CLINICAL CASE

MANAGEMENT OF POST-SURGICAL COMPLICATIONS IN A NEONATE CHYLOTHORAX: A CASE REPORT

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Abstract

Post-surgical chylothorax in neonates is a life-threatening condition which requires immediate diagnostic and treatment. The diagnosis is based on pleural fluid examination, which contains high levels of triglycerides and high cell count, mainly lymphocytes. The treatment consists in drainage, dietary changes, Somatostatin or its analogue Octreotide, pleurodesis or even surgical management. There are no evidence-based guidelines in chylothorax management, which makes the treatment election heavier. We report a newborn with chylothorax developed after primary repair of esophageal atresia with tracheo-esophageal fistula. The aim of this case report is to highlight the significance of post-surgical monitoring, anticipating the outcomes and rapidly managing the post-surgical complications.

Keywords*: chylothorax, octreotide, esophageal atresia*

Introduction

Chylothorax in neonates is a rare condition mostly associated with iatrogenic traumatisms as a complication of thoracic surgeries. The leading cause of chylothorax is esophageal surgery. The most common clinical presentation is respiratory distress. Usually the effusion is unilateral, right sided. The diagnostic consists in chest radiograph or pleural echography, showing an effusion and it is confirmed by thoracentesis. The mortality rate is high without properly management. It can lead to complications which have to be kept under control. Treatment can be conservative or it can require surgical approach [1,2].

Case presentation

A full term male baby, who underwent surgery for esophageal atresia (EA) with tracheo-esophageal fistula (TEF) in another hospital was referred to us at 38 days of life with severe respiratory distress worsened during feeding. The Barium swallow showed esophageal stenosis and gastroesophageal reflux disease (GERD), with no anastomotic leak. At admission he presented facial asymmetry during crying (Figure 1), severe respiratory distress (Figure 2), oxygen desaturations after feeding and respiratory acidosis. It was initiated conventional mechanical ventilation and he underwent surgery with resection and end-to-end anastomosis of the esophagus. In the postoperative period the baby presented

frequent desaturations and required higher ventilatory parameters.

The asymmetry of the lower lip was neurologically evaluated. The initial thought was facial palsy, but it was clinically infirmed by the symmetric wrinkling of the forehead, the complete and symmetric eye closure and the presence of both nasolabial folds. Therefore, it was further investigated and it proved to be a hypoplasia of the Depressor Angularis Oris muscle, a malformation associated in literature with gastrointestinal malformations.

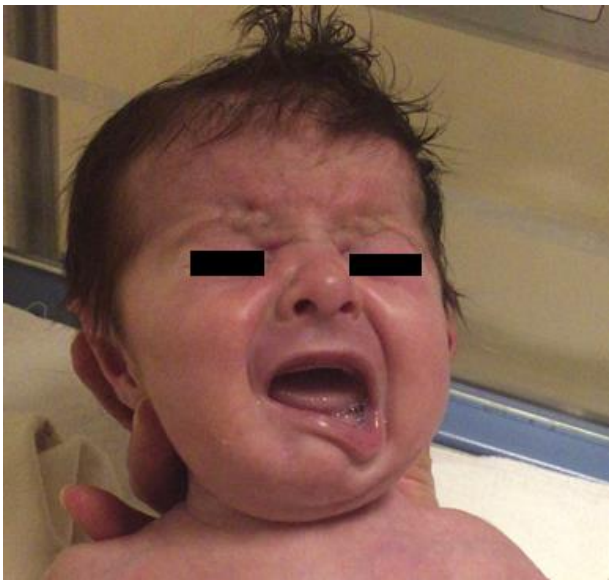

Figure 1 - Asymmetry of the lower left lip while crying

13 days after the second surgery the baby developed severe oxygen desaturation (SO2=40%) and bradycardia (HF=60bpm) with little response at balloon-ventilation. Moreover, his condition worsened with progression to SO2=20%, HF=50bpm and MAP=50mmHg, which required resuscitation with external cardiac massage and administration of Adrenaline and Dobutamine. Capillary blood gases revealed severe mixed acidosis with pH=6.577, pCO2=151.6mmHg and HCO3-=13.8mmol/l, immediately corrected with Sodium Bicarbonate. The Chest X-ray showed both hemithoraces opaque, sign of pleural effusion (Figure 3). Half hour later the CXR showed massive right pneumothorax, which was drained. He developed another episode which required resuscitation and left pneumothorax, also drained (Figure 4).

The pleural echography showed a large quantity of pleural liquid on the right side massively increased after feeding and the pleural drainage revealed 100 ml of serous and sanguinolent liquid (Figure 5).

The laboratory examination of the fluid revealed chylothorax (Table 1).

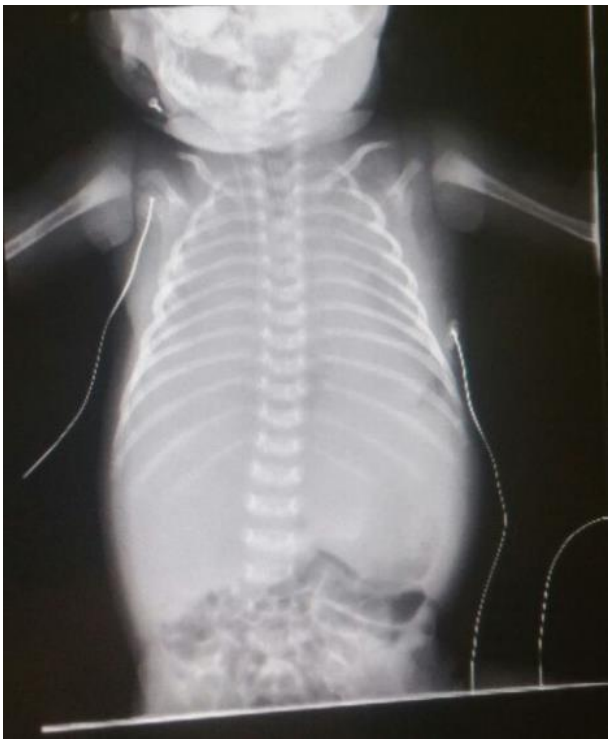

Figure 3 - Both right and left hemithorax completely opaque

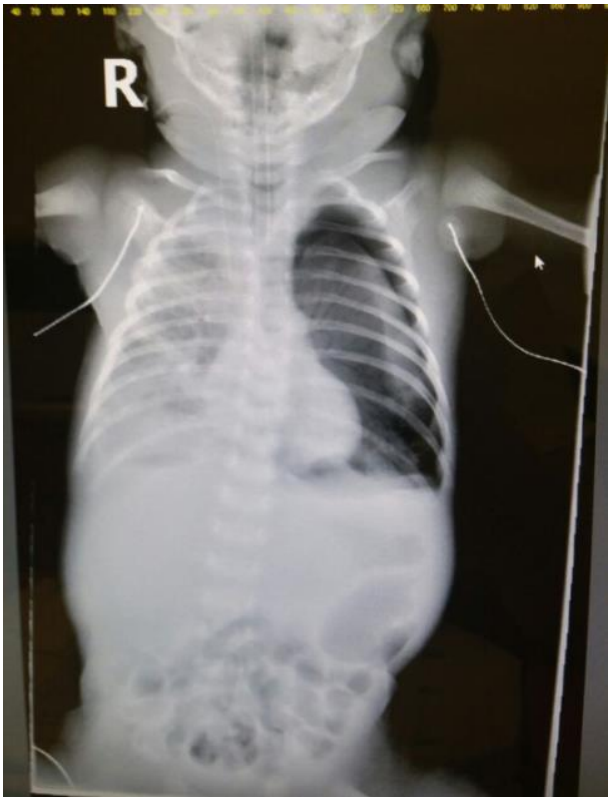

Figure 4 - Pleural effusion on the right side, pneumothorax on the left side

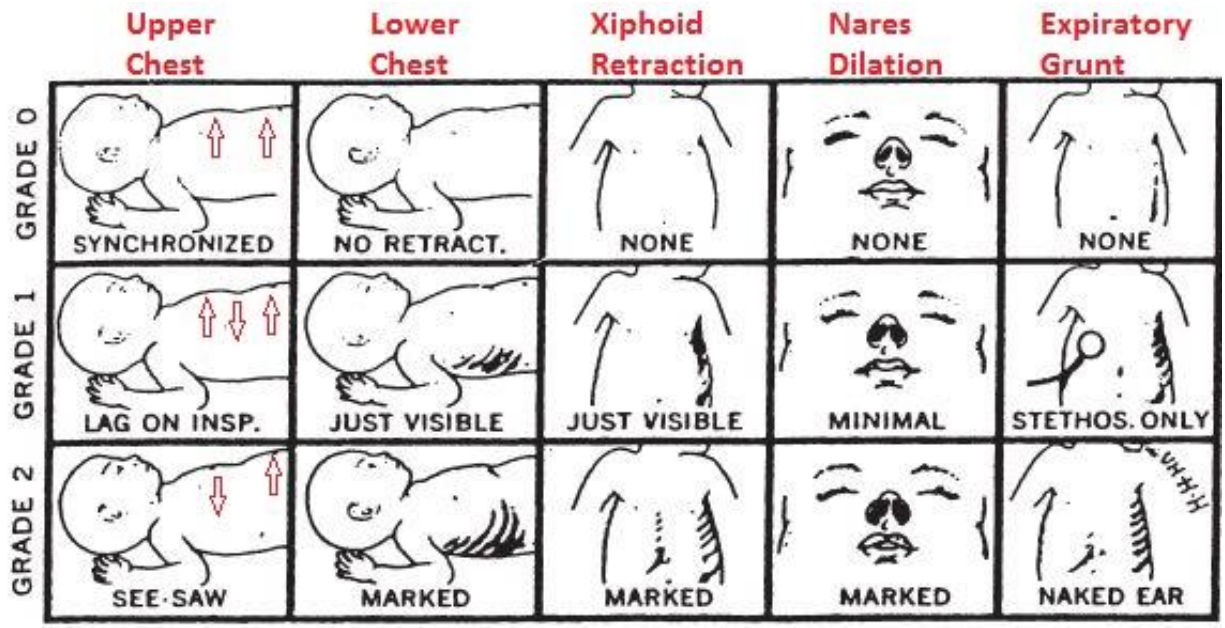

Figure 2 - Silverman Score. (Adapted from Silverman W.A., Andersen D. H Pediatrics 17 (1956) 1-10)

Figure 5 - Serous and sanguinolent pleural liquid

Feature	Patient
Milky colour/ clear (in starvation)	Clear
Sterile	Yes
Triglyceride level >110mg/dl	177.62mg/dl
High cell count	6516cell/mm^3
Lymphocytes predominance	90%

Table 1 - Pleural fluid features meet the chylothorax diagnostic criteria [3]

There are seven principles of management:

- drainage
- changing of the diet
- Somatostatin or analogues
- prevent and treat complications
- etiological treatment
- pleurodesis (with fibrin glue, talc, OK-432, etc)
- surgical treatment [3].

The first step we did was the diagnostic thoracentesis. Because we could not treat the underlying cause of the chyle leakage (damage of the thoracic duct during surgery) and the effusion was likely to reoccur, we placed chest tubes for continuous drainage (Figure 6). At the same time, the enteral nutrition was stopped. When the condition of the baby permitted, it was used a fat free diet with addition of medium-chain triglycerides (MCT). The baby received Octreotide (Somatostatin analogue) for 30 days via a central line - internal right jugular vein.

The effusion of the chyle stopped, so the surgical treatment was not needed. If the conservative treatment would not have been solved the chyle effusion in 4 weeks, an interventional method would have been required. This includes pleurodesis with fibrin glue, talc or OK-432 (a preparation of Streptococcus pyogenes) or surgical treatment - direct ligation of the thoracic duct or a pleuro-peritoneal shunt [4]. Another important component of the management is the prevention and the treatment of the complications, mainly derived from the chyle loss. Chyle is a liquid containing fat, electrolytes, proteins, glucose

and abundant leucocytes. The continuous drainage can produce malnutrition with important loss of calories, proteins, electrolytes, glucose, which need immediate replacement. Another complication is secondary immunodeficiency with higher risk of infection, therefore the baby was permanently screened for infections [5].

Figure 6 - The baby during continuous drainage

Because he had episodes of tracheal flow obstructions with desaturation and bradycardia even after completing the treatment of the chylothorax, a tracheal problem was suspected and the tracheoscopy revealed severe tracheal stenosis with impossible dilatation. The CT showed a tracheal stenosis of 2.7mm, which was surgically treated in Italy with costal cartilage tracheoplasty.

Discussions

Although rare, chylothorax was suspected from the beginning because of the thoracic surgery. The milky appearance of the pleural fluid, usually associated with chylothorax, is not always present. In our patient case, the pleural liquid was serous because of the improper alimentation due to the severe condition of the baby and sanguinolent because of the traumatic etiology.

Immediate treatment is vital, diet being an important part of the management. The primary components of chyle are long-chain triglycerides (LCT), therefore they must be significantly reduced in order to decrease the chyle formation. The diet can be improved using MCT, which contain saturated fatty acids of 8-12 carbon chain length. These bypass the lymphatic drainage, entering directly in the portal venous system [3]. Therefore, it was used a milk formula poor in LCT and rich in MCT.

Somatostatin is the only pharmacologic treatment which has proved its efficiency in chylothorax. It is a hormone involved in the gastrointestinal function, but the mechanisms incriminated in lymph formation are not entirely known. The most probable pathway is through vasoconstriction of splanchnic circulation, reducing the intestinal blood flow and, subsequently, lymph production. Another explanation may be that Somatostatin decreases the gastrointestinal motility, the gastric, pancreatic and biliary secretions, reducing the lymphatic flow [6]. Octreotide is a synthetic Somatostatin analogue, more convenient because of its longer half-life and greater potency. It seems to be well tolerated and no important side effects were registered [3].

The pneumothoraces our patient developed are most likely from both massive chylothorax and mechanical ventilation.

Conclusions

The post-surgical complications in a neonate can be extremely serious, with increased morbidity and mortality. The individual case management is required and permanent monitoring in a Neonatal Intensive Care Unit is mandatory.

Chylothorax can be difficult to manage and there is no consensus in its treatment. A prompt diagnosis is the key to a successful management. Adequate diet and prevention of the complications are also essential. The mortality associated with chylothorax is 50% and if it is post-surgical, the 30-day mortality increases five times [7]. Further studies are

required in order to find the best treatment algorithm.

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