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Case Report

Pneumoperitoneum in an Extremely Preterm Neonate. Can it be an Innocent Finding? Case Report

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Abstract

We report a case of extremely preterm neonate 24 weeks, who presented with abrupt deterioration of her clinical condition with abdominal distension associated with bluish discoloration of her anterior abdominal wall due to sudden onset pneumoperitoneum. In view of worsening distension and compromise of her ventilation support, peritoneal drain was inserted initially that revealed serous peritoneal fluid with air bubble. Later on exploratory laparotomy confirmed normal bowel wall integrity. Concomitant chest x ray showed pulmonary interstitial emphysema. To our knowledge this is the 1st reported case of pneumoperitoneum 2ry to pulmonary air leak in 24 week extremely preterm neonate found to be associated with pulmonary interstitial emphysema with good survival and outcome.

Keywords: *Pneumoperitoneum; Preterm Neonate, Pulmonary interstitial emphysema, pulmonary air leak syndromes*

Introduction and Case Presentation

An extremely preterm Female baby was born at 24+2 week with birth weight of 630 gram. She was born to 25 years old mother, with no antenatal follow up in our tertiary referral hospital. Mother is Gravida 4, Para 0+3 with 3 consecutive miscarriages, she had pregnancy induced hypertension, diabetes mellitus type 1 on insulin, her GBS screen was unknown, her serology as well as COVID screening was -VE. She is rubella immune, presented to emergency department with abdominal pain and bleeding, she received one antenatal course of betamethasone.

A micro preemie baby was born cephalic by vaginal delivery, soon covered with plastic bag, kept under warmed resuscitator, she was flat, started with positive pressure ventilation then intubated in view of bradycardia and poor respiratory efforts and she was transferred

to NICU on portable ventilator. Her Apgar score was 2/5/7 at 1/5/10 minutes respectively. Golden hour applied, received caffeine, started on total parenteral nutrition as well as ampicillin and gentamicin, umbilical lines were inserted.

Her birth weight 630 gram, head circumference: 23 cm, length: 29 cm.

On admission to NICU, she was given the 1st dose of Survanta followed by another 2 doses in view of serve respiratory distress syndrome. She was started on intraventricular haemorrhage (IVH) care bundle which is part of normal care of such micro preemie in our unit. Her initial brain ultrasound scanning in the 1st day of life (DOL) was normal.

In view of abnormal intestinal gas distribution pattern with paucity of intestinal gases on the initial abdominal x ray at the end of the 1st day of life, feeding was not started initially. As she had delayed passage of meconium on 3rd DOL, abdominal Ultrasound was done that showed minimal free peritoneal fluid, fortunate enough on the 4th DOL, she passed meconium spontaneously. Worth mentioning that her repeat brain ultrasound scan after completion of IVH bundle on the 3rd DOL showed IVH grade I-II on the right side. Follow up abdominal X ray on 5th DOL showed better intestinal gaseous distribution. She was started on trophic feed on 6th DOL that was so far tolerated.

On 8th DOL, it was noticed that she became lazy with no response, completely knocked down tiny neonate; she had abdominal distension with bluish discoloration of the anterior abdominal wall. Urgent abdominal x ray showed free intraperitoneal air both in AP and Cross table (Figure 1 with Cannon Ball appearance and figure 2 with air under the anterior abdominal wall). Pediatric surgical consultation was obtained, abdominal drain was inserted that revealed serous fluid with a lot air bubble. after stabilization, getting parental consent and preparation of blood products for the operative intervention, she underwent exploratory laparotomy that revealed clean peritoneal cavity, with no meconium, no malrotation, the duodenojejunal and the cecum all in normal position, intact whole healthy bowel loops, no perforation seen with normally dilated bowel with equal caliber and presence of meconium in the colon. she underwent closure of the abdominal wall with no stoma formation.

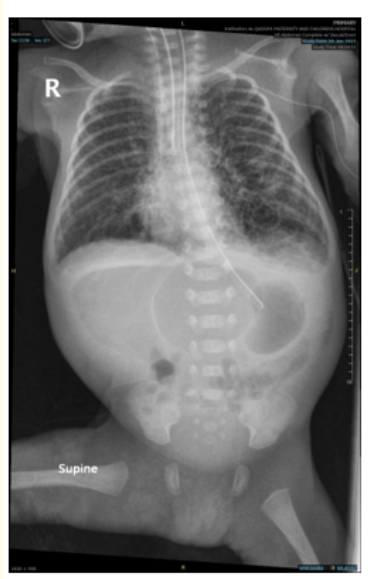

Figure 1

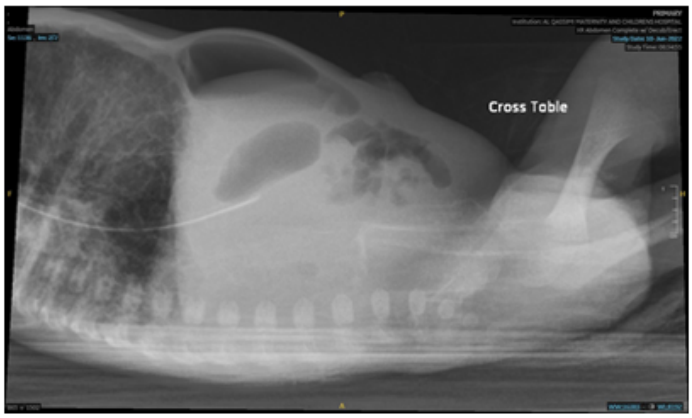

Figure 2

On the review of her chest X ray that was done on the same day of the incident, it was noticed that there was evidence bilateral of pulmonary interstitial emphysema which was more on the left side that later on become more apparent on the repeat chest x ray on 9th and 10th DOL (Figure 3 and 4). Concomitantly with the onset of perforation, she developed neonatal seizure necessitating starting loading and maintenance of phenobarbitone, she had repeat brain ultrasound screen that showed IVH grade IV on the right side.

Her postoperative period was uneventful and after few days she was started on trophic feed that was tolerated and increased gradually to full feeds. She remained on invasive respiratory support on SIMV+ VG+ PS with maintaining of blood gas. Trial to extubate her to noninvasive respiratory support was unsuccessful initially but she was finally extubated to noninvasive respiratory support on the 43rd DOL in the form of NIPPV and then CPAP then HFNC. Her condition was complicated with development of bronchopulmonary dysplasia

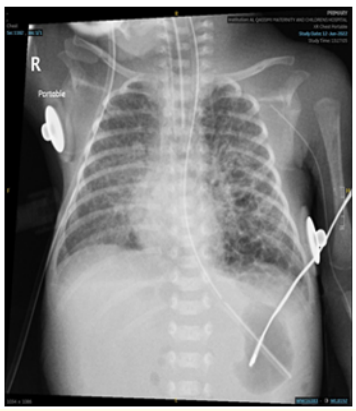

Figure 3

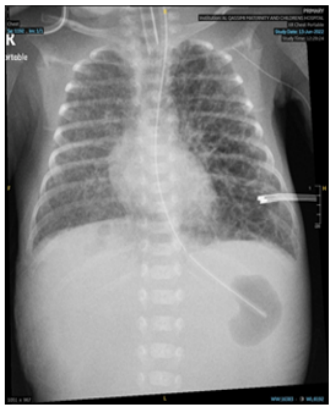

Figure 4

for which she received short and interrupted course of oral diuretics. Follow up repeat brain ultrasound scan showed resolution of her grade IV IVH with no complication related to the IVH and she was gradually weaned of phenobarbitone.

She steadily gained weight and was discharged home on 107 DOL (Corrected age of 1 day) in stable general condition with weight of 1.8 kg for neonatal clinic follow up.

Case Discussion and Conclusion

Pneumoperitoneum denotes the presence of free air in the peritoneal cavity. It can be classified as surgical and nonsurgical (Medical) pneumoperitoneum with the surgical causes account for more than 90% of cases [1].

Surgical pneumoperitoneum is a catastrophic event as it is associated in most cases with intestinal perforation. The perforation is usually caused by necrotising enterocolitis or spontaneous perforation of a viscus in 90% of cases. Also, it could arise from a perforated hollow viscus or a preceding abdominal operation.

On the other hand, pneumoperitoneum can be nonsurgical or medical which can occur in 1% - 3% of mechanically ventilated infants, depending on the ventilation modality [1].

Pneumoperitoneum can be categorized into five major categories: abdominal, gynecological, thoracic, pseudopneumoperitoneum and idiopathic [2].

The rare cases of idiopathic pneumoperitoneum have been described in the literature however its underlying causes and pathophysiology remain uncertain [3].

Leonidas., *et al*. [4] studied 222 mechanically ventilated infants and observed that of the 9 infants who developed pneumoperitoneum, 4 of them were non-surgical.

Zerella., *et al*. [5] distinguished the 'medical' pneumoperitoneum from the 'surgical' pneumoperitoneum in his 10 critically ill infants with respiratory disease and no evidence of an intestinal perforation.

Gupta., *et al*. [6] suggested that the entity of benign pneumoperitoneum should be recognised and laparotomy avoided.

Al-lawama., *et al*. [7] described a case of a neonate who developed major postoperative complications including a grade III intraventricular haemorrhage and a patent ductus arteriosus following an unnecessary laparotomy for a benign idiopathic pneumoperitoneum.

Pulmonary interstitial emphysema (PIE) is one type of the Pulmonary air leak syndromes that occurs when there is dissection of air into the perivascular tissues of the lung from alveolar overdistension or overdistension of the smaller airways. It may be the precursor of all other types of pulmonary air leaks [9].

The pulmonary air leak syndromes compromise pneumomediastinum, pneumothorax, pulmonary interstitial emphysema [PIE], pneumatocele, pneumopericardium, pneumoperitoneum, and pneumoretroperitoneum. Very rarely air can enter pulmonary vessels (pulmonary veins) and cause air embolism. Air can also leak into the subcutaneous layers of the skin, especially skin of the chest, neck, and face, causing subcutaneous emphysema.

The pathogenic mechanism underlying the development of pneumoperitoneum following PIE is that when there is PIE, the air may leak from the ruptured alveoli of non-compliant lungs, tracking downwards along sheaths of vessels in the mediastinum and anatomical defects in the diaphragm, resulting in a collection of free air in the abdomen [9].

Another explanation could be that the air from ruptured alveoli can flow transdiaphragmatically along the great vessels and esophagus into the retroperitoneum. When air accumulates in the retroperitoneum, rupture into the peritoneal cavity can occur [10].

The mechanism of pneumoperitoneum in our case is mostly related to pulmonary interstitial emphysema. the pulmonary air leak syndromes with its types which include pneumomediastinum, pneumothorax, pulmonary interstitial emphysema PIE have been proposed by many authors and can be possible explanation for the development of pneumoperitoneum [5].

There will be still argument that our knowledge about the physiology and anatomy of extremely premature as well as growth-restricted infants remains very limited and further research is required [8].

To our knowledge this is the 1st reported case of pneumoperitoneum 2ry to pulmonary air leak in 24 week extremely preterm neonate found to be associated with pulmonary interstitial emphysema (PIE) with good survival and outcome.

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