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RESEARCH ARTICLE

Gynaecological Surgery

Surgical management of pudendal nerve entrapment after sacrospinous ligament fixation

Eva V. Vodegel[1] | **Kim W. M. van Delft**[1] | **Charlotte H. C. Nuboer**[1] | **Claudia R. Kowalik**[1,2] | **Jan-Paul W. R. Roovers**[1,2]

[1]Department of Obstetrics and Gynaecology, Amsterdam Reproduction & Development Research Institute, Amsterdam UMC, University of Amsterdam, Amsterdam, the Netherlands

[2]Bergman Clinics – Vrouw, Amsterdam, the Netherlands

Correspondence
Eva V. Vodegel, Amsterdam UMC, University of Amsterdam, location AMC, Department of Obstetrics and Gynaecology, Meibergdreef 9, 1105 AZ, Amsterdam, the Netherlands.
Email: e.v.vodegel@amsterdamumc.nl

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Abstract

Objective: To analyse the efficacy of sacrospinous ligament (SSL) suture removal on the reduction of pain symptoms in the case of suspected pudendal nerve entrapment after sacrospinous ligament fixation (SSLF).

Design: Retrospective cohort study.

Setting: Tertiary referral centre, the Netherlands.

Population: A cohort of 21 women having their SSLF sutures removed because of SSLF-related pain symptoms.

Methods: Clinical record review.

Main outcome measures: The primary outcome was reduction of pain after SSL suture removal. Secondary outcome measures were time interval between suture placement and suture removal, complete suture removal, adverse events and recurrence of pelvic organ prolapse (POP).

Results: A total of 21 women underwent SSL suture removal for severe and/or persistent pain, which was confirmed on clinical examination: 95% of the women (20/21) reported pain reduction after suture removal, and 57% reported complete pain relief. The time interval between suture placement and suture removal was at a median of 414 days (range 8–1855 days). Sutures could be completely removed in 86% of cases (18/21). One woman had excessive blood loss (520 ml) without blood transfusion. At 6–8 weeks after surgery, 10% of the women (2/21) had renewed symptomatic POP, stage ≥ 2, for which additional POP surgery was indicated.

Conclusions: When performed by an experienced clinician, SSL suture removal is feasible and efficacious, with low morbidity. In addition, the risk of recurrent POP in the short term appeared to be low.

KEYWORDS
pain relief, POP recurrence, pudendal nerve entrapment, pudendal neuralgia, sacrospinous ligament fixation, sacrospinous ligament suspension, suture removal

Tweetable abstract: The surgical removal of sacrospinous ligament sutures is safe and efficacious for pain relief, even remote from initial placement.

1 | INTRODUCTION

Sacrospinous ligament fixation (SSLF), first described by Sederl in 1958, is a frequently used surgical technique to correct uterine and vaginal vault prolapse via extraperitoneal suspension of the cervix or vaginal apex to the sacrospinous ligament (SSL).[1,2] Although it has a high success rate,[3] entrapment of the pudendal nerve is a complication that may occur.

Sometimes, the nearby inferior gluteal nerve and/or sacral nerves are also affected. This nerve entrapment can cause pain or numbness in the gluteal and posterior thigh on the ipsilateral surgical side.[4]

The literature reveals that the majority of women (55%–84%) experience some gluteal or thigh pain in the immediate postoperative period,[5,6] and that 3%–15% of women experience transient pain at 4–6 weeks following SSLF.[4,6–8] This is probably linked to injury or traction of one of the small nerves that run through the coccygeal–SSL complex, or results from dissection and/or swelling of the pararectal space. Typically, this injury is self-limiting and completely resolves within 6 weeks–6 months postoperatively.[3,4,7,9] However, in the case of severe and/or persistent pain, this may indicate entrapment or trauma of a nerve from the sacral nerve plexus (in most cases the pudendal nerve) by the sutures that fix the cervix or vault to the SSL.[6,10–12] Subsequent gluteal, perineal and lower extremity pain can be debilitating, and is accompanied by paraesthesia and hyperalgesia of the buttock, perineum and/or external genitalia. This pudendal neuralgia may have a devastating impact on a woman's life in the long term, restricting the usual tasks of daily life, particularly when sitting is involved.[13]

The true incidence of pudendal nerve entrapment is unknown but is estimated to be approximately 1%–2%.[14] Treatment of nerve entrapment generally includes the surgical removal of the SSL sutures to accomplish pain relief and, when necessary, the repositioning of new suspension sutures, preferably at a different anatomical location like the contralateral SSL. Studies that evaluate the outcomes of re-operation after nerve entrapment following SSLF are scarce and are limited by the small number of subjects.[5,10,15–17] We intended to perform a study that provides insight into the efficacy of suture removal in women with severe and/or persistent pain following SSLF.

2 | METHODS

2.1 | Study design

A retrospective cohort study was performed in women indicated for SSL suture removal as a result of severe and/or persistent pain symptoms following transvaginal SSLF for apical pelvic organ prolapse. The study was carried out at the Amsterdam University Medical Centre and Bergman Clinics Vrouw Amsterdam in the Netherlands, which are both tertiary referral centres for pelvic floor disorders. Women were included between 1 January 2014 and 1 January 2019. Women were identified through a thorough search of the hospital's computerised surgical database, based on the 10th revision of the International Statistical Classification of Diseases and Related Health Problems (ICD-10) codes for SSL suture removal.

Inclusion criteria for this study was the removal of SSL sutures because of severe and/or persistent buttock and/or lower extremity pain, numbness, weakness, dysaesthesia or a combination of these symptoms. There were no exclusion criteria. Electronic medical records were reviewed to collect baseline demographics, medical history, clinical examination, intraoperative findings during suture placement and suture removal, and postoperative follow up. We used the original referral letters obtained from the referring hospitals, including operating notes if available.

2.2 | Outcome measures

The primary outcome was reduction of pain after SSL suture removal, which was evaluated during the standard outpatient visit at 6–8 weeks after surgery. In the medical record pain was reported as 'completely resolved', 'improved with sequelae' or 'no improvement of pain complaints'. Secondary outcome measures were the time interval between suture placement and removal, complete suture removal, adverse events and recurrence of pelvic organ prolapse at 6 weeks after suture removal. Data collection was completed at least 6 months following the last surgical procedure.

2.3 | Surgical technique

The removal of the SSL sutures was performed using a vaginal approach. All women received antibiotic prophylaxis (cefalozin 1000 mg or metronidazole 500 mg) and had an indwelling catheter during the procedure. Hydrodissection with adrenaline 1:200 000 and xylocaine 2% was applied to the posterior vaginal wall and towards the concerning SSL. The posterior vaginal wall was opened and the rectovaginal fascia was dissected until about 2 cm below the posterior fornix or until the cervix. Using Metzenbaum scissors and blunt dissection, the pararectal space was opened and the sutures at the SSL were identified by palpation. The tissue surrounding the sutures was dissected. After identification and visualisation of the sutures they were cut at the knot, and then complete

suture removal was pursued. The posterior vaginal wall was closed in the usual manner using resorbable suture material. Sometimes the approach for suture identification started from the cervix (or vaginal cuff) if the sutures could not be identified at the SSL. Surgery was performed in a specialised pelvic floor centre by three experienced pelvic floor surgeons.

2.4 | Ethics

The Medical Ethics Review Committee of the Academic Medical Centre Amsterdam (AMC) confirmed that the Medical Research Involving Human Subjects Act (WMO) did not apply to this study and therefore official approval by the committee was not required (ref. no. W21_497 # 21.551). For this study, data were obtained from medical records only, and no additional data have been collected from the women. All women were informed that their medical data are used for healthcare evaluation, data analysis and subsequent publication. All women provided their consent for the use of their (anonymous) medical data for publication.

2.5 | Statistical analysis

Descriptive statistics were used for all included women. Statistical analysis was performed using simple descriptive techniques (mean with standard deviation for variables of normal distribution, and median with range and interquartile range for variables not of normal distribution). Categorical variables were reported as number (%). All data were systematically recorded in an electronic case report form using the web-based data management software Castor EDC (Castor Electronic Data Capture, Amsterdam, the Netherlands, http://castoredc.com). To improve data quality, range checks were incorporated in the electronic case report form. Data handling was performed with anonymous data.

3 | RESULTS

In the inclusion period of 5 years, 21 patients met the inclusion criteria. As a tertiary referral centre, 15 patients were referred for suture removal because of severe and/or persistent pain after SSLF. These women had an SSLF procedure in another hospital. An additional six patients were included in this cohort; they had an SSLF procedure at our own institution (Amsterdam UMC or Bergman Clinics Vrouw Amsterdam), with subsequent severe and/or persistent pain. This makes a total of 21 cases with SSL suture removal between 1 January 2014 and 1 January 2019.

3.1 | Baseline characteristics

The baseline characteristics prior to the SSLF procedure show a median age of 53.5 years, including both premenopausal and postmenopausal women. For all details, see Table 1. All of the women had an SSLF procedure performed at the right SSL with a posterior surgical approach, and in 79% a simultaneous surgical procedure was performed (see Table 2). After the SSLF procedure, pain was reported in multiple areas, with right buttock pain being the most reported site (see Table 3).

3.2 | Suture removal: time interval, complete removal and adverse events

The median time between the SSL suture placement and removal was 414 days (interquartile range, IQR, 216–705 days; range 8–1855 days). In 71% of the women (15/21), complete suture removal was obtained at the first attempt. In the remaining 29% of the women (6/21) with incomplete suture removal, 14% were pain free (3/21) and 14% experienced partial pain relief (3/21). Those last 14% of women underwent a second attempt for complete SSL suture removal to improve the outcome. Complete removal was obtained in all of these cases ($n = 3$). As a result, two of the three women were pain free and one woman obtained no further pain relief (see also Figure 1).

All details of the surgery performed for SSL suture removal are summarised in Table 4. One major adverse event occurred during SSL suture removal: excessive blood loss (520 ml), which did not require blood transfusion.

TABLE 1 Baseline characteristics of the study cohort prior to SSLF

Characteristic	Median or frequency %, $n = 21$
Age, years (median, IQR)	53.5 (45.4–55.7)
BMI, kg/m^2 (median, IQR)	23.9 (21.3–27.4)
Parity (median, range)	2 (1–4)
Current smoker, % (no./total no.)	23.5 (4/17)
Menopausal status, % (no./total no.)	
Premenopausal	42.1 (8/19)
Postmenopausal	47.4 (9/19)
History of uterus extirpation and <55 years old	10.5 (2/19)
Prior pelvic surgery, % (no./total no.)	47.4 (9/19)
Vaginal uterus extirpation	31.6 (6/19)
Anterior colporrhaphy	26.3 (5/19)
Posterior colporrhaphy	15.8 (3/19)
Stress incontinence surgery	5.3 (1/19)
History of micturition complaints, % (no./total no.)	
Urinary incontinence	46.2 (6/13)
Overactive bladder	7.7 (1/13)
Obstructive micturition	7.7 (1/13)
Dysuria	7.7 (1/13)
Clinical assessment, % (no./total no.)	
Pelvic floor hypertonia	0 (0/6)
Pain when palpating the SSL	16.7 (1/6)

Abbreviations: IQR, interquartile range; SSL, sacrospinous ligament.

TABLE 2 Intraoperative characteristics of SSLF procedure

Characteristic	Frequency % (no./total no.), *n* = 21
Surgical approach to SSL	
Anterior	0.0 (0/12)
Posterior	100.0 (12/12)
Sacrospinous ligament used	
Right	100.0 (21/21)
Left	0.0 (0/21)
Suture placement technique – under visualisation	
Breisky retractor + needle holder	50.0 (6/12)
Breisky retractor + device assisted (Capio)	8.3 (1/12)
Suture placement technique – digital approach	
Device assisted only (Capio)	41.7 (5/12)
Type of suture material	
Prolene monofilament	93.3 (14/15)
Mersilene multifilament	6.7 (1/15)
Simultaneous surgical procedures	78.9 (15/19)
Anterior colporrhaphy	63.2 (12/19)
Posterior colporrhaphy	36.8 (7/19)
Perineorrhaphy	10.5 (2/19)
Enterocele repair	5.3 (1/19)
Midurethral sling	10.5 (2/19)
Bilateral salpingo-oophorectomy	5.3 (1/19)
Antibiotic prophylaxis	88.9 (8/9)
Perioperative complications	
>500 ml blood loss[a]	5.6 (1/18)
Postoperative complications	
Immediate postoperative pain	61.1 (11/18)
Haematoma	11.1 (2/18)
Post-void residual >150 ml	11.1 (2/18)

[a]700 ml blood loss.

TABLE 3 Reported pain symptoms after SSLF

	Frequency % (no./total no.), *n* = 21
Pain with or without numbness and/or dysaesthesia	
Right buttock	55.6 (10/18)
External genitalia	16.7 (3/18)
Posterior thigh	16.7 (3/18)
Back	16.7 (3/18)
Lower abdomen	16.7 (3/18)
Coccyx	11.1 (2/18)
Entire pelvic floor	11.1 (2/18)
Perineum	5.6 (1/18)
Groin	5.6 (1/18)
Lower extremity	5.6 (1/18)
Clinical examination	
Pelvic floor hypertonia	45.5 (5/11)
Pain when palpating the SSL	100.0 (18/18)
Sexual activity and dyspareunia	
Sexually active	66.6 (10/15)
Dyspareunia	90.0 (9/10)

3.3 | Reduction of pain after SSL suture removal

After a median follow-up visit of 48 days (IQR 43–55 days) after surgery, 57% (12/21) of the cohort was pain free, 38% (8/21) reported pain reduction and 5% (1/21) reported persistent pain after SSL suture removal (see also Figure 1). No correlation was found between the duration of the SSL sutures being in situ and the reduction of pain.

3.4 | Recurrence of pelvic organ prolapse

At a median follow-up visit of 50 days (IQR 44–57 days) after SSL suture removal, six women (29%) had an anatomical POP with a pelvic organ prolapse quantification (POP-Q) stage of ≥2.[18] Four of those women (19%) had symptomatic POP for which additional POP surgery was indicated. It should be noted that in two women the POP was already identified before the SSL sutures were removed. In the remaining two women (10%), one had a prolapse of the apical compartment and one had a prolapse of the anterior compartment following suture removal, where the SSL sutures have been in situ for 86 and 8 days, respectively.

4 | DISCUSSION

4.1 | Main findings

Our study shows that SSL suture removal in women with severe and/or persistent pain following SSLF – performed in a specialised pelvic floor centre – is safe and efficacious. In 95% of cases SSL suture removal is effective to obtain complete or partial pain relief. The SSL sutures were surgically removed after a median time of 414 days, and in 86% of cases complete suture removal was achieved. During suture removal one adverse event occurred, with excessive blood loss (520 ml) but without the need for blood transfusion. At 6–8 weeks after surgery, 10% of women had renewed symptomatic POP following suture removal, for which additional POP surgery was indicated.

4.2 | Strengths and limitations

This is the largest cohort study on the outcomes of the surgical management of pudendal nerve entrapment after SSLF. Although only 21 women are included, this cohort should be considered as a large cohort because of the

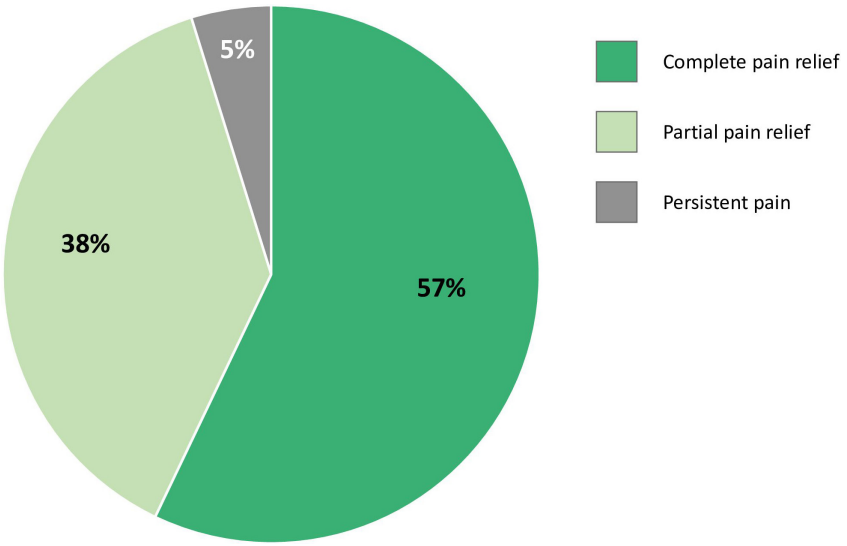

FIGURE 1 Effect of SSL suture removal on pain. In the group with complete pain relief, 14% of the women (3/21) had incomplete suture removal and 10% (2/21) had complete suture removal after two surgical attempts, resulting in complete pain relief. In the group with partial pain relief, 5% of the women (1/21) had complete suture removal after two surgical attempts, resulting in some pain relief but not obtaining complete pain relief

rarity of this complication. In addition, the evidence from this study provides a much stronger base for its current clinical practice, as previous research was based on case reports and small case series only. Finally, the data from this cohort were systematically recorded in an electronic case report form (Castor EDC), including range checks to improve data quality.

We are aware that our research has its limitations. The most important limitation is the retrospective design of this study. For this reason, there was no standardised preoperative and postoperative assessment for objective and subjective outcome measures. Also, not all the outcome measures of interest could be evaluated, such as the impact on quality of life and sexual health. Additionally, sometimes the data were incompletely or poorly reported, resulting in missing data, with a subsequent risk of bias in the estimation of parameters. To elaborate, a number of women from this cohort received conservative treatment measures before or after SSL suture removal to attempt pain reduction, such as pelvic floor physiotherapy, anaesthetic pudendal nerve block, Botox injection at the SSL, dry needling, anticonvulsants and consultation with a sexologist. Unfortunately, as the effect of these conservative treatment measures lacked detailed reporting, it was not possible to examine the additional value of those measures. This also applied to the identification of potential risk factors that might be predictive or associated with pain, including preoperative dyspareunia, pelvic floor hypertonia or pain when palpating the SSL, as well as the surgical approach and suture placement technique used for initial suture placement. For example, direct visualisation of the ligament has been reported to better protect nerve structures from injury.[16,19] On the other hand, this approach requires more dissection, which may result in additional morbidity.[16] But as stated by David-Montefiore et al., even though the device-assisted digital approach might seem less aggressive, because of its small dissection surface, it is still a blind technique with a subsequent risk for entrapment.[19] Unfortunately, as a result of the retrospective nature of this study, with missing data, we were not able to identify such risk factors.

Finally, the results on the recurrence risk of POP following suture removal are limited as the follow up was only at 6–8 weeks after surgery and lacks any long-term follow up.

4.3 | Interpretation

The true efficacy of SSL suture removal on the reduction of pain symptoms is unknown. Current studies on its efficacy are limited by the small number of subjects (n = 1–4) and poor outcome reporting.[5,10,15–17,20] Nevertheless, those studies do report partial or complete pain relief after suture removal, which is consistent with our findings. In our study, 57% of women had complete pain relief and 38% of women had partial pain relief.

To the best of our knowledge, there are no studies that reported on the duration of the SSL sutures being in situ and its effect on pain reduction after removal. In addition, information on the recurrence rates for POP after suture removal is lacking.

However, our findings demonstrate that the duration of the sutures being in place does not have any effect on the degree of pain reduction. This study even showed that complete pain relief can be obtained when sutures are removed after 5 years.

Furthermore, it could be hypothesised that when sutures are removed after a longer period of time the uterine/vaginal vault support is not compromised because of

TABLE 4 Intraoperative characteristics of SSL suture removal

Characteristic	Frequency % (no./total no.), $n = 21$
Total SSL suture removal	85.7 (18/21)
Complete SSL suture removal at first attempt	71.4 (15/21)
Complete SSL suture removal at second attempt	100 (3/3)
Simultaneous placement of new SSL sutures	0.0 (0/21)
Simultaneous procedures	42.9 (9/12)
Manchester Fothergill	9.5 (2/21)
Anterior colporrhaphy	9.5 (2/21)
Perineorrhaphy	14.3 (3/21)
Lateral vaginal wall repair	4.8 (1/21)
Portio amputation[a]	4.8 (1/21)
Partial removal of midurethral sling	4.8 (1/21)
Surgery time, min (median, range)	67.5 (25–110)
Blood loss, ml (median, range)	50 (10–520)
Antibiotic prophylaxis	100 (21/21)
Perioperative complications[b]	
>500 ml blood loss[c]	4.8 (1/21)
Postoperative complications[b]	
Haematoma	14.3 (3/21)
Urinary tract infection	9.5 (2/21)
Post-void residual of >150 ml	4.8 (1/21)
Bacterial vaginosis	4.8 (1/21)
Fever	4.8 (1/21)
Vaginal wall adhesion	4.8 (1/21)

[a]Portio amputation indication: cervical fibroid with adjacent dyspareunia.
[b]There were no perioperative and postoperative complications in the three repeated SSL suture removal procedures.
[c]520 ml blood loss.

sufficient scar tissue fixing the cervix/vaginal cuff. This factor may be responsible for our results, where 10% of women had renewed symptomatic POP, requiring a surgical procedure, which occurred in women who had their sutures removed relatively soon after the initial placement. However, this should be interpreted with the utmost caution, because this did not apply to all women with a relatively short period of suspension sutures being in situ, as well as the fact that the postoperative follow-up period was short, which might suggest a higher recurrence rate than reported by this study. Moreover, one could discuss performing (prophylactic) concomitant procedures at the time of suture removal to prevent the recurrence of prolapse. Based on this study, we cannot provide a recommendation with respect to this topic. However, in our institutions we do not like to perform a simultaneous apical repair at the time of suture removal because we primarily focus on the effect of suture removal for pain symptoms. When a new apical suspension procedure is performed at the time of suture removal, it obscures how efficacious the suture removal has been. On the other hand, the simultaneous repair of the anterior and/or posterior compartment is acceptable as it is unlikely to affect postoperative pain.

In clinical practice, both absorbable (polydioxanone suture or Vicryl) and non-absorbable (prolene or braided polyester sutures) sutures are used for SSLF. Similar to our study, non-absorbable suture material is often used to achieve long-lasting repair and lower recurrence rates. But one question that needs to be raised is whether absorbable suture material does lead to lower postoperative pain, given the temporary entrapment until the suture is resolved. Although lower pain-related complications seem likely, Dangal et al. shows that absorbable sutures can also result in persistent buttock pain at 6 months after surgery.[21] However, the data are limited and the use of different suture materials for SSLF requires more evaluation. Nevertheless, in the case of severe direct postoperative pain, we would recommend removing the SSL sutures, regardless the type of suture material used.

To minimise nerve entrapment and postoperative pain as far as possible – without reducing the overall success rate of the SSLF procedure – it is recommended to place the suture at the mid segment of the ligament, 2–3 cm from the ischial spine, and not too deep into the SSL.[22–24] As stated by Florian-Rodriguez et al., suture placement too close to the sacrum may lead to S4 nerve entrapment and placement too close to the ischial spine may lead to pudendal nerve and/or vessel injury. On the contrary, anatomical studies show that despite meticulous suture placement, nerve injury could not be avoided. Including the course of the (proximal) pudendal nerve, running dorsal to the SSL, where high variability in the branching patterns has been demonstrated.[10,11,14,25,26]

For instance, Mahakkanukrauh et al. noted that pudendal nerve branching already takes place at level of the SSL in 44% of cases, instead of beyond the SSL level. In 11% of cases, a pudendal nerve branch was even found to run through the SSL. Equally important, branches of S3 and S5 also seem to have a close anatomical relationship with the SSL.[11,12,24,26] As an example, Roshanravan et al. wrote: 'In this anatomic study of 21 female cadavers, innervation to the coccygeus and levator ani muscles arose individually or in combination from S3 to S5. In the majority of dissections (89%), branches to the coccygeus and/or levator ani muscles coursed over the midportion of the SSL, just in the area where SSLF sutures are placed'. Those anatomical studies provide evidence that the entrapment of sacrospinous nerves cannot be avoided entirely by any specific technique of suture placement. Therefore, nerve entrapment remains an unpredictable complication of the SSLF procedure. Considering the fact that different sacrospinous nerves could be involved, it explains the widespread complaints reported by the women from both this study and from literature concerning this complication. Nevertheless, our study demonstrates that suture

removal is an effective treatment regardless of the particular nerve involved.

5 | CONCLUSION

This is the largest cohort study that analysed the outcome of the surgical management of nerve entrapment after SSLF. Nerve entrapment resulting in severe and/or persistent pain after SSLF is a rare but relevant complication, causing significant impairment of quality of life. Surgical removal of the SSL sutures is a safe and efficacious treatment for pain relief, even long after the initial placement. The risk of symptomatic POP following the removal of the SSL sutures can be discussed during counselling. Given the rarity of the procedure and the technical challenges, we recommend that the removal of the SSL sutures is performed by experienced hands.

CONFLICT OF INTERESTS

None declared. Completed disclosure of interests form available to view online as supporting information.

AUTHOR CONTRIBUTIONS

All authors were responsible for the conception and design of this study. EV, KvD, CN and CK were responsible for data collection and data management. KvD and EV were responsible for conducting the analyses. All authors were involved in the interpretation of the data. EV drafted the initial manuscript, which was reviewed and critically revised by all authors. All authors approved the final version for publication.

ETHICAL APPROVAL

The Medical Ethics Review Committee AMC confirmed that the Medical Research Involving Human Subjects Act (WMO) did not apply to this study and therefore official approval by the committee was not required (ref. no. W21_497 # 21.551).

DATA AVAILABILITY STATEMENT

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

ORCID

Eva V. Vodegel https://orcid.org/0000-0001-6897-9495

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SUPPORTING INFORMATION

Additional supporting information may be found in the online version of the article at the publisher's website.

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