Case Reports in Women's Health 39 (2023) e00524

Contents lists available at ScienceDirect

Case Reports in Women's Health

journal homepage: www.elsevier.com/locate/crwh

Mid-trimester spontaneous rupture of a bicornuate uterus: A case report

Heba Abu Saleem [a], Yara Edweidar [b], Mutaz Abu Salim [c], Ismaiel Abu Mahfouz [d,*]

[a] The Specialty Hospital, Amman, Jordan
[b] Al-Bashir Hospital, Amman, Jordan
[c] Faculty of Medicine, Al Yarmouk University, Irbid, Jordan
[d] Faculty of medicine, Al-Balqa Applied University, Salt, Jordan

ARTICLE INFO

Keywords:
Bicornuate uterus
Malformation
Primigravida
Uterine rupture

ABSTRACT

Bicornuate uterus (BU) is a rare congenital anomaly that may present with various obstetric complications, and very rarely may be a risk factor for uterine rupture, even of an unscarred uterus.

A 21-year-old primigravida woman, at 19 weeks and 5 days of gestation, presented with severe abdominal pain and features of hypovolemic shock. Urgent laparotomy showed a large hemoperitoneum, a ruptured left horn of a BU and a dead fetus in the abdominal cavity. Excision of the ruptured left horn was performed and the uterine defect was sutured. Postoperative recovery was uneventful. Bicornuate uterus is a rare uterine anomaly and is associated with various obstetric complications at different gestational ages. Ruptured uterus should be considered in the differential diagnoses of acute abdominal pain and a picture of hypovolemia in women with mid-trimester pregnancies.

1. Introduction

Bicornuate uterus (BU) is a rare congenital uterine anomaly that results from failure of fusion of the two Mullerian ducts during embryogenesis, and is related to significant adverse reproductive outcomes [1]. The prevalence of BU varies according to the study population, being 0.4% in the general population and from 1.1% to 4.7% among women with miscarriage and sub-fertility [2]. Women with BU may also present with various obstetric complications, such as mid-trimester miscarriages, preterm labor and delivery, malpresentation, and antepartum and postpartum hemorrhage. The live birth rate among women with BU is only 60% [3].

Uterine rupture is an obstetrical emergency that is associated with high morbidity and mortality and is usually related to previous uterine surgery, such as prior caesarean section and myomectomy; therefore, it is very rarely reported in primigravida women with no prior uterine scarring [4].

A series from the United States reported the incidence of uterine rupture in primigravidae with no prior uterine incision to be from 1 in 7643 to 1 in 16,849 deliveries [5].

However, while rare, BU can be the only risk factor for uterine rupture in primigravida women. Walsh et al. [5] in a review of all case reports, spanning 60 years, of uterine rupture in primigravida women identified 36 cases, of which 25 were with an unscarred uterus, and among them four were women who had a BU.

Here we report the presentation and management of a primigravida woman who had second-trimester spontaneous rupture of the left-sided horn of a BU.

2. Case Presentation

The case concerns a 21-year-old healthy primigravida woman, who previously had had no surgical operations and was at 19 weeks and 5 days of gestation. The pregnancy was unplanned and she had irregular antenatal care visits. The woman presented to the emergency room with severe abdominal pain of two hours' duration, which was associated with progressive abdominal distension, dizziness, and shoulder tip pain; she had no associated vaginal bleeding.

On arrival at the emergency department, she was unconscious, her blood pressure was 70/40 mmHg, pulse rate 130/min and respiratory rate 24/min. Abdominal examination showed a distended abdomen reaching the xiphoid process and generalized abdominal tenderness. Bedside abdominal ultrasound revealed fluid filling the abdominal cavity with clots and a non-viable fetus in the abdominal cavity with fetal biometry in keeping with 20 weeks of gestation. The clinical suspicion was of a ruptured uterus. Immediate resuscitation was started,

* Corresponding author at: Department of Obstetrics and Gynaecology, Faculty of Medicine, Al Balqa Applied University, P O Box 19117, Al-Salt 19117, Jordan.
E-mail address: ismaiel.mahfouz@bau.edu.jo (I.A. Mahfouz).

https://doi.org/10.1016/j.crwh.2023.e00524
Received 23 June 2023; Received in revised form 27 June 2023; Accepted 28 June 2023
Available online 30 June 2023

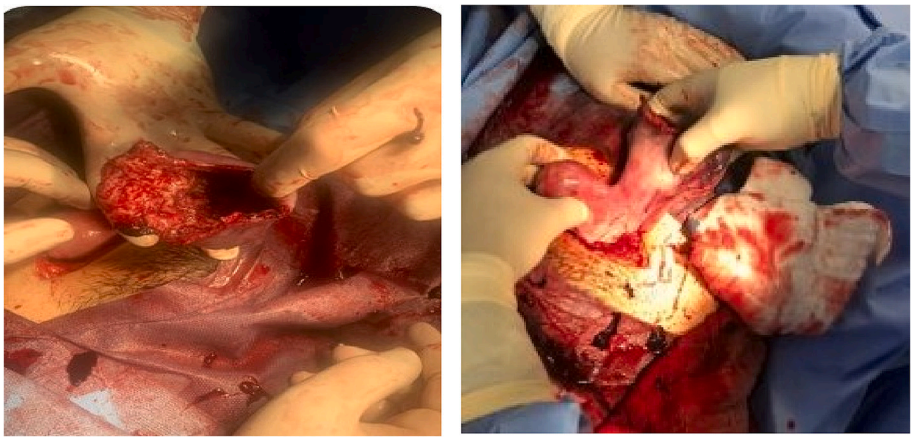

Fig. 1. (a,b). Ruptured left horn at the fundus.

and a decision was made for emergency laparotomy.

Intraoperative findings (Fig. 1) showed a massive hemoperitoneum of around two liters of blood and blood clots, a bicornuate uterus with a ruptured left horn at the fundus, in addition to non-viable normal looking fetus in the abdominal cavity. The weight of the fetus was 300 g.

Excision of the ruptured left horn was performed at its junction with the right horn and the defect resulting from the excision was sutured in 2 layers using delayed absorbable sutures, and an intraabdominal drain was inserted. The woman was transfused four units of packed red blood cells and four units of fresh frozen plasma and was transferred to the intensive care unit (ICU), where she stayed for one night. The woman had an uneventful recovery and was discharged home on the fourth postoperative day. Outpatient follow-up showed an uneventful recovery.

3. Discussion

Bicornuate uterus is a rare malformation that is considered a risk factor for uterine rupture even in the absence of prior uterine scar, as in our case [3]. A suggested explanation is the presence of the fibrous band that connects the two uterine corpora, which limits uterine expansion, and this predisposes to uterine rupture as gestation advances.

The vast majority of women with BU are asymptomatic and the diagnosis is usually made during pregnancy or delivery when complications arise, or during abdominal surgeries such as hysterectomy [6]. In our case, the uterine anomaly was identified intraoperatively, reflecting failure to diagnose the anomaly earlier.

The clinical presentation of uterine rupture at an earlier gestational age is usually nonspecific; therefore, the diagnosis is likely to be more difficult. Symptoms and signs may include sever acute abdominal pain, vaginal bleeding, features of hypovolemia such as dizziness, loss of consciousness, in addition to hypotension and tachycardia. However, not all cases of uterine rupture present with these classical features; therefore, it is critical to keep a high index of suspicion when managing women who present with some or all of these symptoms and signs regardless of the parity or gestational age [7]. The woman in our case presented to the emergency department with severe abdominal pain and distension with loss of consciousness, hypotension and tachycardia, which were suggestive of hypovolemia, and bedside ultrasound scan confirmed the presence of hematoperitoneum.

The recommended surgical treatment of ruptured BU is either resection of the ruptured horn followed by repair or hysterectomy [8]. Our management included immediate blood, blood products and fluid replacement therapy aiming for maternal hemodynamic stabilization, followed by exploratory laparotomy excision of the ruptured left uterine horn and repair of the uterus.

Psychological support and contraceptive advice are essential in postoperative management because pregnancy is discouraged for at least one year after surgery [9].

4. Conclusion

Bicornuate uterus is a rare uterine anomaly and is associated with various obstetric complications at different gestational ages. Ruptured uterus should be considered in the differential diagnoses of acute abdominal pain and a picture of hypovolemia in women with mid-trimester pregnancies.

Contributors

Heba Abu Saleem contributed to conception of the case report, acquiring and interpreting the data, undertaking the literature review and drafting the manuscript.

Yara Edweidar contributed to patient care, conception of the case report and acquiring and interpreting the data.

Mutaz Abu Salim contributed to drafting the manuscript and undertaking the literature review.

Ismaiel Abu Mahfouz contributed to drafting the manuscript, undertaking the literature review and revising the article critically for important intellectual content.

All authors approved the final submitted manuscript.

Funding

No funding from an external source supported the publication of this case report.

Patient consent

Consent was obtained from the patient to publish the clinical details and the images included.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

References

[1] A. Ludwin, S.M. Pfeifer, Reproductive surgery for müllerian anomalies: a review of progress in the last decade, Fertil. Steril. 112 (3) (2019 Sep) 408–416, https://doi.org/10.1016/j.fertnstert.2019.07.005.

[2] Y.Y. Chan, K. Jayaprakasan, J. Zamora, J.G. Thornton, N. Raine-Fenning, A. Coomarasamy, The prevalence of congenital uterine anomalies in unselected and high-risk populations: a systematic review, Hum. Reprod. Update 17 (6) (2011 Nov-Dec) 761–771, https://doi.org/10.1093/humupd/dmr028.

[3] B. Nitzsche, M. Dwiggins, S. Catt, Uterine rupture in a primigravid patient with an unscarred bicornuate uterus at term, Case Rep. Womens Health 15 (2017 Mar 28) 1–2, https://doi.org/10.1016/j.crwh.2017.03.004.

[4] D.A. Miller, T.M. Goodwin, R.B. Gherman, R.H. Paul, Intrapartum rupture of the unscarred uterus, Obstet. Gynecol. 89 (5 Pt 1) (1997 May) 671–673, https://doi.org/10.1016/s0029-7844(97)00073-2.

[5] V. Catanzarite, L. Cousins, D. Dowling, S. Daneshmand, Oxytocin-associated rupture of an unscarred uterus in a primigravida, Obstet. Gynecol. 108 (3 Pt 2) (2006 Sep) 723–725, https://doi.org/10.1097/01.AOG.0000215559.21051.dc.

[6] S. Jayaprakash, L. Muralidhar, G. Sampathkumar, R. Sexsena, Rupture of bicornuate uterus, BMJ Case Rep. 2011 (2011 Oct 28), bcr0820114633, https://doi.org/10.1136/bcr.08.2011.4633.

[7] K.S. Toppenberg, W.A. Block Jr., Uterine rupture: what family physicians need to know, Am. Fam. Physician 66 (5) (2002 Sep 1) 823–828.

[8] P.K. Heinonen, Clinical implications of the didelphic uterus: long-term follow-up of 49 cases, Eur. J. Obstet. Gynecol. Reprod. Biol. 91 (2) (2000 Aug) 183–190, https://doi.org/10.1016/s0301-2115(99)00259-6.

[9] N. Singh, U. Singh, M.L. Verma, Ruptured bicornuate uterus mimicking ectopic pregnancy: a case report, J. Obstet. Gynaecol. Res. 39 (1) (2013 Jan) 364–366, https://doi.org/10.1111/j.1447-0756.2012.01914.x.