Spontaneous peripartum liver haemorrhage presenting as foetal distress

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Abstract

We present the case of a 32-year-old woman who presented at 37 weeks’ gestation with foetal bradycardia. At caesarean delivery, an intra-abdominal haemorrhage was noted before the uterine incision was made. A subsequent exploratory laparotomy revealed the source of the bleeding to be an extensive ruptured haematoma involving both lobes of the liver. Radiologic investigations revealed the presence of multiple cavernous haemangiomata in the liver. The perioperative management of this patient is described, with a brief review of the literature.

Keywords

Peripartum liver haemorrhage; foetal distress.

Introduction

Spontaneous peripartum liver haematoma is a rare, but well-described complication of pregnancy with a reported incidence of between 1 in 45,000\textsuperscript{1} and 1 in 250,000\textsuperscript{2} deliveries. In more than 95\% of cases, it is associated with hypertensive disorders of pregnancy\textsuperscript{3}, with or without the HELLP (haemolysis, elevated liver enzymes, low platelets) syndrome. In a minority of cases, the focus of haemorrhage is a hepatic neoplasm. Traditional management was surgery\textsuperscript{4} and mortality was high, but other treatment options have been described including intensive supportive care\textsuperscript{5} and selective hepatic artery embolization\textsuperscript{6}. We describe the presentation and perioperative management of a parturient with a ruptured liver haemangioma whose only presenting symptom was foetal distress, and in whom intensive supportive care was successful.

Case report

A 33 year-old gravida 2 para 1 with a previous spontaneous vaginal delivery presented at 37 weeks’ gestation with the onset of uterine contractions. She had no previous medical history and in particular had no evidence of gestational hypertensive disease. No signs or symptoms attributable to the liver were elicited on initial assessment. Foetal monitoring was initiated and a
prolonged foetal bradycardia was noted. She was immediately transferred to the delivery unit for caesarean delivery.

The patient was haemodynamically stable, with a heart rate of 85 beats/min and blood pressure of 115/75 mmHg. Routine blood samples were taken, and the results, which were not available before the commencement of surgery revealed haemoglobin 10.5 mg/dl; white cell count 16,600/dl; platelets 98,000/dl; prothrombin time 9.8 s (normal 9–12 s), activated partial thromboplastin time 29.9 s (normal 28–33 s); albumin 24 g/l, bilirubin 11 nmol/l, aspartate transaminase 234 IU/dl (normal range 5–45 IU/dl) and alkaline phosphatase 102 IU/dl (normal 40–110 IU/dl). The renal profile was normal. Specific tests for haemolysis were not performed on admission and were not possible later due to massive transfusion.

Anaesthesia was induced with a classic rapid sequence technique, and maintained with isoflurane in a 50:50 nitrous oxide/oxygen mix. Surgery was commenced as soon as the airway was secure. The abdomen was opened through a Pfannensteil incision and on entry into the peritoneal cavity, but before uterine incision, copious altered blood was found within the pelvis. A single live male foetus was delivered through a lower segment uterine incision, with Apgar scores of 7 and 9 at 1 and 5 min, respectively. A 50% posterior placental abruption was noted; there was no significant occult intra-uterine haemorrhage. A degree of haemodynamic instability developed after the delivery, and the patient was aggressively resuscitated with a total of 3 l of crystalloid, 4 units of packed cells and 4 units of fresh frozen plasma. After uterine haemostasis was achieved, the pelvis and lower abdomen were thoroughly explored but no focus of bleeding was identified. A surgical site drain was inserted and the abdomen closed. The patient was transferred to the intensive care unit under sedation. Estimated intra-operative blood loss was 2 l.

A steady haemorrhage continued over the next 12 h, with a total loss of 2 l from the abdominal drain. The patient was referred for a general surgical opinion and a laparotomy performed. A large ruptured subcapsular liver haematoma was found, involving most of the superior surface of the left lobe. A trickle of ongoing haemorrhage was noted, but as the patient was by this time haemodynamically stable. It was decided to abandon surgery in favour of further imaging and consideration of selective hepatic artery embolization.

A computed tomography (CT) scan of the abdomen was performed and revealed a large area of low attenuation corresponding to the left lobe haematoma (white arrow), but also an area of mixed attenuation in the right lobe (black arrow) (Fig. 1). This was thought to represent a second haematoma. No specific predisposing lesion was identified by CT scans, and a magnetic resonance imaging (MRI) scan was obtained, which demonstrated a haematoma and inferior to it a cavernous haemangioma (Fig. 2). A haemangioma was also noted in the right lobe. Selective embolization was not possible due to the bilateral nature of the haemangiomas and the patient was transferred back to the intensive care unit for ongoing supportive care.

Blood products were transfused as indicated by laboratory parameters, and the patient remained haemodynamically stable. A continuous infusion of aprotinin was commenced, and the

![Fig. 1. CT scan through the liver demonstrating a large area of low attenuation corresponding to the left lobe haematoma (white arrow), but also an area of mixed attenuation in the right lobe (black arrow).](image)
ongoing haemorrhage ceased within 12 h. During the perioperative period, a total of 18 units of packed cells, 18 units of fresh frozen plasma, 2 standard adult doses of platelets (each containing pooled platelets from 10 donors), and 2 units of cryoprecipitate were infused. Extubation was uncomplicated, and the patient was discharged from the intensive care unit after a total of 5 days. She was discharged from hospital 6 days later, and has suffered no complications after 6 months follow-up.

Discussion

Spontaneous liver rupture in pregnancy was first described by Abercrombie in 1844[7], and over 200 cases have been reported. The most useful survey of peripartum liver rupture in the obstetric literature is the review of Rinehart et al.[3] of 141 cases since 1961. The patient profile that emerges is of a 30-year-old woman with pre-eclampsia (in \( >95\% \) of cases) with a history of multiple pregnancies presenting with either epigastric pain (69.2%), hypertension (65.6%) or haemodynamic compromise (56%). Shoulder pain is a less sensitive sign, present in only 20.5% of cases, but it may be more specific. Barton and Sibai[8] found radiologic evidence of liver haemorrhage or infarction in 16 of 33 patients with HELLP syndrome who complained of right upper quadrant pain with neck or shoulder pain. The severity of derangement of liver function tests did not correlate with the severity of the liver injury when assessed radiologically. It is difficult to assess the incidence of HELLP syndrome in the series reported by Rinehart et al.[3], as the liver rupture itself causes transaminitis and low platelets and the results of specific tests for haemolysis are seldom recorded.

When presenting with typical symptoms, the presence of a hepatic haematoma may be confirmed on cross-sectional imaging studies. Haemangiomas are well characterized on MRI, being typically hyper-intense on T2-weighted MR imaging, with a centripetal filling pattern after administration of gadolinium[9]. In the current case, no symptoms were present on presentation and emergent caesarean delivery was performed due to suspected foetal compromise. The
subsequent exploratory laparotomy was also performed as an emergency due to continued blood loss and concern with regard to a missed intra-abdominal haemorrhage.

The pathophysiology in these cases is still unclear, but subcapsular haemorrhage may be secondary to periportal fibrin deposition, which causes microvascular congestion and infarction\[^{10}\]. Spontaneous rupture of liver neoplasms, including cavernous haemangioma\[^{11}\], has also been described in the absence of pre-eclampsia toxaemia (PET), and the pathophysiology in these cases may be varied. Liver haemangioma are common benign tumours of capillary endothelium, found in up to 20% of autopsy examinations\[^{12}\]. They are known to enlarge during pregnancy\[^{13}\], probably as a result of increases in circulating volume as well as of humoral changes. It is possible for the haemangioma itself to become a predisposing factor for haemorrhage; in cases of large or multiple haemangiomata, platelets may become sequestrated and activated in the tumours, resulting in a consumptive coagulopathy and disseminated intravascular coagulation (the Kasabach-Merrit syndrome)\[^{14}\].

In this case, none of the diagnostic criteria for PET was met. The HELLP syndrome may present in the absence of hypertension and proteinuria\[^{15}\]; this patient had a significant thrombocytopenia and increased liver enzymes at presentation, and so may have had atypical HELLP. However, the thrombocytopenia and increased transaminases might have been a consequence rather than a cause of the liver haematoma. The diagnosis is complicated further by the possibility that the coagulopathy with which the patient initially presented may itself have been the primary cause of the haemorrhage, due to the Kasasbach-Merritt syndrome. Hence, unravelling the sequence of cause and effect is very difficult. We suspect that the initial focus of haemorrhage in each liver lobe was a cavernous haemangioma, which was obliterated by the haemorrhage, and thus not identifiable on the initial liver imaging.

This case illustrates the difficulty of diagnosing this life-threatening, but fortunately rare complication of pregnancy. The patient complained of no symptoms suggestive of liver rupture, and this is not unusual in such cases; typically, the presenting symptoms are epigastric pain and hypertension, which are relatively common in the at-risk population. It also demonstrates the possibility of successful non-surgical management of liver rupture; in cases where the patient remains haemodynamically stable, radiologic intervention or intensive supportive care may be more appropriate, and may allow transfer of the patient to a specialist centre. This case raises the possibility of a diagnosis for increased liver enzymes and low platelets other than the HELLP syndrome; if this patient's liver haematoma had not ruptured, it would likely have gone unnoticed, and the thrombocytopenia and deranged liver function tests might have been attributed to the HELLP syndrome. In Barton and Sibai's series\[^{9}\], 9 of 16 cases had an unruptured subcapsular or intraparenchymal haematoma; without liver imaging these would presumably have gone undiagnosed. We wonder how many other such lesions escape notice. This reinforces the need to have a low threshold for scanning the liver if any symptoms attributable to it are present; their absence, however, may be falsely reassuring.

References