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Case Report

The Umbilico-Bilious Fistula: A Diagnostic Challenge

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Abstract:

Background: Hepato-biliary system is known for congenital variations. An acquired entity secondary to a pathology/infection is a rare phenomenon.

Case: A 3 months old baby presented with bilious leak from peritoneum secondary to complicated umbilical sepsis. On further work up, a fistulous communication was found between umbilical ligament and biliary tree.

Conclusion: Umbilico-bilious fistulization is a rare presentation of umbilical sepsis. It should be kept in mind while dealing with umbilical sepsis cases.

keywords: heart failure; morbidity; mortality; perioperative risk; surgery

Introduction

Anatomical variations in the anatomy of the liver are common. The variations in arterial supply to the liver are most frequent among these, but biliary system developmental variations are also common. We present here a rare case of umbilico-bilious fistula encountered in an infant with umbilical sepsis, which was confirmed by contrast study and the difficulties encountered in diagnosis and management.

Case Report

A 3 months old male baby presented to emergency with denuded supra-umbilical skin and greenish discharge for 1 day. There was history of swelling and redness just above the umbilicus for one week. There was no history of fever or vomiting. Baby had decreased breast feeding for 2 days. History of umbilical swelling since birth and reducing at own was elicited, most likely suggesting umbilical hernia. Baby was admitted in a private hospital for 1 month post birth for preterm gestation and low birth weight. Mother was on anti-tubercular treatment for pulmonary tuberculosis. At presentation, baby was 1.95 kg in weight. On examination, baby was dehydrated with heart rate of 157/min, respiratory rate of 28/min and capillary refill time of > 3 seconds. Icteric tinge was present. Per abdomen examination had denuded supra-umbilical skin, exposing the underlying bowel with bile staining. Baby had an ultrasound outside whose report mentioned of umbilical hernia and thrombosis of portal and umbilical vein. After fluid resuscitation, USG at our center revealed normal liver and gall bladder with multiple tortuous vessels in region of portal vein and heterogenous echogenic area around 1x1 cm near portal vein and traversing towards anterior abdominal wall abutting skin upto umbilical area. Mild free fluid was noted in peritoneal cavity. Routine investigations at admission revealed hemoglobin of 9.7 g/dl, total leucocyte counts of 37300/uL and platelets of 3 lakhs/uL. C-reactive protein levels were 113 mg/L. The total bilirubin levels were 9.04 mg/dl with direct component of 4.81. International normalized ratio was 1.2. In view of sepsis, piperacillin tazobactum was started (broad spectrum). The baby was taken for emergency exploratory laparotomy with suspicion of bowel/ biliary tract perforation in view of biliary disharge. On exploration, whole bowel was intact with some bile staining near the lesser sac. On further search, the falciform ligament was found to have a

tract with rent near the hepatic surface and clear bile discharge. [Figure 1] A 6-Fr Foley catheter was inserted into the tract for future contrast study, which revealed communication to the biliary tree. [Figure 2] The tract was ligated in a second surgery (for burst abdomen). Sadly, the baby succumbed to sepsis.

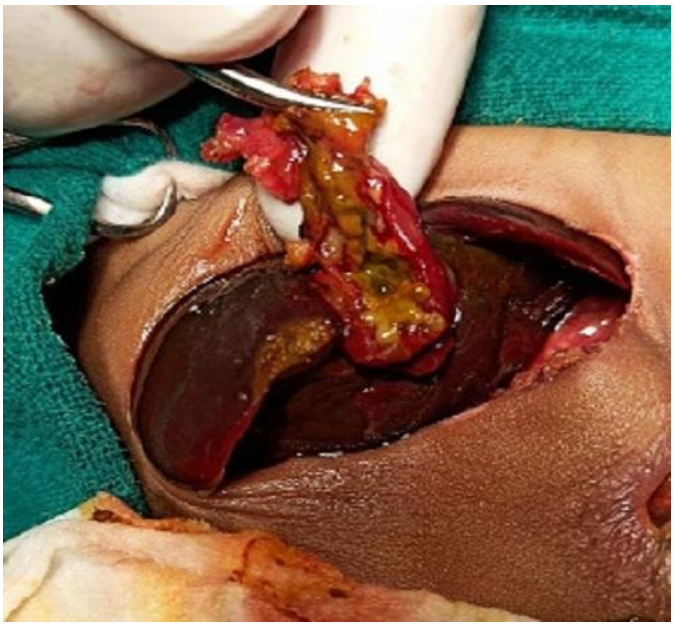

Figure 1: Intra-operative picture of bilious discharge from the umbilical ligament.

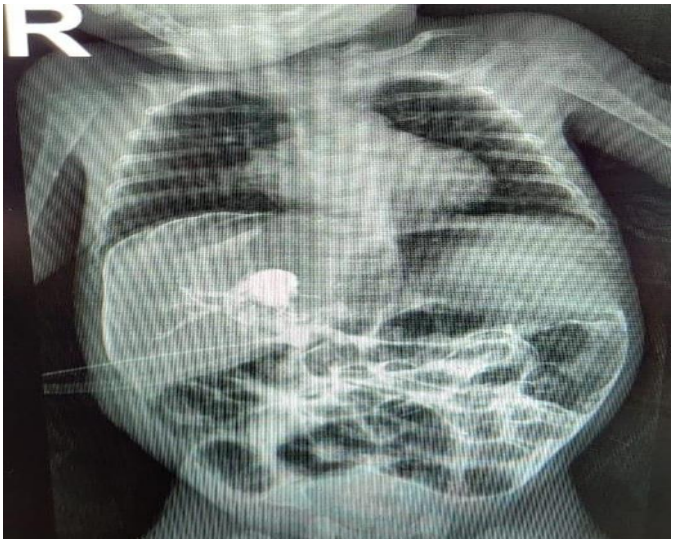

Figure 2: The contrast study showing fistulous communication with the biliary tree.

Discussion

Liver develops as ventral hepatic bud from foregut. The endodermal cells of bud invades septum transversum and form hepatoblasts sheets. The hepatoblasts and mesenchyme cells form biliary tree. The hepato-biliary system is known for variations from normal anatomy. These variations arise from aberrations in embroyological development. Most of these involve extra-hepatic system, but cases are reported in literature for internal communication to other systems like respiratory or gastro-intestinal [1,2,3]. The aberrant duct opening into left triangular ligament has also been reported [4].

The index case presented as biliary peritonitis post denudation of umbilical skin after umbilical sepsis. The bile leak was from falciform ligament at liver surface. It may have been a congenital umbilico-bilious fistula, which got burst into the peritoneum via umbilical ligament or an inflammatory reaction of ligament caused secondary infection/ inflammation leading to fistulization. Only one such case is reported in the literature [5].

Conclusion

Umbilico-bilious fistulization is a rare presentation of umbilical sepsis. It should be kept in mind while dealing with umbilical sepsis cases.

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