

Case Report

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A Parasitic Twin Diagnosed Prenatally As a Sacrococcygeal Teratoma: A Case Report

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ABSTRACT

Parasitic twinning is a rare entity. It is an asymmetric conjoined twinning in which a poorly formed twin, known as the parasite, is attached to a fully developed co-twin, known as the autosite. The parasite depends on the autosite for blood supply. It is diagnosed prenatally with ultrasound, and treatment involves resection of the parasite postnatally. We describe a case that was prenatally diagnosed on 2D ultrasound as a sacrococcygeal teratoma at 25 weeks. No other anomaly was detected. Caesarean delivery was conducted at 34 weeks on account of failure to progress following spontaneous preterm labour. At delivery, we found that the sacrococcygeal mass was a parasitic twin with a poorly formed body, lower limb, and hair. These fetal components were not demonstrated on the prenatal 2D ultrasound. Postnatal assessment revealed no other abnormality. The mass was successfully resected on day 3 of life. The resection was done early on account of ulceration of the mass/ parasite. The histopathological assessment revealed more fetal parts, including brain tissue and lungs. Prenatal diagnosis is essential as it identifies associated anomalies and helps with prognostication and planning of obstetric and surgical management by a multidisciplinary team.

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Introduction

A parasitic twin is a type of twin in which tissues of an incompletely formed twin, known as the parasite, are attached to and dependent on a fully developed co-twin, known as the autosite [1]. It is a rare type of conjoined twins with an incidence of 1-2 per million live births [2]. Reports in the literature have described this with various terminologies, including spinal harmatoma, mature teratoma, rudimentary parasitic twin (heteropagus), and disorganization-like syndrome [3]. The parasite is typically linked to the autosite by a soft tissue pedicle containing large blood vessels and has fetal components that are remarkably recognizable [1]. The union of the twins can occur at various sites. It is known as rachipagus when spinal fusion takes place above the sacrum.

It is virtually entirely found in developing countries, according to geographic distribution [4]. It is unclear exactly what causes parasitic twins. Its geographic clustering may indicate the influence of the environment or genetics. Rachipagus parasites are strongly linked to neural tube defects. A review by Zewdie et al discovered that 35 out of 37 cases had an autosite with neural tube abnormality of some kind [4].

Prenatal diagnosis is important because early detection of anomalies with poor prognosis affords the mother a chance for termination of pregnancy or aim for delivery at a tertiary facility. Prenatal diagnosis is better achieved with a three-dimensional (3D) ultrasound. In a report by Terata et al, a two-dimensional (2D)

ultrasound of a fetus at 28 weeks demonstrated omphalocele, ascites, hydrothorax, and congenital cardiac anomaly. A 3D ultrasound of the same fetus revealed, in addition, a distinct extrafetal body with two lower limbs joined to the fetal chest [5].

Since there is no organ sharing, it is usually possible to separate parasitic twins with simple resection. On the other hand, associated anomalies could complicate resection and repair [4]. In contrast to symmetric conjoined twins, the prognosis of parasitic twins may be generally good due to fewer extensive vascular and visceral connections between the autosite and the parasite. However, unsurprisingly, congenital cardiac defects in the autosite are associated with a worse prognosis [5].

Case Report

A 40-year-old G4P2+1 had a routine obstetric ultrasound at 25 weeks gestation that incidentally identified a sacrococcygeal mass. She was referred to the Maternal-Fetal Medicine Unit of the Korle Bu Teaching Hospital, Ghana, for further assessment. A 2D ultrasound assessment at our unit demonstrated a live female fetus at 25w5d with a solid sacrococcygeal mass with a cystic extension (104mls and 75mls, respectively) [Figure 1 (a) and (b)], with only the solid component demonstrating flow on colour doppler. No other gross anomaly was demonstrated in the fetal anatomy survey. The fetal cardiac assessment was satisfactory, with a normal cardiothoracic ratio. There was no evidence of fetal hydrops, and the middle cerebral artery peak systolic velocity was 1.18 MoM. Fetal biometry yielded an estimated fetal weight on the 26th centile.

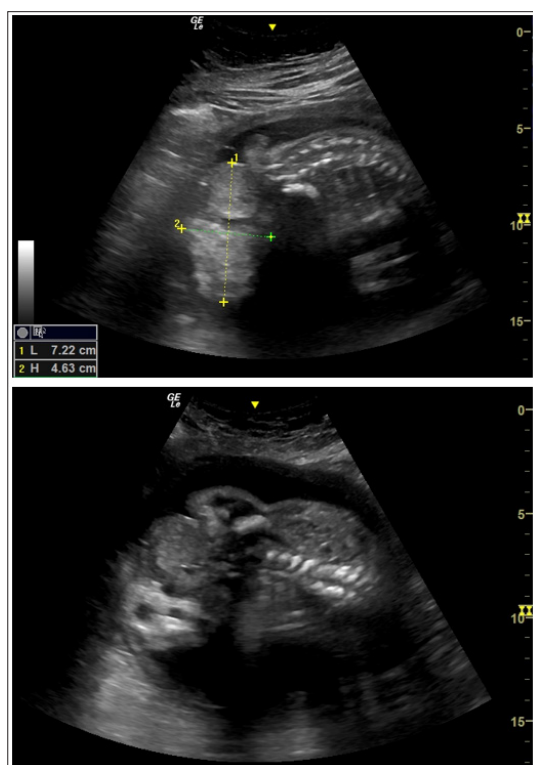


Figure 1: (a) sagittal view and (b) coronal view of the solid component of sacrococcygeal mass at 25w5d.

She did an oral glucose tolerance test (OGTT) at 25 weeks, which diagnosed diabetes in pregnancy and was managed on metformin and mixtard insulin. The pregnancy was with a new partner and was unplanned. Her first two pregnancies were uneventful, and both children are doing well. She has a history of twinning in her family, but no history of congenital anomalies. She does not take alcohol or recreational drugs. She was not on any medication before the pregnancy.

A follow-up scan at 31 weeks demonstrated mild polyhydramnios and an increase in the size of the sacrococcygeal mass. The solid and cystic components measured 183.24ml (75% increase) and 147.32ml (97% increase). She presented with spontaneous preterm labour at 34 weeks and had an emergency caesarean section because of failure to progress. She delivered a live female with a birth weight of 3445g and Apgar scores of 8/10 and 9/10 at 1 and 5 minutes, respectively.

The baby had a sacrococcygeal mass with a poorly formed body, lower limb, and hair [Figures 3 and 4]. There were no other dysmorphic features besides the sacrococcygeal mass and no oedema. Cardiovascular and respiratory system examination was normal. She was diagnosed as a parasitic twin. The differential diagnosis was sacrococcygeal teratoma with mature components. A postnatal ultrasound scan revealed no deep extension of the sacrococcygeal mass and no involvement of pelvic organs. On day 3 of life, she had surgical excision of the parasitic twin [Figures 5 and 6], which had become ulcerated. The procedure was uneventful. Her weight after surgery was 2100g. Histopathology assessment revealed a parasitic twin weighing 950g with a focus of ulceration measuring 80mm in diameter. Identifiable structures included a foot, leg, and hair. In addition, the cut surface showed a cystic area filled with soft tan polypoid tissue (brain) with yellow and tan spongy areas (lungs) as well as adipose and bony tissue [Figure 7]. On microscopy, sections of the brain showed islands

of neuroblasts within the background of the neuropil. Sections of the lungs showed alveoli with thick interalveolar septa. Sections of adipose tissue showed mature adipocytes with histologically normal skin covering. No malignancy was seen.



Figure 2: Cystic Component of Sacrococcygeal Mass At 25w5d



Figure 3: Sacrococcygeal Mass with A Lower Limb and Large Cyst with A Thick Wall



Figure 4: Sacrococcygeal Mass with A Large Cyst, Lower Limb, Hair, And Mouth



Figure 5: Bagged Sacrococcygeal Mass Excised



Figure 6: Repair After Excision of the Sacrococcygeal Mass

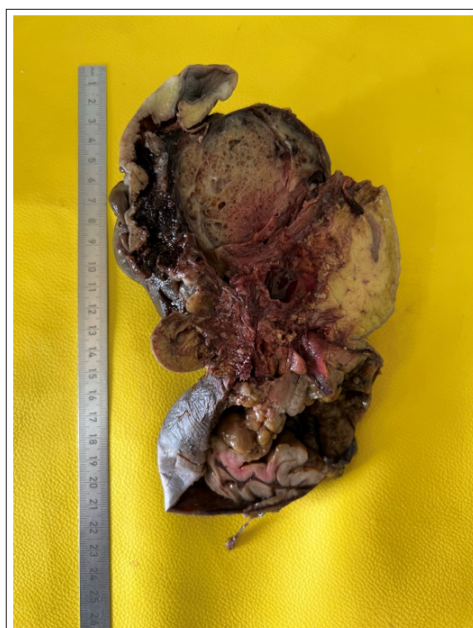


Figure 7: Parasitic Twin Measuring 170 X 130 X 110mm With Cut Surface Demonstrating Brain Matter (Blue Arrow) In the Cystic Extension as Well as Lung Tissue (Red Arrow)

Discussion

Parasitic or heteropagus twinning is rare, and most published data are in the form of case reports and a few case series. There are various hypotheses regarding pathogenesis. The “fission theory” suggests a late unequal incomplete division of the embryo resulting in conjoined twins [6]. On the other hand, the “fusion theory” proposes that at the third or fourth gestational week, the neural folds of the two different embryos in mono chorionic monoamniotic twins can merge if the skin covering the neural tube gets damaged [7]. Another proposed theory is “faulty secondary neurulation”, where an overproduction of neural tube fluid can leak into subcutaneous tissue and differentiate into various tissue types [1]. The latter theory only explains rachipagus (joined dorsally and facing away from each other) parasitic twinning and is usually associated with spinal cord anomalies. Koen postulated that a disruption in the differentiation of pluripotent caudal cell mass might be responsible [8]. In heteropagus twinning, vascular insult in the parasite leads to selective ischaemia of tissues and dependence on collateral blood supply from the autosite. This accounts for the asymmetry between the two [3]. Lorgono et al. have demonstrated potential allelic differences between heteropagus twins, suggesting that they could originate from dizygotic twins with subsequent fusion [3]. In addition, Dejene et al. reported two cases where the female autosites carried parasites with phalluses but no scrotum. This corroborates the possibility of dizygosity in parasitic twinning. However, they could not confirm dizygosity with DNA testing [1].

A differential is a teratoma, which is a true tumour arising from the uncontrolled growth of pluripotent stem cells. Teratoma may contain mature tissues such as respiratory epithelium, fat tissue, hair follicles, fingers, teeth, and jawbones. However, these bones are scattered throughout the tumour and never resemble the real limb [7]. Most reported cases are diagnosed postnatally. The role of antenatal diagnosis is crucial as it identifies associated anomalies, which aids with prognostication and planning of obstetric and surgical management by a multidisciplinary team. The literature shows that 3D ultrasound is superior to 2D in its diagnosis [5]. This may explain why we did not pick all the components on our 2D ultrasound. Some cases report much later in life due to poor access to healthcare and socio-cultural factors. Gocken and Washimo reported on a girl who lived with a heteropagus twin in Ethiopia for 17 years before seeking healthcare. She had a successful surgical excision [9].

Most cases are delivered vaginally. Caesarean delivery is reserved for obstetric indications or in cases where the parasitic twin is large and likely to cause obstructed labour. In the case series by Dejene et al., four out of five cases were delivered vaginally [1]. Although any structure can theoretically be found in the heteropagus twin, limbs are the most frequent. Zewdie et al. reviewed 4 cases and 25 reports on rachipagus heteropagus twins. The most common contents of the parasites were limbs and phallus. The most common anomalies in the autosites were neural tube defects [4]. In our case, the most well-defined content of the parasite was the lower limb. A thorough workup before surgery to identify associated anomalies in the autosite is very important since this impacts prognosis.

Since organs are not usually shared, preoperative imaging is usually restricted to ultrasound, computerized tomography (CT), or magnetic resonance imaging (MRI). Angiography is not required. Echocardiography is recommended [1]. Our case had a postnatal ultrasound scan that ruled out deep extension and pelvic organ involvement.

Management involves surgical excision. Surgery is generally simple and uncomplicated because there is no sharing of organs. In most of the cases in the study by Zewdie et al., the separation was uneventful [4]. In our case, the surgical excision was uneventful as well. Aseptic handling of the parasite during the procedure is crucial to minimize sepsis. In our case, the parasite or sacrococcygeal mass was covered in a sterile bag during the surgery. Post-op complications include recurrence of teratoma at the excision site and incisional hernia [3].

Regarding the timing of separation, delayed surgery is generally favoured except in cases where the parasite is infected or is large enough to cause haemodynamic compromise of the autosite. Bansal et al. recommend surgery be delayed until 6-12 months, since the prognosis is better with physical maturity. Surgery done in the neonatal period carries a mortality rate of 50% [10]. Zewdie et al. also recommend delaying surgery till three months of age to avoid anaesthesia-related complications [4]. Dejene et al. had three out of five cases operated on in the neonatal period, two of which were because of an infection of the parasite. All three had a favourable overall outcome. However, they proposed that surgery be undertaken sooner as the parasite could have untoward effects on respiration and the growth of the autosite, as well as a cosmetic and psychosocial burden [1]. In our case, the surgery was done on day 3 of life because the parasite got ulcerated.

Prognosis is generally good as fewer visceral and vascular structures are shared between the two, compared to symmetrical conjoined twins. Following surgery, the mortality rate for the autosite is 31%. The most typical causes of death are cardiorespiratory failure secondary to congenital heart disease and sepsis [3]. Pre- and post-operative multidisciplinary input between neonatologists, intensivists, and paediatric surgeons is recommended for best outcomes.

Conclusion

Parasitic twinning is very rare. Prenatal diagnosis is crucial and is best done with 3D ultrasound. Some contents of the parasite are not readily discernible on 2D ultrasound. We found a sacrococcygeal mass with solid and cystic components on 2D ultrasound examination at 25 weeks gestation, but could not demonstrate any distinguishable fetal parts. Sacrococcygeal teratoma is a close differential. However, the fetal parts, including a foot, leg, hair, brain, and lungs, revealed on gross and histologic examination following excision postnatally confirmed a parasitic twin rather than a teratoma. We diagnosed our case prenatally, which gave us ample time to plan the delivery and subsequent surgical resection with a multidisciplinary team. The resection was done as early as day 3 of life because the parasite got ulcerated. The procedure was uneventful.

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