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Catastrophic Haemorhage: A Case Report on Spontaneous Rupture of Splenic Hemangioma in Pregnancy

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Authors' contributions

This work was carried out in collaboration between all authors. Author SNSI managed the literature review and wrote the first draft of the manuscript. Authors WFM and NBM compiled the case. Author MFA contributed the intraoperative findings while author NAA contributed the pathology report. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Spontaneous splenic rupture in pregnancy is an uncommon event in the absence of trauma. Since the diagnosis is indirect, the treatment is often delayed. In this report, we highlighted the case of a twenty-six years old Chinese, a primigravida at 28 weeks gestation who presented with hypovolemic shock preceded by vague abdominal discomfort at home. Both physical and sonographic examination revealed massive hemoperitoneum with intrauterine death most likely due to splenic rupture. The surgery done was emergency laparotomy and hysterotomy with abdominal exploration. Intraoperatively, active bleeding was identified at the lateral pole of the spleen consistent with splenic rupture. Through this report, we attempt to illustrate the importance of immediate recognition of this surgical emergency to prevent catastrophic outcome.

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1. INTRODUCTION

Spontaneous splenic rupture (SSR) is an uncommon entity. It carries the incidence of 0.1-0.5% in general population. The first case of splenic rupture during pregnancy ever described in the literature was in 1803 [1] and there were merely 38 reported cases from 1950 to 2011 [2]. Yakub R et al cited that the greatest incidence of SSR in pregnancy occurred in the third trimester and the puerperium and it usually occurs in a diseased spleen. It has been suggested that splenic enlargement, increased blood volume and reduced peritoneal cavity in pregnancy could be implicated in the pathogenesis of splenic rupture [2]. Some of the aetiologies include haematological malignancies such as lymphoma or leukaemia, systemic infections like infectious mononucleosis or malaria, local inflammatory disorder and primary splenic neoplams such as angiosarcoma and haemangioma [3]. Despite the fact that haemangioma is the commonest primary neoplasm of the spleen, only a few cases of its rupture were described in pregnancy or puerperium [4]. We are highlighting a case involving spontaneous rupture of splenic haemangioma in a Chinese primigravida at the third trimester.

2. CASE REPORT

A twenty-six-year-old Chinese primigravida at 28weeks gestation with no previous medical illness referred to our center with maternal collapse secondary to suspected placental abruption with intrauterine death. She presented with irreversible hypovolemic shock after the sudden onset of generalised abdominal pain and loose stool; denied any pervaginal bleeding.

Her abdomen was tense, uterus consistent with 28 weeks gestation and no cervical changes upon vaginal assessment. Initial investigations revealed Hb 54 g/dl with severe metabolic acidosis and acute kidney injury. Ultrasonographic examination revealed a non viable intrauterine fetus corresponding to 28 weeks with no evidence of retroplacenta clot. There was an extensive hemoperitoneum; spleen was enlarged with mixed echogenicity possible of blood clot consistent with features of the splenic lesion. She desaturated and was intubated with endotracheal tube size 7, anchored at 20 cm for impending respiratory collapse. The patient was resuscitated with seven pints blood and two litres of crystalloids. Surgical team was alerted and after reviewing the patient, we agreed to proceed with laparotomy.

Emergency exploratory laparotomy was performed and showed a hemoperitoneum of 4.5 litres. Active bleeding was identified with an area of contusion at splenic bed; splenic haematoma at lateral lower pole with 3cm rupture area. Splenectomy was performed and haemostasis was secured. Hysterotomy was done and a lifeless fetus was delivered weighing 1.2 kg. There was no evidence of placental abruption. Intraoperatively she was transfused with another four pints of blood and two cycles of DIC regime. Post operatively the patient required ionotropic support and was stabilized in Intensive Care Unit. Despite vigorous resuscitation with massive blood transfusion, she remained oliguric and was planned for Continuous Veno-Venous Hemofiltration (CVVH). She died on day 3 of admission due to multiorgan failure. Histopathology reported a spleen weighing 235 g measuring 115 x 98 x52 mm. There was ruptured capsule area between the inferior and anterior border. Microscopically there was intraparenchymal haemorrhage with multiple abnormal dilated vascular spaces seen near capsule and adjacent to the haemorrhagic area, lined by single layer of flat endothelium. These findings were consistent with ruptured spleen secondary to cavernous haemangioma.

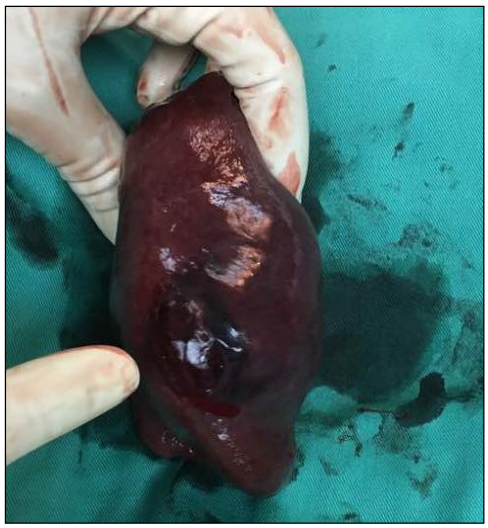

Fig. 1. The image shows anterior aspect of the spleen with area of haematoma measuring 5x6 cm with central area of rupture 3 cm

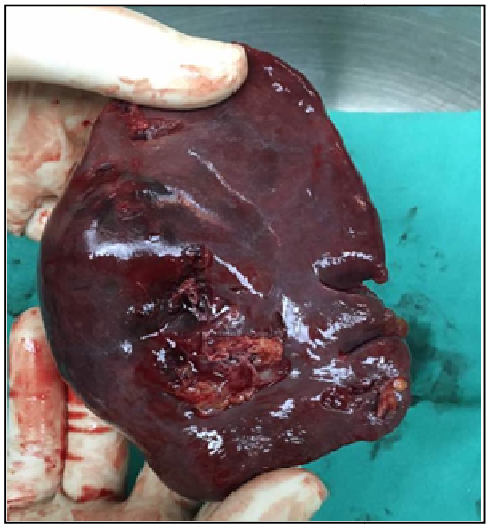

Fig. 2. Active bleeding from ruptured part of splenic haematoma

3. DISCUSSION

Splenic rupture in pregnancy is clinically challenging to diagnose since it has similar symptoms and signs with other life threatening condition such as placental abruption, uterine rupture and ruptured abdominal pregnancy [2]. Maternal death in splenic rupture is most likely due to haemorrhagic shock. This lead to an acute reduction in uteroplacental perfusion resulting in fetal distress and subsequently fetal death[3]. Rapid multidisciplinary management may enhance the patient's chance of survival [5,6].

The clinical features at presentation depend on the severity and timing from onset; ranging from mild vague left upper quadrant abdominal pain, generalized tenderness and rigidity, to a full blown hypovolemic shock with pallor, hypotension and tachycardia.

Diagnosis is most often performed by clinical features and several imaging tests are cited in the literature. The use of paracentesis to ascertain the likelihood of intraperitoneal haemorrhage is previously described [3]. However it has been replaced by abdominal ultrasound which is non-invasive and more practical to demonstrate the intraperitoneal fluid accumulation as justified by Gedik et al. They further illustrated the simplicity of ultrasound, which can be used at the bedside at Emergency Unit. Computed Tomography (CT) on the other hand is more precise in predicting rupture and has the ability to assess the severity of the bleeding, however, it is more beneficial for haemodynamically stable patients. Radiographic features include an enlarged spleen, haemoperitoneum and perisplenic haematoma [7]. Confirmatory diagnosis is made by surgery as well as histopathology; Kocael et al. described a splenectomy materials weighing >200 g and measuring 110 x 70 x 50 mm in size were considered as splenomegaly [8]. In gross cut surface, the spleen contained cystic, blood-filled spaces of varying sizes while histologically, these vascular spaces are lined by a single flat layer of endothelium [9].

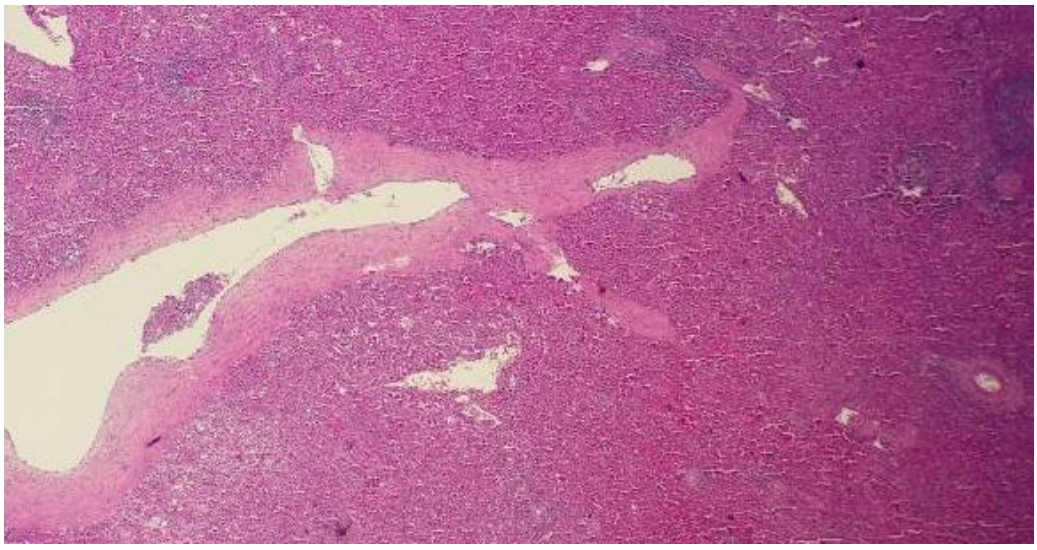

Fig. 3. Microscopic view of the spleen showed multiple thick and thin walled blood vessels with abnormal branching pattern

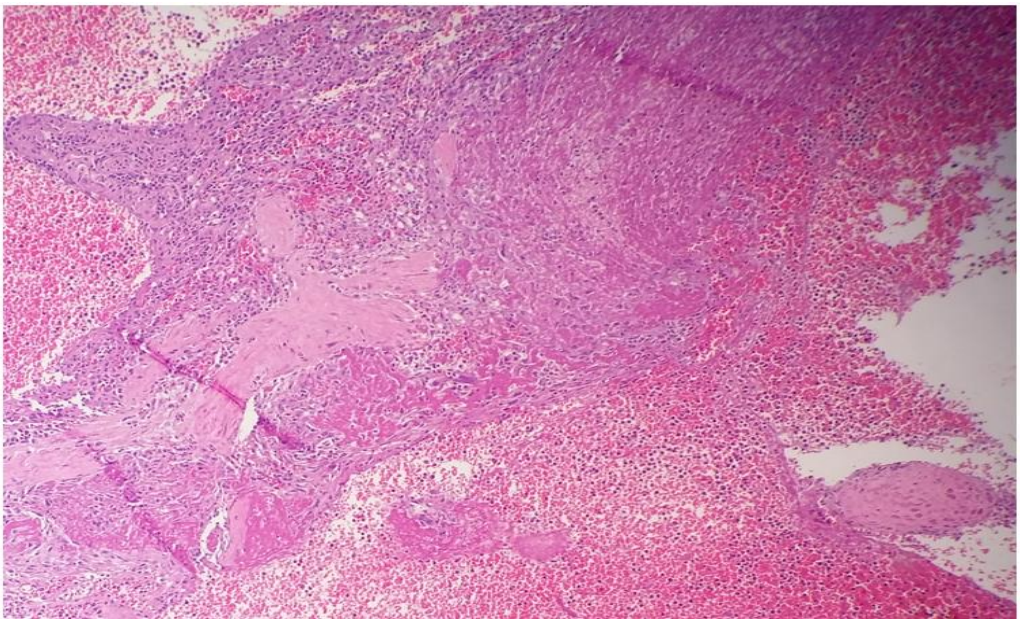

Fig. 4. Image shows thrombosis within abnormal blood vessels

Due to paucity of data in the literature regarding definite management of splenic rupture in pregnancy, total splenectomy remained as the standard of care. However, Yakub et al suggested an alternative approach in hemodynamically stable patient [10]. Conservative management can be adapted in patients with minimal blood loss. In fact, Kocael et al quoted the success rate of conservative treatment in SSR induced by infectious disease is about 80% [8]. Another option would be interventional radiology, which may spare the risk of surgery. When surgical approach is necessary, laparoscopic surgery is preferable to preserve the immune function of the spleen [10].

4. CONCLUSION

In conclusion, the presentation of splenic rupture in pregnancy can be puzzling thus, a high index of suspicion accompanied with aggressive resuscitation and prompt management may improve the outcome for both mother and fetus.

CONSENT

As per international standard or university standard, patient's consent has been collected and preserved by the authors.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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