CASE REPORT

Neonatal Esophageal Perforation with Esophageal Atresia: A Case Report

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ABSTRACT

Esophageal perforation (EP) in a neonate is very rare and is often iatrogenic. Preterm neonates with esophageal atresia (EA) are at more risk, as EP can occur during any test or intervention at pharyngeal region. We are presenting the management of EP in a preterm neonate with EA who presented with pneumomediastinum.

Key words: Esophageal atresia; Esophageal perforation; Pneumomediastinum

INTRODUCTION

Esophageal perforation (EP) in a neonate is often iatrogenic, can occur during repeated and forceful nasogastric/orogastric (NG/OG) tube placement, oropharyngeal suctioning, and endotracheal intubation [1]. In neonates, the pharyngoesophageal junction is the most frequent site of perforation likely due to anatomical narrowing as well as constriction due to instrumentation.

Prematurity with esophageal atresia (EA) makes a newborn more prone for EP due to weak musculature of blind-ending upper esophageal pouch [1,2]. The combination of EA in a premature complicates the situation. A high index of suspicion for EP is warranted in infants with a sudden deterioration of respiratory status, especially following procedures involving the pharyngeal region like NG tube placement [3].

CASE REPORT

A premature neonate with EA was referred to the pediatric emergency unit of a tertiary care center, 24 h after birth in a state of shock and respiratory distress. The neonate was born at 33-week gestation with a birth weight of 1.5 kg, by vaginal delivery due to premature labor. Mother was non-diabetic and pregnancy was unsupervised. The newborn had excessive salivation immediately after birth. There was a history of repeated attempts to insert an OG tube. The baby was intubated immediately and resuscitated. X-ray chest was suggestive of pneumomediastinum (Figure 1) and a chest tube was inserted on the left side. The baby was shifted to the neonatal surgical intensive care unit and put on the ventilator with all other supportive measures and broad-spectrum antibiotics. The condition optimized after 18–20 h of resuscitation and the baby was shifted for surgery with high-risk consent. Through a right posterolateral thoracotomy, EA with tracheoesophageal fistula (TEF) was confirmed. The gap between the upper and lower pouch was 2 cm. The upper pouch was friable and inflamed. The size of perforation was 4–5 mm and it was 7–8 mm above and medial to the distal-most end of upper esophageal pouch. During upper pouch mobilization, there was diffuse oozing of blood and it was initially difficult to identify the proper plane between the upper pouch and trachea. The lower pouch was thin, TEF which was just above the carina was ligated and primary esophageal anastomosis was performed over a 5 Fr feeding tube in a satisfactory manner. The perforation site was incorporated with the anastomosis. The post-operative course was very stormy and serial arterial blood gas analysis showed respiratory and metabolic acidosis. The blood sugar level was persistently high and had required insulin infusion for maintaining normal levels. The baby required triple vasopressor support and high ventilator settings. Despite all supportive and corrective measures, the newborn succumbed after 24 h of surgery.

DISCUSSION

A neonate with EP has different presentations, coughing, choking, bloody aspirates, respiratory distress,

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cyanosis after a feed, excessive salivation, and fever. The site of EP also decides the specific symptoms; thoracic EP may present with subcutaneous emphysema, tachycardia, tachypnea, and grunting while intra-abdominal EP often presents with signs of peritonitis. Chest X-ray can show pneumothorax, pleural effusion, pneumopericardium, and pneumomediastinum [4]. Furthermore, abnormal NG/OG tube location; too high, too low, or having variable or eccentric positions, should alert one to the possibility of EP [4,5].

Premature newborns are more vulnerable to iatrogenic EP due to their fragile tissue. The underlying anatomical abnormality like EA further increases the risk of EP when a forceful NG/OG tube placement had been attempted as in our case [3]. The management is not straightforward and it depends on the size of the perforation, its location, the severity of the infection, and the general condition of the neonate. Basic principles of management are as shown in Figure 2. There are certain peculiar issues with EP in an EATEF child, and due to these, conservative management has no role, repeated suctioning may increase the esophageal rent, distal TEF can worsen the respiratory distress, salivary contamination may cause mediastinitis, associated cardiac anomaly can worsen the situation, and finally, esophageal continuity cannot be established without surgical intervention.

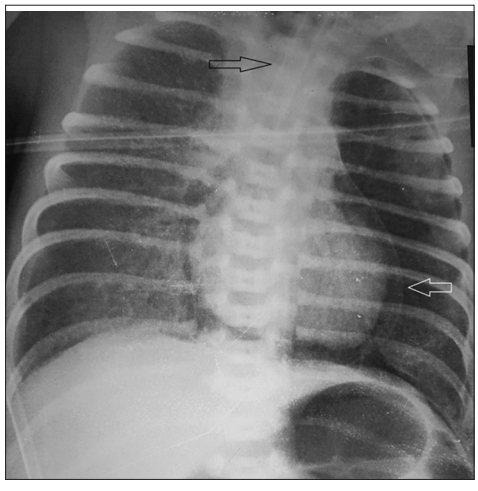

Figure 1: X-ray howing pneumomediastinum (white arrow) and endotracheal tube (black arrow)

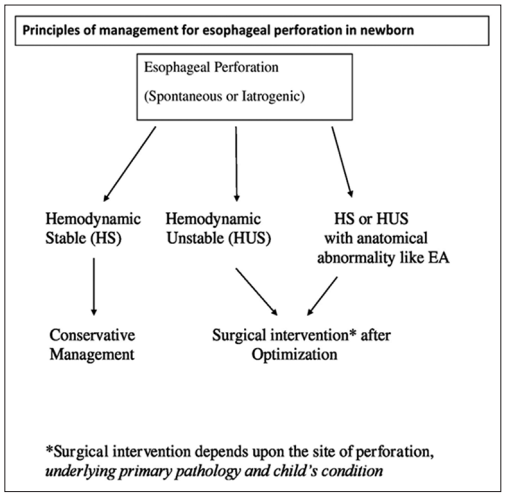

Figure 2: Principles of neonatal esophageal perforation management

Another important point to remember is that EP can mimic the presentation of EA that further adds dilemma in diagnosis. Management of EP, EA, and "EP in EA" is different and so differentiation between these entities is very important before treatment planning [6,7]. Aoun et al. reported a case of EP diagnosed as EP in EATEF during thoracotomy, there was no EA and it was EP only [5].

Perforation of the esophagus causes normal flora of the esophagus to leak into the mediastinal space, which can result in inflammation and infection. Contamination of the mediastinum with bacteria leads to cardiorespiratory embarrassment, shock, major fluid losses, and fulminating infection. Such infection can lead to sepsis and organ failure with a high mortality rate if the diagnosis is missed and left untreated [8].

Principles of management for EP include rapid diagnosis, appropriate hemodynamic monitoring and support, antibiotic therapy, total parenteral nutrition, control of extraluminal contamination, and restoration of luminal integrity or proper diversion either through conservative management or operative approaches. The initial management is to prevent further deterioration and spillage of secretions into thoracic cavity/mediastinum by suctioning and drainage of secretions. Prompt chest tube insertion should be done on the affected side. Once the child is stabilized, the correction of EA should be considered. The esophageal diversion for pure EA and anatomical repair for EATEF can be done, depending on the child's condition and underlying anatomy [2].

In our case, thoracotomy was planned with intent of fistula ligation along with drainage of mediastinum, and following this, we would consider esophageal diversion or primary esophageal repair according to the local anatomy. Despite the presence of inflamed and friable tissue of the upper esophageal pouch, we were able to obtain a satisfactory primary anatomical repair. As the clinical condition was deteriorating postoperatively, despite all supportive measures suggested that irreversible injury was already established due to mediastinitis and septicemia that led to multiorgan dysfunction.

A premature neonate with EA/TEF would as such have a poorer prognosis as compared to his/her term counterpart; the presence of EP would further complicate issues and worsen the situation. The mortality in the literature for EP is mentioned to be approximately

30% and when it is associated with EATEF, mortality increases significantly [2,4,5].

CONCLUSION

Premature neonates with EA are more prone to EP; any intervention in the pharyngeal region can cause iatrogenic EP, especially after forceful and repeated attempts. The treating physician should be aware of this condition and use utmost gentleness in a newborn when performing any test or intervention (NG/OG/ET tube placement) in the pharyngeal region.

REFERENCES

1. Hesketh AJ, Behr CA, Soffer SZ, Hong AR, Glick RD. Neonatal esophageal perforation: Nonoperative management. J Surg Res. 2015;198:1-6.
2. Parelkar S, Mundada D, Joshi P, Sanghvi B, Kapadnis S, Oak S. Iatrogenic perforation of upper pouch in pure esophageal atresia: A rare complication and review of literature. Eur J Pediatr Surg Rep. 2013;1:21-3.
3. Suryawanshi P, Dahat A, Nagpal R, Malshe N, Kalrao V. A rare case of accidental esophageal perforation in an extremely low birth weight neonate. J Clin Diag Res. 2014;8:1-2.
4. Gander WE, Berdon RA. Cowles. Iatrogenic esophageal perforation in children. Paediatr Surg Int. 2009;25:395-401.
5. Aoun JB, Gasmi M, Jemai R, Sghairoun N, Sahli S, Hamzaoui M. Iatrogenic esophageal perforation in the neonate. Tunis Med. 2012;90:72-4.
6. Seefelder C, Elango S, Rosbe KW, Jennings RW. Oesoph ageal perforation presenting as oesophageal atresia in a premature neonate following difficult intubation. Paediatr Anaesth. 2001;11:112-8.
7. Meeralebbae SM, Zain SM, Basir AO, Ahmed AM, Abdullah AM. Misdiagnosing esophageal perforation as esophageal atresia. Saudi Med J. 2002;23:1287-90.
8. Kassem MM, Wallen JM. Esophageal perforation, rupture and tears. StatPearls. Treasure Island (FL): StatPearls Publishing; 2018. Available fromttp://www.ncbi.nlm.nih.gov/books/NBK532298. [Last accessed on 2018 Oct 03].