The Heart Surgery Forum #2020-3043
23 (3), 2020 [Epub June 2020]
doi: 10.1532/hsf.3043

Online address: http://journal.hsforum.com

Incidence of Postoperative Chylothorax Following Congenital Heart Surgery in Pediatric Patients: A Single-Center Experience

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ABSTRACT

Background: Although postoperative chylothorax following congenital heart surgery occurs rarely, it is associated with substantial morbidity and mortality. The incidence of postoperative chylothorax has been reported as 2% to 5%. Therefore, we aimed to evaluate the incidence of postoperative chylothorax at our center and compared our results with those of other studies.

Methods: Between January 2009 and December 2018, there were 2,515 congenital heart repair surgeries performed at our center. Thirty-six patients with postoperative chylothorax were enrolled in this study, and their medical records retrospectively were reviewed. We calculated the overall and surgery-specific incidences. We compared the data of the medical management group with those of the surgical management group.

Results: The overall incidence of postoperative chylothorax was 1.4%. The incidence was highest for patients who underwent vascular ring repair (3/32). Moreover, the incidence was higher for single ventricle-related procedure than bi-ventricle-related procedures (5.6% versus 1.0%, $P < .0001$). Chylothorax was predominantly found on the left side (20/36). Among these cases, six patients died; three of these six were in the surgical management group.

Conclusions: The postoperative chylothorax incidence at our center was comparable to those of other centers. However, a reasonable protocol for postoperative chylothorax management to improve outcomes is necessary.

INTRODUCTION

Postoperative chylothorax following congenital heart surgery is rare; however, it could lead to substantial morbidity and mortality [Lopez-Gutierrez 2014]. The mechanisms of postoperative chylothorax are well known, including direct injury to the lymphatic channel or thoracic duct, central venous hypertension, and venous thrombosis [Efrati 2002]. Therefore, this complication could occur after any type of surgery requiring mobilization around the thoracic duct, mainly the patent ductus arteriosus and coarctation of the aorta. Moreover, postoperative chylothorax could occur in patients undergoing bidirectional or total cavopulmonary shunt, due to increased venous pressure. Continuous leakage of the lipid-rich and protein-rich chyle leads to nutritional complications, volume depletion, and immune suppression. Furthermore, it could contribute to longer intensive care unit or hospital stays.

The management of chylothorax consists of medical and surgical methods. Medical therapies are aimed to improve the lung condition through chest tube insertion or aspiration of the chyle and to decrease the intestinal lymphatic drain through a low fatty acid and medium-chain triglyceride (MCT) diet or total parenteral nutrition (TPN). Octreotide treatment and therapies using similar drugs are also management options for chylothorax; however, their mechanisms still are unclear. Surgical management includes direct ligation of the thoracic duct, simple covering of the fistula with a fibrin-sealing agent, pleurodectomy, or pleurodesis [Liu 2005].

The incidence of postoperative chylothorax has been reported as 2% and 5% [Chan 2005; Nguyen 1995; Zuluaga 2012]. Mery et al [Mery 2014] obtained data from the Pediatric Health Information System database and reported that the occurrence of chylothorax was 2.8% and that the incidence of chylothorax increased from 2.0% in 2004 to 3.7% in 2011. Cavopulmonary shunt, correction of the transposition of the great arteries, and heart transplantation were associated with a higher incidence of chylothorax.

We evaluated the incidence of postoperative chylothorax and compared the data of patients who received conservative management and those of patients who received surgical management. We also compared our postoperative chylothorax incidence and treatment methods with those of other research studies.

MATERIALS AND METHODS

Patients

This study was approved by the ethics committee/ institutional review board of our institution. Between January 2009 and December 2018, a total of 2,515 congenital heart repair surgeries were performed at our center. We reviewed

Received April 28, 2020; received in revised form May 12, 2020; accepted May 17, 2020.

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Table 1. Incidence of postoperative chylothorax according to the surgical approach

	N/total	%
Overall	36/2515	1.4
PDA ligation	8/196	4.1
Vascular ring repair	3/32	9.4
CoA repair	4/173	2.3
Norwood operation	2/28	7.1
BCPS	3/84	3.6
Fontan	6/73	8.2
ToF repair	4/220	1.8
TAPVR repair	1/68	1.5
Other	5/1636	0.3

PDA: patent ductus arteriosus, CoA: coarctation of the aorta, BCPS: bidirectional cavopulmonary shunt, ToF: tetralogy of Fallot, TAPVR: total anomalous pulmonary venous return

Table 2. Comparisons of the medical and surgical management groups

	Medical management group (N = 31)	Surgical management group (N = 5)	*P*
Age (months)	10.6 ± 14.7	0.27 ± 0.28	<.001
Body weight (kg)	6.5 ± 3.7	4.0 ± 3.7	.170
Sex (male)	23	3	.603
Neonate	12	5	.016
Single ventricle	12	1	.634
CPB use	10	2	1.000
Thoracotomy	9	2	.631
Redo*	18	4	.628
Side †	17	3	1.000
Octreotide start (day)	26.8 ± 25.6	10.9 ± 9.9	.239
Octreotide usage (day)	11.2 ± 11.5	16.6 ± 8.6	.321
Hospital stay	44.5 ± 33.9	190.2 ± 184.5	.152
Mortality	3	3	.024

*Included the second and third surgery
†Excluded both sides
CPB: cardiopulmonary bypass, octreotide start (day): the time interval from the day of initial surgery to the day of starting octreotide

the medical records of all patients younger than 18 years, who received treatment for postoperative chylothorax after congenital heart surgery. However, patients with incomplete or inaccurate data were excluded.

Statistical analysis

Data analyses were performed using IBM SPSS statistics, version 20.0 (IBM Corp., Armonk, NY). Continuous data are presented as means with standard deviations and ranges. Frequencies are presented as absolute numbers and percentages. We used the chi-square test to compare categorical variables, and t-tests to compare continuous variables. A *P*-value of <.05 was set as the level of statistical significance.

Literature review

We searched for related articles about postoperative chylothorax that were published in PubMed since 2010. We searched for "cardiac surgical procedure" (MeSH) or "heart defects, congenital" (MeSH) and "chylothorax" (MeSH), and we limited the articles to those that involved patients younger than 18 years and were published in English only. We found 156 articles and reviewed their abstract; we excluded case reports, animal studies, studies with a different scope, and studies with fewer than 2,000 patients.

RESULTS

A total of 36 cases of chylothorax were identified following congenital heart repair surgery during the study period. The mean patient age at the time of congenital heart surgery was 9.1 months, and the mean body weight was 6.16 kg. During the study period, the overall incidence of postoperative chylothorax after congenital heart repair surgeries was 1.4%. Table 1 shows the incidence of postoperative chylothorax, according to the type of surgery performed (Table 1). The incidence was the highest in the vascular ring group (3/32). Moreover, the incidence was higher for single ventricle-related procedures than for bi-ventricle procedures (5.6% versus 1.0%; $P < .0001$). The thoracotomy incision was applied for 11 patients, and 22 patients had no previous operative history. Chylothorax predominantly was found on the left side. (20/36). Postoperative chylothorax occurred on a mean of 13.2 days after surgery.

Initially, all patients were managed as nil per os (NPO) and TPN with octreotide at least a week, if there was no change in the chest tube drainage. Then, the patients were started on MCT milk. The mean duration of octreotide usage was 11.9 days. Medical treatment was not successful for five out of 36 patients. These patients underwent thoracic duct ligation at 42.0 days after the initial surgery and 15.2 days after the start of octreotide treatment. Table 2 lists the potential risk factors for surgical treatment, which have been analyzed in our database (Table 2). There were six deaths (16.7%); of these, two died during the medical management, one died of another cause at 2.8 years later, and three died after the surgical management because of sepsis, severe bronchopulmonary dysplasia, and multi-organ failure at one month, eight months, and two days, respectively.

Ten previous studies within our scope were included, and we reviewed their data to obtain information, regarding the incidence of chylothorax. The postoperative chylothorax incidence was varied, depending on the study group. The incidence of postoperative chylothorax of the 10 studies ranged

Table 3. Data of studies reporting the incidences of postoperative chylothorax

No.	Author	Year	Total no. of patients	Patients with Chylothorax	Incidence (%)
1	Pego-Fernandes	2011	3092	64	2.07
2	Hoskote	2012	3924	133	3.39
3	Mery	2014	77777	2205	2.84
4	Haines	2014	3528		3.2
5	Law	2015	2189	112	5.12
6	Buckley	2017	4864	185	3.80
7	Christofe	2017	4099	87	2.12
8	Day	2018	2273	121	5.32
9	Hirata	2018	13685	47	0.34
10	Buchwald	2018	2255	106	4.70

Year: year of published

from 0.34% to 5.32%, with a mean incidence of 3.3%. The incidence of our study was 1.4%.

DISCUSSION

Despite the improvement in cardiac surgical techniques, cardiac anesthesia, and postoperative care, the incidence of postoperative chylothorax has not decreased because of the increasing number of complicated cardiac surgeries during the neonatal period and palliative surgeries of single ventricle [Beghetti 2000]. In this study, the incidence of postoperative chylothorax was 1.4%, which was comparable to those of previous reports (0.34%-5.32%) (Table 3). The thoracic duct typically extends from the cisterna chyli to the left jugulovenous angle [Higgins 1971]. However, the thoracic duct has many anatomic variations. It is estimated that only 40% to 60% of patients have the typical anatomic course of the thoracic duct [Chen 2011]. Lymphatic system development is similar to that of the vessel system; therefore, it develops from hemangioblastic stem cells at the end of the sixth week of life [van der Putte 1980]. Lymphatic clefts eventually form an extensive plexus between each other and fuse to create a larger pathway for lymphatic drainage, including the right and left thoracic ducts of the embryo. The formation of lymph nodes mainly begins during the ninth week of life, with the simultaneous regression of many previously formed plexus. Through the selective regression within the thorax and abdomen, a single conduit remains that drains the lymph fluid from the lower body and left side of the upper body; that fluid then flows into the left subclavian vein and left internal jugular vein. The right-side lymphatic fluid flows into the right side of the corresponding vessels. The many anatomic variants of the thoracic duct occurred because of the disturbance or developmental impediment, during the selective regression process [Chen 2006; Kwon 2002]. This indicates that performing surgery with more attention focused on the left side does not guarantee good outcomes; instead, we should be more careful when handling the thoracic duct during surgery.

Some studies performed a risk factor analysis of postoperative chylothorax. Although we did not perform this analysis, we used the reported risk factors in our study. The single ventricle was the only risk factor showing a statistically significant association with the incidence of postoperative chylothorax. Single-ventricle palliation, such as the bi-directional cavopulmonary shunt or Fontan procedure, has been related to postoperative chylothorax [Zuluaga 2002]. In this study, neonatal age was a risk factor for the surgical management of postoperative chylothorax.

There is no standard treatment for postoperative chylothorax. However, its current management aims to reduce the lymph flow through the thoracic duct to allow spontaneous healing of the thoracic duct. Management of postoperative chylothorax includes medical, surgical management, and interventional treatments. Medical and surgical treatments are performed in continuity. If the patient does not respond to the medical treatment, then surgical treatment is considered. Generally, medical management consists of NPO or enteral nutrition with MCT milk, or TPN, followed by the use of octreotide. Octreotide is used sporadically, but its mechanism has not extensively been studied. Moreover, during surgical management, mass ligation of the right thoracic duct via open thoracotomy was usually performed, even though both sides of the chest are involved [Liu 2005]. However, Bang et al [Bang 2015] recommended that left-side periaortic mass ligation should be considered for chylothorax that continues after right-side thoracic mass ligation because the path of the thoracic duct may vary in patients. Alternate treatment methods, such as pleuro-peritoneal shunting or interruption of the thoracic duct with interventional radiological techniques, also have been suggested. Milsom et al [Milsom 1985] and Cummings et al [Cummings 1992] reported the effectiveness of pleuro-peritoneal shunting methods. Chen et al [Chen 2011] showed that interventional treatments for chylothorax resulted in good outcomes. Savla et al [Salva 2017] recommended lymphatic embolization as the first-line treatment for

postoperative chylothorax, especially for patients with traumatic leakage and pulmonary lymphatic perfusion syndrome. To determine the cause, they performed dynamic contrast-enhanced magnetic resonance lymphangiography.

Czobor et al [Czobor 2017] reported their therapeutic algorithm for the treatment of chylothorax. Initially, they applied medical management, such as NPO, MCT/TPN, octreotide use, and drainage. Then, they started the MCT diet first, and if the patient showed no improvement for 24 hours, TPN was started. After four days, they used octreotide. If the octreotide was ineffective, they then considered surgical management. Some centers recommended surgical management within five to seven days to decrease the risk of morbidity and mortality [Ismail 2014; Chalret 2011]. At our center, we have no management protocol for chylothorax. After the diagnosis of chylothorax, patients were placed on NPO for one week with octreotide. The octreotide dosage was increased to 10 μg/kg/hour. After one week, if the patient's condition was improving, then MCT milk was initiated for three months without octreotide. However, if there was no improvement in the patient's condition but the patient had stable vital signs, then we continued with medical management. If the patient's condition still did not improve, we then considered surgical management. Therefore, the duration of octreotide usage was relatively long: approximately 12 days and 16.6 days for the surgical management group. Moreover, the results of surgical treatment were not good. We considered that this was due to the delayed timing of surgical treatment, which was relatively late compared with that of other institutions. Based on our data, we will try to establish a management protocol for postoperative chylothorax to improve the patient outcomes.

Study limitations: This study had some limitations, owing to its retrospective nature. First, the number of patients in the surgical management group was small. It was difficult to conclude the risk factors. Second, our study was a single-center study, and the population was heterogeneous in terms of age and type of congenital heart surgery, which could have influenced the results. We need more follow up to establish a management protocol.

CONCLUSION

The incidence of postoperative chylothorax in this study was comparable to those of other studies. However, a suitable protocol for the management of postoperative chylothorax still is needed to decrease the risk of morbidity and mortality.

ACKNOWLEDGMENTS

This study was supported by Research Institute for Convergence of biomedical science and technology, Pusan National University Yangsan Hospital.

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