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Case Report

Iatrogenic Pharyngo-Esophageal Perforation in Neonate: A Rare Yet Significant Challenge in Modern Neonatal Care

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Abstract

Iatrogenic pharyngo-esophageal perforation (IPEP) in neonates is an uncommon yet critical medical complication. IPEP poses an exceptionally high risk for preterm neonates born at less than 28 weeks of gestation or with a birth weight below 1500g. Neonatal IPEP primarily stems from medical procedures such as nasogastric or orogastric tube insertion, multiple attempts at endotracheal intubation, and pharyngeal suctioning. We present a preterm neonate born at 24 weeks and six days of gestational age with pyriform sinus perforation, a phenomenon previously unreported in this population, diagnosed with the use of point-of-care ultrasound (POCUS). The management of neonatal IPEP is shifting from surgical intervention to conservative strategies that prioritize nutritional support, antibiotic therapy, and the utilization of POCUS. This manuscript emphasizes the critical need for safe medical procedures and careful monitoring in neonatal care to promptly recognize IPEP and minimize its associated morbidity and mortality.

Keywords: *Esophageal Perforation; Pharyngeal Perforation; Pyriform Sinus Perforation; Preterm; Neonates; Iatrogenic; Point-of-Care Ultrasound*

Abbreviations

IPEP: Iatrogenic Pharyngo-Esophageal Perforation; POCUS: Point-of-Care Ultrasound; NGT: Nasogastric Tube; NICU: Neonatal Intensive Care Unit; CXR: Chest Radiography; DOL: Day of Life; PMA: Postmenstrual Age; NPO: Nil Per Oral; NIS: National Inpatient Sample

Introduction

Iatrogenic pharyngo-esophageal perforation (IPEP) in neonates, particularly preterm infants, is a rare but critical medical condition. The case of an extreme preterm neonate, as described in this manuscript, sheds light on the unique challenges and complexities surrounding IPEP. This case highlights the importance of vigilance, early detection, and effective management to mitigate the risks associated with neonatal IPEP. Pyriform sinus perforation has not previously been documented among neonates. Furthermore, the utilization of point-of-care ultrasound (POCUS) can provide valuable diagnostic insights when traditional radiological imaging fails to deliver a conclusive diagnosis. Overall, this manuscript emphasizes the critical need for safe medical procedures and careful monitoring in neonatal care to promptly recognize IPEP and minimize its associated morbidity and mortality.

Case Presentation

A preterm neonate was delivered at an outside hospital at a gestational age of 24 weeks and six days to a gravida 5 para 2 mother. The maternal history revealed no significant issues; all prenatal laboratory results were reassuring. This neonate was the first twin of a diamniotic dichorionic twin pregnancy and delivered vaginally due to preterm labor and premature preterm rupture of membranes. The mother received a single dose of betamethasone during delivery as labor progressed. The neonate, weighing 587g (13th percentile) and measuring 29 cm in length (7th percentile), had Apgar scores of 4, 6, and 7 at 1, 5, and 10 minutes, respectively. In the delivery room, the infant initially received positive pressure ventilation and required intubation due to insufficient respiratory effort and hypoxic respiratory failure. Multiple attempts were made to place the endotracheal tube. A dose of surfactant was administered due to a persistently high oxygen requirement before admission to the Neonatal Intensive Care Unit (NICU). Several attempts were made to place a nasogastric tube (NGT); however, the NGT could not be advanced and was eventually removed. Upon NICU admission, the neonate was placed on conventional mechanical ventilation. Chest radiography (CXR) revealed a left pneumothorax, necessitating needle aspiration in the second intercostal space. Given the need for more specialized care, the infant was transferred to our tertiary care level 4 NICU for further evaluation and management.

Upon admission to our NICU, repeat CXR and POCUS lung revealed a large, left-sided pneumothorax. The CXR also showed an atypical course of the NGT, which was positioned laterally to the mediastinum, with the distal tip projecting over the gastric bubble suspicious for esophageal perforation (Figure 1). The NGT was removed, and he received a 10-day course of intravenous piperacillin-tazobactam. He was placed on high-frequency jet ventilation to manage the air leak and treated with inhaled nitric oxide. Serial CXRs indicated complete resolution of pneumothorax within 48 hours.

Figure 1: *Chest radiography on day 1.*

On the fifth day of life (DOL), a second attempt was made to place an NGT. The CXR showed an equivocal course of the NGT in the left paraspinal region, with the tip in the left upper abdomen (Figure 2A). The position of the NGT, combined with the worsening left pneumothorax, suggested a malpositioned NGT. POCUS confirmed the tracking of the NGT in the prevertebral space (Figure 2B), which lateral CXR further validated (Figure 2C). The NGT was then promptly removed. Bedside flexible laryngoscopy performed by the Pediatric Otolaryngology team revealed a circular area in the left piriform sinus with air bubbles, suggesting perforation (Figure 3). Based on recommendations from Pediatric Otolaryngology, we avoided instrumentation of the oropharyngeal area for one week to facilitate healing.

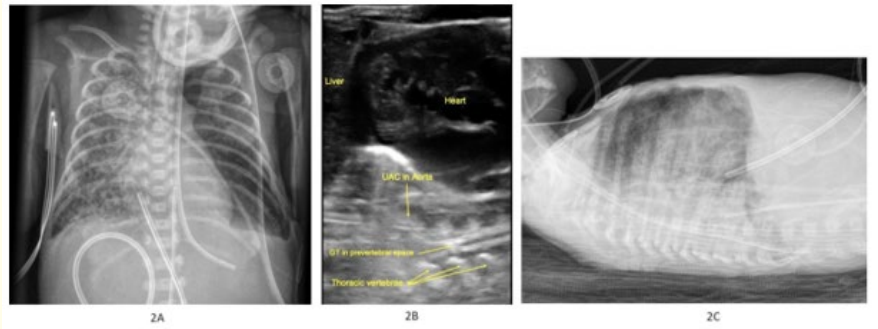

Figure 2: *Radiographic imaging on day 5. (2A: Chest radiograph, anteroposterior view, 2B: POCUS; 2C: Chest radiograph, lateral view).*

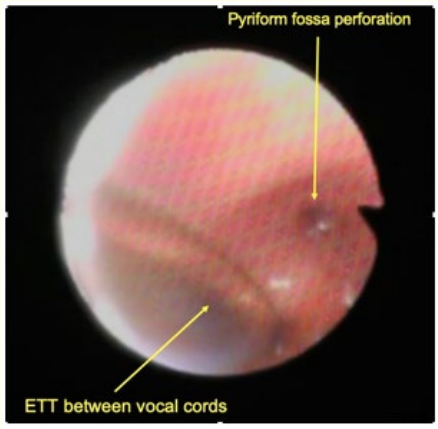

Figure 3: *Flexible laryngoscopy.*

On DOL 12, successful NGT placement was achieved under video laryngoscopy guidance with position confirmed with CXR and POCUS (Figure 4). Repeat flexible laryngoscopy by Pediatric Otolaryngology confirmed pyriform recesses with no obvious mucosal abnormalities, perforations, or ulcerations.

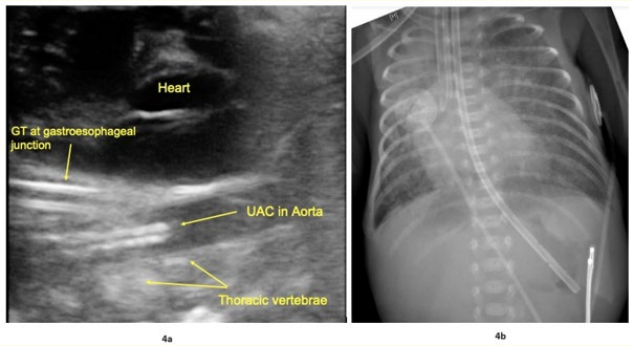

Figure 4: *Radiographic imaging on day 12. (4A: POCUS, 4B: chest radiograph anteroposterior view).*

During the NICU stay, the infant experienced a range of prematurity-related complications, including bronchopulmonary dysplasia, intraventricular hemorrhage, parenteral nutrition-associated liver dysfunction, apnea of prematurity, osteopenia of prematurity, retinopathy of prematurity and anemia of prematurity. The patient transitioned to noninvasive ventilation at a postmenstrual age (PMA) of 31 weeks, was subsequently weaned to continuous positive airway pressure at a PMA of 39 weeks, and finally to a nasal cannula at a PMA of 43 weeks when transferred to a long-term rehabilitation facility for ongoing care and management.

Discussion

Iatrogenic pharyngo-esophageal perforation (IPEP) in newborns, particularly in preterm infants, is a rare but critical medical concern [1,2]. Iatrogenic injury is the most common cause of IPEP. IPEP poses an exceptionally high risk for preterm neonates, born at less than 28 weeks of gestation, or neonates with a birth weight below 1500g [3]. The incidence of IPEP in extremely preterm infants of < 28 weeks is estimated to range from 0.8 to 1% [4]. However, this risk increases in newborns weighing less than 750 grams [5,6]. Over the last two decades [2], using the National Inpatient Sample (NIS) database, Elgendy., *et al*. found a significant upward trend in the incidence of IPEP concurrent with increased focus on managing extremely premature infants and those with extremely low birth weight. Neonatal IPEP is associated with increased mortality, with rates as high as 30%, mainly from prematurity-related complications rather than the iatrogenic injury [4,7,8].

Neonatal IPEP perforations are usually iatrogenic and stem from medical procedures. These procedures include enterogastric tube insertion, multiple attempts at airway intubations, laryngoscopy, suctioning, the use of a laryngeal mask airway, trans-esophageal echocardiography probe insertion and digital manipulation of the neonatal head during breech delivery [9-12]. Due to anatomical narrowing, neonatal IPEP is most often found in the cervical esophagus near the cricopharyngeal muscle or pharyngoesophageal junction [12]. In contrast, pediatric patients typically experience perforations in the thoracic esophagus [13]. Our case report is noteworthy because the infant had a perforation in the pyriform sinus, marking the first documented instance in neonates. The pyriform sinus, a fragile, pear-shaped fossa in the hypopharynx, lies on both sides of the laryngeal opening in a posterolateral position [17]. Remarkably, pyriform sinus perforations had not been reported in neonates previously, although they have been documented in pediatric and adult patients [14-16].

The clinical presentation of IPEP is frequently asymptomatic in neonates and is usually detected through radiological imaging. In some cases, noticeable resistance may be encountered while passing a feeding tube. Alternatively, the infant may experience a clinical deterioration with symptoms like respiratory distress, cyanosis, choking, excessive drooling, vomiting, and indications of sepsis, such as fever. Moreover, subcutaneous or mediastinal emphysema might also be observed.

Diagnosis of IPEP may be made by chest radiography and POCUS. With proper positioning, NGT should follow a specific path along the mid to the left side of the spine and the trachea in a non-rotated chest radiograph, curving left toward the stomach as it passes through the diaphragm [18]. Deviation from this expected route should prompt suspicion of IPEP, underscoring the critical importance of imaging in early diagnosis and timely intervention in neonatal care. For this neonate, the chest radiograph revealed left pneumomediastinum and pneumothorax, with the NGT located outside the esophageal column and appearing to traverse posteriorly, possibly within the left pleural space. This finding prompted consideration of a possible diagnosis of IPEP and the necessity to remove the NGT.

Over the last two decades, the approach to managing neonatal IPEP has transitioned from surgical to conservative strategy. Surgical intervention is reserved for cases of persistent clinical deterioration despite conservative approaches [7,19]. The goal of managing IPEP is to prevent further infection and inflammation, restore the integrity of the alimentary tract, and enhance the patient's overall clinical and nutritional well-being. Neonates diagnosed with IPEP are put on Nil Per Oral (NPO) status and provided with Total Parenteral Nutrition. The healing typically takes place in seven to ten days [8]. In most cases, broad-spectrum antibiotics are prescribed for 7 - 14 days to

prevent local and systemic infections, although the precise antibiotic regimen varies [12]. This conservative approach has demonstrated success in over 85% of cases, leading to recovery without significant complications [4]. For our neonate, the IPEP healed within 10 - 12 days to heal and was treated with a 10-day course of broad-spectrum antibiotics.

Preventing IPEP involves a comprehensive approach encompassing several key measures. Healthcare providers should exercise caution and expertise when using entero-gastric tubes. Proper training in safe insertion techniques and the use of lubricants during insertion is crucial. Vigilance for warning signs, such as resistance to passage, bleeding, or signs of respiratory distress after tube insertion, is essential for early recognition and intervention. Alternative methods like fluoroscopic placement or laryngoscopy (direct or video) should be considered if initial attempts are challenging. Confirming tube placement through 2-view chest radiographs, anteroposterior and lateral, especially for extreme preterm neonates, is crucial [18,20]. POCUS can be utilized to confirm the position of gastric tubes. Ultrasonography offers a faster, more cost-effective, and radiation-free alternative. POCUS provides the potential for real-time visualization of the NGT during insertion and after achieving its final position [21]. We utilized video laryngoscopy for nasogastric insertion, followed by confirmation with POCUS and 2-view chest radiographs to ensure accurate placement. When intubation is necessary, providers should limit attempts to a maximum of two per provider and exercise caution when using the stylet to prevent injury [22]. Identifying and closely monitoring neonates who have experienced multiple intubation attempts is vital as these cases may warrant extra attention.

Conclusion

This case of IPEP in this preterm neonate underscores the multifaceted challenges and risks inherent in neonatal intensive care. While neonatal IPEP remains rare, its potential consequences demand a high level of vigilance, early detection, and adept management. The evolution of management strategies from surgical intervention to conservative approaches has shown improved outcomes for these fragile patients. Moreover, this case report contributes to the medical literature by documenting a unique instance of pyriform sinus perforation in a neonate, expanding our understanding of potential IPEP locations. Additionally, the report strengthens the current evidence suggesting that POCUS, including its application in cases of IPEP, is an effective diagnostic tool for various neonatal pathologies. Ultimately, the key takeaway from this case is the paramount importance of minimizing iatrogenic risks through safe medical procedures, prudent monitoring, and ongoing education for healthcare providers in the neonatal care setting.

Conflict of Interest

The authors have no conflicts of interest to disclose.

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