

Successful dilation and evacuation for second trimester conjoined twin: A case report and systematic review of the literature

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Abstract

Conjoined twins are a rare clinical event. There is limited evidence as to the optimal method of pregnancy termination, particularly in cases of advanced gestational age. We report a successful dilation and evacuation done for conjoined twins at 22 weeks of gestation.

Keywords

Conjoined twins; Dilation and evacuation; Surgical abortion; Thoraco-omphalopagus; Second trimester

Key Clinical Message

Dilation and evacuation can be safely performed for carefully selected cases of conjoined twins beyond 20 weeks' gestations. Adequate cervical preparation, pain control and ultrasound guidance during the procedure are critical for optimal outcomes.

Introduction

Conjoined twins are extremely rare, occurring in about 1 per 50,000 pregnancies and 1 per 250,000 live births. Though prognosis of conjoined twins depends on the degree and location of union, it is generally associated with high perinatal mortality and patients may request termination of pregnancy^{1, 2}. However, there is limited evidence as to the optimal method of pregnancy termination particularly in cases of advanced gestational age. Though medical terminations of conjoined twin pregnancy have been documented up to late second trimester, use of surgical methods is not widely reported^{3, 4}. Here we aim to report a case of conjoined twins successfully managed with dilation and evacuation (D&E) and systematically review previously reported cases to analyze methods of pregnancy termination for conjoined twins in the second trimester.

Case history

A 20-year-old primigravid woman was referred to our hospital at 22 weeks of gestation with a diagnosis of large fetal intraabdominal cysts identified during routine ultrasound examination. In our center, detailed fetal 2D ultrasound anatomic scanning was done, revealing two fetal heads at a fixed position, facing each other (Figure 1). There was a fused chest and abdomen with a single shared distorted heart and one aorta. A single umbilical cord was noted. There was a single shared liver. The kidneys appeared enlarged with multiple non-communicating cysts and thinned-out cortical tissue. Two separate spines were visualized on either side of the uterine cavity (Figure 2). A conjoined, thoraco-omphalopagus twin pregnancy was diagnosed. Fetal karyotyping was offered but declined by the family.

Assessment and counseling were done by a multidisciplinary team composed of obstetricians, fetal medicine specialist, family planning specialist and anesthetists. After discussion on prognosis and options of management, the patient opted for termination of pregnancy. Different options of termination were discussed and patient consented for D&E, with the possibility of reverting to hysterotomy should intraoperative difficulty is encountered.

We performed a two-day cervical preparation. On Day 1, 200 mg oral mifepristone was administered and 5 *Laminaria* were inserted. On Day 2, the patient was admitted and a new set of 10 *Laminaria* were inserted. On the morning of the procedure, she was provided with 400 µg sublingual misoprostol and 200 mg oral doxycycline. After 2 hours she was transferred to the operating room and spinal anesthesia was given. D&E was done under ultrasound guidance. We started the procedure by rupturing the membranes to bring down fetal parts to the lower uterine segment. Initial extraction of fetal parts was done by disarticulating and removing the extremities. Decompression of the thoracic and abdominal cavity allowed further descent and separation of the thoracopagus. The presenting calvarium was decompressed with suction and delivered. Finally, the second twin and placenta were delivered intact. Procedure was completed without complications. Post-procedure tissue examination showed two calvaria and spines, 4 well-formed upper limbs, single thorax and abdomen, and two well-formed and two fused primitive lower limbs. Patient recovered well and was discharged after 24 hours. Follow-up phone call after 2 weeks revealed uneventful course.

Discussion

Conjoined twins are rare. The available management options are usually complex and ample experience with case-management is limited to few centers worldwide¹. Recent advances in antenatal imaging techniques, such as 3D ultrasonography, doppler studies and MRI, enable diagnosis as early as 12 weeks' gestation. In addition, detailed prenatal anatomic scanning will define the extent of organ sharing and inform prognosis^{2, 5}. Early diagnosis followed by thorough counseling on the likely prognosis are crucial for optimal management^{3, 6}. However, as in our case, early diagnosis can be missed and the pregnancy may advance into the second trimester. Other reports from developing countries also show the diagnosis of conjoined twins may be delayed until the third trimester or even up to the time of labor and delivery^{7, 8}.

Conjoined twins with a shared heart are associated with extremely poor prognosis and separation and survival of both twins (or even one) is unlikely^{2, 9}. Given this, our patient decided to terminate her pregnancy.

Pregnancy termination for conjoined twins in later gestation is often accomplished through hysterotomy due to perceived difficulty in vaginal delivery¹⁰. Though details of the methods employed were not described, Brizot et al reported 12 vaginal terminations for second trimester conjoined twins⁹. Similarly, Mitchell et al reported 2 successful induction of late second trimester conjoined twins. However, both patients underwent 2 sessions of laminaria placement prior to administration of uterotonics⁴.

We conducted a systematic search of the electronic databases of MEDLINE, EMBASE and Google Scholar using MeSH and keywords from inception of data base until July 31, 2020 (Additional file1). Bibliographies of the relevant articles were reviewed and then cross-searched to identify further relevant studies. We included all publications in English that specify method of pregnancy termination for conjoined twins in the second trimester (14-28 weeks).

Two authors (FAA and THT) independently performed study screening and data extraction. Titles and abstracts were screened to identify eligible articles, and full text was obtained if both reviewers judged a citation to be potentially eligible. Standardized screening and data extraction forms were created prior to data collection. Extracted data include author, year of publication, the specific type of conjoined twin, gestational age at termination of pregnancy, method of pregnancy termination and adverse maternal outcome or procedure related complications (hemorrhage, blood transfusion, uterine rupture, sepsis or death). Any discrepancies were resolved through discussion with a third reviewer (MDF).

Our initial search identified 512 publications. There were 392 articles after duplicates were removed. Examination of title and abstract led to the exclusion of 264 articles. The remaining 128 articles were assessed for

eligibility by examining the full-text. Of these, 95 were excluded as they did not meet the review inclusion criteria. Thus, our search identified 33 relevant publications with 47 previously reported cases to be eligible. With the addition of the present case we therefore included a total of 48 cases from 34 publications for this review. Figure 3 presents the PRISMA flow diagram illustrating the systematic selection process.

Most authors resort to medical induction of labor resulting in vaginal delivery, 75% of reviewed cases delivered vaginally through medical induction while 18% underwent cesarean section (Table 1).

Successful induction of labor has been reported for thoracopagus conjoined twins at 27 weeks of gestation^{10, 11}. Nevertheless, we identified a few cases of cesarean section performed as early as 20 weeks¹²⁻¹⁴.

None of the papers reviewed report adverse maternal outcomes. However, Mitchell et al reported a case complicated by chorioamnionitis. The patient underwent two sessions of laminaria insertion 24 hours apart and was provided with prophylactic antibiotics. Chorioamnionitis was diagnosed based on high grade fever and tachycardia. She was treated with intravenous antibiotics and was discharged 2 days after successful induction labor⁴.

There are limited data on utilization of surgical abortion for conjoined twins. To our knowledge there is only one report describing successful D&E for conjoined twins after 20 weeks¹⁵. Although D&E offer shorter procedure time and avoid the need for induction or hysterotomy, it is not of course without complications particularly at later gestations. Thus, it should be reserved for specialized centers with experienced providers³.

When performing D&E, adequate cervical preparation is an important intervention to reduce the risk of procedure-related complications including uterine trauma and cervical laceration. This is especially true in advanced gestational age or, as in our case, when difficulty is anticipated^{16, 17}. We achieved adequate cervical preparation with two days preparation, using a combination of medical and mechanical methods.

The routine use of ultrasound during surgical abortion is controversial. However, ultrasound guidance has been shown to increase safety and facilitate completion of the procedure in difficult cases¹⁸. We utilized ultrasound throughout the procedure to localize fetal parts and guide our instruments in the uterus.

Conclusion

Even though this is experience from a single case, D&E can be safely performed for carefully selected cases of conjoined twins beyond 20 weeks' gestations. Adequate cervical preparation, pain control and ultrasound guidance during the procedure are critical for optimal outcomes.

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Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conflict of interest

The authors declare no conflicts of interest.

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Author contribution

FAA, THT, MAS drafted the initial manuscript. FAA and THT did the literature search and analysis. MAS, MDF and SP interpreted the data. WG, DB and SP critically revised the manuscript for important intellectual content. All authors approved the final version for submission and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Data availability statement

The authors confirm that the data supporting the findings of this study are available within the article and its supplementary materials.

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Figure legend

Figure 1. Axial ultrasound image showing two normally shaped fetal heads facing each other.

Figure 2. Sagittal section of the fetal chest and abdomen showing two fetal spines, a single distorted heart

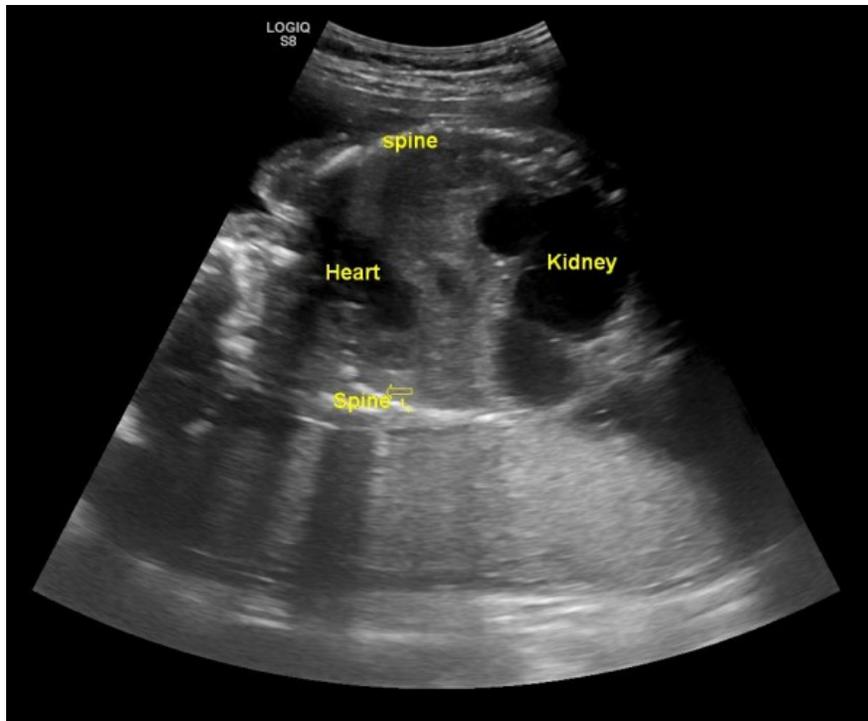
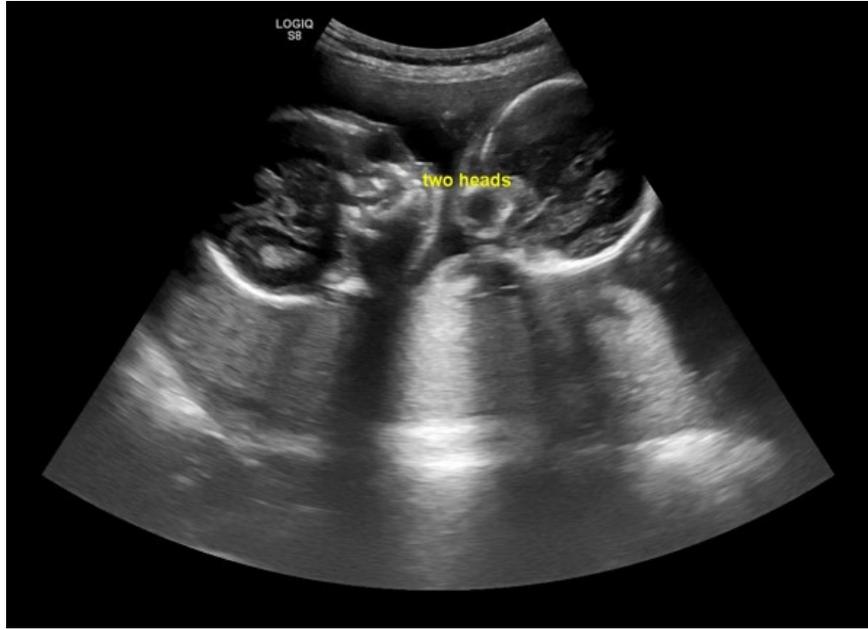
and multi-cystic kidneys.

Figure 3. PRISMA flow diagram study screening and selection

Table 1- Methods of pregnancy termination for second trimester conjoined twins

Author, year	Type of union	Gestational age at termination	Method of termination
Kattel P, 2018 ¹⁹	Parapagus dicephalus	27 weeks 6 days	Cesarean section
Sakala EP, 1986 ¹⁰	Thoracopagus	27 weeks	Vaginal (Pitocin induction)
Chatkupt S, 1993 ²⁰	Dicephalus	21 weeks	Vaginal (Saline induction)
Zoppini C, 1993 ²¹	Omphalopagus	23 weeks	Cesarean section (Classical)
Van den Brand SF, 1994 ²²	Thoracopagus and Omphalopagus	Thoracopagus- 21 weeks and Omphalopagus- 17 weeks	Both vaginal (prostaglandin induction)
Balakumar K, 1995 ¹²	Thoraco-omphalopagus	20 weeks	Cesarean section
Aquino DB, 1997 ²³	Craniopagus parasiticus	20 weeks	Vaginal (prostaglandin induction)
Sen C, 2003 ⁶	Thoraco-omphalopagus	19 weeks	Vaginal (Misoprostol induction)
Esenkaya S, 2004 ²⁴	Dicephalus	17 weeks	Vaginal
Tansel T, 2004 ²⁵	Parapagus (dicephalus tetrabrachius dipus)	22 weeks	Vaginal
Maymon R, 2005 ²⁶	Thoracopagus	16 weeks	Dilation and evacuation
Hassani AA, 2005 ²⁷	Dicephalus dibrachius	16 weeks	Vaginal
Khanna PC, 2005 ²⁸	Cephalothoracopagus janiceps	24 weeks	Vaginal
Özkur A, 2006 ²⁹	Cephalopagus	24 weeks	Vaginal (Misoprostol induction)
Singla V, 2009 ¹¹	Thoracopagus	27 weeks	Vaginal (Misoprostol induction)
Sabih De, 2010 ³⁰	Dicephalus	24 weeks	Cesarean section
Deveer R, 2010 ³¹	Craniothoracopagus	26 weeks	Vaginal (Misoprostol induction)
Mete A, 2010 ³²	Dicephalic parapagus	16 weeks	Vaginal (Misoprostol induction)

Author, year	Type of union	Gestational age at termination	Method of termination
Camuzcuoglu H, 2010 ¹³	Dicephalic parapagus	19 weeks	Cesarean section
Pandey S, 2011 ³³	Thoraco-omphalopagus	15 weeks	Vaginal (Misoprostol induction)
ML Brizot, 2011 ⁹	A total of 13 cases- Thoracopagus-9 Thoracopagus dibrachius tripus-1 Parapagus dibrachius dipus-1 Parapagus dicephalus tribrachius dipus-1 Omphaloischiopagus-1	Gestational age ranging from 18 weeks 3 days to 27 weeks 4 days	One case underwent cesarean section at 27w4d. The other 12 had vaginal delivery.
Pătrașcu A, 2013 ³⁴	Dicephalus dipus dibrachius	21 weeks	Cesarean section
Mitchell T, 2014 ⁴	Thoraco-omphalopagus and Pygopagus	Thoraco-omphalopagus-23 weeks 6 days and Pygopagus-25 weeks 1 day	Both vaginal. Inductions were initiated with Laminaria and augmented with vaginal misoprostol or oxytocin
Wu Y, 2014 ³⁵	Thoracopagus	24 weeks	Vaginal
Vaidya M, 2014 ³⁶	Diprosopus	26 weeks	Vaginal (prostaglandin induction)
Krawczyk J, 2015 ³⁷	Thoraco-omphalopagus	16 weeks	Vaginal
Lu Q, 2016 ³⁸	Thoracopagus	25 weeks	Vaginal (prostaglandin induction)
Biso MP, 2017 ³⁹	Ischiopagus	21 weeks	Vaginal
Ozcan HC, 2017 ¹⁴	Thoracoomphalopagus	17 weeks	Cesarean section
Eris Yalcin S, 2018 ⁴⁰	Cephalopagus	14 weeks	Vaginal
Al Yaqoubi HN, 2019 ⁴¹	Craniopagus parasiticus	17 weeks	Vaginal (Misoprostol induction)
Hern WM, 2019 ¹⁵	Thoracopagus	26 weeks	Dilation and evacuation
Vegar-Zubović S, 2020 ⁴²	Cephalothoracoomphalopagus	21 weeks	Cesarean section
Our report	Thoracopagus	22 weeks	Dilation and evacuation



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Figure_3 PRISMA .docx available at <https://authorea.com/users/352675/articles/476828-successful-dilation-and-evacuation-for-second-trimester-conjoined-twin-a-case-report-and-systematic-review-of-the-literature>