CASE REPORTS

# A UNIQUE SHUNT FOR TREATMENT OF A POSTOPERATIVE CHYLOTHORAX

Rita Costa<sup>1</sup>, Tiago Magalhães<sup>2</sup>, Gustavo Rocha<sup>3</sup>, Pedro Fernandes<sup>1</sup>, Elson Salgueiro<sup>1</sup>

Department of Cardiothoracic Surgery, University Hospital Center of São João, EPE, Porto, Portugal Department of Pediatrics, University Hospital Center of São João, EPE, Porto, Portugal Neonatal Intensive Care Unit, University Hospital Center of São João, EPE, Porto, Portugal

\* Corresponding author: rita2ac@hotmail.com

## **Abstract**

Postoperative chylothorax can be a serious complication. We report on the case of a neonate who had a postoperative chylothorax immediately after esophageal surgery that did not respond to conservative measures or the first two attempts of surgical management of chylothorax. Lastly, a successful pleuroperitoneal shunt was placed and the patient was discharge at 3-months-old. A pleuroperitoneal shunt is usually the last surgical option but may be a curative measure.

Keywords: Chylothorax; Shunt pleuroperitoneal; Pediatric

### INTRODUCTION

Pediatric chylothorax can be either congenital or acquired. Postoperative chylothorax occurs mostly due to trauma to lymphatic vessels. It can be a serious complication since it can promote nutritional deficiencies, prolong the duration of mechanical ventilation, induce electrolyte imbalance, lymphopenia and immunosuppression, increasing the patient vulnerability to infections and mortality rate<sup>1</sup>. We report the usefulness of a pleuroperitoneal shunt (PS) for treatment of refractory chylothorax.

#### **CLINICAL CASE**

A 37+2 weeks gestation male neonate, with prenatal diagnosis of esophageal atresia was delivered by C-section due to breech presentation, to a healthy primigravida. The birth weight was 2035g, and the Apgar scores at the 1st, 5th and 10th minutes of life were 3/7/8, respectively. The fetal genetic evaluation included a karyotype and an array comparative genomic hybridization (aCGH) study that were normal. No other congenital anomalies were detected

On the 2nd day of life (DOL), he was submitted

to a right thoracotomy with sequential anastomosis of the esophageal tops and ligation of a tracheoesophageal fistula. This intervention was complicated with right chylothorax (confirmed by biochemical analysis of pleural effusion). He started conservative treatment for chylothorax (total parenteral nutritionand octreotide perfusion up to 10 mcg/kg/h). Due to respiratory instability, he performed an CT-angiography that showed the presence of a vascular ring with a right aortic arch and a persistent arterial duct. He was referred to our center to be submitted to an aortopexy and closure of the arterial duct via median sternotomy by Cardiothoracic surgery.

After correction of the double aortic arch, chylothorax persisted in spite of medical measures. On the 22ndDOL, he was submitted to a right posterolateral thoracotomy over the 8th intercostal space. Intraoperatively, the thoracic duct was identified near the aortic hiatus and it was oversewn with nonabsorbable suture A mechanical pleurodesis was performed and the chest drain was replaced.

Although an initial reduction in pleural drainage, the patient had a relapse requiring replacement of fluids



with 5% albumin and administration of bicarbonate and immunoglobulins. A lymphangiography was inconclusive. The presence of a thrombus in the superior vena cava was excluded by echo-Doppler.

On the 32ndDOL he was re-submitted to a right thoracotomy over the 5th intercostal space. Intraoperatively, multiples lymphatic pathways in the superior right hemithorax were oversewn with nonabsorbable suture and clips. Since the 36th day of life the infant was fed with an enteric formula with medium chain triglycerides.

Due to the persistence of a chylous drainage (300/400ml/24h) a PS was placed, on the 51st DOL. Since a PS was unavailable, a ventriculoperitoneal shunt with a unidirectional valve (opening pressure of 70 cm H2O) was placed instead. The pressure gradient necessary to open the valve can be overcome by compressing the bulb which houses the one-way valve, forcing the flow from chest to abdomen. The pleural and abdominal parts of the catheter were introduced into the respective cavities under direct viewing, and the shunt was tunneled under the skin with the pumping chamber lodged in a subcutaneous pocket overlying the costal margin (figure 1).

The pleural drainage significantly reduced, but the bulb which houses the one-way valve was pressed several times per hour. The infant started oral feeding seven days after the surgical placement of PS. It was well tolerated and regular abdominal ultrasounds were performed without evidence of ascites.

The pleural drain was removed 20 days after the last surgery. He remained under invasive mechanical venti-

lation until 56thDOL, later under BiPAP support.

The patient was discharged home at 3 months-old without relapse of pleural effusion or ascites. He is currently at home, under milk formula of high energy content, and still maintains the PS.

#### **DISCUSSION**

The treatment of chylothorax should be stepwise, initially with conservative therapies including respiratory support, bowel rest and complete parenteral nutrition or a diet with medium-chain triglycerides. Octreotide and somatostatin have been used to reduce lymph flow. Drainage of pleural effusion is usually required, although a prolonged significant continuous drainage can cause nutritional deficits. The surgical approach presents itself as the next step in the therapeutic algorithm and includes thoracic duct ligation, pleurodesis, pleurectomy and PS¹.

Timing for surgical intervention is variable, with strategies ranging from 2–4 weeks after conservative management, to early intervention depending on whether there is a large-volume chylothorax, severe metabolic and nutritional complications or a well identified site of chyle leak.

Different approaches had been described for thoracic ligation, which is the most definitive treatment if the site of rupture was previously identified by lymphangiography<sup>2</sup>.

Although PS is a less invasive option with equal security and safety compared to other surgical procedures (58 pediatric cases treated with PS were previously reported with a success rate of 79%), it is usually the last surgical option. PS has the advantage of not losing the chyle and

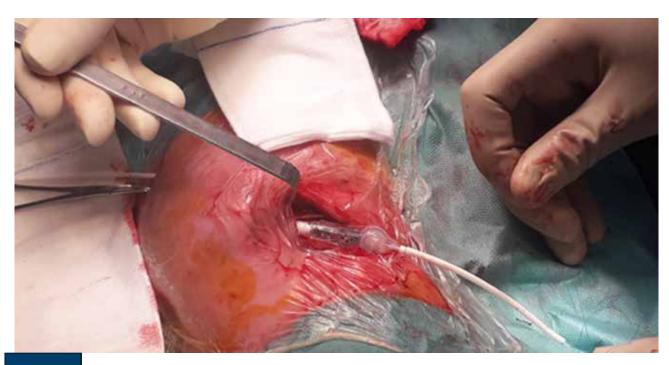


Figure 1

Placement of the pleuroperitoneal shunt



reducing or avoiding malnutrition and immunosuppression. The most frequent complications include shunt closure and ascites<sup>3</sup>. Indications about ideal timing for shunt removal are not clear.

Our PS is still working and timing for removal will be discussed with the multidisciplinary team.

Pediatric postoperative chylothorax can be challenging. PS can be a good solution since it is technically easy to perform, and can allow the reduction of pleural drainage, without loss of proteins and nutrients, while the injured lymphatic vessels heal. PS may also be curative in some patients <sup>1-3</sup>.

#### REFERENCES

- 1. Costa KM, Saxena AK. Surgical chylothorax in neonates: management and outcomes. World J Pediatr. 2018 Apr;14(2):110-115. doi: 10.1007/s12519-018-0134-x.
- 2. Soto-Martinez M, Massie J. Chylothorax: diagnosis and management in children. Paediatr Respir Rev. 2009 Dec;10(4):199-207. doi: 10.1016/j.prrv.2009.06.008.
- 3. Shiraga K, Terui K, Ishihara K, Shibuya K, Saito E et al. Pleuroperitoneal shunt for refractory chylothorax accompanied with a mediastinal lymphangioma: a case report. Ann Thorac Cardiovasc Surg. 2014;20 Suppl:654-8. doi: 10.5761/atcs.cr.12-02244.

