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CASE REPORT

Ischiopagus Conjoint Twins: A Case Report

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Background: The birth of conjoined twins is an extremely rare neonatal condition, occurring in approximately 1 in 100,000 live births. Ischiopagus, a form of conjoined twinning where the twins are joined at the pelvis, accounts for 6–11% of all conjoined twins. Ischiopagus conjoined twinning is a congenital anomaly. The surgical separation of conjoined twins is challenging due to the intricate anatomy and physiology involved. Thorough preoperative evaluation, meticulous planning, and a skilled surgical team are essential for ensuring a successful outcome. This condition significantly requires separation surgeries due to the unique anatomical challenges it presents. Clinical outcomes can vary depending on the healthcare facility and resources available. The development of conjoined twins consistently draws significant attention from researchers and clinicians. Aside from the rarity of such cases, the manner in which these twins develop remains a topic of ongoing debate. This report describes a case of ischiopagus conjoined twins, characterized by two heads (dicephalus), four upper limbs (tetrabrachius), and a shared pelvis with single umbilical cord. The mother of the twins was referred from a primary hospital Somali Sudanese Specialized Hospital for specialized care during her twin pregnancy, where a cesarean section was performed.

Case Summary: The twins delivered by 34 years old gravida 9 para 7, no previous congenital anomalies, the mother was diabetic and she was on anti-diabetic drugs. Both parents had no family history of birth defects or exposure to known teratogens. Imaging (MRI) revealed that that the twins had separate hearts, lungs, kidneys with severe hydronephrosis in one of them, stomach, liver, spleen, and anal canals but shared the bowel loops, a single placenta with one umbilical vein and two umbilical arteries with single umbilical cord. Additionally, they have uterus-like structure with the absence of external genitalia. Despite multiple associated cardiovascular anomalies, there were no external craniofacial, limb, or brain abnormalities. The twins admitted for the neonatal ICU and observed with normal meconium and urinary passage from same opening (cloaca).

Keywords: conjoint twins, ischiopagus, twins

Introduction

Conjoined twins are among the rarest congenital anomalies, also known as Siamese twins, occurring with an estimated incidence of approximately 1 in 50,000 to 1 in 200,000 births. Female conjoined twins are more common than male twins, with a ratio of 2:1 to 3:1.[1]

Conjoined twins develop from the fertilization of a single ovum by a single sperm, followed by incomplete separation of the embryo during the early stages of embryonic development, typically beginning around the third week of intrauterine life. Alternatively, they can form through the fusion of two previously developed monozygotic twins. The classification of conjoined twins is based on the site of fusion, with the most common types being thoracopagus (chest fusion), omphalopagus (abdomen fusion), thoraco-omphalopagus, ischiopagus, and parapagus. In addition to the fusion sites, conjoined twins may have varying limb configurations, such as dicephalus (two heads), tetrabrachius (four upper limbs), and bipus (two lower limbs), which represent some of the rarest forms of conjoined twins.[2]

Various factors, such as alcohol consumption, drug use, radiation or chemical exposure, lack of folic acid supplementation during pregnancy, and family history, are known risk factors for congenital anomalies. However, the exact cause of conjoined twinning remains unclear. Theories often cited in the literature include the theory of fusion and the theory of fission to explain the embryological basis of conjoined twinning.

The fusion theory suggests that conjoined twins arise when two identical twins merge at a specific site during development. The fission theory, on the other hand, posits that conjoined twins are the result of an incomplete division of the embryo. The timing

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of this division significantly affects the extent of shared embryonic structures and fetal membranes, including whether the twins share a single amnion, chorion, or placenta. If the separation occurs at the bilaminar embryonic disc stage, the twins may share a placenta and common chorionic and amniotic membranes. If the division happens later, after the formation of the germ layers (around the third to fourth week of development), the twins may share specific organs.[2]

Ischiopagus conjoined twinning representing only 6% of all conjoined twins is a congenital defect that presents significant challenges in surgical separation due to the complex anatomy and physiology involved along with associated malformations, a thorough clinical assessment, supported by detailed radiological imaging, as well as careful planning and collaboration among medical teams, are essential for the successful separation of conjoined twins.[1]

Ischiopagus twins are fused at the pelvis, sharing lower gastrointestinal systems, genitalia, and urinary tracts, often with a duplication of the genitourinary system. Ischiopagus twins can further be classified by the number of legs they share—tetrapus (four legs), tripus (three legs), or bipus (two legs)—with tetrapus being the most common type. The first successful separation of ischiopagus tetrapus twins was reported in 1966 by Eades and Thomas. Since then, only a few cases have been documented globally.

A careful preoperative assessment, thorough planning, and a skilled surgical team are essential for achieving a successful outcome.[3]

Case Report

The mother of the twins was referred from a primary hospital to the tertiary hospital for specialized care during her twin pregnancy after uneventful course of pregnancy an elective cesarean section was performed. Figure 1

The twins were delivered by a 35-year-old woman, gravida 9 para 7, with no history of previous congenital anomalies. Apgar score after delivery was 9.

Figure 1 Ischiopagus conjoint twins with single umbilical cord.

Figure 2 MRI of the conjoint twins.

The mother had diabetes and was on anti-diabetic medication metformin for a couple of years. Both parents have no family history of birth defects or exposure to known teratogens. Imaging (MRI) Figure 2 revealed that the twins had separate hearts, lungs, kidneys with severe hydronephrosis in one of them, stomach, liver, spleen, and anal canals, but shared the bowel loops, a single placenta with one umbilical vein and two umbilical arteries. Additionally, they have uterus-like structure, with the absence of external genitalia.

Despite multiple associated cardiovascular anomalies, there were no external craniofacial, limb, or brain abnormalities perinatal course. The twins were admitted to the neonatal intensive care unit (NICU) and observed to have normal meconium and urinary passage from a shared opening (cloaca), and treated with early onset of neonatal sepsis.

The patient discharged with good medical condition and referred to high specialized centre in Saudi Arabia for surgical separation and further assessment.

Discussion

Conjoined twinning is a rare developmental anomaly with an uncertain etiology. Some researchers attribute it to incomplete division of the embryonic disc, while others suggest it results from the secondary fusion of two originally separate mono zygotic embryonic discs.[2]

Conjoined twins (CT's) originate from a single fertilized ovum and are always monochorionic (sharing a placenta) and mono-amniotic (sharing an amniotic sac). They occur in approximately 1 in 50,000 to 200,000 deliveries. The term "Siamese twins" was first used to describe the famous pair Chang and Eng Bunker, who lived until the age of 63 and passed away in North Carolina in 1874.

Studies suggest that 70% to 95% of CTs are female.[4] though one study reported an almost equal male-to-female ratio. Spencer classified CTs based on their site of union, distinguishing between ventral (anterior) types—cephalopagus, thoracopagus, omphalopagus, ischiopagus, and parapagus—and dorsal (posterior) types—craniopagus, pyopagus, and rachipagus. Potter and Craig provided a simpler classification based on the most common forms of twinning.[5]

Ischiopagus conjoined twinning, one of the most complex forms, accounts for only 6% of cases. These twins are typically aligned along a longitudinal axis with their heads facing opposite directions. They share a common umbilicus, with fusion occurring below this level, affecting the lower abdomen and pelvis. Approximately 50% of ischiopagus cases

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have four separate lower limbs, one-third have three limbs (two separate and one fused), and one-fifth involve a parasitic twin [1–4[2]. Our case falls into the category of ischiopagus tetrapus (Figure 1).

The magnetic resonance imaging (MRI) is preferable to computed tomography (CT) during the antenatal period due to the absence of radiation exposure. Advances in imaging technology continue to improve diagnostic capabilities. The ultrasonographic (USG) criteria for diagnosing CT's include:

1. Absence of a separating amniotic membrane
2. Inseparable fetal bodies
3. Lack of positional changes in the fetal bodies and heads across repeated examinations
4. Alignment of heads at the same level and body plane
5. Unusual proximity or hyperextension of spines
6. Close positioning of limbs
7. A bifid appearance of the fetal pole in the first trimester
8. Presence of more than three umbilical vessels

Polyhydramnios is observed in 50% to 76% of cases. Diagnosing CTs is challenging and requires the expertise of an experienced perinatologist. Monochorionic-monoamniotic CTs, in particular, Early USG examination is crucial for detection. Three-dimensional Doppler USG is an effective tool for providing families with a clearer understanding of the condition, helping guide decisions regarding pregnancy continuation or termination.

Ischiopagus twins typically share internal organs, including the liver, lower gastrointestinal tract (GIT), and genitourinary structures. The most common GIT fusion occurs at the terminal ileum (near Meckel's diverticulum), with a shared bowel distally. In rare cases, only the colon is shared. The rectum and anus may be single or imperforate. Emergency surgery may be necessary in cases of obstructed GI or genitourinary tracts, ruptured omphalocele, severe respiratory distress, or cardiac failure in one of the twins.

In this case, the twins had two perineal openings through which urine was expelled, but a single urinary bladder.

Diagnostic evaluation includes skeletal surveys, ultrasonography, contrast imaging of the gastrointestinal and urinary systems, endoscopy, computed tomography, and MRI. Additional investigations such as angiography and radioisotope liver and renal scans can provide further anatomical details. A complete cardiac assessment is essential, as congenital heart anomalies are common in all variants of conjoined twins.

A comprehensive diagnostic workup was performed to delineate external and internal structures and assess associated anomalies in preparation for potential surgical separation. However, despite lack of medical facilities the twin referred to high specialized centre in Saudi Arabia.[4]

The prognosis of conjoined twins largely depends on the structures they share and the site of their union. This outcome was likely influenced by the presence of associated cardiac anomalies in both twins. While some cases of conjoined twins have survived either after surgical separation or while remaining joined, most are stillborn or succumb in the early neonatal period.[6]

In conclusion, this case represents one of the rare forms of conjoined twinning, possibly the first reported case in Somalia, featuring twins with two heads, four upper limbs, and two lower limbs. Early diagnosis through antenatal care is crucial in identifying conjoined twins, allowing families to make informed decisions regarding pregnancy management, including the option of early termination. In this case antenatal scan did not detect the anomaly. Additionally, comprehensive antenatal care—including folic acid supplementation—should be made accessible to all pregnant mothers to help reduce the risk of congenital anomalies.[6] The case referred high specialized centre in Saudi Arabia due to lack of facilities and resource limitation in Somalia.

Conclusion

In conclusion, Early diagnosis through antenatal care is crucial for identifying conjoined twins and allowing families to make informed decisions, including the option of early pregnancy termination in cases of severe anomalies. Additionally, comprehensive antenatal care, including folic acid supplementation, should be provided to all pregnant mothers to help reduce the risk of congenital abnormalities.

The prognosis of conjoined twins depends on the extent of organ sharing and the presence of additional malformations. Overall, survival rates remain low, as many CTs have severe cardiac, pulmonary, abdominal, or neurological anomalies, even in structures that are not directly shared.

Data Sharing Statement

Available upon request from the correspondence author.

Ethics Approval

Ethics approval is not applicable and institutional approval was not required for the publication of this case.

Consent for Publication

Written informed consent was obtained from the patient's next-of-kin for publication of this case report and any accompanying images.

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors declare that they have no competing interests in this work.

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