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Neonatal acute appendicitis: a proposed algorithm for timely diagnosis

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Abstract

Background: Neonatal appendicitis (NA) is a rare disease with a high mortality. The diagnosis has never been reported preoperatively and is notoriously difficult to make.

Methods: Charts since 1995 were retrospectively reviewed for discharge or death diagnoses of appendicitis in neonates younger than 28 days. We report 3 cases of NA seen at our institution during this period.

Results: All 3 infants were previously well, born at term, and presented with signs consistent with abdominal sepsis. The first 2 diagnoses were not made until autopsy. The third case survived after having an urgent computed tomographic scan, exploratory laparotomy, and appendectomy.

Discussion: The literature summarizing common presenting features of NA is reviewed. We present an algorithm to guide the workup of these neonates to facilitate earlier diagnosis and potentially improve outcomes.

Neonatal appendicitis (NA) is a very rare condition with a high mortality rate. There has been a significant reduction in mortality over the last century. However, mortality remains as high as 28% [1]. Neonatal appendicitis remains a difficult diagnostic challenge with no reported cases of preoperative diagnosis. It is usually associated with prematurity or comorbidities: primarily inguinal hernias, Hirschsprung disease, cystic fibrosis, cardiac defects, and tracheoesophageal fistula [2]. Here, we report 3 cases from Alberta Children's Hospital, Calgary, Canada, during the last 15 years.

1. Methods

Given the rarity of this condition, a recent death from NA prompted a morbidity and mortality review, which led to a change in our policy regarding neonates with unexplained abdominal sepsis. With this revised policy in place, an additional case was seen, with a favorable outcome; this prompted the present review of the hospital medical records. With ethics board approval, we retrospectively reviewed the medical records from 1995 with a discharge or autopsy diagnosis of appendicitis in patients younger than 28 days. Demographic information and details of the patient's symptoms, diagnostic, treatment, and outcome were collected.

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2. Results

The cases are summarized in Table 1. All 3 cases demonstrated rapid onset of symptoms and deterioration. None of the cases had bilious emesis or hematochezia. Case 1 was seen several times in a peripheral emergency department and discharged before presenting in septic shock and transferred to Alberta Children's Hospital. On presentation, she was severely acidotic with presumed abdominal catastrophe, and despite resuscitative efforts, she died within 3 hours of transfer.

Case 2 was admitted to the intensive care unit and had an initial ultrasound that showed a normal appendix base, no signs of local inflammation, and diffuse ascites. The fluid was tapped and seen to be clear with a high protein and cell count. The diagnosis was thought to be primary peritonitis, and she was treated with antibiotics. As the child deteriorated, surgery was planned, but she had a cardiac arrest and was unable to be resuscitated. At autopsy, the tip of the appendix was perforated, and the base, as suggested on ultrasound, was essentially normal. Based on this, we then instituted a policy that unexplained sepsis, with a clinical suggestion of an abdominal source, which would be investigated by laparotomy/laparoscopy or computed tomographic (CT) scan.

Case 3 presented with similar findings of sepsis and worsening distension that did not fit with the typical clinical presentation of necrotizing enterocolitis (NEC), nor was there an improvement with intravenous antibiotics and supportive care. Accordingly, a CT scan was done, which was verbally reported as normal with no evidence of NEC. This patient was immediately taken to the operating room, which revealed a grossly perforated appendicitis with no evidence of other bowel pathology (Fig. 1).

Fig. 1 Intraoperative photograph of appendix for case 3.

Table 1 Summary data of NA cases

Case	1	2	3
Year of presentation	1995	2009	2010
Age (d)	9	13	15
Sex	Female	Female	Male
Gestational age at birth (weeks)	42	40	40
Comorbidities	None	None	None
Shock	Yes	Yes	Yes—compensated
Fever	No	Yes	Yes
Gastrointestinal symptoms	Vomiting, obstruction	Distension, peritonitis	Pain, tenderness, diarrhea
WBC on arrival (×109/L)	82.2	42.7	18.3
Diagnosis	Autopsy	Autopsy	Intraoperatively
Perforated	No	Yes	Yes
Survival	No	No	Yes
Abdominal x-ray	Thickened small bowel, no free air, no distal air. Consistent with small bowel obstruction	Mild dilated loops of bowel, no free air	Bowel wall thickening with air in rectum, no free air
CT scan	Not done	Not done	Inflammatory changes seen in the right lower quadrant with small amount of free air. Appendix was not visualized
Pathology	Significant transmural inflammation with ischemic necrosis of the appendiceal wall predominated by neutrophilic infiltration with coagulative necrosis and neutrophil serosal exudate. No perforation noted	Heavy transmural acute neutrophilic infiltrate with focal transmural necrosis. Perforation noted at the tip	Acute suppurative appendicitis with microscopic perforation

WBC indicates white blood cell count.