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Case Report

Prenatal Diagnosis of a Rare Dicephalic Parapagus Twin at 21 Weeks: A Case Report

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Abstract

Background: Dicephalus parapagus twinning (DPT) is the rarest conjoined twinning entity of all conjoined twins. The presence of DPT is always associated with mortality before or after delivery. Antenatal diagnosis is crucial for planning labor, assembling a team, and providing counseling. ***Case presentation***: A 23-year-old patient (G2P1A0) at 21 weeks of pregnancy attended the ultrasound clinic for routine care. A monoamniotic-monochorionic pregnancy was observed; the twin fetuses had two heads but one body, which corresponded to dicephalus parapagus twinning. A detailed ultrasound and fetal echocardiography reveal one heart without congenital anomalies. The liver, kidneys, and bladder appeared to be normally developed. Fetal MRI and karyotyping were not carried out after counseling the parents regarding the poor outcome. A multidisciplinary team followed the case; the patient chose to continue the pregnancy. A planned elective cesarean delivery was done after 36 weeks in a tertiary medical facility. A 3,200 g newborn baby boy was delivered alive; however, he quickly developed respiratory insufficiency and failure of heart function. He deceased within 4 hours from birth; an autopsy was declined. ***Conclusion***: The prenatal diagnosis of DPT helps manage the case thoroughly and carefully to avoid maternal complications. All cases of DPT have very poor prognosis. Prenatal confirmation permits structured, proactive management rather than emergent, multidisciplinary responses.

Keywords: Dicephalic parapagus twin; MRI; Perinatal diagnosis; Ultrasound.

Key Message

Dicephalus parapagus twinning carries an almost universally poor prognosis. Early prenatal diagnosis is essential for counseling and management planning. Multidisciplinary care is critical to ensure optimal maternal outcomes.

تشخيص ما قبل الولادة لتوأم نادر ثنائي الرأس في الأسبوع 21 من الحمل: تقرير حالة

الخلاصة

الخلفية: توائم ديسيفالوس باراباغوس (DPT) هي أندر حالات توأم ملتصق بين جميع التوائم الملتصقة. يرتبط وجود DPT دائما بالوفيات قبل أو بعد الولادة. التشخيص قبل الولادة ضروري لتخطيط الولادة، وتجميع فريق، وتقديم الإرشاد. **عرض الحالة**: حضرت مريضة تبلغ من العمر 23 عاما (G2P1A0) في الأسبوع الحادي والعشرين من الحمل عيادة الموجات فوق الصوتية للرعاية الروتينية. لوحظ حمل أحادي السلى-أحادي المشيمة؛ كان للجنين التوأم رأسان لكن جسد واحد، مما يتوافق مع التوأم ثنائي الشكل الباراباغوس. كشفت الموجات الصوتية التفصيلية وصوت القلب الجيني عن قلب واحد بدون تشوهات خلقية. بدت الكبد والكلى والمثانة متطورة بشكل طبيعي. لم يجر فحص الرنين المغناطيسي للجنين والتصوير الكاريوتايبلازمي بعد استشارة الوالدين بشأن النتيجة السيئة. تابع فريق متعدد التخصصات الحالة؛ اختارت المريضة الاستمرار في الحمل. تم إجراء ولادة قيصرية اختيارية مخطط لها في الأسبوع 36 في منشأة طبية ثالثية. تم توليد طفل رضيع حديث الولادة يزن 3,200 جرام على قيد الحياة؛ ومع ذلك، ظهرت بسرعة صعوبات في الجهاز التنفسي وفشل في وظائف القلب. توفي خلال 4 ساعات من ولادته؛ تم رفض إجراء تشريح الجثة. **الخلاصة**: يساعد التشخيص قبل الولادة لعلاج DPT في إدارة الحالة بشكل شامل وحذر لتجنب المضاعفات للأم. جميع حالات DPT لها توقعات سيئة جدا. يسمح التأكيد قبل الولادة بإدارة منظمة واستباقية بدلا من الاستجابات الناشئة متعددة التخصصات.

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INTRODUCTION

Conjoined twinning is a rare condition caused by the abnormal division of an embryo into two. The rarest version is called “dicephalus parapagus," meaning two heads but one body joined to the side. Dicephalus parapagus (DPT) accounts for approximately 11–13% of all conjoined twins, with a frequency of conjoined twinning estimated to be 1 in 50,000 to 100,000 births, with males predominating [1]. DPT is classified according to the number of arms and legs: dibrachius, tribrachius, or tetrabrachius for the upper limbs; dipus or tripus for the lower limbs. This classification directly influences prognosis and potential surgical treatment decisions [2]. The prognosis of DPT depends primarily on the state of the cardiovascular system and brain. The survival of such twins past the first days of life is unusual, especially in cases sharing only one heart. Separation surgeries are almost impossible to conduct due to fused neural structures [3]. In prenatal diagnostics, once the diagnosis is suspected, advanced tests such as targeted ultrasonography (US), fetal echocardiography, and fetal MRI become necessary to provide a detailed evaluation of the internal structures. This is fundamental to having a realistic prognosis, planning of care, and decision-making for delivery [4]. Due to the scarcity of documented cases, every case helps in developing better strategies. Herein, we report a prenatally diagnosed case of DPT twins at 21 weeks who deceased within four hours after delivery.

Case Presentation

A 23-year-old woman, G2P1A0, with a previous Cesarean section attended an obstetric US at 21 weeks of gestation for routine antenatal follow-up. She denied any history of a relevant personal or familial background. There was no consanguinity between the parents, no drug abuse, no substance exposure, and she did not receive assistance from fertilization techniques. Pregnancy was conceived naturally and has been completely healthy until now. Ultrasound results: The ultrasound (US) depicted a monochorionic monoamniotic twin pregnancy with extremely abnormal head formation characteristic of dicephalus parapagus (Figure 1A).

Figure 1: (**A**) 2-dimensional US image shows the fetus's head and fusion line (yellow arrows); (**B**) Parapagus dicephalic conjoined twins in the anterior view that shows well-differentiated two heads that are fused at the checks (yellow arrow), a single neck and trunk, and normal-looking limbs; (**C**) Close anterior view of the fetus's head showing the fusion line.

Two discrete heads were visualized with independent brains, cerebellum, and thalamus lying next to each other while the neck and body were shared amongst both. In the chest and abdominal cavity, a single heart without any duplication was present. A single heart without structural defect was confirmed on echocardiography. The liver, kidneys, and bladder appeared grossly normal. Twin classification based on their limb anatomy was dicephalus parapagus. Diprosopus was excluded by the presence of two independent cranial vaults and brain structures; craniopagus was ruled out by the absence of two separate bodies below the cranium. There was no increase in amniotic fluid index and no placental abnormalities. The patient was advised to have an MRI and karyotyping, but it was declined after she knew the poor outcome. Multi-disciplinary counseling was performed by a team comprising the following: Obstetricians, neonatologists, pediatric surgeons, and perinatal psychology experts. The anomaly was discussed with the patient along with the inevitable death of the newborn in this condition, the impossibility of separation, and the plan for palliative care measures postpartum. The patient chose to continue the pregnancy. Serial US on alternate weeks was conducted to rule out polyhydramnios or preterm labor. Elective delivery by caesarean section was scheduled at 36 weeks in a tertiary hospital facility. The surgery was uneventful; a live male newborn weighing 3,200 grams was delivered (Figures 1B and C). Self-respiration was initially observed but rapidly progressed to deteriorating respiration, with infant demise 4 hours after birth. As per patient request, the autopsy was declined. The patient was offered psychological support post-bereavement by perinatal psychology experts. 3 years later the patient conceived spontaneously and delivered a normal, healthy female, now aged 2 years.

DISCUSSION

The main focus after the diagnosis of DPT would be differentiating dicephalus parapagus from diprosopus and craniopagus twinning (Table 1) [5].

Table 1: Differential diagnosis of cephalic conjoined anomalies showing the e embryologic, morphologic, and prenatal imaging criteria

Condition	Embryologic/Morphologic definition	Cranial–Neural configuration	Body configuration	Key prenatal imaging criteria
Dicephalus parapagus	Lateral union of monozygotic twins it results from incomplete division of the embryonic disc. The result is two heads on a single trunk.	Two distinct cranial vaults and independent intracranial structures (separate cerebra, cerebella, and thalami).	Single trunk with inconstant duplicated thorax, abdomen, and limbs	US/MRI establish two fully formed cranial compartments that shar a common body axis.
Diprosopus (craniofacial duplication)	Craniofacial duplication It is caused by abnormal signaling during craniofacial patterning rather than a true embryonic twinning.	Single cranial vault with single brain. The duplication involves facial structure only.	Single trunk & axial skeleton.	Imaging reveals one skull and brain in spite of duplicated facial components (i.e., orbits, nose, or mouth).
Craniopagus twins	Symmetrical conjoined twinning Characterized by fusion restricted to the cranial vault.	Two distinct brains and skulls merged at the calvarium with possible shared venous sinuses.	Two separate complete bodies below the cranium level .	Prenatal imaging displays two fetal bodies with cranial fusion only, without shared trunks.

This case highlights the major challenges associated with the prenatal diagnosis and management of DPT, an exceptionally rare disorder. DPT is a form of parapagus conjoined twinning characterized by the lateral fusion of twins that produces two heads sharing one body [6]. The degree of facial union is variable, but in the present case, the fusion was limited to the upper face, with two distinct craniums and separate central nervous systems. The use of obstetric US is the mainstay investigation in diagnosing DVT. It may even be done as early as the end of the first trimester of pregnancy. However, the accurate delineation of what organs are shared or duplicated, such as the heart, liver, and genitourinary system, necessitates further investigation via second-level US together with fetal MRI imaging [7]. MRI provides enhanced imaging of soft tissue structures and precise identification of

neurovascular anatomy, thus playing an important role in prognosis and treatment planning [8]. Lastly, fetal echocardiography is vital in determining the state of the heart as either one or two hearts; a summary of reported cases in the last decade was presented in Table 2. The obstetric management of DPT requires individualized planning (Figure 2).

Table 2: A summary of published case reports in the last decade showing diagnostic investigations , the twin morphology , and autopsy notes

Author (year)	Diagnostic Investigation	Twin Morphologic Features	Autopsy/Special Notes
Harma *et al*. (2005) [9]	Prenatal ultrasound (US)demonstrating absent inter-twin membrane and inseparable bodies; adjunct imaging: 3D US, MRI, echocardiography	Dicephalic parapagus, two heads, two vertebral columns, fused at pelvis; suspected common thoraco-abdominal organ.	Autopsy not done. Notes: cardiac and abdominal anomalies.
Camuzcuoglu *et al*. (2010) [10]	Prenatal US and MRI identifying shared cardiovascular system and internal organs.	Two heads, & common trunk Two upper limbs and three lower limbs (one rudimentary); fused hearts but with separate chambers.	Autopsy reveals hearts fused at apex via a fibrous band.
Kaveh *et al*. (2011) [11]	Postnatal anatomical examination of thoraco-abdominal structures.	Dicephalic parapagus with substantial thoraco-abdominal deformities.	Autopsy reveals anatomical malformation of internal organs
Yadav and Yadav (2014) [12]	Not mentioned	Two heads; two upper and lower limbs; Single thorax and abdomen; associated neural tube defect.	Autopsy not reported.
Gothwal *et al*. (2018) [8]	Early prenatal US at 13-weeks.	Two heads attached to a single trunk Severe congenital anomaly.	Diagnosis confirmed after termination; autopsy not done.
N'Guessan *et al*. (2019) [13]	Early Prenatal US; diagnosis	Two heads; single trunk	Autopsy not reported.
Banjarnahor et al. (2022) [14]	Obstetric US showing cardiac and visceral anomaly	Case 1: Two heads, fused heart, fused liver, four kidneys. Case 2: Two heads with single heart and liver; limb deformity	Pregnancy terminated Autopsy not reported.
Guerrero Cevallos et al. (2023) [15]	Detailed obstetric US and MRI confirmed anatomical defects.	Two heads, single trunk. Two spinal columns, single heart; anomalies including cleft lip& palate, imperforate anus, and clubfoot.	Severe polyhydramnios noted. Autopsy not reported.
Febrianda and Sriyanti (2024) [16]	Fetomaternal ultrasound, MRI, and 3D CT confirming diagnosis at ~20 weeks gestation.	Two heads with single body.	Autopsy not done.

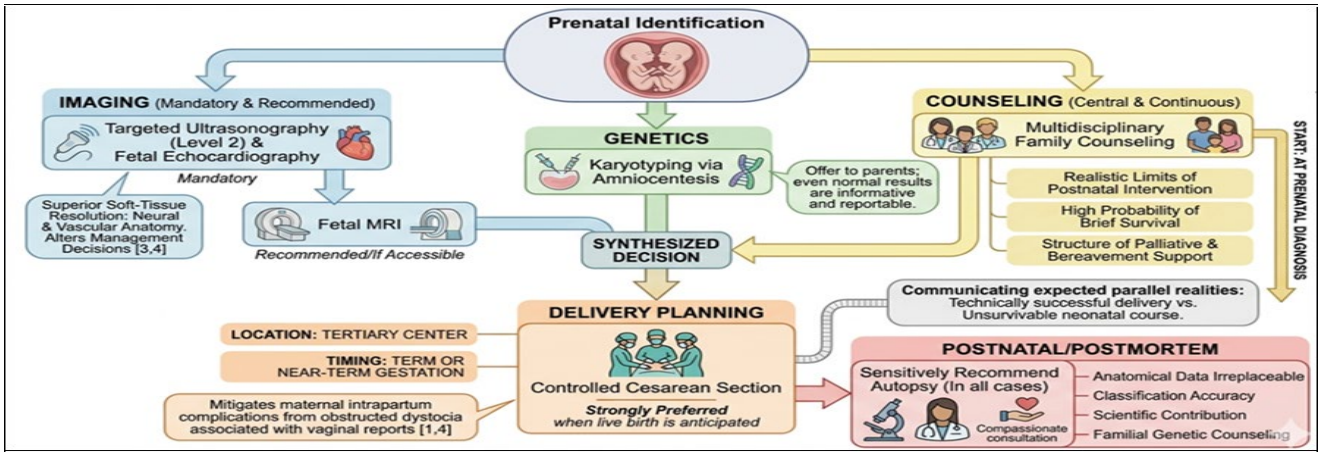

Figure 2: Suggested management paradigm of cases with PDT.

The majority of affected pregnancies result in intrauterine fetal demise, stillbirth, or neonatal death within hours of delivery [9]. The diagnosis should be communicated promptly to the parents together with all the information about the low number of viable treatment options, high mortality rates, and preparation for palliative and bereavement care prior to birth [17]. Pregnant women should be closely followed for the risk of preterm birth and polyhydramnios. Vaginal delivery carries substantial maternal risk due to the enlarged and dysmorphic presenting anatomy. The prognosis is deemed poor for such cases, and for that an early prenatal diagnosis should be combined with multidisciplinary team counseling with early termination of pregnancy as advice to avoid unnecessary maternal complications. Should the termination be declined or if the diagnosis was delayed until delivery, then it is recommended that the delivery be conducted at a tertiary center, keeping in mind that vaginal delivery may be feasible in selected cases [18-20]. A postmortem autopsy should be encouraged since postmortem data is necessary for correct classification, medical documentation, and genetic counseling [21]. In this case, having normal prenatal imaging did not confer viability. The shared neural and cardiopulmonary architecture imposes fatal constraints that cannot be fully characterized before birth. The infant survived only four hours; consequently, even with the presence of normal-looking anatomy, the prognosis should not be optimistic, but it is almost always very poor [8]. We have summarized 4 cases for Arabic women that were reported during the last 2 decades in (Table 3**)** [22,23,9]. The principal strength of this case is prenatal diagnosis, which enabled prospective rather than reactive management—

referral, multidisciplinary team assembly, delivery planning, and parental preparation all occurred in an orderly framework rather than as an emergency response [8,9]. The earlier diagnosis enabled a meaningful contribution to the standard of care in a condition where late or intrapartum diagnosis carries substantial maternal risk [3]. The absence of fetal MRI limits the complete definition of shared neural and vascular anatomy [20]. The absence of a postmortem examination prevents definitive confirmation of the visceral and neuropathological findings that would advance the scientific record of such cases and permit meaningful cross-case comparison and refine the prognostic literature, while karyotyping tends to assess chromosomal status and supports parental genetic counseling; however, most conjoined twins demonstrate a normal karyotype [21].

Table 3: Comparison of reported cases in Arab people

Parameter	Case 1	Case 2	Case 3	Case 4
Author's name and year	Bouya *et al*. (2021) [22]	Bouya *et al*. (2021) [22]	Pelayo *et al*. (2019) [23]	Harma *et al*. (2005) [9]
Nationality	Tunisian	Tunisian	Moroccan	Arabian women living in Turkey
Maternal age	34 years	28 years	25 years	34 years
Obstetric history	G3P2	G1P0	G1P0	G5P4
GA at diagnosis	12 weeks	13 weeks	12+6 weeks	Diagnosed at delivery (37–38) weeks
Diagnostic modality	Transabdominal ultrasound	Transabdominal and transvaginal ultrasound	2D / 3D / 4D ultrasound	Postnatal radiography (X-ray)
Cephalic anatomy	2 separate heads	2 separate heads	2 separate, fully developed heads	2 heads, 2 vertebral columns
Fusion level	Thorax and abdomen	Neck, thorax and abdomen	Thorax and abdomen	Pelvis (lower half)
Cardiac state	2 hearts	2 heartbeats	2 hearts	Not specified
Limbs number	4 upper, 2 lower	3 arms, 2 legs	2 arms, 2 legs	2 arms, 2 feet
Pathological confirmation	Fetal pathology	Fetal pathology	Clinical/imaging	Clinical/radiography
Outcome	Terminated at 13 weeks by misoprostol	Terminated at 13 weeks by misoprostol	Pregnancy interrupted	Stillbirth (Apgar 0); vaginal delivery; birth weight 3,500 g

Conclusion

Dicephalus parapagus twins are extremely rare, lethal congenital malformation. The cornerstone of diagnosis confirmation is the ultrasound, which enables timely counseling and intervention. Fortunately, most cases are diagnosed prenatally and are terminated early. Still, those who continue to term need a multidisciplinary team and planned delivery. Histopathological classification is vital to enrich the limited data on the topic.

Conflict of interests

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N/A

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