Infected Bilateral Chylothorax in a Problematic Case

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Abstract

Within this report we describe a problematic patient who developed infected bilateral chylothorax. A 38 year-old female who had a berry aneurysm at the basilar artery was admitted to neurointensive care unit after coil embolization procedure. On third day, her chest X-ray disclosed bilateral massive pleural effusions. Bilateral intercostal drainage tube insertions were performed. The pleural fluid was clearly. Cultures of the chylous fluid revealed *Acinetobacter calcoaceticus-baumanii complex*. On 10th day, she expired. We concluded that chylothorax in adults may rarely occur without a solid reason; and in the treatment may also be difficult especially when it is contaminated with microorganisms.

Keywords: Aneurysm; Catheterisation; Chylothorax; Thoracic duct

Introduction

Chylothorax represent chylous liquid (ie. enriched with fat and its products) in the pleural cavity [1]. Although it is the most common aetiological factor of the pleural effusions in neonates and fetuses, in adults it is very rare with an incidence of 3-5% [2-4].

Within this report we describe a problematic patient who developed infected bilateral chylothorax after endovascular embolization of a berry aneurysm at the tip of the basilar artery.

Case Report

A 38 year-old female was admitted to emergency room with severe headache, naeusea and vomiting of sudden onset. On neurologic exam, she had neck stiffness and mild right hemiparesis. Cranial CT scan disclosed subarachnoid haemorrhage (Fisher grade IV) (Fig. 1). MR-angiography and four-vessel cerebral angiography revealed a berry aneurysm at the tip of the basilar artery (Fig. 2). She was referred for endovascular surgery. After the embolization procedure, she was admitted to the neurointensive care unit (ICU). Immediately after the general anesthetic effect, she became agitated.

Figure 1. Cranial CT scan discloses subarachnoid haemorrhage.
and disoriented. Considering that an unstable hemodynamic situation would be harmful for the brain, the patient was intubated and mechanically ventilated. Cranial CT scan after sedation revealed obstructive hydrocephalus (Fig. 3). A ventricular drainage catheter was inserted at right Kocher’s point. As part of ICU routine a right subclavian catheter was inserted for nutrition and 3H (hypervolemic, hypertensive haemodilution) treatment. On third day at the ICU, the patient became pyrexic and chest X-ray revealed bilateral massive pleural effusions (Fig. 4), particularly on the right side. Chest CT disclosed mediastinal enlargement (Fig. 5). Bilateral intercostal drainage tubes were inserted and approximately 2000 mL milky fluid was drained. The biochemical analysis of chylous fluid revealed a high level of triglyceride and a low level of total cholesterol with a value of 3534 mg/dL (50 - 200, normal serum ranges), 32 mg/dL (125 - 200, normal serum ranges), respectively. 15 mg twice a day subcutaneous octreotide was initiated.

Transthoracic echocardiography, lower limb venous doppler-US, oesophago-gastro-duodenoscopy and abdomen CT scan were normal except for bilateral surrenal hyperplasia. Urine analysis and routine blood biochemistry were normal. Elevated fibrinogen, D-dimer, Factor VIII and IX (Christmas) levels were pathological laboratory findings. Her total parenteral nutrition was continued with low chain fatty acids; and subcutaneous low molecular weight heparin were also started. Further immunological studies of serum showed antibodies for lupus anticoagulant antigen 1(+) (normal reference range < 4(+) and antinuclear antibodies (ANA) at a titer of 1/100 (normal reference range < 1/360), but test for anti-ds DNA, anti-SSA/Ro, anti-SSB/La,anti-Sm and anti RNP antibodies all gave negative results. Factor V Leiden mutation analysis was normal. Culture of the chylous fluid revealed Acinetobacter calcoaceticus-baumannii complex which was resistant to piperacillin, ceftazidime, ciprofloxacin, trimethoprim-sulfamethaxazole, and tazobactam but not to amicasin, ticarcillin and imipenem, 1 gm thrice a day meropenem and 500 mg twice a day vancomycin intravenous (IV) was initiated. Chylous fluid cytology showed numerous

Figure 2. MR-angiography reveals a berry aneurysm at the tip of the basilar artery.

Figure 3. CT scan reveals obstructive hydrocephalus.

Figure 4. Chest X-ray shows bilateral massive pleural effusions.

Figure 5. Chest CT discloses mediastinal enlargement.
leukocytes without atypical features and its tuberculous-
PCR was negative. Three days later both catheters were re-
moved. On 8th day after admission, she developed left sided 
hemiplegia. Diffusion and perfusion weighted cranial MR 
showed bilateral posterior parietal, and right medial frontal 
hemispheric ischemia; and MR-angiography disclosed vas-
sospasm at right anterior cerebral and both bilateral middle 
cerebral arteries (Fig. 6). Chest MR revealed re-collection 
of the right pleural effusion, persistent mediastinal enlarge-
ment related to haemorrhage like mediastinal collection; and 
thrombus in the azygous vein. All main mediastinal vessels 
except for left inferior pulmonary artery and azygous vein 
were patent. Percutaneous thoracentesis was performed and 
1500 mL chylous fluid was drained. On 9th day, her right 
pupil enlarged and became unresponsive to light. Cranial CT 
showed bilateral cerebral oedema with subfalcine herniation 
towards to left side (Fig. 7). An intracranial pressure (ICP) 
monitor probe was inserted at the left side Kocher’ point, 
and the initial ICP was 69 mmHg. Then she underwent right 
fronto-temporo-parietal decompressive craniectomy and 
dural release. Postoperative ICP was down to 10 mmHg. 
On 10th day body temperature was 40 °C and ICP levels 
increased to 40 mmHg. Her blood pressure level decreased 
steadily despite medical treatment; and she expired. The 
family declined a postmortem study.

Discussion

The aetiology of chylothorax in adults includes trauma to the 
thoracic duct (blunt or penetrating trauma), thoracic surgery, 
central venous catheterisation, extensive venous thrombo-
sis in the neck, neoplasms, compression of the mediastinal 
lymph nodes, hepatic chirois, idiopathic causes (such as 
Down’s syndrome), some connective tissue diseases (such as 
Behçet’s disease and systemic lupus erythomatosus (SLE)), 
infectious diseases (such as flariasis, tuberculosis and his-
toplasmosis), lymphangioleiomyomatosis, and congestive 
heart failure. If there is no history of trauma or surgery, ma-
lignencies should be excluded [1- 3]. There was no causative 
factor for our patient’s chylothorax except nausea and vom-
iting, and central subclavian catheterisation. Transthoracic 
echocardiography, oesophago-gastro-duodenoscopy and 
abdominal CT scan in our patient excluded malignancy or 
infection. Chest CT disclosed mediastinal enlargement and 
bilateral pulmonary microemboli. Further immunological 
studies of serum excluded possibility of SLE. On the other 
hand, fibrinogen, D-dimer, Factor VIII and IX (Christmas) 
levels were elevated. These parameters would elevate in 
most of conditions such as pulmonary thromboembolus, dis-
seminated intravascular coagulation or septic shock. In our 
patient, there were bilateral pulmonary microemboli and 
mediastinal expansion on chest CT scan which could not be 
distinguished from abscess or haemorrhage. On the other 
hand she had a long line inserted into the right subclavian
vein and through this line 3H treatment and hyperalimentation was continued. Central venous catheterisation may lead to venous thrombosis in the neck, and this procedure may obstruct the drainage of the chyle into the subclavian vein which will then result in uni-or bilateral chylothorax especially when venous entry is into the left subclavian vein. Even in such a case, a latency period of 2 to 7 days exist between the time of venous injury and chylothorax, and chyle accumulates first in the posterior mediastinum [1, 5]. In the literature reported complications related to central venous catheterisation and hyperalimentation treatment are mediastinal haematoma and leakage of intravenous fluid because of the displacement of the tip of the central venous catheter [6, 7]. Chest MR of our patient revealed persistent mediastinal enlargement related to haemorrhage like mediastinal collection; and thrombus in the azygous vein. This condition could be explained by the central venous catheterisation procedure, however except the left inferior pulmonary artery and azygous vein, other mediastinal main vasculatures were all patent; and mediastinal enlargement could not be explained by haemorrhage, chylous fluid or intravenous fluid accumulation. Bipedal lymphangiography and/or lymphoscintigraphy has been recommended to clarify the cause and source of the leak of the chyle [1, 2]. Also to demonstrate the aetiology of the mediastinal enlargement the mediastinal endoscopy could be done. However, we did not perform these radiologic or surgical procedures because of the lack of the time, and worse health condition of the patient. Also there was no postmortem investigation to demonstrate the thoracic duct and mediastinal anatomy. On the other hand, in our knowledge association of intracranial aneurysm and chylothorax has not been reported in English medical literature. In patients with SLE, intracranial berry aneurysm and chylothorax have been reported to occur simultaneously [8, 9], but our patient did not have SLE.

Continued loss of proteins, immunoglobulins and lipids into the pleural spaces may lead to secondary immunodeficiency; and these may predispose the patient to opportunistic microorganisms such as Histoplasma spp. However, infection of chylous fluid itself is very uncommon because it is inherently bacteriostatic [1]. In our patient, chylous fluid cytology showed numerous leukocytes without atypical features and Gram (-) bacilli whose cultures revealed Acinetobacter calcoaceticus-baumanii complex. In the literature chylous fluid infected with different types of microorganisms were reported [1], but we were unable to cite Acinetobacter spp as the agent. It is not possible to conclude that chylothorax had been caused by, or contaminated with this microorganism because the blood cultures were negative for Acinetobacter spp. Besides the infection can be referred as nosocomial infection, when we take the resistance pattern of the microorganism into consideration.

**Conclusion**

As conclusion, sometimes the aetiology of chylothorax in adults may be uncertain and/or complicated; and in the treatment may also be difficult especially when it is contaminated with microorganisms. In such a case hypervolemia and hypertension may further aggravate the ventilation-perfusion of damaged lungs.

**References**