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

Ferid A. Abubeker1*, Tesfaye H. Tufa1, Matiyas Asrat Shiferaw1, Mekdes Daba Feyssa1, Wondimu Gudu1, Delayehu Bekele1 and Sarah Prager1,2

Abstract

Background: Conjoined twins are a rare clinical event occurring in about 1 per 250,000 live births. Though the prognosis of conjoined twins is generally low, 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 (D&E) done for conjoined twins at 22 weeks of gestation.

Case presentation: A 20-year-old primigravid woman was diagnosed with a conjoined, thoraco-omphalopagus twin pregnancy after undergoing a detailed two-dimensional (2D) fetal ultrasound anatomic scanning. Assessment and counseling were done by a multidisciplinary team. The team discussed the prognosis and options of management with the patient. The patient opted for termination of pregnancy. Different options of termination were discussed and the patient consented for D&E, with the possibility of reverting to hysterotomy in case intraoperative difficulty was encountered. A 2-day cervical preparation followed by D&E was done under spinal anesthesia and ultrasound guidance.

Conclusion: In this patient, D&E was done successfully without complications. Adequate cervical preparation, pain control, and ultrasound guidance during the procedure are critical for optimal outcomes. A literature review of methods of pregnancy termination for conjoined twins in the second trimester revealed 75% delivered vaginally through medical induction while 18% underwent cesarean section. Only one other report described successful D&E for conjoined twins after 20 weeks. D&E can be safely performed for carefully selected cases of conjoined twins beyond 20 weeks’ gestations avoiding the need for induction or hysterotomy.

Keywords: Conjoined twins, Dilation and evacuation, Surgical abortion, Second trimester, Case report
Case presentation

A 20-year-old primigravid woman was referred to our hospital at 22 weeks of gestation with a diagnosis of large fetal intra-abdominal cysts identified during a routine ultrasound examination. In our center, detailed fetal two-dimensional (2D) ultrasound anatomic scanning was done, revealing two fetal heads at a fixed position, facing each other (Fig. 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 (Fig. 2). 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 specialists, family planning specialists, and anesthetists. After discussion on prognosis and options of management, the patient opted for termination of pregnancy. Different options of termination were discussed and the patient consented for D&E, with the possibility of reverting to hysterotomy in case intraoperative difficulty was encountered.

We performed a 2-day cervical preparation. On day 1, 200 mg mifepristone was administered orally and five 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 misoprostol sublingually and 200 mg doxycycline orally. 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 thoraco-pagus. The presenting calvarium was decompressed with suction and delivered. Finally, the second twin and placenta were delivered intact. The procedure was completed without complications. Post-procedure tissue count showed two calvaria and spines, four well-formed upper limbs, single thorax and abdomen, and two well-formed and two fused primitive lower limbs. The patient recovered well and was discharged after 24 hours. A follow-up phone call after 2 weeks revealed an 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 worldwide [1]. Recent advances in antenatal imaging techniques, such as three-dimensional (3D) ultrasonography, Doppler studies, and magnetic resonance imaging (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 is 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 because of perceived difficulty in vaginal delivery [10]. Though details of the methods employed were not described, Brizot et al. reported 12 vaginal terminations for second trimester conjoined twins [9]. Similarly, Mitchell et al. reported two successful inductions of late second trimester conjoined twins. However, both patients underwent two sessions of laminaria placement prior to administration of uterotonics [4].

We conducted a systematic search of the electronic databases of MEDLINE, EMBASE, and Google Scholar using MeSH and keywords from the inception of the databases until November 30, 2020 (see Additional file 1). Bibliographies of the relevant articles were reviewed and then cross-searched to identify further relevant studies. We included all publications in English.
that specify the 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 weeks [12–14].

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 on the basis of high-grade fever and tachycardia. She was treated with intravenous antibiotics and was discharged 2 days after successful induction labor [4].

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 weeks [15]. Although D&E offers a shorter procedure time and avoids 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 providers [3].

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 2 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 cases [18]. 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.

Supplementary Information
The online version contains supplementary material available at https://doi.org/10.1186/s13256-021-02815-4.

Additional file 1: Search strategy.
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