Rare disease

Pleural effusion in a neonate

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Correspondence to Dr Sandeep Krishnanand Shetty, shettysandeep93@yahoo.co.in**Summary**

A premature neonate who developed respiratory distress in the first few days of life was found to have a pleural effusion, which reaccumulated following drainage. The effusion was demonstrated to be a chylothorax. He required multiple chest drains and was started on a medium chain triglyceride formula feed. This brought about a full resolution of the effusions and he made a complete recovery.

BACKGROUND

Pleural effusions in neonates are relatively uncommon, correct identification of the aetiology ensures the most appropriate management is undertaken.¹ This case highlights potential pitfalls in identification of an effusion, discusses chylothorax, its aetiology and management. Chylothorax is the most common cause of pleural effusion in a neonate, though it is most commonly post

surgical, it may also be associated with lymphatic disorders, certain syndromes, prematurity, or may occur in isolation.^{1–5}

CASE PRESENTATION

The patient is a premature infant born by emergency caesarean section at 32 weeks gestation for maternal pre-eclampsia. He was born to unrelated Caucasian parents

Figure 1 Chest x-ray showing right sided pleural effusion with underlying collapse.

Figure 2 Chest x-ray showing effusion with chest drain.

after an otherwise uneventful pregnancy, booking blood tests and scans had been unremarkable.

He was born in good condition, but developed respiratory distress in the hours after birth and required continuous positive airway pressure for a short period, but he had been self-ventilating in air from day 2 of life. Enteral feeds of preterm formula had been started cautiously on day 3 of life due to absent end diastolic flows on antenatal Doppler studies. He received parenteral nutrition via a long line in his right upper limb from day 2 until day 8 of life, this line was removed after he was established on full enteral feeds. He had been making good progress until the night of day 9, when he had been noted to be more lethargic and with increasingly frequent desaturations.

Enteral feeds were stopped, bloods were taken for a partial septic screen, antibiotics started and a chest x-ray was ordered. The chest x-ray showed a significant right sided pleural effusion with underlying collapse.

INVESTIGATIONS

Chest x-rays prior and post drain insertion. (figures 1–3)

Initial culture and sensitivity report: scanty pus cells, too few for a differential cell count, and scanty growth of fully sensitive staphylococcus epidermis (likely a contaminant).

Though a sample of the fluid was sent for biochemistry, triglycerides were not measured.

DIFFERENTIAL DIAGNOSIS

- ▶ Total parenteral nutrition (TPN) extravasation
- ▶ Chylothorax
- ▶ Parapneumonic effusion
- ▶ Empyema
- ▶ Haemothorax

TREATMENT

Because of increasing desaturations and apnoeas the decision was made to intubate and ventilate the neonate prior to drain insertion. This was done and respiratory status stabilised. The chest drain was inserted and immediately drained 35 mls of a clear yellow fluid, similar in appearance to TPN (figure 2).

The fluid was sent for microscopy, culture and sensitivities. Following drainage the neonate's respiratory status improved dramatically and he was extubated without difficulty.

After 24 h the drain was clamped, but a repeat chest x-ray showed reaccumulation (figure 3). The drain remained in situ for 4 days in total, until the effusion had resolved clinically and on chest x-ray, it was removed on day 13. Enteral feeds were reintroduced on day 10 and fully re-established by day 15.

OUTCOME AND FOLLOW-UP

The neonate remained well for the next week, however on day 21 of life he was noted to have developed increased

Figure 3 Chest x-ray showing reaccumulation of fluid – chylothorax.

work of breathing and desaturations. A chest x-ray again revealed a significant right sided effusion, confirmed on ultrasound scan. A chest drain inserted on this occasion drained over 50 mls of a thick milky fluid. On this occasion analysis of the fluid triglycerides and cholesterol revealed the fluid to be chyle.

The neonate's feeds were changed to a medium chain triglyceride (MCT) based formula, and a chest drain remained in situ over the next 8 days, removed on day 29. Following this he remained well until discharge 2 weeks later. He continued the MCT based formula for a further 4 weeks. He has remained well with no recurrence.

DISCUSSION

In retrospect the diagnosis seems obvious, but in an otherwise normal infant, who had no evidence of hydrops at delivery, a normal chest x-ray post delivery and at the time of long line insertion, and an effusion that appeared so similar to TPN, on the side where a long line had been in situ, it is understandable that the initial suspicion fell here. A delayed and incomplete analysis of the initial sample did not help point the team in the correct direction.

The appearance of the first effusion was due to the neonate's main source of nutrition being TPN, and the more typical chylous appearance of the second effusion, having been established on (standard preterm) formula feeds.

Chylothorax in neonates is most commonly a result of cardiac surgery, or associated with congenital abnormalities

of the lymphatics. Though it has been described in association with various causes such as, direct injury to the thoracic duct, thrombosis and/or high venous pressure in the superior vena cava for any reason.^{1–6}

In this case the chylothorax may have been to congenital abnormality or may have been precipitated by his treatment on the neonatal unit.

The neonate's effusions were successfully treated with switching of feeds to a MCT based formula. This reduces the production of chyle because MCTs passively diffuse from the gastrointestinal tract to the portal system bypassing the lymphatics. The reduced volume of chyle leads to spontaneous closure of the communication with the pleural space.^{3 4}

Though not required in this case, the next step in management would have been treatment with octreotide, a synthetic somatostatin analogue. Though the mechanism of action is not fully understood, it is known to reduce splanchnic blood flow, decrease thoracic duct flow and the triglyceride content of bile, decreasing the production of intestinal lymph.^{4 7–11}

A recent cochrane review by Das *et al*¹¹ looked at the published data to date on the use of octreotide, and though there are a multiple case reports, generally demonstrating resolution of chylothorax with octreotide, the initiation, dose, duration and frequency of doses varied significantly between authors, and there were no randomised controlled trials identified. They concluded that: 'No practice

recommendation can be made based on the evidence identified in this review. A prospective registry of chylothorax patients and a subsequent multicenter randomised controlled trial are needed to assess the safety and efficacy of octreotide in the treatment of chylothorax in neonates.⁷

In children who do not respond to MCT feeding or octreotide, the third option is surgery, previously chemical pleurodesis by intrapleural injection of povidone-iodine was used, but this must be avoided in infants due to the risk of potentially catastrophic renal failure.^{4 10 12}

There are several different procedures used for resistant chylothorax depending on its origins, previous procedures and surgical preference and experience; including pleural abrasion, pleurectomy and thoracic duct ligation.¹² In some difficult cases pleuroperitoneal shunts are still used.¹⁴ Le Nue *et al*¹² propose early surgical management for babies with significant prematurity, low birth weight and massive chylothorax with drainage of over >50 mL/kg/day, as these are less likely to respond to medical management alone.

Several authors have published cases of infants who have developed TPN related pleural effusions, though a very rare event, it may have serious consequences.^{15 16} Al-tawil *et al*⁵ looked at long term outcomes for a number of cases of congenital chylothorax presenting to two large neonatal centres over 13 years. They describe associations and the division between those cases resolving spontaneously, (62%) with conservative management (10%) or requiring surgery (28%), though they do not feature any patients who were treated with octreotide. They note an excellent survival rate, even in patients presenting with hydrops.

Though all the previous cases have looked at the issue in the neonatal period, the confusion between a TPN related effusion or a chylothorax may be encountered in adult practice also.

Wolthius *et al*¹⁷ describe a case of TPN leakage that was misdiagnosed as chylothorax and suggest analysis of potassium and glucose in addition to lipids in order to aid diagnosis.

Competing interests None.

Patient consent Obtained.

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Learning points

- Pleural effusion is an unusual presentation in a neonate, accurate diagnosis of the origin of the fluid is crucial to definitive management.
- The appearance of chyle is determined by the source of nutrition.
- Congenital chylothorax may resolve spontaneously, a MCT formula can aid resolution and avoid the need for somatostatin analogues or surgery.

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