Polish Annals of Medicine

Journal homepage: https://www.paom.pl

Case report

Uterine rupture mimicking Wunderlich's syndrome in pregnancy: An unfortunate case

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Article info

Article history
Received January 17, 2021
Accepted: March 18, 2021
Available online: November 18, 2021

Keywords
Acute abdomen
Haemorrhagic shock
Nephrectomy
Pregnancy complications

Doi
https://doi.org/10.29089/2021.21.00201

Abstract

Introduction: Wunderlich's syndrome (WS), characterized by non-traumatic renal haemorrhage into the subscapular and perinephric space is a rare entity in pregnancy.

Aim: This article highlights the incidental discovery of a pregnant woman with WS that resulted in emergency nephrectomy.

Case study: A 31-year-old gravida 4 para 3 female with 3 previous caesarean sections presented with acute abdomen and was in shock. The abdominal ultrasound revealed gross haemoperitoneum. With the preoperative diagnosis of a uterine rupture, surgical exploration was done showing an extensive right perinephric hematoma and active bleeding from the renal hilum. No renal tumor or pseudoaneurysm of the renal hilum was noted. Emergency nephrectomy was performed. Unfortunately, the foetus did not survive the ordeal.

Results and discussion: WS occurs as a result of renal neoplasms, idiopathic causes, vascular diseases, infection, and miscellaneous. Ultrasonography can help to identify the perinephric hematoma, meanwhile, colour and/or spectral Doppler can aid in the detection of vascular pathologies. Contrast-enhanced computed tomography is still the imaging modality of choice. In pregnancy, a magnetic resonance imaging would be a better modality, avoiding radiation exposure to the foetus and consequent foetal malformations. Treatment includes arterial embolization and/or operative management such as nephrectomy.

Conclusions: WS in pregnancy is a rare clinical entity requiring a high index of clinical suspicion for diagnosis. WS needs to be considered in pregnant patients presenting with shock with the presence of perinephric hematoma. A multidisciplinary approach is essential in providing optimum care.

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1. INTRODUCTION

Wunderlich's syndrome (WS), characterized by non-traumatic renal hemorrhage into the subscapular and perinephric space is a rare entity in pregnancy. It was first described by Carl Reinhold August Wunderlich in 1856, is a rare and life-threatening condition characterized by non-traumatic renal hemorrhage into the subscapular and perinephric space.[1] The classic signs of WS include Lenk's triad of flank pain, abdominal mass and hypovolaemic shock. WS is a very rare clinical entity, more so in pregnancy.[1]

2. AIM

We describe a case of WS in pregnancy and the management approach.

3. CASE STUDY

A 31-year-old gravida 4 para 3 at 19 week's gestation presented to the emergency department with an acute onset of lower abdominal pain radiating to the back, which was associated with pre-syncopal attack. She denies per vaginal bleeding, haematuria or any history of trauma. She has 3 previous lower segment caesarean sections for her past pregnancies. On physical examination, she was evidently in shock. She was pale, tachycardic (110 bpm) and hypotensive (80/44 mm Hg). Her abdomen was soft, with appropriate distension for her gravid state and she had marked general abdomen tenderness, more so over the Pfannensteil scar. A focused obstetric ultrasound showed gross free fluid in all four quadrants of the abdomen, with a viable intrauterine pregnancy corresponding to 18 weeks of gestational age. Initial investigations showed a haemoglobin of 7 (normal: 10–12 g/L), white blood cell count of 19.6 (normal: 4–11 $\times$ 10^9/L), platelet of 86 (normal: 140–400 $\times$ 10^3/L) and international normalized ratio of 1.75. Her renal profile was within normal limits.

The patient was resuscitated with the appropriate fluids and blood products but was not responded. She was subsequently brought to the operating theatre for emergency exploration with the diagnosis of uterine rupture. Intraoperatively, gross haemoperitoneum was noted with fresh blood mixed with clots. Intraperitoneal hematoma was noted, extending from the pelvis to the right retroperitoneal zone 2 area. The uterus however, was intact with no evidence of uterine rupture. The patient continued bleeding profusely despite ligating the right suspensory ligament of ovary which was thought to be the culprit. The surgical unit was consulted on-table and the source

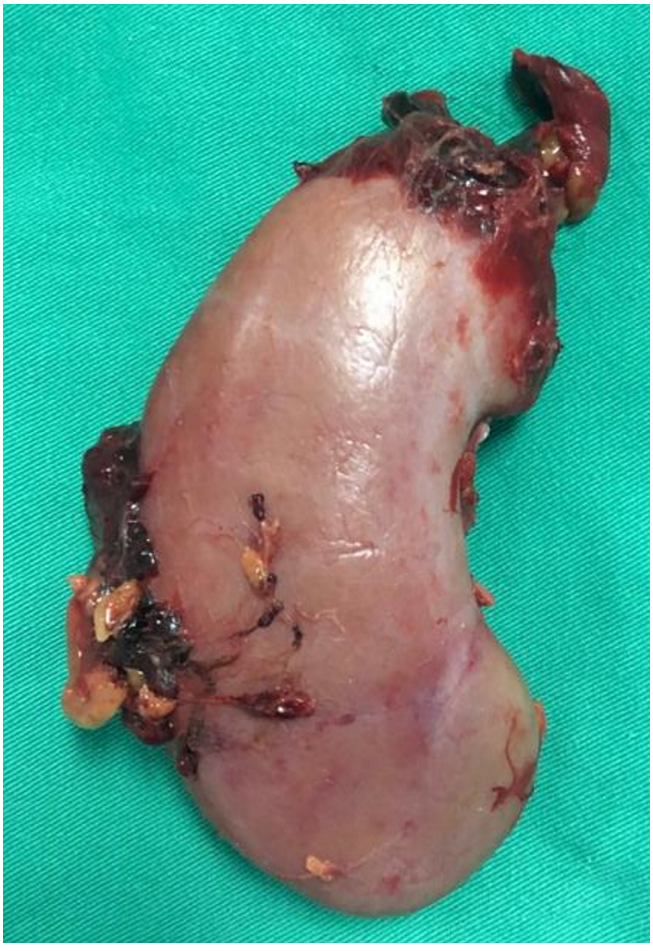

Figure 1. Intraoperative specimen of the right kidney; lateral view. Renal parenchyma is grossly normal.

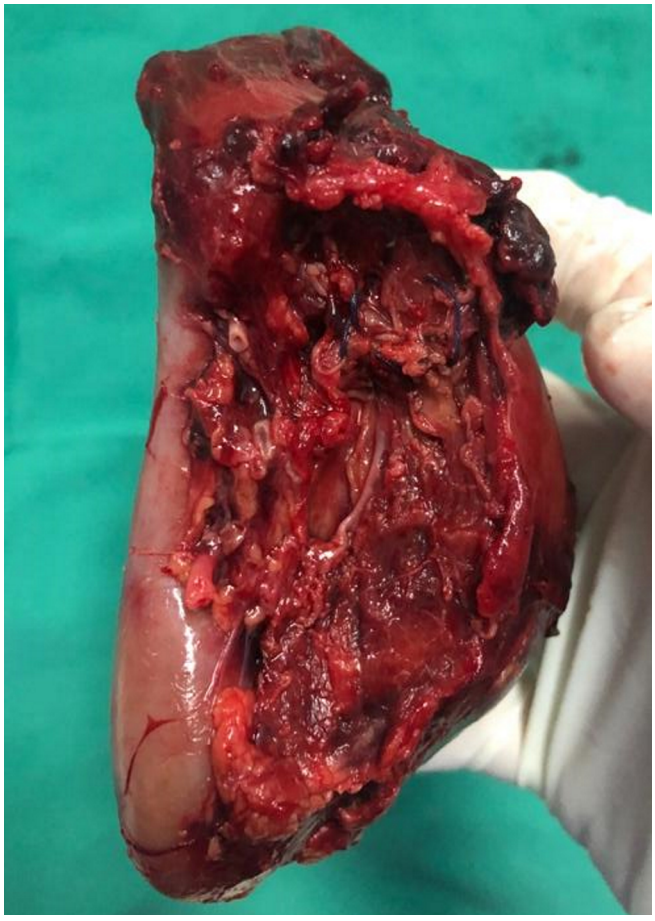

Figure 2. Medial view of the right kidney with visible renal hilar vessels. Breach in the renal capsule due to a large subcapsular haematoma extending to the perinephric region.

of bleeding was found to be originating from the right renal hilum. There was extensive right perinephric haematoma with active bleeding from the renal hilum. There was no pseudoaneurysm over the hilum, and the renal parenchyma appears grossly normal with no evidence of a renal tumor, parenchyma laceration or rupture. Emergency nephrectomy was performed as the patient was hemodynamically unstable. Once haemostasis was achieved, on-table ultrasonography showed that the foetus did not survive the ordeal. Hysterotomy was performed to remove the foetus. The patient was sent to the intensive care unit postoperatively and recovered well with no complications. She was discharged 5 days later. Histopathological examination of the right renal concluded that the kidney was normal with no evidence of tumor such as angiomyolipoma. The visualized renal hilar (Figure 1 and 2) vessels appeared normal with no evidence of a pseudoaneurysm.

4. RESULTS AND DISCUSSION

The aetiology of WS includes renal neoplasms (61.2%), idiopathic causes (38.0%), vascular diseases (17.0%), infection (2.4%), and miscellaneous (12.7%).[2,3] Majority of WS in pregnancy are due to renal angiomyolipoma. Our reported case is an idiopathic cause as it showed neither renal neoplasms nor vascular abnormalities. Although ultrasonography may identify a perinephric hematoma, color and/or spectral Doppler can aid in the detection of vascular pathologies. Contrast-enhanced computed tomography is still the imaging modality of choice. However, in the context of pregnancy, a magnetic resonance imaging would be a better modality, avoiding radiation exposure to the foetus and consequent foetal malformations.[4,5] In our patient, WS was never the initial diagnosis that was entertained. Instead, her preoperative risk factors of multiparity and caesarean sections in addition to gross haemoperitoneum on sonography, the clinical suspicion of uterine rupture was very high.

Treatment should be initiated as a life-saving procedure before identification of the definite aetiology. Initial management is targeted at fluid resuscitation, blood transfusion and reversal of coagulopathy.[6–8] For patients in shock, or ongoing haemorrhage, definitive treatment is required by arterial embolization and/or operative management such as nephrectomy.[5,6] Selective angiographic embolization with preservation of functional organ parenchyma can be performed with the presence of interventional radiological expertise.[9] In contrast to embolization, surgery as in our case has the important advantage of allowing for pathologic evaluation for a definitive diagnosis.[10] Foetal distress or matured gestation may make nephrectomy more favourable as it allows for concurrent delivery.[7]

5. CONCLUSIONS

WS in pregnancy is a rare clinical entity requiring a high index of clinical suspicion for diagnosis. WS needs to be considered in pregnant patients presenting with shock with the presence of perinephric hematoma. A multidisciplinary approach is essential in providing optimum care.

Conflict of interest
Authors declare that there is no conflict of interest.

Funding
None declared.

Acknowledgements
We would like to thank the Director General of Health Malaysia for his permission to publish this article as a case report. In addition, we also thank for those who are directly or indirectly involved in managing this patient.

Ethics
Informed consent was obtained from the patient.

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