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Case Report

Idiopathic pneumoperitoneum without gastrointestinal perforation in a low-birth weight infant: A rare type of air leak syndrome ☆,☆☆

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ABSTRACT

Neonatal free air on X-ray images is generally due to intestinal perforation, and requires surgical intervention. However, some cases without intestinal perforation show free air on X-ray images. Pneumoperitoneum without perforation is caused by an air leak syndrome. We present here the case of a low-birth-weight infant with free air on X-ray images, who had no evidence of intestinal perforation intraoperatively.

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Case report

A female infant weighing 1972 g was born to a healthy mother at 33⁺⁶ weeks of gestational age by emergency caesarean section for decreased fetal movement and fetal pleural effusion. She was intubated and the pleural effusion improved soon thereafter. She was placed on high-frequency oscillatory ven-

tilation (HFOV) with nitric oxide (NO) insufflations and received repeated surfactant insufflations into the tracheal tube to treat respiratory distress syndrome. Nasal tube feeding was initiated on her third day of life. She developed abdominal distention on the 9th day of life, and her X-ray images on the 11th day revealed abdominal free air (Fig. 1). She was transferred

Abbreviations: HFOV, high-frequency oscillatory ventilation; NEC, necrotizing enterocolitis; NO, nitric oxide; RDS, respiratory distress syndrome.

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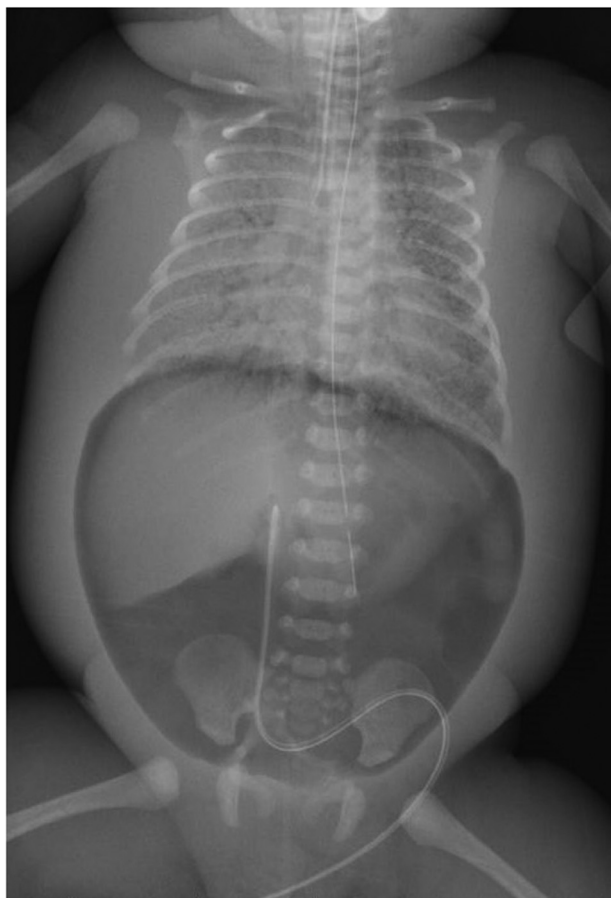


Fig. 1 – Chest X-ray image showing abdominal free air on the 11th day of life.

to the neonatal intensive care unit of our institute for surgical management with a diagnosis of intestinal perforation. On admission, her vital signs showed a heart rate of 80 beats per minute, her SpO₂ was 90%.

She underwent an emergency operation. At laparotomy, a large volume of air came out and the abdomen became flat. We found a small amount of serous peritoneal fluid with no fecal peritonitis evidence. The gastrointestinal tract was in good condition without perforation or pneumatosis. We injected crystal violet solution into the stomach, but found no blue dye leakage from the gastrointestinal tract. Overall, we found nothing wrong, and we completed the operation after inserting an intraperitoneal drain. The bacterial culture of peritoneal fluid was negative, and the postoperative discharge from the drain was serous.

On the 22nd day of life (11th postoperative day), we injected contrast agent with blue dye into her stomach for an X-ray examination. We found no peritoneal extravasation signs. We started the infant on tube feeding on her 23rd day of life (12th postoperative day). The next day, we removed the drain. We found no signs of pneumoperitoneum afterward.

We reviewed the preoperative X-rays again, and found a pneumomediastinum just after surfactant insufflation on the 11th day of life (Fig. 2) that we suspected to have occurred due to an extrapulmonary air leak.

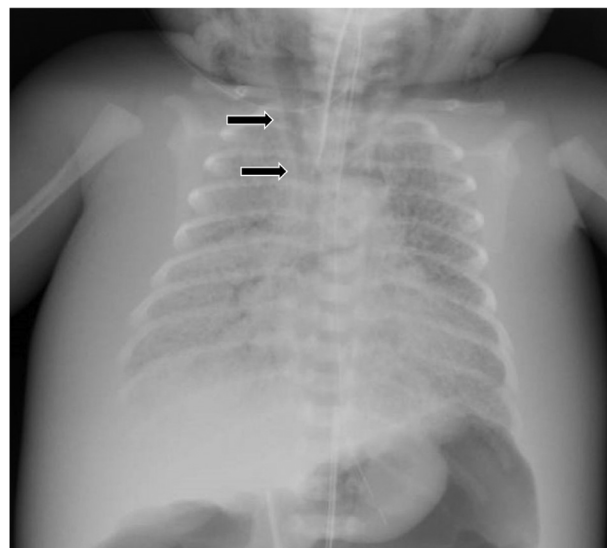


Fig. 2 – Preoperative chest X-ray image showing pneumomediastinum (black arrows) just after surfactant insufflation.

The infant underwent tracheostomy due to chronic lung disease and developed tracheomalacia at the age of 9 months. She died from severe sepsis at the age of 2 years and 11 months.

Discussion

A common cause of pneumoperitoneum in low-birth-weight infants is necrotizing enterocolitis (NEC) [1]. Other causes include isolated gastrointestinal perforations, mechanical ventilation, endoscopic procedures, and pneumatosis cystoides intestinalis [2]. The use of mechanical ventilation (as in nasal continuous positive airway pressure or high-frequency oscillatory ventilation) and exogenous surfactant supplement have improved the general outcome for neonates with severe respiratory failure. However, air leak syndrome is still present in critically ill neonates. Air leak syndrome occurs when air escapes from the tracheobronchial tree and collects in various body spaces where it is not normally present [3]. Types of air leaks include pulmonary interstitial emphysema, pneumothorax, pneumomediastinum, pneumopericardium, pneumoperitoneum, subcutaneous emphysema, and systemic air embolism. X-ray images reveal characteristic findings in each of these clinical conditions. The cases of ventilation-related pneumoperitoneum have increased due to neonatal respiratory support advances. Causes of neonatal pneumoperitoneum include ventilation-related (1.1%) and idiopathic (8%) barotraumas [4]. Other causes include NEC (52.3%), isolated gastrointestinal perforations (20.6%), anorectal malformations (15.1%), meconium ileus (2.3%), and congenital band obstruction (1.1%) [4]. Reported cases include a series of 9 air leak cases including pneumothorax, pneumomediastinum, and pneumoperitoneum [5].

Our case highlights 2 important clinical features: First, surfactant insufflations for infants suffering respiratory distress syndrome may cause pneumomediastinum and pneumoperitoneum. X-ray images revealed abdominal free air on the 11th day of life that suggested a gastrointestinal perforation, but we found no such perforation during the operation. After a later date, we found a pneumomediastinum on the X-ray after a surfactant insufflation. When reviewed in hindsight, we suspect that the surfactant insufflations were the cause of pneumomediastinum with air getting into the peritoneal cavity. We found no similar reports in the literature.

Second, peritoneal discharge can be a sign of a pneumoperitoneum that can be easily misdiagnosed as a gastrointestinal perforation. This is a clinical pitfall, as serious conditions like NEC or gastrointestinal perforations may require rapid laparotomy interventions. Conversely, in the absence of perforation or peritonitis, a laparotomy is an invasive unnecessary intervention. Thus, ruling out severe conditions like the intestinal perforation is important to prevent unnecessary invasive surgeries.

If the “free air” in the abdomen is found radiographically, the general condition of the patient should be checked at first. In the absence of other abnormal findings like unstable vital signs, ascending white blood cell (WBC) or C-reactive protein (CRP) on the blood exam, or intestinal distention on the X-rays, checking the co-existing pneumothorax or pneumomediastinum is important. Examinations such as abdominal puncture or cultivation of its fluid can help in cases with difficult diagnosis to choose a course of treatment [6]. The presence of little fluid with relatively abundant air is likely to be due to an air leak.

In summary, our patient had abdominal distention and the X-ray image revealed free air. However, we found no intraop-

erative evidence of a gastrointestinal perforation during the operation. Retrospectively, the pneumoperitoneum appears to have been an air leak syndrome probably due to intense surfactant insufflation. “Free air” in the abdomen on a radiographic image needs to be carefully considered depending on the presence of other abnormal findings to avoid unnecessary surgical interventions; and intestinal distention alone may be a sign of a co-existing pneumothorax or pneumomediastinum.

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