

# Cervical ectopic pregnancy: a case report and review of the literature

## Ektopinis nėštumas gimdos kaklelyje: klinikinis atvejis ir literatūros apžvalga

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### Background

Cervical pregnancy is a rare pathology with unclear etiology. This condition can be life-threatening and in certain situations be treated with total abdominal hysterectomy, although due to improving diagnostics, approach to cervical pregnancy's management has been changing towards fertility preserving treatment.

### Case report

38 years old patient with vaginal bleeding and lower abdomen pain was admitted to Klaipėda Seamen's hospital. After thorough evaluation, clinical diagnosis of cervical pregnancy was confirmed. Different approaches were considered and total abdominal hysterectomy was determined as most suitable method. Surgery was successful and post-operative period was uneventful.

### Discussion

Ultrasonography is the main diagnostic tool used to observe cervical ectopic pregnancy. Speculum and bimanual examination usually show prominent findings, assisting cervical pregnancy's diagnose. Prior to improved diagnostic tools, total abdominal hysterectomy was the main approach treating cervical pregnancy. Now conservative management using methotrexate is considered as the first-line treatment and frequency of other fertility preserving procedures as suction curettage and tamponade or uterine artery embolization are increasing.

### Conclusion

Data is limited because of cervical pregnancy's rarity and every case of this condition has to be assessed separately. Desire to preserve fertility and patient's condition must be taken to account before making decision on how cervical pregnancy will be managed.

**Key words:** cervical pregnancy, ultrasonography, total hysterectomy

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## **Įvadas**

Nėštumas gimdos kaklelyje yra reta patologija, kurios tiksli etiologija nežinoma. Ši būklė gali būti gyvybiškai pavojinga ir gydoma totalia laparotomine histerektomija, tačiau tobulėjanti diagnostika leidžia ektočinį nėštumą gimdos kaklelyje gydyti išsaugant vaisingumą.

## **Klinikinis atvejis**

38 metų pacientė buvo paguldyta į Klaipėdos Jūrininkų ligoninę dėl kraujavimo iš makšties ir skausmo apatinėje pilvo dalyje. Nuodugniai ištyrus pacientę, buvo diagnozuotas nėštumas gimdos kaklelyje. Apsvarsčius skirtingas gydymo taktikas, totali laparotominė histerektomija buvo pasirinkta kaip tinkamiausias gydymo būdas. Operacija buvo sėkminga, pooperacinis laikotarpis sklandus.

## **Diskusija**

Ultragarso tyrimas yra pagrindinis nėštumo gimdos kaklelyje diagnostikos metodas. Skėtikliais ir bimanualinio tyrimo metu dažniausiai randami svarbūs pakitimai, kurie padeda pagrįsti diagnozę. Totali laparotominė histerektomija buvo pagrindinis gydymo metodas, tačiau tobulėjant diagnostikos metodams metotreksatas tapo pirmos eilės pasirinkimu kaklelinio nėštumo atveju. Kiti konservatyvūs gydymo metodai, pavyzdžiui, gimdos ir gimdos kaklelio abraziacija, lydima tamponavimo Foley kateeteriu, ar gimdos arterijos embolizacija, tampa vis populiariesni.

## **Išvados**

Duomenys apie kaklelinį nėštumą yra riboti dėl šios patologijos retumo ir kiekvienas atvejis turi būti vertinamas individualiai. Noras išsaugoti vaisingumą ir pacientės būklė yra pagrindiniai rodikliai, į kuriuos būtina atsižvelgti nusprendžiant, kokią gydymo taktiką pasirinkti esant nėštumui gimdos kaklelyje.

**Reikšminiai žodžiai:** nėštumas gimdos kaklelyje, ultrasonografija, totali histerektomija

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## **Introduction**

Cervical pregnancy (CP) is a rare ectopic pregnancy with an incidence ranging from 1:2500 to 1:18 000 and occurs in less than 1% of all ectopic pregnancies [1–3]. Etiology of CP is still not clear, but previous miscarriage treated by dilatation and curettage is associated with up to 70% of the cases [3]. Other risk factors include caesarean delivery, in vitro fertilization and intrauterine devices – procedures, which damage endometrial lining [4]. Although rare, CP is considered to be a life-threatening condition due to risk of fertilized ovum penetration into the uterine vessels, resulting in a massive hemorrhage.

Improvement in the quality of ultrasonography images and new ways of treatment have drastically changed the way CP has been approached in recent years. Ultrasonography is the main diagnostic tool for this rare condition and conservative treatment, which preserves the potential childbearing, is desired [4]. Total hysterectomy is usually considered as the last resort treatment, although it reduces the risk of hemorrhage.

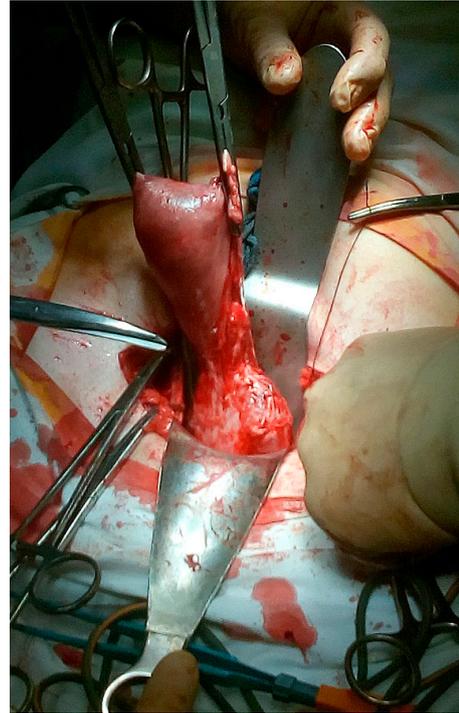
We present you a clinical case of CP treated with total hysterectomy. In light of this report, we also provide a literature review, highlighting diagnostics and management of this rare condition.

## **Case report**

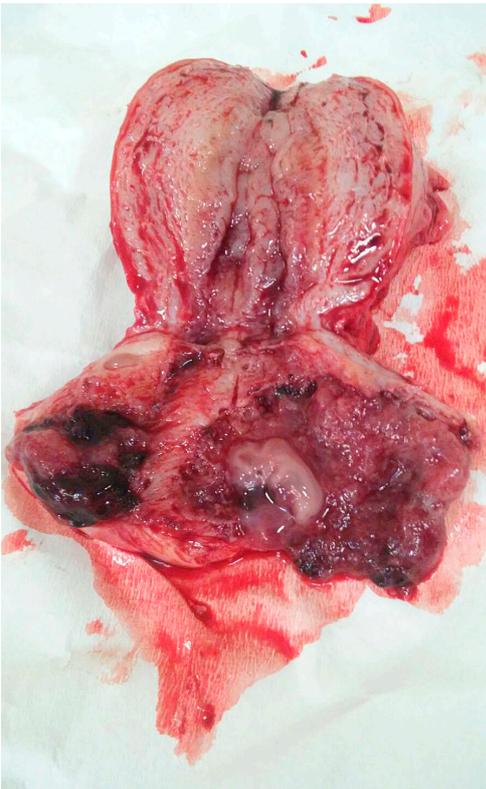
A 38-year-old female, gravida 4 with a history of two cesarean sections and one abortion by vacuum suction curettage, presented to a clinic with mild abdominal pain, three days lasting vaginal bleeding and eight weeks of amenorrhea. Patient was previously diagnosed with chronic anemia. Her visit to a clinic was concerning abortion, but during the examination an ectopic cervical pregnancy was suspected and patient was sent for further inspection to the hospital. During vaginal speculum examination a tight, oval-shaped, cyanotic lesion, highly vascular cervix and ballooned-out cervical canal were observed. Transvaginal ultrasound examination (Figure 1) showed an empty uterine cavity, without tubal and ovarian pathology or free fluid in rectouterine pouch. During the examination, a pregnancy in cervix with a 91 × 44 × 56 mm gestational sac, present fetal cardiac activity and embryo crown-rump length (CRL) of 18 mm were observed. According to the last menstrual period and ultrasonography, gestation was dated as 8 weeks and 2 days. On the first day of admission β-human chorionic gonadotropin (β-hCG) value was 2573 mUI/mL, the following day – 4947 mUI/ml.



**Figure 1.** A gestational sac in the cervix and empty uterine cavity is seen



**Figure 2.** An “hour-glass” shaped uterus observed during the surgery



**Figure 3.** Cut section of the uterus showing a damaged enlarged cervix. Embryo is located in the cervix (arrow)



**Figure 4.** Embryo separated from the cervix

After assessing the risk factors, possible hemorrhage and counseling the patient about the risk-benefit ratio, a total abdominal hysterectomy was performed (Figure 2, 3). During the surgery an embryo-looking specimen was separated from the cervix (Figure 4). Surgery lasted one hour and twenty five minutes and no complications occurred. Postoperative period was uneventful.

## Discussion

CP is defined as a rare form of ectopic pregnancy in which the implantation of pregnancy is in the lining of the endocervical canal. Most common risk factors are dilatation and curettage (70%) and cesarean section (35%) – both of which were performed previously to our presented patient [3,5]. Usual symptoms are painless vaginal bleeding (91%), followed by a period of amenorrhea, and inguinal pain (28%) [6, 7]. During speculum examination a soft, enlarged cervix, open or closed external os and a cystic lesion on the cervical lip should be observed. Bimanual examination should be avoided due to the risk of hemorrhage, although if it is performed, an enlarged cervix compared to the uterus, which is called “an hour-glass” or eight-shaped uterus, is a prominent finding [4, 8, 9].

Ultrasonography, which is the main tool for the observation of CP, has sensitivity of 87.5% when diagnosing this type of ectopic pregnancy. Gestational sac or placenta within the cervix, normal endometrial stripe, hourglass (figure of eight) shaped uterus with ballooned cervical canal are sonographic criteria for CP diagnosis. In addition, an embryo with cardiac activity is a significant discovery [10, 11, 12]. If the case is unusual or complicated and diagnosis is uncertain, further evaluation using magnetic resonance imaging (MRI) facilitates for prompt and accurate diagnosis [13, 14]. Elevated  $\beta$ -hCG level is an assisting diagnostic test and ultrasonography followed by  $\beta$ -hCG test is the most accurate method to diagnose ectopic pregnancy [15].

Prior to ultrasonography and availability of rapid  $\beta$ -hCG test, CP was usually misdiagnosed and treated with dilatation and curettage, resulting in profuse hemorrhage, which was managed performing urgent hysterectomy. Due to widened diagnostic possibilities, a safer and more conservative strategies became more accepted as first-line treatment. Data about a rare condi-

tion as CP is limited and no criteria for selecting a right treatment approach have been determined, but in recent years methotrexate (MTX) has become the most used method [16]. Hung et al meta-analysis reported that MTX single dose treatment, when CP presented with  $\beta$ -hCG  $\geq 10\,000$  mIU/ml, gestational age  $\geq 9$  weeks, embryonic cardiac activity and CRL  $>10$  mm is associated with increased failure rate [17]. According to the same Hung et al meta-analysis, overall success rate using single dose of MTX was 62%, although when cardiac activity was not present 91% of cases were successful and when cardiac activity was present – only 40%. More recent studies show overall MTX success rate as high as 81.3–83% [18]. More advanced pregnancies, when cardiac activity is discovered, should be treated with MTX and potassium chloride in addition to cause fetal death and facilitate resorption of larger gestational sac [19].

In attempt to avoid hysterectomy and preserve fertility, suction curettage and balloon tamponade can be considered. Procedure begins with the injection of vasoconstricting agent, followed by placement of an untied cervical suture, which is tied only if bleeding occurs. Without dilatation suction curettage is passed into the endometrial cavity, Foley catheter is placed and 30ml balloon is inflated within the cervical canal. Balloon is slowly deflated after 24 hours. If bleeding occurs, balloon is re-inflated for later removal. This technique was precisely described by Fylstra in his study where 13 cases were treated with suction curettage and balloon tamponade [20]. All cases were successful, without intraoperative or postoperative bleeding or tying the cerclage suture. Since not all case reports of CP treated with curettage and tamponade show favorable outcomes, sometimes urgent laparotomy might be necessary [21].

Vaginal ligation of cervical arteries, internal iliac artery or uterine artery ligation, internal iliac or uterine artery embolization (UAE) can be used to prevent hemorrhage. UAE is a known method in obstetrics and gynecology and in case of CP it is usually used in combination with MTX or dilatation and curettage [22, 23], although there have been reports about successfully treating CP with UAE only [24, 25].

Hysterectomy can be considered as a first-line approach for women who have completed their families and do not desire fertility, have additional uterine pathology,

are prone to avoid risk of hemorrhage or CP is diagnosed in second or third semester trimester [7, 26]. In other cases hysterectomy is used as a last resort, when more conservative treatment has failed, or in urgent situations, when bleeding is severe and can be life-threatening.

## Conclusions

CP is an unusual, potentially life-threatening form of ectopic pregnancy and every case has to be evaluated

and treated individually. Due to CP's infrequency, no strict criteria for its treatment has been agreed upon and management of this condition lays on patient's wish to preserve or not to preserve fertility. Ultrasound and advancing conservative treatment is changing the approach to CP's management, however, radical surgery such as hysterectomy, which was successfully applied in our case, cannot be dismissed in the treatment of this rare condition.

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