Necrotizing Cellulitis Sepsis In A Pregnant Patient With Antiphospholipid Syndrome Under Treatment

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ABSTRACT
Introduction: *The term "antiphospholipid syndrome" (APS) describes the association between antiphospholipid antibodies (AFA) and a clinical picture of hypercoagulability characterized by repeated thrombosis and recurrent abortions.*
Objective: *To present a case of severe perineal cellulitis in a pregnant patient with APS under treatment with hydroxychloroquine.*
Case: *A 39-year-old pregnant patient with APS treated with hydroxychloroquine and anticoagulation developed a severe perineal infection. She was treated with termination of pregnancy at term, aggressive drainage of the perineum and extensive antibiotic treatment with good evolution.*
Conclusion: *The association of hydroxychloroquine treatment, pregnancy and a septic complication is uncertain. The immunosuppressive treatment is not standard and could have favored the poor prognosis of the clinical condition.*

Key words: *Cellulitis; Fournier; Pregnancy; APS*

INTRODUCTION

The term "antiphospholipid syndrome" (APS) describes the association of antiphospholipid antibodies (AFA) with a clinical picture of hypercoagulability characterized by repeat thrombosis and recurrent abortions. The treatment is based on the use of platelet antiaggregant drugs or anticoagulants.[1,2]

CASE REPORT

The case of a 39-year-old, 35 weeks pregnant woman, with a history of APS and five previous spontaneous abortions is presented. She had a history of hydroxychloroquine, and anticoagulation with low weight heparin treatment for APS during pregnancy. She presented at the emergency department with pain, heat and redness in the left gluteal area (fig. 1). WBC count was 18,440/ mm3. Ultrasound showed no evident collection. Aspiration puncture was obtained serous liquid with subsequent negative culture. Broad spectrum antibiotics were prescribed. Drainage and opening of the space with cellulitis signs was performed, without evidence of collection (fig. 2). On the third day, she had WBC count 25,960/mm3, Dimero D 1,643 ng/ml, negative blood cultures but worsening of the general condition, and developed a systemic inflammatory response syndrome. Also had increased local erythema, swelling, and signs of septic shock, so that an urgent cesarean section was decided. A new necrosis resection was performed at the operating room (fig. 3). Computed tomography was performed to assess the extent of the manifestation (fig. 4). On the 5th day, lab results were CRP 14.5 mg/dl, Procalcitonin 2.4 ng /ml, and Pseudomona aeruginosa carbapenems- resistant in cellulitis culture.

A new lavage, drainage, and tissue resection of the gangrene site was performed (figs. 5 and 6). Biopsy of resected tissue showed coagulation necrosis. Antibiotic treatment consisted of meropenem, vancomycin

and clindamycin. She evolved favorably with outpatient wound treatment and granulation by second intention. The hospital stay was 10 days. Perineal MRI at long term showed ad integrum restitution. Figures 7 and 8 show clinical and imaging ad integrum restitution.

Figure 1: Day 1. Initial cellulitis explored by ultrasound.

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Figure 2: Day 2. Cellulitis progression.

Figure 3: Day 3. Surgical treatment of cellulitis.

DISCUSSION

Cutaneous manifestations of APS appear in the 49% of cases and may constitute the initial manifestation of the condition. Circumscribed (4%), and extensive (2%) cutaneous necrosis are infrequent. The risk of thrombosis is significantly higher during pregnancy, and in pregnant patients with SAF it ranks from 5% to 12%.[1,2]

The association of treatment with hydroxychloroquine, immunosuppression and a septic complication is uncertain. Although immunosuppressive treatment is not standard, it could have favored the poor prognosis. Similar as-

Figure 4: Last surgical resection.

Figure 5: Day 14.

Figure 6: Day 21.

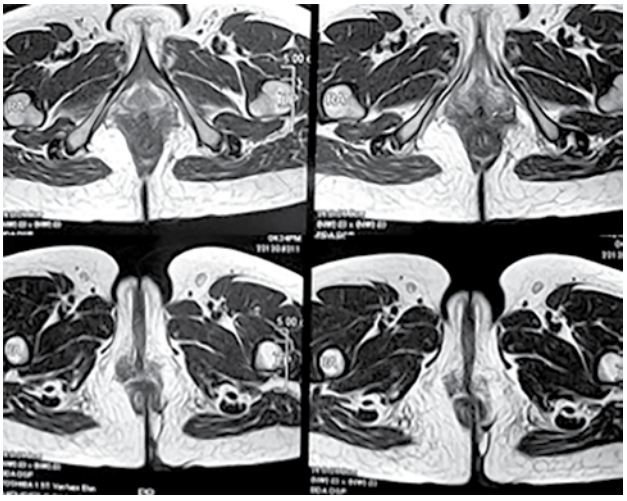

Figure 7: Computed tomography. Ad-integrum restitution.

Figure 8: Clinical ad-integrum restitution.

sociation was not found in the literature search.

CONCLUSION

During pregnancy severe soft tissue infections of the perineum without previous pathology are infrequent.

The immunosuppressive treatment for APS was a determining factor for the establishment of a severe septic clinical picture.

Aggressive treatment and termination of pregnancy at term were fundamental for the favorable resolution of the pathology.

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COMMENT

The cutaneous manifestations associated with antiphospholipid syndrome (APS), although common, are extremely diverse. Thrombosis, its main complication, can affect vessels of different sizes, such as those of the dermis. According to a descriptive study of 200 patients with APS, cutaneous manifestations are frequently the first presentation of the disease, being reticular livedo the most commonly observed (Arthritis Rheum 2005;52 (6):1785-93). Dr. Hugo Amarillo et a. present an infrequent and interesting clinical case of a 39-year-old woman and 35 weeks of gestation with necrotizing fasciitis secondary to APS in immunosuppressive treatment with hydroxychloroquine. The rapid and correct decision-making coupled with a progressively aggressive and consistent surgical approach over time, allowed the patient to progress favorably. In agreement with the authors, the immunosuppressive treatment may have favored the development of coagulation necrosis. Such association is not described in the available bibliography.

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