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# A case of frozen embryo transfer and thoracopagus diagnosed at time of nuchal translucency

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## Abstract

We present a case of conjoined twins diagnosed after single euploid, day 5 frozen embryo transfer. The diagnosis was made at the 12-week nuchal translucency ultrasound via vaginal sonography. A repeat ultrasound at 13 weeks revealed thoracopagus twinning with a single heart and possible abdominopagus with sharing of the liver. There were two separate craniums and all 4 extremities of each fetus were seen moving freely. Large cystic hygromas were noted for both fetuses. The patient underwent therapeutic abortion at 14 weeks. Given the rarity of conjoined twins in the setting of early frozen embryo transfer, this case significantly adds to the body of literature.

## Keywords

Embryo transfer; Thoracopagus; Conjoined twins; In vitro fertilization (IVF); Monozygotic; Ultrasound; Pregnancy; Obstetrics.

## Introduction

A conjoined twin pregnancy is a rare phenomenon that is difficult to diagnose via ultrasonography in the first trimester. It is estimated to occur in 1 in 100,000–200,000 pregnancies [1] as a result of monozygotic twinning. Monozygotic Twins (MZT) pregnancies occur in 0.4% of pregnancies conceived spontaneously [2]. Notably, in Assisted Reproductive Technology (ART), the rate of MZT is estimated as 3.2% of pregnancies, which is eight times the background rate [3]. Thus, ART is a risk factor for MZT and subsequently, conjoined twins. The diagnosis of a conjoined twin pregnancy carries a poor prognosis: 40% are stillborn and 50% of surviving twins demise in the neonatal period [4]. We present a case of an advanced maternal age patient, who underwent blastocyst-stage Frozen Embryo Transfer (FET), and was diagnosed with thoracopagus twinning. There are only a few reported cases similar to our experience, whereby conjoined twins were diagnosed after early FET. Our case offers additional support for the fission

theory of pathogenesis, the importance of early ultrasonography and thorough counselling for those with risk factors.

## Case Report

The patient is a 38-year-old G1P0 with a history of infertility who presented at 12 weeks and 2 days (dated by embryo transfer) for her nuchal translucency and was found to have conjoined twins. She had undergone egg retrieval, followed by pre-implantation genetic testing-aneuploidy (PGT-A) and embryo cryopreservation. A single euploid, day 5 frozen embryo, in the blastocyst stage was transferred. She had a viability scan at 6w3d, showing a single viable intrauterine pregnancy consistent with embryo transfer dating. At her scheduled nuchal translucency scan at 12w2d it was noted that conjoined twins were present. For further characterization, she was referred for ultrasound with Maternal Fetal Medicine, which showed a 13-week pregnancy with thoracopagus twinning. The twins were joined ventrally, involving a portion of the abdominal wall, with a shared heart (Figure 1) and liver. There were two separate craniums (Figure 2) and all 4 extremities per each fetus were seen moving freely. Large cystic hygromas were noted for both fetuses (A measured 7.8 mm and B measured 13.0 mm) (Figure 3). Her medical history was otherwise relevant for hypothyroidism, endometriosis, 7 years of infertility (male and uterine factor) and cervical dysplasia. With regards to her endometriosis, she was diagnosed with stage IV endometriosis and underwent a robotic assisted-myomectomy for a 9 cm myoma. She had no relevant family history. She endorsed using marijuana for morning sickness in the first trimester. At the time of diagnosis, she was on conjugated estrogen vaginal inserts, 100 mcg of levothyroxine, progesterone 100 mg capsules and vitamin D. After counseling, she elected for termination of pregnancy. Preoperative Laminaria were placed and she underwent dilation and evacuation at 14 weeks gestation. Intraoperatively, her uterus was 16-week sized, all products of conception were removed and estimated blood loss was 1 liter. This was attributed to a moderate amount of bleeding throughout the procedure in the setting of a twin pregnancy and advanced gestational age. The pathology report was consistent with products of conception.

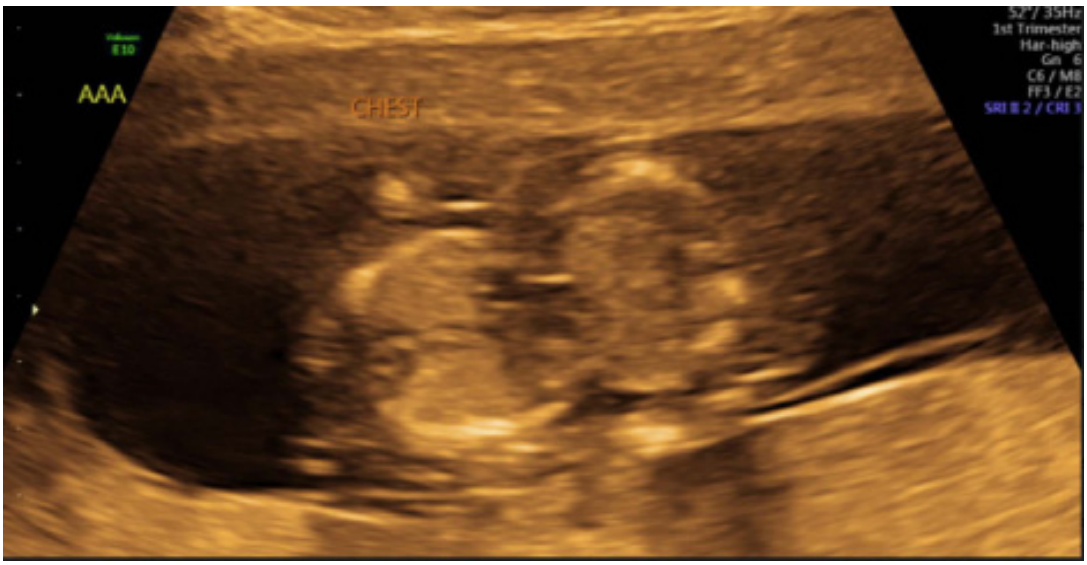


**Figure 1:** Thoracopagus twinning with a single heart.

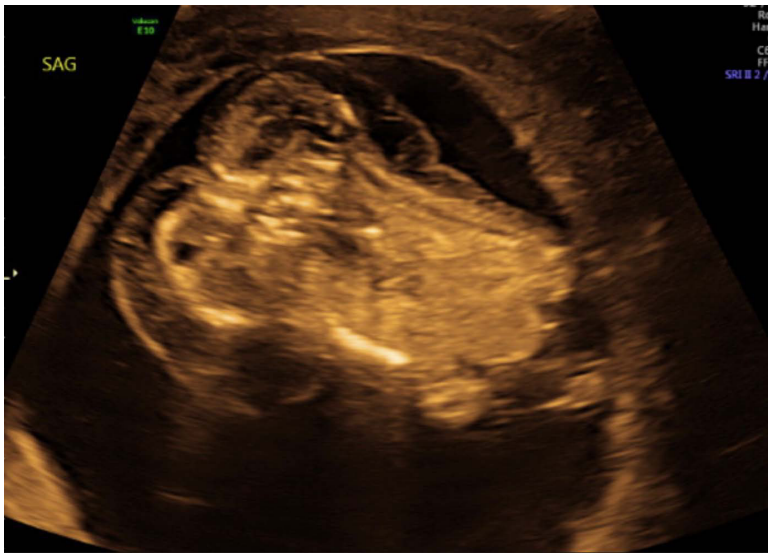


**Figure 2:** Sagittal view of two craniums with cystic hygroma and thoracopagus.

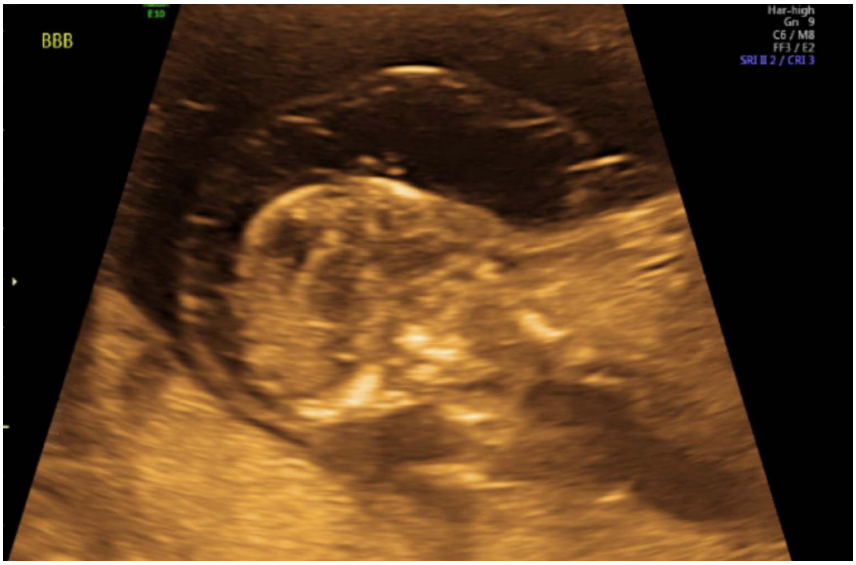


**Figure 3:** Cystic Hygroma.

## Discussion

The phenomenon of conjoined twin pregnancy following embryo transfer was first reported in 1987, with the case of derodidymic monosomic stillborn calf derived by embryo transfer [3]. In 2004, in Australia, two embryos were transferred at four-cell stage on day 3 and at 5 weeks, 1 gestational sac was seen. At 10 weeks, abnormalities were noted on ultrasound and pathology after termination revealed v-shaped mass with two ocular organs, digestive tracts, and livers [4]. In Japan in 2009, two day 5 blastocysts were transferred and at the eighth week of gestation, a triplet pregnancy was diagnosed. There were two gestational sacs: one with a single fetus, and the second with thoracopagus conjoined twins with a shared cardiac structure [5]. In 2010, three additional cases were reported with thoracopagus conjoined twins after frozen embryo transfer at early blastocyst stage (days 2-3) diagnosed via transvaginal ultrasound in the first trimester of pregnancy [6-8]. All pregnancies above failed or were terminated within the first trimester given poor prognosis. There are two theories regarding how conjoined twins come to be: fission

and fusion. The fission theory hypothesizes incomplete separation of embryonic discs at around 13 to 15 days after fertilization [9]. Fusion would occur between two embryos that were initially separated. Some proposed risk factors for monozygotic pregnancy after ART includes maternal age, Intracytoplasmic Sperm Injection (ICSI) [10], ovulation induction [2], blastocyst-stage transfer [11], culture conditions [12], and assisted hatching [13]. There are theories that suggest that some techniques involved as part of ART may produce small punctures in the zona pellucida that allow herniation or pinching and division of the embryo through these openings [5,8]. In this case, the patient was of advanced maternal age and underwent IVF including ovulation induction,

ICSI, and blastocyst-stage transfer. Patients who undergo ART should have early and careful surveillance via ultrasound, as ART is known to increase the risk of MZT. Possible detection of conjoined twins may occur as early as 7 weeks of gestation, although diagnosis in the first-trimester is challenging as fetal movement is limited and it is difficult to discern specific fetal body parts. Careful ultrasound examination should be performed as early as possible after blastocyst transfer with heightened suspicion for conjoined twinning. If conjoined twinning is suspected, it is critical to use ultrasonography to determine the sites of attachment and identify any associated malformations for prognostic determination. Conjoined twins are defined by primary attachment site with suffix "-pagus"; thoracoomphalopagus (28%), thoracopagus (18.5%), and omphalopagus (10%) are the most common types [14]. If termination is not elected, fetal echocardiogram should be performed due to a higher risk of congenital heart disease. Recently, three-dimensional ultrasound imaging has been proposed to provide more information on the extent of twin fusion. Later in pregnancy, it has been proposed that MRI is superior to ultrasonography for fetal assessment, particularly in cases where immediate post-delivery surgical management in indicated [15].

## Conclusion

Appropriate counselling should be provided, particularly when multiple known risk factors for conjoined twins following ART are present. In these cases, early ultrasonographic assessment should be conducted and, if there is concern for conjoined twins further imaging should be conducted for prognostic determination.

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