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A clinical and organizational model entirely devoted to pelvi-perineal health: a new model of care

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ABSTRACT

Pelvi-perineal disorders represent a wide spectrum of diseases involving different disciplines. A patient-centered care and a coordinated approach, extended from functional to oncological diseases, may be achieved through the planning of a "pelvi-perineal network". The network is a new clinical and organizational model, entirely devoted to pelvi-perineal health through a multidisciplinary multiprofessional approach, coordinated and synergistic. The network is a new mental approach, without walls, to treat different diseases in the same patient and not a concrete management unit for single disease. In the network the team care approach is critical to get a more in-depth understanding of the symptoms cascade and to define a coordinated integrated management.

Keywords: Care model; chronic pelvic pain; network; pelviperineology; urogenital tract; vulvodynia

Introduction

Pelvi-perineal disorders represent a wide spectrum of diseases involving different disciplines. Until recent years, urologists, gynecologists, proctologists, and colorectal surgeons had worked independently in their pelvic compartments neglecting the other specialties.

The presentation in 1990 of a ligament-based theory as a universal theory of pelvic floor function and dysfunction - the Integral Theory (IT), represents the first attempt to overcome the boundaries between the traditional areas of specialization that become barriers to optimal best practice management of the patients having a pelviperineal disease.[1]

The implication of the Integral Theory

This theory has permitted to bring back the attention of the international scientific community on the pelvi-perineal unit: according to anatomy and physiology, the perineum represents a single embryological, anatomical and functional entity. An example of this can be found when considering the epidemiological and physiopathological data on pelvic statics disorders: connective, muscular and neurological lesions involve the three levels of the perineum - urological, gynaecological and ano-rectal - according to the location and relationships between the pelvic organs.

It is no longer achievable for any physician dealing with pelvi-perineal disorders to reach an in-depth expertise in more than one or two of the rapidly expanding subspecialist fields. In addition, communication between different specialists is frequently sub-optimal and too often, the patient has to follow a fragmented management. The quality, the location and relationships between the pelvi-perineal organs require healthcare providers speaking the same language and

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considering that a global approach to pelvi-perineal disorders is necessary.

The new patient-centered paradigm

In recent years, new models of care, even if limited to lower female genital tract, have been proposed in order to overcome the fragmented management of pelvi-perineal disorders.[2] According to these proposals, we are convinced that the unitary view of the pelvi-perineal compartment is the only way to ensure more rational and effective solutions to the patients.

A patient-centered care and a coordinated approach, extended from functional to oncological diseases, through the planning of a "pelvi-perineal network" is the way to reach a solution of the fragmented clinical optimal management. The value of this new concept is not to create a novel specialty but to sensitize and gather the different specialists dealing with pelvi-perineal disorders.

The network is a new clinical and organizational model, entirely devoted to pelvi-perineal health through a multidisciplinary approach, coordinated and synergistic. The network is a new mental approach, without walls, to treat different diseases in the same patient and not a concrete management unit for single disease.

Pelvi-perineal compartment is now to be considered as a composite group of different organ systems that interact with one another principally from a functional point of view. Consequently, a single disease may affect more than one organ system, and comorbidities may interact.

Examples of conditions covered by the new approach

A localized endometriosis of bladder, urether, recto-vaginal septum requires a multi-modal approach starting from the correct diagnosis to the combined mini-invasive surgical approach.

An anterior and posterior pelvic organ prolapse must require different approaches in terms of diagnosis and treatment, and the proctologist cannot restrict his competence to the posterior compartment; similarly, the urologist, gynaecologist and general surgeon when evaluating pelvic floor diseases, must take into account the posterior compartment.

Pelvi-perineal pain represents serious health problem for any patient and also a challenge for any physician; behind this symptom, a variety of overlapping conditions may exist: pelvidynia, vulvodynia, interstitial cystitis, somatization disorders, pelvic floor dysfunction, bowel disorders, and myofascial disorders.

An advanced rectal cancer, involving adjacent organs, is to be considered a multiorgan disease requiring a combined concomitant medical and surgical management; a multispecialistic coordinated approach avoids time-consuming repeated procedures, limiting patient's discomfort and psychological distress.

Proctologist, urologist, urogynecologist, physiatrist, sexual therapist, psychologist, enterostomal therapy nurse, rehabilitation nurse, trained midwife, and physical therapist have to interact in solving the progressive disorders occurring in such complex cases.

These few examples clearly show the complexity and the comorbidity of the pelvi-perineal disorders, encompassing pelvic organ prolapse, lower urinary tract dysfunction, anorectal dysfunction, vulvar diseases, bowel and urological cancers, infections, sexual interest and arousal disorders, pelvi-perineal pain, urinary and fecal incontinence. These disorders, alone or combined, may affect all the different episodes of a patient's life, requiring a proper continuity of care.

The team care

The team care approach is critical to get a more in-depth understanding of the symptoms cascade and to define a coordinated integrated management.

It is mandatory to better understand the interactions between the pelvi-perineal compartments, to create a common language, and to use randomized controlled studies with long follow-up to evaluate the anatomical and functional results of the different combined therapeutic approaches.

In addition, the development of social implications of pelvi-perineal associated disorders has increased the number of patients referred for these diseases and the medical consideration of this compartment.

Today, in a patient-centered health-care approach, the pelvi-perineal disorders require careful clinical assessment, appropriate investigations, and counselling before embarking on a well-defined management pathway, including behavioral and lifestyle changes, conservative treatments (pelvic rehabilitation, physical therapy, and biofeedback), pharmacotherapy, and minimally or invasive specialized surgery.

Good clinical care and governance are to be reached through a team approach. The past compartmentalization of the pelvi-perineal unit in independent sites and, consequently, the partitioning of the patients into urological, surgical, proctological, gynecological, or anorectal approach, depending on the patients' presenting symptoms, are no longer advisable.

The first step in organizing the pelvi-perineal network is to define the fields of competence, among the different specialists (nurses

and physicians) that may converge in a shared overlapping knowledge, avoiding disagreement.

The second step is defining the multidisciplinary clinical pathway for each disease to be managed, achievable together with the designation of a coordination group. This team of specialists, individually involved in pelvi-perineal diseases, represents the core of the network.

A key-role is played by the outpatient services managed by trained nurses, to permit the correct assessment of the patient and access to the network. After the hospital multidisciplinary treatment approach, the outpatient ostomy sub-network and the pelvi-perineal rehabilitation sub-network strongly interact in order to guarantee the correct care of the patients.

Particular importance is given to continued education: clinical case multidisciplinary discussion, interdisciplinary course organization about pelvi-perineal disorder management, self-education in specific topic (rehabilitation, psychosexual therapy, and minimally invasive surgery) permit to achieve a high standard and uniformity in the different multi-specialty procedures.

Strenghts and limitations

Complex multidisciplinary care is often fragmented and limited by "clinical linkage deficiency". Many articles have stressed the importance of implementing patient-centered care by building networks, but despite progresses in understanding what the networks are, how they are structured and operate, little is known about their effectiveness and sustainability in clinical practice.

A systematic review on "health professional networks" reveals that only 26 articles on 1560, resulting for title and abstract review, have a focus on the structure of health professional networks.[3]

Anecdotal experiences in "patient-centered network approach" are reported in projects on fertility care or cancer care, without systematization.[4]

Despite the lack of studies on the clinical application of this new concept of care, we are convinced that it is more efficient in respect of the traditional approach in contents, time investments, professional motivations, even if further researches are needed to demonstrate this efficiency.

This approach would seem dedicated to patients traveling numerous complex pathways only, but the characteristic and the complexity of the "patient-centered network" care are important in facilitating greater levels of quality of care and patient safety, allowing professionals from different disciplines to embrace this new theory as a common paradigm. In this way, the quality-related performance, also in patients travel a clear monodisciplinary pathway, will be increased.

The pelvi-perineal disorders management requires the involvement of intensive collaborating professionals and liable patients. The team participants experience is one of the success factors in realizing the network and maintaining the inter-professional teamwork across commonly occurring organizational divides (professionals, genders) is the real challenge. Being in an effective network, encouraging communications and interactions, promoting education with continuous learning, are the building blocks of an effective and resilient professional patient-centered network.

Pelvi perineal network methodology

The first example of this new "pelvi perineal network" organization model, starting from the project of a "lower female anourogenital network" is defined in Italy. The Azienda Sanitaria Locale di Ivrea, Ciriè e Chivasso (ASLTO4) covers a catchment area of more than half a million people and encompasses five hospitals and outpatient divisions related to pelvi-perineal management; urological, gynecological, and colorectal surgical wards and outpatient services; perineal rehabilitation subnetwork, ostomate subnetwork; community nurses subnetwork; physical therapy services; psychological and sexual services.

Particularly, the recruitment protocol starts in outpatient services, where the patient with a pelvi-perineal disorder receives the first assessment and the access to the network (Level 1). After the identification of the main and eventual secondary associated disorders, the patient is assigned to the hospital specific clinic (urological, uro-gynaecological, proctological, etc – Level 2) receiving the hospital multidisciplinary management approach. Level 3 is reserved for complex cases requiring expert decision management (combined multi-specialty surgical procedures).

After the hospital management approach, the patient is assigned to the sub-networks (perineal rehabilitation, ostomate, community nurses) to receive the individualized care. The clinical pathways are updated and re-discussed timely.

This strategic functional and structural pelvi-perineal coordinated network, with trained nurses and specialized physicians, has permitted us to achieve patient's health improvement in both prevention and management, human resource rationalization, expense reduction, and knowledge diffusion.

Contributions

Concept: F.B., C.O., Data Collection or Processing: F.B., C.O., Analysis or Interpretation: F.B., C.O., Literature Search: F.B., C.O., Writing: F.B., C.O.

Ethics

Peer-review: Internally peer-reviewed.

DISCLOSURES

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REFERENCES

1. Petros PE, Ulmsten U. An Integral Theory of female urinary incontinence. Acta Obstetricia et Gynecologica Scandinavica 1990; 69: 7-31.
2. Bogliatto F, Boraso F. A new model of care: the lower female anourogenital network. J Low Genit Tract Dis 2016; 20: 105-6.
3. Cunningham FC, Ranmuthugala G, Plumb J, Georgiou A, Westbrook JI, Braithwaite J. Health professionals networks as a vector for improving healthcare quality and safety: a systematic review. BMJ Qual Saf 2012; 21: 239-49.
4. Den Breejen EME, Hilbink MAWH, Nelen WLDM, et al. A patient-centered network approach to multidisciplinary-guideline development: a process evaluation. Implementation Science 2014; 9: 68.

New section: Vulvodynia

Pelviperineology Journal deals with many aspects of pelvic floor malfunction. One such aspect is pain. With this issue we start a section dedicated to vulvodynia. Vulvodynia is defined as: "vulvar pain of at least three months' duration, without clear identifiable cause, which may have potential associated factors".[1] It is still an enigma. Its etiology, pathophysiology, and treatment have not yet been elucidated. The most perplexing presentation is pain during intercourse, sometimes so severe as it to prevents the possibility to have intercourse. Unfortunately, so far, treatment of vulvodynia has been unsatisfactory, leading to despair of many patients. However, recently, with the understanding that vulvodynia has associated factors,[1] and that some of the factors are common to pelvic floor dysfunction,[2] the paradigm has changed. Currently, the treatment should be picked according to the associated factor. The study of the associated pelvic floor conditions has now led to novel research on the interaction between neuroproliferation which is commonly found in vulvodynia, inflammatory and immune processes, and pelvic floor pathology.

Since the management of vulvodynia is multidisciplinary, it is discussed by Gynecologists, Pelvic floor specialists, Physical therapists, Dermatologists, Psychologists, Sex therapists, and more. However, so far, there is no journal that devoted a specialized section to vulvodynia.

Quite a few papers on that topic have already been published in Pelviperineology over the years. Nevertheless, acknowledging the significance of the topic, we decided to start a unique section, that will appear with each issue of Pelviperineology, and will be dedicated to publication of studies regarding vulvodynia etiology, pathophysiology, research and treatment.

In the present issue, two very important aspects of vulvodynia are discussed, in two separate publications:

First, Dr. Joana Lyra et al.[3], meticulously reviewed the treatment of provoked vulvodynia, formerly called: "vestibulitis", by surgery. Surgery is a very successful treatment of provoked vulvodynia, and its success may now promote it to a first line approach in certain cases.

In the other paper, Dr. Ewa Baszak-Radomańska et al.[3] propose an outstanding diagnostic approach to the pain and malfunction associated with vulvodynia, taking in account many aspects of the condition.

I hope that devoting a specified section to this condition raises the interest and research and I hereby invite submissions to this section.

Professor Jacob Bornstein,
Editor in chief, Pelviperineology

References

1. Bornstein J, Goldstein AT, Stockdale CK, et al. Consensus vulvar pain terminology committee of the International Society for the Study of Vulvovaginal Disease (ISSVD), the International Society for the Study of Women's Sexual Health (ISSWSH), and the International Pelvic Pain Society (IPPS). 2015 ISSVD, ISSWSH, and IPPS Consensus Terminology and Classification of Persistent Vulvar Pain and Vulvodynia. J Low Genit Tract Dis 2016; 20: 126-30.
2. Schonfeld M, Petros P, Bornstein J. Mechanically Supporting Uterosacral Ligaments for the Relief of Provoked Vulvodynia: A Randomized Pilot Trial. J Pain Res. 2021; 14:1281-1288.
3. Joana Lyra J, Joana Lima-Silva J, Pedro Vieira-Baptista P, Preti M, Bornstein J. Surgical treatment for provoked vulvodynia – Where do we stand? A narrative review. Pelviperineology 2021; 40:120-127.
4. Baszak-Radomańska E, Wańczyk-Baszak J, Paszkowski T. VAMP (Vulva, Anus, pelvic Muscles and Paraurethra) protocol for physical examination of pelvic floor in vulvodynia. Pelviperineology 2021; 40:128-144.

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Surgical treatment for provoked vulvodynia – Where do we stand? A narrative review

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ABSTRACT

The treatment of vulvodynia remains challenging. Surgery (vestibulectomy) is an option for localized vulvodynia, but it is often considered only after failure of conservative approaches. The authors reviewed the available literature to establish the role, indications, complications, and success rates of surgical procedures.

We conducted a literature search of all the papers published and indexed in PubMed since 2011 on the surgical treatment of vulvodynia.

Women with localized provoked vulvodynia (LPV) form are the best candidates for the surgical treatment of vulvodynia. Success is associated with secondary LPV, improvement with lidocaine, premenopausal status, and intermittent rather than constant pain. While medical/conservative treatment should generally be the first option, if a neuroproliferative etiology is suspected, surgery can be a first-line treatment. The available data do not allow us to draw conclusions about the best surgical technique. Efficacy (defined in different ways) is high (52%–97%). The complication rate is low, cosmetic results are good, and vaginal delivery seems possible.

Vestibulectomy is a safe and effective treatment for vulvodynia when delivered to appropriately selected women.

Keywords: Neuroproliferation; provoked vulvodynia; surgery; vestibulectomy

INTRODUCTION

In 2015, the International Society for the Study of Vulvovaginal Disease (ISSVD), the International Society for the Study of Women Sexual Health, and the International Pelvic Pain Society published a unified definition of and terminology for vulvodynia. Vulvodynia is defined as vulvar pain of at least three months' duration without a clear identifiable cause that may have potential associated factors.[1]

Vulvodynia affects all age groups at a prevalence of 6.1%–20.8%[2-4] in premenopausal women, when this condition appears more frequently.[5] Besides its recognized prevalence, it is believed to be underdiagnosed, with only 60% of symptomatic women seeking help and 40% of these patients never receiving a diagnosis.[6] As a chronic disorder, vulvodynia is associated with significant health and psychological burden as well as considerable costs.[7-9]

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We can further categorize vulvodynia in terms of location (localized to the vestibule or the clitoris, also named vestibulodynia or clitorodynia), generalized, or mixed; provocation (upon contact or spontaneous); onset (primary or secondary); and temporal pattern (intermittent or constant). These descriptors were defined in a more recent document published by the same consensus group.[10] Despite attempts to further categorize vulvodynia, this is a difficult task, as we still lack important information about its etiology. It is considered a multifactorial condition that envelopes a large spectrum of symptoms and pathophysiological events.[11] In fact, its diagnosis is one of exclusion. The main advantage of the 2015 consensus terminology is that it leads to a paradigm shift in treatment from a trial-and-error approach to tailoring the treatment according to the associated factors; if a significant musculoskeletal factor is documented during the physical examination, physical therapy is proposed. When a psychological factor is revealed, the treatment should consider it.[12]

The new paradigm has not been studied in large cohorts; therefore, most data published to date on treatment options are of low quality. In addition, the studied treatments were often performed without proper patient selection.[13,14] In the past, a multi-step approach starting with conservative and medical treatments was advocated.[5,12] Surgery, since it is a more invasive and definitive option, is usually viewed as a last resort. Surgery should be offered earlier to women with peripheral neurologic associated factors, that is, neuroproliferation. This phenomenon can be a consequence of increased inflammation in vestibule, although the association between the latter and mucosal allodynia remains to be proven.[11,12] Regarding the inflammatory milieu, the most consistent finding is an increased number of mast cells in the vestibular tissue and a reduced systemic number of natural killer cells,[9,15,16] which may be triggered by yeast infections.[17] Some genetic polymorphisms can also explain different inflammatory responses to the same insults.[5,18,19] Inflammation can lead to angiotensin II formation, which induced angiotensin AT2 receptor–mediated neuronal cells, eventually driving nociceptor axon sprouting.[20]

The present study reviews the literature in the field and summarizes the current state of the art of surgical treatment of vulvodynia.

Materials and methods

We performed a literature search for all studies published and indexed in PubMed using the terms “vulvodynia,” “vestibulodynia,” “clitorodynia,” “surgical treatment,” “surgery,” and “vestibulectomy.” We limited our search to papers and guidelines written in English for which the full text was available. We included papers published in the last ten years (2011–2021, with 38 articles retrieved). If relevant, older references found in this set of papers were included in the review (the oldest reference included was dated 1983). We included case reports, case series, review articles, randomized control trials (RCTs), systematic reviews, and meta-analyses. All the included studies were performed in humans, and we excluded those performed in animals.

Ethical approval was not required for this study since it was a review of the existing literature.

Considering the new ISSVD classification for vulvodynia, particularly its new approach to diagnosis and treatment, we revisited and reviewed the most relevant literature with a focus on surgery as a treatment option for vulvodynia.

Results

Is there an ideal surgical candidate?

LPV is most likely type of vulvodynia to benefit from surgical treatment. From the recognized factors that are associated with this phenotype, a peripheral neurologic mechanism with neuroproliferation seems to represent the group of patients who are the best surgical candidates.[12,21,22]

Pure LPV is restricted to a portion of the vulva (vestibule and clitoris) and triggered only by physical contact. It may represent primary or secondary provoked vulvodynia according to the timing of onset at the first contact or after a period of painless contact. The latter classification may be relevant for predicting treatment response since the likelihood of improvement is reportedly higher in cases of secondary vulvodynia.[10,12,23] This is the most prevalent type of provoked vulvodynia, and its etiology and pathophysiology are the most commonly studied.[8,13,24]

LPV can be associated with an increased density of nerve endings in the vestibular stroma-neuroprolifetation.[12,25,26] This condition is characterized by an increased density of C-afferent nociceptors in the vestibular mucosa, leading to allodynia.[12,21,27] The phenomenon of neuroproliferation was recently confirmed by histological examination of vestibulectomy specimens. This study also reported that women with neuroproliferation-associated vestibulodynia treated conservatively presented higher failure rates than surgery.[21]

Furthermore, some factors can help predict the surgical response (Table 1). Secondary LPV has higher success rates than primary LPV[28] LPV that improves with the application of lidocaine is apparently associated with better surgical results;[9] women who have pain beyond the vestibule (generalized

Table 1. Predictive factors for surgical outcome in treatment of LPV	
Predictive factors of surgery results	
Predictors of better surgical outcome	Secondary LPV
	LPV that improves with lidocaine application
Predictors of poor surgical outcome	Generalized vulvodynia
	Postmenopausal woman
	LPV with constant pain
LPV: Localized provoked vulvodynia	

vulvodynia) triggered during a cotton-tipped test or other type of contact and those with severe comorbidities can experience less improvement with surgery;[29] surgery in postmenopausal LPV plays a limited role;[5] and constant pain is associated with a higher risk of surgical failure.[30]

When should we consider surgery for provoked vulvodynia?

After identifying a possible surgical candidate, the potential timing of surgery must be determined. Considering that surgery is a definitive treatment, in cases in which reservations exist about the pathophysiological mechanisms underlying vulvodynia, most experts would unfortunately recommend proceeding to surgery only once less invasive interventions fail.[5,22] Surgery boasts similar pain and sexual function outcomes to those of conservative interventions, reinforcing that medical treatment should be considered before surgery.[31]

On the other hand, there is some consistent evidence about the success rates of vestibulectomy for LPV and, since the new 2015 consensus guiding the therapeutic approach considering associated factors, in cases of neuroproliferative LPV without other associated factors, surgery can be considered as an early effective treatment.[12,32]

Variations in surgical techniques and other practical issues

Anatomical remarks: The vestibule has a ring shape that extends laterally from the hymenial ring to Hart's line localized on the inner surface of the labia minora. It travels anteriorly to the clitoris frenulum and posteriorly to the fourchette. It contains the urethral meatus, vaginal introitus, and vestibular gland openings (Skene and Bartholin's glands). It originates from the urogenital sinus; as such, it has endodermal tissue. It is covered by nonkeratinized squamous epithelium.[12,18,26] Another important remark is the lower level of estrogen and progesterone sensitivity in the vestibule than in the vagina.[33]

Surgical techniques: In 1983, Woodruff and Parmley were the first authors to describe vestibulectomy. It consisted of the excision of a semicircular segment of the perineal skin, posterior vestibular mucosa, and posterior hymenial ring. Subsequently, the defect was closed by approximation of the undermined vaginal mucosa to the perineum.[34]

Over the years, new techniques have been described that aim to improve the success rate and decrease complications. The only systematic review that compared the various techniques found no differences in outcomes and complication rates, so the technique should be the one most familiar to the surgeon that allows for the removal the entire painful area.[5,18]

It is difficult to systematize the different techniques, as few variations exist between them, and most studies did not fully explain all of the surgical steps.

Simplified vestibulectomy: Removing painful areas using a skinning technique and not extending beyond Hart's line. The hymen remains intact.[18]

Posterior/modified vestibulectomy: Only the posterior part of the vestibule is excised (from 2 o'clock to 10 o'clock, inside Hart's line). It could be an option for women reporting only posterior pain.[8,9,12,27] The hymen is not routinely excised. The posterior part of the vestibule is skinned to a depth of 2–4 mm and the hymen is used as a surgical flap with vaginal advancement (1–2 cm) if necessary.

Total vestibulectomy with vaginal advancement: The anterior and posterior vestibules are excised (sometimes used even in the absence of pain in the anterior area to reduce the risk of recurrence), accompanied by a vaginal advancement flap. In addition to removing the parameatal area (1 o'clock to 4 o'clock and 8 o'clock to 11 o'clock), excision extends 5 mm past the hymenial ring all around, resulting in total hymenectomy.[35] Posteriorly, the resection area has a wedge shape (from 4 o'clock to 8 o'clock) extending to the perineum 0.5–1 cm beyond Hart's. The vestibule is skinned to a depth of 2–3 mm and removed with the hymen. A vaginal flap is created by dissection of the vaginal mucosa from the rectovaginal fascia with at least 2 cm to allow closure of the defect without tension.

In 2019, Wu et al.[26] published a detailed description of surgical technique, advocating extensive vestibule removal, a vaginal dissection sufficient to remove as many pain fibers as possible, and the importance of a tension-free defect closure.[26] Still focusing on the technique, other details described in the literature that can facilitate the procedure include the following:[18,27]

- Use a pen to mark the area that should be removed;
- Catheterize the urethra;
- Inject adrenaline with or without lidocaine solution into the mucosa to improve the hemostasis and postoperative pain;

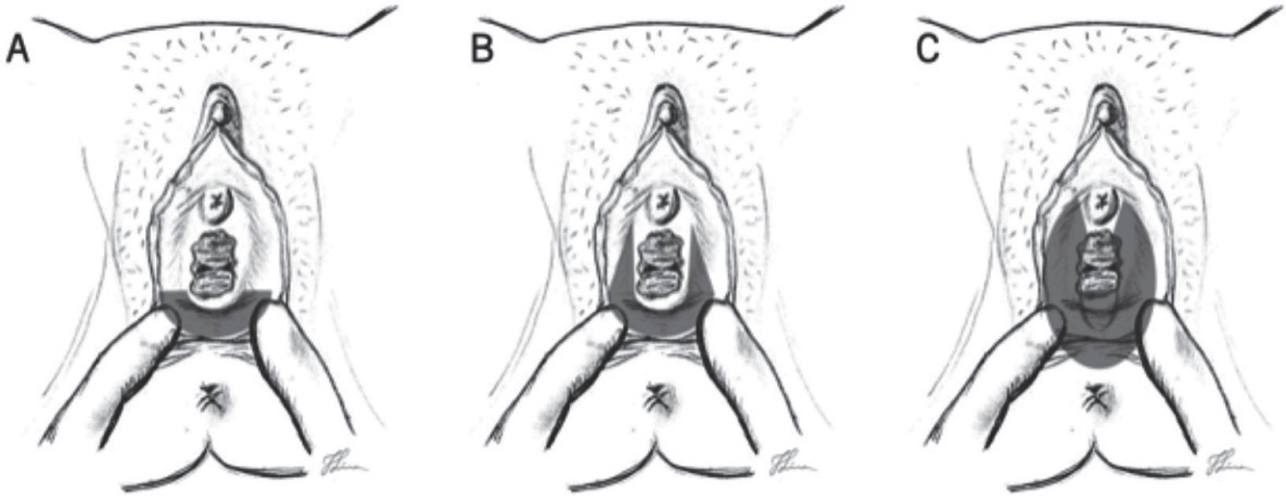

Figure 1. Representation of the three surgical techniques: **A)** Simplified vestibulectomy/vestibuloplasty; **B)** Posterior/Modified vestibulectomy; **C)** Total vestibulectomy with vaginal advancement

- Use two rows of 3-0 Vicryl U-shaped mattress stitches to approximate the vaginal mucosa from the new skin line. The sutures should penetrate the vaginal mucosa backward through the rectovaginal fascia and back again through the vaginal mucosa; they should be placed from anterior to posterior (craniocaudal) to avoid narrowing the introitus. These sutures were not placed anteriorly to avoid urethral injury; and

- Approximate the tissues with interrupted stitches with 4-0 Vicryl and close the anterior defect with a running interlocked 4-0 Vicryl suture.

Postoperatively, the use of ice packs can help prevent swelling and patients should limit physical activity for 4–6 weeks. After complete healing, the use of vaginal dilators could be advocated to stretch the introitus.

Other procedures such as Vestibuloplasty have also been described, but a randomized controlled trial revealed that it failed to improve symptoms.[36]

Complications: The most frequent complications associated with these procedures are bleeding and consequent hematoma formation, dehiscence, scar tissue formation, increased pain, and Bartholin's gland cyst formation.[37] Despite the cosmetic result usually being excellent, there are reports of women unsatisfied with their postoperative vulvar appearance.[38] The use of appropriate surgical techniques and some of the precautions mentioned above can diminish the occurrence of these complications. The incidence of immediate postoperative complications seems low.[18] Recurrence rates are variable, ranging from 0% to 13%.[18]

The Bartholin's gland openings are adjacent to the hymen at the 5 o'clock and 7 o'clock positions. The occurrence of cysts in this location after vestibulectomy is reportedly 5%–9%, and they are responsible for pain and bulging symptoms. The need for surgical repair was present in two-thirds of recurrent cases, most occurring from 1.5 months until 2 years postoperative.[18] For this reason, some authors favor the excision of Bartholin's glands during vestibulectomy to prevent postoperative complications.[12] However, this is not consensual, as the risk of cyst formation is low and Bartholin's gland removal could be a complex surgery with increased operative time, blood loss, and risk of pudendal nerve injury.[39]

Is surgery sufficient or should it be combined with other therapeutic measures?

Despite the ability of surgery to control pain, other concerns persist, and associated symptoms can remain undertreated. For example, there could be decreased vulvar sensitivity after surgery that can negatively affect sexual function. In addition, the prolonged cycle of pain can lead to other psychological dysfunction that limits the full experience of the new painless state achieved with surgery. A combination of physical and psychosexual therapy can amplify the improvement experienced by these patients.[9,12,32,40] In addition, neuropathic pain medication has been continued postoperatively to maximize the surgical results, both in terms of quality of life and sexual function.[41]

Success rates and evidence supporting surgery

Surgery is the most studied treatment for LPV.[5] Vestibulectomy has a success rate of 65%–90%[12,42,43] (Table 2), with an 85% likelihood of permanent improvement when performed by an experienced surgeon in selected patients.[29] The use of different variations of the same procedure does not seem to impact the global surgery success rate.[18] The outcomes evaluated in previous studies were dyspareunia (improvement in 79–89%),[18,44]

Table 2. Summary of the studies, surgical techniques, and surgical success rates				
Surgery and study	**n**	**Study design**	**Outcomes**	**Success rate**
Simplified vestibulectomy/vestibuloplasty				
Bornstein, 1995[36]	21	Randomized control trial	Self-reported symptoms	0/10 vs 9/11 (perineoplasty)
Goetsch, 1996[53]	12	Case series (prospective)	Self-reported dyspareunia	83%
Lavy, 2005[54]	59	Case series (retrospective)	Self-reported dyspareunia	73.6%
Goetsch, 2007[55]	111	Case series (retrospective)	Self-reported dyspareunia	64%
Goetsch, 2008[56]	119	Case series (retrospective)	Self-reported dyspareunia	68%
Goetsch, 2009[39]	155	Case series (retrospective)	Vestibular tenderness (touch test)	83%
Posterior/modified vestibulectomy				
Kehoe, 1999[57]	57	Case series (prospective)	Self-reported dyspareunia	61.1%
Gaunt, 2003[58]	42	Case series (retrospective)	Pain score (objective, subjective findings)	90%
Eva, 2008[59]	110	Case series (retrospective)	Self-reported dyspareunia	83%
Goldstein, 2006[60]	104	Case series (retrospective)	Patient satisfaction	93%
Tommola, 2011[61]	70	Retrospective cohort study	Patient satisfaction	91%
Tommola, 2012[31]	39	Case-control study	Patient satisfaction	89%
Total vestibulectomy				
Bergeron, 1997[62]	38	Case series (retrospective)	Self-reported dyspareunia	63.2%
Bergeron, 2001[45]	78	Randomized control trial	Pain and sexual function	68.2%
Schneider, 2001[63]	54	Case series (prospective)	Self-reported dyspareunia	83%
Rettenmaier, 2003[64]	27	Case series (retrospective)	Self-reported dyspareunia	62%
Traas, 2006[65]	126	Case series (retrospective)	Pain and sexual function	Sustained improvement
Bergeron, 2008[46]	51	Prospective cohort study	Self-reported dyspareunia	52%
Bohm, 2008[66]	67	Case series (retrospective)	Self-reported dyspareunia	56%
Bornstein, 2020[51]	32	Retrospective cohort study	Pain, sexual function, satisfaction	87.5%–97%

n: Number

pain and tenderness of the vestibule evaluated with the cotton-swab test (improvement in 70%–85%), patient satisfaction with the procedure (90%),[18] and improved sexual function.[21]

An RCT of the treatment of provoked vulvodynia that compared surgery with electromyography biofeedback or cognitive-behavioral therapy, reported greater pain reduction with vestibulectomy,[45] but no differences in dyspareunia were reported after surgery versus cognitive-behavioral therapy at 2.5-year follow-up.[46]

Vestibulectomy failure may be associated with preservation of the anterior part of the vestibule since sensitivity may develop there later with the use of techniques such as posterior/modified and simplified vestibulectomy.[12]

Despite the considerable evidence of surgical success reported by case series and RCTs, well-designed comparative studies incorporating a larger number of patients with LPV selected for surgery are lacking. Studies that use a precise definition of the diagnosis and associated factors, provide a detailed description of the technique used, detail the patient selection criteria, and include adequate follow-up are of major importance for validating future results.[24,27,47] The need for agreement about validated outcome measures is of the utmost importance before researchers continue to investigate the therapeutic effects for LPV.[23,48]

Pregnancy and delivery after vestibulectomy

There is little evidence about pregnancy and the best delivery route after vestibulectomy. Burrows et al.[49] reported that, among 44 women with at least one term pregnancy after vestibulectomy, 21 had a cesarean section and 23 had a vaginal delivery. Among the latter, 48% (11) had no perineal lacerations, 13% (3) required a midline episiotomy, and 4.4% (1) experienced a fourth-degree laceration. The authors considered that vaginal delivery after vulvar vestibulectomy seems safe with perineal morbidity similar to that of the general population. In addition, it alone is not an indication for cesarean section.[49]

According to Bornstein, perineal tears during delivery after vestibulectomy are rare. This may be due to excision of the perineal tissue during the procedure, which removes tension from these tissues.[12]

CONCLUSION

Our knowledge about vulvodynia has evolved, and we have gained a new perspective the past few years, mainly after the revised ISSVD consensus terminology and classification were published in 2015.[1] However, considering the relatively recent systematic approach to the classification of this disease, level 1

evidence is still lacking, the availability of which would provide better insight into the best treatment options, with surgery being no exception.[14]

Since we still poorly understand the mechanisms of vulvodynia, it is not simple to rely on its pathophysiology for choosing a treatment option. When we can easily identify a trigger or reason for the pain, treatment will focus on its correction. However, most clinicians will adopt a stepwise methodology, with non-invasive medical treatments coming first and surgery being reserved for refractory cases.[5,14,31,50] However, not all experts agree that surgery should be considered a last resort. For example, in the specific case of vulvodynia associated with neuroproliferation of the nerve endings in the vestibular endoderm tissues[1] – a recognized alteration that leads to provoked vulvodynia – surgery may be the only indicated treatment.[12] Successful rates range from 52%–97% depending on the series and the most frequent complications are bleeding and consequent hematoma formation, dehiscence, scar tissue formation, increased pain, and Bartholin's gland cyst formation. There is no consensus or sufficient evidence on the best technique, so it should be used the one most familiar to the surgeon that allows for the removal the entire painful area. Overall, surgery is considered a safe and successful option for vulvodynia treatment, mainly when performed in selected women. Furthermore, symptomatic improvement after surgery is maintained for long periods, and in cases where symptoms do not immediately subside after surgery, it is described a gradual improvement over the years.[51]

This paradigm shift, in which the treatment choice is determined by the identified associated factors rather than a standardized methodology that fits all women with vulvodynia, is mainly a consequence of the 2015 ISSVD consensus and our best understanding of vulvodynia.[12,21]

Despite the controversies discussed above, there is consensus about the importance of involving patients in the treatment steps and goals. In most cases, there is no single treatment that will completely resolve symptoms, and it may take time for the patient to experience significant relief.[12,52] Therefore, discussing and explaining treatment goals and expectations is essential for patient compliance and satisfaction.

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Ethics

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REFERENCES

1. Bornstein J, Goldstein AT, Stockdale CK, et al. 2015 ISSVD, ISSWSH and IPPS consensus terminology and classification of persistent vulvar pain and vulvodynia. Obstet Gynecol 2016; 127: 745-51.
2. Harlow EGS. A population-based assessment of chronic unexplained vulvar pain: have we underestimated the prevalence of vulvodynia? J Am Med Womens Assoc 2003; 5-8: 82-8.
3. Mitro SD, Harlow SD, Randolph JF. Chronic vulvar pain in a cohort of post-menopausal women: atrophy or vulvodynia? Women Midlife Heal 2016; 2: 4.
4. Vieira-Baptista P, Lima-Silva J, Cavaco-Gomes JB. Prevalence of vulvodynia and risk factors for the condition in Portugal. Int J Gynecol Obs 2014; 127: 283-7.
5. Vieira-baptista P, Donders G, Margesson L, Edwards L. Diagnosis and management of vulvodynia in postmenopausal women. Maturitas 2017; 108: 84-94.
6. Harlow BL, Wise LA, Stewart EG. Prevalence and predictors of chronic lower genital tract discomfort. Am J Obs Gynecol 2001; 185: 545-50.
7. Bornstein J, Preti M, Radici G, Stockdale CK. Vulvodynia: a neglected chronic pain diagnosis. Pain 2019; 160: 1680-85.
8. Sadownik LA. Etiology, diagnosis, and clinical management of vulvodynia. Int J Womens Health 2014; 6: 437-49.
9. Stenson AL. Vulvodynia diagnosis and management. 2017; 44: 493-508.
10. Bornstein J, Preti M, Simon JA, et al. Descriptors of vulvodynia : a multisocietal definition consensus (International Society for the Study of Vulvovaginal Disease, the International Society for the Study of Women's Sexual Health, and the International Pelvic Pain Society). J Low Genit Tract Dis 2019; 23: 161-3.
11. Bergeron S, Reed BD, Wesselmann U, Starke NB. Vulvodynia. Nat Rev Dis Prim 2020; 6: 36.
12. Bornstein J. Vulvar Disease Breaking the Myths. Switzerland: Springer International Publishing AG, Part of Springer Nature, 2019.
13. Vieira-Baptista P, Lima-Silva J, Pérez-López FR, Preti M, Bornstein J. Vulvodynia: a disease commonly hidden in plain sight. Case Reports Women's Heal 2018; 20: e00079.
14. Goldstein AT, Pukall CF, Brown C, Bergeron S, Stein A, Kellogg-spadt S. Vulvodynia: assessment and treatment. 2016; 13: 572-90.
15. Chalmers KJ, Madden VJ, Hutchinson MR, Moseley GL. Local and systemic inflammation in localized, provoked vestibulodynia: a systematic review. Obs Gynecol 2016; 128: 337-47.

16. Regauer S, Eberz CBS. Mast cell inflitrates in vulvodynia represent secondary and idiopathic mast cell hyperplasias. APMIS 2015; 123: 452-6.

17. Päivi Tommola M, Ralf Bützow P, Leila Unkila-Kallio P, Jorma Paavonen P, Seppo Meri P. Activation of vestibule-associated lymphoid tissue in localized provoked vulvodynia. AJOG 2015; 212: 476.E1-8.

18. Tommola P, Unkila-Kallio L, Paavonen J. Surgical treatment of vulvar vestibulitis: a review. Acta Obstet Gynecol Scand 2010; 89: 1385-95.

19. Havemann LM, Cool DR, Gagneux P, et al. Vulvodynia: what we know and where we should be going. J Low Genit Tract Dis 2017; 21: 150-6.

20. Liao Z, Chakrabarty A,Muet Y, et al. A local inflammatory renin-angiotensin system drives sensory axon sprouting in provoked vestibulodynia. J Pain 2017; 18: 511-25.

21. King MA, Mitchell LS, Belkin Z, Goldstein AT. Vulvar vestibulectomy for neuroproliferative-associated provoked vestibulodynia: a retrospective analysis. J Gynecol Surg 2018; 34: 58-62.

22. Rosen NO, Dawson SJ, Brooks M, Kellogg-Spadt S. Treatment of vulvodynia: pharmacological and non-pharmacological approaches. Drugs 2019; 79: 483-93.

23. Davenport RB, Voutier CR, Veysey EC. Outcome measurement instruments for provoked vulvodynia: a systematic review. J Low Genit Tract Dis 2018; 22: 396-404.

24. Henzell H, Berzins K, Langford JP. Provoked vestibulodynia: current perspectives. Int J Womens Health 2017; 9: 631-42.

25. Westrom LV WR. Vestibular nerve fiber proliferation in vulvar vestibulitis syndrome. Obs Gynecol 1998; 91: 572-6.

26. Wu C, Goldstein A, Klebanoff JS, Moawad GN. Surgical management of neuroproliferative-associated vestibulodynia: a tutorial on vestibulectomy with vaginal advancement flap. Am J Obstet Gynecol 2019; 221: 525.e1-5.e2.

27. King M, Rubin R. Current uses of surgery in the treatment for genital pain. Curr Sex Heal Rep 2014; 6: 252.

28. Bornstein J, Maman M AH. Primary versus secondary vestibulitis - one disease, two variants. Am J Obs Gynecol 2001; 184: 28-31.

29. Edwards L. Vulvodynia. Clin Obstet Gynecol 2015; 58: 143-52.

30. Bornstein J, Goldik Z, Stolar Z, Zarfati D. Predicting the outcome of surgical treatment of vulvar vestibulitis. Obs Gynecol 1997; 89: 695-8.

31. Tommola P, Unkila-Kallio L PJ. Long-term well-being after surgical or conservative treatment of severe vulvar vestibulitis. Acta Obs Gynecol Scand 2012; 91: 1086-93.

32. Nunns D, Murphy R. Assessment and management of vulval pain. BMJ 2012; 344: 38-42.

33. Hodgins MB, Spike RC, Mackie RM, et al. An immunohistochemical study of androgen, oestrogen and progesterone receptors in the vulva and vagina. Br J Obstet Gynaecol. 1998. 105: 216-22.

34. Woodruff JD. Infection of the minor vestibular gland. Obs Gynecol 1983; 62: 609-12.

35. Goldstein A. Surgical techniques: surgery for vulvar vestibulitis syndrome. J Sex Med 2006; 3: 559-62.

36. Bornstein J, Zarfati D, Goldik Z. Perineoplasty versus vestibuloplasty for severe vulvar vestibulitis: a randomized comparison. Br J Obs Gynecol 1995; 102: 652-5.

37. Marinoff SC TM. Vulvar vestibulitis syndrome: an overview. Am J Obs 1991; 165: 1228-33.

38. Foster DC, Butts C, Shah KV. Long-term outcome of perineoplasty for vulvar vestibulitis. J Womens Heal 1995; 4: 669-75.

39. Goetsch MF. Incidence of Bartholin's duct occlusion after superficial localized vestibulectomy. Am J Obs Gynecol 2009; 200:688.e1-6.

40. Dias-Amaral A, Marques-Pinto A. Female genito-pelvic pain/penetration disorder: review of the related factors and overall approach perturbação de dor gênito-pélvica e da penetração: revisão dos fatores associados e abordagem geral. Rev Bras Ginecol Obs 2018; 40: 787-93.

41. Stockdale CK, Lawson HW. 2013 vulvodynia guideline update. J Low Genit Tract Dis 2014; 18: 93-100.

42. Ventolini G. Measuring treatment outcomes in women with vulvodynia. J Clin Med Res 2011; 3: 59-64.

43. Andrews JC. Vulvodynia interventions—systematic review and evidence grading. Obstet Gynecol Surv 2011; 66: 299-315.

44. Swanson CL, Rueter JA, Olson JE, et al. Localized provoked vestibulodynia: outcomes after modified vestibulectomy. J Reprod Med 2014; 59: 121-6.

45. Bergeron S, Binik YM, Khalifé S, et al. A randomized comparison of group cognitive-behavioral therapy, surface electromyographic biofeedback, and vestibulectomy in the treatment of dyspareunia resulting from vulvar vestibulitis. Pain 2001; 91: 297-306.

46. Bergeron S, Khalifé S, Glazer HI, Binik YM. Surgical and behavioral treatments for vestibulodynia: two-and-one-half year follow-up and predictors of outcome. Obs Gynecol 2008; 111: 159-66.

47. Pérez-López FR, Bueno-Notivol J, Hernandez AV, Vieira-Baptista P, Preti M, Bornstein J. Systematic review and meta-analysis of the effects of treatment modalities for vestibulodynia in women. Eur J Contracept Reprod Heal Care 2019; 24: 337-46.

48. Sadownik LA, Yong PJ, Smith KB. Systematic review of treatment outcome measures for vulvodynia. J Low Genit Tract Dis 2018; 22: 251-9.

49. Burrows LJ, Sloane M, Davis G, Heller DS, Brooks J. Parturition after vestibulectomy. J Sex Med 2011; 8: 303-5.

50. Davis SN, Bergeron S, Binik YM. Women with provoked vestibulodynia experience clinically significant reductions in pain regardless of treatment: Results from a 2-year follow-up study. J Sex Med 2013; 10: 3080-7.

51. David A, Bornstein J. Evaluation of long-term surgical success and satisfaction of patients after vestibulectomy. J Low Genit Tract Dis 2020; 24: 399-404.

52. Shah M, Hoffstetter S. Vulvodynia. Obstet Gynecol Clin North Am 2014; 41: 453-64.

53. Goetsch MF. Simplifed surgical revision of the vulvar vestibule for vulvar vestibulitis. Am J Obs Gynecol 1996; 174: 1701-5.

54. Lavy Y, Lev-Sagie A, Hamani Y, Zacut D. Modifed vulvar vestibulectomy: simple and effective surgery for the treatment of vulvar vestibulitis. Eur J Obs Gynecol Reprod Biol 2005; 120: 91-5.

55. Goetsch MF. Surgery combined with muscle therapy for dyspareunia from vulvar vestibulitis: an observational study. J Reprod Med 2007; 52: 597-603.

56. Goetsch MF. Patients' assessments of a superficial modified vestibulectomy for vestibulodynia. J Reprod Med 2008; 53: 407-12.

57. Kehoe S. Vulvar vestibulitis treated by modified vestibulectomy. Int J Gynaecol Obs 1999; 64: 147-52.

58. Gaunt G, Good A. Vestibulectomy for vulvar vestibulitis. J Reprod Med 2003; 48: 591-5.

59. Eva LJ, Narain S, Orakwue CO. Is modified vestibulectomy for localized provoked vulvodynia an effective long-term treatment? A follow-up study. J Reprod Med 2008; 53: 435-40.

60. Goldstein AT, Klingman D, Christopher K, Johnson C. MS. Surgical treatment of vulvar vestibulitis syndrome: outcome assessment derived from a postoperative questionnaire. J Sex Med 2006; 3: 923-31.

61. Tommola P, Unkila-Kallio L, Paavonen J. Long-term follow up of posterior vestibulectomy for treating vulvar vestibulitis. Acta Obs Gynecol Scand 2011; 90: 1225-31.

62. Bergeron S, Bouchard C, Fortier M, Binik YM. The surgical treatment of vulvar vestibulitis syndrome: a follow-up study. J Sex Marital Ther 1997; 23: 317-25.

63. Schneider D, Yaron M, Bukovsky I, et al. Outcome of surgical treatment for superficial dyspareunia from vulvar vestibulitis. J Reprod Med 2001; 46: 227-31.

64. Rettenmaier MA, Brown JV. Modified vestibulectomy is inadequate treatment for secondary vulvar vestibulitis. J Gynecol Surg March 2003; 19: 13-7.

65. Traas MA, Bekkers RL, Dony JM, et al. Surgical treatment for the vulvar vestibulitis syndrome. Obs Gynecol 2006; 107: 256-62.

66. Bohm-Starke N. Surgery for localized, provoked vestibulodynia: a long-term follow-up study. J Reprod Med 2008; 53: 83-9.

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VAMP (vulva, anus, pelvic muscles and paraurethra) protocol for physical examination of pelvic floor in vulvodynia

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ABSTRACT

Vulvodynia is a functional chronic pain disorder. The etiology is unclear, although pelvic floor muscle (PFM) dysfunction is suspected as one of the main causes of vulvar discomfort. There are no standardized techniques for the quantification of pain arising from PFM overactivity. The severity of pain can be ascertained by examining four anatomical regions. The two external regions are examined using the cotton swab test, first around the vestibule of the vulva (V), and the second around the anus (A). The two internal regions, both of which are examined bilaterally using digital palpation, include the levator ani muscles (M) and the paraurethral area (P). For simplicity, only one maximum pain score was recorded for each given area, using the numerical rating scale (NRS). The four scores are then recorded under the VAMP acronym. Three of the regions (VMP) may be painful on application of pressure in vulvodynia women. Based on these findings a physical examination schedule is proposed for women presenting with vulvodynia. Confirmation of PFMs dysfunction is essential for conservative management of pain involving pelvic floor physical therapy, general myofascial therapy and biofeedback. The introduction of the VAMP protocol for vulvodynia cases is outlined on the basis of recent literature.

Keywords: Gynecological examination; physical pelvic examination; pelvic floor muscles; vulvar pain; vulvodynia

INTRODUCTION

Vulvodynia is a chronic condition involving vulvar pain of at least 3-month duration, without an identifiable cause, which may have associated pathophysiological factors, as outlined in the 2015 consensus terminology.[1] Apart from spontaneous pain, vulvodynia is vestibular pain that results from intercourse or touch and is called provoked vulvodynia (PV), or combined, as a mixed form.[1]In the general population vulvodynia is estimated to occur in 8.3%–16% of adult women at any one time and more than 25% of women at some point in their lifetime.[2,3] The etiology of vulvodynia is still unclear, although dysfunctional, overactive pelvic floor muscles (PFMs) are suspected as a source of chronic pain. Pelvic floor dysfunction is found in 80%–90% patients with vulvodynia,[4] although other chronic pain factors i.e., systemic, psychosocial, neuroproliferative, inflammatory, genetic, and central nervous system related factors may also be present.[1] It is emphasized that peripheral sensitization and psychological predisposition leads to central sensitization and chronification of pain in some patients.[3-10]

The International Urogynecological Association (IUGA)/ International Continence Society (ICS) reported on the standardization of terminology of PFM function and dysfunction. It proposes the label of overactive PFMs, when pelvic muscles do not relax, or may even contract, when relaxation is functionally needed, as during micturition or defecation.[11]

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Dysfunction of PFM in the form of overactive, non-relaxing muscles can contribute to vulvodynia, including PV, chronic urogenital pain (CUP), bladder pain syndrome (BPS), irritable bowel syndrome (IBS) and chronic pelvic pain (CPP), accounting for most chronic anourogenital pain syndromes.[1,6-10]

The bimanual pelvic examination (BPE) is part of any gynecological assessment of chronic pain patients. It encompasses an examination of vulva, vagina and internal pelvic organs, PFMs are not routinely assessed. Many scales are available to document strength, tone, and tenderness, yet all these scales are subjective and not validated.[9,12-14] As a result, quantification of PFM function is not easy. There is a lack of easy to use and reliable measurement techniques and a lack of cut-off values for pathological conditions. Furthermore, the reproducibility of testing is questionable.[6,7] Without evident and simple descriptors, they are not recommended for clinical practice.[9,14]VAMP protocol (vulva, anus, pelvic muscles and paraurethra) for PFM overactive state examination was assessed and published as a pilot study.[15] On the basis of review from the literature according to BPE requirements,[14] VAMP protocol is presented in the article.

Physical examination for pain in PFM dysfunction

The most important associated feature of overactive PFM is pain on pressure application to the vulva, a form of localized pain, sometimes considered as referral pain.[8,16-18] Deep muscles also reproduce pain when examination is carried out internally via vaginal or rectal access.[8,11,14,18]

A Numeric Rating Scale (NRS) is a preferred instrument for the assessment of pain and has been used in 68% of studies where PFMs were examined in women presenting with pain.[14] According to the NRS, pain is graded using an 11-point numeric scale of 0 to 10, with 0 representing “no pain at all” and 10 “the worst possible pain”.[19]

The NRS was used for research purposes in the Integrated Pain Mapping and Assessment Protocol (IMAP).[8,17,18,20] IMAP evaluated pain severity in women with CUP: dysuria, BPS symptoms and vulvodynia. The IMAP consists of three areas. The first region, for the assessment of pain arising from vulva, pubis, perineal and anal area, using a Q-tip; The second, for digital assessment of pelvic floor points; and the third region, for palpation of specific points in the paraurethral area. A total of 54 points were evaluated and pain scores were recorded for research purposes.[17,18,20]

Vulvar sensitivity and pain were established on the basis of the external examination. The **base** of the **hymenal** remnant has been an important part of the diagnostic criteria for vulvodynia since first proposed by Friedrich[21] in 1987, and was an important element in the assessment of vulvar vestibulitis syndrome, although 13.8% of women with vulvodynia had no increased sensitivity on cotton swab testing.[22] Diagnosis of vulvodynia is not based only on increased vulvar sensitivity.[1,22] In addition, benefits of using the cotton swab test versus other PFM examination is unclear.[12,14,22]

According to the IMAP assessment, Q-tip pressure to the anus is not painful, even though vulvodynia patients sometimes complain of spontaneous pain in the anal area. The internal pelvic muscle pain assessment has been developed to standardize the internal examination procedure, with specific palpation points identified.[8,18] Analysis of all pain scores showed that the most reliable points for the diagnosis of CUP came from palpation of the navicular fossa of the vestibulum and urethral external meatus, the left ischial spine and right puborectalis muscle and the left paraurethral area. The diagnosis of CUP can be made reliably, on the basis of these six points, as derived from the IMAP research.[8,18] Although the IMAP successfully localizes pain in a urogenital pain cohort, it is a research tool and is time consuming because it requires precise pressure on all points, many of which are not relevant from a clinical perspective. Different schedules of physical and internal examination have been proposed in literature for the assessment of PFM status over recent years,[12,14,16] including some that have specifically focused on vulvodynia patients,[12,16] although the lack of cut-off level of pain and quite difficult protocol are the obstacles in the application to clinical practice.

Short version of IMAP: the VAMP protocol

The VAMP protocol is a short, abbreviated version of IMAP, that needs to be evaluated, in order to simplify the pain mapping protocol for clinicians. Chronic pelvic floor pain is assessed on the basis of four anatomical regions of the pelvis; two external (using a cotton swab test) and two internal (using digital palpation to the pressure level accepted by the patient). The external regions include the cotton swab pressure of vulva at the **base** of the **hymenal** remnant (V) and anal area (A); the two internal include the pelvic muscles (M), with bilateral digital palpation of the levator ani muscle, and finally the palpation of the paraurethral area (P), as outlined in Figure 1.

Only one maximum pain score is recorded for a given area, using the NRS. Pain on cotton swab or digital pressure of three regions: VMP (with exception of A) are relevant to pelvic floor overactive dysfunction in vulvodynia women. To draw the conclusion of PFM dysfunction, a recorded VMP score cut-off ≥3 in anyone area, constitutes a PFM overactive state.[15] Although anal area is not painful on examination, it should not be overlooked.

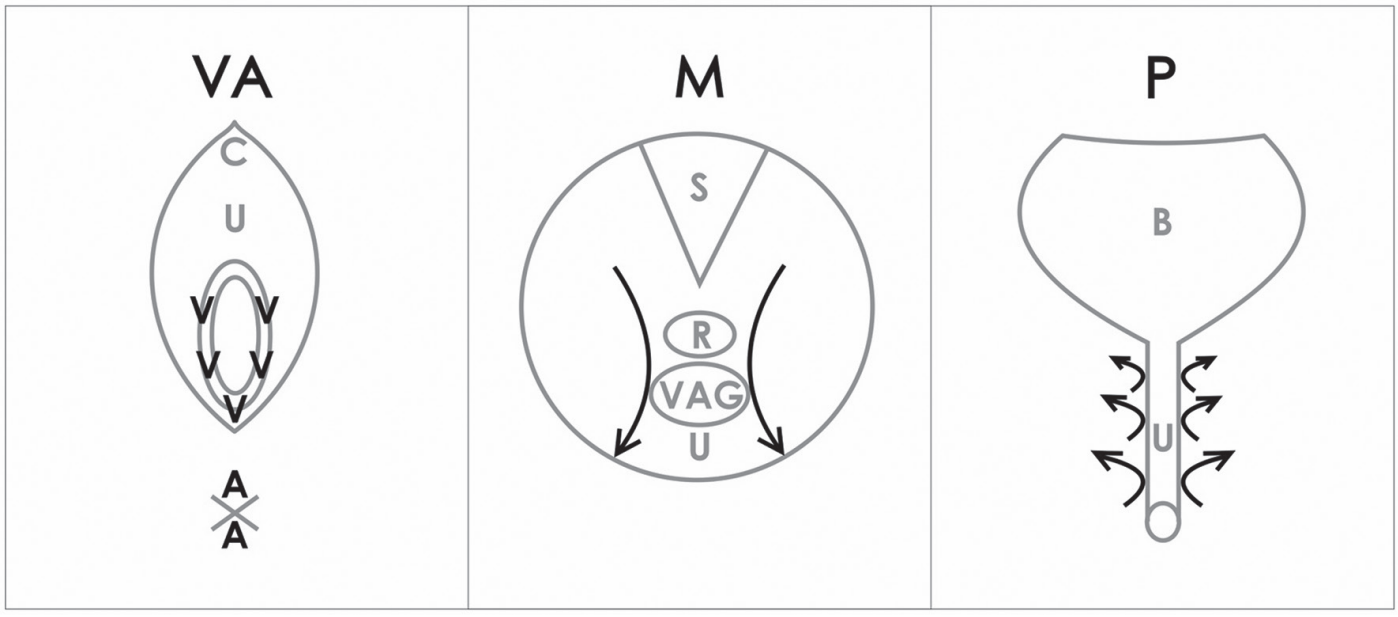

Figure 1. VAMP examination diagram (reproduced from 22) permission of the Polish Society of Gynecologists and Obstetricians (arrows shows the direction of the internal digital examination)

V: vulva 5 points cotton swab pressure; A: anus 2 points cotton swab pressure; C: clitoris; U: urethra; S: sacrum; R: rectum; VAG: vagina; B: bladder; VAMP: Vulva, anus, pelvic muscles and paraurethra

VAMP protocol can be applied during gynecological examination or used separately by healthcare providers as part of the diagnosis of persistent vulvar pain or dyspareunia in patients.

Examination schedule in Vulvodynia

In the diagnosis of persistent vulvar pain in patients, the first step is a detailed history, with particular questions directed to identifying comorbidities.[1,7,13] The second step is gynecological exam to find the cause of pain as mucosa and skin diseases, vaginal infections, or others if visible. On the basis of exclusion other sources of vulvar pain of at least 3 months duration, diagnosis of vulvodynia (in 4 categories i.e., pain location, provocation, onset, and temporal pattern) is undertaken.[1] The third step is focused on PFM status, as the potential source of chronic or recurrent anourogenital pain what is specifically required for PV diagnosis. [7,8,12,13] In vulvodynia patient, when PFM overactive state is not confirmed, other potential associated factors should be taken under consideration.[1]

There are ten important points that need to feature in an examination schedule:

An explanation must be given to the patient that the gynecological examination will focus on pain. Verbal consent needs to be obtained. The patient is forewarned in relation to each step to minimize their fear.

Ask a patient to position themselves in a gynecological chair or couch in a dorsal lithotomy position in preparation for the examination.

Visually examine the vulvar, perineal and perianal area to exclude any pathology as a cause of vulvar pain, itch or discomfort.

Explain the Numeric Rating Scale (NRS) before the examination, and ask the patient for a pain rating of each examined point.

Using a dry cotton swab, gently press the vulva, at the base of hymenal remnant in five different points (in a random or consecutive sequence), avoiding the urethral external meatus area (12 o'clock). Note the maximum NRS rating as a score for V (Vulva) as per Figure 1.

Cotton swab pressure is then applied to the anus at two different points, in the same manner as with the vestibulum area. The maximum NRS score is noted as a score for A (anus), as in Figure 1.

Next a speculum examination is conducted, if it is possible and/or acceptable. Pathological vaginal discharge should be excluded, any other pathology noted.

Any discharge (taken from lateral vaginal wall) should be obtained and assessed for pH, amine odor on application of (5%-10%) KOH (whiff test), microscopic examination for Candida presence is advocated in the office or culture swab may be taken (not obligatory).[22]

A lubricated, gloved index finger is inserted for bimanual transvaginal or rectal examination, for purposes of excluding pelvic inflammatory disease or pelvic mass. Rectal route is indicated if hymen does not permit access, or if the vestibule is too painful or there is significant catastrophizing and vaginal

approach is not possible. Single digital gynecological examination (instead of two fingers insertion) is preferable in every patient and strictly advised in patient with vulvar pain.

The index finger then examines the PFMs (vaginally or rectally). Laterally, progressing from the posterior section, on each side of the rectum (from as far back as possible), along the muscle belly of the iliococcygeus muscle to the anterior portion of the puborectalis muscle, but avoiding the rectum. This is repeated bilaterally with marked tension applied to muscles, but within the acceptable pain threshold of individual patients. This allows for assessment and differentiation of pain severity in superficial PFMs (bulbospongiosus, ischiocavernosus, painful on pressure the most externally) and deep PFMs (levator ani). These muscles are examined in same manner, although precision as to which muscle is painful is not relevant. Maximum NRS rating is noted as a score for M (Muscles) in the medical records (as shown in Figure 1).

The index finger then examines the paraurethral area examination, lateral to the urethra, compressing against the pubic bone. Examination of the urethra is from the distal to the proximal area (from outside to inside) on both the right and left sides. Pressure is applied cautiously with particular attention to patient's level of pain tolerance. Maximum NRS rating is noted as a score for P (paraurethral area) (as on Figure 1).

In the patient's medical history, the pelvic physical exam result is recorded under the VAMP acronym (for example VAMP 3048, although A point is generally not painful, "0" may be skipped). These scores reflect the maximum NRS pain ratings for the four areas: vulva, anus, muscles and paraurethra. At times the examiner may form an impression that the pain rating is overestimated or underestimated, but because it is always a subjective score, it is noted as the patient rates it. Pelvic physical examination using the VAMP protocol is not time consuming for a physician and for a patient when it is carried out as part of the gynecological examination.

Pelvic physical exam requirements

According to Meister et al.[14], based on a systematic review of literature (55 studies since 1946), the following eight recommendations are made in relation to myofascial pain examinations in women;

Document counseling and consent: verbal consent needs to be based on thorough explanation. Vulvar and pelvic examination becomes part of a regular gynecological exam that is performed in patients with pain symptoms. In 45.5% of studies, palpation of the pelvic floor was included in the bimanual physical exam.

Position: a dorsal lithotomy position was chosen in 78.9% of publications.

Numbers of digits inserted: single digit palpation was used in 61,8 % of articles, using gloves and lubrication. A vaginal approach was preferred in 85% of papers, over a rectal approach.

Utilize clock-face orientation (with the 12 o'clock for - symphysis pubis and 6 for PFM).

Preferred order of the examination was mentioned in 30.9% of studies: with 35% beginning with the superficial muscles and then proceeding to deep muscles.

Identifying muscles location: the superficial muscles (2 and 10 o'clock), ischiocavernosus (1 and 11 o'clock), transverse perineal muscles (3 and 9 o'clock), deep layer: pubococcygeus (left: 7 and 11 o'clock, right: 1 and 5 o'clock), iliococcygeus (4 and 8 o'clock) and coccygeus (5 and 7 o'clock, and requires deeper digital insertion).

Examination technique: single sites mid-belly muscle technique is preferable, with a bilateral examination of the levator ani muscle, and obturator internus (reported in only 50% of studies). In some studies, the muscles examined were not specified, but in the greater majority (89.1%) muscles were identified. One third of studies recommend examining levator ani muscle 69.1% of isolated location, 41% in general, 52,7% specified which muscle component should be identified and palpated. Piriformis muscles may not be possible to reach by internal palpation. Lack of description of examination of PFM techniques was identified in 47.3% of articles, with no standard given for the amount of pressure being applied to the PFM in 87.3% of articles (no defined pain/pressure threshold).

Quantifying self-reported pain upon local pressure: the NRS was used in 68% of studies.

According to the VAMP protocol authors, identification of specific pain location during internal pelvic exam is not necessary to draw the conclusion of PFM overactive state.

Only 41.8% of studies incorporate other areas (anus, abdomen, urethra) in pelvic exam. The IMAP research authors[8,18] emphasized the paraurethral area for examination and did not focus on the paraurethral fascia as a possible source of pain. The literature indicated that the anal region was an irrelevant area for assessing PFM pain in conjunction with hypertonic condition. Some suggested that palpation of this area might help identify patients who overestimated the level of pain as may be the case in catastrophic patients. For healthcare practitioner this may be an opportunity to assess the anal area, to exclude comorbid anal pathology (anal fissure, lichens or hemorrhoidal disease) and to use as a means of assessing the validity of the pain scores given by

individuals.To differentiate woman with pelvic pain arising from overactive PFM dysfunction remains a challenge for researchers and practitioners. The most important issue is to distinguish a woman with vulvodynia and other chronic pain patients who may benefit from pelvic myofascial based therapies.[8,14,15,23,24] Once an evidence based, standardized examination is established, the effort can then turn to promoting physician education.[14]

Limitations

The usefulness of VAMP protocol requires reliability, validity of the outcome measures, what is already scheduled in randomized clinical trial by the study authors. The reliability of the participant's pain score can be a source of bias, as perceived pain is always a subjective experience. Furthermore, the examination was performed unblinded, and was based on digital and cotton swab pressure application, without use of calibrated instruments, for purpose of simplifying the diagnostic protocol and ease of clinical practice.

CONCLUSION

The VAMP protocol is proposed as a simple tool for physical examination. A total of four anatomical areas should be examined by the gynecologist, for the sake of a more reliable assessment than the widely used cotton swab test used in the assessment of vulvodynia women. For the patient and healthcare provider the VAMP protocol is not time consuming along with a gynecological examination to obtain information about potential PFM pain arising from overactive muscle dysfunction, as a contributor to vulvodynia. Three of the regions (VMP) are known to be painful on application of pressure in vulvodynia women.[15] Based on these findings a physical examination schedule is proposed for women presenting with vulvodynia. Confirmation of PFMs pain and dysfunction are essential in order to recommend conservative management involving pelvic floor physical therapy, general myofascial therapy and biofeedback. The findings give significant credence to peripheral mechanisms of pain, in which pain of soft tissue origin is examined and potentially responsive to myofascial therapies.[8,23,24]

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Ethics

Peer-review: Internally peer-reviewed.

DISCLOSURES

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

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REFERENCES

1. Bornstein J, Goldstein AT, Stockdale CK, et al. Consensus vulvar pain terminology committee of the International Society for the Study of Vulvovaginal Disease (ISSVD), the International Society for the Study of Women's Sexual Health (ISSWSH), and the International Pelvic Pain Society (IPPS). 2015 ISSVD, ISSWSH, and IPPS Consensus Terminology and Classification of Persistent Vulvar Pain and Vulvodynia. J Low Genit Tract Dis 2016; 20: 126-30.
2. Reed BD, Harlow SD, Sen A, et al. Prevalence and demographic characteristics of vulvodynia in a population-based sample. Am J Obstet Gynecol 2012; 206: 170.e1-9.
3. Harlow BL, Stewart EG. A population-based assessment of chronic unexplained vulvar pain: have we underestimated the prevalence of vulvodynia? J Am Med Womens Assoc 2003; 58: 82-8.
4. Reissing ED, Brown C, Lord MJ, Binik YM, Khalife S. Pelvic floor muscle functioning in women with vulvar vestibulitis syndrome. J Psychosom Obstet Gynaecol 2005; 26: 107-13.
5. Messelink B, Benson T, Berghmans B, et al. Standardization of Terminology of Pelvic Floor Muscle Function and Dysfunction: Report from the Pelvic Floor Clinical Assessment Group of the International Continence Society. Neurourol Urodyn 2005; 24: 374-80.
6. Nunns D, Mandal D, Byrne M, et al. Guidelines for the management of vulvodynia. Br J Dermatol 2010; 162: 1180-5.
7. Goldstein AT, Pukall CF, Brown C, Bergeron S, Stein A, Kellogg-Spadt S. Vulvodynia: Assessment and Treatment. J Sex Med 2016; 13: 572-90.
8. Jantos M. A Myofascial Perspective on Chronic Urogenital Pain in Women. In: Santoro GA, Wieczorek AP, Sultan AH. Springer 2nd ed Springer Nature Switzerland AG 2021
9. Pastore EA, Katzman WB. Recognizing Myofascial Pelvic Pain in the Female Patient with Chronic Pelvic Pain. J Obstet Gynecol Neonatal Nurs 2012; 41: 680-91.
10. Butrick CW. Pelvic Floor Hypertonic Disorders: Identification and Management. Obstet Gynecol Clin North Am 2009; 36: 707-22.
11. Haylen BT, de Ridder D, Freeman RM, et al. An International Urogynecological Association (IUGA)/International Continence Society (ICS) joint report on the terminology for female pelvic floor dysfunction. Int Urogynecol J 2009; 21: 5–26.
12. Zolnoun D, Bair E, Essicks G, Gracely R, Vinita G, Maixner W. Reliability and Reproducibility of Novel Methodology for Assessment of Pressure Pain Sensitivity in Pelvis. J Pain 2012; 13: 910-20.
13. Grimes W. Pelvic floor dysfunction. Updated 6/28/2020; https://www.statpearls.com/articlelibrary/viewarticle/40639/#ref_31804265

14. Meister MR, Shivakumar N, Sutcliffe S, Spitznagle T, Lowder JL. Physical examination techniques for the assessment of pelvic floor myofascial pain: a systematic review. Am J Obst Gynecol 2018; 219: 497.e1-13.

15. Baszak-Radomanska E, Wanczyk-Baszak J, Paszkowski T. Pilot study of testing a clinical tool for pelvic physical examination in patients with vulvodynia. Ginekol Pol 2021; 92: 1-7.

16. Sarton J. Assessment of the Pelvic Floor Muscles in Women with Sexual Pain. J Sex Med 2010; 7: 3526-9.

17. Jantos M, Johns S, Torres A, Baszak-Radomanska E. Mapping chronic urogenital pain in women: rationale for a muscle assessment protocol - the IMAP, Part 1. Pelviperineology 2015; 34: 21-7.

18. Jantos M. Pain mapping: A mechanisms-oriented protocol for the assessment of chronic pelvic pain and urogenital pain syndromes. Pelviperineology 2020; 39: 3-12.

19. dos Santos Calderon P, Peixoto RF, Gomes VM, et al. Concordance among different pain scales in patients with dental pain. J Orofac Pain 2012; 26: 126-31.

20. Jantos M, Johns S, Torres A, Baszak-Radomanska E. Mapping chronic urogenital pain in women: insights into mechanisms and management of pain based on the IMAP, Part 2. Pelviperineology 2015; 34: 28-36.

21. Friedrich EG Jr. Vulvar vestibulitis syndrome. J Reprod Med 1987; 32: 110-4.

22. Reed BD, Plegue MA, Harlow SD, Haefner HK, Sen A. Does degree of vulvar sensitivity predict vulvodynia characteristics and prognosis? J Pain 2017; 18: 113-23.

23. Aguilar VC, White AB, Rogers RG. Updates on the diagnostic tools for evaluation of pelvic floor disorders. Curr Opin Obstet Gynecol 2017; 29: 458-64.

24. Wallace SL, Miller LD, Mishra K. Pelvic floor physical therapy in the treatment of pelvic floor dysfunction in women. Curr Opin Obstet Gynecol 2019; 31:485-93.

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Assessment of the pelvic floor exercises with iPelvis® app. for treatment of women with urinary incontinence: a randomized controlled trial

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ABSTRACT

Objective: To analyze the effectiveness of an application iPelvis® "app" for pelvic floor exercises, alone and associated with physiotherapy, in reducing urinary symptoms and improving the quality of life of incontinent women.

Materials and Methods: This is a longitudinal randomized controlled clinical trial study. The Kings Health Questionnaire (KHQ) and the International Consultation on Incontinence Questionnaire-Short Form (ICIQ-SF) were used as outcome measurements. The participants were divided into four groups: application + physiotherapy (AP + PHYSIO); home exercise sheet + physiotherapy (P + PHYSIO); only application (AP); only home exercise sheet (P).

Results: From the 138 women who were evaluated, 77 (who presented a mean age of 48.31 years) completed the proposed treatments. In the KHQ domains and overall scores, the only ones who presented significant difference between groups were: the General Health Perception (P + PHYSIO higher than P, p=0.008), Social Limitations (P + PHYSIO higher than AP, p=0.04; P higher than AP, p=0.05) and Part I Score (P + PHYSIO higher than P, p=0.04). The analysis of the ICIQ-SF showed P + PHYSIO higher than P (p=0.01). Between the other groups there were no significant differences. The other nine variables analysed no significant differences in all groups.

Conclusion: The use of the application alone and associated with physiotherapy, produces better resulted in reducing urinary symptoms and improving the quality of life of incontinent women, in most variables, but it is not significantly superior in related to the other groups.

Keywords: Applications; iPelvis® app. exercise; pelvic floor; quality of life; urinary incontinence

INTRODUCTION

Pelvic floor dysfunctions are health problems that negatively impact the quality of life of hundreds of thousands of women worldwide.[1] Urinary incontinence (UI), in particular, has a variable prevalence of 13.1% to 70.9% in different populations.[2]

Currently, the first-line therapy for UI is pelvic floor muscle training (PFMT), which is level one evidence and grade A recommendation,[3] indicated by the International Consultation on Incontinence (ICI).[2,4] A recent review by Cochrane confirms that PFMT is effective in curing or improving symptoms of stress urinary incontinence (SUI) and improving quality of life.[5] Despite

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this, there is no consensus on the literature on the administration of this therapy, i.e., if it should be performed by a professional or self-administered.[6]

It is known that the success of PFMT depends on the understanding of the commands given by the therapist and their incorporation into the patient's daily activities.[7] Because of this, one important aspect that should be taken into account when dealing with unsupervised PFMT is adherence, that is defined as "the degree to which a person correctly follows the medical or health professional advice".[8]

With the increased availability of smartphones, the number of applications is growing widely, offering new possibilities for health care provision. Although these applications "apps" are considered capable of increasing adherence to medical treatments, only few have proven their effectiveness in scientific studies.[9]

The PFMT available by the chosen application, follow the protocol developed by Bo et al.[10] and is represented in five stages, respecting the chronological order of motor learning.[11-13] Its differential is the creation of a playful, friendly and encouraging character, who acts as the facilitator in the learning process.[11]

The objectives of this study are: to analyze the effectiveness, of an application for pelvic floor exercises, alone and associated with physiotherapy, in reducing urinary symptoms and improving the quality of life of incontinent women.

MATERIALS AND METHODS

This is a longitudinal, randomized, controlled clinical trial with four parallel groups (1:2) developed in the facilities of Faculdade Inspirar, in Curitiba, Paraná, south of Brazil. We recruited women with stress (SUI), urgency urinary incontinence (UUI), and mixed urinary incontinence (MUI), aged between 18 and 59 years, and with objective demonstration of urinary loss.

We searched for female volunteers in two environments. First, participants were recruited from lists of patients waiting for surgery at the Hospital de Clínicas of the Federal University of Paraná (HC-UFPR). Also, urologists and gynecologists, who work both at HC-UFPR and in their private practices, indicated patients followed in private outpatient clinics who would fit the study. This criterion was important so that there would be no difference in the medical conduct.

Excluded were: pregnant women, women up to six months postpartum, women with prolapse equal or greater than Stage III according to the Pelvic Organ Prolapse - Quantification (POP-Q),[14] with urinary infection, with intrapelvic tumors, with pelvic pain that prevented the performance of the available therapies or who had undergone pelvic surgery in a period shorter than six months, and carriers of pacemakers or intrauterine devices. In addition, participants who missed physical therapy twice in a row were also excluded.

Before performing the evaluation and treatment procedures, each volunteer was informed about the objectives of the study and signed the Informed Consent Form. The research project was approved by the Research Ethics Committee of Faculdade Inspirar, AX – Centro de Estudos da Saúde LTDA. – EPP (opinion number 1,833,987); as well as by the Co-participant Institution, HC-UFPR – Hospital de Clínicas of Federal University of Paraná (opinion number 2,520,073). The study was registered at ClinicalTrials.gov ID: NCT04484753.

The initial evaluation was performed as described in the Clinical Practice Guide of the Brazilian Association of Pelvic Physiotherapy (ABFP), translated from the guide updated and recently published guide of the Royal Dutch Society for Physical Therapy.[15,16] In addition to this evaluation we also applied and analyzed the results from the International Consultation on Incontinence Questionnaire-Short Form (ICIQ-SF),[17,18] and Kings Health Questionnaire (KHQ).[19]

Other ramifications of our research have the functions of pelvic floor muscles as object. Therefore, all participants underwent a physical examination, among other assessment instruments. Due to the fact that about 30% of women are unable to perform contraction only through verbal commands, and that the performance of the inverted maneuver or simultaneous contraction of muscles such as the gluteus, hip adductors and abdominal muscles are very common,[20] all women who presented any difficulty received instruction along with vaginal pelvic floor examination.

After the initial evaluations, each participant went through a drawing to define their participation groups. The groups received the following interventions, without modifications during the research period: application + physiotherapy group (AP + PHYSIO): performed 12 pelvic physiotherapy sessions, in group, once a week and received the iPelvis® Application with PFMT guidelines to be performed at home; home exercise sheet + physiotherapy group (P + PHYSIO): they did 12 Pelvic Physiotherapy sessions, in group, once a week and received a sheet with PFMT guidelines to be performed at home; application "app" only group (AP): received the iPelvis® Application with PFMT guidelines to be performed at home; home exercise sheet only/control group (P): received a sheet with PFMT guidelines to be performed at home. After 3 months all participants were re-evaluated. Many of the women waiting for surgery at HC-UFPR had low financial conditions, hence we offered to cover their transportation costs in order to enable their access to the appointments. The division of the groups is shown in Figure 1.

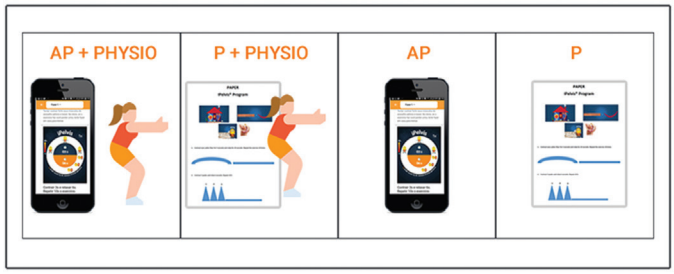

Figure 1. Division of the groups
Source: The Author (2020). AP + PHYSIO: application + physiotherapy; P + PHYSIO: home exercise sheet + physiotherapy; AP: only application; P: only home exercise sheet

The randomization scheme was carried out by a member of the research team, who placed balls with different colors in a box, each one representing one of the intervention groups. Each volunteer took a ball out of the box and from there she was directed to the corresponding group. Initially, groups with physiotherapy (AP + PHYSIO and P + PHYSIO) were allocated, and, after completing these groups, a sequence was given to randomize the groups AP and P. In order to reduce the number of losses of volunteers in the AP and P groups, the answers of the questionnaires from those who could not attend the face-to-face re-evaluation were collected through telephone.

The volunteers drawn for the groups with applications "apps" (AP + PHYSIO and AP), but who did not have a compatible cell phone for their installation, were redirected to the groups with a sheet: from the AP + PHYSIO group they went to the P + PHYSIO group and, from the AP group went to the P group. This was done so that we would not be left with a very small sample and so that the volunteers would have the opportunity to receive intervention.

The team of researchers presented the iPelvis® application to the AP + PHYSIO and AP groups, as well as instructed its installation on the cell phone of each participant, who then received a password. A blocking system was created in order to disable the use of the application in other devices, thus individualizing the system. During the development of the study, the available version of the iPelvis® Application was 1.0, which included 6 phases that lasted 15 days each, totaling 3 months. The P + PHYSIO and P groups received an exercise sheet that contained exactly the same training instructions and life hygiene tips offered by the iPelvis® application.

In the application there are dynamic exercises with sound and image that show how many times each exercise should be performed, the level of strength, and how many seconds to contract and relax the pelvic floor, as shown in Figure 2. The exercises performed in Physiotherapy sessions were the same as those offered within the application and on the exercise sheet.

Figure 2. Example of PFMT in the iPelvis® application
Source: iPelvis® application
PFMT pelvic floor muscle training

Primary outcomes

The primary outcome measures were defined through the ICIQ-SF (a validated Portuguese translation), a tool that assesses the impact of UI on the quality of life and severity of urinary symptoms. The questionnaire is composed of four questions related to frequency, severity of urinary loss and its impact on quality of life.[17,18] The ICIQ score is the sum of the scores from questions three, four and five and ranges from 0 to 21. The impact on quality of life was defined according to the score of question 5: (0) none; (1-3) mild; (4-6) moderate; (7-9) severe; (10) very severe.[21]

Secondary outcomes

The secondary outcomes measure is KHQ, a questionnaire that analyzes the presence of UI symptoms and their impact on various aspects of individuality in quality of life. The questionnaire is composed of 30 questions subdivided into nine domains and a Symptom Severity scale.[19] According to Hebbar et al.[22], KHQ has three Parts Overall Scores: Part 1 Score contains general health perception and incontinence impact domains; Part 2 Score contains role limitations, physical limitations, social limitations, personal relationships, emotions, sleep and energy and severity measures domains; Part 3 Score is considered as a single item where in there are 10 different bladder symptoms. The domains scored between 0 (best) and 100 (worst). The Symptom Severity scale is scored from 0 (best) to 30 (worst).[22]

Sample size

The required sample size of 139 women was calculated taking into consideration the world's female population (3,704,194,620, coutrymeter.com) when the study was conducted, a sampling error of 5% at a 95% confidence level, and a minimum prevalence

of 10% of both urinary and sexual dysfunction. The calculation performed was for prevalence analysis, disregarding the size of the clinically significant effect for the two instruments used.

Statistical analysis

The chi-square test of independence was used to analyze the homogeneity of the groups regarding anthropometric and sociodemographic factors. For age and body mass index, the Snedecor test was used.

The Wilcoxon test for paired data was used to verify whether there were differences between the pre- and post-intervention moments in the ICIQ-SF, KHQ domains and Overall Scores of Parts I, II and III. Analysis of Variance (ANOVA) was used to identify possible effects of the groups under the difference of the means of pre- and post-intervention across ICIQ-SF, all the KHQ domains and Overall Scores of Parts I, II and III. For those variables where there was a significant difference, the Tukey test was applied to identify which groups were significantly different from each other. Statistical analyses were performed using the R software. A significance level of 95% was used ($p<0.05$).

RESULTS

A total of 685 phone calls were made to invite women to participate in this study, from which 177 appointments were scheduled, but only 138 women attended the evaluations that occurred between January 2017 and April 2018. The volunteers evaluated were randomized among the four treatment groups, as shown in Figure 3.

Of the 138 randomized volunteers, 77 completed the treatment and entered the statistical analysis. According to the ICIQ score, 27.3% had mild UI (21/77), 25.9% moderate UI (20/77), 18.2% severe UI (14/77), and 28.6% very severe UI (22/77).

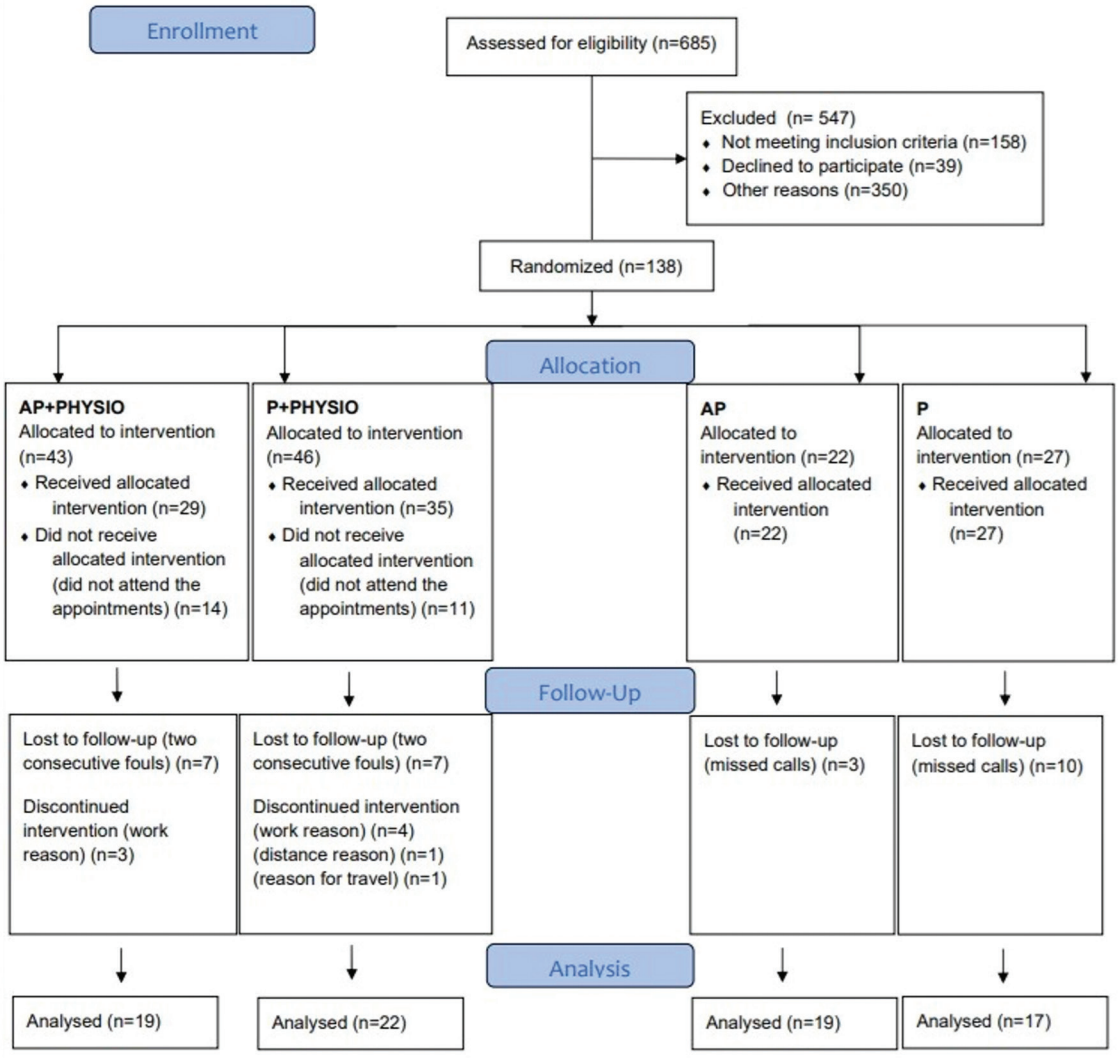

Figure 3. Flow diagram of the study

AP + PHYSIO: application + physiotherapy; P + PHYSIO: home exercise sheet + physiotherapy; AP: only application; P: only home exercise sheet; n: Number

The anthropometric and sociodemographic characteristics of the 77 participants who completed the treatments are shown in Table 1.

Primary outcomes

In the follow-up, AP + PHYSIO, P + PHYSIO and AP groups reported statistically significant reduction in urinary symptoms: (mean reduction in ICIQ-SF score: -4.42 (±4.4); -5.5 (±3.96) and -3.63 (±3.95), respectively), while group P did not present a statistically significant reduction (mean reduction in the ICIQ-SF score): -0.64 (±5.79). The Analysis of Variance showed significant differences between the mean differences of at least two groups, with p=0.01. Tukey's test was performed to identify the significantly different groups. A significant difference was found between the mean differences of the groups P + PHYSIO and P, with p=0.01. The other differences were not significant (Table 2).

Secondary outcomes

The Analysis of Variance demonstrated significant differences between the mean differences of at least two groups in the following KHQ domains and Overall Scores: General Health Perception (P + PHYSIO higher than P, p=0.008), Social Limitations (P + PHYSIO higher than AP, p=0.04; P higher than AP, p=0.05) and Part I Score (P + PHYSIO higher than P, p=0.04). The other differences were not significant. The other domains and Parts did not present significant differences between the groups (Table 3).

Table 1 – Baseline demographic and clinical characteristics.

		AP + PHYSIO (19)	P + PHYSIO (22)	AP (19)	P (17)	p-value
Age	Average	46.73	51.4	45.84	48.82	0.11
	Standard deviation	9.01	6.62	6.26	9.4	
BMI	Average	26.63	28.47	27.51	28.14	0.57
	Standard deviation	3.75	5.12	4.77	3.55	
Type of incontinence	SUI	12 (63%)	6 (27%)	10 (52%)	5 (29%)	0.07
	UUI	0	0	1 (5%)	2 (11%)	
	MUI	7 (36%)	16 (72%)	8 (42%)	10 (58%)	
Marital status	Married	13 (68%)	15 (68%)	11 (57%)	9 (52%)	0.70
	Separated	3 (15%)	4 (18%)	6 (31%)	3 (17%)	
	Single	1 (5%)	2 (9%)	2 (10%)	4 (23%)	
	Widowed	2 (10%)	1 (4%)	0	1 (5%)	
Ethnicity	Caucasoid	19 (100%)	21 (95%)	18 (94%)	15 (88%)	0.31
	Mongoloid	0	0	1 (5%)	2 (11%)	
	Negroids	0	1 (4%)	0	0	
Schooling	1 to 5 years	1 (5%)	5 (27%)	0	0	0.003*
	6 to 10 years	10 (52%)	12 (54%)	4 (21%)	7 (41%)	
	>10 years	8 (42%)	5 (22%)	15 (78%)	10 (58%)	
Income	<2	7 (36%)	13 (59%)	3 (15%)	5 (29%)	0.08
	3 to 4	4 (21%)	3 (13%)	4 (21%)	7 (41%)	
	5 to 6	6 (31%)	6 (27%)	8 (42%)	5 (29%)	
	>6	2 (10%)	0	4 (21%)	0	
Urinary infection (last year)	Yes	9 (47%)	4 (18%)	7 (36%)	7 (41%)	0.22
Smoking	Yes	0	1 (4%)	4 (41%)	1 (5%)	**
Alcoholism	Yes	10 (52%)	3 (13%)	11 (57%)	4 (23%)	0.04*
Sedentarism	Yes	12 (63%)	14 (63%)	9 (47%)	13 (76%)	0.35

*p-value <0.05, rejecting the hypothesis of independence between the groups and the variables schooling and ethylism; ** The low frequency of smokers volunteers made it impossible to perform the test of homogeneity; BMI: body mass index; SUI: stress urinary incontinence; MUI: mixed urinary incontinence; urgency urinary incontinence

Table 2. Primary outcomes measures baseline and at the 3-month follow-up.

Primary outcome	Treatment group	Baseline (SD)	3-month follow-up (SD)	Difference (SD)	Within group p-value (Wilcoxon)	Between groups p-value (ANOVA)	Different groups (Tukey)
ICIQ-SF score	AP + PHYSIO	10.68 (5.37)	6.26 (3.75)	-4.42 (4.14)	0.0009*	0.01*	P and P + PHYSIO (p=0.01)
	P + PHYSIO	13.09 (4.81)	7.59 (4.43)	-5.5 (3.96)	0.00006*		
	AP	9.89 (5.01)	6.26 (6.14)	-3.63 (3.95)	0.001*		
	P	11.82 (5.41)	11.17 (6.18)	-0.64 (5.79)	0.71		

*p-value <0,05; ICIQ-SF: International Consultation on Incontinence Questionnaire - Short Form; SD: standard deviation; ANOVA: Analysis of Variance;

DISCUSSION

Since the first contact with the volunteers, we noticed their anguish and their desire for a fast and resolute treatment due to the fact that many of the women invited to participate in this study were waiting for a surgical procedure and were not considering PFMT as a treatment option. We observed this situation in the analysis of the number of volunteers who attended the initial assessment, but did not initiate the appointments and follow-up with the physiotherapy groups, claiming that they preferred to wait for surgery (32% in the AP + PHYSIO group, 14/43; and 23% in the P + PHYSIO group, 11/46).

Those who participated in the research had the opportunity not only to learn about a new form of treatment, but also to improve their quality of life. However, some of them reported difficulty to be absent from work, which was the main reason for quitting the attendance in physiotherapy groups (7% in the AP + PHYSIO group, 3/43; and 8.7% in the P + PHYSIO group, 04/46).

Sjöström et al.[23] reported a loss of 12% in 4 months and 38% after two years of follow-up,[24] while in our study we observed a loss, including withdrawals and exclusions, of 44%. We believe that the difference is due to the presence of two face-to-face groups in our study, while the Sjöström study did not have any.

Regarding anthropometric and sociodemographic characteristics, we obtained homogeneity in the great majority of variables between groups, except for schooling. P and AP groups had a higher number of participants with more than 10 schooling years, while AP + PHYSIO and P + PHYSIO groups had more participants with low schooling. Despite these differences, the literature describes that the educational level does not seem to affect the ability to learn or perform correct pelvic floor muscle contractions,[25] and does not have association with success in treatment with PFMT.[26] However, the groups can be considered homogeneous regarding income (p=0.08), assuming that not necessarily a higher income indicates a higher education.

The KHQ analysis was performed individually for each of the nine domains and three Parts Overall Scores. The first domain with differences between groups was General Health Perception. All groups showed significant improvement before and after treatment, except the P group. The only significant difference was between the groups P and P + PHYSIO (p=0.008), that is, there is no significant difference between the P + PHYSIO and AP + PHYSIO or AP groups, all of which improve General Health Perception. These data lead us to consider that the use of the application alone or as an aid to Pelvic Physiotherapist promotes improvement of General Health Perception, while only the home exercise sheet with instructions without the physiotherapist's intervention does not produce significant differences.

Social limitations decreased significantly in the AP + PHYSIO, P + PHYSIO and P groups. The AP group was the only one without significant difference. The groups that were significantly superior were P + PHYSIO and P in relation to the AP (p=0.04 and p=0.05, respectively). In this domain, women were asked about their social limitations or meetings with friends resulting from to their bladder problem. It was noticed that the AP group had the lowest initial average in relation to the other groups. Possibly a more careful strategy regarding randomization could present different results.

In the Part I Overall Score all groups showed significant improvement before and after treatment, except the P group. The only significant difference was between the groups P and P + PHYSIO (p=0.04); that is, there is no significant difference between the P + PHYSIO and AP + PHYSIO or AP groups, all of which improve. The other seven domains (incontinence impact, role limitations, physical limitations, personal relationships, emotions, sleep/energy and severity), Parts II and III did not present significant differences between the groups. In general, except group P in Part I, all groups showed improved quality of life, demonstrating that the application is a good option for treating female UI.

Sjöström et al.[23,24] in 2013 and 2015 conducted a study on women with SUI based on PFMT. Two hundred and fifty volunteers were treated, 124 in the internet group and 126 in the postal group. They also studied quality of life with ICIQ-LUTSQoL. In their study they found short- and long-term significant life quality

Table 3. Secondary outcomes measures baseline and at the 3-month follow-up

Secundary outcomes (KHQ domains)	Treatment group	Baseline (SD)	3-month follow-up (SD)	Difference (SD)	Within group p-value (Wilcoxon)	Between groups p-value (ANOVA)	Different groups (Tukey)
General health perception	AP + PHYSIO	18.42 (16.3)	10.52 (15.2)	-7.89 (11.9)	0.01*	0.01*	P and P + PHYSIO (p=0.008)
	P + PHYSIO	36.27 (22.8)	21.59 (20.8)	-14.77 (16.7)	0.002*		
	AP	27.63 (14.2)	21.05 (17.2)	-6.57 (11.3)	0.03*		
	P	26.47 (28.6)	29.41 (28.3)	2.94 (24.8)	0.66		
Incontinence impact	AP + PHYSIO	47.36 (35.7)	14.03 (25.6)	-33.3 (33.3)	0.002*	0.2	No significant differences
	P + PHYSIO	68.18 (33.3)	39.39 (31.9)	-28.3 (31.4)	0.001*		
	AP	56.13 (33.4)	31.57 (34.2)	-24.6 (21.8)	0.001*		
	P	56.86 (25.7)	43.13 (25.7)	-13.7 (23.7)	0.08		
Role limitations	AP + PHYSIO	30.7 (32.5)	11.4 (17.6)	-19.29 (22.4)	0.005*	0.16	No significant differences
	P + PHYSIO	49.24 (35.4)	25 (30.3)	-24.24 (21.7)	0.0004*		
	AP	32.45 (34)	25.43 (33)	-7.01 (14)	0.09		
	P	54.9 (37.6)	35.29 (31.1)	-19.6 (37.8)	0.05*		
Physical limitations	AP + PHYSIO	36.84 (29.7)	15.79 (14.1)	-21.05 (25.4)	0.004*	0.13	No significant differences
	P + PHYSIO	52.27 (26.9)	27.27 (26)	-25 (23.4)	0.0004*		
	AP	36.84 (32.7)	27.19 (34.3)	-9.64 (19.5)	0.06		
	P	60.78 (38.2)	33.33 (25.7)	-27.44 (30.6)	0.005*		
Social limitations	AP + PHYSIO	22.81 (31.3)	3.22 (8.5)	-19.59 (28.7)	0.014*	0.026*	AP and P + PHYSIO (p=0.04); AP and P (p=0.05)
	P + PHYSIO	39.65 (32.7)	18.18 (25.1)	-21.46 (26.6)	0.003*		
	AP	12.57 (14.7)	11.4 (13.8)	-1.17 (3.5)	0.371		
	P	40.85 (38.4)	18.63 (18.1)	-22.22 (28.4)	0.008*		
Personal relationships	AP + PHYSIO	27.27 (33.6)	6.67 (14.0)	-12.28 (30.3)	0.223	0.071	No significant differences
	P + PHYSIO	56.25 (27.8)	28.12 (36.4)	-20.45 (30.8)	0.014*		
	AP	35.19 (41.2)	27.78 (33.3)	-3.51 (11.9)	0.371		
	P	42.86 (36.2)	23.8 (33.1)	-25.49 (27.7)	0.097		
Emotions	AP + PHYSIO	36.84 (29.4)	24.56 (25.5)	-12.28 (32.1)	0.22	0.13	No significant differences
	P + PHYSIO	60.6 (30.8)	32.32 (29.3)	-28.28 (21.6)	0.00005*		
	AP	27.48 (22.9)	16.37 (21.7)	-11.11 (12.8)	0.007*		
	P	49.01 (40.6)	36.6 (31.4)	-12.41 (37.9)	0.13		
Sleep/Energy	AP + PHYSIO	42.1 (25.7)	18.42 (19.2)	-23.68 (22.4)	0.001*	0.57	No significant differences
	P + PHYSIO	46.21 (26.7)	28.03 (23.8)	-18.18 (17.7)	0.0009*		
	AP	31.57 (24.1)	19.29 (25)	-12.28 (20.7)	0.02*		
	P	49.99 (36.8)	31.37 (25.6)	-18.62 (36.7)	0.09		
Severity	AP + PHYSIO	56.14 (26.1)	25.96 (17.2)	-30.17 (20)	0.0003*	0.48	No significant differences
	P + PHYSIO	66.66 (21.8)	42.72 (30.1)	-23.94 (19.6)	0.0002*		
	AP	45.96 (22.3)	24.56 (26.1)	-21.4 (19.4)	0.001*		
	P	62.35 (29)	42.35 (24)	-19.99 (26.6)	0.011*		
Part I	AP + PHYSIO	32.9 (21.4)	12.3 (14.6)	-20.6 (18.9)	0.001*	0.04*	P and P + PHYSIO (p=0.040)
	P + PHYSIO	52.3 (22.7)	30.5 (22.2)	-21.8 (20.2)	<0.001*		
	AP	41.9 (20.3)	26.3 (22.9)	-15.6 (12.6)	0.001*		
	P	41.7 (24.2)	36.3 (26.1)	-5.4 (21.7)	0.247		

Part II	AP + PHYSIO	36.31 (21.2)	15.77 (10.8)	-14.76 (18.4)	0.0003*	0.108	No significant differences
	P + PHYSIO	51.97 (23.8)	28.51 (23.5)	-22.06 (13.5)	0.0000005*		
	AP	30.84 (19.1)	20.21 (20.8)	-7.62 (9.3)	0.0007*		
	P	50.8 (31.69)	31.63 (21.9)	-17.3 (24.3)	0.003*		
Part III	AP + PHYSIO	9.21 (5.1)	4.26 (4.2)	-4.95 (4.7)	0.001*	0.241	No significant differences
	P + PHYSIO	12.82 (5.5)	7 (6.3)	-5.82 (5.4)	0.0004*		
	AP	8.58 (6.5)	5.63 (5.3)	-2.95 (3.4)	0.002*		
	P	12.94 (7.0)	7.88 (6.0)	-5.06 (4.1)	0.0006*		

*p-value <0,05; KHQ: Kings Health Questionnaire; SD: standard deviation; ANOVA: Analysis of Variance; AP + PHYSIO: application + physiotherapy; P + PHYSIO: home exercise sheet + physiotherapy; AP: only application; P: only home exercise sheet

improvements in both groups, but no differences between them. In our study, we also did not find significant differences between the application and home exercise sheet groups, but we noticed some superiority of the groups with pelvic physiotherapy. In congruence with the literature, we highlight the possibility of using the application as another work tool option with good results, even better when associated with the work of an expert.

Also, in the study of Sjöström et al.[23,24] in 2013 and 2015, the participants were questioned about the use of incontinence absorption materials. They observed a significant reduction in both groups, internet and postal, but with superiority of the Internet group (p=0.02). In this study, the use of cloths was questioned in the "Severity Measures" domain of **KHQ** and all groups showed significant improvement between the pre- and post-intervention periods, without significant differences between the groups.

Asklund et al.[27] performed in 2016 an PFMT-based study of 123 women with SUI, in which 62 received an application and 61 had their treatment postponed. They also studied quality of life with ICIQ-LUTSQoL. As a result, they found a mean score reduction of 4.8 points in the application group and 0.7 in the control group, with the application group being significantly better than the control group. The groups were also significantly different regarding the use of strategies for coping with incontinence (p=0.023).[27] Despite significant differences, the main limitation of the aforementioned study was to not compare the results with other active or proven treatment. In our study, expectations regarding treatment, weight change and self-rated improvement of pelvic floor muscle strength were significantly associated with a successful outcome of SUI treatment with the application aid.[26]

Also, the analysis of **ICIQ-SF** in our study was performed by questioning the participants on how often and how much they lose urine, as well as how this loss interferes with their daily lives. The groups that showed statistically significant difference were AP + PHYSIO, P + PHYSIO and AP. The only group without significant difference was P. A significant difference was observed between the groups P + PHYSIO and P (p=0.01), that is, there is no significant difference between the P + PHYSIO and AP + PHYSIO or AP groups, all of which improve urinary symptoms. We noticed that the application alone or as an aid to the pelvic Physiotherapist is capable of improving women's bladder weakness symptoms, and that only a home exercise sheet with instructions does not promote such improvement.

Sjöström et al.[23] in 2013 obtained as primary results in ICIQ-SF, pre- and post-intervention, mean differences in the internet group of -3.4, and in the postal group of -2.9. After two years of follow-up, Sjöström et.[24] al in 2015 published mean differences in the internet group of -3.5 and the postal group of -3.4, demonstrating that the two programs produce significant improvement in urinary symptoms, but present no significant differences between themselves in the short or long term. In the study by Asklund et al.[27] in 2016, a mean score reduction of -3.9 points was found in the application group and -0.9 in the control group. Bokne et al.[28] in 2019 described mean differences in the internet group of -3.4 and the postal group of -2.6. In our study, similar results were found for the AP group (-3.63).

The ICI recommends supervised training.[4] The positive results obtained in groups with Pelvic Physiotherapy in our study corroborate the evidences, suggesting that the application is a good tool option for incontinent women when guided by a specialist, proving to be as effective as current treatments. In addition, according with the studies published so far, applications are instruments of easy access to the patient and that present a positive cost-benefit ratio.[29,30]

In our study, when used alone, analyzing the differences between groups, the application was significantly inferior to the exercise sheet in just one domain of the KHQ (social limitations).

Moreover, exercise sheet alone showed significantly inferior in the primary outcome the ICIQ-SF, General Health Perception and Part I Score of the KHQ, while the application alone was statistically significant. An adequately powered randomized controlled study comparing the application only with the home

exercise sheet only may shed some more light on any superiority in results between stand-alone intervention. This is important because, so far, about ninety percent of women with UI do not have any supervised treatment. Next to this, the comparison between the application as a stand-alone intervention and the application additional to supervised physiotherapy, again adequately powered, is of great interest related to home maintenance programs and adherence. Finally, we suggest future research with a larger sample in each group and greater segmentation of age of women to investigate whether or not younger populations have better acceptance of using the application.

CONCLUSION

New versions of the iPelvis® Application have been launched and other studies are in progress, with the main objective of improving the applicability and adherence of patients to PFMT.

The application, alone or associated with pelvic physiotherapy, improves urinary symptoms and quality of life for incontinent women, in most variables, but it is not significantly superior in related to the other groups. The groups that underwent physiotherapy presented better results to the groups that underwent only application or only home exercise sheet.

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The creators of the iPelvis® Application were Maura Regina Seleme and Claudia Veloso Mueller. Currently the developers of the new versions are Maura Regina Seleme and Bary Berghmans.

Contributions

Concept: C.E.F.B., R.D.F., M.R.S., Data Collection or Processing: C.E.F.B., R.D.F., M.R.S., Analysis or Interpretation: C.E.F.B., R.D.F., M.R.S., B.B., Literature Search: C.E.F.B., R.D.F., M.R.S., B.B., Writing: C.E.F.B., R.D.F., M.R.S., B.B.

Ethics

Ethics Committee Approval: The research project was approved by the Research Ethics Committee of Faculdade Inspirar, AX – Centro de Estudos da Saúde LTDA. – EPP (opinion number 1,833,987); as well as by the Co-participant Institution, HC-UFPR – Hospital de Clínicas of Federal University of Paraná (opinion number 2,520,073). The study was registered at ClinicalTrials.gov ID: NCT04484753.

Informed Consent: Before performing the evaluation and treatment procedures, each volunteer was informed about the objectives of the study and signed the Informed Consent Form.

Peer-review: Internally peer-reviewed.

DISCLOSURES

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

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REFERENCES

1. DeLancey JOL. The hidden epidemic of pelvic floor dysfunction: Achieavable goals for improved prevention and treatment. American Journal of Obstetrics and Gynecology 2005; 192: 1488-95.
2. Abrams P, Cardozo L, Wagg A, Wein A, eds. Incontinence. 6th International Consultation on Incontinence Tokyo, September 2016, ICS-ICUD 2017, 17-35.
3. Berghmans B. Pelvic Floor Muscle Training: What is Important? A Mini-Review. Obstet Gynecol Int J 2017; 6: 00214.
4. Abrams P, Andersson KE, Birder L, Brubaker L, Cardozo L, Chapple C, Fourth International Consultation on Incontinence. Fourth International Consultation on Incontinence Recommendations of the International Scientific Committee: Evaluation and treatment of urinary incontinence, pelvic organ prolapse, and fecal incontinence. Neurourol Urodyn 2010; 29: 213-40.
5. Dumoulin C, Cacciari LP, Hay-Smith EJC. Pelvic floor muscle training versus no treatment, or inactive control treatments, for urinary incontinence in women. Cochrane Database Syst Rev 2018; 10: CD005654.
6. Lukacz ES, Santiago-Lastra Y, Albo ME, Brubaker L. Urinary Incontinence in Women: A Review. JAMA 2017; 318: 1592-604.
7. Felicíssimo MF, Carneiro MM, Souza ELBL, et al. Fatores limitadores à reabilitação da musculatura do assoalho pélvico em pacientes com incontinência urinária de esforço. Acta Fisiatr 2007; 14: 233-6.
8. Sacomori C. Fatores que influenciam a aderência aos exercícios do assoalho pélvico domiciliares em mulheres adultas com incontinência urinaria. [Dissertação] Universidade do Estado de Santa Catarina - FLORIANÓPOLIS, SC 2014 [Article in Portuguese].
9. Free C, Phillips G, Galli L, et al. The effectiveness of mobile-health technology-based health behaviour change or disease management interventions for health care consumers: a systematic review. PLoS Med 2013; 10: e1001362.
10. Bo K, Berghmans B, Møkved S, van Kampen M, eds. Evidence based physical therapy for the pelvic floor. Bridging science and clinical practice. Edinburgh: Churchill Livingstone Elsevier 2015; 446p.
11. Latorre GFS, de Fraga R, Seleme MR, Mueller CV, Berghmans B. An ideal e-health system for pelvic floor muscle training adherence: Systematic review. Neurourol Urodyn 2019; 38: 63-80.
12. Berghmans B, Seleme M. The '5 F's Concept for Pelvic Floor Muscle Training: From Finding the Pelvic Floor to Functional Use. J Women's Health Dev 2020; 3: 131-4.

13. Berghmans B. Physiotherapy Assessment to Identify The '5 Fs': A Concept of Innovative Pelvic Floor Training. Int J Phys Med Rehabil 2020; 8: 556.

14. Persu C, Chapple CR, Cauni V, Gutue S, Geavlete P. Pelvic Organ Prolapse Quantification System (POP-Q) - a new era in pelvic prolapse staging. J Med Life 2011; 4: 75-81.

15. Palma PCR, Berghmans B, Seleme M, et al. Guia de Prática Clínica da Associação Brasileira de Fisioterapia Pélvica em Urofisioterapia: aplicações clínicas e técnicas fisioterapêuticas nas disfunções miccionais e do assoalho pélvico. 2 ed. São Paulo: AB Editora 2014; 574 p. [Article in Portuguese].

16. Berghmans B, Seleme MR, Bernards ATM. Physiotherapy assessment for female urinary incontinence. Int Urogynecol J 31 2020; 917-31.

17. Tamanini JTN, Dambros M, D'ancona CAL, Palma PCR, Netto Jr, Rodrigues N. Validation of the "International Consultation on Incontinence Questionnaire – Short Form" (ICIQ-SF) for portuguese. J Public Health 2004; 38: 438-44.

18. Tamanini JTN, Dambros M, D'ancona CAL, et al. Responsiveness to the Portuguese version of the International Consultation on Incontinence Questionnaire-Short Form (ICIQ-SF) after stress urinary incontinence surgery. Int Braz J Urol 2005; 31: 482-9.

19. Tamanini JTN, D'Ancona CAL, Botega NJ, Netto Jr NR. Validação do "King´s Health Questionnaire" para o português em mulheres com incontinência urinária. Rev Saúde Pública 2003; 37(2):150-58. [Article in Portuguese].

20. Bo K. Effect of antenatal pelvic floor muscle training on labor and birth. Obstet Gynecol, United States 2009; 113: 1279-84.

21. Riccetto CLZ, Palma P, Herrmann V, et al. Is there correlation between urodynamic findings and International Consultation on Incontinence Questionnaire - Short Form (ICIQ-SF) Score? J Urol 2005; 173: 357.

22. Hebbar S, Pandey H, Chawla A. Understanding King's Health Questionnaire (KHQ) in assessment of female urinary incontinence. Int J Res Med Sci 2015; 3: 531-8.

23. Sjöström M, Umefjord G, Stenlund H, Carlbring P, Andersson G, Samuelsson E. Internet-based treatment of stress urinary incontinence: a randomised controlled study with focus on pelvic floor muscle training. BJU Int 2013; 112: 362-72.

24. Sjöström M, Umefjord G, Stenlund H, Carlbring P, Andersson G, Samuelsson E. Internet-based treatment of stress urinary incontinence: 1- and 2-year results of a randomized controlled trial with a focus on pelvic floor muscle training. BJU Int 2015; 116: 955-64.

25. Henderson JW, Wang S, Egger MJ, Masters M, Nygaard I. Can women correctly contract their pelvic floor muscles without formal instruction?. Female Pelvic Med Reconstr Surg 2013; 19: 8-12.

26. Nyström E, Asklund I, Sjöström M, et al. Treatment of stress urinary incontinence with a mobile app: factors associated with success. Int Urogynecol J 2018; 29, 1325-33.

27. Asklund I, Nyström E, Sjöström M, Umefjord G, Stenlund H, Samuelsson E. Mobile app for treatment of stress urinary incontinence: a randomized controlled trial. Neurourol Urodyn 2017; 36: 1369-76.

28. Bokne K, Sjöström M, Samuelsson E. Self-management of stress urinary incontinence: effectiveness of two treatment programmes focused on pelvic floor muscle training, one booklet and one Internet-based. Scand J Prim Health Care 2019; 37: 380-7.

29. Sjöström M, Umefjord G, Lindholm L, Samuelsson E. Cost-effectiveness of an Internet-based treatment program for stress urinary incontinence. Neurourol Urodyn 2015; 34: 244-50.

30. Sjöström M, Lindholm L, Samuelsson E. Mobile App for Treatment of Stress Urinary Incontinence: A Cost-Effectiveness Analysis. J Med Internet Res 2017; 19: e154.

Editor's comment

Applications are now introduced to help with the management of heath issues. They have a special role, mainly in these times of periodic lock-downs, making the regular appintments with health care providers difficult to resume. The significance of the treatment of urinary incontinence is known to the readers of this Journal. Still, we asked Dr. Andrea Ambrosetti of the Centro Italiano Studio e Sviluppo Psicoterapia a Breve Termine (CISSPAT) Associazione Italiana Sessuologia e Psicologia Applicata (AISPA), to write a discussion elaborating on that issue.

Professor Jacob Bornstein,
Editor in Chief, Pelviperineology

Invited Discussion

Given the continuous development of the interaction between human relations and the smartphone, the aim of this study is to verify how an app could become a valid tool for the treatment of the urinary incontinence.

The pelvic floor is a body district composed of muscles and ligaments. It is located at the base of the abdominopelvic cavity and it performs different functions such as urination, defecation, reproduction, and sexual satisfaction. The malfunction of this area may lead to a condition that can affect everyday life and sexuality.

The pelvic floor is not just a muscle. On the contrary, it is an active part of the human body for the whole life of every human being. The proposed article underlines an important problem: the urinary

incontinence in a relevant sample of women between 18 and 59 years old. The purposes of this study were to analyze the efficacy of an application of exercises of the pelvic floor, both alone and associated with physiotherapy, in reducing the urinary symptoms in order to improve the life quality of incontinent women.

The results of this study show that the intervention of a professional of the pelvic floor combined with the use of the app improve the lifestyle and quality of life patients. The pelvic floor is not only an organ or a function but it should be considered also from a psychological and sexological point of view.

The pelvic floor is an intimate part of the body, usually protected, often "forgotten" and untreated, sometimes even unknown. An intimate part of the body invested with symbolical meanings according to the relationship with ourselves, the self-perception, the attributed meanings, the beliefs, the relationship with the other, the culture, the religion, the experiences that may modify the psychological convictions and the events, for example, what happened, the personal life, or the symptom.

Avoiding the problem from the beginning may worsen the physical condition with an aggravation of the psychological state. The urinary incontinence influences in a decisive way all aspects of a woman's life and for this reason, her self-esteem may be affected over time, by activating feelings of inadequacy.

The concern with one's own incontinence may impact different day-to-day moments of everyday life. Even at work, a woman may be constantly worried about frequently going to the toilet or checking her clothes in case of leakages. Moreover, the quality of sleep and night rest may be compromised by the need of getting up frequently to urinate, affecting the wakefulness in the daytime.

Urinary incontinence and sexuality may be closely related: those who suffer from leakage during sexual intercourses may experience shame, enough in some cases to renounce this important part of the couple's relationship. It may happen that leakages may occur during sexual intercourses due to pressure on the bladder, with fear of judgment. Places without a restroom, the idea of not being able to get changed and the fear of external judgment may create strategies of avoidance in the social environment, preventing people from going out and from spending time with other people, out of shame of letting the others know their difficulty.

The lack of a correct diagnosis made by specialized doctors, the tendency to underestimate the pathological status, for example, believing that the situation will heal on its own, like with a regular delivery, the coexistence with the pathology and the delay of its treatment due to a lack of resources may worsen the disfunction or make it chronic.

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Interstitial cystitis symptoms as defined are indistinguishable from posterior fornix syndrome symptoms cured by uterosacral ligament repair

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ABSTRACT

Objective: To test our hypothesis that female interstitial cystitis (IC) and Posterior Fornix Syndrome (PFS) are one and the same.

Materials and Methods: A retrospective study. We extracted raw CPP data from patients who had TFS surgery for cure of uterine/apical prolapse, along with bladder, pad and urodynamic data. We critically compared known IC phenotypes with PFS symptoms to check our hypothesis for truth or falsity. We used a validated Integral Theory System Questionnaire (ITSQ), "simulated operations" testing with the speculum test (which reduces urge and pain if USLs are weak), transperineal ultrasound, urodynamics, surgery and post-operative urodynamics.

Results: Forty-six patients with CPP had 157 urinary symptoms. The cure rate was CPP 76%, urge incontinence 74%, abnormal emptying/retention 80%, nocturia 75%, frequency 50%.

Conclusion: PFS data accord exactly with the ICS definitions for IC, except that PFS patients were cured or improved by USL repair, IC patients, not. Further testing of our hypothesis will require wide-ranging testing with the ITSQ (which diagnoses both PFS and IC), and the simulated operations "speculum tests" to confirm that USL weakness is indeed the cause.

Keywords: Interstital cystitis; posterior fornix syndrome; chronic pelvic pain; uterosacral ligaments; urge incontinence; nocturia; urinary retention

INTRODUCTION

Recently, the first surgical cure Interstitial Cystitis (IC) of histologically validated Hunner's ulcer by Tissue Fixation System (TFS) minisling repair of cardinal/uterosacral ligaments was reported,[1] except that cure occurred by using the diagnostic and surgical protocols of another paradigm, the Posterior Fornix Syndrome,[2] itself part of the Integral Theory of Female Urinary Incontinence.[3] This discovery is potentially transformative, because, if further evidence can be provided that the bladder and pain symptoms which comprise IC according to it definitions, IC, a condition which afflicts 8%–10% of women, may become generally curable by uterosacral ligament (USL) repair, a fairly minimal procedure.[1,4]

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Though only a singular case,[1] the IC cure was extremely well validated. Symptoms consistent with IC were recorded from a validated questionnaire.[2] The diagnosis of Hunner's ulcer was histologically confirmed, with findings of large numbers of mast cells within the muscular layer. Glomerulations were noted on bladder filling. Pre- and post-operative urodynamics showed increased urodynamic bladder capacity, from 300 ml to 700 ml. Repeat cystoscopy following cure showed no evidence of Hunner's ulcer.

PFS was first described in 1993.[2] It comprises symptoms of chronic pelvic pain (CPP), frequency, nocturia, urgency, abnormal emptying/retention, caused by USL laxity, and cured or improved by USL repair, either native USL plication,[2] or posterior sling.[1,4]

The authors,[1] did not set out to cure IC. They followed the standard protocol for diagnosis and management of the PFS. They concluded that IC may be a manifestation (or phenotype) of PFS.[1]

IC, renamed "Bladder Pain Syndrome" (BPS) by ESSIC, the International Society for the Study of BPS, is a debilitating condition, said to occur in up to 8%–10% of women of all ages.

The current International Continence Society (ICS) definition for IC/BPS[5] is *"persistent or recurrent CPP, pressure, or discomfort perceived to be related to the urinary bladder, accompanied by at least one other urinary symptom such as an urgent need to void or urinary frequency diagnosed in the absence of any identifiable pathology which could explain these symptoms."* Hunner's lesion, and glomerulations, previously hallmarks of IC, are no longer considered essential for diagnosis.[5]

Even at ESSIC, there is considerable debate about the very concept of IC. In 2018, Jane Meijink,[6] president of the Interstitial Cystitis society ESSIC stated *"With all patients being bundled together as one 'syndrome,' with no subtyping, study data have been unreliable and even meaningless. This is further exacerbated by the fact that there is still no international consensus on nomenclature, terminology, and definitions nor on diagnostic criteria. This is damaging for research, data sharing, and comparing and ultimately for the patient and treatment since the evidence from drug studies is erratic and inconsistent."*

Butrick,[7] in his classical paper of IC manifestations in accord with ICS definitions, sounded a note of caution as regards the broadening of the definition of IC, as it may have led to reports of higher IC prevalence. Butrick commented,[7] *"Some authors reported the prevalence to be as high as 26%. It is clear that BPS/IC represents a spectrum of bladder and pelvic pain disorders that are quite heterogeneous, yet many clinicians consider this higher symptom-based prevalence to be excessive and worry that the new diagnosis is overly inclusive. Patients who are ultimately diagnosed to have PBS/IC may present with various chief complaints such as recurrent bladder infections, dyspareunia, urinary frequency, or CPP. A bladder component of pain is found in 38%–85% of women with CPP. Patients with BPS/IC often have more than one pain generator and can present with chief complaints not related to the bladder at all. They often report multiple pain syndromes that involve not only the pelvis but also other parts"*

Butrick[7] described 408 patients based on ICS definitions,[6] who presented to a referral urogynecologic pain centre with varied pelvic floor disorders, Table 1, who, after an initial evaluation, were diagnosed as BPS/IC based on ICS symptom-based definitions and diagnostic criteria.[5]

Table 1. Interstitial cystitis (n=408) specific complaints

Bladder installations: improvement; no cure
Bladder pain/interstitial cystitis (n=157)
Chronic pelvic pain (n=98)
Vulvodynia dysfunction (n=70)
Dyspareunia (n=54)
SUI (n=24)
POP (n=21)
Hunner's ulcer (n=18)

SUI: stress urinary incontinence; POP: pelvic organ prolapse; n: Number

Following publication of the IC cure,[1] we extracted raw data, patients who had CPP, from a previous surgical study which used the TFS tensioned mini-sling to cure pelvic organ prolapse. Our aim was to answer the research question, are PFS and IC similar conditions? If they are, patients with IC *who have a positive speculum test* are at least theoretically, potentially curable by USL repair?'

MATERIALS AND METHODS

A retrospective study. We examined data from 46 women who had CPP and USL repair using the TFS for prolapse repair. Surgery was performed by both surgeons PP and PR either at Royal Perth Hospital or Bentley Hospital Perth, WA between 2004 and 2009.

Patient characteristics: the median age of the women was 66 years (range: 35–87) Median parity was 3 (range: 0–8), and median weight was 70 kg (range: 38–117 kg). Of the 46 patients, 18 had only 1st degree prolapse, and 28 had 2nd degree or greater prolapse. The mean number of previous incontinence or vaginal repair operations was 1.6 per patient (range: 0–6 operations). Only 12 patients had not had prior incontinence or vaginal surgery; 25 patients (50.4%) had undergone prior hysterectomy.

Pre-operative assessment

All patients completed a 24-hour urinary diary, and a self-administered Integral Theory System Questionnaire (ITSQ),[8] which was completed at home in the patient's own time. Pre-operatively, a speculum test, Figure 1, and 24-hour pad test were also administered. Symptoms derived from the questionnaire were ticked off in a diagnostic algorithm, Figure 2. The algorithm visually condenses the diagnosis from the ITSQ as regards ligament and fascial damage to three zones of the vagina, anterior, middle or posterior. In this classification. anterior zone extends from external urethral meatus to bladder neck, middle zone from bladder neck to cervix or hysterectomy scar, posterior zone from apex to perineal body.

Inclusion/exclusion criteria

Patients who had stress urinary incontinence (SUI) were excluded from the study.

Inclusion criterion for surgery were patients with typical PFS symptoms as per the red rectangle, Figure 2, symptoms of urgency, frequency, nocturia, abnormal emptying, urinary retention, pelvic pain, and who, like Scheffler's study, had a positive speculum test for urge or pain.

Objective assessment

Using the symptom-based algorithm as a guide, the three zones of the vagina were examined for evidence of ligament or fascial damage; anterior zone: SUI; middle zone, evidence of high cystocele (cardinal ligament damage) central cystocele, perineal body, posterior vaginal wall (rectovaginal fascia), and for the degree of prolapse of the vaginal vault or uterus. (USL damage). Transperineal ultrasound was performed with a Toshiba 3.5 Mhz curvilinear probe to check organ position at rest and straining and to confirm clinical diagnosis of prolapse. Urodynamic testing was performed with Gaeltec microtransducers to assess for maximal urethral closure pressure, flow, residual urine, and "overactive bladder". The patients were monitored at 6 weeks, and at 3 monthly intervals thereafter using a 24-hour urinary diary, structured self-administered questionnaire, cough stress test, 24-hour pad test, transperineal ultrasound, and urodynamic investigation. Residual urine was assessed by catheter after the patient had voided.

Statistical analysis

We applied the McNemar x^2-tests to test for significance changes in the symptoms' incidence-frequency from baseline (preoperative) to the postoperative phase. For each symptom the null hypothesis H_0: P(baseline) = P(12 months after surgery) versus H_1: P(baseline) =P(12months after surgery) was tested, with P indicating prevalence or incidence rate. An $\alpha = 0.05$ was accepted as the nominal level of significance. Because of multiple testing the p-values of the tests were compared to a Bonferroni corrected α (say α^*) for keeping the type I error less or equal to 0.05.

Ethics

This is a retrospective study of previous surgical data. There are no Ethics Committee issues.

Surgical technique

The posterior TFS sling consists of two polypropylene soft tissue anchors through which is inserted an adjustable polypropylene tape. A transverse full thickness vaginal incision was made 1 cm below the hysterectomy scar, or cervical ring. The uterosacral ligament was identified by Allis forceps. A 4-5 cm channel was dissected immediately lateral to the uterosacral ligaments. Using a special applicator, the TFS anchors were inserted into the uterosacral ligaments. The tape was tightened via a one-way system at the anchor base, and this reduced the apical/ uterine prolapse to a normal anatomical position.

The criteria for symptomatic improvement: nocturia: change in patients having >2 episodes per night; abnormal emptying: self-assessed improvement >80%, using a 0–100 Visual Analogue Scale (VAS) expressed as a percentage; urge incontinence: change in the number of times a patient wet per day; frequency: change in patients having >8 episodes per day; pelvic pain and fecal incontinence: self-assessed improvement >80%, also a 0–100 VAS scale; otherwise objective measures such as pad weights and residual urine measured by catheter were used.

RESULTS

Forty-six patients with CPP had 157 urinary symptoms, urge incontinence, frequency, nocturia, abnormal emptying/ retention, Table 2 This equates to three urinary symptoms per pain symptom, fulfilling the ICS diagnosis for IC in all 46 patients in this study. Mean hospital stay was 1.5 days (1–3 days). One patient was lost to follow-up.

Mean post-operative review time was 12 months. The operation results are summarized in Table 2. There was one recurrence of prolapse which needed re-operation.

CPP: of 46 women with CPP, 35 (76%) reported >80% improvement of their pain and 3 (7%), >50% improvement.

Nocturia: there was a reduction from 168 episodes to 61 episodes. Based on ICS definition, less than 2 episodes/night, cure rate was 27/36 (75%).

Table 2. Symptom outcome (n=46)						
		Symptom change with surgery			% cure in brackets	
	Fecal incontinence (n=12)	Frequency >8/day (n=45)	Nocturia >2/night (n=40)	Urge incontinence >2/day (n=33)	Abnormal emptying (n=39)	Pelvic pain (n=46)
%	100	50	75	74	80	76
p	<0.005	0.05	0.005	0.005	0.005	0.005
n: number						

Urge incontinence: the number of women who had urge incontinence episodes reduced from 33 to 9. Cure rate was 74%. The total number of episodes (getting up at night counting even once per night) within these women reduced from 86 to 17.

Frequency: based on ICS definitions, frequency being >8/night, reduced from 27 women to 14 (50%). The total reduction in episodes in the 27 women was from 385 to 125/day.

Fecal incontinence (FI): a total of 12 women had FI. Nine were 100% cured and 3>80% cured.

Severe urge incontinence (UI): was present in 17 women, mean loss 215gm/24-hour pad test (range: 20 gm–644 gm). At 6 weeks review, 10/17 patients said they were >95% cured. Their mean 24-hour urine loss was 6 gm (range: 0–12gm). The results were statistically significant (p=<0.05) (Student's 2 tailed t-test). In the other 7/17 patients from this group with OAB who were not considered cured of their symptoms, their mean 24-hour loss reduced from 320 gm (range: 25–388 gm) to 223 gm (range: 35–720 gm). Of these seven patients, four were cured of nocturia, three of daytime frequency, and six of other symptoms such as fecal incontinence, pelvic pain and abnormal emptying.

Abnormal emptying and residual urine: symptomatically, 23/39 patients reported more than 80% improvement, with another 11 reporting more than 50% improvement. A residual urine >50 ml (50–600 ml) was seen in 29 patients. In this group, the mean reduction of residual urine was from 271 ml preoperatively to 53 ml post-operatively (p=0.005). One patient who self-catheterized four times daily pre-operatively, was restored to normal emptying. Mean emptying time for this group (n=29) decreased from mean 41 seconds (12–130 secs) to 31 seconds (7–130 secs) (p=0.005). There was no significant change in peak flow, (41 ml/sec pre-operatively to 37 ml/sec post-operatively).

Urodynamics: forty-six patients underwent urodynamic testing preoperatively, and 43 postoperatively. Two glomerulations were noted, but no Hunner's ulcers. There were eight cases of "overactive bladder" (OAB) as defined by the International Continence Society associated with significant urine loss. Five of these eight patients considered themselves more than 95% cured, and a sixth more than 80% cured (mean change from 288 gm–17 gm). Two reported no improvement at all.

Prolapse repair: mean post-operative review time was 12 months. The operation results are summarized in Table 2. There was one recurrence of prolapse which required reoperation.

DISCUSSION

Forty-six patients with CPP had 157 urinary symptoms, urge incontinence, frequency, nocturia, abnormal emptying/retention, all PFS symptoms, Table 2. This result equates to three bladder symptoms per pain symptom and therefore, consistent with the ICS definition for IC.[5] Our pain data comprises many different phenotypes of CPP, including vulvodynia, paraurethral pain, and those specified in the 1996 paper attributing CPP to originating from visceral plexuses,[9] lower abdominal pain, contact dyspareunia, coccygeal pain, paraurethral pain. Whereas it is possible to attribute symptom cure in Table 2 as a direct result of ligament reinforcement by a TFS USL sling, the pathway for symptom occurrence in Tables 1 and 2, needs to be elucidated more scientifically, in order to prove that IC as defined,[5] and PFS[2] are one and the same condition.

Normal bladder function for retention, evacuation and urge control[3,10-19]According to Petros and Ulmsten[3] and Petros and Bush[10-19], the key to understanding dysfunctions of closure (stress incontinence), urge control for OAB and emptying dysfunction known as "retention", "underactive bladder" (UAB), is the interaction of three the reflex opposite directional forces and the ligaments pubourethral (PUL) and uterosacral (USL) which they contract against. Note the opposite muscle force action of the reflex muscles in Figure 3 and Video 1: US three directional forces "https://www.youtube.com/watch?v=3vJx2OvUYe0"

During micturition, m. pubococcygeus (PCM) relaxes (broken circle, Figure 3). Levator plate (LP) and conjoint longitudinal muscle of the anus (LMA) contract against USL to actively open out the urethra (broken white lines).[10-19] This action exponentially reduces the urethral resistance to flow inversely by the 4th power of the radius (Poiseuille's Law), Figures 4 and 5.

Video 2 micturition shows active opening of the posterior

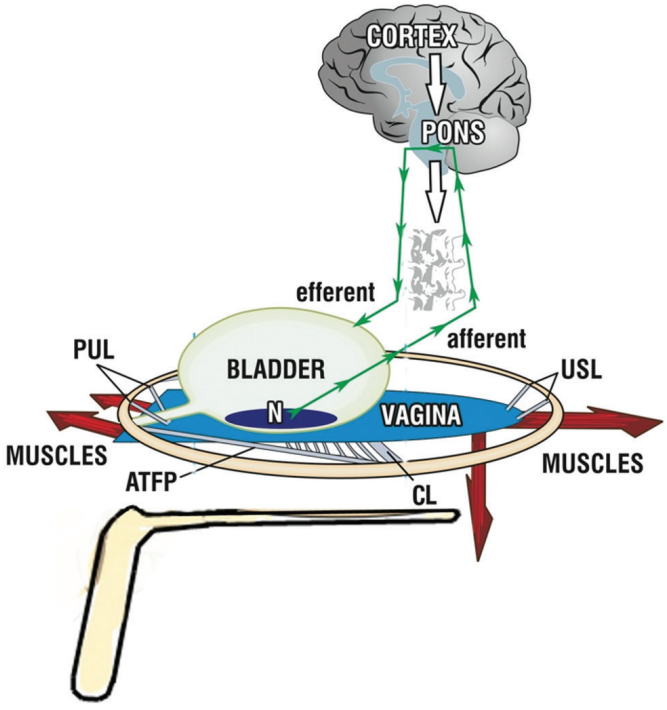

Figure 1. Speculum test. This is a 3D view of the bladder sitting on the anterior vaginal wall. The vagina is suspended from the pelvic brim by ligaments, pubourethral (PUL), cardinal (CL) and uterosacral USL. A speculum inserted into the posterior fornix mechanically supports lax USLs and the nerve plexuses S2-4, T11-L2 supported by USLs. Firm USLs restore the contractile strength of the posterior muscle forces (arrows) which contract against them. These now tension the vagina like a trampoline to support the stretch receptors "N", decreasing the afferent impulses which on passing a critical mass, are interpreted as urgency.
USL: uterosacral

urethral wall by the posterior muscle forces. Note how the anterior lip of LP is pulled down to open the posterior urethral wall. Micturition Video 2 "

https://www.youtube.com/watch?v=eiF4G1mk6EA&feature=youtu.be"

If USLs are weak, LP/LMA contractile force weakens, the posterior urethral wall is not optimally opened out, and the detrusor has to contract against a partially unopened urethra. The detrusor may not empty adequately (retention). The patient experiences this as "obstructed micturition", inability to evacuate, "stopping and starting" etc.

In this next section, specific data from Butrick's[7] classical IC study are compared to data from this work. The pathogenesis detailed is based on original scientific studies.

Urinary retention

Butrick[7] Urinary retention and/or voiding dysfunction was reported by 70% of his cohort, and urodynamics demonstrated

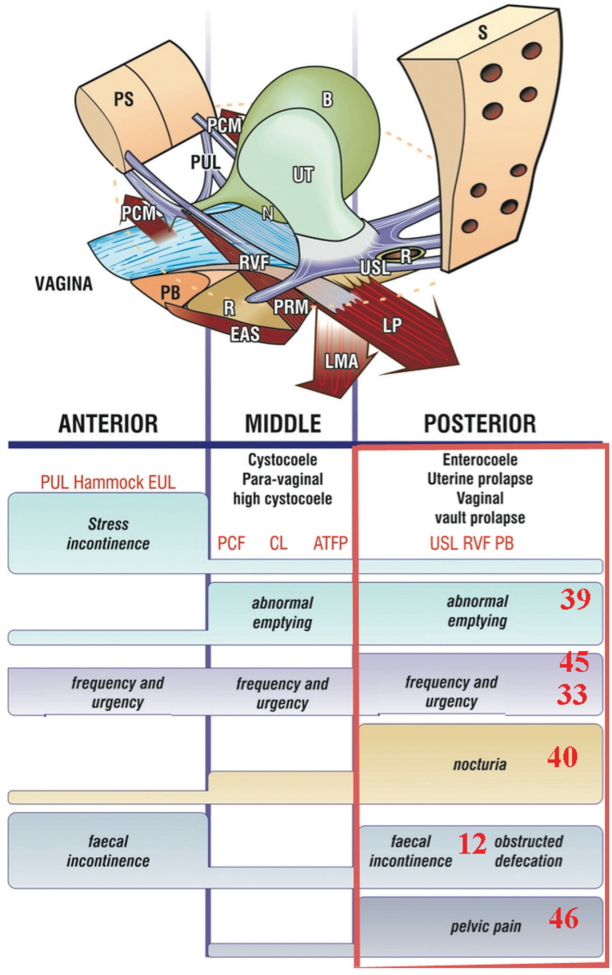

Figure 2. The pictorial diagnostic algorithm. Symptoms indicate which ligaments are damaged
The numbers in red indicate the number of symptoms co-occurring with the 46 symptoms of chronic pelvic pain to confirm the diagnosis of USL damage. The three reflex muscle forces (arrows) tension the organs and the vagina to support urothelial stretch receptors "N".
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.
Anterior zone: external meatus to bladder neck PUL: pubourethral ligament; hammock: suburethral vagina; EUL: external urethral ligament
Middle zone: bladder neck to anterior cervical ring. CL: cardinal ligament; PCF: pubocervical fascia; ATFP: arcus tendinous fascia pelvis.
Posterior zone posterior cervical ring to perineal body (PB): USL uterosacral ligaments; RVF: rectovaginal fascia; PB. The rectangle indicates the symptoms associated with USL laxity and the posterior fornix syndrome.
Chronic pelvic pain and nocturia are uniquely caused by uterosacral (USL) ligament laxity.

voiding dysfunction in 80%. Butrick[7] also reported women with recurrent urinary tract infections, a known consequence of inability to empty.

This study, Table 1 Following USL repair, in women with raised residual urine (>50 ml), there was a mean reduction from 271 ml to 53 ml (p=0.005). One patient who self-catheterized 4 times

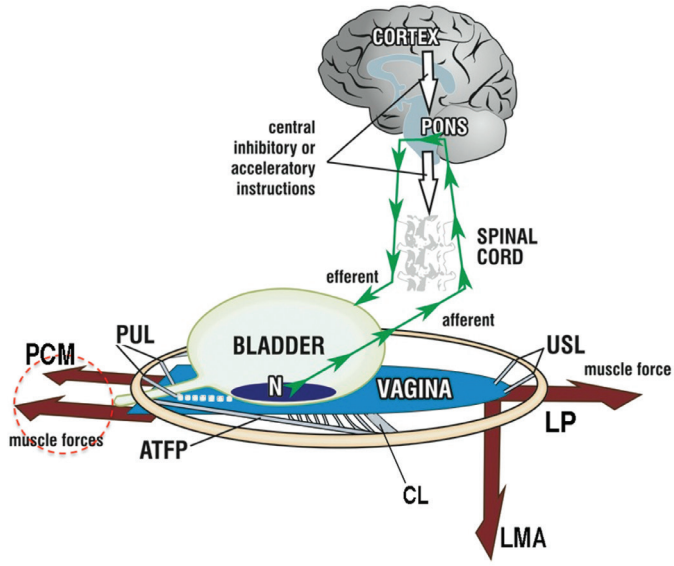

Figure 3. Control of bladder is binary. Schematic 3D sagittal view. System in normal closed mode.

Cortical control: In a woman with no bladder dysfunction, the afferent impulses from the stimulated stretch receptors 'N' are reflexly suppressed cortically (white arrows). When the bladder requires to evacuate, the cortex activates the micturition reflex.

Peripheral control is by a cortically controlled musculo-elastic mechanism which responds to cortical efferents output (small arrows).

The three directional muscles (large arrows), forward, pubococcygeus muscle "PCM ", backward, levator plate" LP", and downward, conjoint longitudinal muscle of the anus "LMA" contract against the supporting ligaments, PUL (pubourethral) and USL (uterosacral), to stretch vagina tightly, much like the membrane of a drum. The stretched vagina supports the pressure of the urine column. This prevents activation of the stretch receptors "N", thereby decreasing afferent impulses to the cortex.

Micturition: When convenient to empty, central control (white arrows) relaxes, as does the forward contractile force of PCM (broken circle); this relaxation allows the posterior muscles LP and LMA to unrestrictedly open out the posterior wall of urethra (white broken lines) immediately prior to bladder evacuation by global detrusor muscle contraction (actually spasm- see micturition video). CX: cervix; CL: cardinal ligament; ATFP: arcus tendineus fascia pelvis.

Dysfunction If there is weakness in the reflex muscles PCM, LP, LMA and/or the ligaments they contract against, PUL, USL, the peripheral control mechanism cannot adequately close the urethra (incontinence), open it to empty (obstructed micturition) or control micturition by bilateral stretching of vagina by the three opposite muscle forces to support "N" (urge incontinence).

daily pre-operatively, was restored to normal emptying. Mean emptying time (n=29) decreased from mean 41 seconds (12–130 secs) to 31 seconds (7-130 secs) (p=0.005).

Cure of retention: The TFS (or other) tape irritates the tissues to create new collagen to repair the damaged USL [20–30], Table 2, restore the contractile force of LP/LMA, to open the urethra and restore urine flow.

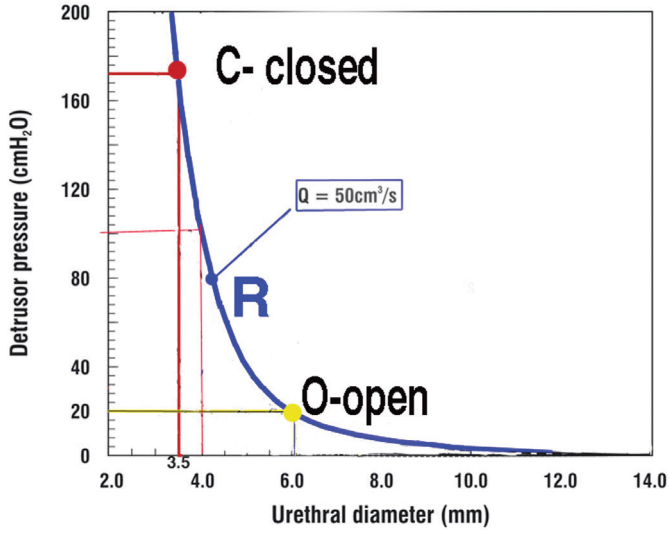

Figure 4. Urine flow is inversely and exponentially related to urethral radius. For a flow rate of 50 ml/sec (thick blue line), opening the urethral diameter from 3.5 mm to 4 mm reduces the head of pressure required by the detrusor to expel urine from the bladder from 172 to 100 cm water. Expanding to 6 mm (yellow lines), reduces the head of pressure to 20 cm water. The blue line represents the total urethral resistance to flow, which is composed of dynamic and frictional flow components.

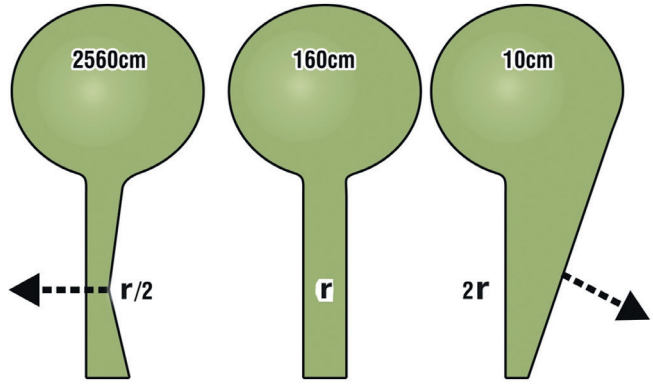

Figure 5. Effect of the external striated muscle mechanism (arrows) on urethral diameter during closure (r/2 left) and opening (2r right) exponentially alters urethral resistance and therefore head of pressure required for the detrusor to drive the urine through the changed diameter. Pressures indicated are based on a nominal pressure of 160 cm for radius 'r'; calculated pressure apply the 4th power law of Poiseuille. Halving the radius (r/2) increases the expulsion force required by a factor of 16, to 2,560 cm H_2O. Doubling the radius (2r), decreases that force by a factor of 16 to 10 cm H_2O.

Retention If we take the front arrow to represent overcompensation by the distal urethral closure mechanism in IC or PFS patients who have loose USLs, very little extra activity is required to close the urethra sufficiently to make it impassable to urine flow (retention). After Petros PE, The Female Pelvic Floor, 3rd Ed Springer, 2010.

Pathogenesis of urge and nocturia ("OAB")[9-30] with reference to Figure 3, Weakness in the muscles PCM, LP, LMA and/

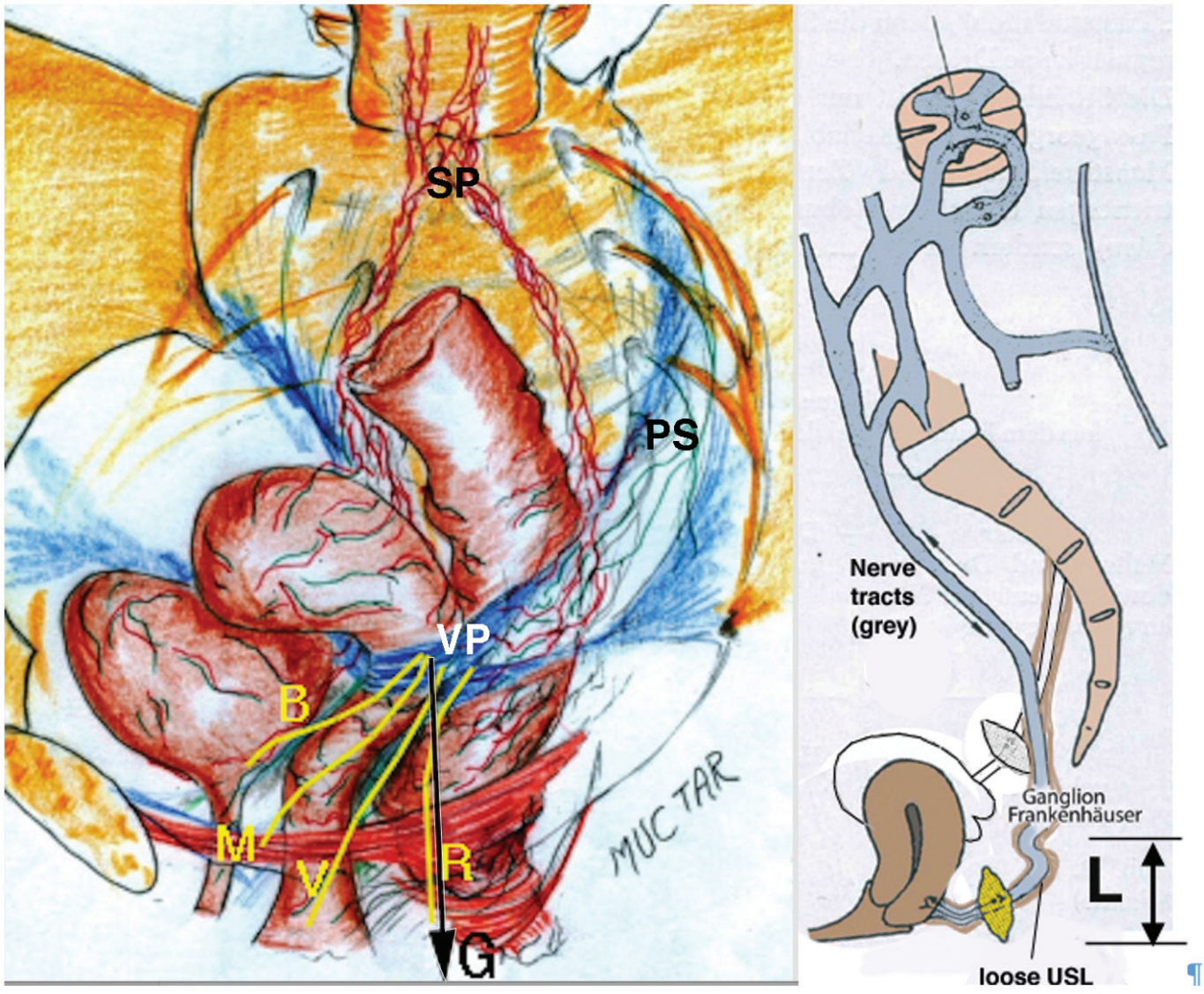

Figure 6. Pathogenesis of chronic pelvic pain from unsupported visceral plexuses
Left figure: Visceral plexus (VP) with components SP (sympathetic T11-L2) and parasympathetic (S2-4). Endorgan afferent nerves M (muscles), B (bladder), V (vagina), R (rectum) travel to VP which serves as a type of relay junction. G signifies forces of gravity acting on these nerves in the upright position.
Right figure: Ganglion Frankenhauser (sympathetic) supported by USLs (uterosacral ligaments). Laxity 'L' in USLs means they no longer can support VPs, so they can be stimulated by 'G'. Arrows upwards: afferent impulses from VP to brain. Right figure by permission, Muctar. S. Left figure after Martius H.
Video 1. https://www.youtube.com/watch?v=3vJx2OvUYe0
Video 2. https://www.youtube.com/watch?v=eiF4G1mk6EA&feature=youtu.be

or the ligaments they contract against, PUL, USL, will affect the ability of the three opposite muscle forces to support the stretch receptors "N" from below. These send increased afferent impulses to the micturition centre. If the impulses cannot be suppressed cortically, the micturition reflex may be activated. The cortex perceives this as urge incontinence. If this occurs at night, the patient may be woken with a need to empty her bladder. Another characteristic of "OAB" is a smaller bladder capacity. After USL repair, Scheffler increased bladder capacity from 300 ml to 700 ml.

Cure of OAB/nocturia: The TFS (or other) tape irritates the tissues to create new collagen to repair the damaged USL,[18-30] Table 2, restore the contractile force of LP/LMA, restore stretching of the vagina to support stretch receptors "N" from below, to decrease afferent impulses to the micturition centre.

Pathogenesis of chronic pelvic pain, Figure 6 The hypothesis of lax USLs causing CPP was first described in the German literature in 1938 by Heinrich Martius[31] and in the English literature by Peter Petros[9] in 1996.

With reference to Figure 6 (left) the visceral plexus (VP) comprises component nerves SP (sympathetic T11-L2) and parasympathetic

(S2-4). VPs are mechanically supported by USLs at their lower end. The yellow lines represent the afferent and efferent visceral nerves from the endogans, muscles, bladder, vagina, rectum. The afferent nerves proceed to the visceral plexus "VP", fig6, which is best regarded as a type of nerve junction. If USLs are loose, the force of gravity of muscle movements may stimulate afferent axons from specific end organs, often more than one at a time. (This explains co-occurrence of several sites of CPP). The afferents pass up to the brain which (falsely) interprets them as an injury (pain) coming from the particular end organ(s). As well as pain, in both vulvodynia and IC, actual inflammatory cells may be seen at the end organs themselves, mast cells, T cells etc. This can be explained by the brain sending efferent signals via the visceral nerves to the end organ, to activate resident "sleeper cells" to produce mast and other inflammatory cells, even the neuroproliferations sometimes seen on biopsy in vulvodynia and in Hunner's ulcer. One could postulate that an ulcerated Hunner's ulcer could be an extreme manifestation of this inflammatory reaction perhaps aided by ischaemia from cutting off low pressure venous drainage by a full bladder descending as a cyctocele. The latter was the explanation accorded for glomerulations.[1] Scheffler cured both Hunner's ulcer, and glomerulations with TFS sling ligament repair.[1]

Cure CPP: With reference to Figure 6, the tape creates mechanical support which prevents stimulation of the nerves within "VP", Table 2. The speculum test works by mechanically supporting USLs. This support temporarily restores the structural integrity of USL to decrease "pain" afferents and likewise "urge" afferents by supporting the urothelial receptors "N", Figure 3.

Muscle spasm and/or pain

With reference to Figure 3, the forward and backward muscle forces are balanced and equal. If USLs are weak, the striated muscles which contract against them also weaken. The system becomes unbalanced. The forward muscles forces (m. pubococcygeus) may over-react to close the urethra excessively. This explains the high urethral pressure described by Butrick[7] More importantly, the excessive contraction may cause pain and spasm to explain the "myofascial pain and hypertonic pelvic floor dysfunction with BPS/IC, well, over 70%." described by Butrick[7]. However, Wu et al.[32] reported immediate improvement in paraurethral pain with the speculum test and the first author (P.P.) after successful posterior sling surgery. At this stage such spasm and pain relief remains a hypothesis to be further tested.

CONCLUSION

We were to show that, at least as regards the ICS definition, IC and PFS are substantially one and the same condition. The descriptions of Butrick regarding the different manifestations of pain and bladder dysfunctions, bear a striking similarity to the PFS as originally described[2] is part of the 1993 Integral Theory.[3] Direct proof of CPP, OAB, nocturia, retention, fecal incontinence cure by a posterior sling to reinforce USL as described, Table 2, must be accepted as proof of USL causation. We were largely able to explain the pathogenesis of these conditions by reference to several of the basic science discoveries of the Theory, [3,33] and the basis for ligament surgery, creation of an artificial collagenous ligament to explain dysfunction and return of function.[34]

Contributions

Surgery and data: P.P., P.R., Concept: P.P., P.R., Analysis of data: P.P., Writing: P.P., P.R.

Ethics

Ethics Committee Approval: This is a retrospective study of previous surgical data. There are no ethics committee issues.

Informed Consent: Written consent was obtained from all patients.

Peer-review: Externally 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.

REFERENCES

1. Scheffler KU, Oliver W, Hakenberg OW, Petros PE. Cure of interstitial cystitis and non-ulcerating Hunner's Ulcer by cardinal/uterosacral ligament repair. Urol Int 2021; 105: 920-3.Petros PE, Ulmsten U. The posterior fornix syndrome: a multiple symptom complex of pelvic pain and abnormal urinary symptoms deriving from laxity in the posterior fornix. Scandinavian Journal of Urology and Nephrology, 1993; 27(Supp)153: 89-93.
2. Petros PE, Ulmsten U. An Integral Theory and its Method, for the Diagnosis and Management of female urinary incontinence. Scand J Urol Nephrol 1993; 27(Suppl)153: 1-93.
3. Petros PE. Severe chronic pelvic pain in women may be caused by ligamentous laxity in the posterior fornix of the vagina. Aust NZ J Obstet Gynaecol 1996; 36: 351-4.
4. Doggweiler R, Whitmore KE, Meijlink JM, et al. A standard for terminology in chronic pelvic pain syndromes: a report from the chronic pelvic pain working group of the international continence society. Neurourol Urodyn 2017; 36: 984-1008.
5. Meijink J. 2018 ESSIC meeting. Florence, Italy; 2018 29 Nov–1 Dec. Available at: https://www.essicmeeting.eu/

6. Butrick CW, Sanford D, Hou Q, Mahnken JD. Chronic pelvic pain syndromes: clinical, urodynamic and urethelial observations. Int Gynecol J Pelvic Floor Dysfunct 2009; 20: 1047-53.

7. Wagenlehner FM, Fröhlich O, Bschleipfer T, Weidner W, Perletti G. The integral theory system questionnaire: an anatomically directed questionnaire to determine pelvic floor dysfunctions in women. World J Urol 2014; 32: 769-81.

8. Petros PE. New ambulatory surgical methods using an anatomical classification of urinary dysfunction improve stress, urge, and abnormal emptying. Int Urogynecol J Pelvic Floor Dysfunct 1997; 8: 270-8.

9. Petros PEP, Bush M. A feedback control system explains clinical and urodynamic bladder instability in the female. Pelviperineology. 2016; 35: 90-3.

10. Petros PE, Ulmsten U. Role of the pelvic floor in bladder neck opening and closure: I muscle forces. Int Urogynecol J 1997; 8: 74-80.

11. Petros PE,Ulmsten U. Role of the pelvic floor in bladder neck opening and closure: II vagina. Int Urogynecol J 1997; 8: 69-73.

12. Bush MB, Petros PEP, Barrett- Lennard BR. On the flow through the human urethra. Biomechanics. 1997; 30: 967-9.

13. Petros PE, Bush MB. A mathematical model of micturition gives new insights into pressure measurement and function. Int Urogynecol J 1998; 9: 103-7.

14. Bush MB, Moron C, Messner-Pellenc L, Petros PE, Millard R. A mechanical model for the opening of the human female urethra. Proceedings of Biomedical Engineering 2005, Austria, K.-P. Adlassnig and M. Bracale (eds), Acta Press, 2005: 210-3.

15. Petros PE, Bush M. Active opening out of the urethra questions the basis of the Valentini–Besson–Nelson (VBN) mathematical model. Int Urogynecol J. 2013; 24: 1585-6. Bush M, Wagenlehner F, Liedl B, Petros P. A finite element model validates an external mechanism for opening the urethral tube prior to micturition in the female. World J Urol 2015; 33: 1151-7.Petros P, Lynch W, Bush M. Surgical repair of uterosacral/cardinal ligaments in the older female using the tissue Fixation system improves symptoms of obstructed micturition and residual urine. Pelviperineology. 2015; 34: 112-6.

16. Petros PEP, Bush M. Rapid bladder and bowel evacuation- an evolutionary survival mechanism? Techniques in coloproctology. Int J Urogynecology 2015; 19: 661-2.

17. Wagenlehner F, Muller-Funogea, IA, et al. Vaginal apical prolapse repair using two different sling techniques improves chronic pelvic pain, urgency and nocturia : a multicentre study of 1420 patients. Pelviperineology 2016; 35: 99-104.

18. Liedl B, Goeschen K, Yassouridis A, et al. Cure of underactive and overactive bladder symptoms in women by 1,671 Apical Sling 1operations gives fresh insights into pathogenesis and need for definition change. Urol Int 2019; 103: 228-34. Goeschen K, Gold DM. Surgical cure of chronic pelvic pain, associated bladder & bowel symptoms by posterior sling in 198 patients validates the Pescatori iceberg principle of pelvic symptom co-occurrence. Pelviperineology 2017; 36: 84-8.

19. Liedl B, Inoue H, Sekiguchi Y, et al. Is overactive bladder in the female surgically curable by ligament repair?. Cent European J Urol 2017; 70: 53-9. 24.Inoue H, Kohata Y, Sekiguchi Y, Kusata T, Kukuda T, Monnma, M. The TFS minisling restores major pelvic organ prolapse and symptoms in aged Japanese women by repairing damaged suspensory ligaments:12-48 month data. Pelviperineology 2015; 34: 79-83.

20. Inoue H, Kohata Y, Fukuda T, Monma M, et al. Repair of damaged ligaments with tissue fixation system minisling is sufficient to cure major prolapse in all three compartments: 5-year data J Obstet Gynaecol Res 2017; 43: 1570-7.

21. Abendstein B, Brugger BA, Furtschegger A, et al., Role of the uterosacral ligaments in the causation of rectal intussusception, abnormal bowel emptying, and fecal incontinence-a prospective study. Pelviperineology 2008; 27; 118-21.Petros PEP, Richardson PA TFS posterior sling improves overactive bladder, pelvic pain and abnormal emptying, even with minor prolapse –a prospective urodynamic study. Pelviperineology 2010; 29: 52-5.

22. Himmler M, Rakhimbayeva A, Sutherland SE, Roovers JP Yassouridis A, Liedl B. The impact of sacrospinous ligament fixation on pre-existing nocturia and co-existing pelvic floor dysfunction symptoms; Int Urogynecol J 2021; 32: 919-28.Richardson P. Surgical cure of nocturia using 4 different methods based on strengthening the structural supports of the vaginal apex – a short review. Pelviperineology 2015; 34: 92-3.

23. Liedl B, Goeschen K, Sutherland SE, Roovers JP, Yassouridis A. Can surgical reconstruction of vaginal and ligamentous laxity cure overactive bladder symptoms in women with pelvic organ prolapse? BJU Int 2019; 123: 493-510.

24. Martius H. Über einen häufigen gynäkologischen, Symptomkomplex. Arch Gynecol Obstet 1938; 166: 332-5.

25. Wu Q, Luo L. Petros PEP Case report: Mechanical support of the posterior fornix relieved urgency and suburethral tenderness. Pelviperineology 2013; 32: 55-6.

26. Petros PE, Ulmsten U. An Integral Theory of female urinary incontinence. Acta Obstet Gyne Scan 1990; 69(Suppl)153: 1-79

27. Petros PE, Ulmsten U, Papadimitriou J. The Autogenic Neoligament procedure: A technique for planned formation of an artificial neo-ligament. Acta Obstet Gyn Scan 1990; 69(Suppl)153: 43-51.

ORIGINAL ARTICLE

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Reconstructive surgery in 1,014 consecutive patients with complex sphincter ani rupture: principles and practice

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ABSTRACT

Objective: To describe the anatomic fecal continence mechanism, the mechanism of action, the complex trauma itself and the reconstructive principles and to present the evidence-based results in a large scale study of a comprehensive approach to the repair of complex sphincter ani rupture.

Materials and Methods: A total of 1,014 women with sphincter ani rupture were operated by a personal operation technique exactly in the opposite order of the trauma, first the internal sphincter, then the external sphincter in end-to-end fashion and lastly the perineal body with meticulous attention to detail.

Results: At first attempt the sphincter ani rupture had healed in 924 patients (91%) and at final attempt 988 patients (98%) had healed with incontinence in 10 and a failure was noted in 26 patients out of whom 10 were inoperable.

Conclusion: This operation technique is highly effective since the normal functional anatomy is reconstructed and restored along with the physiology. It can be recommended to the low-, middle- and high-income world as evidence-based standard.

Keywords: Sphincter ani rupture; complex trauma; reconstructive surgery; prospective large scale study; evidence-based results

INTRODUCTION

Up until today, the fresh, intermediate or old obstetric sphincter ani rupture constitutes a challenge to the obstetrician and the surgeon in the low-, middle- and high-income world since it is a complex trauma with extensive involvement of the anatomic fecal continence mechanism as a Kees IIb rectovaginal fistula.[1-4]

The results after surgery are not optimal because the functional anatomy is complicated, the mechanism is poorly understood, the trauma itself is complex and the surgical skills are not always up to standard combined with traction upon the repair from different directions/structures; as demonstrated in this study since 70% of the patients had been operated already from 1 to 12 times, and also by the fact that even in the author's hands the failure rate per operation is 9%.

Through continuous theoretical, clinical and surgical research and evidence-based results since 1984, the author developed a systematic reconstructive surgery approach with meticulous attention to detail keeping the additional surgical trauma to a minimum.[5]

The anatomic stool continence mechanism

The anatomic stool continence mechanism consists of the distal anorectum as smooth muscle internal sphincter, the external sphincter ani striated muscle as supported posteriorly

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by the puborectalis muscle and anteriorly by the perineal body as centrum tendineum into which the bulbocavernosus muscles radiate as posterior median (in)direct union and the transversus perinei muscles radiate as median (in)direct union as demonstrated in Figure 1.

The mechanism of action

The anterior obstetric sphincter ani rupture is a cut-through trauma whereby the head of the infant cuts through the stiff perineum in the following order: posterior distal vagina wall, the perineum with perineal body, the external sphincter ani, the prerectal fascia and the anorectum (as internal sphincter); since the perineal body is disrupted the posterior median union of the bulbocavernosus muscles and the median union of the transversus perinei muscles are disrupted as well; however without tissue loss.

The cut through is by precipitous delivery of the infant's head either as associated with a wide pelvis and wide pubic arch and then isolated with a live infant or after the obstruction within the birth canal has been overcome in a normal or narrow pelvis and then combined with a pressure necrotic vesicovaginal fistula (VVF) with a stillborn infant.

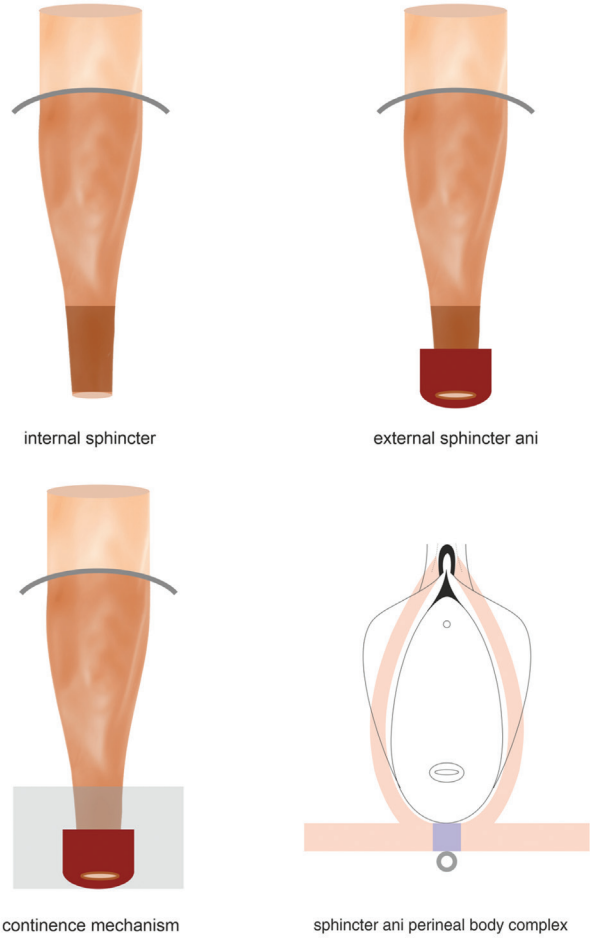

Figure 1. Stool continence mechanism

Seldomly it is due to pressure necrosis in between the fetal head and the maternal sacrum and coccyx bones and then combined with extensive pelvis soft tissue loss which makes most of these fistulas inoperable.

The posterior obstetric sphincter ani trauma is caused by pressure necrosis in between the fetal head and the maternal coccyx bone.

The penetrating trauma like median episiotomy, impalement or rough sex could also be considered as a cut-through trauma.

Rarely it is caused by infection like lymphogranuloma venereum, perianal abscess formation or necrotizing gangrene.

The complex trauma of anterior sphincter ani rupture

The trauma consists of a tear in the distal posterior vagina wall with bilateral retraction, a rupture of the perineum with disruption of the perineal body with bilateral retraction since the posterior median union of the bulbocavernosus muscles and the median union of the transversus muscles are disrupted as well with retraction of these muscles, a rupture of the external sphincter ani muscle with bilateral circular retraction and a rupture of the prerectal fascia and anorectum (internal sphincter) with bilateral circular retraction.

The sum of all these tractions in different directions results into the typical inverted V configuration of the anterior sphincter rupture whereby the anterior anus endings are pulled into the vagina in the direction of the sacrum bone.

Lateral or posterior sphincter ani ruptures have their own configurations as based on traction in their specific location.

MATERIALS AND METHODS

Since 1983 up till today a total of 1,237 women with a sphincter ani rupture were treated out of whom 97 healed spontaneously including 13 with a posterior rupture, 91 were completely stool/flatus continent but came for leaking urine and 45 did not return for repair after VVF repair.

The remaining 1,014 sphincter ruptures were operated personally by the author with the following relevant characteristics and data.

The location of the rupture was anterior in 1,007 or >99%, lateral in three and posterior in four. The cause was obstetric in 978 or 96.5%, penetrating trauma in 22, iatrogenic in 16 and infection in three. The rupture was combined with a VVF in 548 or 54%. A previous operation, ranging from 1 to 12 times, was noted

in 702 or 70% resulting into mutilation in 221 or 22% whilst 16 were considered inoperable. A colostomy was still present in 11 patients. The fistula size, i.e. longitudinal tear of the anorectum, was small (<2 cm) in 469 or 45.5%, medium (2–3 cm) in 410 or 39.8%, large (4–5 cm) in 90 or 8.7% and extensive (>6 cm) in 61 or 5.9%. Out of these patients, 10 developed the rupture within the age group 0–9 years, 639 within the age group 10–19 years, 255 within the age group 20–29 years, 98 within the age group 30–39 years and 12 within the age group 40–49 years. The duration of the fistula on operation day was less than 3 months in 329, from 3 months to 1 year in 300 and from 1 to over 20 years in 385 patients. The vagina was shortened (<9 cm) in 108 with stenosis in 90 and stricture in 22 patients.

The index parity in the 978 obstetric fistula patients varied from 1 to 14 with the majority of 570 or 60% at para 1. The great majority of 657 patients or 67% delivered in a hospital and 63 by caesarean section probably after failed vacuum or forceps trial. Out of the 984 infants born, since 6x twin delivery, 671 were males and 313 were females with a male to female sex ratio of 7:3; whilst 470 were born alive. A foot drop was noted in 765 patients or 78%. A repeat obstetric sphincter ani was found in 27 patients: 23 presented with a second, three with a third and one with a fourth obstetric rupture.

Preparation

As soon as the wounds are clean the patient is considered to be suitable for operation unless her general condition does not permit it. Since it is not possible to give enemas for whatever reason the preoperative instructions are no food the night before and the morning of the operation, bowel movement the morning of operation and drinking until the operation.

Personal reconstructive operation technique (see https://youtu.be/QzVUPvfUtWA)

All operations are performed under spinal anesthesia and in the lithotomy position with the surgeon and assistant standing. Just before the operation is started and under spinal anesthesia, a final assessment is made of the complex trauma with special attention to eventual pressure necrotic lesions.

The operation is executed in the opposite order of the trauma and can be divided into the following different steps:

1. Incision and dissection

Stretching the anorectum trauma into a straight horizontal line by bilateral fixation of the skin next to both external sphincter ani ends onto the inner sides of the legs. Then deep straight horizontal dissecting incision from one sphincter end to the other in between the posterior vagina wall and distal anorectum edge freeing the posterior vagina wall from the prerectal fascia with freshening of the sphincter ends; but only minimally to such an extent that closure becomes possible without tension; extensive dissection is avoided because it is not necessary and will lead only to bleeding; if needed sharp/blunt freeing of adhesions between the rectum and surrounding tissue and/or sphincter ends and surrounding tissue to achieve a tension-free repair; this may be found if the patient has been operated before. Then the stretching sutures are being removed otherwise they would hinder the closure of the rectum, anus and sphincter by pulling exactly the opposite way.

2. Reconstruction of the anorectum (internal sphincter) with repositioning of the anterior anus

Longitudinal closure of the distal anterior anorectum and anterior anus by a layer of interrupted inverting polyglycolic acid sutures thru the prerectal fascia and muscularis (= internal sphincter) starting 1 cm proximally from the anorectum defect and up to the very distal end of the anterior anus; special care has to be given to the most distal suture at the anocutaneous junction which should start inside the mucosa thru muscularis of one side and then thru muscularis/mucosa on the other side and back; if this suture has been tied inside the anus the internal sphincter has been adapted over its full length with repositioning of the anterior anus and the inner ring of the external sphincter should be adapted (as check if the anterior anorectum defect really has been totally repaired); this layer is for strength of the internal sphincter repair

The first layer is inverted by a second layer of continuous polyglycolic acid for an air-tight closure, and then the smooth-muscle internal sphincter has been repaired over its full length.

This part of the reconstruction is the most important since it is the internal sphincter which is predominantly responsible for a continuous closure of the anus due to the tonus of its smooth-muscle circular arrangement.

If there is anything that would endanger the repair, e.g. major tissue loss due to pressure necrosis, the operation is ended as a first stage and the reconstruction of the external sphincter postponed as a second stage as done in 36 patients.

3. Reconstruction of the external sphincter ani

The striated-muscle external sphincter ends are identified and without any further dissection the already freshened sphincter ends are united in an end-to-end manner by three separate polyglycolic sutures (superficial, deep and subcutaneous part). Then patency of the anus is checked.

4. Reconstruction of the perineal body (with re-union of transversus perinei and bulbocavernosus muscles)

To restore the anatomy and shape of the vulva/perineum and to support the sphincter mechanism, the perineal body is repaired by 3-0 polyglycolic acid sutures taking deep bites starting para-anally; at the same time there will be indirect re-union of the transversus perinei and of the bulbocavernosus muscles. After tying these sutures there should be a normal-shape vulva with the perineum adapted; if it does not look normal then the repair is not alright, and this should be corrected.

5. Median transverse adaptation of the posterior vagina wall

Only transverse adaptation of the posterior vagina wall in the midline is needed, together with the last perineal body suture, according to the principles of septic surgery. The vagina and the perineum are always contaminated and thus ensuring free drainage of bacteria, wound fluid and small blood clots.

Figure 2. Sphincter ani rupture

6. Check upon hemostasis

After careful check upon hemostasis, the patient is transferred to the postoperative ward (Figure 2-5).

Remarks

The art is to reconstruct the stool continence mechanism step by step, slowly but very visible after each step, the normal functional anatomy is being restored and the (anterior) anus comes to lie secured in its anatomic distal position, outside the vagina and slightly protruding; everything should look normal.

If the sphincter ani rupture is combined with a vesicovaginal fistula (VVF), the VVF should be repaired first and then the sphincter rupture, either in the same session as done in 427 patients in this study, or if too complicated, the sphincter repair should be performed after the VVF has been repaired and healed.

The operation time of sphincter ani reconstruction is 20 plus/minus 5 minutes and of the combined VVF/RVF reconstruction 40 minutes plus/minus 10 minutes.

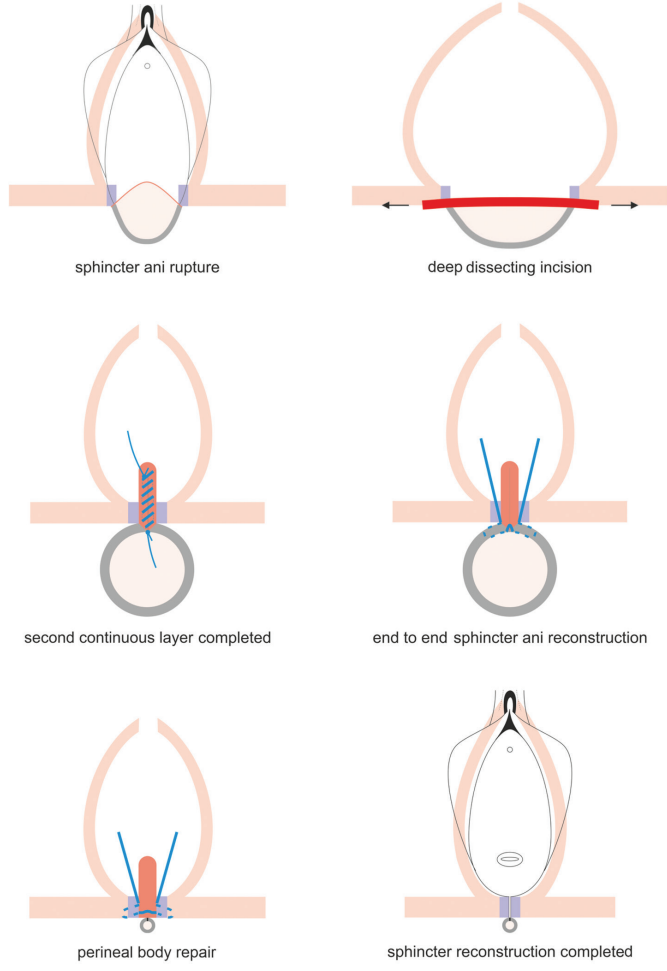

Figure 3. Reconstruction principles

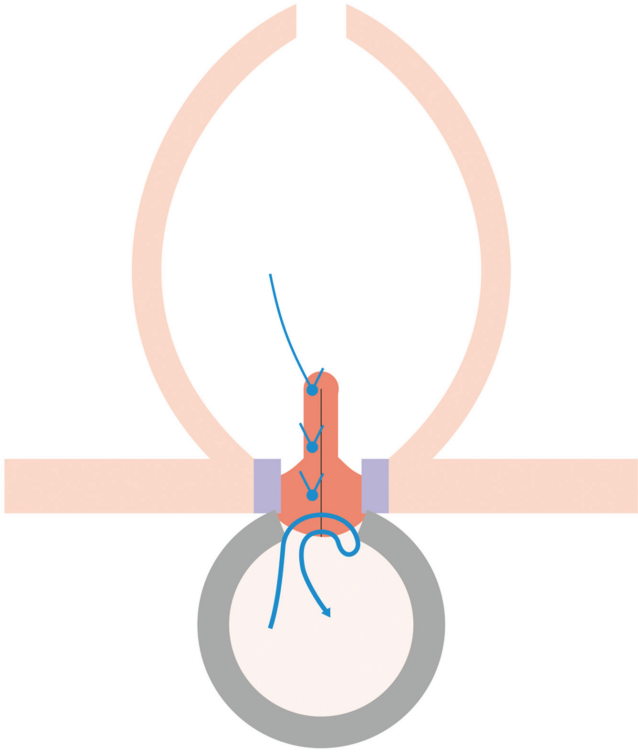

Figure 4. Crucial suture anocutaneous junction

If the repair was not successful, either in closure or in continence, a repeat operation was performed according to the same principles as the first repair.

Postoperative instructions and follow-up

Full mobilization should be started the morning after operation day, no solid food for 10 days and a stool softener for 10 days. After each bowel movement the perineum/anus has to be cleaned carefully by water and then completely dried. Sitzbaths are contraindicated, since they will soften the repaired tissue and infection will be more frequent; normally the less one does about an operation wound the better it heals.

10 to 14 days postoperatively the patient will be asked about defecation and stool/flatus (in)continence with complete inspection/examination of the operation site.

If it has healed, she will be discharged, instructed to refrain from sex for at least 3–4 months and told to come back one month later, when the same examination will be repeated up to 5–6 months postoperatively.

If it has not healed, she will also be discharged and instructed to come back after 3 months for another repair which is done according to the same principles as if it were the first.

If it has healed with slight incontinence, she is re-assured this will improve over 2–3 months since the tissues need time to heal completely due to strengthening and re-arrangement under physiologic stress.

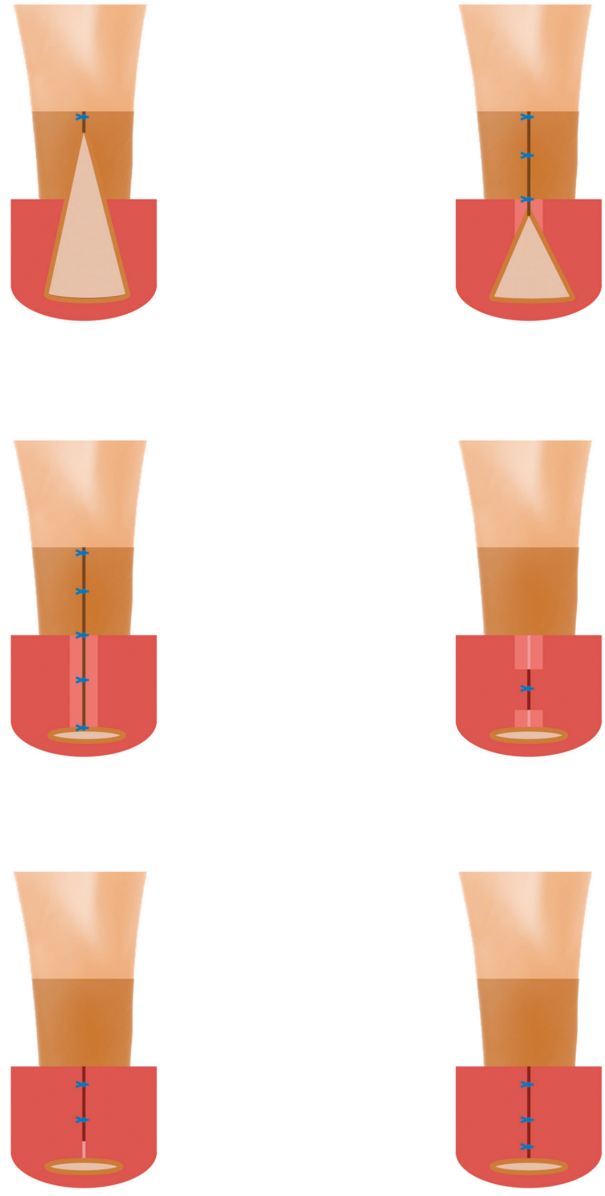

Figure 5. Internal and external sphincter reconstruction

If the patient complains of gross flatus/stool incontinence a meticulous examination is done to exclude a minute blow-out fistula or a loose external sphincter ani, and action taken accordingly.

Normally, the highly qualified and well trained operating theatre staff is responsible for the follow-up; only if there are problems the surgeon will see and examine the patient.

All things are meticulously documented by computerized operation reports including all relevant data, by prediction of outcome in healing and continence on a 5% scale from 5% to 95%, by schematic drawings, by digital pre-, intra- and/or postoperative photos and by written down postoperative check-ups until six months postoperatively; that is the real strength of the programme since it provides evidence-based results.

RESULTS

After first attempt by the author 924 patients (91%) were healed with stool/flatus continence in 895 (97% of the healed patients) whilst 90 had a complete breakdown including 10 of the 16 inoperable patients. During the first six months after repair 12 patients returned with a new sphincter ani rupture probably due to early sexual intercourse.

After another 157 repeat operations by the author finally 988 patients (98%) had healed completely with full stool/flatus continence in 977 (99% of the healed repairs) and 26 had failed out of whom 10 were inoperable.

Unfortunately, one patient died suddenly and unexpectedly at day 1st and another patient died from hepatorenal failure after taking native drugs at day 9th postoperatively.

During long-term follow-up 113 patients returned whilst pregnant with full stool/flatus continence and 31 patients returned with a repeat sphincter ani rupture after a subsequent delivery.

DISCUSSION

Though the perineum is torn as well, the sphincter ani rupture cannot be considered to be a fourth degree perineum tear since a perineum tear is only a tear, whilst the sphincter ani rupture is a highly complex trauma of all the different stool continence structures. In a compound bone-fracture there is a skin tear as well, but nobody considers it as a fourth degree skin tear. However, there is a fluid transition from a perineum tear into a complex sphincter ani rupture, the mechanism is the same and the symptoms may be the same.

The presented technique is a minimum-invasive straightforward approach with the objective to reconstruct only the affected individual structures, under the philosophy that in surgery only the necessary has to be done, nothing more but also nothing less; however, this has to be done very well with meticulous attention to detail. In repairing the distal anorectum,the first phase of the reconstruction it is the muscularis (= internal sphincter) together with the prerectal fascia that is being taken up by the sutures and not the mucosa as being described in other studies; otherwise, the internal sphincter being the most important part of the anal continence mechanism would not be reconstructed. Since the anorectum is composed of very delicate tissue, instrumentation and tissue handling is of utmost importance whilst care should be taken that minimum tension is applied in tying the sutures, bringing the tissues together. If at the end of this stage, the anterior anus is not in anatomic position, and/or the inner ring of the external sphincter ani is not adapted. it means there a defect remains in the distal part of the internal sphincter which may lead to incomplete anal continence.

In 1993 the author used the overlapping sphincter technique once but gave it up immediately since it is non-physiologic with additional surgical trauma.

Though these parts cannot be identified macroscopically, an attempt is made to unite the superficial, deep and subcutaneous parts of the sphincter ani muscle, by different placement of the sutures.

The reconstruction failure rate of 9% may be explained by traction upon the repair from different directions and structures, circular by the internal sphincter, circular by the external sphincter, transverse by the transversus perinei muscles, and anteriorly by the bulbocavernosus muscles whilst previous repairs made their contribution as well.

However, the final reconstruction success rate of 98% with incontinence in only 10 (1%) of the healed patients is excellent.

There still remain few patients, in whom the reconstruction of the anorectum and external sphincter and perineal body is not possible, due to subtotal loss of these and other intravaginal structures. This remind us that there are limits to reconstructive surgery nevermind how skilful and resourceful the surgeon may be.

The reason that some patients with a sphincter ani rupture, even with a large anorectum trauma, claim to be fully stool/flatus continent, may be due to strong action of the puborectalis muscle at the anorectal angle level.

The author never used a colostomy, never united the levator ani muscles and never used the gracilis muscle, in his obstetric trauma surgery since first there is no need for these, and second - the theoretic and practical aspects are not sound. The third reason is that these procedures are mutilating and not contributing to the quality of life.

CONCLUSION

This operation technique is highly effective since the normal functional anatomy is reconstructed/restored and with it the physiology; it can be recommended to the low-, middle- and high-income world as evidence-based standard.

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Ethics

Ethics Committee Approval: This article does not require ethics committee approval since the previous patient records were used as data.

Informed Consent: Not necessary for this type studies.

Peer-review: Internally and externally peer-reviewed.

DISCLOSURES

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REFERENCES

1. Zetterström J, López A, Anzén B, Norman M, Holmström B, Mellgren A. Anal sphincter tears at vaginal deliveries; risk factor and clinical outcome of primary repairs. Obstet Gynecol 1999; 94: 21-8.
2. Okeahialam NA, Thaktar R and Sultan AH. Early secondary repair of obstetric sphincter injuries (OASIs): experience and review of the literature. Int Urogynecol J 2021; 32: 1611-22.
3. Sultan AH, Monga AK, Kumar D and Stanton SI. Primary repair of obstetric anal sphincter rupture using the overlap technique. Br J Obstet Gynaecol 1999; 308: 887-91.
4. Waaldijk K. Obstetric trauma surgery; art and science; 025 kees classification of stool fistulas. Printmarkt, 2020, 22-30. Available at:https://www.urotoday.com/recent-abstracts/pelvic-health-reconstruction/fistula/127612-obstetric-trauma-surgery-art-and-science-setting-standards-by-evidence-based-practice.html
5. Waaldijk K: Obstetric trauma surgery; art and science; 030 complex sphincter ani rupture. Printmarkt, 2021, 50-80.

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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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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 was 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 issue[1-4] 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 test[11] – 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 emptying[7] 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.

REFERENCES

1. Petros PE, Ulmsten U. The posterior fornix syndrome: a multiple symptom complex of pelvic pain and abnormal urinary symptoms deriving from laxity in the posterior fornix. Scan J Urol Nephrol 1993; 27(Suppl 153): 89-93.

2. Goeschen K. Posterior fornix syndrome: comparison of original (2004) and modified (2015) post-PIVS anatomic and symptomatic results – a personal journey. Pelviperineology 2015; 34: 85-91.

3. Liedl B, Goeschen K, Durner L. Current treatment of pelvic organ prolapse correlated with chronic pelvic pain, bladder and bowel dysfunction. Current Opinion in Urology 2017; 27: 274-81.

4. Tuchenhagen K, Liedl B, Yassouridis A, Witczak M. Coexisting symptoms of overactive bladder, abnormal bladder emptying, fecal incontinence, obstructive defecation and pain in women with pelvic organ prolapse. J Urol 2020; 203(Suppl4): e42.

5. Liedl B, Goeschen K, Sutherland SE, Roovers JP, Yassouridis A. Can surgical reconstruction of vaginal and ligamentous laxity cure overactive bladder symptoms in women with pelvic organ prolapse? BJU Int 2019; 123: 493-510.

6. Himmler M, Rakhimbayeva A, Sutherland, SE, Roovers JP, Yassouridis A, Liedl B. The impact of sacrospinous ligament fixation on pre-existing nocturia and co-existing pelvic floor dysfunction symptoms. Int Urogynecology J 2021; 32: 919-28.

7. Himmler M, Kohl M, Rakhimbayeva A, Witczak M, Yassouridis A, Liedl B. Symptoms of voiding dysfunction and other coexisting pelvic floor dysfunctions: the impact of transvaginal, mesh-augmentated sacrospinous ligament fixation. Int Urogynecology J 2021. doi: 10.1007/s00192-020-04649-y [Online ahead of print].

8. Tuchenhagen K. Gruppierte Symptome bei prolapsbedingten Beckenbodendyfunktionen der Frau. Analyse des Eisbergphänomens“ unter Verwendung der Daten der Propel-Studie. Dissertation LMU München 2021. Available at: https://edoc.ub.uni-muenchen.de/28257/7/Tuchenhagen_Kristina_Marie.pdf

9. Liedl B. Inoue H, Sekiguchi Y, Gold D, Wagenlehner F, Haverfield M, Petros P. Update of the Integral Theory and System for Management of Pelvic Floor Dysfunction in Females. Eur Urol 2018; 17(Suppl): 100-8.

10. Wu Y, Luo L, Petros P. Mechanical support of the posterior fornix relieved urgency and suburethral tenderness. Pelviperineology 2013; 32: 55-6.

11. Zarfati D, Petros P. The Bornstein Test - a local anaesthetic technique for testing uterosacral nerve plexus origins of chronic pelvic pain. Pelviperineology 2017; 36: 1-3.

12. Liedl B, Goeschen K, Grigoryan N, et al. The association between pelvic organ prolapse, pelvic pain and pelvic reconstructive surgery using transvaginal mesh: A secondary analysis of a prospective observational cohort trial. J Clin Gynecol Obstet 2020; 9: 79-95.

13. Petros P, Richardson PA. TFS posterior sling improves overactive bladder, pelvic pain and abnormal emptying, even with minor prolapse. A prospective urodynamic study. Pelviperineology 2010; 29: 52-5.

14. Petros P. Severe chronic pelvic pain in women may be caused by ligamentous laxity in the posterior fornix of the vagina. Aust NZ J Obstet Gynecol 1996; 36: 351-4.

15. Goeschen K. Role of uterosacral ligaments in the causation and cure of chronic pelvic pain syndrome. Pelviperineology 2015; 34: 2-20.

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.

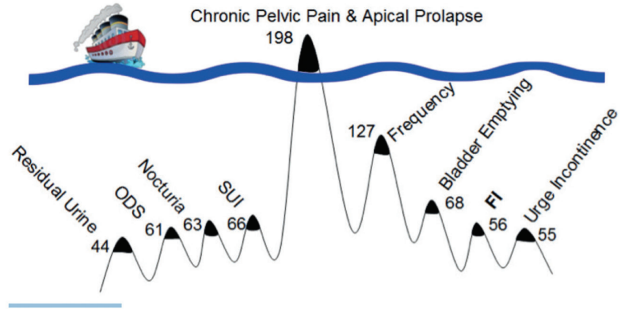

Figure 1. Pescatori Iceberg applied to Chronic Pelvic Pain (CPP). The symptom prevalence is graphically indicated in the iceberg diagram, latent symptoms below the waterline. All symptoms derived from the Integral Theory Symptom Questionnaire (ITSQ). Numbers indicate combined total of the hysterectomy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

REFERENCE

1. Goeschen K, Gold DM. Surgical cure of chronic pelvic pain, associated bladder & bowel symptoms by posterior sling in 198 patients validates the Pescatori Iceberg principle of pelvic symptom co-occurrence. Pelviperineology 2017; 36: 84-8.

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.

REFERENCES

1. Liedl B, Goeschen K, Yassouridis A, Inoue H, Abendstein B, Müller-Funogea I-A A Caliskan A. Cure of Underactive and Overactive Bladder Symptoms in Women by 1671 Apical Sling Operations Gives Fresh Insights into Pathogenesis and Need for Definition Change. Urol Int 2019; 103: 228-34.

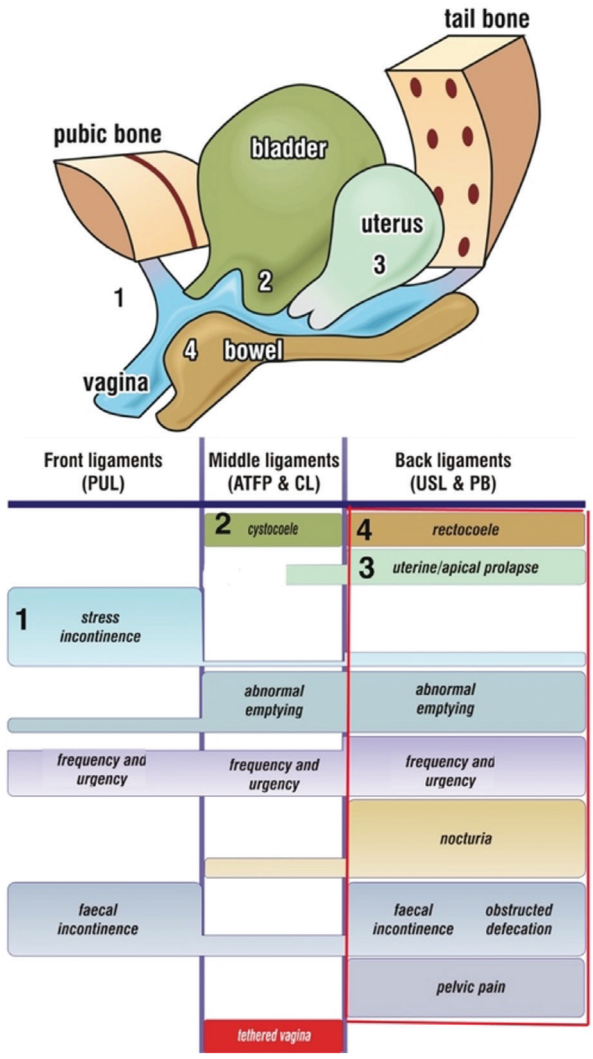

Figure 2. Pictorial algorithm

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 form 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 data[3-5] 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 invariable 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.

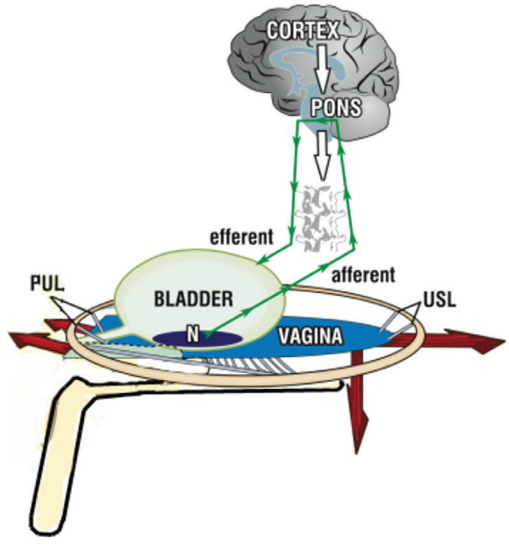

Figure 3. Diminution of urge by "simulated operations"

"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.

REFERENCES

1. Petros PE. Letter. Urinary incontinence during pregnancy: a function of collagen depolymerisation by relaxin? Eur J Obstet Gyn Reprod Biol 2015; 186: 111-2.
2. McCredie JP, Skilling PM. Are psychiatric' findings in patients with Chronic Pelvic Pain primary or secondary? Pelviperineology 2018; 37: 24-7.
3. Petros PE, Skilling PM. Pelvic floor rehabilitation according to the Integral Theory of Female Urinary Incontinence. First report. Eur J Obstet Gyn Reprod Biol 2001; 94: 264-69.
4. Skilling PM, Petros PE. Synergistic non-surgical management of pelvic floor dysfunction: second report. Int Urogynecol J 2004; 15: 106-10.
5. Petros PE, Skilling PM. The physiological basis of pelvic floor exercises in the treatment of stress urinary incontinence. Br J Obstet Gynaecol 1999; 106: 615-6.

Comment from Professor Akın Sivaslioglu:

Akın 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 IVS sling. 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 rectocoele and cystocoele. However, PIVS also cures enterocoele 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 shown[1] 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.

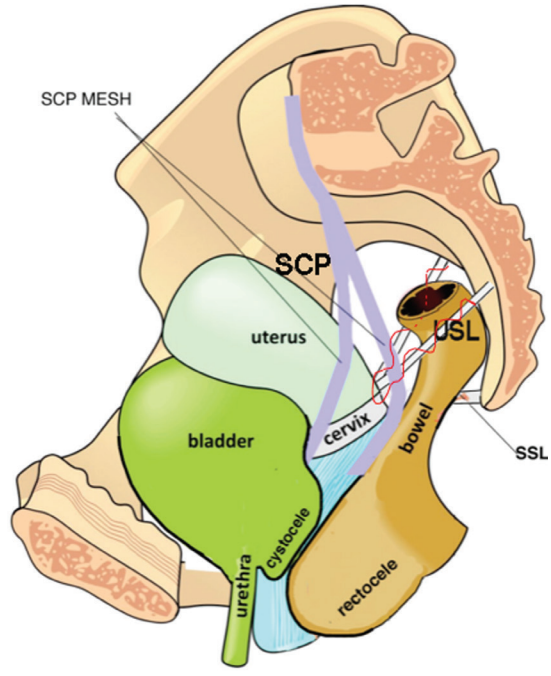

Figure 4. Sacrocolpopexy (SCP) attaches a large mesh laparoscopically from apex to the sacral promontory (lilac colour). Whereas this will hold up a prolapsed uterus or apex, it is not anatomical. The uterus is suspended by uterosacral ligaments (USL) which attach the posterior cervical ring to the sacrum 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.

REFERENCES

1. Sivaslioglu AA, Ilhan TT, Aydogmus S, Uzun M, Dolen I. The comparison of the anatomical and symptomatic outcomes

of sacrocolpopexy and posterior intravaginal slingoplasty. Int Urogynecol J. 2011; 22: 1363-8.

2. Bush MB, Petros PEP, Barrett-Lennard BR. On the flow through the human urethra. Biomechanics 1997; 30: 967-9.
3. Petros PE, Bush MB. A mathematical model of micturition gives new insights into pressure measurement and function. Int Urogynecol J 1998; 9: 103-7.
4. Bush MB, Moron C, Messner-Pellenc L, Petros PE, Millard R. A mechanical model for the opening of the human female urethra. Proceedings of Biomedical Engineering 2005, Austria, KP. Adlassnig and M. Bracale, eds, Acta Press, 2005: 210-3.
5. Petros PEP, Swash M. Sacrocolpopexy may cause difficult defecation by inhibiting the external opening out mechanism. Int Urogyn J 2011; 22: 255.

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 controls[1]. 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.

REFERENCES

1. Vânia Meira e Siqueira-Campos, Rosa Azevedo Da Luz, José Miguel de Deus, Edson Zangiacomi Martinez, and Délio Marques Conde. Anxiety and depression in women with and without chronic pelvic pain: prevalence and associated factors. J Pain Res 2019; 12: 1223-33.
2. Schonfeld M, Petros P, Bornstein J. Mechanically Supporting Uterosacral Ligaments for the Relief of Provoked Vulvodynia: A Randomized Pilot Trial. J Pain Res. 2021; 19; 14: 1281-8.
3. Meister MR, Sutcliffe S, Badu A, Ghetti C, Lowder JL. Pelvic floor myofascial pain severity and pelvic floor disorder symptom bother: is there a correlation? Am J Obstet Gynecol 2019; 221: 235.e1-235.e15.
4. Dixon AM, Fitzgerald CM, Brincat C. Severity and bother of prolapse symptoms in women with pelvic floor myofascial pain. Int Urogynecol J 2019; 30: 1829-34.
5. Liedl B, Goeschen K, Durner L. Current treatment of pelvic organ prolapse correlated with chronic pelvic pain, bladder and bowel dysfunction. Curr Opin Urol 2017; 27: 274-81.
6. Enache T, Bratila E, Abendstein B. Chronic pelvic pain of unknown origin may be caused by loose uterosacral ligaments failing to support pelvic nerve plexuses - a critical review. Cent European J Urol 2020; 73: 506-13.

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.

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