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Idiopathic neonatal pneumoperitoneum, a case report

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ABSTRACT

INTRODUCTION: There have been non-surgical conditions secondary to neonatal pneumoperitoneum (e.g., mechanical ventilation, pulmonary diseases and pneumatosis cystoides intestinalis) that neonates were able to overcome without the need for abdominal exploration. Idiopathic pneumoperitoneum, although similar to perforation of the alimentary tract and the previously mentioned non-surgical conditions, is a rare and benign condition that does not yet have a definite cause. The criteria by which a surgeon decides on which abdomen to open and which one to observe, is ill-defined. Thus, increasing the awareness of neonatologists and surgeons about this condition will help decrease complications due to unnecessary procedures.

PRESENTATION OF THE CASE: We report a case of a neonate with a massive pneumoperitoneum who obtained a surgical intervention with negative finding. We had noted that the bag of the abdominal drain was expanded with gas every 2 days, we replace the bag by new one every 2 days for 10 days postoperative. Nonetheless, the cause of pneumoperitoneum remains unclear.

DISCUSSION: There are five main nonsurgical causes of free air in the peritoneal cavity. These are categorized as follow: pseudopneumoperitoneum, thoracic, abdominal, gynecological and idiopathic. This is a condition in which imaging shows free air in the peritoneal cavity that can either be managed with observation and supportive care alone or results in a negative laparotomy.

CONCLUSION: This case demonstrated that laparotomy is not a true routine in neonates with idiopathic pneumoperitoneum if a timely diagnosis is established. Future research is still necessary to understand the source of the free gas in the abdomen, as well as the underlying causes of delayed postoperative gas underdiaphragm and postoperative abdominal drain bag gas expansion.

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1. Introduction

The term pneumoperitoneum is used to describe the presence of free gas within the peritoneal cavity. In 90% of cases, it is the result of hollow viscus perforation [1]. NP is categorized into two types: surgical and nonsurgical pneumoperitoneum. Idiopathic pneumoperitoneum is classified as nonsurgical and usually has a more favorable prognosis. However, patients with idiopathic pneumoperitoneum who underwent surgery often had negative outcomes. The ability to differentiate between idiopathic pneumoperitoneum from a highly lethal perforation of air containing viscus may reduce surgical intervention and increase the survival rate in neonatal patients [2]. Conservative management, avoiding a laparotomy is the treatment of choice. Future research is still necessary to understand the source of the free gas in the abdomen, as well as the underlying mechanism of pneumoperitoneum.

Herein, we describe a rare case of NP without an established cause; the case obtained a laparotomy with negative finding at Aswan university hospital. Also, we had noted that the bag of the abdominal drain was expanded with gas every 2 days, we had been replacing the bag by new one every 2 days for 10 days postoperative, Future research is still necessary to understand the source of the free gas in the abdominal drain bag. Our case is reported in line with the SCARE criteria [29].

2. Case report

We report a case of premature female infant from Upper Egypt born after 34th weeks of gestation. The baby delivered vaginally from a gravid 2, para2 +0 mother (a women who had been pregnant a total of two times and carried those pregnancies to the age of viability), the baby weighted 1750 g; blood group O negative. The infant admitted to neonatal care unit at west Aswan hospital due to hydrops fetalis and biphasic jaundice. She had an exchange transfusion twice, no oral feeding started, but the infant passed meconium spontaneously at the 2nd postpartum days. Plain erect X ray of the abdomen revealed gas underdiaphragm at the 5th postpartum

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Fig. 1. preoperative gas underdiaphragm.



Fig. 2. gas underdiaphragm at 7th postoperative days.

days, no abdominal ultrasound was done. During this period, the infant was on vancomycin and 3rd generation cephalosporin.

The infant referred to Aswan university hospital at the age of 12 days, presented with abdominal distention, jaundice, septicemia, respiratory distress, pneumonia, gas underdiaphragm, and diagnosed as perforated viscus.

The infant was tachypnic on nasal oxygenation therapy 2 litters, feverish (low grade), white blood cell count of $25,000/\text{mm}^3$, bilirubin total 24.2 mg/dl, direct 16.2 mg/dl, indirect 8 mg/dl. Gastric decompression by mean of nasogastric tube performed.

Abdominal exploration did by experience pediatric surgeon, revealed no pathological finding other than pyogenic membranes at upper abdominal region, stomach, duodenum, colon and small intestine showed any abnormality. We performed abdominal washing with saline; finely pelvic abdominal drain was held by stay suture.

We had noted that the bag of the abdominal drain was expanded with gas every 2 days, we had been replacing the bag by new one every 2 days for 10 days postoperative. No, other than gas, collections in the bag except for a trace of serosanguinous secretion. We did not measure the amount of air at the bag, but most of the size of the bag was expanded with air. We had taken a swab from the abdominal drain at the 10th postoperative days revealed a gram –ve bacilli sensitive to cefepime. No blood culture results were available.

There was difficult transfer of the infant to radiological department for follow up abdominal ultrasound, but, follow up plain erect abdomen x ray at the 5th and 7th postoperative days show gas underdiaphragm.

The patient sustained regular decrease in white blood cell count from $25,000/\text{mm}^3$ to $16,000/\text{mm}^3$ at the 12 postoperative days. The infant was on vancomycin, 3rd generation cephalosporin, carbapenem antibiotic injection started from the time of operative intervention. When the abdominal drain bag was stopped insufflations, the abdominal drain was removed in the 12 postoperative days.

The infant was not passing until the 13th postoperative days, where PR examination was forced the infant to pass thick meconium.



Fig. 3. frequent abdominal drain bag gas expansion.

and no gastrografin enema or followthrough results were available due to poor resources of our hospital.

The infant dies at the 16th postoperative days from respiratory failure, which developed due to pneumonia (Figs. 1–3).

3. Discussion

From reviewing the literature of related subject we have found that, Diesen et al. [3] reported a case of neonatal intestinal perforation caused by necrotizing enterocolitis (NEC) which was spontaneously sealed by omentum, suggesting the possibility that NEC patients are able to heal without laparotomy. Pneumatosis cystoides intestinalis (PCI) is seen in preterm neonates with

NEC [4,5]. NEC is characterized by abdominal distension, gastrointestinal bleeding, abdominal tenderness (even sepsis and shock at advanced stage) and the presence of PCI on abdominal X-ray film [5,6] as tiny free gas within the intestinal wall (linear sign).

In other instances of nonsurgical pneumoperitoneum have been reported, such as pneumoperitoneum secondary to mechanical ventilation and tension pneumothorax [7]. More recently, the relationship between pneumoperitoneum and respiratory diseases has been widely debated [8–10].

Other communication pathways between the chest and abdomen include the periaortic and periesophageal space and congenital defect [11,12] or pleuroperitoneal fistula [13] that permits the air to pass through.

Tao-Zhen He et al. [2] hypothesize that gastric tissue ischemia, secondary to hypoxia, will be results in perforation (range: 2 mm to 4 mm in diameter) at the anterior wall of the stomach is responsible for the etiology of spontaneous neonatal pneumoperitoneum.

As the baby cries (aerophagia), the air in the stomach accumulates until it can enter the intraperitoneal cavity through the leak compressed by gastric peristalsis, hence forming a large pneumoperitoneum. Considering newborns spend most of their time in a supine position, the gastric juices at the bottom of the stomach have a low chance of entering the peritoneal cavity from the anterior wall of the stomach during gastric peristalsis; therefore, no signs or symptoms of peritonitis occur. The gastric leak self-seals, preventing further passage of the air, allowing the intraperitoneal free gas to dissipate gradually.

In our case abdominal exploration revealed no pathological finding other than pyogenic membranes at upper abdominal region. The infant was preterm, feverish, tachypnic, accompanied by sepsis, high white blood cell count, pneumonia which donates that the hypothesis of He TZ et al. may be the cause of idiopathic pneumoperitoneum due to sealed gastric perforation.

Free air after a major abdominal procedure poses a clinical dilemma. Is the retained free air from the operation or does this indicate a perforated viscous or possible postoperative complication? Clearly, these have different forms of management; the former one is observation while the latter requires re-exploration. It is well established that pneumoperitoneum with fever, abdominal tenderness or distension, leukocytosis or signs of peritoneal irritation is a surgical emergency [7,14,26].

Only 5% to 15% of the time, the cause of free air is something other than a perforation and does not require surgery [13,15,16]. This is called “nonsurgical [17,18,26]”, “spontaneous [14,19–22]” or “misleading [23]” pneumoperitoneum. This is a condition in which imaging shows free air in the peritoneal cavity that can either be managed with observation and supportive care alone or results in a negative laparotomy [21,26].

There are five main nonsurgical causes of free air in the peritoneal cavity. These are categorized as follows: pseudopneumoperitoneum, thoracic, abdominal, gynecological and idiopathic [5,10,24–28]. Table 1 lists the pathophysiological mechanisms for causes of nonsurgical pneumoperitoneum [14,25–28].

In our case the cause of delayed postoperative pneumoperitoneum may be due to the presence of abdominal drain up to the 12 postoperative days, or may be due to pneumonia infection which cause basal pulmonary atelectasis give the picture of pseudopneumoperitoneum, or due to intra abdominal sepsis by gas forming organisms as Escherichiacoli, Clostridium, Staphylococcus, Streptococcus, Klebsiella, Candida and Pseudomonas. A swab taken from the abdominal drain on the 10th postoperative days revealed a gram –ve bacilli sensitive to cefprofloxacin. No blood culture results were available.

Table 1
pathophysiological mechanism of some of the causes of nonsurgical pneumoperitoneum.

Non surgical causes	Pathophysiological mechanism
Pseudopneumoperitonium	Adventitial air shadows Overdistension of hollow viscera Undulant configuration of the diaphragm Gas trapped in established wounds Basal pulmonary atelectasis Subdiaphragmatic extraperitoneal fat Interposition of the hepatic flexure of colon between right lobe of liver and diaphragm Interposition of the hepatic flexure of colon between right lobe of liver and diaphragm
Thoracic	Mechanical ventilation High airway pressures Large tidal volumes Noncompliant lungs Preexistent pulmonary disease Cardiopulmonary resuscitation Pneumothorax Pneumomediastinum Rapid decompression (diving accidents) Tracheal rupture Median sternotomy Blast injury
Abdominal	Postoperative retained air after abdominal surgery Peritoneal dialysis Percutaneous endoscopic gastrostomy Endoscopic procedures Pneumatosis cystoides intestinalis Blunt abdominal trauma
Gynecological	Vaginal insufflations Pelvic inflammatory disease Post partum knee chest exercises Coitus Gynecological exams Vaginal douching
Idiopathic	

The cause of frequent expansion of the abdominal drain bag in our case is unknown. Further researches are required to identify possible etiologic factors implicated in such cases.

4. Conclusion

Laparotomy is not a true routine in neonates with idiopathic pneumoperitoneum if a timely diagnosis is established. Thus, increasing the awareness of neonatologists and surgeons about this condition will help decrease complications due to unnecessary procedures.

Future research is still necessary to understand the source of the free gas in the abdomen, as well as the underlying causes of delayed postoperative gas underdiaphragm and postoperative abdominal drain bag gas expansion.

Conflict of interests

No conflict of interest.

Sources of funding

No funding has been used for this research.

Ethical approval

No ethical approval has been applied for this case report study, only the written and oral consent by the relative of the patient.

Consent

A written consent has been obtained from the patient relative for operative intervention and for the publication of this case report and accompanying images and is available for review on request.

Author contributions

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