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CHOLELITHIASIS IN AN INFANT*

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Cholelithiasis is uncommon in childhood. Although intrauterine cholestasis and gallstones have been observed, such cases in infancy are rare[1,2]. Gallstones are encountered more frequently in children with hemolytic anemia – especially the sickle cell variety – and with cirrhosis. Wilson's disease, malformations of the biliary tract, cystic fibrosis, metachromatic leukodystrophy and parasitic diseases[3-7]. Intermittent colicky pain in the right upper quadrant, jaundice and abdominal tenderness are all suggestive of cholelithiasis[1,5]. In the presence of periodic jaundice, acholic stools, hepatomegaly, mildly elevated transaminase levels and cholestasis, cholelithiasis should be suspected. Opaque gallstones are easily seen on direct roentgenographic examination of the abdomen. Oral or intravenous cholangiograms are very helpful in the diagnosis of non-opaque stones. Recently, however, sonography has become one of the preferred laboratory methods for diagnosis[8].

This being an unusual disease, we would like to present this case of an infant with cholelithiasis diagnosed by means of sonographic examination.

Case Report

S.U., a 26 - day - old male, the first child of healthy, nonconsanguineous parents, was admitted to the Hacettepe University Children's Hospital with a history of jaundice, acholic stools and dark urine since the second day of life. His sclerae vere icteric and the liver edge was palpable two to three centimeters below the right costal margin, while the spleen was not palpable; other physical findings did not contribute to the diagnosis.

Laboratory findings. The hemoglobin was 17.25 g/dl, the white cell count was 10.000/µl and there was no reticulocytosis; bilirubinuria was present but there

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was no stercobilin in the stools. The SGOT was 50 U; the SGPT was 55 U, the cholesterol was 295 mg/dl; total lipid was 1000 mg/dl; total bilirubin was 7 mg/dl (conjugated form : 3.9 mg/dl), alkaline phosphate was 17.5 BU. A plain abdominal X - ray did not disclose any abnormality and the dye test, the VDRL and the HBsAg were negative.

Phenobarbital (5 mg/kg) was started. A month later the liver edge was still palpable three centimeters below the right costal margin, the bilirubin was found to be within normal limits (total 1 mg/dl; conjugated bilirubin 0.8 mg/dl) and stercobilin was present in the stool. However, because of elevated SGOT (130 U), and SGPT (155 U) values, a liver needle biopsy was performed. Microscopic findings were compatible with cholestatic hepatitis, therefore prednisolone (2 mg/kg/day) was prescribed for a month. At the end of this period the hemoglobin was 12.45 g/dl; the white cell count was 4200/µ l; SGOT was 34 U; SGPT was 39 U; the total bilirubin was 0.8 mg/dl; alkaline phosphatase was 9.3 BU; fotal lipid was 640 mg/dl and the cholesterol was 162 mg/dl.

At the age of 9 months the patient was readmitted to the hospital with an episode of jaundice. His bilirubin was then 3 mg/dl (conjugated form 2.2 mg/dl), SGOT was 34 U; SGPT was 73 U; alkaline phosphatase was 28 BU; total lipid was 1300 mg/dl and the cholesterol was 400 mg/dl. Sonographic studies showed multiple gallstones. The ductus choledochus was found to be dilated during exploration but the distal part could not be visualized by operative cholangiography. The gallstones were removed and a cholecystectomy with choledochoduedonostomy was performed. The liver wedge biopsy showed some interlobular fibrosis and examination of the gall bladder revealed minimal round cell infiltration in the submocosal layer.

Discussion

Cholelithiasis is rare in children[9]. Since the classical symptom of right subcostal pain cannot be obtained in infancy, the correct diagnosis is often made late. Periodic jaundice, acholic stools and abdominal tenderness are important physical signs. The serum bilirubin, alkaline phosphatase, cholesterol and lipids levels are also usually elevated as in our patient. Increased liver enzyme activity may suggest cholangitis. Cholestatic syndromes, cholestatic hepatitis and sclerosing cholangitis cannot easily be differentiated from cholelithiasis, especially when the stones are silent and non-opaque.

Varios etiologic factors relating to gallstone formation have been considered. In different hemolytic disorders such as sickle - cell anemia[3], thalassemia[10], spherocytosis or elliptocytosis. Pigmented gallstone formation is relatively frequent due to increased bilirubin production[11]. The sluggish flow of sickle cell erythrocytes, which causes focal hepatic necrosis, is an additional factor for

gallstone formation in sickle cell anemia. Increased bilirubin formation as a result of hemolysis may effect gallstone formation in patients with cirrhosis and Wilson's disease[4,9]. Gallstones have been found in children with impaired bile flow due to a prolonged period of dehydration or cystic fibrosis. They often form in patients with cholecystitis following mesenteric lymphadenitis or salmonellosis. Total parenteral nutrition is potentially lithogenic and is a critical factor in noncholesterol stone formation[12]. Furosemide, which acts by reducing bile flow and/or increasing excretion of biliary calcium, should also be considered a potential stone-former[13,14]. Ileal resection is a predisposing factor for cholesterol gallstone formation with the interruption of normal enterohepatic circulation leading to a reduction in the bile salt pool and an increase in hepatic bile lithogeny[15]. Other factors such as borderline enteral intake, low bile output and phototherapy should be considered in babies with gallstones. Our patient did not have any of the above mentioned conditions, but stenosis of the common bile duct, which we have taken to be an etiologic factor, was present.

In uncomplicated cases, the prognosis of cholelithiasis is excellent, but recurrent episodes of jaundice may cause hepatocellular damage and eventually secondary biliary cirrhosis[16]. Gallstones are not always found in conjuction with cholecystitis, and therefore, when the two are present, it may be difficult to decide wihch is cause and which effect[17]. Although treatment with oral chenodeoxycholic acid, especially in cases of radioluscent gallstones, is getting more popular, this form of treatment has not yet been used on infants[18,19]. If a therapeutic method is to be used on young, asymptomatic children, this is the one that should be tried first: However, in the case of our patient, we believe that surgical removal of the gallstones with a cholecystectomy was the best way of dealing with the condition.

Summary

A case of cholelithiasis in an infant is reported. Since this is a rarity in infancy the relevant literature is reviewed and the usefulness of sonographic examination, a recent diagnostic method, is emphasized. Treatment is briefly discussed.

REFERENCES

1. Walker CHM. Etiology of cholelithiasis in childhood. *Arch Dis Child* 32 : 293, 1957.
2. Anderson CM. Cholelithiasis in a neonate. *Arch Dis Child* 50 : 815, 1975.
3. Ariyan S, Shessel FS, Pickett LK. Cholecystitis and cholelithiasis masking as abdominal crises in sickle cell disease. *Pediatrics* 58 : 252, 1976.
4. Rosenfield N, Grand RJ, Watkins JB, et al. Cholelithiasis and Wilson disease. *J Pediatr* 92 : 210, 1978.

5. Roy C, Silverman A, Cozzeto FJ. *Pediatric Clinical Gastroenterology* (2nd ed) St. Louis : CV Mosby Co, 1975, pp 608 - 614.
6. Kirtley JA Jr. Holcomb GW Jr. Surgical management of diseaases of the gallbladder and common duct in children and adolescents. *Am J Surg* 111 : 39, 1966.
7. Hanson BA, Mahour GH, Woolley MM. Diseases of the gallbladder in infancy and childhood. *J Pediatr Surg* 6 : 277, 1971.
8. Buschi AJ. Brenbridge NA. Sonographic diagnosis of cholelithiasis in childhood. *Am J Dis Child* 134 : 575, 1980.
9. Bouchier IA. Postmortem study of the frequency of gallstones in patients with cirrhosis of the liver. *Gut* 10 : 705, 1969.
10. Gürgey A, Altay Ç, Hiçsönmez A. Safra taşı ve spontan yaygın kırıkları olan iki beta talassemi majör vakası. *Çocuk Sağlığı ve Hastalıkları Dergisi* 26 : 85, 1983.
11. Bates GC, Brown CH. Incidence of gallbladder disease in chronic hemolytic anemia (spherocytosis). *Gastroentorology* 21 : 104, 1952.
12. Roslyn JJ, Berquist WE, Pitt HA, et al. Increased risk of gallstones in children receiving total parenteral nutrition. *Pediatrics* 71 : 784, 1983.
13. Whitington PF, Black DD. Cholelithiasis in premature infants treated with parenteral nutrition and furosemide. *J Pediatr* 97 : 647,1980.
14. Boyle RJ, Sumner TE, Volberg FM. Cholelithiasis in a three–week–old small premature infant. *Pediatrics* 71 : 967, 1983.
15. Hill GL. Mair WS, Goligher JC. Gallstones after ileostomy and ileal resection. *Gut* 16 : 932, 1975.
16. Pellerin D, Bertin P, Nihoul - Fekete C, et al. Cholelithiasis and ileal pathology in childhood. *J Pediatr Surg* 10 : 35, 1975.
17. Gravier L, Dorman G, Votteler T. Gallbladder disease in infants and children. *Surgery* 63 : 690, 1983.
18. Coyne MJ, Bonorris GG, Chung A, et al. Treatment of gallstones with chenodioxycholic acid and phenobarbital. *New Engl J Med* 292 : 604, 1975.
19. Dissolving hopes for gallstone dissolution? (Annotation). *Lancet* 2 : 905, 1981.