

Therapeutic Use of Intranodal Lipiodol Lymphangiogram for Chylothorax in Children

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Abstract

Congenital cardiac surgery is the most common cause of chylothorax in children. The majority responds to a staged conservative approach but a few are refractory and requires more aggressive interventions. Lymphangiogram with or without intervention is established for the diagnosis of chyle leaks and there are reports of its additional therapeutic benefits. Its use in children is however limited. We report herewith 3 children with refractory chylothoraces who improved significantly following ultrasound-guided, intranodal lymphangiogram using lipiodol. Our findings suggest that this technique can be therapeutic for children with high flow chylothoraces that fail to respond to conservative therapy.

Keywords: Chylothorax; Children; Cardiac Surgery; Lymphangiogram; Lipiodol; Ultrasound-Guided Lymphangiography; Intranodal Lymphangiography

Introduction

The incidence of chylothorax after congenital cardiac surgery has been reported between 2 - 6.6% [1-3] and carries substantial morbidity and mortality and is associated with a significantly longer length of hospital stay [1]. It is usually managed conservatively with chest tube drainage, dietary restrictions (medium chain triglyceride, parenteral nutrition) and octreotide in a staged protocol [2,3]. Refractory cases would require pleurodesis, thoracic duct embolization and/or thoracic duct ligation. Lipiodol (Guerbet, Villepinte, France) lymphangiography (LAG) was originally used to visualize leakage points [4] or the cisterna chyli during thoracic duct embolization [5]. Its use in children is nevertheless limited. We report herewith the therapeutic significance of ultrasound-guided, intranodal LAG using lipiodol in 3 children with chylothorax after cardiac surgery.

The hospital records and radiological images of all children who developed chylothorax after cardiac surgery were retrospectively reviewed and approved by our institutional review board with a waiver of informed consent. The subjects comprised 3 patients who underwent LAG following refractory chylothorax. In all patients, high-output chylothorax of more than 200 mL/day persisted despite conservative treatment. A milky or creamy colored pleural effusion is considered affirmative of chylothorax. Serous pleural effusions that persists despite usual measures were evaluated for triglyceride and cellular contents. Triglyceride concentration of more than 110 mg/dL (1.24 mmol/L) principally established the diagnosis of a chylothorax and the presence of more than 80% lymphocytes in the effusion was also considered positive [6].

Ultrasound-guided intranodal LAG was undertaken in the interventional suite of the Radiology department. Two patients received endotracheal anesthesia while the third patient was carried out under deep sedation. The inguinal lymph nodes were accessed under

ultrasound guidance by using a 26-gauge needle (Terumo, Tokyo, Japan). Lipiodol injection was fluoroscopically guided to trace the efferent lymphatic fluid or lymph node, and to confirm the needle position. Two to five mls of lipiodol was delivered at a rate of 0.1 ml/min with serial fluoroscopic images obtained during the course of the injection. Intranodal LAG is successful if the target lymph node is positively selected and the lymphatic channels are adequately visualized by using Lipiodol.

Case Reports

From January 2011 to June 2017, there were 836 children who had undergone congenital heart surgery at our institution. There were 54 (6.5%) patients who developed chylothorax with 7 deaths and a mortality rate 13% (mortality in patients who did not carry the diagnosis = 5.6%). A majority resolved following conservative management with 3 patients requiring pleurodesis and one thoracic duct ligation, but in general there were significant drainage and prolonged hospital stays before these measures took effect. Three patients were subjected to LAG and table 1 outlines their characteristics and the outcome following LPG. None of them were syndromic and as part of protocol all had neck ultrasonography which proved negative for deep vein thrombosis.

Pt. #	Age Sex Weight	Surgery	Before LPG			After LPG	
			Days prior to LPG	Average vol/day (mls)	Treatment prior to LPG	Days after LPG	Average vol/day (mls)
1	2 months Male 3.5 kg	VSD Closure + Repair of interrupted aortic arch	53	371	MCT NPO with TPN Octreotide Betadine Pleurodesis	7	220
2	15 months Male 11 kg	Nikaidoh Operation for TGA/VSD/PS	78	200	MCT NPO with TPN Steroids	2	32
3*	21 months Male 12 kg	Fontan operation	25	763	MCT NPO with TPN Octreotide Steroids	7	284

Table 1: Demographics of patients and drainage before/after LPG

* Patient 3 also presents with chylous ascites.

LAG: Lymphangiogram; MCT: Medium Chain Triglyceride; VSD: Ventricular Septal Defect; NPO: nil per os; TPN: Total Parenteral Nutrition; TGA: Transposition of the Great Arteries; PS: Pulmonary Stenosis.

On average, these patients drained 445 mls/day for 52 days before LPG. The volume of effusion decreased to 180 mls/day and resolved at a mean of 5 days after the procedure. For patient #3, both chylothorax as well as the chylous ascites resolved following LAG. The patients tolerated the procedure without any untoward reaction to lipiodol. All 3 patients survived and were discharged home 7 days after LPG.

Discussion

Chylothorax following cardiac surgery is the most common cause in the pediatric population [6]. The mechanisms include direct injury of the thoracic duct, neck vein thrombosis and high central venous pressure (right ventricular diastolic dysfunction, cavopulmonary anastomosis) [3,7]. There is wide variation in its management and as a result, patients are at risk of prolonged, massive losses of pleural fluid that can lead to respiratory, nutritional, immunologic, hematologic and metabolic consequences as well as device utilization and increased length of stays [7]. Although the incidence of chylothorax in our report is fairly high (6.7%), it is within the range reported by others [1-3]. The relatively higher incidence reflects our sensitivity in establishing its presence because this complication has been on the rise in pediatric cardiac surgery, as observed in several reports [1,7]. Our 3 patients are representative of the groups at highest risk of chylothorax; namely neonates, children undergoing multiple procedures and subjects of cavopulmonary anastomosis as described in the analysis of the Pediatric Health Information System database [1]. This latter analysis additionally found mortality twice higher in patients with chylothorax than without [1] as we had equally discovered in our patients (13% vs. 5.6%).

LAG is traditionally conducted via the pedal approach [8]. Nadolski, *et al.* demonstrated the feasibility of intranodal LAG as an alternative to pedal LAG in 2013 [6] and ultrasound-guided intranodal LAG from the groin has been effectively utilized by others [9,10] but use of this modality in children is limited. Inguinal lymph nodes can be readily identified under ultrasound guidance in children and because isolation of lymphatic vessel is not necessary, this minimally invasive procedure is especially fitting for children. Lipiodol has been shown to be therapeutic in relation to its sclerosing and occlusion effects of lymphatics by the contrast agent. Recent reports affirm that the technique is reproducible and free of major complications [8]. We have found the procedure to be effective without any consequent untoward events. We have effectively used it in one patient following failure of pleurodesis and in patients who were poor candidates for surgical interventions.

Conclusion

We conclude that ultrasound guided intranodal lymphangiography can be therapeutic for children with high flow chylothoraces, chylothoraces that fail to respond to conservative therapy, and persistent chylothoraces. Its early use may avoid more aggressive approaches such as thoracic duct embolization and surgical interventions.

Conflict of Interest Statement

The authors declare that they have no conflicts of interest.

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