

An Unusual Presentation of Spontaneous Chylothorax

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ABSTRACT

True bilateral spontaneous chylothorax without any etiology has been reported rarely in the pediatric literature. A 3-year-old male child was detected to have incidental moderate chylothorax on USG thorax done for scrotal swelling. Investigations for infectious, malignant, cardiac, and congenital etiology were unremarkable. Effusion was drained by securing bilateral intercostal drains (ICD) and confirmed to be chyle on biochemical evaluation. The child was discharged with ICD *in situ*, but there was non-resolution of bilateral pleural effusion. Because of the failure of conservative treatment, video-assisted thoracoscopy (VATS) with pleurodesis was done. Thereafter, the child improved symptomatically and was discharged. On follow-up, there is no recurrence of pleural effusion, and the child has been growing well, albeit the etiology remains elusive. Chylothorax should not be missed in children presenting with scrotal swelling. In children with spontaneous chylothorax, VATS should be done after a fair trial of conservative medical management (thoracic drainage) along with continued nutritional management.

Keywords: Chylothorax, Pediatric intensive care unit, Video-assisted thoracoscopy.

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INTRODUCTION

Chylothorax, accumulation of chyle in pleural space, occurs due to various etiologies.¹ Although it is the most common form of pleural effusion in neonates, it is rare in children.² Acquired chylothorax has a definite attributable cause, whereas it is difficult to establish etiology in spontaneous chylothorax. Chylothorax is a potentially life-threatening disorder that can lead to serious metabolic, immunologic, and nutritional complications. The treatment is a low-fat diet, total parenteral nutrition (TPN) and surgical procedures have been described in those unresponsive to medical treatment.³

Twenty-nine cases have been reported in the literature with a diagnosis of spontaneous chylothorax with age-groups ranging from 1 month to 14 years.⁴⁻¹¹ We describe a unique case of a child presenting with scrotal swelling rather than respiratory distress.

CASE DESCRIPTION

A 3-year-old male child was brought by parents with complaints of painless scrotal swelling since 3 days. There was no history of fever, respiratory distress, cough or vomiting, recent trauma, or history suggestive of cardiopulmonary disease. The child's immunization was up to date for age.

Investigations and Treatment

Ultrasonography of the abdomen was suggestive of bilateral pleural effusion (approximately 200 cc on each side), which was an incidental finding. The child was admitted, and ICD were secured bilaterally. Daily drain monitoring was done. Pleural fluid was white and turbid with a high cell count (3200/cu.mm) with predominant lymphocytosis, high protein (5.9 gm/dL), and high triglycerides (1522 mg/dL), and had chylomicrons, suggestive of chyle.² Investigations for infectious, malignant, and cardiac etiology were unremarkable. A magnetic resonance (MR) lymphangiogram was normal with no evidence of a lymphatic leak. Pleural fluid culture grew *Enterococcus faecalis* and *Achromobacter xylosoxidans* which were treated as per sensitivity for 14 days. The child was prescribed a low-fat, high-protein diet with added medium-chain triglyceride formula.

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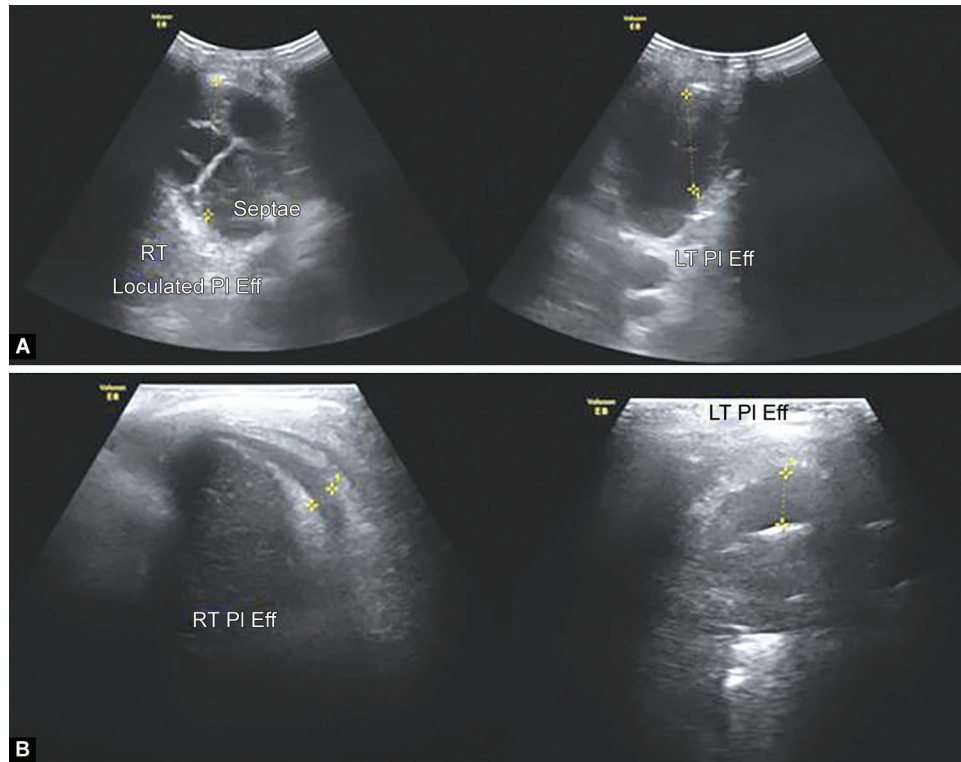
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Clinical condition improved, drain output reduced gradually, and the child was discharged on oral medication with ICD *in situ* after 25 days of hospital stay. The follow-up was done with USG thorax and a complete blood count. Pleural fluid quantity was documented to be reduced as compared to previously reported volumes. The trend of intercostal drain output and total leukocyte count has been shown in [Table 1](#).

Table 1: Trend of total drain output, absolute lymphocytic counts per cubic mm (/cu. mm), and total protein in milligram per deciliter (mg/dL)

Day of illness	Total drain output	Absolute lymphocytic counts	Total protein
1	–	3136	–
6	–	583	5800
13	180	1692	4600
16	590	1864	4260
21	100	1560	4960
46	0	828	5590
VATS with pleurodesis done on day 60 of illness			
70	0	2159	–
77	0	2840	7550

VATS, video-assisted thoracoscopy



Figs 1A and B: Ultrasonography thorax bilateral: (A) Pre-VATS and (B) Post-VATS reduced bilateral pleural effusion (PI Eff)

The child was asymptomatic till 1 month after discharge. After 1 month, child was readmitted with complaints of respiratory distress, fever, and generalized weakness for 8 days. On examination, the child was febrile with respiratory distress in form of tachypnea, nasal flaring, and mild subcostal retractions. Chest X-ray showed homogeneous opacity obscuring bilateral cardiophrenic angle and extending along bilateral lung fields suggestive of pleural effusion, which was confirmed on USG. Video-assisted thoracoscopy with pleurodesis was done with povidone-iodine [povidone iodine 0.1% + normal saline (1:1) ratio, volume: 25–30 mL] was done on both sides on separate occasions due to non-resolution despite adequate conservative treatment. Histopathology report showed acute necrotizing inflammation suggestive of empyema. Post-op X-ray and USG thorax showed complete resolution of effusion (Fig. 1). Intercostal drains were removed as drain outputs were reduced. On outpatient follow-up, there has been no recurrence of pleural effusion clinically or radiologically, and the child has been growing well with no further complaints.

Outcome and Follow-up

The child is under regular follow-up post VATS for the last 6 months. Follow-up clinical and radiological examinations have consistently demonstrated resolution of pleural effusion bilaterally.

DISCUSSION

Due to a smaller number of reported cases in the pediatric age-group and unknown causes, chylothorax still lacks well-defined treatment protocols. Currently, the treatment instituted is based on clinical acumen and hence individualized.

In the present case, the chylothorax was diagnosed incidentally with no leading cause or relevant history, thus, it represents a true

spontaneous chylothorax. The child presented with a history of scrotal swelling, which could be explained by inadequate drainage of the chyle leading to lymphedema and chylothorax as well. Since there are no guidelines in the literature to guide the management in this unique clinical scenario, we had to individualize the treatment. The objective was to manage chylothorax conservatively as much as possible, evaluate the cause concomitantly, and if non-responsive then switch to surgical treatment. The cause of lymphedema remained obscure despite satisfactory evaluation. The chylothorax transiently reduced in volume after chest drain insertion. There was a drop in absolute lymphocyte count (ALC) along with fever, so an infectious etiology was suspected with chyle as a favorable medium, which explains the growth of pathogenic organisms. The child was discharged with advice to monitor drain output and ALC. The ALC increased as the chyle output decreased, which indicates clinical recovery. One-month post-discharge, the volume again increased since it was complicated with empyema. Video-assisted thoracoscopy was done bilaterally due to failure of conservative treatment done for 35 days and complications with organized empyema. Follow-up did not show any recurrence of effusion on serial monitoring with USG to date.

The largest case series from China consisted of 22 children who received surgical treatment in form of povidone-iodine pleurodesis before being treated conservatively with diet and thoracic drainage. The outcome was favorable for all children with very few transient complications such as fever and chest pain.^{7,12}

This would be among few cases in the literature, to the best of our knowledge, with bilateral spontaneous chylothorax and unknown etiology. Our case was challenging since it was rare, and there are no established guidelines to direct management to obtain optimal outcomes despite a multidisciplinary approach.

Take-home Messages

One differential diagnosis for painless scrotal swelling should be a blockage of lymphatic drainage and should prompt into looking for chylothorax. In children with spontaneous chylothorax, trial of conservative medical management (drainage, nutrition, etc.) should be given for a reasonable time frame, i.e., 4–6 weeks. Video-assisted thoracoscopy and chemical pleurodesis may be performed if there is no response to conservative treatment or if there is superadded infection.

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