



# Pulmonary Hypertension as a Rare Cause of Postoperative Chylothorax

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## Abstract

Chylothorax in adult occurs most commonly in the wake of cardiac and thoracic procedures. Injuries to the common thoracic duct in the thorax or its branches in the mediastinum, injuries to the thymus tissues, dissection of the superior vena cava or ascending aorta, dissection of the aortic arch, disruption of the accessory lymphatics in the left or right thorax, and increased pressure in the systemic vein exceeding that of the thoracic duct (usually in the superior vena cava thrombosis, Glenn Shunt, and hemi-Fontan) have been proposed as the possible causes of chylothorax after surgery for congenital heart disease. However, pulmonary hypertension is an exceedingly rare cause of chylothorax in adults. We present a case of chylothorax after atrial septal defect surgery in a 30-year-old female patient with pulmonary hypertension. The postoperative period was complicated by chylothorax, which was confirmed by the high lipid content of chylous effusion. The patient was treated conservatively with diet therapy, and the effusion was abolished completely after two weeks. No recurrence of chylothorax was detected at 3 months' follow-up.

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## Introduction

Concomitant occurrence of pulmonary hypertension and chylothorax has been reported in the neonatal period as a result of congenital heart disease. Nevertheless, a meticulous search of the literature fails to reveal any such complication following atrial septal defect (ASD) surgery in adults.<sup>1</sup> Closed heart procedures performed in the vicinity of the thoracic duct such as systemic-to-pulmonary arterial shunt insertion, repair of aortic coarctation, and ligation of the patent ductus arteriosus likewise predispose to the development of chylothorax as evidenced in this and previous studies.<sup>2-5</sup> Carmelo Mignosa<sup>6</sup> reported a case

of chylothorax as a symptom of a large left-to-right atrial shunt: increased venous pressure in the right atrium created the hemodynamic basis for a hypertensive thoracic duct, which led to chyle leakage and accumulation in the right pleural space. Joyce<sup>7</sup> and Beghetii<sup>8</sup> showed an increase in the prevalence of postoperative chylothorax to 4.7%, which was higher than the previously reported 1% or less by Verunelli<sup>9</sup> and Allen.<sup>10</sup> Cormack<sup>11</sup> attributed these differences to the increased complexity of the surgery being performed today, whereas Puntis<sup>12</sup> ascribed this complication to the earlier reintroduction of feeding after surgery.

The morbidity and mortality related to excessive lymph drainage are well-documented by Chan,<sup>2</sup> but Pelletier<sup>13</sup>

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demonstrated that a definitive management strategy for postoperative chylothorax remained elusive. Bond and colleagues<sup>14</sup> reported that a period of conservative management with the use of total parenteral nutrition was necessary. Milson<sup>15</sup> and Chenung<sup>16</sup> recommended such surgical interventions as pleurodesis, ligation of the lymphatic ducts, and pleural-peritoneal shunting for non-responders. Begheti<sup>7</sup> suggested that ventricular dysfunction after Tetralogy of Fallot repair might predispose to increased systemic venous pressure and postoperative chylothorax. The postoperative leakage of the lymphatic fluid into the pleural space may result from the surgical disruption of the thoracic duct or one of its main tributaries, or increased pressure within the intrathoracic lymph system.<sup>17</sup> Patients usually remain asymptomatic until accumulation of a large volume of chyle in the pericardial space causes tamponade or loss of this fluid by chest tube drainage can lead to nutritional depletion, fluid and electrolyte loss, hypolipoproteinemia, and reduction of thymocyte cells, which can attribute to immunodeficiency.<sup>18</sup>

## Case Report

Chylothorax occurred in our 30-year-old female patient postoperatively after ASD repair. Preoperative physical examination and laboratory examination revealed no abnormality. Transthoracic echocardiography illustrated moderate pulmonary hypertension (pulmonary artery pressure = 50 mmHg).

Having provided informed consent, the patient was transferred to the operating room and placed on the operating room table in supine position. Upon the induction of general endotracheal anesthesia and placement of indwelling arterial and venous monitoring lines, the patient was prepped and draped in the usual sterile fashion from chin to groins, and a full midline vertical skin incision was performed in the sternum. Dissection was carried out through the deeper planes until the sternum was scored and divided with an oscillating saw. A small portion of the anterior pericardium was procured for the patch closure of the segment of the ASD during the procedure. Purse strings were deployed on the ascending aorta on the right and the atrial appendage. After systemic heparinization, central aorto-bicaval cannulation was performed for cardiopulmonary bypass (CPB). Both caval veins were encircled with Ethibond vascular loops. After mild hypothermic CPB and cross-clamping of the aorta, intermittent cold cardioplegic infusion was administered in an antegrade fashion into the aortic root. Through a standard right atriotomy, the ASD was closed. Deairing was performed through the aortic root and cardiac apex before aortic declamping. The right atriotomy was closed in two layers with running 4-0 Viline sutures. Venous decannulation was followed by aortic decannulation

and administration of protamine sulfate. All the cannulation sites were oversewn with 4-0 Viline sutures, and the cannulation sites were observed to be hemostatic. With good hemodynamics and hemostasis, the sternum was thereafter closed with stainless steel wires. The subcutaneous tissues were closed in layers with reabsorbable monofilament sutures. The patient was transferred in very stable condition to the adult Intensive Care Unit. On the third postoperative day while staying in the ward, she started inspiration and became short of breath. On examination, she was found to have reduced left hemithorax sound and normal palpable peripheral pulses. Plain chest X-ray revealed massive pleural effusion. The case was managed conservatively with chest tube drainage and total parenteral nutrition. The drained fluid was tested biochemically and was found to contain chylomicrons (700 mg/dl) and triglycerides (230 mg/dl). The electrolyte content of the chyle was similar to that of the serum, and the concentration of protein in the chyle was 4.0 gr/dl. Additionally, the chyle also contained 7000 white blood cells/ml, predominantly lymphocytes.

The patient had persistent drainage through a left hemithorax drain of approximately 1000 ml of chylous fluid every day. She was, therefore, given a low-fat, high-protein diet for approximately 2 weeks. This conservative management failed, and she continued to have drainage of more than 900 ml of chyle every day. On the 15<sup>th</sup> postoperative day, decision was made to perform re-exploratory surgery. During this operation, the pericardial cavity was opened through the previous incision and washed. There was no lymphatic leak from the thymic lobe, sub vena cava dissection, or the space between the main pulmonary artery and the aorta, and nor was there any evidence of innominate artery thrombosis or vena cava thrombosis. An interesting point with regard to this complication was that the leakage came from the left pleural space, which was intact. She made an uneventful recovery 2 weeks after surgery and was discharged home after 4 weeks.

## Discussion

The thoracic duct is prone to injury during cardiothoracic thoracic surgery due to its extremely variable anatomical course. Joyce<sup>7</sup> reported that chylothorax following coronary artery bypass graft surgery (CABG) commonly results from the distal dissection of the left or right mammary artery and the transection of the thoracic duct or the collateral lymphatic channels – usually in the proximity of the origin of the left subclavian artery – in consequence of attempts to gain extra length on the left internal mammary artery. However, Shu-yan Chan<sup>19</sup> reported for the first time a case of massive-volume chylothorax, caused by injury to a large aberrant thymic lymphatic duct in the anterior mediastinum. The authors routinely divided the thymus in order to gain adequate exposure to the aorta for cannulation in ASD repair



and proximal anastomosis in CABG. The researchers also reported that multiple accessory lymphatic channels, which were present in the vicinity of the superior vena cava (SVC) and aorta, were injured during the encircling of the SVC or the aorta. Existence of a lymphatic duct is unusual in this anatomical situation; owing to this rarity, the presence of a lymphatic duct in this position could be mistaken for a venous tributary. Electrocautery cannot coagulate the lymph duct and is inadequate to control leakage from the lymphatics because lymph contains fewer coagulation factors such as factor 7 than plasma.<sup>20</sup> Thurer<sup>21</sup> reported that chylothorax following ASD repair could also occur due to duct injury after central line placements. Buttiker V.<sup>22</sup> suggested that the use of tape around the SVC and venous thrombosis could raise the SVC pressure and obstruct the drainage of chyle and subsequent extravasations. The Sibert study<sup>23</sup> documented that the thrombosis of the thoracic duct and subsequent backflow and extravasations through the disrupted lymphatic channels were other mechanisms involved in the creation of this complication. The aforementioned study also reported that while the incidence of postoperative chylothorax was 0.5%, the resultant mortality was as high as 50% - when adequate treatment was not promptly provided.

Successful management of chylothorax following cardiothoracic surgery is dependent on the volume and duration of drainage and also on the metabolic, nutritional, and immune status of the patient. The available options are conservative therapy, thoracic duct embolization, duct ligation, and open surgical exploration. Conservative therapy is generally reserved for low-volume drainage (< 1000 ml/day) and usually involves the use of a low-fat diet with medium-chain triglycerides, which are absorbed directly from the portal venous system, or total parenteral nutrition. Carment<sup>24</sup> was the first to describe an association between pulmonary hypertension and chylothorax. Berkenbosch<sup>25</sup> treated a neonate with pulmonary hypertension and chylothorax with nitrous oxide, and posited that the presence of pulmonary hypertension was a contributing factor in the congenital and refractory postoperative chylothorax development. Carment<sup>24</sup> described congenital chylothorax in neonates with persistent pulmonary hypertension. Mercy<sup>26</sup> reported a case of refractory chylothorax in a neonate with moderate pulmonary hypertension after arterial switch operation. He instituted inhaled nitric oxide and achieved marked improvement insofar as the pulmonary artery pressure dropped considerably with a similar reduction in the chest tube drainage. Karimi<sup>27</sup> and colleagues reported a rare case of chylothorax after left internal mammary usage, which was complicated by chylothorax. A similar complication was reported by Brenner<sup>28</sup> in a rheumatic mitral stenosis patient, who had developed severe congestive cardiac failure. Our patient is unique in that she represents a case of adult ASD concomitant with pulmonary hypertension complicated by postoperative chylothorax. To the best of our knowledge and

according to our exhaustive literature review, a similar case has not been reported to date.

## Conclusion

Although ASD closure eliminates hemodynamic changes of a right-to-left shunt, CPB, cardioplegic arrest, right heart stunning, and right heart failure increased the venous pressure in the right atrium of our patient and created a hemodynamic basis for a hypertensive thoracic duct, leading to chyle leakage from a small superficial plural and lung lymph duct and accumulation in the intact left pleural space.

## References

- Higgins CB, Mulder DG. Chylothorax after surgery for congenital heart disease. *J Thorac Cardiovasc Surg* 1971;61:411-418.
- Chan EH, Russell JL, Williams WG, Van Arsdell GS, Coles JG, McCrindle BW. Postoperative chylothorax after cardiothoracic surgery in children. *Ann Thorac Surg* 2005;80:1864-1870.
- Puntis JW, Roberts KD, Handy D. How should chylothorax be managed? *Arch Dis Child* 1987;62:593-596.
- Cerfolio RJ, Allen MS, Deschamps C, Trastek VF, Pairolero PC. Postoperative chylothorax. *J Thorac Cardiovasc Surg* 1996;112:1361-1365.
- Rasihah SV, Oei J, Lui K. Octreotide in the treatment of congenital chylothorax. *J Paediatr Child Health* 2004;40:585-588.
- Mignosa C, Duca V, Ferlazzo G, Bianca I, Salvo D. Chylothorax: an unusual manifestation of a large atrial septal defect. *J Thorac Cardiovasc Surg* 2001;122:1252-1253.
- Joyce LD, Lindsay WG, Nicoloff DM. Chylothorax after median sternotomy for intrapericardial cardiac surgery. *J Thorac Cardiovasc Surg* 1976;71:476-480.
- Beghetti M, La Scala G, Belli D, Bugmann P, Kalangos A, Le Coultre C. Etiology and management of pediatric chylothorax. *J Pediatr* 2000;136:653-658.
- Verunelli F, Giorgini V, Luisi VS, Eufrate S, Cornali M, Reginato E. Chylothorax following cardiac surgery in children. *J Cardiovasc Surg (Torino)* 1983;24:227-230.
- Allen EM, van Heeckeren DW, Spector ML, Blumer JL. Management of nutritional and infectious complications of postoperative chylothorax in children. *J Pediatr Surg* 1991;26:1169-1174.
- Cormack BE, Wilson NJ, Finucane K, West TM. Use of Monogen for pediatric postoperative chylothorax. *Ann Thorac Surg* 2004;77:301-305.
- Puntis JW, Roberts KD, Handy D. How should chylothorax be managed? *Arch Dis Child* 1987;62:593-596.
- Pelletier GJ. Invited commentary. *Ann Thorac Surg* 2005;80:1870-1871.
- Bond SJ, Guzzetta PC, Snyder ML, Randolph JG. Management of pediatric postoperative chylothorax. *Ann Thorac Surg* 1993;56:469-472.
- Milsom JW, Kron IL, Rheuban KS, Rodgers BM. Chylothorax: an assessment of current surgical management. *J Thorac Cardiovasc Surg* 1985;89:221-227.
- Cheung Y, Leung MP, Yip M. Octreotide for treatment of postoperative chylothorax. *J Pediatr* 2001;139:157-159.
- Baffes TG, Potts WJ. Postoperative chylothorax. *Ann Surg* 1954;139:501-505.
- Cevese PG, Vecchioni R, D'Amico DF, Cordiano C, Biasiato R, Favia G, Farello GA. Postoperative chylothorax. Six cases in

- 2,500 operations, with a survey of the world literature. *J Thorac Cardiovasc Surg* 1975;69:966-971.
19. Chan SY, Lau W, Wong WH, Cheng LC, Chau AK, Cheung YF. Chylothorax in children after congenital heart surgery. *Ann Thorac Surg* 2006;82:1650-1656.
  20. van Straaten HL, Gerards LJ, Krediet TG. Chylothorax in the neonatal period. *Eur J Pediatr* 1993;152:2-5.
  21. Thurer RJ. Chylothorax: a complication of subclavian vein catheterization and parenteral hyperalimentation. *J Thorac Cardiovasc Surg* 1976;71:465-468.
  22. Buttiker V, Fanconi S, Burger R. Chylothorax in children. Guidelines for diagnosis and management. *Chest* 1999;116:682-687.
  23. Seibert JJ, Golladay ES, Keller C. Chylothorax secondary to superior vena caval obstruction. *Pediatr Radiol* 1982;12:252-254.
  24. Carmant L, Le Guennec JC. Congenital chylothorax and persistent pulmonary hypertension of the neonate. *Acta Paediatr Scand* 1989;78:789-792.
  25. Berkenbosch JW, Withington DE. Management of postoperative chylothorax with nitric oxide: a case report. *Crit Care Med* 1999;27:1022-1024.
  26. Mery CM, Moffett BS, Khan MS, Zhang W, Guzmán-Pruneda FA, Fraser CD, Jr, Cabrera AG. Incidence and treatment of chylothorax after cardiac surgery in children: analysis of a large multi-institution database. *J Thorac Cardiovasc Surg* 2014;147:678-686.
  27. Karimi A, SalehiOmran A, Yazdanifard P. Chylothorax after coronary artery bypass and internal mammary artery harvesting: a case report. *East Mediterr Health J* 2010;16:1103-1104.
  28. Brenner WI, Boal BH, Reed GE. Chylothorax as a manifestation of rheumatic mitral stenosis: its postoperative management with a diet of medium-chain triglycerides. *Chest* 1978;73:672-673.