Unexplained Intrauterine Death In The Third Trimester: Poor Predictive Value Of Umbilical Artery Doppler Waveform

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CASE REPORT

We report a case of a 40 year old in her fifth pregnancy following 4 previous term vaginal deliveries including one set of dichorionic twins. The patient and all her children were known to have Ehler's Danlos syndrome (EDS) type 3 (benign hypermobility syndrome) on clinical examination.

She booked at 11 weeks gestation, and had a nuchal translucency scan giving her a low risk of Down syndrome (1 in 521). An anomaly scan at 22 weeks gestation showed no soft tissue markers of aneuploidy or structural anomalies. She presented to the labour ward at 28 weeks gestation with decreased fetal movements, for 24 hrs. A cardiotocography (CTG) trace performed at that stage was unremarkable and ultrasound on the same day showed appropriate growth, normal amniotic fluid index (AFI) and positive end diastolic flow (ED) in umbilical artery. Fetal movements however during scanning were minimal and hyperechogenic bowel was noted. She was admitted for observation and a CTG performed the next morning. This showed reduced variability, with no acceleration; however there were no decelerations. She was taken off the CTG to allow going to the toilet for 10 minutes, but when the monitor was put back, no fetal heart beat was found and fetal demise was confirmed by ultrasound scan (USS) 30 minutes later. This second scan confirmed normal AFI, normal biometry for gestation, but there was fetal hydrops with scalp oedema, bilateral pleural effusion and echogenic bowel.

She delivered vaginally after induction with mifepristone and cervagem 2 days following fetal demise. However an hour after delivery she developed severe pain in left iliac fossa (LIF), which was fully investigated due to worsening symptoms and a fall in haemoglobin to 5.9 gms, with USS and computed tomography (CT) scan. These demonstrated the presence of a spontaneous, discrete left sided broad ligament haematoma. This was initially managed conservatively with antibiotics and analgesia. However, despite stable symptoms for 10 days, she became pyrexial with a raised C-reactive protein, therefore an examination under anaesthesia and laparotomy were performed as well as evacuation of retained products of conception (ERPC). At the laparotomy a clearly defined well-organized broad ligament haematoma was noted, but there were no evidence of infection or abscess formation or active bleeding. It was therefore decided not to evacuate the haematoma as it would reabsorb spontaneously and evacuation would have significantly increased the risks of bleeding. At the ERPC a small amount of products were evacuated. Following the procedure she made a good post-operative recovery becoming non-febrile and eventually was discharged home.

POST MORTEM

Postmortem examination of the fetus confirmed a normally grown male fetus, weighing 4.28 kg with some degree of maceration. There was bilateral pleural effusion, but there was no evidence of any congenital malformation or dysmorphic features or infection. There was no placental abnormality on gross examination and on microscopy. There was a normal 46XY karyotype on CVS prior to delivery.
DISCUSSION

Stillbirths occur in about 1% of pregnancies. There is an increase in risk with maternal age, however it has been estimated that as many as 12.5-50% of stillbirths have no identifiable etiology. The causes of fetal death are often complex, however certain causes of fetal demise have higher incidences during specific periods of gestation. Second and third trimester fetal demise may already have occurred seriously, even though an irreversible insult to the fetus might have occurred. Present recommendations on the management of pregnancies with decreased FMs are based on limited and inconsistent scientific evidence. There is a need for further well-designed studies in order to provide evidence-based guidelines.

The second consideration in this patient was the occurrence of broad ligament haematoma. Broad ligament haematoma is easily diagnosed and revealed in cases of classical subperiheal bleeding related to trauma while ‘Delayed’ type is occurring 24 hours after delivery, and thought to result from pressure necrosis. The two time based varieties present in entirely different manners, with unexplained postpartum collapse and abdominal pain and a mass with the immediate variety, and subacute abdominal signs, postpartum pyrexia and anaemia associated with the subacute type.

Last but not the least, an association between EDS (connective tissue disorder) type III with either IUD or broad ligament haematoma can not be easily explained. In EDS type III group, the main symptom is generalised joint hypermobility, the skin involvement (hypertensibility and/or smooth velvety skin) as well as bruising tendencies are present in variable severity, where as EDS type IV is regarded as more serious form of EDS.

Arterial/intestinal/uterine fragility or rupture commonly arise in EDS type IV group. Spontaneous arterial rupture has a peak incidence in the third and fourth decade of life. Mid-size arteries are commonly involved. Acute diffuse or localised abdominal or flank pain is common presentation of arterial/intestinal rupture. Pregnancies may be complicated by intra-partum uterine rupture, pre and post partum arterial bleeding.

CONCLUSION

Maternal complaint of decreased or absent fetal movement should be taken seriously and should be investigated.
thoroughly. Decrease or absence in fetal movements is often present prior to fetal death, even in the presence of normal Doppler velocimetry in umbilical arteries. Cardiotocography, umbilical/uterine artery Doppler velocimetry have been used for antepartum fetal assessment in pregnancies with decreased fetal movements, but the evidence of a clinical benefit is limited. There is need for further well-designed studies in order to provide evidence-based guidelines in the future.

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**References**

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