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Unusual Case of Vaginal Bleeding in A Teenager Caused by Endometrial Cast: Case Report and Literature Review

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Abstract

Introduction

Endometrial cast, a rare occurrence in gynaecology, is the expulsion of the near entirety of the endometrial lining of the uterine cavity, often maintaining the shape of the uterus. Also known as membranous dysmenorrhoea and largely associated with use of a progesterone-containing contraceptive; some cases following oral contraceptive pills (OCP) use in adolescence for the management of heavy menstrual bleeding (HMB), have been reported.

Case Report

A 16-year-old nulliparous girl presented with a sudden passage of tissue per vagina (PV) following a short duration of PV bleeding and short-lasting crampy abdominal pain, after a 3-month regular use of Rigevidon® (a combined oral contraceptive pill (OCP) containing ethinylestradiol and levonorgestrel): a contraceptive choice. Noted was a previously normal menstrual cycle following menarche at age 12 with an unremarkable past medical history, a negative urine pregnancy test and an empty uterus on ultrasound scan. Histological examination of the 14cm-long flat fleshy mass with an intrauterine outline confirmed an endometrial or decidual cast.

Discussion

Cast formation, with different underlying mechanisms – inadequate estrogen opposition effect on endometrium from dysfunctional oestrogen-progesterone imbalance +/- dysfunctional progesterone-mediated cellular Tissue inhibitors to- and matrix metallopeptidases (TIMP/MMP) – has been associated with HMB, ectopic gestations, miscarriages and secondary postpartum haemorrhage.

Our case had no preceding significant risk factors and inadequate sonographic-evidence- a common occurrence in literature. This led to diagnostic and management challenges, increasing anxiety and treatment morbidity.

Management in young women should be expectant +/- hormonal treatment. Reassurance with individualised progestogens type and dose will aid contraceptive-compliance.

Conclusion

While endometrial/decidual cast is not associated with a deleterious health effect, it can be distressing to patients and a diagnostic challenge to clinicians, unfamiliar with it. More research into underpinning pathways and disease associations is needed.

Keywords: *Adolescent, Menorrhagia, Contraception, Progestogens, Progesterone, Anxiety, Anovulation, Decidualization*

Introduction

Passage of tissue per vagina is not an unusual presentation in gynaecology especially in the context of an early pregnancy. However, differential diagnoses of such a presentation include aborted pregnancy, sarcoma botryoides, polyp, and very

rarely endometrial/decidual cast [1,2]. An endometrial cast is the expulsion of the near entirety of the endometrial lining of the uterine cavity, often maintaining the shape of the uterus [3]. It is also known as membranous dysmenorrhoea due to the gross appearance of the tissue passed and the pain often associated with its passage.

The human endometrium undergoes cyclical dynamic remodelling, increasing in thickness in response to progesterone secreted by the corpus luteum, in the secretory menstrual phase. This results in a thick glandular vascularised endometrium with white blood cells known as decidua in preparation for the implantation of the embryo. The use of progesterone-containing pills may lead to the possibility of developing endometrial/decidual casts via this mechanism [4,5].

This being the aetiology underpinning the many reported cases of passage of endometrial cast following the use of a progesterone-containing contraceptive in some teenage girls as well as those who have used oral contraceptive pills (OCP) for the management of heavy menstrual bleeding (HMB) [6,7].

Case Report

A 16-year-old nulliparous girl presented to the Emergency Gynaecology Unit with a history of sudden passage of tissue per vagina following a short duration of per vagina bleeding and crampy abdominal pain – both of which spontaneously resolved after complete tissue expulsion. She had been on regular use of Rigevidon® (a combined oral contraceptive pill containing ethinylestradiol and levonorgestrel) for three months as a means of contraception. She has had a normal and regular menstrual cycle since attaining menarche at age 12 and there was no past medical history of note.

On examination, she had a normal body mass index. There was neither pallor nor abdominal tenderness; and speculum examination performed, with consent and a chaperone present, showed a normal appearance of the vulva, vagina and a long, closed cervix with no active bleeding. Her observations were completely normal.

Urine pregnancy test was negative and ultrasound scan showed an empty uterus and normal pelvis. Gross examination of the tissue showed a 14cm long flat fleshy mass with an intrauterine outline. Confirmatory histology has been received.

Discussion

The standard use of the combined oral contraceptive pill is expected to be associated with a withdrawal bleed which most women would describe as a 'period'. However, an unexpected vaginal bleed, more so associated with passage of fleshy tissue per vagina, can be quite distressing especially in a teenager.

Incidence

From the literature, the exact incidence of endometrial/ decidual cast is unknown [8]. Whilst reported cases are from women of the reproductive age group, there seems to be a prevalence amongst teenagers. Cast formation, though a rare occurrence with an obscure aetiology – with a link to oestrogen-progesterone imbalance – has been associated with HMB, following ectopic gestations or miscarriages and, infrequently secondary postpartum haemorrhage [9,10]. There are also case reports of OCP use in teenage girls with pre-existing HMB provoking a cast expulsion [1,2,6]. There is no known familial predisposition to formation of endometrial /decidual casts.

Pathophysiology

There are varying theories underpinning the pathophysiology of endometrial or decidual casts formation. However, we postulate there is some degree of endometrial thickening preceding the

shedding of a cast due to inadequate estrogen opposition.

At the cellular level, Estradiol (E2) brings about epithelial proliferation increasing endometrial thickness during the proliferative phase of the menstrual cycle, then Progesterone (P4) inhibits E2-induced proliferation and allows stromal cells to begin decidualization during the secretory phase. When the tightly regulated balance of epithelial-stromal P4 and E2 signaling is lost, this leads to excessive build-up [11]. This mechanism is seen following menarche when the menstrual cycle is initially anovulatory due to poor development of the hypothalamic-pituitary-ovarian axis. Unopposed oestrogen leads to a thickened endometrium (Figure 1).

Fig 1: Schematic representation of anovulation leading to decidual cast formation under the influence of progestogen

This mechanism can also sometimes be seen in OCP use despite intermittent withdrawal bleed.

This inadequate opposition to estrogen effect is seen in case reports where there was documented poor compliance with OCP use i.e. missed pills.

When exogenous progestogens are introduced, there is a complete endometrial sloughing off, leading to a uterine cast expulsion, which is often distressing, sometimes painful and accompanied by painful heavy menstrual loss.

In addition, we postulate that monophasic contraceptive pills may carry a higher association than multiphasic pills. Analysis of most case reports reporting cast formation after contraceptive pill use with no preceding heavy menstrual bleed might indicate some association as a comprehensive literature review from 1910 till date, reflects a paucity of case reports or series historically during time-periods, multiphasic pills were commonly used. Multiphasic pills designed to mimic the rising progesterone levels in menstrual cycles, may provide adequate estrogen opposition and prevent excessive endometrium build-up, resulting in less incidence of uterine cast formation. Definite causation is yet to be determined and a careful risk assessment is needed to mitigate possible unconfirmed benefits versus the potential side effects, before commencing on Multiphasic pills, particularly in a group of patients whose medication compliance may be under question.

The endometrial / decidual cast associated with ectopic gestations or miscarriages is related to the progesterone effect on the endometrium - leading up to a decidualization as often described on histopathological examination. Cast expulsion after a significant drop in progesterone levels, is then thought to be secondary to inadequate tissue apoptosis and degeneration following the sudden necrosis and detachment of the thickened endometrium: secondary to a possible dysfunction of the matrix metalloproteinases (MMPs) and tissue inhibitors of metalloproteinases (TIMPs), both of which are progesterone-mediated [12].

Risk factors

However, our case had no significant risk factors which would suggest a sudden endometrial hypertrophy with shedding all within three months. Despite being a teenager with a history of short-term OCP use, both of which are common factors in reported cases; this case was unusual as there was no preceding HMB before the use of OCP, primarily for contraceptive benefit.

Rather, the presence of progesterone in contraceptives as well as the state of the endometrium are the factors implicated in cast expulsion. In a case series of six patients with cast expulsion [13], all cases had used a progesterone-containing contraceptive with different delivery methods including oral, transdermal and injectables.

Prevention

Patient counselling about the risk of cast formation with subsequent expulsion, is important. The possibility of additional progestogens in the first few cycles when commencing OCP use in adolescence, resulting in the reduction of likelihood of cast formation should be explored. This individualised risk assessment should be offered with caution.

In addition, the role of multiphasic oral contraceptive pills in preventing this condition, particularly in cases related to OCP use should also be explored. Given the paucity of case reports, implying a reduced prevalence of cast expulsion during the time-period of prevalent decreased use or uptake of multiphasic OCPs, a direct causation should be explored of what

is an observed trend.

Diagnosis

Decidual casts raise a diagnostic dilemma prior to cast-shedding. Sonographic evidence might be lacking in the absence of a polyp, as it was in our case, with investigations and management focused on other causes of secondary menorrhagia thereby increasing anxiety. In literature, there is little evidence supporting the diagnostic role of an ultrasound scan. Features of a thickened echogenic intra-uterine material sometimes with fluid within have been described in some case reports, largely in association with ectopic pregnancies or postnatal events [12]. Other imaging studies found to be useful diagnostic tools include magnetic resonance imaging (MRI) and positron emission tomography (PET) scans; both identified an intra-uterine mass with the latter (PET) scan suggesting a possible malignancy [2]. For a benign condition, one could argue that these latter investigations are unnecessary and expensive unless there are other clinical suspicions that would warrant them. A provisional diagnosis is therefore made after the cast has been expelled and subsequently confirmed following histology, as gross macroscopy can sometimes be misleading given the preceding or surrounding circumstances [11].

Management

An expectant management approach is often adequate, once initial symptoms have resolved. Psychologically, it could be quite troubling for a teenager who has expelled a cast especially as those around her, are unlikely to know the diagnosis, which can be made worse if the patient presents to a clinician who has little suspicion or awareness of the diagnosis. Clinician knowledge and/or experience would be invaluable in reassuring such patients pending histology. Particularly with respect to treatment compliance in women using OCPs as their means of contraception, Reassurance is important given the alarming nature of the event. A surgical curettage is often unwarranted.

Disease Associations

Given the potential association between unopposed oestrogen as seen in some anovulatory cycles and cast formation, could women who have experienced shedding of casts be at risk of developing polycystic ovarian syndrome or metabolic syndrome menstrual disorders in the future? In addition, could an endometrial cast be an early feature of occult disease? As the incidence is relatively small, and no longitudinal studies exist, more research would be needed to identify causation and likely associations.

Conclusion

The incidence of endometrial /decidual casts remains unknown and there is no recognized acceptable way to diagnose it before expulsion. An expectant management often suffices once the cast has been expelled. More awareness is needed amongst clinicians. This awareness should also translate into proper counselling of patients who have been prescribed a progesterone-containing contraceptive as this would prevent any unnecessary anxiety should they experience cast expulsion.

References

1. Şen, Y., Cimbek, E. A., & Uğraş, N. S. (2013). Decidual cast after discontinuation of oral contraceptives use in a young girl. Journal of pediatric and adolescent gynecology, 26(6), e127-e129.
2. Ekmekci, S., & Özer, E. (2016). Decidual cast: A case report. Izmir Dr Behcet Uz Cocuk Hastanesi Dergisi, 6(3).
3. Parkes, P., Trainor, J. L., Raval, M., & Dhar, C. P. (2021). Endometrial cast expulsion: A rare cause of pelvic pain case report and review of the literature. Journal of Adolescent Health, 68(5), 1017-1019.
4. Okada, H., Tsuzuki, T., & Murata, H. (2018). Decidualization of the human endometrium. Reproductive medicine and biology, 17(3), 220-227.
5. Hegazy, A. (2014). Clinical embryology for medical students and postgraduate doctors. Lap Lambert Academic Publishing.
6. Torres, A., Baszak-Radomańska, E., Torres, K., Paszkowski, T., Staśkiewicz, G. J., & Wozniakowska, E. (2009). A case of unusual course of adolescent menorrhagia: decidual cast as a side effect of treatment. Fertility and sterility, 92(5), 1748-e5.
7. Malik, M. F., Adekola, H., Porter, W., & Poulik, J. M. (2015). Passage of decidual cast following poor compliance with oral contraceptive pill. Fetal and pediatric pathology, 34(2), 103-107.
8. Nunes, R. D., & Pissetti, V. C. (2015). Membranous Dysmenorrhea Case Report. Obstet Gynecol Cases Rev, 2, 042.
9. Jyoti, Kumari, S., Kumar, D., Gupta, P., Gupta, N., & Kumar, A. (2019). Recurrent decidual cast with membranous dysmenorrhea. International Journal of Reproduction, Contraception, Obstetrics and Gynecology, 8(2), 738-741.
10. Meena, L., & Budania, S. (2021). A case report

on decidual cast following B-lynch compression suture. International Journal of Reproduction, Contraception, Obstetrics and Gynecology, 10(7), 2921-2924.

11. Al-Sabbagh, M., Lam, E. W. F., & Brosens, J. J. (2012). Mechanisms of endometrial progesterone resistance. Molecular and cellular endocrinology, 358(2), 208-215.
12. Reavey, J. J., Maybin, J. A., & Critchley, H. O. (2019). Physiology of menstruation. Inherited bleeding disorders in women, 29-44.
13. Omar, H. A., & Smith, S. J. (2007). Membranous dysmenorrhea: a case series. The Scientific World Journal, 7, 1900-1903.