

Association Of Laparoschisis And Sacrococcygeal Teratoma In A Twin Pregnancy: A Case Report

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Abstract:

Laparoschisis is a rare congenital defect characterized by the evisceration of abdominal viscera through a defect in the anterior abdominal wall, usually to the right of the umbilical cord insertion. It is often isolated but can occasionally be associated with other congenital anomalies. Sacrococcygeal teratomas are common extragonadal tumors arising from pluripotent germ cells. Their co-occurrence in a twin pregnancy is exceedingly rare. We present the first known case of a twin pregnancy complicated by both laparoschisis and a sacrococcygeal teratoma. At 22 weeks of gestation, ultrasound revealed laparoschisis in both fetuses, with evisceration of the intestines, liver, and stomach. One fetus also had a large solid cystic mass in the caudal region, suggestive of a sacrococcygeal teratoma. The pregnancy was complicated by preterm labor at 30 weeks, resulting in the vaginal delivery of two premature neonates who died within two hours due to multivisceral failure. This rare case highlights the challenges in managing twin pregnancies with multiple congenital anomalies. The combination of laparoschisis and sacrococcygeal teratoma complicates prenatal care, delivery, and postnatal outcomes. Despite early diagnosis, the presence of severe visceral involvement and preterm delivery resulted in a poor neonatal outcome. The association of laparoschisis and sacrococcygeal teratoma in a twin pregnancy presents unique clinical challenges. Prenatal imaging and multidisciplinary management are crucial to optimizing outcomes, although complex congenital anomalies often result in poor neonatal prognosis. Further research is needed to better understand the implications of this rare association.

Keywords: Laparoschisis, sacrococcygeal teratoma, twin pregnancy, congenital anomalies, prenatal diagnosis, preterm labor, neonatal Outcome, multivisceral Failure, abdominal wall defect, teratoma

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I. Introduction

Laparoschisis is a rare parietal anomaly associated with median coelomic defects, characterized by the evisceration of abdominal viscera at the right lateral-umbilical level. It should be differentiated from omphalocele or anterior coelomic defects, where the viscera are contained in a sac over which the umbilical cord is implanted [1, 2, 3]. The estimated prevalence of laparoschisis is 2.5 per 10,000 live births (ANDERSON, 2018), and while the prognosis is generally favorable, it is often influenced by ischemic lesions of the digestive tract that can complicate immediate post-birth management. These ischemic lesions are typically the result of the disruption of blood flow to the eviscerated organs, and therapeutic intervention may be limited in some cases.

Currently, no specific genetic cause has been identified for laparoschisis, apart from its association with certain syndromic conditions. Gastroschisis, which is a similar defect, is rarely associated with other congenital anomalies. However, recent epidemiological studies have shown that at least one associated malformation is present in 32% of cases, which may include: small bowel or colon atresia (in which the term "complex gastroschisis" is used), biliary tract anomalies, hydrocephalus, cryptorchidism.

In this report, we present the first known case of laparoschisis associated with a sacrococcygeal teratoma in a twin pregnancy.

II. Clinical Observation

We report a rare case involving a multiparous woman with an uncomplicated medical history, who was under follow-up for a twin pregnancy at our CHU Hassan II Hospital. At 22 weeks of gestation, obstetric ultrasound revealed laparoschisis in both fetuses. The ultrasound findings showed a defect in the abdominal wall, with evisceration of the digestive tract, liver, and stomach. Additionally, a solid cystic mass measuring 12 × 10

cm was identified in the caudal region of one of the fetuses, initially raising the suspicion of a sacrococcygeal teratoma.

At 30 weeks' gestation, the patient presented with rupture of the amniotic sac and experienced abdominal-pelvic pain, which was thought to be due to uterine contractions. Unfortunately, the pregnancy progressed with complications, and two premature neonates were delivered vaginally. Both infants succumbed to multivisceral failure within two hours of birth.

The prenatal diagnosis of both conditions was made through detailed obstetric ultrasound, which is crucial in the management of complex pregnancies like this one. Sacrococcygeal teratomas are often diagnosed antenatally, and their size and vascularity can be assessed using Doppler imaging, which helps in determining the risk of fetal hydrops or other complications. Prenatal diagnosis of laparoschisis is typically made in the second trimester, with the defect being visualized as a gap in the abdominal wall through which the fetal intestines protrude. It is essential to distinguish laparoschisis from other abdominal wall defects, such as omphalocele, as the management of these conditions can differ significantly.



III. Discussion

The association of laparoschisis and sacrococcygeal teratoma in a twin pregnancy is an extremely rare occurrence. While laparoschisis is a relatively well-known congenital defect, characterized by the lateral evisceration of the abdominal viscera, it is typically isolated and does not often present with additional complex malformations. The occurrence of a sacrococcygeal teratoma in the same pregnancy further complicates the clinical picture, as both conditions carry substantial risks to neonatal survival, especially in the context of preterm delivery, as seen in our case.

Laparoschisis is characterized by the herniation of abdominal contents through a defect in the anterior abdominal wall, usually to the right of the umbilical cord insertion. It is differentiated from omphalocele, where the eviscerated viscera are encased in a membranous sac. The prognosis of laparoschisis is often influenced by the degree of visceral involvement, particularly when the intestines, liver, or stomach are exposed. The associated ischemic damage, as a result of impaired blood supply to the herniated organs, can lead to significant morbidity if not managed promptly. Recent literature suggests that laparoschisis can be linked to other congenital anomalies, but the incidence of such associations is relatively low. Sacrococcygeal teratomas are among the most common congenital tumors and are typically located in the sacrococcygeal region. These tumors arise from pluripotent germ cells and contain a variety of tissues, including hair, muscle, and fat. In our case, the presence of a large cystic mass, measuring 12 × 10 cm, located in the Caudal region of the fetus, was initially suggestive of a sacrococcygeal teratoma.



The combination of laparoschisis and sacrococcygeal teratoma in a twin pregnancy represents a highly challenging clinical scenario. In our patient, both fetuses exhibited laparoschisis, which complicated the pregnancy due to the risk of preterm labor and other obstetric complications. The rupture of the amniotic sac at 30 weeks of gestation, accompanied by uterine contractions, further stressed the need for urgent and coordinated care. Preterm delivery in such cases is often associated with poor outcomes, particularly when there are additional anomalies such as sacrococcygeal teratomas and significant visceral involvement.



Several studies have reported the challenges associated with the management of laparoschisis and other abdominal wall defects. In most cases, laparoschisis is surgically corrected soon after birth. However, the outcome can be influenced by factors such as the degree of ischemia, the presence of other congenital anomalies, and the timing of surgical intervention. In our case, the presence of a sacrococcygeal teratoma likely contributed to the rapid deterioration of the twins after birth. The large size of the teratoma could have interfered with fetal development, leading to vascular compromise and potentially contributing to the multivisceral failure observed in the neonates.

Sacrococcygeal teratomas are graded based on their size and the presence of vascular involvement, which can influence the prognosis. Large teratomas, especially those with significant blood flow, pose a risk for fetal hydrops (the accumulation of fluid in fetal compartments), heart failure, and other complications. Although prenatal surgical intervention is sometimes considered for large or complicated sacrococcygeal teratomas, the decision is based on a number of factors, including fetal maturity, tumor size, and the presence of other malformations. In our case, despite the early detection of both anomalies, the combination of preterm birth and multivisceral involvement was overwhelming, and the infants unfortunately did not survive.

The rarity of this case emphasizes the need for a thorough and multidisciplinary approach to managing pregnancies complicated by multiple congenital anomalies. Prenatal counseling is crucial, as families need to be informed about the potential outcomes and the possible need for intensive neonatal care. Early referral to specialized centers with expertise in managing complex congenital anomalies, including both laparoschisis and sacrococcygeal teratomas, is essential for optimizing outcomes. Although laparoschisis and sacrococcygeal teratomas are rare in isolation, their combined occurrence in a twin pregnancy, as in this case, presents unique challenges for both obstetricians and neonatologists.

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