

Surgical outcomes of women undergoing radical resection of deep endometriosis of the sacral plexus: A prospective cohort study

Gernot Hudelist^{1,2,3}  | Ezgi Darici Kurt^{4,5}  | Gábor Szabó⁵ | Dominika Miklos⁵ | Theresa Hudelist⁶ | Attila Bokor⁵ 

¹Department of Gynecology, Center for Endometriosis, Hospital St. John of God, Vienna, Austria

²Rudolfinerhaus Private Clinic and Campus, Vienna, Austria

³Department of Gynecology, Jagiellonian University, Krakow, Poland

⁴Department of Gynecology and IVF, Acibadem University Hospital, Istanbul, Turkey

⁵Department of Obstetrics and Gynecology, Center for Endometriosis, Semmelweis University, Budapest, Hungary

⁶Paracelsus Medical University, Nuremberg, Germany

Correspondence

Gernot Hudelist, Department of Gynecology, Center for Endometriosis Hospital St. John of God, Johannes Gott Platz 1, 1020 Vienna, Austria.
Email: gernot.hudelist@meduniwien.ac.at

Abstract

Introduction: Surgical resection of sacral plexus endometriosis (SPE) is contemplated in severely symptomatic patients not responding to medical or hormonal therapy. However, there is only limited data on the effects of surgery on pain and neurological symptoms. This study aims to report on the surgical outcomes in terms of pain and neurological symptom reduction in women undergoing surgical resection of SPE.

Material and Methods: Thirty premenopausal patients with histologically confirmed SPE who underwent surgical resection of the disease between 2018 and 2024 were included in this multicenter prospective analysis. The primary outcome was the change in neurological symptoms reflected by sacral radiculopathy including dysaesthesia, paraesthesia, hyperaesthesia, and pain. The secondary outcome was post-surgical morbidity reflected by rates of major intra- and postoperative complications.

Results: All patients exhibited DE affecting the sacral roots S1-S4, whereas no case of isolated supracardinal sciatic nerve involvement was observed. Out of 30 patients, one was lost to follow-up leaving 29 patients for final analysis. Six (20%) of the 30 patients underwent partial resection of the sacral root because of endometriotic infiltration of the neural tissue. In all other patients, dissection and shaving with cold scissors were sufficient to remove DE affecting the sacral root. Dysaesthesia was observed in 13/30 (43.3%), paraesthesia in 16/30 (53.3%), hyperaesthesia in 5/30 (16.7%), and secondary motor dysfunction in 4/30 (13.3%), preoperatively. The mean follow-up interval was 25.5 ± 20.2 months showing an overall improvement in sacral radiculopathy in 93.1% (27/29) of the patients. A significant decrease in numeric rating scale (NRS) scores of dysaesthesia ($p=0.003$), paraesthesia ($p \leq 0.001$) and hyperaesthesia ($p=0.068$) were observed post-surgically. Equally, reduced pain symptoms including dysmenorrhea, dyspareunia and dyschezia (all $p \leq 0.001$) with a relevant increase in post-surgical quality of life

Abbreviations: DE, deep endometriosis; NRS, numeric rating scale; QoL, quality of life; SPE, sacral plexus endometriosis.

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scores ($p \leq 0.001$) were recorded. De novo hyperaesthesia and paraesthesia occurred in 6.8% (2/29) and 3.4% (1/29) of the patients, respectively. Major Clavien-Dindo grade III complications occurred in 13.3% (4/30) of the cases.

Conclusions: Radical resection of symptomatic deep endometriosis affecting the sacral plexus reduces neurological and pain symptoms and leads to an increase in quality of life but is associated with high surgical morbidity.

KEYWORDS

deep endometriosis, morbidity, quality of life, radical resection, sacral plexus; neurological and pain symptoms

1 | INTRODUCTION

Deep colorectal endometriosis (DE) affects 5% to 12% of women diagnosed with DE.¹ Women suffering from DE frequently report various cyclic and non-cyclic pain symptoms. These include visceral, non-neuropathic pain² and gastrointestinal dysfunction.³ Visceral pain sensations caused by ovarian, peritoneal, or colorectal DE such as dysmenorrhea, dyschezia, or dyspareunia are predominantly transmitted by autonomous nerve fibers of the inferior hypogastric plexus and typically arise in conjunction with vegetative symptoms.⁴ In contrast, DE involvement of structures of the sacral plexus, that is, sacral nerve roots S1-S4 or fibers of the supracardinal portion of the sciatic nerve will cause somatic and neuropathic pain leading to so-called sacral radiculopathy.^{5,6}

This entity of DE is very rare. Pain results from irritation, infiltration, and/or compression of neural structures.^{5,7} In contrast to the vague, poorly localized character of visceral pain, sacral radiculopathy is described as a clearly localized, sharp, painful, and burning or prickling sensation (dysaesthesia) with electric, shock-like tingling (paraesthesia) and/or excessive physical sensitivity (hyperaesthesia). Radiation of the pain sensation to the lower extremity corresponding to sacral dermatomes is commonly observed. In rare cases, it may also be associated with motor weakness due to sciatic nerve DE or secondary protective posture and motor weakness due to pain. In addition, urinary urgency and frequency caused by irritation of autonomous parasympathetic splanchnic nerve fibers may arise.⁶

In general, these symptoms add up to the visceral pain components in affected patients and may be associated with a relevant decrease in quality of life (QoL). Surgical resection is considered in cases where the patient is not responding to medical, that is, hormonal treatments but is associated with several complicating aspects: First, extensive surgical expertise and neuroanatomical knowledge is necessary to perform procedures involving dissection of the sacral plexus and adjacent structures. These interventions are not considered as standard procedures in surgical gynecology.^{5,8,9} Secondly, surgical interventions including colorectal, vaginal, parametrial resection and dissection or resection of neural structures confer relevant intraoperative and postoperative morbidity and may cause long-term sequelae such as dysfunction of the pelvic autonomous nerve supply. This can be explained by damage to the hypogastric

Key message

Radical resection of symptomatic deep endometriosis affecting the sacral plexus confers reduction of pain and reduction of neurological symptoms but is associated with high surgical morbidity.

plexus which usually has to be sacrificed in cases where resection of unilateral sacral plexus endometriosis (SPE) is necessary.⁹

So far, there are only limited case series and cohorts reporting on post-surgical outcomes of women undergoing surgical resection of SPE. Within this, a review summarized a total of 362 patients following surgical treatment of SPE including 40 eligible studies with the two largest single center series including 267 and 52 patients respectively.^{5,8} The aim of the present study was to report on the surgical outcomes in terms of pain reduction and rates of severe complications and sequelae of women with severely symptomatic SPE treated in two linked tertiary referral centers in order to extend the knowledge on short- and long-term results following resection of SPE.

2 | MATERIAL AND METHODS

2.1 | Study Design and Patients

This prospective cohort study included women scheduled for surgery for severely symptomatic DE affecting the sacral plexus (SPE). Inclusion criteria were as follows: pre-surgical diagnosis of SPE based on typical symptoms (unilateral sacral radiculopathy causing dysaesthesia and/or paraesthesia in the corresponding dermatomes) in combination with visualization of SPE by transvaginal sonography (TVS) as described previously^{7,10} and/or magnetic resonance imaging (MRI).¹¹ Indication for the surgical dissection of the sacral plexus was therapy-resistant decreased QoL—reducing pain and sacral radiculopathy resulting from compression, infiltration and/or irritation of sacral nerve roots S1-S4 of the sacral plexus associated

with symptoms of lower buttock pain, leg pain and lower extremity numbness or tingling in the corresponding dermatomes. Exclusion criteria were history of pelvic malignancy, previous surgery including dissection of the sacral plexus or neurological or orthopedic diseases also leading to sacral radiculopathy.

In all patients included, dissection of the sacral roots was performed combined with additional surgical procedures for concomitant DE at additional anatomical locations (Table 1). This was

TABLE 1 Baseline characteristics and intraoperative findings of patients undergoing surgical resection of sacral plexus endometriosis.

	All patients (n = 30)
Age (years)	36.83 ± 5.51
BMI	23.71 ± 2.68
Sacral nerve root involved ^a	
S1	8 (26.7%)
S2	25 (83.3%)
S3	17 (56.7%)
S4	2 (6.7%)
ENZIAN classification	
ENZIAN A (Vagina/RVS)	15 (50%)
ENZIAN B (USL, Parametrium)	19 (63.3%)
ENZIAN C (Rectum/Sigmoid)	18 (60%)
C1 (<1 cm)	0 (0%)
C2 (1–3 cm)	5 (16.6%)
C3 (>3 cm)	13 (43.3%)
FA	16 (53.3%)
FB	6 (20%)
FU	6 (20%)
Hospital stay (days)	6.31 ± 3.25
Surgical procedures	
Unilateral adnexectomy	6 (20%)
Bilateral adnexectomy	4 (13.3%)
Posterior vaginal fornix resection	17 (56.7%)
Total hysterectomy	2 (6.7%)
Nerve-vessel sparing segmental resection	15 (50%)
Full-thickness discoid resection	2 (6.7%)
Ureter resection	6 (20%)
Bladder resection	4 (13.3%)
Clavien-Dindo major complication rate	4 (13.3%)
Grade III	4 (13.3%)
Rectovaginal fistula	3 (10%)
Pelvic abscess	1 (3.3%)
Grade IV	0 (0%)

Note: Data are presented as mean ± standard deviation or n (%). Abbreviations: BMI, body mass index; RVS, rectovaginal septum; USL, Uterosacral ligament.

^aInvolvement of the nerve alone or in combination with another nerve root.

followed by surgical confirmation and histological proof of endometriotic tissue either fully infiltrating or densely adherent to structures of the sacral plexus (sacral roots S1, S2, S3, or S4). All procedures were performed by G.H. and A.B. in a tertiary referral center setting at the Department of Obstetrics and Gynecology of the Semmelweis University Budapest, Hungary (18 patients), and the Hospital St. John of God, Vienna (12 patients) from August 2018 to January 2024. The goal of surgical treatment was to remove all macroscopically visible DE lesions. In patients with bowel DE >3 cm in diameter as well as in patients with multiple, that is, more than one bowel lesion, nerve-vessel sparing segmental resection was performed.^{12,13} Full-thickness discoid resection was performed for lesions <3 cm in diameter. Initially, all procedures were performed laparoscopically. Surgical steps of nerve-vessel sparing segmental resection, full-thickness discoid resection,¹⁴ and the steps of dissection of the sacral roots S1–S4 have been described previously¹⁵; However, systematic ligation of deep branches of the internal iliac vein/veins of the gluteal or obturator region was only performed in cases where SPE was not accessible due to venous branches crossing the nerval tissue.

The primary outcome of the present study was the post-surgical change of SPE-associated sacral radiculopathy and cyclic pain symptoms assessed by the NRS score. Moreover, the rates of severe complications as defined by Clavien-Dindo grade III–IV¹⁶ were investigated as secondary outcome.

Clinical characteristics including age, body mass index, gravidity, parity, and previous surgical treatments were recorded and are depicted in Table 1. Pain symptoms, in particular dysmenorrhea, dyspareunia, and dyschezia were assessed using numeric rating scale (NRS), in which the patient selects a whole number (0–10) to describe the pain intensity. Similarly, neurological symptoms reflecting sacral radiculopathy including dysaesthesia, paraesthesia, hyperaesthesia, and QoL rated from 0 to 10 were also recorded using the NRS (Table 2). DE was classified using the #Enzian classification.¹⁷ STROBE criteria were followed for reporting.¹⁸

2.2 | Statistical Analyses

Assuming a medium effect of the intervention on symptom reduction ($d=0.50$), with a probability of error of 5% and a power of 0.80, the required sample size was 27 cases. All parameters were not distributed normally except age. All metric and ordinal data are described using mean values and standard deviations, median and quartiles as well as minimum and maximum. Categorical data are presented with frequencies and percentages. The normal distribution was checked using the Kolmogorov–Smirnov test and Shapiro–Wilk test as well as visually using a histogram. Ordinal variables were tested for significant differences between the two measurement times using the Wilcoxon test. Dichotomous variables were tested for significant differences between the two measurement times using the Mc Nemar test. All statistical analyses were performed using the SPSS 28 (SPSS Inc., Chicago, Illinois,

TABLE 2 Pre-surgical and long-term outcomes regarding neurological symptoms, pain symptoms, quality of life scores following resection of sacral plexus endometriosis.

	Pre-surgical	Post-surgical	p-value
Neurological symptoms			
Sacral radiculopathy	29 (100%)	11 (37.9%)	
NRS score sacral radiculopathy	8.10 ± 1.65	1.72 ± 2.76	<0.001
Dysaesthesia	12 (41.4%)	7 (24.1%)	
NRS score dysaesthesia	6.58 ± 2.23	1.58 ± 3.03	0.003
Paraesthesia	16 (55.2%)	9 (31.0%)	
NRS score paraesthesia	6.06 ± 2.62	1.75 ± 2.82	<0.001
De novo paraesthesia	—	1 (3.4%)	n.a.
Hyperaesthesia	5 (17.2%)	3 (10.3%)	
NRS score hyperaesthesia	8.20 ± 1.92	2.40 ± 4.28	0.06
De novo hyperaesthesia	—	2 (6.8%)	n.a.
Motor dysfunction	4 (13.8%)	3 (10.3%)	
Pain symptoms			
Dysmenorrhea	26 (89.7%)	17 (58.6%)	
NRS score dysmenorrhea	8.23 ± 1.42	2.85 ± 2.98	<0.001
Dyschezia	17 (58.6%)	8 (27.5%)	
NRS score dyschezia	6.94 ± 1.78	1.53 ± 2.13	<0.001
Dyspareunia	19 (65.5%)	12 (41.3%)	
NRS score dyspareunia	6.63 ± 2.06	2.37 ± 2.48	<0.001
Dysuria	5 (17.2%)	6 (20.6%)	
NRS score dyspareunia	6.0 ± 2.35	0.80 ± 1.79	0.04
Quality of life NRS score	2.59 ± 1.40	8.10 ± 1.80	<0.001

Note: Data are presented as mean ± standard deviation or *n* (%).
 Abbreviations: NRS, numeric rating scale; n.a., not applicable.

USA) software program. A value of $p \leq 0.05$ was considered to be statistically significant.

3 | RESULTS

A total of 30 consecutive patients underwent surgery for SPE and fulfilled the inclusion criteria. Table 1 shows intraoperative findings and distribution of DE according to the #Enzian classification. Surgical procedures and intra- and post-surgical complications are depicted in Table 1. All patients exhibited involvement of the sacral roots; none of the 30 consecutive patients showed infiltration of the sciatic nerve in its supracardinal portion as described in other case series.⁵ Six (20%) of the 30 patients underwent partial resection of the sacral root because of endometriotic infiltration of the neural tissue. In all other patients, dissection and shaving with cold scissors were sufficient to remove DE affecting the sacral root.

Additional major surgical procedures involved the digestive tract (full-thickness discoid resection and nerve-vessel sparing segmental resection) in 56.7% (17/30) of the cases and the urinary tract in 29.6% (8/30) of the patients. The major complication (Grade III-IV) rate was 13.3% (4/30) according to the Clavien-Dindo classification (Table 1). Three patients had a rectovaginal fistula (10%) and three patients (10%) presented with voiding dysfunction requiring self-catheterization over 3 weeks, one (3.3%) of which required permanent catheterization. This complication may, however, be related to unilateral damage of the inferior hypogastric plexus during resection of parametrial DE rather than due to sacral neurolysis. Furthermore, one patient (3.3%) exhibited pre-sacral abscess formation.

All except one patient were eligible for follow-up. These patients took part in a telephone interview and answered a self-administered questionnaire on neurological and pain symptoms at the time of follow-up. The mean follow-up interval was 25.5 ± 20.2 months. Dysaesthesia was observed in 12/29 (41.4%), paraesthesia in 16/29 (55.2%), hyperaesthesia in 5/29 (17.2%), and motor dysfunction in 4/29 (13.8%), preoperatively. Table 2 and Figures 1–3 depict changes in neurological symptoms, that is, sacral radiculopathy, pain symptoms, and QoL.

An overall improvement of sacral radiculopathy was observed in 93.1% (27/29) patients. In detail, dysaesthesia (6.5 ± 2.2 vs. 1.5 ± 3.0, pre- and postoperative, respectively [$p = 0.003$]), paraesthesia (6.0 ± 2.6 vs. 1.7 ± 2.8, pre- and postoperative, respectively [$p \leq 0.001$]) and hyperaesthesia (8.2 ± 1.9 vs. 2.4 ± 4.2, pre- and postoperative, respectively [$p = 0.068$]) were significantly decreased following surgery.

In addition, NRS scores changed significantly for pain symptoms including dysmenorrhea (8.2 ± 1.4 vs. 2.8 ± 2.9, pre- and postoperative, respectively [$p \leq 0.001$]), dyspareunia (6.6 ± 2.0 vs. 2.3 ± 2.4, pre- and postoperative, respectively [$p \leq 0.001$]) and dyschezia (6.9 ± 1.7 vs. 1.5 ± 2.1, pre- and postoperative, respectively [$p \leq 0.001$]). There was a relevant increase in post-surgical QoL scores as rated by NRS (2.5 ± 1.4 vs. 8.1 ± 1.8, pre and postoperative, respectively [$p \leq 0.00$, Table 2]). De novo hyperaesthesia was reported in two patients (6.8%) and de novo paraesthesia in one patient (3.4%) after surgery. However, no patient experienced postoperative de novo motor dysfunction which also could not be observed at the time of follow-up in patients with previous motor weakness.

4 | DISCUSSION

In the present study, we report the outcomes of the so far third-largest series of surgically treated patients with SPE (Figure 4). This entity is very rare and may cause severe pain symptoms including sacral radiculopathy leading to a relevant reduction of QoL. Our data clearly show that interventions involving dissection of the sacral plexus and resection of SPE entail a relevant risk of intra- and postoperative major complications. In our series, 2 patients

FIGURE 1 Semi-quantitative data (box blots) on changes in sacral radiculopathy following surgery for sacral plexus endometriosis.

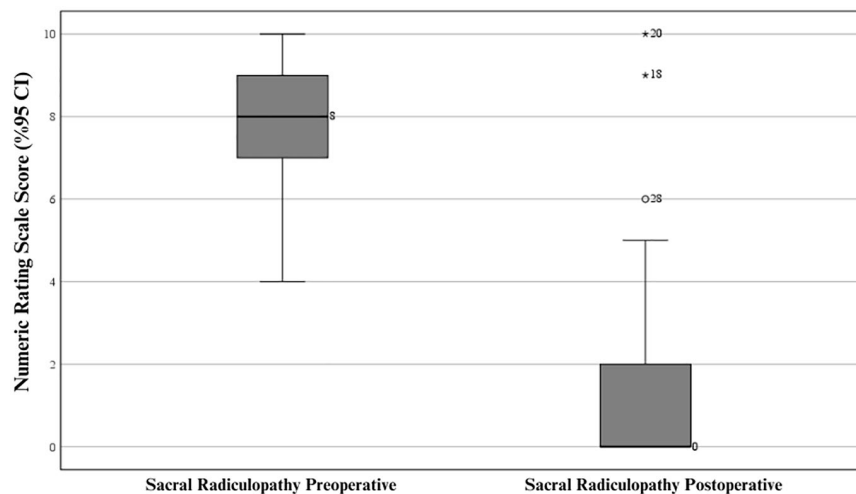


FIGURE 2 Semi-quantitative data (box blots) on changes in dysmenorrhea following surgery for sacral plexus endometriosis.

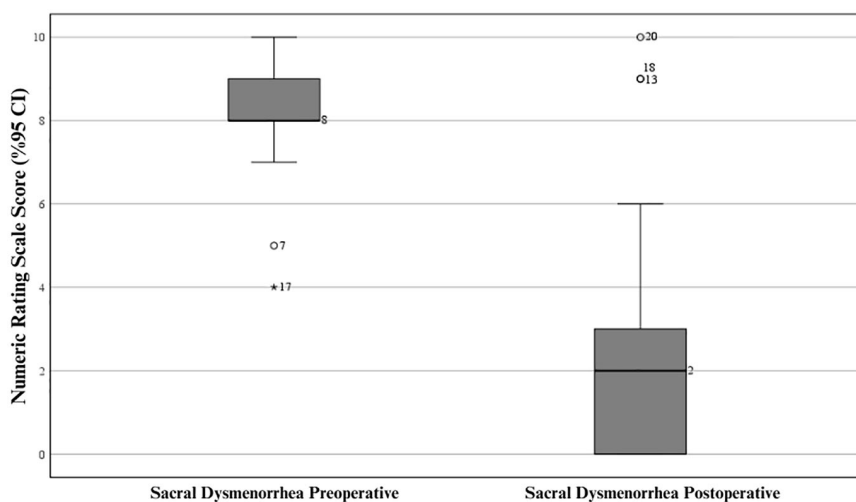
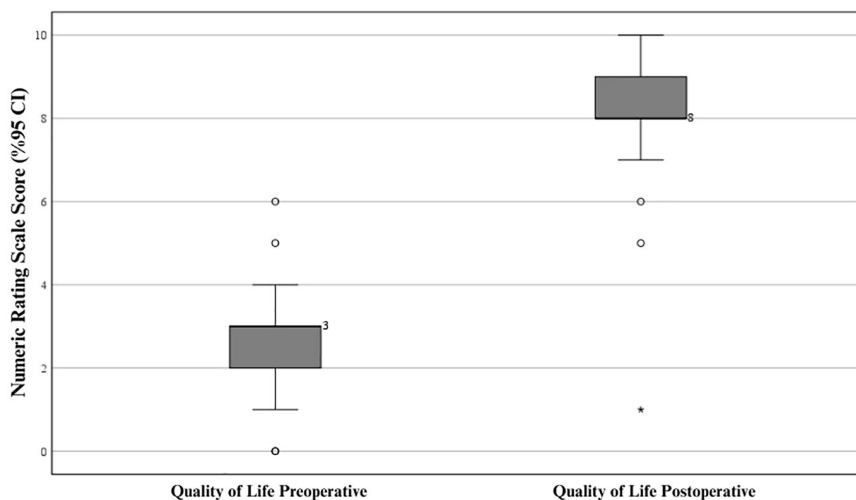


FIGURE 3 Semi-quantitative data (box blots) on quality of life following surgery for sacral plexus endometriosis.



experienced large vessel bleeding necessitating blood transfusions and large vessel ligation. Furthermore, we observed an overall post-surgical major complication rate of 13.8% including three rectovaginal fistulas and one pelvic abscess. All these patients underwent further surgical interventions to manage these

complications. Finally, the predominant post-surgical sequelae were transient and persistent voiding dysfunction with three patients requiring self-catheterization over 3 weeks and one patient with persistent voiding dysfunction necessitating permanent self-catheterization at the time of follow-up.

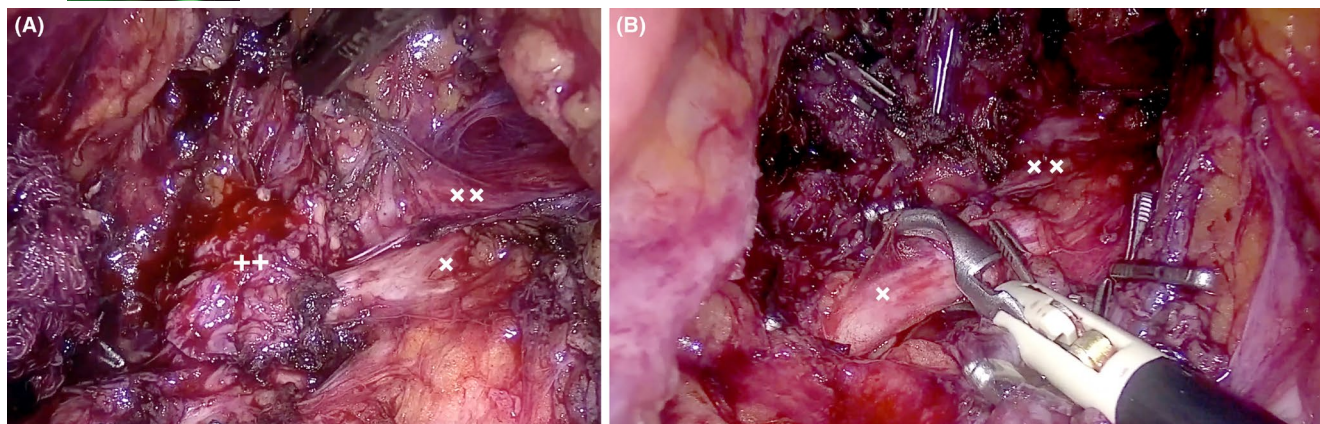


FIGURE 4 (A) Intraoperative situs following dissection of sacral plexus endometriosis, “x” depicts the sacral root S2, “++” shows DE lesion infiltrating the sacral root S2 and partly S3 which is marked as “xx”. (B) Intraoperative situs following resection of sacral plexus endometriosis; “x” depicts the sacral root S2 which has been cleared from the DE lesion by sacral neurolysis and shaving with cold scissors; “xx” depicts sacral root S3.

Our data are in line with the major intra- and post-surgical complication rates reported by Roman et al.⁸ who reviewed 52 consecutive cases undergoing resection of SPE. Similarly, the predominant intraoperative complication was large vessel bleeding, that is, severe hemorrhage due to injury to hypogastric veins. In our series, one patient experienced injury of the internal iliac artery which was then ligated following conversion to laparotomy; the second case was related to injury of the large internal iliac vein which was then controlled by ligation and coagulation of contributing vessels. As already observed by Roman and colleagues,⁸ the predominant post-surgical complication in our cohort was the development of a rectovaginal fistula in 3 women (10%) of our series. Compared to the rate of rectovaginal fistulae following colorectal surgery for DE in other large series without SPE of around 3% to 4%,¹⁹ the rate of 10% in our cohort or up to 13.5%⁸ in patients with SPE and DE at other sites including the rectum and vagina is high. It is, however, in contrast to the numbers published by Possover et al.²⁰ who did not observe any major complications following surgery for SPE in the so-far largest series published including 259 patients. Whether these numbers regarding major complications or sequelae of this single-center series are in any way reproducible or reliable remains to be elucidated.

We presume that the high rate of resection of concomitant vaginal DE and colectomy in over half of our patients also in combination with low rectal resection appears to be the predominant causative factor of this complication. In line with Roman et al., patients with SPE often present with concomitant vaginal and colorectal endometriosis which clearly adds surgical complexity to these procedures.¹⁹

On the contrary, the results of the present analysis demonstrate that despite the high surgical morbidity of these complex interventions, most of the patients experienced a significant relief of their neurological symptoms, that is, sacral radiculopathy which was also accompanied by a large reduction of dysmenorrhea, dyspareunia and cyclic defecation pain. Therefore, severely symptomatic patients with a relevant reduction of QoL not responding to medical therapy

may be offered this treatment option in case these procedures can be performed with the necessary surgical experience and expertise.

Our work does have several limitations. First and foremost, the present analysis is based on a limited number of patients with a very rare entity of DE and was analyzed in the absence of a control group and randomization. Secondly, all these patients presented with a heterogenous anatomical distribution of DE which makes a standardized comparison difficult. Finally, the outcomes of this analysis cannot be translated into the everyday clinical practice of centers treating DE surgically as these interventions require extensive surgical and neuropelvic training and experience. The authors do not support to offer these procedures without sufficient experience in these surgical techniques but also in managing complications and sequelae.

Despite the fact that these interventions entail a relevant risk of intraoperative and postoperative complications and sequelae and require exceptional surgical skills and experience in a multidisciplinary tertiary referral center setting due to their surgical complexity, surgical treatment does offer a therapeutic option for severely symptomatic patients.

5 | CONCLUSION

Radical resection of symptomatic DE affecting the sacral plexus reduces pain and neurological symptoms in the selected patient cohort, but it is associated with high surgical morbidity rates and requires surgical experience in a multidisciplinary tertiary referral center setting.

AUTHOR CONTRIBUTIONS

Gernot Hudelist: Project development. Ezgi Darici Kurt and Theresa Hudelist: Data collection and manuscript writing. Dominika Miklos: Data collection. Attila Bokor: Project development and manuscript writing. All authors commented on the manuscript. All authors read and approved the final manuscript.

CONFLICT OF INTEREST STATEMENT

The authors have no conflicts of interest to declare.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author.

ETHICS STATEMENT

The Institutional Review Board at Hospital St. John of God approved the study on January 20, 2017 (reference number 2017/1-BB).

ORCID

Gernot Hudelist  <https://orcid.org/0000-0002-9424-2208>

Ezgi Darici  <https://orcid.org/0000-0001-9570-1165>

Attila Bokor  <https://orcid.org/0000-0001-9416-2438>

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