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Delayed Concurrent Chylothorax and Chyloperitoneum: Report of a Case after an Old Blunt Trauma

Mohsen Sokouti, Babak Abri Aghdam

Department of Thoracic Surgery, Imam Reza Hospital, Tabriz University of Medical Sciences, TABRIZ – IRAN.

ABSTRACT

A 15- year-old boy was referred to Imam Reza Hospital with a right chest tube and chylothorax for 40 days. The patient had respiratory distress and undergone refractory treatment for chylothorax. The fluid content was chyle-rich in lipids. Computed Tomography of the chest showed a large, incompletely evacuated cyst in the left posterior mediastinum with left pleural effusion. The cyst could not be resected through right thoracotomy, because of the left side location of the cyst. Ligation of the thoracic duct through right thoracotomy was not effective in reducing chylous effusion 4 days later. Left chylothorax exacerbated because of the complication of right thoracotomy. Laparatomy was performed to ligate the thoracic duct 6 days later. On exploratory laparatomy, chylous effusion was detected in the peritoneum. Thoracic duct with all the fibro-fatty tissues was ligated below the diaphragm over the spine at 12th to 2nd vertebral spaces. Right chylothorax was resolved after ligation of thoracic duct transabdominally 1-2 days later. Left chylous effusion was decreased and treated 46 days after laparatomy. One year follow up of the patient showed excellent result. In our knowledge, thoracic duct cyst occurring as a result of a delayed chylothorax and chyloperitoneum has not been reported in the literature. Surgical thoracic duct ligation can be the treatment of choice. (Tanaffos 2011; 10(1): 52-56)

Key words: Thoracic duct, Cyst, Chylothorax, Chyloperitoneum, Trauma

INTRODUCTION

Chylothorax is a rare complication of blunt chest trauma. Longelot reported the first case of traumatic chylothorax in 1663 in German literature and Quincke is given the most credit for defining the disease in 1875(1). The causes of blunt traumatic

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Correspondence to: Sokouti M
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Address: Department of Thoracic Surgery, Imam Reza Hospital, Tabriz, Iran.

Email address: sokouti.mohsen@yahoo.com Received: 18 September 2010 Accepted: 2 November 2010 chylothorax are not clear, and most authors agree that it follows a puncture, rupture, or overstretching of the major thoracic duct by fracture or other injuries of neighboring thoracic spine (2). Here we present a case of delayed concurrent bilateral chylothorax and chyloperitoneum after an 11-year history of blunt trauma with development of thoracic duct cyst in the mediastinum. The patient was successfully treated with surgical thoracic duct ligation. To the best of our knowledge, no similar case has so far been reported in the literature.

CASE SUMMARIES

A 15 year- old boy had a mild respiratory distress, dyspnea and back pain with right thoracostomy tube and chylothorax and admitted to our referral hospital in Tabriz, Iran. In his medical history, he had experienced a trauma caused by a fall 11 years before. The recent patient's history revealed that the right chylous pleural effusion was detected 40 days ago. Physical examination of the patient revealed tachypnea with a respiratory rate of 48/min, decreased respiratory sounds of the left lung and mild cachexia. Chest X-ray showed a biconcave smooth, well-marginated cystic mass in the left border of the heart and aorta. Computed tomography of the chest after right tube thoracostomy showed mild left pleural effusion and a low-density, incompletely evacuated cystic mass in the left posterior mediastinum (Figure 1). Right thoracostomy tube was inserted in the first hospital the patient presented to 40 days ago and 2,200- 2,900 ml of chyle was drained within a day. The characteristic appearance of pleural effusion was milky. Analysis of chylous effusion revealed 100 ml of chyle, with 90% lymphocytes; 5.9 g/dl of protein, total fat of 4.6 mg%, cholesterol 68mg/dl and up to 1,490 mg/dl of triglyceride. Oral intake of regimens containing lowmedium chain triglycerides was not effective in reducing chylous effusion. Because of continuity of massive chylous drainage, the patient was prepared for surgery. Much more creamy diet was started orally at the night of surgery. After right posterolateral thoracotomy, there was much more chylous effusion in the right pleural space; whereas, there was no tearing or disruption in the thoracic duct. In exploration, a large cyst $(10 \times 6 \times 3)$ was discovered in the mediastinum which had developed within 11 years after the blunt trauma. The cyst was torn during excision and chyle was drained into the

pleural cavity (Figure 2). Excision of the cyst was not possible because almost all of it was located in the left posterior hemithorax. Tearing site was ligated at the 11th vertebral level over the vertebrae and above the diaphragm. The day after surgery no chylous drainage was observed in right chest tube but the next day, chylous drainage continued in an amount equal to that of pre-operation. On the 4th day after surgery, left thoracostomy tube was inserted, because of increased volume of chylous effusion, and almost 1000 ml of chylous was drained in one day. On day 6 after thoracotomy, because of no drainage in the volume of chylous effusion, we were certain that the thoracic duct ligation was not effective. The patient was prepared for laparatomy and surgical ligation of the thoracic duct was performed transabdominally. There was about 300 ml of chyle in the peritoneal cavity (chyloperitoneum), and a bulging cystic mass was located on the 12th thoracic vertebrae being extended down to the 3th abdominal vertebrae in the retroperitoneum. Chyle had leaked from visceral mesentery and cystic mass of the retroperitoneum.

Thoracic duct along with its fibro-fatty tissues were ligated below the diaphragm over the spine at 12th vertebral spaces. One Foley catheter was positioned in cul de sac. Chylous drainage of right chest tube stopped the day after surgery. Left chylous pleural effusion was decreased to 500-900 ml/day, whereas chylous drainage of peritoneum continued for 2 months after surgery. Finally left chylous effusion stopped 46 days after laparatomy. The patient was discharged from the hospital 14 days after the operation. Two months later, chest x-ray and CT- scan were taken and no effusion was detected in the hemithorax. Left chest tube was extracted and 6 days later abdominal Foley catheter was also removed. During the one year follow- up, no pleural effusion was detected on chest x-ray (Figure 3). The abdomen looked normal as well.



Figure 1.Computerized tomography after evacuation of right chylothorax with chest tube. Mild left pleural effusion is seen on chest x-ray.



Figure 2 Cyst wall tear during excision through right thoracotomy. A great amount of chyle was present in the cyst and pleural cavity.



Figure 3. Chest x-ray one year later. Mild pleural thickening and linear atelectasis were seen in the right lung.

DISCUSSION

Thoracic duct cyst and chylothorax are rare consequences of blunt thoracic traumas. Traumatic

mediastinal cyst of thoracic duct is rare (3) and most of them are found in the posterior mediastinum (2). It shares similar characteristics in all parts of thoracic duct (4).

Although the pathogenesis of thoracic duct cysts is unclear, congenital weakness of the duct wall and acquired degenerative process induced by trauma, infection or other inflammation have been discussed in the literature (5). Another suggested theory is that the cysts may arise as an aneurysmal dilation of the duct which is related to a traumatic injury of the duct. Knowledge about the anatomy of thoracic duct and its variations is important in shearing of it in trauma and producing chylothorax (6).

Possible mechanisms of duct injury after blunt thoracic trauma include:

(a) Fracture or dislocation of the lower thoracic vertebrae resulting in injury of the duct.

(b) Perforation or tearing of the duct when osteophytes or exostosis exist.

(c) Sudden hyperextension of the lower thoracic spine caused by stretching the duct over the vertebral bodies or by shearing of the duct with right crus of the diaphragm (2).

In our patient a history of trauma was noted 11 years ago, which could account for an injury of thoracic duct.

It may occur anywhere from the thoracic inlet to the diaphragm and can communicate with the thoracic duct (2). It was also observed as concurrent abdominal and cervical cysts (5). Communication of the cyst with the thoracic duct was mentioned in 17 cases of Cervantes-Perez series (7). Some authors believe that thoracic duct cysts do not have communication with the thoracic duct (8). In our patient, an obvious communication was detected between the cyst and the thoracic duct, because after surgical ligation of the thoracic duct, two-sided chylothorax stopped.

CT scan and lymphangiography can show the cystic nature of the lesion and may specify the

anatomic connection of the cyst with the thoracic duct if not evacuated (9).

Definite diagnosis of thoracic duct cyst is based on surgical and histopathological findings (6). Diagnostic findings of our patient were confirmed during surgery as leakage of chyle from thoracic duct cyst and creation of chylothorax with Histopathological chyloperitoneum. findings revealed thin wall with endothelial cells and lymphoreticular tissue (9). Because of difficulties in excision of thoracic duct cyst, pathologic study of the patient became impossible.

Multiple complaints, the risk of life- threatening complications and the need for confirming the diagnosis account for the resection of cysts even in asymptomatic patients (9).

Radiologically, thoracic duct cysts appear as a round or oval, sharply confined mass in the visceral compartment that may extend into the ipsilateral para vertebral sulcus (10). In our patient it was seen with biconcave appearance in left paravertebral sulcus.

Diagnosis is made by appearing in lymphangiography and analysis of fluid obtained by CT-guided needle aspiration, but it is rarely diagnosed before surgery and most of them reveal during thoracotomy (6, 11). Of the eight surgically treated patients in Tsuchiya's series, only one patient was diagnosed by lymphangiography before surgery (10).

Conservative treatment (RST): Re-expansion of the lung by closed tube drainage (R) with dietary starvation (S), and total parenteral nutrition (T) is another choice of treatment in uncomplicated and free symptom cysts of mediastinum (2, 11). Considering such complicated cases as the one reported in this article, efficacy of conservative treatment is questionable.

Surgery and ligation of thoracic duct are indicated in cases with: (a) drainage for more than 14 days (b) daily drainage greater than 1,500 ml in adults or 100 ml a day in children older than 5 yrs (c) metabolic complications (1).

Up to 40% of patients may have double channels. If not ligated, they will cause recurrent build-up of chyle. Complete ligation of the posterior mediastinal tissue surrounding it has also been recommended (1). In our patient; (a) and (b) were present which was reason enough to operate.

Thoracic duct ligation is the treatment of choice for chylothorax. Recurrence has been reported because of anomaly of thoracic duct in the mediastinum or incomplete closure of thoracic duct proximally or distally. When cyst is the primary diagnosis, surgical excision is recommended (9). In this case because of difficulties in terms of location and tearing of the cyst, excision became impossible.

Other treatments include pleural patches to cover the thoracic duct and sealing of the leak with thoracoscopy and fibrin glue application (12).

To the best of our knowledge, delayed and simultaneous occurring of chylothorax and chyloperitoneum has not been reported in the literature. When reviewing the literature, only a case of a 26- year-old woman with chylothorax after blunt chest trauma was reported by Milano and associates which was diagnosed after a latent period of 11 weeks (13).

CONCLUSION

Delayed concurrent chylothorax and chyloperitoneum after a history of blunt trauma and cyst formation have not been reported in the literature. Only a patient with chylothorax after blunt chest trauma was reported by Milano, diagnosed after a latent period of 11 weeks (13).

We described our experience in performing thoracic duct ligation to treat thoracic duct cyst in a patient with delayed chylothorax.

Conflicts of interest: The authors have no conflicts of interests to declare.

REFERENCES

- McCormick J 3rd, Henderson SO. Blunt trauma-induced bilateral chylothorax. *Am J Emerg Med* 1999; 17 (3): 302-4.
- Apostolakis E, Akinosoglou K, Koletsis E, Dougenis D. Traumatic chylothorax following blunt thoracic trauma: two conservatively treated cases. *J Card Surg* 2009; 24 (2): 220-2.
- Hom M, Jolles H. Traumatic mediastinal lymphocele mimicking other thoracic injuries: case report. *J Thorac Imaging* 1992; 7 (3): 78- 80.
- Ikonomidis JS, Boulanger BR, Brenneman FD. Chylothorax after blunt chest trauma: a report of 2 cases. *Can J Surg* 1997; 40 (2): 135-8.
- Gottwald F, Iro H, Finke C, Zenk J. Thoracic duct cysts: a rare differential diagnosis. *Otolaryngol Head Neck Surg* 2005; 132 (2): 330- 3.
- Brauchle RW, Risin SA, Ghorbani RP, Pereira KD. Cervical thoracic duct cysts: a case report and review of the literature. *Arch Otolaryngol Head Neck Surg* 2003; 129 (5): 581-3.
- Mori M, Kidogawa H, Isoshima K. Thoracic duct cyst in the mediastinum. *Thorax* 1992; 47 (4): 325- 6.

- Roberts JR, Kaiser LR. Acquired lesion of the mediastinum: Benign and malignant disease. In: in Fishman A. Fishman's pulmonary disease and disorder. 4th ed. McGraw-Hill Co; 2008; p: 1609.
- Karajiannis A, Krueger T, Stauffer E, Ris H. Large thoracic duct cyst - a case report and review of the literature. *Eur J Cardiothorac Surg* 2000; 17 (6): 754- 6.
- Miller J. Anatomy of the thoracic duct and chylothorax. In: Shields T. LoCicero J Ponn RB, Rush V. General Thoracic Surgery. 7th ed. Philadelphia: Lippincott Williams and Wilkins; 2005: pp. 879-88.
- Morettin LB, Allen TE. Thoracic duct cyst: diagnosis with needle aspiration. *Radiology* 1986; 161 (2): 437-8.
- Shirai T, Amano J, Takabe K. Thoracoscopic diagnosis and treatment of chylothorax after pneumonectomy. *Ann Thorac Surg* 1991; 52 (2): 306-7.
- Milano S, Maroldi R, Vezzoli G, Bozzola G, Battaglia G, Mombelloni G. Chylothorax after blunt chest trauma: an unusual case with a long latent period. *Thorac Cardiovasc Surg* 1994; 42 (3): 187- 90.