CASE REPORT

Successful vaginal delivery following spontaneous adrenal haemorrhage at term

Sally Street,¹ Marloes Dekker Nitert,^{2,3} Leonie K Callaway^{2,3,4}¹Department of Obstetrics and Gynaecology, Redland Hospital, Cleveland, Queensland, Australia²University of Queensland School of Medicine, Herston, Queensland, Australia³University of Queensland Centre for Clinical Research, Herston, Queensland, Australia⁴Department of Obstetric and Internal Medicine, University of Queensland School of Medicine, Herston, Queensland, Australia,

Correspondence to

Dr Sally Street,
sally.lecomte@uqconnect.edu.au

Accepted 1 May 2016

SUMMARY

Spontaneous adrenal haemorrhage (SAH) is a rare event in the general population, estimated to be around 0.3–1.8%. The exact incidence in pregnancy is unknown but rare. Most cases of SAH at or near term have presented with massive haemorrhage and haemodynamic instability, requiring emergency caesarean delivery or intrauterine fetal death. This is the first reported case of a successful vaginal delivery after acute, spontaneous, left adrenal haemorrhage at term.

BACKGROUND

Spontaneous adrenal haemorrhage (SAH) is a rare event in the general population, estimated to be around 0.3–1.8%.^{1 2} The exact incidence in pregnancy is unknown but SAH is rare, seemingly more often right sided and unilateral, but occasionally left sided or bilateral. Since SAH is rare, it is often missed initially. Furthermore, most cases of SAH at or near term have presented with gross haemodynamic instability, resulting in emergent caesarean delivery or intrauterine fetal death. Since SAH is uncommon, the optimal mode of delivery for those women in whom SAH occurs without haemodynamic instability is equivocal. We present a case of successful vaginal birth in the setting of acute SAH at term as well as a literature review and discussion regarding differential diagnoses, approach to excluding underlying causes, mode of delivery/intrapartum care and postpartum follow-up.

CASE PRESENTATION

A 30-year-old gravida 7, para 4 presented to the birth suite of a peripheral centre at 38+4 following the acute onset of left-sided abdominal pain. Her pain was constant, sharp in nature, 9/10 in severity and located in the left upper quadrant with radiation into her back. She had associated nausea and two episodes of emesis but otherwise no headache, visual changes or history of elevated blood pressure. Her pregnancy had been otherwise uncomplicated. She had a background of four vaginal births at term with no antenatal or postnatal complications. On examination, her blood pressure was 112/78 mm Hg, pulse rate 78 bpm, temperature 37.1°C and oxygen saturation equal to 98% on room air. Her abdomen and uterine fundus were soft, with tenderness only in the left upper quadrant and flank. The uterine fundus was equivalent to her dates and fetal movements were felt. She had no vaginal bleeding and she was not contracting. Her cardiotocography was normal.

INVESTIGATIONS

Formal urine microscopy and culture was unremarkable, and she had no proteinuria. Full blood examination, renal function test, liver function test, inflammatory markers and coagulation studies were normal. She had a duplex Doppler of both lower limbs which was negative for deep vein thrombosis. An obstetric ultrasound confirmed fetal well-being. Abdominal ultrasound revealed a 12×10×10 cm mass in the left upper quadrant thought to be either a degenerative fibroid or an adrenal mass pathology. This was followed by an MRI which revealed an 11 cm adrenal haemorrhage with no obvious mass or lesion underlying it (figure 1).

DIFFERENTIAL DIAGNOSIS

Adenoma, pheochromocytoma, carcinoma, adrenal vein thrombosis, placental abruption, pyelonephritis, trauma, eclampsia/pre-eclampsia, pulmonary embolism.

TREATMENT

Vaginal examination found her cervix to be 2–3 cm dilated and 1 cm long. A multidisciplinary team discussion was conducted and it was decided that she was suitable for a trial of vaginal birth, given that she was ready for amniotomy, was haemodynamically stable at the time, and was of higher order parity. It was therefore likely that she would have a successful vaginal birth in a reasonable time frame. It was, however, felt that birth should take place at a tertiary centre where there was access to advanced obstetric/neonatal intensive care as well as emergency endocrine surgery and interventional radiology. Transfer was arranged for the same day. On arrival, her haemoglobin (Hb) was 89 mg/dL, a drop from 101 mg/dL 12 h earlier. Later that evening, her Hb dropped to 76 mg/dL and she was transfused with 2 units of red blood cells. An artificial rupture of membranes was performed the next morning, followed by an immediate epidural block. At the time of the artificial rupture of membranes, her Hb was 90 mg/dL, decreasing to 74 mg/dL after 2 h, and she received 2 further units of red blood cells. Given the evidence of ongoing bleeding, a second multidisciplinary team discussion was held to debate continued expectant management versus emergency caesarean delivery. By this time, she was 8 cm dilated and there was no fetal-maternal compromise. The consensus was that continued expectant management was likely to result in timely vaginal delivery that would avoid the additional morbidity of a caesarean section. Thus, the patient was allowed to continue with vaginal

CrossMark

To cite: Street S, Dekker Nitert M, Callaway LK. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-215096

Figure 1 MRI (A–C) of the upper abdomen showing a normal right kidney and adrenal. The left kidney is normal with an 11 cm mass in the left adrenal consistent with haemorrhage, no tumour or other underlying cause being evident.

delivery with her informed consent. She received hydrocortisone intrapartum to cover for presumed adrenal insufficiency, and was prepared for an instrumental second stage but the baby was delivered with one push. The APGAR scores were 9 and 9 at 1 and 5 min and there was no need for any resuscitation. Her pain resolved after delivery and she was discharged on day 3 with endocrine and surgical follow-up arranged.

OUTCOME AND FOLLOW-UP

All investigations to exclude an underlying cause during her inpatient stay were normal. The patient was lost to follow-up after discharge.

DISCUSSION

Any decision regarding mode of delivery requires careful consideration of a woman's preferences plus medical, surgical and obstetric history. However, in the setting of acute SAH at term, there are few cases to help guide delivery decisions when all other factors permit either mode.

The first case of SAH in pregnancy was reported in 1942. By 1962, only one of a further 34 reported cases featured survival of the patient.² Of the 14 cases of SAH with survival of the patient since 1962, the 12 published in English are included in this review.^{3–4} Of those 12 cases;

- ▶ Four required an emergency caesarean for maternal–fetal compromise.^{5–8}
- ▶ Two resulted in intrauterine fetal death prior to presentation.^{2,9}
- ▶ One case occurred immediately after the vaginal birth of twins.¹
- ▶ Three cases occurred in the first and second trimesters, and all had vaginal deliveries without exacerbation of haemorrhage closer to term.^{10–12}
- ▶ In one case, after stabilisation of the initial haemorrhage at 33 weeks, labour was induced at 41 weeks and an emergency caesarean section performed due to failure to progress in the second stage, but no exacerbation of adrenal haemorrhage occurred during labour.¹³
- ▶ In one other case, an elective caesarean section was performed for a woman whose haemorrhage occurred at 35 weeks. In this case, the initial haemorrhage was stabilised but after 2 weeks she developed haemorrhage in the contralateral gland and adrenal insufficiency. Therefore, an elective caesarean was performed for reasons of repeat and theoretical risk of further haemorrhage during labour.¹⁴

The current case presents the first of a successful vaginal birth in the setting of acute haemorrhage at term. It is recognised that she was an ideal patient for such a 'trial' given that she was full

term, ready for amniotomy and a higher order multipara with a history of straightforward vaginal births previously. Interestingly, her pain and bleeding resolved spontaneously following delivery of the baby.

While the mechanism of spontaneous adrenal haemorrhage is unknown, it is thought that the physiological hypervascularity in pregnancy could lead to an imbalance between the delivery of arterial blood and the venous drainage capacity. The increased delivery of arterial blood would result in extravasation of blood into the gland from the engorged capillary sinusoids. The hypervascularity resolves postpartum and this could either resolve or stabilise the haemorrhage. The risk of recurrence is unknown, but counselling of patients after delivery should include mention that a history of SAH theoretically means a higher likelihood of recurrence in a subsequent pregnancy. Postpartum counselling should include a discussion of the potentially increased risk for SAH in the next pregnancy and warning that it may present as more significant maternal–fetal haemorrhage ± maternal–fetal death.

Learning points

- ▶ Spontaneous adrenal haemorrhage (SAH) is a rare cause of abdominal cause in pregnancy and is often missed initially.
- ▶ SAH often presents with gross maternal–fetal compromise (including intrauterine fetal death) requiring an emergency caesarean delivery.
- ▶ In cases where there is no gross maternal–fetal compromise, the optimal mode of delivery is unclear; however, this case illustrates a successful vaginal birth in the setting of acute haemorrhage at term.
- ▶ If vaginal birth is not counterindicated, a tertiary centre with access to 24 h surgical, obstetric, obstetric medical, neonatology and intensive care unit/anaesthetic services is advisable.

Contributors LC was also involved in the patient's clinical care. All authors have contributed to the concept, planning, drafting and revision and have agreed on the final submission.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Wani MS, Naikoo ZA, Malik MA, et al. Spontaneous adrenal hemorrhage during pregnancy: review of literature and case report of successful conservative management. *J Turk Ger Gynecol Assoc* 2011;12:263–5.

- 2 Young H Jr, Burt RL, Johnstone F. Spontaneous adrenal hemorrhage in pregnancy. *Obstet Gynecol* 1962;19:440–3.
- 3 Kazarians B, Kausch I, Gellissen J, *et al.* [Spontaneous hemorrhage of the adrenal gland during pregnancy]. *Aktuelle Urol* 2007;38:403–5.
- 4 Hoeffel C, Legmann P, Luton JP, *et al.* [Spontaneous unilateral adrenal gland hemorrhage and pregnancy]. *J Gynecol Obstet Biol Reprod (Paris)* 1993;22:875–8.
- 5 Imga NN, Tutuncu Y, Tuna MM, *et al.* Idiopathic spontaneous adrenal hemorrhage in the third trimester of pregnancy. *Case Rep Med* 2013;2013:912494.
- 6 Bolla D, Schyrba V, Drack G, *et al.* Spontaneous rupture of an adrenal artery in pregnancy: a case report. *Case Rep Obstet Gynecol* 2012;2012:859068.
- 7 Anagnostopoulos A, Sharma S. Spontaneous adrenal haemorrhage in pregnancy. *BMJ Case Rep* 2011;2011.
- 8 Chrisite J, Batool I, Moss J, *et al.* Adrenal artery rupture in pregnancy. *BJOG* 2004;111:185–7.
- 9 Mweemba DM, Beck I, Tuffnell DJ. Spontaneous maternal adrenal haemorrhage as a cause of intra uterine foetal death. *J Obstet Gynaecol* 2005;25:194–5.
- 10 Keizer AL, Peters LW, de Vries C, *et al.* Spontaneous adrenal haemorrhage in early pregnancy. *BMJ Case Rep* 2013;2013.
- 11 Shen JZ, Stranks SN, Ho JT. Spontaneous adrenal haemorrhage in pregnancy. *Intern Med J* 2011;41:362–3.
- 12 Shantha N, Granger K. Spontaneous adrenal haemorrhage in early pregnancy. *J Obstet Gynaecol* 2008;28:449–51.
- 13 Gavrilova-Jordan L, Edmister WB, Farrell MA, *et al.* Spontaneous adrenal hemorrhage during pregnancy: a review of the literature and a case report of successful conservative management. *Obstet Gynecol Surv* 2005;60:191–5.
- 14 Bockorny B, Posteraro A, Bilgrami S. Bilateral spontaneous adrenal hemorrhage during pregnancy. *Obstet Gynecol* 2012;120:377–81.

Copyright 2016 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- Submit as many cases as you like
- Enjoy fast sympathetic peer review and rapid publication of accepted articles
- Access all the published articles
- Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow