ACUTE CHOLECYSTITIS PRESENTING AS INTRUSUSCPEPTION IN AN INFANT: CASE REPORT AND LITERATURE REVIEW

Siba P Paul1, Rowena Remorino

INTRODUCTION

It has traditionally been believed that cholelithiasis in infancy is very rare and that all such cases have a predisposing factor such as haemolysis, prolonged fasting, congenital abnormality of the biliary tract or administration of TPN; with severe morbidity and mortality. Early clinical suspicion and imaging is necessary to diagnose the condition. In this article we describe a case of acute cholecystitis in an infant, review the literature and discuss the possible complications.

CLINICAL COURSE

A healthy 6 month old male presented with one day history of intermittent, insoluble cry, abdominal pain and drawing up of legs. The infant was born at term and was growing well at 25th percentile. He refused feed at presentation and had one episode of non-bilious vomiting.

His examination was normal, with a soft abdomen when settled. He was admitted for observation and discharged home the next day as he remained asymptomatic. The infant re-presented 3 days later with intermittent bouts of screaming and vomiting. The stool colour was reported normal. A working diagnosis of intussusception was made, although the abdomen was soft on palpation. Abdominal ultrasound (fig. 1) showed no evidence of intussusceptions but revealed a 6 mm highly echogenic focus in the gall bladder. Blood investigations showed bilirubin of 45 mmol/L and ALT 244 IU/L. The infant was seen by the paediatric surgical team on the 6th day from initial presentation. The repeat abdominal ultrasound revealed 2 gallstones in the neck of the gallbladder with thickened walls and prominent biliary tree with no obstruction.

A diagnosis of acute cholecystitis was made and he was initially managed with IV fluids and Co-amoxiclav. There was a quick recovery and the infant

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1 Corresponding Author: Dr Siba Prosad Paul, Department of Paediatrics, St Richard’s Hospital, Spitalfield Lane, Chichester, West Sussex PO19 6SE
was recommenced on breast feeds. All the symptoms resolved except the passage of one pale creamy stool.

Haemoglobin electrophoresis, G6PD and cystic fibrosis screen were negative. The blood film showed no evidence of hereditary spherocytosis.

Specialist ultrasound in the tertiary paediatric hepatology clinic showed only one stone in the gallbladder with resolution of the bile duct dilatation. ALT at the time was 32 IU/L. Cholangiopancreatography MRI (MRCP) scan showed no gall stones.

He is currently well and under yearly follow up at the hepatology clinic with a diagnosis of idiopathic gallstones.

DISCUSSION

Definition and Aetiology

Acute cholecystitis (1,2) is termed as inflammation of the gallbladder with gallstones present during the painful episodes. Acalculous cholecystitis (1) occurs in children with other major medical problems and infections.

European studies (3) in paediatric patients estimate an overall prevalence of 0.13% to 1.9% for gallstone disease in children up to 19 years of age. In England (4) the age-standardised hospital admission rate for cholelithiasis in the 0-14 years age group was 1.1 per 100000.

It has been found that for gallstone formation some or all of the following need to occur (1): alteration in the proportion of bile constituents, nucleation, alteration in the gall bladder motility or infection.

Congenital anomalies (1,5) e.g. – multi-septate gallbladder may predispose the children to stasis and subsequent cholestasis. Sickle cell disease (SCD) is the most common cause of cholelithiasis in children (6).

TYPES OF GALLSTONES IN CHILDREN

There are three major types of gallstones:

1. Cholesterol gallstones (1) – requires hypersecretion of cholesterol into the bile. A study of 18 children with gallbladder aspiration at laparotomy for non-hepatobiliary disease, revealed that the infants had more dilute bile than children, but it was more saturated with cholesterol.

2. Black pigment gallstones (1,3) - is the most common type and is associated with a large number of diseases wherein excess secretion of conjugated bilirubin into the bile occurs.
   - haemolytic disease eg – sickle cell disease, hereditary spherocytosis;
disorders of dysfunctional erythropoiesis eg – prolonged TPN, cancers;
diseases that cause bilirubin to undergo enterohepatic circulation eg – ileal resection in infancy, Crohn’s disease, cystic fibrosis.

3. Brown pigment gallstones (1) – are rare in children and in the Western world. It occurs in the presence of obstruction and subsequent infection.

CLINICAL FEATURES OF CHOLECYSTITIS

The presentation of gallbladder disease depends on the underlying disease process. It is not clear why most children with gallstones remain asymptomatic for long periods while others have recurrent biliary colic. Differential diagnosis includes (1,5) pancreatitis, cholangitis, intussusception or genitourinary pathologies.

Patients with acute cholecystitis can have fever, right upper quadrant pain and may have positive Murphy’s sign. Leucocytosis is common. Biliary colic often recurs. The acute episodes are however, of short duration rather than ongoing.

Children with chronic cholecystitis can have a conglomeration of symptoms such as indigestion, abdominal discomfort and heart burn.

INVESTIGATIONS OF CHOLECYSTITIS

Clinical suspicion and radiological investigations help in preventing long term morbidity.

Ultrasoundography is the investigation of choice in diagnosing gallstones (1-6). It is best performed after 4 hours of fasting. The gall bladder is examined for evidence of stones, sludge, masses, distension, gangrene and pericholecystic fluid. Cholecystitis is often associated with a thickening of the wall of the gallbladder (>3mm).

Plain abdominal films are helpful in cases presenting as acute abdomen. 10 – 15% of gallstones are radiopaque and are detectable. A rare condition called “milk of calcium cholelithiasis” can have dramatic findings on abdominal films.

MRCP is the investigation of choice in bile duct obstruction and images the anatomy of the hepatobiliary tree and pancreatic ducts (1,8).
MANAGEMENT OF CHOLECYSTITIS

Surgical management\(^1\) is the mainstay of treatment for the paediatric gallstones \((1,9)\) and laparoscopic cholecystectomy is currently the procedure of choice. Open cholecystectomy is done where laparoscopic cholecystectomy is relatively contraindicated or cannot be safely completed.

Medical management \((1,4,5)\) consists of either a wait and watch approach, administration of bile salts or extracorporeal shock wave lithotripsy (ESWL). The benefits may be limited.

CONCLUSION

This case illustrates the importance of being aware of the presence of gallstones in infants presenting with signs of acute abdomen. Appropriate referral to paediatric surgical team and involvement of hepatologist is necessary to prevent long term complications. We hope this article leaves the reader with a heightened awareness about gallstones which are not just in the "fat, fair, fertile, female of forty" (5Fs) but also in the babies who are “born early, have blood disorders, bowel diseases or had bags of parental nutrition” (the 4Bs!)

KEYPOINTS

1) Gallstones need to be considered as a differential diagnosis in infants presenting with acute abdomen.

2) Ultrasound is the investigation of choice in diagnosing gallstones and have led to increased frequency of diagnosing gallstones.

3) Black pigment gallstones is the commonest type and are associated with excess secretion of conjugated bilirubin into the bile.

4) Sickle cell disease is the commonest cause of gallstones in children.

5) Paediatric surgeons and hepatologist needs early involvement.

6) Babies who are “born early, have blood disorders, bowel diseases or had bags of TPN” (the 4Bs! not just the 5Fs) are more at risk of developing gallstones.

KEYWORDS

1) Gallstones
2) Ultrasound
3) Black pigment gallstones
4) Cholecystectomy
5) Intussusception
6) Sickle cell disease

REFERENCES