

An Unusual Complication of Twin-to-Twin Transfusion Syndrome: Case of Intrauterine Limb Gangrene

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ABSTRACT

Introduction: Twin-to-twin transfusion syndrome (TTTS) results from abnormal vascular anastomoses within the placenta, creating an imbalance in blood flow between the twins. TTTS only occurs in monozygotic twins with a monochorionic placenta. Twenty percent of all twin pregnancies are monochorionic, and TTTS affects up to 17% of monochorionic twins. The incidence of thrombosis in one of the twins is five percent.

Case report: Here, we report a case of intrauterine limb gangrene secondary to TTTS. A preterm twins at 33 weeks of gestation were born to a second gravida mother. Antenatal ultrasonography showed monochorionic diamniotic placenta with polyhydramnios in one sac and oligohydramnios in the other. Twin A, which was the recipient twin, had a birth weight of 1.782 kg and was polycythemic (Hb- 26 gm/dl and HCT- 89%). Twin B, which was a donor twin, had a birth weight of 1.502 kg and was anemic (Hb- 9.0 gm/dl and HCT- 27%). Twin A, on examination, had bluish discoloration and absent peripheral pulses in the left upper limb (radial and ulnar arteries) and lower limb (popliteal, posterior tibial, dorsalis pedis arteries). Ultrasound Doppler revealed a thrombus occluding the left radial, ulnar, and femoral artery). A partial exchange transfusion was done for Twin A and was started on low molecular weight Heparin. However, twin A had a stormy course and succumbed to disseminated intravascular coagulation and intraventricular hemorrhage. Twin B was transfused packed cells for anemia and was subsequently discharged.

Discussion: Intrauterine thromboembolic phenomena, gangrene, and peripheral limb ischemia in the recipient twin are very rare complications of TTTS. Various causes of intrauterine gangrene have been described, the commonest being polycythemia leading to thrombosis in the recipient twin, which was evident in our case. Other causes include thromboembolic phenomenon due to thrombosis of placental vessels, as a complication of laser photocoagulation done antenatally, or as a result of umbilical artery steal syndrome.

Conclusion: Antenatally diagnosed monochorionic twin gestation should be evaluated closely for TTTS and complications like heart failure, thromboembolic phenomena, and skin necrosis or ischemia. These complications can be prevented by in-utero interventions such as amnioreduction, fetoscopic laser photocoagulation, and septostomy..

1. INTRODUCTION

Twin-to-twin transfusion syndrome (TTTS) is a serious complication that arises exclusively in monochorionic twin pregnancies due to vascular anastomoses that create an imbalanced blood flow between the twins [1]. This syndrome is reported to occur in approximately 10–17% of monochorionic diamniotic pregnancies [2], accounting for significant morbidity and mortality if left untreated [3]. The recipient twin is at risk of polycythemia, hypervolemia, and cardiac overload, while the donor twin often suffers from anemia and hypovolemia [4]. The vascular imbalance can cause a range of complications, including intrauterine growth restriction (IUGR), cardiac dysfunction, and, in rare cases, thromboembolic events [5]. Intrauterine thrombosis in the recipient twin has an estimated incidence of about 5% and can lead to peripheral ischemia or even gangrene [6]. Although cases of cerebral thromboembolic events in TTTS are documented, reports of intrauterine limb gangrene are exceedingly rare and often signal an advanced disease stage or critical hemodynamic disturbance [7].

Case Report

Here, we present a case of early preterm (33 weeks) monochorionic diamniotic twins, where antenatal ultrasonography revealed polyhydramnios in one sac and oligohydramnios in the other, indicative of TTTS. A cesarean section was performed, delivering Twin A (recipient) and Twin B (donor). Twin A weighed 1.782 kg and was polycythemic with a hemoglobin level of 26 g/dL and hematocrit of 89%. Twin B weighed 1.502 kg and was markedly anemic with a hemoglobin of 9 g/dL and hematocrit of 27%. Upon examination, Twin A displayed bluish discoloration, as seen in Figure 1, and absent peripheral pulses in the left upper and lower limbs. Doppler ultrasound confirmed thrombosis in the left radial, ulnar, and femoral arteries. Twin A was managed with partial exchange transfusion and initiated on low molecular weight heparin (LMWH). Despite aggressive management, he developed disseminated intravascular coagulation (DIC) and intraventricular hemorrhage (IVH) and succumbed on the second postnatal day. Twin B received packed red cell transfusion and was discharged in stable condition after 2 weeks of NICU care.



Figure 1: Discoloration of upper and lower limbs of the recipient twin.

2. DISCUSSION

Twin-to-twin transfusion syndrome (TTTS) remains a significant obstetric complication in monochorionic twin pregnancies, with a spectrum of consequences ranging from mild imbalances to severe, life-threatening outcomes. The hallmark of TTTS is the abnormal placental anastomoses that result in unbalanced blood flow between the fetuses, leading to conditions such as polycythemia in the recipient twin and anemia in the donor twin [1]. The development of thromboembolic complications in TTTS, including intrauterine limb gangrene, although rare, highlights the critical nature of this syndrome and its potential for severe morbidity.

The incidence of thrombosis in the recipient twin has been reported to be around 5%, often manifesting as peripheral ischemia or, in more severe cases, gangrene [6]. The case presented here underscores the severe manifestation of TTTS in the form of intrauterine limb gangrene, a condition typically seen in advanced cases of the syndrome. Polycythemia in the recipient twin contributes to increased blood viscosity and subsequent thrombotic events. In our case, Twin A, the recipient twin, developed thrombosis in the left radial, ulnar, and femoral arteries, leading to ischemia and gangrene of the left upper and lower limbs. The pathophysiology of such thromboembolic phenomena is often linked to the hyperviscosity state created by polycythemia, which predisposes to thrombosis, further compounded by the imbalance in blood flow that defines TTTS [4].

Intrauterine gangrene in TTTS is an uncommon, but severe, complication, as it represents a critical stage of the disease, often associated with significant hemodynamic instability in the recipient twin [5]. Reports of intrauterine limb gangrene are sparse, and the majority of documented cases involve polycythemia-induced thrombosis [4]. Other less common causes include thrombosis of placental vessels or complications arising from antenatal treatments such as laser photocoagulation for TTTS [7]. Although laser therapy is often utilized to treat TTTS by cauterizing the abnormal placental vessels, it can, in rare instances, induce thromboembolic complications [6].

This case highlights the importance of early detection and management of TTTS, as well as the need for close surveillance of both twins throughout the pregnancy. The presence of polyhydramnios and oligohydramnios, as seen in this case, is a critical indicator of TTTS, and early intervention can significantly improve outcomes [2]. Additionally, the early identification of complications such as polycythemia and anemia in the respective twins can guide appropriate interventions, such as partial exchange transfusions and anticoagulation therapy [1]. Although the recipient twin in our case ultimately succumbed to disseminated intravascular coagulation (DIC) and intraventricular hemorrhage (IVH), Twin B, the donor twin, received appropriate transfusion therapy for anemia and was discharged in stable condition.

Antenatal management of TTTS includes various interventions such as amnioreduction, fetoscopic laser photocoagulation, and septostomy, all of which aim to correct the imbalanced blood flow between the fetuses [8]. These therapies, when applied early in the course of TTTS, can mitigate the risk of severe complications, including thromboembolism and ischemic damage. In addition, postnatal care for both twins must involve vigilant monitoring for signs of polycythemia, thrombotic events, and other sequelae of TTTS. Prompt management, including transfusions and anticoagulation therapy, can help avert further complications [3].

3. CONCLUSION

Monochorionic twin pregnancies require close surveillance for early signs of TTTS. Timely recognition of complications such as anemia-polycythemia imbalance and prompt intrauterine interventions like laser ablation or amnioreduction may prevent severe sequelae, including thromboembolism and limb gangrene. Postnatal management must include vigilant monitoring for signs of polycythemia and thrombotic events, and immediate interventions such as exchange transfusion and anticoagulation when needed

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