



A Case Of Cow’s Milk Protein Allergy Presenting As Infantile Gangrenous Appendicitis With An Incidental Ovarian Cyst –A Rare Entity Posing A Diagnostic Challenge

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Abstract

Background: Acute appendicitis is rare in neonatal or post neonatal period with an incidence rate of 0.25% of all surgical emergencies (1,3). Perforation of the appendix is common which leads to higher morbidity and mortality. Although it is very challenging to diagnose acute appendicitis in infants due to non specific symptoms and atypical clinical presentation. This case of appendicitis with an etiology of cow milk protein allergy is presented for its rarity and to emphasis the importance of history taking as a tool for effective clinical diagnosis and treatment ,thereby preventing infantile morbidity and mortality(2) .

Case Report: A one and half month old female baby presented with high grade fever and incessant cry since 2 days. H/o cows milk ingestion 2 days back . USG abdomen showed focal peritonitis with infected collection in right iliac fossa. So proceeded with open Laparotomy and removed gangrenous appendix and a small ovarian cyst (incidental finding). We received a swollen appendix and external surface shows blackish discolouration and fibrinous exudation ,with a small perforated area at the tip . C/s lumen appears necrotic. Also received a tiny greyish brown tissue mg 0.5cm (collapsed cyst). Microscopically Extensive ulceration of lining mucosa, dense transmural neutrophilic infiltration with extensive areas of ischemic necrosis extending upto the serosa . Section from the collapsed cyst wall shows cyst lined by flattened epithelium with underlying unremarkable ovarian stroma. No necrosis noted. Histomorphological features were consistent with a clinical diagnosis of Perforated Gangrenous Appendicitis and Benign simple Developmental Cyst.

Conclusion : Acute appendicitis is rare in neonates and infants. Delay in diagnosis and treatment results in perforation of appendix and peritonitis. Cow milk protein allergy is one of the common etiological factor in infants and children which results in gastrointestinal manifestations varying from mild to severe life threatening conditions. Its very essential for an accurate diagnosis and active intervention is needed for reducing morbidity. And histopathological examination helps in confirming final diagnosis and for a further follow up.

Keywords: NIL

Introduction

Acute appendicitis is rare in neonatal or post neonatal period with an incidence rate of 0.25% of all surgical emergencies (1,3). Perforation of the appendix is common which leads to higher morbidity and mortality. Although it is very challenging to diagnose acute appendicitis in infants due to non

specific symptoms and atypical clinical presentation. Delayed diagnosis can result in higher perforation rate and increased morbidity(2). So clinical diagnosis and radiological evaluation helps in effective management of the condition. Quick and accurate

diagnosis may increase survival rates and favourable outcome(3).

Case Report

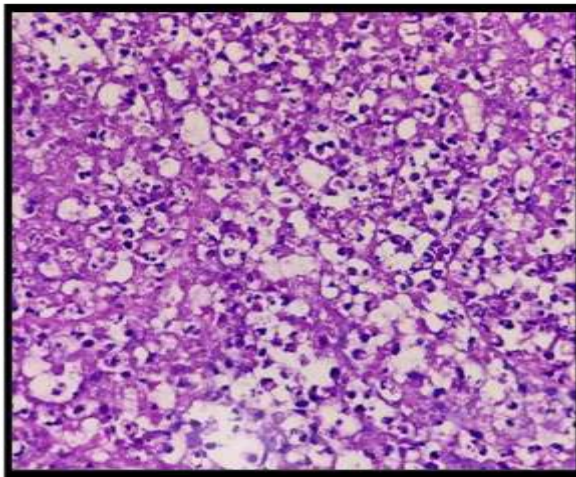
A one and half month old female baby came to emergency department with complaints of high grade fever since 2 days, irritable cry and poor feeding since one day. Baby was on exclusive breast feeding and there was history of recent introduction of cows milk present. Neonatal history was late preterm, small for gestational age (SGA) and low birth weight baby (1.42kg). No history of congenital abnormalities/drug allergy seen. On clinical examination there was pallor and distended abdomen. Complete blood count showed Haemoglobin 7.5gdl, RBC -4millions/cumm , WBC- 6900 cells/cumm and platelet count was 1.9 lakhs/cumm. RFT and serum electrolytes are within normal limits. LFT showed elevated alkaline phosphatase and CRP was high (386). Xray showed dilated bowel loops. USG abdomen showed inflammatory changes with omental and mesenteric edema/thickening in right iliac fossa and lumbar region – suggestive of focal peritonitis with infected collection. Clinical diagnosis was septic ileus with perforation peritonitis. So proceeded with emergency laparotomy.

Intraoperative findings were perforated gangrenous appendix and multiple erythematous patches over entire length of terminal ileum surrounded by flakes. In our histopathology laboratory, we received two biopsy samples, specimen A and specimen B. Specimen A was necrotic appendix and Specimen B was ovarian cyst which was incidentally detected during surgery. On gross examination, appendix measuring 3cm in length. External surface of appendix appears blackish in colour with exudates and a perforated area seen at the tip of appendix measuring 0.2cm in length. Specimen B was ovarian cyst right and we received a collapsed tiny cyst with greyish brown appearance measuring 0.5x0.4x0.2 cm. Microscopy of specimen A, appendix showed extensive ulceration of lining mucosa and dense transmural neutrophilic infiltration extending upto serosa with areas of necrosis. Serosa shows congested vessels and fibrinous exudates Microscopy of Specimen B ovarian cyst right, shows a cyst lined by flattened epithelium with underlying unremarkable ovarian stroma. No evidence of necrosis noted. Histomorphological pictures were consistent with a clinical diagnosis of Perforated Gangrenous Appendicitis and Benign simple Developmental cyst.

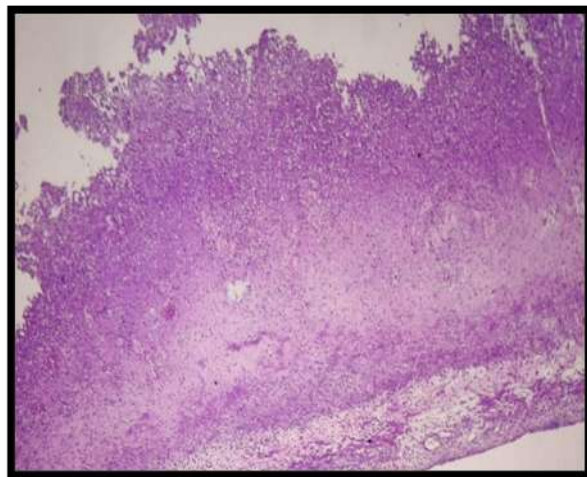
Figure 1a) Gross appearance of appendix



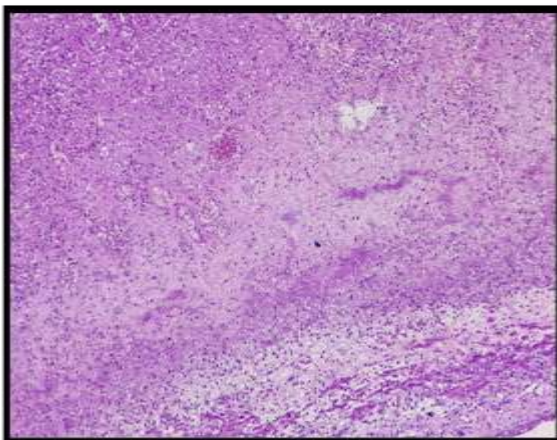
Figure 1 b) Gross appearance of collapsed ovarian cyst



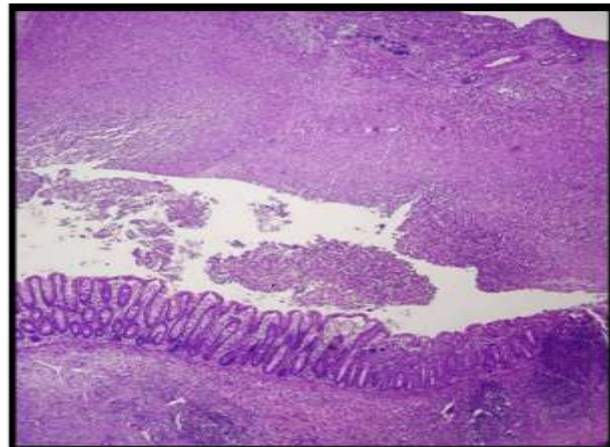
H&E 400X - Dense neutrophil infiltration appendix



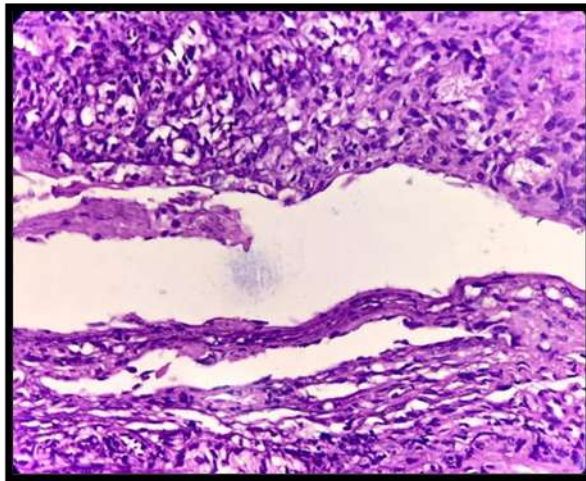
H&E 100X Appendix with extensive ulceration



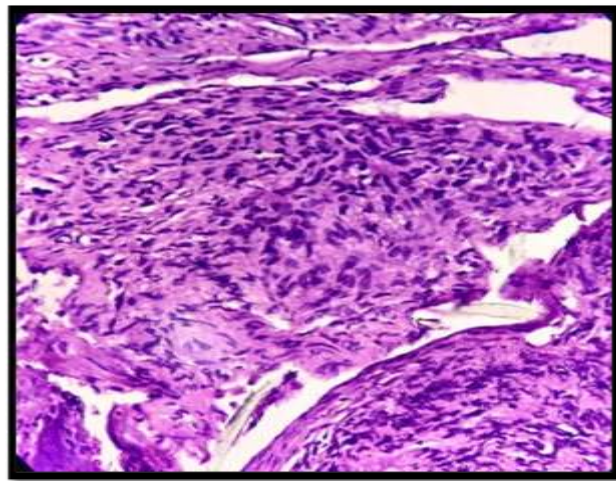
H&E 400X - Areas of ischaemic necrosis



H&E:100X- Appendix with extensive ulceration



H&E400x – Flattened epithelium ovarian cyst



H&E 100X – Ovarian cyst

Discussion

Acute appendicitis is the most common cause of abdominal pain and indicative of surgery in paediatric age group(5). But it is uncommon in infants and neonates (6). Appendicitis is an acute inflammatory process involving the appendix. The cause of the appendicitis is an obstruction of appendix either from the inflammation of the wall or faecoliths or due to lymphoid hyperplasia(6). Inflammation of the appendix can occur due to any foreign substance or allergens (3). One of the allergic factor causing inflammation of colon or appendix is beta-lactoglobulin which is present in cows milk (2).

Cow milk protein allergy (CMPA) is the most prevailing food allergy seen in infants and young children. It usually affects 2 to 4% of children, with the highest prevalence during the first year of life(7). It can present as mild to moderate symptoms like wheezing, vomiting and varying gastrointestinal symptoms. Infants usually present with vomiting, abdominal distension, dysentery due to allergic enterocolitis and proctocolitis. Cow milk protein allergy can result in gastrointestinal symptoms varying from mild colitis to severe life threatening conditions like ileal perforation or perforated gangrenous appendicitis which leads to peritonitis (1,4).

Ovarian lesions are cystic in nature in newborns and infants. Cystic lesions are common in infants due to maternal hormonal stimulation. Asymptomatic ovarian cysts are incidentally detected by ultrasound. Most of the cysts are benign only with less than 1 cm in diameter and spontaneously decrease over the first 6 months. Complications associated with ovarian cyst are high chances of torsion and haemorrhage (8,9). In this case, because of the localised peritoneal collection associated with peritonitis on the right side, missed the right side ovarian cyst on the ultrasound examination.

Conclusion

Acute appendicitis is rare in neonates and infants. Delay in diagnosis and treatment results in perforation of appendix and peritonitis. Cow milk protein allergy is one of the common etiological factor in infants and children which results in gastrointestinal manifestations varying from mild to severe life threatening conditions. Its very essential for an accurate diagnosis and active intervention is needed for reducing morbidity. And histopathological examination helps in confirming final diagnosis and for a further follow up.

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