

CASE REPORT

Hepatic Subcapsular Hematoma: Two Neonates With Disparate Presentations

Maliyackel Aiyappanpillai Anjay^{a,*}, Chaniyil Krishnan Sasidharan^b, Parameswaran Anoop^c

^a Department of Paediatrics, Great Ormond Street Hospital, London, UK

^b Department of Neonatology, Baby Memorial Hospital, Calicut, India

^c Department of Paediatric Haematology, Great Ormond Street Hospital, London, UK

Received Aug 9, 2009; received in revised form Jan 22, 2011; accepted Jan 31, 2011

Key Words

birth trauma;
hepatic subcapsular
hematoma;
neonatal jaundice

Subcapsular hematoma of the liver rarely occurs in neonates and the diagnosis is often missed or delayed. We report two babies who had this uncommon condition in the early neonatal period. In the first baby, the hematoma was associated with ventouse delivery and presented with abdominal distension and worsening jaundice. In contrast, the other baby was relatively well, with progressive pallor as the only clinical finding. The former had no other identifiable risk factors, whereas the latter was confirmed as having classical hemophilia. The literature is briefly reviewed with regards to incidence, etiology, diagnosis and management. Awareness of this unusual entity coupled with a high index of suspicion is essential for early identification and stabilization of such babies.

Copyright © 2012, Taiwan Pediatric Association. Published by Elsevier Taiwan LLC. All rights reserved.

1. Introduction

Collection of blood between the liver and its capsule is a rare manifestation of birth trauma. The diagnosis is frequently made only at autopsy. In an analysis of 755 perinatal autopsies by Singer et al. in 1999, hepatic subcapsular hematomas were encountered in 21 neonates (2.8%).¹

We report two neonates who had this unusual condition, with varying etiologies and presentations.

2. Case Reports

2.1. Case 1

A 4-day-old male infant presented with poor feeding, lethargy and jaundice. He had been delivered by ventouse at term, following a prolonged second stage of labor. He weighed 3600 g at birth and was well during the first 3 days.

* Corresponding author. 4 Foulis Terrace, London SW7 3LZ, UK.
E-mail address: anjayma@gmail.com (M.A. Anjay).

Jaundice with rapid progression was noted from the third day. Subsequently, he developed poor feeding, fast breathing and abdominal distension. Examination revealed a sick, tachypnoeic baby with a heart rate of 180/minute and a systolic blood pressure of 60 mmHg. He was pale and deeply jaundiced, with abdominal distension and firm hepatomegaly. No other abdominal masses were palpable and bowel sounds were heard normally.

The hemoglobin level was 8 g/dL (80 g/L) with a normal platelet count and coagulation profile. He had indirect hyperbilirubinemia (total bilirubin 17.5 mg/dL, direct 1.6 mg/dL), with no evidence of hemolysis. Sepsis screen was negative. Ultrasonogram of abdomen revealed a moderate-sized anechoic area separating the anterolateral edge of the right lobe of liver from its capsule, with a few internal echoes towards the dependent part, suggesting a subcapsular hematoma (Figure 1).

There was no history of bleeding diathesis in the family. The baby was managed with blood transfusion and supportive care. The clinical condition improved gradually. The maximum measured dimensions of the hematoma were 1.9 × 4.6 cm and serial ultrasonograms demonstrated resolution of the lesion. He recovered uneventfully and is currently under follow-up.

2.2. Case 2

A term neonate was born following uncomplicated pregnancy and delivery. He was noted to be lethargic and not feeding well at 8 hours of life. Clinical examination showed mild pallor and otherwise normal systemic examination. Complete blood count done as a part of partial septic screen showed hemoglobin of 13.1 g/dL (131 g/L) with normal white cell count, platelets and blood film.

The baby continued to refuse feeds and had increasing pallor. Cranial ultrasonogram ruled out intracranial hemorrhage. He was started on intravenous 10% dextrose in addition to first-line antibiotics. A repeat full blood count showed a significant drop in hemoglobin to 9.2 g/dL. Coombs' test was negative, serum bilirubin was within normal limits and blood film examination was unremarkable. An ultrasonogram of abdomen showed a curvilinear fluid collection with some echogenicity between the liver

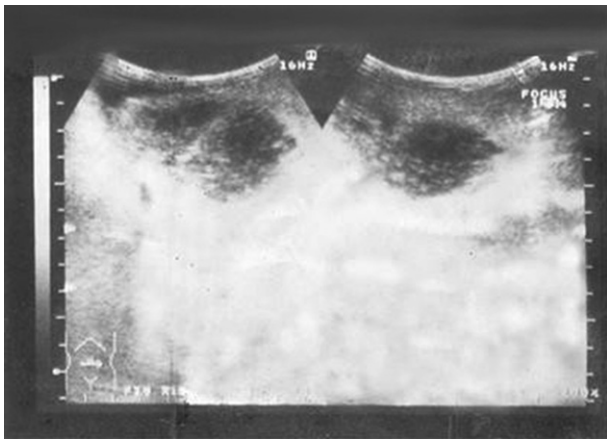


Figure 1 Ultrasonogram showing hematoma under the liver capsule.

and its capsule, consistent with a subcapsular hematoma. The maximum recorded dimensions were 1.3 × 2.9 cm.

A subsequent coagulation screen showed a prolonged activated partial thromboplastin time of 129 seconds (normal 35–90 seconds), normal prothrombin time of 16 seconds (normal 13–17 seconds) and fibrinogen level of 3.3 g/L (normal 1.5–3.5 g/L). Further investigations were performed to establish the cause for the bleeding diathesis. The factor VIII level was noted to be strikingly low at 0.03 U/mL (3%; normal 1.00 U/mL ± 0.39), thus confirming a diagnosis of severe classical hemophilia. There was no family history of bleeding disorder, which is the case in approximately one-third of patients with classical hemophilia.

The baby was managed conservatively with cryoprecipitate, transfusion of factor VIII and packed red cells. Serial ultrasonograms demonstrated resolution of the hematoma and he is on hematological follow-up.

3. Discussion

The process of labor is a combination of tractional forces, torsion, compression and contractions. Injuries to the infant resulting from mechanical forces during delivery are categorized as birth trauma. Although there has been a considerable decline in their incidence following advances in obstetric management and reduced use of instrumentation, birth injuries continue to complicate many deliveries. Subcapsular hematoma of liver is a collection of blood under the Glisson capsule. Though known to occur in adults following blunt abdominal trauma, it is a very unusual manifestation of birth injury. This should be distinguished from an intrahepatic hematoma, where bleeding is usually less extensive and occurs within the liver parenchyma. Proposed predisposing factors for a subcapsular hematoma include traumatic labour, coagulopathies, prematurity, very low birth weight, hypoxia, sepsis, pneumothorax and umbilical venous catheterization.² Most reported cases in the literature are in babies born early with very low birth weight and the condition is extremely rare in term healthy neonates.

Acute massive bleeds can present in the immediate neonatal period with signs of hypovolemia and shock. Slowly progressing hematomas manifest with pallor, jaundice, irritability or respiratory distress. They can rarely present as abdominal masses without clinical signs of bleeding and may mimic tumors.³ Ultrasonography is the investigation of choice. It can delineate the lesion well, differentiate it from neoplasms, rule out rupture and aid in serial follow-up.^{3,4}

The neonatal liver has limited ability to achieve spontaneous hemostasis, because of the weaker parenchymal connective tissue framework and poor contractility of hepatic veins. In addition, compression of the thoracic cage during delivery may lead to stretching and damage of the coronary ligament of the liver, which is attached to the inferior surface of the diaphragm. In Case 1, presence of birth trauma may have augmented these inherent risks even in the absence of other risk factors. We hypothesize that the considerable negative pressure exerted by the ventouse may lead to significant pressure changes in the

body cavities and great veins. This, when transmitted to solid organs like liver, can potentially cause hemorrhage.

Severe hemophilia is well known to present as bleeding in the neonatal period, most commonly as intracranial bleeds. Even in the absence of a positive family history, hemophilia should be suspected in neonates with unusual major bleeding manifestations as new mutations account for 30% of patients, as in Case 2. Although other forms of bleeding like muscular hematoma following injections and bleeding from venepuncture sites have been reported, subcapsular hematoma of liver has seldom been identified as a manifestation of hemophilia in neonates.^{5,6} Early diagnosis is essential as the subcapsular hematoma may progress and rupture and result in catastrophic hemorrhagic shock in the absence of prompt factor VIII replacement therapy.

It is interesting to note that the two cases discussed here had strikingly dissimilar clinical features. The presentation in Case 1 was as a sick neonate with jaundice and abdominal distension with hepatomegaly. In contrast, Case 2 was a relatively stable baby with unexplained worsening pallor alone as the clinical manifestation. Awareness of the varying spectrum of severity and the diverse etiology will help in clinical suspicion of a hepatic subcapsular hematoma at birth.

Management is mainly conservative, including blood transfusion, correction of coagulopathies and avoiding

excessive handling of the baby. Surgery is reserved for rupture of the hematoma into the peritoneum. In view of the potential for life-threatening complications, timely suspicion and use of appropriate investigative modalities are essential to minimize morbidity and mortality from this rare entity.

References

1. Singer DB, Neave C, Oyer CE, Pinar H. Hepatic subcapsular hematomas in fetuses and neonatal infants. *Pediatr Dev Pathol* 1999;**2**:215–20.
2. French C, Waldstein G. Subcapsular hemorrhage of the liver in the newborn. *Pediatrics* 1982;**69**:204–8.
3. Mouratidis B, Antonio G. Sonographic diagnosis of subcapsular liver hematoma mimicking a tumor in a neonate. *J Clin Ultrasound* 2000;**28**:53–7.
4. Mouzard A, Cohen JY, Huault G. Ultrasonography in subcapsular hematomas of the liver in the newborn. *Pediatrics* 1982;**70**:1016–8.
5. Le Pommelet C, Durand P, Laurian Y, Devictor D. Haemophilia A: two cases showing unusual features at birth. *Haemophilia* 1998;**4**:122–5.
6. Hamilton M, French W, Rhymes N, Collins P. Liver haemorrhage in haemophilia: a case report and review of the literature. *Haemophilia* 2006;**12**:441–3.