J. Fetal Med. (December 2015) 2:183–186
DOI 10.1007/s40556-015-0069-4

BRIEF COMMUNICATION

Conjoined Twins Following Assisted Reproduction Technique

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Received: 2 August 2015 / Accepted: 8 December 2015 / Published online: 21 December 2015

Abstract The incidence of conjoined twins as reported in a worldwide epidemiological study is 1.47 per 100,000 births. It varies from 1 in 45,000–200,000 births. Conjoined twins following assisted reproduction are even rarer and we report one such case. A 36-year-old woman who conceived by intracytoplasmic sperm injection, was referred to the feto-maternal unit for nuchal translucency scan. A diagnosis of thoracopagus conjoined twins was made on ultrasound at 12 weeks 3 days of gestation. The couple was counseled in detail and they opted for termination. Pregnancy was terminated medically and the ultrasound diagnosis was confirmed. Assisted reproduction techniques involve a risk of conjoined twins and the present case report supports this. Early diagnosis of conjoined twins and determination of the type of fusion is possible. This aids in discussing the prognosis and counseling the couple regarding options of termination or continuation of pregnancy.

Keywords Conjoined twins · Intracytoplasmic sperm injection (ICSI) · Thoracopagus · Omphalocele

Introduction

Conjoined twinning is a rare complication of monozygotic twin pregnancy. Its high rate of morbidity and mortality makes early prenatal diagnosis significant. Prognosis depends on the type and site of fusion, degree of sharing of the vital organs, and associated anomalies.

Report of Case

A 36-year-old primigravida who conceived by intracytoplasmic sperm injection (ICSI) after 11 years of marriage, was referred to the fetomaternal unit for nuchal translucency scan. A diagnosis of thoracopagus conjoined twin was made on ultrasound at 12 weeks 3 days of gestation.

Ultrasonography showed two heads in the same plane very close to each other with no membrane in between. The fetuses were positioned face to face, bodies fused from upper chest with a single heart, with parallel arrangement of the spines. The fetal bodies were not separable and lacked independent movement. They maintained similar position even after observing for a prolonged time (Fig. 1).

The couple was counseled in detail and they opted for termination. The pregnancy was terminated medically and the ultrasound diagnosis was confirmed. The fetuses were males and they were united at the thoraces. There was an omphalocele and a single umbilical cord (Fig. 2). The couple refused autopsy.

Discussion

The incidence of conjoined twins as reported in a worldwide epidemiological study is 1.47 per 100,000 births [1]. It varies from 1 in 45,000–200,000 births. Conjoined twins following assisted reproduction are even rarer. It has been documented that assisted reproductive technology (ART) might increase the risk of conjoined twins [2]. Multiple factors might play a role in this, like ovulation induction,

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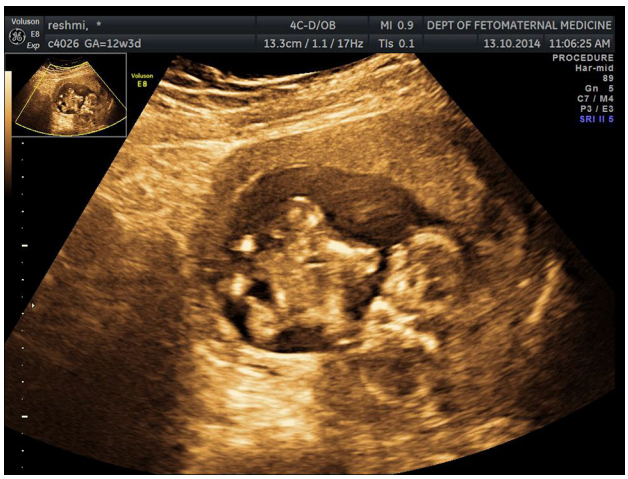

Fig. 1 Ultrasound image showing fetuses facing each other, two heads in the same plane with bodies fused from upper chest

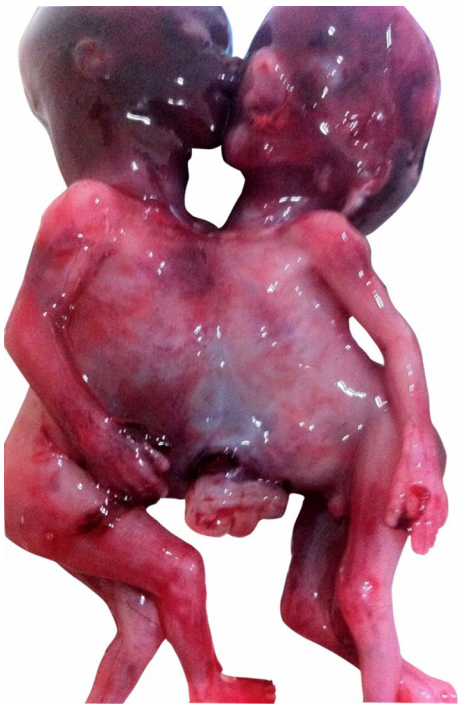

Fig. 2 Thoracopagus male conjoined twins with omphalocele

ICSI, assisted hatching, in vitro culture condition, etc. [3]. There is a female to male ratio of 3:1 [4].

There are two theories of origin:

Theory of fission: According to this theory, incomplete division of the monozygotic embryonic disc occurs after day 13 of conception.

Theory of fusion: The theory of fusion states that the previously separated embryonic discs undergo secondary fusion. The fusion theory is more accepted recently [5].

It has been reported that the rate of monozygotic twins, after ART, has risen about 2.25 times above the background rate [6]. The occurrence of monozygotic twinning after assisted reproduction has been linked by different authors to ovulation induction [7], gonadotropin treatment [8], maternal age either younger [9] or older [10], culture conditions [11], day 5 embryo transfer [12–15], and prolonged culture due to abnormal hatching [16] or apoptosis in the embryo [17]. Recently, Sobek et al. [18] have proposed that the high incidence of monozygotic twins following assisted reproduction techniques is conditioned by hereditary factors and that good ovarian function only facilitates the expression.

First trimester ultrasound enables accurate determination of chorionicity and amnionicity which is of great value in assisted reproduction. Apart from being an effective method of screening for aneuploidies, the 11–14 weeks scan can also identify many major nonchromosomal abnormalities [19]. Though feasible, early first trimester diagnosis of conjoined twins is associated with false positive rates owing to limited fetal movements at this gestation. Also, it has been reported that first trimester 3D imaging does not add any advantage to 2D [20]. A classification of conjoined twins based on the site of fusion was put forward by Winkler et al. [21] (Table 1).

The incidence of major congenital anomalies in conjoined twins is 10–20 %, excluding the defects at the site of fusion [22]. The anomalies include congenital heart defects, spina bifida, cystic hygroma, limb defects, abdominal wall defects, and diaphragmatic hernia [23]. Conjoined twins, especially the ones with major congenital anomalies, have a high mortality [24, 25]. About 60 % die in utero and another 35 % are stillbirth or die within 24 h of birth [21]. The success of separation surgeries depends on the complexity of fusion, degree of sharing of the organs, severity of abnormalities, and the post-operative clinical conditions.

Conclusion

ART has increased the rate of conjoined twins as is evident from this rare case following ICSI. Early diagnosis of conjoined twinning and determination of the type of fusion

Table 1 Classification of conjoined twins modified from reference [21]

Type	Description	Incidence (%)
Ventral union		
Cephalopagus	Fused from head to umbilicus, lower abdomen and pelvis separated. Each twin with two extremities	11
Thoracopagus	Thoraces are fused with a shared heart or a single interatrial vessel	20–40
Omphalopagus	Similar to thoracopagus without shared heart or a single interatrial vessel	18–33
Ischiopagus	Joined at the pelvis. External genitalia and anus always shared	6–11
Dorsal union		
Craniopagus	Joined at the skull except the face and foramen magnum. Skull, brain and meninges shared	2
Pyopagus	Joined at the sacrococcygeal and perineal regions. Anus shared and rectums separate. Spinal cord may be shared	18–28
Rachipagus	Fused dorsally above the level of sacrum	Rare
Lateral union		
Parapagus	Twins have side-by-side connection with shared pelvis and variable cephalad sharing defined as follows:	28
Dithoracic	Separate thoraces and heads	
Dicephalic	Separate heads, fused thoraces	
Diprosopus	Single head with two faces on the same side	

is possible. This aids in discussing the prognosis and counseling the couple regarding options of termination or continuation.

Compliance with Ethical Standards

Conflict of interest None.

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