Ectopic pregnancy after hysterectomy may not be so uncommon: A case report and review of the literature

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ABSTRACT

Background: Ectopic pregnancy after hysterectomy is a very uncommon event, but its frequency is increasing. Since first reported by Wendler in 1895, 71 cases of post-hysterectomy have been reported.

Case: A woman, 2 years after an abdominal supracervical hysterectomy, presented with a ruptured fallopian tube ectopic pregnancy.

Conclusion: Any woman, even after hysterectomy but with ovaries in situ, who presents with an acute abdomen or abdominal–pelvic pain should be screened for pregnancy.

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1. Introduction

Ectopic pregnancy after hysterectomy is a very uncommon event, but its frequency is increasing. Since first reported by Wendler in 1895 [1], fifty-six cases of post-hysterectomy ectopic pregnancies were reported by this author in 2009 [2]. Since that publication, 11 subsequent cases have been reported, and other additional cases have been found through a bibliography review. The total published number of cases is now 71. This author has treated the 72nd (Table 1).

2. Case

A 32 year old woman, Gravida 5, Para 4, with a history of a prior abdominal supracervical hysterectomy presented with the acute onset of severe abdominal pain, nausea and vomiting, and vaginal bleeding. A pregnancy test was positive. A serum human chorionic gonadotropin level was 2279 mIU/ml. Her last delivery, 2 years prior, was her third cesarean, complicated by post-operative bleeding requiring abdominal re-exploration and an emergent supracervical hysterectomy. Very dense pelvic adhesions were described during this operation. Upon presentation she was found on abdominal ultrasound to have a large volume hemoperitoneum. Her abdomen was explored via laparotomy finding a ruptured, bleeding right fallopian tube ectopic pregnancy with a 2000 ml hemoperitoneum. The right fallopian tube and ovary were densely adherent to the residual cervical stump. The right adnexum and the cervix were removed. Post-operative transfusion was required, but she recovered uneventfully.

3. Comment

Thirty of the now 72 cases of ectopic pregnancies after hysterectomy occurred in the immediate period after hysterectomy, “early presentation,” suggesting that a pregnancy, or a potential for pregnancy, existed at the time the hysterectomy was performed. An immediate prehysterectomy pregnancy test would not be expected to be positive under such circumstances, and an early pregnancy diagnosis would be unlikely. This has occurred after all types of hysterectomy [3–32]. This is presumed to occur because an unrecognized, preclinical (luteal phase) pregnancy existed at the time of hysterectomy: a preimplanted fertilized ovum was in transit and confined to the fallopian tube, or sperm was present within the fallopian tube when the hysterectomy was performed in a periovulatory period, allowing postoperative fertilization and tubal implantation.

Because the symptoms of ectopic pregnancy can be mimicked by common immediate complications after hysterectomy, such as protracted abdominal pain, pelvic hematoma formation, vaginal cuff infection, and vaginal bleeding, ectopic pregnancy is rarely expected in most cases until additional imaging or repeat operation confirms the diagnosis [2,22,26,27,29,30]. Therefore, the prevention of “early presentation” ectopic pregnancy after hysterectomy is the prevention of pregnancy before hysterectomy. As previously recommended, hysterectomy, like tubal sterilization, should be avoided in the luteal phase of the menstrual cycle in those women not previously sterilized or not using reliable contraception, unless no vaginal intercourse has occurred during the preoperative period. Women should be preoperatively counseled as such. Any woman who has undergone hysterectomy and

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### Table 1
Pregnancy after hysterectomy.

| Year published | Type of hysterectomy | Time to diagnosis |
|----------------|----------------------|-------------------|
| **Early presentations** | | |
| Knaus [3] 1937 | vag | 57 days |
| Girone [4] 1952 | abd | 53 days |
| Adams and Schreier [5] 1957 | abd | 86 days |
| Claus [6] 1959 | abd | 29 days |
| Smythe [7] 1961 | abd | 40 days |
| Graffagnino [8] 1963 | vag | 59 days |
| Ledger and Daly [9] 1963 | abd | 96 days |
| Moayer [10] 1965 | vag | 35 days |
| McDaniel and Gullo [11] 1968 | vag | 59 days |
| Wells [12] 1970 | vag | 39 days |
| Grunberger [13] 1971 | vag | Unknown |
| Bradner and Vigilante [14] 1973 | vag | 54 days |
| Niebyl [15] 1973 | vag | 79 days |
| Alexander and Everidge [16] 1979 | vag | 41 days |
| Cocks [17] 1980 | vag | 26 days |
| Jackson [18] 1980 | abd | 36 days |
| Buchan [19] 1980 | abd | 6 weeks |
| Zdravkovic [20] 1980 | abd | 5 weeks |
| Marut and Zucker [21] 1981 | vag | 55 days |
| Williams [22] 1981 | abd | 7 weeks |
| Zoli and Rocho [23] 1982 | abd | 15 weeks |
| Nehra and Loginsky [24] 1982 | vag | 30 days |
| Meizner et al. [25] 1982 | abd | 12 weeks |
| Arora [4] 1983 | vag | 47 days |
| Reese et al. [27] 1989 | vag | 24 days |
| Gaeta et al. [28] 1993 | abd | 2 months |
| Allen and East [29] 1998 | LAVH | 6 weeks |
| Weisenfeld and Guido [30] 2003 | abd | 12 weeks |
| Binder [31] 2003 | vag | 13 weeks |
| Fader et al. [32] 2007 | abd | 12 weeks |
| **Late presentations** | | |
| Wendler [1] 1895 | vag | 6 years |
| Grigg [33] 1920 | abd SC | 1 year |
| McMillan and Dunn [34] 1921 | abd SC | 1 |
| McMillan and Dunn [34] 1921 | abd SC* | 2 |
| Weil [35] 1938 | vag | 5 years |
| Connors et al. [36] 1943 | abd SC* | 4 years |
| Brown and Shields [37] 1944 | abd SC | 1 year |
| Frech [38] 1948 | vag | 9 years |
| Lyle and Christianson [39] 1955 | vag | 11 years |
| Gordy and Otis [40] 1961 | abd | 14 months |
| Zaczek [41] 1963 | abd | 7 months |
| Hanes [42] 1963 | vag | 9 months |
| Kornblatt [43] 1968 | vag | 12 months |
| Altinger [44] 1973 | vag | 2 years |
| Sims and Letts [45] 1973 | vag | 2 years |
| Schnell and Sinn [46] 1982 | vag | Unknown |
| Heidenreich et al. [47] 1983 | vag | 1 year |
| Salmi et al. [48] 1984 | vag | 3 years |
| Beuthe and Wemken [49] 1985 | vag | Several years |
| Culpepper [50] 1985 | vag | 6 years |
| Casco et al. [51] 1992 | vag | 5 years |
| Isaacs et al. [52] 1996 | vag | 8 years |
| Adeyemo et al. [53] 1999 | LAVH | 2 1/2 years |
| Brown et al. [54] 2002 | C-hyst | 12 years |
| Pasic et al. [55] 2004 | LSH | 4 months |
| Nnochiri and Warwick [56] 2007 | vag | 1 year |
| Tagore et al. [57] 2007 | abd | 9 years |
| Babikian et al. [58] 2008 | abd SC | 3 years |
| Rosa et al. [59] 2009 | vag | 5 years |
| Fylstra [60] 2009 | C-hyst | 6 years |
| Barhate et al. [61] 2009 | vag | 2 years |
| Bansal et al. [62] 2010 | abd SC* | 4 years |
| Ramos et al. [63] 2010 | vag | 5 months |
| Hitti et al. [64] 2010 | abd SC* | 7 years |
| Anupama et al. [65] 2012 | abd SC | 11 |
| Friedman et al. [66] 2013 | vag | 5 years |
| Villegas et al. [67] 2013 | abd SC | 2 |
| Anis et al. [68] 2013 | abd SC | 6 years |
| Cook and Davies [69]. 2014 | abd | 2 years |
| Yesilyurt et al. [70] 2014 | C-hyst | 3 years |
had not previously undergone tubal sterilization or had a partner vasectomy, or was not using reliable contemporaneous contraception, should be considered at risk for this diagnosis, should otherwise unexplained postoperative pain or bleeding occurs.

Interestingly, no additional early post-hysterectomy pregnancies have been reported since this author’s 2009 report.

Ectopic pregnancy has been reported to occur as late as 12 years after hysterectomy, “late presentation,” and 42 such cases have now been reported [1,33–67], including this latest case treated by this author. This can only develop because the sperm have gained access to the peritoneal cavity through a fistulous tract between the vagina and the peritoneal cavity. Although this has occurred after all types of hysterectomy, 50% follow vaginal hysterectomy [1,35,38,39,42–53,56,59,61,63,66], and this would suggest a causal relationship. Although the operative narrative for the hysterectomy was seldom available to the physicians treating the ectopic pregnancy after hysterectomy, observations thought to increase the chance for vaginal-peritoneal fistula formation include an open vaginal cuff closure technique, vaginal cuff infection or hematoma formation after hysterectomy, vaginal cuff granulation tissue, and a prolapsed fallopian tube [51–56,59].

The usual method of vaginal cuff closure differs between vaginal hysterectomy and abdominal hysterectomy. The adnexal structures are brought into closer proximity of the vaginal cuff with vaginal hysterectomy cuff closure, and can even be incorporated into the peritoneal closure, increasing the chance for a prolapsed fallopian tube into the vaginal cuff or the development of a vaginal-to-peritoneal or tubo-vaginal fistula [47,49]. “Late presentation” ectopic pregnancies after total abdominal hysterectomy have been reported, indicating that vaginal-to-peritoneal fistula can even develop after this procedure.

However, the small number of such cases would suggest that it is less likely to occur, presumably because the residual fallopian tubes and ovaries are more distant from the vaginal cuff during abdominal hysterectomy cuff closure, and the commonly used technique of closure of the pelvic floor parietal peritoneum over the vaginal cuff isolates the vagina from the peritoneal cavity [47]. These numbers of ectopic pregnancies and the hysterectomy method differences are suggestive that the risk would be greater after vaginal hysterectomy, but this is not based on any proven medical evidence.

Subtotal hysterectomy has increased in the United States in the past decade, estimated to now make up 7.5% of all hysterectomies performed [70–72]. Fourteen cases of “late presentation” ectopic pregnancy have followed supracervical or cesarean hysterectomy, including the current case [33,34,36,37,56,59,61,63,65–68]. Leaving a remnant of the cervix or the epithelialization of a much larger vaginal cuff closure area because of cervical dilation at the time of cesarean hysterectomy may increase fistulous tract formation [54,73]. With the now more commonly performed laparoscopic supracervical hysterectomy, this author and other investigators are concerned about a potential increase in the incidence of ectopic pregnancy after hysterectomy. The commonly used technique of cauterizing the residual proximal cervical canal to prevent cyclic vaginal bleeding after hysterectomy at the time of laparoscopic supracervical hysterectomy may not be adequate to prevent patency of the cervical canal. Pathologic identification of such a communication through a residual cervix has been documented [58]. Cautery of the cervical canal and cervical stump at the time of laparoscopic supracervical hysterectomy has also failed to prevent a patent cervical canal and an ectopic pregnancy after hysterectomy [55].

It may not be possible to prevent all “late presentation” ectopic pregnancies after hysterectomy, but its prevention is the prevention of vaginal-to-peritoneal cavity communication. Vaginal cuff closure, regardless of operative technique, should be sure not to incorporate the fallopian tube into the vaginal cuff, and postoperative vaginal cuff granulation tissue, a very common finding, must be differentiated from a portion of prolapsed fallopian tube, with biopsy, if necessary [74]. When the cervix is left in situ, techniques should be used to obliterate or isolate the residual cervical canal, thus preventing a patent cervical canal allowing the sperm access to the peritoneal cavity.

Ectopic pregnancy after hysterectomy is very rare. An estimated 600,000 hysterectomies are performed each year in the United States, and one-third of all US women will have had a hysterectomy by age 60 years [70–72]. Only 72 cases of ectopic pregnancy after hysterectomy have now been reported in the world’s literature since 1895. This incidence is very small, but may be on the increase because of supracervical hysterectomy. This author recommends that any woman, even after hysterectomy but with ovaries in situ, who presents with an acute abdomen or abdominal–pelvic pain should be screened for pregnancy. A urine pregnancy test is readily available and inexpensive, and although ectopic pregnancy after hysterectomy has been very uncommon until now, only a high index of suspicion will make the diagnosis.

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Notes to Table 1:

vag: vaginal hysterectomy.
abd: abdominal hysterectomy.
abd SC: abdominal supracervical hysterectomy.
LAVH: laparoscopic assisted vaginal hysterectomy.
LSH: laparoscopic supracervical hysterectomy.
C-hyst: cesarean hysterectomy.
Diagnosed at 23 weeks and delivered electively a healthy infant at 36 weeks.
Second case of post-hysterectomy ectopic in the same woman.
Vaginal delivery at 6 months living infant. Supracervical hysterectomy later followed by trachelectomy.
Emergent subtotal hysterectomy for PPH after the 3rd cesarean.
Vaginal removal, vaginal cuff opened and tubal pregnancy removed.
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