Cardiology in the Young

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Chylothorax: a rare postoperative complication in paediatric cardiac surgery patients – a 15-year retrospective study from a tertiary care centre in a developing country

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Original Article

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Abstract

Chylothorax, a postoperative complication of CHD surgery, involves chyle accumulation in the pleural cavity, posing challenges in diagnosis and management. This retrospective study investigates the prevalence, aetiology, management, and outcomes of postoperative chylothorax in paediatric patients undergoing cardiac corrective surgery at a tertiary care centre over 15 years. Medical records of paediatric patients who underwent cardiothoracic surgery at the Children's Heart Center at the American University of Beirut Medical Center between 2007 and 2022 were retrospectively reviewed. Data collection included demographic characteristics, blood parameters, chylous fluid characteristics, diagnostic criteria, treatment modalities, and hospitalisation details. Ethical approval was obtained, and descriptive statistics were employed using SAS 9.4. Among 2,997 children who underwent cardiothoracic surgery, nineteen cases of postoperative chylothorax were identified. The majority were females (63.2%) with a median age of 9 months. Glenn, Fontan, and Blalock–Taussig shunt-related surgeries were the most common operations associated with chylothorax. Single ventricle physiology was the predominant CHD observed (58%). Diagnosis relied primarily on clinical presentation, imaging studies, and triglyceride levels in pleural fluid. Treatment options included conservative dietary modifications, medical therapy such as octreotide, and surgical intervention if necessary. No mortalities were reported, and patients were adequately followed up. This study sheds light on postoperative chylothorax in paediatric cardiac patients, offering insights into its epidemiology, aetiology, clinical features, and treatment outcomes. While conservative and medical approaches effectively managed chylothorax in this group, larger studies are needed to develop standardised diagnostic and treatment protocols, improving outcomes in paediatric patients with postoperative chylothorax.

Introduction

Congenital heart disease (CHD) is the most prevalent birth defect, significantly contributing to infant morbidity and mortality.[1,2] CHD encompasses a range of structural or functional abnormalities in the cardio-circulatory system.[3] Despite improvements in diagnostics and surgical techniques pertaining to the management of CHD, postoperative complications persist, with varying severity.[4] Lung complications, notably chylothorax, are among the most common, highlighting ongoing challenges in CHD management.[5]

Chylothorax, also referred to as a chylous pleural effusion, is defined by the presence and accumulation of chyle in the pleural cavity.[6] Two major lymphatic ducts exist in our body: the left lymphatic duct, commonly known as the thoracic duct, and the right lymphatic duct. The thoracic duct, being the larger of the two, exhibits considerable variability in its anatomy.[7] It ultimately drains into systemic circulation at the junction of the left subclavian vein and the left jugular vein.[8] These anatomical details, alongside associated structures, are illustrated in Figure 1, which represents the classic anatomy. However, complex cardiac defects have been frequently associated with variations in the anatomy and path of the thoracic duct.[9]

When the integrity of the thoracic duct is compromised, the milky lipid-rich chyle may leak into its vicinity, leading to an accumulation of chyle in the pleural space, especially considering that approximately 2.4 litres of chyle are produced daily.[10] In cases where the thoracic duct is

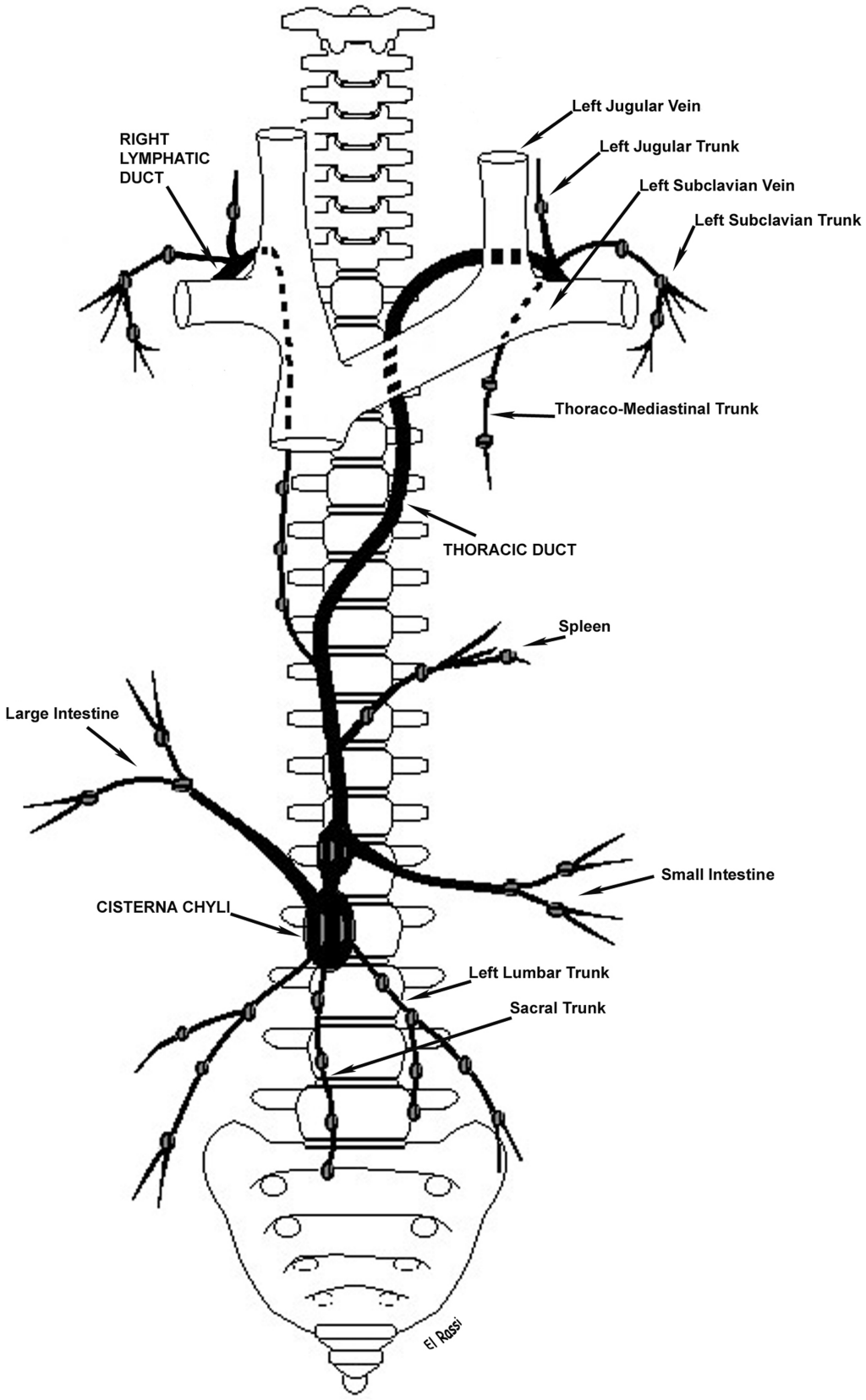

Figure 1. Anatomy and trajectory of the thoracic duct and major lymphatic vessels.

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injured or obstructed, the resulting chylothorax is termed traumatic. It often occurs post-operatively following manipulation in the thoracic cavity, such as Fontan and Glenn surgeries, coarctectomy, esophagectomy, lung resection, and mediastinal lymph node dissection.[11–13] While surgical injury to the thoracic duct and its major afferents is a well-recognised cause, the pathophysiology goes beyond mechanical trauma. Increased venous pressure plays a crucial role, particularly in patients undergoing procedures such as Fontan and Glenn surgeries. In these cases, elevated venous pressure in the upper body or throughout the circulatory system contributes significantly to the development of chylous leaks.[14] This occurs through disruption of lymphatic channels, as the high pressure impairs lymphatic drainage, leading to the leakage of lymphatic fluid into the pleural space, resulting in chylothorax. Additional non-traumatic causes also exist, mainly malignancy-related.[11] Specifically, lymphomas account for the majority of malignant chylothoraces, with other causes including infections, systemic disorders, diseases in the lymph vessels, and miscellaneous factors like retrosternal goitres and cirrhosis being less common.[15] Another subcategory of nontraumatic chylothorax is idiopathic chylothorax, where no identifiable cause is present.[16]

Since chyle consists primarily of lymphatic fluids and fats, including long-chain triglycerides and chylomicrons,[10] its presence outside the circulation requires careful management to minimise additional risks and potential fatalities. Treatment options, generally categorised into conservative, medical, and surgical approaches, depend on severity and other factors.[17] Given that CHD affects approximately 0.8% to 1.2% of live births globally,[18] optimising surgical outcomes and minimising postoperative complications are paramount. This study aims to investigate the prevalence of post-operative chylothorax, along with its aetiology, management, and other characteristics, among paediatric patients undergoing cardiac surgery at a tertiary care centre over a 15-year period.

Methods

In this study, we conducted a retrospective review of medical records pertaining to paediatric patients (< 18 years old) who underwent cardiothoracic surgery at the Children's Heart Center at the American University of Beirut Medical Center. The Children's Heart Center performs around 200 surgeries per year, distributed between simple to complex surgeries and among all the age groups, from neonates to adult patients with CHD. Among 2,997 records over a 15-year period (2007–2022), nineteen patients were identified as having developed postoperative chylothorax. This study received approval from the Biomedical Institutional Review Board (BIO-2023-0054) of the medical centre and was conducted in compliance with the Declaration of Helsinki, the Nuremberg Code, and the Belmont Report, adhering to all relevant ethical standards.

The adopted standard diagnostic criteria for chylothorax include pleural effusion with a triglyceride level exceeding 110 mg/dL or visibly white/turbid drainage from the pleural drain. Aetiological data regarding the type of CHD and the specific surgical corrections were extracted from the patient medical records. Additionally, we collected data on various parameters, including demographic details (age, gender, height, and weight), haematologic parameters (complete blood count, prothrombin time, and partial thromboplastin time), chylous fluid characteristics (triglyceride level and the development of chylous effusion post-surgery), chest tube characteristics (qualitative and quantitative drainage of chest tubes, number of days with chest tube, and location of test tube), and hospitalisation specifics (length of stay and hospital service). Hospital stay specifically refers to the duration from the diagnosis of chylothorax until discharge. Thus, it reflects the time required for the effusions to resolve.

As for the treatment options, standard conservative and medical solutions were provided for the patients. The conservative approach comprised a low-fat/fat-free diet supplemented with medium-chain triglycerides or total parenteral nutrition through a central venous line. The medical option primarily consisted of administering octreotide to the patients. Daily drainage of chylous fluid was carefully measured. Typically, this conservative management lasted for 3–4 weeks, depending on the severity of the case and after ensuring that the in-hospital conservative management of chylothorax could have been extended to a home setting, provided that the patient was stable, and the clinical team deemed it safe to do so. If both options proved ineffective, surgical intervention would have been considered.

Descriptive statistics are presented for continuous variables as either medians with interquartile ranges or as means and standard deviations, depending on the normal distribution. Normality was assessed using histograms. Categorical variables are presented as frequencies with percentages. All analyses were conducted using SAS 9.4 (SAS Institute, Cary, NC, USA).

Table 1. Demographic characteristics

	Patients with chylothorax (N = 19)
Incidence rate (per 100 patients)	0.63
Age (months) median (IQR)	9 (6.5, 42)
Females n (%)	12 (63)
Weight (kg) median (IQR)	8 (6.5, 16.6)
Height (cm) median (IQR)	74 (64, 95)
Median time lapsed between surgery and development of chylothorax (days) (IQR)	19 (8, 30)

Results

At our centre, 22 cases of chylothorax were diagnosed in 19 patients. Three children suffered recurrent chylothorax. Of the identified 19 patients, 12 (63.2%) were females and 7 (36.8%) were males. The median (interquartile range) age was 9 (6.5–42) months, with a median (interquartile range) body weight of 8 (6.5–16.6) kg and a median (interquartile range) height of 74 (64–95) cm. Table 1 outlines the demographic characteristics of these patients.

All reported cases in this study were classified as postoperative chylothorax. The most common surgeries associated with thoracic duct rupturing and subsequent chylothorax were Glenn, Fontan, and BT-shunt insertion/removal surgeries. The combination of Glenn and BT shunt-related procedures accounted for the highest proportion of postoperative chylothorax (32%). Other surgeries mainly included atrial septal defect (ASD) closures and a balloon atrial septostomy. Detailed data are provided in figure 2. CHDs and defects for which these procedures were performed were categorised into five major groups found in figure 3.

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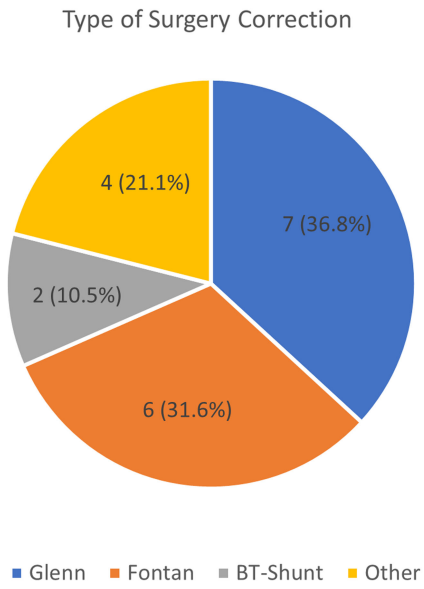

Figure 2. Distribution of correction surgeries at our institution.

As for the diagnosis of postoperative chylothorax, it was primarily based on clinical presentation, imaging studies (using chest X-rays and ultrasound), laboratory evaluation, and aetiological assessment of congenital anomalies. In chest X-rays, a homogenous opacity in the pleural space indicated the presence of fluid. A triglyceride level above 110 mg/dL confirmed the diagnosis, with a median triglyceride level of 667 mg/dL observed in our patients. Additionally, chylomicrons were detected in 6 patients. Details of the pleural fluid analysis are presented in table 2.

The median duration for postoperative chylothorax resolution was 10.5 days. The majority of chylothorax cases occurred on the left side of the chest, occurring in 12 patients (63.2%). Chylothorax was less frequently observed on the right side of the chest, occurring in 6 (31.6%) of our patients. Only one patient exhibited chylothorax of the pericardium (chylopericardium). The median chylous drainage was 10 mL/Kg/24h. Chylous effusions were diagnosed at a median of 19 days post-surgery. The median length of hospital stay for our paediatric patients was 16 days, with the starting point being the clinical diagnosis of chylothorax. Our patients were properly managed with a fat-free or low-fat diet. We typically remove chest drains on the second day post-surgery, provided that the drainage volume is minimal. We have found that waiting until enteral feeding begins can unnecessarily prolong the duration the chest tube is in place, especially since restarting feeding can take several days. Our approach helps in reducing patient discomfort and potential complications associated with prolonged chest tube placement, without significantly impacting the diagnosis or management of chylothorax. Additionally, one of the patients was managed with steroids, and two other patients were managed with octreotide. The choice between steroids and octreotide often depends on the specific clinical scenario and the patient's response to initial interventions. Our protocol typically includes initiating treatment with a fat-free diet supplemented with medium-chain fatty acids and monitoring the patient for signs of improvement over a 1–2-week period. If this dietary approach does not show the desired decrease in lymphatic drainage, we consider pharmacological options. Octreotide, being a somatostatin analogue, is usually our drug intervention of choice due to its efficacy in reducing chyle flow. Steroids are reserved for cases where there is persistent chylous output despite the use of octreotide. In one instance, the patient continued to present with significant lymph drainage despite initial treatments, prompting the use of steroids. This intervention proved effective, significantly reducing lymph output and improving the patient's condition. We do not routinely use anticoagulation and vascular ultrasound solely for chylothorax management unless there is another indication for its use. No mortalities were reported, and all patients (100%) were adequately followed up. Also, none of the patients at our institution underwent surgery to resolve chylothorax because conservative and medical management were successful. Additional details can be found in table 2. As for the three patients with recurrence, they were successfully managed using a medium-chain triglyceride diet.

Discussion

The global prevalence of paediatric postoperative chylothorax remains undocumented, yet several studies have shed light on its incidence within specific regions. The Pediatric Cardiac Critical Care Consortium (PC4) reported a prevalence range of 0 to 10.1%,[19,20] indicating the potential for mitigating factors to reduce its occurrence. A more recent study by Rudrappa and Paul reported a prevalence of 0.2% to 1% following various cardiothoracic surgeries.[21] Another multicentre database analysis ($n = 2205$) by Mery et al. showed that the incidence rate was 2.8%.[22] Similarly, by Lee & Choi reported an overall incidence of 1.4% in their study ($n = 36$).[23] Other studies reported incidence rates ranging between 0.5% and 6.5%,[12] 0 and 4%,[24] and 0.25% and 5.3%.[25] Notably, a recent multicentre study by Buckley et al. that included 4864 surgical hospitalisations from 15 centres found the rate to be 3.8%.[26] Our retrospective review of paediatric patients who underwent cardiac surgery identified nineteen cases of postoperative chylothorax among 2,997 patients from 2007 to 2022, resulting in an incidence rate of approximately 0.63%. This rate is lower than the commonly reported incidence of chylothorax following paediatric cardiac surgeries. To understand this discrepancy, we examined both the numerator (number of chylothorax cases) and the denominator (total at-risk population) in detail. Our denominator accurately reflects all paediatric cardiac surgeries performed at our institution over this 15-year period, encompassing patients who were theoretically at risk for developing postoperative chylothorax. Given that all cases of cardiac surgery were included, we believe this provides a reliable base for calculating incidence. However, our relatively low numerator (nineteen cases) may be influenced by several institutional and procedural factors.

One key consideration is the case mix at our institution. Compared to multicentre reports, our patient population may have a different distribution of CHDs, surgical complexity, and preoperative risk factors. Certain high-risk procedures may have been performed at lower volumes in our centre compared to high-volume paediatric cardiac surgery programs. This may subsequently result in a lower proportion of patients at high risk for chylothorax. Additionally, differences in perioperative management, including fluid administration, nutritional strategies, and chest tube management, may contribute to variations in incidence rates across institutions.

First, surgical expertise may play a significant role. Our centre's surgeries were performed by a single senior paediatric cardiothoracic surgeon with extensive experience, which could contribute to fewer postoperative complications, including chylothorax.

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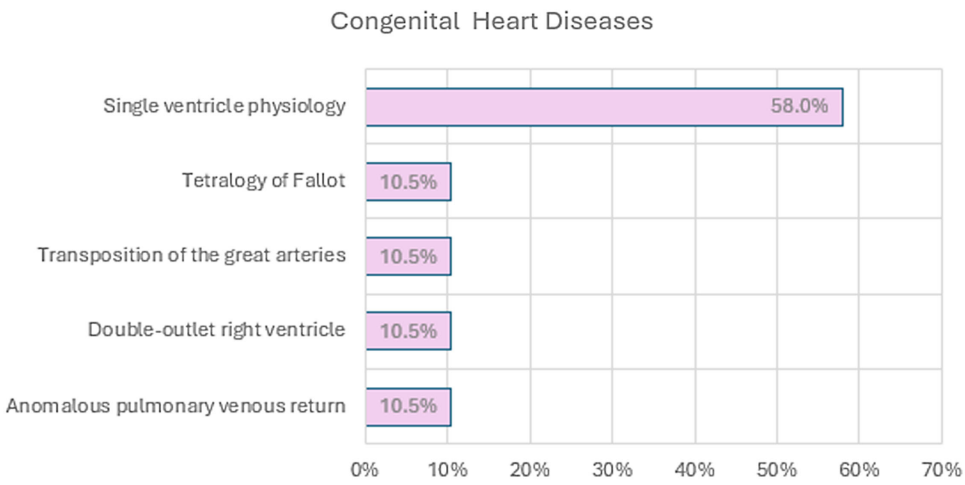

Figure 3. Prevalence of CHDs at our institution.

Second, variations in diagnostic practices may have impacted case identification. It is possible that mild or transient cases of chylothorax were underdiagnosed, particularly if the condition resolved spontaneously or did not meet clinical thresholds for diagnosis during routine follow-up. Additionally, our institution's specific postoperative management protocols and patient follow-up processes may differ from those in centres reporting higher incidence rates, potentially influencing the frequency with which chylothorax cases are detected and recorded. In our cohort, the age range was wide, with the eldest being 16 years old and the youngest just a day-old full-term neonate. Notably, the second oldest patient was 7 years old, while ten patients were under a year old, resulting in a median age of 9 months. Lin et al. found a similar pattern, reporting a mean age of 11.5 months in their study with comparable data parameters.[27] However, additional research is needed to fully understand the age-related differences in susceptibility to chylothorax. Regarding gender, existing literature does not indicate a specific predisposition for males or females to develop postoperative chylothorax. In our study, most of our patients were females ($n = 12$).

While the primary risk factor for postoperative chylothorax in our cohort was the type of surgery, other contributing factors should be considered. Elevated central venous pressure, prolonged mechanical ventilation, and extensive mediastinal dissection may predispose patients to chylous effusions. Additionally, patient-specific factors, including younger age, lower body weight, and pre-existing lymphatic anomalies, have been implicated in increased susceptibility to postoperative chylothorax.[23] In our cohort, although single ventricle physiology was the predominant CHD observed, the relatively low incidence of chylothorax suggests that institutional surgical expertise, postoperative fluid management strategies, and early dietary modifications may also influence risk. However, a formal comparison between patients with and without chylothorax would be required to establish predictive factors definitively. Future studies incorporating a larger control group without chylothorax could provide further insights into non-surgical predictors and guide preventative strategies.

In our study, operations involving cavopulmonary anastomoses emerged as the primary procedures associated with chylothorax. Specifically, Glenn, Fontan, and BT shunt-related surgeries were linked to postoperative chylothorax. The Glenn procedure involves the creation of an anastomosis between the superior vena cava and the right pulmonary artery. The current modified BT-shunt procedure establishes a systemic-pulmonary shunt without sacrificing major arteries. In the Fontan procedure, modern techniques involve either an extracardiac conduit or a lateral tunnel to reroute systemic deoxygenated blood to the pulmonary vasculature.[28] This observation aligns with findings from Mery et al.'s multicentre study, which highlighted cavopulmonary anastomoses as having the highest incidence of postoperative chylothorax.[22]

The three procedures mentioned are frequently employed in correcting single ventricle aetiology, the most prevalent CHD in our cohort ($n = 11$). In a study tracking patients undergoing total cavopulmonary connection procedures, 50% of those with post-operative chylothorax also had single ventricle physiology, compared to 36.8% of those without chylothorax.[29] Similarly, the correction of Tetralogy of Fallot has been associated with postoperative chylothorax.[30] Other CHDs identified in our patients included transposition of the great arteries (10.5%), double-outlet right ventricle (10.5%), and anomalous pulmonary venous return (10.5%). Multiple studies have linked these anomalies with an increased likelihood of developing post-operative chylothorax.[31–33]

Most patients presented with dyspnoea, coughing, and respiratory distress, with dullness on percussion during physical examination, indicative of fluid accumulation in the chest cavity. All patients had chest X-rays and/or CT-scans before the thoracocentesis to assess the characteristics of the fluid, including density and location. Diagnosis of chylothorax depends on several criteria, with the triglyceride level being a key determinant. A pleural fluid triglyceride level above 110 mg/dL (1.24 mmol/L) is highly suggestive of chylothorax, particularly when combined with other criteria. Many studies, including one by Rehman and Sivakumar, suggest that a pleural fluid cholesterol level below 200 mg/dL (5.18 mmol/L) is highly indicative of chylothorax.[15] Our study satisfied both of those criteria, where the pleural fluid's median triglyceride level was 667 mg/dL, and its median cholesterol level was 77 mg/dL, as can be seen in table 2. Additionally, fluid chylomicrons were observed in 6 patients, a crucial marker for confirming chylothorax, as demonstrated by lipoprotein electrophoresis.[6]

The majority of thoracostomy tubes post-chylothorax were positioned on the left side of the chest for 63.2% of patients, likely reflecting the anatomical location of the thoracic duct. Injuries above the fifth thoracic vertebrae typically result in left-sided

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Table 2. Clinical characteristics and management outcomes

	Patients with chylothorax (N = 19)
Cholesterol (mg/dl) median (IQR)	77 (42.5, 97)
Triglycerides (mg/dl) median (IQR)	667 (312, 1294)
Chylomicrons detected n (%)	6 (31.6)
Median number of days with chest tube (IQR)	10.5 (8, 16.5)
Location of chest tube n (%)	
Left side of chest	12 (63.2)
Right side of chest	6 (31.6)
Pericardium	1 (5.2)
Median chylous drainage in mL/Kg/24h (IQR)	10 (8.2, 11.1)
Median length of hospital stay (days) (IQR)	16 (12.3, 23)
Recurrent chylothorax n (%)	3 (16)

chylothorax, while those below it can lead to bilateral or right-sided chylothorax.[34] Notably, none of our patients presented with bilateral chylothorax. The median duration of chest tubes placement post-chylothorax was 10.5 days, with a median chylous drainage of 60 mL, which can be classified as low-output drainage.[35] Due to chylothorax, patients had a median hospital stay of 16 days, knowing that chylothorax developed after a median of 19 days post-surgery. Knowing that all patients underwent echocardiography testing prior to discharge, this discrepancy highlights the importance of additional testing such as chest radiography post-CHD surgery prior to discharge and when there is an index of suspicion. Despite these findings, no clear trend was observed correlating these statistics with other parameters such as age, gender, heart disease, and surgery correction.

To promote healing of the underlying injury and decrease chyle production, modifying the diet is recommended as an alternative to invasive treatments. Suggestions for patients with chylothorax may include lipid restriction or temporarily cessation of oral intake.[36] All our patients were enterally fed at the time of diagnosis. At our institution, all 19 patients were managed with chest tube drainage. Unlike conventional fats, medium-chain triglycerides are absorbed directly into the portal system without chylomicron formation, reducing lymphatic flow along the thoracic duct and minimising chyle output.[37] Two patients received octreotide therapy, and one received standard steroid treatment. Octreotide, a synthetic somatostatin analog, has been shown to effectively reduce drainage volume and improve outcomes in postoperative chylothorax cases.[38] Despite the success of conservative and medical approaches in our cases, surgical intervention remains an option when conservative methods are inadequate or direct intervention is necessary. Two notable surgical techniques include thoracic duct ligation, which involves tying the thoracic duct to prevent chyle from entering the pleural space, and thoracic duct embolisation, which entails lymphangiography followed by catheterisation of the cisterna chyli.[39,40] Methylene blue can be employed in surgical procedures to highlight chyle leaks to the surgeon, as the fluid can be difficult to notice if left undyed.[41] However, it is important to note that thoracic duct ligation in patients with venous congestion, such as after a Fontan procedure, can resolve chylothorax but may redirect lymphatic fluid, leading to complications like abdominal ascites or protein-losing enteropathy.[42] Elevated venous pressures in these patients exacerbate lymphatic drainage issues, making such complications more likely. However, none of the patients at our institution underwent thoracic duct ligation or alternative surgical procedures given the successful treatment with other conservative means. Follow-up to the patients via phone calls was performed by staff at our centre, with intervals ranging from days to weeks. However, we do recognise that there could be a possibility of underdiagnosis, particularly in cases where venous and lymphatic pressures evolve over time.

This study was limited by its retrospective design and the relatively small cohort size. In addition, individual patient medical conditions influenced some aspects of their management plans. At our centre, we do not usually test for chylothorax unless the chest tube drainage exhibits characteristics suspicious of chyle. This could underestimate the true prevalence of chylothorax in neonates specifically and in all patients who remain intubated and fasting for a long period after the surgery. Nevertheless, this research study stands as one of the few conducted in the region, and it highlights the need for standardised guidelines pertaining to the diagnosis and treatment of postoperative chylothorax, while acknowledging the uniqueness of every patient's situation.

Conclusion

In conclusion, our retrospective study provides insights into the epidemiology, clinical characteristics, and management outcomes of 19 patients with postoperative chylothorax. Accurate diagnosis, coupled with an understanding of clinical aetiology, is paramount. While our study addresses these aspects, further research with larger, multicentre cohorts is warranted to delve deeper into the associations of postoperative chylothorax. Such studies can inform the development of treatment plans that follow international guidelines and are tailored to individual paediatric patients.

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Author contributions. *FB and MA conceived the presented idea and the study framework. BS, JAK, and SY performed the data collection. AM performed the data analysis and validation. BS, JAK, SY, CER, RZ, MO, IER, and AM wrote the first draft of the manuscript. BS, JAK, and IER constructed the figures. MO, IER, FB, and MA supervised the project and* did the final editing. All authors contributed to corrections and adjustment of subsequent iterations of the manuscript. All authors approve and agree with the content.

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Competing interests. The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Ethical standard. This study was approved by the institutional review board (IRB) at the American University of Beirut. The IRB has waived the need for obtaining consents for this study. All procedures performed in the study involving human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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