Case Report

A Rare Case of Ischiopagus Dicephalus Conjoined Twins with Tetrabrachius Tripus: Prenatal Diagnosis and Management: A Case Report

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Abstract
Objectives: This article reports a rare case of conjoined twins with ischiopagus dicephalus and tetrabrachius tripus, emphasizing the significance of early prenatal imaging and interdisciplinary therapy.
Case Report: Conjoined twins are a rare congenital anomaly resulting from the incomplete division of a single fertilized ovum in monozygotic, monochorionic, monoamniotic pregnancies. Ischiopagus dicephalus is an exceptionally rare subtype characterized by two heads on a single body, with fusion of the lower thorax and pelvis. We report a 28-year-old primigravida who was diagnosed at 18 weeks' gestation with ischiopagus dicephalus tetrabrachius tripus. Transabdominal and 3D ultrasonography revealed a unique anatomical combination: two separate heads and two independent hearts and thoracic cavities, yet fused at the lower thorax and pelvis, with a shared liver and gastrointestinal tract and a tri-podal lower-limb configuration. Following multidisciplinary consultation, the "dual-heart yet single-pelvis" anatomy confirmed non-separability and a poor prognosis, leading to the decision for medical termination. A safe second-trimester vaginal delivery was achieved without maternal morbidity.
Conclusion: The key learning point is that identifying the specific dual-heart, single-pelvis configuration is a definitive prognostic marker for non-separability. This case emphasizes the critical role of early, accurate prenatal imaging in detecting rare variants of conjoined twins, enabling comprehensive multidisciplinary counseling, informed ethical decision-making, and safe maternal management.

Keywords: Conjoined twin, dicephalus, ischiopagus, prenatal diagnosis, ultrasonography

Laporan Kasus Langka Kembar Siam Ischiopagus Dicephalus dengan Tetrabrachius Tripus: Diagnosis Prenatal dan Tatalaksana: Laporan Kasus

Abstrak
Tujuan: Artikel ini melaporkan kasus langka kembar siam dengan tipe *ischiopagus dicephalus* dan *tetrabrachius tripus*, serta menekankan pentingnya pencitraan prenatal dini dan penatalaksanaan multidisiplin.
Laporan Kasus: Kembar siam merupakan kelainan kongenital langka yang terjadi akibat pembelahan tidak sempurna dari satu ovum yang telah dibuahi pada kehamilan monozigot, monokorionik, dan monoamniotik. *Ischiopagus dicephalus* adalah subtipe yang sangat jarang, ditandai dengan dua kepala pada satu tubuh dengan fusi pada toraks bawah dan pelvis. Seorang perempuan primigravida berusia 28 tahun didiagnosis pada usia kehamilan 18 minggu dengan ischiopagus dicephalus tetrabrachius tripus. Ultrasonografi transabdominal dan 3D menunjukkan kombinasi anatomi yang unik: dua kepala terpisah dan dua jantung/rongga dada yang independen, namun menyatu pada toraks bawah dan pelvis dengan fusi hati, saluran pencernaan, serta konfigurasi tiga ekstremitas bawah (tripus). Berdasarkan konsultasi multidisiplin, anatomi "dua jantung namun satu pelvis" mengonfirmasi kondisi janin yang tidak dapat dipisahkan dan prognosis buruk sehingga diputuskan untuk terminasi medis. Persalinan pervaginam trimester kedua yang aman berhasil dilakukan tanpa morbiditas maternal.
Kesimpulan: Poin pembelajaran dari kasus ini adalah bahwa identifikasi konfigurasi spesifik dua jantung dan satu pelvis berfungsi sebagai penanda prognostik definitif untuk ketidakmungkinan pemisahan janin. Kasus ini menegaskan peran krusial pencitraan prenatal yang dini dan akurat dalam mendeteksi varian langka kembar siam sehingga memungkinkan konseling multidisiplin yang komprehensif, pengambilan keputusan etik yang tepat, serta penatalaksanaan maternal yang aman.

Kata kunci: *Dicephalus*; *ischiopagus*; kembar siam; *prenatal diagnosis; ultrasonography*

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Introduction

Conjoined twins are a rare congenital defect resulting from incomplete division of a single fertilized ovum, occurring only in monozygotic, monochorionic, and monoamniotic (MCMA) twin pregnancies.[1] The estimated incidence ranges from one in 50,000 to one in 200,000 live births, with female newborns showing a higher prevalence. The condition is associated with high rates of prenatal morbidity and mortality, which are largely determined by the type and extent of organ sharing.[2]

Ischiopagus is an uncommon form of conjoined twinning characterized by ventral fusion of the pelvis and lower abdomen. The dicephalus subtype features two separate heads on a single torso. The anatomical variety dicephalus tetrabrachius tripus has two heads, four upper limbs, and three lower limbs, indicating partial duplication of cranial and thoracic features with a common lower body. This complex morphology is frequently associated with shared pelvic organs, gastrointestinal tract malformations, and variable degrees of cardiovascular and genitourinary fusion.[3,4]

Ultrasonography and fetal magnetic resonance imaging (MRI) are commonly used to make prenatal diagnoses because they help define anatomical linkages, measure organ sharing, and guide parental counseling.[5,6] Early and accurate diagnosis is critical, as conjoined twins with ischiopagus dicephalus are typically considered non-separable due to substantial shared structures, resulting in a poor prognosis.[6]

We report a rare case of MCMA ischiopagus dicephalus conjoined twins with tetrabrachius tripus, emphasizing the anatomical features, clinical consequences, and prenatal imaging findings. The novel contribution of this article to the existing literature is a detailed, early second-trimester sonographic roadmap (combining two-dimensional and three-dimensional imaging) for diagnosing an extremely rare subset of conjoined twins, along with a safe, multidisciplinary approach for medical termination and vaginal delivery.

Case Report

A 28-year-old primigravida (G1P0A0) was referred to our maternal-fetal medicine center at 18 weeks and 3 days of gestation for further evaluation of a possible conjoined twin pregnancy. She had been receiving routine antenatal care and was initially referred from a secondary hospital after ultrasonographic findings at 17 weeks of gestation suggested conjoined twins.

The patient had no notable medical history. She denied a history of allergies, asthma, diabetes, heart disease, or hypertension. Additionally, the patient reported no history of maternal infections, genetic predispositions, or exposure to environmental teratogens or chemical hazards throughout the pregnancy. There was no personal or familial history of twin pregnancies or congenital abnormalities. Her obstetric history was unremarkable, as this was her first pregnancy. She regularly took calcium, iron, and folic acid supplements during her pregnancy. At presentation, she reported no uterine contractions, abdominal pain, vaginal bleeding, fluid leakage, or abnormal vaginal discharge. Fetal movements had begun to be perceived. Upon assessment, the patient was in good overall condition, with stable vital signs and normal laboratory results. Two fetal heart rates were measured at 149 and 133 beats per minute. The rest of the physical examination was unremarkable.

Transabdominal Ultrasonography revealed a monochorionic monoamniotic twin pregnancy, with two distinct fetal heads positioned side by side on a single shared torso. The fetuses had four upper and three lower limbs, indicating partial duplication of

the lower extremities. The twins were joined in the lower thoracic and pelvic regions, with fusion extending into the upper abdomen. Each fetus had its own thoracic cavity and lungs. Abdominal imaging revealed fusion of several visceral organs, including segments of the gastrointestinal tract and liver, as well as two distinct urinary bladders. The placenta was located posteriorly within a single amniotic sac, with two distinct fetal heartbeats identified (Figure 2).

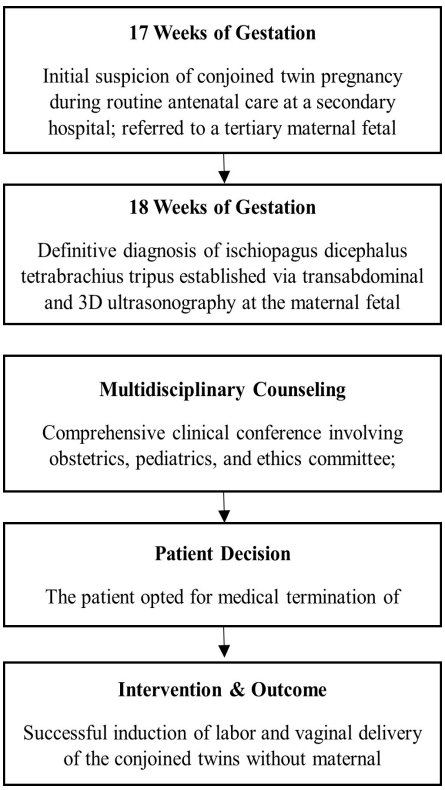

Figure 1 Clinical Management Flowchart

Furthermore, a three-dimensional ultrasound examination confirmed the diagnosis, showing twins with two separate heads conjoined at the lower thoracic and pelvic regions, with fusion extending through the abdomen and liver (Figure 3). These findings are consistent with ischiopagus, dicephalus conjoined twins, and tetrabrachius tripus.

The anatomical uniqueness of this case is defined by its complex dicephalus tetrabrachius tripus configuration. The twins exhibited a single torso with two distinct heads and four upper limbs, each supported by its own thoracic cavity, lungs, and heart, which is a rare finding given the extensive abdominal fusion. The anatomy became singular in the upper abdomen, with a shared liver and gastrointestinal tract, ending in a fused pelvis, two separate urinary bladders, and a unique tripodal (three-limbed) lower body. This specific combination of dual vital thoracic organs with a singular, tripodal lower axis represents a novel anatomical variant in the scant literature on ischiopagus twins.

A multidisciplinary clinical conference involving obstetrics and gynecology, maternal-fetal medicine, pediatric surgery, neonatology, radiology, and the hospital ethics committee was held. Further assessment with fetal magnetic resonance imaging (MRI) was recommended to evaluate abdominal organ involvement and likely resectability, given the complexity of organ sharing and the poor prognosis. However, the MRI was not performed because the 2D and 3D ultrasound findings provided sufficient diagnostic clarity regarding extensive organ sharing and non-resectable anatomy. Given the extremely poor fetal prognosis and the parents' decision for immediate termination, additional imaging for surgical planning was deemed unnecessary. Pregnancy termination was recommended in consultation with a tertiary referral center if the twins were found to be non-resectable and had a poor prognosis.

After counseling regarding the pregnancy condition and the poor fetal prognosis, the patient decided to terminate the pregnancy. Consent was obtained for medical termination of pregnancy (MTP) in the second trimester. The patient was induced for a second-trimester abortion as planned. She was admitted for inpatient care and underwent induction with a 70 cc Foley catheter balloon, combined with vaginal

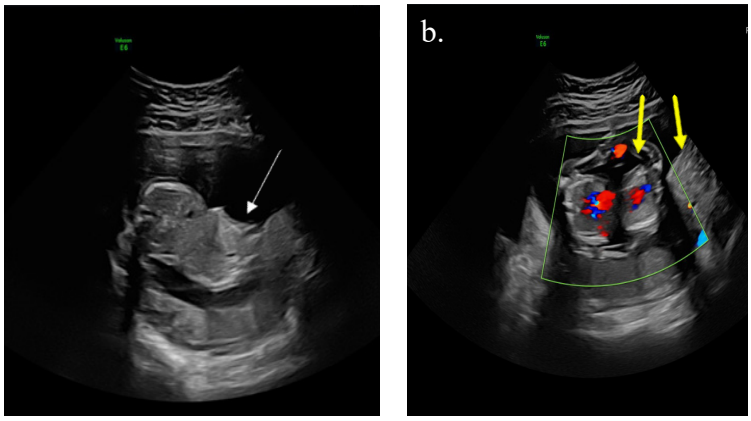

Figure 2 **Two-dimensional ultrasound image. Figure A (white arrow) illustrates fusion of both fetuses in the abdominal region, whereas Figure B (yellow arrow) shows separation in the thoracic region, with each fetus having its own heart**

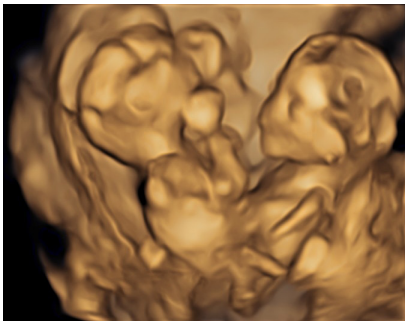

Figure 3 **Three-dimensional ultrasound image of twins showing that the fetuses face each other, each with two upper extremities, and that there is fusion of the umbilical region extending caudally**

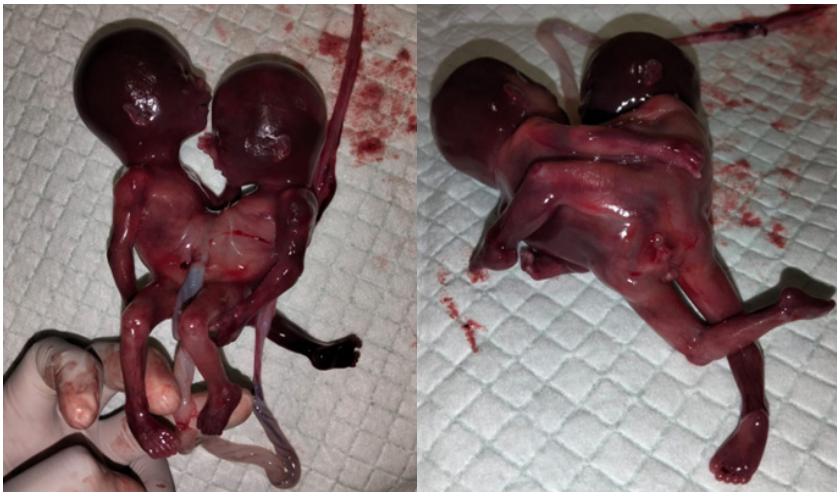

Figure 4 Intact ischiopagus dicephalus conjoined twins and tetrabrachius tripus after abortion.

administration of Misoprostol 200 mcg every 4 hours, based on the clinical rationale of minimizing maternal surgical risk compared with hysterotomy, especially given the early 18-week gestation and the soft, non-ossified nature of the conjoined fetuses. Prophylactic oral Amoxicillin 500 mg was administered every 8 hours to prevent ascending infection during the prolonged induction process involving an intrauterine balloon, which can act as a potential nidus for bacteria. The conjoined twins were delivered vaginally one day after induction, weighing 280 grams at birth, with a single umbilical cord on the fused abdomen extending down to the pelvis, and presenting with four upper limbs and three lower limbs (figure 4).

The patient remained stable throughout induction and delivery. The postpartum course was unremarkable, with normal vital signs and minimal vaginal bleeding. There were no immediate or delayed maternal complications, such as postpartum hemorrhage, infection, or uterine injury. The patient was discharged in good overall condition.

Discussion

Conjoined twins are an uncommon and complex congenital condition caused by the incomplete division of a single fertilized egg during early development. This condition occurs only in monochorionic monoamniotic pregnancies and is linked to severe perinatal morbidity and death, with outcomes largely determined by the type of fusion and the extent of shared organs.[1,3]

Ischiopagus dicephalus conjoined twins are among the rarest types and are typically associated with a very poor prognosis due to extensive fusion of key tissues. Ischiopagus conjoined twins account for roughly 6% of

Table 1 Classification of Conjoined Twin

Types	Definition
Cephalopagus	There are two faces that extend from the top of the head to the umbilicus.
Thoracopagus	They are linked face-to-face from the upper thorax to the upper abdomen and invariably involve the heart.
Omphalopagus	The umbilical region of the lower thorax is frequently fused, although the heart is never involved.
Ischiopagus	The union typically involves the lower abdomen, the fused pelvic bones, the external genitalia, and the anus.
Parapagus	Parapagus conjoined twins are laterally connected and commonly share the pelvis. They come in three varieties: parapagus dithoracic (separated thoraces), parapagus dicephalus (one trunk with two separate heads), and parapagus diprosopus (one trunk, one head, and two faces).
Craniopagus	They are joined by the skull, share meninges but rarely the brain surface, and exclude the face and trunk.
Pygopagus	Dorsally fused, sharing the perineal and sacrococcygeal regions, with a single anus but two rectums.
Rachipagus	The dorsolumbar vertebral column, and in rare cases, the cervical vertebrae and occipital bone, may be affected by a dorsal fusion deformity.
Other symmetrical	Some publications describe conjoined twins differently, and a number of unusual variants of symmetrical conjoined twins are included.
Asymmetric	Parasitic conjoined twin and fetus in fetus

all conjoined twin cases. Ischiopagus twins have umbilical fusion that extends caudally, resulting in a large, fused pelvis. Each twin has a separate spinal cord, and the bodies can be oriented face-to-face or end-to-end, with the vertebral columns aligned in a straight line. Based on pelvic architecture and shared features, ischiopagus twins are classified as tetrapus (four limbs), tripus (three limbs), or bipus (two limbs). The most common form is Ischiopagus tripus, in which the third limb is typically deformed.[7,8,9]

The differential diagnosis for this case included other ventral and lateral fusion types, such as thoracopagus and parapagus. Thoracopagus twins are fused at the thorax and almost always share a single heart, whereas this case featured two distinct hearts. Parapagus twins are laterally fused and share a single trunk, which can mimic ischiopagus; however, the pelvic-to-pelvic fusion and the specific tripodal (tripus) limb arrangement in our case definitively confirmed the ischiopagus dicephalus classification.[10,11]

The exact cause of conjoined twinning remains unclear and is still debated in embryology. Conjoined twins are thought to result from aberrant development during monozygotic twinning in early embryogenesis. Two major theories have been proposed: the fission theory, which holds that a single embryo incompletely separates into two individuals, and the fusion theory, which posits the secondary merger of two previously separate embryonic discs. Conjoined twinning is regarded as a rare developmental aberration caused by disrupted mechanisms in early embryonic organization rather than by a clearly established genetic or environmental factor.[10]

Prenatal Diagnosis

Early prenatal diagnosis is critical for effective management and parental counseling. Ultrasonography remains the primary method for detecting conjoined twins and can be used as early as the first trimester. Although ultrasonography is the primary diagnostic tool, it can be limited by maternal habitus, fetal positioning, and lower soft-tissue contrast compared with MRI. In this case, however, early second-trimester timing and the use of 3D modalities provided sufficient clarity for a definitive diagnosis. Key sonographic findings include the absence of an intertwin membrane, the persistence of close proximity between the fetal bodies, the inability to differentiate fetal shapes, and shared anatomical structures.[6,11]

In the present case, transabdominal ultrasound revealed a monochorionic monoamniotic pregnancy. This was characterized by two distinct fetal heads on a single body, fusion at the lower thorax and pelvis, and shared abdominal organs, including the liver and digestive system. The presence of four upper and three lower limbs confirmed the diagnosis of conjoined twins with ischiopagus dicephalus and tetrabrachius tripus. Importantly, each fetus had a separate thoracic cavity and heart, as well as separate urinary bladders, indicating partial organ duplication.

Three-dimensional ultrasonography played a vital role in confirming the diagnosis and providing a clearer image of the fusion plane, fetal position, and limb configuration. This imaging modality enhances spatial awareness of complex anatomy and improves diagnostic confidence, particularly for rare and unusual types of conjoined twinning.[6,12]

In this case, fetal magnetic resonance imaging (MRI) was recommended during a multidisciplinary discussion to assess the extent of abdominal organ sharing, even though the prognosis was deemed poor based solely on ultrasonographic findings. MRI is advised as a second-trimester adjunct to further delineate shared organs and vascular anatomy, particularly for surgical planning and assessment of potential separability.[5,13]

Prenatal Management

Conjoined twin pregnancies must be managed collaboratively by obstetricians, maternal-fetal medicine specialists, radiologists, pediatric surgeons, neonatologists, and ethics committees. Comprehensive counseling is necessary to educate parents about the anatomical findings, prognosis, potential postnatal therapies, and maternal risks.[4,14]

Ischiopagus dicephalus conjoined twins are often considered inseparable due to substantial fusion of pelvic organs, a shared gastrointestinal system, and intricate vascular connections, making postnatal survival uncertain.[4,5] As a result, termination of pregnancy is frequently suggested as a treatment option, especially when the diagnosis is made in the second trimester, and the prognosis is poor.[7]

In this case, after receiving extensive counseling on the severity of fetal defects and predicted outcomes, the patient chose medical termination of pregnancy. A second-trimester termination was successfully performed using a Foley catheter balloon and misoprostol, a routine obstetric procedure. Vaginal delivery was successful with no maternal complications, demonstrating that early detection allows for safer maternal treatment and avoids obstetric morbidity associated with advanced gestation.

Preconception Counseling

Preconception Counseling for Future Pregnancies. As part of comprehensive maternal care, counseling about future pregnancies was a critical component of the patient's management. Because conjoined twinning is a rare, idiopathic developmental error, the patient was reassured that the recurrence risk in subsequent pregnancies is virtually negligible, equivalent to that of the general population. For future preconception preparation, standard guidelines should be followed, including daily folic acid supplementation and optimization of maternal nutritional status prior to conception. Most importantly, early first-trimester ultrasonography (between 11 and 13 weeks of gestation) is highly recommended for her next pregnancy. Although the recurrence risk is extremely low, early sonographic evaluation is essential to confirm normal embryonic development, rule out structural anomalies, and provide vital psychological reassurance to alleviate parental anxiety following this traumatic loss.[6,7,11]

Clinical Implications

This case has significant clinical implications for the management of complex conjoined twins. Early, precise sonographic mapping of critical organs, such as identifying dual hearts alongside a single fused pelvis, serves as a definitive prognostic marker of non-separability. Recognizing this configuration early in the second trimester is crucial, as it shifts the clinical paradigm from planning complex postnatal separation surgeries to focusing entirely on minimizing maternal morbidity. By opting for a medically induced vaginal delivery at an early gestational age, the severe maternal risks associated with classical hysterotomy, which can compromise future fertility, were successfully avoided.

Limitations

This report has several limitations that should be acknowledged. First, although the multidisciplinary team suggested it, a fetal MRI was not performed. The early second-trimester timing and clear 3D ultrasound findings provided sufficient evidence of non-separability, leading to the immediate parental decision to terminate, thereby rendering further imaging for surgical planning unnecessary. Second, the parents declined a post-mortem autopsy for personal

and cultural reasons. Consequently, detailed histopathological confirmation of internal organ sharing, particularly the exact vascular connections within the fused liver and gastrointestinal tract, could not be obtained beyond the macroscopic evaluation.

Ethical Consideration

Ethical considerations are critical in managing conjoined twin pregnancies. Medical feasibility and fetal prognosis should inform decision-making while respecting parental autonomy. Multidisciplinary case conferences and ethics committee involvement, as seen in this case, are strongly recommended to promote ethically sound, patient-centered care.[3,15,16]

Conclusion

This case underscores the importance of early, precise prenatal detection of rare conjoined twin variants using modern imaging modalities. Detailed anatomical assessment enables informed counseling, appropriate multidisciplinary planning, and tailored management. For ischiopagus dicephalus conjoined twins with substantial organ sharing, prenatal detection facilitates timely decision-making and improved maternal outcomes. Explicitly identifying the unique "dual-heart, single-pelvis" anatomy is a novel contribution to the literature and serves as a definitive prognostic marker for non-separability.

Conflict of interest

All authors confirm that there are no conflicts of interest.

Ethical approval and consent to participate

Written informed consent was obtained, and the patient was voluntary.

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