Histopathological study of ectopic endocervical pregnancy—a rare case report

Rimi Pandey¹, Padam Kumari Agarwal^{2*}, Nivedita Yadav¹, Vaishali Jain³

European Journal of

Medical Case Reports Volume 3(2):83–86 © EJMCR. https://www.ejmcr.com/ Reprints and permissions: https://www.discoverpublish.com/ https://doi.org/10.24911/ejmcr/ 173-1549867490

ABSTRACT

Background: Ectopic endocervical pregnancy is a rare clinical dilemma, threatening to life.

Case Presentation: A 30 years young lady presented with painless vaginal bleeding of short duration without the history of amenorrhea. On radiological examination, she was diagnosed as a case of cervical polyp/fibroid. Provisional clinical diagnosis was as a case of retained products of conception. She had a history of three previous Caesarian sections; hence, hysterectomy was performed. Histopathologically, the uterus was found to have adenomyosis, while endocervical canal was embedded by well-formed chorionic tissue from internal os to the external os. The ectocervix was spared of invasion by chorionic tissue.

Conclusion: Based on histopathological findings, a diagnosis of ectopic endocervical pregnancy associated with adenomyosis uterus was established. In given clinical scenario, ectopic pregnancy in endocervical region may be included in differential diagnosis of painless vaginal bleeding in young ladies.

Keywords: Aetiopathogenesis, ectopic pregnancy, ectopic endocervical pregnancy.

Received: 06 March 2019	Accepted: 01 May 2019 Correspondence to: Padam Kumari Agarwal		
Type of Article: CASE REPORT		* Former Senior Consultant, Department of Pathology, Vivekananda Poly- clinic and Institute of Medical Sciences, Lucknow 226020, India.	
Funding: None		Email: madamagarwal@gmail.com	
		Full list of author information is available at the end of the article.	
Declaration of conflicting interest	s: The authors declare that there is		
no conflict of interest regarding the publication of this article.			

Background

The ectopic pregnancy of the endocervical region is an extremely rare condition, the commonest being ectopic tubal pregnancy. The incidence of endocervical pregnancy varies from less than 1% of all the ectopic pregnancies [1,2] and ranges from 1 in 1,000 [3] to 1 in 18,000 [4] amongst all the gestations. Its etiology is not yet known. However, in some cases reported in the literature, prior history of damage to the endometrial lining as a result of cesarean sections, intrauterine device, or in vitro fertilization has been documented [5]. The cervical pregnancy has been associated with higher morbidity, because of non-availability of the imaging techniques in the past. The first case of ectopic endocervical pregnancy was diagnosed on ultrasound by Raskinin [6]. Till date, these cases were managed based on radiological diagnosis, but in the present case, hysterectomy was performed which provided an opportunity to study the histopathological changes in the uterus and also the aetiopathogenesis of ectopic endocervical pregnancy.

Case Description

A 30-year-old lady, P4 A1 L2 presented with complaints of continuous painless bleeding per vaginum for last 20 days and urinary retention associated with fever for last 2 days. The patient had undergone dilatation and curettage 3 days back at a peripheral hospital. Records of the procedure were not available, except the sonographic report, which showed hyper-echogenic mass of complex echo-texture in the lower part of uterus.

On clinical examination, she was thin built, febrile, had pallor and normal blood pressure. Systemic and per abdomen examination was within normal limits. Per speculum examination revealed bleeding through external os. On vaginal examination, the uterus was firm, non-tender with restricted mobility. Cervix was enlarged and barrel shaped.

Her obstetric history revealed that she had previous three cesarean sections at term due to contracted pelvis. Her first pregnancy was 14 years back and she delivered a full term male baby, she was also diagnosed as pregnancy induced hypertension in this pregnancy. Second pregnancy was about 12 years back and delivered a full term female child, while the third pregnancy was about 10.5 years back and again delivered a male child. Now, she had irregular menstrual cycles since almost 7 years, without any history of amenorrhea.

Ultrasound (US) abdomen showed uterus normal in size and shape and a hypo-dense lesion with internal hard echoes of $5.2 \times 4.7 \times 6$ cm in size occupying the endocervical canal (Figure 1a). Computerized tomography (CT) scan abdomen confirmed the US findings, i.e., there was a low attenuating mass in endocervical canal (Figure 1b), which was widened. Mass had cauliflower like appearance

(Figure 1b). Following the imaging studies, a diagnosis of cervical polyp/fibroid was made.

Laboratory investigations revealed that hematological parameters, except for anemia (Hb 7.6 g%), and serum biochemistry were within normal range. Also, the urine pregnancy test at the time of admission was negative, but quantitative serum beta-human chorionic gonadotrophin (beta hCG) was high (56.34 ml U/ml; normal range <35 ml U/ml).

Total abdominal hysterectomy with adhesionolysis was done. Per-operative findings revealed that the uterus was slightly bulky. Cervix was found ballooned up. Cut section showed blood clots and probably products of conception, which were firmly adhered to the endocervix. The hysterectomy specimen was submitted for histopathological examination. Postoperative period was uneventful, and the patient was discharged from the hospital in a stable condition.

On gross examination, specimen consisted of uterus and cervix which measured $11 \times 6 \times 5$ cm in size. Uterus appeared normal in size and shape, but endocervix was enlarged (Figure 1c). Ectocervix was hypertrophied. Horizontally cut surface of uterus revealed slit like and smooth endometrial cavity. It was 3 cm in length. Endocervical canal measured 4.5 cm in length. The cut surface was covered by organized blood clots, which were invading the wall of endocervix resulting in thinning of the wall of endocervix (Figure 1d).

Microscopic examination

Sections from uterus revealed that the myometrium was lined by normal proliferative endometrium. Islands of

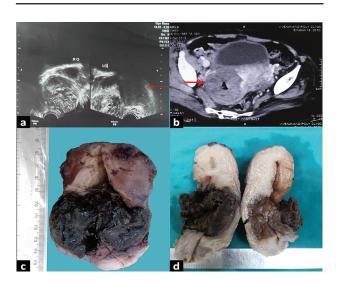


Figure 1. (a) Transvaginal ultrasound showing a heterogeneous mass lesion (arrow) compressing the endocervix, (b) CT scan of lower abdomen showing a low attenuating mass lesion (arrow) lying in the endocervical canal, (c) Gross appearance uterus is of normal size. Endocervix is enlarged and covered by blood clots above the level of external os, (d) horizontally cut specimen showing slit like endometrial cavity; endocervix eroded by blood clots from internal to external os and ectocervix is normal.

endometrial tissue were embedded in the myometrium, i.e., adenomyosis was present in the uterus. Sections from endocervix revealed that the surface was covered by blood clots, enclosing well-formed chorionic villi (Figure 2a and b). The chorionic tissue was embedded in the muscles of endocervix (Figure 2c and d). The endocervical glands were proliferated and were lined by tall columnar mucous secreting epithelium. Based on histopathological findings, diagnosis of "Adenomyosis Uterus associated with Ectopic Pregnancy in the Endocervical Canal" was established.

Discussion

Endocervical canal is a very rare site for ectopic pregnancy amongst all the ectopic gestations; the reported incidence varies from 1 in 1,000 to 16,000 to all the ectopic pregnancies [7]. In the author's Padam Kumari Agarwal (PKA) experience, this is the first case seen after 40 years of histopathological reporting on approximately 25,000 hysterectomy specimens at a medical college and then in a tertiary care hospital.

Patients with cervical pregnancy classically present with painless vaginal bleeding during the first trimester of pregnancy [1]. The present case presented with painless continuous bleeding per vaginum for 20 days. On ultrasound, a hypo dense lesion with hard internal echoes within the endocervical canal was reported. The uterine cavity was empty and endometrial surface was smooth. The mass in the cervical canal was radiologically diagnosed as endocervical polyp. CT scan showed uterus of normal shape and size and a low attenuating mass lesion in cervical canal which was widened. Keeping these investigations in consideration, four differential diagnoses were made, i.e., cervical pregnancy, retained products of conception, cervical polyp, and cervical fibroid. The final diagnosis was settled on histopathological examination of the hysterectomy specimen which revealed adenomyosis in the uterus, along with involvement of the endocervix which was covered by blood clots enclosing well-formed chorionic tissue invading the wall of endocervix. The absence of evidence of pregnancy in the endometrium

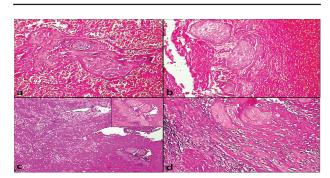


Figure 2. Microphotographs of uterus and endocervix (a&b) sections showing blood clots enclosing chorionic tissue (H: E X 40), (c) section showing wall of endocervix embedded by chorionic tissue (H:E X 40, inset X 100), (d) chorionic tissue embedded in the wall of endocervix (H:E X 40).

with normal myometrium and presence of chorionic issue within the endocervical canal favored the diagnosis of ectopic endocervical pregnancy.

In endocervical pregnancy, the implantation occurs in the endocervical canal, at or below the internal os. The erosion of the wall of endocervix by trophoblastic tissue resulting in thinning and distension of the canal had been reported earlier on radiological examination [8]. No histopathological confirmation of ectopic endocervical pregnancy has been reported in the literature to the best of the authors' knowledge.

The etiopathogenesis of ectopic pregnancy, is not well understood, some local pathology in the uterus, such as leiomyomas, adenomyosis, or previous surgical interventions such as previous cesarean sections, dilatation & curettage, and intrauterine devices [9] may be the responsible causes for ectopic pregnancy. In the present case, the patient had previous three cesarean sections. Also, she had adenomyosis which may be responsible for ectopic endocervical pregnancy as reported earlier in the literature [4]. In all, the etiology of ectopic pregnancy in the endocervical canal in the present case appears to be a consequence of previous three cesarean sections and associated adenomyosis of the uterus.

In the present case, total abdominal hysterectomy was performed as a choice of treatment which allowed studying the histopathological changes in such cases with associated pathology in the uterus. Histopathology confirmed the diagnosis of ectopic endocervical pregnancy by demonstrating the chorionic tissue embedded in the endocervical canal above the external os with adenomyosis uterus. To the best of our knowledge till date, cases of ectopic endocervical/cervical pregnancy were diagnosed by a radiologist and the patients were managed accordingly. An additional, information on thorough histopathological features in a case of ectopic endocervical pregnancy has been provided in this case. The patient is hail and hearty after surgery, after 1 year of follow up.

Conclusion

This case provides a message, that though the cervical/ endocervical pregnancy is a very rare condition, this can be life threatening if not diagnosed and managed early. Such patients should always be investigated thoroughly to confirm the diagnosis for proper and timely management. Primary obstetric care providers should always keep in mind, the possibility of cervical pregnancy in females presenting with first trimester painless vaginal bleeding, especially with poor obstetric history.

Acknowledgment

Authors are grateful to Dr. Shyam Kumar Maurya, HOD(Pathology), Vivekananda Polyclinic and Institute of

Medical Sciences, Lucknow, India for giving permission to publish this interesting and rare case. Authors are also grateful to Mr. S. K. Shukla and Ms Maya Singh for technical assistance to prepare the histopathology slides.

List of Abbreviations

CT Computerized tomography

Consent for publication

Informed consent was obtained from the participants.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

Author details

Rimi Pandey¹, Padam Kumari Agarwal²*, Nivedita Yadav¹, Vaishali Jain³

- 1. Senior Resident, Department of Pathology, Vivekananda Polyclinic and Institute of Medical Sciences, Lucknow 226020, India
- 2. Former Senior Consultant, Department of Pathology, Vivekananda Polyclinic and Institute of Medical Sciences, Lucknow 226020, India
- 3. Head, Department of Gynae and Obstetric, Vivekananda Polyclinic and Institute of Medical Sciences, Lucknow 226020, India

REFERENCES

- Marcovici I, Rosenzweig BA, Brill AI, Khan M, Scommegna A. Cervical pregnancy: case reports and a current literature review. Obstet Gynecol Survey. 1994;49(1):49–55. https://doi.org/10.1155/2011/858241
- Frates MC, Benson CB, Doubilet PM, Salvo DN, Brown DL, Laing FC, et al. Cervical ectopic pregnancy: results of conservative treatment. Radiology. 1994;191:773–5. https:// doi.org/10.1148/radiology.191.3.8184062
- Shinagawa S, Nugayama M. Cervical pregnancy as a possible sequela of induced abortion. Am J Obstet Gynecol. 1969;105(2):282–4.
- Dees HC. Cervical pregnancy associated with uterine leiomyomas. South Med J. 1966;59(8):900.
- Ushakov FB, Elchalal U, Aceman PJ, Schenker JK. Cervical pregnancy: past and future. Obstet Gynecol Surg. 1996;52(1):45–59. https://doi. org/10.1097/00006254-199701000-00023
- Raskin MM. Diagnosis of cervical pregnancy by ultrasound: a case report. Am J Obstet Gynecol. 1978;130(2):234– 235. https://doi.org/10.7439/ijbar
- Yitzhak M, Orvieto R, Nitke S, Neuman-Levin M, Ben-Rafael Z, Schoenfeld A. Cervical pregnancy-a conservative stepwise approach. Human Reprod. 1999;14(3):847–9. https://doi.org/10.1093/humrep/14.3.847
- Gun M, Mavrogiorgis M. Cervical ectopic pregnancy: a case report and literature review. Ultrasound Obstet Gynecol. 2002;19(3):297–301. https://doi.org/10.104 6/j.1469-0705.2002.00559
- Mohebbi MR, Rosenkrans KA, Luebbert EE, Hunt TT, Jung MJ. Ectopic pregnancy in the cervix: a case report. Case Rep Med. 2011;6(1):1–3. https://doi. org/10.1155/2011/858241

Summary of the case		
Patient (gender, age)	1	30 years, Female
Final diagnosis	2	Ectopic endocervical pregnancy
Symptoms	3	Continuous painless vaginal bleeding per vaginum
Medications /procedure	4	
Clinical procedure	5	Total abdominal hysterectomy with adhesionolysis
Specialty	6	Gynecology