



Cure of interstitial cystitis and Hunner's ulcer by TFS ligament repair brings new insights into pathogenesis and management

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This report concerns the first cure of Interstitial Cystitis (IC) and histologically validated Hunner's ulcer in the literature by Tissue Fixation System (TFS) mini-sling repair of cardinal and uterosacral ligaments. The cure was unintentional. A 73-year-old woman was managed according to the protocol for Posterior Fornix Syndrome (PFS)*: validated Integral Theory System Questionnaire, diagnostic algorithm which uses symptoms to locate ligament damage, vaginal examination to confirm uterosacral ligament damage and finally, "simulated operation", mechanical support of the apex with a speculum to test for relief of urgency and pain.

The significance of this cure goes far beyond a case report. It demonstrates that IC is not incurable as was generally believed. Furthermore, the criteria for PFS conforms exactly to the European/International Society for the Study of Interstitial Cystitis (ESSIC) definition for IC which opens opportunities for further research in order to define the differences, if any, between PFS and IC.

*PFS was first described in 1993 as part of the 2nd iteration of the Integral Theory of female Urinary Incontinence.¹ PFS comprises predictably grouped symptoms of urge, frequency, nocturia, abnormal emptying/retention, and chronic pelvic pain

caused by uterosacral ligament (USL) laxity, cured or improved by native uterosacral ligament plication.

As he sailed into the Swan River, Western Australia, on January 10, 1697, Willem Hesselsz de Vlamingh saw a black swan. This disproved forever what had been accepted for thousands of years as incontestable fact, that all swans were white. Thus, it is with the unintentional discovery of a surgical cure of interstitial cystitis (IC) and Hunner's ulcer by ligament repair.² It can no longer be said that IC is incurable. Scheffler's Case was consistent with the ESSIC definition for IC:³ "*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.*" and also, previous IC criteria, glomerulations and histologically validated Hunner's ulcer.

Clearly, it cannot be claimed that ligament repair can cure every case of IC. This editorial limits its aim to seeking insights for pathogenesis and management.

Scheffler did not set out to cure IC. He followed standard protocol for cure of the Posterior Fornix Syndrome (PFS)*.¹ The first assessment was carried out in March 2016. Scheffler

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reported: *"She was assessed by the validated Integral Theory System Questionnaire** and a diagnostic algorithm which uses symptoms to locate ligament damage. On examination, a moderate cystocele and an entero/rectocele were seen with laxity of the cardinal/uterosacral ligaments (USLs), lax pubocervical and rectovaginal fascia. "Simulated operation"***; mechanical support of the anterior vaginal wall/apex with a speculum led to almost complete relief of urgency and pain."*

**The validated ITSQ (Integral Theory System Questionnaire) is specific for the PFS diagnostic protocol. Specific symptoms locate which damaged ligaments may be causing the prolapse and symptoms. Chronic pelvic pain and nocturia are uniquely caused by USL laxity.

***Simulated operation is a screening test for PFS: a speculum gently inserted into the posterior fornix mechanically supports lax USLs and nerve plexuses S2-4 and T11-L2 in order to relieve pain and urgency.

The pre-operative assessment for PFS being positive, Scheffler proceeded to Urodynamic examination. Maximum capacity was 430 ml (limited by pain), and the detrusor was stable.

Compliance was normal, and there was no urine leakage. According to the cystoscopy: bladder capacity was 300 ml, there was diffuse distension bleeding, chronic cystitis with patchy mucosal reddening, glomerulations, and 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" with a tampon inserted in the vaginal apex to support the USLs again improved urge and pain. Satisfied that lack of apical support was the pathogenic factor, Scheffler proceeded to repair the cardinal and uterosacral ligaments with TFS tensioned tape. Post-operatively, there was good anatomical correction of the apical descensus and cystocele. Symptoms gradually disappeared 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. Bladder capacity was now 700 ml. Following this, the patient was seen regularly with no further symptom recurrence.

Scheffler's discovery raised two important questions.

- (1) Was IC a one-off cure?
- (2) Is IC a manifestation of PFS caused by USL weakness?

Similarities with Table 1 indicate that it may not have been a one-off cure. The symptoms in the Goeschen's study of 198 PFS

women who underwent a USL sling⁴ conform in every way to the ESSIC definition of IC and are virtually identical with those in Butrick's classic IC study of 408 women⁵ (Table 1). Butrick treated his patients with bladder installations with no cure. Goeschen performed a USL sling and achieved significant anatomical and symptomatic cure.⁵

The 2nd question can only be answered by an RCT, where one group has bladder installations, and the other a USL repair, or a USL sling if post-menopausal.⁴

Hypotheses for the pathogenesis and cure of IC and Hunner's ulcer² are detailed below. Both the speculum test and the sling mechanically support the USLs and the visceral nerve plexuses (VPs). Unsupported, the VPs are susceptible to stimulation by gravity or muscle movement and so send afferent impulses to the cortex. These are interpreted as pain from an injury to the end organ, the bladder. Thus alerted, the cortex sends efferent signals via visceral nerves to special resident cells at the end-organ. These activate inflammatory cells such as mast cells to initially form non-ulcerating Hunner's ulcer. If the inflammatory response is excessive, the inflammation may cause ulceration. Overactive bladder (OAB) cure is explained differently. Repair of loose USLs restores the muscle forces which stretch the vagina to support the urothelial stretch receptors from below, thus diminishing the afferent impulses which, beyond a threshold which is different for each patient, are interpreted by the cortex as urge.

Finally, a 3rd question needs to be asked: "How can this method be applied?"

For premenopausal women, a simple USL plication done vaginally as reported,³ or laparoscopically, should give good results for both pain and urge symptoms. As postmenopausal women generally have ligaments which are collagen deficient, we have found a sling is generally required for a longer-term cure. Whatever the surgical option, it is imperative to have a positive speculum test prior to any surgery.

Keywords: Hunner's ulcer; interstitial cystitis; TFS mini-sling

ETHICS

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DISCLOSURES

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Table 1. Similarity between interstitial cystitis symptoms and the Posterior Fornix Syndrome

Similarity between interstitial cystitis symptoms and the Posterior Fornix Syndrome	
Butrick Interstitial cystitis (n=408)	Goeschen Posterior Fornix Syndrome (n=198)
Bladder installations NO CURE	Uterosacral sling % cure in brackets
Bladder pain/interstitial cystitis (n=157)	Voiding dysfunction (n=68) (54%)
Chronic pelvic pain (n=98)	Urge incontinence (n=55) (80%)
Vulvodynia/dyspareunia (n=40)	SUI (n=66) (95%)
Voiding dysfunction (n=70)	POP (n=198) (90%)
Dyspareunia (n=54)	Chronic pelvic pain (n=198) (74%)
SUI (n=24)	Nocturia (n=63) (79%)
POP (n=21)	
Hunner's ulcer (n=18)	No Hunner's ulcer reported
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