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Case Report

Utilization of Distilled Water Lavage for Localized Fluid Collection After Combined Hepatectomy and Cyst Fenestration for Polycystic Liver Disease

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Ascites necessitating persistent drainage or paracentesis after drain removal has been found among early postoperative complications after combined hepatectomy and cyst fenestration (CHCF) for polycystic liver disease (PLD). It has been reportedly observed in 20% to 70% of cases and seemed to easily cause recurrent symptoms unless properly treated. We utilized distilled water lavage for treating localized fluid collection after combined hepatectomy and cyst fenestration for PLD. A 63-year-old female patient underwent CHCF for PLD, which caused severe abdominal fullness. Early postoperative course was uneventful until 10 days after surgery when the patient suffered sudden abdominal fullness and resultant severe anorexia because of right subphrenic massive localized monolocular fluid collection diagnosed by abdominal computed tomography—although total liver volume was reduced to less than half of that before surgery. Percutaneous drainage relieved symptoms immediately, but the drainage tube could not be removed because of massive outflow. Then we utilized distilled water lavage for treating this condition. After that, drain outflow dramatically reduced and the drainage tube was successfully removed. Total liver volume of the patient continued to reduce up to 1 year after surgery and retained less than one-third of preoperative total liver volume thereafter. Sustained reduction of total liver volume in the present case suggested a sclerosant effect of hypotonic cytocidal property of distilled water for cyst endothelium

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and/or retrieved effectiveness of fenestration. Hence, we consider this approach to be useful for patients with PLD receiving CHCF and thus present it here.

Key words: Polycystic liver disease – Combined hepatectomy and cyst fenestration – Postoperative complication – Distilled water lavage – Total liver volume

Polycystic liver disease (PLD) itself is benign, but morbidity brought on by PLD is sometimes life-threatening.[1–4] Thus, considerably invasive procedures, including combined hepatectomy and cyst fenestration (CHCF) and even hepatic replacement, are necessary despite its benign nature.[5–7] Ascites necessitating persistent drainage or paracentesis after drain removal is found among early postoperative complications after CHCF for PLD.[5–7] It has been reportedly observed in 20% to 70% cases and seemed to easily cause recurrent symptoms unless properly treated.[5–7] Diuretics has been often used for this condition. However, its effectiveness seemed limited and thus prolonged drainage was usually required.[1,2,5–7]

We present a case of massive localized noninfectious fluid collection after CHCF for a patient with PLD, which was relieved by distilled water lavage (DWL). Furthermore, total liver volume (TLV), including an area where massive localized fluid collection treated with DWL existed, continued to reduce up to 12 postoperative months and thereafter retained less than one-third of preoperative volume more than 2 years after surgery. Sustained reduction of TLV suggested a sclerosant effect of hypotonic cytocidal property of distilled water for cyst endothelium[8] and/or retrieved effectiveness of fenestration. Hence, we consider this approach to be useful for patients with PLD receiving CHCF, and thus present it here.

Materials and Methods

Written informed consent was obtained from the patient for publication of this report and any accompanying images. Institutional review board of our institution approved this manuscript to be published. Use of DWL for the present case was approved by the Ethics Committee of the Yokohama Ekisaikai Hospital because its use was considered in accordance with the ethical guidelines for epidemiologic research in Japan based on previous reports regarding use of DWL in clinical settings.[8–13]

A 63-year-old postmenopausal woman with PLD developed severe abdominal fullness and edema of lower extremities. Her PLD was not accompanied by polycystic kidneys because no cystic lesions were found in her kidneys. On the other hand, the patient did not have any history of receiving oral contraceptives or estrogen replacement therapy. Genetic disorders causative of PLD[3] were not investigated because any PLD carriers were not proven in her blood relatives and the patient did not hope to receive genetic analyses. Her TLV measured by CT volumetry, including cysts and normal parenchyma, was 3600 mL at presentation, although her standard liver volume was 1014 mL calculated by Urata's formula.[14] Her blood tests, including carcinoembryonic antigen and carbohydrate antigen 19-9, did not show any abnormality except for a slightly elevated alkaline phosphatase with 345IU/L. Indocyanine green retention rate at 15 minutes was normal with 8% and no finding of portal hypertension was found. Her PLD was classified as type II of Gigot's classification,[2] grade 4 of Qian's classification,[3] and type C of Schnelldorfer's classification[5] (Fig. 1). Because numerous large cysts existed in the liver and thus a means for treating as many cysts as possible at once was considered preferred, we considered radiologic intervention including aspiration and/or sclerotherapy to be impractical. Therefore, we selected hepatectomy and fenestration of remnant cysts for the patient. The right posterior section of the liver was almost totally replaced by the cysts, although a major part of the right anterior section and left medial section was filled with cysts. However, the patient's left lateral section was not affected as much by the cysts (Fig. 1). Thus, the right posterior sectionectomy combined with fenestration of remnant cysts under laparotomy was performed. (Liver anatomy and method of hepatectomy was described according to the Brisbane 2000 system of nomenclature of liver anatomy and resections.[15]) We first planned to place an intraoperative prophylactic drain because ascites necessitating persistent drainage or paracentesis is a common early postoperative complication after CHCF for PLD.[5–7] However, fenestration is expected to cause the intracystic fluid outflow into the peritoneal cavity to be absorbed through the peritoneum. Namely, indiscreet drain placement

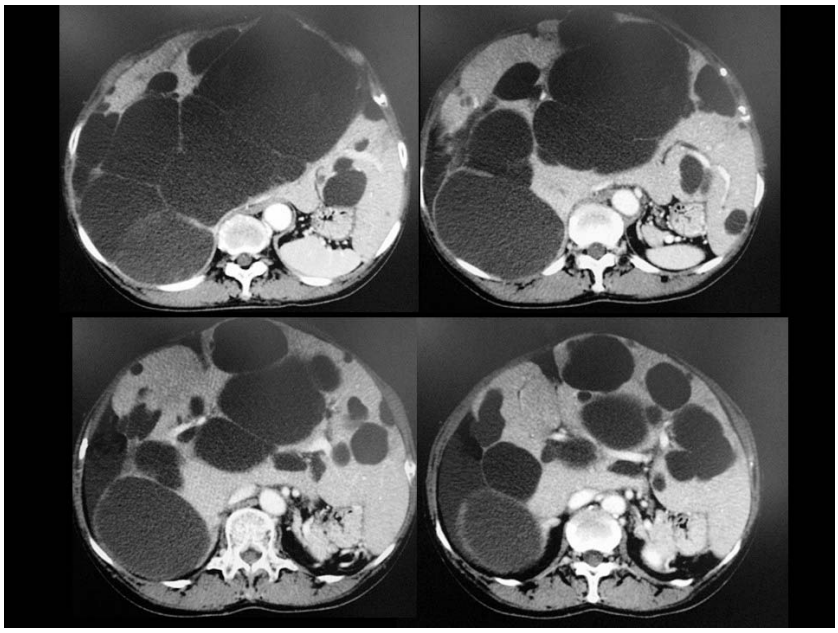

Fig. 1 Abdominal CT findings before combined hepatectomy and cyst fenestration. Right posterior section of the liver was almost totally replaced by the cysts, and a major part of the right anterior section and left medial section was filled with cysts. However, the left lateral section was not affected as much by the cysts. The type of polycystic liver disease in the present case was classified in type II of Gigot's classification, grade 4 of Qian's classification, and type C of Schnelldorfer's classification.

around the liver might isolate the outflow of intracystic fluid and impair the effect of fenestration due to narrowed peritoneal surface exposed to effluent through fenestration orifices. Therefore, an intraoperative prophylactic drain was not placed. Early postoperative course was uneventful until 10 days after surgery when sudden abdominal fullness and resultant severe anorexia developed. An emergent abdominal CT revealed massive monolocular fluid collection in the right subphrenic space where most severely occupying lesions that must have been eradicated by surgery were housed. Although TLV measured by CT volumetry (including the fluid collection) was reduced to 1700 mL at that time (Fig. 2), other than the fluid collection, only mild ascites was observed. Therefore, urgent percutaneous drainage for the right subphrenic collection was undertaken (Fig. 2). The content of the fluid collection was proven to be neither biliary nor infectious. Percutaneous drainage relieved the symptoms immediately, but the drainage tube could not be removed because of massive outflow with more than 500 mL a day consistently for the following 7 days. Mean duration of intra-abdominal drain placement after CHCF has been reported to range 7 to 21 days.[1,2,5–7] Regarding drain outflow, criterion of prophylactic intra-abdominal drain removal in these reports was less than 200 mL a day. Based on these previous reports, we considered that drain outflow of the present case, which exceeded 500 mL a day at postoperative day 17, was extraordinarily large. Furthermore, the effectiveness of diuretics, which has been reportedly used widely for mitigating this condition, was considered limited.[5–7] Thus, we decided to apply DWL via percutaneous drainage tube to this condition as a sort of sclerotherapy.[8] Ethanol is commonly used as a sclerosant of sclerotherapy for liver cysts.[16] However, ethanol was considered too strong and rather harmful if exposed to the cut surface of the liver and thus we decided to use distilled water. Actual procedures of DWL were as follows. All procedures were performed under fluoroscopic guidance. Furthermore, if distilled water leaked through fenestration orifices outside the cysts, the effect of fenestration might be disabled by the adhesive sealing due to the sclerosant effect of distilled water. Thus, the body position of the patient was in semi-Fowler and left-half side-lying position during the procedures. That was intended to prevent the distilled water from outflowing from the cysts and being exposed to the abdominal wall. First, fistulography via drainage tube was performed for measuring the volume of cavity. Special attention was paid to confirm the volume that contrast media did not leak through fenestration orifices. After complete aspiration of contrast media, distilled water was injected into the cavity with the volume confirmed by preceded fistulography (Fig. 2). Irrigation and aspiration with 5-minute interval of distilled water was performed using another

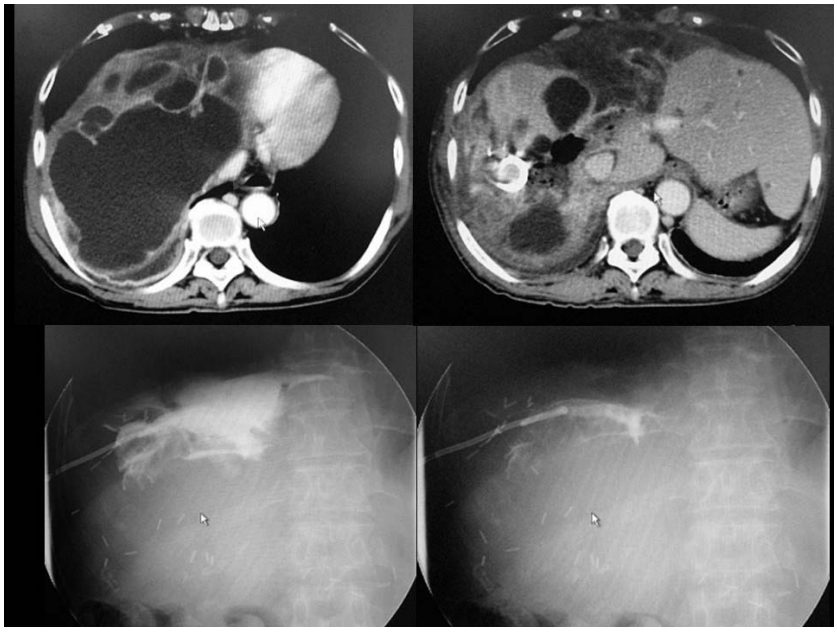

Fig. 2 CT findings and fistulography of massive right subphrenic fluid collection. Upper left: abdominal CT taken on postoperative day 10 when sudden abdominal fullness and resultant severe anorexia developed. Upper right: abdominal CT taken 3 days after percutaneous drainage for right subphrenic massive fluid collection. Lower left: fistulography via percutaneous drainage performed for measuring the volume of cavity. Lower right: fistulography after complete aspiration of contrast media. Abdominal CT revealed massive fluid collection in the right subphrenic space 10 days after surgery (upper left). Massive fluid collection was eradicated by percutaneous drainage (upper right). DWL for massive fluid collection via percutaneous drainage was performed under fluoroscopic guidance. First, fistulography via drainage tube was performed for measuring the volume of cavity (lower left). After complete aspiration of contrast media used volume measuring (lower right), distilled water was injected into the cavity with the volume confirmed by preceded fistulography.

volume of distilled water for each set repeatedly 3 times.

Results

The first session of DWL was performed postoperative day 17 and each subsequent session was performed every 2 days. The day after the 1st session, outflow through a drainage tube was dramatically decreased to 200 mL/day. After completion of 3 sessions, the outflow reduced to less than 50 mL/day and subsequent discontinuation of the DWL session did not result in increasing outflow. Hence, we removed percutaneous drainage on postoperative day 25. After that, symptoms did not recur and the patient was thereafter doing well.

The patient has been currently doing well without recurrence of symptoms 2 years after surgery. Abdominal CT study was performed every 3 months up to 1 year after surgery and thereafter every 6 months in fear of extraordinary expansion of the liver. However, TLV continued to reduce up to 1 year after surgery and thereafter retained less than one-third of preoperative TLV that was almost equivalent to her standard liver volume calculated from Urata's formula (Figs. 3 and 4).[14]

Discussion

Distilled water has been known to have hypotonic cytocidal property and been used usually with chemotherapeutic agents for mitigating symptom of carcinomatous peritonitis or pleuritis.[9,10] Furthermore, it has been reportedly utilized as peritoneal/pleural lavage perfusate after surgery for various cancers, including lung cancer,[10] esophageal cancer,[11] ruptured hepatocellular carcinoma,[12] gastric cancer,[9] and colorectal cancer,[13] with expectation for preventing peritoneal/pleural metastases due to its cytocidal potential. With respect of use for benign condition, Shin *et al*[8] reported that they successfully treated corneoscleral cyst with distilled water

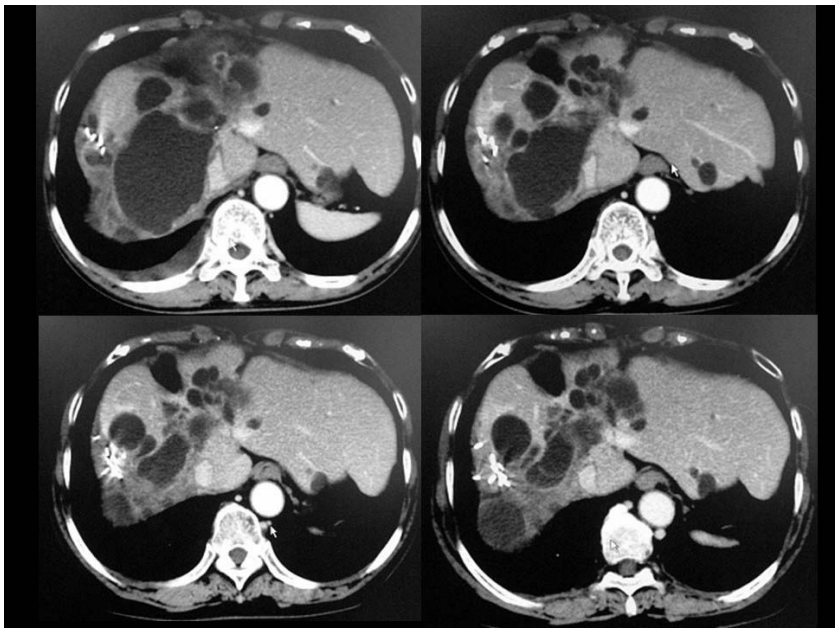

Fig. 3 Chronologic changes in the CT appearance of the right subphrenic massive fluid collection after removal of percutaneous drainage. Upper left: 6 months after surgery. Upper right: 9 months after surgery. Lower left: 12 months after surgery. Lower right: 24 months after surgery. Right subphrenic fluid collection seemed to increase once after the removal of percutaneous drainage (upper left). However, the fluid collection continued to reduce thereafter up to 12 postoperative months (upper right, lower left). Furthermore, it seemed that gained reduction of the fluid collection until 12 postoperative months retained at 24 postoperative months (lower right).

injection. As to the Shin's report, they utilized distilled water as a sclerosant for a sort of sclerotherapy for benign fluid collection. Sclerotherapy for symptomatic liver cyst(s) has been reportedly effective for mitigating symptoms and even for cure in certain occasions.[16] Based on these reports, we decided to use DWL for localized fluid collection in the present case. Ethanol is commonly used as a sclerosant of sclerotherapy for liver cysts because it has robust potential to destroy the inner cell lining with the property of strong chemical stimulant.[16] In the present case, however, strong sclerotherapy around fluid collection was considered unfavorable because strong sclerotherapy might cause severe adhesion around the liver if it resulted in strong sclerosant leaks through fenestration orifices into the abdominal cavity near the liver. Consequently, fenestration orifices on the liver surface may be sealed with adhesion and disabled, rendering liver cysts symptomatic again due to re-expansion. According to the above-stated articles regarding the usage of distilled water for mitigating symptoms of fluid collection, distilled water does not cause severe adhesion despite its cytocidal property.[9–13] In these reports, any severe complications associated with use of distilled water have not been described other than mild serum mineral imbalances caused by dilution of serum due to distilled water absorbed through peritoneum/pleura. Because sclerosant use of distilled water requires a much smaller amount compared to perfusate use, sclerosant use of distilled water seems quite safe. Therefore, we used distilled water for the present case. The fact that fenestrated cysts and TLV in the present case continued to decrease up to 1 year after surgery indicate that DWL was effective for mitigating acute localized fluid collection and did not cause severe adhesion, which can disable the fenestration, around the liver.

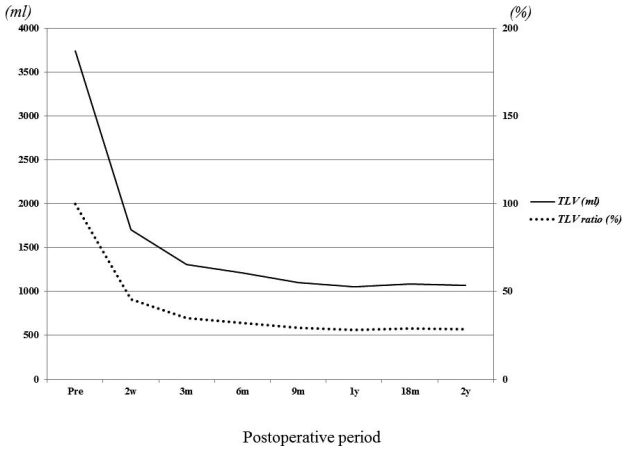

Fig. 4 Chronologic changes in total liver volume calculated by CT volumetry. Abdominal CT study was performed every 3 months up to 1 year after surgery and thereafter every 6 months. Total liver volume continued to reduce up to 1 year after surgery and thereafter retained. Total liver volume ratio was calculated by the following formula: (total liver volume calculated at each postoperative month/preoperative total liver volume) × 100%. TLV ratio reached less than one-third of preoperative level and retained up to 24 postoperative months.

Persistent drainage of ascites and/or progression of ascites necessitating paracentesis have been reportedly the commonest complications after surgery for PLD.[5–7] These conditions were considered caused by the following 2 different entities: content of cysts and postsurgical exudate.[5–7] If the cause of ascites is the former, fluid should be reabsorbed through the peritoneum if the fenestration works,[2,4–6] whereas ascites must be usually relieved spontaneously if the cause of ascites is the latter. However, it is nearly impossible to discriminate between these 2 entities in daily clinical practice. Furthermore, if the ascites originates from the content of cysts, extraperitoneal drainage should isolate the space in which the drain is placed, from the other peritoneal cavity, as a means of disabling the fenestration. We considered that reexpansion of the fenestrated cysts would be unavoidable if the fenestration does not work. Furthermore, a considerable number of the patient population receiving CHCF for PLD reportedly suffered symptom recurrence, although rate of symptom recurrence varied to range 0% to 100% among reports.[1–7] Although any previous reports did not use the term "localized fluid collection," we consider that "localized fluid collection" can be rephrased by "recurrent cyst." In other words, untreated "localized fluid collection" seemed easily to cause symptom recurrence. Thus, we had to take countermeasures against the acute localized fluid collection as soon as possible in the present case. As stated above, the use of a strong sclerosant seemed unfavorable in this situation because fenestration is disabled by severe adhesion caused by a strong sclerosant such as ethanol. To the best of our knowledge, there have not been any reports regarding use of distilled water as a sclerosant other than the Shin's report.[8] We believe that 2-year relief of symptoms of PLD in the present case was a result of our choice of distilled water as a sclerosant. In addition, we consider that the appropriateness of our choice is supported by the fact that fenestrated cysts and TLV in the present case continued to decrease up to 1 year after surgery and thereafter retained less than one-third of the preoperative volume.

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