

Successful Separation of Rachipagus Parasiticus Neonate: Case Report and Review of the literature

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Abstract

Background Data: Lumbosacral Rachipagus Parasiticus is an extremely rare condition. Separation of a parasitic twin that is attached at the lumbosacral spine in the neonates is even less frequent. In this case the spinal cord was herniated through a spina bifida into the parasite.

Purpose: The aim of this report is to present a case of Rachipagus Parasiticus in which surgical separation was successfully performed.

Study Design: Case Report and review of the literature.

Patients and Methods: The child was 28 days old. Computed tomography, magnetic resonance imaging of the whole spine focusing on the lumbosacral spine provided the information necessary to perform surgery. The duration of surgery was 150 minutes. The parasitic twin hadn't neural attachment with the myelomeningocele. The child received 120 CC of blood. After separation of the parasitic leg, the dura was repaired using autogenous graft. Skin and subcutaneous tissue were closed directly after application of a wound drain for 24 hours.

Received at:

July 22th, 2016

Accepted at:

September 2nd, 2016

Results: The parasitic twin was successfully separated, and the patient was discharged from the hospital after 4 days, moving her lower limbs proximally with some weakness in both feet.

Conclusion: Rachipagus parasitic twin is a rare form of conjoined twin with a favorable outcome. Detailed perioperative evaluation and management planning, as well as multidisciplinary microvascular approach, are integral to perform the best separation surgery. (2016ESJ096)

Keywords: Rachipagus, Parasite, Conjoined twinning, Spinal dysraphism

Introduction

Conjoined twins have always been considered as a rare challenging condition, of which, the parasitic type are rarer. The rarest of the rare, is the Rachipagus type. It's derived from the Greek words Rachi- (spine) and -pagus (fixed). According to a worldwide collaborative epidemiological study of the International Clearinghouse for Birth Defects Surveillance and Research (ICBDSR), the total prevalence of conjoined twins was 1.47 per 100,000 births, the parasitic type form 5% of conjoined twins. The Rachipagus type represents 1% from those 5%.¹⁵

Conjoined twins are always joined at homologous sites, and the clinical classification is based on the most prominent site of union. There are eight recognized nomenclatures in this field, including thoracopagus (chest), omphalopagus (umbilicus), ischiopagus (hip), pygopagus (rump), rachipagus (spine), craniopagus.²⁵

The parasitic type is a grossly defective fetus, or fetal parts, attached externally (perhaps with internal connections as well) to a relatively normal twin (the autosite) in one of the same areas in which intact conjoined twins are united.²⁶ The parasitic twin can contain any structure. However, limbs bone are more frequently present. In some lumbar parasites, the glandular tissue, intestine, rudimentary genitalia, or anal orifice can be observed.⁶ We present one additional case of parasite Rachipagus on the lumbosacral level.

Case Report:

A baby girl 28 days old delivered by Cesarean section at 38 weeks of gestation due to difficult normal vaginal delivery. The baby was referred to our service from Libya with a mass in her back. The condition was not discovered at the anti or perinatal period. There was positive consanguinity with no family history of any twins or congenital anomalies. The child had 3 healthy siblings, and there was no history of maternal illness, drug intake, alcohol, or smoking during pregnancy.

A complete healthy baby girl weighing 3.5 kgms presented with one lower limb attached to her back at the lumbosacral region. A parasitic twin formed of one completely formed lower limb (with extra digit at the heel) attached to rudimentary pelvic girdle and rudimentary genitalia anal dimple with very bad odor. The parasitic limb hadn't any motor or sensory function. There were area of capillary hemangioma and dimpling of the skin above the attached mass.

Plain x-ray has shown unfused posterior vertebral arches between L1-S1 segments. The accessory limb was attached to a rudimentary posterior vertebral arch by rudimentary hemipelvis at L5-S1 level. The leg was arising from lumbosacral region in the midline and containing two long bones resembling femur, tibia, and foot bones. MRI examination of the spine disclosed a low-lying conus with a lumbosacral meningocele herniating in the parasite (Figure 1). CT scan

of the Brain was within normal limits with no associated cerebral anomalies.

Surgery Procedure:

The child was 28 days old when the surgery was performed. Anesthesia was induced by sevoflurane 7%, fentanyl 1.5 µg/Kg intravenous, atracurium besylate 0.5 mg/Kg and endotracheal tube 3.5 mm was inserted. Monitoring was conducted by ECG, Non Invasive Blood Pressure (NIBP), oxygen saturation and temperature monitoring through a rectal probe. Prone positioning and controlled ventilation with tidal volume 7ml/Kg, Respiratory Rate 20/min. Maintenance by sevoflurane 2% in oxygen & muscle relaxants as needed. Fluid therapy in the form of paedimaint solution to replace deficit and maintenance fluid needed. An elliptical skin incision was made at the base of the swelling, with superior and inferior extensions. A well-demarcated avascular plane could be found between the spinal canal of the autosite and the parasite, except for the meningocele and small arteries going from autosite to parasite (which was the main arterial supply to the parasite) with no major vessels was identified. The lipomatous element of the parasite was completely limited to the outside of the dura of the autosite. When the dura was opened,

the partially unfolded posterior cord in the region of the parasite's attachment was found adherent to the overlying dura, but at this stage no detethering of the cord was attempted. Neural tissue microscopic dissection was the most serious problem, as there wasn't neuro-monitoring available for this age, but the parasitic twin hadn't neural attachment with the meningocele. After separation of the parasitic legs, the dura was repaired using autogenous graft. Skin and subcutaneous tissue were closed directly after application of a suction drain for 48 hours, with good dressing compression to avoid any CSF or seroma collection. Duration of surgery was about two and half hours, bleeding was the second serious problem, blood loss was about 140 cc and replaced by packed RBCs about 120 cc. Warming device was used and reversal of muscle relaxant by neostigmine and atropine. Good recovery and hemodynamically stable.

The baby was discharged from the hospital after 4 days, moving her lower limbs proximally with weakness in both feet. Suture was removed two weeks postoperatively. The wound was clean with no CSF leakage or subcutaneous collection (Figure 2).

Table 1. Reported Cases of *Rachipagus Parasiticus* in the Literature.

Series	Cases added	Sex (No.)	Autosite (No.)	Parasitic Organs (No.)
Spencer et al, 1995 ²⁷	24	M(4), F(14), NR(6)	C(4), T(8), TL(4), L(4), LS(4)	UL(8), LL(16)
Krishna et al, 1999 ¹²	9	F(8), M(1)	1C, 3T, 2L, 2LS, 1S	UL(4), LL(5)
Chadha et al, 2004-2005 ^{4,19}	2	F(2)	LS(1), TL(1)	LL(2)
Sharma et al, 2008 ^{2,23}	2	M(1), F(1)	L(1), LS(1)	UL(1), LL(1)
Bayri et al, 2014 ³	4	M(2), F(1), NR(1)	T(1), TL(1), LS(1), S(1)	UL(1), LL(3)
This study, 2005-2016 ^{1,6,9,16,17,20,22,24,30}	11 (including our)	M(5), F(6)	T(4), TL(2), L(1), LS(4)	UL(4), LL(6), Breast(1)
Total	52	F(31), M(14), NR(7)	C(5), T(17), TL(7), L(8), LS(13), S(2)	(18) UL, (33)LL, Breast(1)

M: Male; F: Female; NR: not reported; C: Cervical; T: Thoracic; TL: Thoracolumbar; L: Lumbar; LS: Lumbosacral; S: Sacral; UL: upper limb; LL: lower limb

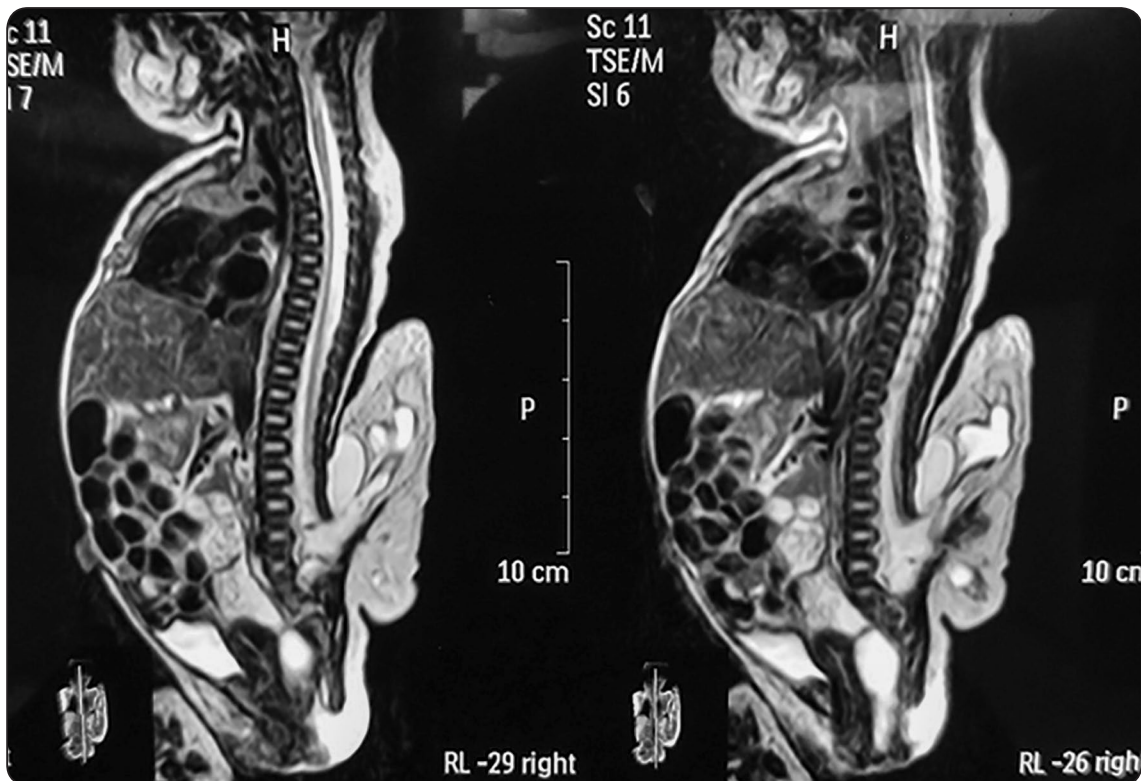


Figure 1. Pre-Operative MRI T2 Midsagittal Cuts, showing the lumbosacral attachment of the parasitic twin with the autosite, with a low-lying conus with a lumbosacral meningocele herniating in the parasite.



Figure 2. (A,B): photograph showing our lumbar parasitic Rachipagus twin pre operatively. (C,D): photograph showing the girl one week post operatively.

Discussion

Most scientists agreed that conjoint twin (CT) originate from monozygotic twin (MZ). The partition of the embryonic axis into two parallel ones, gives origin to the monoamniotic monochorionic type of placentation of MZ twins which is characteristic of CT. Currently, it is accepted that CT originate from a failure in the development of primitive structures at later stages of development, that is, Carnegie stage 6 (days 12–15), or the primitive streak stage of human development.¹¹ However, the exact mechanisms of CT remain obscure.

Up till now, there is no definite theoretical explanation of conjoined twinning. Although, there are many theories, two opposing theories have been suggested to explain the sequence of events of CT “Fission versus Fusion”. Spencer proposed the Fusion Spherical Theory, it is proposed here that two early monovular embryonic discs may lie adjacent to one another at various angles and planes, as though floating on the surface of one sphere (the yolk sac) or on the inside of another (the amniotic cavity), and may become secondarily united rostrally, caudally, laterally or dorsally, symmetrically or asymmetrically, but always homologously.²⁸ In contrast, supporters of the fission theory suggest

that incomplete fission of the blastocyst inner cell mass during the primitive streak stage, 13 to 15 days post fertilization, results in 2 centers of axial growth that retain a connection at some point.¹⁵

There is more than one hypothesis tried to explain the etiopathogenesis of asymmetrical (parasitic) dorsally attached conjoined twins. For group of authors rather than conjoined, it is considered parasitic because it is incompletely formed and wholly dependent on the body functions of the complete fetus. It results from embryonic death of one twin, leaving various portions of the body vascularized by the surviving autosite. It is postulated that the dorsally united twins are joined in some (rachipagus in the mid-portion of the neural tube) portion of the neural tube before the neural folds have completely closed. Neither the abdomen nor the umbilicus is ever involved in the union. The union in all of these twins involves the appropriate portion of the osseous cranio-vertebral axis, the dura, and not infrequently, the neural tube itself.^{26,28}

On the other hand, the authors considering the condition as an accessory limb, believe in the hypotheses of Gardner⁷ and Egar⁵ that lipomyelomeningoceles are secondary neural-tube defects that occur due to rupture of the neural tube under intact ectoderm. The leakage of the proteinaceous neural tube fluid acts as an abundant source of Schwann cells, which collect under the skin and de-differentiate. The majority of de-differentiated cells develop into a lipomatous mass that may extend intradurally. Sometimes cartilage, bone, muscles, and nerves may also develop in this lipomatous mass. On rare occasions, these components may grow in an organized manner and develop into an accessory limb. The stimulus for this organized growth remains uncertain.

First case reported on the literature dated back to 1889, by Jones and Larkin.¹⁰ Up till 1996, there wasn't standardized nomenclature of this condition such as that presented by Spencer.²⁵ Where, she unified the system of nomenclature and classified conjoined twins into 8 types based on the theoretical site of union. She also differentiated the parasitic conjoined twins from other conditions that are differentially diagnosed, fetus in fetu and teratoma. A fetus in fetu is a fetus or fetus-like structure with a vertebral axis seen in the body of its twin. Teratoma is an accumulation of pluripotential cells.²⁶

However, up till now in the literature review, we found this topic is presented with different key words. The most commonly used was Rachipagus,^{4,19,22,25,27} dysraphic appendages and accessory limb.^{3,12} Every group adhere to his terminology, as it comes from his theoretical explanation of the embryology of the condition. But after research we found that Spencer classification was devised to facilitate the identification of the pertinent anatomy in these patients, and will result in more accurate identification of conjoined twins, an increase in the value of future publications, and less confusion for the reviewer.

From our search using Pubmed (including Ovid, Springer, and Science direct publication) and Google scholar we found that there are some cases missed and others added. In this report we tried to collect all the cases from previous reviews and we added the cases that wasn't mentioned (either new or old) in addition to our case (Table 1). We used key words Rachipagus, accessory limb, and parasitic twins.

Spencer in 1995,²⁷ made the first literature review concerning rachipagus parasitic twin, 20 dorsally attached parasitic twins were reevaluated and reclassified as rachipagus.

Two had extensive vertebral fusion and 18 were united in the dorsal midline, all with a meningocele, vertebral anomaly, and/or bony or neural connection. Later in her wider review on parasitic twins, she added four more cases.²⁶ Krishna et al,¹² presented his own experience with four cases. They added a review of more five cases in the twenty century. Eight were females, and all have one or more neural tube lesion. Chadha et al,⁴ added two more female cases (including his case) and made a more broad literature review including the above studies. Sharma et al,²¹ in his review of the wide array of heteropagus twins, he mentioned Chadha and Ratan cases, and added two more cases. One was male and the other was a female. Bayri et al,³ with presenting his male case report, he made a limited review on the cases as accessory limbs and added three more cases one male, one female and one not reported.

In our review on all cases, whatever the title, we found 11 more cases including ours. Three of these cases were the first to be mentioned from Africa. Five were males and six were females.^{1,9,17,20,22,24,30} From the analysis of the data above, most of the authors believe that the condition results form a MZ twins. However, Krishna and Lal¹² in his review found one female autosite with a rudimentary scrotal tissue. But, he didn't confirmed this with chromosomal analysis. Also, Lorongo et al,¹³ in his case report, found that Fluorescent in situ hybridization (FISH) studies suggested that the parasitic twin was male, and DNA typing studies demonstrated dizygosity. But this was point of debate between him and Machin.¹⁴

Those who believe in the hypothesis of fusion, found that parasitic twins is caused by death of the embryo with no heart formation and depending mainly on the autosite for vascular supply of the organs remains. This

was confirmed when Hirokazu et al,⁸ in a case of pygopagus, although there is a complete attached twin, but because the heart was not working, the twin died with cyanosis upper part, while the viable lower part is supplied from the autosite by a sizable artery confirmed by angiography.

On the other hand, from the beginning those who accepted Humphrey hypothesis, there is no heart or brain tissue formation. But how come, there are cases reported with two completely formed twins attached at the back,²⁷ and not only limb differentiation? Also, the presence of colon-like tissue in one case¹² and primitive heart like saccules in another.²⁹ They say these cases may represent an extreme degree of de-differentiate of the blastemal cells, but this is more baffling.

Although the ICBDSR epidemiological study found male predominance in parasitic twins¹⁵, and Sharma et al,²¹ in his review stated that the sex difference is less pronounced in heteropagus twins. In our review on rachipagus parasitic twins, there is significant female predominance. 31 cases were females (59.6%), while 14 were males (26.9%), and seven cases (13.4%) the sex was not mentioned or determined.

According to the site of attachment, there was 17 cases attached at the thoracic spine (whether upper or lower), and the least were the cervical with just four cases. There is only one case with two parasite at different levels, with a fully formed hind limb attached dorsally at lumbar region with a small soft mass covered with scrotal skin, and an under developed upper limb was attached dorsally at the thoracic region.¹⁷

Most parasites consisted only of supernumerary limbs, except one case the parasite was formed of a well formed breast and scapula attached to the lower dorsal spine.³⁰

The parasites located in the rostral portion of the vertebral column usually have upper limbs and those found in the caudal portion have lower limbs. There was 33 cases (63.5%) having lower limb, and 18 cases (36%) upper limb.

In all cases there is a bony defect in the autosite spine at the site of attachment of the parasitic twin. Also, it is anticipated that, in parasitic rachipagus twins, the commonest abnormalities in the autosite involve the central nervous system (CNS) and the vertebral column, usually at or close to the site of union. Authors explained these findings as resulting from the on sequences of fusion of the neural folds and the spinal column of the parasitic twin and the autosite, where 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.^{1,19,27}

From the review of all previously published cases, there is an overall positive results that can be obtained in treating this condition and that the presence and degree of cardiac involvement have a major influence on the prognosis. To achieve best results from your surgery, most surgeons who face a case of rachipagus twins, recommended: (1) recognize to what extent the conjoinment of neural structures exist by preoperative imaging examinations including CT, MRI; (2) in cases of spinal cord involvement neurophysiological monitoring is mandatory, to avoid or minimise post-operative neurological deficit; (3) identification of the vascular supply of the parasitic twin (if sizable) and closure with piecemeal haemostasis; (4) the dura is repaired and closed water tight using autogenous graft from the fascia, followed by pediculated muscular flab covering the dura to prevent postoperative CSF leak.^{8,18}

Conclusion

Rachipagus parasitic twin is a rare form of conjoined twin with a favorable outcome. Obstetrician should be aware of the existence of a parasitic twin during prenatal examinations. Detailed perioperative evaluation and management planning, as well as multidisciplinary microvascular approach, are integral to perform the best separation surgery. Avoid traction during separation. MEP is a must if available.

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الملخص العربي

نجاح فصل توأم متطفل متصل بالعمود الفقري الصدري القطني: تقرير حاله

البيانات الخلفية: التوائم الطفيلية المتصلة بالعمود الفقري القطني العجزي حالة نادرة للغاية. هذه هي المحاولة الأولى في الأدب لمحاكمة فصل التوائم الطفيلية المتصلة بالعمود الفقري القطني العجزي في حديثي الولادة مع الحبل الشوكي فتق من خلال ثقب خلفي في الفقرات لقطنية الي راخل التوأم الطفيلي.

الغرض: والهدف من هذا التقرير هو تقديم حالة نادرة جدا تم فيها الفصل الجراحي بنجاح، ولا يزال الطفل حيا وبصحة جيدة.

تصميم الدراسة: دراسته لحالات اكلينيكيه لحاله نادرة و مراجعه للدراسات العلميه

المرضى و الطرق: وكان الطفل عمره ٢٨ يوما عند إجراء الجراحة. كان عمر المريض، والتخدير، والنزيف أثناء العملية، والتصاق أي الأنسجة العصبية، عوامل خطر كبير أثناء الجراحة. تم عمل الاشعة المقطعية، والتصوير بالرنين المغناطيسي على العمود الفقري كله مع التركيز على العمود الفقري القطني العجزي لتوفير المعلومات اللازمة لإجراء عملية جراحية. من خلال التقليل من وقت الجراحة، وذلك باستخدام تقنية المجهرية والتحكم الكافي و الحد من النزيف أثناء العملية، تم تحقيق الفصل الجراحي الناجح. كانت مدة الجراحة ساعتين ونصف. كان تشريح الأنسجة العصبية المشكلة الأكثر خطورة، حيث لم يكن هناك جهاز كهروفيسيولوجي لمتابعة الاعصاب المتاحة لهذا العصر، ولكن التوأم الطفيلي لم يكن متصل بالاعصاب. كان النزيف هو المشكلة الثانية الخطيرة، تم نقل ١٢٠ سم من الدم للطفل. كان المصبر الرئيسي للشريان الطفيلي عن طريق شرايين صغيرة من الرضيع. بعد فصل الساقين الطفيلية، تم إصلاح الام الجافية باستخدام انسجة ذاتية المنشأ. تم إغلاق الجلد والأنسجة تحت الجلد مباشرة.

النتائج: تم بنجاح فصل التوأم الطفيلي مع ترقيع الام الجافية، خرج المريض من المستشفى بعد ٤ أيام، تتحرك أطرافها السفلية بشكل طبيعي مع ضعف في كلا القدمين.

الاستنتاج: التوأم الطفيلي الملتصق بالعمود الفقري هو شكل نادر من التوأم الملتصقة مع نتائج جراحية جيدة. التقييم المفصل حول الجراحة والتخطيط لها ، فضلا عن نهج الأوعية الدموية بدقة شديدة، هي جزء لا يتجزأ من أداء أفضل لجراحة الانفصال.