BMJ Case Reports

Successful Outcome After Spontaneous First Trimester Intra-amniotic Haematoma and Early Preterm Premature Rupture of Membranes.

Journal:	*BMJ Case Reports*
Manuscript ID	bcr-2018-224596.R2
Manuscript Type:	Rare disease
Date Submitted by the Author:	13-Aug-2018
Complete List of Authors:	Bakalis, Spyros; Guy's and Saint Thomas' NHS Foundation Trust, Obstetrics, Maternal and Fetal Medicine David , Anna ; University College London, EGA Institute for Women's Health; NIHR University College London Hospitals Biomedical Research Centre,
Keywords:	Obstetrics and gynaecology, Pregnancy < Obstetrics and gynaecology

BMJ Case Reports

Full cases template and Checklist for authors

Full clinical cases submission template

TITLE OF CASE ***Do not include "a case report"***

Successful Outcome After Spontaneous First Trimester Intra-amniotic Haematoma and Early Preterm Premature Rupture of Membranes.

SUMMARY ***Up to 150 words summarising the case presentation and outcome (this will be freely available online)***

Spontaneous intra-amniotic haematoma is a rare cause of preterm premature rupture of the membranes (PPROM) but can have significant fetal and maternal consequences. It has been previously been reported to occur in the second and third trimesters but not in an earlier gestation. We present a case that presented acutely in the first trimester of pregnancy, which lead to early PPROM at 15 weeks and spontaneous preterm delivery at 28 weeks of gestation. There were no maternal complications during the pregnancy.

BACKGROUND ***Why you think this case is important – why did you write it up?***

Intra-amniotic haematoma, in the absence of trauma or amniocentesis, is a very rare event. It is thought to occur due to either a subchorionic or subamniotic haematoma which dissects through the amnion and into the amniotic cavity. A few case reports of spontaneous intra-amniotic haematoma occurring in the second and third trimester describe significant maternal compromise, threatened preterm labour, and the intra-amniotic haematoma mimicking fetal abnormalities or as an incidental finding. We report a case of acute intra-amniotic haematoma detected in the first trimester leading to early preterm premature rupture of membranes (PPROM) and subsequent preterm delivery with a good maternal and neonatal outcome.

CASE PRESENTATION ***Presenting features, medical/social/family history***

A 30-year-old Caucasian gravida 2 para 0 was referred for fetal medicine specialist assessment at 14+1 weeks of gestation due to detection of an anechoic area (41x30x26mm) within the maternal isthmus of the uterus during the first trimester ultrasound examination (12+0 weeks) for

PLEASE DELETE THIS PAGE BEFORE SUBMITTING YOUR ARTICLE

BMJ Case Reports

combined screening (1:2700 Down syndrome risk, βhCG 1.08 MoM and PAPP-A 0.46 MoM) (figure 1). The patient had attended hospital 3 days earlier with vaginal bleeding and abdominal pain, where ultrasound examination of the fetus and uterus showed no abnormality.

Transvaginal ultrasound was performed due to the poor image quality transabdominally. The fetal growth velocity was normal, the bowel was echogenic but there were no other fetal structural abnormalities or soft markers were detectable. Floating within the amniotic fluid attached to the superior end of the placenta was an area of heterogeneous echogenicity (45x37x28mm), which resembled an organised blood clot within the amniotic sac (figures 2 and 3). This area moved with the placenta, whilst Doppler studies revealed no active blood flow within it. The patient was advised of the risks of further bleeding, PPPROM and subsequent preterm birth.

Follow up was planned for 3 weeks later, however, she attended hospital 6 days later describing a brown vaginal discharge and wetness. A speculum examination allowed visualisation of pooling liquor in the posterior fornix on Valsalva. An ultrasound scan confirmed oligohydramnios, a live fetus and the previously observed blood clot (now 30x37x22 mm). Based on the history of 'wetness', findings on speculum examination, and oligohydramnios, a diagnosis of PPROM was made, and the patient was counselled about the possible maternal and neonatal outcomes. The couple declined termination of pregnancy. The patient was commenced on Erythromycin 500mg BD for 10 days as per our department's policy to give it after PPROM, even this early, to prevent infection and delay delivery. In addition, she was offered serial weekly outpatient monitoring for maternal and fetal wellbeing.

Between 16 and 18 weeks of gestation the bleeding and amniotic fluid leakage ceased, fetal growth was appropriate, and two pools of amniotic fluid were consistently seen (7mm and

BMJ Case Reports

1.4mm). At 20+1 weeks, detailed anomaly ultrasound examination revealed normal fetal growth and anatomy; the fetal stomach, kidneys and bladder were visualized and fetal movements were observed. Over the following seven weeks' small pockets of amniotic fluid remained around the fetus, fetal growth continued with normal fetal Doppler studies. At 28+0 weeks, the patient experienced vaginal bleeding and was admitted to hospital where maternal dexamethasone injection was administered to mature the fetal lungs. Spontaneous preterm labour ensued at 28+2 weeks and she delivered a live baby girl by breech vaginal delivery weighing 1.19kg with Apgars of 3 at 1 minute. The baby was initially intubated for 7 days and required a further 18 days of continuous positive airway pressure but could breathe spontaneously at 3 weeks of age. A chest x-ray showed pulmonary hypoplasia (small lung volume and poor lung expansion). Though initially treated as septic, all neonatal cultures were negative and antibiotics were stopped after 2 days and the baby was discharged to her local neonatal unit on day 27.

Histological analysis of the placenta showed florid amnion nodosum consistent with longstanding oligohydramnios, but with no acute inflammation. There were haemosiderin laden macrophages in the chorion which suggested an amniotic haemorrhage. The cord and membranes looked otherwise normal, with no signs of villitis or intervillositis.

INVESTIGATIONS ***If relevant***

DIFFERENTIAL DIAGNOSIS ***If relevant***

TREATMENT ***If relevant***

OUTCOME AND FOLLOW-UP

BMJ Case Reports

The child has been seen recently aged 3. She is walking and talking and ahead in all her milestones. There are no ongoing concerns resulting from her prematurity.

DISCUSSION ***Include a very brief review of similar published cases***

Intra-amniotic haemorrhage, unrelated to amniocentesis or trauma, is a very rare event with an unknown incidence, and only a handful of cases described in the literature (Table 1). Intra-amniotic haemorrhage is thought to result from either a subchorionic or subamniotic haematoma which dissects through the amnion and into the amniotic cavity. This bleeding can either small echogenic particles floating in the amniotic fluid which represent fibrin strands, or form a mass which can mimic fetal abnormalities [1].

Gilboa et al., 2012 presented a case of an incidental finding of intra-amniotic haemorrhage prior to a planned Caesarean section with no adverse effect on the mother or neonate [2]. Ustuner et al., 2013 reported a patient at 20 weeks who presented for specialist fetal medicine review with a raised serum AFP concentration and a normal scan at 18 weeks. In this case, the intra-amniotic haemorrhage mimicked an anterior abdominal wall defect and required a fetal MRI to confirm the diagnosis [3]. Intra-amniotic haemorrhage has presented in two cases in the third trimester with uterine contractions, vaginal bleeding and anaemia [4,5] In one case at 36 weeks, ultrasound imaging revealed real time rapidly increasing polyhydramnios, and a transplacental tear was visible with a freely mobile flap of placental tissue extending into the amniotic cavity. Pathology examination revealed a circumvallate placenta [4]. In the second case at 32 weeks, there was reduced beat to beat variability on the cardiotocogram, and ultrasound scan showed polyhydramnios and a blood clot around the umbilical cord. Amniocentesis revealed heavily blood stained amniotic fluid and immediate Caesarean section was performed with delivery of a healthy neonate. Placental histological analysis revealed chorioamnionitis [5]. Finally, intra-

BMJ Case Reports

amniotic haemorrhage has also presented with maternal collapse, tachycardia, hypotension and anaemia at 40 weeks of gestation (Sijanovic et al., 2007). Ultrasound examination showed a live, well grown fetus with a large intra-amniotic homogenous echogenic mass (120x80mm). She developed haemorrhagic shock and underwent immediate Caesarean delivery. The placental histological examination was normal with no evidence of an abruption, but there was a 20cm diameter 450g blood clot detected [6].

Our case is unique in the literature due to its early gestational age at diagnosis of intra-amniotic haemorrhage and associated PPROM. Though there were no effects to the maternal cardiovascular system, the vaginal bleeding and intra-amniotic blood clot were early signs of the pathology present. The lack of sepsis and chorioamnionitis, both clinically during the pregnancy and on histological analysis of the placenta is the most likely reason that the pregnancy progressed. The haemosiderin laden macrophages in the chorion confirmed the presence of amniotic haemorrhage, however, the defect in the placenta was not seen, either as it was too small or because it had already sealed up.

Though intra-amniotic haemorrhage is a rare occurrence, its consequences can be severe for both mother and fetus. Its identification at any gestation should be taken seriously and acted upon as appropriate for the presenting symptoms and gestational age.

Author	GA (wks)	Presenting Complaint	Ultrasound Findings	Placental Findings	Outcome

BMJ Case Reports

Gilboa et al.,	38	Routine prior to LSCS.	'Sludge' and a 70x20mm cylindrical mass.	No gross pathology.	LSCS – PPH (not related to intra-amniotic haemorrhage).
Ustuner et al.,	20	Raised AFP. Possible of anterior abdominal wall defect and oligohydramnios.	A highly echogenic, partially solid and partially cystic mass measuring 28x30mm.		PPROM miscarriage.
Cutillo et al.,	36	Uterine contractions, APH. Maternal anaemia.	Real time rapidly increasing polyhydramnios. Transplacental tear with decreased echogenicity and irregular borders. A freely mobile flap of placental tissue extending from the tear.	Placenta circumvalate with blood-stained membranes, extensive intervillous fibrin deposition and focal villous fibrosis.	Artificial rupture of membranes revealed dark blood-stained amniotic fluid proceeding to LSCS.
Kurata et al.,	32	Uterine contractions, vaginal bleeding and maternal tachycardia and anaemia. Cardiotocogram showed reduced variability. A diagnostic	A clot around the umbilical cord and polyhydramnios.	A 150g old clot was found around the umbilical cord with no signs of abruption. Analysis of the placenta revealed chorioamnionitis.	LSCS.

BMJ Case Reports

		amniocentesis revealed heavily blood stained amniotic fluid.			
Sijanovic et al.,	40	Maternal collapse and weakness. Maternal tachycardia, hypotension and anaemia	A homogenous echogenic mass measuring 120x80mm within the amniotic fluid.	Fibroid matter at the edge of the placenta.	LSCS

Table 1. Presenting complaint, ultrasound findings, placental findings, pathology and outcome of 5 cases of intra-amniotic haemorrhage.

LEARNING POINTS/TAKE HOME MESSAGES ***3 to 5 bullet points – this is a required field***

Spontaneous intra-amniotic haematoma is a rare cause of preterm premature rupture of the membranes (PPROM).

It can have significant fetal and maternal consequences.

With appropriate conservative management, a successful outcome is possible.

REFERENCES ***Vancouver style (Was the patient involved in a clinical trial? Please reference related articles)***

1. Trop I, Levine D. Hemorrhage during pregnancy: sonography and MR imaging. AJR Am J Roentgenol. 2001 Mar;176(3):607-15.

2. Gilboa Y, Duvdevani N, Yinon Y, Achiron R. A case of spontaneous intra-amniotic hemorrhage in an asymptomatic patient at near term pregnancy. Fetal Diagn Ther. 2012;31(1):73-5.

3. Ustüner I, Güven ES, Balık G, Sentürk S, Ustüner E, Atman ED, Avşar AF. Spontaneous

BMJ Case Reports

intraamniotic hemorrhage in the second trimester mimicking an abdominal wall defect. J Turk Ger Gynecol Assoc. 2013 Jun 1;14(2):109-12.

4. Cutillo DP, Swayne LC, Schwartz JR, Dise CA, Faux RG. Intra-amniotic hemorrhage secondary to placenta circumvallate. J Ultrasound Med. 1989 Jul;8(7):399-401.

5. Kurata H, Sekizuka N, Kato R, Yoshizawa H, Tanaka K. Intra-amniotic maternal hemorrhage in preterm labor: a case report. J Perinat Med. 1995;23(3):229-32.

6. Sijanovic S, Selthofer R, Abicic-Zuljevic K, Milojkovic M, Topolovec Z, Sijanovic I, Kulas T. A case of intra-amniotic maternal hemorrhage in term pregnancy. Fetal Diagn Ther. 2007;22(4):299-301.

The patient was not involved in a clinical trial.

FIGURE/VIDEO CAPTIONS ***figures should NOT be embedded in this document***

Table 1. Presenting complaint, ultrasound findings, placental findings, pathology and outcome of 5 cases of intra-amniotic haemorrhage.

Figure 1: Intra-amniotic haemorrhage resembling an organised blood clot.

Figure 2: Intra-amniotic haemorrhage resembling an organised blood clot within the amniotic sac adjacent to the fetal head.

Figure 3: Intra-amniotic haemorrhage resembling an organised blood clot.

PATIENT'S PERSPECTIVE ***Optional but strongly encouraged – this has to be written by the patient or next of kin***

The initial diagnosis was devastating. We really appreciated the care we received, particularly the time that the team took to talk us through the available evidence on PPROM and its potential consequences. Having that information enabled us to make an informed decision. We felt hugely supported by the fetal medicine team.

The remaining 13 weeks of the pregnancy were very difficult. We learned to live with a great deal of uncertainty, trying to balance hope with realism. A high-risk pregnancy is an isolating experience, although we were helped and encouraged through this period by family, friends, and the medical team. We also found the support from people online who had had similar experiences very valuable.

Despite a difficult time on the neonatal unit, our daughter is now a bright, lively, funny three-year old. When we see her running around the park, we remember a conversation with a neonatal consultant shortly before she was born, who told us that in the best-case scenario, she might never be able to run very far. We are amazed and so grateful to have had the outcome we did.

BMJ Case Reports

Date: 23/06/2018

PLEASE SAVE YOUR TEMPLATE WITH THE FOLLOWING FORMAT:

Corresponding author's last name and date of submission, eg,

Bakalis_June_2018.doc

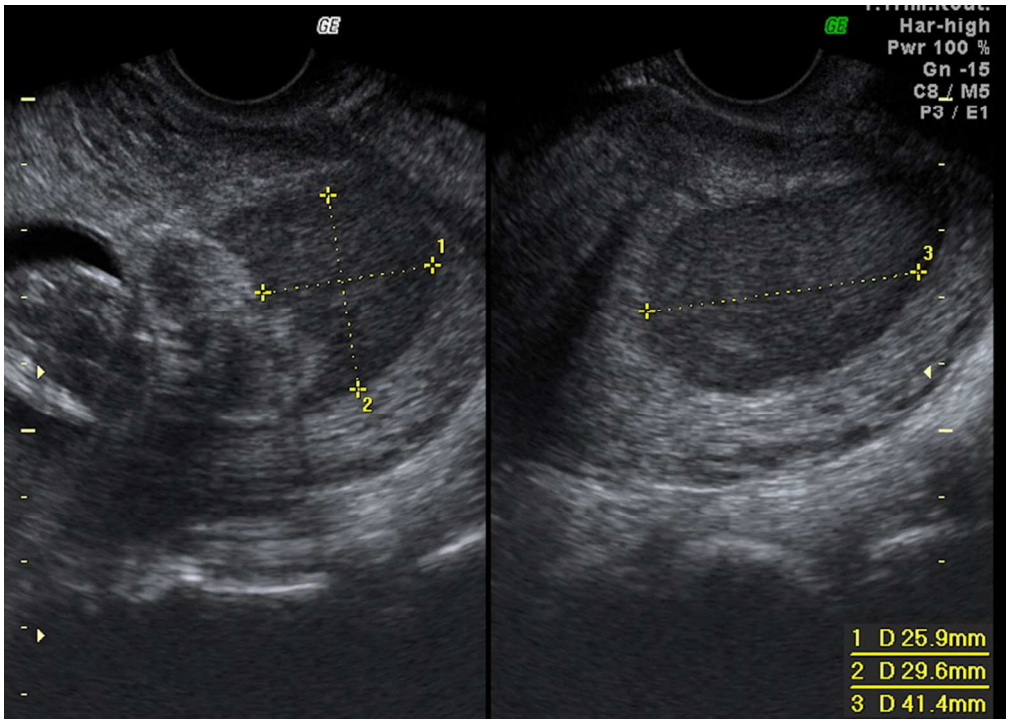

Figure 1: Picture from initial scan at 12 weeks showing anechoic area (41x30x26mm) within the maternal isthmus.

90x64mm (300 x 300 DPI)

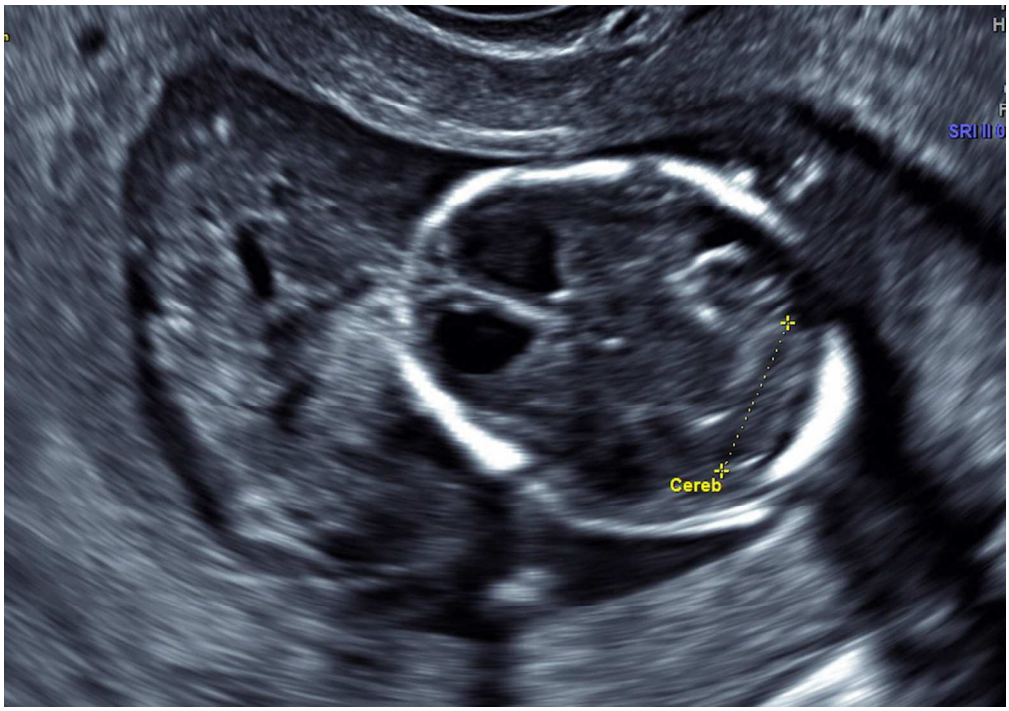

Figure 2: Intra-amniotic haemorrhage resembling an organised blood clot within the amniotic sac adjacent to the fetal head.

99x70mm (300 x 300 DPI)

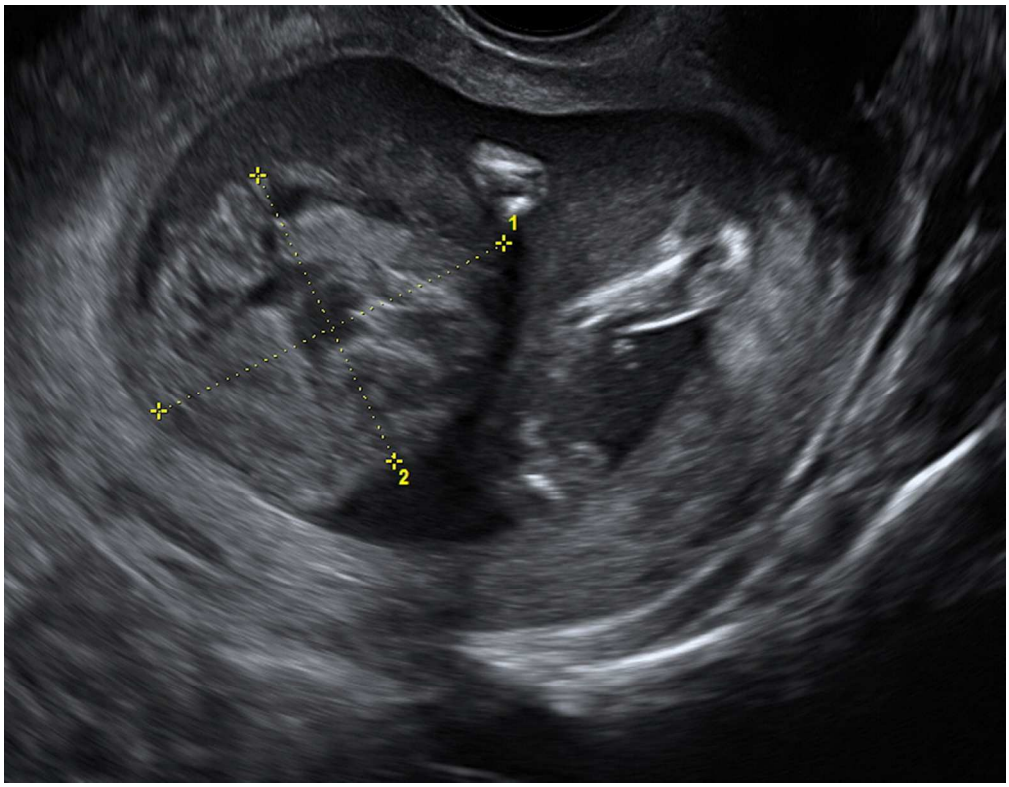

Figure 3: Intra-amniotic haemorrhage resembling an organised blood clot.

90x70mm (300 x 300 DPI)