Preterm with appendicular abscess and subcapsular liver haematoma: an unreported association

Mohamed S. Shalaby[a,b] and Gregor Walker[a]

Neonatal appendicitis is a rare yet serious condition. Known reported associations are Hirschsprung's disease, necrotising enterocolitis (NEC) and cystic fibrosis. The occurrence of subcapsular liver haematoma in preterm neonates can be life threatening. We present an unreported association of these two serious conditions in a preterm infant with appendicular abscess and subcapsular liver haematoma. She presented with sepsis, enlarged liver and tender right iliac fossa mass. Laparotomy was performed for possible complicated NEC. Intraoperatively, there was an appendicular abscess with a small subcapsular liver haematoma and no evidence of NEC. Appendectomy and peritoneal toilet were performed. She had a stormy postoperative period due to sepsis, thrombocytopaenia and expansion of her liver haematoma. The pathology of her appendix was in keeping with appendicitis, with no evidence of NEC. Her liver haematoma gradually resolved. Suction rectal biopsy showed normal ganglia. She was discharged after 7 weeks and remained well since then. ***Ann Pediatr Surg*** **10:95–96 © 2014 Annals of Pediatric Surgery.**

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[a]Department of Paediatric Surgery, Royal Hospital for Sick Children, Glasgow, United Kingdom and [b]Department of Paediatric Surgery, Ain Shams University, Cairo, Egypt

Correspondence to Mohamed S. Shalaby, FRCS Paed Surg, FEBPS, Department of Paediatric Surgery, Royal Hospital for Sick Children, Glasgow, G3 8SJ, United Kingdom
Tel: +44 797 384 6651; fax: +44 141 232 1977;
e-mail: mshalaby@doctors.org.uk

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Introduction

Neonatal appendicitis is a rare yet serious condition [1]. Known reported associations are Hirschsprung's disease (HSD), necrotising enterocolitis (NEC) and cystic fibrosis [2]. The occurrence of subcapsular liver haematoma in preterm neonates can be life threatening [3]. We present an unreported association of these two serious conditions in a preterm infant with appendicular abscess and subcapsular liver haematoma.

Case report

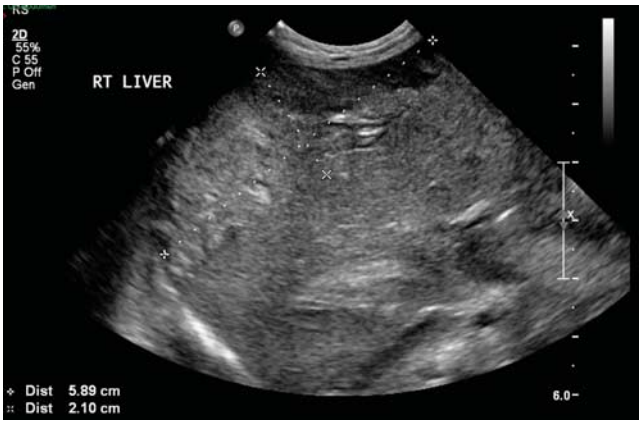
A 1.4-kg preterm baby girl was born by spontaneous vaginal delivery at 30 weeks gestation, with normal antenatal history. She required brief respiratory support for the first 2 days of life then was self-ventilating in air. On 12 days of age, she had an episode of sepsis with coagulopathy that was initially treated with broad-spectrum antibiotics, but she subsequently developed abdominal distension with a palpable right iliac fossa mass. Plain radiographs showed no pneumatosis but dilated thickened bowel loops displaced with a mildly enlarged liver. Following fluid resuscitation and correction of the coagulopathy, her condition deteriorated with abdominal wall erythema and tenderness; hence, an emergency laparotomy was performed for suspected complicated NEC. The intraoperative findings were a perforated appendix with an appendicular abscess but no evidence of pneumatosis in the adjacent bowel. Appendectomy and peritoneal lavage were performed. A small subcapsular liver haematoma was noted intraoperatively.

Four hours postoperatively, she developed acute cardiorespiratory instability, and haematological investigations showed anaemia and thrombocytopaenia. There was marked abdominal distension but before considering re-exploration, an urgent ultrasound scan was performed, demonstrating rapid expansion of the subcapsular liver haematoma (Fig. 1).

Repeat blood product transfusions were required to correct the coagulopathy, and her clinical status improved after 72 h with no further increase in the size of the subcapsular haematoma. Over the next 4 weeks, the haematoma gradually involuted, although cholestatic jaundice developed, presumably secondary to prolonged parenteral nutrition and liver haematoma.

The pathology of the appendectomy showed an acutely necrotic appendix in keeping with appendicitis, with no evidence of NEC. She was discharged after 7 weeks after a suction rectal biopsy excluded HSD. At last follow-up (14 weeks corrected age), she was thriving, with normalization of liver enzymes and ultrasonographic resolution of the liver haematoma.

Fig. 1



Postoperative ultrasonograph showing large subcapsular liver haematoma compressing the right lobe of the liver.

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Ethical adherence: no ethics approval required; however, consent from parent has been obtained for publication.

Discussion

Neonatal appendicitis is a rare yet serious condition. Over a century, there has been a total of 141 reported cases [4]. Male-to-female ratio is 3 : 1; 52% are preterm babies and 74% had appendicular perforation [4]. Diagnosis is usually made intraoperatively or at autopsy [4].

Appendicitis in neonate is thought to be rare because of the funnel-shaped wide base of the appendix, together with the liquid diet and the recumbent position for neonates [5]. This with the nonspecific signs at presentations and the more common causes of neonatal sepsis causes the frequent delay in management, and hence the increase in mortality. In our case, both the intraoperative findings and the histopathology were in keeping with appendicitis, with no signs suggestive of NEC.

Jancelewicz *et al*. [2] classified the reported associations into three main groups: (i) conditions associated with impaired immunity or systemic infection such as prematurity and NEC; (ii) conditions associated with vascular insufficiency or hypoxia such as cardiopulmonary failure and hyaline membrane disease and (iii) conditions associated with intestinal obstruction such as HSD, inguinal hernia and cystic fibrosis. There is no published report describing the association of appendicular abscess in preterm patient with liver haematoma.

The incidence of subcapsular liver haematoma of autopsy in infants varies in the literature between 1.2 and 15%, with 10% incidence of haemoperitoneum in those infants [3]. Risk factors include trauma following delivery, cardiopulmonary resuscitation, umbilical venous catheterization, thrombocytopaenia and coagulation disorders [3]. The pathogenesis of subcapsular liver haematoma in sepsis is probably related to disseminated intravascular coagulopathy and shock [3]. Emma *et al*. [3] concluded that the development of subcapsular liver haematoma in very low birth weight infants is multifactorial due to their 'fragile' nature [3]. The diagnosis is often missed or delayed, and a high clinical suspicion is necessary for early identification and stabilization of affected babies [6]. Our case was complicated by the presence of intra-abdominal sepsis with deterioration requiring emergency laparotomy.

Acute appendicitis in preterm babies has always been considered as a life-threatening disease, necessitating operative management [1,2]. As the patient had an enlarged liver clinically and radiographically, particular care was taken to avoid direct manipulation of the small subcapsular haematoma intraoperatively. We believe that the haematoma was secondary to the preoperative sepsis and coagulopathy, but rapid expansion during the early postoperative period was heralded by a decrease in haemoglobin and deteriorating coagulopathy. This was managed by paralysis and prolonged period of intensive cardiorespiratory support until the haematoma started to show signs of liquefaction and regression on serial ultrasonographies.

In our case, there were clinical signs of intra-abdominal sepsis with right iliac fossa tenderness, mass and erythema, which prompted urgent laparotomy. An ultrasound scan would have confirmed the diagnosis of an abscess in the right iliac fossa and alerted us to the presence of the subcapsular liver haematoma preoperatively, but it would not have avoided the need for a laparotomy. There are no published reports of successful nonoperative management of perforated appendicitis in the neonate. Other case reports detail clinical deterioration with attempted conservative management requiring laparotomy, and, in our case, this would have resulted in performing a laparotomy on an unstable neonate with a larger subcapsular haematoma.

Schwartz *et al*. [1] recommended an algorithm to avoid delay in operating on babies with appendicitis based on ultrasonography and/or computed tomography. They advocate that, if there are no definite signs of NEC on both or any of them, and the baby is unwell with abdominal distension and sepsis, then laparotomy or laparoscopy should be performed to rule out appendicitis. Although we agree that ultrasonography can be particularly helpful in patients with unexplained abdominal sepsis, we do not agree with the suggested role of computed tomography. We advocate that, in neonates with localized right iliac fossa signs, appendicitis should be considered, ensuring timely operative intervention.

Conclusion

In neonates with localized right iliac fossa signs, appendicitis should be considered. No operative intervention is needed apart from appendectomy and peritoneal toilet.

Ultrasonography may be a useful adjunct and may highlight the presence of other associations such as liver haematoma. Gentle intraoperative handling during neonatal laparotomies especially in preterms is important to avoid iatrogenic liver injury.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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