

# Intrauterine fetal death after multiple umbilical cord torsion—complication of a twin pregnancy following assisted reproduction

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## Abstract

**Background** Patients requiring assisted reproductive techniques may have a higher rate of congenital malformations. Some rare complications of pregnancy might be related to such abnormalities. Torsions of the umbilical cord resulting in fetal death have previously been described exclusively in pregnancies following spontaneous conception.

**Case** The case of 37 year old gravida I, para O woman with a twin pregnancy after intracytoplasmatic sperm injection and intrauterine death of one twin at approximately 30 weeks' gestation is presented. The surviving twin was delivered by cesarean section at 31 weeks after spontaneous onset of labor and recurrent fetal bradycardia. The intra-operative situs showed that the demised twin had suffered from multiple umbilical cord torsions leading to intrauterine hypoperfusion.

**Conclusion** Umbilical torsion leading to fetal death might represent a previously unrecognized complication in women requiring assisted reproductive techniques, but this problem is known to occur in pregnancies achieved by natural methods.

**Keywords** Intracytoplasmatic sperm injection · Fetal hypoxia · Multiple pregnancy · Stillbirth

## Introduction

Intrauterine fetal death is a devastating complication of pregnancy. The overall incidence of stillbirth is reported to be around 1.2% with an incidence after 20 weeks of 1%, after 28 weeks of 0.4% and at term of 0.2% [1]. The underlying reasons can be divided in maternal, fetal and placental causes. Umbilical cord complications are the most common cause for fetal demise in the third trimester [2]. Unfortunately these complications are regarded as unpredictable and unpreventable. Nuchal umbilical cords, knots and insertion anomalies like velamentous insertions are relatively frequent findings and not necessarily associated with a negative fetal outcome. Whereas an umbilical cord prolapse occurs under labor, cord constrictions can occur at anytime during second or third trimester pregnancy. Constrictions are rather rare events which, when complete and persistent, lead to acute intrauterine asphyxia.

Here the case of a di-chorionic, di-amniotic twin pregnancy is reported during which intrauterine death of one twin occurred due to umbilical cord constriction following multiple umbilical cord torsions. This complication has not been previously described in a pregnancy following assisted reproduction techniques.

## Case report

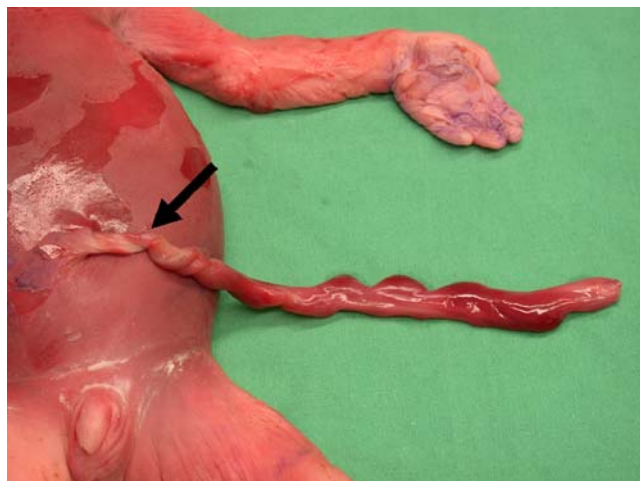
A 37 year old gravida 1, para O woman at 28 weeks' gestation was admitted to hospital after she had noticed reduced fetal movements. This twin pregnancy resulted

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from intracytoplasmic sperm injection (ICSI) and transfer of two embryos. Prior amniocentesis was performed in the 16th week and showed a normal karyotype in both fetus (female and male). Starting during the 17th week the mother developed contractions. During a routine ultrasound the intrauterine death of the male fetus was diagnosed. Within 1 week the mother experienced contractions with increasing intensity and subsequent shortening of the cervix. Also fetal heart rate monitoring showed alterations in the vital twin so that cesarean section was indicated and performed. Intraoperatively multiple torsions of the umbilical cord of the demised twin were found which had led to a critical shortening in fetal blood supply. A complete constriction had occurred approximately 7 cm proximal to the fetal insertion side (see arrow Fig. 1). The surviving female twin had a birth weight of 1,480 g and an APGAR score of 7/8/8, the arterial pH was 7.46.

The post mortem examination of the male fetus showed a weight of 1,115 g, a crown–rump length of 27 cm and a foot length of 5.5 cm in accordance with a gestational age of 27 weeks according to Patten and Streeter [3]. The fetus did not reveal any abnormalities or dysmorphic signs. Internal organs showed a concordant development with open fetal blood vessels and an orthologic lung development in its final stage. The umbilical cord had a diameter between 1 and 2 cm and a length of 37 cm containing three blood vessels. The histologic examination of the placenta showed a regular dichorial diamniotic twin placenta without signs of inflammation.



**Fig. 1** Complete constriction of the umbilical cord approximately 7 cm from the fetal insertion (arrow)

## Discussion

Umbilical cord torsions are considered to be a sporadic event and a rare cause for intrauterine fetal death. The actual obstruction can occur at any site of the umbilical cord and has been reported to occur in the second and third trimester. A case study reporting this phenomenon in three subsequent pregnancies in one woman suggested a potential familial clustering [4]. To our knowledge this is the first report on this problem during a twin pregnancy following in vitro fertilization. All other published cases of umbilical cord torsion and subsequent fetal death occurred in pregnancies following spontaneous conception. Umbilical cord torsions can either lead to chronic hypoxia with critically reduced blood flow, oligohydramnios and fetal growth retardation [5] or can acutely obstruct fetal–placental circulation with subsequent death. The fact that the fetus showed a grossly concordant growth before intrauterine death was diagnosed suggests a more acute obstruction event. The underlying mechanism for umbilical cord torsions remains obscure. Previous reports emphasized the absence of Wharton's jelly leading to a narrowing of the umbilical cord and the vascular lumen [6]. However, this finding was not present in our case. The reported lengths of the obstructed cords at the time of death were variable ranging from 34 to 120 cm making it unlikely that this feature is of considerable importance [7]. Interestingly, other reports also mentioned prior amniocentesis during the pregnancy where the complication occurred [6]. Fetal mobility is facilitated by increased amounts of amniotic fluid, whereas oligohydramnios and amniotic bands are associated with pulmonary hypoplasia [8] and contractures due to the lack of fetal movements [9]. Increased mobility due to polyhydramnios is associated with a higher incidence of true knots of the umbilical cord [10].

In summary, umbilical cord torsion with intrauterine fetal death is a rare but fatal complication of a twin pregnancy. The prevention of this complication is virtually impossible. The general consideration of umbilical cord complications as underlying cause for chronic intrauterine hypoxia with reduced fetal movements, growth retardation or oligohydramnios should be mandatory. However, this case shows, that a multitude of torsions can occur without apparent sonographic signs of fetal retardation and that also in the case of multiple torsions intrauterine fetal death might be the first clinically apparent manifestation of this severe complication. Since this is the first case of umbilical cord torsion and subsequent fetal death following assisted reproductive techniques, we cannot draw reliable conclusions regarding the frequency of this condition.

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