

Challenges of Thoraco-Omphalopagus Conjoined Twins in Resource Limited Settings: A Case Report

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Case Report

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Abstract

Background

Conjoined twins are a rare and challenging congenital malformations resulting from incomplete separation of a monochorionic monoamniotic zygote, leading to fusion and sharing of various body parts. They may be symmetric or asymmetric and occur sporadically with uncertain etiology. Thoraco-omphalopagus conjoined twin is the most common type of conjoined twinning, in which the twins are fused at the lower thorax and upper abdomen.

Case presentation

: Here, we reported a case of 21days old female thoraco-omphalopagus conjoined twins with associated omphalocele who shared a pericardium and had a shared dilated bowel at the gastro-duodenal region, a fused liver at the midline, and a hepatic vein draining from the liver of one twin into the contralateral inferior vena cava. The diagnosis was made at intrapartum and both neonates were having sepsis taking empirical antibiotics and oxygen in the neonatal intensive care unit.

Conclusions

Though these malformations impose a substantial financial and emotional burden on parents in resource-limited settings, where surgical intervention is often nearly impossible; advances in neonatal and surgical care have made successful separation feasible in selected cases, with favorable outcomes.

Background

Conjoined twins (CT) are a rare, sporadic, and complex embryological malformation resulting from incomplete division of monochorionic monoamniotic zygote at 13–15 days fertilization with uncertain etiology. The prevalence of CTs is varies and is estimated to be between 1 in 50,000 and 1 in 100,000 births. They are characterized by physical fusion of the twins, involving shared organs or body structures(1–4). Although more live-born are females, with a female-to-male ratio of approximately 3:1, this condition appears to occur more frequently in male fetuses, as evidenced by its higher rates among male stillbirths(5).

Conjoined twinning is thought to result from incomplete cleavage or axial duplication occurring after 13th days of fertilization and classified into different types according to the site of fusion. The most common forms are thoraco-omphalopagus, thoracopagus, omphalopagus, heteropagus and craniopagus(1).

The diagnosis of CTs can be established as early as the first trimester with ultrasonography or magnetic resonance image (MRI), which can assist parents in decision-making and enable Physicians to anticipate

and prepare for potential complications. However, in resource-limited settings, the diagnosis may be delayed until the time of delivery because of inadequate access to proper maternal care(3, 6, 7).

The overall management of CTs is challenging and requires an individualized multidisciplinary approach, including adequate prenatal care, well-planned delivery, intensive care in early neonatal life, advanced surgical intervention, and consideration of ethical issues(5). Despite comprehensive multidisciplinary management, the overall outcome of all CTs remains poor(8). Separation of CTs poses significant challenge to surgeons, including determining the optimal timing of surgery, making decisions in situations where the life of one co- twin may need to be sacrificed to save the other with a better prognosis, and obtaining detailed imaging to accurately delineate anatomy before surgery(9).

Case Presentation

A 21-day-old pair of conjoined twins born to a para-two 32-year-old mother from Dilla, Ethiopia, presented to our hospital. The mother did not recall the date of her last normal menstrual period but reported having been amenorrheic for the preceding 9 months. Delivery was by cesarean section for cephalopelvic disproportion (CPD), resulting in the birth of live conjoined twins with a combined birth weight of 5,400 gram. The mother had attended antenatal care at our hospital and had been informed that she was carrying a twin pregnancy.

After delivery, the twins were transferred to the neonatal intensive care unit (NICU) with a diagnosis of conjoined twins and respiratory distress. At presentation, both infants were in respiratory distress. On physical examination, their pulse rates ranged from 130 to 148 beats/min, respiratory rates from 64 to 75 breaths/min, and they exhibited nasal flaring and chest retractions. Heart sounds were normal. The twins were symmetrically fused below the nipple line and at the upper abdomen, with an associated omphalocele and a single umbilical cord (Fig. 1). Both infants were female, and neonatal reflexes were normal.

With the initial diagnosis of *Term + NBW + AGA + EONS + thoraco-omphalopagus conjoined twins + omphalocele*; complete blood count, echocardiography, and abdominal ultrasonography were requested. Both neonates received intranasal oxygen, broad-spectrum antibiotics (initially ampicillin and gentamicin), and nasogastric feeding, and their clinical condition was closely monitored by the NICU team. After 48hurs, as their clinical condition was the same, antibiotics was changed to ceftriaxone and vancomycin that continued for about 10days; gradually their clinical condition showed good improvement.

Echocardiography revealed a shared pericardium with two separate structurally normal hearts and major vessels. Abdominal ultrasonography demonstrated fusion of the anterior abdominal walls with extension of bowel loops from one neonate to the other through the junctional area. In the subsequent days, chest and abdominal computed tomography (CT) scans were performed to further delineate the extent of the fusion (see description and Fig. 2 below).

During subsequent days of follow-up in the NICU, both neonates showed good clinical progress. Referral for surgical separation abroad was planned because of the limited experience in Ethiopia.

Contrast-enhanced chest and abdominal computed tomography (CT) was performed and revealed the following findings: *The twins were fused face-to-face across the midline at the lower thorax and upper abdomen, with separate sterna, a fused diaphragm, and a fused liver. A hepatic vein drained from the liver of one twin into the contralateral inferior vena cava (IVC). They shared a dilated bowel loop at the gastro-duodenal region and a common pericardium, while the hearts were separate and abutted centrally. Otherwise, each twin had a separate pancreas, a pair of kidneys, suprarenal glands, lungs, and lower gastrointestinal and genitourinary tracts. The IVC, pulmonary artery, aorta, and their major branches were separate in each co-twin.*

The overall radiologic impression was thoraco-omphalopagus conjoined twins with a fused liver, shared bowel loops, a hepatic vein draining from one twin into the contralateral IVC, and a shared pericardium (Fig. 2).

Discussions

Conjoined twins are a rare monozygotic twin who are physically fused in utero and remained joined at birth because of incomplete division of single fertilized ovum, resulting in shared body organs or anatomical structures with uncertain etiology, and the prevalence has been estimated to be 1 in 50,000 to 1 in 100,000 births. They are thought to result from late cleavage (≥ 13 days after fertilization) of a single zygote following fertilization(1, 2, 8).

Conjoined twins are classified according to the site of fusion, including thoraco-omphalopagus (the most common type), thoracopagus, omphalopagus, heteropagus (parasitic), craniopagus, pyopagus, rachipagus (involving sacrum and perineum), ischiopagus (involving lower abdomen and pelvis), cephalopagus (extending from the head to the umbilicus) (1).

In our reported cases, the neonates were fused at lower thorax, sharing the only pericardium, and at the upper abdomen, with a shared dilated bowel loop at gastro-duodenal region, a fused liver at midline, and a hepatic vein draining from one twin into the contralateral inferior vena cava (IVC).

Cases of CTs are usually diagnosed during the early stages of pregnancy using transabdominal sonography. A case of thoracopagus CTs with shared single heart, reported from China, was diagnosed at 8 weeks of gestation(7). Another case demonstrated that omphalopagus CTs were diagnosed at 17 weeks of gestation(9). Similarly, a case of thoraco-omphalopagus CTs were diagnosed at 19 weeks of gestation, in which the twins shared the sternum, fused diaphragm, and upper abdominal wall(8).

Failure to diagnose CTs during pregnancy may lead to serious complications for the mother and neonates and can cause significant emotional and financial distress to the family. In our case, the condition was initially diagnosed as a normal twin pregnancy during antenatal care follow-up and CTs

were identified intrapartum, resulting in cephalopelvic disproportion (CPD) and necessitating an emergency cesarean section.

Similarly, a case of ischiopagus and dincephalic parapagus CTs reported in Ethiopians settings were diagnosed intrapartum after being initially diagnosed as normal twin pregnancies. Unfortunately, all neonates died at 36hrs and 30minutes of age, respectively(4, 10). Another case reported by Sisay Wodeyes described a mother who had not undergone obstetric ultrasonography during antenatal care. She presented in labor and was evaluated ultrasonography, which suggested a twin pregnancy, leading to continuation of labor. The first twin was delivered but was found to be attached to the second twin, which remained in the uterus. An emergency laparotomy was subsequently performed to deliver the second twin in abdominal-vaginal approach. Both neonates developed birth asphyxia and unfortunately died at 1hour and 20 hours after delivery, respectively(11). These cases highlight the importance of comprehensive antenatal care, including careful obstetric ultrasonographic evaluation of twin pregnancies by experienced clinicians to exclude the possibility of conjoined twins.

In addition to the shared body organs or structures, CTs may present with associated congenital anomalies involving the cardiovascular, gastrointestinal, genitourinary, central nerve system, and musculoskeletal systems. In our case, the twins had an associated omphalocele. A case of spinal bifida associated with CTs was reported by Solomon et al. (12), while Kakembo et al. (13) described a case of dextrocardia with CTs. Alansari et al.(14) reported CTs with a common heart and atrioventricular cushion defect, and Safira et al. (6) described transposition of great arteries in one of the twins. Alene & Abebe also reported CTs with a common imperforate anus and a single penis with sub-coronal hypospadias(4), whereas Amuabunas et al. described a case in which co-twin-I had dextrocardia(5).

Surgical separation of CTs remains an extraordinary challenge, particularly in resource-limited settings(15). To date, there are no published reports of successful surgical separation of complex CTs in Ethiopian settings. However, a case of non- complex parasitic CTs associated with spinal bifida was successfully managed surgically(12), and another parasitic CTs was treated successfully in Uganda(13).

A report from west African countries described local surgical separation of nine sets of CTs, resulting in the survival of fourteen infants, while three died due to postoperative infections(15). These experiences suggest that establishing local surgical facilities equipped with multi- disciplinary teams capable of managing complex neonatal surgical conditions could lead to favorable outcomes and reduce the financial and emotional burden on families. Parents of our patients are scared with the diagnosis of their children, treatment outcome and financial constraints related with separation procedure.

Conclusions and Recommendations

Although the occurrence of conjoined twins (CTs) is sporadic, maintaining a high index of suspicion is essential to avoid missed diagnoses, particularly in twin pregnancies in resource-limited settings. Early and meticulous antenatal ultrasonographic evaluation is crucial for timely diagnosis, appropriate counseling of parents, and planning of delivery at specialized centers.

The outcome of surgical separation is influenced by the type and extent of organ sharing, the site of fusion, and the availability of comprehensive care provided by a multidisciplinary team, including obstetricians, neonatologists, pediatric surgeons, anesthesiologists, radiologists, and intensive care specialists. Advances in neonatal and surgical care have made successful separation feasible in selected cases, with favorable outcomes.

Strengthening prenatal diagnostic services, improving access to obstetric ultrasonography, and expanding multidisciplinary neonatal surgical services in resource-limited settings are imperative to improve survival, optimize clinical outcomes, and reduce the emotional and financial burden experienced by affected families.

Abbreviations

Appropriate for Gestational Age (AGA)

Computed Tomography (CT) scan

Conjoined twins (CTs)

Cephalo

Pelvic Disproportion (CPD)

Early Onset Neonatal Sepsis (EONS)

Inferior Vena Cava (IVC)

Magnetic Resonance Imaging (MRI)

Normal Birth Weight (NBW)

Neonatal Intensive Care (NICU)

Declarations

Ethics and consent to participate: Ethics approval is not applicable. Written informed consent to participate was obtained from the patient's parent.

Consent to publish: informed written consent was obtained from babies' father for publication of this case report and accompanying images. A copy of the written consent is available for journal review if requested.

Clinical trial number: not applicable

Availability of data and materials: All relevant data supporting the findings of this case report are included within the article. Additional information is available from the corresponding author upon reasonable request.

Competing interest: the author declares that there are no competing interests.

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Author's contribution: HM conceived the idea for this case report, collected and interpreted the clinical data, obtained written informed consent for publication from the patient's parents, and drafted and revised the manuscript. The author read and approved the final manuscript.

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Figures



Figure 1

A case of Thoraco- omphalopagus conjoined twins with omphalocele

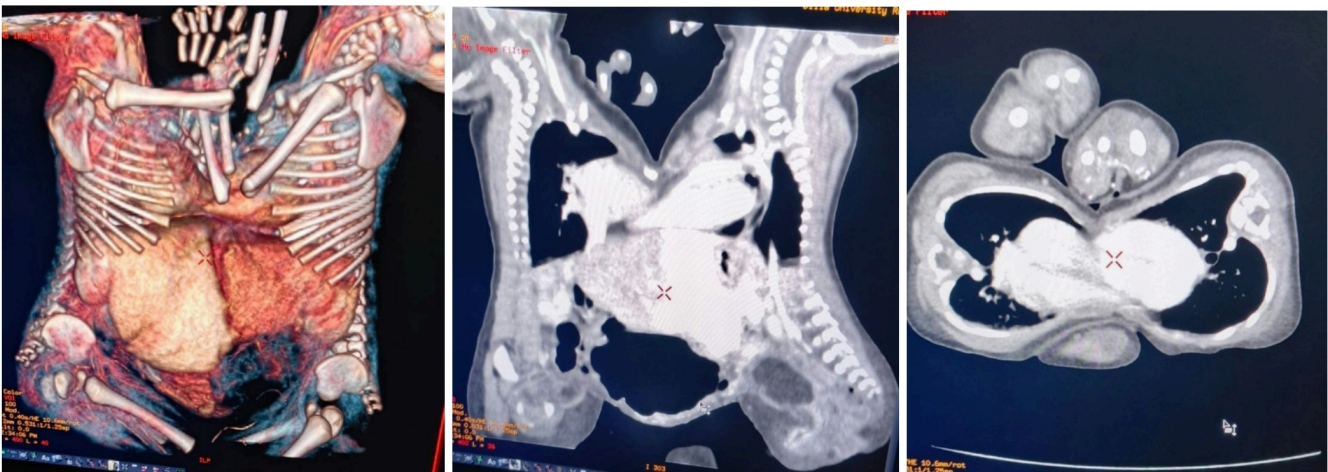


Figure 2

chest and abdominal CT scan of thoraco-omphalopagus conjoined twins

Supplementary Files

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