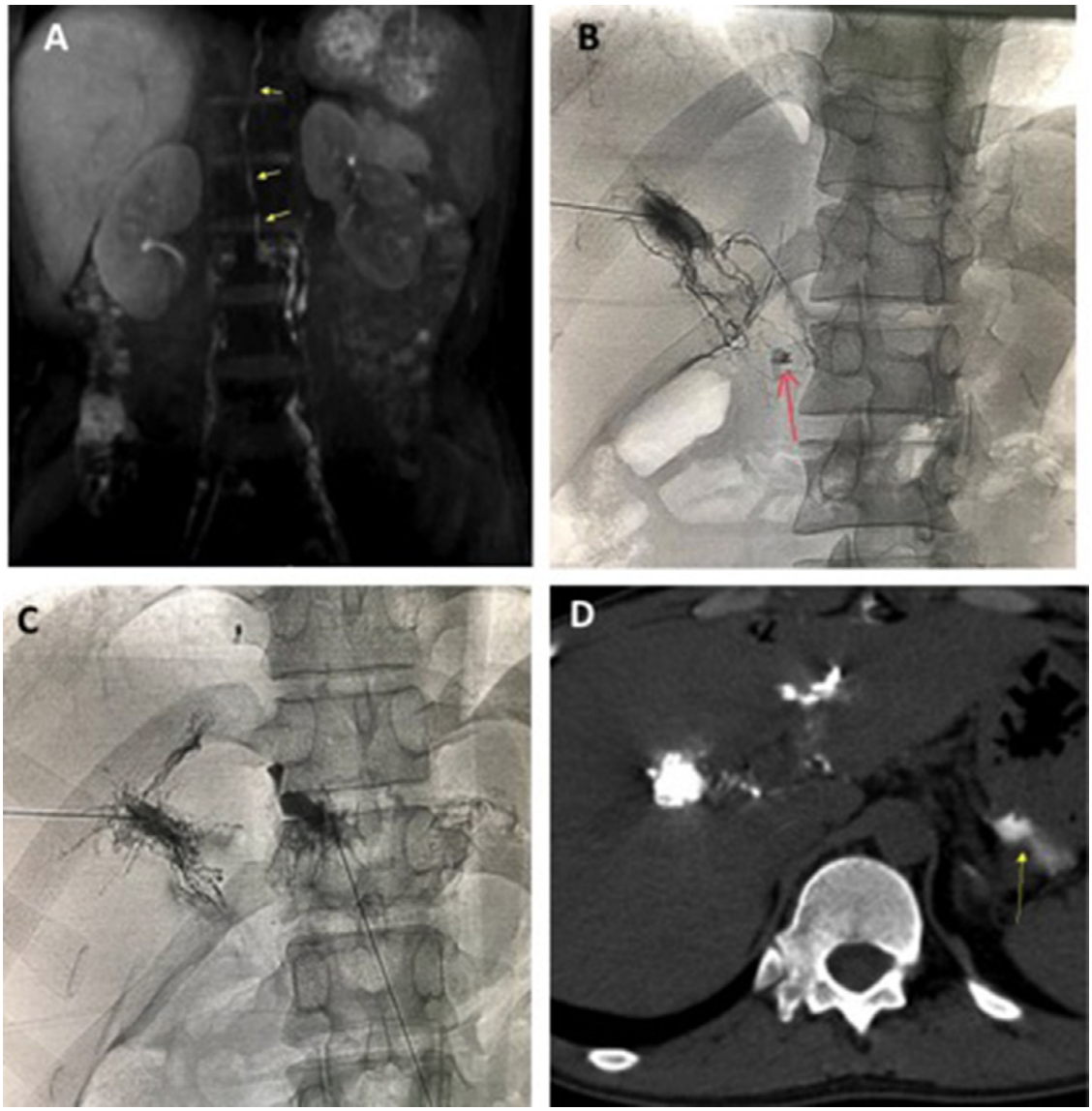

Fig. 1 – Case 1. Panel A: MRI lymphangiography showing normal thoracic duct and no extravasation of contrast. Panel B and C: Transhepatic lymphangiography showing an extravasation of contrast at the liver hilum (arrow). No contrast extravasation was seen when the left hepatic lymphatic channel was opacified. Panel D: Noncontrast CT scanner just after the procedure revealing presence of contrast in the peritoneum.

drained through the gastroduodenal ligament by tortuous tiny lymphatic canals. So, any surgery with dissection of that ligament can cause hepatic lymphorrhea.

Diagnosis of postoperative hepatic lymphorrhea is based on biochemistry's profile of peritoneal fluid and exclusion of other causes of ascites (carcinomatous, tuberculous, or other infectious peritonitis). Hepatic lymph usually appears clear-colored, on the contrary of the milky appearance of chylous ascites, and it contains high concentration of protein, albumin but very low level of triglycerides [5]. In our series of patients, the level of protein in paracentesis fluid was much higher than that of triglyceride. Because of protein and albumin loss through ascites, the level of these parameters was lower than normal. This observation was true in all our 3 patients.

After demonstration that the peritoneal fluid results from lymphorrhea, finding the leakage point permits to precisely guide the treatment. On the contrary of MR lymphangiography, percutaneous transhepatic lymphangiography is the only method to depict the hepatic lymphatic vessels. It is a minimally invasive technique and can be performed under local anesthesia.

Few case reports in literature described treatment of postoperative lymphorrhea [1, 2]. Inoue et al described peritoneovenous shunt to treat hepatic lymphorrea [6]. Guez et al [3] embolized intrahepatic lymphatic vessels by Onyx. Bartoli et al [2] treated the patient by intermittent opening of the drainage and the patient hospitalized for 181 days. Recently, Kojima et al [1] used OK-432 for sclerosing the lymphatic collection at the hepatic concaves.

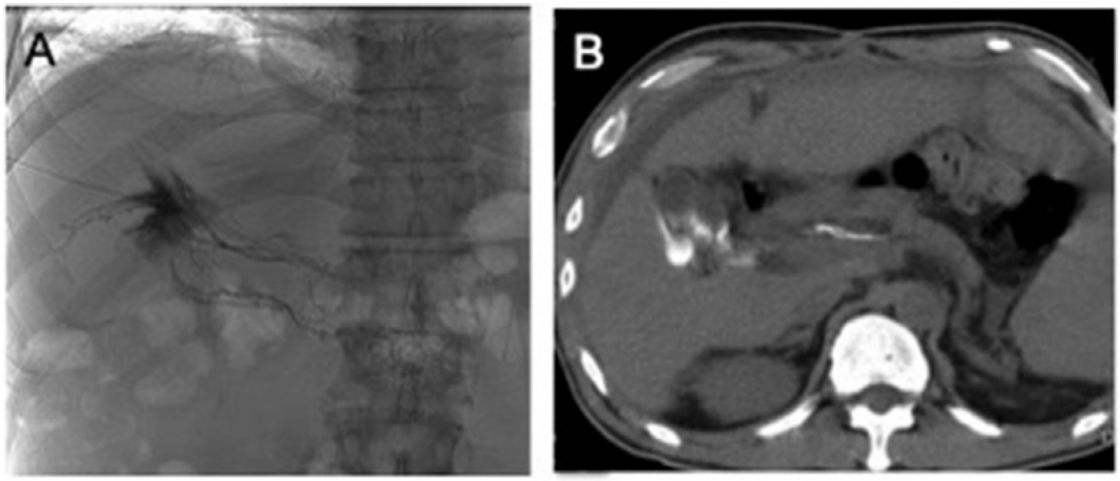

Fig. 2 – Case 2. Panel A: Transhepatic lymphangiography showing no abnormality. Panel B: Noncontrast abdominal CT-scan just after lymphangiography showing presence of intraperitoneal contrast.

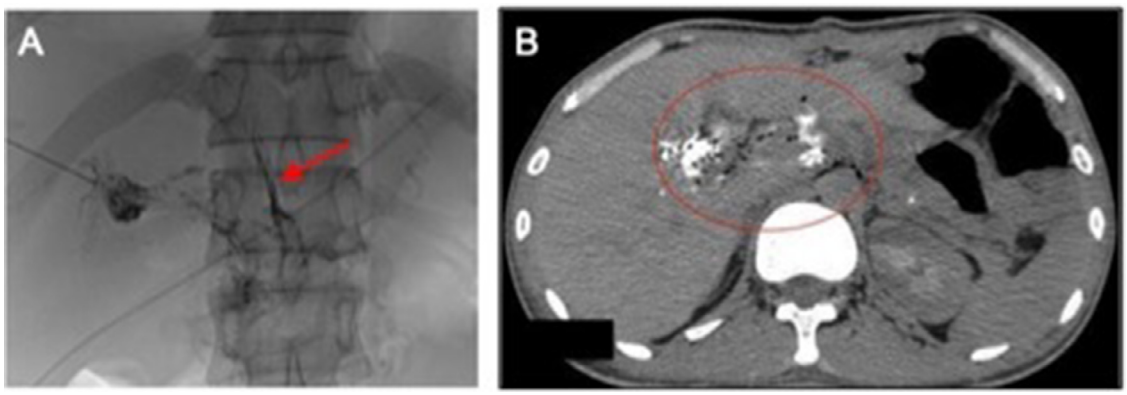

Fig. 3 – Case 3 Panel A: Transhepatic lymphangiography showing extravasation of contrast into the abdomen (arrow). Panel B: Conbeam CT was done after injecting foam, showing presence of air bubbles in lymphatic vessels at the hepatic hilum and lymphatic vessels retroperitoneal due to reflux (in the circle).

In theory, embolization of intrahepatic lymphatic vessels might be complicated with liver dysfunction. A case report in which lymphatic vessels were embolized by nonadhesive glue (Onyx, EV 3) [3], described a remaining normal liver function after the procedure, as in our 3 patients. Thus, embolization of hepatic lymph vessels appears to be safe; however, further studies with a larger number of patients are needed to conclude.

We used sclerotic foam injection into lymphatic vessels in order to stop leakage point at the liver hilum. The advantage of foam is that it can flow easily into the small lymphatic vessels and make these vessels occluded because of sclerotic reaction. This procedure can be performed in repeated sessions to achieve total occlusion of the leakage point. In our patients, we needed maximum 2 sessions.

After embolization of the lymphatic vessels at hepatic hilum, circulation of lymphatic fluid in the liver may change. There are numerous lymphatic channels at hepatic hilum and other lymphatic vessels may work as collaterals. Another theory based on superficial hepatic lymphatic vessels. There are 2 hepatic lymphatic systems: one deep with lymphatic vessels running around portal branches in the portal space that drain lymph into cisterna chyli; and one superficial with lymphatic vessels running along the hepatic vein or the subcapsular part of liver that drain lymph into central venous system [7]. Therefore, after embolization of lymphatic vessels at hepatic hilum, the superficial lymphatic system might develop into collateral circulation. So we considered that there was no change in liver function at the short time following up.

Conclusion

Transhepatic lymphangiography followed by sclerotherapy appears to be a safe and effective method to localize and occlude the source of postoperative lymphatic ascites.

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