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BILATERAL CHYLOTHRAX IN A 5-YEAR-OLD MALE WITH END STAGE RENAL DISEASE

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Abstract:

Background*: Chylothorax is a rare but serious complication of chronic venous lines. Here we are presenting a 5-year old with End stage renal disease who developed bilateral cylothorax secondary to right internal jugular and left subclavian vein due to the use of central hemodialysis lines*

Case presentation: *This s a 5 year old mare witj ESRD secondary to alport syndrpme who presented to emergency department with shortness of breath. Chest X ray showed right sided plural effusion and chemical analysis indicated that it's a chylothoax . Patient improved clinically and discharged in a good condtion.One month later he presented again to emergency department with severe shotnss of breath and chest X ray and plural fluid analysis indicted a bilateral chylothorax. Bilateral pigtail inserted. Patient managed conservatively with total parental nutrition.*

Conclusion: *Although Chylothorax is a rare but it is serious diagnosis that's require an attention. Patients on chronic venous lines are at particular risk to develop such a complication*

Background: *Chylothorax is an accumulation of chyle fluid in the plural cavity. It is are complication thats can develop secondary to venous stenosis or obstruction. Underlying mechanisms include developing of a thrombosis intitmal injury (1). Another potential mechanism for chylothorax is injury to thoracic duct in open heart surgery (1). Paints with chronic kidney disease are at particular rsil becuase of chonic use of central venous lines to undergo hemodialsysis Here we are presenting a first case in the literature for child with ESRD who developed a bilateral cylothorax.*

Key words: *Chylothorax, Alport syndrome , End stage renal disease*

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CASE PRESENTATION:

The patient is a 12-year old Yemeni boy with a known case of end-stage renal disease (ESRD) secondary to Alport syndrome and family history of ESRD in two of his brothers. One of the brothers died while the other underwent a renal transplant.

The patient has been on regular hemodialysis by central line through right internal jugular vein since September 2015.On June 20, 2017 the patient presented with a poorly functioning line which was changed through a guidewire on the same side.

On June 26, 2017 the patient presented to the hospital with fever and shortness of breath. On examination, patient was edematous, tachypneic, tachycardic, and hypertensive, with a blood pressure of 150/90mmHg.Chest auscultation revealed decreased breath sounds on the right side.

The patient underwent laboratory testing which showed the following results:K^+:6.7 mmol/l,Na^+:131 mmol/l,Mg^{2+}:1.07 mmol/l,and a pH of 7.27, indicating metabolic acidosis. Complete blood count testing revealed a white blood cell count of15.95 $*10^9$/L, C-reactive protein level of 89 mg/l,urea level of 28.6 mmol/land a creatinine level of1020 µmol/l.The patient was admitted to the pediatric intensive care unit (PICU) as a case of septic shock with central line infection and respiratory distress. A chest X-ray was done, which showed right pleural effusion (See picture 1).

In PICU, there was no backflow from his central right internal jugular vein. A temporary femoral line was inserted for urgent dialysis, cefepime and vancomycin were given, and another central line was inserted in the left subclavian vein through which hemodialysis continued.

On the second day in PICU the patient's condition deteriorated;both a central line and chest drain were inserted under sterile conditions. The central line was inserted in the right lower intercostal space, while the chest drain was inserted and drained around 460 mL of milky effusion. This fluid was sent for analysis, which revealed an increased level of triglycerides (9.65 mmol/L) and prompted the diagnosis of right chylothorax.

The patient’s symptoms improved immediately after the chest tube insertion.Chest tube drainage reduced progressively over the next days; the chest tube was removed after two days and the patient was discharged in a stable condition.

The patient presented to hospitala gain after 35 days with severe shortness of breath over the last 3 days.Chest examination revealed bilateral decreased air entry and bilateral coarse crepitations with a SpO_2 of 90% on room air.

An urgent Chest X-ray was done which showed bilateral pleural effusion; the patient was admitted to PICU as a case of bilateral pleural effusion with impending respiratory failure.

On the next day, the patient was drowsy and not fully awake, with a respiratory rate of 60 per minute progressing respiratory distress. Respiratory acidosis was diagnosed based on a venous blood gas PCO_2value of78 mm of Hg.A non rebreather mask was applied but respiratory distress remained with a SpO_2 of 90%. Bilateral size 14 pigtail chest drains were inserted under sedation and underwater seals were established which drained around 1300 mLof milky fluid which was sent for analysis, which showed the following results:LDH =106 U/L,TG=7.9,glucose =7.4 mmol/l,protein =43 g/l, Cl^- =104 mmol/l, K^+=5 mmol/l,and Na^+ =142 mmol/l.As a result, bilateral severe chylothorax was diagnosed.

A CT angiogram was done which showed occlusion of the central veins, except for the distal parts of the left internal jugular and subclavian veins which were narrowed. Bilateral moderate pleural effusion was also noted, multiloculated on the right. On the left, there were no definite signs of loculation (see picture 3). The patient was shifted from hemodialysis to peritoneal dialysis on August2017 due to extensive jugular vein thrombosis andtreated conservatively with nutritional support. After 5 weeks, chest drainage was stopped, and pigtail needles were removed. His chest X-ray showed resolution of bilateral effusion and the patient was discharged in a good condition See picture 4 .

DISCUSSION:

We reported a case of an ESRD patient secondary to Alport syndrome on chronic hemodialysis who presented to our ER with moderate bilateral pleural effusion. Chylothoraxis uncommon in hemodialysis patients; the diagnosis depends on pleural fluid analysis showing elevated triglyceride concentrations >6 mmol/L (or the detection of chylomicrons). Few of chylothoraces show the classic milky appearance. It may result from iatrogenic vascular trauma conducive to venous thrombosis and stenosis as hemodialysis catheter requires long-term indwelling. Generally other causes of chylothoraxmay include malignancy, cardiac failure,sarcoidosis, goiter, and thoracic or upper gastrointestinal surgery.[1]

Our case was associated with hemodialysis-induced right central vein obstruction as well as left central vein stenosis. In conclusion, two major factors were summarized, as they may have been the cause of central venous stenosis. First, having temporary central venous catheterization for hemodialysis. Second,the creation of arteriovenous shunt increased the turbulence of some regions causing high-flow state.[2]

Obstruction/stenosis of the innominate and central veins can cause lymphatic drainage impairment due to an increase in pressure in the thoracic duct and lymphatics. As a result, chylous effusion leaks into eitherinto the pleural cavity causing chylothorax or into the peritoneum causing chyloperitonieum.. [3]

Central venous obstruction is a serious complication that is difficult to manage and resolve. Usually, patients are asymptomatic. Elevated venous dialysis pressure is a sensitive indicator of central venous obstruction. Clinical signs and symptoms aspain in the arm, massive edema, or ipsilateral venous hypertension may precipitate in marked disability of the limb, which is responsible for thrombosis of the access site and incompetence to carry out proper dialysis. These downsides are aggravated in patients requiring chronic hemodialysis, with multiple surgical interventions to alternate the access site.[4]

A successful outcome was achieved in our patient with pleural drainage of chylous effusion as the patient was then shifted from chronic hemodialysis to automated peritoneal dialysis (APD). Studies have showed remarkable outcomes of the re-infusion of chylous effusion. Chyle is an immunoglobulin-/ albumin-/T-cell-rich fluid that, when evacuated and lost at a certain rate, may produce serious nutritional derangement, further compromising the host's immune system.As local or systemic infections are infrequent in patients with this disorder, chylous effusion has bacteriostatic properties. Despite the bacteriostasis exerted by the chylous fluid, long-term indwelling pleural drainage catheters could lead to infection, creating serious morbidity and possible mortality. [5]

Conservative treatment of axillary and subclavian thrombosis is usually palliative and ineffective. Combined endovascular procedures and stent placements has improved rates of permeability. Still, total occlusive lesions are difficult to resolve percutaneously.. [4]

Vascular access in pediatric hemodialysis patients is a challenging but mandatory undertaking for the practitioner. As the surgical approach and decision-making procedure remains poorly defined, proper advance planning is warranted to assure that the best permanent access is placed. A multidisciplinary team communication involving nephrologists, nurses, surgeons, and interventional radiologists, and ongoing monitoring is vital to ensure its long lifespan. [6]. Studies show that angioplasty appears to be advantageous for hemodialysis patients with central venous stenosis as it helps preserve functional access in the affected limb.However, stent placement in the central veins of dialysis patients has a high success rate resulting in symptomatic relief and maintenance of access. Taking into consideration cost-effectiveness, it is likely that the surgical techniques may be the best therapeutic option in some circumstances.[4]

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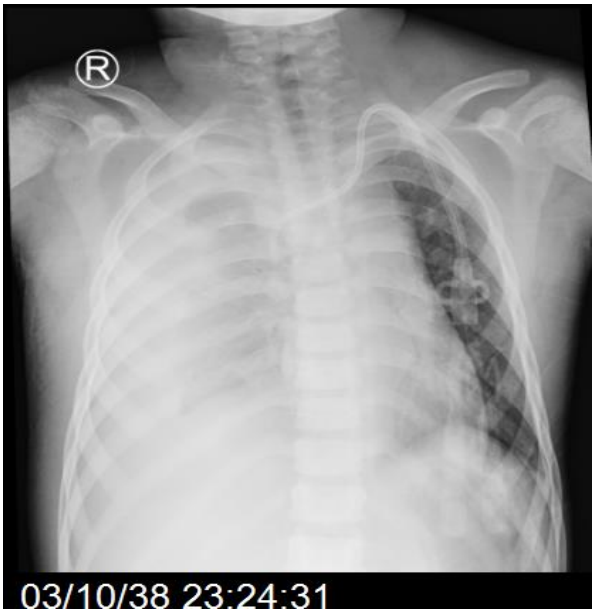
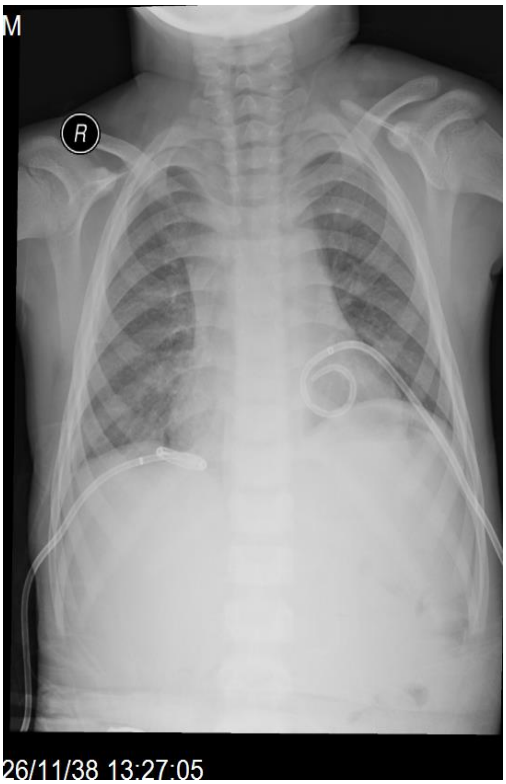
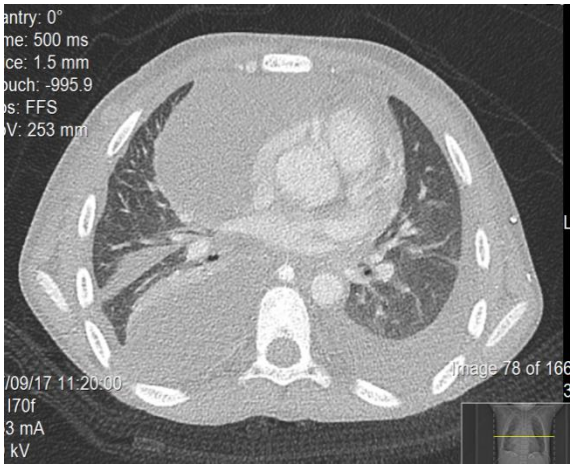
PICTURE 1: right sided plural effusion



Picture 2: resolution of right sided chylothorax after insertion of chest tube insertion



Picture 3: A CT angiogram was done which showed occlusion of the central veins, except for the distal parts of the left internal jugular and subclavian veins which were narrowed. Bilateral moderate pleural effusion was also noted, multiloculated on the right. On the left, there were no definite signs of loculation



Picture 4: Chest X ray showed improvement of bilateral plural effusion after drainage by pigtail size 14