

Case Report

Strangulation of umbilical cord by an amniotic band: A rare cause of still-birth in the third trimester

Hakan Timur, Cantekin Iskender*, Halil Korkut Daglar, Oktay Kaymak, Ayse Kirbas, Nuri Danisman

Department of Perinatology, Dr Zekai Tahir Burak Research and Training Hospital, Ankara, Turkey

Abstract

The amniotic band syndrome (ABS) is a relatively rare entity with an estimated incidence of 1 in 1200 to 15000 livebirths. Strangulation of the umbilical cord can also occur in ABS. While it is assumed that umbilical cord is involved in 10% of cases, severe perinatal morbidity or mortality is rarely reported due to strangulation of the umbilical cord. Here, we report a case of umbilical cord strangulation causing still birth at term without preceding abnormalities in fetal well being tests. The fetus exhibited normal growth and had normal modified biophysical profile. The patient did not report decreased fetal movement except for the day of admission. In the post-mortem examination, we observed that there was no well established constriction ring or edema distal to the point of strangulation. The present case shows that umbilical cord involvement in ABS may lead to fetal death at term without antecedent signs.

Key words:

Amniotic band, placental pathology, umbilical cord strangulation

Introduction

The amniotic band syndrome (ABS) is a relatively rare entity with an estimated incidence of 1 in 1200 to 15000 livebirths [1]. The exact etiology of ABS is yet to be defined. But a popular theory is an early rupture in the amniotic membrane and subsequent strangulation and ischemia of fetal parts [2]. Fetal limbs are frequently affected in ABS while a variety of other fetal deformities such as anencephaly, cleft lip, gastroschisis, omphalocele have also been demonstrated [1,3]. Strangulation of the umbilical cord can also occur in ABS. While it is assumed that umbilical cord is involved in 10% of cases, severe perinatal morbidity or mortality is rarely reported due to strangulation of the umbilical cord [4-9]. Here, we report a case of umbilical cord strangulation due to ABS causing still birth at term.

Article history:

Received 16 10 2014

Accepted 24 03 2015

* **Correspondence:** Cantekin Iskender,
Adalet sitesi-2 Kehrubar Sk 19-a,14 B.Esat, Ankara, Turkey,
E-mail: c_iskender@yahoo.com
Phone: +905334505230

Case presentation

A 39-year-old gravida 3 para 2 woman with a previous cesarean section woman admitted with a complaint of reduced fetal movements at the 39th gestational week according to her last menstrual period. Her past medical and obstetrical history was unremarkable with two previous healthy term deliveries. Her current pregnancy was uneventful with low risk first trimester combined screening and second trimester triple screening tests for aneuploidy and major congenital malformations have been excluded by fetal ultrasonographic scan. Her 50 grams oral glucose challenge test was 116 mg/dl. Modified biophysical profile tests performed weekly after 36th gestational weeks were normal and she did not report decreased fetal movements until the day of admission. On admission, sonographic examination revealed absent fetal cardiac activity. The fetus was at vertex presentation, amniotic fluid index was normal. Her complete blood count, blood biochemistry and coagulation profile were normal. She underwent cesarean delivery on the day of admission. A 3280 gram exitus female fetus was delivered.

An amniotic band was detected at the right lower extremity 2 cm distal to the knee which also trapped the umbilical cord (Figure 1a). There was no constriction ring at the point of strangulation or distal edema (Figure 1b). The pressure marks on the right limb at the site of strangulation faded soon after we relieved the strand. There were no additional gross morphological abnormalities of the fetus or placenta.

Figure 1.



Strangulation of umbilical cord by amniotic band 2 cm distal to right knee (a) and acute pressure marks at the site of entrapment (b).

Discussion

In the present case, amniotic band has resulted in stillbirth without preceding abnormalities in fetal well being tests. The fetus exhibited normal growth and had normal modified biophysical profile. The patient did not report decreased fetal movement except for the day of admission. In the post-mortem examination, we observed that the right lower extremity was normal without signs of prolonged vascular disruption. There was no well established constriction ring or edema distal to the point of strangulation. The pressure marks on the right limb at the site of strangulation faded soon after we relieved the strand. These findings suggest that the strangulation took place within hours. An autopsy was not performed due to denial of parents.

The diagnosis of amniotic band can be easily made with prenatal ultrasonography. In 10% of the cases the band may involve umbilical cord [5]. Despite the involvement with umbilical cord is not rare in patients with amniotic bands, complications related to umbilical cord strangulation were rarely reported previously. To the best of our

knowledge, there had been less than 60 cases of stillbirth due to umbilical cord strangulation. The majority of cases occurred in the second trimester [4]. More rarely, it has also been defined in the third trimester and at term [7-9].

In patients with prenatally detected amniotic band, serial follow-up is warranted with particular attention to the extremities. In the initial phase, strangulation leads to distal edema which is followed by vascular disruption eventually leading to limb amputation [10]. The series of events leading to limb reduction is well established and can be easily detected during follow-up in these patients. However, recognition of umbilical cord strangulation can be difficult. In most cases, prenatal diagnosis was not possible except in only a single case previously [5]. But in this case, the diagnosis was made during fetoscopy and neither prenatal ultrasonography nor magnetic resonance imaging led the authors to suspect umbilical cord strangulation.

In affected cases, reliable predictive features are often absent. In a previous report by Sifakis et al., elevated alpha fetoprotein and human chorionic gonadotropin (hCG) was reported in a case of amniotic band syndrome resulting in second trimester fetal demise [11]. The authors have suggested that elevated hCG was seen due to placental response to fetal hypoxia. Severe intrapartum fetal distress and intrauterine growth restriction and fetal death have been described in term pregnancies previously [9]. However, fetal demise has been recorded to occur without preceding findings that suggest fetal hypoxia as in our case [8].

The strangulated cord segment can be adjacent to a fetal extremity or can be a free cord loop [6,8]. Umbilical cord strangulation must be suspected in amniotic band syndrome cases in which the cord is persistently adjacent to the affected limb. Additionally, through ultrasonographic evaluation of the umbilical cord is necessary. Fetoscopic examination could be considered when umbilical cord strangulation is suspected as the consequences can be catastrophic [12]. In conclusion, the present case shows that amniotic band syndrome involving umbilical cord may lead to fetal death at term without antecedent signs.

Conflict of Interest

Authors declare no conflict of interest

References

1. Marino T. Ultrasound abnormalities of the amniotic fluid, membranes, umbilical cord, and placenta. *Obstet Gynecol Clin North Am* 2004; 31: 177–200.
2. Torpin R. Amniochorionic mesoblastic fibrous rings and amniotic bands: associated constricting fetal malformations or fetal death. *Am J Obstet Gynecol* 1965;91:65-75.
3. Hata T, Tanaka H, Noguchi J. 3D/4D sonographic evaluation of amniotic band syndrome in early pregnancy: a supplement to 2D ultrasound. *J Obstet Gynaecol Res.* 2011;37:656-660.
4. Heifetz SA. Strangulation of the umbilical cord by amniotic bands: Report of 6 cases and literature review. *Pediatr Pathol* 1984; 2: 285–304.
5. Peiró JL, Carreras E, Soldado F, et al. Fetoscopic release of umbilical cord amniotic band in a human fetus. *Ultrasound Obstet Gynecol.* 2009;33:232-4.
6. Hallak M, Pryde PG, Qureshi F, Johnson MP, Jacques SM, Evans MI. Constriction of the umbilical cord leading to fetal death. A report of three cases. *J Reprod Med* 1994; 39:561–565.
7. Duckworth HL, Leather AT, Jessop F. Intrauterine death and growth restriction at term, secondary to an umbilical cord amniotic band. *J Obstet Gynaecol.* 2011;31:547.
8. Lurie S, Feinstein M, Mamet Y. Umbilical cord strangulation by an amniotic band resulting in a stillbirth. *J Obstet Gynaecol Res.* 2008;34:255-7.
9. Larciprete G, Montagnoli C, Fusco P. Severe fetal distress and umbilical cord strangulation. *Case Rep Med* 2011;2011:645487.
10. Tadmor OP, Kreisberg GA, Achiron R, Porat S, Yagel S. Limb amputation in amniotic band syndrome: serial ultrasonographic and Doppler observations. *Ultrasound Obstet Gynecol.* 1997;10:312-5.
11. Sifakis S, Mantas N, Konstantinidou A, Koukoura O, Avgoustinakis E, Koumantakis E. A stillborn fetus with amniotic band syndrome and elevated levels of alpha-fetoprotein plus beta-human chorionic gonadotropin: a case report. *Fetal Diagn Ther* 2008;24:111-114.
12. Keswani SG, Johnson MP, Adzick NS, et al. In utero limb salvage: fetoscopic release of amniotic bands for threatened limb amputation. *J Pediatr Surg* 2003; 38: 848–851.