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Case Report

Favorable fetal outcome in a high-risk Rh-alloimmunized pregnancy with high indirect coombs titre complicated by systemic sclerosis: a rare case report

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ABSTRACT

Rh alloimmunization remains an important cause of fetal and neonatal morbidity despite anti-D prophylaxis. Pregnancy in diffuse systemic sclerosis is uncommon and associated with significant maternal and fetal risks. The coexistence of these two high-risk conditions is rare. Case Presentation: 25-year-old G2P1D1 with diffuse systemic sclerosis and Rh-negative blood group was managed with serial antenatal surveillance. Indirect Coombs titres progressively increased from 1:256 at 32 weeks to 1:512 at 34 weeks. Serial ultrasonography showed no evidence of hydrops fetalis and until delivery, fetal surveillance findings remained reassuring, with reactive third-trimester non-stress tests. Elective cesarean section was performed at 34+2 weeks for severe Rh alloimmunization with breech presentation. A healthy male neonate weighing 2.5 kg was delivered and managed successfully with phototherapy for neonatal hyperbilirubinemia. Conclusion: Favorable outcomes can be achieved through multidisciplinary care, close surveillance, and timely delivery.

Keywords: Rh alloimmunization, Diffuse systemic sclerosis, Indirect Coombs test, High-risk pregnancy, Neonatal hyperbilirubinemia, Case report

INTRODUCTION

Rh alloimmunization continues to be an important contributor to fetal anemia and hemolytic disease of the newborn.[1–3] Systemic sclerosis is a chronic autoimmune connective tissue disorder characterized by fibrosis, vasculopathy, and immune dysregulation.[4–6] Pregnancies complicated by systemic sclerosis are associated with preterm birth, fetal growth restriction, and hypertensive disorders.[4,5] The coexistence of severe Rh alloimmunization and diffuse systemic sclerosis is rarely reported and presents unique clinical challenges.

CASE REPORT

A 25-year-old gravida 2 para 1 death 1 (G2P1D1) woman was admitted at 34 weeks of gestation for antenatal evaluation and further management of a high-risk precious pregnancy. Her last menstrual period was dated 19/03/2025 with an expected date of delivery of 26/12/2025. The pregnancy was spontaneously conceived after 2.5 years of married life.

The patient had been regularly attending antenatal clinic visits at our tertiary care hospital throughout the

pregnancy. Routine antenatal investigations and serial obstetric assessments were performed during follow-up.

Her previous obstetric history revealed an institutional full-term normal vaginal delivery one year earlier. The neonate had died within five minutes of birth due to meconium aspiration syndrome, contributing to significant emotional and obstetric concern in the current pregnancy.

The patient was diagnosed of diffuse systemic sclerosis in current pregnancy and was under regular dermatological follow-up during pregnancy. She had clinical features suggestive of diffuse cutaneous involvement including skin tightening and cutaneous sclerosis involving the extremities and trunk. Associated dermatological manifestations included areas of skin thickening with pigmentary changes and reduced skin flexibility.

Dermatology consultation and expert dermatological reference had been obtained during antenatal care for assessment and monitoring of systemic sclerosis-related manifestations and disease progression during pregnancy. Ongoing surveillance for systemic involvement including vascular and connective tissue manifestations was performed throughout the antenatal period.

She was receiving treatment with topical Fusidic cream, topical moisturizer and Tablet nifedipine (NF 10 mg once daily), which was continued during pregnancy under supervision. Nifedipine was maintained considering possible vasospastic manifestations and peripheral vascular symptoms associated with systemic sclerosis.

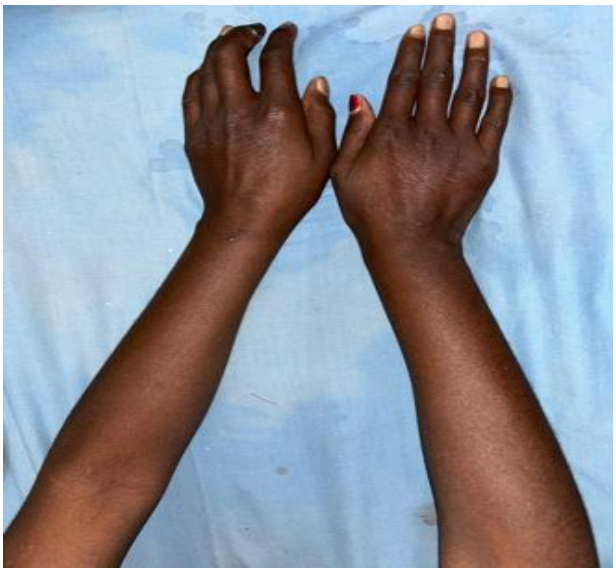

Figure 1: Bilateral upper limbs showing skin tightening and induration with tapering of the fingers, consistent with sclerodactyly.

Clinical monitoring for progression of systemic sclerosis features was performed throughout the antenatal period. Multidisciplinary care involving obstetricians, dermatologists, and neonatologists was planned considering the coexistence of autoimmune disease and severe Rh alloimmunization.

Figure 2: Palmar aspect of both hands demonstrating digital ischemic changes, fingertip ulceration, and pigmentary alterations associated with systemic sclerosis-related vasculopathy.

Antenatal evaluation

Maternal investigations revealed an A Rh-negative blood group with a strongly positive indirect Coombs test (Grade 4+). Serial indirect Coombs titres demonstrated progressive sensitization, increasing from 1:256 at 32 weeks to 1:512 at 34 weeks. Liver and renal function tests were within normal limits, and the coagulation profile was also within normal limits.

In view of already established Rh sensitization with markedly elevated antibody titre, prophylactic anti-D immunoglobulin administration was not expected to provide therapeutic benefit during the current pregnancy. Therefore, management was focused on close maternal and fetal surveillance, serial antenatal assessment, neonatal preparedness, and timely obstetric intervention.

The patient was on regular antenatal follow-up. Indirect Coombs testing showed strong positivity (4+), suggestive of significant maternal alloimmunization. Quantitative antibody titre estimation was not available at our center. Despite this, serial fetal surveillance revealed no sonographic evidence of hydrops fetalis, and fetal well-being remained reassuring until delivery at 34+2 weeks gestation. In view of Rh incompatibility and breech presentation, elective lower segment cesarean section was planned and performed at 34.2 weeks of gestation after Steroid Coverage.

A live male neonate weighing 2.5 kg was delivered on 16/11/2025 at 1:01 PM. The neonate cried immediately after birth and had APGAR scores of 8, 9, and 9 at 1, 5,

and 10 minutes respectively. No gross congenital anomalies were identified on neonatal examination.

Neonatal investigations

Neonatal investigations revealed an O Rh-positive blood group with a negative direct Coombs test. Peak serum bilirubin was 15.54 mg/dl, with a reticulocyte count ranging from 6.8% to 8.5%. G6PD levels, thyroid profile, and CRP were within normal limits.

At 24 hours of life, icterus extending up to the legs was noted. Serum bilirubin was 8.13 mg/dL, for which phototherapy was initiated. Subsequent bilirubin monitoring showed a peak total bilirubin level of 15.54 mg/dL at 42 hours of life, necessitating continuation of intensive phototherapy.

Further investigations including thyroid profile and G6PD assay were within normal limits. Repeat bilirubin at 66 hours of life decreased to 11.69 mg/dl and phototherapy was discontinued. The neonate remained hemodynamically stable with adequate feeding and no evidence of severe hemolysis. The baby was discharged in stable condition on exclusive breastfeeding, vitamin D supplementation, and routine immunization advice.

Maternal postpartum course

The mother remained hemodynamically stable during the postoperative and postpartum period. Postpartum recovery was uneventful with no evidence of hypertensive complications, excessive bleeding, thromboembolic events, worsening vascular symptoms, or progression of systemic sclerosis manifestations.

Regular postpartum monitoring was continued with multidisciplinary input. Dermatological follow-up was advised for ongoing evaluation of diffuse systemic sclerosis and continuation of medical management. The patient was counseled regarding future pregnancy risks, Rh isoimmunization, neonatal follow-up, and the importance of long-term rheumatological and dermatological surveillance. Mother and baby were discharged in satisfactory condition on Postoperative Day 10 after complete removal of Cesarean Section Sutures.

DISCUSSION

Rh alloimmunization continues to pose substantial risks to fetal and neonatal health. High indirect Coombs titres are associated with increased risk of fetal anemia and hemolytic disease of the newborn.[1–3] However, clinical severity may vary depending on antibody load, fetal antigen expression, and timing of intervention.

Systemic sclerosis during pregnancy is itself associated with increased obstetric complications due to vasculopathy and immune-mediated tissue involvement.[4–6] The coexistence of diffuse systemic sclerosis with severe Rh alloimmunization is rarely described in literature.

In the present case, despite markedly elevated indirect Coombs titre of 1:512, the fetus achieved a favorable outcome. Although the neonate developed significant hyperbilirubinemia requiring intensive phototherapy, exchange transfusion was avoided and neonatal recovery was satisfactory.

This favorable outcome may be attributed to close antenatal monitoring, timely obstetric intervention, multidisciplinary care involving obstetricians, pediatricians, and dermatologists, and prompt neonatal management. Fetal surveillance with Doppler ultrasonography is an established approach for assessing the risk of fetal anemia in Rh alloimmunized pregnancies.[7] The present case emphasizes that successful fetomaternal outcome is achievable even in pregnancies complicated by multiple high-risk factors when managed appropriately at a tertiary care center.

CONCLUSION

Pregnancy complicated by severe Rh alloimmunization with high indirect Coombs titre in the background of diffuse systemic sclerosis represents a rare and challenging clinical scenario. Early diagnosis, close surveillance, multidisciplinary management, and prompt neonatal care contributed to favorable maternal and fetal outcomes in this case.

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