

Unexpected extensive hemorrhage from a subcapsular hematoma of the liver during emergent laparotomy in a premature neonate

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ABSTRACT

غالبًا ما يتم العثور على ورم دموي تحت الكيسية أثناء تشريح الجنين عند الرضع الميتين وليس سريريًا. وفي الغالب لا تظهر الأعراض إلا إذا تمزق. وبالتالي، غالبًا ما يتم تأخير التشخيص أو تفويتها. يؤدي تمزق ورم دموي تحت الكيسية في حديثي الولادة المبكرة إلى حدوث نزيف كبير داخل البطن، والذي يرتبط بارتفاع معدل الوفيات. وبالتالي، لا بد من الإدراك المبكر والعلاج لتجنب التمزق. نستعرض في هذا التقرير حالة نزف تهدد الحياة من ورم دموي تحت الكبد من خلال فتح البطن الناشئ لعرقلة ميكانيكية في وليد حديث الولادة يزن 860 غرام ومناقشة التحضير قبل الجراحة المناسب وإدارة التخدير لهذه الحالة.

A subcapsular hematoma of the liver is often found during autopsy in stillborn infants rather than clinically. It is usually asymptomatic unless ruptured; thus, the diagnosis is often delayed or missed. Rupture of a subcapsular hematoma in a premature neonate causes massive intraabdominal hemorrhage, which is associated with high mortality. Thus, early recognition and treatment to avoid rupture are imperative. We describe a case of life-threatening hemorrhage from a subcapsular hematoma of the liver during emergent laparotomy for mechanical obstruction in an 860 g premature neonate and discuss the appropriate preoperative preparation and anesthetic management for this case.

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A subcapsular hematoma of the liver (SHL) is a rare disease wherein blood accumulates under Glisson's capsule of the liver. It should be distinguished from an intrahepatic hematoma that occurs in the liver parenchyma and in which the amount of bleeding is limited.¹ Subcapsular hematoma of the liver in neonates is rare with few reported cases in the literature. However, the prevalence in stillborn infants was 2.8%-15%,² suggesting that it is an important cause of stillbirth. Massive bleeding in small neonates can be catastrophic and is difficult to manage if there is insufficient preparation. Herein, we reported a case of unexpected life-threatening bleeding of SHL during emergent operation for mechanical obstruction in a premature neonate. We aim to discuss the appropriate preoperative preparation and anesthetic management for this case.

Case Report. Patient information. A preterm and extremely low birth weight (ELBW) neonate was scheduled for emergent laparotomy for a mechanical obstruction at age 28 days. He was born weighing 880 g by an emergent breech cesarean section at a gestational age of 24 weeks + 5 days (Figure 1).

Clinical findings. Immediately following birth, he was intubated and treated with surfactant and mechanical ventilation. For inotropic support, 8 ug/kg/min of dopamine was administered continuously. His comorbidities were sepsis, acute renal failure, and bilateral intraventricular germinal matrix hemorrhage. He also had a patent ductus arteriosus for which indomethacin was given.

One day before the surgery, the neonate developed abdominal distention and deteriorated rapidly with

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evident metabolic acidosis and acute renal insufficiency. Preoperative abdominal ultrasonography revealed severe mechanical obstruction, with an incidental small smooth ovoid hepatic mass of 1.1 x 0.5 cm² in the anterior portion of the left liver, which was echogenic and well demarcated (Figure 2). The radiologist reported this hepatic lesion suspicious for a liver abscess.

Diagnostic assessment and therapeutic intervention.

On the day of surgery, the infant's body weight was 860 g and baseline vital signs just before administering anesthesia were as follows: heart rate (HR) 159 beats/min, blood pressure 38/15 mmHg, and oxygen saturation (SpO₂) 100%. We continuously monitored the invasive arterial blood pressure and goal-directed fluid management was administered with continuous monitoring of pulse pressure variations. The neonate had a percutaneous central venous catheter (PCVC) in situ and a 24 G peripheral venous cannula was secured. The umbilical vein access was removed for the operation. As soon as the peritoneum was opened, a red swelling

was observed in the liver which suddenly ruptured. There was no direct trauma to the liver, but extensive hemorrhage occurred. We immediately initiated a blood transfusion, but the degree of bleeding was very extensive. The patient's blood pressure dropped and cardiac arrest developed soon despite rapid resuscitation. Even after recovery of spontaneous circulation, surgical hemostasis was not successful and massive transfusion continued. The surgeon applied surgical hemostasis for approximately 2 hours, but eventually failed to control the bleeding. Tremendous amount of hemorrhage persisted. The surgeon and anesthesiologist concluded that the patient had a subcapsular hepatic rupture that rarely occurs in premature infants, rather than a simple abscess. The liver was packed with gauze, without further surgical hemostasis. The surgery for mechanical obstruction was hastened for completion. The meconium plug was squeezed out, and the involved small bowel was resected. The total surgical time was 230 min and the estimated blood loss was

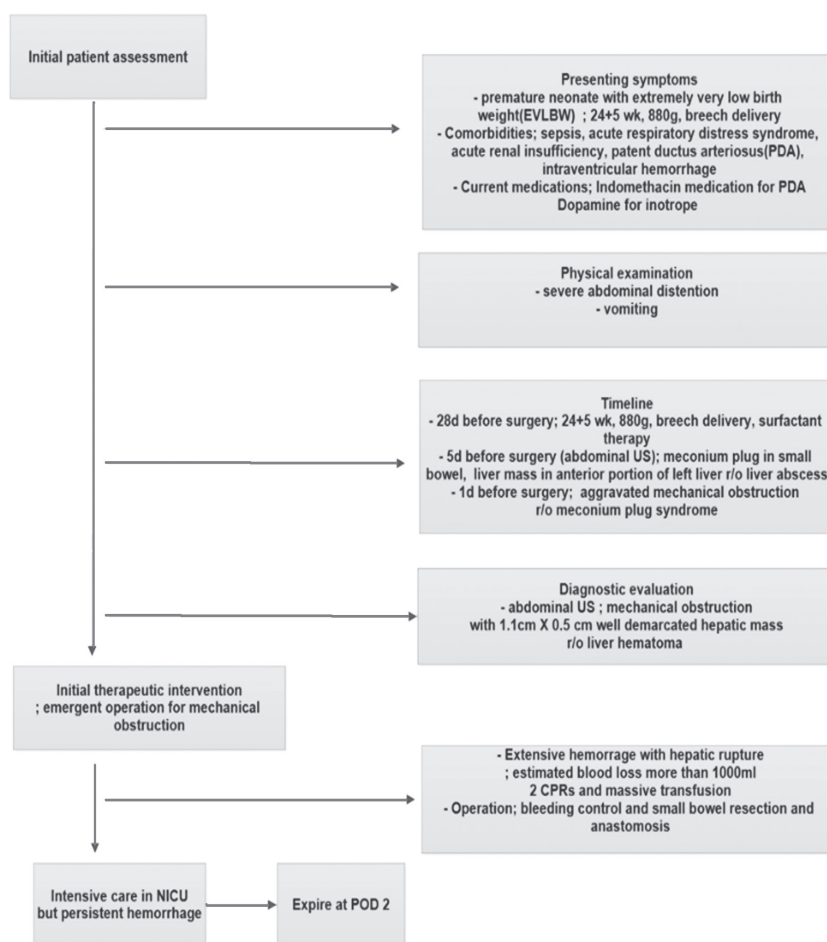


Figure 1 - Patient's flow chart of clinical feature. NICU - neonatal intensive care unit. CPR - cardiopulmonary resuscitation

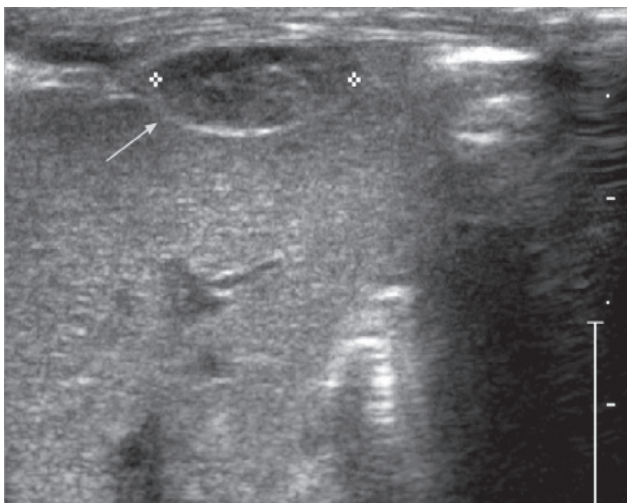


Figure 2 - Preoperative abdominal sonography showed a subcapsular liver hematoma of 1.1 cm x 0.5 cm.

approximately 1000 mL. During peritoneal closure, a second cardiac arrest occurred, with hypoventilation and hypovolemia. The peritoneum was opened again and temporary abdominal closure was performed. The infant was transferred to a neonatal intensive care unit (NICU) under portable ventilator support and a continuous blood transfusion.

Follow-up and outcomes. In the NICU, high-frequency oscillation ventilation with a mean airway pressure of 10 mmHg and 10 Hz frequency was applied. Arterial blood gas analysis showed a PaO₂ of 47 mmHg and base excess of 6.8 mmol/L. A moderate amount of hemoperitoneum and a subcapsular hematoma were observed on the abdominal ultrasound. Hemorrhage persisted, at a rate of >30 mL/h. Brain ultrasonography showed hypoxic brain injury with multifocal hemorrhagic infarcts in both cerebral hemispheres. Despite intensive care, the neonate died 2 days after surgery.

Discussion. We report a case of a ruptured subcapsular hematoma in a preterm neonate undergoing surgery for bowel obstruction.

The incidence of SHL is very low in infants and therefore diagnosis is commonly delayed or misdiagnosed as an abdominal mass or a small cystic tumor. Some cases of SHL have been incidentally found on ultrasonography or computed tomography.^{3,4} In our case, the preoperative hepatic lesion found with ultrasonography was not clearly distinguished from an organizing hematoma, organizing abscess, or any other hepatic mass. Given this, it was possible the radiologist to report the lesion as a hepatic abscess, considering

the clinical features of severe sepsis. Unfortunately, no physician considered that this small hepatic lesion could be a SHL that could result in extensive hemorrhage. Subcapsular hematoma of the liver has a varying spectrum of disease severity. An unruptured SHL is initially asymptomatic. A slowly progressing hematoma manifests with pallor, jaundice, and irritability. These nonspecific manifestations can be followed by sudden catastrophic collapse. If the fragile bond between the hepatic capsule and parenchyma breaks spontaneously or due to direct trauma, massive bleeding can occur, which is usually life-threatening.

Many predisposing factors for SHL have been proposed, such as prematurity, very low birth weight, sepsis, hypoxia, pneumothorax, traumatic labor, traumatic umbilical venous catheterization, cerebral hemorrhage, coagulopathy, and exposure to drugs that lead to a bleeding tendency. The 3 main causes of SHL are fragility of the neonatal liver, trauma, and coagulopathy. The neonatal parenchyma has a scanty fibrous stroma and its capsule is thin, with low elasticity.⁵ The connection between the capsule and parenchyma along the hepatic sinusoids is fragile. Even a small irritation can cause a liver hematoma or rupture. Contractility of the hepatic vein is also poor. Premature or very-low-birth-weight infants are more vulnerable to immaturity; thus, even if there is no trauma, a hematoma may occur naturally.¹

In addition, the birthing process itself is traumatic. Delivery causes compression of the neonatal thoracic cage which pulls or injures the coronary ligaments attached to the inferior surface of the diaphragm. Abnormal conditions during labor such as a breech presentation or oversized infants, increase the incidence of trauma. Traumatic umbilical venous catheterization and cardiopulmonary resuscitation have also been reported to induce hepatic rupture.^{1,2,6} Moreover, many cases of SHL are related to coagulopathy. Congenital abnormalities, such as hemophilia or vitamin K deficiency; poor general conditions, such as hypotension or sepsis and antiplatelet drugs like non-steroidal anti-inflammatory drugs can cause coagulopathy. Associated germinal matrix hemorrhage has also been reported as a risk factor for SHL.² Severely premature infants or underweight babies are also prone to coagulopathy. In our case, several risk factors were present, including premature birth with very low birth weight, breech delivery, sepsis, germinal matrix hemorrhage, and indomethacin exposure for the closure of the patent ductus arteriosus.

Since SHL rupture is extremely rare, a sudden rupture of liver is not usually anticipated. However, if the rupture actually occurs without thorough preparation, it will be very difficult to cope with, as in our case. Therefore, the possibility of SHL should be considered among several diagnoses to be distinguished, when a hepatic lesion is detected in preoperative examination of prematurity.⁷ Most SHLs generally disappear spontaneously or decrease in size on serial ultrasound tracing. Therefore, it was necessary to repeat the ultrasound examinations before the surgery to observe the progress so that a clearer diagnosis could be made.

Management for SHL should be primarily conservative. Transfusion and correction of the coagulopathy is needed, and unnecessarily excessive handling should be avoided. Surgery is reserved for rupture of the hematoma in the peritoneum. However, in the case of an abdominal emergency such as necrotizing enterocolitis or mechanical obstruction, it is difficult to delay surgical correction. Liver injury has been associated with a poor prognosis of abdominal emergencies in premature infants.⁸ Routine abdominal surgical manipulation can result in blunt injury to the liver. Without specific trauma, the SHL can spontaneously tear during opening of the distended abdomen itself. The risk associated with SHL rupture is catastrophic, as evident from this case. Because preoperative diagnosis of SHL may be delayed or missed, the anesthesiologist should confirm the ultrasonic finding of the hepatic lesion. If findings indicative of SHL are present, multidisciplinary discussions among the pediatrician, radiologist, surgeon, and anesthesiologist are required for development of appropriate treatment strategies. The risk of SHL rupture should be carefully considered when determining the treatment strategy. Despite the controversy regarding the treatment modalities for abdominal emergencies in neonates, conservative treatment, such as peritoneal drainage, may be preferred for neonates with SHL.⁹

Once emergent surgery has been decided upon, thorough preparation for anticipated massive bleeding is needed. In ELBW infants weighing less than 1000 g, securing a large bore intravenous line is a significant challenge. In our case, one PCVC and one peripheral intravenous line were secured. However, the long PCVC does not allow rapid infusion of thick blood, and the peripheral line may lose its patency during a massive transfusion. If an emergency laparotomy is scheduled for neonates with the possibility of SHL, a central

venous catheterization or a cutdown vasculotomy is highly recommended. Sufficient blood preparation, as early as at the time of incision, is also necessary.

During the surgery, handling of tissues should be performed carefully. Once liver bleeding begins, surgical hemostasis should be completed as soon as possible. Surface hemostasis is generally known to be ineffective, but pack tamponade has been reported to be effective in some cases.⁵

In conclusion, we presented a case of unexpected fatal hemorrhage associated with SHL rupture in a premature infant. In view of the potential for life-threatening complications, timely detection and multidisciplinary collaboration for the establishment of treatment modalities to avoid rupture are essential to minimize morbidity and mortality. If surgery is unavoidable, anesthesiologists need to be thoroughly prepared for a massive transfusion with substantial vascular access and sufficient blood components to manage catastrophic hemorrhage.

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