



A teaching module on a case of chronic pelvic pain

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Keywords: Chronic pelvic pain; pain; Pelvic floor; vagina

INTRODUCTION

Colleagues, fellows, students

Welcome to another Pelviperineology teaching/discussion module! Pelviperineology seeks to encourage a holistic anatomical approach to dysfunctions of the pelvic floor. It is not possible to slice and dice the pelvic floor into ever smaller subsections, drawing lines between pain, psychology, urology, gynecology, coloproctology, as this case shows. I take this opportunity to thank our founding editor, Professor Dodi, Professor of Coloproctology, University of Padova (est. 1222) who has steered the journal in the most dedicated way, from its founding in 2007 to the end of 2020. At the same time, I warmly welcome our new Editor-in-Chief, Professor Jacob Bornstein, Past President of the International Society of Vulvovaginal Disease and a leader of vulvodynia research, from a somewhat more modern university, Bar-Ilan, Israel.

This Case for discussion on first look is a case of chronic pelvic pain. On closer inspection, it fits multiple diagnostic criteria, chronic pelvic pain of unknown origin, vulvodynia, overactive bladder (OAB), interstitial cystitis and the posterior fornix syndrome which envelops the previously stated conditions. There

are many fascinating aspects to this case which are discussed by our experts, not the least being her marginalization by the medical profession as a “psychological” case, which is the cause of everything. I have directly quoted parts of the history below as discussion points and invited the colleague who provided the case to make some brief comments on each point at the end. I hope our readers will address the comments by our experts in correspondence.

CASE REPORT

Mrs P was 49 years old. She had chronic pelvic pain and she requested referral to the clinic because she had heard that we were achieving good results in patients with pelvic pain. Her General Practitioner, an empathetic and caring man, rang the doctor before she arrived and asked that we “handle her very carefully” as she was severely disturbed psychologically, that this was the reason for her pain and there was nothing anyone could do for her. The first impression we had of this lady did indeed fit the description of her GP. Her face was contorted, she spoke rapidly and with obvious anxiety and depression. She had visited many specialists over the years for her pain. She had undergone several diagnostic laparoscopies were all normal. She

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Received: 16 August 2021 **Accepted:** 30 July 2021

had undergone a hysterectomy as a last resort and had attended a pain clinic.

None of these treatments had helped her pain. The consensus from other specialists as reported to the GP was that her problem was psychological. Her replies to the Integral Theory System Questionnaire (ITSQ) were as follows: she woke up to eight times per night to empty her bladder (nocturia), wore pads continually, as she wet up to 6 times per day (urge incontinence), frequency 20 times per day, had difficulties emptying her bladder and faecal incontinence. We asked her if she had told her GP about her bladder and bowel problems. She said she had only consulted him about the burning pain around her vagina and anus, but on specific questioning she had other sites of pelvic pain, “dragging” back pain and pain low in her right lower abdomen. She said that her vagina was so tender that she couldn’t have sexual intercourse and sometimes had problems sitting. Examination revealed a moderate prolapse of the apex of her vagina. The vaginal introitus was hypersensitive-she recoiled when gently tested with a cotton swab, the classical test for “vulvodynia” (pain at the entrance of the vagina).

We did not claim that we could cure this lady’s pain, as there are many other causes for pelvic pain. Nevertheless, it was explained that her vaginal prolapse needed to be fixed and that there was a strong possibility that some of her symptoms would also improve with a sling inserted into the back part of her vagina, a fairly minor day-care procedure.

Management

The patient had a posterior intravaginal slingoplasty (IVS) operation. This operation restored a prolapsed uterus or apex by a posterior sling which essentially used a posterior sling to reinforce weak uterosacral ligaments.

Post-operative visit at 6th weeks

The first thing we noticed at the six-week post-operative visit was the absence of tension in her face. She was smiling and calm. Her pelvic pain was gone, her entry dyspareunia gone. The frequency was now five times per day. She said her urge and faecal incontinence were now cured. Her nocturia had reduced to two per night and her bladder emptying was “60% improved”.

Comment of Bernhard Liedl:

Bernhard Liedl M.D., Chefarzt, Urologische Klinik Planegg, Zentrum für Rekonstruktive Urogenitalchirurgie, Planegg, Germany

I strongly suppose that all reported symptoms - pain, nocturia, urgency incontinence, abnormal emptying of the bladder and fecal incontinence - are induced by the detected moderate apical pelvic organ prolapse (POP).

From my experience it is typical, that such patients have a long history with many examinations - even by urologists, gynecologists, and psychologists - until the diagnosis of “posterior fornix syndrome” induced by POP could be established.

Despite several publications on this issue¹⁻⁴ many specialists obviously are not aware of this association. Important to know that even first degree and second-degree pelvic organ prolapse can be responsible for this symptom complex.³⁻⁷ These stages only can be detected by careful vaginal examination with full Valsalva. So, without performing the Valsalva maneuver, these stages can be routinely undetected.

It is also typical that such patients only report their main symptom – pain in this case. It is important that with all these patients a systematic assessment of symptoms of stress urinary incontinence, overactive bladder, underactive bladder, anorectal dysfunctions and pain has to be done, as these symptoms often coexist.⁴⁻⁸ After the vaginal examination it has to be proven if POP is responsible for these symptoms. Simulated operations as described by Liedl,⁹ are very helpful and important in diagnosis of stress urinary incontinence, and urgency symptoms. Furthermore, mechanical support of the posterior fornix can relieve urgency and suburethral tenderness.¹⁰ With a local anaesthetic into the uterosacral ligament – the Bornstein test¹¹ – the origin of pain at uterosacral ligament can be tested.

The association of POP and pain at different sites (lower abdomen, lower back, vulvodynia, vagina, bladder) is well known.^{4,12-15} The pathophysiologic pathways of POP and development of symptoms also are well known.^{2,3,9,15} The link for both is ligamentous (connective tissue) laxities.^{4,9}

It has been shown that POP-associated pain symptoms can be improved or even cured in high percentages by vaginal ligamentous POP-repair.^{3,12,14,15} It has been shown that co-existing POP-associated nocturia,⁶ urgency, urgency incontinence, daytime urinary frequency,⁵ abnormal bladder emptying⁷ and fecal incontinence^{3,6,7} also surgically, they can be cured in high percentages.

This woman would need simulated operations to confirm ligamentous causation and a micturition diary to rule out polyuria. Probably I would recommend her to undergo a vaginal ligamentous POP-repair with the option of high cure rates for her chronic pain syndrome and her coexisting symptoms.

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Comment of Professor Klaus Goeschen:

Professor Dr Med Klaus Goeschen, MD PhD, formerly Professor of Gynecology, University of Hannover, Germany

Dr. Liedl has excellently analysed the patient's situation and precisely set the necessary course for diagnostics and treatment. I completely agree with Dr. Liedl on all points and therefore refrain from commenting on my own. I elaborate only on a few points made by Dr Liedl: Symptoms occur in predictable groupings, almost always with one symptom predominating. In this case, it was vulval pain, but she had other sites of pain and several bladder symptoms. See "iceberg figure".¹ I agree that major pain and other symptoms can occur with minimal prolapse and care must be taken to confirm the presence of some prolapse on examination. One useful technique I found in patients with no apparent prolapse is to lift up the anterior vagina wall with a speculum blade and ask the patient to strain down. Appearance of an enterocele bulge is diagnostic of weakened USLs. "Simulated operations" such as the speculum test are very useful, but not invariable. I have seen women with negative speculum tests whose urge and pain nevertheless being cured by repair of their prolapse.

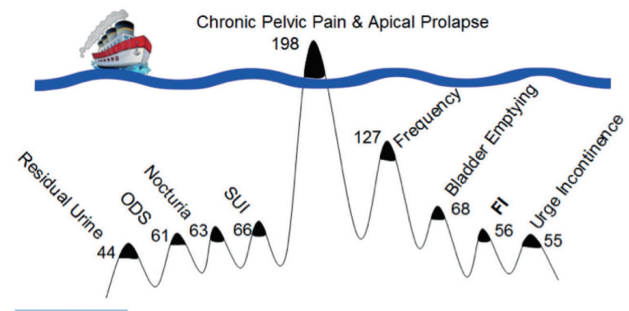


Figure 1. Pescatori Iceberg applied to Chronic Pelvic Pain (CPP). The symptom prevalence is graphically indicated in the iceberg diagram, latent symptoms below the waterline. All symptoms derived from the Integral Theory Symptom Questionnaire (ITSQ). Numbers indicate combined total of the hysterectomy and nonhysterectomy cohorts

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Comment of Dr. Ion-Andrei Mueller-Funogea:

Dr. Ion-Andrei Mueller-Funogea MD, PhD, Gynecologist & Urogynecologist, Aachen, Germany

Firstly, I discuss the Posterior Fornix Syndrome (PFS) and after that, its connection to Interstitial Cystitis and Hunner's ulcer.

My background is relevant, as I obtained my PhD by a thesis on the posterior fornix syndrome (PFS) in 2015. It was based on an intensive study on 453 PFS patients over 8 years, with specific attention to diagnostic criteria, diagnosis & surgical treatment.

My initial comment is that the case is a typical case of PFS, very similar to what I met many, many times in those 453 cases, all with USL laxity and/or after hysterectomy!

The PFS concepts crosses many disciplines, although acceptance is difficult (see algorithm, Figure 2):

1. The interdisciplinarity of PFS: proctologists sometimes cannot accept that obstructed defecation is caused by weak USLs and urologists sometimes find it hard to admit that nocturia is explained by posterior ligament pathology of the pelvic floor.
2. We must know and accept the non-linearity of the pelvic floor, that means minor anatomical disorders may cause massive functional disturbances (and vice-versa).
3. We should know and accept the Integral Theory (role of ligaments and muscles in normal and abnormal function of the pelvic floor, and later the integral system, which is an anatomical system of diagnosis and surgery).

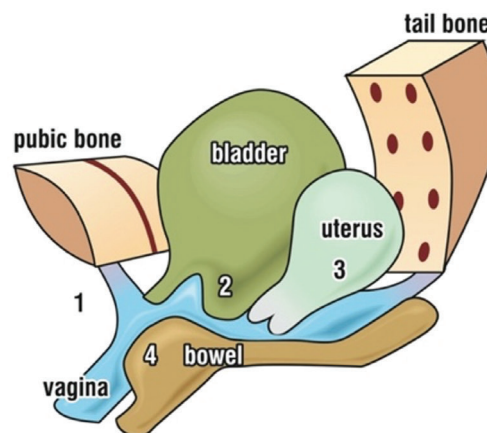
We still have a long way to go in understanding and accepting this paradigm change, but it moves forward.

I also comment on interstitial cystitis (IC) and Hunner’s ulcer. I am not greatly experienced in interstitial cystitis (IC), but I have seen hundreds of women who had chronic pelvic pain and several bladder symptoms as in the rectangle of the diagnostic algorithm, who were very successfully treated by me as PFS. I understand that this description fits the International Continence Society (ICS) definition for IC.

Finally, I encourage you all to read the ground-breaking paper by Liedl et al.¹ of 1671 cases.

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Front ligaments (PUL)	Middle ligaments (ATFP & CL)	Back ligaments (USL & PB)
	2 cystocele	4 rectocele
		3 uterine/apical prolapse
1 stress incontinence		
	abnormal emptying	abnormal emptying
frequency and urgency	frequency and urgency	frequency and urgency
		nocturia
faecal incontinence		faecal incontinence obstructed defecation
		pelvic pain
		leathered vagina

Figure 2. Pictorial algorithm

1. Symptoms indicate which ligaments are damaged. The numbers indicate the sites of damage. The height of the bar indicates probability of association of a symptom with a particular zone. The connective tissue structures causing prolapse and pelvic symptoms fall naturally into three zones.
2. **Anterior zone:** external meatus to bladder neck pubourethral ligament (PUL);
3. **Middle zone:** bladder neck to anterior cervical ring. cardinal ligament (CL); arcus tendineus fascia pélvis (ATFP).
4. **Posterior zone posterior cervical ring to perineal body (PB):** USL uterosacral ligaments; PB: perineal body. The rectangle indicates the symptoms associated with USL laxity and the posterior fornix syndrome.
5. Chronic pelvic pain and nocturia are uniquely caused by uterosacral (USL) ligament laxity.

Comment by Professor Petre Bratila:

Professor Petre Bratila, Medical Director, Euroclinic Hospital, Bucharest Romania. Supervisor of surgical activities in General surgery and Ob-Gyn.

The case presented above is a “classical” description of the “Posterior fornix syndrome” caused by apical support laxity, accompanied by neuromuscular dysfunction of pubococcygeus muscle. We must remember that pubococcygeus muscle receive innervation from the superior rectal nerve branch of pudendal nerve in 30% of cases.

In this case I propose that apical suspension by a posterior sling inserted to the uterosacral ligaments will be associated postoperatively by a high frequency stimulation of levator ani.

Comment by Dr Shuqing Ding:

Dr Shuqing Ding, LAc. CMD. Ph.D. Pelvic floor specialist, Colorectal surgeon (China). Dr Ding Shuqing is 10th generation Chinese Medicine trained and also, in Western Medicine with a specialist degree in colorectal surgery.

1. Review from the perspective of colorectal surgeons for diagnosis and evaluation:

1. For the perianal pain: check the skin around the anus and vagina for any skin damage or scars, then test the anal skin reflex, using a needle to pitch the anus skin, whether the external anal sphincter has contraction after stimulating; If there is, it means the spinal reflex is normal and there is no nerve injury.
2. Digital examination in rectum and vagina for testing muscle tone and contraction. To see if it is a spasm pain from pelvic floor muscles tightness, or if it has some trigger points, and testing the pain aggravation when pulling puborectalis muscles; then do Valsalva maneuver to observe the puborectalis muscle coordination to confirm if it is pelvic floor dyssynergia. If the tension of the anus or vagina is normal or low, and the pelvic floor muscle coordination is normal, then the pelvic floor laxity from muscle or ligaments should be considered.
3. According to the Integral theory. Determine if it is “posterior fornix syndrome”. Examine the patient in a semi-recumbent position; support the vaginal fornix with a speculum, and asked whether the patient’s pain or urgency improved.
4. Communicate with patients, observe the patient’s cognition and the severity of anxiety and depression and quality of life, and clarify the expectations of patients.

2. From a Chinese Medicine and acupuncturist perspective (non-surgical). Non-surgical treatments include acupuncture and biofeedback.

The role of acupuncture:

1. Reduce the overactive excitement of pain in the cerebral cortex and generate the natural enkephalin to promote the good feedback effect: this is achieved by stimulating acupoints of the head and auricular.
2. Regulating sacral nerve reflex and interacting with the brain to form benign feedback effect: it is achieved by stimulating S2-4 sacral nerve. A needle of 0.35 x (75 mm–125 mm) is passed through the sacral foramen at the back, which needs to enter the anterior sacral foramen from the posterior sacral foramen, and the patient had a heavy feeling around the rectum and vaginal area and deep in the pelvis.
3. Use of an electrical stimulator clamped on the needle, and continuous electrical stimulation was performed for 20 minutes at 2 Hz, two to three times a week. The curative effect can be seen after six times in general.

Role of pelvic floor biofeedback

1. For pelvic floor dyssynergia This method is the first choice, and the training plan can be made after the sEMG evaluation.
2. For the pelvic floor laxity The long-term effect of biofeedback with acupuncture and electrical stimulation is satisfactory.

Other non-surgical Chinese Medicine options include:

1. Pelvic floor muscles massage and stretch through the vagina.
2. Traditional Chinese medicine sitz bath: can promote blood circulation and relieve pain, increase local blood circulation, reduce pain substances, and improve symptoms. Generally, some patients have benefits.
3. According to the patients’ pain characteristics and personal body constitutions, herb formula may be taken after syndrome differentiation.

Surgical option: From Integral theory, it is related to USL, Rectovaginal fascia.

I prefer transvaginal native ligament repair surgery, but only after having exhausted all non-surgical options.

Comment by Patricia M Skilling:

Patricia M Skilling, former Director Pelvic Floor Rehabilitation, Kvinno Centre Perth Australia.

My question is, are ‘psychiatric’ findings in patients with Chronic Pelvic Pain, primary or secondary? I comment specifically on the “psychological cause” of the patient’s condition.

The 2005 Cochrane Review summarizes Chronic Pelvic Pain as follows “*Chronic pelvic pain (CPP) is common in women in the*

reproductive and older age groups and causes disability and distress. Often investigation by laparoscopy reveals no obvious cause for the pain. As the pathophysiology of chronic pelvic pain is not well understood its treatment is often unsatisfactory and limited to symptom relief. Currently the main approaches to treatment include counselling or psychotherapy, attempts to provide reassurance by using laparoscopy to exclude serious pathology, progestogen therapy such as medroxyprogesterone acetate, and surgery to interrupt nerve pathways”

During the time I was Medical Director for pelvic floor rehabilitation (PFR) at the Kvinno Centre Perth Western Australia, we assessed some hundreds of women whose primary complaint was chronic pelvic pain. Several women had similar stories to Mrs P. Our view was that these “psychiatric” symptoms were real, but secondary to the pain. The one constant in the story of such women as Mrs P who had had children, was that most of them never had pain or bladder problems prior to having children. It followed logically from this, that something happened in childbirth to cause these problems. According to the Integral System.¹

In an Opinion piece in 2017,² we wrote “traditionally hysterectomy has been recommended as a cure for chronic pelvic pain by a significant body of specialist opinion”, and “the present trend to psychiatric evaluation and treatment appears to have arisen as a consequence of previous studies which showed a high correlation between CPP and psychiatric disturbances”.

Mrs P is an obvious case of Posterior Fornix Syndrome, predictable groupings of chronic pelvic pain, urge, nocturia, abnormal emptying caused by uterosacral ligament (USLs) weakness.

Our data³⁻⁵ showed that we achieved 50% improvement in pain and bladder symptoms with our squatting-based PFR protocols in a majority of premenopausal women. We attributed this to strengthening their reflex pelvic muscles and the ligaments they contract against.

Mrs P had been hysterectomized for her pain. Our view at the Kvinno Centre was that this was an unnecessary operation with not insignificant surgical complications. It is anatomically impossible for the uterus per se to cause “chronic pelvic pain of unknown origin”, if there is “no obvious cause”, as in Mrs P’s case. Furthermore, it is now being recognized that hysterectomy is associated with increased incidence of prolapse and incontinence developing after the menopause. Mrs P had gone to numerous psychiatrists and pain clinics to no avail. **Patients tell me “If you have never experienced severe pelvic pain, you would never know how disabling it is. Of course, you will have anxiety and depression”. Would squatting pelvic exercises help Mrs P? It seems her psychological state was so extreme, only a**

“quick fix” would do. Our surgeons at the Kvinno Centre were achieving up to 80% cure rates with uterosacral ligament slings in women who had positive “simulated operations”: relief of CPP and urge by the speculum test, as in Figure 3. We would have advised her that as she was close to menopause, the squatting PFR was probably worth doing in the same sense as going to the gym, but our results for symptom were very poor after the menopause. Given the severity of her symptoms, we would have advised a posterior sling.

It is unfortunate that many doctors, including this lady’s General Practitioner, were not aware that this type of pelvic pain is associated with loose ligaments. Because the pain is often constant and unremitting, the patient is invariably anxious and depressed. If an obvious cause cannot be found, the doctor seeks another cause, usually “psychological”. Our experience is that when such patients are cured by surgery, the cure is literally instant - they awake from the anesthetic with no pain. Hopefully this teaching module will help educate all health professionals that anxiety and depression from chronic pelvic pain in women is rarely primary. It is secondary, caused by ligament damage, after childbirth or menopause, because of collagen breakdown in ligaments after the menopause.

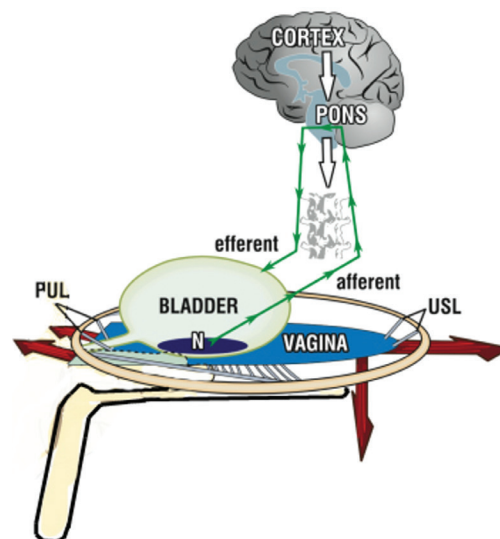


Figure 3. Diminution of urge by “simulated operations”

“Simulated” operations work by mechanically supporting loose or damaged ligaments. The speculum is very gently inserted into the apex of the vagina. This stretches the vagina and supports the uterosacral ligaments (USL). It relieves urge by restoring the strength of the opposite pelvic muscles which stretch the vagina to support the bladder base stretch receptors “N” from below; it relieves chronic pelvic pain by supporting the USLs which mechanically support the Frankenhauser and sacral plexuses.

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Comment from Professor Akin Sivaslioglu:

Akin Sivaslioglu, Immediate Past President of ISPP

Symptom Cure for PFS is specific to uterosacral ligament repair, not SCP.

With relevance to the case report, it is clearly a case of posterior fornix syndrome (PFS): chronic pelvic pain, OAB, nocturia, evacuation difficulties, which I would normally manage with a posterior IVSsling. I have published several papers demonstrating cure of PFS symptoms with a posterior IVS sling, in turn, based on the concepts of the Integral Theory. The question I always had was, if the anatomical basis of cure of PFS (chronic pelvic pain of unknown origin, urge, frequency nocturia abnormal emptying and or urinary retention) is weak apical support, why would any operation which repairs uterine or apical prolapse not also cure symptoms of PFS?

We performed a comparative study,¹ comparing abdominal sacrocolpopexy (ASCP) with posterior intravaginal slingplasty (PIVS) in terms of lower urinary tract symptoms and anatomical healing. Ninety-two patients underwent PIVS and 98 patients underwent ASCP. Anatomically, ASCP and PIVS appeared to have same efficiency for the treatment of vault prolapse in the long term. PIVS improved lower urinary tract symptoms more efficiently when compared with ASCP. Both operations have good anatomical healing for rectocele and cystocele. However, PIVS also cures enterocele as an additional advantage over ASCP, evident from examination of the figure: ASCP does not repair USL. It creates an artificial strut to suspend the uterus or apex.

CONCLUSION

We have shown¹ that symptom cure with PFS symptoms is not a matter of restoring the apex. It is specific to repairing USL. "Repair the structure and you will restore the function"- **Integral Theory.**

A mesh attachment from apex to the sacral promontory does not repair the USL (structure) in the same precise way as a posterior sling. Basic science and computerized finite element studies at the School of Mechanical and Mathematical Engineering at the University of Western Australia,^{2,5} have determined that both urethral closure and opening (micturition) are exponentially determined and governed by complex flow mechanics, inversely proportional to 4th power of the radius (Poiseuille's law). This means that the urethral and anorectal closure and opening pelvic muscles rely on a precisely tensioned USL to contract against. Reference to Figure 4 shows such accuracy cannot easily be achieved by an ASCP mesh, as compared to USL repair (red wavy lines). Furthermore, it is known that an overtight ASCP can sometimes cause severe evacuation problems for both bladder (obstructed micturition) and anorectum (obstructive defecation).⁵ Such complications have never been reported from a posterior sling which accurately repairs USL.

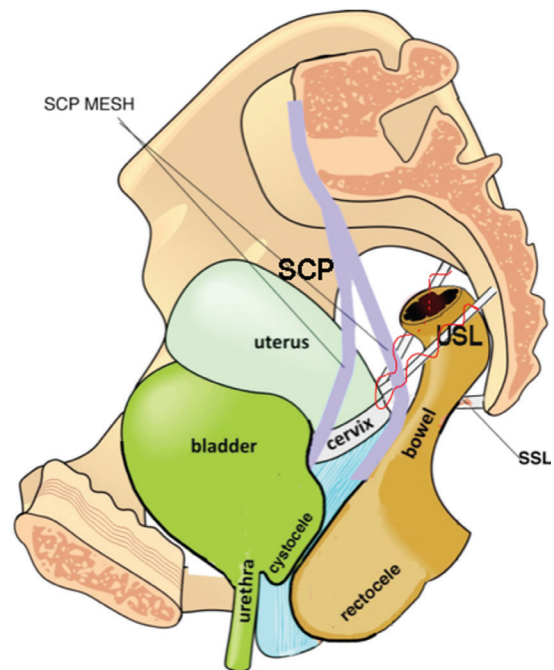


Figure 4. Sacrocolpopexy (SCP) attaches a large mesh laparoscopically from apex to the sacral promontory (lilac colour). Whereas this will hold up a prolapsed uterus or apex, it is not anatomical. The uterus is suspended by uterosacral ligaments (USL) which attach the posterior cervical ring to the sacrum S2-4. Repair of USLs either by a posterior sling (indicated by red wavy lines) or even by plication, mimics the correct anatomy of USL. SCP, clearly does not mimic the natural anatomy of USL. It does effectively resuspend the apex, however.

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Comments of Dr Traian Enache:

Dr Traian Enache, Spitalul Clinic de Obstetrica si Ginecologie "Prof Dr. Panait Sirbu" Bucharest Romania

Apparently unexplained chronic pelvic pain in women may cause severe psycho-social problems. Social impact might lead to social reclusion, depression, or professional decline. A recent article published in 2019 in *Journal of Pain Research* underlined that the prevalence of anxiety, depression and mixed anxiety and depressive disorder was higher in women with CPP compared to the pain-free controls¹. Among the wide variety of pelvic pain, vulvodynia is an underestimated and an almost ignored aspect. Most physicians pass over it if there is no vulvar lesion. However, Schonfeld et al.² published in 2021 a study where they find an association between vulvodynia and vaginal apical defect. They stated that applying mechanical support to the posterior fornix temporarily alleviates provoked vulvar pain in some women.

Evaluation of female pelvic pain is often a difficult task, but we must always search some other symptoms and signs, such as pelvic floor myofascial pain was common in patients seeking evaluation for pelvic floor disorder symptoms.³

One in three women with a diagnosis of pelvic organ prolapse (POP) were found to have pelvic floor myofascial pain. On average, when pain was present, women had a lower stage of prolapse and were more severely bothered by their pelvic floor symptoms.⁴ Therefore, we must pay attention to even a slight POP degree.

The associated symptomatology of that patient (nocturia, urge incontinence, faecal incontinence) suggests even stronger a pelvic floor disorder as the trigger factor of her condition. As many authors suggest, we must a cure a specific defect (objectively diagnosed) in order to release specific symptoms.⁵ In this case I think the main problem is the apical vaginal defect.

Most probably, all the symptoms she has, were associated with it. Chronic pelvic pain, bladder and bowel incontinence occur in predictable symptom groupings, which are associated with apical prolapse. USL repair, whether native tissue or, preferably, using a posterior sling, has the potential to improve clinical practice, quality of life for women and open new research directions.⁶

So, I propose a surgical cure for apical vaginal defect. In these cases, with hysterectomy anteriorly performed, I suggest a posterior polypropylene patch, attached to the vaginal vault, and suspend it bilaterally to the sacrospinous ligaments.

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Contributions

Surgical and Medical Practices: M.N., A.S., Concept: M.N., A.S., Design: M.N., A.S., Data Collection and/or Processing: M.N., A.S., Analysis and/or Interpretation: M.N., A.S., Literature Search: M.N., A.S., Writing: M.N., A.S.

Ethics

Peer-review: Internally peer-reviewed.

DISCLOSURES

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.