Review

A 20 year experience of microsurgical removal of the Bartholin's glands for refractory vulvodynia

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Abstract: Since the 1980s, a flood of women have presented with introital dyspareunia and chronic unexplained vulvar discomfort, for which no clear-cut somatic diagnosis can be found. Three decades of research has failed to attribute this syndrome to a structural disease process. Pathogenesis reflects an interaction between individual hypersensitivity and an external irritant, creating a self-sustaining pain reflex. This reflex has well defined afferent (sensory), central (dorsal horn) and efferent (motor) arms. As such, vulvodynia fits the model of a complex regional pain syndrome. Most women respond to conservative treatment, involving membrane stabilizing drugs (to downregulate pain transmission through the dorsal horn) and a biofeedback-controlled exercise program (to stabilize electrical transmission within the efferent arm the spinal pain reflex). In refractory cases, there is sometimes a need to excise the tissue from which the most intense pain signals are arising. This has been most commonly done by vestibulectomy. This article describes a microsurgical technique for Bartholin's glands excision, as a less 'anatomy-altering' and more effective alternative. During calendar years 1999-2014, 99 patients were adjudged suitable for microsurgical removal of the Bartholin's glands. Of these 99 women, pain free intercourse was re-established in 93 (94%) patients, and another two (2%) had partial improvements. There was a 4% failure rate (four women). No major complications were encountered.

Keywords: Vulvodynia; Bartholin's gland; Surgery; Chronic pelvic pain; Pain.

INTRODUCTION

Historically, complaints of chronic vulvar pain were readily attributable to somatic diseases (Table I). Since the 1980s, however, there has been a flood of women presenting with introital dyspareunia and chronic unexplained vulvar discomfort, for which no clear-cut somatic diagnosis can be found.

Chronic and seemingly incurable pain is a potent cause of anxiety, anger and reactive depression, even in mature and socially successful individuals. Vulvodynia primarily affects nulliparous women in their formative years. Symptoms of intractable vulvar pain place key interpersonal relationships under duress and may even impact the ability to work, thus explaining the emotional distress that is so manifest in these young women. Not surprisingly, subsequent research found these psychological symptoms to be the result (not the cause) of the chronic sexual pain in vulvodynia[1,2].

There is now a consensus that vulvodynia is actually a chronic pain disorder[3-5]. It fits the model of a *complex regional pain syndrome* (in past terminology, a "sympathetically maintained pain syndrome" or a "reflex sympathetic dystrophy") consistent with a self-sustaining spinal reflex, with afferent (sensory), central (dorsal horn) and efferent (motor) arms.

The pathogenesis of vulvodynia is thought to reflect the interaction of hypersensitivity (i.e., a genetic susceptibility to an excessive inflammatory response) and an external irritant (i.e., exposure to an extrinsic factor that triggers an idiosyncratic inflammatory cascade) (Table 2).

• *Hypersensitivity:* Many clinical investigators have recognized that 90% of affected women have a fair complexion and a tendency to "sensitive" skin. Goetsch[6] reported that one third of vulvodynia patients have a female relative with dyspareunia or tampon intolerance, thus highlighting the extent to which vulvodynia runs in families. Witkin et al[7] reported that 53% of vulvodynia patients (versus 5% of controls) had a genetically impaired ability to switch off interleukin 1 mediated inflammatory responses[7].

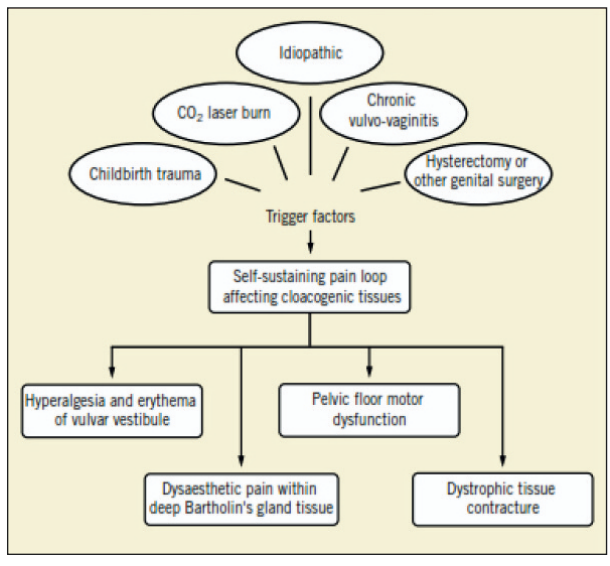
• *External Irritants:* It is likely that the pain reflex might be triggered an external irritant, usually by bacterial vaginosis and chronic candidiasis. However, specific external irritants cannot always be identified. Vulvodynia is not sexually transmitted, and is not a form of subclinical papillomavirus infection[8].

TABLE 1. Classic differential diagnosis of chronic vulvo-vaginal pain.

- Cutaneous vulvar infections (cyclic candidiasis, seborrhoeic dermatitis)
- Secondary irritation from a vaginal discharge (bacterial vaginosis, trichomoniasis)
- Contact dermatitis secondary to chemical or drug irritations (acute and chronic eczematoid dermatitis)
- Vulvar dermatoses (lichen sclerosus, lichen planus, psoriasis, pemphigus)
- Neoplastic epithelial disorders (vulvar intraepithelial neoplasia, Paget's disease, melanoma)
- Vulvar scarring (fibrotic hymen, fourchette contracture band)
- Urethral pain (diverticulum, posterior urethritis)
- Vulvar dysaesthesia (causalgia, pudendal neuralgia)

The end result is a complex regional pain syndrome with several distinct elements. Namely, a nociceptive element (triggered by physical contact with hyperalgesic foci surrounding the Bartholin's and Skene's ostia on the vestibule); a neuropathic element (secondary to sympathetic-somatic nerve fibre cross-talk within foci of surface erythema or the hyperalgesic Bartholins' glands); and a myal-

TABLE 2. A proposed model of pathogenesis.



gic element (secondary to an ischaemic lactic acid build up within chronically hypertonic pelvic floor muscles).

Most women benefit significantly from conservative therapy. The primary thrust is to break the causative self-sustaining pain reflex. This objective can potentially be achieved in several ways:

1. The afferent limb of this spinal reflex is largely sustained by cross-talk between hyperaesthetic perivascular sympathetic nerve fibres and the adjacent somatic pudendal nerve fibres. Dye laser photothermolysis has the capacity to selectively destroy the engorged blood vessels within the painful erythema on the vulvar vestibule[3], thus disrupting this pain loop.
2. Nociceptive and neuropathic pain transmission through the dorsal horn can be impeded with local anaesthetic and membrane stabilizing drugs, such as lignocaine, amitriptyline, gabapentin or pregabalin[9]. Medication can be delivered either orally or topically.
3. Residual neuropathic pain can often be further downregulated with remedial massage of painful trigger points within the anterior vaginal wall[10,11].
4. Myofascial pain within the chronically hypertonic pelvic floor musculature can be targeted using biofeedback, to stabilize electrical transmission within the efferent arm the spinal pain reflex. Biofeedback is a specialised technique, which measures baseline muscle function via surface EMG electrodes, and then transmits these signals to a television monitor in real time. By studying the monitor, patients can learn to isolate and break their chronic pelvic pain loop[12].
5. Sexual pain at this point in life can have damaging emotional consequences. Some couples also benefit from relationship counselling for associated psychosexual disorders.

In a prospective observational study of 725 women with vulvodynia over a 5 year period, selective FEDL photothermolysis of hyperalgesic erythema on the vulvar vestibule secured lasting pain relief in 70.2% (118 of 168) of subjects[3]. Significantly, 43 of the 50 photothermolysis failures reported a severe deep pain on palpation of their Bartholin's glands. Following a lead from Miklos and Baggish[13], our group pioneered a minimally invasive technique for Bartholin's glands excision, using a microsurgically adapted carbon dioxide (CO_2) laser. Gland removal increased final success rate to 93%. However, the development of the Glazer protocol for breaking the vulvodynia pain loop with muscle relaxation obviated the need to use of FEDL photothermolysis beyond 1996[12]. Nonetheless, in refractory cases, there can be a need to excise foci of hyperaemic tissue from which the most intense pain signals are arising. This has been most commonly done by vestibulectomy[8,14]. However, I have continued to rely on Bartholin's glands excision, as a less 'anatomy-altering' and more effective alternative.

This paper reports on my experience with microsurgical Bartholin's glands excision over the last two decades, 1997 to retirement 2016.

CLINICAL ASSESSMENT OF THE REFRACTORY VULVODYNIA PATIENT

The degree of disability associated with vulvodynia varies from nuisance level to sexually and emotionally disabling. The predominant symptom is always introital dyspareunia, sometimes accompanied by unprovoked vulvar discomfort, usually described as a sense of rawness or dryness. Symptoms do not respond to topical antifungal or antibacterial therapies. Vulvodynia symptoms tend to remain relatively constant day to day, meaning that the severity of the patient's presenting complaints provide a guide to the likely treatment response.

• The usual history is that of *secondary vulvodynia*, meaning the sudden occurrence of sexual pain after years of pleasurable intercourse. Provided that any ongoing trigger factors are controlled, such cases usually respond well to muscle relaxation therapy.

• A minority of women *primary vulvodynia,* due to the presence of occult pathology from a young age. Presenting complaint is that of severe and enduring introital dyspareunia from the time of first intercourse, whenever that occurs.

• Occasional vulvodynia patients present with *vaginismus* but no signs of surface vulvodynia.The true nature of the problem is revealed by the finding of a characteristic lancinating pain on Bartholin's glands palpation.

In my experience, the women with primary vulvodynia and unexplained vaginismus are more likely to fail conservative protocols, and may eventually become candidates for surgery.

Examination evaluates four main physical phenomena:

• Chronic dysaesthetic pain, emanating from inflammatory foci on the mucosa of the vestibule.

• Similar inflammatory foci within the tissues of the Bartholin's glands themselves.

• Secondary to this chronic inflammation, the levator muscles exhibit chronic painful spasm[3].

• There can also be dystrophic shrinkage of adjacent tissues (notably hymenal fibrosis, posterior fourchette contracture bands and stenosis of the inflamed Bartholin's ducts). A small YV flap is an easy option for resolving posterior fourchette contracture, and is much superior to a Fenton's procedure[15].

TREATMENT RESULTS

During calendar years 1999-2014, approximately 500 patients were referred to my private gynaecology practice in Sydney for the management of intractable dyspareunia. Patients were first evaluated to identify and grade their various pain components, and to resolve whatever trigger factors might be helping sustain their vulvodynia[16,17]. A conservative therapy regimen was then employed, in an attempt to break the self-sustaining pain reflex driving the vulvar hyperalgesia syndrome. This regimen combined membrane stabilizing drugs to downregulate pain transmission through the dorsal horn[9] and a biofeedback-controlled program to stabilize electrical transmission within the efferent arm the spinal pain reflex. Biofeedback is a specialised technique, which measures baseline muscle function via surface EMG electrodes, and then transmits these signals to a television monitor in real time. By studying the monitor, patients can be taught to isolate and break their chronic pelvic pain loops within their chronically hypertonic pelvic floor muscles[12]. Occasionally, such additional measures as remedial massage for refractory myofascial pains in secondary muscle groups or hormone creams for symptomatic skin fissuring were needed. Some couples also benefited from relationship counselling for associated psychosexual disorders.

About 85% of patients responded permanently to conservative therapy. However, small subsets of patients require a combination of medical and surgical treatment. Surgery was used in two circumstances:

• To break an otherwise refractory local pain reflex, by excising an area of tissue from which the most intense pain signals are arising. This objective is traditionally approached by vestibulectomy. However, in that these foci of painful, hyperemic vessels are consistently seen to emanate

from the ostia of the Bartholin's glands. I have preferred to continue with the strategy of microsurgical gland removal[3]. The procedure requires learning some new operative skills, but has the advantage of being less 'anatomy-altering' and more effective than simple vestibulectomy.

• Reconstructive surgery was sometimes required to release a painful introital narrowing. Small contracture bands are easily and cosmetically corrected by YV advancement. With more severe deformities, I relied on vulvoplasty with transposition of a pair of sensate skin flaps (based on the terminal distribution of the posterior labial artery and nerve)[15]. This aspect is not further discussed in this paper.

Over the course of this 15 year period, 99 patients were adjudged suitable for microsurgical removal of the Bartholin's glands. Beginning in the fourth postoperative week, all patients began the use of a #4 or #5 glass vaginal dilator, to forestall the scar contracture that attends a inevitably attends the maturation of a semicircular peri-orificial scar. It is vital to begin dilator use while the scar is still in the elastic *red scar stage*. During this stage, these immature scars can always be stretched. If dilator use is delayed until the inelastic *white scar stage*, dilator use will only evoke tearing and further cicatrization. As such, at least half of these women will develop severe scar dyspareunia. Patients were also counselled that surgery is not, in itself, curative. Rather the objective of surgery is to remove sites of intractable pain messaging, so that the other elements of their pain loop can be resolved. Hence, all patients were instructed to continue with their biofeedback pelvic floor program until normal muscle tone had been restored. Of these 99 women, pain free intercourse was re-established in 93 (94%) patients, and another two (2%) had partial improvements. There was a 4% failure rate (four women). No major complications were encountered.

OPERATIVE TECHNIQUE FOR MICROSURGICAL REMOVAL OF THE BARTHOLINS' GLANDS

(a). Proper exposure of the operative field: The patient is prepped and draped in lithotomy position. A Lone Star retractor is applied and the frame anchored to the patient's body (by placing two sutures through the groin skin and tying them across the retractor screws) (Fig 1). An outer ring of six elastic hooks are applied at the level of the pilosebaceous line, to evert the vestibular and stabilise the surface epithelium. It is important not to insert hooks into the glaborous skin of the interlabial sulci or labia minora, as they will tear out.

(b). Staining of the Bartholin's ducts and glands: Using surgical loupes or an operating microscope, a search is made for the ostia of the Bartholin's ducts. If not immediately visible, gentle digital pressure over the Bartholin's fossa will often express a droplet of clear mucous from the ostia. Unfortunately, the ducts will have atrophied shut about one quarter of patients, making subsequent dissection much more difficult. If the ducts are still patent, the underlying gland can be made easier to locate and dissect by instilling a few drops of Bonney's blue or gentian violet through a paediatric intravenous cannula. It is vital not to use methylene blue as the staining agent, because this dye rapidly spreads across the gland capsule. End result will be diffuse staining of the surrounding tissues, thus totally masking all surgical landmarks. Once the ducts have been stained, the submucosal erectile tissue is carefully infiltrated with local anaesthetic and adrenaline. Because of the vascularity of this region, these injections should be very slow and the blood pressure must be watched carefully because of the risk of rapid systemic absorption.

(c). Making the incision: The sulcus just distal to the hymen on the side to be dissected first is circumscribed with either a focused carbon dioxide (CO_2) laser beam or a #15 blade. Depth of the incision is kept very shallow, to expose the shiny white and relatively tough fascia (Gallaudet's fascia) overlying the Bartholin's fossa. This is a key landmark that must be identified and cleanly incised, to facilitate accurate closure after gland removal. Failure to do this will cause unaesthetic introital gaping, even in young nulliparas.

(d). Establishing tension and countertension throughout the surgical field, to facilitate sharp dissection: It is impossible to microdissect a flaccid wound. Good technique for dissecting the Bartholin's fossa begins by retracting the

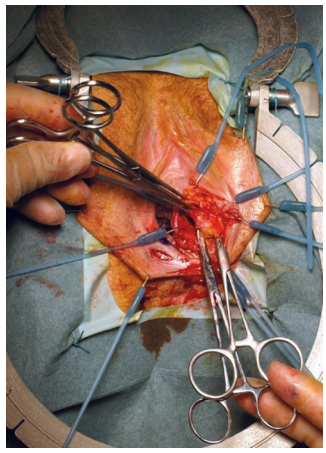


Figure 2. – Mobilizing the posterior pole of the Bartholin's gland.

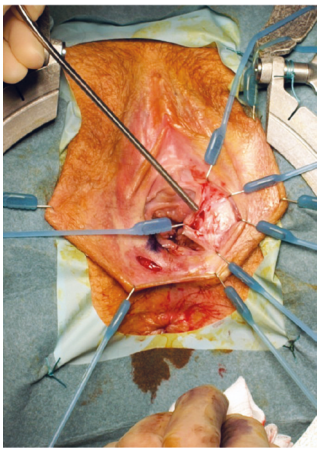


Figure 1. – Use of the Lone Star retractor to expose operating site and create tension-countertension.

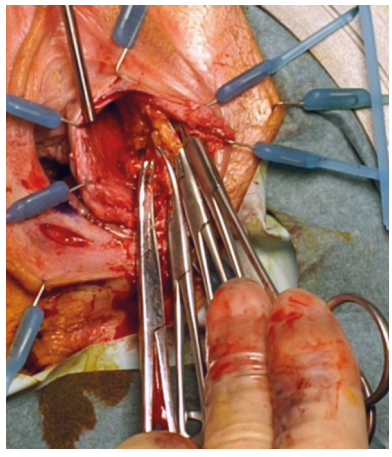


Figure 3. – Blood vessels clamped ready for suture ligation.

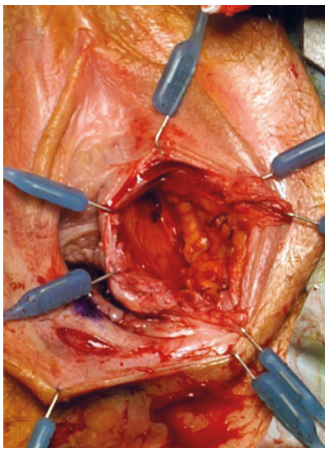


Figure 4. – Pedicles ligated, but Bartholin's fossa not yet closed.

mucosal incision margins with four more hooks. This second row of hooks widely exposes the Bartholin's fossa, providing unrivalled visibility and freeing the assistant to keep tis very vascular dissection dry with an ENT suction. More importantly, these four hooks create the tension-countertension needed for gland identification and removal.

(e). Identifying the Bartholin's glands: The mucosal incision is taken down through the thin superficial (Colles') fascia, to expose the point where the blue-stained Bartholin's ducts descend on the medial side of the gland, to penetrate the deep (Gallaudet's) fascia. Incising the thick Gallaudet's fascia exposes the inferior (superficial) surface of the Bartholin's glands. Key anatomic relations are as follows: Medial (investing fascia of vaginal wall); Lateral (bulbocavernosus muscle); Anterior: the gland is attached to the vestibular bodies; Posterior (the ischioanal fossa); Superior/deep (perineal membrane/inferior fascia of urogenital diaphragm and pubococcygeus muscle). The easiest identification point is the posterior pole, which can be grasped and elevated with a paediatric Allis forceps (Fig. 2). Note that an adult size is totally unsuited to this task. The gland is vascularized by small blood vessels that penetrate the capsule both medially and laterally. As such, there is a potential space between the superior (deep) surface of the gland and the perineal membrane, bounded medially and laterally by the vascular bundles. A closed haemostat can be slid into this space. The lack of resistance to the forward passage of the haemostat is highly characteristic, and provides confirmation that the gland has been correctly identified.

(f). Excising the Bartholin's glands: Mosquito forceps are inserted into the space between the perineal membrane and the deep surface of the gland, to clamp the lateral and medial vascular bundles emanating from the urogenital diaphragm (Fig. 3). After transection, these pedicles are suture ligated with Vicryl 000 on an 18 gauge round bodied needle (Fig. 4). Finally, the posterior part of the vestibular body is clamped and ligated, allowing gland removal. This technique will forestall significant bleeding. However, any dissection of erectile tissues may present some persistent oozing from other parts of the Bartholin's fossa. Haemostasis is improved by suturing together the adjacent Bartholin's fossa soft tissues, and completed by closing Gallaudet's fascia.

(g). Wound closure: In order to leave this important sexual anatomy essentially undisturbed, the edges of the Gallaudet's fascia incision must be accurately re-approximated with Vicryl 000 sutures (Fig. 5). This fascial closure must be done with interrupted stitches, because of the risk of ischaemia and consequent wound infection with a continuous suture. Not repairing the deep vulvar fascia will cause introital gaping, thus eroding some of the advantage that microsurgical gland removal has over vestibulectomy. Finally, the hymen must be excised before mucosal closure with 000 Vicryl Rapide. Preserving the hymen carries an unacceptable risk of painful introital scarring and a need for surgical revision.

DISCUSSION

Acute and most chronic pain seen in surgical practice is *nociceptive pain*. Some form of tissue damage (e.g., trauma, infection, malignancy) activates specialised pain receptors, resulting in the conduction of an impulse from the site of trauma to the interneurons in the dorsal horn. From here, the signal is transmitted to pain projection neurons in the brain stem, where these signals are recognised by the cortex as pain. Nociceptive pains usually have distinctive symptom patterns. Pain is typically sharp and highly localised. Severity is proportionate to the force of the initiating insult, and worsens steadily if tissue trauma continues to increase. A biopsy from a site of nociceptive pain will usually identify the responsible histological process. Histochemical studies have identified a specific inflammatory cascade involving prostaglandins, substance P and other vasoactive peptides. As such, pain can be relieved (to some extent) by corticosteroids, nonsteroidal anti-inflammatories and opiates. Although the initial trauma may provoke transient hyperaesthesia at the site of injury, nociceptive pain is associated with a minimum of sustained autonomic afferent activity. Finally, nociceptive pain always resolves when the eliciting process is removed.

There is, however, a completely different system of pain generation, known as *sympathetically-maintained pains (SMPs)* or *complex regional pain syndromes (CRPS)*. Although generally unfamiliar to gynaecologists, these neuropathic pain disorders are a major reason for referral to a pain clinic. SMPs generally arise as a disabling but idiosyncratic response to a trivial trauma. The pathophysiology involves pain dysregulation in both the sympathetic and central nervous systems, with likely genetic, inflammatory and psychological contributions[1,2,6,7]. SMPs are generally described as a diffuse burning pain which the patient cannot localize. Clinical features classically follow a regional (rather than a dermatomal or peripheral nerve) distribution and tend to favor the distal part of the affected region. Biopsy shows normal tissue, apart from nonspecific features like round cell infiltration, oedema, vascular ectasia and trophic changes. Unlike nociceptive pain, SMPs do not respond to steroids, nonsteroidal anti-inflammatories and

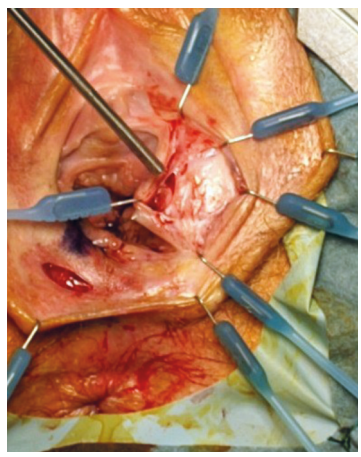


Figure 5. – Gallaudet's fascia closed, but vulvar skin edges still retracted.

opiates. However, neuropathic pains will often respond to antidepressants, anticonvulsants and sympathetic blockers, Another important contrast is the presence of diffuse autonomic dysfunction, such as hyperalgesia, allodynia, sudomotor and vasomotor abnormalities.

Pain severity is disproportionate to the degree of tissue injury and symptoms persist beyond the expected time for tissue healing.

The features of vulvodynia resemble an SMP rather than a nociceptive pain. Specifically, vulvodynia begins as a sudden idiosyncratic response to an often trivial tissue insult – such as a yeast infection or childbirth trauma. Once established, the syndrome manifests as a disabling, poorly localised pain centred on Bartholin's and Skene's glands. Vasomotor instability begins as patches of painful erythema on the vestibular surface, extending down into the gland itself in more severe cases. Oedema, hymenal fibrosis and fourchette contracture are common. In essence, vulvodynia is a self-sustaining pain loop (passing from vulvar sensory afferents to the dorsal horn neurons, then synapsing with the upper neuronal tracts, before terminating at pelvic floor muscles). Experience has shown that symptoms can be abolished by measures that interrupt any part of the pain loop. Prior to the discovery that the efferent arm of this pain loop could be suppressed with pelvic muscle relaxation therapies[9], we had successfully used the pulsed dye laser to disrupt the afferent arm[3]. Yellow (577 nm) light was used to selectively coagulate hyperemic vessels around the Bartholin's ducts, without damaging the vulvar epithelium. These painful vessels subsequently underwent vascular sclerosis, thus shutting down crosstalk between the hyperaesthetic perivascular sympathetic fibres and adjacent somatic afferents. During the course of this study, persistent inflammation deep within the Bartholin's gland was identified as the dominant factor in failed FEDL photothermolysis therapy.

CONCLUSIONS

Bartholin's gland removal is a technically difficult operation, but one with low morbidity and excellent success rate. It works on a similar principle to vestibulectomy (namely, by decreasing the intensity of chronic pain signaling generated from the inflamed vulvar skin and vestibular glands).

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