Aegean Journal of Obstetrics and Gynecology 5/2

Aegean Journal of Obstetrics and Gynecology

Case Report

Hysteroscopic management of infected and partially discharged desidual cast in an adolescent: A case report and review of the literature in adolescents

Elcin Telli [a, †], Mehmet Surhan Arda [b]

[a] *Department of Obstetrics and Gynecology, Eskisehir Osmangazi University, School of Medicine, Eskisehir, Turkiye*

[b] *Department of Pediatric Surgery, Eskisehir Osmangazi University, School of Medicine, Eskisehir, Turkiye*

A B S T R A C T

Objective: Decidual cast in other words membranous dysmenorrhea is a rare entity. Hereby, we report the first infected case of an adolescent caused by drospirenon containing contraceptive use and managed successfully by hysteroscopy.
Case presentation: A 13 years-old girl started on drospirenon containing contraceptive theraphy since her menarche resulted in menorrhagia and anemia that requires transfusion. On follow up, she developed a partially discharged infected mass revealed to be a decidual cast on pathological review.
Discussion: Membranous dysmenorrhea occurs more frequently in the adolescent and young adult population. All cases in this review were on hormonal theraphy except one occured spontaneously. Cast formation of the immature uterus after menarche is an exaggerated response to dose-independent increased progestin levels.
Keywords: decidual cast; hysteroscopy; infection; membranous dysmenorrhea; oral contraceptives

A R T I C L E I N F O
Doi: 10.46328/aejog.v5i2.148

Article history:
Received: 06 June 2023
Revision received 10 July 2023
Accepted 29 July 2023

Introduction

"Decidual cast'' or ''membranous dysmenorrhea'' is referred to discharge of endometrial cast in the shape of endometrial cavity per the uterine cervix amd vagina [1]. It is a rare entity mentioned in the literature by case reports and case series. The differential diagnosis of tissue passed through vagina in an adolescent includes aborted pregnancy, rhabdomyosarcoma (sarcoma botryoides), foreign body, polyp and a very rarely decidual cast. Pathologic examination is the only way for diagnosis [2]. Theories put forward are due to an increase in the production of estrogen and progesterone, infection, exaggerated decidual reaction of menstrual cycle, intense development of spiral arterioles and high blood levels of endogenous or exogenous progesterone [1]. Menorrhagia is one of the common menstrual disorders encountered in adolescent girls and is diagnosed in 34% to 37% of the adolescent population. Oral contraceptives are frequently used to treat adolescent menorrhagia. Treatment with hormonal contraceptives provides hemostasis and endometrial stabilization [3]. Decidual cast formation can be seen as an unusual side effect during or after cessation of treatment [4]. Hereby we aimed to present an infected and partially discharged case caused by drospirenon containing oral contraceptive use and managed successfully by hysteroscopic approach. enign neoplasms of the ovaries are of epithelial origin in 50%. Mucinous tumors are the second most common type of epithelial tumors and comprise 8–10% of ovarian tumors. They may macroscopically reach massive dimensions, although the size of the tumor is not included as a criterion for malignancy. All of mucinous neoplasms, 80 % are benign. Ovarian mucinous cystadenomas are characteristically unilateral, with only 5% presenting bilaterally [1].

Benign mucinous tumors typically have a lobulated, smooth surface, and contain mucoid material within the multilocular. Laparoscopy has become the standard of care in the management of ovarian cysts, because of the lower morbidity rate, improved postoperative recovery. Conservative procedures such as ovarian cystectomy may be preferred in patients with ovarian benign tumors who desire to retain their fertility. Recurrent mucinous cystadenoma after optimal excision is very rare.However, when faced with a huge mass, saving the ovarian tissue may be difficult. . If the cystectomy procedure is not completed thoroughly, recurrences may occur. The data on recurrence of benign ovarian mucinous cystadenomas are limited. A literature search resulted in 11 cases from the first report in 2001 to the present.
This report a 22-year-old nulliparous. women with a huge benign mucinous cystadenoma managed by laparoscopic cystectomy, followed by recurrence within 2 years. Left salpingo-oophorectomy was performed on a repeat laparoscopy. This report discusses a patient, who underwent a laparoscopic unilateral salpingoophorectomy after recurrent mucinous cystadenoma.

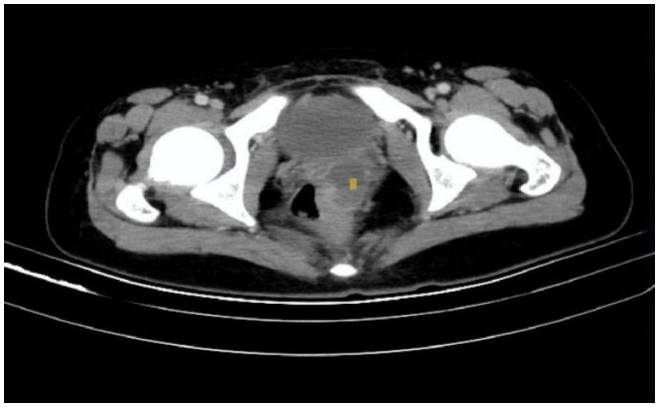
Case Report

A 13 year-old girl presented to our emergency department with a complaint of heavily bleeding lasting for 20 days after menarche. Based on laboratory findings, hemoglobin level was 8.9 gr/dL whereas platelet count, prothrombin time, activated partial prothrombin time, international normalized ratio and bleeding time were in the normal range.

† Corresponding author.
E-mail: drelcinuzmez@hotmail.com
Orcid ID: 0000-0002-3228-8809

Regarding her history, three units of erythrocyte suspension were transfused and she started on drospirenon and ethinyl estrodiol containing oral contraceptive for the treatment of menorrhagia in another medical unit. Her family history was noncontributory. She was consultated by a pediatric hematology department and called for follow up. Her bleeding disorder workup (von Willebrand antigen, factor VIII antigen, coagulation panel, and thyroid panel) was negative. After a month, she admitted to the emergency department with complaints of heavily bleeding, abdominal pain and vomiting on the 6th day of her cycle. Her blood tests revealed anemia (hemoglobin 8.7 gr/dL), leukocytosis (15.4 × 109/L), and elevated acute phase reactants (procalcitonin was 0.25 ng/mL(normal range 0-0.046), CRP was 10.1 mg/L (normal range 0-5)). Serum human chorionic gonadotropin level was within non pregnant levels. On computed tomography imaging, heterogenous fluid collection in the endometrial cavity and cervix was reported (Figure 1).

Figure 1. Computed tomography image of uterus (endometrial cavity is pointed)

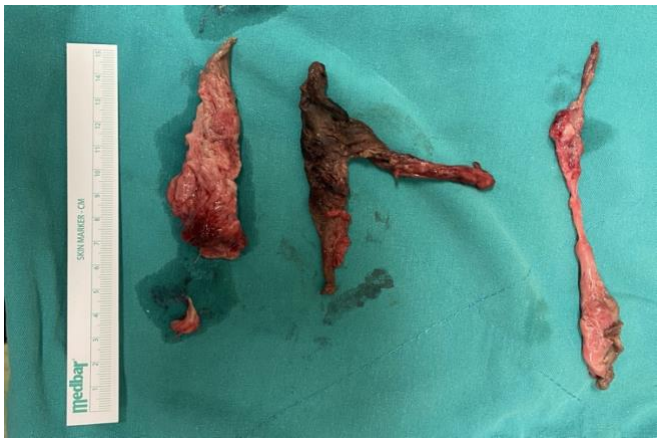


On gynaecologic consultation foul smelling bleeding was recorded. A mass protruding per hymenal ring was inspected. When provoked to cough, a part of the mass passed the hymenal ring and a piece was excised for pathological examination. A transabdominal ultrasound revealed an enlarged uterus(10 × 7 × 6.5 cm) and distended cervix filled with soft tissue extending per vagina. An urgent pathological review revealed decidualizated endometrium. She was hospitalized and administered broad spectrum antibiotherapy. During the 48 hour of theraphy, foul smell disappeared and an examination under general anesthesia was planned (Figure 2).

Figure 2. Adherent mass bulging from vaginal introitus.

On examination, taking care of the hymenal ring, the protruding tissue was carefully held by an instrument and pulled through the vagina. Since the tissue broke off via pulling, a cystoscope no: 11 was inserted through the intact hymenal ring to observe the vagina and the uterine cavity. The remaining part of the tissue through cervical canal was removed spontaneously by the extander effect of running water (Figure 3).

Figure 3. Excised decidual cast material.



On final view, uterine cavity and vagina were normal. She was discharged and oral antibiotheraphy was prescribed for a week. However, she failed to attend her follow up.

Discussion

To the best of our knowledge this is the first case of an infected decidual cast managed successfully by hysteroscopy and also the first case reported while on drospirenon use among adolescents.

On the literature search, terms such as 'decidual cast'' and ''membranous dysmenorrhea'' were used among adolescents (aged 10-19 years) and only 16 cases were found [4-14] (Table 1). The average age of the cases was 14.9 (range 10.8-19) years. Membranous dysmenorrhea occurs more frequently among the adolescent and young adult population. In a review including all ages, 12 out of 21 cases were under the age of 20 [7]. All of the cases were after menarche. Progesterone levels tend to rise incrementally after menarche. Although the progesterone and progesterone containing agents induce decidualization and cycle stability , the mystery of membranous dysmenorrhea still remains unclear [15].Only thirteen cases were found to suffer from pain, a type of secondary dysmenorrhea, which occurs from the passage of tissues through the non-dilated cervix [16].

Physiologic and supraphysiologic doses of progestin causes an exaggerated response of the immature uterus resulting in excessive endometrial growth and cast formation [4-14]. An immature uterus differs from a mature uterus by immature uterine natural killer (uNK) cells. uNK cells matures with each cycle to form effective spiral artery and placental vasculature development during pregnancy. Increased progesterone on uNK cells may lead to an ineffective spiral artery development and makes adolescents more sensitive to form an endometrial cast [5]. Depomedroxyprogesterone acetate has been reported in the decidual cast development and was cited in 50% of reported adolescent cases. Oral contraceptives are related with another 25% of cases whereas the hormonal patch is in 12.5%. All cases were on hormonal therapy except for one that occured spontaneously This demonstrates that cast formation of the immature uterus after menarche is not attributable to the dose given but rather to an exaggerated response due to increased progestin levels [5].

Table1. Demographic and clinical features of the cases

Case	Age (years)	Symptom	Suspicious for Etiology	Pathological confirmation	Additional history/finding	Management	Follow-up
Parkes et al.2021 (5)	12	dysmenorrhea	COC for heavy bleeding	Yes	Menorrhagia	Discontinued COC	NR
Van Gaal et al.2016 (6)	18	painful vaginal tissue loss	COC	Yes	NR	NR	NR
Topçu et al.2015 (7)	17	dysmenorrhea and painful tissue passage	none	Yes	leukocytosis (14.8 × 109/L) attiributed to inflammatory substances of pain	Follow up	8 months, uneventful
Rabinerson et al. 1995 (8)	18	NR	COC	NR	NR	NR	NR
Omar et al.2007 (9)	12	Vaginal tissue discharge and dysmenorrhea	estrogen and progestine patch for dysmenorrhea	Yes	Cerebral palsy, dysmenorrhea	DMPA for dysmenorrhea	3 years, uneventful
Omar et al.2007 (9)	13	Painful vaginal discharge	DMPA for dysmenorrhea	Yes	Dysmenorrhea	Continued to use DMPA	4 years, uneventful
Omar et al.2007 (9)	15	Painful vaginal discharge	Transdermal patch for contraception on the 5th cycle	Yes	NR	Continued to use transdermal patch	NR
Omar et al.2007 (9)	16	Painful vaginal bleeding	DMPA for contraception on 15th month	Yes	NR	Continued to use DMPA	3 years, uneventful
Omar et al.2007 (9)	17	Painful vaginal discharge	COC for contraception on 7th month	Yes	NR	Discontinued COC, started DMPA	2 years, uneventful
Omar et al.2007 (9)	16	Painful vaginal discharge	DMPA postpartum	Yes	NR	Continued to use DMPA	2 years, uneventful
Sen et al.2013 (4)	10.8	Painful vaginal discharge	Oral contraceptive	Yes	Menorrhagia	Discontinued oral contraceptive	4 months, uneventful
Appelbaum. 2010 (10)	16	Cramping, tissue discharge and irreguler bleeding	norethindrone acetate	Yes	Endometriosis and mullerian abnormality	Discontinued norethindrone acetate	Multiple surgeries and hormonal agents for four years
Torres et al.2009 (11)	13	Vaginal tissue discharge after bleeding	monophasic oral contraceptive	Yes	Heavy bleeding	Continued OC until withdrawal then cyclic progestogen	4 months, uneventful
Malik et al.2015 (12)	13	Vaginal tissue discharge after cramping and bleeding	COC on 10th month	Yes	menorrhagia	Discontinued COC	NR
Ekmekci et al.2016 (13)	13	Pain and abnormal bleeding	COC	Yes	Abnormal uterine bleeding	Hysteroscopic view, spontaneous discharge on follow up	NR
Strauss.2018 (14)	19	Spotting,vaginal tissue discharge	DMPA	NR	Sexually active	NR	NR

There are no long-term effects of the situation. The longest follow up period was four years with DMPA. Fourteen out of 16 cases were confirmed by pathology. Treatment options were controversial. Five cases under contraceptives had discontinued taking contraceptive pills probably because of recurrence fear but no recurrences were found despite ongoing hormonal contraceptives or progesterones.As a result, this suggests that patients can safely continue hormonal contraceptive therapy. Only one case report presented the use of hysteroscopy to view the mass that was later spontaneously discharged on follow up [13].

We conclude that membranous dysmenorrhea should be kept in mind as a differential diagnosis of adolescents with lower abdominal pain, especially for ones under hormonal contraceptive therapy. The infection risk of undischarged tissue is not rare and may cause a misdiagnosis of malignancy and treatment delay. Hysteroscopic approach is safe for partially discharged cases.

Disclosure

Authors have no potential conflicts of interest to disclose.

References

[1] Greenblatt, R. B., Hammond, D. O., & Clark, S. L. Membranous dysmenorrhea: studies in etiology and treatment. American journal of obstetrics and gynecology.1954; 68(3):835-844.

[2] Badalyan V, Burgula S, Schwartz RH. Congenital paraurethral cysts in two newborn girls: differential diagnosis, management strategies, and spontaneous resolution. J Pediatr Adolesc Gynecol.2012;25(1):e1-e4.

[3] Borzutzky C, Jaffray J. Diagnosis and Management of Heavy Menstrual Bleeding and Bleeding Disorders in Adolescents. JAMA Pediatr. 2020;174(2):186-194.

[4] Sen Y, Cimbek EA, Uğraş NS. Decidual cast after discontinuation of oral contraceptives use in a young girl. J Pediatr Adolesc Gynecol. 2013;26(6):e127-9

[5] Parkes P, Trainor JL, Raval M, Dhar CP. Endometrial Cast Expulsion: A Rare Cause of Pelvic Pain Case Report and Review of the Literature. J Adolesc Health.2021;68(5):1017-1019.

[6] van Gaal N, van Krimpen CK, Zwart CA. Een jonge vrouw met abnormaal vaginaal weefselverlies [A young woman with abnormal vaginal tissue loss]. Ned Tijdschr Geneeskd. 2016;160:A9515.

[7] Topçu HO, Topçu S, Kokanalı D, Memur T, Doğanay M. Spontaneous Membranous Dysmenorrhea in an Adolescent Girl: A Case Report and Literature Review. J Pediatr Adolesc Gynecol.2015;28(5):e139-41.

[8] Rabinerson, D., Kaplan, B., Fisch, B., Braslavski, D., & Neri, A. Membranous dysmenorrhea: the forgotten entity. Obstetrics & Gynecology.1995;85(5): 891-892.

[9] Omar, Hatim A., and Shawn J. Smith. Membranous dysmenorrhea: a case series. TheScientificWorldJournal.2007;7:1900-1903.

[10] Appelbaum, H. Membranous dysmenorrhea: a complication of treatment for endometriosis. Obstetrics & Gynecology.2010;116(Part 2): 488-490.

[11] Torres, A., Baszak-Radomańska, E., Torres, K., Paszkowski, T., Staśkiewicz, G. J., Wozniakowska, E. A case of unusual course of adolescent menorrhagia: decidual cast as a side effect of treatment. Fertility and sterility.2009;92(5):1748-e5.

[12] Malik, M. F., Adekola, H., Porter, W., & Poulik, J. M. Passage of decidual cast following poor compliance with oral contraceptive pill. Fetal and Pediatric Pathology.2015;34(2):103-107.

[13] Ekmekci, S., Özer, E. Decidual cast: A case report. Izmir Dr Behcet Uz Cocuk Hastanesi Dergisi.2016;6(3):234-36.

[14] Strauss, L. Fleshy mass passed vaginally by a young woman. American Family Physician.2018;98(7):449-450.

[15] Brosens I, Muter J, Gargett CE, Puttemans P, Benagiano G, Brosens JJ. The impact of uterine immaturity on obstetrical syndromes during adolescence. Am J Obstet Gynecol. 2017;217(5):546-555.

[16] Upasham, P. S., Sirmukaddam, S. V., & Sharan, A. Dysmenorrhea membranacea: A case report and review of the literature. Fertility Science and Research.2014; 1(1):56.