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CHOLELITHIASIS IN AN INFANT WITH HYPERTROPHIC PYLORIC STENOSIS

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ABSTRACT:

Objective: To report a case of cholelithiasis with hypertrophic pyloric stenosis in a young infant and review the literature.

Methods: A case of hypertrophic pyloric stenosis with cholelithiasis diagnosed during ultrasonographic evaluation is described.

Results: A 3 month old infant presented with vomiting from day 7 of life was diagnosed as hypertrophic pyloric stenosis with cholelithiasis. Predisposing factor for cholelithiasis was starvation and dehydration secondary to pyloric stenosis. The patient was managed with open pyloromyotomy with cholecystectomy.

Conclusion: Cholelithiasis in childhood is considered a rare condition, usually associated with hemolytic disease, TPN, starvation etc. The increase in the use of ultrasonography has contributed to an increase in the diagnosis among newborns and young infants. This case report alerts to the possibility of gallstone formation following prolonged starvation.

KEYWORDS: Cholelithiasis, hypertrophic pyloric stenosis

INTRODUCTION

Cholelithiasis in infancy has been reported as a rare condition.1-4 The occurrence of cholelithiasis in the neonatal period and in young infants has been more frequently described due to the increase in use of abdominal ultrasonography.4,5 Cholelithiasis in childhood is usually associated with hemolytic disease.6 In the neonatal period, however, the following predisposing factors have been reported: prematurity; sepsis; total parenteral nutrition; long-term use of furosemide; hemolysis; and congenital anomalies of the biliary tree.1,4,7,8 Neonatal cholelithiasis may be characterized by incidental diagnosis and possible spontaneous resolution.8-11 Our objective is to report a case of asymptomatic cholelithiasis in a young infant diagnosed incidentally during evaluation for pyloric stenosis. The patient underwent cholecystectomy at the same time of pyloromyotomy.

CASE REPORT

A 3 month old female infant a product of consanguineous marriage, born as a full term normal delivery, presented with vomiting of feeds from 7th day of life. The vomiting progressively increased and became projectile. The patient presented late to us at the age of 3 months with features of dehydration and hypochloremic metabolic alkalosis. The electrolyte imbalance normalized over 24 hours with N/2 Saline in D5% and KCl (1:100). Laboratory investigations showed following:

Hb	9.4		CRE	0.17
HCT	31.3		Bil	1.08
Bil conjugated 0.2				
WBC	4950		ALT	70
PLT	159,000	ALP	159	
Ret	5.79		TP	5.24
PBF	Macrocytes, crenated RBC, Schistocytes seen		Alb	3.4
pH	7.595			
Na	129			
K	3.5			
Cl	89			
Coaglogram	Normal			
Blood culture	Sterile			

Ultrasonography showed multiple stones in gall bladder with pyloric canal length of 15 mm, thickness 12 mm and muscle thickness of 7 mm suggestive of hypertrophic pyloric stenosis. Patient was subjected to exploratory laprotomy through a right subcostal incision. Pyloro myotomy and cholecystectomy was done. (Because patient was operated for hypertrophic pyloric stenosis and it is recommended that if in a child on laprotomy you will find icidental **Cholelithiasis** you should do cholecystectomy) Post operative period was uneventful. Normal feeding was resumed in 24 hours. Patient was discharged on 3rd post operative day. Patient is asymptomatic and is on follow up. On analysis the stones were found to contain bile salts (30%), bilirubin (30%), biliverdin (20%) and hematin (20%).

DISCUSSION

Cholelithiasis in infancy is considered rare.1-4 During the recent years , incidence of cholelithiasis in infancy has been increasing due to the increase in the use of abdominal ultrasonography in this age group.4,5,8 Usually, diagnosis of cholelithiasis is incidental, observed in the investigation of other pathologies or in routine examination at neonatal Intensive Care Units.8-10 Cholelithiasis in childhood is associated with hemolytic disease, congenital anomalies of the biliary tree, infection, and adolescent pregnancy.4,6,12-15 Neonatal cholelithiasis is rarely associated with hemolytic disease3,4 The following predisposing factors have been reported in cholelithiasis in childhood: prematurity, prolonged fasting, long-term total parenteral nutrition, long-term use of furosemide, sepsis, dehydration, phototherapy, congenital anomaly of the biliary tree, Down's syndrome, TORCH, family history, and antibiotic therapy.1,4,5,7,8,10,11,16-18 The literature also reports association of maternal predisposing factors, such as preeclampsia and diabetes, with neonatal cholelithiasis.19

Up to 43% of children receiving TPN develop gall stones. The mechanism is probably change of composition of bile and stasis. Because only some patients receiving TPN develop gallstones so other factors also play a role. Sepsis, dehydration, chronic furosemide therapy,cystic fibrosis , short bowel syndrome may also be contributing factors. It is possible that all of the mentioned factors, associated with immaturity of enterohepatic circulation of bile acids, could determine biliary stasis - the main mechanism in formation of gallstones during the neonatal period.20-22 Other studies have indicated that neonatal cholelithiasis may be a temporary condition due to biliary stasis.4,11 The temporary characteristic of cholelithiasis could explain prenatal ultrasonography diagnosis of gallstone in fetuses with subsequent involution during intrauterine life.4,23 Prolonged fasting and minimal enteral nutrition may yield biliary stasis, whereas a full diet stimulates bile flow and, thus, inhibits biliary stasis. Prolonged fasting also inhibits secretion of intestinal hormone, which is responsible for normal enterohepatic circulation of bile acids. The lack of hormone may alter bile composition and, consequently, lead to stasis.7,11,16

In premature children, total parenteral nutrition is associated with cholestasis and formation of sludge and gallstones.11 Amino acids play a serious toxic role, since most enzyme pathways of its metabolism are still immature, causing accumulation of intermediary metabolites.24 The

literature discusses the fact that total parenteral nutrition is usually associated with prolonged fasting and minimal enteral nutrition in immature newborn infants.25,26 Different authors, however, have observed that cholestasis, with consequent lithogenic disorders and formation of gallstones, is a late complication in total parenteral nutrition, especially among premature babies.27-30 Biliary stasis favors proliferation of bacteria, and thus causes an increase in the production of beta-glucuronidase enzyme, which is a product of bacteria. This

enzyme is responsible for hydrolysis of conjugated bilirubin into unconjugated bilirubin. A greater amount of unconjugated bilirubin increases saturation of the bile, causing formation ofgallstones. 8,17,21,31 The use of diuretics, especially of furosemide, inhibits transportation of sodium into the cell interior, which may cause a reduction in excretion of bile acids and, consequently, the formation of gallstones. Also, furosemide may increase excretion of calcium into the bile; a mechanism which is similar to what occurs in the kidney and leads to formation of nephrocalcinosis and gallstones.10,16,32-34 According to the literature, incidentally diagnosed asymptomatic neonatal cholelithiasis usually has a benign course, with about a 50% chance of spontaneous resolution of cases during the first 6 months of life.7 Resolution of cholelithiasis occurs after the dissolving of gallstone or after the fragmenting and passing of gallstone through the biliary tree.35 Long-term follow-up is recommended for children persisting with gallstones after 6 months of age, as long as they have remained asymptomatic.7,35 Surgical intervention should be reserved for symptomatic cases or those in which calcification of gallstone occurs.36,37 The case presented in this report is in agreement with the literature with respect to presence of the following risk factors: prematurity, sepsis, total parenteral nutrition, longterm use of furosemide, delayed introduction of enteral nutrition, use of phototherapy, and mother with history of preeclampsia. The diagnosis of our patient was incidental; it occurred during a routine follow-up ultrasonography examination of nephrocalcinosis. Formation of gallstone after the 16th week of life, however, is in opposition with the study by Randall et al.,34 in which formation of gallstones in risk-group patients occurs primarily during the first 12 weeks of life. In our case, the patient remained asymptomatic, thus allowing observation of the case. Table 1 was adapted from Morad et al.7 and presents cases of incidentally diagnosed neonatal cholelithiasis described in the literature. In the event of incidental diagnosis of asymptomatic cholelithiasis in neonates or young infants, it is recommended to simply observe the case and carry out periodical control ultrasonographies. Usually, spontaneous resolution of gallstone occurs during the first 6 months of life. In cases of asymptomatic cholelithiasis... –

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