Case report

NEONATAL APPENDICITIS: AN EXPERIENCE WITH 2 CASES AT HUE CENTRAL HOSPITAL

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ABSTRACT

Background: *Acute appendicitis is extremely rare in the neonatal periods. The number of cases published in the last century is just over 100. Mortality and morbidity are still high due to diagnostic problems because there are no specific clinical features and reliable investigation for the diagnosis. Herein, we present two patients to remind physicians that the diagnosis of neonatal appendicitis should always be kept in mind.*

Case presentations: *The first case presented at Neonatal Intensive Care Unit with an 18-hour-history of irritability, vomiting, and abdominal distension. With high white blood cell count and C-reactive protein level, the baby was initially diagnosed with neonatal sepsis. The ultrasound performed on day 3 after hospitalization found peritonitis but not seen appendicitis. Post-operative diagnosis confirmed acute perforated appendicitis withperitonitis. The baby was well covered without complication. The second case was a 6-year-old full-term infant with Down syndrome and Pulmonary atresia with ventricular septal defect. The patient underwent surgery and postoperative critical care. However, he died at post-operative day 5 due to worsening sepsis and decompensated hemodynamic instability.*

Conclusion: *It is a fact that acute appendicitis in neonates and infants may not be diagnosed easily and quickly as in older children because there are no specific clinical features and reliable investigation for the diagnosis. Delay in diagnosis and treatment often results in appendicular perforation and peritonitis. The main safeguard against mortality and morbidity remains a high index of suspicion.*

I. INTRODUCTION

Acute appendicitis is the most frequent cause of acute surgical abdominal emergency in children and adults, but extremely uncommon in newborn infants [1] with the incidence between 0.04% and 0.2% [2]. Although neonatal appendicitis has been reported for over a century, the total cumulative cases published are approximately 100, of which most cases presented with local or general peritonitis [3].

Diagnosis and management of neonatal appendicitis is challenging due to the rarity together with lack of specific clinical features and low index of suspicion, which may lead to delay in diagnosis and surgical intervention [4]. The diagnosis is

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usually made when perforation has occurred and confirmed intraoperatively. These may result in a high risk of complications such as perforation, thus leading to peritonitis may be life-threatening in neonates [2,5] with the high mortality rate as 34% [6]. Herein, we presented two patients to remind that the diagnosis of neonatal appendicitis should always be kept in mind.

II. CASE PRESENTATION

Case 1

This baby was born full-term via cesarean section delivery without perinatal complications. He presented in the Neonatal Intensive Care Unit of Hue Central Hospital on May 8, 2020 with an 18-hour-history of irritability, vomiting, and abdominal distension when he was 12 days old. These conditions had resulted in decreased breastfeeding, prompting his parents to bring him to our center. Upon arrival, the patient was febrile to 38.3°C, tachycardia (145bpm), and normal breathing (40 bpm). Physical examination demonstrated a distended abdomen. The sign of tenderness were hardly seen. White blood cell count was 16.6 K/μL (neutrophil 37.9%). C-reactive protein and procalcitonin level was high (63.8 mg/L and 2.56 ng/ml, respectively). Serum lactate, liver enzyme, renal function tests, and electrolytes were within normal ranges.

In the suspect of neonatal sepsis, antibiotic and supporting therapy were started, and an abdominal ultrasound was done, but no ecographic typical signs were found.

Three days later, general clinical as well as abdominal condition was not improved. Therefore, we performed an abdominal ultrasound again and found a mixed-echo mass (19 x 18 mm) with gas and fluid inside. The inflammation phenomenon was seen around the mass. The ultrasound findings demonstrated peritonitis but did not confirm any abnormalities concerning the appendix. This was considered the reason for the acute surgical abdomen, and a standard open surgery was immediately started. Surgical findings showed turbid fluid in the peritoneal cavity and gangrenous appendicitis with a perforation at the tip **(Figure 1)**.

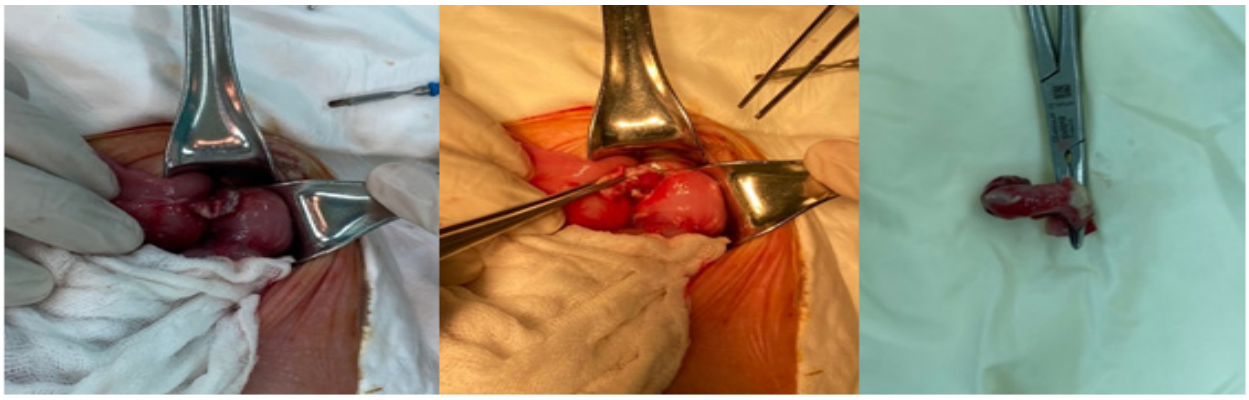

Figure 1: The acute perforated appendicitis was confirmed intraoperatively with peritonitis.

Postoperative management was regular with antibiotic and supporting therapy. On postoperative day 3, he was starting to have breastfed with the return of bowel function. Blood tests switched back to normal limits. Peritoneal fluid culture from the day of operation returned positive for Escherichia coli, which was sensitive to antibiotics.

The patient continued to cover without any complication, and no additional studies were necessary. He was discharged on postoperative day 7.

Case 2

A 3000-g boy was born by Cesarean section at 38 4/7 weeks of gestation to a 35-year-old mother (PARA 2022). The patient was diagnosed by prenatal ultrasound at five months of gestation with Down syndrome, pulmonary atresia with ventricular septal defect. The mother had no antenatal history of infection or other medical or surgical illnesses. The patient appeared icteric and cyanotic at birth with SpO2 of 78% on room air. Continuous intravenous

prostaglandin at 10 mcg/kg/min was used to maintain the ductus arteriosus patency. He was transferred to the neonatal intensive care unit. He was passing meconium normally. Initial blood test showed normal complete blood count, unconjugated hyperbilirubinemia in accordance with physiologic jaundice, mildly elevated C-reactive protein of 10.0 mg/L, normal renal function test and mild electrolyte disturbance.

On the 5th day, he developed abdominal distention and refused to feed. On examination, he developed a fever of 38.50C. There was no tachypnea. The oxygen saturation was at 60-70% on 3 liters of oxygen. The lung was clear to auscultation. The abdomen was distended without obvious tenderness or erythema. Bowel sounds were hypoactive. Abdominal Xrays and ultrasound were performed and revealed no pneumoperitoneum or free fluid, no intestinal pneumatosis or portal venous gas **(Figure 2)**. His blood test showed a normal leukocyte count of 4.21 K/µL. A diagnosis of necrotizing enterocolitis (NEC), Bell stage IA was made and he was put on empiric intravenous antibiotics with ceftriaxone, amikacin and metronidazole. He was kept nil by mouth and a nasogastric tube was inserted with intermittent suction.

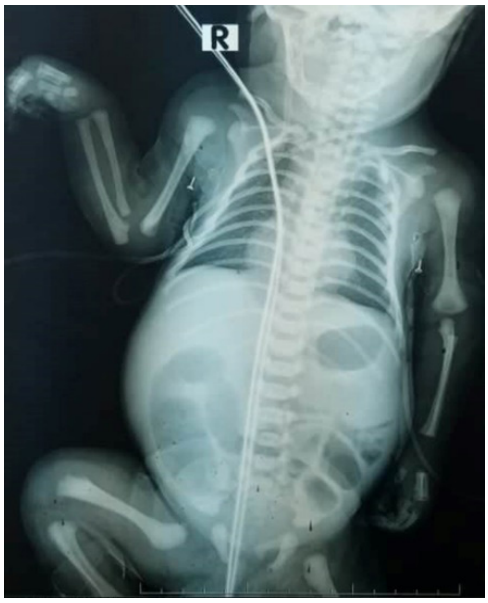

Figure 2: Plain radiograph showed dilated bowel loop, no pneumatosis intestinalis, free gas or portal venous gas.

On the 6th day, the abdominal distention increased. There was generalized tenderness and erythema of the abdomen on examination (Figure 3). No blood was found on per rectal examination. A repeat abdominal ultrasound was made and revealed pneumoperitoneum and moderate ascites. The bowel loops were distended up to 28 mm. No portal venous gas or pneumatosis intestinalis was detected. A repeat blood test showed a leukocytosis of 11.78 K/µL with 68.1% neutrophils. The C-reactive protein was markedly elevated at 224.3 mg/L. A diagnosis of peritonitis due to perforated hollow viscus was made and he was rapidly transferred for exploratory laparotomy.

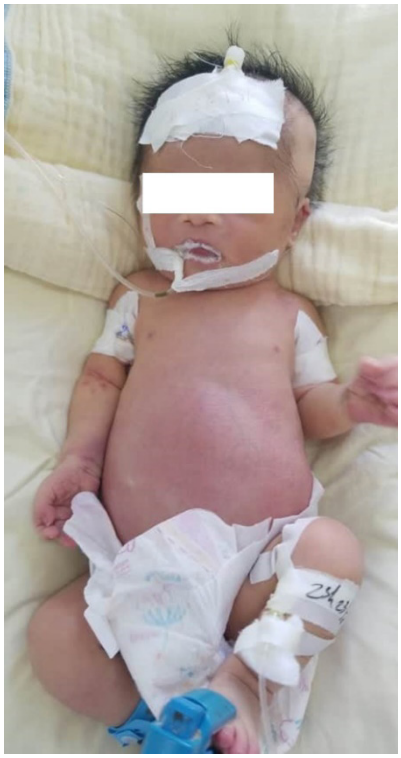

Figure 3: The abdomen was distended and abdominal wall erythema was present

A midline incision was made. The entire bowel loops (small intestine, colon) were distended. The peritoneal cavity was filled with turbid and yellowish fluid at all four quadrants. Fibrinous exudate was distributed mainly at the right iliac fossa around the cecum **(Figure 4)**. The appendix was inflamed and perforated at its middle portion. Other organs in the

abdomen were carefully examined and no further lesions could be found. Vicryl 2.0 tie was used to secure the base and the appendix was transected by scalpel. The peritoneal cavity was irrigated with a large amount of warm normal saline and peritoneal fluid was sent for culture. Peritoneal drainage was placed at the Douglas pouch. The abdomen was closed by interrupted suture.

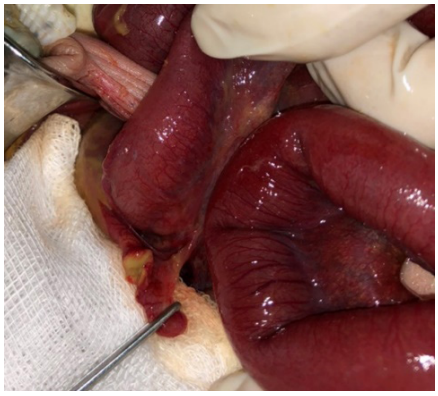

Figure 4: Intraoperative image of perforated appendicitis at the middle third of the appendix. Adjacent yellowish fluid with fibrinous exudate can be seen

Pathologic analysis of the appendix demonstrated a grossly inflamed appendixmeasured 4mm in diameter and 2.5cm in length. Microscopic analysis showed marked infiltration of inflammatory cells including degenerated neutrophils and lymphocytes. The appendiceal mucosa was completely necrotic **(Figure 5)**.

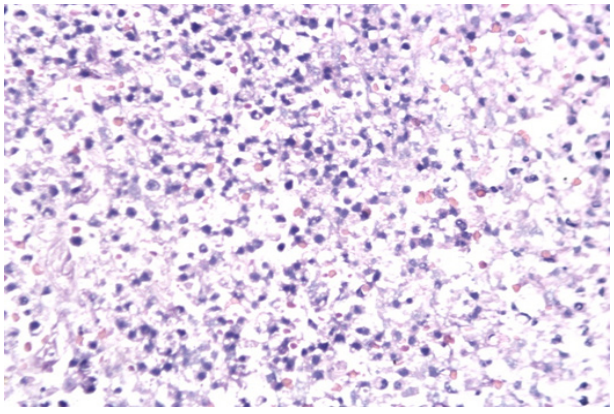

Figure 5: Histopathologic examination revealed infiltration of neutrophils and widespread necrosis of the appendix (HE x 400)

The patient was transferred to the surgical intensive care unit. Empiric broad-spectrum antibiotic therapy was continued using meropenem and amikacin. However, the patient was died at postoperative day 5 due to worsening sepsis and decompensated hemodynamic instability.

III. DISCUSSION

Despite the frequency of acute appendicitis in children and adults, only 0.04-0.2% of cases are reported in neonates [2]. These patients are approximately three times as likely to be perforated compared to adult patients at the time of diagnosis [5]. About 85% of the newborn infants had perforated appendicitis at laparotomy [1]. Due to clinical features are nonspecific, preoperative diagnosis is challenging, and most of the neonatal appendicitis have been diagnosed intraoperatively. Raveenthiran reviewed 52 neonatal cases with abdominal manifestation and found that the definitive diagnosis of appendicitis was made clinically in three cases and at autopsy in two neonates. All other cases were retrospectively diagnosed after operation [1]. This was similar to our case that was initially diagnosed of neonatal sepsis, and the definitive diagnosis was made intraoperatively about 42 hours after hospitalization.

The fact that all the pediatricians and surgeons emphasize the forgotten diagnosis due to both the nonspecific presentation and the extreme rarity of acute appendicitis at this age [2,7]. Generally, the patients may present irritability, wriggling, distressed breathing, swelling of the scrotum, abdominal distension, a right lower quadrant palpable mass, bilious vomiting, and also fever, anorexia, and leukocytosis [8]. According to Raveenthiran, in newborn infants, abdominal distension (accounted for 89%), vomiting (seen in 54%), abdominal tenderness (found in 48%), restlessness or lethargy (counted for 36%) and fever (about 31%) were the most common signs and

symptoms [1]. Flat abdominal X-ray alone is usually not helpful if pneumoperitoneum is not present [7]. Despite the diagnosis of acute appendicitis, further studies must be performed to rule out other diseases such as cystic fibrosis, Hirschsprung's disease and necrotizing enterocolitis [9].

The low incidence of appendicitis in neonates can be explained by multiple factors such as wider based appendix in infants, which may lead to a decreased risk of luminal obstruction [10]. Newborn infants have breastfed or intake soft consistency foods. They also spend in the recumbent position for days. And finally, newborn infants often have less common infections, which may result in a very low inflammation rate of peri-appendiceal lymphoid tissue [3].

To our acknowledgment, this is the first-two case of neonatal appendicitis in our hospital, and no similar cases were reported in Vietnam. At the time of hospitalization, we suspected of neonatal sepsis due to the clinical features of fever, vomiting, abdominal distension, and the elevation of white blood cells, C-reactive protein, procalcitonin. Besides, the first abdominal ultrasound did not find any echographic abnormalities concerning the acute abdomen. Even at the late stage, when the ultrasound was considering peritonitis, but appendicitis was not mentioned preoperatively.

Acute appendicitis with perforation in neonates and infants has been correlated with increased morbidity and mortality. The mortality of neonatal appendicitis was high (around 78%) before 1975. It then declined to 33% between 1976 and 1984 due to the advances in diagnostic modalities, neonatal intensive care, and antibiotic therapy. Further down in mortality to 28% between 1985 and 2003 was just a modest development [1]. This is mostly because of the delay in diagnosis [3,10].

Laparoscopy is more and more accepted for appendectomy in both the infant and neonatal populations. However, in our cases, the preoperative diagnosis was peritonitis and did not confirm acute appendicitis. Thus, we indicated an open surgery to fully explore the acute abdominal causes. Post-operative events in our cases were similar to neonatal patients who underwent a laparoscopic appendectomy reported in other studies [1,11,12]. Our first patient was well covered without any complication and discharged on postoperative day 7, however, the second case died at postoperative day 5 due to worsening sepsis and decompensated hemodynamic instability.

There were some limitations in our cases concerning diagnosis. We did not perform an abdominal computed tomography scan preoperatively when the ultrasound answered a negative result. It could be helpful for a timely diagnosis. Postoperatively, the surgeon did not take a histology examination to confirm the diagnosis of acute perforated appendicitis.

IV. CONCLUSION

It is a fact that acute appendicitis in newborns is not diagnosed easily and quickly as in older children because there are no specific clinical features and reliable investigation for the diagnosis. Delay in diagnosis and treatment often results in appendicular perforation and peritonitis. The main safeguard against mortality and morbidity remains a high index of suspicion.

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