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An anencephalic heterophagus or parasitic twin. A case report with literature review

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ABSTRACT

A case of 6-week-old female twins is presented. Twin 1 was fully formed, whereas twin 2 was parasitic, adhered to the left flank, and had anencephaly and complete upper limbs. Depending on the twins' overall condition, the recommendation is to separate conjoined twins as soon as possible for a better outcome.

Keywords
anencephaly, heteropagus, parasitic twins

INTRODUCTION

With an estimated prevalence of fewer than 0.1 per 100,000 births, heteropagus or parasitic twinning is a very unusual disorder. Ambroise Pare, a 16th-century French surgeon, described an acephalus body linked to the abdomen of one of his patients, which is perhaps the earliest plausible mention of the ailment.[1] The symmetry, site of fusion, and grade of duplication of conjoined twins are characterised, with thoracopagus being the most common kind (40%), followed by thoraco-omphalopagus (28%). Asymmetrically conjoined twins also known as heteropagus or parasitic twins are difficult to categorise.[2]

According to Spencer[3], parasitic twins are conjoined twins that are linked in one of the same anatomical sites as "complete" conjoined twins. One of the twins has a significant disability and is referred to as a "parasite." While certain congenital anomalies, such as heart malformations, may be present, its corresponding "autosite" must be largely intact. To describe parasitic twins, the terms "heteropagus," "asymmetric conjoined," "partial or incomplete conjoined," "parasitic," and "exo-parasitic" are all used interchangeably.[1]

The largest documented series on the incidence of heteropagus twinning comes from a database of 7.9 million births collected over an eight-year period in the 1970s in the United States. The true incidence of heteropagus twins was estimated to be 0.05 to 0.1 per 100,000 in this US study.[5] An incidence of 0.02 per 100,000 was discovered in a more recent European research of approximately 5 million newborns.[4] The frequency of heteropagus pairs as a fraction of all conjoined twins has been estimated to be between 4.5 and 15%.[5] In symmetric conjoined

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twins (72%), there is a significant female preponderance; however, in heteropagus twins, the gender difference is less prominent. In a 2001 study of 157 heteropagus twins, 51% were female.[3]

Case presentation

Female twins, 6 weeks old, were presented to us at the emergency department. The twins were conjoined and parasitic. There was no significant history of any antenatal checkups during the pregnancy. No record of any antenatal ultrasound was available. Twins were born at full term and without any medical assistance at home. According to the family, the twin 1 (complete twin) cried after a few minutes of birth. The birth weight of the twins was unknown. On presentation to the emergency, the twins were ordered to have baseline investigations, X-rays and then an Magnetic Resonance Imaging (MRI) lumbar spine without contrast. On examination, the complete twin had a well-formed head, torso, upper limbs, and lower limbs. A parasitic twin was attached to the left flank and presented with an anencephalic head and normal upper limbs as shown in Figure.1 (A,B&C). On MRI, a scoliotic deformity of the thoracolumbar spine was noted in twin 1. There was a convexity towards the left side of the spinal cord terminating at L3/L4 level.

Figure 3. Twin 1: Complete twin (Autosite), Twin 2: Partial twin (Parasite).

There was a partial widening of the spinal canal along with partial defective posterior elements. Twin 2, the partial twin, had non visualisation of brain matter as well as the spinal canal. After sufficient radiological reports and clinical evaluation, elective surgery was scheduled to separate the conjoined twins. Before the surgery could take place, there were episodes of repeated apnea attacks. Even though sufficient resuscitative measures were taken, unfortunately, the twins died before the planned surgery.

Discussion

The spontaneous occurrence of twinning, both monozygotic and dizygotic, is being highlighted. Conjoined twinning, on the other hand, happens only when the twinning event occurs during the primitive streak stage of development, which occurs around 13-14 days after conception in humans, and is only associated with the monoamniotic monochorionic form of placentation. Humans are said to be the species with the highest rate of conjoined twinning. While monozygotic twinning can be created in animals in the lab using a number of chemicals, the mechanism of spontaneous twinning in humans is unknown. All agents that can cause twinning are teratogenic, and they most likely do so by interfering with the spindle apparatus.[2]

According to the findings of the greatest study to date, the spontaneous birth rate is around 10.25 per million. Thoraco-omphalopagus (28%), thoracopagus (18.5%), omphalopagus (10%), parasitic twins (10%), and craniopagus (6%) were the most common types seen. Approximately 40% of these babies were stillborn, while 60% were born alive, albeit only about 25% of those who made it to birth lived long enough to be surgical candidates. Conjoined twinning is caused by the incomplete division of the embryonic axis, and all conjoined twins are symmetrical, with the exception of parasitic conjoined twins, and "the same parts are always attached to the same parts."[1] Table.1 shows a comprehensive evaluation of the literature on heteropagus twins from 2001 to 2022.

Conjoined twins are characterised based on their symmetry, fusion site, and duplication grade. In contrast to conjoined twins, who have the same locations fused, parasitic twins are a subset of conjoined twins who join "asymmetrically," with their own varieties and classification.[1] The anencephalic parasitic twin was connected to the left flank of the autosite in our reported case, an unusual presentation that has never been defined or classified before.

Table 1. Literature review on heteropagus twins from 2001 to 2022

Authors	Year	Type of heteropagus	Gender	Autosite abnormalities	Parasite features
EK Cury[13]	2001	1.Epigastric heteropagus 2.Epigastric heteropagus 3.Epigastric heteropagus	1.M 2.M 3.F	1.ASD 2.Omphalocele 3.Absent 1/3rd sternum	BUE, BLE, bowel, bladder, kidney, ureter, pelvis, external genitalia & patent urethra
DK Gupta[14]	2001	1.Epigastric heteropagus 2.Epigastric heteropagus	1.M 2.M	1.Omphalocele 2.VSD, PDA	UUE
P Raynal[15]	2001	Epigastric heteropagus	M	Omphalocele	Size: 12×10 cm, BLE, bowel, pelvis & imperforate anus
JK Mahajan[16]	2002	Ischiopagus	F	Omphalocele, single ovary, hemiuterus	BLE, bowel, bladder, ovary×1, hemiuterus, pelvis, external genitalia & patent urethra
DA De Ugarte[17]	2002	1.Epigastric heteropagus 2.Epigastric heteropagus	M	Inferior sternal cleft, VSD, PFO, TGA, PDA, COA, omphalocele, meckel diverticulum	Rudimentary BUE, complete BLE, palpable spine, liver, gallbladder, kidney, ureter, bladder, pelvis, testes×2 (undescended) & external genitalia
Martinez[1]	2003	1.Epigastric heteropagus 2.Acardiac acephalus	-	-	-
A Tongsin[18]	2003	Epigastric heteropagus (4)	-	Congenital heart defect, omphalocele	-
JR Corona-Rivera[19]	2003	Ischiopagus	M	Small left diaphragmatic defect, omphalocele, exstrophy of the cloaca, and lumbar meningomyelocele	Complete left LE, hemipelvis, lumbosacral vertebral column, spinal cord, and one kidney with ureter & adrenal gland
AP MacKenzie[20]	2004	Epigastric heteropagus	F	Bilateral choroid plexus cysts, dextrocardia, omphalocele	BLE & pelvis
AT George[21]	2004	Epigastric heteropagus	M	Talipes equinovarus	BLE, accessory abdominal compartment, bowel, liver, gallbladder, cystic duct, pelvis, testes×2, phallus & empty scrotum
SK Ratan[22]	2004	Rachipagus	F	Spinal dysraphism, tethered cord	Size: 8×8 cm, ULE (fused feet), vertebrae×2, bowel, pelvis, bladder & fallopian tissue
AK Bangroo[23]	2004	Epigastric heteropagus	M	-	UUE, BLE & phallus

K Fujimori[24]	2004	Omphalopagus	F	Single ventricle, hypoplastic aorta	BUE (limb buds), alobar holoprosencephaly, cleft palate/lip & hypoplastic lung
RC Ribeiro[25]	2005	Xipho Omphalopagus	M	Unilateral renal agenesis	Hydrocephalus, hypertelorism, choanal atresia, micrognathia, trachea, rudimentary heart, bowel & bladder
M Bhansali[26]	2005	1.Epigastric heteropagus 2.Epigastric heteropagus 3.Epigastric heteropagus	1.F 2.M 3.F	1.Omphalocele 2.Omphalocele 3.Unknown	1.BUE, BLE, atretic rectum, absent anus, kidney×2, pelvis 2.BLE (limb buds) 3.BLE, pelvis, external genitalia
H Rode[27]	2006	Ischiopagus (4) Not explained (5)	F	-	-
R Chadha[28]	2006	Rachipagus	F	Spinal dysraphism	Size: 11×8 cm, anal dimple, external genitalia, empty scrotum & digits×3
Shibata[1]	2006	Craniopagus	F	None	Multiple cranial malformation, hair, scalp, rudimentary trachea, lungs, esophagus, stomach & bowel
Y Kanamori[29]	2006	Epigastric heteropagus	F	Abducted/flexed LE	BUE, BLE (fused), lung, stomach, bowel, liver, pancreas, kidney, ureter, bladder, pelvis, external genitalia & ovaries×2
J Hager[30]	2007	Epigastric heteropagus	F	VSD	BUE, BLE, bowel, imperforate anus, multicystic kidney, pelvis, external genitalia & blind urethra
CM Snelling[31]	2008	Rachipagus	M	High arched palate, retrognathia, posteriorly set ears, simian crease, spinal dysraphism, meningocele, hydromyelia, split, tethered cord	Size: 4×4×2 cm, amorphous with skin, bone, digit, clavicle & scapula
G Albert[32]	2008	Rachipagus	F	Spinal dysraphism, myelocystocele, teratoma	Size: 8×9.5× 2.5 cm, amorphous
E Satter[33]	2008	Omphalopagus	F	-	Size: 15×9 cm, amorphous with skin, hair, bone, rudimentary URT, bowel & liver

XL Hu[34]	2009	Epigastric heteropagus	M	-	Limbs, abdomen, buttocks & male external genitalia
S Sanoussi[35]	2010	1.Rachipagus 2.Rachipagus	1.M 2.M	1.Meningocele, spinal lipoma 2. Spina bifida	1.Complete LE & rudimentary genitalia 2.Dysmorphic UE fused at the midline
AM Abubakar[36]	2011	Epigastric heteropagus	M	Ventral hernia of a healed exomphalos	BLE, rudimentary UUE, pelvis, external genitalia & imperforate anus
S Das[37]	2011	Epigastric heteropagus	M	Omphalocele	Rudimentary limbs, auricles, and hairs on the scalp
J Zhang[38]	2011	Rachipagus	F	Spina bifida, diplomyelia, scoliosis, tethered cord, & cardiac VSD	Soft-tissue mass including fully developed breast glandular tissue with an external nipple-areola complex, uterine tube & duct, bone & cartilage
M Qasim[39]	2011	Epigastric heteropagus	M	-	Size: 6x4 cm, rudimentary UE, LE, head & external genitalia
H Ozkan-Ulu[40]	2011	Epigastric heteropagus	M	Cardiac malformation	UUE & rudimentary external genitalia
RV Reddy[41]	2011	Pyopagus	-	-	Rudimentary bony hemipelvis, BLE with gross deformity of right UE, intergluteal fold without anal orifice, partial gastrointestinal system & rudimentary bladder
A Solak[42]	2012	Rachipagus	F	Spina bifida	Dysmorphic & immobile UE with 4 small buds at the base
JT Xie[43]	2012	1.Epigastric heteropagus 2.Epigastric heteropagus	1.M 2.M	1.None 2.Omphalocele	1.BUE, BLE, pelvis, scrotum, and penis 2.Immobile LE, buttocks, perineum & male genitalia
K Kesan[44]	2013	Epigastric heteropagus	M	Omphalocele	-
G Komla[45]	2013	1.Epigastric heteropagus 2.Epigastric heteropagus 3.Omphalopagus	1.F 2.M 3. -	1.Omphalocele 2.Omphalocele and fibrous mitral valves 3.Systolic murmur	1.BUE & BLE 2.BUE & ULE 3.BUE & ULE
DR Calderoni[46]	2014	Thoraco-omphalopagus	M	Omphalocele	BUE, BLE, external genitalia, scrotum & anus
Y Bayri[47]	2014	Rachipagus	M	Lumbosacral meningocele, unfused	Accessory LE & rudimentary hemipelvis

				vertebral arches between L1 & S1	
EC Gokcen[48]	2015	Ischiopagus	F	-	BLE, rudimentary LE
D Baskaran[49]	2015	Epigastric heteropagus	M	-	Gastroschisis, BUE, BLE, pelvis
AA Navaei[50]	2015	1.Rachipagus 2.Rachipagus	1.F 2.M	1.None 2.Neurogenic bladder	1.Fully formed LE & hemipelvis 2.Soft-tissue mass with rudimentary scrotum & penis as well as bony structures resembling a scapula & rib
A Sahlu[51]	2016	Rachipagus		Lipomyelomeningocele and tethered cord	Foot, ankle, lower leg & a small sinus that resembled an anus
K Ahmed[52]	2016	Thoracic heteropagus	M	Giant omphalocele	Hypoplastic BUE, thorax, misshapen trunk, small abdomen, pelvis, BLE, male genitalia, empty scrotum & imperforate anus
KN Rattan[53]	2016	Rachipagus	F	-	Ulcerated, multicystic mass with rudimentary limb-like structures including bone & cartilage
P Raj[54]	2017	Epigastric heteropagus	M	-	BLE, rudimentary UE, pelvis, scrotum, penis & imperforate anus
A Raheja[55]	2017	Rachipagus	-	-	-
A Khushdil[56]	2018	Epigastric heteropagus	M	Omphalocele	LE, UE, pelvis & male genitalia
M Malik[57]	2018	Epigastric heteropagus	M	Omphalocele	BLE, rudimentary UE, external genitalia, scrotum & absent testes
N Khavanin[58]	2018	Rachipagus	F	-	BLE, cervicothoracic diplomyelia, pelvis & rudimentary scapula
AR Muhelo[59]	2018	Ischiopagus	F	-	BLE
MS Falyoun[60]	2020	Epigastric heteropagus	M	Omphalocele	BLE, external genitalia, scrotum, absent testes & anus
AB Pati[61]	2020	Rachipagus (2)	-	-	-
RCS Alves[62]	2020	Asymmetrical	F	Anencephaly	BUE, BLE, rudimentary external genitalia & pelvis

MH Takrouney[12]	2020	1.Asymmetrical 2.Epigastric heteropagus 3.Epigastric heteropagus	1.F 2.M 3.M	1.Unexplained 2.Omphalocele 3.Omphalocele	1.Incomplete head & soft tissue swelling 2.BLE, rudimentary UE & external genitalia 3.BLE, rudimentary gut

"Fission" and "Fusion" are the two basic ideas for the embryologic origin of conjoined twins. Incomplete fission of the blastocyst inner cell mass at the primitive streak stage, 13 to 15 days postfertilization, results in two foci of axial growth that preserve a connection at some point, according to proponents of the former theory.[6] "Fusion," on the other hand, refers to the coalescence of two initially separate inner cell masses at a later stage.[3] Donitz et al.[7] proposed the most widely accepted theory for the formation of asymmetry between autosite and parasite after these occurrences. They hypothesised that vascular impairment causes the parasitic twin's tissue to become reliant on autosite collaterals. The starved section of the parasite's body experiences selective ischemic atrophy.[1]

Asymmetric conjoined twins, like their symmetric counterparts, were traditionally thought to be universally monozygotic. Recent studies that used deoxyribonuleic acid (DNA) polymerase chain reaction to analyse renal tissue and blood cells from the autosite and parasite support this conclusion.[8,9,10] One group, on the other hand, has discovered a parasite with scrotal skin that is related with a female autosite.[11]

Depending on the location of the attachment and the shared inner parts, surgery to separate conjoined twins might be simple or complex. Rectal manometry can only be employed if the anal canal is impacted or implicated during separation, and the surgery often leads in the death of one or both of the conjoined twins, especially if they are attached at the head or share a vital organ.[12] For a better outcome, we urge that conjoined twins be separated as soon as feasible, depending on their overall condition. Recently, endoscopic minimally invasive techniques like laparoscopy and robotic surgery have been used in the preoperative and operational management to examine internal organs and determine whether or not connections exist, as well as to treat connections. The mainstay of the separation procedure for external connections is open surgery.[12]

Conclusion

An anencephalic parasitic twin attached to the autosite's left flank has never been classified or reported before. Separating conjoined twins as soon as possible, depending on their general condition, is recommended for a better outcome. Depending on the location of the attachment and the shared inner parts, surgery to separate conjoined twins might be simple or complex. Endoscopic minimally invasive procedures can now be employed for internal organ assessment and surgery planning, thanks to recent advancements.

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