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Thoracic duct–azygos vein anastomosis in an infant with superior vena cava syndrome and recurrent chylothorax

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Abstract

In this study, we describe an intrathoracic microsurgical lymphatico-venous anastomosis as an alternative surgical technique for the treatment of refractory chylothorax in an infant. This procedure allowed us to restore enteral nutrition within days of surgery. At 3-year follow-up, there was no recurrence of pleural effusion.

Keywords: Chylothorax • Surgery • Infant • Venous • Thrombosis

INTRODUCTION

Chylothorax, the most common cause of pleural effusion in neonates, can be caused by increased thoracic duct pressure secondary to superior vena cava (SVC) syndrome and often leads to prolonged hospitalization [1, 2]. In children, conservative treatment (thoracic drainage and parenteral nutrition) is successful in over 80% of patients [1]. Surgical options for refractory chylothoraces include chemical or surgical pleurodesis, ligation or percutaneous embolization of the thoracic duct [1]. Recently, intrathoracic lymphatico-venous anastomosis has been described in adult oncological patients [3] and in children [4]. Herein, we report a successful case of a 9-month-old girl who underwent a lymphatico-azygos anastomosis using microsurgery. This innovative surgery resulted in complete patient recovery.

CASE REPORT

A female neonate with antenatal diagnosis of laparoschisis underwent a three-step surgical repair. Written informed consent was obtained from the parents. By day 12, she developed SVC syndrome and bilateral chylothoraces. By ultrasound, there was no evidence of venous thrombosis at the site of the indwelling catheter. On day 30, repeated Doppler ultrasound showed SVC thrombosis extending into the innominate vein. Neither heparin nor thrombolysis reversed the thrombotic process.

In addition to chest drainage, the patient received parenteral nutrition for 45 days, but this conservative approach failed. Somatostatin was similarly unsuccessful. Over the ensuing months, she required multiple chest tube insertions for recurrent chylothoraces (9 times).

At 9 months, weighing 5 kg, chest angio-computed tomography scan demonstrated the segmental SVC thrombosis, well-developed venous collaterals with a patent dilated azygos system draining in the SVC near its cavoatrial junction. Isotopic lymphography showed collateral circulation at the peritoneal and mediastinal level. There was no drainage via the thoracic duct. Surgical options were thoracic duct ligation, pleurectomy or an attempt of thoracic duct 'rerouting'. After multidisciplinary meetings, we decided to explore the patient with the purpose of performing an intrathoracic lymphatico-venous anastomosis.

Through thoracotomy, the posterior mediastinum was exposed. There was a marked parietal and visceral pleural thickening with 'transpleural' lymphatic oozing.

The azygos vein pressure was 5 mmHg. The azygos vein and the thoracic duct were dissected at the level of T5. Heparin was administered, the duct was ligated and the azygos vein clamped proximally and distally. Following a 5-mm-length venotomy, a microscope-assisted 'end-to-side' anastomosis was performed by 9–0 polypropylene interrupted stitches (Fig. 1). This resulted in distension of the duct with good permeability tests (Video 1). The chest was closed and drained.

The chest tube was removed 1 week after surgery. Within the following 3 months on a normal diet, she corrected her weight's curve slope. At 3-year follow-up, there was no recurrence of pleural effusion.

DISCUSSION

The presence of a central venous indwelling catheter is among the greatest risk factors for central venous system thrombosis in neonates and infants [2].

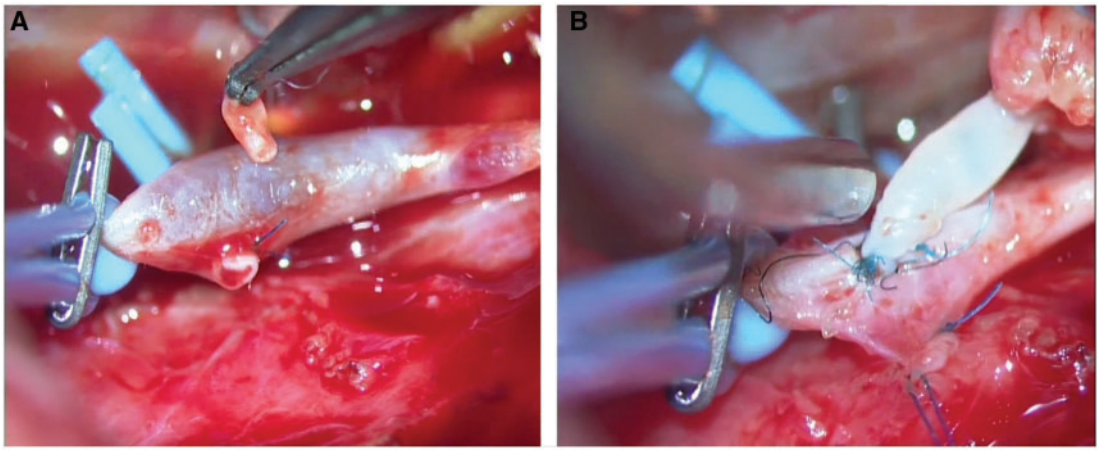
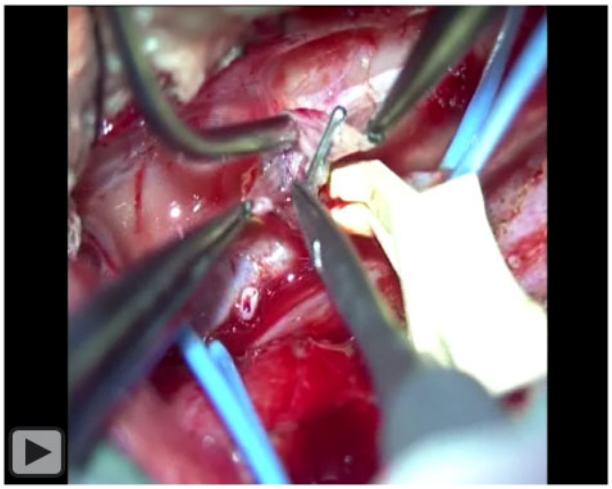


Figure 1: The azygos vein was dissected from T7 to T5, temporarily occluded with a silastic loop, and the thoracic duct was mobilized to allow an end-side anastomosis. Before (**A**) and after completion (**B**) of the lymphatico-venous anastomosis.



Video 1: Lymphatico-venous anastomosis.

In our patient, both therapeutic anticoagulation and thrombolysis were performed without success. After months of repetitive chest tube drainage and hospitalization, a surgical approach needed to be considered. However, no guidelines exist on the type and/or timing of surgical intervention. Besides, lymphography demonstrated the absence of drainage via the thoracic duct reflecting distal obstruction.

Conventional surgical options included (i) ligation of the thoracic duct, (ii) chemical or surgical pleurodesis or (iii) percutaneous embolization of the thoracic duct [1].

In our patient, surgical exploration confirmed diffuse pleural oozing of lymph and hence neither thoracic duct ligation nor pleurectomy was deemed a reasonable option. Therefore, we decided to perform a microsurgically guided intrathoracic lymphatico-venous anastomosis.

Lymphatico-venous anastomosis was initially reported to treat chronic lymphoedema secondary to lymphadenectomy. More recently, the technique was applied to prevent/treat chylothoraces in oesophageal surgical patients [3]. Such a microsurgical repair had never been reported in infants at the time we discussed the available options for our 5-kg infant.

This technique was recently described in 2 paediatric cases [4], with one early failure requiring long-term chest tube drainage. In our patient, this innovative surgical alternative to thoracic duct ligation resulted in complete patient recovery with >3 years of follow-up.

In their meta-analysis on the surgical management of chylothorax, Lindenblatt *et al.* [5] identified only 5 patients with anastomosis between the thoracic duct and the venous system, with an excellent outcome in 4 patients.

CONCLUSION

SVC thrombosis following central venous catheter placement can cause refractory chylothorax. Our case report highlights the potential value of lymphatico-venous anastomosis as a surgical alternative in the case of thoracic duct obstruction even in small infants.

Conflict of interest: none declared.

Reviewer information

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