

Umbilical Endometriosis after Unprotected Removal of Uterine Pieces Through the Umbilicus

Philippe R. Koninckx, M.D., Gilbert Donders, M.D., and Hilde Vandecruys, M.D.

Abstract

We reviewed the frequency of umbilical endometriosis after laparoscopic-assisted subtotal hysterectomy with unprotected removal of uterine segments through the umbilical incision. Retrospective analysis of surgical records was followed by recall and clinical examination of 10 patients. Two women developed umbilical endometriosis. One was the only patient operated during menstruation. The other was one of two women operated in the late luteal phase and with subsequent adequate exposure to endogenous or exogenous estrogens. From 1976–1997, 22 reports of umbilical endometriosis were published: 18 cases were spontaneous, 2 occurred after ring sterilization, and 2 after diagnostic laparoscopy. These data lend support to the concept that implantation and the potential development of menstrual endometrium are increased after surgery. It is speculated that procedures that expose menstrual endometrial cells to nonepithelialized areas could be associated with an increased risk of endometriosis.

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The physiopathology of endometriosis is incompletely understood, with transplantation¹ and metaplasia theories^{2,3} supported experimentally.^{4,5} For pelvic endometriosis most efforts were initially aimed at explaining the implantation and growth of retrograde regurgitated endometrial cells. Since it is apparent that retrograde menstruation is a rather universal phenomenon occurring in all women,^{6–8} the question is why all women do not develop endometriosis; that is, why these cells do generally not attach, implant, and develop into endometriosis. Some mechanisms involved seem to be the expression of adhesion molecules⁹ and integrity of the mesothelial layer,¹⁰ expression of paracrine factors such as angiogenic factors¹¹ as vascular endothelial growth factor

(VEGF),^{12,13} modulation by the immune system (macrophages, natural killer cells),¹⁴ and overall hormonal milieu as stressed by luteinized unruptured follicle syndrome hypothesis.⁶

In experimental transplantation of endometrial cells into the peritoneal cavity of primates, luteal endometrium is less effective at inducing endometriosis than menstrual endometrium, lending support to concepts such as expression of adhesion molecules and VEGF in the menstrual endometrium. The feasibility of transplanting endometrium to the abdominal wall was proved experimentally in primates. In humans, since 1976, 168 case reports or small series described umbilical endometriosis or endometriosis in the epiotomy or abdominal or cannula puncture scar after

From the Department of Obstetrics and Gynecology, Division of Endoscopic Surgery, University Hospital Gasthuisberg, Leuven, Belgium (Drs. Koninckx and Vandecruys); Nuffield Department of Obstetrics and Gynecology, University of Oxford, Oxford, United Kingdom (Dr. Koninckx); and Department of Obstetrics and Gynecology, H.-Hartziekenhuis, Tienen, Belgium (Dr. Donders).

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Address reprint requests to Philippe R. Koninckx, M.D., University Hospital Gasthuisberg, Department of Obstetrics and Gynecology, Herestraat 49, Leuven, B-3000 Belgium; fax 321 634 4238.

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surgery. The prevalence is low and the risk was estimated to be 0.1%^{15,16} after cesarean section and 1%¹⁷ after hysterotomy.

Umbilical endometriosis is a rare but well-known pathology. Two cases that occurred after laparoscopic-assisted subtotal hysterectomy (LASH) prompted us to evaluate whether they were correlated with phase of the menstrual cycle at the time of surgery.

Materials and Methods

We reviewed records of 10 women (mean age 41 ± 8 yrs) who underwent LASH in 1993 and 1994. The uterus was cut longitudinally in large segments that were removed through the umbilical incision without protection. These fragments obviously exposed the endometrium to the incision.

In 9 of the 10 women the presence of umbilical endometriosis was evaluated by physical examination and clinical symptoms. Women were considered free of the disorder when periodical umbilical pain and/or swelling were absent despite prolonged exposure to adequate endogenous or exogenous estrogens. The records of these women were scrutinized to ascertain the phase of the menstrual cycle during which LASH was performed, (especially premenstrual and menstrual phases) and the presence of natural or artificial postoperative menstrual cycles. Hormone exposure was assumed adequate when women were premenopausal at the time of surgery, no ovariectomy was performed, and no subjective menopausal discomfort was experienced after surgery or when sequential hormone replacement therapy was given.

A MEDLINE search spanning 1976 to 1998 was performed using Knowledgefinder and Winspirs as search engines.

Statistical Analysis

Statistical significance was evaluated by the χ^2 test of the SAS package.¹⁸

Results

Our Series

Two of the 10 patients developed umbilical endometriosis. Two years after LASH a 40-year-old woman experienced vague periumbilical pain. Clinical examination was equivocal, as was the ultrasound scan, on which some vague irregularities were interpreted as

scar tissue. The patient's complaints increased, including intermittent swelling of the umbilicus. The diagnosis of umbilical endometriosis was obvious by clinical examination, ultrasound, magnetic resonance imaging (MRI), and thin-needle aspiration of brownish fluid. Excision confirmed an endometriotic nodule 1 cm in diameter between fascia and peritoneum. Microscopically the lesion consisted of endometrial glands and stroma surrounded by fibrotic tissue and numerous hemorrhagic cysts, 1 to 2 mm in diameter. The other woman reported a similar history, with pain starting 2 years after LASH. She underwent excision of the umbilical swelling at another hospital and pathology showed endometriosis.

Of the nine women available for follow-up, umbilical endometriosis occurred in the only woman who was operated during menstruation, and in one of two who were operated in the late luteal phase and who probably had subsequent cyclic ovarian activity. The other seven patients were free of the disorder.

Literature Review

Since 1976, only 1 review and 25 case reports of umbilical endometriosis were found, with a total of 136 patients. Of these, 131 women had no history of surgery and the condition was considered spontaneous. Of 34 patients, 3 had a history of ring sterilization,^{19,20} and 2 had undergone diagnostic laparoscopy for pelvic endometriosis.^{21,22} Endometriosis in surgical scar tissue, especially after cesarean section (15 patients since 1996^{15,22-34}) is reported more frequently. For both umbilical and scar endometriosis in case reports or small series, discussions focused on diagnostic difficulty, but no attempt was made to correlate onset of endometriosis with menstrual phase during surgery.

Discussion

It is tempting to suggest from this small series that surgery that exposes menstrual endometrium to tissues increases the risk of implanting endometrial cells and thus of endometriosis. Indeed, umbilical endometriosis developed in the only patient operated during menstruation, in one of two operated in the late luteal phase, and in none of the seven remaining women. No statistical significance was reached with χ^2 with Yates continuity correction for small numbers or by Fisher's exact test ($p = 0.067$). This does not, however,

contradict the hypothesis that menstrual endometrial cells have an increased potential for implantation, as suggested from observations in baboons that after experimental transplantation, menstrual endometrium is more likely to develop into endometriosis than follicular or luteal phase endometrium.³⁵ Another factor to be considered is difference in implantation potential between functionalis and basalis layers of endometrium. In fact, endometrial cells from the basalis were also involved in these two patients and in menstrual curettings injected in baboons.

This observation is surprising since in our experience removing over 500 large deep endometriotic nodules³⁶ and some 800 cases of cystic ovarian endometriosis of the wall through umbilical incisions, not a single case of umbilical endometriosis has occurred. This is even more surprising since all specimens were removed without protection; most through the cannula, but at least 50 to 100 through the umbilical incision after dilatation, with direct contact between scar tissue and endometriotic tissue. We could speculate that the outer layer of these excised specimens was no longer viable since excision was performed with CO₂. Or perhaps the implantation potential of endometriotic cells after leuteinizing hormone-releasing hormone agonist pretreatment is lower, since most of the women in our series with very large nodules received the agents.

A more plausible hypothesis is that specifically menstrual endometrium has a higher implantation potential, and that deep and cystic endometriosis differ from menstrual endometrium because of the dif-

ferent endocrine environment of the peritoneal cavity. This could moreover explain the low frequency of endometriosis in scar tissue, taking into account the ubiquitous presence of living endometrial cells in peritoneal fluid. A review of the literature confirmed that the frequency of umbilical endometriosis after endoscopic surgery for endometriosis must be low, since no specific case reports were found. Of the 34 patients reported since 1976, only 2 had undergone diagnostic laparoscopy and 2 ring sterilization, and in none of them was endometriosis mentioned specifically.

Scar endometriosis seems to be slightly more common after cesarean section or hysterotomy, consistent with the high frequency in baboons after hysterotomy. This also is surprising since it suggests that decidual endometrium has a higher implantation capacity, or that the hormonal milieu of pregnancy would favor implantation.

The difficulty diagnosing umbilical endometriosis is confirmed by this report. The became clinically apparent because of cyclic pain and menstrual swelling,³⁷ but was missed when the nodule was smaller. Ultrasound imaging is reported to be equivocal, as in our patients, and can easily be mistaken for scar tissue, especially in obese women. Magnetic resonance imaging was reported to be helpful in the diagnosis of umbilical endometriosis,^{38,21} but it remains unclear whether it can diagnose small, vaguely suspected nodules. In rectovaginal endometriosis MRI can detect larger deep nodules, but not smaller ones that are apparent only during menstruation; nor can it predict lateral extension of these nodules.³⁹ Umbilical

TABLE 1. Umbilical Endometriosis after LASH with Unprotected Removal of Uterine Segments Through the Umbilical Incision

Year of Surgery	Age (yrs)	Menstrual Cycle Phase	Ovariectomy	Hormone Replacement Therapy	Umbilical Endometriosis
1993	30	EF	-		-
1993	47	LL	+	-	-
1993	43	Sec. am.	+	-	-
1993	43	LL	-		-
1993	55	Sec. am.	+	+	-
1993	40	LF	-		-
1993	37	LL	-		+
1994	45	M	-		+
1994	29	Oligomen	-		-
1994	41	Oligomen	-		-

EF = early follicular; LF = late follicular; LL = late luteal;

M = menstrual; Sec. am. = secondary amenorrhoea; oligomen = oligomenorrhoea.

endometriosis should be differentiated from a hernia and cannula port metastasis.⁴⁰ Table 2 reviews case reports of the disorder from 1976 to 1995.^{19–22,37,41–57}

Conclusion

To the best of our knowledge this is the first attempt to correlate the etiology of scar endometriosis with the phase of the menstrual cycle in which an intervention is performed. Although the series was small, the data are consistent with the observation that menstrual endometrium has a high potential to develop into endometriosis. Although it would be premature to recommend that endoscopic and other abdominal surgery be avoided during menstruation in women with open tubes and/or endometriosis, the results suggest that these patients should be followed specifically to prove or refute that menstrual surgery is a risk factor for developing scar endometriosis.

TABLE 2. Case Reports of Umbilical Endometriosis, 1976–1995

Year	Number of Patients	Spontaneous	Postoperative Procedure
1976 ⁴¹	1	+	
1977 ⁴²	1	+	
1978 ⁴³	1	+	
1980 ⁴⁴	1	+	
1980 ⁴⁵	1	+	
1980 ⁴⁶	1	+	
1981 ⁴⁷	1	+	
1983 ⁴⁸	6	6 +	
1985 ⁴⁹	1	+	
1987 ¹⁹	1		Laparoscopic ring sterilization
1990 ⁵⁰	1	+	
1990 ⁵¹	2	2 +	
1992 ⁵²	1	+	
1992 ⁵³	1	+	
1993 ⁵⁴	1	+	
1993 ²⁰	2		Laparoscopic ring sterilization
1994 ³⁷	1	+	
1994 ²¹	2	1 +	Diagnostic laparoscopy
1995 ⁵⁵	1	+	
1995 ²²	2	1 +	Diagnostic laparoscopy
1996 ⁵⁶	1	+	
1997 ⁵⁷	1	+	

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