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CASE REPORT

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Successful management of a chylothorax in infancy using a pleurectomy

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Abstract The management of chylothorax is described in three infants. Because none of them had had a direct injury to the thoracic duct, it was suspected that the chylothoraces had a significant chance of recurrence after simple ligation of the duct. To avoid recurrence, formal stripping of the parietal pleura was performed using a technique not previously reported in the literature. The operation was simple to perform and resulted in resolution of the chylothorax in all infants. The three cases and details of the surgical procedure are described.

Key words Chylothorax · Hydrothorax · Surgical treatment · Pleurectomy

Introduction

Chylothorax in infancy can arise from a variety of etiologies, the most common being direct trauma to the thoracic duct during a surgical procedure [1, 2]. The management of a traumatic chylothorax is usually initiated by a modification of diet (e.g., total parenteral nutrition [TPN] and the use of medium-chain triglycerides for enteral fats) and repeated thoracenteses or placement of a thoracostomy tube [3]. Should the chylothorax be persistent, direct ligation of the thoracic duct is performed [1, 2, 4, 5]. Other forms of chylothorax are associated with either congenital derangement of the lymphatic flow or occlusion of the superior vena cava [6, 7]. This latter form has no specific opening in the thoracic duct, but occurs due to diffuse leakage secondary to abnormalities in the drainage of the duct. Because no direct trauma to the thoracic duct had occurred in our three patients, this appeared to be the etiology in these cases.

Each of the three chylothoraces failed to resolve on conventional medical management and required surgery. Because no specific site of leakage could be identified, we suspected that simple ligation of the duct would lead to recurrence. We therefore elected to perform a pleurectomy with low ligation of the thoracic duct and pleurodesis. A formal pleurectomy may be associated with large amounts of blood loss and is generally reserved for recurrent chylothoraces following failure of simple thoracic duct ligation. We, however, modified the technique of pleurectomy in these infants, making it a part of the initial surgical procedure with virtually no blood loss and requiring little additional operative time.

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Case reports

Case 1 A 2.5-kg, 32-week gestational age infant in whom prenatal ultrasound showed a right-sided pleural effusion had a pigtail catheter placed in utero. A chest tube was placed for respiratory distress and removed at 24 h of age. The infant was discharged at 3 weeks of age with a normal chest radiograph, but readmitted at 5 weeks of age with respiratory distress. A chest radiograph showed a large fluid collection in the right chest. A tube thoracostomy was performed with drainage of a chylothorax. Despite 3 weeks of conservative treatment, he continued to have large amounts of drainage and subsequently underwent a right thoracotomy with decortication, pleurectomy, and pleurodesis. All mediastinal lymphatic tissue was ligated. The pathology report showed lymphangiectasia and acute lymphadenitis. He was treated with 2 weeks of IV antibiotics and then discharged home. Two weeks later he was readmitted to the hospital with right middle lobe pneumonia and a cystic lesion of the chest, for which he was treated successfully with antibiotics. The patient is currently well 30 months following surgery.

Case 2 A 3.7-kg, full-term female was born with a left congenital diaphragmatic hernia (CDH) that was repaired at 12 h of age at an outside hospital. She required extracorporeal life support (ECLS) post-operatively for 9 days. Her postoperative course was complicated by the development of two recurrent diaphragmatic hernias. During the surgery for her third repair, 30–40 ml milky, white fluid was evacuated from the left hemithorax. Evaluation of the fluid demonstrated 12,150 white blood cells/mm3 with a differential of 96% lymphocytes, 1% neutrophils, and 4% histiocytes, pH 8.59, specific gravity 1.016, triglyceride level >10,000, and a negative culture. She remained fasting and was supported nutritionally with TPN, however, the chest tube output remained at 40–60 ml/day. On postoperative day 14 she underwent a left exploratory thoracotomy with pleurectomy and pleu-

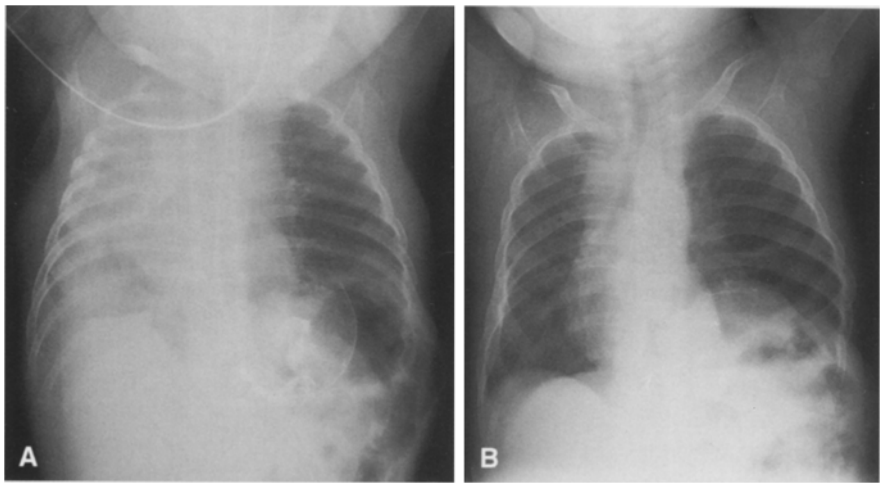

Fig. 1 **A** Recurrent right-sided chylothorax in case 3 despite placement of multiple chest tubes. **B** Chest radiograph 5 months after pleurectomy and ligation of thoracic duct. Note abscence of any effusion (breakdown of diaphragmatic hernia repair was subsequently reoperated)

rodesis. Empiric excision and ligation of a portion of the mediastinal lymphatic tissue in the region just superior to the diaphragm was also performed. Minimal chest tube drainage was noted postoperatively. Feedings were initiated on postoperative day 5, initially with Portagen and subsequently with breast milk. The chest tube was removed on postoperative day 8. A follow-up chest radiograph showed expansion of the left lung without evidence of a pleural effusion. She is currently assymptomatic 18 months after surgery.

Case 3 A female infant was born at 38 weeks gestational age with a left-sided CDH and a right pneumothorax. She underwent repair of the CDH and required ECLS support following surgery for 5 days. She did well until 7 weeks of age when she was admitted for respiratory distress requiring tracheal intubation. A chest radiograph at that time showed a presumed right-sided pleural effusion. A thoracostomy tube was placed with the removal of chylous fluid, and the infant was placed on TPN. Ten days after admission she was extubated and placed on a Portagen enteral diet. The chest tube was removed 5 days later followed by recurrence of the effusion despite her modified diet (Fig. 1A). A new chest tube was placed and she was again placed on TPN. The chest tube output continued to be elevated, and after 5 weeks of unsuccessful treatment the child underwent operative therapy. An extrapleural dissection was performed with ligation of the thoracic duct and excision of lymphatic tissue just above the diaphragm followed by resection of the pleura and a pleurodesis. She was extubated on the same day, was discharged from the hospital with a clear chest X-ray film on the 13th postoperative day. A recurrent left-sided CDH was repaired at 18 months of age. She is currently asymptomatic 24 months following her surgery (Fig. 1B).

Operative technique

A standard posterolateral thoracotomy incision is performed. The 5th intercostal space is incised and an extrapleural dissection bluntly performed with moistened cotton-tip applicators. Incision at this level appears to offer the best exposure of the chest cavity. Initially, dissection is performed posteriorly and then superiorly over the apex of the lung and inferiorly down to the diaphragm; following this, it is carried anteriorly. In general, this extrapleural dissection is as easy as that for an esophageal atresia despite previous placement of multiple chest tubes. Once the extrapleural dissection is complete, virtually all the parietal pleura is separated from the chest wall except for some portions on the diaphragm and at the hilum of the lung. The mobilized pleura is resected along the perimeter of the pleural dissection using cautery. By this means, the extrapleural and pleural spaces are completely joined. The thoracic duct is identified and ligated as close to the diaphragm as possible. Although each of our patients had heavy cream administered by nasogastric tube in the immediate preoperative period, no evidence of a chylous leak or discrete injury to the thoracic duct could be identified. A pleurodesis is then performed with either a surgical sponge or a Bovie scraper pad on all aspects of the lung. A chest tube of relatively large size is placed and the chest is closed in the standard fashion. The chest tube is removed after the initiation of feedings and when drainage is minimal.

Discussion

The most common cause of chylothorax in infancy is trauma due to a previously performed cardiac or thoracic procedure [1, 2, 8, 9]. Most chylothoraces can be managed nonoperatively with pleural drainage and dietary manipulation. If the chylothorax persists or recurs after 2 to 3 weeks of management, an operative approach should be undertaken since continued medical therapy is associated with high morbidity and mortality [10]. A chylothorax in infancy may be associated with mortality ranging from 15% to 83% despite operative management with thoracic duct ligation and pleurodesis [1, 6, 11, 12].

In this review, we present three patients with chylothoraces that were refractory to treatment with thoracostomy tube placement and discontinuation of enteral feedings. The etiology was idiopathic in one child (case 1); in the other two the chylothorax may have been due to the associated CDH itself or to partial superior vena caval occlusion after internal jugular vein cannulation for ECLS. The correction of a chylothorax in such patients may not be amenable to thoracic duct or other lymphatic vessel ligation since a diffuse lymphatic leak is usually present [2]. Because of our concern that the chylothoraces might recur, we elected to perform a pleurectomy and pleurodesis in addition to ligation of the thoracic duct. In combining these techniques, we hoped that the development of adhesions between the chest wall and visceral pleura would prevent recurrence. In all three cases, the chylothoraces resolved: there was no evidence of recurrence on long-term radiographic follow-up in all three patients.

Pleurectomy is usually utilized only in the management of recurrent chylothoraces because it can be associated

with blood loss and technical difficulty [9]. However, in these cases we utilized an operative approach that is used routinely in the correction of a tracheoesophageal fistula with esophageal atresia. The extrapleural dissection was easy to perform, resulted in minimal blood loss, and required little additional operative time. Despite our initial concerns that the previously placed chest tubes would make dissection technically challenging, this proved not to be the case, as the extrapleural dissection was performed without difficulty. Despite the apparent absence of a leak, ligation of the thoracic duct was performed in all cases to prevent excessive flow and subsequent accumulation of chyle through the involved thoracic cavity. Our current recommendation is to treat a chylothorax, regardless of etiology, with medical management. Should it persist for greater than 2 to 3 weeks or recur, the surgical technique presented here should be performed.

In summary, we describe an operative technique to manage patients with chylothoraces utilizing a combined approach of extrapleural dissection, resection of the parietal pleura, ligation of the thoracic duct at the level of the diaphragm, and subsequent pleurodesis. Using this approach, we successfully managed three patients with chylothorax and encountered no excessive operative time or technical difficulty.

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