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Case Report

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Cure of Interstitial Cystitis and Non-Ulcerating Hunner's Ulcer by Cardinal/Uterosacral Ligament Repair

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Keywords
Interstitial cystitis · Hunner's ulcer · Posterior fornix syndrome · Tissue fixation system sling · Lax uterosacral ligaments

Abstract
A serendipitous cure in a 73-year-old woman of Hunner's ulcer, urge, nocturia, apical prolapse by a tissue fixation system tensioned minisling (TFS) which reinforced the cardinal, and uterosacral ligaments (USLs) led us to analyse the relationship between Hunner's ulcer and known pain conditions associated with USL laxity. The original intention was to cure the "posterior fornix syndrome" (PFS), uterine prolapse, and associated pain and bladder symptoms by USL repair. A speculum inserted preoperatively into the posterior fornix alleviated pain and urge symptoms, by mechanically supporting USLs. Hunner's ulcer, along with pain and other PFS symptoms were cured by USL repair. The concept of USL laxity causing chronic pelvic pain and bladder problems is not new. It was published in the German literature by Heinrich Martius in 1938 and by Petros in the English literature in 1993. These findings raise important questions. As PFS symptoms are identical with those of interstitial cystitis (IC), are PFS and IC similar conditions? If so, then patients with IC who have a positive speculum test are at least theoretically, potentially curable by USL repair. These questions need to be explored.

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Introduction

Interstitial cystitis (IC)/painful bladder syndrome in all its manifestations, from definitions to diagnosis to management remains an enigma. In 2009, Hanno and Dmochowski [1] summarized the state of knowledge as follows: "*Standardization of nomenclature, definition, and evaluation of interstitial cystitis/bladder pain syndrome/painful bladder syndrome was and remains a problem. Hunner's ulcer is not necessarily an essential part of the diagnosis.*"

The current definition for interstitial cystitis/bladder pain syndrome [2] is "persistent or recurrent chronic pelvic pain, 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, previously a hallmark of IC, is no longer considered essential for diagnosis. In fact, the very

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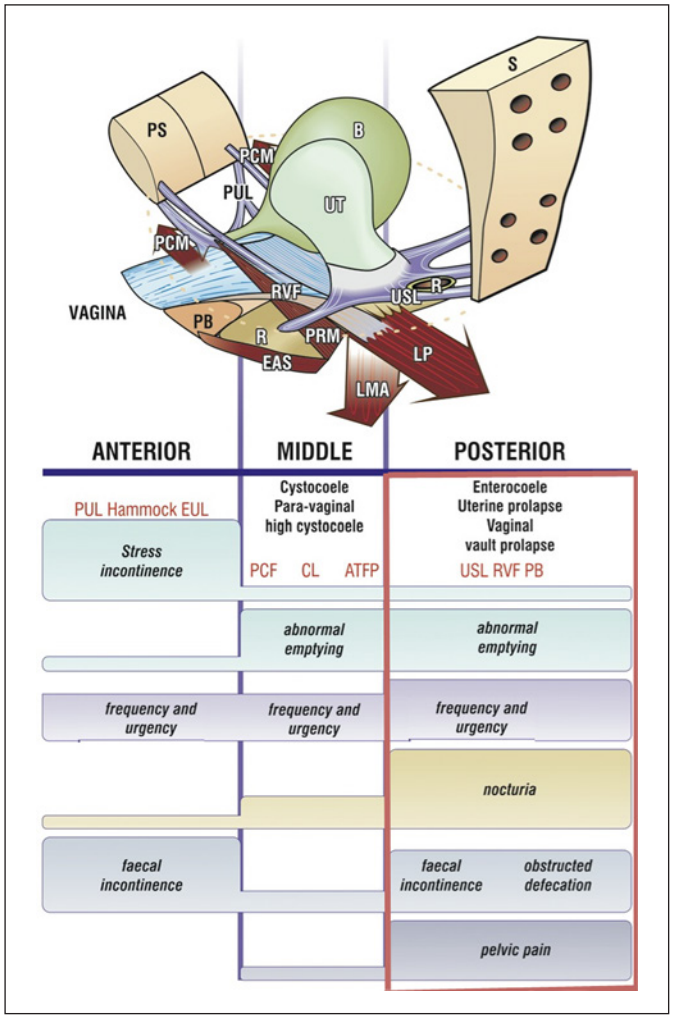

Fig. 1. The pictorial diagnostic algorithm. Symptoms indicate which ligaments are damaged. The height of the bar indicates probability of association of a symptom with a particular zone. The connective tissue structures causing prolapse and pelvic symptoms fall naturally into 3 zones. Anterior zone: external meatus to bladder neck. Middle zone: bladder neck to anterior cervical ring. Posterior zone: posterior cervical ring to PB. The rectangle indicates the symptoms associated with USL laxity. Chronic pelvic pain and nocturia are uniquely caused by USL laxity. PUL, pubourethral ligament; hammock, suburethral vagina; EUL, external urethral ligament; CL, cardinal ligament; PCF, pubocervical fascia; ATFP, arcus tendinous fascia pelvis; PB, perineal body; USL, uterosacral ligaments; RVF, rectovaginal fascia; USL, uterosacral ligament.

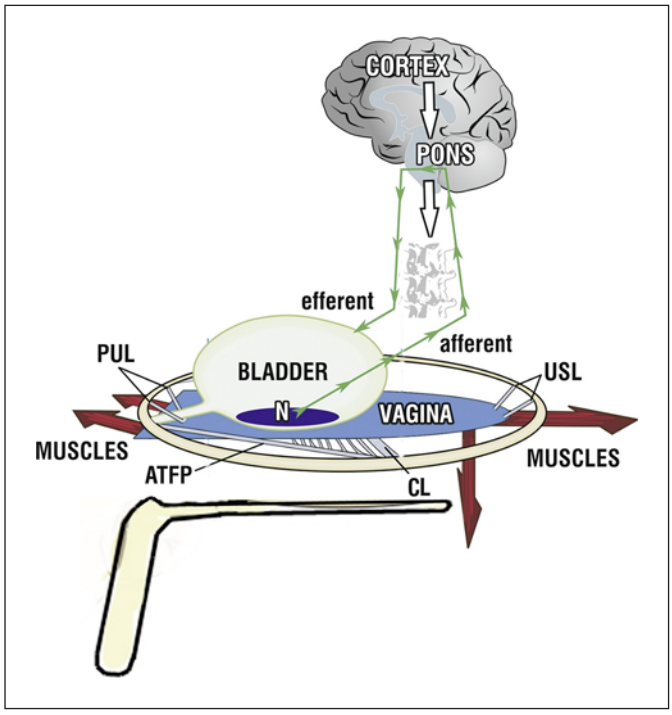

Fig. 2. Speculum test. 3D view of the bladder sitting on the anterior vaginal wall which is suspended from the pelvic brim by ligaments, PUL, cardinal CL, uterosacral USL. A speculum inserted into the posterior fornix mechanically supports lax USLs and with it the nerve plexuses S2-4 and T11-L2. Firm 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. PUL, pubourethral ligament; CL, cardinal ligament; USL, uterosacral ligament.

concept of IC is in doubt. In 2018, Meijink [3], 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." Unsurprisingly, there is no single accepted treatment. The American Urological Association lists 6 "levels of treatment" each containing several totally different options.

Case Report

Frau C., a 73-year-old lady presented in March 2016 with urgency, half-hourly frequency and nocturia twice/night. There was no wetting, and urinalysis was normal. She was assessed by the validated Integral Theory System Questionnaire [4] and diagnostic algorithm (Fig. 1), which uses symptoms to locate ligament damage. On examination, a moderate cystocele and an entero/

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rectocele were seen with laxity of the cardinal/uterosacral ligaments (USLs) and pubocervical/rectovaginal fascia. "Simulated operation," mechanical support of the anterior vaginal wall/apex with a speculum (Fig. 2), led to almost complete urgency and pain resolution as per Wu et al. [5]. Urodynamic examination showed a maximum capacity of 430 mL (limited by pain), detrusor stability, normal compliance, and no leakage. On cystoscopy under general anaesthesia, bladder capacity was 300 mL showing diffuse distension bleeding, chronic cystitis with patchy mucosal reddening, glomerulations, and a non-ulcerating Hunner's ulcer. Resection biopsies showed large numbers of mast cells within the muscular layer. Further clinical testing with a 2nd "simulated operation," a tampon inserted into the vaginal apex to support the USLs considerably improved both urge and pain. In June 2016, an apical repair with a tissue fixation system (TFS) tensioned tape was performed.

Post-operatively, there was good anatomical correction of the apical descensus. The patient reported that her symptoms were gradually disappearing with voiding reduced to 5–6 times daily and nocturia once.

A follow-up cystoscopy in September 2016 showed complete healing of the Hunner's ulcer, no bleeding, and no glomerulations. The bladder capacity was 700 mL. Since then, the patient has been seen regularly twice yearly and is free of any symptoms suggestive of IC.

Discussion

Frau C. was treated according to the integral theory paradigm (ITP) [6], specifically, the "posterior fornix syndrome (PFS)": grouped symptoms of urge, frequency, nocturia, chronic pelvic pain, and abnormal emptying caused by uterosacral ligament laxity (Fig. 1). Inserting the lower blade of a bivalve speculum confirmed lax USLs were causing the pain and urge [5] (Fig. 2). The speculum stretches the vagina to mechanically support USLs. Instantaneous disappearance of urge and pain symptoms supports the concept of cure by restoring the integrity of muscle/ligament complex (http://www.cms.galenos.com.tr›Uploads›Pelviperineology-32-55-En" xmlns:xlink="http://www.w3.org/1999/xlink"www.cms.galenos.com.tr›Uploads›Pelviperineology-32-55-En).

Management according to the ITP

Non-surgically, a >50% improvement in pain, urge, frequency, and nocturia has been reported using squatting-based exercises which strengthen the 3 directional forces and ligaments they contract against [7]. Surgically, experience with younger women with chronic pelvic pain of unknown origin and presumably good USL collagen has shown good cure rates for PFS symptoms (rectangle, Fig. 1) with native tissue USL plication [6]. For postmenopausal women, poor results probably because of collagen deficiency in ligaments necessitated application of a posterior sling to reinforce USLs, achieving high cure rates for chronic pelvic pain, urge, nocturia [8–11], and classic symptoms of IC [2]. Long-term cure at 60 months in a group of 70-year-old women using a TFS CL/USL sling [11] has been reported for prolapse, pelvic pain, and urge.

Our findings with Frau C. support the hypothesis of Wyendale that bladder pain may arise from bladder unmyelinated C-fibres. This may be due to an inability of the 3 reflex muscle forces (arrows, Fig. 2) to tension the bladder base sufficiently to support the stretch/pain receptors "N" because of looseness in the suspensory ligaments (USL, Fig. 1) they contract against. This may cause the now unsupported stretch receptors "N" to fire off excess afferent urge/pain impulses, perceived by the cortex as urge and bladder pain. Alternatively, it is known that chronic pelvic pain may be due to inability of loose USLs to support the T11-L2 and S2-4 visceral nerve plexuses located 2–3 cm before their attachment to cervix [12].

The glomerulations may have been caused during bladder filling by the weight of the bladder contents creating a downward rotation of the bladder into the vagina, to cause mechanical obstruction of low pressure vesical veins. Glomerulations were not present post-operatively. We attribute this to the TFS cardinal ligament sling curing the cystocele, as predicted, (Fig. 1). We hypothesize that repeated mucosal venous outflow obstruction over time may have caused sufficient hypoxia to induce inflammatory mast cell infiltration. Alternatively, unsupported visceral nerve fibres supplying the bladder may have sent afferent signals to the cortex "tricking it" to believe the impulses came from the end organ itself. The cortex sends inflammatory mast and other cells to the endorgan (in this instance bladder).

Conclusions

Statements from the Interstitial Cystitis Society itself indicate a chaotic situation in the very concept of IC: definitions, symptomatology, treatment, unknown pathogenesis, and no known cure. Frau C. was treated according to a different paradigm, the Integral Theory paradigm, ITP, as a typical case of posterior fornix syndrome, PFS, which was successful. All the descriptions of IC in the literature to date indicate equivalence to PFS, at least

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in women. If so, then patients with symptoms of IC who have a positive speculum test may, at least theoretically, be potentially curable by USL repair, ligament plication for the young, and sling for the old. These questions need to be explored.

Statement of Ethics

The study was conducted according to the WMA Declaration of Helsinki. Written informed consent was obtained from the patient, and ethical approval was obtained for publication of this case report.

Conflict of Interest Statement

None for any author.

Funding Sources

None declared.

Author Contributions

All authors contributed to conceptualization and writing. Surgery: K.S.; figures: P.P.

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