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Case report

Couvelaire uterus in a previable pregnancy: Complication in abruptio placenta, case series from Tanzanian tertiary hospital

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ABSTRACT

Introduction and importance: Couvelaire uterus, also previously known as uteroplacental apoplexy. This is a life-threatening condition resulting from bleeding into the myometrium that may extend to the parametrium and peritoneum. Couvelaire uterus is typically associated with abruptio placentae, the premature separation of the placenta. This syndrome can only be diagnosed by direct visualization during caesarean section or biopsy (or both). For this reason, its prevalence is under-reported and underestimated in the literature.

Cases findings: We present a rare case series of two patients with Couvelaire uterus in previable pregnancy at Aga Khan Hospital, Dar es salaam. This combination is a rare occurrence and there are no cases reported in sub-Saharan Africa to the best of our knowledge.

Clinical discussion and conclusion: Couvelaire uterus is a rare manifestation to find in a previable pregnancy. The incidence of Couvelaire uterus is difficult to estimate since the diagnosis can only be reached intra-operatively. In most cases it occurs with abruptio placentae which develops due to a disruption in the vessels within the placenta allowing for blood to seep into the decidua basalis leading to premature separation of the placenta and bleeding into the myometrium which may extend to the parametrium and peritoneum. Clinicians should be vigilant when dealing with vaginal bleeding in a pre-viable pregnancy and placental separation is considered as an important differential to avoid the maternal morbidity and mortality that may ensue.

1. Introduction

Couvelaire uterus, also previously known as uteroplacental apoplexy. First described in the medical literature by Dr. Alexandre Couvelaire, a French obstetrician in 1912.This is a life-threatening condition resulting from bleeding into the myometrium that may extend to the parametrium and peritoneum [1].

Couvelaire uterus is typically associated with abruptio placentae, the premature separation of the placenta. This condition is usually diagnosed by direct visualization of the uterus during caesarean section. For this reason, its prevalence is perhaps underreported and underestimated in the literature [2].

Couvelaire uterus is a potentially fatal severe form of placental abruption. This is a significant cause of antepartum haemorrhage that is commonly associated with maternal and neonatal morbidity and mortality [2,3].

We present a rare case series of Couvelaire uterus in previable pregnancy in Aga Khan Hospital, Dar es salaam. This combination is a rare occurrence and there are no cases reported in sub-Saharan Africa from our literature search using various search engines. This paper has

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been reported in line with the SCARE 2020 criteria [7]. This article has been registered with the Research Registry with identification number researchregistry8362 and can be found through the following hyperlink Browse the Registry - Research Registry.

2. Case presentation

2.1. Case 1

A 37-year-old, Gravida 4 Para 3 living 3 at 19 weeks' gestation age with three previous caesarean scars, a known patient with chronic hypertension. She presented at our facility with complain of vaginal bleeding for 4 h prior to arrival, which was characterized by minimal bleeding with clots, she changed two pads. This was accompanied by lower abdominal pain that was intermittent and cramping in nature. It was non progressive with no specific periodicity. She had no preceding vaginal discharge or leakage. She did not report of awareness of heartbeat, loss of consciousness, or dizziness. On review of other system was uneventful. She has no known drug or food allergy.

She booked at 12 weeks and was prescribed methyldopa 500 mg thrice daily for her chronic hypertension, which she has been using with good compliance and her blood pressure was well controlled. The pregnancy was uneventful until the time of admission. She is a non-smoker and reports no alcohol use during pregnancy.

On examination: she was ill looking, alert, afebrile, not pale, not dyspnoeic with no lower limb oedema. Her vitals on admission were a blood pressure reading of 139/86 mmHg, a pulse rate of 86 beats per minute, a temperature of 36.6 °C saturating 99 % on room air.

Her abdominal examination revealed a gravid abdomen, had an old sub-umbilical midline incision scar, fundal height was 20 cm, with variable lie. The abdomen was not tense, and mild tenderness was elicited. Foetal heart rate was 168 beats per minute. Sterile speculum examination was done a healthy cervix was visualized which was closed with fresh blood oozing from the cervical OS. Review of other systems was unremarkable.

Obstetric ultrasound was performed initially, showed a single viable foetus with estimate weight of 330 g, foetal heart rate were 172 beats per minutes, posterior placenta with a heterogenous echotexture extending to anterior aspect (Figs. 1 and 2).

Initial blood work up revealed a Haemoglobin level of 11.5 g/dl, Haematocrit of 36 %, Platelets count of 161 $\times$ 10^9/l. Our provisional diagnosis at this point was threatened abortion at 19 weeks and chronic hypertension with differential diagnosis of low-lying placenta.

She was initiated on Progesterone (ethyl oleate progesterone) 100 mg intramuscular, intravenous Tranexamic acid 1000 mg, Intravenous paracetamol 1 g.

7 h post admission, she was reviewed once more by a resident of Obstetrics and Gynaecology reported she had developed per vaginal bleeding, with soaked one maternity pad and accompanied with awareness of her heartbeat. On examination: she was pale, tachycardic with a heart rate of 120.

Foetal heart rate was not picked on doppler assessment, other vital signs were stable.

Per abdominal exam revealed fundal height had significantly increased from 20 cm to 30 cm and tenderness elicited over the uterus.

Repeat Full blood picture showed Haemoglobin of 8.8 g/dl, Haematocrit of 25.7 % and Platelets count of 47 $\times$ 10^9/l. She was transfused with two units of packed red cells and four units of platelets. Our current diagnosis at this point was an Abruptio placenta. She consented for emergency exploratory laparotomy and was taken for surgery. Intra-operative under aseptic technique abdominal incision was made through old sub-umbilical midline incision scar and findings were

1. Hemoperitoneum of about 200 ml with fresh blood and clots
2. Uterus found measuring about 30 cm fundal height
3. Uterus had a distinct white and blueish discoloration resembling that of a Couvelaire uterus (Fig. 5)
4. Ovaries and fallopian tubes appeared normal
5. The urinary bladder was normal, but the bladder peritoneum was adhered to the lower part of the uterus.

Hysterotomy performed and delivery of a 260 g foetus with no signs of life. There were multiple heavy clots in the uterus, about 1000 ml of clots were evacuated.

Uterine muscles were lax and there was intractable bleeding despite administration of 40 IU oxytocin in 500 ml of Normal Saline.

Given that the levels of Platelet count pre-operatively was 57 and the continued bleeding seen at the incision site the decision was made for subtotal hysterectomy. Subtotal hysterectomy was done, the uterine stump sutured, and haemostasis achieved. There was significant bleeding from the left ovarian vessels therefore left oophorectomy was done as well. The abdomen was then closed in layers.

Patient was the sent to the Intensive Care Unit for close monitoring whilst receiving two units of packed red cells, four units of Platelets and

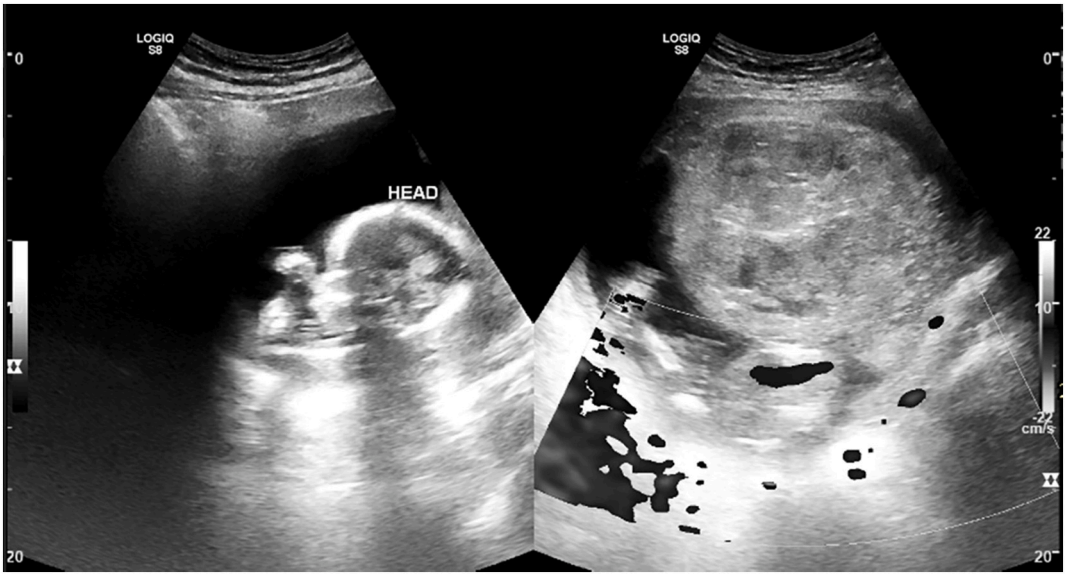

Fig. 1. Thickened and heterogeneous placenta.

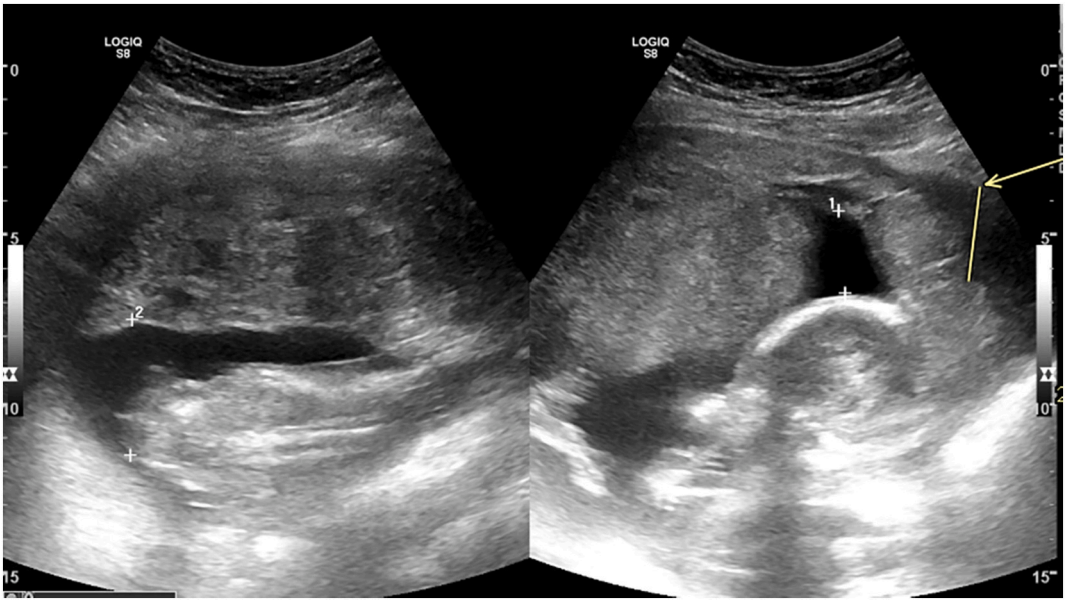

Fig. 2. Separated inferior aspect of the placenta.

two units of fresh whole blood. On day four post operation she was discharged in good condition. Her outpatient follow up was uneventful.

2.2. Case 2

A 38-year-old female, Gravida 5 Para 3 + 1 Living 2 at 25 weeks 6 days gestational age with three previous scars, also a known patient with chronic hypertension on medication. She presented at our facility with profuse vaginal bleeding for which she had changed four pads fully soaked 3 h prior to admission. This was preceded by severe abdominal pain. She reported the pain to be generalized and constant, reported normal foetal movements, no history of trauma, no other associated symptoms. She had a history of previous placental abruptio in her second pregnancy. On review of other system was uneventful. She has no known drug or food allergy.

She booked her Antenatal clinic at 12 weeks' gestation age, vitals and initial necessary investigations at booking were all normal except for a positive serology test for hepatitis B surface antigen. She was initiated on aspirin 150 mg daily, methyldopa 500 mg thrice daily and calcium 1tablet daily to reduce risk of hypertension in pregnancy. Her first obstetric scan was normal with placenta at fundal posterior. The rest of the pregnancy was uneventful until the day of admission.

On admission the patient was afebrile, not pale, with blood pressure reading of 148/90 mmHg, pulse rate of 69 beats per minute. Per abdomen examination revealed a fundal height of 26 cm, uterus was woody hard, and tender, foetal heart rate on Doppler picked at 120 beats per minute. The sterile speculum exam was inconclusive because of profuse bleeding. Other systemic examination was normal.

Initial blood work up showed Haemoglobin level of 11.1 g/dl with a Haematocrit level of 32.6 %, mild thrombocytopenia of 113×10^9/l, not previously noted on prenatal investigations. Coagulation profile was normal. Liver function test- showed elevated transaminases twice the

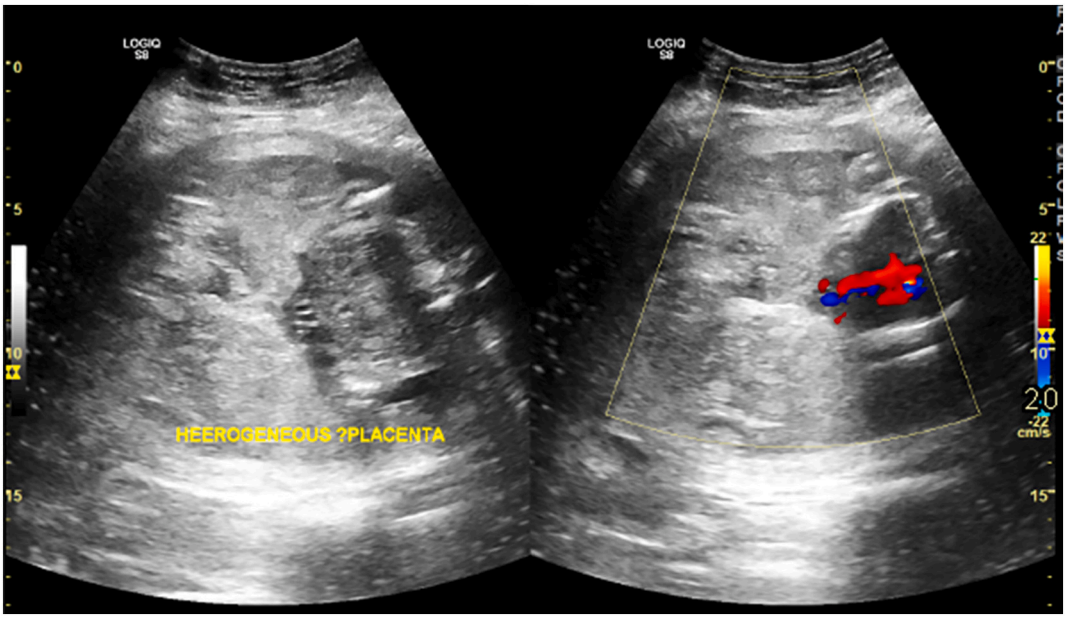

Fig. 3. Heterogenous Fundal Placenta.

upper limit. Urine protein was found to be plus three on urine dipstick. Creatinine levels were elevated to 176.2 umol/l (reference range 45–84 umol/l). Urgent ultrasound scan showed features suggestive of abruptio placenta, foetal heart rate at 150 beats per minute, estimated weight was 593 g. (Figs. 3 and 4).

At this point we had a diagnosis of placenta abruptio secondary to severe superimposed preeclampsia on chronic hypertension with acute kidney injury.

The patient was prepared for emergency caesarean hysterotomy given her history after she consented for the procedure. Under aseptic technique a Pfannenstiel incision was made on the abdomen and abdominal was opened in layers.

1. she had a peritoneal fluid which was blood stained
2. Uterus found measuring about 24 cm fundal height
3. Uterus had dark purple patches with ecchymosis and indurations of a Couvelaire uterus (Fig. 6)
4. Bladder, ovaries and fallopian tubes appeared normal

The foetus was delivered with a weak pulse with a birth weight of 610 g upon which resuscitation was attempted and transferred to Neonatal Intensive care Unit (NICU) for continuity of care.

Haemostasis was achieved with difficulty due to delayed clotting time. She developed postpartum haemorrhage of which she lost approximately two litres of blood, she then received two units of blood and fresh frozen plasma.

She was transferred to Intensive Care Unit (ICU) for further care. Post procedure her haemoglobin levels dropped to 7.7 g/dl of which she had already received 2 units of blood, she had worsening acute kidney injury creatinine levels of 472.06 umol/l (reference range 45–84 umol/l), Blood urea nitrogen of 13.17 mmol/l (reference range of 1.79–6.43), with electrolyte imbalance-hyperkalaemia of 6.92 mmol/l (reference range 3.5–5.1).

Nephrologist was consulted at this point and advised to continue with blood transfusion at least three units and review again after 48 h whereby renal function and potassium levels improved significantly after blood transfusion to creatinine levels of 102.06 umol/l, blood urea nitrogen of 5.7 mmol/l and potassium level of 4.2 mmol/l.

Day 2 post procedure patient was faring well and was transferred to normal ward where she continued with care and was stable for discharge on day six, whereby all blood work had improved significantly, and she was vitally and clinically stable. Unfortunately, the neonate passed away on day two of life.

3. Discussion

Couvelaire uterus is a rare, severe and possibly fatal complication of abruptio placentae. This was first described in the early 1900s as uteroplacental apoplexy by Couvelaire [1]. It is rare to find in a previable pregnancy. Few case reports have been published on this matter, most of the literature briefly references to Couvelaire uterus when discussing abruptio placentae but not as an entity in and of itself [2]. Which is similar presentation in our patients, both had an abruptio placenta together with Couvelaire uterus diagnosed intra operatively.

The true incidence of Couvelaire uterus is difficult to estimate since the diagnosis can only be made intra operatively, given that most cases abruptio placentae are delivered vaginally. It is possible that the true incidence is unknown due to this [3]. Given that our patients went for delivery by hysterotomy due to their multiple previous scars and haemodynamic instability; it was possible to identify their fatal complication of abruptio placenta which was a Couvelaire uterus.

The true aetiology remains unknown however it has been intricately linked to preeclampsia, chronic hypertension, amniotic fluid embolus, coagulopathies, maternal smoking and alcohol use [4]. In both of our case series, the patients had chronic hypertension and were on medication with good compliances nevertheless ended up with abruptio placenta.

Abruptio placentae develop due to a disruption in the vessels within the placenta allowing for blood to seep into the decidua basalis leading to premature separation of the placenta. This leads to clot formation between the placenta and the uterine wall that may stay as is or continue to expand leading to partial or total separation of the placenta. The presentation may be painful vaginal bleeding or painless vaginal bleeding with uterine hypertonus accompanied by non-reassuring foetal heart rate tracings [2]. There may be overt vaginal bleeding or concealed bleeding, with concealed bleeding this may lead to delays in diagnosis and management which is similar what occurred to our first patient.

To our knowledge, there has been one case report of Couvelaire uterus in a previable pregnancy of 21 weeks' gestation age [5]. Their

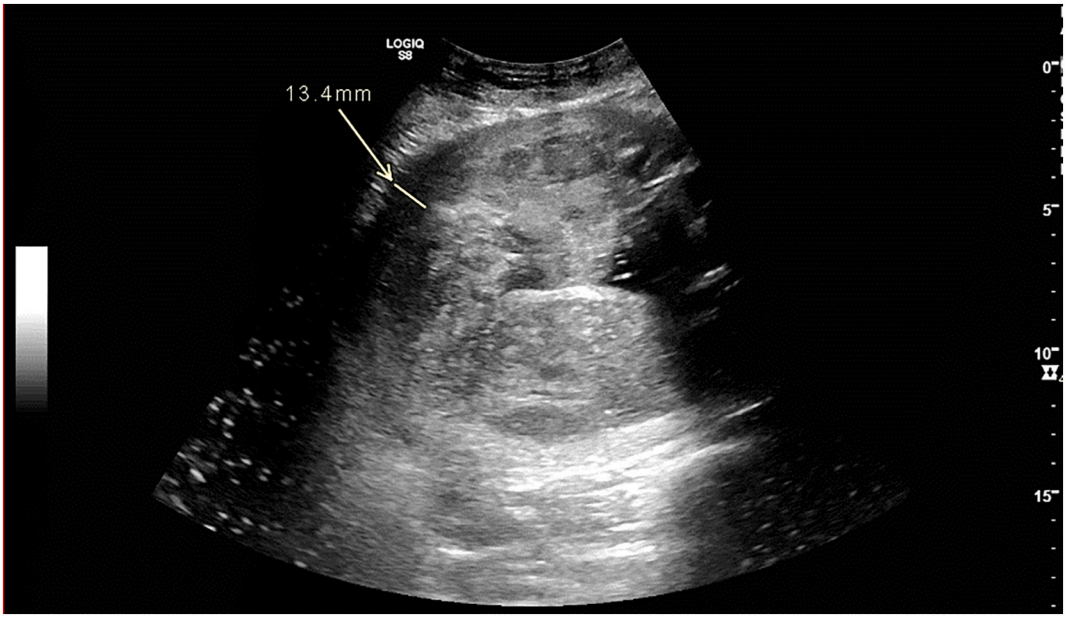

Fig. 4. Suspected placental separation at the fundus.

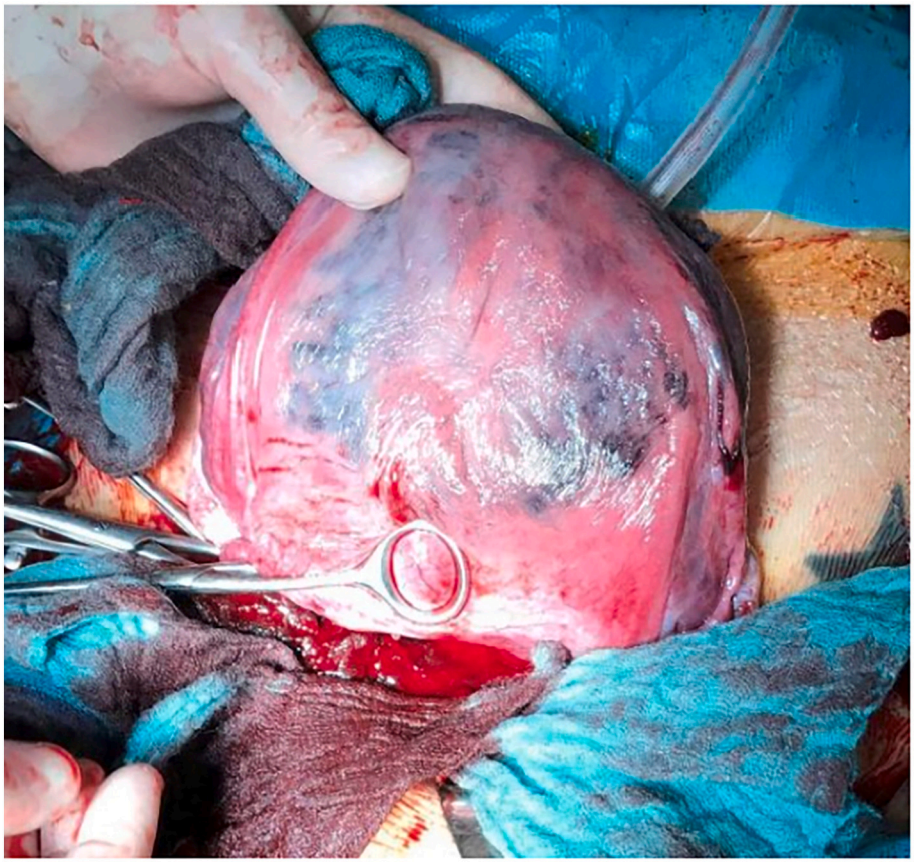

Fig. 5. Case 1. (Couvelaire uterus because of acute intradecidual haemorrhage produced by the rupture of the uterus-placental spiral arterioles which produce ecchymosis discolorations, secondary to extravasation of blood into myometrium and serosa).

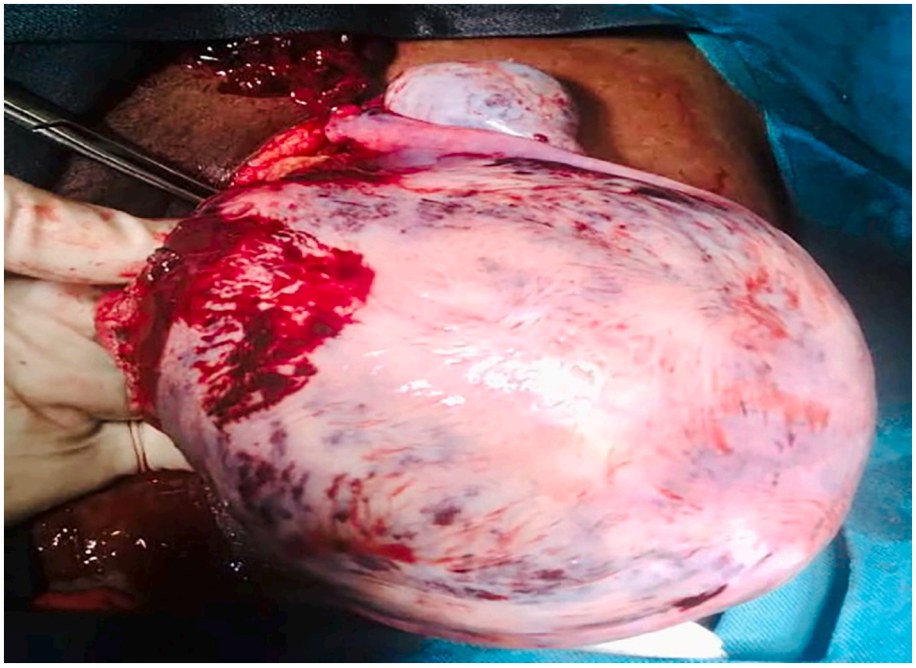

Fig. 6. Case 2. (Picture showing uterus having dark purple patches with ecchymosis and indurations). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

patient presented similarly to ours but developed acute abdomen and deranged lab work which is what lead them to operative management. In their case, a hysterotomy was done and uterus was closed in layers and haemostasis achieved. This goes in line with the current school of thought that despite extravasation of blood into the myometrial layer the uterus retains its ability to contract, and a hysterectomy is not the mainstay of treatment. [3]

In both of our cases, the patients presented with an episode of overt bleeding followed by cessation of bleeding. The level of suspicion for concealed bleeding increased once we noted the significant increase in fundal height and new onset derangement in her lab work suggestive of continued haemorrhage and impending disseminated intravascular coagulopathy. Given the history of previous scars in our patients and deranged bleeding indices, we were unable to induce labour and proceed with conservative management.

Couvelaire uterus is meant to be managed conservatively and hysterotomy is reserved for obstetric indications and maternal haemodynamic instability among other reasons. A hysterectomy is usually not indicated as the uterine muscles retain their ability to contract despite blood exsanguinating to the myometrium [3]. In the pre-viable pregnancy, there is no standard recommendation on the mode of delivery, therefore decision lies with the attending Obstetrician, and it is individualized.

In the instance of the first case, the earlier gestational age and the cessation of bleeding along with the patient being vitally and clinically stable delayed diagnosis leading to total placental abruption and disseminated intravascular coagulopathy which led to the sequence of events that followed. Had the team on the ground entertained a possible differential of abruptio placenta operative management may have been instituted earlier and avoided the hysterectomy and further morbidity suffered by the patient. It is worth remembering that a bleeding pregnant woman should be reviewed frequently, even after cessation of bleeding, vigilance is required to avoid potential adverse outcomes.

Couvelaire uterus is known to cause significant morbidity and mortality from postpartum haemorrhage (PPH), disseminated intravascular coagulopathy (DIC), Intensive Care Unit (ICU) admissions, need for blood transfusion, prolonged hospital stay and adverse neonatal outcome including foetal death [6]. Which are the similar events that have occurred to both of our case series. Patients had post-partum haemorrhage, disseminated intravascular coagulopathy (DIC) ending to ICU admission and receive multiple blood transfusions. The second patient also developed acute kidney injury due to extensive bleeding and this is worth noting that multiple disciplines should be involved in the care of a patient with significant bleeding and haemodynamic instability.

4. Conclusion

Vigilance is needed when dealing with vaginal bleeding in a pre-viable pregnancy and placental separation should be considered as a differential to avoid maternal morbidity and mortality that may ensue.

Consent

Written informed consent was obtained from the patients for publication of this case series and its accompanying images. A copy of the written consent has been retained by the hospital as the patients have opted to remain anonymous.

Ethical approval

Case series are exempt from ethical approval from the research committee of the Aga Khan Hospital, Dar-es-Salaam.

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Author contribution

VK: Involved in the acquisition of data, data collection, manuscript drafting and its revision.

BM: Involved in the acquisition of data, data collection, manuscript drafting and its revision.

WK: Involved in the acquisition of data and manuscript drafting.

AJ: Involved in the acquisition of data and manuscript revision.

MM: Involved in the clinical care of the patient.

MK: Involved in the clinical care of the patient, manuscript revision and supervision.

Guarantor

Dr. Munawar Kaguta, Obstetrician and Gynaecologist, Aga Khan Hospital.

Registration of research studies

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Provenance and peer review

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Declaration of competing interest

The authors report no conflicts of interest for this work.

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