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Case Report

Dorsolumbar parasitic twin associated with lipomyelomeningocele: a case report from a tertiary teaching hospital, Ethiopia, East Africa☆

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ABSTRACT

A parasitic or heteropagus twin is a grossly defective fetus (or fetus part) attached externally, with or without internal connections and is dependent on the cardiovascular system of the other twin (autosite) for survival. The estimated incidence is approximately 1 per 1 million live births. To date according to the authors' knowledge; there are a few case reports published in the literature. Here we present a case of dorsolumbar parasitic twin with associated lipomyelomeningocele.

Introduction

A parasitic or heteropagus twin is a grossly defective fetus (or fetus part) attached externally, with or without internal connections, to a relatively normal twin in one of the same 8 areas in which symmetrical twins are united [1]. Classification of conjoint twins is based on the site of the union. In the thoracoomphalopagus types, they are attached at the chest and abdomen (74% of cases), in the pygopagus types, they are joined at the buttocks (18% of cases), in the ischiopagus varieties, they are attached at the ischium (6% of cases), and the craniopagus ones at the level of the head (2% of cases). A rachipagus parasite is one where the fusion occurs at the spinal level [2–4]. It is a rare entity with an estimated incidence of less than 0.1 in 100,000 live births [5]. The atrophic embryo which is called the parasitic twin becomes atrophic and parts of it will survive attached to the normal embryo (autosite) at the junction area [6]. Any structure can be found in the parasite. Bones of the limbs are frequently present. Rarely the heart and neural tube structures can be found in the parasite. In some lumbar parasites, glandular tissue, intestine, or anal orifice can be observed [7]. We present a rare case of rachipagus parasite at the dorsolumbar level with a review of contemporary literature.

☆ Competing Interests: The authors hereby notify the editors that they have nothing to declare and there is no conflict of interest.

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Case report

A 4-day-old neonate was brought to our hospital by its parents who reported a strange anomaly of accessory lower limbs on the right lateral back. The neonate was born to a 28-year-old para I lady. The parents confirmed they did not belong to the same family tree. Antenatal course was uneventful and no history of maternal medication or harmful drug use or exposure to radiation was reported during pregnancy. *The antenatal report documented that prenatal ultrasound showed twin pregnancy with little or no specific details.*

On physical examination, a well-formed parasitic twin part *was* attached to the back, in the dorso-lumbar region. *The parasitic attachment consisted of well-formed lower limbs (paired femoral, tibial, fibular, and foot bones, with knee, and ankle joints; there were poorly formed pelvic bones and a male external genitalia.*

Thoracolumbar MRI, abdomino-pelvic CT and abdominal ultrasound *were* performed. On ultrasound, the right ischial bone and both femora of the parasitic twin were visualized. The autosite had posterior thoracolumbar vertebral bone defect with herniation of a cystic mass containing neural elements in the right posterolateral surface *which was* closely related with the parasitic twin. The right lateral abdominal wall muscle of the autosite showed continuation with the parasitic twin at which level the right kidney herniated through. The arterial supply of the parasitic twin arose from the infrarenal thoracic aorta which was also confirmed on CT scan. MRI and CT showed widening of the spinal canal with posterior vertebral bone defects from Th10 to L5 and hemivertebrae of T12 and L2 (Fig. 1). There was fat in the lower portion of the mass. A fluid filled herniated structure containing neural elements and fat was visualized (Fig. 2). The parasitic twin had partially formed pelvic bones including the right iliac bone and bilateral ischial bones, femora, tibia and fibula, and foot bones (Fig. 3). Male external genitalia were visualized on the 3D reconstruction on CT but no pelvic visceral organs were seen (Fig. 4). There was no osseous connection or major visceral organ sharing.

A successful surgical removal of the parasitic twin was done. The autosite developed right lower limb monoparesis which was likely iatrogenic.

Discussion

The earliest description of heteropagus (parasitic) twining was in the 16th century by a French surgeon Ambroise Pare. In another report, as described by spencer the twins were conjoined where one of them (the parasite) was severely defective and depended on the other (the autosite) for survival [5]. Besides the label parasitic twin, various other names had been given to this condition such as heterotopic redundancy, aborted twinning, teratoma, tripedus, disorganization-like syndrome, spinal hamartomas, mature teratoma, mid-

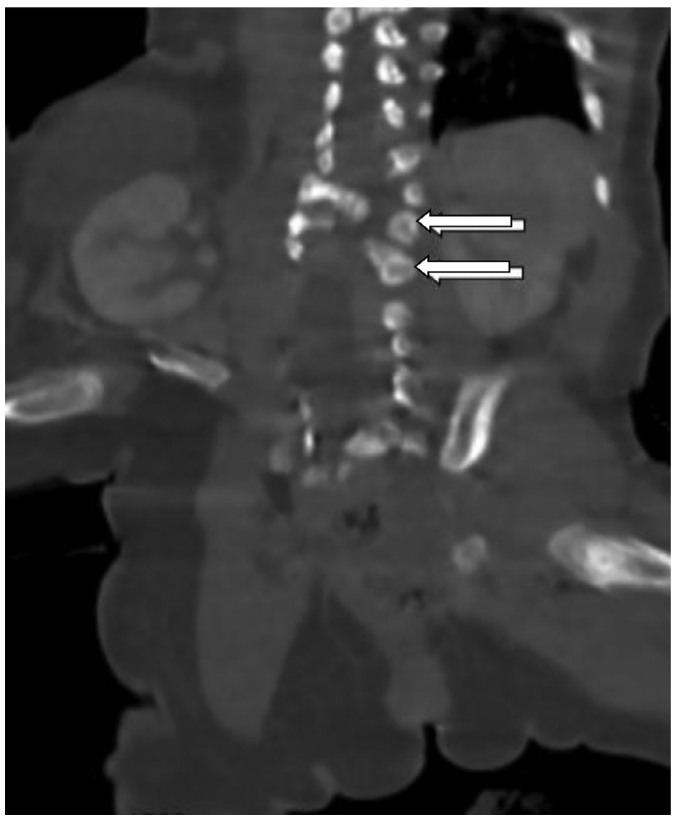

Fig. 1 – Coronal CT scan bone window showing T12 and L1 hemivertera (white arrows). TASH, 2020.

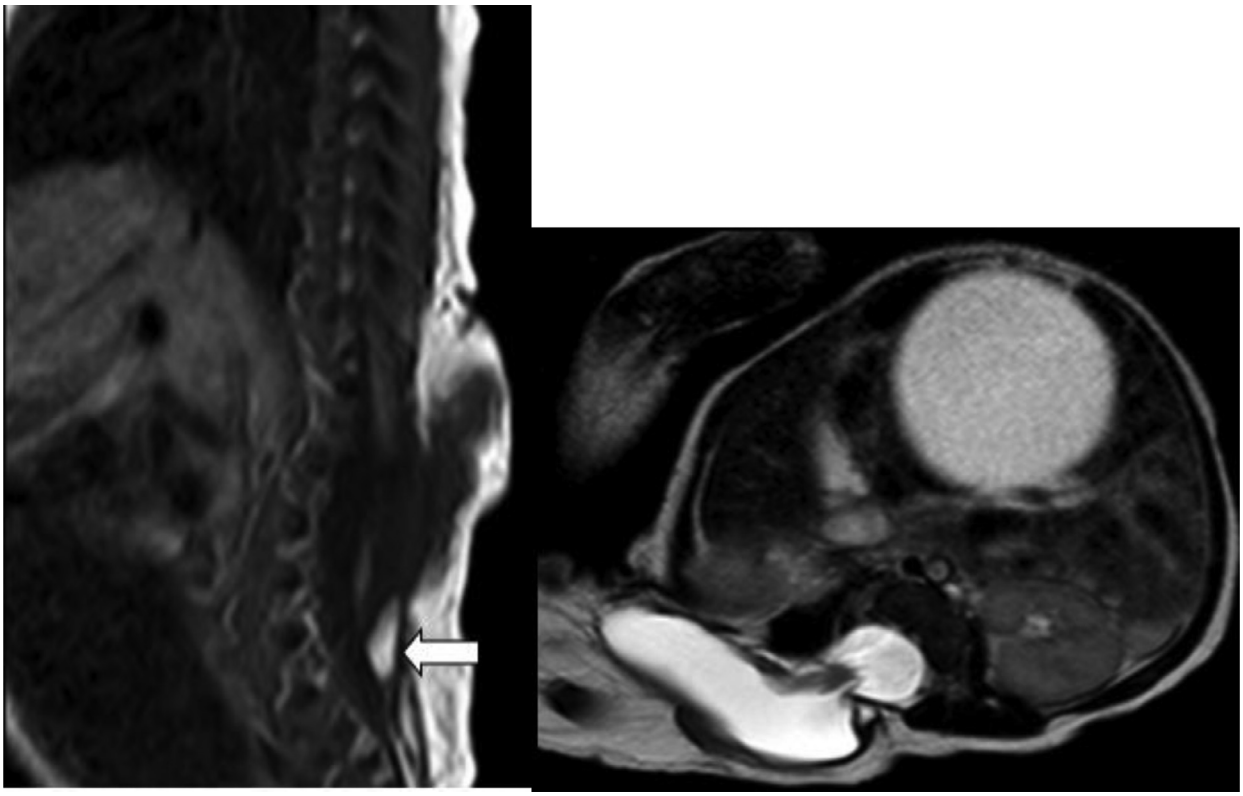

Fig. 2 – Coronal T1 showing the posterior lumbar defect fat (arrow) in the lower portion and axial T2 WI showing the posterior spinal defect containing the neural elements. TASH, 2020.

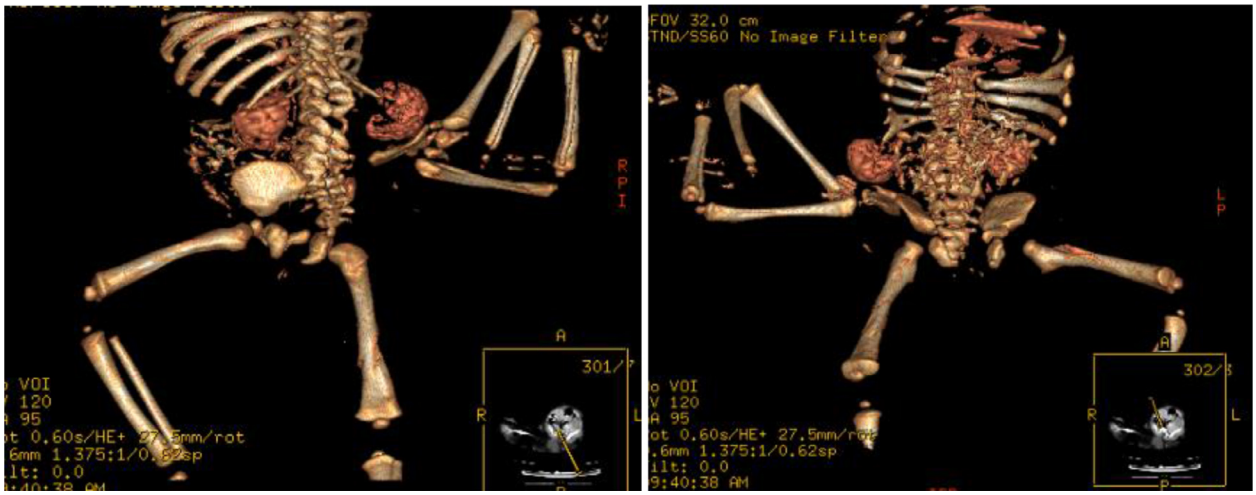

Fig. 3 – Surface extraction showing the partially formed pelvic and lower limb bones of the autosite and parasitic twin. TASH, 2020.

line dorsal appendages, and dorsal accessory limbs [1]. Conjoined twinnings are classified in terms of the attachment site of the body: thoracopagus (thorax), ischiopagus (pelvis), cephalopagus (face), omphalopagus (abdomen), and craniopagus (cranium). Rachipagus describes a parasitic twin joined dorsally at the vertebral column. If the conjoined region is located at the lumbosacral spine, it is referred to as pygopagus [8].

No particular risk factors for developing heteropagus twins have been reported to date [9]. The most widely accepted theory explaining the embryogenesis of this abnormal condition is the fusion hypothesis. In the early fetal life, embryonic discs of monozygotic monoamniotic twins are located in the same amniotic cavity. At the third or fourth gestational weeks, the neural folds of the 2 different embryos can merge if the skin covering the neural tube gets damaged. If the 2 embryos de-

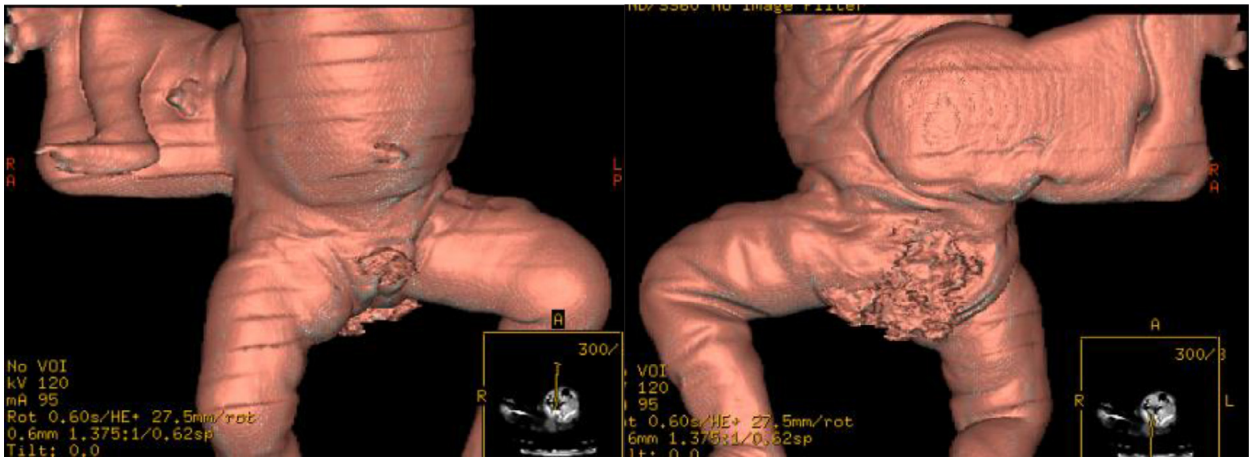

Fig. 4 – 3D reconstruction showing both lower limbs & the external male genitalia of the parasitic twin attached to the autosite. TASH, 2020.

velop equally, 2 complete but conjoined fetuses arise [10]. The attached part composed of primitive embryonic tissue prevents closure of the neural tube during later development, resulting in spina bifida, or other neural tube defects [6, 9]. Important abnormalities involving other organ systems are rare in the living fetus. In patients with rachipagus, a lipoma at the base of the parasitic mass with an intraspinal extension had frequently been noticed [6].

Neural tube defects associated with ectopic limbs have been reported in a few cases. There was a case report of accessory legs associated with spina bifida and rudimentary external genitalia [11]. There was also a report of 2 cases with spina bifida and hemivertebrae [12]. In the case presented here, there was thoracolumbar spina bifida and hemivertebra with lipomyelomeningocele and a parasitic twin with well-formed lower limbs and external genitalia. No other anomalies in the autosite were diagnosed.

Conclusion

Parasitic twin attachment in the lumbosacral region is rare; imaging is of paramount importance in diagnosing such anomalies and provides detailed morphological descriptions that help in accurate surgical intervention and further follow up of the autosite.

Patient consent

We have obtained the parents' consent and they fully agreed with the authors that the case can be reported and published for educational purposes as long as the identity of the patient and family is kept anonymous.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2022.07.079.

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