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Clinical Research

Chylothorax after Surgery for Congenital Cardiac Disease: A Prevention and Management Protocol

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Background: Chylothorax after congenital heart surgery is not an uncommon complication, and it is associated with significant morbidity. However, consensus treatment guidelines are lacking. To improve the treatment outcomes of patients with postoperative chylothorax, we implemented a standardized management protocol at Severance Hospital in September 2014.
Methods: A retrospective review of patients treated at a single center was done. All corrective and palliative operations for congenital heart disease performed at our institution between January 2008 and April 2018 were reviewed. The incidence and treatment outcomes of postoperative chylothorax were analyzed.
Results: The incidence of chylothorax was 1.9%. Sixty-one percent of the patients could be managed with a low-fat diet, while 28% of the patients required complete restriction of enteral feeding. Thoracic duct embolization was performed in 2 patients and chest tube drainage decreased immediately after the procedure. No patient required thoracic duct ligation or pleurodesis. After implementation of the institutional management protocol, the number of chest tube drainage days decreased (median, 24 vs. 14 days; p=0.45).
Conclusion: Implementing a strategy to reduce postoperative chylothorax resulted in an acceptable incidence of postoperative chylothorax. Instituting a clinical practice protocol helped to curtail the treatment duration and to decrease the requirement for surgical treatment. Image-guided embolization of the thoracic duct is an effective treatment for postoperative chylothorax.

Keywords: Chylothorax, Congenital heart defects, Postoperative complication, Postoperative care

Introduction

Postoperative chylothorax after surgical procedures for congenital heart disease poses a challenging clinical situation. Its incidence varies across centers, with a reported range from 2% to 5% [1-4]. Postoperative chylothorax is associated with poor nutrition, an electrolyte imbalance, and a higher risk of thrombus development, and its clinical impact includes increased mortality, increased length of hospital stay, increased length of intensive care unit stay, prolonged mechanical ventilation, and a higher likelihood of extracorporeal membrane oxygenation support [4].

From a surgical perspective, the mechanism of postoperative chylothorax can be categorized as traumatic or non-traumatic. Traumatic chylothorax is caused by direct injury of the thoracic duct or one of its lymphatic branches. Non-traumatic chylothorax is caused by unfavorable hemodynamics, lymphatic malformation, or congenital syndromes such as Noonan syndrome and Turner syndrome. Traditionally, treatment options included diet modification and octreotide infusion as conservative management, and thoracic duct ligation and pleurodesis as surgical management [5-7]. In recent years, new imaging modalities such as magnetic resonance lymphangiography have improved our understanding of chylothorax, and studies on thoracic duct embolization demonstrated an acceptable success rate in patients in whom thoracic duct ligation failed [8-10].

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Although many treatment options for postoperative chylothorax exist, there is no generally accepted standardized treatment guideline. At Severance Hospital, a standardized management protocol for postoperative chylothorax was developed and has been implemented since September 2014. The aim of this study was to determine the incidence of postoperative chylothorax after surgery for congenital heart disease and to evaluate the efficacy of implementing a chylothorax management protocol.

Methods

Patients

This is a retrospective review of experiences at a single center. The medical records of all patients who underwent corrective or palliative operations for congenital heart disease at Severance Hospital from January 2008 to April 2018 were reviewed. This study was reviewed and approved by the Institutional Review Board (IRB approval no., 4-2019-0903). The obligation to obtain informed consent was waived by the institutional review board in view of the retrospective nature of the study.

Diagnosis of chylothorax

Chylothorax was suspected when persistent drainage from an existing chest tube became cloudy when the patient was fed, or when similar milky fluid was found after placing a tube into a newly discovered effusion. The diagnosis of chylothorax was confirmed based on the characteristics of the pleural fluid recovered from each patient. Chylothorax was diagnosed if any of the following criteria was met: (1) the pleural fluid triglyceride level was >100 mg/dL, (2) the percentage of lymphocytes in the pleural fluid was >80%, or (3) the pleural fluid triglyceride level was higher than that of the serum.

Postoperative chylothorax was defined as chylothorax developing within 30 days of surgery. Based on the maximum cumulative output of all chest tubes for a 24-hour period, chylothorax was classified high (≥20 mL/kg/day) or low (<20 mL/kg/day). The recurrence of chylothorax was diagnosed when a drainage tube was re-inserted for chylous effusion. Chylothorax duration was defined as the time interval from the day of diagnosis to the day of chest tube removal.

Institutional policy to reduce postoperative chylothorax

To minimize the development of postoperative chylothorax, we follow our institutional policy. During the operation, we do not remove the thymus routinely. Even in neonates or for aortic arch repair, we preserve the thymus whenever possible. If the thymus is large, by splitting its left and right lobes and retracting the lobes with pericardial tenting sutures, good exposure of the aortic arch and pulmonary arteries can be obtained. Cannulation of the innominate artery for selective cerebral perfusion can be carried out without thymus resection. Dissection of the aortic arch and superior vena cava (SVC) is minimized if not indicated. We routinely place a SVC drainage cannula through the right atrium and avoid direct cannulation of the SVC. The indications for direct SVC cannulation are superior cavopulmonary anastomosis and anomalous pulmonary venous connection to the SVC.

The postoperative management policies are as follows: (1) early removal of the central venous catheter, (2) avoidance of hyperalimentation, and (3) therapy to lower the SVC pressure in patients with physiologically high SVC pressure (e.g., those who have undergone a Fontan operation or received bidirectional cavopulmonary shunt), which includes diuresis, pulmonary toileting, supplemental oxygen, pulmonary vasodilator use, nitric oxide inhalation, and afterload reduction.

Management protocol for postoperative chylothorax

As a quality improvement program, we established a standardized management protocol for postoperative chylothorax. The protocol was developed over 3 months, including a period of literature review and gathering the opinions of multidisciplinary team of experts, after which it was implemented in September 2014. The protocol was then revised in 2017 (Fig. 1).

Based on the chest tube drainage amount, the patients are classified as having low- or high-output chylothorax, according to which a different initial management strategy is applied. The diet of patients with low-output chylothorax is modified to a medium-chain triglyceride (MCT) or low-fat diet. In patients with high-output chylothorax, early adoption of nil per os (NPO) with total parenteral nutrition (TPN) treatment is applied. Surgical thoracic duct ligation, intravenous infusion of octreotide, and pleurodesis are sequentially considered as treatment options for

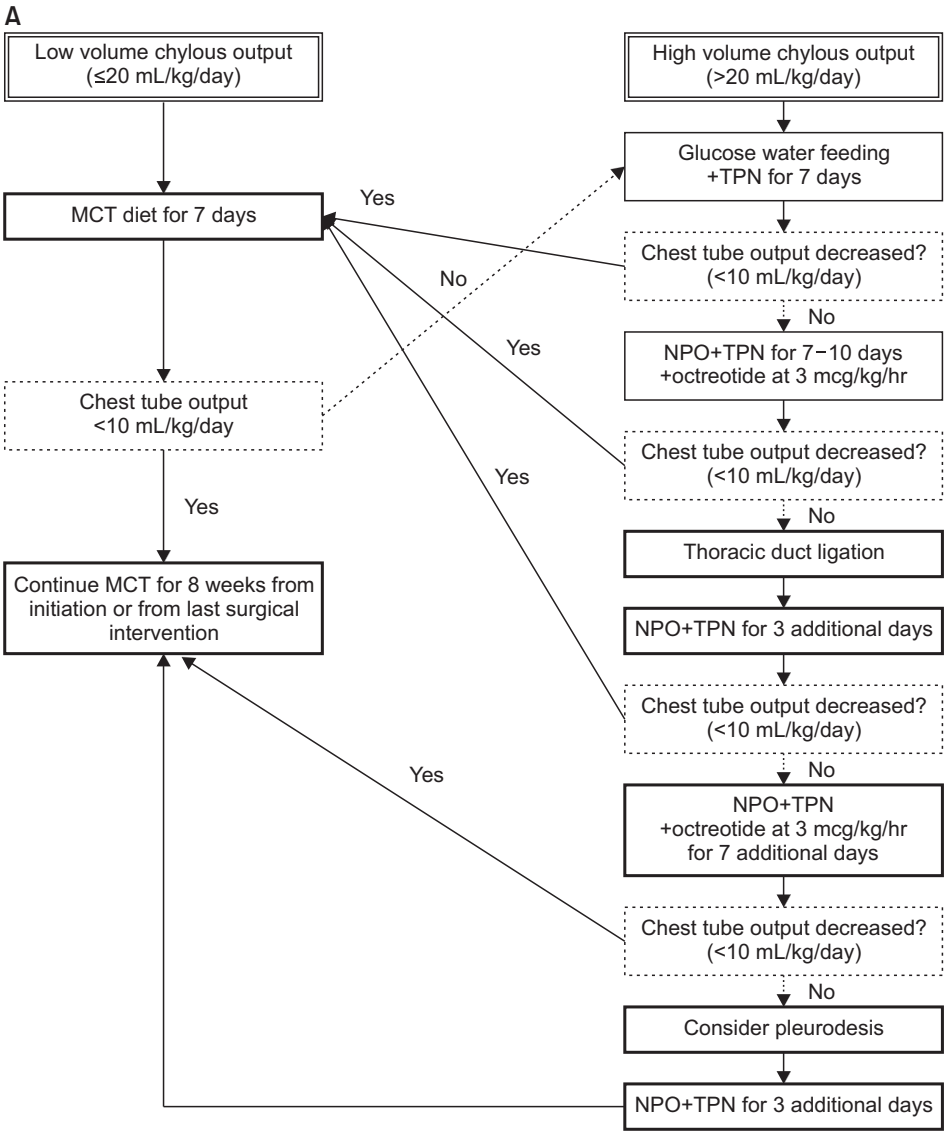

Fig. 1. Management protocol for postoperative chylothorax for children (A) and adults (B). MCT, medium chain triglyceride; NPO, nil per os; TPN, total parenteral nutrition. (Continued on next page).

non-responders. The version of the protocol revised in 2017 incorporates lymphangiography and thoracic duct embolization as the first choice of treatment modality for adult-sized patients. For the NPO and TPN treatment groups, enteral intake of water, a glucose-water solution, or a balanced ion solution with no fat components is allowed.

Lymphangiography and thoracic duct embolization are performed by interventional radiologists. Inguinal lymph nodes are accessed under real-time ultrasound guidance with a 25-gauge needle, and lymphangiography is performed using an oil-based contrast agent (Lipiodol; Guerbet, Bloomington, IN, USA). After visualization of the thoracic duct and identification of a target lymphatic vessel, a 21-gauge long Chiba needle is placed in the target lymphatic vessel under fluoroscopic guidance. Next, a microcatheter is placed in the lymphatic vessel and contrast dye is injected to identify the chylous leak. The thoracic duct is embolized with an embolization coil and/or a mixture of cyanoacrylate (Histoacryl; B. Braun, Barcelona, Spain) and Lipiodol (Guerbet).

Statistical analysis

The incidence of chylothorax was calculated for all corrective or palliative surgical procedures for congenital heart disease. Continuous variables were summarized as median with range. Categorical and ordinal variables were presented as frequency and percentage. Group compari-

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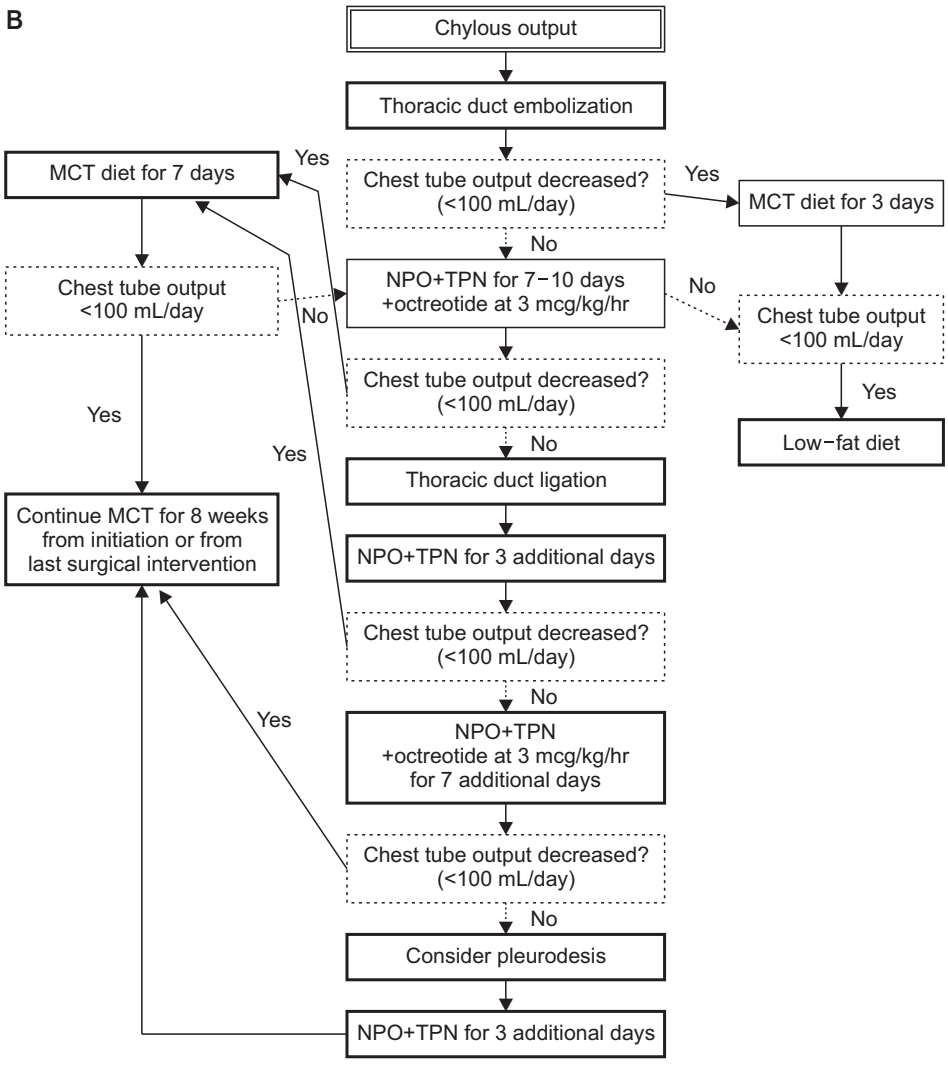

Fig. 1. (Continued; caption shown on previous page).

sons were made using the Wilcoxon rank sum test. All p-values <0.05 were considered to indicate statistical significance. Statistical analysis was performed using IBM SPSS ver. 23.0 (IBM Corp., Armonk, NY, USA).

Results

Incidence of postoperative chylothorax

There were 2,465 corrective or palliative surgical procedures in the study period, and 46 patients developed postoperative chylothorax. The incidence was 1.9%. In 16 patients (35%), chylothorax developed after postoperative day 7. In 15 patients (33%), the chest tube drainage was classified as high-output (>20 mL/kg/day). The characteristics of the patients are summarized in Table 1.

Table 1. Characteristics of patients with postoperative chylothorax (n=46)

Characteristic	Variable
Male sex	29 (63)
Age at operation	2.7 mo (1 day–46 yr)
Body weight (kg)	4.7 (2.6–107)
Single ventricle	12 (26)
Timing of chylothorax occurrence (day)	
<3	7 (15)
3–7	23 (50)
>7	16 (35)
Output of chyle	
High output (>20 mL/kg/day)	15 (33)
Low output (≤20 mL/kg/day)	31 (67)

Values are presented as number (%) or median (range).

Twenty-six percent of cases of postoperative chylothorax occurred in patients with a functional single ventricle. The incidence of postoperative chylothorax was markedly higher after repair of a vascular ring (3 of 11, 27%), the Fontan operation (5 of 21, 24%), and the Norwood operation (2 of 10, 20%) than after other procedures (Table 2).

Table 2. Incidence of postoperative chylothorax based on type of surgery and RACHS category

Variable	Occurrence of chylothorax (%)
Type of surgery	
Ventricular septal defect repair	9 (1)
Coarctation of the aorta repair	2 (4)
Systemic to pulmonary artery shunt	3 (9)
Arterial switch operation	1 (2)
Bidirectional cavopulmonary shunt	3 (5)
Fontan operation	5 (24)
Norwood operation	2 (20)
Vascular ring repair	3 (27)
Total anomalous pulmonary venous connection repair	3 (10)
Tetralogy of Fallot repair	4 (9)
Others	11 (1)
RACHS category	
I	4 (9)
II	19 (41)
III	18 (39)
IV	2 (4)
V	0
IV	2 (4)
Not categorized	1 (2)

RACHS, risk adjustment for congenial heart surgery.

Treatment modalities

In 28 patients (61%), the chylothorax could be managed with only an MCT or low-fat diet. Restriction of food except for water or a glucose-water solution was required in 13 patients (27%). Intravenous somatostatin was administered in 4 patients (9%). Lymphangiography and thoracic duct embolization were performed in 2 adult patients, in whom chest tube drainage decreased immediately after the procedure. No patient required surgical thoracic duct ligation or pleurodesis. Data on the treatment modalities, chylothorax duration, and hospital stay are summarized in Table 3.

Percutaneous thoracic duct embolization was performed in 2 patients, and it was effective for controlling the chylous drainage (Fig. 2). A 46-year-old woman was diagnosed with postoperative chylothorax on postoperative day 16 af-

Table 3. Treatment modalities for postoperative chylothorax

Treatment modality	No. (%)	Chylothorax duration (day)	Hospital stay (day)
MCT or low-fat diet	44 (96)		
MCT or low-fat diet only	28 (61)	9 (6–49)	23 (7–331)
Nil per os	13 (28)	17 (3–66)	28 (15–93)
Octreotide	4 (9)	35 (17–196)	50 (22–196)
Lymphangiography and thoracic duct embolization	2 (4)	6 (5–6)	37 (21–38)
Thoracic duct ligation	0		
Pleurodesis	0		
Protocol based approach	20 (43)	14 (6–39)	21 (9–184)

Values are presented as median (range), unless otherwise stated.
MCT, medium chain triglyceride.

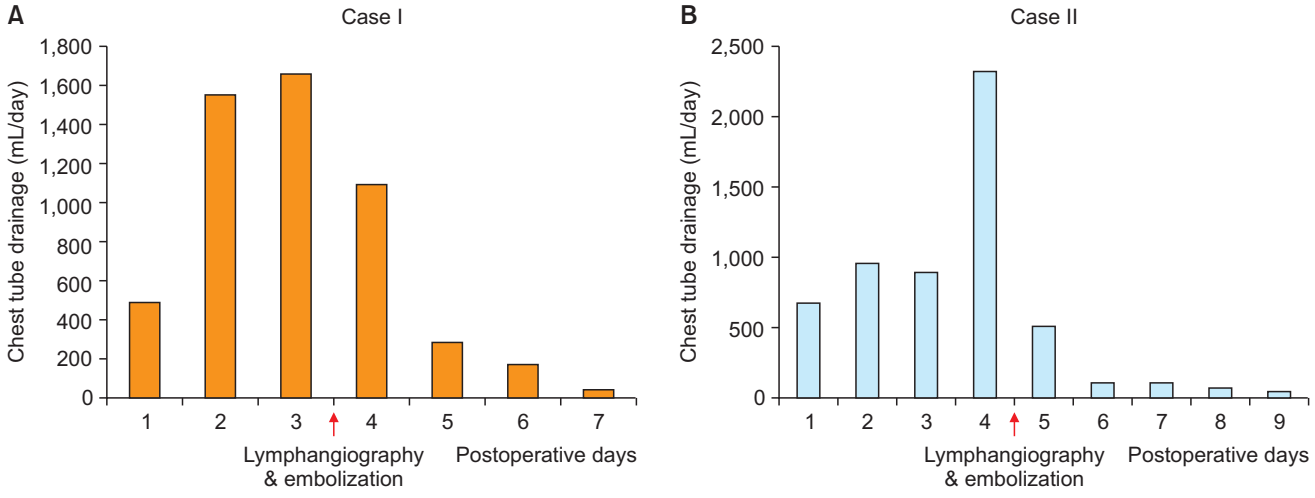

Fig. 2. (A, B) Chest tube drainage amount of the 2 patients who underwent lymphangiography and thoracic duct embolization for postoperative chylothorax.

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Table 4. Impact of a protocol-based approach on outcomes

Variable	Pre-implementation (n=26)	Post-implementation (n=20)	p-value
Era	Before September 2014	After September 2014	
Amount (mL/kg/day)	18 (1.0–115)	13 (0.9–79)	0.48
Chylothorax duration (day)	24 (6–196)	14 (6–39)	0.045
Hospital stay (day)	37 (7–331)	21 (9–184)	0.067
In-hospital death	2	0	0.20
Recurrent chylothorax	2	2	0.78

Values are presented as median (range), unless otherwise stated.

ter the repair of pulmonary valve stenosis. On postoperative day 18, lymphangiography and thoracic duct embolization were carried out. Her chest tube drainage decreased from 1,500 mL/day pre-procedure to 200 mL/day on the day after the procedure. The chest tube was removed 3 days after thoracic duct embolization. The other patient was a 20-year-old man who underwent vascular ring repair. His chest tube drainage was 2,300 mL/day despite full restriction of enteral intake, but decreased to 510 and 110 mL/day on post-procedure days 1 and 2. There were no procedure-related complications or recurrence of chylothorax.

Comparison of clinical outcomes before and after implementation of the treatment protocol

The median chest tube drainage duration of all chylothorax patients was 20 days (range, 6–196 days). In our cohort of 46 patients with postoperative chylothorax, 26 (57%) were diagnosed before September 2014, when our management protocol was implemented. The number of chest tube drainage days decreased after implementation of the protocol (median, 24 versus 14 days; p=0.045). The length of hospital stay also decreased (median, 37 versus 21 days; p=0.067), with marginal statistical significance. Chylothorax recurred in 2 patients both before and after implementation of the protocol (p=0.78). There were 2 in-hospital deaths in the pre-implantation period, whereas no in-hospital mortality occurred in the post-implementation period (p=0.20) (Table 4).

Discussion

In our patient cohort, the incidence of postoperative chylothorax for congenital heart surgery was 1.9%. This is lower than has been reported in other previous reports [1-4]. Therefore, we think that our intraoperative and postoperative management strategy to reduce postoperative chylothorax is effective. To prevent traumatic damage to lymphatic channels, we minimize the dissection of the structures around the lymphatic pathway. In neonates and infants, the thymus is normally large, which limits the exposure of the great vessels. The fissure between the left and right lobes of the thymus can be easily dissected, and lateral retraction of the right and left lobes with pericardial tenting provides good exposure of the aortic arch and the branch pulmonary arteries. In most cases, neonatal aortic arch repair or repair of tetralogy of Fallot can be performed without resection of the thymus. Another surgical precaution is dissection around the SVC. To avoid dissection around the SVC, we do not directly cannulate the SVC unless indicated. The SVC is directly cannulated only for repair of an anomalous pulmonary venous connection to the SVC or superior cavopulmonary anastomosis. Creation of a superior cavopulmonary connection or a Fontan procedure leads to elevated SVC pressure and can result in lymph leakage. These operations are known to be high-risk procedures for postoperative chylothorax. In our cohort, 24% of the patients developed chylothorax after the Fontan operation, but the incidence was only 5% after placement of a bidirectional cavopulmonary shunt. This suggests that our strategy of minimal intraoperative dissection and postoperative SVC pressure-lowering management is effective for reducing the incidence of chylothorax. In our cohort, 35% of the cases of chylothorax developed later than postoperative day 7. This suggests that non-traumatic causes are an important factor in chylothorax development. In this regard, postoperative management is also important to prevent postoperative chylothorax. Central venous lines should be removed as early as possible [11] and medical management should have the goal of hemodynamic stability and lowering the central venous pressure.

Protocol-based management has been proposed to improve the outcomes of chylothorax, and studies have shown that implementing such management strategies led to reductions in chest tube utilization time and improvements in clinical outcomes [12-14]. We developed a management protocol and implemented it in September 2014. The number of chest tube utilization days decreased (median, 24 to 14 days; p=0.045), and the length of hospital stay also decreased (median, 37 to 21 days; p=0.067). There was no significant difference in in-hospital mortality or the recurrence of chylothorax. Even before implementing the protocol, physicians at our institution treated the patients in a

similar way to that described in the protocol. However, the protocol more specifically stipulated the relevant details, which helped to decrease the variability in practice among physicians and led to a shortened treatment duration.

Recently, percutaneous lymphatic embolization has been reported as a successful treatment option for patients with postoperative chylothorax [10]. In our cohort, lymphangiography and interventional thoracic duct embolization were performed in 2 patients. These patients were adults who underwent vascular ring repair and pulmonary valve replacement. In both patients, the daily chest tube drainage was more than 1,500 mL, but after embolization, the chylous drainage decreased immediately and the chest tubes could be removed in 4 days. Based on our experience, we revised our management protocol to consider lymphangiography and lymphatic embolization as the first treatment option in adult-sized patients. Our experience is limited to adults, but according to the case series published by Savla et al. [10], lymphatic embolization was successfully performed in young infants, suggesting new possibilities for chylothorax treatment, even for younger patients.

The main limitation of this study is that it presents a retrospective analysis of a limited number of patients. We compared clinical outcomes between before and after the implementation of the treatment protocol. The protocol helped to decrease variability in practice, but the management of patients did not always strictly adhere to the protocol. Each patient's characteristics and medical conditions influenced small details of the management. With the accumulation of more data and advances in medical practice, the protocol should be revised further to obtain the best treatment results for this postoperative problem.

In conclusion, the utilization of a strategy to reduce postoperative chylothorax after surgical procedures for congenital heart disease resulted in an acceptably low incidence. Implementation of a standardized treatment protocol for postoperative chylothorax helped to reduce the treatment duration and to improve treatment outcomes. A strategy focusing on protocol-guided conservative management and image-guided embolization of the thoracic duct helped to reduce the need for surgical treatment for postoperative chylothorax.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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