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SPONTANEOUS CHYLOTHORAX IN A CHILD: A RARE CASE PRESENTATION

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KEYWORDS

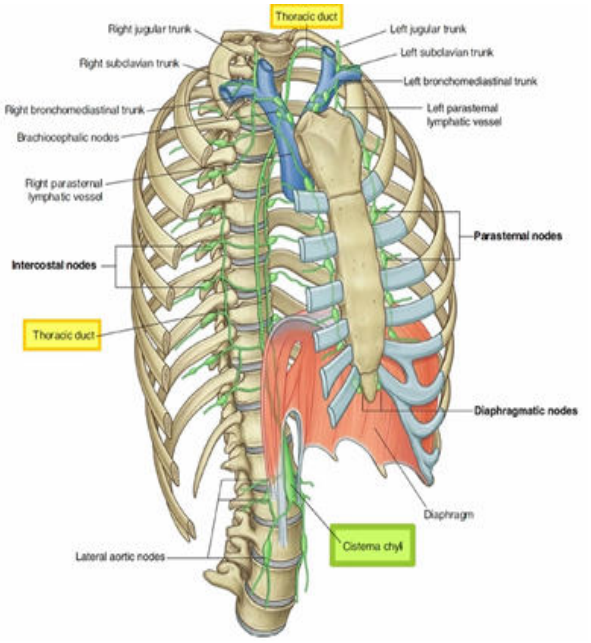
INTRODUCTION

Chylothorax is a rare disease, defined as an abnormal accumulation of chylous lymphatic effusion in thoracic cavity, with a high mortality rate in pediatric patients. At present, there are few studies on the treatment of pediatric chylothorax, and conservative treatments like somatostatin and pleurodesis are performed empirically. SST has been used for treating pediatric chylothorax over 20 years, and povidone-iodine,talc powder chemical pleurodesis . chylothorax is a rare disease resulting from the abnormal accumulation of lymphatic fluid in the thoracic cavity, and it is one of the most common causes of pleural effusion in young children. Although congenital chylothorax demonstrates prevalence in pediatric patients, occurring with an incidence of 1:6,000 to 1:24,000 live births, the mortality associated with this disease could reach 60%, uniquely combining with premature birth, low birth weight, fetal edema, lung hypoplasia, hypoproteinemia, or continuous chylous exudation (1)

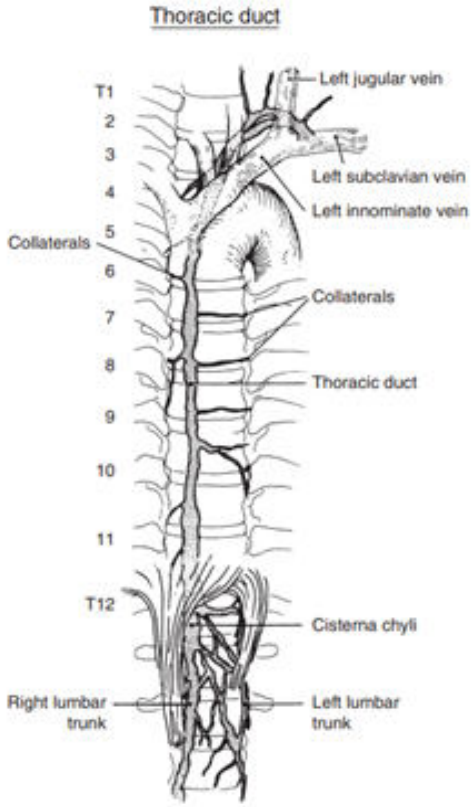
Depending on the level of chylous effusion, fluid accumulations can occur at any place in the thoracic cavity. Accumulation in the pleural space is typical but also chylopericardium and chylous ascites may result after injuring the chyle-transporting thoracic duct (3)

Thoracic Duct Anatomy

- The thoracic duct develops from outgrowths of the jugular lymphatic sacs and the cisterna chyli.
- originates in the abdomen at the cisterna chyli located over the second lumbar vertebra.



- extends into the thorax through the aortic hiatus and then passes upward into the posterior mediastinum on the right before shifting toward the left at the level of the fifth thoracic vertebra.
- It then ascends posterior to the aortic arch and into the posterior neck to the junction of the subclavian and internal jugular veins.
- contains smooth muscle sufficient force to propel lymph upward toward the jugular venous junction at a rate of 50 to 200 mL/hour



PRESENTATION AND COURSE IN HOSPITAL

A 7-year-old female child presented with fever and pain in neck for two months, and swelling on the right side of neck, axilla and chest wall since few days later.

The child was apparently well two months ago when she developed fever. it was it was intermittent in nature associated with chills, relieved with medications. the pain in neck was dull aching in nature used to aggravate intermittently and with neck movement. It also used to radiate to chest wall and upper arm.

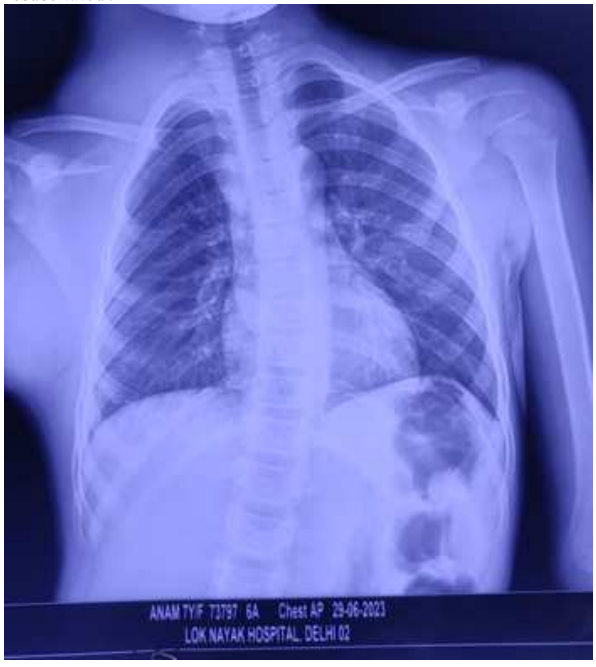
For these complain she underwent massage of the neck, after which she developed swelling in the right side of neck which rapidly increased in size to involve the right axilla, right upper arm and scapular region. this was associated with further decrease in neck movement.

To relieve the tension in the neck incision was taken on the lateral most dependent portion of the neck on left side following which her neck became more mobile.

Child then developed sudden respiratory distress after 2 days then immediate chest xray was done which was suggestive of right lung pleural effusion and immediately right chest icd was inserted which caused drainage of chyle fluid of around 300ml,al the chyle fluid was quantified daily, and child was kept npo and started on tpn AND SOMATOSTATIN . however there was no significant improvement in

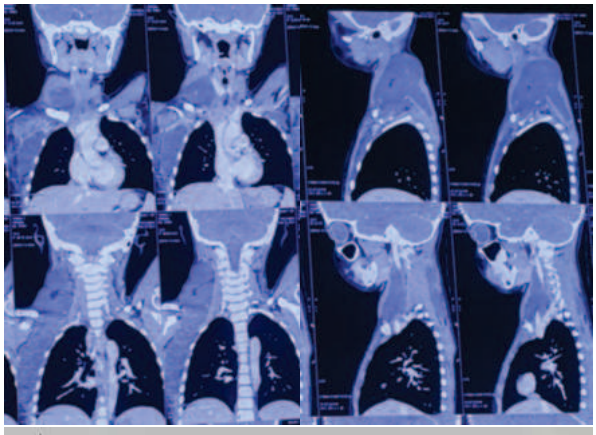
childs morbidity and hence she was planned for VATS pleurodesis .

Child s pleurodesis was done and ,but on post op day 2 she again developed respiratory distress and on auscultation no air entry was found and icd on left side was immediately inserted and then chyle fluid was coming out from left icd .but during the post op course child deteriorated and developed hypovolemic shock but couldn't be resuscitated .



CXR

CECT neck and chest-



CT NECK-

A well defined smoothly margiated nonenhancing fluid density lesion is seen in the subcutaneous plane and along the myofascial plane of the right lateral aspectof the neck extending from the level of the C1 Vertebra tracking inferiorly the axilla supraclavicular space along the right lateral chest wall .Maximum transverse and AP dimension of lesion in neck is 62mm x 73mm and maximum transverse and AP dimension of lesion in axilla and chest wall is 60mm x 75mm .The craniocaudal extent of the entire lesion in the neckaxilla and supraclavicular fossa and lateral chest wall on right side is 250mm.There are few thin septa and vessels seen through the lesion.No phleboliths, thick nodular enhancing septa solid enhancing component fat or rim enhancement is seen in the lesion.

Lymphoscintio Graphy

Showed significant tracer accumulation in B/L axillary, supraclavicular, cisterna chyli; S/O lymphatic leak and accumulation in subcutaneous neck.

Treatment Algorithm- aspiration of fluid from swelling revealed white coloured fluid with high levels of triglycerides

↓

Incisions were given on both sides of neck was done with aim to drain out the the fluid and swelling to settle.

↓

The swelling reduced in size, but used to increase again as the wounds got blocked.

↓

Attempt to seal, the fistula was done by instilling bleomycin in the neck wound

↓

Several attempts were made to dilate the wound and keep it patent for draining the fluid.

↓

Developed respiratory distress, CXR showed pleural effusion. USG guided pleural tapping of chylous fluid was done.

↓

↓

Right sided ICD was secured. Drain output continued 4-600 mL/day for several days.

↓

Planned for VATS. Lymphoscintigraphy was done

↓

VATS with Pleurodesis done with talc

CONCLUSION-

this case report presents neck massage as a probable cause of rupture of neck lymph which further aggravated and reached to thoracic Duct causing its rupture .there is lacunae in literature regarding treatment algorithm and strategies in children.

conservative treatment like multiple organ support, thoracic drainage,

somatostatin (SST), and pleurodesis are the first line treatment. Few children will finally be suggested to accept surgery, including ligation of the thoracic duct when conservative treatment fails (2).

The theory of PLEURODESIS and SOMATOSTATIN treatment is also different. PLEURODESIS works in closing the pleural cavity and induces the reconstruction of the lymphatic circulation by causing pleurisy (4,5), and SSOMATOSTATIN mainly promotes the self-healing of the lymphatic fistula by reducing the generation of lymph fluid and the pressure of lymphatic vessels.

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