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Case Report

Fusion vaginal wall in a pediatric patient with post hematopoietic stem cell transplantation

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ABSTRACT

Aplastic anemia is a rare condition characterized by bone marrow failure and reduced blood cell production. Hematopoietic stem cell transplantation (HSCT) is the preferred treatment when a matched donor is available. This condition carries a graft-versus-host disease (GVHD) risk, affecting 30-70% of patients. GVHD can manifest in various areas, including the mouth, skin, and genitals, with vaginal symptoms present in 26% of cases. These may include dryness, itching, discharge, and labial fusion. A 14-year-old female treated with HSCT, developed skin and vaginal GVHD. She experienced amenorrhea and abdominal pain, a subsequent MRI revealed a normal uterus but a lobed vagina with multiple septa. The patient underwent vaginal exploration and laparoscopic surgery, revealing a fusion in labia minora and a significant vaginal cavity obstruction. Approximately 600 ml of blood was drained. Female genital GVHD in pediatric patients has a low incidence of 5.9%. Symptoms often include vulvar pain (37%), dysuria (37%), and pruritus (26%), with many patients remaining asymptomatic. A study found limited occurrences of vaginal stenosis, emphasizing the rarity of severe presentations. Routine gynecological evaluations are recommended for patients post-HSCT. In cases of colpohaematometra, draining and addressing stenosis may be necessary. Vaginal GVHD is a common complication of HSCT, ranging from vulvar pruritus to severe vaginal stenosis or fusion. Management may involve surgical liberation, vaginal molds, and treatment with estrogens and corticosteroids. Gynecological consultations are recommended for all patient's post-transplant.

Keywords: Aplastic anemia, Vaginal fusion, Stem cell, Hematometra

INTRODUCTION

Aplastic anemia is a condition where the bone marrow fails, resulting in a suppression of function and pancytopenia with at least two blood lineages of cells. The incidence of this disease is rare, with 1 or 2 cases per million.[1] The treatment of choice in case of having a direct matched donor is the HSCT. T cells function is preserved in these patients, elevating the risk of GVHD that happens in 30-70% of patients; this complication is a multi-system immune disorder with an exaggerated inflammation, wherein donor lymphocytes interact pathologically with foreign antigens, affects the morbimortality of the patients being worst in the acute GVHD with 15% of mortality that happens in the first 100 days, for the chronic version of the complications there have been reported certain risk factors like previous acute GVHD and the use of supplemental buffy coat infusions.[2]

The GVHD can be manifested in the mouth, skin, eyes, liver, intestines, and genitals; vaginal manifestations are reported in 26% of cases, the first manifestations are vulvar dryness, itching, burning sensation, vaginal

discharge, burning and swelling complicated with sclerotic changes, mucosal erosion, fissures, fusion of the labia, vaginal synechiae and vaginal stenosis with or without hematocolpos.[3,4] The time presentation is 7 to 10 months, which is more common in patients with peripheral blood stem cell versus bone marrow transplantation.[5] A special relation between skin and vaginal manifestations showed a significant positive correlation ($p<0.01$) with a 90% probability of vaginal coexistence with other organ involvement as reported in the mouth or skin.

CASE REPORT

A 14-years-old female patient with a previous diagnosis of epilepsy treated with levetiracetam and a diagnosed aplastic anemia that was treated sixteen months before with HSCT with development of skin GVHD and a simultaneous vaginal GVHD treated one month earlier with the liberation of vaginal synechia and hymenectomy by another institution. The patient had her menarche at nine years old with regular menstruation every 28 days and four days long with eumenorrhea; four months after the transplant, she manifested amenorrhea, with the consecutive presentation of abdominal mass pain at palpation.

She underwent an abdominal ultrasound (Figure 1) with the uterus and ovaries not appropriately visualized; it was reported that a vaginal dilatation of 8.9 cm and an abundant amount of hypoechogenic content, which is why we performed a magnetic resonance for better visualization. The magnetic resonance reports a uterus in anteversion of 5.9×4.8×4.3 centimeters, a homogeneous myometrium, a distended endometrial cavity, it is observed a lobed vagina with multiple septa of 12.2×9×7.6 cm, and a total volume of 435 milliliters, with an incomplete septum at 3.6 cm of introitus (Figure 2).

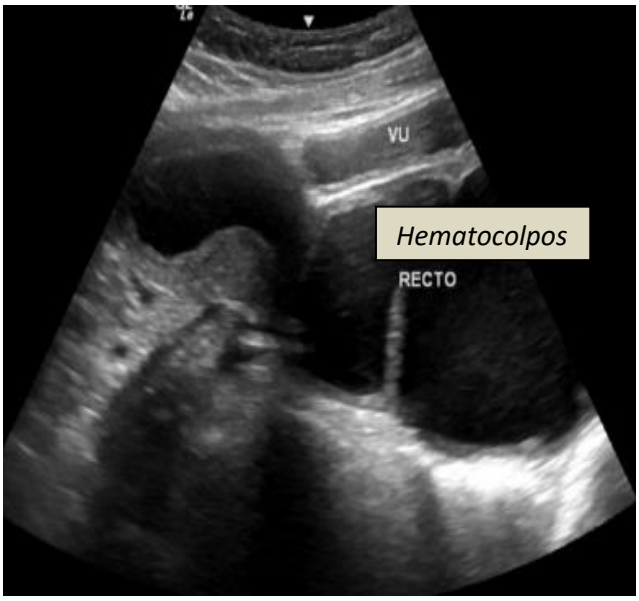

Figure 1: Abdominal ultrasound with the visualization of hematocolpos and hematometra.

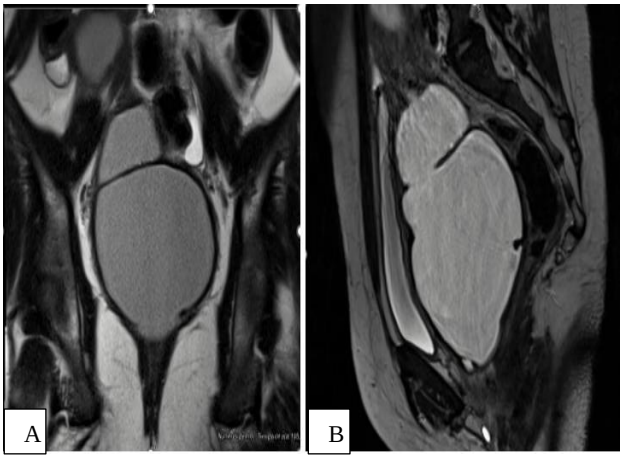

Figure 2 (A and B): Magnetic resonance showing a lobulated vagina, with a 400 cc hematocolpos.

In this clinical context, the patient was scheduled for a vaginal exploration under anaesthesia with simultaneous laparoscopic exploration; at initial visualization, the labia minora were fussioned (Figure 3), there were located a membrane, and the vaginal access began with a vertical incision with the draining of approximately 600 ml of blood, the vaginal walls were dissected finding severe vaginal fusion at the distal third of the vagina, with 2 vaginal synechiae, until the cervix was found; with a 10cm vaginal cavity, the remanent vaginal fold was suture with a 2-0 polyglycolic acid to vaginal walls, a vaginal mold was placed. At the laparoscopic approach, the distended uterus was progressively drained, and the uterine volume was diminished at an exponential rate.

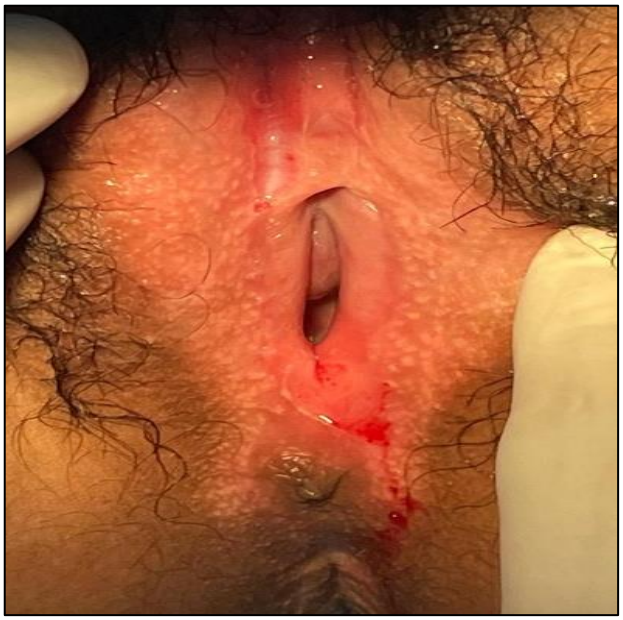

Figure 3: Fusion of labia minora.

On the first post-operative day, the patient presented a spontaneous expulsion of the vaginal mold; a second vaginal exploration was performed with the finding of

vaginal fusion. It has decided the administration of exosomes (ReGenesis®) in vaginal mucosa and a new vaginal mold application with estrogens.

The vaginal mold was extracted at the 48 post-surgeries with dilation by the patient for 2 weeks; after this period, the patient presented regular menstruation that lasted 6 days with normal flow in volume and characteristics. One month later, a simultaneous administration of exosomes and mesenchymal cells in the oral cavity under anesthesia was performed; we found a conserved vaginal patency with 10 centimeters of length (Figure 4), with very few and lax adhesions, an improvement was seen in the oral cavity as well. The patient reported an important recovery.

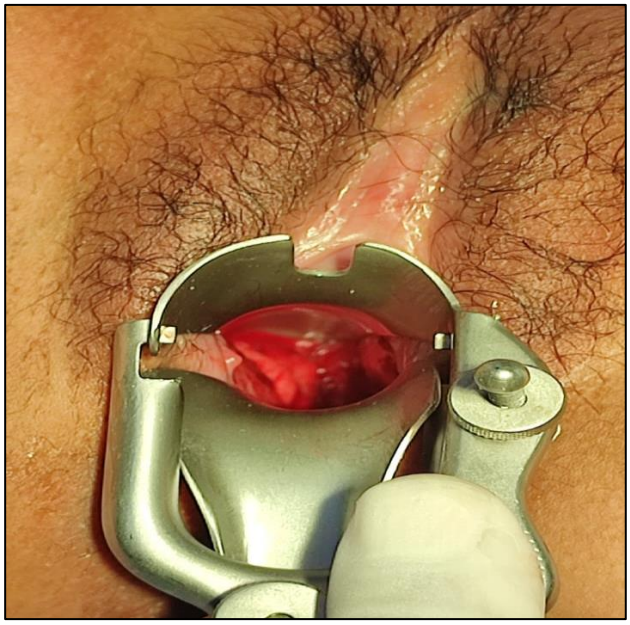

Figure 4: Vaginal length of 10 centimeters at one month later.

DISCUSSION

The female genital GVHD in the pediatric population is reported in very few cases, with an incidence of 5.9%; 42% of patients are asymptomatic, other manifestations like vulvar pain is reported in 37%, dysuria in 37%, and vulvar itching in 26%. Some of the treatments described are corticosteroids such as clobetasol, topical immune modulators such as tacrolimus, vaginal topical estrogens, and, like in this case, exosomes, in severe cases, surgical liberation and lysis of the adhesive tissues.[4] In a study conducted by Yaraghi et al they analyzed 55 patients with vaginal GVHD. Only one patient had vaginal stenosis in two-thirds, while two others had one-third of the upper vagina. Constantini et al reported a case series with eight patients, all above 25 years old, and no pediatric cases were reported; three of these patients presented colpohaematometra with surgical draining; after this management, five patients presented vaginal patency. The other patients lost the follow-up.[6]

A previous gynecological check-up can be recommended for patients treated with HSCT.[4] For the cases with colpohaematometra, a draining and liberation of stenosis can be considered with a complement vaginal dilatation as this case report presents; in the pediatric population, this severe presentation is poorly reported in medical literature, and the spontaneous laparoscopic approach offers a unique opportunity of watching this disease with direct visualization, the surgical technique implied the drainage of a heavy amount of hematocolpos and hematometra, it is essential to mention that the time of progression was only one month of evolution for this patient, it can be inferred that the presence of vaginal synechiae was an essential factor for recurrence.[6] This complication needs to be prevented, and vigilance must be an essential point to watch in the post-transplantation period, as recommended by the national institute of health.[7] Even though this patient is not sexually active, the repercussions for the long term can be significant, and a subsequent follow-up by gynecology must be considered.

CONCLUSION

Vaginal GVHD is a non-infrequent complication related to HSCT; it can vary from vulvar itching to a severe case of vaginal stenosis or fusion, as presented in this case report. A successful approach for this complication can be the liberation and use of vaginal mold with dilatations at the same time with medication like estrogens and treatments like exosomes that began to show promising outcomes for these patients. A gynecological consultation for all patients with HSCT must be in consideration.

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REFERENCES

1. Miano M, Dufour C. The diagnosis and treatment of aplastic anemia: a review. Int J Hematol. 2015;101(6):527-35.
2. George E, Georges R. Stem cell transplantation for aplastic anemia. Int J Hematol. 2001;72(2):141-6.
3. Appiah L. Moravek M, Hoefgen H, Rotz S, Childress K, Samis J. Reproductive late effects after hematopoietic stem cell transplant in pediatric, adolescent, and young adult cancer survivors. Pediatr Blood Cancer. 2023;5:e30551.
4. Yaraghi M, Mokhtari T, Mousavi A, Hazari V. Vaginal complications of graft-versus-host disease after hematopoietic stem cell transplantation: a cross-sectional study. Ann Med Surg. 2012;86(7):3924-8.
5. Shanis D, Merideth M, Pulanic K, Savani B, Battiwalla M, Stratton P. Female long-term survivors after allogeneic hematopoietic stem cell transplantation: evaluation and management. Seminars Hematol. 2012;49(1):83-93.
6. Costantini S, Chiodi S, Spinelli S, Bosi S, Marchiolé P. Fulcheri E. Complete vaginal obstruction caused by

chronic graft-versus-host disease after haematopoietic stem cell transplantation: diagnosis and treatment. J Obstetr Gynaecol. 2044;24(5):591-5.
7. Klasa Ł, Sadowska-Klasa A, Piekarska A, Wydra D, Zaucha J. The management of gynecological complications in long-term survivors after allogeneic hematopoietic cell transplantation-a single-center real-life experience. Ann Hematol. 2020;99(6):1361-8.

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