

# Cervico-isthmic ectopic pregnancy: A case report

Makukhina Tatiana, Knyazeva Natalia

## ABSTRACT

**Introduction:** Due to the rarity of cervico-isthmic ectopic pregnancy (CIP) the specialists are insufficiently informed about the diagnosing methods and preferable treatment patterns. **Case Report:** We report a case of misdiagnosed CIP manifested by repeated heavy bleeding after curettage. The patient hospitalized at Perinatal Center with complaints of vaginal spotting. Ultrasound scan showed shortened cervix, chorion frondosum presentation just above cervical M-echo and subchorionic hematoma in endometrial cavity above the gestation sac. Attempt of medicamentally induced abortion failed, and dilatation and curettage (D&C) were performed. Massive vaginal bleeding suddenly occurred two weeks later. Trophoblastic invasion into cervix by color Doppler was suspected. Combined management with systemic methotrexate administration and bilateral uterine artery embolization (UAE) was used. Four days after D&C under US control was performed again and no pathology was found two weeks later. The diagnosis could have been formulated on the first admission at the hospital, in spite of low implantation and chorion frondosum presentation. Retrospective analysis of US-video files just before the patient discharge

the Perinatal Center shows the high echogenic lesion in the cervico-isthmic area, which was considered a blood clot, but was not confirmed by color Doppler. Only combinations of methods used (UAE, methotrexate administration with subsequent curettage) stopped bleeding. **Conclusion:** Early diagnosis of CIP requires US mapping of low implantation. The question of CIP prolongation should be decided individually. CIP termination needs hemorrhage prevention. color Doppler is important as a diagnostic and monitoring tool to exclude chorion persistence after treatment.

**Keywords:** Cervico-isthmic ectopic pregnancy, Chorion persistence, Color Doppler, Ultrasound diagnosis

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## INTRODUCTION

Due to the rarity of cervico-isthmic ectopic pregnancy the specialists are insufficiently informed about the diagnosing methods and preferable treatment patterns.

## CASE REPORT

A 33-year-old female hospitalized at a regional Perinatal Center with complaints of vaginal spotting.

She gave one term birth 10 years ago, two first-trimester miscarriages three and one years ago. Oligomenorrhea caused by uterine synechiae has developed afterwards and was surgically destroyed twice, the last time - two months before the described pregnancy.

On admission, the Ultrasonography (US) showed gestational sac (GS) with embryo CRL 16 mm without heartbeat, cervix shortened up to 21 mm, chorion frondosum presentation just above cervical M-echo and subchorionic hematoma in endometrial cavity above the GS (Figure 1 a,b). Clinical diagnosis: “Missed recurrent miscarriage of eight-week uterine pregnancy. Subchorionic hematoma”.

Attempt of medicamentally induced abortion failed, and D&C were performed. Instantaneous blood loss of 600 ml took place. Vaginal packing was conducted. In 24 hours the hemorrhage was repeated and one unit of fresh frozen plasma and HES-INFUZIA 6% 500 ml were transfused. According to the histological examination «Missed pregnancy. Trophoblast proliferation, with possible partial hydatid mole” and the second-opinion pathologist confirmed it. At the day of discharge (13th after D&C) blood test showed hemoglobin 8.8 g/dl,  $\beta$ -hCG level 1601, 5 mIU/ml. Endometrial cavity thickening was 4 mm, M-echo of cervix was 14 mm (Figure 2a).

Two days later massive vaginal bleeding suddenly occurred and patient presented at the emergency gynecology department of the Municipal Hospital. Due to heavy bleeding US was performed on portative scanner so the images were not stored. The endometrial cavity appeared to be empty; in the proximal part of cervix tissue of heterogeneous background with irregular cystic areas volume 45 ml with abundant low-resistant blood flow was found. Outlines of cervix were clear and surrounded tissues were intact. Trophoblastic invasion was suspected. After the informed consent, emergency UAE under epidural anesthesia was performed. Also methotrexate was administered systemically intramuscular at 50 mg per m<sup>2</sup> of body square area. The patient ceased bleeding.

Four days after UAE  $\beta$ -hCG dropped to 106 mIU/mL, focal lesion volume decreased to 8.5 ml. and peripheral blood flow was persistent (Figure 3a–b). The D&C under US control was repeated. Pathological report: “Decidual tissue and chorionic villi. Hydatid mole not found”. The independent pathology report confirmed it. Final diagnosis according to ICD-10-CM was “O00.8. Complications following ectopic and molar pregnancy.”

Outpatient follow-up: volume of hematocervix decreased (Figure 4). Further normal US appearance and subliminal  $\beta$ -hCG level were documented two weeks after hospital discharge.

## DISCUSSION

Cervico-isthmic ectopic pregnancy (CIP) is more common whereas true cervical pregnancy is rare and in addition more lethal as the maternal risk of massive

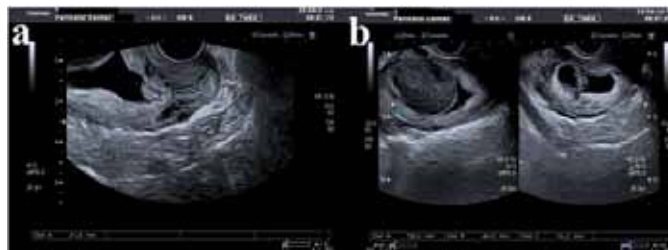


Figure 1: Ultrasound on the day of the patient admission at the Perinatal Center: (a) cervix shortening, chorion frondosum presentation just above cervical M-echo, (b) subchorionic hematoma in uterine cavity above the gestation sac.



Figure 2: Ultrasound just before discharge of the patient from the Perinatal Center: (a) Absence of cervical contraction after D&C, (b) Heterogeneous tissue in the cervico-isthmic area (arrow).

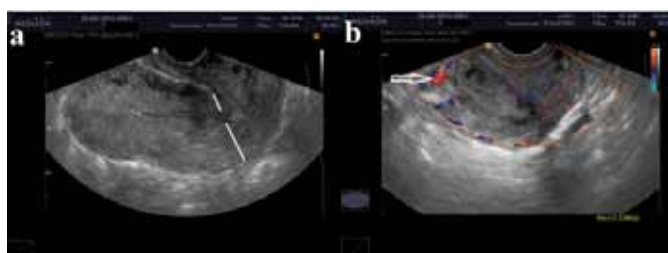


Figure 3: Ultrasound after UAE and methotrexate administration before repeated D&C in the Municipal Hospital: (a) Asymmetry of the walls thickened at the isthmus level (lines), (b) Persistent peripheral blood flow (arrow).

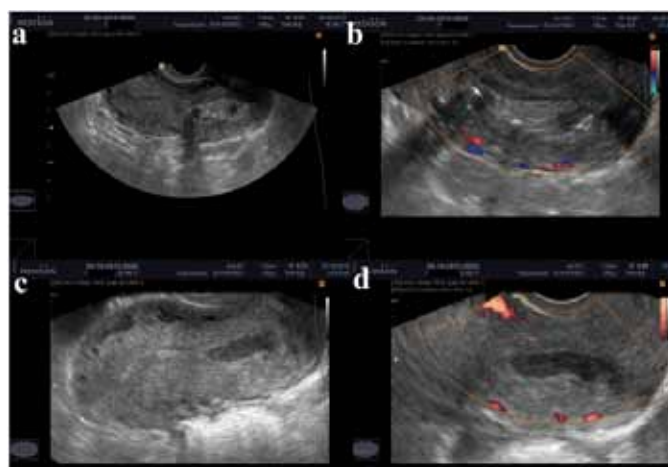


Figure 4: Ultrasound after repeated D&C: (a, b) one day later: cervical canal dilated up to 13 mm with heterogeneous avascular masses volume up to 4 ml; (c, d) 10 days later: volume of hematocervix decreased up to 1 ml.

hemorrhage increases with the advancement of the gestational period [1, 2]. Up to the present day, no consensus exists for the terminology, diagnostic criteria and disease management. A number of authors [3, 4] stress the importance of formulating US diagnosis for treatment policy. Due to the rarity of this pathology, most specialists have little personal experience of it [4, 5]. We reviewed current medical literature by searching PubMed files using the keywords “cervico-isthmic pregnancy” finding 24 sources: 21 publications were case reports, 2 sources – without abstracts, one prospective study [4].

CIP is viewed as a variation of ectopic pregnancy [3–6]. Some regard CIP as a sub-type of cervical pregnancy (CP) [2], or “intrauterine ectopic pregnancy” [7], others associate this localization with cesarean scar pregnancy (CSP) as pregnancy located below the internal os [3–5].

Recent advances in high-resolution ultrasound have made the diagnosis of early CIP easier [4, 6]. Whereby it is important to distinguish cervico-isthmic and true CP [8], because without connection between GS and endometrial cavity prolongation of pregnancy until the terms of fetus viability is impossible and life-threatening. Sonographic criteria of CIP include shortened cervix [8], low implantation [6, 8], ‘empty endometrial cavity’ [6, 8]. Jurkovic D. [5] specifies criteria for CP and CSP: GS located below the level of the internal os; evidence of functional trophoblastic/placental circulation on color Doppler examination; negative ‘sliding organs sign’, defined as the inability to displace the GS from its position at the level of the internal os. In the difficult situations, the use magnetic resonance imaging is recommended [8].

Color Doppler may be useful for differentiation between cervical phase of intrauterine miscarriage and ectopic pregnancy [5]. Power Doppler provides additional information for monitoring and quantifying neovascularization change for the initial diagnosis and during follow-up period but is not essential in most cases for the initial diagnosis [4].

The optimal management has not yet been established [6] because of the rarity of the condition. Reports of successful CIP leading to delivery of healthy neonates are present in the literature; almost all cases require blood transfusion and hysterectomy [1, 2]. Only two reports of cases of delivery with preservation of uterus were found [9, 10]. Early diagnosis of CIP leads to conservative management helping to preserve the woman’s future fertility [6, 7].

The present case report had the “classical” CIP signs: catastrophic hemorrhage in attempt of curettage, repeated incidents of bleeding despite conservative therapy, absents of cervical contraction, persistent chorion villi into cervix confirmed histologically. Only combinations of methods used (UAE, methotrexate administration with subsequent curettage) stopped bleeding.

The diagnosis could have been formulated on the first admission at the hospital, in spite of low implantation and chorion frondosum presentation, which suggested its growing into cervix. Although there is evidence of

successful curettage as an independent treatment method of CIP [4], it is associated with high risk of massive bleeding because of rich blood supplies, and necessary to provide preventive hemostasis [3]. Persistent high  $\beta$ -hCG level and absence of cervical contraction needs Color Doppler, which would allow to detect trophoblastic tissue at the cervix and reconsider the treatment policy. Retrospective analysis of US-video files just before the patient discharge the Perinatal Center show the high echogenic lesion in the cervico-isthmic area, which was considered a blood clot, but was not confirmed by color Doppler (Figure 2b).

## CONCLUSION

For early diagnosis of cervico-isthmic ectopic pregnancy (CIP) suspected during the US due to cervix shortening and low implantation it is necessary to map out the point of bifurcation a. uterine to specify the internal os placement: placental involvement of the cervix can cause erosion of cervical blood vessels and massive bleeding may be present. The ‘empty endometrium cavity’ sign is not obligatory: subchorionic hematoma may fill the cavity, as well as submucosal myoma. The question of viability of CIP prolongation should be decided individually. Information on ectopic implantation should be present in the US conclusion, as, unfortunately, lots of obstetricians do not read the description. CIP termination needs hemorrhage prevention measures depending on the technical infrastructure of the hospital. Color Doppler is important as a diagnostic and monitoring tool to exclude chorion persistence after treatment.

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## Author Contributions

Makukhina Tatiana – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Knyazeva Natalia – Acquisition of data, Drafting the article, Final approval of the version to be published

## Guarantor

The corresponding author is the guarantor of submission.

## Conflict of Interest

Authors declare no conflict of interest.

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