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Case Report

A case report on rare ultrasound finding of spontaneous chorioamniotic membrane separation

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ABSTRACT

The separation of the chorion and amnion before 14 weeks of gestation is physiologically normal. The amnion and chorion usually fuse between 14 and 16 weeks, and any chorioamniotic separation (CAS) that persists after 16 weeks is uncommon and anomalous. CAS can occur spontaneously or after an intrauterine intervention such as amniocentesis where it may be detected on follow-up ultrasound. Another cause of chorioamniotic membrane separation (CAMS) can be chromosomal abnormalities which present with the absence or delayed fusion of the two layers. It is associated with poor perinatal outcomes such as preterm delivery, preterm prelabour rupture of membranes (PPROM) and amniotic band syndrome. As cases of spontaneous CAMS (sCAMS) are exceptionally rare, perinatal outcomes associated with it are ambiguous. This poses a difficulty for clinicians who are managing the case to determine appropriate antepartum care. It also hinders the clinician's ability to counsel patients. We hope our case report helps in better recognition of the abnormality and encourages more reporting and studies of similar cases to be able to have a better understanding of the full implications of such sonographic findings. Moreover, increased reporting will help obstetricians to mitigate the fetal and maternal risks associated with sCAMS.

Keywords: Preterm labour, PPRM, CAMS, Amniotic band syndrome

INTRODUCTION

The fetal membrane is a bilayer structure composed of thick cellular chorion covering the thinner amnion. During development chorion and amnion arise from different germ layers. It is physiological and a normal sonographic finding for the chorion and amnion to be separated by chorionic fluid during the first trimester of pregnancy. Complete fusion of the two layers occurs by 14-16 weeks of gestation.¹

CAMS is a new separation of chorion and amnion after its fusion beyond 16 weeks of gestation. This is pathological and can either occur spontaneously or following an

invasive procedure like amniocentesis or fetal surgery. Another cause of CAMS can be chromosomal abnormalities which present with absence or delayed fusion of the two layers. In complete CAMS, the chorion and amnion are completely separated except at the insertion site of the umbilical cord.²

From a systematic review of case series and case reports it is noted that iatrogenic CAMS is a known complication of interventions like amniocentesis or in-utero fetal surgery. It is associated with poor perinatal outcomes like preterm delivery, PPRM, amniotic band formation, umbilical cord compromise and intrauterine fetal death. Though there is no standardised protocol for the management of these patients, local hospitals implement plans such as

admission, fetal heart rate monitoring and serial ultrasound.¹⁻⁵

As cases of sCAMS are exceptionally rare, perinatal outcomes associated with it are ambiguous. This poses a difficulty for clinicians who are managing the case to determine appropriate antepartum care. It also hinders the clinician's ability to counsel patients. A retrospective cohort study including 64 cases of sCAMS demonstrated that preterm delivery occurred in 51.6% of the cases and preterm premature rupture of membranes in 36.7% of the cases.⁷

We present to you a case of sCAMS diagnosed at 28 weeks of gestation.

CASE REPORT

This is the case of a 37-year-old lady in her third pregnancy. The first two pregnancies unfortunately ended up with miscarriages around 8 weeks of gestation. This pregnancy was conceived spontaneously. She was known to have PCOS, asthma, which was well controlled on medication and lymphedema since childhood. She was also diagnosed to have a bicornuate uterus.

She had routine antenatal care with increased surveillance in preterm clinic (due to bicornuate uterus) with serial cervical length scans.

She had her first trimester screening and was at a 1/44 risk of Downs syndrome. She declined all invasive testing but did have NIPT. The results of NIPT were reassuring and she was deemed low risk for Downs syndrome.

Anomaly scan at 19+2 weeks was essentially normal but could not assess the cardiac and brain anatomy and therefore was referred to the fetal medicine consultant for a detailed scan. No obvious defects or markers of chromosomal anomalies were found on scan. Cervical length was 28.5 mm. The uterine artery doppler showed high PI resistance and hence she was deemed screen positive for PET/FGR.

She was diagnosed with preeclampsia at 28+3 weeks of gestation and commenced on antihypertensive medications.

She was referred to fetal medicine due to severe growth restriction (EFW<1st centile) suspected to be secondary to placental insufficiency due to PET and that is when she was diagnosed with CAMS (Figure 1).

On a scan performed by the fetal medicine consultant, it was found that the baby was measuring small, and the estimated fetal weight was <1st centile. Umbilical artery PI was above the 90th percentile. There were signs of marked redistribution. Liquor was reduced. Good fetal movements were noted.

Irregular amniotic sac with trabeculations and adhesions between amnion and chorion with some fine amniotic bands and encysted fluid between amnion and chorion. The amniotic fluid also appeared cloudy. A provision diagnosis of partial sCAMS was made and a perinatal alert was put in place to assess the baby for any signs of amniotic band sequence.

It was explained to her that this is most likely an early rupture of amnion with no obvious sequence. The baby would be assessed after delivery for probable signs of amniotic band syndrome.

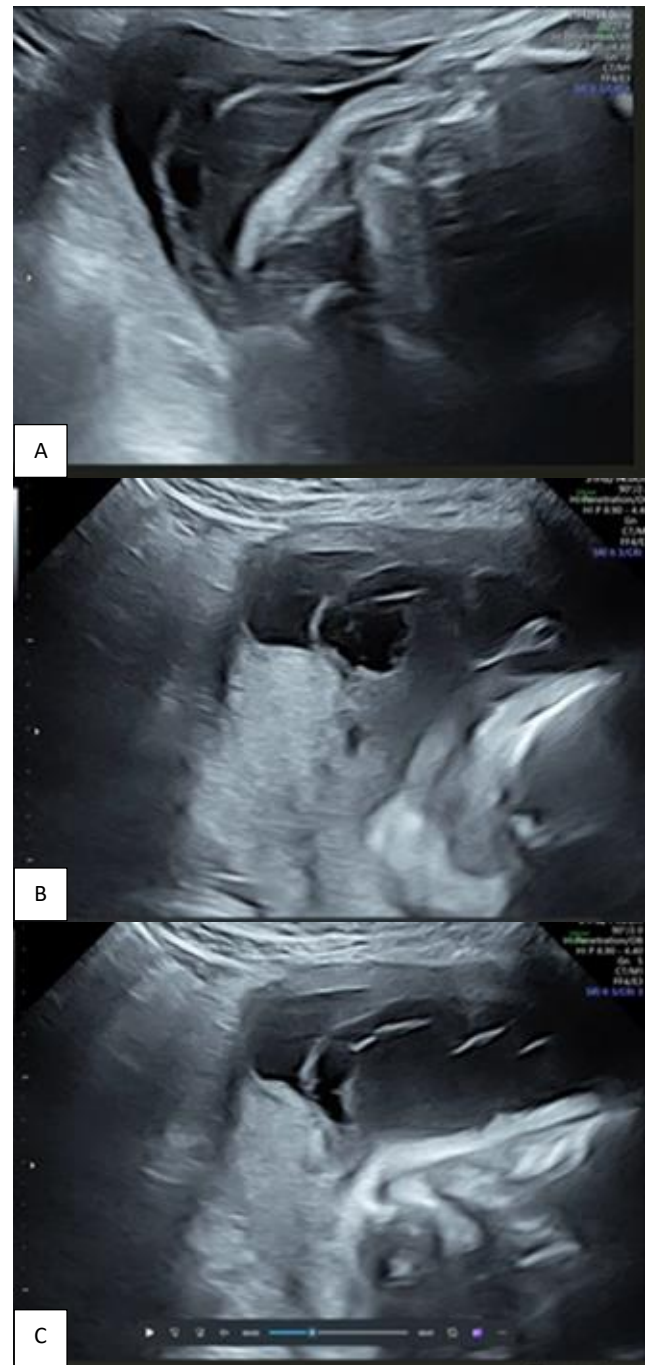


Figure 1 (A-C): Scan images at 28 weeks showing partial chorio-amniotic membrane separation.

The plan was to deliver this baby via a caesarean section at 29+5 weeks after a course of steroids and magnesium sulphate for neuroprotection. She was then admitted to the labour ward with raised BP and symptoms of severe pre-eclampsia. She was started on a higher dose of antihypertensives and started on magnesium sulphate. She had cCTG 4 hourly. She also received 2 doses of steroids.

She was delivered at 29+4 weeks by emergency C-section due to suspected fetal compromise on CTG. The baby had a prolonged period of admission in NICU and had some birthmarks on the body, possibly because of the amniotic fluid bands.

A macroscopic examination of the placenta showed partial sCAMS with no evidence of cord occlusion by amniotic bands. Maternal vascular malperfusion was suggested in histopathology by the presence of accelerated villous maturation.

DISCUSSION

CAMS can be diagnosed after 16 weeks of gestation, as chorion and amnion fusion are normally complete before that. Iatrogenic CAMS is more common and is known to be associated with preterm delivery and other adverse perinatal outcomes. sCAMS are rare and poorly understood.

According to a systematic review of sCAMS in singleton gestations, sCAMS seem to be associated with an increased risk of preterm birth compared with the general population, along with other adverse perinatal outcomes including structural anomalies, aneuploidy, amniotic bands, and IUFD.⁸ We saw similar results in our case report with preterm delivery, which was indicated due to fetal concerns and amniotic bands on the baby.

According to Levine, membrane separation causes decreased mechanical resistance since the chorion and amnion normally reinforce one another.^{9,10} In iCAMS, resistance decreases when membranes are traversed during a procedure, reducing membrane integrity and predisposing to PTB. Weakened membranes may also increase subclinical infection risk, further increasing spontaneous preterm birth risk.

In sCAMS, an intrinsic membrane resistance defect may weaken and separate membranes. Furthermore, the amnion is an avascular structure dependent on surrounding structures including the decidua underlying the chorion for nutrients.^{11,12}

Initial separation may reduce the membranes' nutritional supply, weakening them and diminishing their integrity and barrier function. This increases the risk of infection and, consequently, raises the overall risk of preterm delivery, especially spontaneous preterm birth.

sCAMS have been linked to a higher risk of intrauterine fetal demise (IUFD) and spontaneous miscarriage with varying incidences reported. According to a systematic review of literature, most IUFDs in sCAMS cases resulted from amniotic band-mediated cord strangulation, a complication also associated with iCAMS.⁸ Our case report also had an amniotic band, with the baby presenting signs of an amniotic band postnatally.

Skin and connective tissue disorders, including ichthyosis prematurity syndrome, epidermolysis bullosa with pyloric atresia, and restrictive dermopathy, may be connected to sCAMS due to the shared ectodermal origin of the epidermis and amnion.^{12,14} These disorders result in impaired skin development leading to thin rigid skin (restrictive dermopathy), epidermal sloughing (ichthyosis prematurity syndrome), or blistering (epidermolysis bullosa).¹⁵⁻¹⁷ Due to their shared embryological origin, the mutations causing these disorders might also negatively impact the normal development of fetal membranes. This association further supports the hypothesis that an intrinsic membrane defect underlies sCAMS.

Structural anomalies in sCAMS were also investigated. Limb abnormalities frequently resulted from amniotic bands. Other anomalies in sCAMS include contractures and club feet. These findings can be explained by restricted fetal movement due to extrinsic compression. Membrane separation alone can limit movement if the fetus becomes trapped against a surface.¹⁸

CONCLUSION

In conclusion, spontaneous CAS may be connected to an increased risk of preterm delivery and negative perinatal outcomes, including amniotic bands, structural anomalies, aneuploidy, and IUFD. As there are no large-scale studies and optimal follow-up guidelines for cases with scam, inpatient management after viability, including fetal heart rate monitoring and serial ultrasounds, may be considered for this high-risk population. The current evidence for this association is quite limited, as most studies to date are case reports and case series. To fully understand the risks associated with sCAMS and its varied underlying causes. We hope our case report helps in better recognition of the abnormality and encourages more reporting and studies of similar cases to be able to have a better understanding of the full implications of such sonographic findings. Moreover, increased reporting will help obstetricians to mitigate the fetal and maternal risks associated with sCAMS.

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