Successful Management of Three Consecutive Cervical Pregnancy in the Same Patient - a Case Report

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Abstract

Ectopic cervical pregnancy is an infrequent complication of the early gestation. Despite tremendous development in the management of ectopic pregnancy, there still lies dilemma in the diagnosis and management of cervical ectopic pregnancy. Traditionally dilation and evacuation has been used in the successful management of these kind of ectopic gestation. In this case report we describe an uncommon case of recurrent ectopic pregnancy and subsequent management.

1. Introduction

Cervical pregnancy accounts for < 1% of all ectopic gestations. It is the rarest form of ectopic pregnancy [1, 2] with incidence varying between 1: 18 000 to 1: 1000 [2]. It is an uncommon form of ectopic pregnancy implanted within the cervical mucosa, the first case was described in 1817 and first named as such in 1860 [3].

It can be a life-threatening complication because of the risk of severe hemorrhage. Earlier, it diagnosed when dilatation and curettage for a provisional diagnosis of incomplete abortion led to massive hemorrhage. Nowadays, with the progression of ultrasonography (USG) along with the ready availability of beta hCG values, early detection of cervical pregnancy is feasible. This has helped improve the number of pre-operatively diagnosed cases from 35% (1978-1982) to 87.5% (1991-1994) [4]. Still, there is no recommendation in the literature for well a defined treatment protocol for management/ fertility preservation in cases of cervical pregnancy.

2. Case Presentation

25 years old, G6P1040, presented to the emergency unit of our institute at 8+2 weeks gestation as a referral case of ectopic pregnancy. Early USG at 5+6 weeks gestation had reported a gestational sac in the lower part of the uterus near its anterior wall. Repeat USG after the first episode of bleeding per vaginum showed a gestation sac in the cervical canal, with a live fetus of 8+3 weeks, probably cervical pregnancy.

In her obstetrical career, she had uneventful surgical evacuation (S&E) for incomplete abortion in her first conception and emergency cesarean for fetal distress in her second pregnancy. One year later she underwent uneventful S & E for incomplete abortion. In her fourth conception, suction evacuation for incomplete abortion at her native place resulted in massive hemorrhage necessitating referral to a higher center, where she received 8 units of blood transfusion, uterine tamponade with foley's catheter bulb for immediate control of bleeding followed by laparotomy and uterine artery ligation for uncontrolled hemorrhage. The post-operative diagnosis assigned was cervical pregnancy. Fifth gestation was again diagnosed to be a cervical pregnancy on USG at 6 weeks. She was managed conservatively with two doses of methotrexate one week apart. Surgical evacuation following medical management was carried out after applying hemostatic sutures to occlude descending cervical artery at 3 & 9’0 clock position,
received 3 units blood and 2 units fresh frozen plasma as replacement for blood loss. In the index pregnancy, she presented to us at 8 weeks gestation with history of spotting and suspicion of cervical pregnancy. She was hemodynamically stable. Per speculum examination revealed minimal bleeding through the external os and a ballooned-out cervix. On transvaginal sonography (TVS), uterine cavity was empty and a gestational sac with a live fetus corresponding to 8 weeks was seen in the cervical canal, below the internal os. (Figure 1) As she had two previous cervical pregnancies, and a history of excessive bleeding and a stormy clinical course of events, a decision was taken for systemic methotrexate (MTX) along with uterine artery embolization (UAE) for prevention of hemorrhage for managing recurrent cervical pregnancy in the same patient.

Her serum $\beta$hCg levels were 1,31,438 iu /ml on day one of MTX and was given a single dose of MTX as 50 mg/m$^2$. UAE revealed hypertrophied bilateral uterine arteries, tortuous arcuate arteries (Right>Left). Bilateral uterine arteries were embolized with ploy vinyl alcohol (PVA) particles. Day 4 $\beta$hCg showed a significant fall to 23,246 iu /ml, which further came down to 17,200 on day 7 and was undetectable at the end of five weeks. Normal menstrual cycles resumed after 7 weeks. She was advised barrier contraception till hysteroscopy.

3. Discussion

The usual presentation of cervical pregnancy is like threatened abortion in almost all cases. Clinically, the findings of a soft enlarged cervix with open external os which usually results in profuse vaginal bleeding when manipulated during examination and may need an emergency hysterectomy [1, 3]. However, these signs are not always diagnostic, and imaging is essential to confirm the diagnosis if cervical pregnancy is suspected. Radiologically, one needs to differentiate between a cervical pregnancy, cervical abortion, and early intrauterine pregnancy. As proposed by Raskin [5] there are 4 criteria by ultrasound for diagnosing cervical pregnancy: cervix and enlargement of the uterus, diffuse intrauterine echoes, and absence of intrauterine pregnancy. This was followed by more stringent criteria by Timor-Tritsch et al [6]: the placenta and entire chorionic sac containing the live pregnancy must be below the internal os and cervical canal must be dilated and barrel-shaped. The role of color doppler for the demonstration of peritrophoblastic blood flow remains controversial. Routine use of magnetic resonance imaging does not improve the diagnostic benefit. The causes of cervical pregnancy remain elusive, but instrumentation and manipulation of the endocervical canal, intrauterine device insertion, di-ethyl stilbesterol (DES) exposure, in-vitro fertilization (IVF) conception [3] have been commonly cited. Although in a study [7], the authors have reported that only 4% of cervical pregnancies were a consequence of IVF while the majority (96%) were because of spontaneous conception, as was in our patient also who had spontaneous cervical pregnancy thrice, though she had predisposing risk factor as well such as repeated cervical dilatation and curettage. Serum beta hCG as high as 1,28,700 IU, with fetal cardiac activity is found in 80% cases. Day 1 Beta hCG of our patient was 1,31,438 IU.

Currently, there are no clear treatment protocols for this entity. Management of this rare but potentially life-threatening complication is usually a combination of systemic therapy with MTX alone or injecting
potassium chloride (KCL) locally. As it occurs in relatively young women, the uterine preserving mode of treatment is the preferred one compared to the traditional teaching of total abdominal hysterectomy [2]. Non-surgical management with MTX has now become an effective alternative [1]. Early diagnosis is crucial for the management of cervical pregnancy to preserve fertility. Surgical evacuation with tamponade has a success rate of 62.5% but is associated with blood loss. Procedures to reduce hemorrhage include ligation of cervical branches of uterine arteries – as was done for previous cervical pregnancy in the index case, or uterine artery ligation or embolization, as was also done in our patient in the present pregnancy. Although Mitrani [8], Pisarski [9] have reported live births in cervical pregnancy, but hysterectomy was done for control of bleeding. Hysterectomy leads to loss of future reproductive potential. Hysterectomy may be considered if the patient does not desire fertility, has intractable hemorrhage or gestational age is > 12 weeks [4].

Fertility preserving options consists of use of systemic MTX, Injection KCl into fetal heart with USG alone, or, in combination with UAE to decrease risk of bleeding. MTX alone or combined with KCI was successfully used by the authors for the management of cervical pregnancies [2, 10]. Intracardiac injection of feticidal agents under ultrasound-guidance may be considered superior maternal chemotherapeutic agents alone [11]. Similarly for control of hemorrhage, irrigation of uterine cavity with 3.5% H2O2 through transcervical foley’s catheter followed by successful hysteroscopic removal of cervical gestation has also been described [12]. Bilateral uterine artery ligation laparoscopically combined with intraamniotic MTX helps reduce the blood loss and therefore preserve fertility [13]. Another fertility preserving option for early gestation cervical pregnancy described is uterine artery embolization before dilation and curettage[14]. Non-surgical management should be undertaken only after informed written consent in the settings where advanced medical facilities for resuscitation and emergency surgery are available.

In our patient, a viable fetus of 8 weeks gestation was noticed in the cervical canal on ultrasonography which looked ballooned out on speculum examination. The uterine cavity was empty which differentiated this condition from a miscarriage in the process of expulsion. Three-dimensional ultrasonography may aid in confirming the diagnosis [2] Spontaneous recurring cervical pregnancy is even rare, with two case reports only with a mean gestational age of 7.5 weeks at diagnosis, bleeding as the chief presenting feature in 80%, as was seen in our patient also, but with a higher Beta hCG value. Previous curettage and previous cesarean delivery were the commonly identified risk factors, both of which were also present in our patient [1].

In another case report of recurrent cervical pregnancy, cervical pregnancy was erroneously diagnosed as a spontaneous abortion, for completion of which curettage led to brisk hemorrhage. The clinical diagnosis of cervical pregnancy was made. Cervical and vaginal packing for local pressure had to be resorted to for control of bleeding. Pressure tamponade with locally inflated foley’s balloon tamponade was shown to be the most effective local treatment for hemorrhage as was done in second consecutive cervical pregnancy [15], while the first cervical pregnancy, in the same patient was successfully treated
with MTX and UAE. Both these treatment modalities were also found to be of help in our patient in our patient.

4. Conclusion

To conclude, though cervical pregnancy is a rare catastrophic complication, early diagnosis, and combination of invasive as well as non-invasive treatment options, as described in the literature, usually results in a successful outcome with the least maternal morbidity and preservation of reproductive potential.

Declarations

1. Funding: Not Applicable

2. Conflicts of interest/Competing interests: None

3. Ethics approval: Not applicable

4. Consent to participate: Obtained. A written informed consent of the patient has been obtained for participating and furnishing data for research process. The identity of the patient has been kept confidential.

5. Consent for publication: Obtained. A written informed consent of the patient has been obtained for participating and furnishing data for research process. The identity of the patient has been kept confidential.

6. Availability of data and material: Available (all important clinical data and images have been included in the case report itself)

7. Code availability (software application or custom code) : Not applicable

8. Authors' contributions:

Seema Chopra: responsible for concept and design of the case report.

Neha Agarwal: responsible for drafting the manuscript.

Neelam Aggarwal: responsible for revision and supervision

Nayana Gaba: responsible for acquisition of the data

References


Figures
Figure 1

On transvaginal sonography (TVS), uterine cavity was empty and a gestational sac with a live fetus corresponding to 8 weeks was seen in the cervical canal, below the internal os.

Supplementary Files

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