Differential Diagnosis of Postpartum Hemorrhage in a Patient with Sickle Cell Disease: Case Report and Literature Review

Diagnóstico Diferencial de Hemorragia Pós-Parto em uma Paciente com Doença Falciforme: Relato de Caso e Revisão da Literatura

Diagnóstico Diferencial de Hemorragia Posparto en una Paciente con Enfermedad de Células Falciformes: Informe de un Caso y Revisión de La Literatura

RESUMO

Introdução: A doença falciforme é a hemoglobinopatia genética mais comum no Brasil e representa um fator de risco para complicações gestacionais, incluindo a hemorragia pós-parto (HPP). O diagnóstico preciso e o manejo adequado são essenciais para reduzir a morbimortalidade materna. **Relato de caso:** Gestante de 37 anos, portadora de anemia falciforme (AF) e gestação gemelar dicoriônica, evoluiu com síndrome HELLP, sendo submetida à cesariana com necessidade de transfusões sanguíneas. No 13º dia de pós-operatório, apresentou quadro de HPP secundária grave. Exames revelaram uma malformação arteriovenosa, tratada por meio de embolização arterial. Devido à recorrência do sangramento, foi realizada histerectomia total abdominal. A paciente evoluiu com endometrite, recebendo antibioticoterapia. O exame histopatológico confirmou placenta acreta. No total, foram transfundidas 7 unidades de concentrado de hemácias, e a paciente recebeu alta após estabilização clínica. **Discussão:** A AF intensifica a resposta hematológica frente a condições hemorrágicas, dificultando o manejo transfusional e o diagnóstico de causas subjacentes, como a placenta acreta. A sobreposição de síndromes obstétricas, anemia crônica e complicações infecciosas demanda uma abordagem multidisciplinar. **Conclusão:** A vigilância no puerpério de pacientes com AF deve ser intensificada, com atenção especial a causas incomuns de HPP. A detecção precoce e o manejo direcionado são fundamentais para prevenir desfechos maternos adversos.
DESCRITORES: Doença falciforme, anemia falciforme, hemorragia pós-parto, diagnóstico diferencial.

ABSTRACT

Introduction: Sickle cell disease is the most common genetic hemoglobinopathy in Brazil and represents a risk factor for gestational complications, including postpartum hemorrhage (PPH). Accurate diagnosis and appropriate management are essential to reduce maternal morbidity and mortality. **Case report:** A 37-year-old pregnant woman with sickle cell anemia (SCA), carrying a dichorionic twin pregnancy, developed HELLP syndrome and underwent a cesarean section requiring blood transfusions. On the 13th postoperative day, she presented with severe secondary PPH. Examinations revealed an arteriovenous malformation, for which she underwent arterial embolization. Recurrent bleeding led to a total abdominal hysterectomy. She developed endometritis and received antibiotic therapy. Histopathology confirmed placental accreta. In total, she received 7 units of packed red blood cells and was discharged after stabilization. **Discussion:** SCA exacerbates the hematological response to hemorrhagic conditions, complicating transfusion management and the diagnosis of underlying causes, such as placental accreta. The overlap of obstetric syndromes, chronic anemia, and infectious complications requires a multidisciplinary approach. **Conclusion:** Postpartum surveillance of patients with SCA should be intensified, with special attention to unusual causes of PPH. Early detection and targeted management are essential to prevent adverse maternal outcomes.
DESCRIPTORS: Sickle cell disease, sickle cell anemia, postpartum hemorrhage, differential diagnosis

RESUMEN

Introducción: La enfermedad de células falciformes es la hemoglobinopatía genética más común en Brasil y constituye un factor de riesgo para complicaciones gestacionales, incluida la hemorragia posparto (HPP). El diagnóstico preciso y el manejo adecuado son esenciales para reducir la morbimortalidad materna. **Reporte de caso:** Mujer embarazada de 37 años, portadora de anemia falciforme (AF) y gestación gemelar di-

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coriónica, desarrolló síndrome HELLP y fue sometida a cesárea con necesidad de transfusiones sanguíneas. En el decimotercer día postoperatorio presentó HPP secundaria grave. Los exámenes revelaron una malformación arteriovenosa, tratada mediante embolización arterial. Debido a la recurrencia del sangrado, se realizó histerectomía abdominal total. La paciente presentó endometritis y recibió tratamiento antibiótico. El examen histopatológico confirmó placenta acreta. En total, se transfundieron 7 unidades de concentrado de glóbulos rojos, y la paciente fue dada de alta tras su estabilización clínica. **Discusión:** La AF intensifica la respuesta hematológica frente a condiciones hemorrágicas, dificultando el manejo transfusional y el diagnóstico de causas subyacentes, como la placenta acreta. La superposición de síndromes obstétricos, anemia crónica y complicaciones infecciosas requiere un abordaje multidisciplinario. **Conclusión:** La vigilancia posparto de las pacientes con AF debe ser intensificada, con especial atención a causas inusuales de HPP. La detección precoz y el manejo dirigido son fundamentales para prevenir desenlaces maternos adversos. **DESCRIPTORES:** Enfermedad de células falciformes, anemia falciforme, hemorragia posparto, diagnóstico diferencial.

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INTRODUCTION

Sickle cell disease (SCD) is a hemoglobinopathy representing the most common genetic disorder in Brazil, with an estimated prevalence of 60,000 to 100,000 cases and an overall mortality rate of 0.22 per 100,000 inhabitants between 2014 and 2020[1]. The most severe form is sickle cell anemia (SCA), an autosomal recessive condition caused by a mutation in the beta-globin gene that results in the predominant formation of hemoglobin S. This variant has a higher propensity to polymerize, alter membrane structure, induce cellular deformation, and increase susceptibility to hemolysis, leading to the clinical manifestation of the disease[2,3]. SCA is also a risk factor of complications during pregnancy and the postpartum period, including severe anemia, thromboembolic events, infections, preeclampsia, intrauterine growth restriction, and miscarriage[4,5].

A critical pathological entity in this context is postpartum hemorrhage (PPH), defined as blood loss exceeding 500 mL after vaginal delivery, more than 1,000 mL post-cesarean section, or any bleeding that leads to signs or symptoms of hypovolemia[6]. PPH is classified as primary (within the first 24 hours postpartum) or secondary (between 24 hours

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and six weeks following birth). While the former is more prevalent, secondary PPH is a significant cause of hospital readmission in the postpartum period[7,8].

In patients with SCD, hemorrhagic events are relevant, mainly due to chronic anemia and history of previous blood transfusions. Timely identification and accurate diagnosis of these events are also essential for effective management and reduction of morbidity and mortality in affected patients. Therefore, this study aimed to report the case of postpartum hemorrhage in a patient with sickle cell anemia.

CASE REPORT

A 37-year-old patient with SCD (diagnosed during childhood), secundigravida with previous cesarean delivery, presented with dichorionic-diamniotic twin pregnancy. She received prenatal care at a specialized service for patients with SCD. During the first trimester of gestation, after initiating specialized follow-up, she developed a pain crisis requiring hospitalization and intravenous vancomycin due to methicillin-resistant *Staphylococcus aureus* bacteremia. Discharge occurred after a negative control blood culture.

At 33 weeks of gestation, she developed altered blood pressure with elevated liver enzymes and bilirubin, which were twice her baseline values (Table 1). After the diagnosis of HELLP syndrome (hemolysis, elevated liver enzymes, and thrombocytopenia), a cesarean delivery was performed without complications.

Table 1. Results of laboratory tests during hospitalization.

	Baseline prenatal routine	Before cesarian	First hours post-cesarean	9th day post-cesarean	13th day post-cesarean	Before embolization	After embolization	Before hysterectomy	After hysterectomy	Hospital discharge
Hb/Hct (g/dL/%)	8,4/27	8,1/25,3	6,4/20,2	7,0/21,6	6,1/19,2	7,0/21,9	5,9/18	6,4/19,8	7,8/24	7,8/25,1
Plaquetas (mm3)		321.000	257.000	785.000						962.000
AST/ALT (U/L)	76/47	181/100	231/140	34/30						46/46
Total/Indirect bilirubin (mg/dL)	2,23/0,98	3,36/1,13	3,08/1,05	0,71/0,31						0,72/0,07
Urea/Creatinine (md/dL)	21/0,53	x/0,4	20/0,4	08/0,4						08/0,4
LDH (U/L)	588	805	894	370						220
Fibrinogen			366							
INR			1,14		1,07					

Hb = hemoglobin; Hct = hematocrit; AST = aspartate aminotransferase; ALT = alanine aminotransferase; LDH = lactate dehydrogenase; INR = international normalized ratio.

The patient required intensive care postoperatively with magnesium sulfate for HELLP syndrome and received one unit of packed red blood cells for anemia. Additional blood transfusion was required after being transferred to the rooming-in care and before discharge. On postoperative day 13, the patient returned with severe genital bleeding, systemic symptoms (fatigue, weakness, and headache), and low hemoglobin levels (6.1 g/dL), necessitating a new blood transfusion. The ultrasonographic investigation revealed findings compatible with arteriovenous malformation, confirmed by magnetic resonance imaging; thus, uterine artery embolization was performed on postoperative day 17 (Figures 1 and 2). Three days later, recurrent bleeding with hemodynamic compromise prompted urgent total abdominal hysterectomy without complications. Intraoperatively, purulent pelvic secretion was observed and treated with intravenous clindamycin (2,700 mg/day) and gentamicin (320 mg/day) due to endometritis. The patient achieved discharge fitness seven days after the surgery. Seven units of packed red blood cells and eight days of intravenous antibiotics were required during hospitalization.

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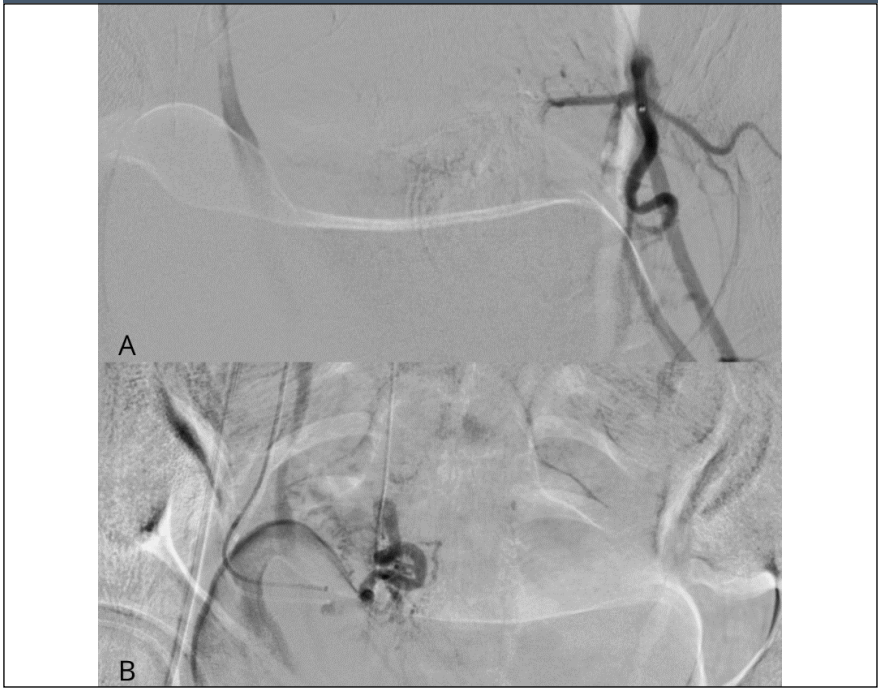
Figure 1: Arteriography before uterine artery embolization. A: Initial arteriography of the left internal iliac and uterine artery. B: Initial arteriography of the right uterine artery.

Figure 2: Arteriography after uterine artery embolization. A: Control arteriography post-embolization of the left uterine artery. B: Control arteriography post-embolization of the right uterine artery.



Outpatient follow-up showed good general condition without complications. The histopathology identified retained placental tissue partially infiltrating the myometrium, consistent with placental accretism, left fallopian tube with congestive changes, and acute suppurative peritonitis.

The patient provided informed consent for this case report, which was approved by the institutional research ethics committee (CAAE: 87027725.9.0000.5201; no 7.496.719).

DISCUSSION

The World Health Organization recognizes SCD as a global public health problem[9,10]. Severe complications, including infections, vital organ damage, stroke, respiratory and renal diseases, bone marrow suppression, growth impairment, and cognitive disorders, may lead to high maternal and fetal mortality rates[10].

SCD is an important risk factor for increased morbidity and mortality during pregnancy and the puerperium[4]. Patients frequently present a history of multiple blood transfusions, hindering emergency transfusions in hemorrhagic situations due to alloimmunization and blood incompatibility[4]. People with SCD commonly exhibit chronic low hemoglobin levels, requiring rigorous clinical surveillance, especially during high-risk situations (e.g., surgical procedures)[11], to prevent adverse outcomes.

Given the complexity of cases, the precise identification of severe anemia and its causes becomes essential for appropriate therapeutic approaches. In this case report, placental accretism may have caused intense hemorrhage, which may occur even without suggestive imaging findings and particularly in patients with risk factors, such as previous cesarean sections[12,13]. However, the literature review revealed no direct association between SCA diagnosis and placental accretism occurrence.

In this context, differential diagno-

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sis and management of intense hemorrhage with postpartum anemia is challenging, considering the inherent complications of SCD. This emphasizes the importance of careful diagnostic evaluation, adequate management, and early intervention when facing postpartum hemorrhage in this population.

Despite healthcare advances in pregnant woman care, maternal and fetal morbidity and mortality rates remain high. Managing PPH in patients with SCA requires strengthened surveillance and maternal-fetal care during the puerperal period. The early identification of risk factors, including placental accretism without suggestive imaging findings, is also fundamental in women with relevant obstetric histories, while effective care protocols and continuous patient monitoring are indispensable for reducing morbidity and mortality. Therefore, investing in specialized and comprehensive postpartum care represents a clinical necessity and a commitment to the health and life of these women.

INFORMED CONSENT

Informed consent was obtained from the patient for publication of this case report.

CONTRIBUTION STATEMENT

Conceptualization: NAM, GFAS, AIS. Data curation: GFAS. Investigation: NAM, GFAS, NFM, LS. Methodology: NAM, GFAS, AIS, ASRS, LK. Project administration: AIS, GFAS. Supervision: AIS, ASRS, LK. Roles/Writing - original draft: GFAS, AIS. Writing - review and editing: All the authors.

CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

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