Umbilical Cord Constriction by an Amniotic Band after Amniocentesis Resulting in Foetal Demise

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ABSTRACT

Amniotic band syndrome (ABS) is defined as fibrous strands entangling foetal parts, resulting in malformation, deformation, disruption, or even foetal demise. Early diagnosis is necessary for timely intervention and to lower undesirable pregnancy-related events. We report a 28-year pregnant woman (gravida 2, para 0) who presented at 39 weeks of gestation for foetal demise. The pregnancy history was unremarkable except for an early second-trimester amniocentesis. Medical induction was conducted. The clinical examination of the foetus showed an amniotic band tightly constricted to the umbilical cord at 6 cm near the placental origin, leading to the interruption of blood flow. Although rare during pregnancy, clinical suspicion of amniotic band syndrome is necessary, especially in women who undergo invasive prenatal procedures. Prenatal diagnosis of ABS remains challenging for obstetricians. However, selected women with increased risk may benefit from intensive ultrasonographic evaluation.

Key Words: *Amniotic band syndrome, Foetal demise, Amniocentesis, Prenatal diagnosis.*

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INTRODUCTION

Amniotic band syndrome (ABS), also known as constriction ring syndrome (CRS), is a group of congenital malformations that refers to the entanglement or entrapment of foetal parts by fibrous strands in the amniotic sac.[1] ABS occurs sporadically with an incidence ranging from 1 in 1,200 to 1 in 15,000 live births.[2] There is no augmented risk of genetic or chromosomal abnormalities. ABS can affect any part of the foetal body.[3-5] Most of the reported cases mainly focus on three categories: (A) Defects of limb, (B) craniofacial defects, and (C) defects of visceral organs.[5] The umbilical cord is rarely encircled by amniotic bands, however, it can cause catastrophic events, including foetal distress or even foetal loss. Early diagnosis is important for clinical intervention. However, antenatal diagnosis of ABS remains challenging for obstetricians. Despite that, a presumptive diagnosis may be made based on the ultrasound features. Most of the time, the amniotic strand cannot be clearly displayed, especially in cases involving the umbilical cord. Most cases of ABS are diagnosed after birth.

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Here, we report a case of ABS with foetal demise at term after second-trimester amniocentesis by the amniotic bands entangling the umbilical cord, intending to call attention to this syndrome among the obstetricians and spotlight some significant points in the assessment and management of ABS.

CASE REPORT

A 28-year woman gravida 2 para 0, at 39 weeks and four days of gestation, presented at the obstetric emergency ward with a complaint of diminished foetal movements for four hours. The patient followed up exactly on the schedule of prenatal care examination in our hospital and underwent amniocentesis during second trimester since she was a carrier of haemophilia A. The amniocentesis results showed that the baby had a haeterozygous variation on the *F8* gene. The patient had no history of teratogenic or addictive drug intake. Prenatal examination did not indicate any abnormalities.

Her general condition was normal. Obstetric examination revealed no uterine contractions, no vaginal bleeding, and no cervical dilation, but disappearance of foetal heart rhythm. Intrauterine foetal death was then confirmed by an ultrasound scan. After the informed consent of the couple, induction of abortion with mifepristone and oxytocin was attempted. The patient gave birth to a female stillborn baby with a birth weight of 3110 g. The clinical examination of the foetus showed that the umbilical cord was loosely wrapped around the neck for two cycles. The umbilical cord measured 105 cm in length and was twisted for about 35 turns. Amniotic bands were found to be tightly constricted to the umbilical cord at 6 cm near the placental origin, leading to blood flow occlusion (Figure 1 A-D). The

patient refused to do further foetopathological examinations. The patient recovered well without any complications and was discharged on the second day after delivery.

Table I: Pathogenesis of amniotic band syndrome.

Pathogenesis	Year	Reference
Embryonic dysplasia theory (endogenous theory)	1930	Streeter *et al.*[6]
Lack of mesodermal development in constriction bands	1961	Patterson *et al.*[4]
Early amnion rupture sequence (exogenous theory)	1965	Torpin *et al.*[3]
Vascular disruption theory	1981	Van Allen *et al.*[7]

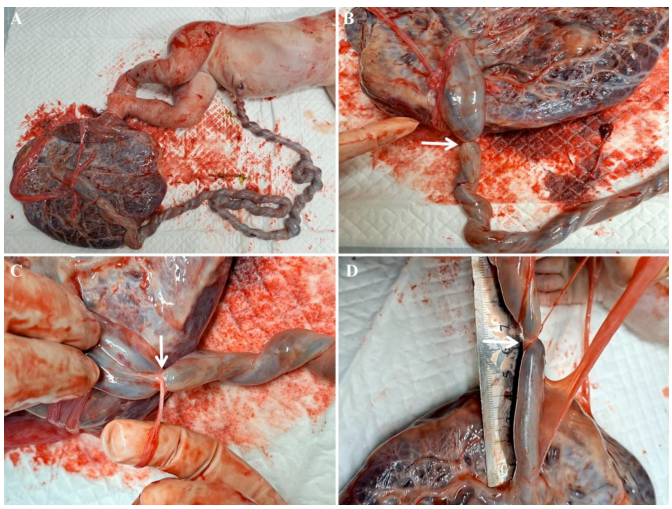

Figure 1: Findings of the placenta and cord. (A) The placenta and umbilical cord. The full length of the cord was 105 cm. The cord twisted about 35 turns (B-D) Amniotic bands tightly encircled the umbilical cord causing blood flow interruption (white arrow).

DISCUSSION

It is of vital significance to appropriately diagnose and treat ABS as it is related to a broad spectrum of clinical manifestations ranging from minor scarring to severe tissue deformities, and even foetal demise. We report a rare presentation of ABS affecting the umbilical cord which resulted in an irreversible and fatal outcome of the foetus. Most of the reported cases of ABS are sporadic without any chromosomal anomalies. Regarding the pathogenesis of ABS, several theories have been elucidated (Table I).[3,4,6,7] Among these theories, the early amnion rupture sequence (exogenous theory) remains the most widely accepted one.[3] Mesoblastic fibrous bands emanate from the chorionic side of the ruptured amnion, thus entangling or entrapping the foetal parts, causing decreased blood supply, oedema, necrosis, and amputation. In most cases, severe anomalies arise from an early amnion rupture and milder malformations result from the late amnion rupture. Although Patterson's theory is not widely accepted, the classification of amniotic strands in limbs is based on Patterson's criteria.[4] He pointed out that these constriction bands are abnormal skin creases due to a lack of mesodermal development.

Additionally, several high-risk factors have been reported, including the use of recreational medicines, the use of misoprostol for termination in the first trimester, chorion villous sampling, amniocentesis, trauma, uterine malformations or placental abnormalities, etc.[8,9] In this case, we hypothesised that ABS might be explained by the exogenous theory. The amniocentesis performed in her second trimester led to amnion rupture, thus emanating fibrous bands that tightly encircled the umbilical cord and eventually blocked the blood flow. However, none of these theories or risk factors could fully explain the variety of manifestations noted in foetuses affected by ABS.

Due to the rarity and variable presentations of ABS, developing a standardised diagnostic and treatment algorithm is quite challenging for the obstetricians. Early diagnosis is indispensable for timely management. Exclusion of ABS is recommended when gross foetal anomalies are present on foetal ultrasound. It is quite challenging to detect the amniotic strands in the first trimester, especially when extremities are involved. In the second and third trimesters, it is relatively easier to visualise the representative abnormalities caused by ABS. The 3-dimensional imaging, computed tomography (CT), or magnetic resonance imaging (MRI) may improve diagnostic accuracy. However, a CT scan is not recommended as the first choice due to its radiation risks to the foetus. Yet it is worth noting that diagnosis is prone to be missed in the antenatal period, especially in cases with minor malformations or in parts difficult to detect.[10] Taking this case as an example, umbilical cord constriction by amniotic strands is not diagnosed prenatally, since it is not feasible to systematically inspect the umbilical cord in every aspect by ultrasonic technology. But Doppler colour-flow mapping and monitoring of vascular resistances may help. With regard to the management of cases of umbilical cord strangulation by amniotic bands, continuous foetal heart rate tracing is considered an effective method. However, it is not a routine practice to perform continuous monitoring of foetal heart-rate during prenatal examinations. So far, no standard diagnostic tool is of high sensitivity and specificity to predict ABS. Further studies are needed to develop more effective diagnostic and monitoring methods.

Both the treatment and the prognosis of ABS depend on the timing of diagnosis and the severity and the sites of the malformations. Most of the cases receive surgical correction after birth. Though fetoscopic technology remains a relatively new field, some cases may benefit from foetal endoscopic intervention.[5,10] However, medical-induced termination is suggested if severe craniofacial and visceral malformations or foetal demise are indicated.

In conclusion, ABS is remarkably a rare but dangerous embryofoetopathy during pregnancy, which can lead to adverse foetal outcomes. Prenatal diagnosis of ABS remains challenging for obstetricians; however, selected women with increased risk may benefit from intensive ultrasound assessment and foetal heart-rate tracing. As invasive antenatal testing is becoming more and more common, obstetricians need to focus on this potential complication so that such cases can be offered special monitoring during the entire pregnancy and labour. The obstetrical treatment should be individually conducted. Medical-induced abortion should be proposed in cases of severe and lethal malformations.

PATIENT'S CONSENT:
The patient provided her written informed consent to publish this case and allowed for the publication of any accompanying images.

COMPETING INTEREST:
The authors declared no conflict of interest.

AUTHORS' CONTRIBUTION:
XT: Drafted and revised the main manuscript.
LJ, GD: Collected the data.
LF: Prepared Figure 1 and Table I.
JS: Designed the study and revised the final paper.
All authors approved the final version of the manuscript to be published.

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