Membranous dysmenorrhea: a rare or a forgotten entity?
Dismenorreia membranosa: uma entidade rara ou esquecida?

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Abstract

Membranous dysmenorrhea is a rare entity, characterized by the vaginal extrusion of a uterine cavity-shaped structure. We described the case of a healthy 35-year-old woman under combined oral contraception who presented with vaginal extrusion of a decidual cast.

Keywords: Dysmenorrhea, Decidual cast, Hormone therapy.

Resumo

A dismenorreia membranosa é uma entidade rara caracterizada clinicamente pela extrusão por via vaginal de uma estrutura com formato de molde uterino. Apresenta-se o caso clínico de uma mulher de 35 anos, saudável, sob contraceção oral combinada com extrusão vaginal de um molde decidual.

Palavras-chave: Dismenorreia, Molde decidual, Terapia hormonal.

INTRODUCTION

Membranous dysmenorrhea is a rare entity, first described in the 18th century, being a subtype of dysmenorrhea, clinically characterized by the extrusion through the vagina of a uterus mold-shaped structure[1,2]. Epidemiologically, there are no data on its prevalence or incidence, but it must be underdiagnosed, having as a possible cause the lack of knowledge about this entity and the absence of anatomopathological study[2].

Most of the cases described in the literature are associated with hormonal treatment and it is thought that they are associated with endometrial stimulation by unregulated progesterone[1,2]. It may also be associated with 1st trimester miscarriages or ectopic pregnancy[1]. Histologically it is defined as decidualized endometrial stroma, with the absence of other cell lineages[1,2].

CLINICAL CASE

Afro-Brazilian female, 35 years old, multiparous, with no relevant personal or surgical history, medicated with combined oral contraception (Gestodene 0.075 mg + Ethinylestradiol 0.03 mg), with regular withdrawal bleeding, lasting 2-3 days, without associated dysmenorrhea. She went to the Emergency Service of Gynaecology referring to withdrawal bleeding, lasting for two days, in slight amounts and an acute episode of intense dysmenorrhea, associated with spontaneous vaginal expulsion of a red fragment of spongy tissue, which had the shape of the uterine contour (Figure 1). The genital exam was normal and the

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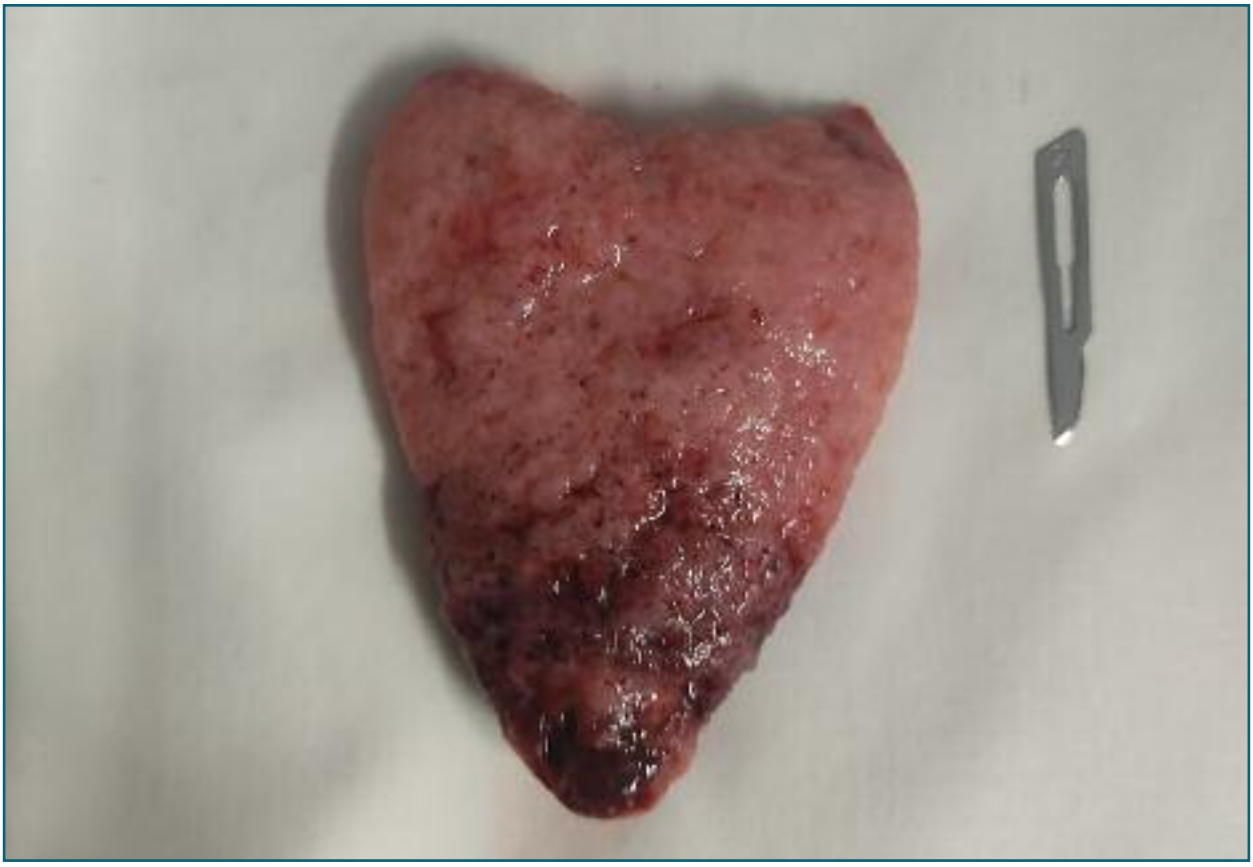

FIGURE 1. Decidual mold (fresh) measuring 7.1×5.5 cm with the shape of the uterine contour.

gynaecology ultrasound showed a virtual uterine cavity with a 4 mm thick endometrium. A Beta--human Chorionic Gonodotrophin (hCG) assay was performed to exclude pregnancy, which was negative and the spongy tissue was sent for histopathology exam. The patient was discharged and referred to Gynaecology outpatient Unit. The histological result showed a mass measuring 7.1×5.5 centimeters, the presence of predecidual stromal cells with a predominance of lymphocytes and a reduced epithelial component, composed by tubular endometrial cells, without evidence of hyperplasia or atypia; the presence of chorionic villi was excluded, and the histological findings were compatible with a decidual mold. At the reassessment consultation, the patient was asymptomatic, without recurrence of the episode and satisfied with the contraceptive method she used prior to the episode of membranous dysmenorrhea.

DISCUSSION

Membranous dysmenorrhea is described in the literature as a rare cause of secondary dysmenorrhea[1]. The pain is explained by detachment and passage of the decidual mold through a non-dilated cervix[2]. Its prevalence is unknown, being described in textbooks and some case reports in the literature. Like the clinical case presented, it is more prevalent in women aged between 20 and 40 years, but there are cases described in adolescence[2,3].

The cause of membranous dysmenorrhea is still unknown, however, there are some mechanisms that can explain the development of this entity. It seems to be associated with increased estrogen and progesterone, with hyperprogesteronism figuring to be the main risk factor, or a result of withdrawal of exogenous hormones in combined oral contraceptive users[3,4].

High levels of progesterone seem to contribute to the transcriptional activation of corticotrophin-releasing hormone, leading to greater decidualization of stromal cells and lower levels of relaxin, responsible for endometrium fragmentation[5]. There are theories that indicate an infectious cause and association with increased production of prostaglandins[4,5]. Another possible mechanism for this process of endometrial degradation is attributed to endometrial mast cells that upregulate matrix metalloproteinases, possible contributing to endometrial shedding[3].

Cases described in the literature are scarce and potential risk factors are very variable, reason why no conclusions can be reached about its etiology[2]. There are reported cases of patients without hormone therapy, women with ectopic pregnancies, women on low-dose combined oral contraceptives and progestin only pill[2,5]. Likewise, no recurrence was observed in patients who maintained the same treatment as previously performed[2].

The treatment of membranous dysmenorrhea is controversial due to the unknown etiology and rare recurrences, even maintaining the ongoing therapy[4].

In the clinical case presented, there was no isolated stimulation with progestative and being the first episode reported in this woman, it was decided to maintain combined oral contraception. The presented case aims to remind of a rare entity that should be part of the differential diagnosis in cases of vaginal expulsion of organic tissue in women of reproductive age. If any potentially modifiable risk factor is identified, its removal or replacement may be attempted to reduce the recurrence of this entity, which is rare.

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REFERENCES

1. Sharmila V and Thrinuvukkarasu A. Spontaneous membranous dysmenorrhea: a rare clinical entity. International Journal of Reproductive, Contraception, Obstetrics and Gynecology. 2019. Oct:8(10);4094-4096.

2. Maciel R, Rodrigues S, Inocêncio G, Saraiva J, Montalvão M. Membranous Dysmenorrhea: a rare and unknow entity. Acta Obstet Ginecol Port 2014;8(4):402-404.

3. Mamede R, Costa C, Djokovic D. A rare case of en bloc endometrium shedding. The International Society for Gynecologic Endoscopy. TheTrocar 2023;2(4):32-35.

4. Rabinerson D, Kaplan B, Fisch B, Braslavski D, Neri A. Membranous Dysmenorrhea: the forgotten entity. The American College of Obstreticians and Gynecologists. 1995; Volume 85, Number 5, Part 2, 891-892.

5. Malik M, Adekola H, Porter W, Poulik M. Passage of decidual cast following poor compliance with oral contraceptive pill. Fetal and Pediatric Pathology. 2015.34;103-107.

AUTHORS CONTRIBUTIONS

Conceptualization: Joana Araújo Pereira, Luísa Silva, Rosália Coutada. Methodology: Joana Araújo Pereira Writing – Original draft: Joana Araújo Pereira Writing – Review: Joana Araújo Pereira, Luísa Silva, Rosália Coutada.

INFORMED CONSENT

The patient gave informed consent for publication.

CONFLICTS OF INTEREST

Authors declare they have no conflicts of interest.

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