Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Postpartum chylothorax: Two case reports and literature review ☆

Zhentao Yan[a], *Junqing Xi*[b], *Jingui Li*[b], *Zhuochao Li*[a], *Xiangjun Han*[a,*], *Zhengqiang Yang*[b,*]

[a] *Department of Interventional Radiology, The First Hospital of China Medical University, Shenyang, Liaoning, China*
[b] *Department of Interventional Therapy, National Cancer Center/National Clinical Research Center for Cancer/Cancer Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing, China*

ARTICLE INFO

Article history:
Received 17 October 2023
Accepted 24 October 2023

Keywords:
Postpartum chylothorax
Chylothorax
Lymphangiography
Interventional therapy
Case report

ABSTRACT

Postpartum chylothorax is an infrequent complication of delivery that is sometimes overlooked. We presented 2 cases of chylothorax in primiparous women who developed chest tightness and breath shortness after vaginal birth, probably due to increased pressure in the thoracic ducts during labor. Lymphography with iodine oil revealed leakage at the T4 level of the thoracic duct in 1 patient but not in the other. Only trace amounts of iodized oil were deposited in the thoracic cavity. There was a significant decrease in postoperative drainage. However, the treatment did not yield the anticipated curative effect in either case. Eight incidences of postpartum chylothorax were identified in the reviewed literature. Patients with refractory chylothorax may benefit greatly from lymphography since it can detect structural changes and determine whether there is a leaking in the thoracic duct. Lymphography-guided therapy for chylothorax with a verified leak has the potential to be both effective and safe. Lymphangiography can serve as a useful tool in selecting the optimal surgical strategy.

Introduction

Chylothorax is an infrequent medical disorder primarily attributed to thoracic duct damage, which subsequently leads to the buildup of chyle inside the pleural space. This buildup can cause chest tightness, dyspnea, and significant pleural effusion. Diagnosis can be determined by radiographic findings or by performing thoracentesis and obtaining a milky fluid sample with a triglyceride content over 110 mg/dL [1]. Chylothorax arises from either traumatic or nontraumatic causes. Chylothorax resulting from tumors and esophagectomy is frequent, but spontaneous chylothorax in postpartum pregnant women is rarely documented in the literature [2]. The etiology of chyle buildup in the pleural cavity of pregnant individuals may be attributed to many pathophysiological mechanisms.

Abbreviations: LAG, Lymphangiography; CCT, Congenital chylothorax.

☆ Funding Information: Cancer Foundation of China LC2020A02. Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

* Corresponding authors.
E-mail addresses: xjhan@cmu.edu.cn (X. Han), ntdoctoryang@hotmail.com (Z. Yang).

https://doi.org/10.1016/j.radcr.2023.10.057

These mechanisms include Valsalva movements performed during birthing or iatrogenic causes such as external abdominal pressure, which increases intra-abdominal and thoracic lymphatic pressures. Abnormal thoracic duct anatomy is present in approximately 35% of the population. Patients undergoing surgery for thoracic or esophageal cancer are more susceptible to injury, and due to their unique anatomical location, they may also develop nontraumatic celiac disease [3]. The most effective therapy for chylothorax is yet undetermined. Previous studies have shown that thoracic duct ligation has similar efficacy to thoracic duct embolization in the treatment of traumatic and nontraumatic celiac disease [4,5]. However, in cases of refractory celiac disease, the efficacy of these interventions can be greatly reduced, as the exact site of air leakage remains challenging to determine. This study presents 2 primiparous women with postpartum chylothorax treated with lymphography. Additionally, we conducted a comprehensive search of the available literature, which revealed a limited number of reports of eight cases of postpartum celiac disease. Therefore, it is essential to concentrate on the pathophysiological causes and therapeutic strategies of postpartum chylothorax.

Case presentation

Case 1

In January 2023, a 28-year-old primiparous female patient presented with chest tightness and shortness of breath 1 month after delivery. After 6 months, her symptoms worsened. The presence of pleural effusion in the left thoracic cavity and insufficient extension of the left lower lung lobe was seen using a chest X-ray and CT scan (Figs. 1A and D). A thoracentesis was performed under local anesthesia, and a milky fluid was withdrawn. The presence of chyle in the fluid was verified by laboratory testing, leading to the diagnosis of chylothorax. The drainage tube discharged a daily volume ranging from 800 to 1000 milliliters. In August 2023, a lymphangiogram procedure was conducted under the administration of a local anesthetic. The right inguinal lymph node was punctured using a 1ml syringe needle under the supervision of ultrasonography. The imaging (Fig. 1B) confirmed the accurate positioning of the needle into the lymph node. The infusion pump administered iodine oil gradually at 6 milliliters each hour. During the operation, a series of images were captured at 10-minute intervals to enable real-time monitoring of the dynamic movement of iodine oil inside the lymphatic channels. The lymphatic vessels were discernible after the injection of 6 ml of iodized oil. On thoracic fluoroscopy, the thoracic duct was visualized entering the left subclavian vein. Multiple congenital variant branches were found in the thoracic duct, while no apparent fistula was detected. An extra volume of 1 milliliter of iodine oil was administered through injection. However, no evidence of contrast agent overflow was found in the thoracic duct region (Fig. 1C). The results of the lymphangiogram revealed that the patient's chylothorax was a consequence of persistent leakage of lymphatic fluid from the thoracic duct. The precise location of the leak site could not be determined for thoracic duct embolization or ligation. The patient was placed on conservative measures such as fasting, and daily celiac drainage was recorded. During the first 3 days after the operation, the drainage tube discharged around 300 mL of fluid per day. A computed tomography (CT) scan revealed no change in the fluid accumulation in the pleural space, accompanied by observable deposition of iodine oil inside the thoracic cavity (Fig. 1E). Between days 4 and 7, an increase in drainage was observed to approximately 700 mL. Additionally, a CT scan revealed a noteworthy decrease in pleural effusion, accompanied by the re-expansion of the left lung (Fig. 1F). The chylothorax resolved after treatment with retrograde femoral vein embolization at another facility. No cases of recurrence were seen throughout the 3-month follow-up period.

Case 2

In October 2020, a 27-year-old primigravida gave birth to a son by vaginal delivery. After 3 months, she developed symptoms of chest tightness and shortness of breath. The lung CT scan results indicated the presence of a substantial volume of pleural effusion inside the right thoracic cavity. A drainage catheter was appropriately placed into the pleural space, extracting a milky chyle with a daily volume ranging from 500 to 1000 mL. In March 2021, a lymphangiogram revealed the leakage of iodine oil from the thoracic duct at the T4 level after entering the right thoracic cavity (Figs. 2A–C). Following the lymphangiography procedure, there was a noticeable reduction in the drainage fluid amount, ranging from around 100 to 600 mL each day. The pleural effusion exhibited a reduction after the thoracic duct dredging procedure, and no recurrence was seen throughout the 6 months of post-treatment monitoring.

Systematic review

Literature searches were performed in databases including PubMed, Web of Science, and China National Knowledge Infrastructure (CNKI), using the following keywords: "postpartum," "after childbirth," "pregnant women," "chyle," and "chylothorax." Additionally, a comprehensive examination of all references cited in the literature was also conducted. Among the original 80 papers, 71 were eliminated after deduplication and abstract evaluation. The majority of these deleted studies consisted of case reports and reviews on neonatal chylothorax, chylothorax during pregnancy, and chylous ascites. Subsequently, a total of 9 studies were acquired and comprehensively examined. One of the study papers was not accessible for download, and another pertains to the topic of pregnancy, and the last is a literature review. The previously mentioned were excluded. Additionally, a supplementary search was performed on the references cited in the selected literature, including 2 studies that satisfied the predetermined criteria for inclusion. Eventually, eight studies were included in the research [6–13]. Figure 3 is a flow chart that visually represents the selected research materials. Table 1 provides a summary of the critical features of the studies included in the analysis.

Table 1 – The literature review of postpartum chylothorax.

Study	Year	Design	n	Age	Reason	Time	Diagnostic	Chylous fluid	Effusion volume	Location	Treament	Complications
Tornling [6]	1987	Case report	1	23	High pressures	2 wk	X rays	TAG:1807 mg/d Protein:8.5 g/dL LDH:80 IU/L	1000-1100 mL/d	Left thoracic cavity	Thoracic duct ligationb	Recovery
Burlew [7]	1991	Case report	1	34	LAM	8 mo	X-rays	TAG:1262	Total:2000 mL	Left thoracic cavity	Progesterone	Recovery
Cammarata [8]	1991	Case report	1	20	Valsalva; High pressures	Childbirth	X rays	TAG:2730mg/dL Protein:3.8 g/dL LDH:197IU/L	Total:3000 mL	Right thoracic cavity	Thoracic duct ligation	Recovery
Bai [9]	1997	Case report	1	25	Malformation of thoracic duct	10 d	Symptom	NA	800 mL/d	Left thoracic cavity Right abdomen	Thoracic duct ligation	Recovery
Antonio [10]	2006	Case report	1	28	High pressures	2 wk	X rays	TAG:863 mg/d	Total:7000 mL	Right thoracic cavity	Talc pleurodesis	Recovery
Momose [11]	2008	Case report	1	24	High pressures	2 mo	SPECT	TAG:3135 mg/d Protein:3.6 g/dL LDH:80 IU/L	Total:2200 mL	Right thoracic cavity	Thoracic duct ligation	Recovery
Rahimi-Rad [12]	2008	Case report	1	32	NA	11 d	X rays	TAG:562 mg/d Protein:6.8 g/dL LDH:116 IU/L	NA	Left thoracic cavity	NA	NA
Huang [13]	2017	Case report	1	22	NA	Childbirth	X rays	TAG:118 mg/dL	1500-2000 mL/d	Right thoracic cavity	Thoracic duct ligationb VSD	Recovery

LAM, lymphangiomyomatosis; LDH, lactate dehydrogenase; TAG, triglyceride; VSD, vacuum sealing drainage.

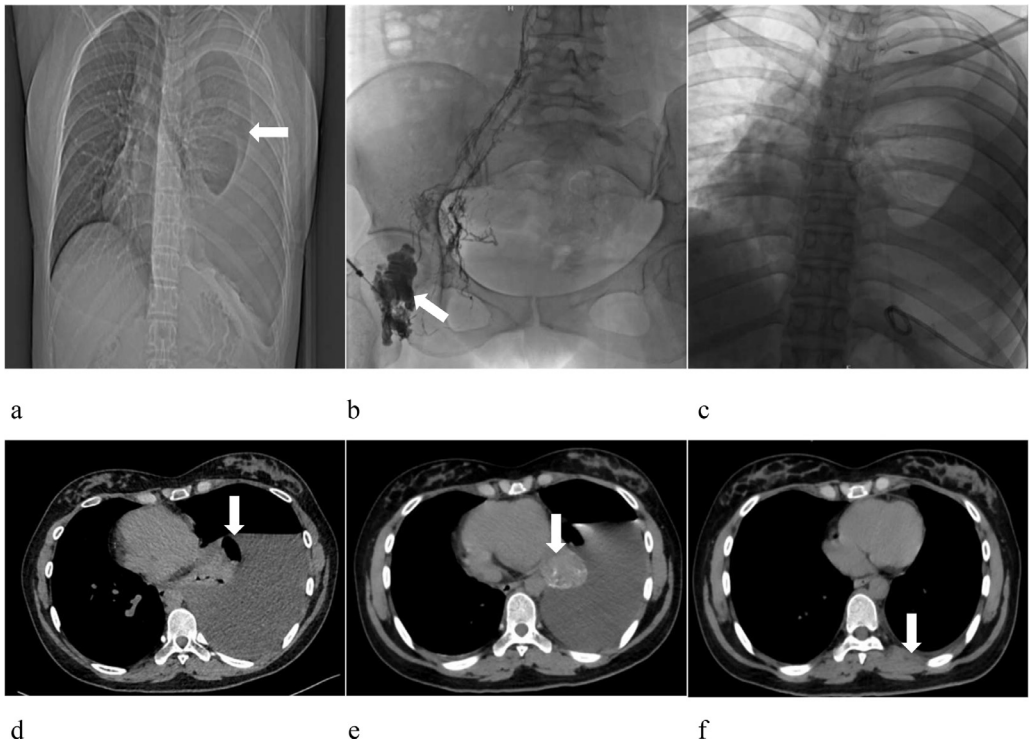

Fig. 1 – (A) Chest X-ray showing significant pleural effusion on the left side. (B) Ultrasound-guided puncture of the right inguinal lymph node. (C) Intraoperative DSA image of the thoracic duct. (D) Chest CT image upon admission. (E) Chest CT image on the first day postoperation. (F) Chest CT image on the seventh day postoperation.

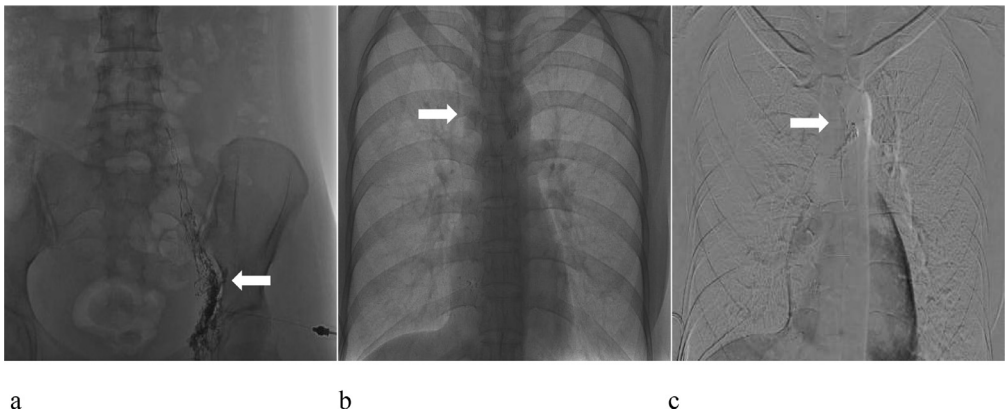

Fig. 2 – (A) Ultrasound-guided puncture of the right inguinal lymph node. (B) Intraoperative DSA image of the thoracic duct. (C) Iodine oil leakage from the thoracic duct at the T4 level.

Within the included investigations, eight individuals were examined, with an average age of 28 and a range of ages from 20 to 32. Postpartum, all patients exhibited the occurrence of spontaneous chylothorax, which was confirmed by identifying certain characteristics on chest X-rays and triglyceride levels. Chylothorax was seen in 4 cases as a result of elevated intrathoracic pressure following vaginal birth [6,8,10,11]. One had a congenital anatomical variant of the thoracic duct [9], and the other experienced pulmonary lymphangioleiomyomatosis [7]. All patients underwent thoracic duct ligation or pleurodesis after failure of conservative therapy. None of the patients underwent surgery such as lymphangiography. All patients who received treatment exhibited a complete recovery within 1 year.

Discussion

Postpartum chylothorax is less common than pregnancy chylothorax, often manifesting 2 weeks after delivery. It is characterized by a sudden and inconspicuous appearance. The oc-

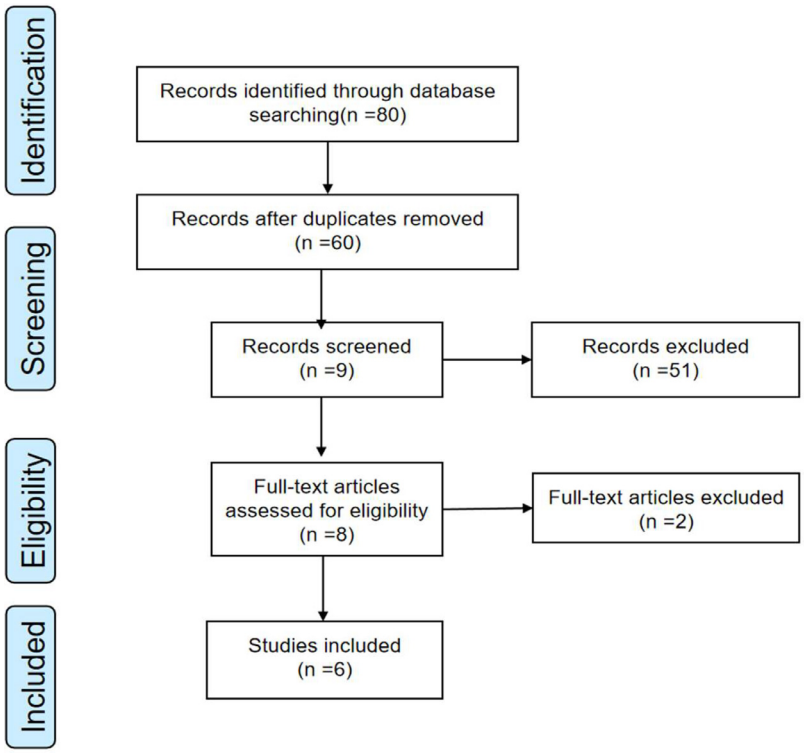

Fig. 3 – Flow diagram of the study selection process.

currence of chylothorax during delivery may potentially be attributed to factors such as extreme physical activity during labor, Valsalva procedures, or the application of external pressure on the abdomen. These actions might result in a notable elevation of lymphatic pressure, thereby contributing to the development of chylothorax. Compression, stretching, or damage to the thoracic duct may lead to the exposure of lymph fluid and the development of chylothorax, resulting in fluid loss and impairment of cell-mediated immunity [6,8,10–12]. Moreover, Bai et al. [9] documented a case study of spontaneous postpartum celiac disease in which contralateral celiac disease and celiac ascites appeared in the right lower abdomen after ligating a thoracic duct. This observation implies the existence of significant interconnections between the lymphatic systems of the thoracic and abdominal regions. The existing literature provides evidence to support the notion that chylothorax resulting from cirrhosis may occur when chylous ascites migrate to the pleural cavity [14,15]. The case documented by Bai has similarities with our first report of a patient with chylothorax, as both had congenital structural anomalies of the thoracic duct. The limited efficacy of surgical and interventional treatments and the marked tendency to recurrence in the treatment of this intractable disease has led to the term refractory celiac disease. The prevalence of chylothorax patients exhibiting symptoms on the left side is mainly attributed to the anatomical peculiarities of the thoracic duct. There was no difference in the site of onset between the summarized cases and the reported cases. The diagnosis of chylothorax was performed by Momose et al. [11] with the use of SPECT/CT lymphatic imaging. Technetium-99m-HSA-DTPA was injected subcutaneously into the dorsum of the foot. This methodology presents a novel strategy for observing of the lymphatic pathway in patients with chylothorax who exhibit anatomical changes in the thoracic duct or to verify the location of leaks.

Furthermore, in conjunction with the aforementioned pathophysiological pathways, many postpartum conditions have the potential to induce chylothorax. A woman presented 8 months postpartum with respiratory distress and celiac disease (a condition characterized by the accumulation of celiac in the pleural cavity). This occurrence was attributed to pulmonary lymphangioleiomyomatosis, a rare lung disease, which may be linked to increased levels of estrogen during pregnancy [7]. Another woman developed pancreatitis and celiac disease within a week of delivery. The patient had completed a cesarean section without experiencing considerable physical strain, Valsalva maneuvers, or trauma during the birth process. This case may be attributed to the inflammation of the thoracic lymphatic system resulting from pancreatitis, which subsequently obstructed lymph circulation, ultimately resulting in the development of chylothorax [16]. The pathophysiologic mechanisms of postpartum celiac disease are closely related to those of celiac disease after thoracic surgery or during pregnancy, so it is clinically relevant to study the pathophysiologic mechanisms of postpartum celiac disease.

Additionally, our findings indicate that the administration of contrast agents, including iodine oil during lymphangiography, has the potential to reduce symptoms in patients and sustain their recovery for a significant duration. The precise

mechanism behind reducing chyle secretion by iodine oil remains uncertain. There are many factors that may contribute to the observed phenomenon, one of which is the persistent administration of contrast chemicals under high pressure. This continuous injection could lead to the occlusion of lymphatic reflux, decreasing the lymph flow. Furthermore, it is worth noting that iodine oil has the potential to obstruct lymphatic leak spots [17]. In their study, Constantin Cope et al. [18] documented the effective management of 29 individuals diagnosed with chylothorax using percutaneous lymphangiography and thoracic duct embolization. This groundbreaking interventional approach represents a significant advancement in the treatment of chyle leakage. Lymphangiography is a novel interventional radiological technique used in the nonvascular domain, which serves to ascertain the precise site of chyle leakage and afterward address the leakage by means of sealing. Nevertheless, the efficacy of this approach is contingent upon the specific anatomical site of the lymphatic lesion and the individual's general health status [19]. This groundbreaking interventional approach represents a significant advancement in the treatment of chyle leakage. It is anticipated that lymphangiography will increasingly be considered the primary option after surgical operations. These patients are more likely to seek interventional therapies for financial and aesthetic reasons. These patients, driven by economic and aesthetic considerations, are more likely to seek interventional therapies. Nevertheless, it is crucial to acknowledge that lymphangiography has limited efficacy in the management of refractory chylothorax resulting from congenital structural anomalies of the thoracic duct and lymphatic fluid leakage [20]. The symptoms of these 2 patients were only mitigated after lymphangiography without attaining a complete resolution. However, it is valuable to learn about this treatment approach's use in patients with postpartum chylothorax, and the utilization of lymphangiography pictures might provide insights for future therapeutic interventions.

The optimal therapeutic approach for individuals with postpartum chylothorax remains uncertain. In most cases, first attempts are made to use conservative treatment methods. Strict parenteral nutritional therapy may be required for high-volume celiac disease patients who do not respond to enteral dietary therapy. Following an extended period of conventional therapy, some patients can attain a level of leak closure that is deemed adequate [21,22]. The timing and reasons for surgical intervention still need to be standardized. It should be predicated upon considerations such as the kind and location of the chyle leak, the clinical presentation of the patient, and several other pertinent aspects [23]. According to Wasmuth-Pietzuch et al., surgical intervention is recommended for chyle leaks that persist for 5-7 days with a volume above 500 mL per day or for leaks that continue for 2 weeks [24]. Therapeutic thoracentesis and negative pressure sealed drainage have been shown to relieve respiratory distress and reach a specified cure rate temporarily. However, it is essential to note that these procedures have the potential risk of complications such as starvation, electrolyte imbalance, and infection [25]. It is advisable to use multidisciplinary methodologies while taking into account the patient's tolerance and desire for surgical intervention.

Conclusion

In essence, the main limitation of this systematic review is that only case reports were included and the patient sample size was relatively limited. Nevertheless, conducting large-scale retrospective investigations on postpartum chylothorax poses significant difficulties due to its infrequent occurrence. Based on our findings, it can be inferred that lymphangiography offers a distinct benefit in detecting leaks and anatomical deviations of the thoracic duct in patients with postpartum refractory chylothorax. This information may serve as a valuable resource for selecting appropriate surgical procedures. In the 2 cases we reported, patients with postpartum spontaneous chylothorax mostly received interventional therapy using lymphangiography. Additional research is necessary to validate the extent and dependability of its clinical use. Furthermore, further investigation is required to elucidate the underlying pathophysiological processes of postpartum spontaneous chylothorax.

Patient consent

The patient and the patient's family have signed an informed consent for the publication of their cases for clinical research.

REFERENCE

[1] McGrath EE, Blades Z, Anderson PB. Chylothorax: aetiology, diagnosis and therapeutic options. Respir Med 2010;104(1):1–8.
[2] Totadri S, Trehan A, Bhattacharya A, Bansal D, Attri SV, Srinivasan R. Chylothorax in children with cancer: a milky predicament. Indian J Cancer 2017;54(4):691–4.
[3] Nair SK, Petko M, Hayward MP. Aetiology and management of chylothorax in adults. Eur J Cardiothorac Surg 2007;32(2):362–9.
[4] Kim PH, Tsauo J, Shin JH. Lymphatic interventions for chylothorax: a systematic review and meta-analysis. J Vasc Interv Radiol 2018;29(2):194–202 e4.
[5] Brinkmann S, Schroeder W, Junggeburth K, Gutschow CA, Bludau M, Hoelscher AH, et al. Incidence and management of chylothorax after Ivor Lewis esophagectomy for cancer of the esophagus. J Thorac Cardiovasc Surg 2016;151(5):1398–404.
[6] Tornling G, Axelsson G, Peterffy A. Chylothorax as a complication after delivery. Acta Obstet Gynecol Scand 1987;66(4):381–2.
[7] Burlew BP, Shames JM. Lymphangiomyomatosis: hormonal implications in etiology and therapy. South Med J 1991;84(10):1247–9.
[8] Cammarata SK, Brush RE, Hyzy RC. Chylothorax after childbirth. Chest 1991;99(6):1539–40.
[9] Shutang B, Zhengguo Li, Liping G. A case of spontaneous chylothorax and chylous ascites. Chin J Surg 1997;34(07):417 [in Chinese].
[10] Honguero Martínez AF, Arnau Obrer A, Pérez Alonso D, Estors Guerrero M, Cortés Alcaide CM, Cantó Armengod A. Bilateral chylothorax after delivery: an infrequent case treated with videothoracoscopic talc pleurodesis]. Cir Esp 2006;80(6):400–2.

[11] Momose M, Kawakami S, Koizumi T, Yoshida K, Kanda S, Kondo R, et al. Lymphoscintigraphy using technetium-99m HSA-DTPA with SPECT/CT in chylothorax after childbirth. Radiat Med 2008;26(8):508–11.
[12] Rahimi-Rad MH. Chylothorax after childbirth in a mother. Indian J Med Sci 2008;62(1):19–20.
[13] Xiu-rong H, Xiao-hong L, Li-cai D. Vacuum sealing drainage for spontaneous chylothorax during puerperium: a case report. Chin J Clin New Med 2017;10(03):272–3.
[14] Romero S, Martín C, Hernandez L, Verdu J, Trigo C, Perez-Mateo M, et al. Chylothorax in cirrhosis of the liver: analysis of its frequency and clinical characteristics. Chest 1998;114(1):154–9.
[15] Tsauo J, Shin JH, Han K, Yoon HK, Ko GY, Ko HK, et al. Transjugular intrahepatic portosystemic shunt for the treatment of chylothorax and chylous ascites in cirrhosis: a case report and systematic review of the literature. J Vasc Interv Radiol 2016;27(1):112–16.
[16] Smędra A, Barzdo M, Krupińska J, Klemm J, Machała W, Szram S, et al. Chylothorax as a rare complication of acute pancreatitis in a 25-year-old woman after cesarean section. Arch Med Sadowej Kryminol 2015;65(3):182–9.
[17] Matsumoto T, Yamagami T, Kato T, Hirota T, Yoshimatsu R, Masunami T, et al. The effectiveness of lymphangiography as a treatment method for various chyle leakages. Br J Radiol 2009;82(976):286–90.
[18] Cope C, Kaiser LR. Management of unremitting chylothorax by percutaneous embolization and blockage of retroperitoneal lymphatic vessels in 42 patients. J Vasc Interv Radiol 2002;13(11):1139–48.
[19] Juszczyk K, Waugh R, Sandroussi C. Lymphangiography as therapeutic management of chylothorax. J Med Imaging Radiat Oncol 2013;57(4):460–1.
[20] Stecker MS, Fan CM. Lymphangiography for thoracic duct interventions. Tech Vasc Interv Radiol 2016;19(4):277–85.
[21] Agrawal A, Chaddha U, Kaul V, Desai A, Gillaspie E, Maldonado F. Multidisciplinary management of chylothorax. Chest 2022;162(6):1402–12.
[22] Merigliano S, Molena D, Ruol A, Zaninotto G, Cagol M, Scappin S, et al. Chylothorax complicating esophagectomy for cancer: a plea for early thoracic duct ligation. J Thorac Cardiovasc Surg 2000;119(3):453–7.
[23] Ur Rehman K, Sivakumar P. Non-traumatic chylothorax: diagnostic and therapeutic strategies. Breathe (Sheff) 2022;18(2):210163.
[24] Martucci N, Tracey M, Rocco G. Postoperative Chylothorax. Thorac Surg Clin 2015;25(4):523–8.
[25] Wasmuth-Pietzuch A, Hansmann M, Bartmann P, Heep A. Congenital chylothorax: lymphopenia and high risk of neonatal infections. Acta Paediatr 2004;93(2):220–4.