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Case report

Surgical repair of hepatic hydrothorax caused by diaphragmatic fistula

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ABSTRACT

A 65-year-old woman visited our hospital complaining of dyspnea several days before admission. A chest X-ray showed massive right-sided pleural effusion, which was not observed 1 month previously. Although the patient had never been diagnosed with cirrhosis at regular visits, the patient was diagnosed with primary biliary cholangitis at admission. Hepatic hydrothorax was suspected because pleural effusion was transudative. A diaphragmatic fistula was confirmed and closed by thoracoscopy. Pleural effusion did not reappear after this procedure. Existence of a diaphragmatic defect should be confirmed under direct vision if pleural effusion accumulates acutely or becomes beyond control.

1. Introduction

Hepatic hydrothorax is generally defined as pleural effusion of ≥500 ml in patients with cirrhosis [1]. This is an uncommon complication of uncompensated cirrhosis with an estimated prevalence of 5%–12% [2]. The mechanism of hepatic hydrothorax has only been proven by thoracic abdominal communication due to diaphragmatic fistula. However, diaphragmatic fistula confirmed under direct vision is unique. Because thoracoscopy is rarely performed in cases of transudative pleural effusion, such as heart failure and hypoalbuminemia, this condition often only improves with medical treatment. However, thoracoscopy is performed for diagnosing cases of exudative pleural effusion. Additionally, cases of improved pleural effusion by closing diaphragmatic fistulas are uncommon. We report a rare case of hepatic hydrothorax due to primary biliary cholangitis, which was confirmed by diaphragmatic fistulas and treated by closure.

2. Case report

A 65-year-old Japanese woman with chronic renal failure due to diabetic nephropathy presented with a history of coughing and dyspnea on exertion for a few days. The patient had never been diagnosed with cirrhosis. On physical examination, the patient showed decreased breathing sounds and percussion dullness in the lower right lung field. Palmar erythema and vascular spider were absent. The liver was not palpable at the right upper abdomen. A laboratory evaluation showed that alkaline phosphatase and gamma-glutamyl transpeptidase levels were elevated at 876 U/L and 241 IU/L, respectively. Total bilirubin and albumin levels and prothrombin activity were within the normal range at 0.6 mg/dL, 3.5 g/dL, and 100%, respectively. There was no thrombocytopenia. A chest radiographic examination showed a large amount of pleural effusion on the right side that was not noticed 1 month previously (Fig. 1A). Chest and abdominal computed tomography showed right-sided pleural effusion, right lower lobe atelectasis and no abnormal findings of the right diaphragm. There were also a small amount of ascites fluid, right hepatic atrophy, and no splenomegaly (Fig. 2A,B). The patient had yellow-clear transudative pleural effusion at diagnostic thoracentesis. Hepatic hydrothorax was suspected from the patient's medical history. Thoracoscopy showed a bleb on the diaphragmatic surface (Fig. 3). Therefore, we thought that ascites may have been transferred to the thoracic cavity through diaphragmatic fistulas. The patient was ultimately diagnosed with asymptomatic primary biliary cholangitis at the decompensated stage from an examination of various antibody tests and a liver elastography test. Therefore, the patient was placed on a salt-restricted diet and provided with a diuretic and ursodeoxycholic acid. However, pleural fluid was difficult to control. Accordingly, we decided to search for diaphragmatic defects. Under general anesthesia, when water was stored in the thoracic cavity and air

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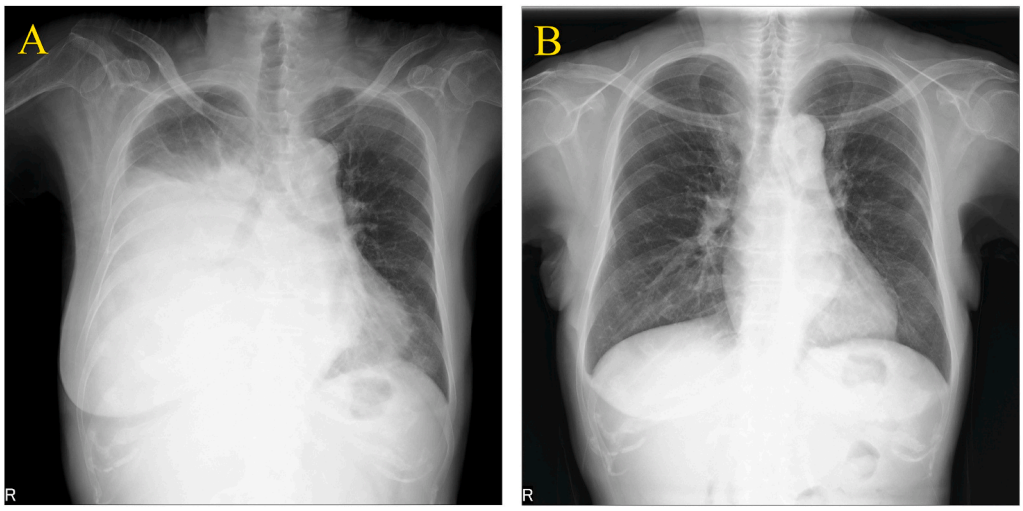

Fig. 1. Chest radiographic examination. (A) Massive pleural effusion on the right side at the first visit was observed. (B) Pleural effusion disappeared after closure of the fistula.

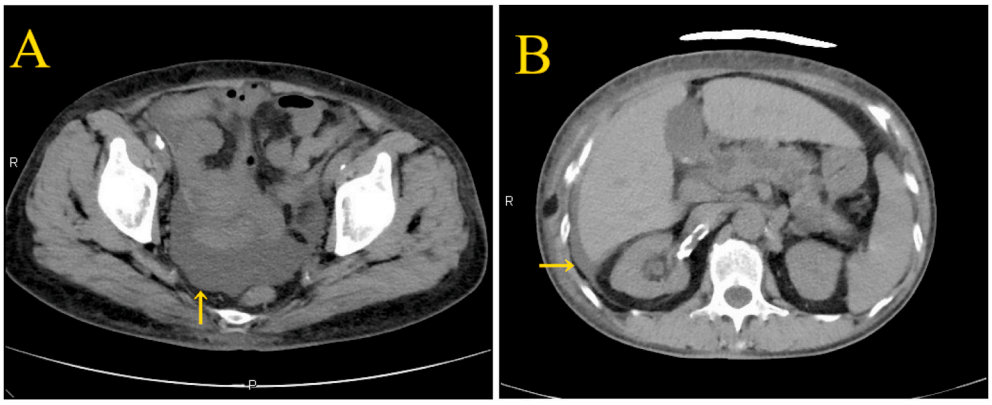

Fig. 2. Abdominal computed tomography scan. (A) Minimal ascites was observed. (B) There are hepatic atrophy and minimal ascites on the surface of the liver.

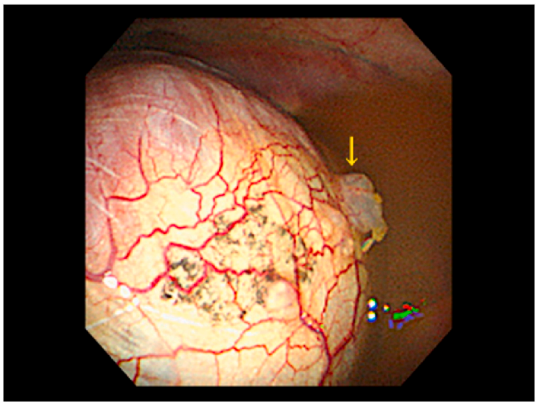

Fig. 3. Thoracoscopic view shows a bleb on the surface of the right side of the diaphragm.

was delivered from the abdominal cavity, the air leaked from part of the diaphragm. Therefore, thoracoscopic repair of diaphragmatic defects was performed. After surgery, pleural effusion disappeared. The pleural effusion never increased after this time and the ascites were well controlled by adjusting diuretics (Fig. 1B).

3. Discussion

There are few cases in which diaphragmatic defects can be seen directly in hepatic hydrothorax. This is because thoracoscopy is often performed to search for the cause of exudative pleural effusion, but it is rarely used in transudative pleural effusion. In our case, the presence of diaphragmatic traffic was suspected because of right unilateral large transudative pleural effusion, despite the minimal amount of ascites. Initial attempts were made to inject the radioisotope intraperitoneally, but safely injecting the radioisotope was difficult because of the small amount of ascites. Therefore, we performed thoracoscopy and were able to find the lesion in the diaphragm. Diaphragmatic defects need to be identified, even in cases with minimal or no ascites, if there is a large amount of transudative pleural effusion.

Hepatic hydrothorax is a complication in patients with decompensated cirrhosis who do not have cardiac or pulmonary disease. In a previous study, a diagnosis other than hepatic hydrothorax accounted for 18% of pleural effusion in patients with cirrhosis [3]. Therefore, a chest computed tomographic scan, pleural effusion cytology, and biopsy if necessary should be performed to exclude pulmonary, mediastinal, or pleural disease.

The causes of hepatic hydrothorax are reported to include diaphragmatic communication, leakage due to hypoalbuminemia, an increase in azygous vein pressure and flow leading to leakage of plasma, and leakage through the lymphatic pathway of the diaphragm. In most cases, hepatic hydrothorax develops on the right side, while 13% of

cases occur on the left side, and 2% are bilateral [2]. Histologically, the hemidiaphragm has a embryological developmental defect, and the tendinous portion eventually becomes the muscular components during development. When intra-abdominal pressure increases due to ascites or coughing, the muscle fibers are displaced, causing herniation from the abdominal cavity to the thoracic cavity. Therefore, a diaphragmatic fistula appears when there is herniation of less than 1 cm of rupture [2, 4]. The left hemidiaphragm is thicker and more muscular than the right and is less likely to have a diaphragmatic defect. Epidemiology explains the right-sided predomination of hepatic hydrothorax.

Hepatic hydrothorax is almost moderate to large in size according to radiological criteria [5]. Half of patients with hepatic hydrothorax have minimal-small ascites, 42% have moderate-large ascites, and only 9% do not have ascites. Negative intrapleural pressure during inspiration compared with pressure of the peritoneal cavity facilitates one-way transfer of fluid through diaphragmatic fistulas [6].

In diagnosis of diaphragmatic defects, intraperitoneal injection of 99mTc-sulphur colloid or 99mTc-human serum albumin can be helpful. Movement of the radioisotope from the peritoneal cavity to the pleural cavity can confirm diaphragmatic communication [7]. However, testing facilities are limited. Intraperitoneal injection of indocyanine green may be considered [8]. Thoracoscopy may also reveal diaphragmatic defects, which are considered highly invasive and have a high threshold.

Treatment of hepatic hydrothorax is generally a sodium-restricted diet, diuretics, and therapeutic thoracentesis [2]. Refractory hepatic hydrothorax is defined as when pleural puncture is frequent or when pleural effusion reappears rapidly after puncture. Surgical treatment of refractory hepatic hydrothorax includes continuous thoracentesis, pleurodesis, a transjugular intrahepatic portosystemic shunt, surgical repair of diaphragmatic traffic, and liver transplantation [4]. In cases of rapid accumulation of hepatic hydrothorax or those with difficult medical control, finding and treating diaphragmatic defects may improve the patient's distress.

4. Conclusion

We experienced a case of hepatic hydrothorax due to primary biliary cholangitis caused by diaphragmatic defect and treated it by closure. In cases of hepatic hydrothorax that are relatively acute or difficult to control, the presence of diaphragmatic defects should be confirmed.

Declaration of competing interest

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.rmcr.2020.101325.

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