

Results of treatment of perineal endometriosis at Viet Duc University Hospital

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Abstract

Introduction: Perineal endometriosis (PEM) greatly affects patients quality of life, especially chronic pelvic pain associated with menstruation. Around the world, there are many studies on endometriosis, but PEM is less mentioned. In Vietnam, there is no study that evaluates the results of surgical treatment for this disease.

Patients and methods: This was a descriptive, retrospective study of 16 patients operated for PEM at Viet Duc Hospital during the period 2019 - 2020.

Results: All 16 patients had a history of vaginal delivery and episiotomy. The mean incubation time was 50.6 ± 44.8 months. Among 16 patients operated for a complete resection of PEM mass, 7 patients presented with anal sphincter invasion requiring anal sphincter repair. The mean length of hospital stay was 8 ± 2.6 days. The average duration of follow-up was 17.43 months. 14/16 (87.5%) patients had good results. 2 patients (12.5%) had recurrence of the painful mass after surgery.

Conclusion: Surgical treatment of PEM, especially in patients with sphincter invasion, provided good results.

Keywords: Perineal endometriosis; Chronic perineal pain; Episiotomy; Anal sphincter.

Introduction

Endometriosis is a disease characterized by the presence of endometrial glands and stroma outside the uterus. This is a very common gynecological disease that presents among females of childbearing age with an incidence of 5 to 10% of this population. Perineal endometriosis (PEM) is less common, covers only 0.31% among female patients treated by surgery at hospital, according to Lan Zhu research (2009) [1]. PEM in the study is manifested with the presence of endometrial

tissue in perineal region. This rare disease might heavily disrupt patients quality of life, especially chronic pelvic pain associated with menstruation. The preferred treatment is to perform surgery with the aim of getting rid mostly damaged endometrial tissues while preserving the adjuvant structures. If there has anal sphincter involvement, the surgery should be performed by experienced proctologist to avoid the recurrence and the risk of damaged anal sphincter leading to defecating disorder. Although globally there has many researches on

endometriosis, PEM is much less concerned. In Viet Nam, besides clinical reports from author Le Tu Hoang and Nguyen Sy Lanh (2005) [2], we haven't seen any studies that evaluate surgical results from this disease. Therefore, this study was conducted to demonstrate clinical and subclinical manifestations of PEM as well as evaluate the surgical results in Viet Duc hospital.

Materials and methods

Research subjects:

We included 16 female patients who were diagnosed PEM and treated by surgery at the Center of colorectal surgery of Viet Duc University Hospital from 2019 to 2020.

Method:

Study design: Descriptive case study

Data collection: It was collected through patients' medical records, post-operative re-examination and telephone interview.

We retrospectively collected patient clinical data (including medical history, incubation period, symptoms, treatment, outcome and observed early post-operative complications such as pain after surgery (evaluated using VAS- Visual Analogue Scale 0-10), infection, post-operative bleeding and incontinence. Moreover, some late complications such as post-operative unsightly or painful scars, relapse or defecating disorder. The incubation period was defined as the period from the last delivery to the time when symptoms appeared.

Post-operative re-examination and result evaluation: followed up 1 month after hospital discharge and telephone interview at 3 months, 6 months after discharge until May 2021.

Data analysis: Data was analyzed with REDcap software via Excel.

Research ethics:

Study was strictly conducted following all principles of research ethics including, commit to no conflicts of interest in research, protect patient confidentiality, report research honestly and precisely, collected data can only be used for research purposes.

Results

Demographic characteristic:

Table 1. Demographic characteristic

Characteristic	Average	Min	Max
Age (year)	34,0 ± 5,3	25	46
BMI (kg/m ²)	20,3 ± 2,3	16	25.5
Incubation period	50,6 ± 44,8	7	168

Clinical and sub-clinical characteristics:

Nodules were detected around perineal scars, accompanied by chronic menstruation-related pain (100%) (right side : 11/16 cases (68,75%, left side: 5/16 (31,25%))

Medical history: 100% patients had vaginal delivery and were treated for episiotomy. 2/16 patients have already been treated for PEM but not sure whether there had anal sphincter involvement. After surgical treatment, these two patients relapsed and presented with nodules around perineal region: 1 patient relapsed after 10 months, 1 patient relapsed after 23 months.

MRI Findings: 15/16 cases performed pre-operative MRI examination.

Table 2. MRI findings

Findings	MRI (N =15)		
	High signal	Equivalent signal	Low signal
T1 (Fat-suppressed + gadolinium-enhanced)	14 (93,33%)	-	-
T2	10 (66,67%)	2 (13,33%)	3 (20,00%)
Average size of lesion	22,6 mm (from 7 to 46 mm)		
Invasion rate	5/15 (33,33%)		

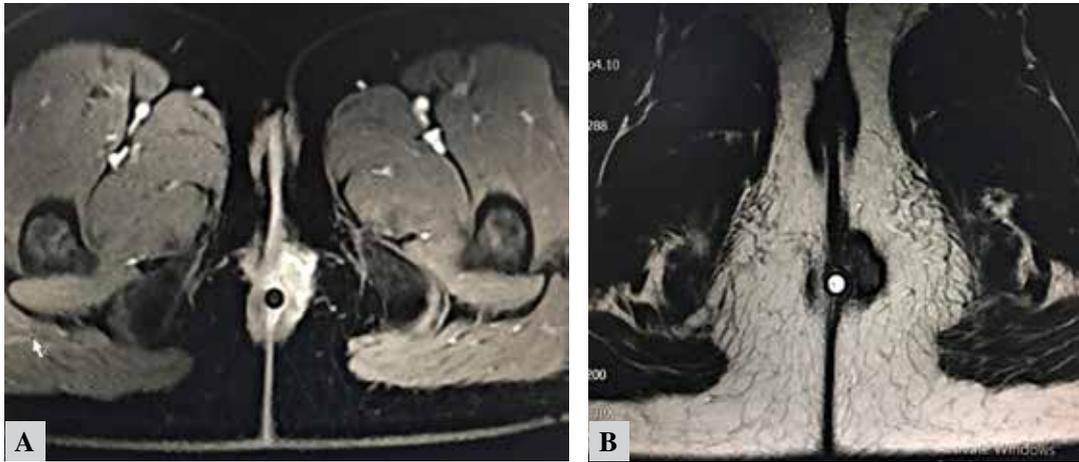


Figure 1. Patient Đỗ Thị Thu H. 46 years old: lesion with high signal on T1 with fat-suppressed (A) and patchy high signal on T2 (B)

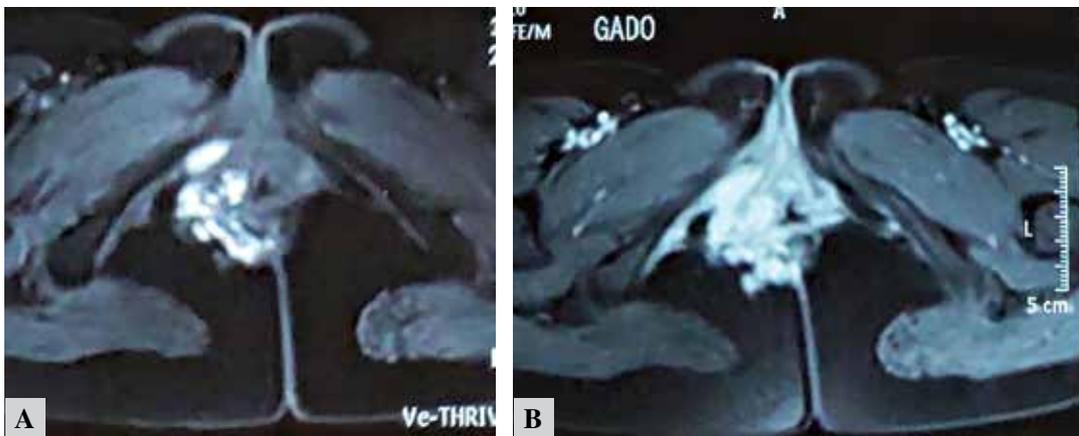


Figure 2. Patient Nguyễn Thị N. 34 years old: PEM lesion invaded anal sphincter
 A. T1 fat-suppressed without gadolinium-enhanced, B. T1 fat-suppressed with gadolinium-enhanced.

All patients were treated by surgery under spinal anesthesia during all the procedure.

Observed lesion during operation: Average size of the lesion was $2,7 \pm 1,0$ cm (from 1 to 5 cm). 7/16 (43,75%) patients who had lesion invading anal sphincter were indicated to perform anal sphincter repair during surgery

Treatment Results:

Early post-operative outcomes:

Pain evaluation at the first day after surgery: Average VAS score $4 \pm 1,4$ (from 3 to 7). Paracetamol was used for pain relief in 11/16 cases (68,75%), whereas Morphine was used in 2/16 cases (12,5%).

Urinary catheterization and parenteral nutrition

were indicated for all the patients to keep surgical wound clean and prevent the risk of infection.

Table 3. Post-operative treatment characteristics

Early post-operative observation (N = 16)	Average	Min	Max
VAS score (first day after surgery)	$4,0 \pm 1,4$	3	7
Duration of urinary catheterization (days)	$3,7 \pm 2,7$	1	9
Duration (days)	$2,7 \pm 2,0$	1	7
First defecation (days after surgery)	$4,0 \pm 2,3$	2	9

Early complications after surgery: 14/16 patients were normal. one patient who had defecating disorder was treated with Forlax. One patient who had mild swelling at incision site was treated with antibiotic including Betalactam, Metronidazole and Ciprofloxacin. No reports about other complications such as surgical wound bleeding, removal of suture for severe wound infection. Discharged patients were all stable.

Histopathology examination: All the patients presented with PEM in histopathological examination, fortunately there had no malignancy.

Average length of stay was $8 \pm 2,6$ days (from 5 to 14 days).

Outcomes after hospital discharge:

Average follow-up period: 17,4 months (from 7 to 27 months)

14/16 cases (87,5%) showed no unusual symptoms: no unsightly scar, no recurrence of post-surgical pain, no fecal incontinence. 2/16 cases (12,5%) appeared recurrence of post-surgical pain at one month and 6 months

Discussions

In our series, the mean age was $34,0 \pm 5,3$ (range, 25-46) years. This result matched with study from Na Chen (2012) [3] in which mean age of 31 cases was 33,4 (range, 26-43) years as well as report of Juanqing Li (2015) [4] in which mean age of 17 patients was 34,35 (range, 26-57) years.

The average BMI was $20,3 \pm 2,3$ kg/m² (16-25,5 kg/ m²). Correlation analysis from Yan Liang research (2021) [5] showed that the incubation period of PEM was unrelated to the age of onset, delivery age, breastfeeding period, and the time of return to menses, but it was negatively correlated with BMI at delivery and BMI within one month after delivery ($R_2 = 0,53 / 0,86$, $P < 0,05$). In our study, we did not evaluate this correlation.

In our series, all the patients had vaginal delivery and were treated for episiotomy as well as accompanied by chronic menstruation-related pain. This finding was also found in many other clinical

case reports or large scale studies around the world [6-9].

The incubation period was defined as the period from the last delivery to the time when symptoms appeared or when lesions were found in perineal area. In our study, average incubation period of 16 patients was 50,6 months (from 7 to 168 months). This result was quite higher than in Na Chen research (2012) [3] which was 36 months and in Yu Liu report (2020) [10] was 42,44 months.

15/16 patients were indicated for MRI with contrast to find deep lesions and evaluate whether there has anal sphincter involvement. 14/15 cases (93,75%) occurred with high signal on fat-suppressed T1 via gadolinium-enhanced. Signal on T2 diversely manifested: low signal (20%), equivalent signal (13,3%) and high signal (66,67%). These results corresponded to various stages of hemoglobin degradation: acute, sub-acute or chronic [11].

All the patients were treated surgically under spinal anesthesia. Post-operative histopathological examination detected PEM in 16/16 cases. 7/16 patients (43,75%) who had lesions invading anal sphincter were indicated for anal sphincter repair during surgery. Torn sphincter can be repaired using overlap repair technique or approximation repair. Up to date, only one randomized clinical trial study of Fitzpatrick [12] compared these two techniques and showed that there had no significant differences in fecal incontinence rate.

Our study determined average size of perineal lesions was $2,7 \pm 1,0$ cm (from 1 to 5 cm). This result corresponded to Lan Zhu study (2009) [1] in which average perineal lesions size was 2,7 cm (from 0,5 to 6 cm) or in Juanqing Li study (2015) [4] was 2,38 cm (from 1 to 4 cm).

Rates of anal sphincter involvement were 7/16 (43,75%). This rate was lower than collected data from several studies from 1990 to 2010, 72,2% according to Lan Zhu study (2009) [1] and 48,4% in Na Chen study (2012) [3]. However, when compared with some researches from 2010 to 2020,

for instance, Juanqing Li report (2015) [4] and Yu Liu (2020) [10] with 35,3%, 28,57% cases of lesions invading anal sphincter, respectively, the rate in our study was relatively higher.

During the hospitalization, patients were treated for pain relief based on 3 stages: mild pain (paracetamol or NSAIDs), moderate pain (tramadol) or severe pain (opioid). Paracetamol was used in most cases (11/16, 68,75%) to relieve acute pain after surgery. Morphine was used in 2/16 cases (12,5%). All the patients were indicated for urinary catheterization and parental nutrition to avoid urine and feces presented in perineal region which might result in surgical wound infection, especially accompanied by anal sphincter repair. Average urinary catheterization period was 3,7 days, while average parental nutrition period was 2,7 days (from 1 to 7 days).

We also recorded several early post-operative complications such as bleeding, surgical site infection, fecal and urinary incontinence. Results showed one case presented with mild surgical site infection (N.T.N 34 years old) (6,25%) and was compromised with Ciprofloxacin and removal of sutures. This patient had lesions invading anal sphincter in the 7-11 o'clock direction and on the right side of Bartholins glands. Although lesion size of this patient was also the biggest among 16 cases in research (5cm x 5cm x 4cm), no urinary and fecal incontinence occurred after surgery. Furthermore, this patient was discharged in normal condition after 14-day hospital stay.

We also observed several late complications such as post-operative unsightly scar, pain at surgical site, recurrence of perineal lesions and fecal incontinence at one month after surgery, at three months and 6 months after hospital discharge until May 2021. Average follow-up period was 17,43 months (from 7-27 months). Results showed almost no patient (87,5%) had unusual symptoms. Only two cases were found with recurrence of perineal lesions at one month and 6 months after surgery.

If only assessed post-operative relapse in Viet Duc University Hospital, the rate was 2/16 (12,5%). However, if we concerned all the treatment methods that were applied to every single patient in study, relapse in perineal region occurred in 3/16 cases (18,75%) after the first operation. This rate was higher than in Michail Matalliotakis study (2020) which was 2/14 cases (14,28%) [9]. It was also comparable to relapse rate in 3 out of 16 patients (18,75%) that were not treated by GnRH after surgery in Yu Liu et al study (2020) [10] with average follow up period was 49 months (from 7 to 86 months), whereas higher than the group treated by GnRH (1/13 = 7,69%). Although relapse rate of PEM increased in patients whom were not treated for endocrine therapy after surgery, this result needed to be further analyzed in future study with higher sample size.

Clear cell carcinoma derived from PEM was found in several reports with very few cases [13] [14] [15]. Due to the limited follow-up period (from 7 to 27 months), we couldnt report any cases that had this complication.

Conclusions

Clinical and subclinical features: The average age of patients was 34 years (from 25 to 46). Average incubation period was 50,6 months. Average size of lesion on MRI was 22,6mm (from 7 to 46mm). Rate of cases involved lesions invading anal sphincter was 7/16 (43,75%).

Surgical outcomes:

Post-operative outcomes: Average length of stay was 8 days. Average VAS score at the first day after surgery was 4/10. Cases that didnt have early post-operative complications were 14/16 (87,5%). 1/16 patients (6,25%) had mild surgical site infection. 1/16 (6,25%) had fecal incontinence but was normal condition after hospital discharge.

Follow up outcomes: Average follow up period was 17,4 months (from 7 to 27 months). 14/16 (87,5%) cases achieved satisfactory outcomes, whereas 2/16 (12,5%) cases relapsed with perineal lesions.

Conflict of interest: The authors declare that they have no conflict of interest.

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