CASE REPORT

Rare case of massive bilateral chylothorax

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SUMMARY

We present a case of a 48-year-old woman who was referred from an outside hospital. There, she had initially underwent hysterectomy and left salpingo-oophorectomy for tubo-ovarian abscess. She later developed a colovaginal fistula and perforation of sigmoid colon and underwent Hartmann's procedure along with drainage of a left subphrenic abscess. Subsequently, she had to be intubated for acute respiratory failure and was transferred to our hospital. At our hospital, she was found to have massive bilateral pleural effusions. Bilateral small-bore chest tubes were inserted that drained milky fluid. Pleural fluid analysis was consistent with bilateral chylothorax. Thereafter, patient's respiratory status improved and she was extubated. The mechanism of chylothorax was thought to be either secondary to the multiple abdominal procedures or alternatively as a complication of the right subclavian catheter that was placed at the outside hospital. Her chest tubes were removed eventually, and she had a slow but definite recovery.

BACKGROUND

Chylothorax is an uncommon medical condition characterised by accumulation of chylous fluid in the pleural space. Thoracic surgery is one of the leading causes of chylothorax, but it has been reported to occur after abdominal surgeries as well. Also, chylothorax has very rarely been reported as a complication of central venous catheter placement. The precise pathogenesis of the development of chylothorax in these scenarios is subject to speculation. Chylothorax more commonly occurs unilaterally; we describe an interesting case of massive bilateral chylothorax.⁸

CASE PRESENTATION

A 48-year-old woman presented from an outside hospital. She had initially presented at the outside hospital with severe pelvic pain. There, she was found to have a left adnexal mass on examination, and subsequent imaging revealed a left tubo-ovarian abscess. Five days before transfer to our hospital, she underwent total abdominal hysterectomy and left salpingo-oophorectomy. Three days after the procedure, patient's condition started deteriorating, and she started passing stool and flatus through the vagina. She was diagnosed with colovaginal fistula and was taken back to the operating room for repair. During the laparotomy, she was also found to have perforation of the sigmoid colon. Sigmoidectomy and end colostomy (Hartmann's procedure) was performed, and a left subphrenic abscess was

drained. It is unclear if these findings were complications from the previous surgery or progression of the primary disease process. In the following 2 days, patient's respiratory status worsened, and she had to be intubated due to acute respiratory failure. She was subsequently transferred to our hospital for further management. Her other past medical, surgical, social and family histories were unremarkable.

On admission to our intensive care unit, patient was found to be hypotensive and was requiring pressor support. She had a right subclavian venous catheter at the time of transfer, which was removed and replaced with a left internal jugular venous catheter. She was febrile and had a white cell count of $21\,400/\text{mm}^3$. She was initially started on meropenam. The cultures obtained from intraoperative tissue samples in the outside hospital were found to be growing *Escherichia coli*, and the antibiotic was eventually switched to ertapenam. The precise reason for the acute respiratory failure was unclear at this moment. Notably, the chest X-ray performed after left internal jugular catheter placement revealed moderate to large bilateral pleural effusions. Thereafter, a CT scan of the chest was performed, which confirmed the presence of bilateral massive pleural effusions (figure 1). Thereafter, small-bore chest tubes were inserted in the pleural space on both sides. On insertion, the right-sided chest tube drained 2000 mL of fluid, whereas the left-sided chest tube drained 1200 mL. The fluid was found to have a milky appearance.

INVESTIGATIONS

Pleural fluid analysis revealed the following: pH=7.66, protein=0.2 g/dL and lactate dehydrogenase (LDH)=66 U/L. Notably, the pleural fluid triglyceride level was 880 mg/dL, which was highly consistent with the diagnosis of chylothorax. Cell count analysis showed 367 nucleated cells per cubic millimetre of which 95% were polymorphonuclear cells. Cytology, gram stain and bacterial culture of the pleural fluid were negative. Lymphoscintigraphy was contemplated to define the precise site of leak for possible cardiothoracic surgical intervention, but it was deferred as patient's chest tube output gradually trended down over the next 2 days.

DIFFERENTIAL DIAGNOSIS

The exact mechanism of chylothorax was unclear. One possibility that was considered was iatrogenic trauma of the thoracic duct at its junction with cisterna chyli secondary to its handling during the extensive abdominal surgeries. The second consideration was thoracic duct damage secondary to

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Figure 1 Images taken from the CT scan of the chest showing massive bilateral pleural effusions (marked with asterisk) with compressive lower lobe atelectasis. (A) Transverse view. (B) Sagittal view.

central venous catheter insertion. Unfortunately, a lymphoscintigraphy to localise the site of leak was not warranted in her case and was thus not performed.

TREATMENT

As highlighted before, the bilateral chylothorax was treated with bilateral chest tube insertion. After this intervention, her respiratory status improved dramatically and she was extubated the very next day. CT chest repeated 2 days later showed marked interval improvement bilaterally. She was subsequently transferred to general medical floor and started on a fat-restricted diet. Thereafter, the output from her chest tubes reduced gradually to about 100 mL per day on day 8, and they were subsequently removed. The general surgery team closely followed the patient, and she had to undergo another exploratory laparotomy for revision of the end colostomy and drainage of the pelvic abscess. Ertapenem was continued to complete a total of 14 days of antibiotics.

OUTCOME AND FOLLOW-UP

Patient's overall condition gradually improved. After removal of the chest tubes, her chief concern was pain control that was addressed with opioid medications. She was eventually discharged on day 21 of admission with a close outpatient follow-up planned. She was advised to continue with fat-restricted diet for a total duration of 6 weeks. She was followed up at the general surgery office up to 6 weeks after discharge and was found to be doing well with almost complete resolution of pain and normal bowel movements.

DISCUSSION

The thoracic duct begins at the T12 vertebra as a continuation of cisterna chyli. It then ascends through the aortic hiatus of the diaphragm, which is also present at the level of T12 vertebra. As the thoracic duct ascends through the thoracic cavity, it is situated mainly on the right side until around T3/T4 vertebra where it crosses the midline and turns to the left. It finally drains into the venous system at the junction of left internal jugular and left subclavian veins. Four litres of chyle is drained into the venous system every day. Chylothorax is the result of disruption of thoracic duct or one of its main tributaries. As highlighted by Talwar and Lee,¹ the most common causes of chylothorax

are neoplasm, thoracic surgery and direct trauma. Chylothoraces most commonly occur on the right side as the largest part of the duct is in the right hemithorax. Left chylothorax can occur if the superior portion of the duct is involved. When the leakage occurs where it crosses over to the midline, a bilateral chylothorax can occur. Regardless of its aetiology, when the duct leaks, a collection of chyle is formed adjacent to the pleura. This has been described in literature as 'chyloma'. This soon ruptures and empties into the pleural space leading to chylothorax.

Although the possibility of thoracic duct damage during thoracic surgeries is intuitive, chylothorax has also been described secondary to abdominal surgeries. One mechanism involves development of chylous ascites and leakage of chylous fluid through diaphragmatic defects.² However, in some cases such as ours, chylothorax has been shown to occur without the occurrence of chylous ascites. Griffo *et al*³ presented three cases of chylothorax after abdominal surgeries with no evidence of chylous ascites in any of the cases. Notably, all cases had right-sided chylothorax. Cestero *et al*⁴ presented another case where right-sided chylothorax was caused by an abdominal gunshot injury that required surgical exploration. The mechanism of chylothorax in these cases has not been clearly defined. It can be contemplated that traction of the thoracic duct at its origin, at the level of the aortic hiatus, may cause disruption of supra-diaphragmatic portion of the duct. This may subsequently lead to chylothorax. It should also be noted that although not explicitly mentioned, most of these patients likely underwent central venous cannulation, which itself has been described to cause chylothorax.

Chylothorax after central venous catheter insertion has also been reported; Saxena *et al* compiled a total of 22 cases in their review.⁵ Interestingly, 9 out of the 18 cases where the side of chylothorax was documented had bilateral chylothorax, which is far more than the overall proportion of bilateral chylothoraces. The side of chylothorax was not mentioned in the remaining four cases. Various mechanisms have been proposed including direct trauma to the thoracic duct by the catheter tip causing leakage of chyle in the upper mediastinum and slow passage into the pleural cavity. Of note, our patient had a right subclavian catheter placed at the outside hospital before he was transferred. It is not known if that catheter was placed under ultrasound

guidance. This can certainly be attributed as the cause of the chylothorax in our patient, especially considering the fact that the chylothorax was bilateral.

The initial management of chylothorax is conservative by decompression of the pleural space with chest tube placement. Fat-restricted oral diet or total parenteral nutrition should be considered to reduce chyle production.⁶ If the chest tube continue to have high output, surgical options can be considered as a last resort. Lymphoscintigraphy done prior to surgery can help localise the site of leak, but it is a technically challenging procedure.⁷ In conclusion, development of chylothorax in the setting of abdominal surgeries as well as central catheter placement has been described in the literature, but its mechanism in these scenarios is still speculative. More data are needed to

develop a more robust model of pathogenesis of chylothorax in these two scenarios.

Contributors All authors contributed to the manuscript as follows: GST and YA were both part of the intensive care unit team that admitted the patient. DMZ resumed care of the patient in the second week. Hence, all authors were directly involved with patient care. GST and DMZ did the literature review and manuscript write-up. YA revised the manuscript critically for important intellectual content. All authors gave their final approval of the version to be published.

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Learning points

- ▶ Chylothorax can very rarely occur as a complication of abdominal surgery as well as central venous catheter insertion. More research is needed to gain further mechanistic insights into its pathogenesis in these scenarios.
- ▶ Chylothorax secondary to central venous catheter insertion is more likely to be bilateral.
- ▶ Early drainage of chylothorax with chest tubes can lead to dramatic recovery. Fat-restricted oral diet or total parenteral nutrition reduces chyle production.
- ▶ Lymphoscintigraphy and surgical treatment should be reserved in cases refractory to conservative management with persistent chest tube output.

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