

Chylothorax in a pediatric patient secondary to stab wound

Quilotórax secundario a herida por arma blanca

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What do we know about the subject matter of this study?

Chylothorax is the accumulation of chyle in the pleural space, secondary to thoracic duct injury. The most frequent etiology in children is iatrogenic after cardiac surgery. Treatment depends on the chyle output and the patient's clinical condition.

What does this study contribute to what is already known?

We present a case of chylothorax secondary to direct injury of the thoracic duct after a knife wound. Extrathoracic traumatic etiology is uncommon, especially in children. The case's uniqueness is further compounded by the unusual wound location, which made surgical treatment (pleurodesis, thoracoscopy) unlikely to be effective. This presented a therapeutic challenge due to the high chyle output, yet resolution was achieved with conservative management.

Abstract

Thoracic duct injury is usually the consequence of direct trauma following thoracic surgery, with external trauma mechanisms being exceptional in the pediatric population. Massive chylothorax requires pleural drainage and, when it presents clinical decompensation, it may require surgical repair of the thoracic duct injury as well. **Objective:** To report a case of chylothorax of uncommon cause in pediatrics, and to describe how efficient but conservative management allowed complete recovery. **Clinical Case:** A 10-year-old male patient, without previous clinical history, suffered from a stab wound to the left supra-clavicular region. Three days after the incident, due to respiratory distress, he was diagnosed with left massive pleural effusion which required tube thoracostomy drainage. The milky appearance and the presence of chylomicrons in the drained fluid oriented the diagnosis towards a chylothorax. The patient was admitted to the pediatric intensive care unit and received treatment with intravenous (IV) octreotide and a low long-chain triglyceride diet, supplemented with medium-chain triglyceride oil. Despite the initial improvement, a high chyle output persisted,

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requiring immunoglobulins and albumin replacement. After optimizing conservative management with fasting, parenteral nutrition for 15 days, octreotide at the maximum dose, and respiratory physiotherapy, a complete recovery was achieved, preventing the need for surgical intervention. **Conclusion:** Chylothorax caused by penetrating trauma is a rare condition in the pediatric population that requires customized management. Surgical repair of the thoracic duct is an aggressive surgery and currently, there is limited expertise for the pediatric patient. Adequate conservative management may result in complete recovery, as it was described in this case report.

Introduction

Chylothorax is the accumulation of chyle in the pleural space secondary to altered lymph flow through the thoracic duct¹. The origin of chylothorax can be traumatic, congenital, neoplastic, infectious, or idiopathic. Within the traumatic origin, it is usually caused by an iatrogenic lesion of the thoracic duct during intrathoracic surgery, accounting for 49.8% of the total and being the most frequent overall cause². In contrast, chylothorax due to external trauma or penetrating injury is a rare phenomenon, being extremely rare in the pediatric age group.

A delay in diagnosis or treatment can lead to malnutrition, due to the lack of fat absorption and protein loss, immunosuppression with risk of sepsis, due to hypogammaglobulinemia and lymphopenia, as well as respiratory complications, and even death³.

There are basically two management options: conservative and surgical. Conservative treatment is aimed at reducing chyle flow with a low-fat diet supplemented with medium-chain triglycerides since they are absorbed directly at the portal level without accessing the lymphatic system. Somatostatin or its long-acting analog (octreotide) reduces the pressure of the lymphatic system by reducing chyle production so they can be used as an adjuvant⁴. Surgical management is an option when chylothorax persists for > 2-4 weeks with an output > 1500 mL/day or when nutritional or metabolic complications occur^{5,6}.

The objective of this article is to report a case of chylothorax of uncommon etiology in pediatrics and to describe how efficient conservative management allowed complete recovery.

Clinical Case

A 10-year-old male patient with no history of interest attended the emergency department of his regional hospital immediately after an accident with a 20-cm kitchen knife in the left supraclavicular region. At the time of consultation, the physical examination was normal except for mild respiratory distress. Pul-

monary auscultation showed good bilateral air entry with no pathological sounds and oxygen saturation by pulse oximetry > 95%. The rest of the vital signs were stable; he was afebrile (36.5°C), with 25 breaths per minute (bpm), 120 beats per minute (bpm), and blood pressure 110/60 mm of mercury. On examination, an incised open wound, apparently superficial, 3-4 cm in diameter with no active bleeding was observed in the suprasternal region. A chest X-ray showed a left pneumothorax, so the study was completed with a chest CT scan (Figure 1A,1B,1C,1D) which confirmed the pneumothorax and subcutaneous emphysema, and given their small size, a watchful waiting approach was adopted. After suturing the wound, the patient was admitted to the pediatric ward for 48 hours, showing clinical improvement, therefore he was discharged.

After 24 hours, the patient consulted again in the emergency room of the same center due to dyspnea, orthopnea, and pleuritic chest pain on the left side. Pulmonary auscultation revealed hypophonia in the left hemithorax. The rest of the physical examination showed no pathological findings. Vital signs showed tachycardia at 140 bpm, tachypnea at 35 bpm, and desaturation of up to 88%. Chest X-ray showed massive left-sided pleural effusion with mediastinal shift (Figure 2).

A 12 French multi-perforated chest tube was inserted in the 5th intercostal space, draining 225 mL of milk-like fluid, with improvement in respiratory symptoms. The chest tube was connected to a continuous suction system with a water seal chamber. The drained pleural fluid had a pH of 7.44, cholesterol of 98 mg/dL, triglycerides of 2321 mg/dL, and the presence of chylomicrons confirming the diagnosis of chylothorax. He was transferred to the referral pediatric intensive care unit (PICU).

The patient was placed on fasting with total parenteral nutrition (TPN) and octreotide was started with a continuous infusion at 3 mcg/kg/h. The chyle output during the first few days was around 2 liters per day (65 ml/kg/day). On the third day, a low-fat diet was gradually introduced with a supplement of medium-chain triglycerides and fat-soluble vitamins. After initial improvement, the chyle drainage remained around 30

mL/kg/day without decreasing over the following 10 days of hospitalization. Laboratory tests showed moderate lymphopenia of $0.42 \times 10^9/L$ (normal value (NV): $1.5-6.7 \times 10^9/L$) and a decrease in IgG with 383 mg/dL (NV: 600-1230 mg/dL); the other immunoglobulins were within normal values: IgA 90 mg/dL (NV: 30-200 mg/dL), IgM 89mg/dL (NV: 50-200 mg/dL), and IgE 38 IU/mL (NV: < 200 IU/mL), therefore, intravenous non-specific human immunoglobulin (0.6 g/kg) was administered with subsequent stabilization, without requiring new doses. He required albumin replacement due to hypoalbuminemia of up to 2.3 g/dL (NV: 4.1-4.8 g/dL). Regarding lipid metabolism, triglycerides were in the low normal range with 58 mg/dL (NV: 44-197 mg/dL).

Due to the high chyle output (> 30mL/kg/day) without improvement and the metabolic-nutritional consequences after 15 days of conservative management, an interventional approach was considered together with pediatric surgery, evaluating options for ligation or embolization of the thoracic duct and pleurodesis. However, the location of the injury made successful surgical repair challenging with potentially serious complications, so it was decided to optimize conservative management. Oral intake was suspended again, TPN was resumed, and octreotide infusion was increased to a maximum of 15mcg/kg/h. As ventilatory support, humidified nasal CPAP was maintained and respiratory physiotherapy was intensified.

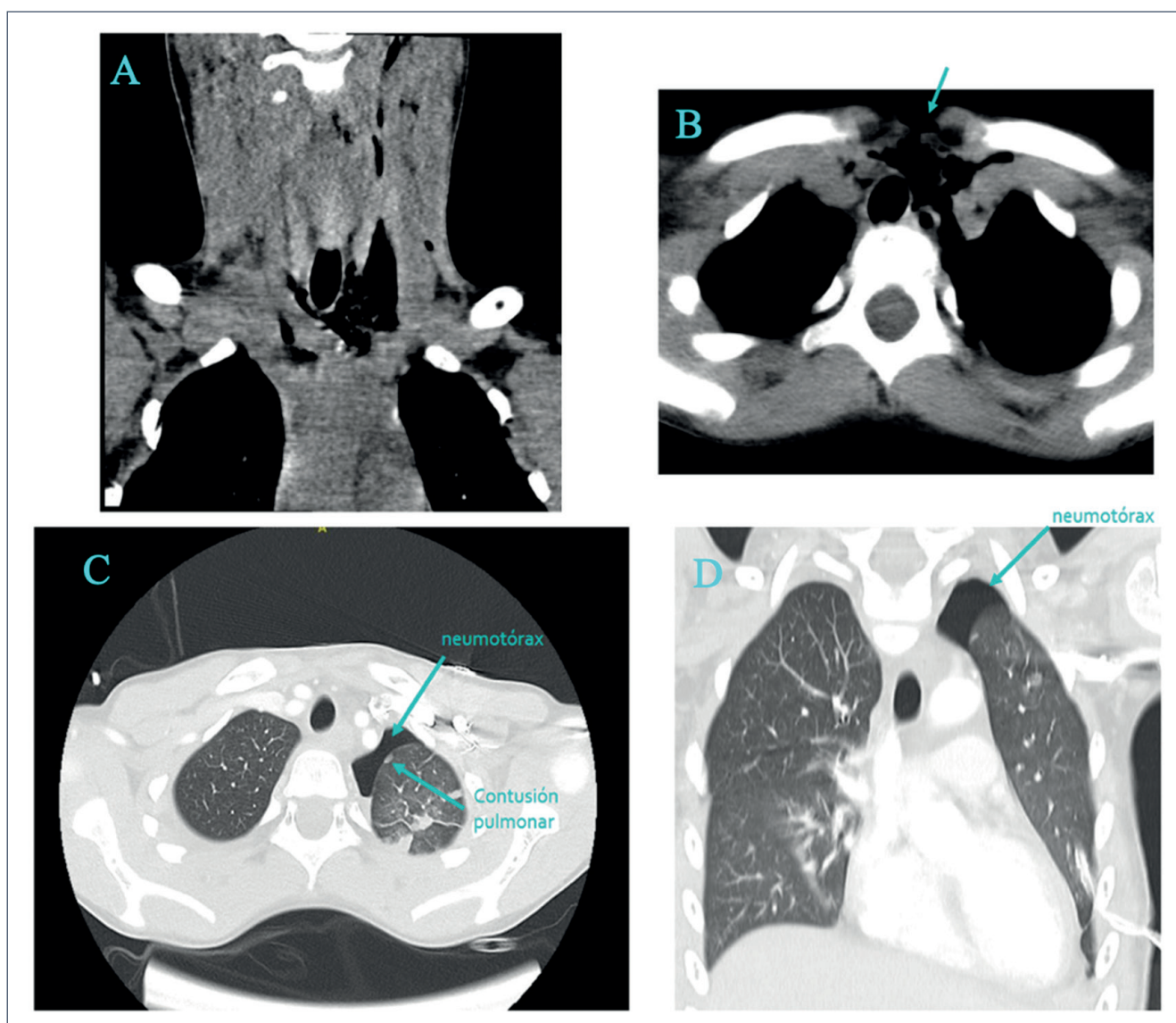


Figure 1. Computed axial tomography performed at the first medical visit. **A:** Sagittal projection, mediastinal window. Emphysematous bullae in the left cervicothoracic region. **B:** Axial projection, mediastinal window. Emphysematous bullae in the left cervicothoracic region. **C:** Axial projection, lung window. Left apical anterior pneumothorax. Pulmonary contusion is observed. **D:** Sagittal projection, lung window. Left apical anterior pneumothorax.

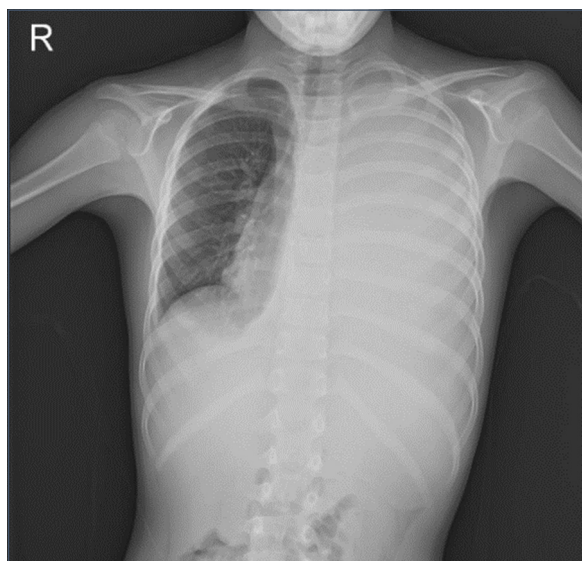


Figure 2. Chest X-ray performed at the second consultation, showing massive left pleural effusion with mediastinal shift.

After 24-48 hours, the output rate decreased. The patient remained for a total of 17 days on a fasting regimen. Enteral nutrition was restarted with a polymeric formula based on whey protein, low in long-chain triglycerides, and rich in medium-chain triglycerides, without recurrence of chylothorax. After 37 days of admission, the pleural drainage tube was removed after draining up to 25 liters of chyle during his stay in the PICU (Figure 3). After discharge, he was followed up for 3 months, remaining asymptomatic and without sequelae in pulmonary function, evaluated by spirometry, plethysmography, and diffusion.

Discussion

We present a clinical case of a pediatric patient with chylothorax caused by a stab wound, an extremely infrequent scenario, and, in the literature reviewed, we only identified 6 published cases in adult patients in the last 20 years^{2-5,7-8}. Stab wounds are usually seen in adults, particularly young males in the context of an assault. In children, they are infrequent⁹⁻¹⁴ and may be, as in our case, the result of accidents. Not only because the mechanism of injury is infrequent, but also because of the anatomical variability of the thoracic duct, chylothorax requires a high level of suspicion in the presence of pneumothorax or hemothorax. Therefore, after this type of trauma, it is suggested to perform a chest X-ray before discharge and schedule the patient later for follow-up. In fact, especially in children, observation in intensive care and periodic monitoring with imaging tests would be indicated in most cases. Given

its low incidence, pediatricians and pediatric surgeons tend to have less experience in its management, making it difficult to standardize measures.

According to the review by Heffner JE¹⁵, conservative management would be chosen in low output chylothorax, considering as such < 1000mL/day (in children < 100 mL/age/day or < 10 mL/kg/day for 5 days)¹⁶⁻¹⁸. In the series by Santos LLD. et al, with 497 patients with lymphatic duct injury after cardiac surgery, they propose a more aggressive management from diagnosis since conservative management has greater morbidity and admission time¹⁹. On the other hand, the review by Kakamad FH. et al.²⁰ included 30 articles on chylothorax secondary to extrathoracic trauma, with a total of 39 adult patients, a mean age of 35.8 years, and was decided conservative management which was successful in 71.4% of these cases, so they concluded that surgery would be indicated after failure of conservative management. The duration of non-surgical treatment before resorting to invasive measures is controversial but, in general terms, it has been established at about 15 days^{15,16,21}.

Conservative treatment aims to reduce the passage of chyle through the thoracic duct and achieve pulmonary re-expansion²² and is usually based on reducing enteral intake and administering drugs that reduce intestinal chyle production, such as octreotide. The most effective first-level measure is to reduce lymphatic flow through the thoracic duct, thus facilitating its healing. This can be achieved by maintaining a fasting regimen with TPN or a low-fat diet supplemented with medium-chain triglycerides²³, which reduces comorbidity associated with prolonged TPN, such as cholestasis, dyslipidemia, catheter-associated infections, psychological disturbance, and malnutrition. As for pharmacological treatment, octreotide has shown effectiveness in patients with very high outputs despite dietary treatment²⁴. However, there is no consensus on the timing of treatment initiation, dosage, or duration of therapy. It is usually proposed to start at 1-4 mcg/kg/h with a progressive increase if the patient does not present side effects such as hyperglycemia, hypotension, and/or abdominal distension²². Some studies suggest starting early treatment with higher doses (3-4 mcg/kg/h)^{5,7}.

As for invasive management, there are no universally accepted indications due to the multiple factors that intervene in the response to conservative treatment, such as age, etiology, symptoms, functional status, and chyle accumulation rate²⁵. It is suggested surgical or interventional repair when the output is > 40 mL/kg/day²⁶ or conservative treatment if it fails after 15 days⁶. This surgical repair consists of ligating the lymphatic duct by thoracoscopy or open surgery and may be associated with pleurodesis in patients with high-volume chyle leakage¹⁶. Despite being a de-

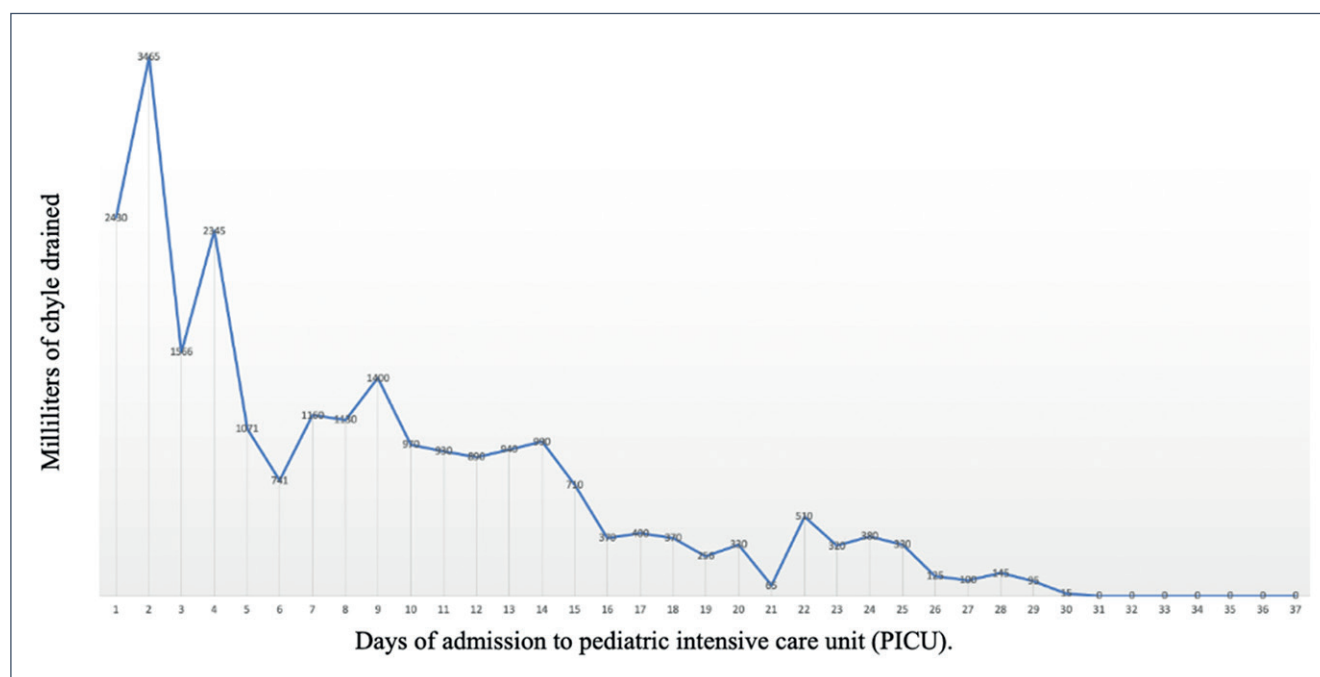


Figure 3. Evolution of the milliliters of chyle drained through the chest tube during the days of admission to pediatric intensive care (PICU).

finitive treatment, it may present short-term complications such as prolonged mechanical ventilation, air leak, or atrial fibrillation^{27,28}, as well as the inherent risk of any thoracic surgery due to injury to neighboring structures, especially the subclavian and internal jugular veins or contralateral chylothorax. Long-term complications such as diarrhea, lower limb and abdominal edema, or malnutrition have also been described^{27,28}.

As a less aggressive alternative, there is the possibility of embolizing the thoracic duct by interventional radiology. It is considered safe, effective, and with a high success rate, although variable according to studies, from 71% in the series of Cope C. and Kaiser LR.²⁹, 73.5% reported by Itkin M. et al.³⁰, and up to 87% by Alexandre-Lafont E. et al.³¹. It should be noted that all these data correspond to an adult population. The procedure consists of identifying the leakage point by lymphography, followed by embolization with a microcatheter. There are antegrade or retrograde variants, with the former being the most commonly used in children, typically via an inguinal or abdominal approach^{32,33}, especially in postoperative chylothorax cases³⁴⁻³⁶. Transvenous retrograde embolization is a less widespread technique; it was described for the first time by Mittleider D. et al.²¹ in 2008 in adult patients. In the pediatric population, only one case has been published in a 5-year-old patient with chylothorax after the Fontan procedure³⁶. The main complication associated with embolization is the appearance of edema in the lower limbs due to

malfunction of the lymphatic duct²⁹. It should be noted that surgical closure or embolization does not mean the immediate end of the output, so it does not necessarily imply a reduction in hospital stay²⁰.

Finally, regardless of the cause or therapeutic approach, in high-output chylothorax, the nutritional status of the patient may be affected, especially in terms of fat and protein absorption. At present, there is no consensus about the nutritional approach of the patient with chylothorax, but we recall that the exclusive consumption of medium-chain triglycerides does not prevent the deficiency of essential fatty acids, which are long-chain fatty acids. In an exempt diet of more than 7 days, a strategy to avoid malnutrition and deficiency of essential fatty acids should be foreseen. It will be essential to periodically check nutritional status during hospitalization, which will mark the nutritional support and corrections of albumin or other deficiencies resulting from nutrient loss through the lymph³⁷.

Conclusion

We describe a pediatric patient with chylothorax secondary to a stab wound section of the thoracic duct. It is an exceptional mechanism that requires personalized and consensual treatment since current guidelines are based on chylothorax secondary to complications after intrathoracic surgery.

Despite the limitations inherent to the description of only one clinical case, we observed that the low-fat diet supplemented with medium-chain triglycerides did not allow the reduction of lymph flow as did the fasting regimen with TPN, along with intensive ventilatory physiotherapy, ventilatory support with humidification, and adequate analgesia. Treatment with octreotide was effective and without side effects in infusion at 15 mcg/kg/h, but further studies would be necessary to know the effective doses of octreotide for the treatment of chylothorax in pediatric patients. In our case, optimal conservative management achieved complete resolution of the condition avoiding the possible risks derived from surgery or embolization by interventional radiology.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

Financial Disclosure

Authors state that no economic support has been associated with the present study.

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