Case Report

Spontaneous rupture of cervical hydatidiform molar pregnancy in 53 years old woman: A case report and brief review of the literature

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Abstract
The incidence of molar pregnancy and ectopic pregnancy are 1 and 10 per 1000 pregnancies, respectively. An ectopic molar pregnancy is a very rare occurrence. The differentiation of molar ectopic and non-molar ectopic pregnancy is very difficult. A 53 years old gravida 5, parity 2, dilatation and curettage 1, abortus 1 woman was referred from emergency service due to excessive vaginal bleeding and pelvic pain. Pelvic examination revealed significant vaginal hemorrhage from a lesion on anterior cervical lip. Quantitative β-hCG test was 41.842 mU/ml. Ultrasonographic findings revealed no bilateral adnexal or uterine pathology but cervical ectopic pregnancy. As the patient’s hemoglobin level was 4.7 gr/dL, total abdominal hysterectomy and bilateral salpingo-oophorectomy was performed immediately. Final pathologic examination revealed cervical hydatiform molar pregnancy. No peri or postoperative complication was detected and the patient was uneventful.

Key words:
Pregnancy, cervical ectopic pregnancy, uterine cervix, hydatiform mole

Introduction
Ectopic pregnancy occurs when the developing blastocyst becomes implanted at a site other than the endometrium of the uterine cavity. The prevalence of ectopic pregnancy among women who go to an emergency department with first trimester bleeding, pain, or both ranges from 6 to 16 % [1]. However, the overall incidence of ectopic pregnancy increased during the mid-twentieth century, plateauing at approximately almost 20 per 1000 pregnancies in the early 1990s, the recent national data were reported by the Centers for Disease Control [2]. Previous ectopic pregnancy, tubal pathology and surgery, in-utero DES exposure, previous genital infections, intrauterine devices, infertility, multiple sexual partners, smoking, in vitro fertilization, vaginal douching, age are known risk factors for ectopic pregnancy.

Cervical pregnancy is a rare form of ectopic pregnancy in which the trophoblast implants in the cervical tissue of the endocervical canal. Cervical pregnancy incidence is approximately 1/9000 [3,4]. Although the etiology is still unknown, there is evidence for its association with cervico-uterine instrumentation [3], and in particular vaginal termination of pregnancy [5]. Several case reports have suggested that invitro fertilization may also increase the risk of cervical ectopic pregnancy [6,7].

Gestational trophoblastic disease is a proliferative disorder of trophoblastic cells. It defines a heterogeneous group of interrelated lesions arising from the trophoblastic epithelium of the placenta. The World Health Organization classification of gestational trophoblastic disease includes complete and partial hydatiform mole, invasive mole, choriocarcinoma, placental site trophoblastic tumor, epitheloid trophoblastic tumor, exaggerated placental site and placental site nodule [8].

Hydatiform molar degeneration of the placenta tissue can be classified in partial and complete hydatiform mole pregnancy. Hydatiform mole pregnancy is considered as a sporadic and genetic disorder with a recurrence rate of 1%. Risk factors for the development of hydatiform molar pregnancy are maternal age, history of previous molar pregnancy, smoking, alcohol abuse, oral contraceptives [9,10]. Normally molar pregnancy presents in the first trimester and is associated with a wide array of clinical symptoms, most commonly vaginal bleeding in combination with excessive β-HCG levels. However at the time of presentation, 60 % of molar gestation had ruptured [11].

Cervical hydatiform molar pregnancy is not common and only four case reports are presented in the literature [12-15]. Here is a presentation one more case of cervical molar pregnancy in a 53 year old woman.
Case presentation

A 53 years old gravida 5 parity 2 dilatation and curettage 1, abortus 1 woman was admitted to our emergency department with complaint of vaginal bleeding for 15 days and pelvic pain. Pelvic examination revealed significant vaginal hemorrhage from ruptured lesion on cervical lip. Ultrasonographic findings revealed no bilateral adnexal or uterine pathology but cervical ectopic pregnancy. Materials in the ruptured lesion on cervical lip were removed for pathologic examination and gentle aspiration curettage was performed to cease bleeding. As the patient’s quantitative β-hCG test was 41.842 mU/ml and hemoglobin level were 4.7 gr/dL, total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed. On gross examination 2x1 cm area on the serosal surface of left side of cervix was ruptured (Figure 1). Microscopic evaluation revealed endoservical glands and stroma consisting of large cells with eosinophilic cytoplasm with hyperchromatic nuclei (Figure 2). Immunohistochemically, these cells expressed cytokeratin AE1-AE3 but not vimentin (Figure 3). The findings supported that these cells were trophoblastic in nature. Histologically, materials derived from ruptured cervical lesion showed cisternal formations in the stromal parts of the chorionic villi. Moreover, there were significant abnormal proliferations in the trophoblastic cells with areas displaying circumferential growth from the villous surface (Figure 4). Five weeks after the operation, β-HCG level was 4.7mU/ml and the patient was uneventful.

Discussion

Incidence of cervical ectopic pregnancy is approximately 1/9000 [3, 4]. It may be fatal due to excessive vaginal bleeding; fortunately, early diagnosis and effective treatment methods significantly decrease the mortality rates. No recent deaths due to cervical pregnancy have been reported [1]. Ectopic molar pregnancy is an extremely rare condition. The incidence of ectopic gestational trophoblastic disease can be estimated at approximately 1.5 per 100000 births [11].

To the best of our knowledge, this is the fifth case of cervical molar pregnancy in PUBMED as shown in Table 1 [12-15]. Two of these cases were partial molar pregnancy [12, 14]. In these two cases, dilatation and curettage was performed and bleeding ended after operation. The third case was a complete hydatiform molar pregnancy of the cervix presenting with vaginal bleeding. In this case methotrexate treatment was attempted first, but heavy hemorrhage necessitated evacuation. Moreover, patient had demonstrated raised β-HCG levels that remained elevated for up to 4 weeks, was given a chemotherapy treatment consisting of seven successive doses of methotrexate given in combination with folinic acid [13].

Fourth case was complete molar ectopic pregnancy localized to the ectocervix. This case occurred two months after curettage of a missed abortion [15]. As the patient in the present case was hemodynamically unstable and in menopausal transition period, total abdominal hysterectomy and bilateral salpingo-oophorectomy was performed.

In conclusion, physicians should keep in mind that, cervical ectopic molar pregnancy may cause excessive vaginal bleeding in a woman even at her fifth decade.
Case presentation

β-HCG level was 4.7 mU/ml and the patient was uneventful. Five weeks after the operation, abnormal proliferations in the trophoblastic cells with cisternal formations in the stromal parts of the lesion showed that these cells were trophoblastic in nature. Microscopic evaluation revealed endosalpingeal glands and stroma consisting of large cells with eosinophilic cytoplasm with hyperchromatic nuclei. Immunohistochemistry for cytokeratin AE1-AE3 but not vimentin (Figure 3). The findings were consistent with a diagnosis of complete molar pregnancy.

Conflict of interest statement
The authors declare no conflict of interest.

<table>
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<tr>
<td>K Chapman [12]</td>
<td>35</td>
<td>Elevated vaginal bleeding for the last 2 days that has been occurring for the past 3 weeks, lower abdominal pain</td>
<td>Partial mole</td>
<td>D&amp;C</td>
<td>After 6 weeks, β-hCG &lt; 5 mIU/L</td>
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<td>Wee et al., [13]</td>
<td>36</td>
<td>Heavy vaginal bleeding following the evacuation done 2 weeks ago</td>
<td>Complete mole</td>
<td>Mtx, evacuation, and bimanual compression. Afterwards, chemotherapy with mtx, 7 cure.</td>
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<tr>
<td>Aytan et al., [14]</td>
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<td>Heavy vaginal bleeding</td>
<td>Partial mole</td>
<td>D&amp;C, bimanual compression</td>
<td>3 weeks later, β-hCG &lt; 5 mIU/ml</td>
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<tr>
<td>Schwentner et al., [15]</td>
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<td>Heavy vaginal bleeding following the abortion done 2 months ago</td>
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<td>Exfoliation and conservative surgery</td>
<td>After 3 months, beta-β-hCG undetectable.</td>
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<td>Heavy vaginal bleeding</td>
<td>Complete mole</td>
<td>TAH+BSO</td>
<td>After 5 weeks, β-hCG &lt; 5 mIU/ml</td>
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D&C = Dilatation and curettage, Mtx = Methotrexate, TAH+BSO = total abdominal hysterectomy and bilateral salpingo-oophorectomy
References