A PSYCHOPHYSIOLOGICAL PERSPECTIVE ON VULVODYNIA

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Thesis submitted in fulfilment of the requirements for the Degree of Doctor of Philosophy

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Jantos, M. (2009). Electromyographic assessment of female pelvic floor disorders. In *Cram's*

Introduction to Surface Electromyography (2nd edition). Sudbury, MA: Jones & Bartlett,

(In Press).

Abbreviations

BDI   Beck Depression Inventory

BMI   Body Mass Index

DSM   Diagnostic and Statistical Manual of Mental Disorders

fMRI   functional Magnetic Resonance Imaging

GSI   Global Severity Index

IASP   International Association for the Study of Pain

IBS   Irritable Bowel Syndrome

IBQ   Illness Behaviour Questionnaire

IC   Interstitial cystitis

ISSVD   International Society for the Study of Vulvovaginal Disease

MUI   Mixed urinary incontinence

NRS   Numerical Rating Scale

NVA   National Vulvodynia Association

PFM   Pelvic floor muscles

PSDI   Positive Symptom Total

QST   Quantitative sensory testing

SCL-90-R   Symptom Checklist-90 Revised

SEMG   Surface electromyography

SUI   Stress urinary incontinence

UI   Urinary incontinence

UUI   Urge urinary incontinence

Declaration

Name: Marek Jantos   Program: Doctor of Philosophy

I hereby declare that this submission is my own work, and that, to the best of my knowledge and belief, it contains no material which has been accepted for the award of any other degree or diploma in any other tertiary institution and, to the best of my knowledge and belief, contains no material previously published or written by another person, except where due reference has been made in the text.

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<center>**Preamble**</center>

The study of human sexuality opens an intriguing window into human existence and the individual's sense of wellbeing. From a population standpoint, sexuality is an integral part of human reproduction, but from the perspective of the individual, sexuality can also be a source of pleasure and the basis of interpersonal bonding and happiness (O'Donohue & Geer, 1993). Sexuality crosses many dimensions of the human experience, any difficulties encountered in the expression of emotional and physical intimacy can have a wide and multidimensional impact (Basson, 2005).

Vulvodynia is a prevalent chronic pain condition in women affecting the lower urogenital tract (Moyal-Barracco & Lynch, 2003; Harlow & Stewart, 2003). It interferes with an array of activities, including sexual intimacy, and compromises quality of life (Sandownik, 2000; Jantos & Burns, 2007; Arnold et al., 2006). The disorder can significantly undermine self-esteem, impacts on relationships, and engenders significant psychological distress in the form of anxiety and depression (Jantos & White 1997; Masheb et al., 2005; Jantos & Burns, 2007; Jantos, 2008). Various causes have been theorized and many treatments proposed, but there is no consensus on its classification and management (Masheb et al., 2000; Bachmann et al., 2006). The practical and theoretical implications of a proper understanding of the disorder and its accurate classification are important (Lynch, 2008; Binik, 2005).

Structure and outline of the thesis

This thesis consists of three publications which include two research studies (Chapter 3 & 4) and a discussion paper (Chapter 5). The two research papers assess the psychosexual and psychophysiological profile of a cohort of women diagnosed with vulvodynia and the discussion paper focuses on conservative management of vulvodynia and other conditions linked to pelvic floor dysfunction.

As an introduction to the three publications, the first chapter discusses the role of sexuality in general wellbeing. It provides a historical background to the study of vulvar pain and introduces classification systems that address the issue of painful sexual intercourse. It also reviews current perspectives on the nature of the disorder, its prevalence, etiology and management.

Chapter 2 outlines the rationale, aims and structure of the research studies. Vulvodynia is a poorly understood but prevalent disorder that has been identified as a neglected women's health problem (Binik, 2003; Haefner et al., 2000). Therefore, further information is needed about the population affected by the disorder, about the impact on wellbeing, and about the potential mechanisms mediating symptoms of pain, so that protocols can be developed for its effective management.

Chapters 3, 4 and 5 are reprints of the three publications forming the core of this thesis. Chapter 3, entitled *Vulvodynia: Development of a Psychosexual Profile,* is a retrospective study of seven hundred and forty-four patients. It examines the age of symptom onset, the age distribution of the clinical cohort and the age related risk of developing the condition. The study identifies a range of psychosexual changes associated with the disorder. Chapter 4, entitled *Vulvodynia: A Psychophysiological Profile Based on Electromyographic Assessment,* is also a retrospective study. It is based on five hundred and twenty-nine patient cases and sought to assess the location and temporal characteristics of chronic vulvar pain, the relationship between psychological and physiological variables in the experience of pain, and the role of dysfunctional pelvic muscles in mediating the symptoms of vulvodynia. Both of these research studies make an original contribution to the current understanding of vulvodynia and present data derived from the largest clinical sample studied to date.

Chapter 5 entitled *Electromyographic Assessment of Female Pelvic Floor Disorders,* is a review of literature examining pelvic floor anatomy and physiology and views pelvic muscle dysfunction as the primary mechanism mediating a spectrum of pelvic disorders, including vulvodynia. It reviews research on the applications of surface electromyography (SEMG) in relation

to vulvodynia and a range of pelvic disorders associated with both hypertonic and hypotonic muscle states. Guidelines are suggested for conservative management of these disorders.

Chapter 6 discusses the contribution of the three publications to the current understanding of vulvodynia and draws on the research findings of this thesis, and other literature, to arrive at several key recommendations in relation to its classification and management.

The primary recommendation of the thesis is that vulvodynia should be seen as a chronic pain syndrome rather than a psychiatric disorder or a sexual dysfunction and that the management of pain should be the foremost priority.

A Psychophysiological Perspective on Vulvodynia

Introduction

This chapter introduces some of the current issues in the study of vulvodynia. Vulvodynia, as a form of chronic vulvar pain, is a poorly understood and negelected area of women's health. There is considerable controversy in relation to the classification of chronic vulvar pain and a lack of consensus in regards to its management. The chapter will review a range of issues pertinent to the understanding of vulvodynia.

Centrality of sexuality to wellbeing

When asked about the importance of sexuality, 94% of adults indicated that sexual pleasure significantly adds to their quality of life (Marwick, 1999). Yet, the prevalence of sexual problems in the general population and the level of personal dissatisfaction with sexual wellbeing is high (Phillips, 2000).

If sexual pleasure is indeed so fundamental to happiness and quality of life, it is not surprising to find that almost 90% of women, diagnosed with vulvodynia, attend therapy, motivated by the desire to increase the frequency of sexual activity in their relationships (Jantos & Burns, 2008). Without diagnosis and assistance, vulvodynia patients and their partners experience higher levels of sadness, depression and frustration (Jantos & White, 1997; Jantos & Burns, 2007; Desrosiers et al., 2008). Research consistently highlights the fact that vulvodynia significantly undermines the quality of life of women and couples (Arnold et al., 2006, Sargent & O'Callaghan, 2007). In comparison with other vulvar medical problems, the impact of chronic vulvar pain on general wellbeing and sexual function far exceeds that of other problems (Ponte et al., 2009) and is more disabling than other pelvic pain conditions (Meana et al., 1997; Reed et al., 2000). The disorder

diminishes a women's sense of wellness, impacts on relationships and gives rise to isolation and loneliness (Sargeant & O'Callaghan, 2007; Desrosiers et al., 2008; Jantos & Burns, 2007).

In 1975 the World Health Organization defined sexual health as the "integration of the somatic, emotional, intellectual, and social aspects in ways that are positively enriching and that will enhance personality, communication and love" (World Health Organization, 1975, p. 2). Yet, sexual health is one of the last frontiers of wellness to be studied and one of the last disciplines of human physiology to be scientifically investigated (Markos, 2005). The psychological, physiological, relational and social threads that weave together in human sexuality are often overlooked (Schrover & Jensen, 1988). The centrality of sexual health to wellbeing needs to be further recognized within the health care system (Parish & Clayton, 2007).

Assessment of Sexual health

Sexual health can be easily overlooked in health training and medical practice. In part this may be due to patients and doctors being hesitant to communicate about sexual issues, fearing that raising matters of sexuality may cause the other party embarrassment. To avoid the risk, sexual health assessment can be easily neglected.

The need to address sexual health matters, especially in relation to women's sexuality, is highlighted by the prevalence of problems reported in research studies. Estimates of sexual difficulties among women range from 19-50% in "normal" patient populations, and increase to 68-75% when sexual dissatisfaction is included (Phillips, 2000). The high prevalence of sexual problems noted in survey samples is not reflected in patient notes and medical reports. In one study, general practitioners had recorded sexual problems in only 2% of their case notes; while in another study, where physicians were trained to take a sexual history, 53% of patients were noted as having problems. It is evident that when clinicians make inquiries of patients about their sexual health, the prevalence of reported problems increases significantly. In order to detect patient concerns and

difficulties, explicit questions need to be asked by the clinician during routine health assessments (Schultz et al., 2005).

The most common female problems identified in surveys relate to: low desire (77%); low sexual arousal (62%); inability or difficulty achieving orgasm (56%); and vaginal dryness (46%) (Berman et al 2003). The specialists most frequently approached with these problems were: gynecologists (42%); general practitioners (24%); psychiatrists (12%); and urologists (3%) (Berman et al., 2003).

Several surveys have sought to assess the extent of the health care provider's involvement in assessing sexual wellbeing. Findings indicate that both patients and health care providers expressed an unwillingness to raise matters of sexual importance. In a survey of almost four thousand women, 40% expressed a reluctance to seek help from a physician in relation to sexual complaints, even though 54% expressed a desire to do so (Berman et al., 2003). In exploring reasons why patients fail to raise sexual issues, a study found that 75% believed that their physician would dismiss their sexual health concerns, or that such issues would embarrass them (Marwick, 1999). These beliefs appear to be validated, in part, by reports showing that, when patients raised concerns about their sexual health, the physician was unprepared to hear them and they were met with embarrassed silence, misinformation, surprised or shocked reactions, personal discounting or belittling (Berman et al., 2003).

The health care providers, on their part, also cite various reasons for avoiding sexual issues during medical screening. The most common of these include: lack of training, insufficient knowledge, lack of information about treatment options, discomfort with sexual language, apprehension that inquiries of a personal nature may offend the patient, and their own personal feelings of embarrassment (Parish & Clayton, 2007). Yet, 91% of patients were of the view that questions about sexuality were appropriate in the context of health care (Parish & Clayton, 2007).

With such misgivings on the part of the patient and the physician, sexual health issues are neglected and very few doctors ever take a patient's sexual health history (Parish & Clayton, 2007). From a health perspective, such unease is not conducive to the early identification of chronic pain disorders such as vulvodynia (Nuns & Mandal, 1996). Physicians and allied health professionals need to be more proactive in creating an environment where the patient's sexual wellbeing can be discussed (Phillips, 2000).

A multidisciplinary approach to the study of vulvodynia

With pain as its primary symptom, vulvodynia has perplexed medical and allied health professionals for more than a hundred years. To date a multidisciplinary approach to the study of the disorder has enriched discussion and encouraged productive controversy (Binik, 2005). Such controversy has been unavoidable given the nature of the topics involved.

Because vulvodynia is defined as an idiopathic pain condition in which the absence of pathology is a defining feature (Moyal-Borracco & Lynch, 2003), it generates considerable debate regarding its etiologies, management and classification. In other chronic pain conditions the lack of pathology is a recognized anomaly and a lack of association between visible pathology, pain severity and disability is not questioned (Steege et al., 1997; Jantos, 2007). However, in relation to urogenital pain, such an absence is viewed with suspicion. As a result, some regard vulvodynia as a sexual dysfunction (APA, 2000; Basson, 2005), while others see it as a somatoform disorder (Dobson & Friendrich, 1978; Lynch, 2008). However, a growing number of researchers and clinicians view it as pain syndrome (Binik et al., 1999; Haefner et al., 2000; Pukall et al., 2003). How the condition is perceived and classified will ultimately influence the way it is managed and will affect the selection of the primary case manager, be it a pain therapist, a sex therapist or a physician (Binik, 2005).

Because chronic vulvar pain impacts upon many aspects of the individual's wellbeing, the discussion of etiology, classification and management inherently involves discussion of the complexities of human sexuality, of chronic pain, and calls for a multidisciplinary outlook (Steege et

al., 1998; Sandownik, 2000; Innamaa & Nunns, 2005). To appreciate the need for a multidisciplinary approach, it is important to briefly reflect on the intricate nature of human sexuality and the complexity of chronic pain.

In relation to sexuality, the human sexual response is an ultimate example of the blending of mind and body and requires more than a discussion of biology and psychology (Schultz et al., 2005). Sexuality consists of an intricate interaction between physiology, emotions, attitudes, values and lifestyle, and requires a detailed scientific approach (Phillips, 2000). To illustrate, at the physiological level it is coordinated by subtle neurologic, muscular, vascular and endocrine system changes (Bachmann & Phillips, 1998). At the emotional level it is associated with self-esteem, personal fulfillment, happiness and interpersonal bonding. At the social level, values, norms and lifestyle practices shape the way in which the most intimate affections find expression (Phillips, 2000). If sexuality, as an idiom for the expression of emotional and physical intimacy is thwarted, the ramifications for personal wellbeing and for relationships are wide-ranging (Sargeant & O'Callaghan, 2007; Desrosiers et al., 2008). Many of these multidimensional aspects of sexuality have been examined in relation to vulvodynia and generate diverse and often inconsistent findings. Some of these will be reviewed in this chapter.

The complexity of studying chronic pain also brings together contributions from many disciplines, each sharing a unique perspective on the topic. Chronic pain is a far more complex phenomenon than acute pain (Steege, 1998; Merskey & Bogduk, 1994). Aristotle considered pain to be a passion of the soul, characterized by a mingling of sensation and emotion (Blackwell, 1989). The modern definition of pain maintains the Aristotelian view defining pain as an "unpleasant sensory and emotional experience"(Merskey & Bogduck, 1994). The word pain derives from the Greek term *poine,* meaning punishment (Blackwell, 1989). In ancient times pain was perceived as a punishment from the gods. In modern times vulvodynia patients are often burdened by guilt, stemming from their own perception that their pain is a form of punishment (Jantos & White, 1997). Hence, the experience of pain is associated with significant emotional distress, anxiety and depression (Jantos &

Burns, 2007). The sensory and emotional aspects of pain need to be recognized and studied in a scientific and multidisciplinary manner. Clinicians who tend to view vulvodynia as an expression of emotional tension or psychosexual conflict, inadvertently reinforce certain misconceptions about chronic pain that add to the patient's distress and may be counterproductive in restoring the individual's sense of wellbeing (Reed et al., 2000; Kaler, 2005).

When the themes of sexuality and chronic pain are brought together, as in the study of vulvodynia, the complexity increases, creating a multifaceted and composite discussion, which often lacks any general consensus. As a result, some have advocated the formation of a new multidisciplinary subspecialty of sexual medicine (Goldstein, 2007), while others have proposed the establishment of a new science of vulvology, with a specific focus on the study of the vulva (Micheletti et al., 2002).

This thesis contributes towards a multidisciplinary approach to the study of vulvodynia and approaches the topic from a psychophysiological perspective. In recognizing pain, as a "sensory and emotional experience" (Merskley & Bogduk, 1994), the thesis seeks to avoid the traditional dualistic perspective in which the physical and the emotional aspects are seen as separate entities and instead examines their unique interaction, as reflected in the experience of the patient.

Historical background to the study of vulvodynia

From ancient Egyptian papayri, to the 1st century writings of Soranus of Ephesus, antiquity provides very early accounts of female dyspareunia akin to modern day vulvodynia (McElhiney et al., 2005). In more recent times, the medical literature of the 18th and 19th century again documents the existence of chronic vulvar pain, which was reported to cause great discomfort and distress in some female gynaecology patients. In 1874, T. Gilliard Thomas in his book *A Practical Treatise on the Diseases of Women* describes hyperesthesia of the vulva with patients experiencing "excessive hypersensibility of the nerves supplying the mucous membrane of some portion of the vulva" (Thomas, 1874, p. 115). In 1889, Skene in his *Treatise on the Diseases of Women*, described a

condition of chronic pain characterised by pain on touch and examination of the vulva (Skene, 1889) and in 1928, Kelly, in his book *Gynaecology*, reported the occurrence of tender areas close to the hymenal ring sufficient to cause a patient to cry out aloud (Kelly, 1928).

In 1976, Weisfogel, speaking at a congress of the International Society for the Study of Vulvovaginal Diseases (ISSVD), introduced a disorder, primarily affecting young women, who complained of a problem described as *"the burning vulva."* Physical examination and laboratory testing showed no evident pathology. Weisfogel described the condition as one in which "I see nothing. I hear nothing. I smell nothing. I feel nothing" (Moyal-Barracco & Lynch, 2003). The ISSVD meeting discussed the problem of the "*burning vulva syndrome*" as the symptoms were reported to be resistant to a wide range of medications (Young et al., 1984). The same year, Pelisse and Hewitt, provided a review of 30 women with hypersensitivity of the vulva (Pelisse & Hewitt, 1976). In 1978, Tovell and Young suggested that the terms *"vulvodynia"* or *"pudendagra"* might best describe the localized sensation of burning reported by women (Tovell & Young, 1978). At the same time, Dodson and Friedrich described a form of vulvovaginal pain and dyspareunia, and named it *"psychosomatic vulvovaginitis"* (Dodson & Friedrich, 1978). In 1982, a task force was established to survey ISSVD membership views concerning the burning vulvar syndrome (Young et al., 1984). Two-thirds of the respondents put forward a range of ideas regarding etiology, suggesting psychogenic, neurogenic, infectious and dermatological causes of the disorder. At the 1983 congress, the ISSVD task force proposed the term *vulvodynia*, to describe this idiopathic form of vulvar discomfort, most often characterised by patient's complaints of burning. The same year, Woodruff and Parmley gave account of 15 patients with infection of the minor vestibular glands causing pain (Woodruff & Parmley, 1983) and in 1986, Peckham and others, reported on 67 women with a condition they called *"focal vulvitis"* (Peckham et al., 1986). In general, *vulvodynia* and the *burning vulva syndrome* were seen as symptomatic of an "*end-stage"* condition that was recalcitrant to most treatments and psychogenic factors were thought to be strongly implicated but not well defined (Young et al., 1984).

In 1987, Friedrich, on the basis of his study of 86 patients, offered the first classification describing the condition of "*vulvar vestibulitis syndrome*" (Friedrich, 1987). The classification suggested by Friedrich was the first to systematize the diagnosis of vulvar pain, providing the basis for further discussion and future comparative studies. Friedrich's criteria were widely accepted and still continue to be used for diagnostic and research purposes (Payne et al., 2007). According to his classification, the syndrome was characterised by three key diagnostic features:

- pain on penetration (entry dyspareunia),

- introital tenderness, and

- mild to moderate introital erythema.

Patients suffering symptoms would commonly describe their discomfort as a sensation of burning, rawness, and discomfort. These symptoms would occur in the absence of any visible clinical or neurological findings. The term *vulvar vestibulitis syndrome* was not only widely accepted but was often used synonymously with vulvodynia.

By 1988, several subtypes of vulvodynia were proposed (McKay, 1988) and were well summarized in a review of then current concepts in vulvodynia (Masheb et al., 2000). The first subtype was vulvar dermatoses (lichen sclerosus, lichen planus, chronic dermatitis, and eczema); the second subtype was cyclic candidiasis/vulvitis, often associated with recurrent candida infections; the third subtype was vulvar papiliomatosis, confirmed by the presence of small papillae found around the vulvar vestibule and possibly associated with human papillomavirus (HPV); the fourth subtype was essential vulvodynia, a form of dysesthesia arising from nerve irritation similar to postherpatic neuralgia; and the fifth subtype was vulvar vestibulitis syndrome (VVS), characterised by hypersensitivity in the vulvar vestibule, but occurring in the absence of physical findings, with the exception of varying degrees of erythema. Later classifications separated chronic vulvar pain into two categories, a category listing vulvar pain due to identifiable medical causes and a category listing types of vulvodynia occurring in the absence of any know medical causes.

In 1991 the ISSVD made further changes and replaced the term "*burning vulva syndrome*" with *"vulvodynia"*. Recognizing the existence of various subsets of chronic vulvar pain, some too difficult to classify, the ISSVD added two additional terms, *essential vulvodynia* and *idiopathic vulvodynia* to describe patients who showed no significant changes on physical examination and for patients reporting a dull continuous pain, originating from areas deeper than the vestibule but with no medical diagnosis (McKay et al., 1991). Discussions on terminology and classification continued through the 1990's until the 2003 classification system was established. In 2003, at the 17th Congress of the ISSVD, the membership of the society voted to accept a reversion to the term *vulvodynia,* and in 2004, the current terminology and classification of vulvodynia was published (Moyal-Barracco & Lynch, 2004).

Classification of lower tract urogenital pain

The development of classification systems for medical diseases and disorders has been an ever evolving phenomenon (Levine, 2005). The classificatrion of chronic vulvar pain illustrates this process. The purpose of classifications is to provide a framework for identifying and grouping conditions, facilitating communication, both spoken and written, and guiding clinical practice as well as research (Merskey & Bogduk, 1994; APA, 2000). To achieve this goal, classification needs to accurately reflect current knowledge in a consistent and reliable manner (Merskey & Bogdul, 1994). However, if the diagnostic classification is erroneous or misleading, or if the foundations of its classification are faulty, treatment will be compromised and research misdirected (Klienplatz, 2005).

Uncertainty in relation to the classification of vulvodynia continues to the present day. To date three perspectives on the classification of vulvodynia have emerged: that of a sexual dysfunction (Basson, 2005; APA, 2000), a somatoform disorder (Mascherpa et al., 2007, Lynch, 2008), and that of a chronic pain syndrome (Binik et al., 1999, Pukall et al., 2003). These perspectives are linked, in varying degrees, to the following classification systems:

- The American Psychiatric Association's Diagnostic and Statistical Manual of Mental Disorders (APA, 2000) classification of pain under sexual disorders and somatoform disorder.

- The International Association for the Study of Pain (IASP), Multi-Axial Classification of Chronic Pain (Merkesley & Bogduk, 1994).

- The International Society for the Study of Vulvovaginal Disease (ISSVD), Terminology and Classification of Vulvodynia (Moyal-Barraco & Lynch, 2003).

Each of these diagnostic systems will now be discussed briefly.

<u>The DSM System</u>

The American Psychiatric Associations Diagnostic and Statistical Manual (DSM) classifies urogenital pain under Sexual Dysfunction or Somatoform Disorders. The text of the fourth edition of the DSM-Text Revision (DSM-IV-TR) version will form the basis of the discussion in this chapter (APA, 2000). Unless otherwise stated, all references will be made to the DSM-IV-TR, but referred to by the simplified acronym of DSM.

The DSM makes reference to two sexual pain disorders; *dyspareunia*, which is characterized by recurrent or persistent genital pain associated with sexual intercourse; and *vaginismus*, which is defined as a recurrent or persistent involuntary spasm of the pelvic musculature interfering with sexual intercourse (APA, 2000). Both of these conditions are discussed in the *Sexual Pain Disorders* section of the chapter on *Sexual and Gender Identity Disorders.*

In the DSM, the experience of pain in conjunction with sexual intercourse is most commonly classified as a sexual dysfunction. A sexual dysfunction refers to any "disturbances in sexual desire and in the psychophysiological changes that characterize the sexual response cycle and cause marked distress and interpersonal difficulty" (APA, p. 535). From the DSM perspective, sexual dysfunction is a disturbance in *sexual desire* and *sexual response*. This creates a conceptual problem, because sexual desire and sexual response in vulvodynia patients is no different to that of non-pain patients (Payne et al., 2007; Reed et al., 2000; Meana et al., 1997; Van Lankveld et al., 1996). It is the

disabling impact of pain that makes intercourse difficult, if not impossible. Furthermore, as has been suggested elsewhere (Phillips, 2000; Marthol & Hilz, 2004), the most common causes of sexual dysfunction include psychological, pharmaceutical, medical, and physiological factors, which can be summarized as follows:

- *Psychological* factors: intrapersonal conflicts arising from religious practices, guilt or social restrictions; historical factors including past or current abuse, rape, sexual inexperience; interpersonal conflict in the form of relationship difficulties, extra-marital affairs, abuse, desire differences, sexual communication problems; and life stressors arising from illness, depression, finances, family or job problems (Philips, 2000).

- *Pharmaceutical* factors: medications that cause disorders of desire, psychoactive, antihypertensive, and hormonal preparations; medications that cause disorders of arousal, including anticholinergics, antihistamines, antihypertensives and psychoactives; medications that cause orgasmic dysfunction, such as tricyclic antidepressants, amphetamines, anorexic drugs, antipsychotics, narcotics and others (Phillips, 2000).

- *Medical conditions*: a wide range of illnesses such as cancer, diabetes, gynecologic surgery, and procedures such as chemotherapy, radiation therapy and others (Phillips, 2000)

- *Physiological changes*: including changes due to menopause, organ atrophy and muscle weakness. These changes can be lifelong, acquired, generalized or situational (Phillips, 2000).

These causes of sexual dysfunction, except in individual cases, have not been shown to be linked to vulvodynia and are not seen as relevant. The only characteristics that distinguish vulvodynia patients from controls and other chronic pelvic pain patients are pain sensitivity during sexual arousal, general severity of pain and the level of disability caused by pain (Payne et al., 2007; Reed et al., 2000; Meana et al., 1997).

Earlier versions of the DMS lacked clarity in relation to the specific role of pain in sexual dysfunction (First, 2005). Subsequently, the Text Revision (TR) qualifies the definition of sexual dysfunction by stating that a "sexual dysfunction is characterized by a disturbance in the processes

that characterize the sexual response cycle *or by pain associated with sexual intercourse"* (APA, p. 535, italics added). Without this qualification, vulvodynia could not be considered a sexual dysfunction, since it does not directly affect any particular phase of the sexual response cycle (First, 2005; Schultz et al., 2005). Pain was identified as a separate cause of sexual dysfunction, quite apart from any other factors disrupting the sexual response cycle. The issue that remains unclear in the DSM classification is how pain, caused by sexual intercourse, can be the defining characteristic of a sexual dysfunction. It is the pain that causes the sexual dysfunction and not the reverse (White & Jantos, 1998; Pukall et al., 2003). As stated elsewhere, "it is tautological to define the pain as the symptom which causes pain" (Moser, 2005). To illustrate the point, it has been argued that lower back pain prevents people from working, but it is not appropriate to conceptualize back pain as a work disorder or a work pain (Pukall et al., 2003).

Finally, in order for a sexual dysfunction to meet the DSM classification criteria, it must cause marked distress and interpersonal difficulty. Vulvodynia, without a doubt, is a source of distress (Jantos, 2008; Pukall et al., 2002), impacting significantly on quality of life and on interpersonal relationships (Arnold et al., 2006; Desrosiers et al., 2008) but is not necessarily the cause of interpersonal difficulty or the result of relationship issues (Van Lankveld et al., 1996; Reed et al., 2000; Jantos & Burns, 2007).

Female sexual dysfunction in the DSM system is generally divided into four categories which are not mutually exclusive:

- Hypoactive Sexual Desire Disorder – a low sexual desire – characterized by diminished interest in sexual activity.

- Female Sexual Arousal Disorder – reflected in an inability to become aroused or maintain arousal during sexual activity.

- Female Orgasmic Disorder – evidenced by a persistent or recurrent difficulty in achieving orgasm following a period of arousal and stimulation.

- Sexual Pain Disorder – where pain is experienced in the urogenital area during sexual arousal or stimulation.

The two sexual pain disorders listed in the DSM are *dyspareunia* and *vaginismus*. *Vaginismus* is defined as an involuntary spasm of the muscles surrounding the outer third of the vagina when intercourse is attempted. *Dyspareunia* (from the Greek term for painful mating) is identified as a pain disorder, with no specific cause listed. These two pain disorders are further subclassified as *Due to Psychological Factors* or *Due to Combined Factors* (APA, p. 537). Pain can also be classified in the category of *Sexual Dysfunction Due to a General Medical Condition* (APA, p. 558).

Numerous psychological and medical conditions have been identified as possible causes of dyspareunia (Schultz et al., 2005), but the DSM provides few options for differentiating between causes of pain and provides limited inclusion and exclusion criteria (Meana et al., 1997; Pukall et al., 2003). By contrast, the current ISSVD definition of vulvodynia specifically states that it is a condition "occurring in the absence of relevant visible findings, or a specific, clinically identifiable, neurologic disorder," and cannot be subtyped under any category for which there is a medical cause (Moyal-Barracco & Lynch, 2003; Lynch, 2008). In light of these exclusions, vulvodynia should not be classified as a sexual dysfunction due to medical causes, or combined factors (psychological and medical). According to the DMS this would leave only one remaining option, namely, to classify the disorder as being due to psychological factors. If that were to be the case all available evidence needs to be weighed to determine if indeed the etiology of vulvodynia is linked with psychological causes.

The DSM classification has been questioned for several other reasons as well. One of the major criticisms of its classification of sexual disorders is that it provides only descriptive information which is not based on empirical evidence (First, 2005; Binik, 2005). The descriptive and pragmatic nature of the classification system has been called into question by a range of research findings and is generally seen as lacking validity (Kaler, 2005; Kleinplatz, 2005; Moser, 2005). This is clearly illustrated in reference to vaginismus and dyspareunia.

The DSM classification lists vaginismus and dyspareunia as two sexual pain disorders. However, evidence shows that the distinction between vaginismus and dyspareunia is difficult to maintain (Binik et al., 2000; Binik, 2005; Ter Kuile et al., 2005; deKruiff et al., 2000; Binik et al., 1999; O'Donohue, 1993). Studies have shown that more than 50% of women presenting with primary

vaginismus also meet the criteria for vulvodynia (Ter Kuile et al., 2005; deKruiff et al., 2000).

Furthermore, in defining vaginismus as a "recurrent and persistent involuntary spasm of the musculature of the outer third of the vagina that interferes with sexual intercourse" (APA, 2000, p. 558), the diagnostic criteria do not specify that pain must be present, even though it is classified as a pain disorder. The DSM definition of vaginismus makes an assumption about the cause of pain. The spasm oriented diagnostic criteria for vaginismus have been widely questioned. Studies utilizing SEMG, have consistently failed to differentiate between normal controls and vaginismic women on the basis of muscle tension or spasm (Van der Velde & Everaerd, 1999; Van der Velde et al., 2001; Van der Velde & Everaerd, 2001). These studies showed no involuntary spasm in vaginismic women, who demonstrated equally good control of pelvic muscles, when compared with controls. The DSM provides no justification of why dyspareunia and vaginismus are listed as separate pain disorders, or why these pain conditions have been designated as sexual dysfunctions (Moser, 2005). The evidence raises questions about the validity of the DSM's classification of sexual pain disorders (Binik, 2005) and other definitions of these disorders have been proposed (Basson et al., 2003).

Another section of the DMS relevant to vulvodynia is the section on Pain Disorder, in the chapter on Somatoform Disorder. This section of the DSM groups all physical manifestations of psychiatric disorders under Somatoform Disorders (Moser, 2005). To avoid the psychoanalytic overtures of the previous editions of the DSM, disorders previously seen as linked to hysteria, were grouped together on the basis of the predominant symptom of pain. Pain conditions, whether acute or chronic, were subtyped on the basis of factors that were seen to play a major role in its etiology. In the Somatoform Disorders section, three options exist: "*Pain Disorder Associated With Psychological Factors*": "*Pain Disorder Associated with Both Psychological and a General Medical Condition*"; and "*Pain Disorder Associated With a General Medical Condition*". Where psychological factors directly contribute to onset of pain the first two subclassifications are relevant. If pain is primarily due to a general medical condition, but psychological factors are present, they are not judged to have a major role in its onset, severity or maintenance. Medical conditions such as musculoskeletal problems, disc herniation, malignancies and neuropathies, are typical causes of pain

due to medical cause. These conditions are judged not to arise from a "mental disorder" (APA, 2000, p. 499). Again, according to the ISSVD definition, if medical causes can be identified, they immediately exclude the diagnosis of vulvodynia. The only subtype remaining is *Pain Disorder Associated with Psychological Factors*. In this section of the DSM, there is no listing of any specific pain conditions, but the criteria apply where pain is shown to have a psychosomatic origin. According to DSM (IV-TR), the diagnosis of Pain Disorder is appropriate where "…psychological factors are judged to have a major role in the onset, severity and exacerbation, or maintenance of the pain" (APA, 2000, p. 499). If vulvodynia is to be classified as a pain disorder, emotional factors would need to be identified as having a primary role in its etiology (Moser, 2005). No such findings have been made to date.

From a psychiatric perspective, some pain conditions are seen as secondary to Axis I disorders (psychiatric disorders), or to an Axis III conditions (general medical conditions), providing there is a temporal relationship between the pain and potential triggers. However, in relation to chronic pain;

> "when chronic pain is the central or predominant feature, it fits somewhat uneasily into the framework... Because of its private and inferred nature, the frequent lack of demonstrable pathology, and its often obscure relationship to emotional stimuli, chronic pain does not fall readily under the traditional psychosomatic rubric...it is unlike other somatoform disorders, although that is the rubric under which it is currently classified." (Blackwell, 1989, p. 1267)

Classification of chronic vulvar pain under the rubric of a somatoform disorder is often justified on the basis of a patient's persistent search for a physical diagnosis, their rejection of psychological explanations, and their sexually dysfunctional status (Dodson & Friedrick 1978; Lynch, 2008). Yet, the search for a diagnosis, and the denial of psychological causes, are both features of patient behaviour that constitute appropriate efforts to resolve a health dilemma and in themselves do not warrant psychiatric labelling (Blackwell, 1989). Suggestions that sexual pain may be a form of anger displacement were also not supported by empirical evidence (Meana & Binik, 1994), which, leads to the conclusion that, "the explanation of sexual pain as a type of somatisation also has its roots in

psychoanalytic theory and also has no empirical support (Binik et al., 1999). Until psychological factors are shown to have a major role in the onset of vulvodynia, its classification as a somatoform disorder is not justified.

It is evident that if vulvodynia is to be classified within the DSM framework, irrespective of whether it is subsumed under the sexual dysfunctions section, or under the pain disorder section, the primary cause of symptom onset, severity and maintenance, would need to be psychological. If indeed it can be shown to be psychological, than such a reclassification would have significant social and clinical implications. Thirty years ago, it was argued that vulvodynia was a psychosomatic pain disorder (Dodson & Friedrich, 1978). This argument was recently restated (Lynch, 2008). However, the research presented in this thesis and the research findings reviewed in the literature do not appear to support such a view. The relevance of the DSM to vulvodynia is questionable, unless the cause can be shown to be predominantly psychological. The DSM lacks empirical support and theoretical rational. As a result other classification systems have been suggested (Basson et al., 2003), with the specific purpose to "aid future research on women's sexual dysfunctions by better delineating the clinical realities of women's sexuality and by helping clinicians to minimize inappropriate classification and pathologizing of women" (Meston & Bradford, 2007).

The IASP multi-axial pain system

The most comprehensive classification of chronic pain was developed by the IASP (Mersky & Bogduk, 1994). This system classifies pain on the basis of body region, body system involved, pain characteristics, intensity and etiology. It lists the known generalized and localized pain syndromes (visceral, muscular, spinal and radicular) and includes pelvic and genital pain in its 700 and 800 code sections. The symptom characteristics in pain syndromes are rated along five axes:

- Axis I: Regions: Identifying the main sites of the body affected by the pain (e.g., genital region).

- Axis II: Systems: Identifying the physiological system whose abnormal functioning produces the pain (e.g., musculoskeletal, cutaneous, or nervous system).

- Axis III: Temporal characteristics of pain and patterns of occurance (e.g., continuous, intermittent, recurring, or irregular).

- Axis IV: Patient statement of intensity and time since onset of pain (e.g., mild or severe, of 6-12 months duration).

- Axis V: Etiology: Identifying potential antecedents to pain (e.g., infection, physical trauma).

The IASP classification system is the most comprehensive taxonomy of chronic pain syndromes. Each category is rated on a nine point scale (0-8). This provides a five digit code which classifies the pain according to each of the five scales.

The IASP classification has been successfully applied to the study of vulvodynia (Pukall et al., 2003). Hypothetically, in the case of vulvodynia, the region of the body is clearly identified, affecting the vulvar region, or more specifically the vestibule, perineum or clitoris (Bergeron et al., 2001; Jantos, 2008). The physiological systems involved in vulvodynia can include the neuromuscular (White & Jantos, 1997; Glazer et al., 1995) and the autonomic nervous system (Payne et al., 2007; Granot et al., 2002). In relation to temporal patterns of occurrence, pain can be spontaneous or provoked by activities such as penetration and last for several hours post-intercourse (Meana et al., 1997; Reed et al., 2000); the quality of the pain is reported as burning, rawness, itching, or stabbing, and rated by most patients as severe (Bergeron et al., 2001; Jantos, 2008). The presumed etiology may be secondary to recurrent infections or inflammation (Friedrich, 1987; Saunders et al., 2008).

The classification of vulvodynia as a pain syndrome is becoming more widely accepted, with more reports referring to vulvodynia as a chronic pain syndrome, rather than a sexual dysfunction or somatoform disorder (Reed et al., 2000; Pukall et al., 2003; Binik, 2005). The IASP pain classification, with its primary focus on pain, enables clinical and research work to focus on the more objectively measurable symptom, than on interference with sexual activity and sexual response. This is a significant shift in focus as pain measures have been shown to be the most important predictors of patient-rated improvement (Bergeron et al., 1999), providing support for a pain-centered conceptualization of vulvodynia (Binik et al., 1999).

The ISSVD definition and classification of vulvodynia

With the prevalence of idiopathic vulvar pain increasing, the International Society for the Study of Vulvovaginal Disease (ISSVD) established a special task force in 1983 to review the terminology and classification of vulvar pain. The first report proposed two terms to designate chronic vulvar pain: *vulvodynia* and *burning vulvar syndrome* (Young et al., 1984)*.* Following consultation with relevant specialist associations, including the IASP, the ISSVD adopted the term vulvodynia as a descriptive term for chronic unexplained vulvar pain (Moyal-Barracco & Lynch, 2004). The anatomical area referred to as the vulva includes the external portion of the female genital tract, consisting of the vestibule, hymen, urethral opening and Skene's ducts, the greater vestibular ducts (Bartholin's ducts), labia minora and majora, the clitoris, mons pubis and the perineum (McLean, 1988). The ISSVD defined vulvodynia as "vulvar discomfort, most often described as burning pain, occurring in the absence of relevant visible findings or specific, clinically identifiable, neurologic disorder" (Moyal-Barracco & Lynch 2004, p. 775). The ISSVD definition of vulvodynia meets the essential criteria for the definition of a chronic pain condition (Williams et al., 2004). It identifies the location of the pain, its sensory qualities, and its unknown but potentially multi-factorial nature. In relation to the location of the pain, the term vulvodynia is descriptive in that it identifies the anatomical area affected – namely the vulva, and the Greek term *odyno* points to pain as the primary symptom. Other terms such as dysesthesia and dysesthetic vulvodynia were not supported. The earlier term *vulvar vestibulitis syndrome* was also discontinued as it wrongly implied that vulvar pain was an inflammatory disorder.

The current classification system for vulvar pain was put forward and accepted at the 17th Congress of the ISSVD in 2003 (Moyal-Barracco & Lynch, 2004). The term vulvodynia was accepted as the most appropriate descriptor for chronic unexplained pain of the vulva. For purposes of consistency, the current ISSVD terminology will be used throughout this discussion, but earlier nomenclature used by authors will be acknowledged in parentheses.

To clearly differentiate between medical causes of vulvar pain and vulvodynia, the ISSVD proposed the following classification;

A) Vulvar pain related to specific disorder

1) Infectious (e.g., candidiasis, herpes, etc.)

2) Inflammatory (e.g., lichen planus, immunobullous disorders,etc.)

3) Neoplastic (e.g., Paget's disease, squamous cell carcinoma, etc.)

4) Neurologic (e.g., herpetic neuralgia, spinal nerve compression, etc.)

B) Vulvodynia

1) Generalized

a) Provoked (sexual, nonsexual or both)

b) Unprovoked

c) Mixed (provoked and unprovoked)

2) Localized (vestibulodynia, clitorodynia, hemivulvodynia,etc.)

a) Provoked (sexual, non sexual or both)

b) Unprovoked

c) Mixed (provoked and unprovoked)

As is evident from the classification of vulvar pain, vulvodynia is a diagnosis of exclusion. Medical causes of vulvar discomfort are classified under four separate categories in Section A, covering infectious, inflammatory, malignant and neurological causes. Unexplained discomfort not accounted for by any of the medical diagnoses is classified in Section B under one of two categories, generalized or localized vulvodynia. Generalized vulvodynia accounts for the involvement of the whole vulva, and localized refers to the involvement of a specific portion of the vulva, such as the vestibule (vestibulodynia), clitoris (clitorodynia), or hemivulva (hemivulvodynia). Under each of the two categories (generalised and localised), provision is made to further identify if the discomfort occurs spontaneously or is provoked by physical triggers such as sexual intercourse, tampons, speculum, or tight clothing. There is very little research and empirical evidence supporting the sub-typing of vulvodynia (Masheb & Richman, 2005) and further work is required to clearly differentiate between different localizations, and to establish the significance of the generalised and localised sub-categories. Some reports have used further sub-classification to differentiate between primary (early onset) and secondary (later onset) vulvodynia, which may need to be considered in future reviews of

terminology (Goetsch, 1991; Jantos & Burns, 2007; Sutton et al., 2009). The early onset of symptoms in primary vulvodynia, often predates, or is associated with, first attempts at intercourse, whereas, later onset in secondary vulvodynia, occurs after a period of pain free intercourse. The significance of a sub-classification based on time of onset needs to be further explored.

In terms of a medical assessment, the diagnosis of vulvodynia is established on the basis of reported discomfort and pain associated with vestibular touch and occurring in the absence of any other diagnosis. Cotton swab testing is used to localise painful areas and the degree of discomfort and pain can be classified as painless, mild, moderate or severe (Haefner et al., 2005). Concerning the common sensory descriptors used by patients, 88.1% chose adjectives that described a thermal quality and 86.6% chose adjectives that described an incisive pressure sensation, highlighting the involvement of peripheral sensory mechanisms (Bergeron et al., 2001; Jantos, 2008).

Finally, the ISSVD definition emphasizes the absence of any specific physical or neurological findings to account for the chronic pain. As a chronic pain condition, vulvodynia typifies the chronic pain syndrome anomaly where pain is not proportional to, or explained by, visible pathology (Steege, 1998). However, some have used the exclusion criteria to argue that if medical causes are excluded, the only alternative is to view psychosexual distress and psychopathology as the most likely cause of vulvodynia (Lynch, 2008; Schultz et al., 2005). The ISSVD classification is being accepted and, as a new classification system, needs to be reviewed and given time to evolve.

Reliability and validity of current classification systems

The DSM classification system in labelling pain in the urogenital area as a sexual dysfunction or a "sexual pain," has inadvertently redefined this regional pain disorder in a manner which lacks validity and reliability (Moser, 2005). It has embedded the problem of pain in psychological concepts and discouraged the study of its major symptom, which is pain (Binik et al., 1999). Even though it has sought to remove the psychoanalytic language of the past versions, it has arbitrarily regrouped many of the disorders in a manner that still reflects its past history (Moser, 2005). As a result a growing number of researchers have been critical of the DSM classification (Kaler, 2005; Kleinplatz, 2005).

The IASP classification is a well validated system, providing the best model for the classification of chronic vulvar pain. It lists pelvic and genitourinary pain syndromes (including vaginismus and dyspareunia) in its classification, and can be adapted for the classification of chronic vulvar pain (Pukall et al., 2003). Many clinicians and researchers have called for the reclassification of vulvodynia as a chronic pain syndrome (Kaler, 2005; Binik, 2005) and have called specifically for the adaptation of the IASP classification in relation to vulvodynia. In recommending the IASP system, they are of the opinion that vulvodynia "fits nicely into this framework" and that a "de-sexualized" approach to pain "will lead to improved understanding and treatment of important and currently neglected women's health problems" (Binik et al., 1999, pp. 230, 231).

For the diagnosis of vulvodynia two physical criteria show good reliability and validity: presence of pain on vaginal penetration and tenderness on pressure application to the vulvar vestibule (Bergeron et al., 2001; Masheb et al., 2004; Reed et al., 2006; Reed et al., 2008). Both of these criteria resulted in over 90% of cases being correctly classified (Bergeron et al., 2001). The IASP classification of chronic pain is focussed on the specific region affected by pain and on the physiological system producing symptoms of pain with an emphasis on objective diagnostic criteria such as reproducible tenderness on pressure application of the affected area. It appears to be the only system which can provide reliability and validity for the classification of chronic vulvar pain.

The ISSVD classification draws a clear distinction between medical and non-medical causes. In doing so it typifies a traditional dualism where visible pathology is equated with medical causes and lack of pathology with non-medical causes, which are often assumed to be psychological (Lynch, 2008). The ISSVD classification fails to make any mention of the possible physiological systems mediating pain, even though considerable evidence exists highlighting the involvement of the neuromuscular and neuropathic mechanisms (Bachmann et al., 2006). The ISSVD classification and definition of vulvodynia fails to address the emotional and subjective components of pain, as reflected in the definition of pain given by the IASP (Merskey & Bogduk, 1994, Jantos, 2008). These omissions contribute to confusion in the context of the current controversy on whether vulvodynia

constitutes a sexual dysfunction, a somatoform disorder, or a regional pain syndrome. The ISSVD classification needs to be further developed and its sub-classifications updated.

Prevalence of vulvodynia

Prevalence studies provide information on current or lifetime baselines whereas incidence studies are more concerned with the rates of increase of a given problem (O'Donohue, 1993). The aim of prevalence studies is twofold: to establish how many women may be experiencing the problem at any one time (point prevalence) or to ascertain how many individuals may have had the problem at some time in their lives (lifetime prevalence). General prevalence studies on vulvodynia have varied significantly in their findings.

T. Galliard Thomas was among the first to comment on the prevalence of chronic unexplained vulvar pain. In his *Practical Treatise on the Diseases of Women* (Thomas, 1874), Thomas stated that: "this disorder, although fortunately not very frequent, is by no means very rare…that it becomes a matter of surprise that it has not been more generally and fully described" (Thomas, 1874, p. 115). Very little interest was shown in the study of chronic vulvar pain for the next century. This trend only changed in the last two decades with a significant growth in the number of clinical, epidemiological and experimental studies into the prevalence and nature of the disorder.

Early prevalence estimates for female dyspareunia have ranged from a low of 3% to over 50% (Meana & Binik, 1994). In European studies, five percent of sexually active women indicated they had experienced pain during intercourse, while an additional 19% reported pain some of the time (Spira et al., 1993, in Meana & Binik, 1994). In an equivalant study in the U.S. 15% of women reported experiencing pain during intercourse for a part of the preceding 12 months (Laumann et al., 1994).

In 1991, one of the first clinical studies on the occurrence of vestibulodynia (in the study referred to as vulvar vestibulitis syndrome– hereafter abbreviated to vvs), reported a 15% prevalence among a clinical sample of gynecology patients (Goetsch, 1991). The study examined a sample of 210 patients and found that 37% met the diagnostic criteria for vestibulodynia. Of the

total group, 50% reported an early onset of symptoms, experiencing discomfort and pain during

their teen years, while 21% reported onset post partum. An unexpected finding revealed that as

many as 32% of the sufferers reported other female relatives experiencing introital dyspareunia,

thereby raising the possibility of a genetic predisposition. However, it is difficult to make any

inference about the prevalence of vulvodynia in the general population on the basis of a single

cohort of patients attending a gynaecology clinic (Green et al., 2001).

A more recent epidemiological study based on a sample of women living in the Boston area

of the United States provided a more reliable prevalence estimate (Harlow & Stewart, 2003). In the

Boston sample of approximately 5000 women, 16% of the women reported a lifetime prevalence of

chronic vulvar pain akin to vulvodynia. When the study group was limited to women who reported

no history of other pelvic disorders (such as endometriosis and polycystic ovaries), the chronic vulvar

pain cumulative incidence was 14%. When the population was further restricted to those who

reported limitations in normal sexual relations, the cumulative incidence was 10%. In relation to the

age of onset, the cumulative incidence was highest before the age of 25 and progressively decreased

to age 44, remaining constant through age 64. In relation to age related risk, the authors reported

that "we can further conservatively estimate that approximately 5% of women will experience this

condition before age 25" (Harlow & Stewart, 2003, p. 87). Furthermore, where vulvodynia was

previously thought to be a disorder primarily affecting Caucasian women, a different ethnic trend

became apparent, with Hispanic women shown to be at greatest risk (22.7%); white and African

American women had the same level of risk (16.2%), while Asian women had the lowest risk (11.1%).

The study also found that only 54% of the women reporting a history of vulvar pain had ever sought

treatment. However, when symptoms became severe enough to limit intercourse, some 64% of

women had sought medical assistance. In summary, the authors suggest that the prevalence of

unexplained vulvar pain, meeting the diagnostic criteria of vulvodynia, may be significantly

underestimated. The authors ended their report by saying that their findings "bring to the forefront

a highly prevalent condition that is associated with substantial disability" (Harlow & Stewart, 2003,

p. 87).

In a quasi-experimental study, 105 women with dyspareunia were compared with 105 non-pain controls (Meana et al., 1997). In terms of medical findings, the dyspareunia group were found to present with more physical pathology at the time of the medical examination and reported more psychological symptomatology of distress, depression, phobic anxiety, and negative attitudes towards sexuality and higher interpersonal sensitivity. The dyspareunia group as a whole did not report a higher than average incidence of physical and sexual abuse, past or present. However, among the dyspareunia group, 54% of the women met the diagnostic criteria for vestibulodynia (in the study referred to as vvs). If vestibulodynia, as a single entity, accounts for more than half of the dyspareunia cases, the finding provides further evidence that vulvodynia is the most prevalent form of dyspareunia among women.

Several other studies reported differing prevalence rates. A clinical study on the prevalence of vulvar pain among women attending family planning services and gynaecologic care in an urban minority population found prevalence to be 11% (Levy et al. 2007). The majority of patients (77%) in this study were under 31 years of age. Another study, based on mailed questionnaires to a sample of almost 5000 women, with a 36.8% return rate, found a much lower prevalence of only 4% (Bachmann et al., 2006). Clinical studies from Africa and Europe showed much higher prevalence rates. A general medical clinic in Ghana found the prevalence to be 22.8% (Adanu et al., 2005), and a study of Swedish adolescents attending an adolescent health centre found it to be 34% (Berglund et al., 2002). Thus, the prevalence reports range from 4% to 34%. It should be noted that different sampling techniques were used, with some studies based on clinical samples, others on general population studies, and others on experimental samples; the ages of respondents also varied considerably. As a result the literature on the prevalence of vulvodynia continues to be equivocal.

One consistent trend in these reports is the high prevalence rate of vulvodynia among the younger population of women (Harlow & Stewart, 2003; Levy, 2007, Jantos & Burns, 2007). Until recently the age-specific incidence of these disorders was largely unknown, and there was very little information available regarding any predisposing factors and the age of onset (Harlow & Stewart,

2003). The age related prevalence and risk of onset were deemed as important issues: "it is crucial to investigate how early this pain really begins and how it develops" (Binik, 2000, p. 66).

A recent clinical study of a cohort of 744 Australian women, diagnosed with vulvodynia, examined the age distribution of patients and studied the age of symptom onset (Jantos & Burns, 2007). The study found that 75% of the vulvodynia patients were under the age of 34 years. Prevalence peaked at 24 years of age. The average age of symptom onset was 22.8 years, but for primary vulvodynia cases the average age of onset was 19 years of age. The age of onset ranged from 5.5 years to 45.2 years. The onset of symptoms occurred before the age of 24 years in approximately 50% of the case. The age related risk of symptom onset coincided with a time when many young women were entering into closer relationships.

An unexpected finding in the Australian study was that a significant number of the women reported the onset of symptoms in their early childhood or shortly after puberty, with commencement of tampon use, first medical exam, or with first attempts at sexual activity. These findings, for the first time, highlight the very early onset of symptoms for at least half of the study population. Given that the onset of vulvodynia appears to be unrelated to sexual activity this finding has important implications for the classification of the disorder. Although most evident with commencement of sexual intercourse, it cannot be attributed to solely psychosexual conflict and it is unlikely to meet the criteria of a somatoform disorder (Jantos & White, 1997). A recent retrospective study of vulvodynia in preadolescent girls confirmed the occurrence of vulvodynia in children between ages four to eleven, with duration of pain varying from several months to seven years (Reed & Cantor, 2008). In an early landmark paper, Friedrich noted the disproportionate prevalence of chronic vulvar pain among patients in the 20 -30 age group, with 65% of his study cohort being between the ages of 20 and 40 (Friedrich, 1987). Two other studies suggested that vulvodynia may predominantly affect women under the age of 35 (Rogstat, 2000; Baggish, 1995).

Other studies also noted that women who reported difficulty and pain with first use of tampons were seven times more likely to report chronic vulvar pain than women who experienced little or no difficulty with pain (Harlow & Stewart, 2003). The triggers and risk factors for such an

early onset are not clear, but most likely are multi-factorial. Further research is required to identify the specific predisposing risk factors. A range of causes of vulvodynia has been proposed, including embryological, immunological, genetic, hormonal, inflammatory, infectious and neuropathic (Haefner et al., 2005). It is unlikely that there is one single cause.

There is general agreement that the incidence of vulvodynia has increased, not only due to a growing awareness of the problem but also to a real increase in the number of women affected (Moyal-Barracco & Lynch, 2003; Damsted Peterson et al., 2008). The cause of the increase is unknown. Some have suggested that because women of reproductive age are more likely to seek gynaecological care this may result in a higher proportion of younger women being diagnosed (Harlow & Stewart, 2003). However, social change and the redefining of gender roles may also be inextricably linked through growing stresses and pressures which in themselves may contribute to increased disorders of chronic pain. Changes in the social milieu have also helped to overcome past prejudices about women's sexuality and have encouraged more research. Clinically, a greater move toward cooperation between health disciplines has facilitated earlier recognition of sexual problems, and new technologies, such as the internet, have enabled women to source more information relating to their health and well-being (Leiblum & Rosen, 2000). Patient centered organizations such as the National Vulvodynia Association (NVA) have become an important source of information.

Clinical management of vulvodynia

In an era of pharmacology where the focus on resolving sexually related difficulties has become "pill-focussed" (Lieblum & Rosen, 2000), chronic urogenital pain has not received the same attention. Current literature on female dyspareunia, and in particular vulvodynia, highlights the paucity of effective clinical protocols for the management of the disorder (Sandownik, 2000). There are signs of greater consensus and consistency in diagnosis and treatment emerging, especially among professionals who are directly involved with research and clinical management of vulvodynia (Reed et al., 2008), but this may not reflect the trend among the wider community of clinicians.

A survey of clinicians seeking to determine practice patterns in relation to management of vulvar pain syndromes found considerable uncertainty in relation to treatment (Updike & Wiesenfeld, 2005). Of 327 providers contacted, 167 responded by completing an extended survey outlining their treatment practices. The majority of responders were tertiary specialists who worked in the following settings: in a private group practice setting (33%); solo practice (26%); university practice (31%); a multidisciplinary group practice (8%). Approximately 20% of the practitioners indicated they saw more than 20 vulvar pain patients per month, while 40% saw between one to five patients per month. Most of the respondents (85%) felt that the treatment of vulvodynia was not addressed adequately in their training programs and that the terminology in relation to the vulvar pain syndrome was confusing. In relation to treatment, therapeutic drugs were the frontline modality. The most common drugs used were tricyclic antidepressants (89%) and the anticonvulsant, gabapentin (68%). Both of these non-specific pharmaceutical agents were used on the assumption that vulvodynia was caused by a form of neuropathy. Only 21% of the respondents felt that vulvar pain had a psychologic cause, with psychiatric care being recommended more often for generalized than for localized vulvodynia. However, most practitioners acknowledged that due to the negative impact of vulvodynia on quality of life and relationships, some form of counselling was warranted. In general the study concluded that "It is impossible to practice evidence-based medicine in the care of women with vulvodynia with so few clinical trials" (Updike & Wiesenfeld, 2005 p. 1406). Until there is prospective comparative data available, clinicians must rely on personal experience and expert consensus.

A large proportion of vulvodynia patients report attending multiple medical consultations before receiving a diagnosis (Jantos & Burns, 2007; Harlow & Stewart, 2003). Patients repeatedly expressed concern that their complaints were being overlooked and not addressed effectively (Green et al., 2001; Sandownik, 2000). Lack of diagnosis, confusion in classification, inappropriate referrals and poor therapeutic outcomes also contributed to a high level of disappointment and frustration among referring doctors,

"Numerous physicians have seen these patients, and numerous interventions were tried. On

average, >64% of the time these interventions made the patients' symptoms no better or

worse. No single treatment or combination of treatments were perceived as consistently

improving symptoms" (Sandownik, 2000).

This has caused some to appraise the general quality of treatment for women with dyspareunia as

"shockingly poor" (Kaler, 2005). Others attribute the lack of progress in diagnosis and management

of vulvodynia to the inappropriate classification of the condition as a psychiatric disorder or a sexual

dysfunction (Binik et al., 1999; Binik, 2005; Pukall at al., 2003). Classifying vulvodynia as a psychiatric

disorder or a sexual dysfunction has resulted in therapy being misdirected:

"...sufferers are referred to mental health care professionals by gynaecologists and other

medical specialists who can find no organic basis for the pain and imply to the women that it

is all in their heads. Mental health care professionals may make a variety of diagnoses and

assumptions about the nature of the pain, ranging from poor sexual technique to

somatisation and then treat the women accordingly. These diagnoses rarely help, and

women find themselves increasingly frustrated by an endless series of referrals and little

pain relief" (Binik, 2003, p. 69).

Patients who persist in searching for a physical cause are likely to meet with spoken or implied

suggestions that the "pain is in their head" (Binik, 2000).

Classifying the problem as psychological has been found to have a detrimental impact on the

patients themselves (Kaler, 2005). How the pain is defined and categorized not only has bearing on

the therapeutic outcomes but on their own self-perception and self-esteem. In a sociological study

examining the effects of vulvar pain on women's self concepts and social relations, the views held by

the women themselves, by their partners, or by the medical professionals were found to have

significant impact on the women (Kaler, 2005). When vulvodynia patients were asked if they had

received unhelpful medical advice or treatment for their pain, 34% mentioned being told that their

problems were "psychological" or "all in the head" as the most unhelpful thing that they had been

told (Kaler, 2005).

Two recent consensus statements specifically addressed questions of terminology, diagnosis and management (Haefner et al., 2005; Bachmann et al., 2006). *The vulvodynia guideline* (Haefner et al., 2005), based on a comprehensive review of literature and cumulative experience of a panel of practitioners, sought to offer clinicians guidance on management of cases from the time of diagnosis through to the selection of treatments. The guideline was set out in the form of an algorithm designed to assist with decision making at various levels of intervention. Some of the optional therapies listed ranged from oral medications to surface electromyography (SEMG) biofeedback, physical therapy, and surgical excisions. However, due to the complexities of chronic vulvar pain, the guidelines acknowledged the reality that no single treatment is successful for all women and no rapid resolution of symptoms is likely.

The most recent survey, approximately three years following the first survey, looked at the diagnostic and treatment practices among vulvodynia researchers and members of the ISSVD (Reed et al., 2008). The survey results reflect an emerging consensus on diagnostic and treatment preferences. For diagnosis, the majority of practitioners find that a history of pain, together with tenderness on examination, described as producing a burning sensation, rawness or stinging, constitute a reliable basis for the diagnosis of vulvodynia. The findings also showed that Friedrich's criteria, published in 1987, are still most widely used and cited. In particular, the first two criteria; reported pain on touch or attempted vaginal entry, and tenderness to pressure localized within the vulvar vestibule, are most commonly used. Little additional value was conferred from the use of the third criterion, vestibular erythema (Bergeron et al., 2001; Masheb et al., 2004; Reed et al., 2008). In relation to treatment, the most commonly used therapies included: continued use of tricyclics antidepressants, pelvic floor therapies, psychological counselling and sex therapy. Non-medical therapies, such as pelvic floor related therapies, were rated as most effective. The majority of survey participants (87.5%) were aware of cases where full remission of symptoms occurred (Reed et al., 2008), signifying a more effective approach to management.

The management of vulvodynia, and the improved outcomes reported, reflect a growing perception that vulvodynia has a physiological and possibly neuropathic basis (Reed et al., 2008).

Further research on pain mechanisms mediating symptoms of chronic vulvar pain are needed (Devoe, 2007). Information regarding the diagnosis and management of vulvodynia needs to be widely disseminated and education and training opportunities provided for primary and tertiary care providers.

A controversial question: Is the pain sexual or is the sex painful?

Characteristically, clinicians and medical specialists are guided in their approach to chronic pain disorders by certain assumptions about the nature of chronic pain. This is well illustrated in the case of gynecology and psychiatry when it comes to lower tract urogenital pain. Traditionally, the strategy in gynaecology was to look for organic causes of pain and in their absence to assume psychogenic etiology (Binik, 2003). In psychiatry, pain was seen as psychogenic unless there was evidence of medical causes. But the gynaecological and psychiatric views of sexual pain differ in one other important way. In gynaecology, the focus is on the anatomical structures affected, whereas in psychiatry it shifts away from the location of the pain and focuses on the activity with which the pain is associated, thus labelling urogenital pain conditions such as vulvodynia as "sexual pain" (Binik, 2003).

The listing of so called "sexual pain" disorders in the DSM classification system, raises the important question of whether a special type of pain exists, that is sexual in nature and that warrants inclusion in psychiatric nosology. The psychiatric classification assumes that such a unique form of pain exists. In questioning this assumption, some have argued that if there is a pain that is indeed sexual in nature, it should be possible to induce the pain not only by sexual activity but also by sexual thoughts and sexually related feelings (Binik et al., 1999). Furthermore, if sexual pain exists, then by implication, other categories of pain may also exist that can be defined by the activity that triggers the pain, including, eating pain, work pain, or sports pain. Yet, sexual pain disorders are the only forms of chronic pain noted in the psychiatric classification system (Binik, 2003). What determines the sexual nature of this pain condition or why it should be considered a sexual

dysfunction is not clear, but its inclusion in the DSM classification system creates much ambiguity and confusion (Moser, 2005).

In relation to dyspareunia, the DSM lists pain as the primary diagnostic feature of the disorder but provides no suggestion as to its causes, or underlying mechanisms. The psychiatric classification system makes no reference to the fact that genital pain can exist in the absence of sexual activity, and can be triggered by other activities that are non sexual, such as the use of tampons, wearing of tight clothing, sitting for prolonged periods, undergoing medical examinations, and other general day-to-day activities (Sandownik, 2000). All of these non-sexual activities are known triggers of vulvar pain and are known to exacerbate the severity of symptoms.

From an historical perspective, it is interesting to note that while medical accounts of chronic vulvar pain from the 18th to early 19th century attributed the pain to such physiological causes as "hyperesthesia" and "abnormal sensitiveness," none of the reports attributed its etiology to psychological factors (Thomas, 1874; Skene, 1898). This is well illustrated in a case study presented by Sims in 1861 and cited in a recent discussion paper (Binik et al., 1999). The case is that of a patient who though married for a quarter of a century, remained a virgin because of her vulvar pain symptoms. In his account, Sims states;

> "Amongst other investigations of her, I attempted to make a vaginal investigation but failed completely. The slightest touch at the mouth of the vagina producing most intense suffering. Her nervous system was thrown into great commotion: there was a general muscular agitation; her whole frame was shivering...She shrieked aloud, her eyes glaring wildly, while tears rolled down her cheeks and she presented the most pitiable appearance of terror and agony. Notwithstanding all these outward involuntary evidences of physical suffering, she had the moral fortitude to hold herself on the couch, and implored me not to desist from any efforts if there was the least hope of finding out anything about her inexplicable condition. After pressing with all my strength for some minutes, I succeeded in introducing the index finger into the vagina up to the second point, but no further. The resistance to its passage was great, and the vaginal contraction so firm, as to deaden the sensation of the

finger, and thus the examination revealed only an insuperable spasm of the sphincter

vaginae." (In Binik, 1999, p. 212).

In this account, the pain was seen as mediated by physiological mechanisms, namely muscular

reactivity of unknown etiology. There is no insinuation that the pain had a psychological origin, nor is

there any allusion to it being a "sexual pain" even though the pain was regional and affected the

vaginal introitus.

The "sexualizing" of urogenital pain appears to be a post Freudian phenomenon (Binik et al.,

1999: Binik, 2005). It reflects a past theoretical perspective which dates back to:

"a less enlightened era of medicine in which women's reports of pain were much more likely

than men's to be met with some form of the 'it must be in your head' explanation from their

doctors, including gynaecologists...Genital pain was thus vulnerable to being read as a

manifestation of disturbances of women's minds and social relationships, rather than an

organic ailment" (Kaler, 2005, p. 35).

The literature from the mid 1900's manifests a trend by which dyspareunia is included under the

general rubric of frigidity, hysteria and manipulative sexual avoidance behaviour (Fenichel, 1945). On

the basis of this perspective it was suggested that: "The dyspareunic patient must be helped to see

for herself that the hyperesthesia is a fiction and that the pain is of her own making" (Malleson,

1954, p. 390). The statement fully attributes the cause of vulvar pain to psychological factors.

In more recent publications, Dodson and Friedrich, in a 1978 paper entitled *Psychosomatic*

Vulvovaginitis argued that vulvar pain is a psychosomatic disorder (Dodson & Friedrich, 1978). They

stated that "Psychosomatic vulvovaginatis is a real clinical entity that should be suspected in any

patient whose vaginal complaints do not correlate with the physical findings." The authors were of

the view that chronic pain must be accompanied by visible pathology; otherwise, its absence is

evidence of psychogenic aetiology. The evidence cited to support their view is as follows:

- the pain was characterized by persistent symptoms of longstanding duration,

- it lacked any demonstrable pathology,

- was typified by sexual inactivity arising from symptoms,

- resulted in unsuccessful consultation with multiple physicians,

- showed reluctance to accept a psychophysiological explanation of its cause,

- showed allergy to many common vaginal preparations and,

- many of the patients exhibited psychological difficulties including emotional lability and dependence.

The authors went on to comment that "The patient often pleads for help but is absolutely resistant to any suggestion that her symptoms might be psychologic in origin" (Dodson & Friedrich, 1978, p. 23s). Furthermore, claims that patients would enjoy sex and would resume normal relations if they were cured of their disease were seen as inconsistent.

Published accounts consistently document a refusal by women to accept the psychogenic origin of their pain and evidence shows that the primary motivation for seeking a diagnosis and treatment is a desire to resume and increase the level of sexual activity (Jantos & Burns, 2007). However, Dodson and Friedrich concluded that patients:

"...manifested signs of neurosis, dependant personality, guilt feelings, emotional lability, while denying psychologic difficulties...these patients receive secondary gain from their symptom complex, i.e., a reason not to engage in sexual activity. As a consequence, they are understandably reluctant to accept any treatment that might destroy the defence mechanism that they have unconsciously constructed...Patients with persistent or incapacitating symptoms however, should be promptly referred to psychiatric care" (Dodson & Friedrich, 1978, pp. 24s-25s).

The statement implies that vulvodynia patients engage in "pain games" and "psychosomatization for secondary gain."

The specific presentation features of vulvar pain reported by Dodson and Friedrick (1978) are consistent with current accounts of vulvodynia, but their conclusions about the psychological origins of the disorder continue to be unsupported.

In less than ten years, Friedrich's perspective on vulvar pain changed significantly, as was evident from his landmark paper *Vulvar vestibulitis syndrome* and his subsequent publications (Friedrich, 1987, 1988). Friedrich's published criteria for the diagnosis of vulvar vestibulitis syndrome focused solely on physiological changes in the vulva and made no allusions to psychological factors in its etiology. His published criteria have aided the development of a more systematic study of vulvodynia.

However, patients still continue to be told that they are "frigid, sexually dysfunctional, repressed, or otherwise sexually abnormal because they experienced pain," and based on their encounters with medical practitioners, the pain appears to be generalized into diagnoses that imply that their entire sexual being is somehow sick (Kaler, 2005, p. 35). Recent publications continue to argue that a lack of demonstrable pathology is the basis for assuming a psychogenic etiology (Schrover et al., 1992; Mascherpa et al., 2007; Lynch 2008). A dualistic perspective of chronic pain appears to lead to the common sexualizing and psychologizing of lower urogenital tract pain.

Furthermore, it is not uncommon for views on female dyspareunia to frequently reflect a gender bias. Even though dyspareunia is a disorder that affects both men and women, there appears to be an implicit assumption that it does not exist in men, and if men do experience pain, it is "real" and therefore should not to be diagnosed as dyspareunia (Townsend, 2005). It has been claimed that in women, dyspareunia is more likely to involve psychological factors than in male dyspareunia (Abarbanel, 1978). Why the same chronic pain condition should have a greater psychological content in its etiology for women than for men, is not clear. Such gender bias has not helped advance the understanding of vulvodynia.

Literature reveals a continued polarization of views on the potential links between "organic" and "psychogenic" contributors to the etiology of vulvodynia symptoms. There is an ongoing failure to see the experience of pain as consisting of both somatic and psychological aspects and to treat it accordingly (Merksley & Bogduk, 1994). Instead, proponents of physiological causes of vulvar pain argue that the prevalence of psychological variables in the etiology of pain is low or negligible (Bohm-Starke et al., 1998; Lowenstein et al., 2004), whereas proponents for psychological causes

minimize the physiological links, asserting a prevalence of psychological variables (Schrover et al., 1992; Lynch, 2008). It appears that the attribution of pain to psychological causes, when pathology is not evident, may arise, in part, on account of a poor understanding of the nature of chronic pain (Steege, 1998; Jantos, 2007); or the physician's inability to make a specific diagnosis, inability to relieve the pain and unwillingness to listen (Lynch, 1986); or the patients visibly depressed and tearful state (Edwards, 1997). However, none of these reasons justify classifying pain as a psychiatric disorder or sexualizing its etiology (Binik, 2005). Pain needs to be classified according to the guidelines applicable to chronic pain nosology and managed according to the primary presenting symptom – which is pain.

The answer to the question of whether the pain is sexual or the sex is painful, appears to depend on the theoretical presuppositions about the nature of pain. In relation to chronic vulvar pain, assumptions about the nature of pain can lead to incorrect classification and ultimately inappropriate treatments. The discussion in the section that follows will review literature which identifies potential organic and psychogenic factors related to the etiology of vulvodynia and will seek to identify any emerging trends in the understanding of the condition.

Research linking organic and physiological factors with vulvodynia

Various potential pathophysiologies have been researched in an attempt to identify possible dermatological, infectious, neurological, or muscular causes. The physiological-organic findings reported include:

- low grade chronic inflammation (due to possible up-regulation of local mast cells),

- viral, bacterial and fungal infections,

- proliferation of local pain fibres (indicating upregulation of the local pain system),

- dysfunctional pelvic muscle states (mostly up-regulation of pelvic muscle tone).

These pathologic findings are acknowledged in the recent state-of-the-art consensus statement on vulvodynia (Bachmann et al., 2006) and will form the basis of the discussion that follows.

The search for inflammatory and skin related causes of vulvodynia produced inconsistent findings. This ultimately led to the conclusion that, by the very definition of vulvodynia, the condition and any positive dermatological findings would automatically result in a medical diagnosis, and exclude the diagnosis of vulvodynia (McKay, 1985; McKay et al., 1991; Byth, 1998; Willsteed, 1995).

Vestibular inflammation, even though reported by patients and noted in some studies (Pyka et al., 1998; Peckham et al., 1986), was not a consistent feature and failed to differentiate between vulvodynia patients and controls (Landquist et al., 1997). Because inflammation was not found to be a reliable factor in the differential diagnosis of vulvodynia, the term "-itis", as in vestibulitis was excluded from ISSVD terminology in relation to vulvodynia (Moyal-Barracco & Lynch, 2003). The presence of erythema, when present, was predominantly confined to the posterior fourchette (Lynch, 1986; McKay, 1988).

Several studies have looked at the potential role of genetic factors in the etiology or susceptibility to vulvodynia symptoms. Some patients were found to possess an uncommon genotype (IL-1RA) that is associated with chronic inflammation (Witkin et al., 2002) and prolonged and intensified inflammation was seen as a potential trigger of other events, including the autonomic nervous system alterations, resulting in increased sensitivity to pain and chronic localized inflammation (Gerber et al., 2003). Women who carry the proinflammatory genetic variants were found to be 4-8.5 times more likely to develop vestibulodynia (Foster et al., 2004). Further studies are required to improve the current understanding of the relevance of genotypes and the pro-inflammatory response at the tissue and cellular level.

Infections

Research also focussed on potential links between sexually transmitted diseases and vulvodynia but no evidence was found to support the link (Marinoff & Turner, 1986). The role of candida infections had been suspected and studied as a possible trigger in the development of vulvodynia (Friedrick, 1988; Peckham et al., 1986, Mann et al., 1992). Although a history of candida

is reported by many women with vulvodynia (Witkin et al., 2002; Jantos & White, 1997) no causal relationship or hypersensitivity to candida has been demonstrated (Marinoff & Turner, 1986). A link between the human papillomavirus (HPV) and vulvodynia was proposed (Marinoff & Turner, 1991) but subsequent research found no evidence linking HPV in the etiology of the disorder (Bergeron et al., 1994).

Neuropathic causes

A suggestion was made that vulvodynia may be a form of neuropathic pain in the form of pudendal neuralgia (pain along the distribution of the pudendal nerve), caused by compression or entrapment of the nerve by the sacrospinous and sacrotuberous ligaments (Turner & Marinoff, 1991). Even though such entrapment can occur, there is no evidence of pudendal neuralgia being a primary cause of vulvodynia.

Further studies focussed on the role of superficial nerves in the vulva. Since the primary symptom of vulvodynia is evoked by touch and pressure application to the vaginal introitus, a number of studies explored the role of superficial nerves in vulvar pain. Morphological studies examining the neural content of biopsied vulvar tissue found evidence of increased density of superficial intraepithelial free nerve endings in vulvodynia (Bohm-Starke et al., 1998). To assess the impact of increased density of nerve endings on pain thresholds, a study of 19 patients diagnosed with vestibulodynia (in the study referred to as vvs), examined the reactivity of patients to heat, cold, mechanical stimulation, vibration and distension of the mucosa (Bohm-Starke et al., 2001). All of the patients demonstrated significantly greater hypersensitivity to mechanical stimulation, temperature, and distension but not to vibration. The study provided some evidence supporting the view that sensory abnormalities in these patients were best explained by peripheral noxious mechanisms involving sensitization and/or proliferation of various types of C-nociceptors (pain fibres know to cause a burning sensation). The authors concluded that, from a clinical perspective, treatment should focus on destroying superficial nociceptive nerve endings, or reducing their hyper-excitability by suitable ion channel blockers. In the opinion of the authors, the sensory abnormalities

associated with vulvar pain had a clear organic basis and could not be explained on the basis of psychological factors.

A further study examining a neuropathic link with vulvodynia used quantitative sensory testing (QST) with vestibulodynia patients (in the study referred to as vvs), to demonstrate that symptom severity was directly related to mechanical and thermal pain thresholds (Lowestein et al., 2004). In this study, sensory tests effectively discriminated between patients and controls, and between levels of pain severity. The evidence was seen as favouring a neuropathic explanation of chronic vulvar pain. However, the significance of this finding was partially called into question by a subsequent study of pain thresholds, in which tests were carried out applying noxious stimuli, not to the vulvar region, but the forearm (Granot et al., 2002). It was noted that enhanced pain perception in patients was not just localized to the genital site but also evident in other regions of the body. The severity of pain was found to be related to patient's anxiety and provided evidence of a generalised pain disorder. Patients demonstrated higher trait and state anxiety, lower pain and unpleasantness thresholds, an increased autonomic reactivity. This study provided evidence that secondary hyperesthesia may be modulated by psychological attribute such as anxiety.

In a follow up study the same research team showed that systemic pain perception was closely related to the choice of treatment and the rated success of treatment, irrespective of treatment modality used (Granot et al., 2004). Women who showed increased pain perception were more likely to choose conservative treatment options, but were less likely to report improvement. Those with a lower pain perception were more likely to choose surgery (vestibulectomy) and report more improvement. Pain perception was found to be related to autonomic dysregulation, with systolic blood pressure positively correlated with level of vulvar pain. Women with lower systolic blood pressure showed increased pain perception and reported less reduction in pain. Those with higher blood pressure showed lower reception of pain and reported more improvement with treatment.

In a study using functional magnetic resonance imaging, the heightened processing of tactile pain sensation was studied (Pukall et al., 2005). The goal of the study was to compare the neural

response to mild and moderate pressure to the posterior portion of the vulva. Participants consisted of 14 women diagnosed with vestibulodynia (referred to in the study as vvs) and were matched with 14 age and contraceptive matched controls. All of the women with vestibulodynia described moderate pressure as painful and unpleasant, whereas only one of the control women found it to be painful and unpleasant. During pressure application described as painful by women with vestibulodynia, significantly higher cortical activation levels were noted in the insular and frontal cortical regions than that noted in controls. The results showed that women diagnosed with vestibulodynia exhibit an augmentation of genital sensory processing, which is similar to that observed in other syndromes characterised by hypersensitivity, including fibromyalgia, idiopathic back pain, irritable bowel syndrome and neuropathic pain.

A recent study on the effects of sexual arousal on genital and non-genital sensitivity in women with vestibulodynia (in the study referred to as vvs) offered further insight into the potential role of vulvar sensitivity in vulvodynia (Payne et al., 2007). Upon establishing baseline touch and pain thresholds, healthy controls and vestibulodynia women viewed erotic film segments while undergoing temperature sensory testing and monitoring their level of subjective sexual arousal. The findings showed that there was no difference between healthy and vestibulodynia women in terms of the level of physiological arousal, and increased sensitivity of the vestibule during the viewing of sexual content. However, in the case of the vestibulodynia group, arousal was associated with increased sensitivity to touch and pain. The results also showed a difference in sensitivity to touch as opposed to pain. The forearm, though more sensitive to touch, was less sensitive to pain than the labia and the labia were also less sensitive to touch then the vulvar vestibule. The results suggest increased sensitivity within the vulvar vestibule that may be the result of vasocongestion, increased muscle tension and heightened sensation associated with the orgasmic platform. From these findings it appears that "The vulvar vestibule may be a particularly unique genital area with sensory properties as different from other genital locations as other more distant peripheral locations, such as the forearm" (Payne et al., p. 298). However, at all locations tested, women with vestibulodynia experienced more sensitivity and pain than healthy participants at all locations tested. These data

confirmed the existence of generalized sensory abnormality, as well as increased catastrophizing, hypervigilance, and fear of both intercourse and non-intercourse pain, as previously reported (Giesecke et al., 2004; Granot et al., 2002; Pukall et al., 2002). These findings imply the need to address issues related to fear, catastrophizing, and pain related hypervigilence as part of any pain management strategy.

Neuromuscular variables

Another area of research where a high degree of consensus has emerged relates to over-activation of the levator ani muscle, the primary muscle within the pelvic diaphragm. Through the use of SEMG, several studies have sought to evaluate the functional status of pelvic muscle in vulvodynia patients. Early studies found consistent SEMG characteristics that could differentiate between patients and controls (White et al., 1997). The SEMG characteristics included:

- Elevated resting baselines in 71% of patients, with readings over 2.0 μV,

- Poor contractile potential in 63% of patients with readings under 17 μV,

- Elevated resting standard deviation greater than 2 μV in 93% of patients,

- Poor recruitment and recovery times of over 0.2 sec in 86% of patients,

- Spectral frequency of less than 115 Hz in 69% of patients.

Among vulvodynia patients, 88% showed at least three of the above criteria, thus providing objective confirmation for the diagnosis of vulvodynia. Subsequent studies confirmed that SEMG can differentiate symptomatic patients from asymptomatic controls (Glazer et al., 1998). Vulvodynia patients showed:

- 46% less amplitude during 3 sec phasic contractions,

- 49% less amplitude during 12 sec tonic contractions,

- 32% more amplitude during pre-test rest,

- 49% more muscle instability during pre-test rest.

Based on these findings, therapy focused on normalization of levator ani muscle function. SEMG assisted normalisation of pelvic muscle function resulted in an 83% reduction in symptoms (Glazer et

al., 1995). It has been proposed that increased muscle resting tone in the pelvic floor can be a response to physical or chemical irritants, inflammation, trauma, or emotional tension (Hong et al., 2008; Hawthorn & Redmond, 1998; Flor et al., 1992; Gevirtz et al., 1996; McNaulty et al., 1994). Chronic over-activation of these muscles can progressively lead to painful decompensation of muscle tissue and peripheral sensitization associated with muscle ischemia (Burton, 1996).

Evidence linking psychological factors with vulvodynia

Literature reports on the role of psychological variables in vulvodynia are diverse in their views. Most of the early etiological conceptualizations of dyspareunia were guided by psychoanalytic constructs, focussing on phobic reactions, anxiety conflicts, hostility, aversion to sexuality, abuse and trauma in childhood (Binik et al 1999). Other reports generalizing from findings on chronic pelvic pain to vulvar pain, reported increased hypochondriasis, hysteria and decrease in ego strength (Ranaer et al 1979). A number of them reported unsatisfactory interpersonal relationships with parents, higher incidence of marital conflict, psychosexual problems, and avoidance of close physical contact with their partners (Beard et al., 1977). Typically, many of the studies placed disproportionate emphasis on somatisation issues. More recent studies have sought to assess the psychological profile of vulvodynia patients and focused on emotional states, in particular level of anxiety, depression, illness behaviour, quality of relationships and history of abuse. These studies will be reviewed next.

<u>Negative emotions and vulvodynia</u>

One of the earliest studies, based on non-structured interviews, found that all older patients, and 80% of the younger ones, showed signs of depression (Lynch, 1986). The study found evidence of obsessive compulsive behaviour, as reflected by the degree of personal neatness, a history of fastidious housecleaning, concern about productivity and other perfectionistic tendencies. It was also noted that while patients expressed anger about their medical care, very rarely did they

express anger or hostility towards their spouses. In general, relationships appeared to be strong and supportive.

Depression amongst various chronic pain patient groups is estimated to range from 30-54% (Sullivan et al 1992). In vulvodynia, current and lifetime prevalence of major depressive disorder was shown to be 17% and 45% respectively (Masheb et al 2005). These reported prevalence rates are lower than rates in other chronic pain patients and confirm earlier findings that showed vulvodynia women not be different to controls in relation to the level of reported depression (Reed et al., 2000).

A study focussed on the psychosexual aspects of vulvar pain examined the role of stress in vulvodynia (Schrover et al., 1992). The study concluded that psychological stress was the primary trigger of chronic vulvar pain. In spite of the fact that patients disagreed with such a link, the authors attributed the cause of pain onset to stressors that included: severe marital conflict, new relationships, break up of an existing relationship, a patient's or their partner's affair, job stress, fear of pregnancy, fear of sexually transmitted diseases, depressive symptoms, or somatisation disorder. However, the findings in this study are problematic in that the results of the psychological tests contradict the conclusions of the authors, and the treatment provided appears to be inconsistent with the assessments made. The study reported that the standardised tests used showed the scores for depression, somatisation and psychological distress to be well within the normal range. The authors acknowledged these discrepancies and sought to clarify this apparent contradiction by stating that "The discrepancy between psychologic testing and the results of interviewer observation is striking," but "this group of patients is very reluctant to label vulvar pain as related to psychologic stress. These women minimize conflict on questionnaires but cannot conceal problems as easily when interviewed in person by an experienced clinician" (Schrover et al., 1992, p. 632).

It is difficult to draw a clear conclusion from these findings because of the degree of internal inconsistency between standardized objective tests and opinions of the authors. Furthermore, the findings are at variance with earlier studies showing no marital conflict, but strong and supportive relationships (Lynch, 1986). The results are also inconsistent with more recent studies highlighting the supportive nature of the majority of relationships (Reed et al., 2000; Jantos & Burns, 2007). The

study is also open to further criticism due to the inconsistency between the suggested psychogenic etiology of vulvar pain and the surgical treatment utilized. The treatment consisted of the surgical excision of purportedly psychologically triggered vulvar lesions. It is widely acknowledged that stress can exacerbate vulvar discomfort in that "...tension or stress can magnify symptoms, particularly when a disorder [vulvodynia] is as mysterious and/or frightening" (McKay, 1985), but a causal relationship between stress and lesions has not been shown.

Other studies also examined the level of psychological distress among patients attending a vulvar health clinic (Stewart & Reicher, 1994). The study found that vulvodynia symptoms interfered more seriously with sexual function than any other forms of pathology seen in non-vulvodynia patients. This conclusion was supported by similar findings in a more recent study (Ponte et al., 2009). Furthermore, the level of disability was found to be related to increased levels of anxiety with patients displaying a heightened awareness of various sensations throughout their bodies, were more likely to attribute serious disease etiologies and consulted more physicians in relation to their pain. The role of anxiety and somatisation was seen as a central issue requiring stress-reduction techniques such as lifestyle changes, exercise, relaxation and psychotherapy. Where anxiety and somatisation were considered to be a manifestation of an underlying depression, antidepressant therapy was recommended (Stewart & Reicher, 1994).

Personality traits, illness behaviour and psychopathology

A study investigating the relationship between medical symptoms, personality traits, illness behaviour, and psychopathology in vestibulodynia patients (in the study referred to as vvs), noted a high prevalence of depression, anxiety and perfectionistic traits (Jantos and White, 1997). The study found insufficient evidence for the diagnosis of somatisation disorder, but patients often reported a history of unconfirmed candidiasis, multiple premenstrual syndrome symptoms, headaches and migraines, urologic and gastrointestinal symptoms and skin allergies. The Illness Behaviour Questionnaire (Pilowsky et al., 1984) showed that, although patients were preoccupied with symptoms, there was no evidence of phobic or hypochondriacal behaviour. Patients did not believe

that their pain was of psychogenic origin, but manifested disappointment and anger with a lack of diagnosis. The elevated levels of anxiety and depression were related to chronicity of symptoms and concern about the unknown cause of the pain.

The quasi-experimental study of 105 women with dyspareunia and 105 non-pain controls mentioned in earlier discussion (Meana et al., 1997) highlighted several findings relevant to this discussion. The dyspareunia patients, as a group, presented with more physical pathology and greater psychological distress than the control group. However, when the vestibulodynia subgroup, making up 54% of the dyspareunia group, was compared with control subjects, the vestibulodynia group showed the highest level of sexual impairment, lower frequencies of intercourse, lower sexual desire, lower arousal, and reduced ability to achieve orgasm. Yet, analysis of the psychological data did not show a higher level of psychological impairment. The study found pain related impairment, but the vestibulodynia group were found to be more resilient with no evidence of a link between psychosexual conflict and etiology of vulvar pain.

Psychosexual profiling

Due to the centrality of psychosexual issues in the discussions on vulvodynia, the relationship between pain symptoms and sexual function was studied in more detail (White & Jantos., 1998). A total of 40 vestibulodynia patients (in the study referred to as vvs) sought treatment for painful intercourse. The average duration of pain was 33 months and the average intercourse pain was rated as 7.5 (on a scale of 0-10, where 0 is no pain and 10 is the most severe pain experienced). A large percentage of the patients (83%) also reported experiencing pain with non-intercourse orgasm, and rated non-penetrative pain as 7 on the same scale. When compared with controls, patients experienced the same levels of sexual desire, but reported a higher rate of refusal to sexual advances and sexual intercourse. They also experienced significantly less physiological arousal and reported a more negative attitude towards sexual activity.

The sexual behaviour in patients was most influenced by the experience of pain. The authors commented that "the pain stimuli compete with sexual stimuli, resulting in decreased sexual desire"

(p. 784) and the "psychological morbidity noted in the vestibulitis cohort" appeared to be "...most influenced by the actual experience of chronic pain..." and "the psychological morbidity noted in the vestibulitis cohort appears to be secondary and a consequence of the chronic sexual pain" (p. 787). Furthermore, they suggested that "sexually related pain, as reported by vestibulitis patients, can be responsible for recurrent disappointment, anxiety, frustration, depression and loss of self-esteem" (p.785). The authors caution that:

> "It may be tempting for the physician to view sexual behavioral changes associated with vulvar vestibulitis syndrome as indicative of sexual dysfunction of psychogenic etiology...It is most important to ensure that the pain is not categorized prematurely as of psychological origin or to fall into the dichotomy of seeing sexual pain as either a solely physical or solely psychological problem" (p. 787).

From this study and others, it is evident that the pain ratings for sexual activity fall into the severe range (Jantos, 2008; Bergeron et al., 2001) and the severity of pain is often emotionally and physically disabling, preventing intercourse from occurring and contributing to emotional turmoil and disappointment.

Most studies examining the potential relationship between psychosexual disturbances and vulvodynia omitted to control for the effect of chronic pain itself. A study using a cross-sectional design evaluated psychological traits, marital satisfaction, sexual history and behaviour and somatic characteristics comparing vulvodynia patients with a chronic pelvic pain group without vulvodynia symptoms, and a control group (Reed et al., 2000). The data showed that in terms of the quality of marital relationships, interest in sex, importance of vaginal sex and frequency of sexual activities, women with vulvodynia were not dissimilar to the other two groups. Upon onset of pain, women with vulvodynia and those with pelvic pain rated the quality and quantity of current sexual activities as significantly more impaired when compared to control subjects and to their own premorbid state. The history and incidence of depression was similar for all three groups; however, the severity of depression and of global affective distress, as measured by the Beck Depression Inventory and the Brief Symptom Inventory and Global Severity Index, was significantly higher for the pelvic pain

group. There were no significant differences between the vulvodynia and control groups. The pelvic pain group had a significantly higher rate of sexual abuse.

The study highlighted that psychological dysfunction was not a prerequisite of chronic vulvar pain. Likewise, there were no indicators to suggest that vulvodynia patients were sexually averse, rather, the symptoms of vulvodynia significantly interfered with sexual functioning which resulted in a decline in frequency and quality of sexual activity. The study also found that the occurance of somatic complaints among the vulvodynia group did not differ from controls, it notes that the failure to understand the multidimensionality of the symptom of pain and the lack of awareness of how pain impacts on women's sexuality needed to be addressed. The findings provided no support for a link between vulvodynia and psychological disorders or lack of marital adjustment, sexual or physical abuse, depression or somatic sensitivity. The authors concluded that a primary psychological cause of vulvodynia is not supported and "suggestions that personal choice or psychological weakness of women have caused vulvodynia are unsubstantiated and may add to the patient's distress" (p. 631). The study emphasizes that "attention needs to continue to be given to the possibility of increased psychological distress in women who are dealing with chronic pain," especially unpredictable genital pain (Reed et al., 2000, p. 631).

In a recent assessment of a psychosexual profile of vulvodynia patients, the connection between vulvodynia symptoms, relationships, and sexual activity was studied (Jantos & Burns, 2007). Of the 516 patients who were sexually active, 80.3% reported a decrease in sexual desire and a marked reduction in frequency of sexual intercourse. The average weekly frequency of sexual intercourse changed from 3.5 times per week prior to onset of symptoms, to 0.6 times per week post symptom onset. Over 50% of patients experienced some difficulties with physiological arousal, and sexual intercourse was prevented by a dry and tight vagina in 41.2% of the cases. Negative emotional reactions to sexual intercourse were experienced by 39.4%, with only 18.3% of the patients indicating that they still found sexual activity enjoyable. Patients also noted a reduction in the frequency with which their partners initiated sexual activity, due to concern that intercourse would cause pain. Indeed, 74.9% of patients indicated that their partners were supportive, 25.1%

indicating that their partners were frustrated, but only 3.5% reported having partners who were angry or did not care. When asked which activities patients would like to increase if pain symptoms abated, the majority expressed a desire to increase sexual activity (87.5%).

The findings confirm that women with vulvodynia experience some negative emotions towards sexual activity because of pain, and concerns about pain lead to lower levels of sexual activity, but generally they enjoy supportive relationships. For those in a marriage relationship, marriage acted as a buffer against emotional distress. The study found no evidence linking vulvodynia with relationship stress or psychosexual conflict.

Incidence of reported physical and sexual abuse

The issue of physical and sexual abuse has often been raised in relation to the etiology of vulvodynia and is pertinent to this discussion. Unlike the earlier findings relating to chronic pelvic pain (Steege, 1998; Reed & Haefner, 2000), research has consistently failed to find any relationship between physical or sexual abuse and vulvodynia. The incidence of sexual abuse in the vulvodynia cohorts was found to be no higher than in the general population (Jantos, 1997; Schrover et al., 1992; Friedrich, 1987). Variations in findings appear to arise on account of different definitions of abuse. In a Canadian study of 300 vulvodynia patients, 9% of women reported non-consensual sexual intercourse (Sandownik, 2000). An Italian study reported non-penetrative sexual abuse in 29% and penetrative abuse in 6.5% of its vulvodynia patients (Graziottin & Brotto, 2004). A US study reported an 8% incidence of reported sexual abuse among its patients (Brotto et al., 2003). In another US study on victimization, 242 patients were compared with 113 controls and the prevalence of victimization was found not to be higher in the patient group when compared with the control group. The authors of the study concluded that "...if anything, there was a trend toward a negative association between vulvar pain and sexual victimization" (Dalton et al., 2002). In a study specifically designed to compare the incidence of sexual and/or physical abuse in women with vulvodynia, questionnaire responses from 89 vulvodynia patients were compared with those of 65

patients with chronic vulvar pain of medical origin and 166 general dermatology patients (Edwards et al., 1997). The analysis showed that there were no differences between the three groups.

In a US study on adult-onset vulvodynia and childhood violence and victimization, a sample of 12,000 women in the Boston area was contacted, and 125 women were identified, who, on the basis of a phone interview, met the ISSVD diagnostic criteria for vulvodynia (Harlow & Stewart, 2005). Through telephone interviews an assessment was made of the medical, psychiatric and sexual histories and of childhood victimization. Results from this non-clinical sample showed that women who never or rarely received family support as children were two to three times more likely to report vulvodynia symptoms. Women who reported severe physical abuse in childhood, and those who experienced severe sexual abuse, were four and six times more likely, respectively, to experience vulvodynia symptoms. The joint effect of severe abuse, childhood endangerment and lack of family support was associated with a 14-fold increased risk of vulvodynia. It is unclear to what extent the strong associations reported in this study may be due to a broader definition of victimization or due to recall bias. However, increased stress during childhood and adolescence and other early life experiences may constitute a risk factor for chronic vulvar pain.

Finally an Australian study assessing the psychosexual profile of 744 vulvodynia patients found that the self-reported incidence of unwanted sexual activity during childhood and adulthood were 11.5% and 12.5%, respectively (Jantos & Burns, 2007). The findings of this study confirm that the incidence of unwanted sexual activity among vulvodynia patients is no higher than among the control groups and the general population.

Somatoform hypothesis

Two recent opinion papers have reactivated discussion of the somatoform hypothesis in relation to the etiology of vulvdynia (Mascherpa et al., 2007; Lynch, 2008). The hypothesis as outlined in the most recent paper (Lynch, 2008), rests in part on a semantic argument stemming from the current wording of the ISSVD terminology and classification of vulvodynia. The author of the opinion paper acknowledges that his is a minority view, but the therapeutic implications of the

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somatoform hypothesis he sees as "immensely important" (p.395). According to the hypothesis, "medical" (neurological, inflammatory and infectious) causes of vulvar pain preclude the diagnosis of vulvodynia and only "idiopathic" or unexplained non-medical causes can form a plausible basis of diagnosis. It is then assumed that the idiopathic causes are primarily psychogenic in nature. The evidence cited in support of the hypothesis is based on findings of predisposing psychological, sexual and social disturbances among patients, and on the co-existence of other chronic pain syndromes (fibromyalgia, irritable bowel syndrome, interstitial cystitis and other idiopathic pain problems such as headache, lower back pain and pelvic pain). Symptoms of vulvar pain are seen as an expression of personal and/or social distress, not on account of inflammatory and neuropathic processes. As a result patients are non-responsive to commonly used analgesics and anti-inflammatory medications, but responsive to non-medical conservative therapies. According to the hypothesis, the psychosexual and social factors, previously seen as predisposing individuals to the development of vulvodynia, are actual precipitating factors. The pervasiveness of psychosexual dysfunction is seen as far exceeding what might be expected in response to organic pathology. The high levels of depression and anxiety, and lower levels of sexual desire, arousal and frequency, together with high levels of marital conflict provide evidence for the pre-existence of psychosexual and social issues which ultimately find expression in the form of pain symptoms. Furthermore, the argument is put forward that the higher than average incidence of abuse among vulvodynia sufferers is also a possible precipitating factor. However, research evidence supporting such a hypothesis is lacking.

In summary, current studies assessing the psychological profile of vulvodynia patients find no evidence for a psychogenic cause of chronic vulvar pain. Negative attitudes toward sexual activity, fear, phobias, depression and generalised distress appear to arise in response to the association of pain with sexual activity (Meana et al., 1997; White & Jantos, 1997; Reed et al., 2000, Jantos & Burns 2007). It is also important to note that patients consistently continue to reject any suggestions that vulvodynia is symptomatic of personal psychological factors (Schover et al., 1992, Jantos & White, 1997; McKay, 1989).

It would appear that much of the confusion surrounding the classification and management of vulvodynia arises from a lack of understanding of the nature of chronic pain. The next section seeks to briefly review the difference between acute and chronic pain, to assist in recognising vulvodynia as a chronic pain syndrome.

Understanding the nature of chronic pain

Pain is a distressing experience for sufferers, but serves the important function of attracting the patient's and physician's attention to an underlying problem (Markos, 2005). Pain is also a complex phenomenon that defies precise definition. Unlike other physiological functions in the body there is no localised centre for the regulation of pain and no simple intervention for its effective management. The experience of pain cannot be measured in any absolute units in an objective manner. Its severity can only be inferred on the basis of the sufferer's communication (Jantos 2007).

Little is known about the causes of chronic pain or its mediating mechanisms. Attributing proportionality, or labelling the physical components of pain as "real," and the emotional as "unreal," is of no clinical value and may compromise the quality of treatment of patients (Steege 1998). Much of the debate regarding vulvodynia stems from a failure to differentiate between the nature of chronic pain and that of acute pain. Chronic pain has gradually emerged as "a distinct phenomenon in comparison with acute pain" (Merskey & Bogduk, 1994, p. ix) and requires greater insight.

Acute Pain versus Chronic Pain

Acute pain is the most common form of discomfort and is often experienced by patients after surgery or soft tissue trauma. It tends to be immediate, severe and short lived. Pain that extends beyond a normal recovery period and lasts longer than 3 -6 months constitutes chronic pain (Merskey & Bogduck, 1994). As inferred in earlier discussion, chronic pain is more difficult to understand because it exists where there is no visible pathology and continues long after soft tissue damage has occurred, persisting well beyond the expected time of healing. In simple terms, chronic

pain occurs when there is little if any reason for it to exist. Yet the pain is real and can significantly affect the patient's quality of life, limiting their daily physical activities and disrupting their ability to rest and sleep (Gordon et al 2003; Sandownik 2000).

When acute pain enters the chronic phase, normal sensory processes are affected by progressive sensitization of the peripheral and central nervous system (Hawthorn and Redmont 1998). Sensitization is an important property of nociceptors and manifests itself in:

- decreased thresholds to nociceptor stimulation,

- increased field of nociceptor reception (progressing from localised to generalised),

- increased nociceptor responsiveness to normally non-noxious stimuli (allodynia),

- increased intensity of response (hyperalgesia),

- prolonged post-stimulus sensations (hyperpathia),

- the occurrence of unexplained spontaneous pain

Such sensory changes are the defining characteristics of chronic pain syndromes. What is important to note is that sensitization can occur at the peripheral or central level (Hawthorn and Redmont 1998). There is no evidence supporting the view that chronic vulvar pain is a form of central sensitization akin to that of fibromyalgia or chronic fatigue syndrome (Mees & Nijs, 2007), but it appears to be mediated by peripheral sensitization and is responsive to conservative therapy (Jantos 2008; Reed et al., 2008).

Sensitization in chronic pain conditions require management strategies that are different to those used in the management of acute pain. While patients often hope that tests will uncover some form of pathology to explain their pain, in chronic pain syndromes, discomfort and hypersensitivity is never proportional to pathology findings. The anomaly is acknowledged in the following statement:

"I'm not aware of a single chronic clinical problem associated with pain in which pain is seen as proportional to tissue damage...most clinicians intuitively or by training look for enough pathology to explain the pain. With pain this proportionality simply does not exist...the intensity of pain is not consistently related (either directly or inversely) to the apparent degree of tissue damage" (Steege 1998, p. 6).

A lack of proportionality between pathology and severity of pain is what distinguishes chronic pain from acute pain. Recognising this fundamental distinction excludes the need to apportion pain to organic or psychogenic causes.

<u>Potential sources of pain</u>

In most cases of chronic pain the prevailing question in the mind of the clinician and patient relates to the source of pain. Generally there are three common origins of pain:

- Somatic origin - arising from skin, muscles and bone tissue. Patients describe this type of pain as a throbbing, stabbing or burning.

- Visceral origin - coming from internal organs. This type of pain tends to be diffuse and more generalised, with patients frequently describing it in more emotive terms.

- Neuropathic origin - arising from damaged nerve fibres. The pain is described as numbness, pins and needles and produces electric current like sensations (Mense & Simons 2001).

Of the three sources of pain, the most common is somatic pain. Somatic pain which arises predominantly from muscle tissue and is sympathetically maintained pain (Schattschneider et al., 2006). Pelvic muscle overactivation (hypertonus muscles) and spasm can lead to chronic pelvic and perineal pain (Travel & Simmons, 1992). Pelvic muscle overactivation has been shown to be characteristic of vulvodynia (Pukall et al., 2006; Glazer et al., 1995; White et al., 1997). Muscle overactivation can arise in response to a range of triggers, including irritants, deep somatic or visceral disease, distressed viscera or iatrogenic causes (Graven-Nielsen et al., 2003; Schattschneider et al., 2006). Triggers of chronic pain are initially acute in nature (e.g. infection or inflammation), but result in chronic muscle overactivation via spinally mediated reflexes (Menses & Simons, 2001; Simons & Travell, 1999; Schattschneider et al., 2006). Such overactivation gives rise to progressive neuromuscular tension by which muscle tissue not only responds to nociceptive triggers, but progressively becomes the primary "initiator of nociception" and the site of chronic pain (Burton, 1996; Cailliet, 1996).

<u>Mechanisms of pain</u>

Several pain mechanisms arise in association with muscular over-activation (often referred to as "wind-up"). The first of these is ischemia. Ischemia (reduced blood flow) and hypoxia (reduced oxygen supply) during increased demand (muscle contraction or over-activation) leads to deep tissue pain of moderate to high intensity (Mense & Simons, 2001; Graven-Nielsen et al., 2002; Coderre et al., 2004; Cram et al., 1998). Ischemic pain is most often described as "stabbing", "burning", "heavy" and "exhausting" pain and leads to lower pressure related pain thresholds whereby patients experience a most pronounced hyperalgesia to touch, consistent with peripheral sensitization (Seo et al., 2008; Coderre et al., 2004). If a muscle is contracted under ischemic conditions, severe pain can develop within a minute (Menses & Simons, 2001)

Hyperalgesia arising from ischemic pain can be reversed through conservative therapy, indicating that peripheral sensitization due to ischemia may be different to sensitization and hyperalgesia mediated through central mechanisms. Peripheral sensitization and central nervous system sensitization require different management strategies. Peripheral sensitization arising from muscle ischemia requires normalization of muscle function, while centralized sensitization may be more complex and dependant on pharmacological therapy. The characteristics of ischemic pain appear to be totally consistent with symptoms of vulvodynia (Bergeron et al., 2001; Jantos, 2008).

A second mechanism of pain that arises from muscle over-activation is mediated by myofascial trigger points (TrPs) which can give rise to myofascial pain syndrome (Simons et al., 1999; Ge et al., 2008). Some estimate that 85% of chronic pain conditions are muscle mediated giving rise to regional pain (Slomski, 1996). TrPs are focal points of tenderness, a few millimetres in diameter, found at multiple sites in muscles and fascia of muscle tissue. TrPs are defined by several primary characteristics (Simons et al., 1999; Menses & Simons, 2001):

- A TrP has a clear and consistent referred pain pattern. Pain from TrPs can be felt not only at the site of its origin but also in areas remote from it. Since the pain originating from a given muscle tends to exhibit a relatively consistent pattern of pain referral, it is often possible to

identify the muscle from which the pain originates if the pattern of pain is clearly delineated,

- TrPs can arise in response to acute and chronic overload, or overuse of the muscle in which it occurs. Such muscle overuse can arise from muscle wind-up following physical trauma or as result of sympathetically mediated tension (anxiety related bracing and guarding/splinting),

- TrPs contribute to motor dysfunction by causing increased muscle tension (the primary function of the muscle spindles is the regulation of tension in muscle tissues), spasm of neighbouring muscles, loss of coordination in affected muscles, substitution patterns in recruitment of muscles and a weakening of affected muscles,

- TrPs cause weakness and limited range of motion. In most cases the patient is only aware of the pain but not of the other dysfunctional aspects of muscles,

- The intensity and extent of the pain depends on the degree of irritability of the TrPs and not on the size or location of the muscle,

- TrPs disturb the proprioceptive, nociceptive and autonomic functions of the affected anatomical region.

Pain from TrPs can go unrecognized unless the clinician is prepared to actively look for and identify the source by palpating muscles that may harbour these points of tenderness. Palpation of the tender spot always evokes discomfort and assists the patient to immediately recognize and identify "their" pain. This simple and reliable means of identifying the pain confirms in the patient's mind that the pain is of muscular origin and not due to other causes. Pelvic musculature is structurally and functionally predisposed to developing myofascial TrPs, due to its work load supporting abdominal and pelvic viscera and maintaining posture and facilitating movement.

The presence of TrPs in pelvic muscles has been well documented (Travell & Simmons, 1992; Weiss, 2001). TrPs in muscles of the posterior half of the pelvic floor are a common source of poorly defined pain in the perineal region and discomfort in the anus, rectum, coccyx and sacrum. TrPs in

muscles in the anterior half of the pelvic floor refer pain to genital structures like the vagina, bladder and clitoris (Wise, 2001). Active TrPs in these muscles can interfere with intercourse by contributing to introital pain and TrPs in the deeper pelvic muscles are associated with deep penetration pain (Travell & Simons, 2001).

Myofascial TrPs and dysfunctional pelvic muscles have frequently been linked to symptoms of interstitial cystitis, urgency and frequency, and pain in the suprapubic, perineal, rectal and vulvar areas (Wise, 2001; Fitzgerald & Kotarinos, 2003a, 2003b). These conditions are the most commonly reported comorbidities in vulvodynia (Peters et al., 2008; Peters et al., 2007). Normalizing pelvic muscle function by reducing pelvic floor hypertonicity and releasing myofascial trigger points has been reported to result in an 83 per cent reduction in IC symptoms (Weiss, 2001). In the case of vulvodynia, where dysfunctional pelvic muscles (high muscle tone, instability, poor contraction and recovery) and tenderness to pressure were noted, soft tissue mobilization and myofascial release produced complete resolution of symptoms or significant improvement (Hartmann & Nelson, 2001; Bergeron et al., 2002).

Psychophysiological perspective on chronic pain

Several psychological factors have been identified as strong modulators of pain perception (Keefe et al., 2004). These factors can be divided into two broad categories, those associated with increased pain, psychological distress and physical disability, and those associated with decreased pain, distress and disability:

- **Factors associated with increased pain, psychological distress, and physical disability**. These include: pain catastrophizing, pain related anxiety and fear, and helplessness. *Pain catastrophizing*; a tendency to negatively evaluate one's ability to cope with pain, accounts for 7% to 31% of the variance in pain ratings, and is associated with higher levels of depression, psychological distress, pain-related disability, lower energy levels, and more negative general health status.

    Pain-related anxiety and *fear of pain*; these patients have a tendency to be anxious

about their pain and engage in fear avoidance behaviours. They report high levels of attention to sensations of pain and over-predict the amount of pain they will experience during physical examination. They score higher on self-report measures of disability, depression, pain behaviour and help-seeking, and lower on measures of pain coping.

Helplessness; refers to a tendency to view negative outcomes as inevitable, unpredictable and uncontrollable, and is associated with higher levels of pain, depression and disability, and much poorer medical therapy outcomes.

- **Factors associated with decreased pain, psychological distress, and physical disability.** These include: self-efficacy, pain coping strategies, readiness to change, and acceptance.

Self-efficacy; refers to a patient's confidence in their ability to accomplish desired outcomes, such as control of their pain. It is associated with lower levels of psychological distress and negative medical outcomes. Most importantly, patients who rated their self-efficacy for managing their clinical pain as high, exhibited significantly higher pain thresholds and pain tolerance than patients who rated their self-efficacy for controlling pain as being low. Those scoring high on self-efficacy for clinical pain also rated the laboratory thermal pain stimuli as significantly less unpleasant.

Pain coping strategies; refers to patient's ability to utilise a variety of strategies to help them cope with pain. Such pain coping strategies may include pain reduction efforts, relaxation, distraction, redefinition, venting emotions, seeking emotional support, and seeking spiritual comfort. Patients who reported the highest level of coping effort experienced decreasing pain.

Readiness to change; refers to the patient's readiness to engage in self-management efforts. Evidence suggests that patients who become actively engaged in self-management efforts are more likely to show improvement in pain management.

Acceptance; refers to the willingness to engage in meaningful activities in life,

regardless of the experience of pain-related sensations. Acceptance refers to a

balanced approach of pursuing change if it is likely to assist, and acceptance when

change efforts are not likely to succeed. Patients scoring high on acceptance

reported significantly lower levels of pain-related anxiety and avoidance, depression,

physical and psychosocial disability, and better utilization of time. Acceptance

measures were found to explain 24% of variance in measures of adjustment.

It is evident that patient attitudes and beliefs (catastrophizing, passive attitude to treatment),

behaviours (active coping strategies, versus high drug intake), emotions (feelings of helplessness,

uselessness, anxiety and fear of increased pain) and level of social support (lack of support or over-

protectiveness) are all important factors, identified as "yellow flags" in at risk patients (Waddell,

1998).

In examining the role of anxiety and fear in the perception of pain in vestibulodynia patients,

cognitive and affective factors were shown to exert a significant influence (Payne et al., 2005).

Vestibulodynia patients, when compared with controls, demonstrated hypervigilance in the form of

selective attentional bias to pain-relevant stimuli, which in the case of vestibulodynia was coital pain.

On self-report measures, patients reported more hypervigilance to pain during sexual intercourse. A

further comparison between the patients and the control group showed that the groups differed on

measures of hypervigilance, state and trait anxiety, and fear of pain; with anxiety and fear of pain

correlating with hypervigilance. However, when controlling for anxiety and fear of pain, the group

differences on hypervigilance disappeared. Such selective focus on pain stimuli can lead to an

increase in the perceived intensity of pain. In relation to sexual function, the authors suggest that "if

attention is preferentially allocated to pain processing during activities such as sexual intercourse,

then theoretically, fewer attentional resources will be available for the processing of sexually

arousing or pleasurable stimuli"(p 436). This may further exacerbate vulvar pain.

With higher levels of anxiety, fear, depression and dysfunction, few models of chronic pain have sought to theoretically conceptualize links between hypervigilance, catastrophizing, fear of activity, avoidance behaviour, disuse, disability and depression (Lame et al., 2005; Boersma & Linton, 2006). Few models have sought to identify the physiological mechanisms that link beliefs, emotions and behaviours to the experience of pain. An extensive discussion of such mechanisms is beyond the scope of this chapter, but a brief discussion will be presented on the mechanisms linking anxiety and depression with pain.

Anxiety can be conceptualized as a sympathetically mediated biological response that prepares the body to threatening situations (Hoehn-Saric & McLeod, 2000). The individual feels tense, experiences increased heart rate, faster but shallower breath, increased skin conductance and increased muscle tension. The expectation that patients who manifest heightened anxiety will also exhibit generalised physiological hyper-arousal at rest has not been confirmed. The majority of individuals show no greater autonomic arousal then normal control subjects, except in relation to stimuli that have been associated with a phobic stimulus (Hoehn-Saric et al., 2000). In relation to a phobic stimulus, anxious subjects were found to be selective in their response and "overreact subjectively and physiologically to stimuli that are anxiety-provoking." At rest, anxiety levels appear to be associated with a tonic increase in stimulus related physiological arousal, which is also associated with a diminished range of autonomic responsivity, labeled as "diminished physiological flexibility."

The relationship between anxiety and muscle tension is well documented. Of all the physiological parameters monitored, it is noted that:

"Elevated muscle tension has been the most consistent finding in psychophysiological studies of patients with anxiety disorder. It cannot be attributed to increased restlessness but rather to a tonic increase in tension...It probably represents a peripheral manifestation of central hyperarousal" (Hoehn-Saric et al., 2000, p. 218).

It is further suggested that the anxious patient may be exhibiting increased muscle tension as an expression of hyperalertness but not necessarily generalised autonomic hyperarousal. The authors

suggest that strong emotional experiences which initially trigger autonomic responses may be capable of subsequently eliciting similar somatic sensations, even in the absence of physiological triggers. This form of "physiological rigidity", though not fully understood, may be "a constitutional predisposition, a physiological adaptation to chronic arousal," or a selective pre-occupation with the patient's pathophysiological condition. On the other hand, the non-anxious individuals that had lower muscle tension at rest tended to respond to test tasks with "greater increases in muscle activity than the already tense anxiety patients, indicating a task-oriented rather than generalized arousal response." With elevated anxiety, the subjective perception of bodily states and muscle tension were found not to be congruent with their physical state, with correlations between subjective and physiological measures predictably low. These findings are consistent with the earlier work of Flor and Turk (1989), showing that chronic pain patients show a symptom-specific psychophysiological response and that physiological dis-regulation tends to occur in the affected system.

There are parallels between these findings and the psychophysiological assessments of vulvodynia patients. Increased muscle activity, lower pain thresholds, lower unpleasantness thresholds, significant increases in systolic blood pressure, enhanced perception and increased autonomic reactivity have been documented (Jantos 2008; Granot et al., 2002; Hoehn-Saric et al., 2000).

In the management of vulvodynia the first line of therapy often consists of the use of tricyclics antidepressants, such as imiprimine (Updike & Wisenfeld, 2005). Evidence shows that tricyclic antidepressants result in a decrease in subjective somatic symptoms, while physiological measures such as heart rate, systolic blood pressure and SEMG activity show significant increase (Hoehm-Saric et al., 2000). Patients report varying degrees of benefit from the use of tricyclic antidepressants. The mechanism by which these drugs assist in the management of symptoms remains poorly understood (Updike & Wisenfeld, 2005). Further research needs to examine the benefits of drug therapy as subjective perception of improvement may in fact mask increased autonomic dysregulation and reactivity.

Conclusion

This chapter reviewed some of the current issues and perspectives in vulvodynia. As a chronic pain condition, vulvodynia poses a number of clinical challanges in relation to classification and management. As more research information becomes available in relation to the psychosexual, psychophysiological and pathophysiological issues in vulvodynia, the less psychogenic and more somatic it appears. It is evident that vulvodynia can arise from multiple causes, activating various pathways that require a range of interventions (Haefner et al., 2005). Like other chronic pain conditions, vulvodynia has "evolved from being conceptualized as a psychological problem to now being viewed by many in the medical world as being a physical problem" (Wylie et al., 2004). However, as with all chronic pain conditions, it is essential to recognise the role of the subjective experience on the sufferer. Biomedical factors alone have limited influence on perception of pain and functional disability (Samwel et al., 2006), whereas psychological factors, though not causal in nature, can modulate the experience of pain to a significant degree (Keefe et al., 2004; Lame et al., 2005; Boersma et al., 2006). One of the activities most affected by symptoms of vulvodynia is sexual intercourse. The reported distress and associated reduction in sexual frequency appear to stem directly from the association of pain with an activity otherwise linked to the experience of pleasure and personal bonding. Excessive psychological attribution in the etiology of chronic vulvar pain stigmatizes the problem, increases the emotional buden of the patient, and compromises its management (Kaler, 2005, Reed et al., 2000, Binik, 2003). A new approach to the management of lower tract urogenital pain is required and evidence suggests that this is progressively evolving (Kaler, 2005, Reed et al., 2008) into one which defines pain as the problem (Binik, 2005; Kaler, 2005) and focuses on the underpinning physiological mechanism mediating symptoms. To this end, the research publications and discussion found in the following chapters provide further evidence to enhance understanding and management of vulvodynia.

Rationale, Aims and Structure of Studies Undertaken

Vulvodynia is recognised as the most common cause of dyspareunia in premenopausal women (Harlow & Stewart, 2003). It is considered to be a neglected issue in women's health (Binik, 2003; Reed et al., 2000). The condition affects women of all races, ages, educational and socioeconomic backgrounds, with an estimated lifetime prevalence of 16 percent (Harlow & Stewart, 2003). It undermines quality of life (Binik et al., 2000; Sargeant & O'Callaghan, 2007), causes psychological distress (Jantos, 2008) and contributes to relationship difficulties (Desrosiers et al., 2008). Because the pathophysiology of vulvodynia is largely unknown (Haefner et al., 2005), opinion is divided on whether vulvodynia should be classified as a somatoform disorder (Dobson & Friedrich, 1978; Lynch, 2008), a sexual dysfunction (APA, 2000), or a chronic pain syndrome (Pukall et al., 2003; Binik, 2005).

To improve the current understanding of vulvodynia and the women affected by the disorder, together with the mechanisms mediating symptoms of discomfort and pain, further research is needed. It is important to establish the age related risk of developing the disorder; the age distribution of patients presenting with symptoms; the efficacy of diagnosis; and to assess the impact of the disorder on the general wellbeing of women, in terms of their physical, emotional and sexual health. Furthermore, to develop more effective management strategies, research needs to focus on the possible physiological mechanisms of pain by examining the role of the neuromuscular system in mediating symptoms of vulvar pain and to examine the interaction between somatic and emotional variables in vulvodynia.

To address these issues two research studies and a discussion paper were prepared and submitted for publication. Both research studies focus on women with vulvodynia, the first on an

assessment of the psychosexual profile, and the second on an assessment of the psychophysiological profile. The discussion paper focuses on a range of pelvic disorders and discusses the role of SEMG in the management of conditions linked to pelvic muscle dysfunction.

Research study 1: Vulvodynia: Development of a psychosocial profile

Aims

1. Analyse the age distribution of patients diagnosed with vulvodynia and identify the age-specific incidence of the disorder.

2. Identify the age of symptom onset based on patient self-reports.

3. Evaluate the age related risk of developing chronic vulvar pain.

4. Estimate the proficiency of diagnosis by analysing the time lapse between symptom onset and confirmation of diagnosis.

5. Examine the impact of symptoms on the formation of social relationships.

6. Examine the impact of symptoms on the emotional wellbeing of women.

7. Assess the role of reported unwanted sexual activity in the etiology of vulvodynia.

8. Assess the extent to which vulvodynia symptoms contribute to changes in sexual desire and how they impact on sexual behaviour.

9. Evaluate the impact of the disorder on the reproductive potential of women.

10. Identify implications for the management of vulvodynia based on evidence arising from the psychosexual profile.

Structure

The first research study assessed the psychosexual profile of a cohort of 744 vulvodynia patients. The psychosexual profile was developed via a retrospective review of patient data consisting of information obtained from questionnaires, tests and interviews of patients who were initially examined and diagnosed by independent tertiary specialists (gynaecologists, dermatologists and sexual health physicians) during the years of 2000-2006. Data used for analysis were obtained

only from patients who provided signed consent for their information to be used for research purposes. The study was approved by the University of Adelaide Human Research Ethics Committee.

Research study 2. Vulvodynia: A psychophysiological profile based on electromyographic assessment

<u>Aims</u>

1. Identify the anatomical region affected by discomfort and pain and review the sensory and temporal characteristic of symptoms reported.

2. Analyse severity of pain related to sexual activity.

3. Explore the relationship between severity of pain, depression, anxiety and general distress.

4. Examine the relationship between SEMG readings and severity of pain.

5. Assess the potential role of pelvic muscle over-activation in relation to symptoms of vulvodynia.

6. Identify potential role of peripheral physiological mechanisms mediating vulvar pain.

7. Identify implications for the management of vulvodynia based on evidence arising from the psychophysiological profile.

<u>Structure</u>

The second study assessed the psychophysiological profile of a cohort of 529 vulvodynia patients. The psychophysiological profile was developed via a retrospective review of SEMG data, psychological tests and questionnaires from patients referred for therapy by tertiary specialists (gynaecologists, dermatologists and sexual health physicians) who diagnosed and referred patients during the years of 2000-2006. Data used for analysis were obtained only from patients who provided signed consent for their information to be used for research purposes. The study was approved by the University of Adelaide Human Research Ethics Committee.

Discussion paper: Electromyographic Assessment of Female Pelvic Floor Disorders

Aims

1. Provide an outline of pelvic floor anatomy and physiology, relevant SEMG applications.

2. Discuss assessment and management of urogenital pain disorders associated with hyper-tonus muscle states.

3. Discuss the assessment and management of pelvic floor disorders associated with hypo-tonus muscle states.

4. Identify risk factors to normal pelvic anatomy and function.

5. Develop guidelines for normalization of pelvic muscle function utilising SEMG.

6. Identify ethical issues in relation to training, qualifications and treatment of pelvic floor dysfunction.

Structure

The third paper is a discussion paper consisting of a review of pelvic floor anatomy and physiology relevant to female pelvic floor disorders. It reviews research relating to the use of SEMG in assessment and retraining of pelvic muscles. Vulvodynia is examined in the context of a range of muscle mediated pelvic disorders arising from hypertonic and hypotonic muscle states. The hyper-tonic states are predominantly associated with chronic pain conditions such as vulvodynia, vaginismus and bladder pain syndrome, commonly referred to as interstitial cystitis. The hypotonic states are associated with pelvic weakness and include urinary incontinence and sexual dysfunction. The paper provides guidelines for normalizing pelvic floor function utilizing down-training protocols for conditions stemming from hypertonic muscle states and up-training protocols for conditions arising from hypotonic muscles. This paper seeks to provide clinicians with research based information which can be used for the development of applied protocols in the management of pelvic muscle mediated disorders like vulvodynia and suggests guidelines in terms of training proficiency and qualifications necessary for treatment of pelvic related disorders.

Nicholas R Burns (Co-author)

I was the supervisor (advisor) for Mr Jantos PhD candidature. My role was advisory with respect to planning empirical studies, suggesting statistical treatments and analyses, and dealing with editing of manuscripts and comments of reviewers.

I hearby give my permission for this paper to be incorporated in Mr Jantos' submission for the degree of PhD in the University of Adelaide.

Signed_____   Date_____

Chapter 3

Vulvodynia: Development of a psychosexual profile

Vulvodynia: A psychophysiological profile based on electromyographic assessment

Abstract

The objective of this study was to explore the relationship between psychological and physiological processes and how these interact in the case of vulvodynia. The study design consisted of a retrospective review of predominantly premenopausal women presenting with vulvodynia via analyses of questionnaires, psychometric tests, sexual history, surface electromyographic (sEMG) assessments, and clinical notes. Five hundred and twenty nine patients with vulvodynia (mean age 27.7 years) were studied. The average age of symptom onset was 22.8 years and the average duration of symptoms was 5.0 years. Patients scored higher than the comparison group on global dimensions of the Symptom Checklist - 90 Revised (SCL-90R), with anxiety and depression scores showing a significant but modest correlation with severity of pain. sEMG data confirmed an association with pelvic muscle dysfunction but there was no correlation with severity of vulvar pain. A negative correlation between sEMG readings and duration of pain was noted and may be due to progressive time-related quieting of electrical activity in muscle tissues, which is commonly associated with the development of a functional muscle contracture. In conclusion, it is important to view chronic pain syndromes like vulvodynia from a psychophysiological perspective which recognizes the potential contribution of psychological and physiological variables in the aetiology of chronic vulvar pain.

Keywords: Vulvodynia, Anxiety, Surface electromyography, Functional muscle contracture.

Vulvodynia: A psychophysiological profile based on electromyographic assessment

Introduction

Growing recognition of the prevalence of vulvodynia and of its impact on the quality of life of women has led to the publication of several major reviews and a consensus statement on current terminology, diagnosis, management and future research (Bachmann, Rosen, Pinn, et al., 2006; Haefner, Collins, Davis, et al., 2005). These publications are positive moves in addressing the complexity of this pain syndrome. However, it is imperative that greater emphasis be placed on understanding the underlying mechanisms of pain and identifying the most successful therapeutic interventions. This need has been succinctly highlighted in a recent editorial comment "…it has been both surprising and disappointing that relatively little scientific effort has been applied to understanding the mechanisms of its causation and to developing effective treatment strategies" (Devoe, 2006). This is a challenging task because chronic pain syndromes like vulvodynia are characterised by a complex interaction of psychological, physiological, and behavioural variables (Jantos & Burns, 2007; Pukall, Reissing, Binik, et al., 2000). A better understanding of the interaction of these variables would have theoretical and applied implications. The aim of this study was to examine (1) the relationship between psychological states of depression, anxiety and level of reported pain; and (2) sEMG readings and the severity of pain.

Pain is a sensory and subjective phenomenon, most commonly defined as an "unpleasant sensory and emotional experience" (Merskey & Bogduck, 1994). Accordingly, the experience of pain involves both sensory peripheral mechanisms and higher cortical functions involved in interpreting and modulating the perception of pain (Cailliet, 1996; Turk & Melzack, 1992).

The International Society for the Study of Vulvovaginal Disease (ISSVD) defines vulvodynia as "vulvar discomfort, most often described as burning pain, occurring in the absence of relevant physical findings or a specific, clinically identifiable, neurologic disorder" (Moyal-Barracco & Lynch, 2004). This definition highlights three key points: the location of the pain; its sensory qualities; and its unknown but potentially multi-factorial nature. In relation to the location of pain, the term

vulvodynia is descriptive in that it identifies the anatomical area affected – namely the vulva and the Greek term *odyno* points to pain as the primary symptom. In regards to the common sensory descriptors used by patients these are of a thermal and incisive nature, highlighting the involvement of peripheral sensory mechanisms (Bergeron, Binik, Khalife, et al., 2001). Finally, the definition emphasizes the absence of any specific physical or neurological findings and in this regard vulvodynia typifies the chronic pain syndrome anomaly where pain is not proportional to, or explained by, visible pathology (Moyal-Barracco & Lynch, 2004; Steege, 1998; Turk & Melzack, 1992).

Unfortunately the ISSVD definition of vulvodynia makes one major omission in that it totally fails to acknowledge the role of emotional factors in the development and maintenance of symptoms. Without recognition of the role of sensory and subjective elements in pain, research has tended to focus on one or the other without scrutinizing the interaction between psychological and physiological variables. At times the absence of identifiable pathology has given rise to the view that chronic vulvar pain may be solely due to psychological factors (Malleson, 1954); however, such an extreme position may be as erroneous as those emphasizing only the physiological aspects of this condition. Both extremes are contrary to current opinion on general chronic pain syndromes, chronic pelvic pain, and vulvodynia (Bachmann et al., 2006; Merskey & Bogduck, 1994; Steege, 1998). There is a high prevalence of depression and anxiety among vulvodynia patients (Jantos & White, 1997; Masheb, Wang, Lozano, et al., 2005), likewise physiological research has shown unique characteristics which include, increased density of superficial nerve endings, increased immunoreactivity, and nociceptor sensitivity (Bohm-Stark, Hillinges, Brodda-Jansen, et al., 1998, 2001; Bohm-Stark, Hillinges, Falconer, et al., 1999; Foster, Sazenski, & Stodgell, 2004). Such physiological peculiarities, when considered in conjunction with commonly reported psychological traits of elevated anxiety and depression (Brotto, Basson, & Gehring, 2003; Granot, Friedman, Yarnitsky, et al., 2002; Masheb, Wang, Lozano, et al., 2005), may potentially constitute a partial predisposition to chronic vulvar pain and possibly moderate the severity of symptoms. It would, therefore, seem appropriate to study this pain syndrome from a psychophysiological perspective

(Hubbard, 1996; Keefe, Rumble, & Scipio, 2004; Severeijns, Vlaeyen, van den Hout, et al., 2005; Sullivan, Lynch, & Clark, 2005; Sullivan, Thorn, Haythornthwaite, et al., 2001; Turner & Aaron, 2001).

The interaction between emotions and physiology can occur at different levels of functional complexity. At the higher cortical level, functional Magnetic Resonance Imaging (fMRI) studies have demonstrated that mechanical/tactile allodynia primarily activates the anterior cingulate cortex and thalamus (Hofbauer, Olausson & Bushnell, 2006). These cortical structures are also known to be involved in the experience of conscious negative emotions (Coghill, Sang, Maisog, et al., 1999). At the peripheral sensory level, a significant interaction has been demonstrated in vulvodynia between anxiety and nociceptor sensitivity. Studies using quantitative sensory testing (QST) in female genital sensation (Vardi, Gruenwald, Sprecher, et al., 2000) showed that vulvodynia patients have lower pain thresholds in the vulvar vestibule (Hoehn-Saric & Mcleod, 2000; Lowenstein, Vardi, Deutsch, et al., 2004; Pukall, Payne, Binik, et al., 2003). A closer analysis of the difference in pain thresholds between vulvodynia patients and controls revealed that the difference was mediated by anxiety (Granot et al., 2002). Vulvodynia patients experienced enhanced pain perception, greater emotional response, increased autonomic reactivity, and scored significantly higher than controls on measures of state anxiety (unpleasant emotional reactions) and trait anxiety (personal tendency to respond with state anxiety), demonstrating the significant role of emotional states on nociceptor sensitivity.

When acute vulvar pain enters the chronic phase (vulvodynia), normal sensory processes are affected by progressive sensitization of the peripheral and central nervous system. Sensitization, is an important property of nociceptors and manifests itself in decreased thresholds to nociceptor stimulation; an increased field of nociceptor reception (from localized to generalized); nociceptor responsiveness to normally non-noxious stimuli (allodynia); increased intensity of response (hyperalgesia); prolonged post-stimulus sensations (hyperpathia); and the occurrence of spontaneous pain (Hawthorn & Redmond, 1998). Such sensory changes are the defining characteristic of vulvodynia. From QST studies (Vardi, Gruenwald, Sprecher, et al., 2000; Lowenstein, Vardi, Deutsch, et al., 2004; Pukall, Payne, Binik, et al., 2003; Granot et al., 2002), there is growing evidence that secondary hyperalgesia of the vulva is in part modulated by anxiety. This finding is

consistent with studies of other pain syndromes showing that the most reliable predictors of hyperalgesia are anxiety, fear, and catastrophizing (Boersma & Linton, 2006; Sullivan et al., 2005; Sullivan et al., 2001).

One of the known physiological correlates of anxiety is elevated muscle tension, a form of inefficient peripheral response to the hyper-alertness commonly seen in chronic pain patients (Hawthorn & Redmond, 1998). Muscles are the body's primary responders to pain, trauma, injury, and negative emotional states (Flor, Birbaumer, Schugens, et al., 1992; Gevirtz, Hubbard, & Harpin, 1996; Hawthorn & Redmond, 1998). Sensory and emotional stimuli can contribute to muscle over-activation, as assessed by sEMG, and can lead to stiffness, spasm and pain (Miller & Layzer, 2005), especially in symptomatic muscle groups (Flor, Birbaumer et al., 1992). sEMG monitored muscle overactivation has, in some conditions, proven to have diagnostic value (Miller & Layzer, 2005; White, Jantos, & Glazer, 1997). This symptom specific involvement of muscles has been extensively documented in vulvodynia studies, linking pelvic muscle hypertonicity (sEMG over-activation) and muscle instability to symptoms of vulvar pain (Glazer, Jantos, Hartmann, et al., 1998; Glazer, Rodke, Swencionis, et al., 1995; White, Jantos, & Glazer, 1997). With the evidence discussed it is important to further examine the relationship between emotions, muscle over activation and pain in the context of vulvodynia.

Method

The psychophysiological profile was developed via a retrospective review of patient files consisting of questionnaires, clinical interviews, psychological evaluations, case notes, and electromyographic assessments. Data were obtained only from patients who provided signed consent for their files to be used for research purposes. The study was approved by the Human Research Ethics Committee at the University of Adelaide.

All patients included in this study were initially examined and diagnosed by independent tertiary specialists working with vulvar pain patients (i.e. gynaecologists, dermatologists and sexual health physicians) and referred for therapy between the years 2000-2006. The diagnosis of

vulvodynia was made on the basis of generalized or localized vulvar pain (provoked or unprovoked) elicited by touch (cotton-tipped swab/speculum examination/manual palpation), or present without any provocation and occurring in the absence of any diagnosable pathology. The 529 patients included in the psychophysiological profile were selected on the basis of the following inclusion criteria: (1) patients were between the ages of 16 and 46; (2) satisfied the ISSVD classification of generalized vulvodynia, localised vulvodynia (previously known as vulvar vestibulitis), or clitorodynia; (3) experienced vulvar discomfort for a minimum period of 6 months; and (4) provided a valid response to the psychometric tests and physiological assessments. The age criterion was selected to reduce (because it was not possible to totally exclude) the potential impact of perimenopausal factors on profile data. The following exclusion criteria were used specifically in relation to the analysis of EMG data: (1) patients who were in recovery from recent pelvic surgery (i.e. within the last month); (2) cases known to be undergoing active treatment for current bacterial or fungal infection. All patients satisfying these criteria were included in this study.

Each participant completed an 82-item questionnaire assessing demographic information, general health, sexual health, use of health care, vulvar pain symptoms, and pain severity. Pain severity was assessed using a Numerical Rating Scale (NRS) where patients were asked to rate their pain from 0 to 10 (11-point scale) with the understanding that 0 represents no pain and a score of 10 represents the worst pain ever experienced. The validity of NRSs has been well documented showing positive and significant correlations with other measures (Jensen & Karoly, 1992). Several sections of the questionnaire were optional and the number of questions answered varied between individuals depending on the medical items reported. Additionally, participants were asked to complete the Symptom Checklist 90-Revised (SCL-90R), a well validated psychometric instrument (Derogatis, 1994), shown to be sensitive to psychological distress among vulvodynia patients (Wylie, Hallam-Jones & Harrington, 2004). The SCL-90R consists of the following nine primary symptom dimensions: somatization; obsessive-compulsive behaviour; interpersonal sensitivity; depression; anxiety; hostility; phobic anxiety; paranoid ideation; and psychoticism. The SCL-90R also provides three additional global indices of distress: a global severity index (GSI), measuring the depth of disorder

and intensity of distress; a positive symptom severity index (PSDI), measuring the symptom intensity; and a positive symptom total (PST), which measures the number of symptoms endorsed. In accordance with the aims of this study only the anxiety, depression and global scales were utilized.

At the time of the first consult, patients were asked to provide a history of their symptoms by means of questionnaires and guided interview; they then received an overview of the assessment procedures and sEMG assisted therapy and underwent an sEMG assessment consisting of a modified Glazer protocol (Glazer et al., 1995); and were introduced to a regime of twice daily sEMG assisted pelvic muscle exercises aimed at normalising general muscle tone according to criteria established by earlier research (Glazer et al., 1998; Glazer et al., 1995; White et al., 1997).

For the sEMG assessment, patients rested in a semi-supine position and readings were taken using a single-user vaginal sensor connected to a MyoTrac 3 encoder and analysed by computerized software (Vaginal sensor T6065, MyoTrac 3 T9900, and software manufactured by Thought Technology, Ltd, Montreal. Canada). Channel bandwidth was 20-450Hz, $\pm$5Hz/$\pm$50Hz and the sampling rate per channel was 2000 Hz. Data from the initial and final assessments were used to compare the amplitude of muscle activation and muscle instability (based on the standard deviation of resting amplitude) in a pre-treatment symptomatic state with that of a post-treatment and asymptomatic state.

At the time of the second appointment, scheduled 3-4 weeks after initial assessment, patients were guided in the use of dilators to measure the size of the introital opening and to gently release its narrowing. The muscle mediated narrowing of the introitus in symptomatic and asymptomatic states, was assessed by conical glass dilators (manufactured by Scientific Glassware, Sydney, Australia). The dimensions of the dilators (height x diameter – with a rounded conical tip) were: Size 1 = 80mm x 22mm; Size 2 = 85mm x 24mm; Size 3 = 95mm x 26mm; Size 4 = 105 x 30; Size 5 = 115mm x 34mm ; Size 6 = 125mm x 38mm.

Subsequent reviews of patients occurred every 3-4 weeks. At the conclusion of sEMG retraining and dilator exercises, psychotherapeutic interventions focussed on issues relating to fear

of sexual activity, loss of sexual desire, arousal disorders, stress management, generalised anxiety, depression and on relationship counselling. The number of psychotherapeutic sessions varied depending on the needs of the patient. Between 1-3 sessions were provided by a qualified psychologist. The criteria for conclusion of therapy included: stabilised sEMG readings of pelvic muscle function; reduction in discomfort and pain; and couple's ability to resume sexual intercourse.

Results

The mean age of the 529 patients was 27.7 years (median age =27.0 years; age range 16-46 years). The average age of symptom onset was 22.8 years (age range 5.5-45.2 years) and average duration of symptoms was 5.0 years (range 0.5-30.0 years).

Table I Summary of patient pain ratings for sexual intercourse

	N	Mean	SD	Range	Skewness
Penetration pain	480	8.0	2.0	1-10	-1.2 (SE=0.11)
Thrusting pain	464	7.6	2.1	1-10	-0.9 (SE=0.11)
Post intercourse pain	419	6.6	2.6	0-10	-0.8 (SE=0.12)
Most severe pain	429	9.2	1.2	3-10	-2.0 (SE=0.12)

The common pain locations listed by patients (many patients identified more than one location) included: the vestibule (48.2%); perineum (33.6%); clitoris (27.4%); urethra (23.3%); and anus (14.4%). The common and multiple descriptors for vulvar discomfort used by patients were: burning (79%); rawness (73.2%); itching (23.6%); sharp stabbing (9.6%); tearing (5.1%); stinging (4.3%); and tightness (4.0%). Other less frequent descriptors were tingling, heaviness, drawing, and throbbing.

In the 82-item questionnaire patients were asked to provide four separate pain ratings for sexual intercourse, reflecting the average level of pain experienced during recent attempts at penetration, with thrusting, post intercourse, and the most severe pain ever experienced. A summary of the four pain ratings is given in Table I. Figure 1A-D indicate the distribution of the sexual pain ratings for penetration, thrusting, post-intercourse pain and most severe pain

experienced. All four sexual pain ratings were significantly negatively skewed; that is, they did not follow a normal distribution of scores. Patients tended to rate pain related to sexual activity at the higher end of the 0 – 10 scale, indicating that it is experienced and perceived by patients as severe, disruptive, and distressing. The four pain ratings were correlated with depression, anxiety and the three global indices of distress from the SCL-90R as summarised in Table II. Significant but modest correlations were found between each of the four pain ratings and the Depression Dimension (p<0.01), Anxiety Dimension (p<0.01), Global Severity Index (0.01), Positive Symptom Distress Index (p<0.01), Positive Symptom Total (p<0.01).

Figure: Penetration Pain Ratings on a 10 point scale

Fig. 1 (a) Penetration pain: -1.18 (SE= 0.11)

Figure: Pain rating with thrusting on a 10 point scale

(b) Thrusting pain: -0.88 (SE= 0.11)

Figure: Post-Intercourse Pain Rating on a 10 point scale

(c) Post-intercourse pain: -0.77 (SE= 0.12)

Figure: Most Severe Pain Rating on 10 point scale

(d) Most severe pain: -2.01 (SE= 0.12)

To assess the potential role of pelvic muscle over-activation in vulvodynia symptoms, sEMG readings from the initial assessment of patients in a symptomatic state were compared with final assessment readings of patients in an asymptomatic state. Pre- and post-sEMG readings included

pelvic muscle resting baseline, standard deviation at rest, and phasic and tonic contraction amplitude. The sEMG readings are summarised in Figure 2 and show a normalization of pelvic muscle function between initial and final assessment. Using a paired samples t-test, all sEMG readings showed changes between initial and final readings as follows: a significant decrease in muscle resting baseline ($t(455)=21.2$, p<.001, $n^2 =.57$); a significant decrease in muscle instability as measured by standard
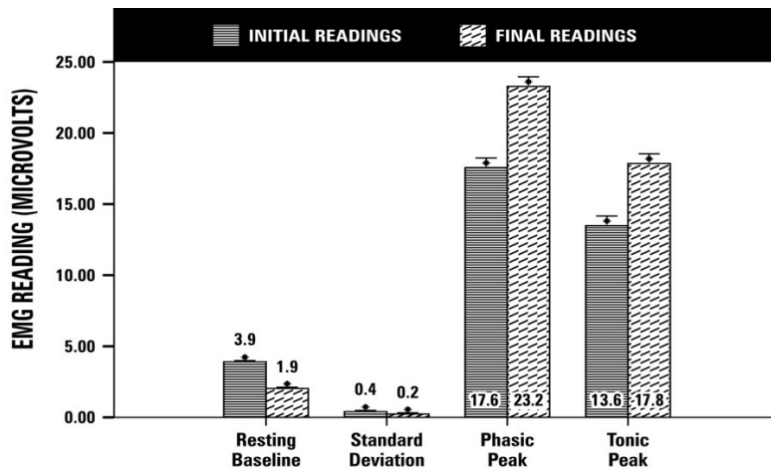
Fig. 2 Summary of sEMG readings for the initial and final evaluations (Error bars represent 1 Standard Error).

deviation ($t(455)=13.0$, p<.001, $n^2 = .34$); a significant increase in phasic contraction amplitude ($t(455)=5.2$, p<.001, $n^2 = .10$); and a significant increase in tonic contraction amplitude ($t(455)=5.9$, p<.001, $n^2 = .13$). The reduction in muscle resting baselines and muscle instability reflects a more relaxed pelvic muscle and the increases in phasic and tonic contractile amplitude reflect improved muscle tone as a result of biofeedback muscle retraining. No correlation was found between pain severity ratings and resting sEMG baselines or standard deviation readings at time of initial assessment. The lack of any correlation between these variables implies that no inference can be made in relation to severity of pain on the basis of sEMG resting baselines and standard deviation scores.

Table 2 Summary of Pearson's correlations (two-tailed) between SCL-90R dimensions and pain

	Penetration Pain	Thrusting Pain	Post-Intercourse Pain	Most Severe Pain
Depression Dimension	.18**	.21**	.15**	.18**
Anxiety Dimension	.17**	.13**	.13*	.11**
Global Severity Index	.19**	.18**	.17**	.16**
Positive Symptom Distress Index	.22**	.20**	.20**	.18**
Positive Symptom Total	.13*	.15**	.18**	.15**

The relationship between duration of pain and initial sEMG readings was also analysed and outcomes are summarized in Figure 3. The mean resting baseline reading and standard deviation scores were calculated for each of seven pain duration groups (i.e., 0.5-1.0 yr; 1-2yrs; 2-3 yrs; 3-4 yrs; 4-5 yrs; 5-10 yrs; 10+ yrs). A one-way ANOVA revealed that there was a significant difference between the groups in resting baseline readings ($F(6,458)=5.1, p<.001$) and standard deviation scores ($F(6,458)=3.5, p<.002$). Bonferroni corrected comparisons revealed that the 1-2 year pain group readings ($M = 5.1$) were significantly higher than the 5-10 year and 10+ year groups ($M = 1.3$, $p<.002$; $M =1.8$, $p<.001$); and the 6-12 month ($M = 4.4$) and the 2-3 year group ($M = 4.2$) had significantly higher readings then the 10+ years group ($M =1.4$, $p=.01$; $M =1.2$, $p=.03$). When looking at all the pain duration groups from 1 year of pain onwards, a two-tailed Pearson correlation showed that the size and significance of the negative correlation remained about the same (baseline rest: $p=-.22$, $p<.001$; baseline SD: $r=-.16$, $p<.001$). A partial correlation controlling for age, showed that the relationship between resting baseline sEMG amplitude and duration of pain was attenuated but still statistically significant ($r=-.18$, $p=.001$) as was the baseline standard deviation of readings ($r=-.10$, $p=.04$). The results show that progressive reduction of sEMG electrical activity could not be attributed to variables associated with ageing. Over time, there was a progressive physiological quieting of electrical activity of muscle tissue, but without any corresponding reduction in severity of pain symptoms.
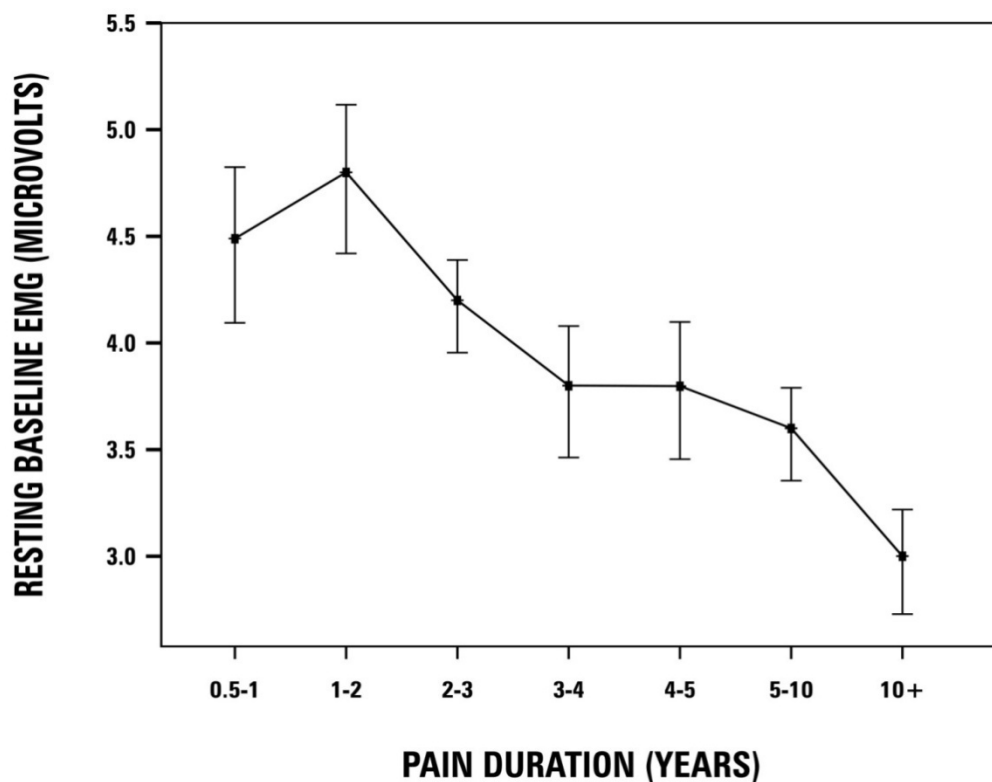
Fig. 3 Relationship between pain duration and initial EMG resting baseline (Error bars represent one Standard Error).

The size of the vaginal introitus was assessed by analysing patient records in relation to initial and final dilator sizes used. The following dilator sizes were initially used by patients; 12.7% commenced with Size 1; 24.2% with Size 2; 38.7% with Size 3; 17.8% with Size 4; 6.3% with Size 5; and 0.2 with Size 6. A total of 75.6% of the patients were initially limited to the use of only small dilator sizes (sizes 1-3). At the time of final assessment 96.7% were able to use the larger dilator sizes (sizes 4-6). A paired-samples t-test showed that there was a significant difference in initial and final dilator size used (t(343)=45.07, p<.001, mean difference =2.67, n^2 = .86). The patient's ability to use significantly larger dilator sizes at the conclusion of treatment indicated muscle tissue lengthening in response to dilator assisted muscle release. Progressive relaxation and re-opening of the introitus facilitated ease of sexual penetration. Due to resumption of pain free intercourse, a number of patients did not provide feedback on final dilator size used, resulting in a smaller statistical sample for analysis of data in relation to dilators.

The functional status at the time of initial assessment was as follows: Of the total study group 84.5% had been sexually active at some time prior to treatment, but 43.5% reported being totally sexually abstinent at the time of treatment, with 80.3% of the group reporting a reduction in sexual desire. The functional status at final assessment: 77.0% had resumed regular sexual activity; 4.0% reported successful conception prior to completion of therapy; 7.4% concluded therapy but remained sexually inactive by choice, or due to lack of partner; and 11.0% completed therapy but abstained from sexual activity by choice even though with a partner.

Discussion

The current study found a significant correlation between vulvar pain, anxiety, depression and global distress (as measured by the three global scales of the SCL-90R), highlighting a positive relationship between pain and negative emotions. Although it is not possible to establish causality on the basis of correlation, the prevalence of anxiety and depression among vulvodynia patients necessitates that further research attention be directed to examining the relationship between severity of vulvar pain symptom and intensity of emotional distress.

Furthermore, the consistently high and negatively skewed pain ratings associated with sexual intercourse confirms that vulvodynia, more so then other chronic pain conditions, has an emotionally and physically disabling impact on patients. Pain, once provoked, can be so intense as to disrupt sexual activity, arouse fear, lead to loss of sexual desire, avoidance, or total abstinence (Jantos & Burns, 2007). Unlike the normal distributions of pain ratings for other chronic pain conditions (Boersma & Linton, 2006; Meana, Binik, Khalife, & Cohen, 1997; Pukall et al., 2003), vulvar pain is consistently rated high on pain scales, confirming that is has a disruptive and disabling impact on the quality of life of the women affected.

The difficult question that arises from these findings is whether the pain contributes to emotional distress or emotions contribute to pain. Anxiety and depression have been shown to be prevalent in most other pain conditions (Keefe et al., 2004; Sullivan et al., 2001), with anticipatory

fear and pain related hyper-vigilance among the most common traits in chronic pain patients (Sullivan et al., 2005). Research with vulvodynia appears to confirm that elevated anxiety may be a contributing factor to secondary hyperalgesia (Granot et al., 2002). In a similar manner, it may be possible that depression, because of its high prevalence (Jantos & White, 1997; Masheb, Nash, & Brondolo, 2000), may act not only as a passive pain coping strategy but another contributing factor in the psychophysiological complexity of this pain syndrome. However, to date there is no conclusive evidence for establishing causality. Further research into predisposing personality and physiological characteristics is required to assist in identifying causal relationships. In general, this study highlights the importance of not only focussing on important physiological variables (Bohm-Stark et al., 2001; Lowenstein et al., 2004; Wylie, Hallem-Jones, & Harrington, 2004) but also taking into consideration emotional variables when studying chronic vulvar pain (Masheb, Wang, Lozano, et al., 2005; Samwel, Evers, Crul, et al., 2006).

Data from sEMG findings in this study appear to provide further information on the potential mechanisms underlying the symptoms of vulvodynia. In discussing sEMG findings it is important to state at the outset that the sEMG signal should always be seen as a correlate of muscle activation, providing insight into muscle function, but not a direct measure of muscle pain (Miller & Layzer, 2005). This study showed no correlation between sEMG resting amplitude or standard deviation in relation to the intensity of pain. This is consistent with most of the earlier research findings (Kendall, Elert, Ekselius, et al., 2002; Miller & Layzer, 2005), but contrary to an earlier vulvodynia study based on a small sample of patients (Glazer et al., 1998). sEMG data from this study provides qualified support for earlier studies showing an association between pelvic muscle over-activation and vulvodynia symptoms (Glazer et al., 1998; Glazer et al., 1995; White et al., 1997). In general, muscle over-activation has been identified as a factor in a range of chronic pain syndromes (Flor, Birbaumer et al., 1992; Flor, Miltner, & Birbaumer, 1992) and the mechanisms by which muscle over activation leads to pain have been extensively discussed. It is accepted that muscle over activation leads to muscles decompensating in a painful manner (Burton, 1996). Irrespective of whether muscle tension is due to emotional or physical triggers, ischemia, hypoxia, build up of neurogenic metabolites (lactic

acid, potassium, arachidonic acid), alterations in intramuscular blood flow, release of sensitizing

agents (such as bradykinin and serotonin), inflammation, erythema, oedema formation, muscular

rigidity, all can ultimately lead to pain. Yet there is no relationship between pain and sEMG

amplitude.

A conceptually challenging finding arising from a further analysis of sEMG data is the

negative correlations between resting amplitude and standard deviation readings, and the duration

of pain. As shown in Figure 3, chronicity of vulvar pain appears to be associated with a progressive

reduction in muscle electrical activity. This paradoxical finding may be associated with a time related

physiological "shut-down" of over activated muscles (Cailliet, 1996). Clinically, it is not uncommon to

see long-term vulvodynia patients who complain of debilitating pain and introital muscle tightness

but who on assessment, show sEMG readings that are exceptionally low and stable, resembling

those of symptom free individuals. This anomaly may be associated with progressive shortening of

over-active muscle tissue and the development of a muscle contracture that is consistently

described by patients as painful and as an obstruction of the vaginal introitus.

Muscle contracture has been previously described as consisting of "an electrically silent,

involuntary state of maintained muscle shortness and decreased extensibility (i.e. increased

stiffness) of the passive elastic properties of the connective tissue" (Cailliet, 1996; Cram, Kasman, &

Holtz, 1998). In relation to vulvodynia, such a contracture appears to be mediated by an anteriorly

oriented tractioning of the pubococcygeus muscle (in particular the pubovaginalis and puborectalis

portions) with anal retraction and in clitoral descent. The loss of pelvic muscle extensibility appears

to be consistent with findings derived from assessments using graded vaginal dilators. At the

commencement of therapy, patients were only able to accommodate the smaller size dilators. This

also appears to coincide with difficulties using tampons and undergoing medical exams. However, it

is also very evident that such pelvic contractures are not permanent but "functional" (Cailliet, 1996)

and can be reversed by conservative therapy involving the use of dilator exercises and sEMG assisted

retraining.

Muscle contractures may also give rise to myofascial "trigger points," tenderness, and pain (Cailliet, 1996; Hubbard, 1998; Simons et al., 1999). Physical pressure on hypertonic and shortened muscles produces symptoms of sharp, shooting and burning pain in the absence of physical evidence. These anomalies are consistent with myofascial pain, which often causes functional difficulties but has no visible pathology.

Vulvodynia symptoms are frequently attributed to, and misdiagnosed as vaginismus. Vaginismus is defined as a "recurrent and persistent involuntary spasm of the musculature of the outer third of the vagina that interferes with sexual intercourse" (APA, 1994). The similarities between the two conditions are that a functional contracture can resemble the acute shortening of muscles in spasm, but the main difference between a contracture and muscle spasm is the striking sEMG characteristic; contractures are electrically silent (Miller & Layzer, 2005). However in vulvodynia there is no sEMG evidence of any spasm-like activity in pelvic musculature. The existence of normal reflexes, such as the bulbo-cavernosus or sacral reflex and their modulation by the involvement of supraspinal sites, should not be confused with vaginismus (McKenna, 2002). Considerable confusion surrounds the classification of vaginismus both in relation to vulvodynia, or as an independent diagnostic entity in itself (Binik, Meana, Berkley, et al., 1999; Reissing, Binik, Khalife, et al., 2003). This study found no evidence of involuntary spasm in vulvodynia and sEMG assessment can assist in differentiating vulvodynia from other diagnostic conditions such as vaginismus.

Vulvodynia patients commonly report urethral and bladder symptoms in conjunction with vulvar pain. Previous studies have explored the possibility of parallel pathologies for vulvar pain and interstitial cystitis (Stewart & Berger, 1997) and there appear to be some similarities in symptoms reported (e.g., discomfort, burning and stinging sensation). Several studies of these co-morbidities have confirmed muscle over-activation, lack of voluntary control, inability to relax, shortening of muscles and trigger point referred pain, as well as hypersensitivity exacerbated by anxiety, sexual and physical activity (Bernstein et al., 1992; Brookoff & Bennet, 2006; Fitzgerald & Kotarinos, 2003a, 2003b; Oemler, Grabhorn, Vahlensieck, et al., 2006; Weiss, 2001). Ultimately, further research into

these co-morbidities is required (Arnold, Backmann, Rosen, et al., 2006) but may show that vestibulodynia, clitorodynia, generalized vulvodynia, urethral syndrome, and abacterial interstitial cystitis may share a common mediating mechanism and may be symptomatic of pelvic muscle dysfunction distinguished only by the locale of the pain.

A clinical protocol focussed on sEMG assisted retraining of pelvic muscles and the release of muscle contracture may alleviate, if not totally relieve the common symptoms of vulvodynia. However, due to the complexity of human sexuality alleviating symptoms of discomfort and pain may not automatically lead to the commencement or resumption of sexual activity. Considering that 11-17% of patients remained sexually abstinent, highlights the need to address psychological difficulties that patients experience. With the prevalence of anxiety, sexual fear and depression, patients also need to receive sexual and relationship counselling and be helped to understand the relationship between emotional states and chronic pain. More than 80% of patients in this cohort were able to resume regular sexual activity as a result of adopting a psychophysiological protocol in the management of vulvodynia.

Conclusion

This retrospective review of vulvodynia patients confirms the need to view chronic pain from a psychophysiological perspective which recognizes the intricate interaction of psychological and physiological mechanisms in the experience of pain. The absence of visible pathology should not be the reason for either seeking psychological explanations or questioning the reality of a patient's pain. One of the anomalies of chronic pain syndromes is the absence of pathology to explain the severity of reported pain. In vulvodynia, the subtle physiological changes associated with over-activation of pelvic muscle may be associated with the development of a functional contracture and may be exacerbated by emotional factors. Conservative therapy, followed by qualified counselling can successfully restore pain free sexual function.

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Chapter Five

Electromyographic assessment of female pelvic floor disorders

Chapter Six

Overview of Findings

The two research studies entitled: *Vulvodynia: Development of a psychosexual profile*; and

Vulvodynia: A psychophysiological profile based on electromyographic assessment; and a discussion

paper: *Electromyographic assessment of female pelvic floor disorders;* provide insight into the nature

of vulvodynia and its impact on women's wellbeing. The information derived from the three papers

contributes to an understanding of vulvodynia by providing information on:

- the clinical population of women affected,

- the impact of the disorder on their emotional, sexual and social wellbeing,

- the interaction of psychophysiological variables in the experience of vulvar pain,

- the potential mechanisms involved, and

- the rationale for the classification of vulvodynia as a chronic regional pain syndrome.

The research presented in this thesis is derived from the largest clinical sample of vulvodynia

patients studied to date. The study sample enabled an extensive analysis of psychosexual and

psychophysiological variables pertinent to vulvodynia. The work is original and derived from the

author's clinical involvement with vulvodynia patients through a network of Australian clinics.

Summary of findings from first study

The first research study, *Vulvodynia: Development of a psychosexual profile* assessed the

profile of a cohort of 744 vulvodynia patients. The study provided data in relation to the following

key points:

- <u>The age specific incidence of the disorder</u>. This study was the first to examine the

    age specific prevalence of vulvodynia. The need for such research was identified in

    earlier studies (Harlow & Stewart, 2003).

The findings revealed that although vulvodynia affects women of all ages, it predominantly affects young women in their teen and early adulthood years with its prevalence peaking at approximately 24 years of age. More than 50% of women affected by vulvodynia were under 25 years of age, and 75% were under 34 years of age. It has been suggested that the higher prevalence among younger women may indicate that women of reproductive age are more likely to seek gynaecological care, and therefore contribute to a higher proportion of clinical cases (Harlow & Stewart, 2003). Other studies have also noted an early onset of the disorder (Rogstat, 2000; Baggish & Miklos, 1995). The cause of the early onset of symptoms needs to be further studied.

Without under-estimating the negative impact of vulvodynia on the wellbeing of women of all age groups, the findings of this study highlight the negative impact of the early onset of symptoms on sexual confidence, on the capacity to enter into intimate relationships and on fertility rates. Onset of symptoms, prior to the commencement of sexual activity, contributes to considerable anxiety, fear and apprehension about entering into relationships, with young women often questioning their suitability as sexual partners. This particularly applies to primary vulvodynia cases who, according to this study, made up at least a third of the vulvodynia population. The younger women, unlike women with secondary vulvodynia, have no reference point by which to judge what constitutes "normal," pain-free, sexual activity. As a result, they experienced negative emotions towards sexuality, delayed relationships, avoided intimate sexual encounters, and often opted to remain single.

- The age of symptom onset. This study was the first to document the age related onset of symptoms. Many of the vulvodynia patients gave accounts of experiencing symptoms during their early childhood, teen years, or early adulthood.

From analysis of data, the age of symptom onset peaked at 19 years of age for primary vulvodynia cases (a subgroup in which onset predates or coincides with commencement of sexual activity) and at 25 years of age for secondary vulvodynia cases (a subgroup in which the onset follows a period of pain-free sexual activity). The ages for onset of symptoms ranged from 5.5 – 45.2 years of age.

Women in this study suffered symptoms for an average period of five years (range 0.5-30 years). When considering the age distribution of patients and the age of symptom onset, it is evident that for a large percentage of women the onset of vulvodynia symptoms is totaly unrelated to commencement of sexual activity.

In the case of primary vulvodynia, symptoms were often noticed during childhood play, with first use of tampons, or first medical examination. Because of the early onset, unrelated to sexual activity, a causal attribution of symptoms to psychosexual conflict, relationship stressors, marital disharmony or sexual dysfunction is not supported and is unwarranted. Since the publication of this study, another report based on a retrospective review of clinical records confirmed the diagnosis of vulvodynia in children as young as 4-11 years of age (Reed & Cantor, 2008).

With onset of symptoms occurring at such an early age, future studies need to further assess the impact of chronic urogenital pain on the development of body image, self-esteem, personal confidence and attitudes toward sexual behaviour in adult years.

- The age related risk of developing chronic vulvar pain symptoms. The data provide further evidence that the prevalence of vulvodynia peaks prior to age 25 and progressively decreases thereafter. This finding is consistent with the report of an earlier study (Harlow & Stewart, 2003). Only a slight increase in prevalence was noted around the peri-menopausal years. Therefore, the risk of developing the disorder is highest during the teen years and early adulthood.

Other studies, based on smaller patient samples, have suggested that "vulvodynia was most strongly correlated with increasing age" (Arnold et al., 2006). These earlier reports were not comfirmed by data from this study. The discrepancy in findings may have arisen on account of sampling error associated with the use of mail questionnaires, and reliance on respondent's for self-diagnosis, whereas the data in this study were derived from a clinical sample of patients who were diagnosed by experienced tertiary specialists.

In a number of earlier studies, the reported average age of patients was significantly higher than the average age reported in this study (Friedrich, 1987; Bachmann et al., 2006; Arnold et al., 2006). This also may have given rise to the impression that vulvodynia is more common in later years. This study confirmed that the risk of symptom onset is greatest prior to twenty-five years of age.

- Proficiency of diagnosis. The average duration of symptoms prior to reported diagnosis was 5 years, with a range of 0.5 - 30.0 years.

  In over 60% of the cases, 3-24 consultations took place prior to diagnosis, and in 16% of cases, more than 24 consultations were required. Some evidence suggests that as few as 1% of women with symptoms receive a diagnosis of vulvodynia (Arnold et al., 2006), and of the women affected by symptoms only about half seek medical assistance (Harlow & Stewart, 2003).

  From the results of this study, it is evident that more information needs to be disseminated to women and clinicians to raise general awareness about the existence of the disorder. For a health issue as prevalent as vulvodynia, it is difficult to justify such long delays in diagnosis, or such frequent misdiagnoses.

- Impact of vulvodynia on social relationships. When primary vulvodynia patients were compared with secondary cases, the early onset of vulvodynia was associated with fewer intimate relationships and postponement of sexual involvement. Some

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patients intentionally avoided relationships to evade potential embarrassment and preserve self-esteem.

Of the women who were in relationships, 75% indicated that their partners were supportive. Those in married relationships suffered less depression and anxiety. This research provided no support for the hypothesis that vulvodynia is the result of relationship tension and psychosexual conflict.

- <u>Impact of vulvodynia on emotional wellbeing.</u> As a group, vulvodynia patients showed increased levels of depression (40% of cases scoring in the clinically significant range), and increased anxiety (20% of the cases scoring in the clinically significant range). Higher scores on depression and anxiety do not necessarily equate with a clinical diagnosis of affective disorder which can only be made on the basis of a more comprehensive assessment.

  Those most affected by depression and anxiety were single young women with a history of early onset of symptoms (primary vulvodynia cases). On the other hand, depression and anxiety levels were lowest among married women. Marriage appeared to be an effective buffer against depression and anxiety. It may be that marriage provided a more secure relationship and a higher level of support. De facto relationships and long-term friendships showed no such protective effect.

- <u>Impact of vulvodynia on sexual behaviour.</u> Over 80% of vulvodynia patients reported a significant decrease in sexual desire. The decrease in sexual desire was associated with:

  1. Reduced frequency of sexual intercourse

  2. Sexual activity being less enjoyable

  3. Increased negative emotions towards sexual activity

  4. Lower physiological arousal

  5. Decreased levels of partner initiation.

Analysis of data derived from the responses of secondary vulvodynia cases showed that the frequency of intercourse changed significantly from a pre-morbid frequency of 3.5 times per week, to 0.6 times per week after symptom onset.

On average primary vulvodynia patients on average gave a significantly lower satisfaction rating for their first sexual encounter when compared with secondary vulvodynia patients (on an eleven point scale of 0-11, the average rating by primary patients was 2.5 and for secondary patients 4.3).

Furthermore, patients reported a notable decrease in their partner's sexual desire and a reduction in the partner's initiation of sexual intercourse, presumably on account of increased worry that intercourse would cause pain.

- <u>Sexual abuse and etiology of symptoms.</u> This study confirms earlier findings that the incidence of sexual abuse is no higher among vulvodynia patients than in the general population (Dalton et al., 2002; Edwards et al., 1997).

  In this study, the reported incidence of unwanted sexual activity in childhood was 11.5% and in adulthood 12.5%. The evidence does not support the view that abuse may be a causal factor in the etiology of the disorder as has been suggested elsewhere (Harlow & Stewart, 2005; Lynch, 2008).

- Desire to increase frequency of sexual activity

  It is evident that the primary reason for the reduction in frequency of sexual activity is the symptom of pain which not only has a significant disabling effect on daily activities but also interferes with the ability to engage in sexual intercourse. When patients were asked which activities they would like to increase if symptoms were resolved (patients could identify multiple activities), 87.5% expressed a desire to increase the frequency of sexual activity, 27.4% to increase physical activity and 8.1% to increase work activity.

  These findings provided evidence that pain-free sexual function is a highly valued activity by women and was a primary motivator for pursuing treatment. There is no

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evidence that the reduction in sexual activity is due to malingering, manipulative behaviour, psychosexual conflict, sexual dysfunction, or other psychological causes. The finding that pain is the primary barrier to frequency of sexual activity is consistent with earlier reports (Meana et al 1997; Reed et al 2000).

- <u>Reproductive potential.</u> Considering that 95% of the patients in this study were of reproductive age, and 73% were nulligravidas, the condition has the potential to significantly impact on general reproductive potential of sufferers.

  Among the vulvodynia cohort were women who could be best described as married virgins because they were unable to sexually consummate their marriages. Approximately 5% of the married women (who made 37% of the total study cohort) were unable to consummate their marriage through sexual intercourse for a period of 2-15 years into marriage. A number of the married couples could only conceive with the assistance of in-vitro fertilization.

- <u>Recommendations based on results of the study:</u>

  1. Because vulvodynia is a highly prevalent disorder and its incidence appears to be increasing, it is essential to disseminate information that would assist women and care-givers to recognise the symptoms, and enable early diagnosis.

  2. Due to the early onset of symptoms, future studies need to examine how the early pain related focus on the urogenital area affects children's and teen's development of body image, self-esteem and attitudes towards sexual behaviour in later adult years. Such developmental issues need to be recognised and addressed in therapy.

  3. Appropriate models for therapeutic intervention, addressing all aspects of health and wellbeing need to be developed focussing on physical, social and emotional wellbeing.

  4. Management of pain needs to be the focal point, especially in the initial stages of therapy. A focus on the physiological aspects of pain will confirm in the patient's

mind that the carer accepts that the pain is "real" and not of psychogenic origin. The emotional and sexual aspects of the disorder can be addressed in later stages of therapy.

5. The least intrusive and conservative therapies should be advocated as first line therapies, leaving the more invasive, surgical procedures, as last line interventions (Haefner et al., 2005).

6. Therapeutic management of vulvodynia requires a multidisciplinary approach (Goldstein et al., 2005) and should be carried out by skilled professionals who have a good knowledge of the lower urogenital tract, and are competent to recognise and address all the issues contributing to the distress and disability associated with the disorder (Jones, 2000). All care providers should be mindful that vulvodynia has a significant impact on social, emotional and sexual wellbeing, and these issues should be addressed in the course of therapy.

The study provided important information on the onset, pervalence and psychosexual impact of vulvodynia on women.

Summary of findings from second study

The second study, *Vulvodynia: A psychophysiological profile based on electromyographic assessment,* assessed the psychophysiological profile of 529 vulvodynia patients. The study examined the interaction of psychophysiological factors, in particular the impact of psychological distress on the experience of chronic vulvar pain. The interaction between emotions and physiology can occur at various levels of functional complexity, involving higher centres in the central nervous system as well as peripheral mechanisms. For approximately two decades, research has sought to identify specific triggers and causes of vulvodynia symptoms. Much of the focus has been on either neuropathic or psychogenic causes of the disorder. However, based on the current scientific knowledge of the intricate links between mind and body, and the symptom specific physiological

responses to pain (Flor & Turk, 1989), it was important to assess the role of peripheral

neuromuscular changes through a psychophysiological assessment of women with vulvodynia.

Since the experience of pain consists of sensory and emotional variables , vulvodynia

patients have been shown to experience enhanced pain perception, greater emotional reactivity,

physiological rigidity, increased autonomic response and dysregulation, and higher scores on trait

and state anxiety (Granot et al., 2002) in a manner consistant with other chronic pain conditions

(Hoehn-Saric & McLeod., 2000). On the basis of this evidence, it was important to explore the

relationship between depression, anxiety and psychological distress and severity of pain; and to

examine the relationship between pain and the functional status of pelvic muscle as assessed by

SEMG.

The study provided important data in relation to the following key points:

- <u>Location of the pain and sensory descriptors.</u> The most common pain areas

  identified by women (involving multiple sites) in the urogenital area, included; the

  vestibule (48.2%); perineum (33.6%); clitoris (27.4%); urethra (23.3%); and anus

  (14.4%). The very precise localization of the pain affected areas confirms that

  vulvodynia is a regional pain syndrome

  The most common pain descriptors used by patients included burning (79%);

  rawness (73.2%); itching (23.6%); sharp stabbing (9.6%); tearing (5.1%); stinging

  (4.3%) and tightness (4.0%). Some of the descriptors used are of a thermal and

  incisive nature and further highlight the involvement of peripheral sensory

  mechanisms (Bergeron et al., 2001).

  Although some studies have identified physiological peculiarities in the form of

  increased density of superficial nerve endings, increased immunoreactivity and

  nociceptor sensitivity in the vulvar tissue of vulvodynia patients (Bohm-Stark et al.,

  1998), it is highly improbable that such physiological parameters alone would fully

  explain the extent and severity of urogenital pain reported by patients (Lynch,

  2008). It is more likely that multiple mechanisms are involved.

- Severity of pain associated with sexual intercourse. The pain ratings given by patients in relation to sexual intercourse were based on an eleven point scale (where 0 represents no pain and a score of 10 represents the worst pain ever experienced). An analysis of the pain scores for penetration ($M = 8.0$), thrusting ($M = 7.6$), most severe pain experienced ($M = 9.2$) and post-intercourse pain ($M = 6.6$), did not follow a normal distribution as previously suggested (Meana et al 1997; Pukall et al 2003), but were consistently negatively skewed. This indicates that the pain experienced by vulvodynia patients is severe and very disruptive. Several other reports have confirmed the severity of vulvar pain, with the subjective ratings of pain by patients falling in the 7.0-7-5 range on an eleven point scale (White & Jantos, 1998; Bergeron et al., 2001).

  The severity of pain is the primary reason for the loss of desire, avoidance of sexual activity, abstinence and decreased satisfaction with sexual activity. Sexual activity was impaired on account of the disabling nature of vulvar pain.These findings were consistent with those of other studies (Reed et al., 2000; Meana et al., 1997). These findings confirm earlier reports which found that for vulvodynia patients, "pain during intercourse is the most significant limitation that vulvodynia places on their daily functioning" (Kaler, 2005, p 34). As has been reported in the first study (Jantos & Burns 2007), the severity of pain results in a dramatic reduction of intercourse frequency (from 3.5 to 0.6 times per week), with 87.5% of patients expressing a desire to increase the frequency of sexual activity once symptoms were resolved.

  In the absence of pain, vulvodynia patients are not different from other women in the quality of relationships, interest in sex, importance of vaginal sex or frequency of sexual activity (Reed et al., 2000; Meana et al., 1997).

- Severity of pain, depression, anxiety and general distress. The severity of pain showed modest but significant correlations to depression, anxiety and three indices

of symptom related distress. As reported in earlier studies, the sensory and emotional aspects of pain do not produce high correlations (Binik et al., 1999; Pukall et al., 2003). However, this moderate correlation indicates that pain severity may be the primary contributing factor, if not the cause, of the emotional distress noted in patients.

Considering the findings of earlier studies, depression appears to be reactive and arises on account of the physically and emotionally disabling effects of chronic vulvar pain (Reed et al., 2000). Although it is not possible to establish a causal relationship on the basis of correlation, the prevalence of depression among vulvodynia patients and the severity of vulvar pain reported may be directly related to the emotional distress experienced by patients (McKay, 1987; Reed et al., 2000). The prevalence of depression among women with vulvodynia is consistent with prevalence of depression in other chronic pain syndromes (Samwel et al., 2006; Bar et al., 2005).

Due to the difference in the impact of anxiety and depression on pain perception, the effect of the two emotional states needs to be studied independently (Bar et al., 2005).

- Relationship between SEMG and severity of pain. Resting baseline SEMG readings of pelvic floor muscles showed no correlation with the severity of pain.

The lack of correlation implies that no direct inference can be made about the severity of pain on the basis of SEMG readings. These findings clearly identify the fact that SEMG signal is a correlate of muscle activation, but should never be seen as a measure of pain.

There is no evidence of generalized physiological dysregulation in chronic pain syndromes (Hoehn-Saric & McLeod, 2000), instead a symptom specific response is common (Flor & Turk, 1989). The data in this study confirms an association between

pelvic muscle dysfunction and symptoms of vulvodynia, but there is no linear correlation between severity of pain and SEMG readings.

- Role of pelvic muscle over-activation and symptoms of vulvodynia. The SEMG readings of pre-treatment symptomatic patients were matched and compared with post-treatment asymptomatic readings. In comparing the pelvic muscle resting baselines, standard deviation of readings at rest, and phasic and tonic contraction amplitude pre-and post-treatment, several significant changes were noted as a result of therapy. These included:

    (1) A decrease in muscle resting baseline

    (2) A decrease in muscle instability as measured by standard deviation readings

    (3) An increase in phasic contraction amplitude

    (4) An increase in tonic contractile amplitude.

The reduction in muscle resting baselines and muscle instability is associated with a relaxed and stable muscle state. Improved phasic (fast-twitch fiber mediated activity) and tonic (slow-twitch mediated activity) contractile amplitude is indicative of improved muscle tone and greater resistance to fatigue. These changes in SEMG readings are associated with normalization of pelvic muscle function and reduction in vulvodynia symptoms.

Muscle tension is a recognised correlate of pain and anxiety (Hoehn-Saric & McLeod, 2000; Flor & Turk, 1989) and can be activated by a range of noxious stimuli (Schattschneider et al., 2006; Ge et al., 2008; Hawthorn & Redmond 1998) as well as negative emotional states (Hubbard, 1996; McNulty et al., 1994).

SEMG assessments noting overactivation of pelvic muscles were shown to be reliable in the diagnosis of vulvodynia (White et al., 1997; Glazer et al., 1995), and in the differential diagnoses of several pelvic floor disorders (Glazer et al., 1998). In the case of vulvodynia, whether muscle over-activation occurs in response to noxious

irritants, infections and inflammatory reactions, physical trauma, anxiety, or a combination of these factors, its presence is not disputed (Bachmann et al., 2006; Haefner et al., 2005).

Neuromuscular overactivation is a physiological feature of vulvodynia and a peripheral physiological system involved in the mediation of pain. This is consistent with the findings in relation to other complex regional pain syndromes (Schattschneider et al., 2006).

- <u>Further findings highlighting potential mechanisms mediating vulvar pain.</u> Findings from this study confirm physiological changes in muscle tissue arising from muscle over-activation.

The negative correlation between resting SEMG baselines and duration of pain reported in this study is a convincing indicator that chronic overactivation of pelvic muscles leads to a progressive quieting of electrical activity in muscle tissue, and a gradual physiological "shut-down" that is typically associated with muscle shortening and the development of a muscle contracture (Calliet, 1996). A muscle contracture is an "electrically silent, involuntary state of maintained muscle shortness and decreased extensibility (i.e. increased stiffness) of the passive elastic properties of the connective tissue" (Calliet, 1996).

Considering that the primary function of the pubococcygeus muscle is the compression of the urethral, vaginal and rectal lumen against the pubic rami (DeLancey, 1999; Ashton-Miller & DeLancey, 2007), its over-activation and shortening results in the narrowing of the vaginal introitus, with increased tension in the urethral and anal sphincter muscles. In vulvodynia, hyperalgesia and restriction of the introitus are the primary symptoms.

The most common comorbidities in vulvodynia include bladder, urethral and rectal pain (Kennedy et al., 2007; Arnold et al., 2006; Peters et al., 2008; Gardella et al., 2008). Yet, pain in any of these localized areas may arise from the same

neuromuscular mechanisms. Current evidence confirms that muscle overactivation and pelvic muscle pain (sometimes referred to as pelvic myalgia) are prominent features in bladder pain syndrome (Peters et al., 2008; Gardella et al., 2008). Further evidence of muscle shortening and loss of muscle tissue extensibility comes from the use of standardized dilator. To evaluate the degree of muscle contracture, patients would select and self-insert a dilator that could be accommodated by the vaginal introitus. Initially patients were only able to use small size dilators, but with progressive normalization of muscle function progressed to larger size dilators. Because muscular contractures are functional and not permanent (Caliet, 1996), and sensitization is peripheral as opposed to central, both the contracture and sensitization can be reversed through conservative therapy. This study showed that by normalizing pelvic muscle function, 80-90% of the patients were able to resume pain free sexual function. This is a significant finding and carries important implications for treatment.

- Recommendations based on results of the study:

  1. This study confirms that vulvodynia is a regional pain syndrome by accurately identifying the bodily region affected by pain, and highlighting certain temporal pain characteristics commonly provided by patients. As a result, vulvodynia needs to be classified as a pain syndrome.

  2. The primary focus of therapy needs to be on the management of pain, not to the exclusion of other aspects of therapy, but as an important part of restoring wellbeing.

  3. SEMG readings provide evidence that the neuromuscular system is one of the primary physiological mechanisms involved in mediating vulvar pain. Chronic muscle over-activation appears to lead to physiological changes in pelvic muscles which can give rise to ischemic pain and myofascial pain. Pelvic muscle normalization should be a primary focus of therapy.

4. When researching and managing chronic pain syndromes like vulvodynia, it is important to take into consideration not only the sensory, but also the emotional aspects of pain, without over ascribing to the psychological issues. Even though there may be certain predisposing personality traits and physiological variables, it is clear that emotions do modulate the experience of pain and interact with neurological and muscular functions (Flor & Turk, 1989).

5. Further studies need to focus in greater detail on the role of muscle ischemia and myofascial trigger points in the onset of peripheral sensitization.

The study assessing the psychophysiological profile of women with vulvodynia provided new evidence pointing to peripheral physiological mechanisms mediating symptoms and confirming that the disorder constitutes a regional pain syndrome.

Summary of themes in discussion paper

The discussion paper entitled, *Electromyographic assessment of female pelvic floor disorders*, provides an extensive review of literature on pelvic disorders mediated by dysfunctional muscles. Pelvic floor disorders are a cluster of pain, incontinence and sexual disorders that arise primarily out of dysfunctional muscle states and structural changes, rather than malfunction of specific pelvic organs (Ashton-Miller & DeLancey, 2007). Following a review of pelvic floor anatomy and physiology, the discussion paper focused on disorders associated with hyper-tonus (over-active) and hypo-tonus (under-active) muscle states. Chronic vulvar pain was examined in the context of a spectrum of pelvic muscle related disorders. The following is a summary of keypoints:

- <u>Disorders associated with hyper-tonus muscle states</u>. Hyper-tonus muscle states are commonly associated with chronic pain conditions such as vulvodynia, interstitial cystitis (IC)/bladder pain, urgency and frequency. The evidence linking muscle over-activation to vulvodynia has been extensively discussed in the research studies (Glazer et al., 1995; White et al., 1997; Glazer et al., 1998; McKay et al., 2001). SEMG characteristics enabled an 88% accurate differential diagnosis of vulvodynia

patients. When compared with controls, SEMG assessments of women with vulvodynia demonstrated elevated resting baselines (hypertonicity), poor contractile amplitude (weakness), higher resting standard deviation readings (instability), poor recovery post contraction (irritability), and low frequency muscle fibre activity (fatigue) (White et al., 1997). These findings were validated by manual assessments carried out by specially trained physical therapists who found that 90% of the women reporting pain with intercourse demonstrated pelvic floor dysfunction and pathology (Reissing et al., 2005).

Other studies provided evidence of overactive and painful pelvic muscles in IC/painful bladder syndrome (Weiss, 2001; Fitzgerald & Kotarinos, 2003). Due to the involvement of a common mechanism, namely overactive pelvic muscles, vulvodynia patients and IC/painful bladder syndrome cases share several common symptoms. As many as 77% of IC patients reported pain with sexual intercourse; 69% described it as burning pain and 85.1% meet the diagnostic criteria for provoked or generalized vulvodynia (Gardella et al., 2008). By contrast, only 23.4% of the IC patients reported bladder pain and 51.1% reported urgency and frequency. From the studies reviewed, it was evident that the more prevalent symptom in IC patients is chronic vulvar pain in the form of vulvodynia. The patients who presented with IC and vulvodynia like symptoms were found to have significantly higher levels of levator pain in which pelvic muscles were tender to palpation, produced referred pain to the suprapubic, perineal area, rectum and labia, and experienced evacuation difficulties and anal fissures (Peters et al., 2007; Reissing et al., 2005; Gardella et al., 2008).

The most effective therapies focused on the normalization of muscle function, through SEMG assisted downtraining of overactive muscles, release of myofascial TrP release, and pelvic muscle stretching. A 65% reduction in SEMG readings was

associated with marked improvement of IC, urgency and frequency symptoms in 73-83% of the cases respectively.

- Disorders associated with hypo-tonus muscle states Hypo-tonus muscle states are characterized by under-activation and weakness, and are associated with symptoms of urinary incontinence and sexual dysfunction (anorgasmia and arousal problems). SEMG studies point to progressive deterioration of the pelvic neuromuscular function in women with urinary stress incontinence (Gunnarson & Mattiasson 1999). Pelvic floor assessments consistently revealed lower levels of SEMG values for resting baselines and rapid and tonic contractions in incontinent women. Women undergoing pelvic floor muscle retraining were seven times more likely to be cured and 23 times more likely to show improvement in symptoms of urinary incontinence as a result of muscle strengthening (Hey-Smith et al., 2001). Pelvic muscle retraining assisted by SEMG was recommended as first line therapy (AHCPR, 1996).

  In relation to orgasmic dysfunction, since the early work of Arnold Kegel, deficiency in the pubococcygeal muscle was seen as a factor in 40% of anorgasmic women (Kegel, 1956). The prevalence of orgasmic disorders significantly increases in women with stress urinary incontinence, with 86% of women reporting sexual dysfunction and 60% specifically reporting anorgasmia (Amareco et al., 1996). These findings highlight the potential role of muscle weakness as a mechanism in both disorders.

- SEMG assisted retraining of pelvic muscles. In the case of both hyper- and hypo-tonus muscles, management of the associated disorders should focus on the normalization of pelvic muscle function. In the case of hyper-tonus muscles, normalization of muscle function requires down-training of over-active muscles, improving tone and coordination of muscle recruitment, restoring muscle extensibility, and elimination of myofascial trigger points.

This comprehensive review of research in relation to SEMG applications in the management of female pelvic floor disorders is intended for clinicians who wish to develop protocols for the

treatment of muscle mediated pelvic floor dysfunctions. It is a comprehensive and original compilation and sets out practical guidelines for therapists focussing on pelvic floor disorders. It will especially assist those who may wish to incorporate SEMG assessments into their clinical work.

The paper concludes with a discussion of training requirements and competence for clinical work with pelvic floor disorders.

Conclusion and recommendation

Vulvodynia is a more prevalent disorder then previously realized (Harlow & Stewart, 2003). As a chronic pain condition it compromises quality of life, impacts on social relationships, sexual behaviour and emotional wellbeing (Ponte et al., 2009; Arnold et al., 2006; Jantos & Burns, 2007). The disorder affects women of all ages, but the age related risk is highest in women under the age of 24 years (Jantos & Burns 2007; Harlow & Stewart 2003). Due to a lack of general awareness, vulvodynia is under-diagnosed and often misdiagnosed. As a result, women suffer disabling chronic pain for periods of time ranging from months, to years, and in some cases, a lifetime. On average the reported delays in diagnosis can average 4-7 years (Jantos & Burns 2007; Jantos & White 1997).

Vulvodynia is a localised pain condition, reproducible during medical exams, consistently described by patients as a burning or stabbing pain, and rated by patients in the severe range of 7.0 to 8.5 on an eleven point pain scale (Bergeron et al., 2001; Jantos & White, 1997; White & Jantos, 1998; Jantos, 2008). The disabling impact of the pain often limits an individual's ability to maintain regular day-to-day activities (Ponte et al., 2009). Sexual activity can be severely affected, with many women abstaining from intercourse and reporting a marked reduction in sexual desire, frequency, pleasure and satisfaction (Jantos & Burns, 2007; Jantos, 2008).

The disabling impact of vulvar pain leads to significant psychological distress, as reflected in increased levels of anxiety, depression and a sense of helplessness (Binik et al., 1999; Jantos & Burns, 2007). The high prevalence of depression and anxiety in vulvodynia and other chronic pain conditions has been well documented (Masheb et al., 2005; Sullivan et al., 1992). In the case of vulvodynia the prevalence of depression prior to onset of symptoms is no higher than other control

groups (Reed et al., 2000; Meana et al., 1997). Without pathology to explain the severity of symptoms, some elect to classify vulvodynia as a sexual dysfunction (Basson, 2005; APA, 2000), others perceive it as a somatoform disorder (Dobson & Friedrich, 1978; Lynch, 2008). Evidence from research presented in this thesis, and from the medical literature in general, does not support either of these two classifications.

Based on the research evidence available to date, the most appropriate classification system relevant to vulvodynia is that of a chronic pain syndrome. The absence of visible pathology in any chronic pain syndrome should not be the basis for either seeking psychological explanations, or questioning the reality of the patient's pain (Jantos, 2008). The predicament that confronts pain patients is the common myth and misconception subscribed to by health care professionals that all pain conditions must have some form of identifiable physical cause (Hawthorn & Redmond, 1998). This is clearly not the case in most chronic pain conditions (Steege, 1998).

Reasons for rejecting the classification of vulvodynia as a sexual dysfunction

From the findings reported in this thesis, and on the basis of other research literature, there is no justification for classifying vulvodynia as a sexual dysfunction. Vulvodynia should not be classified as a sexual dysfunction for the following reasons:

- The DSM text defines sexual dysfunction as a disruption of the sexual response cycle. The four phases of the sexual response include desire, excitement, orgasm and resolution. Localized vulvar pain which interferes with sexual function does not constitute a disruption of the sexual response cycle (First, 2005). The majority of women with vulvodynia experience difficulties with intercourse and would have to be considered as "non-starters," but only on account of pain, not on account of failure in sexual response. Women with vulvodynia are just as responsive to sexual stimuli as women who experience no pain (Payne, 2007).

- The DSM includes *Sexual Pain Disorders* in its classification of sexual dysfunctions. This subclassification assumes that sexual pain arises predominantly on account of

psychological factors, or a combination of psychological and medical factors. There is no evidence to support such an assumption in relation to vulvodynia. Vulvar pain is a localized pain problem with symptoms reproducible during medical examination. In the case of chronic vulvar pain, it is illogical to define pain as the symptom and the cause (Moser, 2005).

- Psychosexual conflict is assumed to be the primary cause of sexual dysfunction. Research provides no evidence linking psychosexual conflict to symptoms of vulvodynia. On the contrary, the majority of women who experienced chronic vulvar pain symptoms, rated their relationships positively, regarding their partners as supportive. Relationships enabled them to cope better with chronic pain (Jantos & Burns, 2007). Marriage relationships, seen in other studies as the source of stress and cause of pain (Schrover et al., 1992; Lynch, 2008), proved to be an effective buffer against psychological distress, depression and anxiety. There are no reports showing relationships to be causal or contributing factors to vulvar pain.

- Where sexual dysfunction may be due to sexual incompetence, there is no evidence that vulvodynia patients are inept in terms of sexual skill. There is no evidence that vulvodynia women are different to other women in terms of sexual frequency, responsiveness, values and interests (Reed et al., 2000; Meana et al., 1997). Vulvodynia women show the same level of sexual arousal as asymptomatic women, but sensitivity to touch and pressure precludes them from sexual intercourse (Payne et al., 2007).

- Abstinence or reduced frequency of sexual intercourse is directly attributable to the severity of pain. From the psychosexual profile study, it is evident that prior to the onset of symptoms, the frequency of sexual activity among vulvodynia women was six times higher than in the symptomatic state (Jantos & Burns, 2007). Not only was there a significant reduction of sexual frequency between the asymptomatic and symptomatic state, but evidence shows that partners of patients also initiated fewer sexual activities out of fear that such activities would provoke pain.

- Where loss of sexual desire may be seen as evidence for classification of vulvodynia as a sexual dysfunction, the reported loss of desire by 82% of the patients was related to the onset of pain. Of the total study cohort, 87% of women expressed a desire to increase the frequency of intercourse once symptoms abate (Jantos & Burns, 2007).

- Chronic vulvar pain is asexual in nature. Evidence that vulvodynia discomfort is commonly triggered and exacerbated by non-sexual activities, such as walking, sitting, bike riding and wearing of tight clothing (Sandownik, 2000), confirms that chronic vulvar pain is not a "sexual pain" and should not be confused with sexual dysfunction.

- Findings that vulvodynia symptoms are often secondary to infections, irritants, and iatrogenic trauma, reinforces the suggestion that pain does not arise from sexual causes (Haefner et al., 2005; Jantos & White, 1997).

- Biopsies and physiological studies show increased density of nerve endings, a genetic pro-inflammatory predisposition, and increased immunoreactivity which may predispose some women to vulvodynia (Bohm-Stark et al., 1998, 1999, 2001; Gerber et al., 2002). These findings point to important variables that are not psychological in nature and take the disorder out of the sexual dysfunction classification.

- The often early onset of vulvodynia (primary vulvodynia), where pain and discomfort is reported in childhood, teen years and early adulthood, prior to the commencement of sexual activities, confirms that onset of vulvodynia symptoms bears no relationship to sexual activity and should therefore not be classified as a sexual dysfunction. Likewise, the late onset, especially during the postmenopausal years, is not likely to be due to psychosexual issues but due to hormonally induced physiological changes (hypoestrogenism and vaginal atrophy) and other non-hormonal factors, not of psychiatric nature (Kao et al., 2008).

To classify vulvodynia as a sexual dysfunction is to confuse the dysfunctional state casued by pain, with the unknown but potentially multifactorial etiology of pain. In general the DSM is considered an inappropriate classification system, based on a grouping of disorders that lacks

validity and objectivity, using a problematic conceptualization of women's sexual response (Meeson & Bradford, 2007). The implications of an incorrect classification and incorrect conceptualization of vulvodynia limit the implied treatment options.

The view of vulvodynia as sexual dysfunction is not supported by evidence from current research studies and the research literature generally.

Reasons for rejecting the classification of vulvodynia as a somatoform disorder

From the findings reported in this thesis, and on the basis of available research literature, there is no justification for classifying vulvodynia as a somatoform disorder. Vulvodynia should not be classified as a somatoform disorder for the following reasons:

- The diagnosis of somatoform disorders is made where psychological factors are judged to have an important role in its onset, severity and maintenance of symptoms. The justification for grouping somatoform disorders into one category is based on a lack of medical explanation for the existence of symptoms. Somatoform disorders include what was historically referred to as hysteria, and today include conversion disorder, hypochondriasis, pain disorder and body dysmorphic disorders. From all of the research reviewed, there is no evidence that psychological factors play a primary role in the etiology of vulvodynia.

- Vulvodynia is seen by some as a form of somatization disorder (a subgroup of somatoform disorders which represent the expression of personal and social distress throught the development of physical symptoms). The criteria for somatization disorder require that the onset of symptoms must occur prior to 30 years of age and last for a period of several years. Vulvodynia affects women of all ages and the onset of symptoms can occur as early as childhood (primary vulvodynia) or postmenopause (secondary vulvodynia). The onset of symptoms in the psychosexual profile study ranged from 5.5 - 45.2 years of age. It is highly unlikely that somatoform disorder would manifest itself at

such an early age or that onset during menopausal years would qualify under the diagnostic criteria.

- Somatization disorder has a high hereditary component. It is observed in 10-20% of female first-degree biological relatives of women suffering from the disorder. Relatives of women with somatization disorder also show an increased risk of antisocial personality disorder and substance related disorders. Personality disorders are commonly associated with somatization disorder. Individuals suffering from the disorder are unable to provide consistent or factual account of their symptoms, rather they describe them in colourful and highly exaggerated terms. In the case of vulvodynia, there is no evidence of such familial hereditary tendencies, nor of other personality disorders (Meana et al., 1996). Patients provide highly consistent accounts of symptoms (Jantos, 2008; Bergeron et al., 2001) and symptoms can be consistently reproduced during medical exams providing for a highly reliable diagnosis (Bergeron et al., 2001; Masheb et al., 2004).

- The estimated prevalence of somatization disorder among women is very low, ranging from 0.2 - 2.0% (APA, 2000), whereas the life-time prevalence of vulvodynia has been reported at 4 - 34% (Harlow & Stewart, 2003; Berglund et al., 2002; Adanu et al., 2005). The prevalence of vulvodynia is 10 - 100 times higher than the rate of somatization disorder and more in keeping with the prevalence of other chronic pain conditions, such as lower back pain or tension headache pain.

- Elevated levels of depression and anxiety do not equate with the common occurance of major depressive disorder or panic disoder seen in somatization patients. The presence of depressive and anxiety-like symptomatology in vulvodynia is more in keeping with chronic pain (Sullivan et al., 1992; Meana et al., 1997).

- Where somatization disorder requires a history of pain related to at least four different body regions or functions, this is not the case with vulvodynia. Due to the fact that vulvar pain affects the urogenital area, pain during sexual intercourse should not be the

primary symptom of diagnosis. Other comorbidities such as bladder, perineal, rectal

pain, or even abdominal and lower back pain, arise on account of common physiological

mechanisms (i.e. neuromuscular system) and should not be seen as separate from the

disorders. No other co-existing pain regions have been identified in vulvodynia that

would satisfy the diagnostic criteria for somatization disoder.

- Because there must be a history of at least one sexual or reproductive symptom other

  than pain, in vulvodynia the symptom of pain is its primary characteristic with no other

  known reproductive symptoms being reported.

- Where there must be at least one pseudoneurological symptom akin to a conversion

  like symptom (e.g., paralysis, hallucinations, blindness, deafness, seizures, amnesia or

  loss of consciousness), there is no evidence of such symptoms in vulvodynia. The

  increased prevalence of depression and anxiety does not qualify as a pseudoneurological

  phenomenon. The level of psychopathology among patients is no higher than among

  control groups of women (Reed et al., 2000; Meana et al., 1997).

- The symptoms must not be intentionally produced or feigned. There is no evidence of

  feigning or malingering among women with vulvodynia. Symptoms of vulvodynia are

  localized and described in a consistent, highly reliable manner and reproducible during

  medical examinations. The same cannot be said for symptoms of somatoform disorder,

  which seems to vary, reoccurring and affecting multiple sites.

- Psychiatric literature confirms that chronic pain conditions do not fit well into the

  somatoform classification (Blackwell, 1989). This applies to the case of vulvodynia.

Since women with vulvodynia show no more psychopathology than control groups, and

there is no other evidence of psychiatric symptoms, a different classification system is required. If

the absence of pathology is the primary ground for classifying vulvar pain as a psychiatric disorder

then all other chronic pain conditions without visible pathology should also be re-classified.

Viewing vulvodynia as a psychiatric disorder or a sexual dysfunction has contributed to much

confusion and hindered its management (Binik, 2005). Patients have been frequently referred for

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psychiatric assessment on the assumption that the etiology of vulvodynia is linked to psychological

tensions, marital disharmony, past abuse, or repressed emotions. Occasionally, they are referred to

sex therapists, where the primary focus has been on improving sexual technique, assuming that

ineptness and lack of compatibility may be a cause of symptoms. Otherwise they are referred to

psychologists who have focussed on pain coping strategies. In all, the primary problem of pain has

been overlooked (Pukall et al., 2003; Binik et al., 1999). This has contributed to the patient's and

referrer's disappointment, leaving the patient with little, if any, pain relief.

Key recommendations

In view of the findings in the studies reported in this thesis, and based on evidence available

in the research literature, the following recommendations are proposed:

Recommendation # 1: *Classify chronic vulvar pain as a regional pain syndrome according to the*

IASP Multiaxial System.

Because the diagnosis of vulvodynia relies exclusively on the women's self-report of pain,

which is localised, reproducible by pressure application to the hyperalgesic area, accounted for using

the same temporal characteristics and triggered by a range of day-to-day activities inclusive of

sexual intercourse but not limited to sexual activity, vulvodynia should be viewed as a regional pain

disorder.

Pain is the primary symptom of the disorder. The symptom of pain and the associated

mechanisms involved should be the primary focus of management. Using the terminology of the

ISSVD, the taxonomy that most precisely accommodates vulvar pain is the multi-axial system for

classification of chronic pain, developed by the IASP (Mersky & Bogduk, 1994). There is no other

classification system that provides a more specific focus on the core symptom which motivates

women to seek help. The reclassification of chronic vulvar pain from a sexual dysfunction or a

somatization disorder does not resolve the pain (Carpenter et al., 2005), but provides a more

rational and informed approach to therapy. According to the IASP classification, the pain is coded on

the basis of region, physiological system, temporal characteristics and patterns of occurrence, and intensity and etiology (where etiology is known).

According to this classification, a hypothetical classification of vulvodynia would be made on the basis of the localized nature of the pain affecting the vulvar region and could be presented as follows:

Regional Vulvar Pain Syndrome

Definition: Vulvodynia is defined as vulvar discomfort, most often described as burning pain, occurring in the absence of relevant visible findings or a specific, clinically identifiable, neurologic disorder.

Site: The vulvar region which consists of the mons pubis, the labia majora and minora, the vestibule of the vagina, the hymen, the greater vestibular glands of Bartholin, the clitoris, the bulbs of the vestibule and the external urethral orifice.

System: The neuromuscular system.

Main features:

Prevalence rates: A lifetime prevalence of 15% is generally acknowledged.

Age of onset: Onset can occur at any age. An early onset can occur in pre-adolescent children and late onset in pre- and post- menopausal women is common. The highest risk of onset is prior to 25 years of age.

Pain quality: Discomfort is most often described as burning, sharp stabbing pain, especially when provoked by pressure application to the area.

Time pattern: Chronic pain with episodes lasting for days, weeks or months, with either a continuous or intermittent pattern.

Intensity: Moderate to very severe, interferes with daily activities, and in particular with sexual intercourse.

Usual duration: Chronic pain varying in duration from months to years, or a lifetime. Rarely subsides spontaneously. Often associated with delays in diagnosis and commonly misdiagnosed. Responsive to therapy.

Associated symptoms: Common comorbidities include bladder pain, urgency and frequency, irritable bowel symptoms, fibromyalgia, severe pre-menstrual symptoms, skeletal muscle tenderness in lower abdominal, gluteus and upper thigh muscles, psychological distress, expressed in depression and anxiety.

Signs and laboratory findings: Hyperesthesia to touch, with notable tenderness elicited using a cotton-tipped applicator just distal to the hymenal ring but proximal to Hart's line. Tenderness can be elicited by digital pressure over the lateral walls of the vagina at the level of the pubococcygeus muscle and over the base of the bladder.

Usual course: Chronic discomfort, may last for years. Some cases show spontaneous remission in symptoms, but not necessarily in pain mechanism. Due to personal embarrassment, some women may be reluctant to seek medical assistance, until severity of pain begins to prevent attendance at work or engagement in sexual intercourse. Responsive to conservative therapy, in some instances may require surgery.

Complications: None. However, patients may have co-existing fungal or bacterial infections, or dermatological problems requiring medical management.

Social and physical disability: Interferes with daily activities, sleep and work. Impacts significantly on sexual relationships.

Pathology: None known. Biopsies may reveal presence of inflammatory cells, but this is not significant diagnostically.

Summary of essential features and diagnostic criteria: Vulvar discomfort described as burning, rawness, stabbing pain, spontaneous or evoked by pressure application.

Differential diagnosis: May be confused with chronic candidiasis. With persistent symptoms and paucity of clinical features and lack of demonstrable pathology, psychogenic etiology has been considered. Current evidence does not support the concept of the pain being of psychological origin.

The IASP classification, through its axial system provides a consistent means of classifying pain on the basis of the location and temporal characteristics of pain. As a recognised and validated

pain classification system it is ideally suited for clinical and research use in relation to chronic vulvar pain.

Recommendation # 2: *Integrate the ISSVD Terminology with the IASP Multiaxial Pain Classification System*.

A workable approach, based on findings and current understanding of chronic vulvar pain, would be to adopt the current terminology of the ISSVD (Moyal-Barracco & Lynch 2003), and encourage its further development by integrating it into the multiaxial pain classification of the IASP. By adopting the ISSVD terminology, the accepted term of *vulvodynia* should be maintained and promoted through educational campaigns and training curricula. The integration of the ISSVD terminology with the IASP multiaxial classification system would encourage a more consistent and systematic study of lower urogenital tract pain. It would encourage a more detailed mapping of pain, provide focus on the mediating physiological systems involved and encourage further study of pain characteristics and patterns of occurrence.

The IASP approach to chronic pain acknowledges the psychophysiological nature of pain, essential to understanding the experience of chronic pain. The integration of the two systems would overcome the shortcomings of the current ISSVD classification, and would provide a superior classification to the current DSM classification, which lacks validity, and places excessive emphasis on psychogenic factors.

Recommendation # 3: *Classify chronic vulvar pain (vulvodynia) on the basis of known underlying mechanisms, not the superficial symptoms reported.*

Substantial evidence exists implicating the neuromuscular system in the etiology and symptom maintenance in vulvodynia. Paradoxically, the label "idiopathic" vulvar pain may have served its purpose and outlived its usefulness. Though the science of mechanisms has not fully evolved, there is no reason for ignoring existing knowledge.

The treatment of all medical disorders should be directed at the etiology and pathophysiology involved (Bachmann et al., 2006). Although the current understanding of the

pathologic processes involved in vulvodynia may not be well advanced or complete, it should be reflected in treatment and management guidelines. Often there is a tendency to adopt the so-called "shot gun" approach to management of medical conditions; however, such an approach does not foster confidence nor convey reliability, or validity, in educational forums. In chronic pain disorders of a non- malignant nature, specific physiological systems should form a part of the classification structure, and direct treatment decisions.

Recommendation # 4: Adopt a psychophysiological view of chronic vulvar pain, discarding the sexualized and psychologised views of vulvodynia.

Chronic pain should always be viewed as a sensory and emotional experience. There is no valid evidence linking vulvodynia to sexual dysfunction or to support views of a psychogenic etiology of chronic vulvar pain. Chronic pain is a mechanism by which the body alerts the patient and the doctor to the existence of an underlying problem. Pain is associated with various degrees of physiological dysregulation and increased rigidity, and these interact with emotional states and psychological traits of the individual. Recognising that the experience of pain is modulated by the interaction of cognitions, emotions and physiology is essential. Pain research highlights the fact that psychological factors have a major bearing on the experience of pain and act as powerful modulators of pain thresholds and of pain perception. On this account, pain must be studied from a psychophysiological perspective, but not necessarily from a *biopsychosocial* approach. For some, the biopsychosocial approach is synonymous with a "serial undisciplinary" approach consisting of ill defined and mixed interventions (Townsend, 2005).

Sexualising vulvar pain adds a stigma to the pain and may ultimately impede appropriate intervention (Kleinplatz, 2005). There are not two types of pain in the body, one sexual and the other non-sexual. Sexualising the problem may cause it to be trivialised and dismissed on account of inexperience and ineptness. To confuse the anatomical area affected with its functional role is not only detrimental to the management of pain but unacceptable from the perspective of classification.

The past history of sexualizing and psychologising vulvar pain has proved to be counterproductive, conceptually and clinically. There is no evidence linking the condition to sexual

dysfunction or to support the hypothesis that vulvodynia arises against a background of psychological, sexual, social or physical distress. To suggest that women with vulvar pain are seeking to avoid intercourse through psychological and psychosomatic complaints is not a valid and reliable reason for classification and should not be permitted to guide diagnosis and treatment (Townsend, 2005).

Recommendation # 5: *Adopt a multidisciplinary approach to the assessment and management of vulvodynia.*

The studies reported in this thesis highlight the need for a multidisciplinary approach to the assessment and management of vulvodynia. Both human sexuality and chronic pain are very complex phenomena. This requires an integrated approach which seeks to address both issues in a balanced manner and avoids traditional dualisms. Assessment needs to focus on the primary symptom of pain and the mechanisms maintaining it, but attention also needs to be directed to the impact of pain on the emotional, sexual and social wellbeing of the patient.

In the majority of cases, pain management needs to be integrated with sexual counselling and/or traditional sex therapy. In some patients, the resolution of pain does not automatically guarantee sexual functionality and wellness. A proportion of patients (approx. 11-17%) who successfully completed therapy, remained sexually abstinent. This may indicate that for some women the pain has few sexually related implications, or, it highlights the need to provide patients with additional psychotherapeutic support, enabling them to attain a fully functional state.

As vulvodynia is known to have significant impact on relationships, social needs must also be addressed. It is not uncommon for young women to seek support and guidance on how to address and discuss their pain disorder with their partner or other care-givers involved. This requires tactful guidance. In cases of existing relationships, enabling women to progress from a position of fear of pain, and avoidance of sexual activity, to a point of increased desire and re-engagement in sexually intimate involvement, calls for special qualification and training. Where the pain disorder has caused disharmony and separation in marital relationships, experienced marriage counselling would be

recommended. Yet, on account of the very personal and private nature of the disorder, it is not recommended nor expected that women engage numerous professionals through a "revolving door" approach, visiting multiple carers. Highly specialised teams are needed where all members of the team have a good appreciation of the disorder; are knowledgeable of the anatomical issues in lower tract urogenital pain, as well as their individualised specialty and can communicate across various professional domains.

Recommendation # 6: Recognise the need to provide individualised therapy.

The etiology of vulvar pain is multifactorial. Each individual will present with a unique medical and personal history. Therapy needs to centre not only on the pain, or the specific activities with which it interferes, but on the individual who is seeking assistance.

Each patient's priorities may be different and will require an individualized case plan. In the development of case plans, women must be given the option of having input into indentifying treatment priorities and preferences (Keefe et al., 2004). Some women seek assistance for relief of pain which interferes with day to day activities, others request help because the pain is destroying their sex life. In the first case the emphasis will be predominantly on resolving pain, but in the latter, pain needs to be addressed in the context of sexual function. Sexuality is highly valued in society, and some women feel defective and sexually inadequate on account of vulvodynia (Payne, 2005). Patients receiving tailored treatment report significantly lower levels of fatigue, depression, and helplessness and higher levels of active coping, social support and compliance (Kleinplatz, 2005; Binik et al., 1999).

From a psychological perspective, unlike headaches and other forms of pain, vulvar pain affects the bodily region said to define womanhood, femininity, and sexuality (Kaler, 2005). Vulvar pain, especially on account of its often early onset, has developmental implications in relations to body image, self-concept and self-esteem. The early onset of pain has been shown to have impact on formation of relationships and the postponement of intimate sexual behaviour. These issues need to be further researched but must be taken into consideration throughout therapy. The

potential impact of early onset may be detrimental and long-lasting and needs to be borne in mind, especially with primary vulvodynia cases. Other issues may be identified by the patient and will need to be addressed.

**Recommendation # 7:** *Advocate the use of conservative therapies as front line interventions.*

From the perspective of the physiological systems involved, dysfunctional pelvic muscles have consistently been shown to be associated with chronic vulvar pain. The normalization of pelvic muscle function, with the assistance of SEMG biofeedback, and application of myofascial release, has been shown to be a highly effective, conservative treatment modality. Muscle overactivation is an inefficient peripheral response to irritants, inflammation and trauma. It can occur and be exacerbated by negative emotional states. With time, muscle overactivation can give rise to ischemia and myofascial changes, resulting in muscles becoming the primary source of pain. Muscle dysfunction is known to cause peripheral sensitisation (Schattschneider et al., 2006; Graven-Nielsen et al., 2003). Peripheral sensitisation in vulvodynia is responsive to conservative therapy (Jantos, 2008; Reissing et al., 2005). The down-training of hypertonic muscles assists in the development of improved muscle function and is associated with a reduction in symptoms of vulvodynia and other common comorbidities. Implicating central sensitization processes (Lynch, 2008) in chronic vulvar pain is not warranted.

Conservative therapies are often used as a last resort, but their usefulness and effectiveness should be better recognised in chronic vulvar pain management. Conservative therapies should constitute first line interventions, followed by progressively more invasive and radical treatments.

Summary

In summary, from the evidence available to date, the etiology of vulvodynia appears to be multifactorial. Research has identified certain predisposing psychophysiological traits, as well as common triggers that may act as antecedents to vulvar pain. The combined effect of the predispositions and triggers, though often unknown, is the overactivation of pelvic muscles,

progressive autonomic dysregulation, physiological rigidity and progressive peripheral sensitization. Though the physiological mechanisms are not fully understood, evidence shows that normalization of pelvic muscle function, together with therapy addressing emotional, social and sexual issues, can reverse the etiological trend from that of a symptomatic state, to an asymptomatic state with renewed functionality and quality of life. This view, based on the findings reported in this thesis and in published literature, can be best summarised diagrammatically as shown in Figure 1.
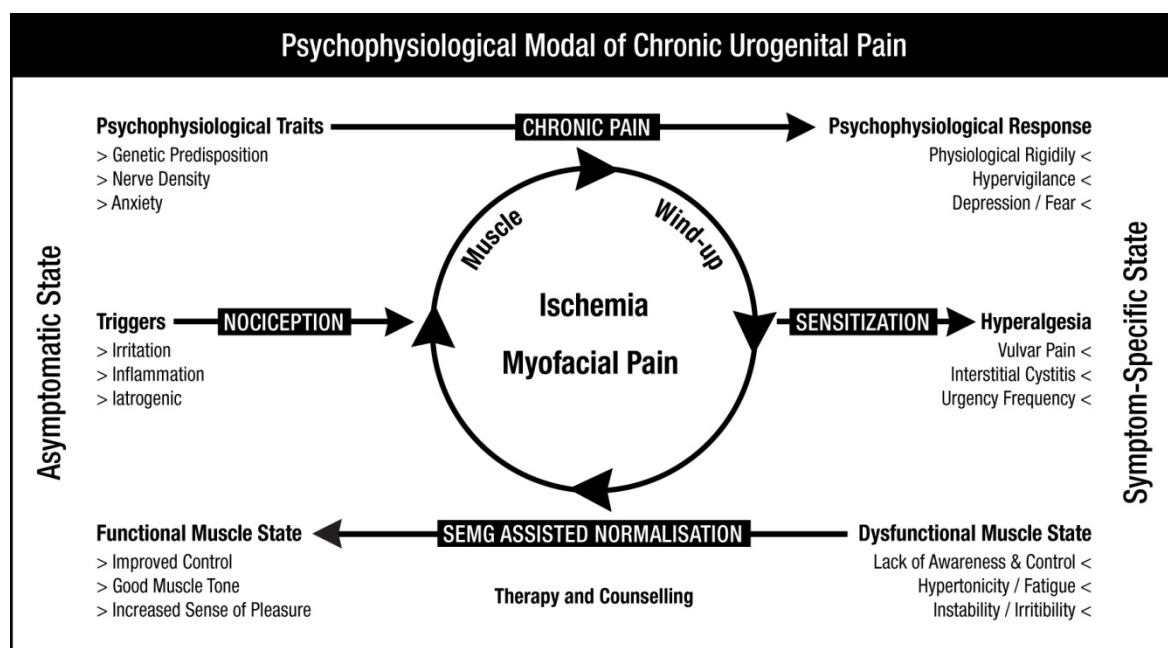
Figure 1 A Psychophysiological Model of Chronic Urogenital Pain.

To facilitate greater understanding of vulvodynia, it is essential to "de-sexualize" the condition and manage it as a regional chronic pain disorder. By doing so, the symptom of pain becomes the focal issue of assessment and management, and the impact of pain on wellbeing and quality of life can be addressed effectively.

The research findings of this thesis and of the literature reviewed provide evidence that has theoretical and applied implications for the understanding and management of vulvodynia. With competent professional intervention, vulvodynia patients can resume normal daily activities, regain normal sexual function and experience a sense of wellbeing and enjoy good quality of life.

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