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## Case Report

# A delicate balance: hereditary hemorrhagic telangiectasia and antiphospholipid syndrome in pregnancy with dual therapy

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## ABSTRACT

Hereditary hemorrhagic telangiectasia (HHT) and antiphospholipid syndrome (APS) are two rare disorders that present a challenge during pregnancy due to the competing risks of bleeding and thrombosis. This case presents a 26-year-old female, gravida 3, para 0-1-1-1, with a past medical history of HHT and APS receiving prophylactic anticoagulation. The patient presented to the antepartum ward (APW) at 32.1 weeks of gestation with complaints of hemoptysis and epistaxis. During this evaluation, nebulized tranexamic acid (TXA) was added as a treatment without interruption of anticoagulation therapy. This management resulted in complete resolution of symptoms without maternal or fetal complications. Therefore, nebulized TXA may represent a safe therapeutic option for hemoptysis in pregnant patients with HHT who require anticoagulation for APS. The findings highlight the importance of individualized, multidisciplinary care in a setting of complex diseases.

**Keywords:** Hereditary hemorrhagic telangiectasia, Antiphospholipid Syndrome, Pregnancy, Hemoptysis, Nebulized tranexamic acid, High-risk pregnancy

## INTRODUCTION

Hereditary hemorrhagic telangiectasia (HHT) is a rare autosomal dominant bleeding disorder characterized by mucocutaneous telangiectasias and visceral arteriovenous malformations, involving the nasal mucosa, lungs, gastrointestinal tract, and brain.<sup>1</sup> One of the most common clinical manifestations is recurrent epistaxis, and pulmonary arteriovenous malformations may lead to symptoms such as hemoptysis, hypoxemia, and complications including paradoxical embolic events.<sup>2</sup> Pregnancy represents a unique physiological state in which increases in blood volume and cardiac output can exacerbate bleeding and increase the risk of pulmonary hemorrhage, particularly in patients with HHT.<sup>2</sup>

Antiphospholipid syndrome (APS) is an acquired autoimmune thrombophilia associated with recurrent

pregnancy loss, placental insufficiency, preeclampsia, and maternal thromboembolism.<sup>3</sup> Standard management during pregnancy consists of prophylactic or therapeutic anticoagulation with low-molecular-weight heparin and low-dose aspirin to reduce maternal and fetal morbidity and mortality.<sup>3</sup> In patients with coexisting APS and HHT, anticoagulation is necessary to decrease thrombotic risk; however, it may exacerbate bleeding from fragile vascular malformations.<sup>3</sup>

Tranexamic acid (TXA) is an antifibrinolytic agent that inhibits fibrin degradation by blocking the activation of plasminogen to plasmin.<sup>4</sup> By binding to lysine receptor sites on plasminogen, it prevents plasmin from degrading fibrin clots, thereby stabilizing clot formation and reducing bleeding.<sup>4</sup> TXA has an established role in the management of obstetric hemorrhage and mucosal bleeding during pregnancy and the postpartum period.<sup>4</sup> However,

nebulized TXA could be an option as a targeted therapy for hemoptysis, given its minimal systemic absorption and ability to achieve adequate local hemostasis.<sup>5,6</sup> Regardless, data on the use of nebulized TXA during pregnancy, particularly in patients requiring concurrent anticoagulation, remains limited.

This case highlights a potential therapeutic strategy for balancing bleeding and thrombotic risks in a rare and high-risk obstetric population.

**CASE REPORT**

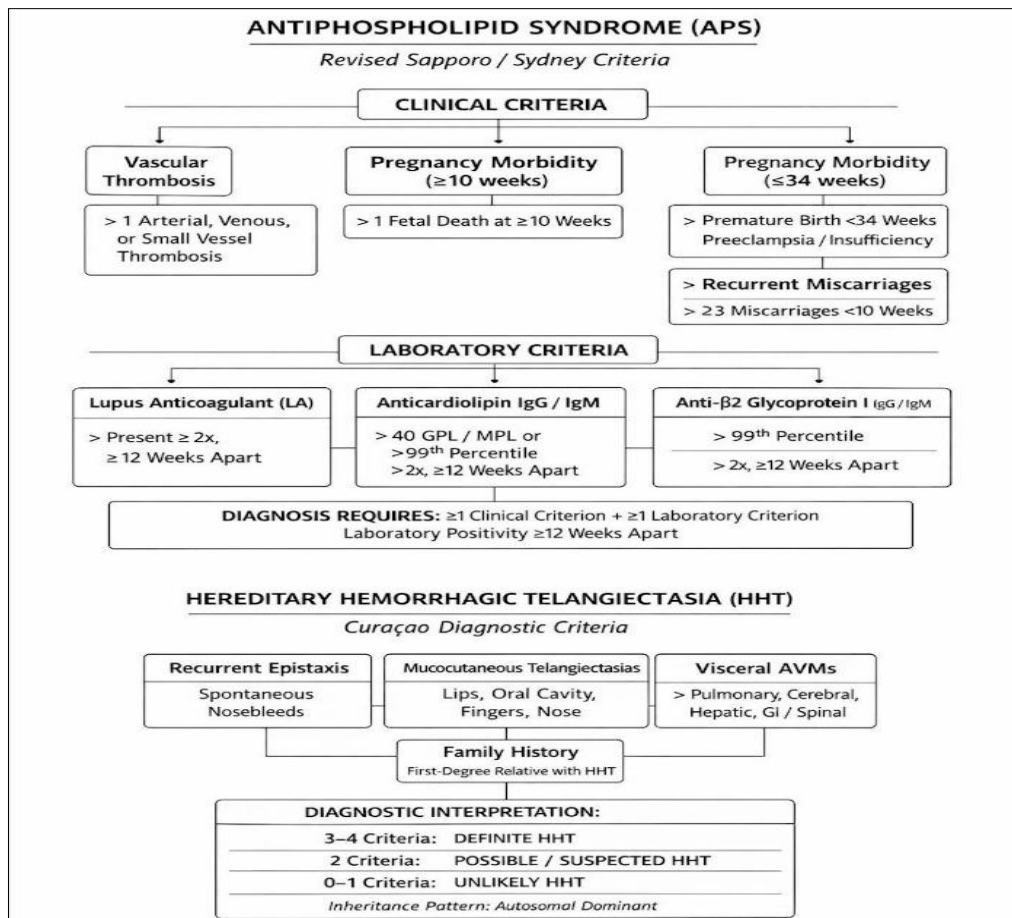
A 26-year-old gravida 3, para 0-1-1-1 woman with a history of APS and HHT presented to the APW at 32 weeks and 1 day of gestation, with a four-day history of recurrent hemoptysis and epistaxis.

Obstetric history was significant for a pregnancy in 2021, during which she presented with similar hemoptysis episodes at 34 weeks of gestation. During that hospital admission, the patient was monitored and initially presumed to have hemorrhagic dengue associated with thrombocytopenia. The patient denied a family history of

coagulopathies, APS, or HHT during that admission. Evaluation for von Willebrand disease was negative. However, physical examination was remarkable for visible arteriovenous malformations (AVMs) in the nasal mucosa, and chest computed tomography demonstrated pulmonary AVMs. Based on her history of recurrent epistaxis and visible nasal AVMs, the patient met 3 of 4 Curaçao criteria, confirming a clinical diagnosis of HHT.

APS was also diagnosed in 2021 based on clinical and laboratory criteria, including a persistently antinuclear antibody (ANA) positive result and a positive lupus anticoagulant test. Since that diagnosis, the patient was managed with prophylactic anticoagulation using Lovenox 40 mg subcutaneously once daily in combination with low-dose aspirin 81 mg daily (Figure 1).

The diagnostic criteria for both APS and HHT are summarized below in Figure 1. APS diagnosis requires at least one clinical and one laboratory criterion, confirmed on two occasions  $\geq 12$  weeks apart. HHT is diagnosed using the Curaçao criteria, including epistaxis, mucocutaneous telangiectasia, visceral arteriovenous formation, and family history.<sup>3,7</sup>



**Figure 1: Diagnostic criteria for APS and HHT.**

At the antepartum ward, the patient reported spontaneous coughing episodes with blood-tinged sputum along with

recurrent nosebleeds. She denied any chest pain, dyspnea, fever, uterine contractions, vaginal bleeding, or fluid

leakage. Physical examination was remarkable for mild active bleeding from both nostrils. Fetal assessment by non-stress test (NST) and ultrasound demonstrated a reassuring biophysical profile. Laboratory evaluation, including complete blood count, fibrinogen, and coagulation studies, was unremarkable. Chest radiography and computed tomography imaging during this admission showed no evidence of acute intrapulmonary pathology or progression of known pulmonary AVMs. She was therefore diagnosed with a flare of HHT.

The case was referred to the services of Pulmonary Critical Care Medicine and Hematology/Oncology, whom recommended continuing prophylactic Lovenox and low-dose aspirin, given her thrombotic risk due to APS. However, due to her continuing episodes of hemoptysis, the pulmonary team recommended nebulized TXA therapy at a dosage of 500 mg every six hours for localized control of bleeding. Antenatal corticosteroids were also administered to promote fetal lung maturation.

After two days of treatment, the patient demonstrated clinical improvement with complete resolution of hemoptysis and epistaxis, at which point nebulized TXA was discontinued. She remained hospitalized for observation without recurrence of symptoms. On hospital day five, she was discharged home, in stable condition, and remained asymptomatic for the remainder of the pregnancy.

At 37 weeks and 3 days of gestation, the patient underwent a repeat caesarean delivery due to no trial of labor after caesarean (NO TOLAC). Delivery timing was guided by APS management recommendations, balancing the risks of early-term delivery against potential maternal and fetal complications, including preeclampsia, intrauterine growth restriction, venous thromboembolism, stillbirth, and maternal morbidity. The postoperative course was uncomplicated, with favorable maternal and neonatal outcomes.

## DISCUSSION

The coexistence of HHT and APS represents a complex clinical challenge, as anticoagulation required for APS can worsen bleeding in HHT, while its interruption increases the risk of thrombosis and adverse pregnancy outcomes. As a result, management must carefully balance these competing risks, often requiring an individualized and multidisciplinary approach. Patients with HHT have vessels that lack normal capillary architecture, leading to fragile, tilted vessels that increase the risk of hemorrhage, particularly under the hemodynamic stress of pregnancy.<sup>1</sup> Furthermore, these vascular malformations may contribute to complications such as pulmonary hypertension, a prothrombotic state, or immune dysfunction.<sup>1</sup>

APS, in contrast, confers a hypercoagulable state with increased risk of thrombosis and adverse pregnancy outcomes, including recurrent pregnancy loss,

preeclampsia, intrauterine growth restriction, and preterm delivery.<sup>3</sup> The recommendation for prophylactic anticoagulation, such as low-molecular-weight heparin combined with low-dose aspirin, stems from the importance of reducing thrombotic and obstetric complications.<sup>3</sup> In this patient, continuation of Lovenox and aspirin was necessary for APS management, even as she experienced an HHT flare manifested by hemoptysis and epistaxis.

Furthermore, a retrospective study by Andofer et al showed that more than 60% of patients with HHT report nosebleeds during pregnancy and approximately 27% of the 60% reported an increase in the severity of nosebleeds compared with their first pregnancy.<sup>2</sup> This exacerbation is likely related to the hemodynamic and hormonal changes of pregnancy, including increased cardiac output and nasal mucosal hyperemia.<sup>2</sup> These studies also highlight the lack of patient education, especially during the family planning period. A cohort study by Andofer et al also demonstrated improved survival in patients with prior awareness of their HHT compared with those unaware of having HHT.<sup>2</sup> In this patient with coexistent APS and HHT, the flare of epistaxis and hemoptysis required rapid evaluation to balance the competing risks of hemorrhage and thrombosis.

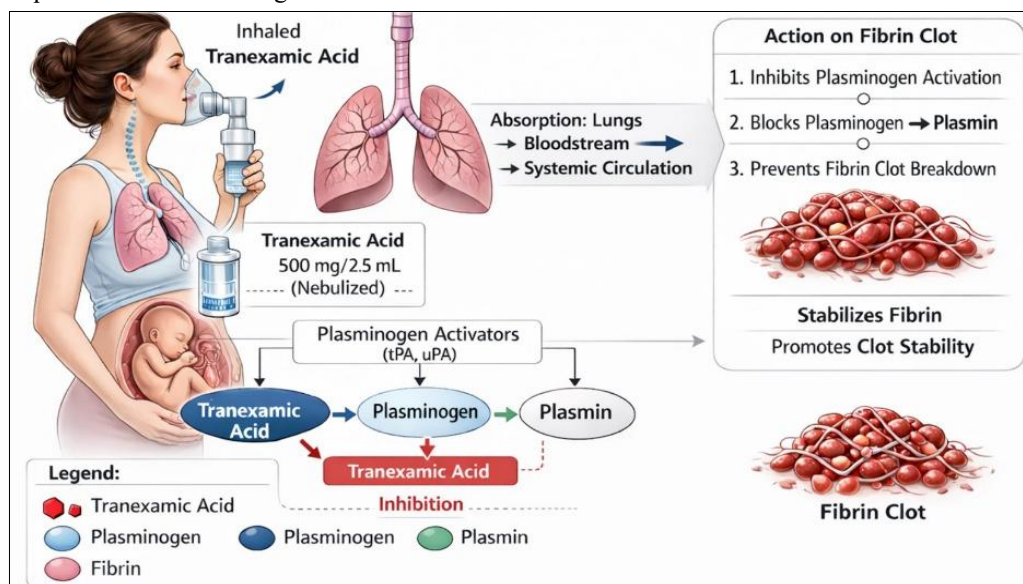
First-line treatment strategies in pregnant patients with HHT have consisted of bipolar cautery, laser therapy, nasal packing, and intravenous TXA.<sup>2</sup> TXA has been evaluated in obstetric populations, where it reduces blood loss and transfusion requirements following cesarean and vaginal deliveries and appears safe for use during pregnancy and postpartum.<sup>4</sup> These findings support the general safety of TXA in pregnancy, providing reassurance for its use as a localized therapy in this patient.

However, nebulized TXA may represent an alternative therapeutic approach due to its localized mechanism of action, allowing clot stabilization at the site of bleeding while minimizing systemic absorption and reducing thrombotic risk.<sup>6</sup> The mechanism of action of inhaled TXA is illustrated in Figure 2. Inhaled tranexamic acid inhibits the conversion of plasminogen to plasmin, thereby preventing fibrin degradation and promoting clot stability at the site of bleeding.

Evidence supporting nebulized TXA includes randomized trials and case reports demonstrating efficacy and safety for pulmonary hemorrhage.<sup>8</sup> For example, a randomized controlled trial by Wand et al. reported resolution of hemoptysis within 5 days of nebulized TXA treatment, along with reduced expectorated blood volume, shorter hospital stays, and fewer invasive interventions.<sup>5</sup> Another study reported effective control of hemoptysis with nebulized TXA in patients on rivaroxaban after failure of oral TXA.<sup>9</sup> This study was relevant to the case of our patient, who was on Lovenox due to an APS diagnosis.

Additional evidence supports nebulized TXA as a safe and effective therapy for pulmonary hemorrhage, with reduced systemic thrombotic risk and minimal adverse effects.<sup>6</sup> It also demonstrates the potential to reduce invasive interventions for the treatment of hemoptysis.<sup>8</sup> Therefore, current evidence supports the use of nebulized TXA as a local therapeutic option for hemoptysis in pregnant patients who require continued anticoagulation.<sup>2,5,6</sup> In this

case, rapid resolution of hemoptysis and epistaxis was observed without systemic adverse effects, confirming the clinical applicability of this approach.<sup>2,5</sup> The case also demonstrates the importance of a multidisciplinary approach involving obstetrics, hematology, and pulmonary specialists in the management of complex vascular disorder.



**Figure 2: Mechanism of action of inhaled tranexamic acid.**

## CONCLUSION

HHT and APS present a significant clinical challenge in pregnancy due to their competing risks of hemorrhage and thrombosis. The use of nebulized TXA, in this patient, proved a possible option to treat symptoms in HHT during pregnancy without exposing the patient to the risk of thrombotic events due to APS. Evidence from prior studies demonstrates the efficacy of this therapy in non-pregnant patients with hemoptysis and epistaxis, reducing the need for invasive interventions. While current evidence supports the effectiveness of TXA in pulmonary and obstetric bleeding, there are no established protocols for managing patients with coexisting HHT and APS during pregnancy using nebulizer TXA. Therefore, cases such as this case highlight the importance of further studies to better define indications, dosing, and long-term outcomes in pregnant populations with competing risks of hemorrhage and thrombosis.

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