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Partial vaginismus - definition, symptoms and treatment

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Cover painting by a young woman with partial vaginismus and vulvar vestibulitis illustrating her vulvar pain (red) and her associated anxiety (blue).

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*They say it is all in my head,
but still it hurts.*

To Anna, Linnea and Hilda

ABSTRACT

Background: Vaginismus is a sexual pain disorder, where spasm of musculature of the outer third of the vagina interferes with intercourse. Vaginismus exists in two forms: total vaginismus, where intercourse is impossible, and the more seldom described partial vaginismus, in which intercourse is possible but painful.

Aim: The aim of the thesis was to develop a useful definition of partial vaginismus for both clinical and scientific purposes; to describe the prevalence of partial vaginismus among women with superficial coital pain; to report on symptoms and clinical findings in women with partial vaginismus; and to present treatment results for women with vaginismus.

Methods and findings: In a clinical sample of 224 women with superficial coital pain, we found a great overlap of the clinical diagnoses of partial vaginismus (PaV) and vulvar vestibulitis (VVS) (nowadays called provoked vestibulodynia); 102 women had both PaV and VVS. All women with VVS had vaginismus. Partial vaginismus was more common in all our samples than total vaginismus.

sEMG of pelvic floor muscles was found to be of no value in distinguishing women with partial vaginismus with or without vulvar vestibulitis (PaV+/-VVS) (n=47) from each other or from an asymptomatic group (n=27).

Women with PaV+/-VVS (n=53) reported not only burning pain but also itch during a standardized penetration situation (sEMG of pelvic floor muscles), while asymptomatic women (n=27) did not. In most cases, the appearance of burning pain preceded the appearance of itch.

In a retrospective interview study, 24 women with PaV+/-VVS reported pain after intercourse more often than pain during penetration at the onset of the problem. When the women ceased having intercourse, both symptoms were equally common. Intensity of pain during penetration increased dramatically from very low at onset of the problem to very high when the women ceased having intercourse, while intensity of pain after intercourse was already high at onset of the problem and increased to very high when the women ceased having intercourse.

Pain after intercourse in women with PaV+/-VVS was described as burning and/or smarting and lasted in mean for two hours, while pain during penetration was described with words like sharp/incisive/bursting and lasted for one minute.

At long-term follow-up (more than three years) of a group of women treated with cognitive behaviour therapy for vaginismus (n=59, response rate 44/59 on a questionnaire), a majority were able to have and enjoy intercourse. The proportion of women with positive treatment outcome was, however, associated to the definition of treatment outcome. An ability to have intercourse at end of therapy was maintained at follow-up. Every tenth women with vaginismus healed spontaneously after thorough assessment.

Conclusion: Partial vaginismus was more common in our studies than total vaginismus, and all women with vulvar vestibulitis had partial vaginismus. Women with PaV+/-VVS reported not only burning pain during standardized penetration but also itch. When the problem started in women with PaV+/-VVS, pain after intercourse was more common than pain during penetration. Pain after intercourse was described as longlasting and burning and/or smarting, while pain during penetration was described as short and sharp/incisive/bursting. Long-term follow-up results of a series of women treated with CBT for vaginismus show good treatment outcome.

LIST OF ORIGINAL PAPERS

I Wijma, B., Jansson, M., Nilsson, S., Hallbook, O. and Wijma, K., Vulvar vestibulitis syndrome and vaginismus. A case report, *J Reprod Med*, 45 (2000) 219-23.

II Engman, M., Lindehammar, H. and Wijma, B., Surface electromyography diagnostics in women with partial vaginismus with or without vulvar vestibulitis and in asymptomatic women, *J Psychosom Obstet Gynaecol*, 25 (2004) 281-94.

III Engman, M., Wijma, K., and Wijma, B., Itch and burning pain in women with partial vaginismus with or without vulvar vestibulitis, *J Sex Marital Ther*, 33 (2007) 171-186.

IV Engman, M., Wijma, K., and Wijma, B., Postcoital burning pain and pain at micturition: early symptoms in women with partial vaginismus with or without vulvar vestibulitis?, Accepted *J Sex Marital Ther*.

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ABBREVIATIONS

BW	Barbro Wijma
CBT	Cognitive behaviour therapy
DSM-IV	Diagnostic and Statistical Manual of Mental Disorders, 4 th ed.
DSM-IV-TR	Diagnostic and Statistical Manual of Mental Disorders, 4 th ed., text revision
ICD-10	International Statistical Classification of Diseases and Related Health Problems, 10 th revision
ISSVD	International Society for the Study of Vulvovaginal Disease
ME	Maria Engman
OR	odds ratio
P25	25 th percentile
P75	75 th percentile
PaPV	partial primary vaginismus
PaSV	partial secondary vaginismus
PaV	partial vaginismus
PV	primary vaginismus
sEMG	surface electromyography
TPV	total primary vaginismus
TSV	total secondary vaginismus
TV	total vaginismus
WL	waiting list
VVS	vulvar vestibulitis (syndrome)

FOREWORD

The work with this thesis started nearly ten years ago when I had the opportunity to be taught by Barbro Wijma one day/week for one year in the investigation and treatment of women with vaginismus. There was a great advantage in meeting the patients together during the clinical work; accepted truths became questioned and alternative explanatory models were discussed. We found a great deal of clinical overlap between the diagnoses vaginismus and vulvar vestibulitis. During this time the case report (Study I) was written. At first, we tried to use vaginal manometry to measure the vaginistic reflex and the increased tone in the pelvic floor muscles, but we found this method to be of little value and charged with technical problems. At this time, one study reported that a diagnosis of vulvar vestibulitis could be confirmed by sEMG measurements of the pelvic floor muscles, and that women with vulvar vestibulitis could be successfully treated with sEMG biofeedback. We conducted an sEMG study of pelvic floor muscles in women with vaginismus with or without vulvar vestibulitis and asymptomatic women in which we expected to find a difference between groups, but we did not. We also started a randomized treatment study of women with vaginismus with or without vulvar vestibulitis. The original design was a randomization to CBT, CBT + sEMG biofeedback or to waiting list and subsequent treatment. The purpose of this design was to see if an addition of sEMG-biofeedback to CBT could reduce the number of treatment sessions needed. The treatments were more time-consuming than expected. The difference in time on the waiting list between the active treatment group and the waiting list group became insignificant, and the waiting list arm had to be omitted. Finally, the randomized treatment study was disrupted due to economic problems of the clinic. The final design for the treatment study was a long-term follow-up study of a series of cases with questionnaires sent to all women with vaginismus who had been treated with CBT according to the pre-formulated study protocol.

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INTRODUCTION

The subject of this thesis is partial vaginismus. Vaginismus is a sexual pain disorder (American Psychiatric Association 2000) where spasm of the musculature of the outer third of the vagina interferes with sexual intercourse.

Historical background

Vaginismus is probably a very old problem. Painful coitus has been described as far back as in ancient Egypt in the Ramesseum Papyri Scrolls (Costa Talens and Colorado Vicente 1971). The first known written description of vaginismus is nearly a thousand years old. Trotula of Salerno described in “The Diseases of Women” “a tightening of the vulva so that even a woman who has been seduced may appear a virgin” (Trotula 1547/1940). The term vaginismus was introduced by the gynaecologist J. Marion Sims in 1862 (Sims 1862). He described a reflex-like contraction of the circumvaginal musculature upon attempts at vaginal penetration, and called this vaginismus. According to Sims, vaginismus was easily treated by surgical division of the muscles of the vaginal opening, followed by a program of exercises with glass dilators*. The surgical intervention soon became disputed, and e.g. Scanzoni and Thorburn suggested that it should be replaced by dilatation under anaesthesia (Scanzoni 1867; Thorburn 1885). Yet it was still the standard treatment in the early decades of the 20th century (Drenth et al. 1996). Nearly a hundred years ago, Waltherhard (Waltherhard 1909) presented theories of the vaginal muscle spasm being a phobic reaction resulting from the woman’s fear of pain, and recommended psychotherapy and education to solve the problem. From the 1940s to 1970s vaginismus was often described by psychoanalytic theories, and defined as a hysterical or conversion symptom (Fenichel 1945; Musaph 1965). Suggested therapeutic approaches, if any, were classical psychotherapy, hypnosis and tranquilizers (van de Wiel 1990). Masters and Johnson (1970) described vaginismus as a classical example of a psychosomatic illness, and claimed excellent success rates with behaviour-oriented sex therapy.

* Sims’ glass vaginal dilators are still (2007) available and can be bought on the Internet.

Aspects on reviewing literature concerning vaginismus

As the knowledge of both vaginismus and dyspaeunia/vulvar vestibulitis has greatly increased recently, I have, wherever appropriate, divided the review into: 1) what was known before we started our studies; and 2) what has been added during the course of the work with this thesis.

A major problem when reviewing the literature of vaginismus is that the definitions of vaginismus are often insufficiently described as to primary (i.e. the woman has never been able to have intercourse) or secondary (i.e. the woman has been able to have intercourse earlier), total or partial (detailed definition on page 2), and generalised or situational vaginismus. Most authors seem to refer to total, primary vaginismus when using the term vaginismus, i.e., the woman has never been able to have intercourse. Partial forms, in which penetration is possible but painful, are seldom discussed (Lamont 1978; Steege 1984; Caplan 1988; Kessler 1988; Wijma and Wijma 1997; Jeng et al. 2006). This review will include literature on vaginismus in general. Other authors’ definitions of vaginismus are, wherever possible, clarified and defined according to our classification: total primary vaginismus (TPV), total secondary vaginismus (TSV), partial primary vaginismus (PaPV), and partial secondary vaginismus (PaSV)

Even before our studies started, some reports discussed a relationship between dyspareunia/vulvar vestibulitis and vaginismus (Spano and Lamont 1975; Lamont 1978; Steege 1984; Shortle and Jewelewicz 1986; Schover et al. 1992; Monif and Belatti 1993; Abramov 1994; Basson 1995; van Lankveld et al 1994; Goetsch 1996; Hassel 1997; Nunns and Mandal 1997; White and Jantos 1998), and recently even more comprehensive studies of this relationship have been presented (de Kruiff et al. 2000; Reissing et al. 2004; Reissing et al. 2005; ter Kuile et al. 2005). Selected studies of dyspareunia/vulvar vestibulitis will also be discussed, as they are important for the discussion of the results of this thesis.

Definitions and operationalisation of terms

Vaginismus

Our studies are based on the definitions in *the Diagnostic and Statistical Manual of Mental Disorders, 4th ed. Text revision (DSM-IV-TR)* (American Psychiatric Association 2000). Vaginismus is here categorized as a subgroup of sexual pain disorders, and defined as: A. Recurrent or persistent involuntary spasm of the musculature of the outer third of the vagina that interferes with sexual intercourse (in the diagnostic features not only vaginal penetration by penis is included but also by finger, tampon or speculum). B. The disturbance causes marked distress or interpersonal difficulty. C. The disturbance is not better accounted for by another Axis I disorder (e.g., Somatization Disorder) and is not due exclusively to the direct physiological effects of a general medical condition. Vaginismus is further divided into lifelong (called primary in this thesis) or acquired (called secondary in this thesis), generalized or situational, and due to psychological or combined factors. At the end of the diagnostic features it is written: "In some females, even the anticipation of vaginal insertion may result in muscle spasm. The contraction may range from mild, inducing some tightness and discomfort, to severe, preventing penetration". Despite the fact that vaginismus is categorized as a sexual pain disorder in DSM-IV-TR, pain is not an obligate condition for the diagnosis.

Study criteria of partial and total vaginismus

To obtain more precise definitions of vaginismus, we added study criteria of partial vaginismus in four of our studies (II, III, V, and VI) and of total vaginismus in Study II.

Partial vaginismus was defined as a reflex contraction of the pelvic floor muscles that partly closes the vagina during penetration or attempt to penetrate (Wijma and Wijma 1997; American Psychiatric Association 2000). The reflex contraction makes penetration difficult, but not impossible; is beyond the control of the woman; and is simultaneously accompanied by burning pain (Engman et al. 2004). The reflex contraction of the pelvic floor muscles was ascertained by palpation with one or two fingers during a pelvic examination, and every reflex contraction of the muscles simultaneously accompanied by the woman's report of burning pain was defined as partial vaginismus (Engman et al. 2004).

In our study definition of partial vaginismus, penetration is not limited to penetration of vagina by a penis.

Our study definition of partial vaginismus does not include increased tension of the pelvic floor muscles.

Total vaginismus was defined as: 1) severe contraction of pelvic floor muscles preventing penetration; 2) the contraction is beyond the control of the women; and 3) by one of two alternative criteria: A) attempts of penetration are simultaneously accompanied by burning

pain, and feared or avoided; B) there is a pronounced fear or avoidance of vaginal penetration excluding all attempts of penetration.

Dyspareunia

In DSM IV TR (American Psychiatric Association 2000), the sexual pain disorders are divided into two subgroups, vaginismus and dyspareunia. According to this diagnostic system, a diagnosis of dyspareunia should not be set if the disturbance is caused exclusively by vaginismus. There is confusion in the literature between the use of the term dyspareunia and the DSM-IV-TR diagnosis of dyspareunia. The term dyspareunia is nowadays generally used for pain related to intercourse (Danielsson 2001). In this thesis, the term dyspareunia refers to “superficial coital pain”, where not otherwise specified.

Vulvar vestibulitis

As a definition of vulvar vestibulitis, we used Friedrich’s first and second criteria (Friedrich 1987): 1) severe pain on vestibular touch or attempted vaginal entry; 2) tenderness to pressure localised within the vulvar vestibule. The first criterion was confirmed by the woman’s history. The second criterion was confirmed by use of the Q-tip test in the vulvar vestibulum. For scientific use, we have dichotomized the second criterion: Only women who reported severe tenderness to the gynaecologist’s pressure within the vulvar vestibule (≥ 5 on a scale from 0-10) were diagnosed as having vulvar vestibulitis.

Friedrich’s third criterion, vestibular erythema, has been questioned in later studies (van Beurden et al. 1997; Bergeron et al. 2001a), and was therefore not included as a criterion of vulvar vestibulitis (see below, Page 4, Erythema as a diagnostic criterion of vulvar vestibulitis)

ISSVD has redefined vulvar vestibulitis as *provoked vestibulodynia*, (Moyal-Barracco and Lynch 2004), but in this thesis, we stick to the more commonly used term vulvar vestibulitis. Provoked vestibulodynia is by ISSVD defined as “discomfort on intromission (introital dysperunia), clothing pressure, tampon insertion, cotton-tipped applicator pressure, fingertip pressure, etc.” (Moyal-Barracco and Lynch 2004).

Generalized unprovoked vulvodynia was used as a term for vulvar pain during everyday life.

Pain was used as a comprehensive term for both burning pain and other types of pain in Study IV.

Severe pain, in our studies, means pain intensity ≥ 5 on a scale from 0-10.

Intercourse was (where not otherwise specified) used as a comprehensive term for both intercourse and attempted intercourse.

Attempted intercourse was defined as intercourse being interrupted due to pain, and/or fear of pain.

Technical intercourse means ability for the woman to let her partner slowly penetrate her vagina without feeling any or minimal pain.

Standardized penetration situation means a penetration situation where a defined “penetrator” is used, and a defined protocol directs the insertion and extraction procedures of the “penetrator” and moments of contraction/relaxation of the pelvic floor muscle.

Self-healing means a written or oral report to the clinic from a woman, who had been assessed and was on the waiting list, but refrained from therapy when she was invited for treatment, due to no or minimal remaining symptoms of pain during intercourse.

Erythema as a diagnostic criterion of vulvar vestibulitis

Bergeron et al. (2001a) stated that erythema does not appear to be a useful diagnostic criterion for a diagnosis of vulvar vestibulitis. The authors found in a sample of 146 women with dyspareunia that of Friedrich's three diagnostic criteria for vulvar vestibulitis, only tenderness to pressure within the vulvar vestibule differentiated dyspareunia patients with and without vulvar vestibulitis. They also found poor inter-rater agreement and test-retest reliability for the presence or absence of erythema (graded from 0-3) in women diagnosed as having vulvar vestibulitis by two independent gynaecologists. Bohm Starke et al. (2001a) found a significant difference in erythema in the mucosa around the vaginal introitus between patients with vulvar vestibulitis (n=20) and controls (n=21), but there was an evident overlap of the two groups. In this study, however, erythema was used as a diagnostic criterion of vulvar vestibulitis.

In addition, women without vulvar complaints may present with vestibular erythema. Van Beurden et al. (1997) reported on 40 women without vulvar complaints and found vestibular erythema in 17 women (43%). The "touch test" was positive in 9 of 17 women (53%) with vestibular erythema.

Prevalence

Primary and secondary vaginismus (unclear if partial vaginismus is included) is thought to be one of the most common female psychosexual dysfunctions (Crowley et al. 2006). The prevalence and incidence in the general population are unknown (Spector and Carey 1990) and dependent on the definition of vaginismus. Prevalence rates from sexual dysfunction clinics vary from 5-17% (Bancroft and Coles 1976; Renshaw 1988; Catalan et al. 1990; Hirst et al. 1996). In a Swedish population group, 1% of women had had vaginismus during the last 12 months (defined as difficult or impossible to penetrate with penis into the vagina) (Fugl-Meyer 1996). "It may be assumed that temporary vaginismus is a common and transient experience in young women when they begin to have coitus" (Wijma and Wijma 1997). Vaginismus would probably often be found among women attending care for coital pain if appropriate diagnostic efforts are made.

Relationship between dyspareunia/vulvar vestibulitis and vaginismus

Despite the division of sexual pain disorders in DSM-IV-TR (American Psychiatric Association 2000) into two main groups, dyspareunia and vaginismus, there has lately been a discussion in scientific literature on a relationship between the two diagnostic groups.

When we started our studies, some articles with a clear definition of vaginismus (difficult/painful/not existing vaginal penetrations, including TPV, TSV, PaPV, PaSV) discussed a relationship between dyspareunia/vulvar vestibulitis and vaginismus (Spano and Lamont 1975; Lamont 1978; Steege 1984; Shortle and Jewelewicz 1986). Studies with more unclear definitions of vaginismus also discussed this relationship (Schover et al. 1992; Monif and Belatti 1993; Goetsch 1996; Hassel 1997; Nunns and Mandal 1997; White and Jantos 1998). Some studies had found both diagnoses among women with lifelong vaginismus (defined as a lifelong history of pain and difficulty with vaginal entry, PaPV and TPV)

(Basson 1995) or among women with vulvar vestibulitis (definition of vaginismus unclear) (Abramov et al. 1994). Other researchers found difficulties in differentiating between vaginismus (definition according to DSM-III, but unclear) and dyspareunia (van Lankveld et al. 1995).

During and after our studies more substantiated studies on this relationship have been presented: de Kruiff et al. (2000) reported on great difficulties in distinguishing between women with primary and secondary vaginismus (not further defined) and women with dyspareunia/vulvar vestibulitis by means of history and/or clinical examination. Reissing et al. (2004) also reported on difficulties in distinguishing between women with vaginismus (inability to experience vaginal intercourse, TV) and women with dyspareunia/vulvar vestibulitis by means of history and/or clinical examination.

Reissing et al. (2005) reported clear pathology of the pelvic floor muscles (including vaginal hypertonicity, and restriction of the vaginal opening) in 90% of women with vulvar vestibulitis. ter Kuile et al. (2005) described pain on touch in the vestibulum in 69% of women with lifelong vaginismus (women who had ever had full intercourse were excluded, TPV).

There exist several different possibilities to theorize about the relationship between dyspareunia/vulvar vestibulitis and vaginismus.

In 1978, Lamont (Lamont 1978) stated that vaginismus (difficult/painful/impossible vaginal penetrations, including TPV, TSV, PaPV, PaSV) and dyspareunia are undeniably linked. "Repeated dyspareunia is likely to result in vaginismus and vaginismus may be the causative factor in dyspareunia". Other authors have also expressed that "it is not possible to differentiate if the tightness of the pelvic floor muscles in women with dyspareunia is due to a vaginismus-like disorder, or is an anticipatory guarding response typically seen in chronic pain conditions" (White and Jantos 1998), "to prevent further penetration and consequent pain and tissue damage" (Graziottin and Brotto 2004).

Some authors believe that vaginismus/abnormal muscle function is the primary problem in the course of the development of dyspareunia and that repeated trauma of coitus in an unaroused state with abnormal muscle tension may establish vulvar inflammation and lesions in the vulnerable woman (Schover et al. 1992; Monif and Belatti 1993). Other authors apprehend the pelvic floor pathology in women with dyspareunia/vulvar vestibulitis as a secondary protective muscle guarding response (Goetsch 1996; Nunns and Mandal 1997; Mariani 2002; Reissing et al. 2005). In none of these studies is there any scientific evidence for the opinion stated.

Both views are thoroughly disputed in a recently published conceptual model for the pathophysiology of vulvar vestibulitis by Zolnoun et al. (2006). In this model, it is stated that "it is plausible that in some women with vestibulitis, pelvic floor muscle dysfunction may act as an initiator of sensory changes in susceptible mucosa; whereas in others, muscle dysfunction may occur in response to mucosal inflammation". "Well-established constructs in neurosensory research support this concept of muscle contraction as either an initiator or a consequence of skin inflammation or an ongoing component of sustained dysfunction" (Graven-Nielsen and Arendt-Nielsen 2002; Zolnoun et al. 2006).

Aetiology

Most articles on the aetiology of vaginismus deal with women with total primary vaginismus. The aetiological studies are often characterised by focusing either on only biological or only

psychological predisposing factors (Wijma et al. 2007), although many authors agree that vaginismus is a psychophysiological problem (Lamont 1978; Steege 1984; Kessler 1988; Scholl 1988; Hawton and Catalan 1990; van de Wiel 1990; Ward and Ogden 1994; Ogden and Ward 1995; van Lankveld et al. 1995; Wijma and Wijma 1997; Reissing et al. 1999; van der Velde and Everaerd 2001; van der Velde et al. 2001; Reissing et al. 2004; Rosenbaum 2005).

According to Masters and Johnson (Masters and Johnson 1970), vaginismus was a classical example of *psychosomatic* illness.

Biological aspects

Lamont (Lamont 1978) stated that any medical problem causing persistent dyspareunia might result in vaginismus (difficult/painful/impossible vaginal penetrations, including TPV, TSV, PaPV, PaSV). Barnes et al. (1984) hypothesised that women with total primary vaginismus (TPV) had a faulty perception of vaginal muscle tone. They fail to distinguish between a relaxed state and spasm and are unaware that tone can be voluntarily altered. This hypothesis is in accordance with later findings where muscle contraction might be a consequence of a sustained dysfunction in the muscles (Graven-Nielsen and Arendt-Nielsen 2002).

Vaginismus as general defence mechanism

Vaginismus has been suggested to be part of a general defence mechanism in both women with and without vaginismus.

This idea is supported by the findings in two studies by van der Velde et al. (van der Velde et al. 2001; van der Velde and Everaerd 2001). Exposure to threatening and sexual-threatening excerpts, but not neutral and erotic excerpts, resulted in increase in muscle activity of pelvic floor muscles (van der Velde and Everaerd 2001), and pelvic floor muscles and postural muscles (van der Velde et al. 2001); as measured by sEMG, both in women with and without vaginismus. There were also positive associations between recorded changes in the pelvic floor muscle activity, measured by sEMG, and experienced threat in both groups (van der Velde and Everaerd 2001). In both studies, vaginismus was defined according to DSM-IV, but not specified as total or partial. The included women were found by advertisements. Only women who were able to insert the vaginal device themselves were included. This aspect makes it reasonable to assume that women with total vaginismus and/or phobic avoidance of vaginal penetration did not participate in the study, which may have influenced the results. In the total vaginismus group, even erotic excerpts of vaginal penetration could be perceived as threatening.

Psychoanalytic theory

In classical psychoanalytic theory, vaginismus (definition unclear) is conceptualized as a conversion disorder caused by unresolved psychosexual conflicts from early childhood (van de Wiel 1990; Reissing et al. 1999). On the basis of these theories, vaginismus (defined according to DSM-III (?) TPV), has been described as a protective symptom and a defence against a perceived fearful violation (Silverstein 1989).

Aetiological factors in family patterns of women with primary vaginismus have also been discussed from a psychoanalytical point of view (Barnes 1986b).

Learning theory/Fear of intercourse/pain

According to these theories, fear of pain and/or fear of penetration may contribute to vaginismus.

Already in 1909, theories were presented of vaginal muscle spasm as a phobic reaction resulting from fear of pain (Waltherhard 1909). Fear of intercourse in general has also been suggested as an aetiological factor, and the fear itself acts as a maintaining factor (Barnes et al. 1984). Wijma and Wijma (1997) suggested that conditioning of fear and/or pain to a penetration situation or to genitals and their functions predisposed to vaginismus (including TPV, TSV, PaTV and PaPV) after which the penetration situation itself or even thoughts of penetration may trigger the reflex. In a randomised study of vaginismus (lifelong, TPV) treatment (three-month CBT or waiting list control), successful outcome (full vaginal penetration by the partner's penis) was partly mediated by changes in fear of coitus and changes in avoidance behaviour (ter Kuile et al. 2006) (fear of pain during and/or after intercourse was not included in the study measurements as a specific item). This finding may support the behavioural model of vaginismus used by this research group, in which "the vaginistic reaction represents a conditioned fear response to certain (sexual) stimuli that can be overcome by exposure therapy" (ter Kuile et al. 2006).

Recently Reissing et al. (2004) has suggested that the spasm-based definition of vaginismus (defined as inability to experience vaginal intercourse, TV) is not adequate. Instead, they suggest that pain and fear of pain, pelvic floor dysfunction, and behavioral avoidance should be included in a multidimensional reconceptualization of vaginismus.

Cultural aspects

Only a few studies, excluding feminist studies, discuss the social and relational aspects of vaginismus.

Some authors express that the cultural milieu — Irish (Barnes 1986b) (TPV) and Turkish (Kabakci and Batur 2003) (definition unclear, DSM-IV) — must be taken into account in studies of vaginismus. "The theoretical frameworks of psychotherapy may not necessarily be appropriate for clients from non-Western cultures" (Kabakci and Batur 2003).

Feminist/gender theories

Much of the literature on feminist theories of vaginismus is written in Dutch and has been summarised in an article by Drenth (Drenth 1988). "This literature emphasises that sex is strongly subject to social mores and conventions, to ineradicable myths about masculinity and femininity, and that sex is defined from the male standpoint in our Western civilisation" (Drenth 1988).

Sexual and physical abuse

Childhood physical and sexual abuse may, according to some of the existing literature, contribute to vaginismus.

Reissing et al. (2003) used a structured interview questionnaire, Sexual and Physical Abuse History Questionnaire (Leserman et al. 1995). They found that women with total vaginismus were twice as likely (23/29) to report childhood sexual interference (attempts at sexual abuse and sexual abuse involving touching) as women in a "no pain" group (11/29). For sexual

interference that occurred during adulthood or for childhood or adult physical abuse, no differences were found between total vaginismus, dyspareunia/vulvar vestibulitis and “no pain” groups.

Twenty-three percent of women with vaginismus (all attempts at intercourse failed, TPV) had had a violent father during childhood compared to 9% in a reference group of women with other sexual problems (Barnes 1986b).

In a retrospective analysis of therapy records of women with vaginismus (DSM-III (?), TPV) 13/22 had experienced intrafamilial abuse (Silverstein 1989).

Women with vaginismus may also show symptoms of vulvodynia. In a study with self-administered surveys in 125 women with adult-onset vulvodynia and 125 age- and community-matched controls, the diagnosis of vulvodynia was strongly associated with childhood physical abuse (OR = 4.1) and childhood sexual abuse (OR = 6.5) (Harlow and Stewart 2005). The prevalence of vaginismus was not reported. In that study, more than 80% of women with vulvodynia reported pain on contact during intercourse or a pelvic examination for three months or longer, and 27% had always experienced pain during intercourse.

Van Lankveld et al. (1995) found no significant differences in prevalence (17-22%) of lifetime sexual abuse (not further defined, interview questions) between women with primary and secondary vaginismus (n=50), dyspareunia, (n=46) and both diagnoses (n=51) (diagnoses according to DSM-III-R) and women in randomised samples from the population.

No differences were found between women with lifelong vaginismus (TPV) (n=117) and women from a general population sample in prevalence of experiences of physical and sexual abuse (van Lankveld et al. 2006), as measured by Sexual and Physical Abuse Questionnaire (SPAQ) (Kooiman et al. 2002) (measuring the severity of hands-on sexual and physical abuse).

Male partners to women with vaginismus

Male partners to women with vaginismus (used as a general term) have been described as passive and unassertive (Reissing et al. 1999). Barnes stated that the partner should always be suspected of having a maintaining role for the symptoms of vaginismus (TPV) (Barnes 1986b). However, no group differences have been established by use of a questionnaire (Crown–Crisp Experiential Index (CCEI), (Crown and Crisp 1979)) when comparing male partners to women with vaginismus (TPV) with a male population sample (Kennedy et al. 1995). By use of a multidimensional indicator of psychopathology (Symptom Checklist, SCL-90 (Derogatis et al. 1973)), no differences were found when comparing male partners to women with vaginismus (definition according to DSM-II, but unclear) with male partners to women with dyspareunia (van Lankveld et al. 1995). Hawton and Catalan (1990) claimed that male partners to women with vaginismus (no definition) have even less sexual dysfunction than male partners to women with other female sexual dysfunctions. Male partners to women with vaginismus (definition according to DSM-III, but unclear) have also been reported to have a lower prevalence of sexual dysfunctions than men in a male population sample (van Lankveld et al. 1995). During the course of therapy for vaginismus, temporary erectile and ejaculatory dysfunction have been described (Lamont 1978; Barnes 1986a).

Evolutionary aspects

We found no articles with an evolutionary aspect on vaginismus. Wijma and Wijma have stated “from an evolutionary point of view, the vaginistic reflex is in the first place an adequate defence reflex by which a woman may be able to avoid a penetration which she fears or dislikes” (Wijma and Wijma 1997) (TPV, TSV, PaPV, PaSV). When conditioning has occurred, the reflex most often does not suit its purpose, as it stops the woman from allowing a penetration that she wishes.

EMG-findings

For theories of EMG measurements see page 14.

From the beginning of our studies, we have searched for a method to measure the vaginistic reflex and the increased tone in the pelvic floor muscles in women with vaginismus. At first, we tried to use vaginal manometry. We found vaginal pressure measurements to be of little value for registering increased tone in the pelvic floor muscles: there were often technical problems during the investigation, for example, the transducer was pressed out of the vagina by the increased tone (Wijma et al. 2000), which invalidated registrations. At the starting time of our sEMG study we knew of one study which asserted that a diagnosis of vulvar vestibulitis could be confirmed by sEMG measurements (White et al. 1997).

Women with vulvar vestibulitis compared to an asymptomatic group

White et al. (1997) proposed that the diagnosis of vulvar vestibulitis can be established by sEMG measurements of the pelvic floor muscles. The authors presented both values of asymptomatic women and characteristics of women with diagnoses of vulvar vestibulitis. The “asymptomatic” group (referred by the authors as “non morbid”) comprised, however, mostly women who had been treated earlier with sEMG-biofeedback because of vulvar vestibulitis, which may have biased the results. The outcome of a diagnostic procedure when applying the proposed sEMG criteria for women with and without a history of vulvar vestibulitis has, to our knowledge, not been published.

Women with dysesthetic vulvodynia compared to asymptomatic women

Glazer et al. (1998) found that an asymptomatic group of women had higher contractile amplitudes registered by sEMG than a group of women with dysesthetic vulvodynia (definition unclear). Baseline level showed lower values in the asymptomatic than in the symptomatic group. According to Glazer, the sample size of the study was insufficient to suggest normal pelvic floor values.

Women with vaginismus compared to women without vaginismus

It is difficult to interpret the results of existing studies where women with and without vaginismus have been examined with EMG of the pelvic floor muscles.

Van der Velde and Everaerd (1999) used a vaginal sensor for the sEMG recordings and found no difference in ability to contract and relax the pelvic floor muscles between women with and without vaginismus. The authors refer to the definition of vaginismus in DSM-IV, but it is unclear if women with partial vaginismus were included, and even more uncertainty is

added by the inclusion criteria as it is stated: “the included women felt no pain or discomfort during the insertion of the sensor”. In our studies, 76% of women with vaginismus reported burning pain during the insertion of the vaginal sensor. It is difficult to tell if this difference is due to different diameters of the vaginal sensors used, since the diameter of the vaginal sensor used by van der Velde is not reported.

Shafik and El-Sibai (2002) used needle-EMG of pelvic floor muscles. They found higher EMG activity at rest and on induction of the vaginismus reflex in seven women with vaginismus (duration one to two months) than in seven healthy volunteers matched by age. The author’s definition of vaginismus is unclear. Vaginismus was in some patients evoked by simply touching the vaginal introitus, and in some by introducing a vaginal dilator (not further described) into the vagina. Time from insertion of the EMG-needles to provocation of the vaginismus reflex was not stated. Due to this lack of information, we do not know to what extent the pain evoked by the needle insertion acted as conditioned stimulus in women with vaginismus and provoked a vaginismus reflex. If so, the increased EMG activity measured at rest might in fact be due to a vaginistic reflex.

Women with vaginismus and women with vulvar vestibulitis compared to a no pain group.

Reissing et al. (2004) compared the sEMG readings in three groups of women: women with a history of total primary or secondary vaginismus (defined as inability to experience vaginal intercourse, TV); women with dyspareunia/vulvar vestibulitis (defined as ability to experience vaginal penetration, but such penetration is painful); and women in a no-pain control group (without vulvar/vaginal/pelvic pain and penetration difficulties). The results were possibly biased by the fact that 46% of the women in the vaginismus group refused to undergo the sEMG evaluations. There were no significant group differences in pelvic floor muscle tone measured by sEMG prior to and after muscle contractions. Both the vaginismus and the dyspareunia/vulvar vestibulitis groups differed significantly from the “no pain” group in strength (amplitude) of contractions.

Pain measurements

Pain in women with vaginismus

In DSM-IV-TR, vaginismus is sub-classified as a sexual pain disorder but pain is not necessary for the diagnosis, and dyspareunia should not be diagnosed if the pain problem is caused exclusively by vaginismus. Most articles on vaginismus do not discuss the pain component at all (Colgan and Beautrais 1977; O’Sullivan and Barnes 1978; Barnes et al. 1984; Barnes 1986b; Barnes 1986a; Elkins et al. 1986; Scholl 1988; Shaw 1994; Kennedy et al. 1995; Ogden and Ward 1995; Drenth et al. 1996; Schnyder et al. 1998; van der Velde and Everaerd 1999; Katz and Tabisel 2001a; Katz and Tabisel 2001b; van der Velde and Everaerd 2001; van der Velde et al. 2001; Kabakci and Batur 2003; ter Kuile et al. 2006). Many of these articles only deal with primary, total vaginismus and these women often avoid attempts at penetration and by this also pain. Other authors express that pain is a prominent feature of vaginismus (Lamont 1978; Reamy 1982; Steege 1984; Shortle and Jewelewicz 1986; Kessler 1988; Hassel 1997; Wijma and Wijma 1997; Ng 1999; Reissing et al. 1999; de Kruiff et al. 2000; Har-Toov et al. 2001; Kaneko 2001; McGuire and Hawton 2001; Reissing et al. 2004; Kimberly et al. 2005; Reissing et al. 2005; Rosenbaum 2005; ter Kuile et al. 2005). The pain in women with partial vaginismus is only described in a few articles (Caplan 1988; Kessler 1988; Wijma and Wijma 1997).

Pain in women with vaginismus compared to pain in women with dyspareunia/vulvar vestibulitis

There are very few articles with any systematic investigation of the co-morbidity of vaginismus and dyspareunia. Basson (Basson 1995) has clinically found that 46% (23/50) of women with lifelong vaginismus (defined as lifelong history of pain and difficulty with vaginal entry, PaPV and TPV) had vestibulitis (pain on vestibular touch) in addition to the vaginismus, and it was impossible by history (including pain history) to distinguish the two groups. De Kruiff et al. (2000) found no differences between women with primary and secondary vaginismus (not further defined, PV and SV, n=14) and women with dyspareunia (n=16) in the reported level of pain during coitus (or attempted coitus), when inserting one finger into the vagina, or during a gynaecological examination. In her study, 81% of women with dyspareunia and 69% of women with vaginismus fulfilled the diagnostic criteria (positive touch test) of vulvar vestibulitis. Reissing et al. (2004) found no differences between women with vaginismus (n=29) (defined as inability to experience vaginal intercourse, TV) and women with dyspareunia/vulvar vestibulitis (n=29) on retrospective pain reports, and on pain measures. Ter Kuile et al. (2005) found pain "on touch" in the vestibulum in 69% and erythema and pain on the same location in 56% of patients (n=91) with lifelong vaginismus (women who had ever had full intercourse were excluded, TPV).

Treatment studies

General findings

Vaginismus is traditionally believed to be simple to treat and with excellent treatment outcome. The lack of valid therapy studies in the field of vaginismus has been clearly demonstrated in a Cochrane report (McGuire and Hawton 2003). Only one randomized study was included in the report (Schnyder et al. 1998). The same discouraging findings of treatment studies of vaginismus have also been found in two comprehensive review articles (van de Wiel 1990; Reissing et al. 1999); hardly any of the publications met even the most basic methodological demands (van de Wiel 1990; Reissing et al. 1999). A definition of the term vaginismus is often unclear or even absent. In our own review of the vaginismus literature we also found that the therapist in almost all the studies evaluates effect of therapy; in some cases, it is even unclear whose evaluation is reported. Interview data or questionnaires are seldom used (Wijma et al. 2007).

Randomized treatment studies

Until today only two randomized treatment studies of women with vaginismus have been presented. In a study by Schnyder et al. (1998), women with vaginismus (defined according to DSM-III; but unclear as to inclusion of both total and partial forms of vaginismus) were randomized to insertion of a vaginal mould by the therapist or by the woman herself during the first therapy session. There was no difference in main treatment outcome between the two groups. In 2006, van Lankveld et al. presented a study where women with lifelong vaginismus (duration of problem 11 years, defined according to DSM-IV including recurrent and persistent spasm, but women who had had full intercourse at any time were excluded, TPV) were randomized to either group therapy or bibliotherapy, both cognitive behaviour therapy oriented, or a waiting list group. The short treatment period of three months may partly explain the low success rates of 21% and 15%, measured as successful intercourse (not further defined) at follow-up (one year), in both therapy groups (van Lankveld et al. 2006).

Therapeutic approaches

Therapeutic approaches for vaginismus often include combinations of systematic desensitisation (in vivo and/or imaginal) together with the use of dilators of increasing diameters (Lamont 1978; Barnes 1985; Barnes 1986b; Scholl 1988; Abramov et al. 1994; Basson 1995; Schnyder et al. 1998; Har-Toov et al. 2001; Seo et al. 2005), electromyographic biofeedback (Barnes 1985; Seo et al. 2005), sex therapy/education (Lamont 1978; Barnes 1985; Barnes 1986b; Kennedy et al. 1995; Drenth et al. 1996; van Lankveld et al. 2006), cognitive behaviour therapy (Scholl 1988; Hawton and Catalan 1990; Drenth et al. 1996; Wijma and Wijma 1997; Kabakci and Batur 2003; van Lankveld et al. 2006), relaxation therapy (van Lankveld et al. 2006), psychodynamic interventions (Barnes 1986b; Kennedy et al. 1995), Kegel exercises (Lamont 1978), physiotherapy (Rosenbaum 2005), hypnotherapy, (Fuchs 1980), treatment with benzodiazepines, in (van de Wiel 1990), botulinum toxin injections (Shafik and El-Sibai 2000; Ghazizadeh and Nikzad 2004). Rapid desensitization with insertion of a vaginal mould during anaesthesia is also used (Biswas and Ratnam 1995; Goonewardene 2005). Surgery is, according to van de Wiel, still used but never reported (van de Wiel 1990). Many authors mixed several forms of therapy.

Aims of therapy and measurement of treatment results

The aims of therapy and measurement of treatment results in a majority of studies on vaginismus have been ability to have intercourse, and mostly with successful intravaginal ejaculation of the male partner (Wijma et al. 2007). As late as in 2005 “full penetration and ejaculation” is used a measurement of treatment outcome (Seo et al. 2005) in women with vaginismus. Consummation as the only “successful” treatment goal has been questioned (Drenth 1988; Shaw 1994; Drenth et al. 1996; Crowley et al. 2006). Psychoanalytically oriented authors have also argued against the fact that most treatment studies are aiming at performance and not at exploring the message of the symptom vaginismus (Shaw 1994). For some women the treatment goal is pregnancy rather than achievement of penetration (Drenth 1988; Drenth et al. 1996).

Many nonrandomized treatment studies on vaginismus present very good effect of therapy, often with success rates above 80%, even 100%, at end of therapy or at follow-up (3–12 months) (Barnes et al. 1984; Barnes 1986; Scholl 1988; Hawton and Catalan 1990; Abramov et al. 1994; Basson 1995; Kennedy et al. 1995; Har-Toov et al. 2001; Kabakci and Batur 2003; Seo et al. 2005; Zukerman et al. 2005; Jeng et al. 2006). Therapy results in these studies are often evaluated by the therapists or it is unclear by whom. Dropout rates are seldom reported (Wijma et al. 2007).

Only a few previous studies report on the women’s ability to enjoy intercourse. One study measured pleasure during intercourse at follow-up (six weeks to three months), with a success rate of 80% (total and partial, primary and secondary vaginismus) (Lamont 1978), and one study measured regular intercourse with orgasm, with a success rate of 83% (total and partial primary vaginismus) (Jeng et al. 2006). None of these studies reported on pain during intercourse. Some studies have reported improvement in follow-up compared to pre-treatment measures of different sexual parameters, e.g. sexual desire (Hawton and Catalan 1990; Schnyder et al. 1998; van Lankveld et al. 2006), sexual arousal (van Lankveld et al. 2006), sexual satisfaction (van Lankveld et al. 2006), orgasm capacity (Schnyder et al. 1998), and pleasant feelings in sexual situations (Hawton and Catalan 1990; Schnyder et al. 1998; van Lankveld et al. 2006).

Long-term follow-up results

Long-term follow-up results (> one year after finished treatment) of women with vaginismus have been presented in two studies during the last 20 years (Scholl 1988; Drenth et al. 1996). Both studies dealt with total primary vaginismus and focus was not the long-term follow-up results. Scholl (1988) used telephone communication for follow-up, and reported that 19 of 20 successfully treated patients (behaviour therapy and systematic sensitization) continued to have coitus at least one year after finishing therapy (follow-up one to four years). Drenth et al. (1996) used a questionnaire focusing on procreation, and reported that 54% of women had reached required results of therapy (behaviour and sex therapy), meaning sexual intercourse and/or pregnancy at follow-up (maximum five years). None of these studies described whether the women had any pain or fear during intercourse, nor whether they were able to enjoy intercourse at follow-up.

Conditioning of the vaginistic reflex

According to a learning-theory framework, the main problem in women with both total and partial vaginismus may be characterized as a conditioned reflex (Schwartz 1989; Wijma and Wijma 1997; ter Kuile et al. 2006; van Lankveld et al. 2006). “From an evolutionary point of view, the vaginistic reflex initially is an adequate defence reflex by which a woman may be able to avoid a penetration which she fears or dislikes” (Wijma and Wijma 1997) (Figure 1).

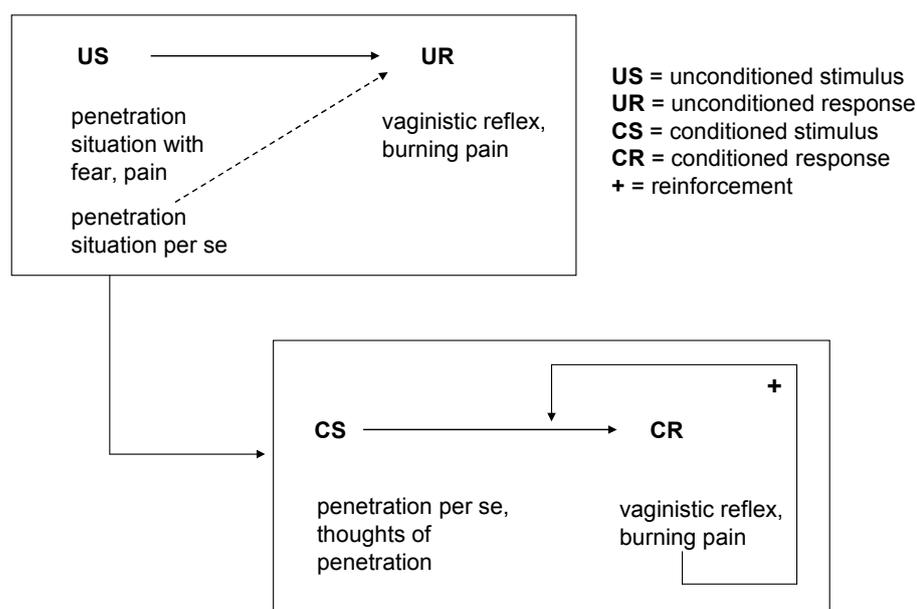


Figure 1. Conditioning of the vaginismus reflex (Wijma and Wijma, 1997), reprinted with permission from Taylor & Frances.

Penetration with fear or pain (US, unconditioned stimulus) can elicit the vaginistic reflex (UR, unconditioned response), which itself can induce burning pain. “Problems occur as soon as conditioning takes place, i.e. the reflex is elicited by conditioned stimuli such as foreplay, touching of the genitals, or by the penetration situation itself” (Wijma and Wijma 1997). In this case the penetration situation per se, or thoughts of penetration (CS, conditioned stimulus), can elicit the vaginistic reflex (CR, conditioned response), which itself is accompanied by pain, and a vicious circle develops. The contractions of the pelvic floor

muscles and the concomitant burning sensation itself act as a discriminative stimulus (S^D) for avoidance/flight reactions and reinforce the vaginistic reflex and the burning pain sensation (Figure 2).

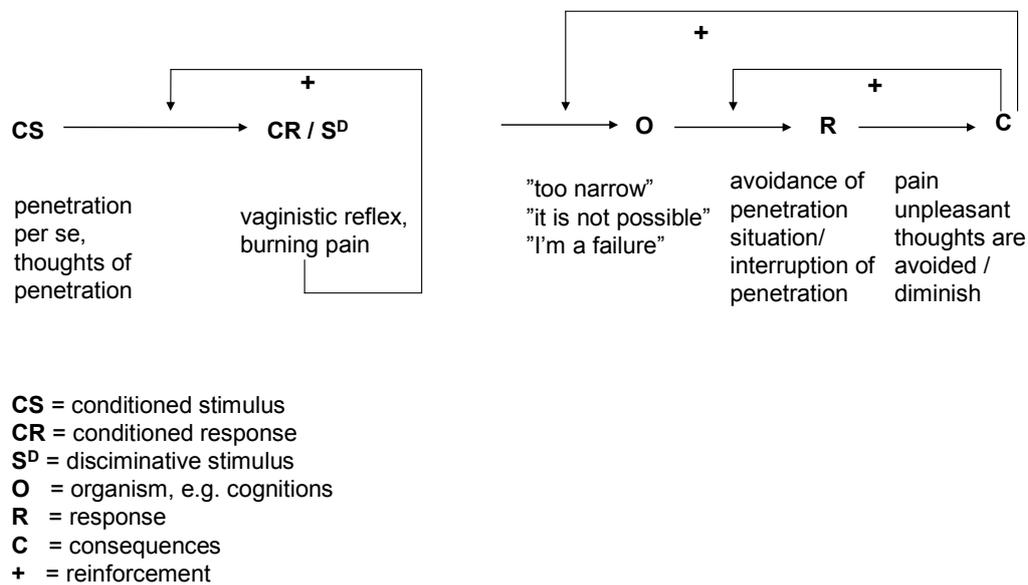


Figure 2. Negative reinforcement of the vaginismus reflex (Wijma and Wijma, 1997), reprinted with permission of the publisher, Taylor & Frances.

“The bodily reaction experienced as burning pain is often wrongly attributed as a signal that something is wrong” (Wijma and Wijma 1997) (O, organism) and leads to a wish to interrupt/avoid the unpleasant feelings/thoughts; see Figure 2. This avoidance behaviour (R, response) acts as a maintaining factor in women with vaginismus. The pain and/or the unpleasant feelings diminish (C, consequence) at the very moment of the avoidance, but the avoidance behaviour reinforces the problem in the long run, as the erroneous cognitions are not corrected by experience. The erroneous cognitions may also act as discriminative stimuli per se and elicit a vaginistic reflex, i.e. thoughts of the problem lead to the conditioned response, a vaginistic reflex. Thoughts of the problem “all the time” lead to a continuous conditioned response and hypertonicity of the pelvic floor muscles.

EMG measurements and muscle physiology

EMG measurements

Electromyography is a method to measure the electric potential field evoked by active muscle fibres. Two main methods exist. Noninvasive surface electromyography (sEMG) with measurements through the intact skin/mucosa, and invasive needle EMG. Almost no studies have compared needle EMG and sEMG recordings with respect to sensitivity and specificity of diagnostic properties (Zwarts and Stegeman 2003). The small measurement area of needle electrodes and the fact that the signal amplitude depends on the exact position of only a few muscle fibres near the needle tip, negatively affect the reproducibility of needle EMG (Zwarts and Stegeman 2003). In the case of sEMG, the signal provides data regarding the integrated activity of a motor unit (several skeletal muscle fibres supplied by one motor neuron) or of a whole muscle. The sEMG measurements are influenced by the distance from the electrode to

the muscle, and by the tissue layers with different properties between the source and the recording sEMG electrode (Merletti et al. 1999a; Merletti et al. 1999b; Blok et al. 2002).

Muscle contraction

A skeletal muscle contraction occurs briefly in the following way: An action potential in a motor nerve initiates an action potential in the muscle fibre membrane that travels along the membrane. The action potential depolarises the muscle membrane — these are the depolarisations that are registered with EMG — and also travels deeply into the muscle fibres. Here it causes the sarcoplasmic reticulum, which surrounds each myofibril, to release large quantities of calcium ions into the myofibrils. The calcium ions initiate the contractile process, and cause the actin and myosin filaments to slide together, producing shortening of the muscle fibre. After this process the calcium ions are pumped back into the sarcoplasmic reticulum, where they are stored until a new action potential comes along (Guyton 1991, pp 56-67). Repeated action potentials in the muscle fibres are necessary to create a muscle contraction. The degree of muscle contraction is dependent on the number of activated motor units and the firing rate of the action potentials.

Muscle tone

A certain level of muscle tone is present in a normal, conscious and relaxed person. However, in the relaxed state there is no EMG activity in normal skeletal muscles (Shumway-Cook and Woollacott 2001). The tone of resting muscles devoid of action potentials has been found to be enigmatic (Simons and Mense 1998). According to a theory by Simons and Mense (1998), muscle tension depends physiologically on two factors: the basic viscoelastic properties of the soft tissues associated with the muscle, and/or on the degree of activation of the contractile apparatus of the muscle. Contractile activity — often identified as spasm when used in the general sense — may occur in three different forms: (1) electrogenic stiffness (muscle tension coming from electrogenic muscle contraction, i.e. observable EMG activity) in normal individuals who are not completely relaxed; (2) electrogenic spasm (observable by EMG) that specifically identifies pathological involuntary electrogenic contraction; (3) contracture, arising endogenously within the muscle fibres independent of any EMG activity (Simons and Mense 1998).

EMG recordings only identify electrogenic contraction (forms 1 and 2 above), as it measures the depolarisation of the muscular membranes, and therefore not endogenous contracture of the contractile apparatus of skeletal muscle. This endogenous contracture is often caused by excessive release or impaired uptake of calcium by the sarcoplasmic reticulum (Simons and Mense 1998).

Pelvic floor muscles

Anatomy

The vagina is normally narrowest in its lower third where it is constricted laterally by the adjacent portions of the levator ani muscles. The levator ani is part of the deep muscle layer of the pelvic floor, and is composed of two portions; pubococcygeus and iliococcygeus (Nichols and Randell 1989; Travell and Simons 1992) (Figure 3). The medial and anterior division of the levator ani muscle, the pubococcygeus, is from the gynaecologist's clinical point of view the most significant component of the levator ani (Nichols and Randell 1989). The

pubococcygeus muscle attaches along the dorsal surface of the pubic bone from the symphysis to the obturator canal. The gap between vagina and anus and between anus and os coccygeus are filled with fibromuscular tissue constituting the perineal and the anococcygeal body. The two halves of the pubococcygeus muscles meet in the midline in these fibromuscular structures, some at the perineal and some at the anococcygeal body (Travell and Simons 1992). Ileococcygeus constitutes the posterior portion of the levator ani. It anchors to the tendinous arch of the levator ani and to the spine of the ischium. Both sides of the muscle fuse, between the anus and coccyx, before they insert into the coccyx (Gould 1991).

Some authors also include the puborectalis (Gould 1991) and/or the ischiococcygeus (coccygeus) muscles (Nichols and Randell 1989) in the levator ani. Puborectalis might also be regarded as a part of pubococcygeus (Nichols and Randell 1989).

In the superficial layer of the pelvic floor muscles, three muscles form a triangle (Figure 3). The medial part, the bulbocavernosus muscle (also termed bulbospongiosus and/or sphincter vaginae), surrounds the introitus of the vagina. The muscle arises posteriorly from the perineal body and inserts into the dorsum of the clitoris and fascial structures. The superficial transverse perinei muscles and fibres from the external anal sphincter are also inserted into the perineal body (Nichols and Randell 1989; Travell and Simons 1992). In women, only a few fibres of the deep transverse perinei muscles cross the midline between the rectum and the vagina (Nichols and Randell 1989).

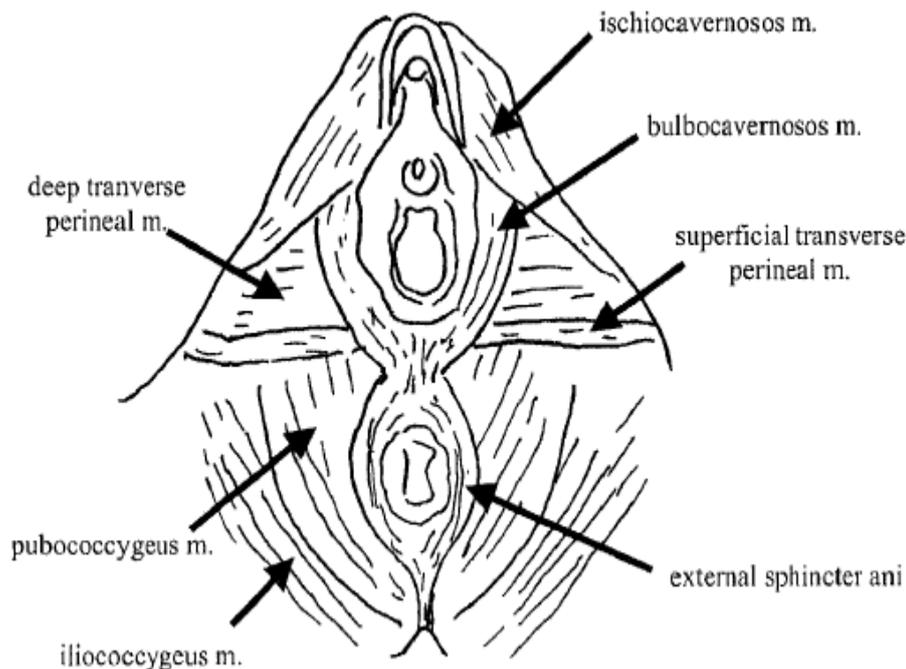


Figure 3. Pelvic floor musculature, (Reissing et al. 2005) reprinted with permission from Journal of Psychosomatic Obstetrics & Gynecology. <http://www.tandf.co.uk/journals>

Muscles included in the vaginismus reflex

Neither in DSM-IV-TR (American Psychiatric Association 2000) nor in most studies of vaginismus are the muscles defined which are supposed to be involved in a vaginismus reflex (Reissing et al. 1999; Payne et al. 2005). Lamont (1994) in (Reissing et al. 1999) suggested that these are the bulbocavernosus muscles and/or the pubococcygeus muscles.

A muscular mechnwork of smooth muscle fibres are described in the vaginal wall (Smouth et al. 1969). It is not known if these smooth muscle fibres take part in the vaginismus reflex.

By use of needle EMG in women with vaginismus (n=7) Shafik and El-Sibai (2002) showed an increased activity at rest in the levator ani, puborectalis and bulbocavernosus muscles. When a vaginismus reflex was provoked, all the examined muscles contracted. This study has several shortcomings discussed above (see EMG findings page 10); besides a limited amount of participants.

In treatment of vaginismus with Botulinum toxin, injections in either bulbocavernosus (n=8) (Shafik and El-Sibai 2000), or in the levator ani muscle (defined as puborectalis), (n=24) (Ghazizadeh and Nikzad 2004), (i.e. in both the deep and the surface muscle layers) have shown satisfactory results. As puborectalis is not adjacent to the vagina, but instead forms a sling posterior to the rectum, the choice of this muscle in one of the studies (Ghazizadeh and Nikzad 2004) is somewhat confusing.

By vaginal palpation, specially trained physiotherapists have registered significantly more hypertonicity of the pelvic floor in women with vulvar vestibulitis than in women without pain during intercourse, both in the superficial (not divided into different muscles) and in the deep muscle layers (pubococcygeus but not iliococcygeus) (Reissing et al. 2005). By anal palpation of the pubococcygeus and the puborectalis, no difference in hypertonicity was found between the groups (Reissing et al. 2005).

Innervations of the female external genitals and vagina

Sensory innervation

Most of the sensory innervation of the vulva is supplied by branches of the pudendal nerve (Gould 1991). The vulvar vestibule is by definition visceral tissue, but is considered to have non-visceral innervation (Cervero 1994); thus sensations to touch, temperature and pain are similar to sensations evoked in the skin (Bohm-Starke et al. 2001b). The upper end of the vagina is part of the internal genitalia and the afferent paths from this part of the vagina go through the pelvic nerves, but probably also through somatic nerve fibres to sacral segments 3-4 (Lundberg 2001). The pudendal nerve innervates the lower end of the vagina. The exact line of demarcation between these two distributions is ill defined (Gould 1991). The distal areas of the vaginal walls have been found to have more nerve fibres than the proximal parts (Hilliges et al. 1995). Free intraepithelial nerve endings, considered to be mainly pain receptors, have only been detected in the regions of introitus vaginae and the hymenal ring (Krantz 1958).

Innervation of pelvic floor muscles

The two layers of striated pelvic floor muscles are innervated from the spinal sacral segments 2-5 (Schroder 1981; Gould 1991). The superficial layer of the pelvic floor muscles is innervated from the pudendal nerve; the levator ani muscles are innervated directly from the sacral nerves (Gould 1991).

Cutaneous sensory receptors and pain mechanisms

Sensory receptors

Sensory receptors in the skin are classified according to whether they have distinct end organs or terminate as free nerve endings. The receptors with end organs are connected to coarse A β -fibres and are associated with the perception of various aspects of touch (Vallbo and Johansson 1984). The free nerve endings include nociceptive, thermoreceptive and tactile receptors.

Nociceptors are defined as receptors responding to stimuli which may cause tissue damage. Some nociceptors respond only to intense mechanical stimuli, others to both mechanical and thermal stimuli, and still others (polymodal nociceptors) to chemical stimulation as well (Lundeberg 1995). A subgroup of C-nociceptors (“silent or sleeping nociceptors”) cannot normally be activated by mechanical or thermal stimuli. Instead, they become responsive only under pathological conditions such as inflammation (Schmelz et al. 1994; Schmidt et al. 1995).

Women with vulvar vestibulitis have significantly more intraepithelial nerve endings in the vestibulum (Weström and Willen 1998; Bohm-Starke et al. 1998; Bornstein et al. 2004), characterized as nociceptors (Bohm-Starke et al. 1999), than healthy women.

Transmission of pain

Pain from skin and mucosa is transmitted in afferent A δ - and C-fibres. The *A δ -fibres* are thin myelinated nerves, with high conduction velocity, that convey discriminative information about nociception, stimulation of which leads to the first, sharp, localized pain. The *C-fibres* are thin unmyelinated nerves, and stimulation of those gives rise to the secondary, aching, and burning pain. The region of pain conveyed by C-fibers is not strictly limited and is often referred to a larger area even if the stimulus itself is localized (Lundeberg 1995; Hansson 1998). Burning pain is mediated by mechano-insensitive C-units (Schmelz et al. 2000)

Sensitization

Where tissue damage results in inflammatory reaction, itself a part of the normal healing process, the response properties of the various components of the nociceptive system change. Nociceptors may become sensitized (peripheral sensitization), with a decrease in response threshold so that they may be activated even by innocuous stimuli (Lundeberg 1995). The increased responsiveness of the sensitized receptors plus the recruitment of previously unresponsive receptors causes a dramatic increase in the afferent inflow to the CNS and a consequent modification of responsiveness of the dorsal horn neurons (central sensitization) (Lundeberg 1995). This central sensitization is characterized by enhanced transmission of nociceptive information, sensitivity to non-nociceptive inputs and spontaneous impulse generation in the spinal cord (Stannard and Booth 2004)

Hyperalgesia

Hyperalgesia is defined as “an increased response to a stimulus which is normally painful” (Merskey and Bogduk 1994). A local cutaneous injury can produce *primary hyperalgesia*, i.e. within the injured area, and *secondary hyperalgesia*, i.e. in the normal surrounding skin. (Baumann et al. 1991; Hansson 1998), (Figure 4). Primary hyperalgesia is characterized by increased responsiveness both to heat and mechanical stimulation (Koltzenburg et al. 1992;

Torebjork et al. 1992), while secondary hyperalgesia is characterized by increased responsiveness to mechanical stimulation (Hansson 1998). In secondary hyperalgesia, central rather than peripheral sensitization accounts for the observed increased responsiveness to mechanical stimulation, and ongoing activity of primary afferents is not required for its maintenance (LaMotte et al. 1992).

Silent C-nociceptors may contribute both to primary hyperalgesia and to secondary hyperalgesia as a consequence of central sensitization (Schmidt et al. 1995)

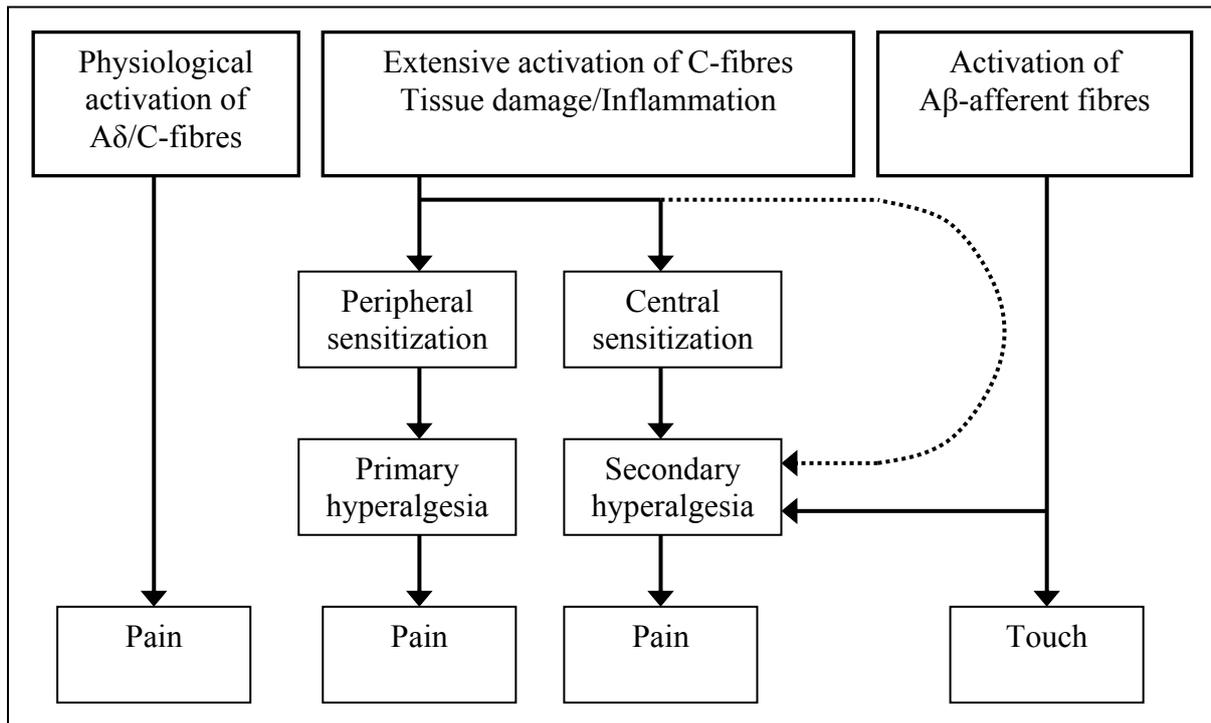


Figure 4. Schematic presentation of the distinctions between physiological pain, primary and secondary hyperalgesia/allodynia. Translated with permission from Hansson (Hansson 1998, p 47). Printed with permission of Pfizer/Pharmacia & Upjohn.

Allodynia

Allodynia is defined as “pain due to a stimulus which does not normally provoke pain” (Merskey and Bogduk 1994). Dynamic mechanical allodynia (to touch and, less frequently, to pressure) is the most common type, and is thought to be mediated by activation of Aβ-afferents (Hansson and Kinnman 1996). This secondary hyperalgesia to touch is mediated by altered central processing of input from myelinated mechanosensitive units (Torebjork et al. 1992) although this altered central processing of mechanoreceptor input also requires ongoing activity in afferent nociceptive C-fibres (Koltzenburg et al. 1992) (Figure 4).

Allodynia and hyperalgesia found in women with vulvar vestibulitis

Allodynia and hyperalgesia in women with vulvar vestibulitis have both been explained with theories of peripheral and central sensitization.

Bohm-Starke et al. (2001b) found both allodynia evoked by stretch and punctate stimuli and heat hyperalgesia in the vestibulum in women with vulvar vestibulitis. Sensory abnormalities in this group are explained by peripheral noxious mechanisms involving sensitization and/or proliferation of various types of C-nociceptors (Bohm-Starke et al. 2001b)

Pukall et al. (2002) found allodynia to light touch in the vestibular region and hyperalgesia on the adjacent labium minus in women with vulvar vestibulitis. According to their theory, peripheral changes in women with vulvar vestibulitis lead to a sensitization in the spinal cord and development of secondary hyperalgesia (Pukall et al. 2002).

By use of tender point examination, (typically used in the diagnoses of fibro muscular syndrome) women with vulvar vestibulitis have reported higher pain intensity and more unpleasant ratings than matched controls. The results suggest that pain mechanisms in women with vulvar vestibulitis may include not only those that are genital specific but also those that are more generalized and possibly centrally mediated (Pukall et al. 2006).

Muscular pain

Pain sensations in muscles are also conveyed by free nerve-ending nociceptors, thin myelinated Group III afferents (A δ -fibres) and thin unmyelinated Group IV afferents (C-fibres). The nociceptors are situated especially in the walls of the arteriole and in the surrounding connective tissue (Hansson 1998). The nociceptors have shown no response to innocuous forms of stimulation (light pressure, muscle contraction and stretching) and required noxious (tissue-threatening) stimuli to become (clearly) activated (Mense and Meyer 1985). Muscle pain is described as aching and cramping, with diffuse and referred localisation (Graven-Nielsen and Arendt-Nielsen 2002). There is also a connection between pain from the mucosa and pain from the musculature. "Pain fibres that are sensitized by mucosal inflammation initiate sensitization and contraction of the underlying musculature. Further sensitization of the muscle pain receptors may in turn, through a process of central sensitization, reduce sensory pain thresholds, resulting in a 'vicious cycle' of inflammation and additional muscle contraction" (Svensson et al. 1998; Graven-Nielsen and Arendt-Nielsen 2002) cited in Zolnoun et al. (2006).

Primary muscle dysfunction and pain: Individuals with temporomandibular disorder have a dysfunction in contraction and relaxation of the facial muscles, which is associated with chronic orofacial pain (Dworkin and LeResche 1992; Zolnoun et al. 2006). "This dysfunction has been traced to an imbalance between central inhibitory and excitatory pathways. In this case, primary abnormalities in muscle function are intimately associated with the development of pain" (Zolnoun et al. 2006).

Itch

Itch has many similarities to pain. Both are unpleasant sensory experiences, but the behavioural response patterns differ; pain elicits a withdrawal reflex, whereas itch leads to a scratch reflex. The definition of itch as "an unpleasant sensation leading to a desire to scratch" (Haffenreffer 1660) is said to be an oversimplification (Greaves and Khalifa 2004). Not all patients with itch scratch to obtain relief from itch.

A stimulus causing itch leads to two distinct responses: first, a well-localised itch at the site of the stimulus, which persists only briefly after the stimulus has been removed; and secondly, a

subsequent diffuse, poorly localised area surrounding the site, which responds with intense itch when exposed to gentle touch (alloknesis) and punctate mechanical stimuli (hyperknesis) (Yosipovitch et al. 2003; Ikoma et al. 2004; Ikoma et al. 2006). Alloknesis is a prominent feature of itch and explains the bouts of intense itching associated with sweating, sudden changes in temperature, dressing and undressing (Yosipovitch et al. 2003).

Itch might be classified into four categories on a pathophysiological basis (Twycross et al. 2003; Yosipovitch et al. 2003; Greaves and Khalifa 2004; Ikoma et al. 2006).

Pruritoceptive itch is generated in the skin due to inflammation, dryness or other skin damage and is transmitted by C nerve fibres, e.g. urticaria or insect bites.

Neuropathic itch arises because of disease located at any point along the afferent pathway, e.g. itch due to herpes zoster neuropathy, multiple sclerosis or brain tumours.

Neurogenic itch originates centrally but without evidence of neural pathology, and is of two types. It could be due to enhanced itch processing in the central nervous system, probably important in patients with atopic eczema; or it could be due to the action of circulating factors, e.g. itch in patients with cholestasis.

Psychogenic itch is poorly understood, with parasitophobia as an example.

These classes of itch are not mutually exclusive; the itch of atopic eczema might be both pruritoceptive and neurogenic.

Reports on specific, histamine-sensitive, mechano-insensitive C-receptors for itch in human skin (Schmelz et al. 1997) and on spinothalamic lamina I neurons selectively sensitive to histamine (Andrew and Craig 2001), indicate that itch is subserved by specific neural elements both peripherally and centrally (Andrew and Craig 2001). The activation of multiple sites in the brain after itch induction suggests that there is not a sole itch centre (Yosipovitch et al. 2003). Antagonism between itch and pain via the spinal cord might be bidirectional: pain inhibits itch, whereas inhibition of pain can cause itch (Nilsson et al. 1997; Yosipovitch et al. 2003; Ikoma et al. 2006; Stander and Schmelz 2006). In patients with chronic pruritus, painful stimuli might be perceived as itch due to central sensitization (Ikoma et al. 2004). Following sensitization by histamine application, central itch neurons can acquire responsiveness to mechanical stimuli. Under these conditions mechanical stimuli might be perceived as itch even if the histamine-sensitive primary afferents are mechanically insensitive (Schmelz 2001).

RESEARCH THEORIES

Here we describe our research theories in their *present* form. During the course of the work with this thesis for nearly ten years, our research theories have been disputed and redefined.

In all our studies, we used a theoretical framework of learning theory. According to learning theory, the main problem in women with both total and partial vaginismus may be characterized as a conditioned response (Schwartz 1989; Wijma and Wijma 1997; ter Kuile et al. 2006; van Lankveld et al. 2006) reinforced by avoidance (phobic) behaviour and flight reactions, e.g. interrupted attempt of intercourse. Pain during penetration and after intercourse in women with partial vaginismus can be explained when regarding the partial vaginismus reflex as a conditioned response. The penetration situation, associated with pain or fear, has become a conditioned stimulus, and elicits the conditioned response, i.e. a vaginismus reflex and a feeling of burning pain. As pain in itself is a stimulus for the vaginismus reflex, a vicious circle is easily created. Generalized unprovoked vulvodynia could also be explained

by theories of conditioning. Thoughts of the problem act as a conditioned stimulus which evoke the conditioned responses: a vaginistic reflex and burning vulvar pain. In this situation the vaginismus reflex clinically appears as hypertonicity of the pelvic floor muscles, which is referred to as persistent involuntary spasm in DSM-IV-TR (American Psychiatric Association 2000). Also itch during penetration and after intercourse in women with partial vaginismus might be explained as a conditioned response to penetration (Wijma et al. 2000).

A main theory in this thesis was that vaginismus exists in two main forms, partial vaginismus and total vaginismus. No results in our studies contradict this theory. We have therefore chosen a model for vaginismus where vaginismus appears in two main forms, partial vaginismus (PaV) and total vaginismus (TV), and further into primary (P) and secondary (S) vaginismus (PaPV, PaSV, TPV, and TSV). According to our clinical experience, vaginismus often has a progressive course. The individual woman's diagnosis may therefore depend on at what stage a diagnostic evaluation is performed. When the problem starts, the woman might be diagnosed as PaSV or PaPV, which, during the course of time, may aggravate to TSV (Figure 5). Vaginismus can, however, also start as partial and remain partial, or start as total.

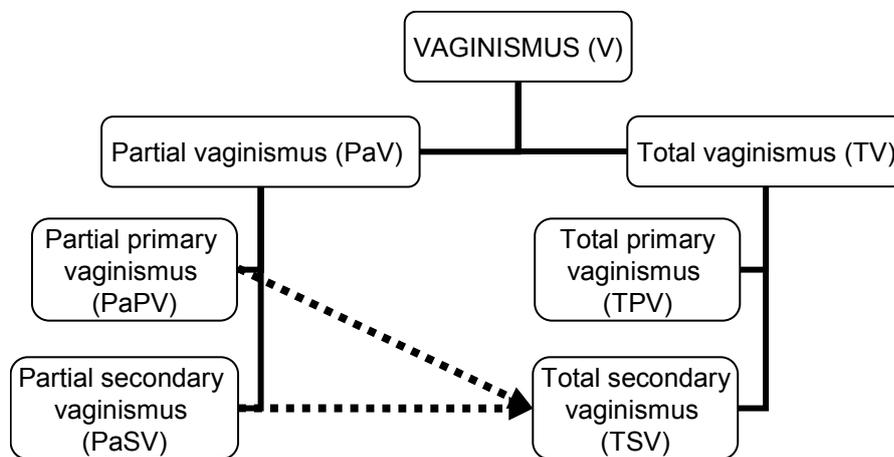


Figure 5. Forms of vaginismus. (Wijma et al. 2007) Reprinted with permission from Parthenon Publishing.

We found that many women could be diagnosed as having both vaginismus and vulvar vestibulitis, and we found no woman with vulvar vestibulitis without vaginismus. Therefore, we have chosen to use the term total/partial vaginismus with or without vulvar vestibulitis.

Our theory for the association between partial vaginismus and vulvar vestibulitis is that, at least in some women, the partial vaginismus reflex comes first, and repeated penetrations or attempts at penetration in an unaroused state with a partly closed vagina lead to the findings and symptoms seen in women with vulvar vestibulitis. The pathophysiology of burning pain in relation to partial vaginismus is not known, as no studies of this subject exist. We assume that pain in relation to the partial vaginismus reflex is the primary pain process in women with partial vaginismus and vulvar vestibulitis, and that symptoms of concomitant vulvar vestibulitis might be explained both by theories of peripheral and central sensitization causing primary and secondary hyperalgesia and allodynia.

AIMS AND RESEARCH QUESTIONS

The main purposes of this study were (1) to develop a useful definition of partial vaginismus for both clinical and scientific purposes; (2) to describe the prevalence of partial vaginismus among women with a diagnosis of superficial coital pain; and (3) to report on symptoms and clinical findings in, and treatment results for, women with partial vaginismus.

Specific aims of Study I-V

- To illustrate some of the theories on which this thesis is based by means of a case report on a woman with both vulvar vestibulitis and vaginismus (Study I).
- To investigate to what extent women with superficial coital pain can be diagnosed with both vaginismus (partial and total) and vulvar vestibulitis (Study II).
- To measure to what extent sEMG of the pelvic floor muscles can distinguish between women with partial vaginismus solely, women with partial vaginismus and vulvar vestibulitis, and asymptomatic women (Study II).
- To measure burning pain and itch sensations as to reported frequency, intensity, and temporal pattern during a “standardized penetration situation” in women with partial vaginismus with or without vulvar vestibulitis and in asymptomatic women (Study III).
- To describe early symptoms and development over time of superficial coital pain in women with partial vaginismus with or without vulvar vestibulitis (Study IV).
- To present long-term follow-up results in a sample of women with superficial coital pain and vaginismus treated with cognitive behaviour therapy (Study V).

Additional aims of the thesis

- To analyse the distribution of women with a diagnosis of vulvar vestibulitis among women with partial vaginismus and total vaginismus in Study II, III, IV, and V.
- To analyse the distribution of women with partial vaginismus and total vaginismus among women with vaginismus in Study II and V.
- To analyse the correspondence between our criteria of the various forms of vaginismus and those in the existing diagnostic systems concerning vaginismus, dyspareunia, vulvar vestibulitis, and provoked vestibulodynia.

Research questions

Study II

What proportion of women with superficial coital pain fulfil both the diagnostic criteria of vaginismus according to DSM-IV (American Psychiatric Association 2000), of partial vaginismus according to Wijma et al. (Wijma and Wijma 1997; Wijma et al. 2000), and of vulvar vestibulitis set up by Friedrich (Friedrich 1987)?

Do women with partial vaginismus with or without vulvar vestibulitis relax and contract their pelvic floor muscles differently compared to women without these problems, as registered in sEMG readings from these muscles?

What is the test-retest reliability of sEMG of the pelvic floor muscles?

Study III

Do women with partial vaginismus with or without vulvar vestibulitis differ in reporting burning pain and itch during a standardized penetration situation, compared to asymptomatic women?

To what extent do women with partial vaginismus with or without vulvar vestibulitis, who experience both burning pain and itch during a standardized penetration situation, report that burning pain precedes itch?

Study IV

How do women with partial vaginismus with or without vulvar vestibulitis describe pain during penetration and after intercourse: when the problem starts; when the women cease having intercourse; in terms of intensity of reported pain symptoms; and choice of words to describe the pain and duration of pain?

Is pain after intercourse in women with partial vaginismus with or without vulvar vestibulitis a more common symptom at onset of the problem than pain during penetration?

Do women with partial vaginismus with or without vulvar vestibulitis have vulvar symptoms in everyday life, and what kind of symptoms do they describe?

Do women with partial vaginismus with or without vulvar vestibulitis have symptoms of pain at micturition after intercourse, and how is this symptom described?

Study V

What are the long-term treatment results for women with superficial coital pain and vaginismus, treated with a CBT-program for vaginismus, with regard to ability to have intercourse without pain and ability to enjoy intercourse?

Is an ability to have intercourse at end of therapy, among women with superficial coital pain and vaginismus treated with a CBT program for vaginismus, maintained at follow-up?

To what extent do women with superficial coital pain and vaginismus, treated with a CBT-program for vaginismus, report fulfillment of treatment goals?

Is there an increase in reported self-worth as sex partner, woman and human being from assessment to follow-up, among women with superficial coital pain and vaginismus treated with a CBT-program for vaginismus?

Do women with superficial coital pain and vaginismus heal while on waiting list?

METHODS

Measures

Evaluation of the diagnoses of vaginismus and vulvar vestibulitis (Study II)

In Study II a thorough diagnostic evaluation was performed in women referred to our department and diagnoses were set according to: (1) DSM-IV-TR (American Psychiatric Association 2000) criteria of vaginismus, including recurrent or persistent, primary and secondary vaginismus, mild contraction inducing some tightness and discomfort, severe contraction preventing penetration; and (2) Freidrich's definition of vulvar vestibulitis (Friedrich 1987) (third criterion of vestibular erythema excluded). We also used described study criteria of partial and total vaginismus which both correspond with the criteria of vaginismus in DSM-IV-TR and cited above.

Use of sEMG of pelvic floor muscles (Study II and III)

In Study II we used sEMG for assessment of resting level, stability, contraction and relaxation in pelvic floor muscles.

In Study III we used the "sEMG of pelvic floor muscles procedure" merely as a standardized penetration situation for measurement of itch, burning pain and other sensations during penetration.

Both studies are based on measures using the following equipment:

- MyoTrac 3, sEMG signal processing unit
- MyoTrac 3 Continence Software, Version 1.2.1
- Perianal surface single EMG electrodes T3404
- Uni-gel reference electrode T3425
- Vaginal surface EMG sensor T6050, ($\varnothing=25.5\text{mm}$), called "the bigger vaginal sensor"
- Rectal surface EMG sensor T6051, ($\varnothing=17\text{mm}$), called "the smaller vaginal sensor" (Thought Technology, Montreal, Quebec, Canada).



Figure 6. Vaginal and rectal sEMG sensors. Reprinted with permission of Thought Technology.

The vaginal sEMG sensor consists of bipolar longitudinal electrode plates mounted in a plastic vaginal probe (Figure 6), and was of the same model as those used in three studies of women with vulvar vestibulitis (White et al. 1997; Glazer et al. 1999; Romanzi et al. 1999) and later in one study of women with vaginismus (Reissing et al. 2004).

The signals from the sensor were recorded at 20 samples per second. The device actually samples the EMG activity 2000 times per second. A RMS (root mean square) calculation is applied to create the recording level of 20 samples/second. The channel bandwidth was 20-450 Hz for both channels.

Surface electromyography of pelvic floor muscles (Study II)

For the sEMG measurements in Study II, we used Protocol No 01 Pelvic Muscle Assessment, “the Glazer protocol” (Glazer et al. 1998). The protocol consists of: (1) A one-minute, pre-test baseline assessment, evaluating the resting level and the resting stability of the pelvic floor. The resting stability is measured as a mean of the standard deviations of the pre-test baseline sEMG registrations; (2) Five rapid contractions “spikes”, each separated by a resting period of 10 seconds, evaluating the fast-twitch muscle fibres; (3) Five tonic contractions of 10 seconds duration with resting periods of 10 seconds in between, evaluating the amplitude of the tonic contractions performed by slow-twitch muscle fibres, and the woman’s ability to change between contraction and relaxation; (4) One maximal endurance pelvic floor contraction of 60 seconds; (5) A one-minute, post-test baseline registration, measuring resting level and the resting stability after the contractions, as in the pre-test baseline assessment.

In eight of 47 symptomatic women (see page 29 for description of sample) we used “the smaller vaginal sensor” (as penetrator into the vagina) due to those women’s pronounced pain reaction when having a pelvic examination with two fingers in the vagina some weeks earlier (burning pain > 5 on a scale from 0 to 10 where 0 = “no pain” and 10 = “the most intense pain she could imagine”).

Measurement of itch and burning pain (Study III)

In Study III we used the surface electromyography (sEMG) of the pelvic floor muscles procedure (Glazer et al. 1998) as a standardized penetration situation for the following reasons: (1) a defined sensor was “penetrator”; (2) a defined protocol directed the insertion and extraction procedures of the sensor and moments of contraction/relaxation of the pelvic floor muscle; (3) no pressure was applied against the vaginal walls except for that of the sensor.

We used a Numerical Rating Scale (Jensen and Karoly 2001) to let the women describe the intensity of their symptoms. The participants were asked at 20 standardized moments to report the intensity of burning pain, other types of pain, itch, sensations of discomfort, fear, and sensation of a need to urinate separately by saying aloud a number between 0 and 10 (0 = “no sensation”, 10 = “the most intense sensation the woman could imagine”). We did not use written list of possible sensations. The researcher (ME) noted the women’s estimates of different sensations by use of symbols in a protocol.

In this study, we used “the smaller vaginal sensor” in seven of 53 symptomatic women (see page 29 for description of sample) for the same reasons mentioned above.

The investigation was divided into four sections: (A) Perianal measurement (400 sec); (B) Insertion of the sensor (150 sec); (C) Vaginal measurement (400 sec); and (D) Extraction of the sensor (150 sec). During the two parts of the study when the women contracted and relaxed their pelvic floor muscles (section A, perianal measurement (without any inserted vaginal sensor), and section C, vaginal measurement (with inserted vaginal sensor)) we used Protocol 1, Pelvic Floor Muscle Assessment according to the “Glazer protocol” (Glazer et al. 1998) described above. During insertion of the sensor into the vagina (section B) and during

extraction of the sensor from the vagina (section D), we used Protocol 3, Open display, where no contraction or relaxation of the pelvic floor muscles took place.

Pain development (Study IV)

In Study IV we were interested in pain development in women with superficial coital pain. We used a semi-structured telephone interview. The questions were organized into two different sections: (A) symptoms at the onset of the problem; and (B) symptoms when the women ceased having intercourse. In each section, the women were asked to report the intensity and duration of genital pain, itch, and any other symptom, at two different time periods of intercourse, i.e. during penetration and after intercourse. They were also asked to report the intensity of pain at micturition after intercourse, and the intensity of sensations of genital pain, itch and any other symptoms in everyday life.

We used a Numerical Rating Scale (Jensen and Karoly 2001) to let the women describe the intensity of their symptoms, by choosing a figure from 0–10, where 0 = “no pain/symptom”, and 10 = “the most intense pain/symptom the women could imagine”.

Further, the women were asked to describe in their own words the different types of pain they had experienced, during penetration and after intercourse. We did not use a list of possible sensations by which they could describe the pain. Women who had ceased having intercourse were asked for the reason/reasons for that decision.

Long-term follow-up results (Study V)

In Study V we used a postal questionnaire for the study of long-term follow-up results. This *treatment evaluation questionnaire* included 32 questions focusing on the situation during the previous two months concerning partnership, attempted intercourse, intercourse, enjoyment of intercourse, effects of therapy, and goal fulfillment. The questionnaire also included questions of self-worth as sex partner, woman and human being at assessment and at follow-up.

The CBT program focused on reinforcement of abilities rather than focusing on existing problems. Questions of pain during intercourse at follow-up were therefore measured as ability to have intercourse without pain.

The women estimated their *ability to have intercourse without pain* at follow-up on a scale from 0-10 (0 = “no ability at all to have intercourse without pain”; 10 = “total ability to have intercourse without pain”).

Enjoyment of intercourse was measured as *ability to enjoy intercourse*, estimated on a scale from 0-10 (0 = “no ability at all to enjoy intercourse”; 10 = “total ability to enjoy intercourse”). An estimate of ≥ 6 was called “ability to enjoy intercourse”.

Fulfillment of treatment goals at follow-up were estimated by the women on a scale from 0-10 (0 = “no fulfillment of treatment goal”; 10 = “total fulfillment of treatment goal”).

Self-worth as sex partner and as woman and human being was estimated before assessment and at follow-up (i.e. partly a retrospective evaluation) on a scale of 0–10 (0 = “lowest possible self-worth”, and 10 = “highest possible self-worth”).

The women were also asked if their problems of vaginismus and superficial coital pain from end of therapy to follow-up had decreased, increased, or if there were unchanged problems.

Example of questions of goal fulfilment

Questions of fulfilment of treatment goals. These questions include various treatment goals that women often express before treatment. Mark to what extent you have reached those goals that you had before you started treatment at our department.

Treatment goal "to have intercourse without pain"

- This was not one of my treatment goals
- This was one of my treatment goals (mark to what extent you have reached the goal today by putting a circle round the figure which best corresponds to how you feel).

0 ----- 1 ----- 2 ----- 3 ----- 4 ----- 5 ----- 6 ----- 7 ----- 8 ----- 9 ----- 10

Goal not
fulfilled at all

Goal totally
fulfilled

Example of questions of self-worth

How do you estimate your self-worth as human being before treatment/contact and today?

Answer by putting a circle round the figure which best corresponds to how you estimate your self-worth as **human being before treatment.**

0 ----- 1 ----- 2 ----- 3 ----- 4 ----- 5 ----- 6 ----- 7 ----- 8 ----- 9 ----- 10

Lowest
possible self-worth

Highest
possible self-worth

Answer by putting a circle round the figure which best corresponds to how you estimate your self-worth as **human being today.**

0 ----- 1 ----- 2 ----- 3 ----- 4 ----- 5 ----- 6 ----- 7 ----- 8 ----- 9 ----- 10

Lowest
possible self-worth

Highest
possible self-worth

Procedures and samples

Case report(Study I)

This study consisted of a detailed case report.

Diagnostic evaluation (Study II)

Sample: The sample consisted of all women (n=224) who were consecutively referred for superficial coital pain to the Division of Psychosocial Obstetrics and Gynaecology, Department of Obstetrics and Gynaecology, University Hospital, Linköping, Sweden from January 1999 to March 2002.

Procedure: The diagnostic evaluations of women with superficial coital pain were performed by ME and BW and included: (1) a brief interview about past medical/psychological history, medications, and obstetric/gynaecological history; (2) a thorough interview concerning the intercourse-related pain; (3) a gynaecological examination including inspection of vulva, cotton-swab examination of labia minora and the vestibular area, degree and extension of

vestibular erythema, vaginismus reflex and associated burning pain, ability to contract and relax the pelvic floor muscles, and resting pelvic muscle tone.

Diagnoses of vulvar vestibulitis, partial vaginismus, total vaginismus and other diagnoses and/or a combination of those diagnoses were set by the examiner according to definitions of vaginismus in DSM-IV-TR (American Psychiatric Association 2000) and study criteria of partial and total vaginismus, Friedrich's definition of vulvar vestibulitis (Friedrich 1987) (third criterion of vestibular erythema excluded). The two examiners (ME and BW) discussed all diagnoses which were set until agreement was achieved.

Diagnostic evaluation in the comprehensive summary

Sample: Eligible for the study were all women who had attended our clinic for superficial coital pain and had been included in any of the studies of this thesis. Women with partial vaginismus were included in four studies (II, III, IV, and V), and women with total vaginismus in two studies (II and V).

Procedure: We analysed Studies II, III, IV, and V for the distribution of women with a diagnosis of vulvar vestibulitis among women with partial vaginismus and total vaginismus, and for the distribution of the diagnosis of partial vaginismus and total vaginismus among women with vaginismus.

Analysis of the correspondence between our study criteria and existing diagnostic criteria

Procedure and sample: Our study criteria of partial and total vaginismus (see Introduction page 2) were compared to the diagnostic criteria of vaginismus and dyspareunia in DSM-IV-TR (American Psychiatric Association, 2000) and ICD-10 (World Health Organisation 2003), and to Friedrich's criteria of vulvar vestibulitis (Friedrich 1987), and to ISSVD's criteria of provoked vestibulodynia (Moyal-Barracco and Lynch 2004).

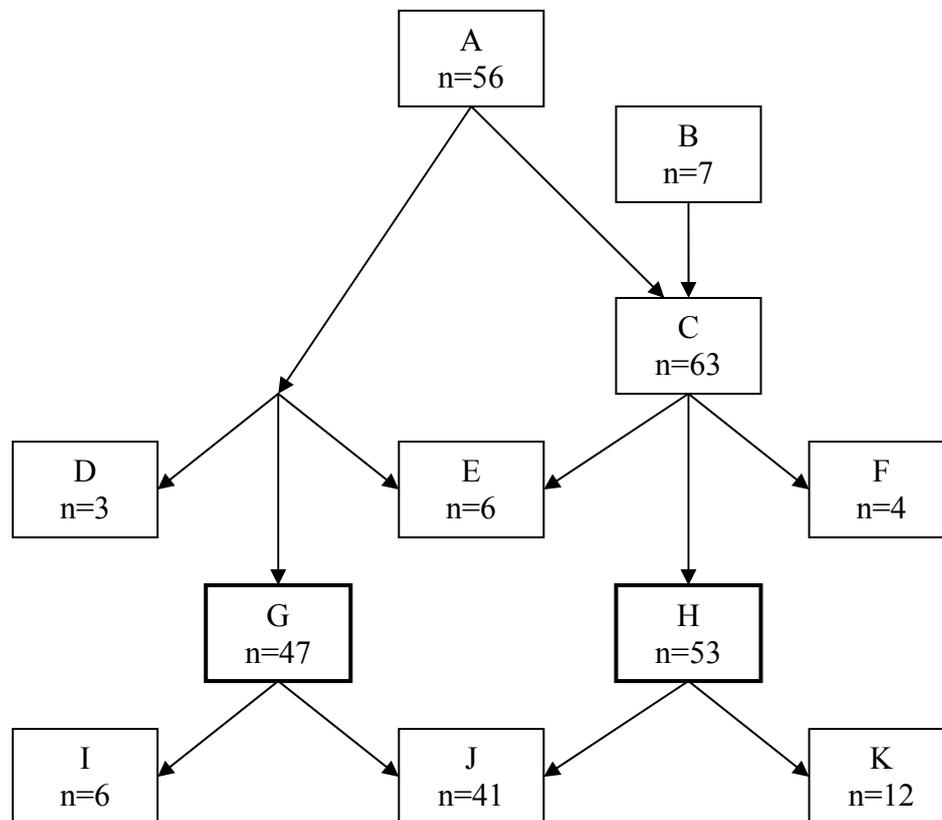
sEMG of pelvic floor muscles (Study II) and measurement of itch and burning pain (Study III)

Symptomatic sample Study II and III: Sixty-three consecutive women with partial vaginismus were included in any investigation of Study II or III. Forty-seven women were included in the sEMG measurement of pelvic floor muscle function in Study II (Figure 7). Fifty-three women took part in Study III (Figure 7) where we used sEMG of pelvic floor muscles as a standardized penetration situation for measuring of itch and burning pain during penetration. Forty-one women were included in both studies (Figure 7). All symptomatic women included in both Study II and III were investigated only once. sEMG measurements and registration of reported levels of burning pain and itch were carried out during the same investigation.

Women with reports of or clinical signs (clinical examination and analyses of wet smear) of ongoing genital infection, especially Candida infections or Bacterial vaginosis were regarded as not eligible for the study. One woman who at assessment session 1 and 2 did not report symptoms of or have clinical findings of Candida infection was initially included in Studies II and III. This woman was afterwards excluded as she later, at assessment session 3, reported such symptoms and had clinical signs of Candida infection.

Women with superficial coital pain mainly caused by cutaneous disorders of the vulva (e.g. lichen sclerosus), (n=41), were not included in Studies II and III.

The mean age of the symptomatic women was 26.9 (Study II) and 24.5 years (Study III); range 18-60 (both studies). One woman included in Studies II and III was 60 years old. This woman was treated with local Estradiol at the moment of the sEMG investigation and the assessments of burning pain and itch. Mean duration of problems was 3.8 (Study II) and 4.2 years (Study III). Eight women in Study II and seven in Study III were parous.



- A Women included in the sEMG (Study II) and itch and burning pain study (Study III)
- B Additional women included in the itch and burning pain study only (Study III)
- C Women included in the itch and burning pain study (Study III)
- D Women excluded from the sEMG study only, due to artefacts in the registrations
- E Women excluded from both studies, due to artefacts in the registrations and incomplete records
- F Women excluded from the itch and burning pain study only, due to incomplete records
- G Study sample sEMG study (Study II)
- H Study sample itch and burning pain study (Study III)
- I Sample of women included in the sEMG study only
- J Sample of women included in both studies
- K Sample of women included in the itch and burning pain study only

Figure 7. Sampling procedure (symptomatic sample) in Studies II and III.

Asymptomatic sample Study II and III: We used the same asymptomatic sample in Studies II and III. The asymptomatic group consisted of 27 women (mean age 27.1, range 20-37 years) without pain during coitus who responded to an invitation in the local press. Exclusion criteria were: (1) having given birth; (2) ongoing pregnancy; (3) post-menopause; (4) dyspareunia;

(5) urinary incontinence; (6) urinary urgency; (7) present urinary infection; (8) present genital infection; (9) cysto- or rectocele; (10) protracted constipation, and (11) pain from other reproductive organs. The exclusion criteria in the asymptomatic group were based on assumptions of factors that might influence the pelvic muscle function, and before we knew the sample characteristics of the symptomatic group. Twenty-six of 27 women in the asymptomatic group were investigated twice with sEMG of pelvic floor muscles (time interval 2-4 weeks). They were instructed not to practise pelvic floor exercises in the interval between the registrations.

Procedure Study II: Study participants in Study II were investigated in the lithotomy position in a pelvic examination chair. We never examined the pelvic floor muscles digitally at the sEMG registration visit. Perianal surface-single EMG electrodes were placed at 2 and 10 o'clock nearby the anus. First we instructed all women how to properly perform the quick and tonic pelvic contractions, using the Open Display. This was not a part of the registration. We avoided repeated instructions, since this might have influenced the results. After the instruction part the women were not allowed to look at the screen during the investigation. Protocol No 01 Pelvic Muscle Assessment was used twice for each investigation. First we used only perianal registration (Channel 1). After the insertion of the vaginal sEMG sensor, for technical reasons we did change the perianal registration to Channel 2 and both perianal (Channel 2) and vaginal (Channel 1) registrations were collected. We also used the perianal sensors to register the muscular activity during the insertion and extraction of the vaginal sensor (Channel 1, Open Display). Only vaginal registrations are reported. The activity of surrounding muscle groups was not investigated.

Procedure Study III: Assessments of burning pain, itch and other sensations in Study III were carried out with study participants in lithotomy position in the pelvic examination chair; see procedure study II. ME, who also instructed the participants how to estimate burning pain, itch and other sensations, made all the investigations. The investigator was aware of the status of the participants (vaginismus or control).

Perianal surface self-adhesive single EMG electrodes were placed at 2 and 10 o'clock near the anus. The vaginal sensor was inserted by the investigator.

Pain development (Study IV)

Sample: Participants in the pain development study were a sample of 35 women with partial vaginismus with or without vulvar vestibulitis, who had applied for help because of superficial coital pain, at three Swedish departments of obstetrics and gynaecology from November 2004 to June 2005. The women were invited by a written letter to participate in a semi-structured telephone interview concerning the development of their pain during intercourse. Twenty-seven women agreed to participate (Linköping University Hospital, n=10, Västervik Hospital, n=13; and Sundsvall Hospital, n=4). At the time of the interview, median age was 23.2 years, and duration of symptoms nearly four years.

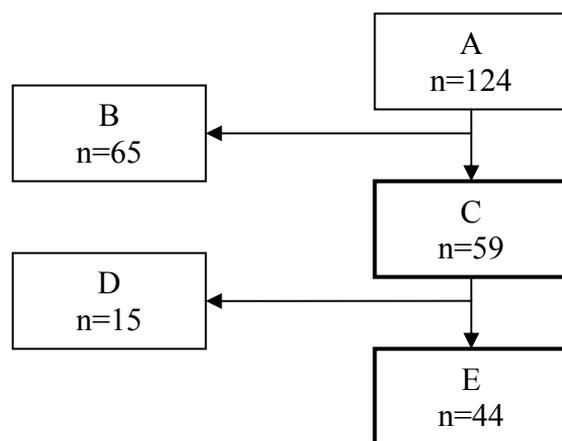
Procedure: All interviews were performed by ME and had a duration of 1-1.5 hours. During the interview, notes were taken on two pre-designed forms. Three women were excluded after the interview, due to insufficient memories of early pain development (response rate 68.6%).

Long-term follow-up results (Study V)

Assessment sample: The assessment sample consisted of 124 women who had consecutively attended the Division of Psychosocial Obstetrics and Gynecology, Linköping University Hospital, from November 1996 to February 2004, for superficial coital pain and had been diagnosed as having vaginismus. All included women had finished their contact with the clinic >1 year ago. We excluded women with vaginismus in whom vaginismus was not the dominating problem, e.g., who suffered mainly from experiences of sexual abuse (n=4) or fear of the pelvic examination (n=10).

Assessments and information given at baseline: All 124 women in the assessment sample had met one of the two therapists in the research team (ME and BW) for one or two assessment sessions. The gynecological assessment included inspection, grading of tenderness to pressure within the vestibule, estimation of tension of the pelvic floor muscles, and presence or not of an involuntary vaginistic reflex. A sexual, psychiatric and abuse history was taken. We made an analysis of the debut and development of symptoms, and possible variables contributing to the origin and maintenance of symptoms.

A treatment rationale was given, and the women were informed of theories about fear (and expectations) of pain as maintaining factors of vaginismus (Wijma and Wijma 1997), and of the importance of eliminating flight and/or avoidance behavior (response prevention). At the end of the assessment, the women were asked to express which aims they wanted to reach during therapy. We gave no explicit advice to the women to refrain from coitus during the time on waiting list.



- A Women with superficial coital pain and vaginismus who finished assessments
- B Women assessed but not eligible for the CBT program
- C Women treated with CBT
- D Non-respondents on follow-up evaluation questionnaire (n=12) and incomplete records (n=3); called non-respondents
- E Respondents on follow-up evaluation questionnaire (=index cases)

Figure 8. Sampling procedure of respondents at follow-up.

Follow-up sample: Among those 59 women who had been treated with CBT according to our study protocol the response rate was 44/59 (74.6%) (Figure 8). Another 65 women had been assessed but were not eligible for the CBT program for the following reasons: 15 women had

healed without treatment, 26 women had other reasons for no treatment (moved, not interested or not enough time, unclear reasons), and 24 women were for practical reasons (e.g. limit of time by the research team) treated by a gynaecologist not involved in the study and not according to the study protocol. The selection procedure to the two treatment options was not in the hand of the research team.

Mean age of the respondents was 25.3 years and duration of symptoms 3.8 years.

Procedure: To women who had finished treatment with CBT more than one year ago we sent a treatment evaluation questionnaire, a letter where we explained the aim of the study, and a pre-paid reply envelope. To those who did not return the questionnaires we sent one reminder.

Intervention methods

Cognitive behaviour therapy (Study V)

Treatment consisted of cognitive behaviour therapy following the guidelines for treatment of vaginismus, described in detail by Wijma and Wijma (1997) with sessions weekly or every other week; using a form of systematic desensitization by stepwise exposure to the penetration situation in a context characterized by total relaxation of the pelvic floor muscles, calmness, and the woman's total control of the situation. The women first learnt how to consciously contract and totally relax their pelvic floor muscles. Thereafter the training was performed with the pelvic floor muscles totally relaxed, in which state neither the reflex nor the pain was appearing. The hierarchy of "reflex provoking situations" were: touching the vestibulum closer and closer to the introitus vaginae, penetration with one, and then two, of the woman's own fingers; partner's one, then two fingers, and last partner's penis (technical intercourse). All training should ideally be performed in such a way that the reflex was not elicited. The women received oral and written instructions for daily homework and the progress of the homework assignments was registered in the Vaginistic Behavior Scale (VABESC) (Wijma and Wijma 1997). In VABESC the woman registers what training she performed and estimates the intensity of e.g. the fear and pain she felt before, during and after training by choosing a figure between 0 and 10. An ideal training session should be reported in VABESC by means of only 0:s. Educational elements became a major component, as the patient and her partner became the true therapists, which is why the patient needed to have fully integrated the rationale for the treatment (Wijma and Wijma 1997). During the course of therapy, the couples were encouraged to refrain from intercourse, until they could have technical intercourse as a natural step of the systematic desensitization procedure.

We regarded therapy as completed when the woman's individual goal of treatment was fulfilled. In most cases the women reached fulfillment of their individual treatment goals when they were able to have technical intercourse. Technical intercourse was therefore used as a comprehensive term for measurement of treatment results at end of therapy.

Therapists in the study were ME and BW.

Ethics

The Ethics Committee at the Faculty of Health Sciences, Linköping University, approved Studies II, III, and V. Study IV was regarded as a clinical assessment and therefore needed no approval by an ethics committee. The participants gave their informed consent to participate in Study I-V.

The most difficult ethical dilemma was to perform the sEMG part of Study II and Study III, where as part of the investigation we induced pain and itch in women with coital pain. Inducement of pain/itch in women with vaginismus might reinforce the problem.

At that stage of the investigation, sEMG biofeedback in combination with cognitive behaviour therapy was part of our investigation and treatment program for women with vaginismus, and only women who were interested in participating in that program were included in the studies. Women with total vaginismus (n=29) were excluded from the studies, as we found that sEMG investigation in this group produced so much pain and fear that we found it unethical to perform such an investigation. These women were treated with cognitive behavioural therapy alone. In women with partial vaginismus, who had had a pronounced pain reaction during a pelvic examination with two fingers in the vagina some weeks before the sEMG investigation, we used “the smaller vaginal sensor” as penetrator into the vagina, to make an sEMG investigation of the pelvic floor muscles possible.

Statistics

Data were analyzed using the Statistical Package of Social Sciences (SPSS) version 10.0, 11.5 or 14.0. As level of statistical significance we used $p < 0.05$.

Study II

In study II parametric tests were used. Descriptive methods were used for analysis of the diagnostic classification. We used analysis of variance (ANOVA) for comparing the three different study groups, women with only partial vaginismus, women with vulvar vestibulitis and partial vaginismus, and asymptomatic women. T-test was used for comparing the results in the symptomatic group (women with partial vaginismus + vulvar vestibulitis, and women with only partial vaginismus) and in the asymptomatic group. Pearson’s correlation coefficient was used for analysing test-retest reliability of sEMG measurements of pelvic floor muscles in the asymptomatic group.

Study III

The burning pain/itch scale readings were analyzed as categorical data and any report (≥ 1) of burning pain and itch was regarded as a positive finding. We used the Mann-Whitney U-Test to test differences of burning pain and itch between: (1) partial vaginismus group and asymptomatic group; (2) group with only partial vaginismus and group with partial vaginismus and vulvar vestibulitis; and (3) women investigated with the smaller or the bigger vaginal sensor. Analysis 1 and 2 were repeated excluding parous women and excluding women more than 30 years old. We used descriptive statistics for calculation of median time from the start of burning pain to the start of itch.

Study IV

In Study IV, the pain rating data were analyzed with non-parametric tests. We used the Wilcoxon Signed Ranks Test for comparison of intensity of pain during penetration and after intercourse when the problem started and when the women ceased having intercourse. We used a McNemar test for calculating the difference between the number of women reporting pain at those two moments. At onset of the problem, we were interested in early and minimal

symptoms of pain. The women's pain ratings were at this moment dichotomized into no pain or pain ≥ 1 . When the women ceased having intercourse, we wanted to get hold of those severe symptoms that had forced the women to stop having intercourse. The women's pain ratings were at this moment dichotomized into pain < 5 or pain ≥ 5 . We used the McNemar test for calculation of use of words to describe the pain during penetration and after intercourse. Descriptive statistics were used to describe sample, pain intensity, and pain duration.

Study V

For sample characteristics, we used mean values for age, duration of symptoms and follow-up time, but median values for number of sessions as this distribution was skewed. We used descriptive analyses for calculation of treatment outcome and for fulfillment of treatment goals in the index cases. We used Pearson Chi square to test a possible difference of: 1) the outcome variables between women with total vaginismus and partial vaginismus; 2) the outcome variables between women with only vaginismus and women with vaginismus and vulvar vestibulitis; and 3) ability to have technical intercourse at end of therapy among the respondents and non-respondents on the follow up questionnaire. Difference between reports of self-worth at assessment and at follow-up were analyzed with Wilcoxon's Rank Sum test.

RESULTS

Case report of a woman with vaginismus and vulva vestibulitis (I).

In our case report in Study I, we present a young woman with history, symptoms and objective findings typical of vaginismus that started as partial secondary vaginismus and developed into total secondary vaginismus with the characteristics of a specific phobia. At onset of symptoms, the woman had burning pain after intercourse. Later on she also developed pain during penetration, and when the problem had existed for 1.5 years, her partner was no longer able to penetrate. At this moment, she also suffered from continuous burning pain and itching in the vestibule (generalized unprovoked vulvodynia). Earlier, during the course of the problem, she had received a diagnosis of vulvar vestibulitis. During an assessment with vaginal manometry, a vaginismus reflex with clear increase in burning pain could be provoked by imagined penetration. The patient was treated with cognitive behaviour therapy developed for vaginismus (Wijma and Wijma 1997). During the course of the therapy, the woman got symptoms of burning pain and itch in relation to the training sessions. The burning pain sensations always preceded the sensations of itch. At the end of therapy, the patient had no burning pain or itch, neither in her daily living nor after the training sessions. At three-months follow-up, the couple could have technical intercourse without pain.

Overlap of diagnoses of vaginismus and vulvar vestibulitis (I, II, III, IV, V)

Summary: A concomitant diagnosis of partial vaginismus and vulvar vestibulitis was a common finding in our studies and varied in the different studies from 57 to 79% among women with partial vaginismus.

In Study II, 224 consecutive women with superficial coital pain were evaluated for the diagnoses of both vaginismus and vulvar vestibulitis. Of those, 39 (17.4%) had total vaginismus, 144 (64.3%) had partial vaginismus, and 41 (18.3%) had other diagnoses. One hundred and six women had vulvar vestibulitis, of whom 102 had partial vaginismus and four had total vaginismus. No woman had vulvar vestibulitis without vaginismus, but 71% of women with partial vaginismus also had vulvar vestibulitis (Figure 9 and Table II).

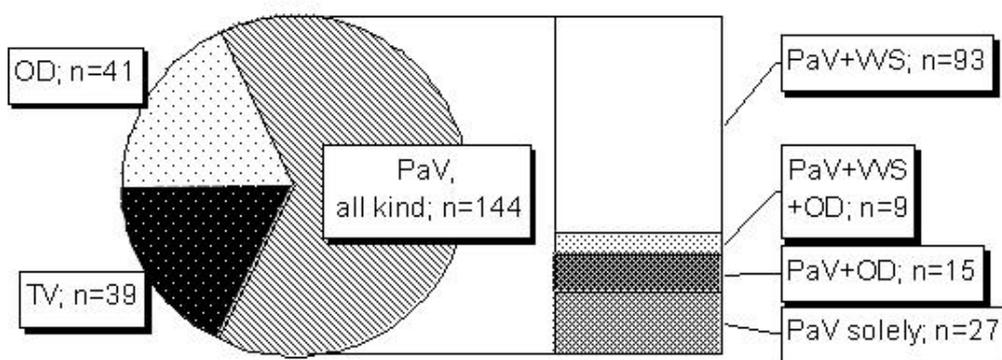


Figure 9. Number of women with partial vaginismus (PaV), total vaginismus (TV), and other diagnoses (OD) among 224 women with superficial coital pain. Women with PaV are divided into subgroups. Reprinted with permission of Parthenon Publishing.

Women with partial vaginismus were included in four of our studies (II, III, IV, and V). In all groups, the women were consecutively attending our clinic for superficial coital pain. The proportions of women with vulvar vestibulitis among women with partial vaginismus, varied in these studies from 57 to 79% (Table I).

Table I. Proportions of women with vulvar vestibulitis among women with partial vaginismus

	Study II^a		Study II^b		Study III		Study IV		Study V^c	
	n=144		n=47		n=53		n=24		n=35	
	N	%	n	%	n	%	n	%	n	%
Diagnoses										
Partial vaginismus and vulvar vestibulitis	102*	70.8	33	70.2	37	69.8	19	79.2	20	57.1
Partial vaginismus solely	42 [†]	29.2	14	29.8	16	30.2	5	20.8	15	42.9

* partial vaginismus and vulvar vestibulitis and partial vaginismus and vulvar vestibulitis and other diagnoses

[†] partial vaginismus solely and partial vaginismus and other diagnoses

a = women with partial vaginismus in the total sample of women with superficial coital pain

b = women with partial vaginismus in the sEMG group

c = women with partial vaginismus among index cases

Defined groups of women with total vaginismus were included in two of our studies (II and V). The proportions of women with vulvar vestibulitis among women with total vaginismus in these studies were 10 and 11% (Table II).

Table II. Proportions of women with vulvar vestibulitis among women with total vaginismus

	Study II^a		Study V^b	
	n=39		n=9	
	n	%	n	%
Diagnoses				
Total vaginismus and vulvar vestibulitis	4	10.3	1	11.1
Total vaginismus solely	35	89.7	8	88.9

a = women with total vaginismus in the total sample of women with superficial coital pain

b = women with total vaginismus among index cases

Proportions of women with partial vaginismus among women suffering from vaginismus (II, V)

Partial vaginismus was much more common in our studies (II, V) than total vaginismus; with the proportions of partial vaginismus amounting to 79 and 80% among women with vaginismus (Table III).

Table III. Proportions of women with partial vaginismus and total vaginismus among women with defined vaginismus

	Study II n=183		Study V ^a n=44	
	n	%	n	%
Diagnoses				
Partial vaginismus	144	78.7	35	79.6
Total vaginismus	39	21.3	9	20.4

a = women with partial and total vaginismus among index cases

Correspondence between our diagnostic criteria of the various forms of vaginismus and those in existing diagnostic systems concerning vaginismus, dyspareunia and vulvar vestibulitis/provoked vestibulodynia

Summary: All the criteria for vaginismus in DSM-IV-TR and two of three criteria for vaginismus in ICD-10 correspond to all our subgroups of vaginismus. No criteria for dyspareunia in DSM-IV-TR and ICD-10 correspond to any of our subgroups of vaginismus. Friedrich’s first and second criteria of vulvar vestibulitis and all ISSVD’s criteria of provoked vestibulodynia correspond to all our subgroups of vaginismus.

Vaginismus

The criteria for vaginismus in DSM-IV-TR (American Psychiatric Association 2000) correspond well to all our subgroups of vaginismus (Figure 10).

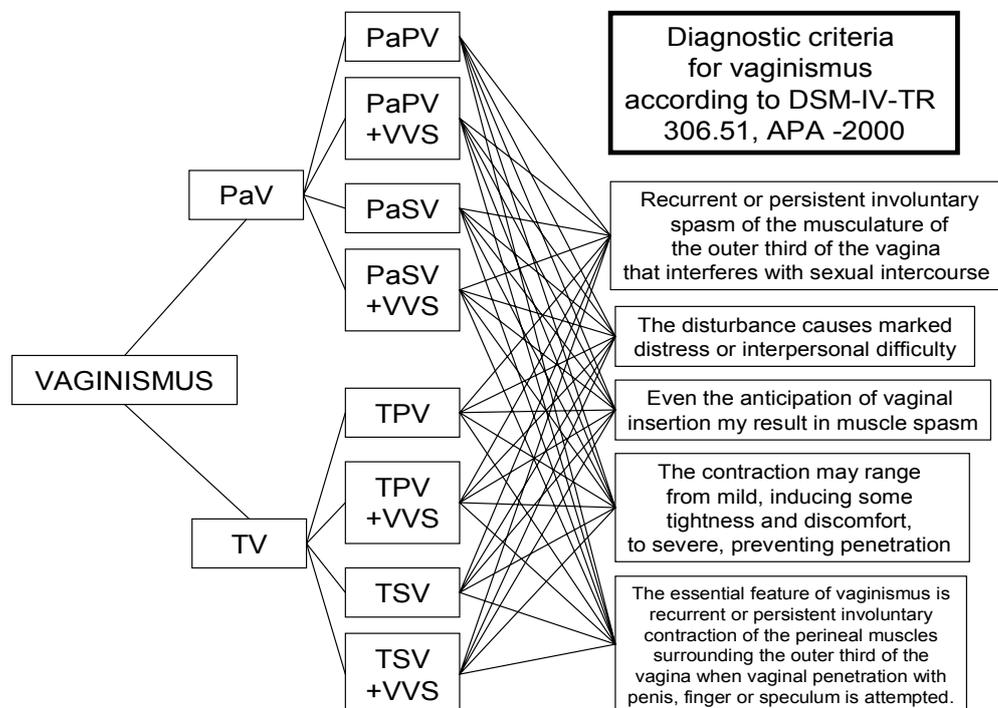


Figure 10. Subgroups of vaginismus according to our model compared to the definition of vaginismus in DSM-IV-TR (American Psychiatric Association 2000). Lines between groups indicate conformity between groups.

In the clinical gynaecological work, the ICD-10 criteria for classification of diseases (World Health Organization 2003) are often used. Two of the criteria for nonorganic vaginismus in ICD-10 correspond well to all our subgroups of vaginismus. The subordinate clause “causing occlusion of the vaginal opening” corresponds only with TV (Figure 11). However, the second sentence “Penile entry is either impossible or painful” is not in concordance with the subordinate clause “causing occlusion of the vaginal opening”. The definition of vaginismus in ICD-10 is therefore unclear.

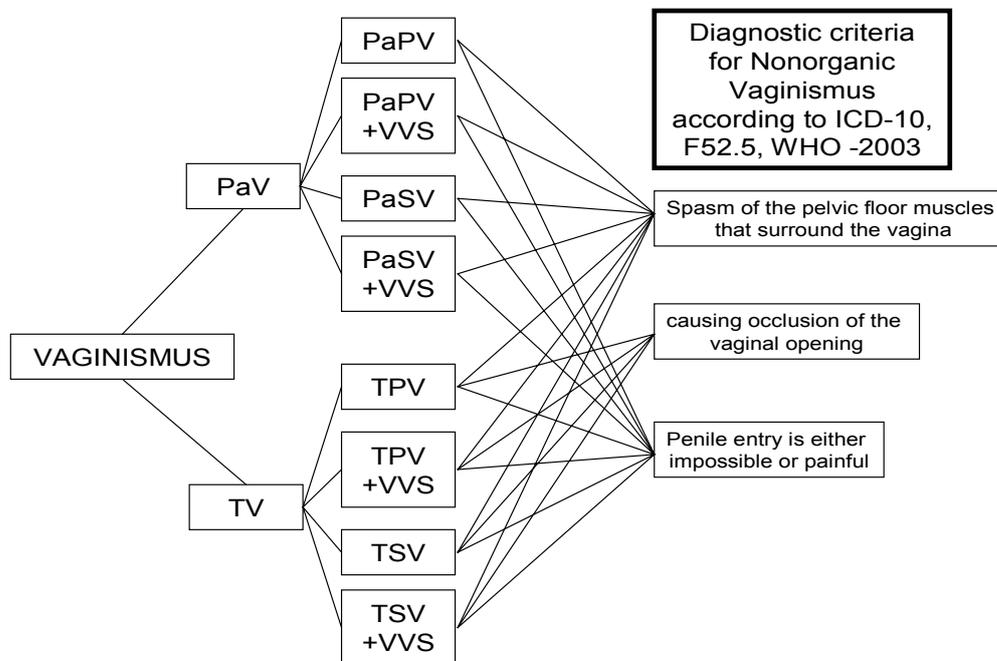


Figure 11. Subgroups of vaginismus according to our model compared to the definition of vaginismus in ICD-10 (World Health Organization 2003). Lines between groups indicate conformity between groups.

Vaginismus and dyspareunia

Clinically there is a difference between total primary vaginismus (TPV), which often has phobic components, and partial secondary vaginismus (PaSV), which has many symptoms in common with chronic pain syndromes. According to our theory and definition of partial vaginismus, all women in this group have penetration-related pain. In women with total vaginismus, attempts at penetration are simultaneously accompanied by burning pain, and feared or totally avoided.

The term dyspareunia is nowadays generally used for pain related to intercourse. Dyspareunia is a symptom, rather than a diagnosis (Danielsson 2001). Women with vaginismus could therefore theoretically be characterised by the symptom of dyspareunia.

The first and second criteria of dyspareunia in DSM-IV-TR, could fulfil a description of a woman with vaginismus according to our model. However, if the genital pain is caused exclusively by vaginismus, the genital pain should instead be classified as vaginismus according to the instruction in DSM-IV (Figure 12).

In ICD-10, the category of dyspareunia is to be used only if there is no primary nonorganic sexual dysfunction (e.g. vaginismus or vaginal dryness) (Figure 13).

None of our subgroups of vaginismus fulfils any of the diagnostic criteria for dyspareunia in DSM-IV-TR or ICD-10 (Figure 12 and 13).

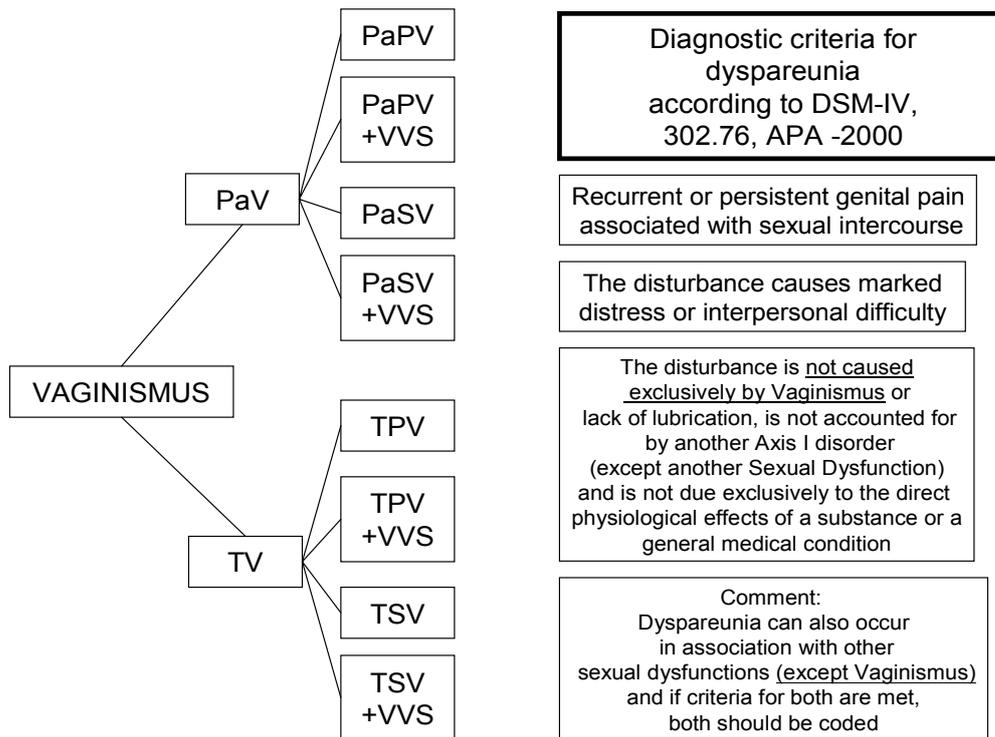


Figure 12. Subgroups of vaginismus according to our model compared to definition of dyspareunia in DSM-IV-TR (American Psychiatric Association 2000). Lines between groups indicate conformity between groups.

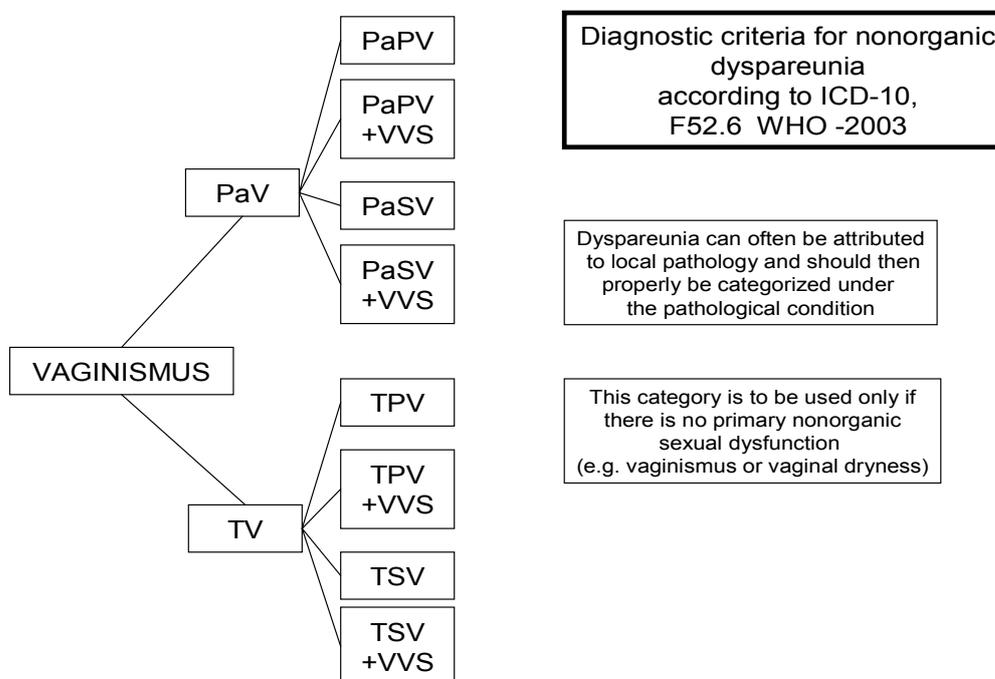


Figure 13. Subgroups of vaginismus according to our model compared to definition of dyspareunia in ICD-10 (World Health Organization 2003). Lines between groups indicate conformity between groups.

Vaginismus and vulvar vestibulitis/provoked vestibulodynia

Both de Kruiff et al. (2000), and ter Kuile et al. (2005) have reported on vestibular pain (defined as pain or no pain) on a cotton swab test in a majority of women with vaginismus. According to the findings in those studies, Friedrich’s first and second criterion of vulvar vestibulitis (Friedrich 1987) correspond to all our groups of vaginismus (Figure 14). Ter Kuile et al. (2005) have also stated that “ pain on touch in the vestibular area has to be included as one of the primary features of lifelong vaginismus”.

Only women who reported severe pain (≥ 5) in the vestibulum on a swab test were diagnosed, according to our study definition, as having vulvar vestibulitis. Friedrich does not use the word severe in the description of the second criteria “tenderness to pressure localised within the vulvar vestibule”. This means that also women with partial vaginismus with pain <5 on a swab test in the vestibulum are included in his definition.

ISSVD (Moyal-Barracco and Lynch 2004) has suggested a new definition of vulvar vestibulitis, called provoked vestibulodynia. All the subgroups of this new definition correspond to all our groups of vaginismus (Figure 15).

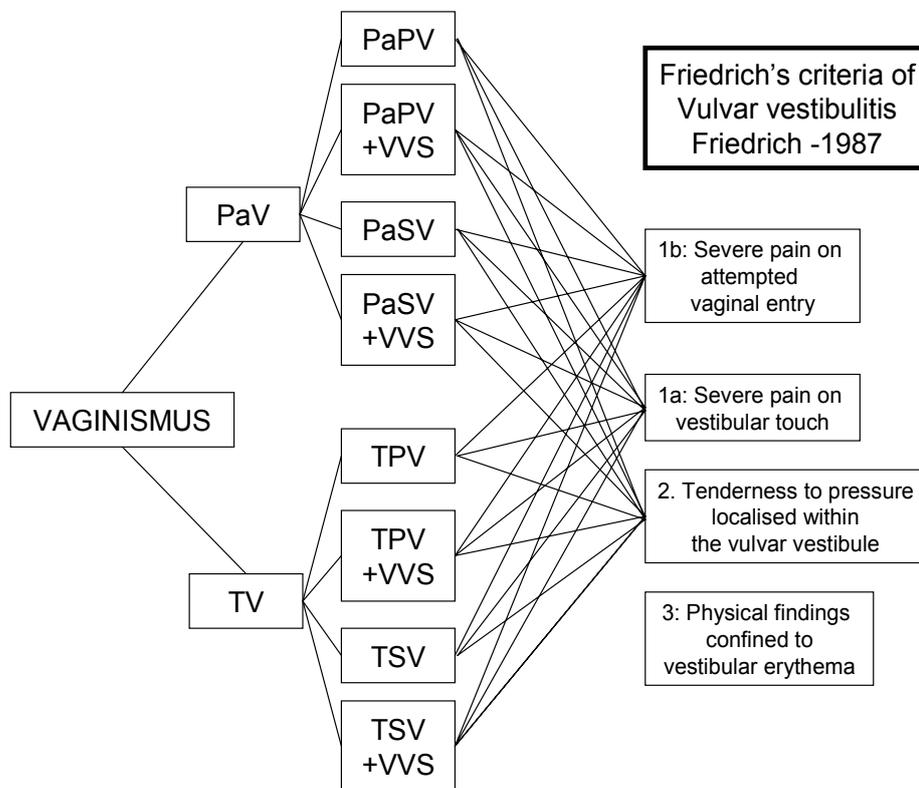


Figure 14. Subgroups of vaginismus according to our model compared to definition of vulvar vestibulitis by Friedrich (Friedrich 1987). Lines between groups indicate conformity between groups.

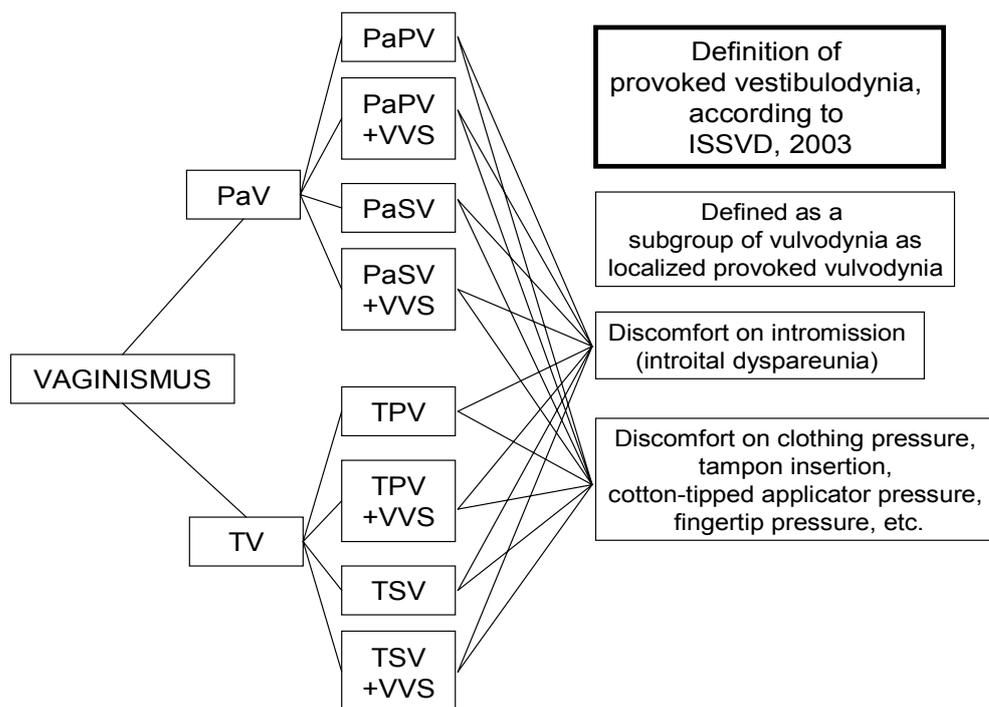


Figure 15. Subgroups of vaginismus according to our model compared to definition of provoked vestibulodynia by ISSVD (Moyal-Barracco and Lynch 2004). Lines between groups indicate conformity between groups.

Surface electromyography of pelvic floor muscles (II)

Summary: sEMG of the pelvic floor muscles revealed no significant differences between women with partial vaginismus solely, partial vaginismus and vulvar vestibulitis, and women from the asymptomatic group. In asymptomatic women, sEMG of pelvic floor muscles had good test-retest reliability.

In Study II we found no significant differences in sEMG measurements of pelvic floor muscles in pre- and post-test baseline; nor in rapid, tonic, and endurance contractions between women with partial vaginismus solely, women with partial vaginismus and vulvar vestibulitis, and asymptomatic women. The level of pre-test resting baseline was 4.55 μV in the group of women with partial vaginismus with or without vulvar vestibulitis, compared to 4.35 μV in the asymptomatic group.

There were no significant differences in the sEMG measurements within the symptomatic group, and between the symptomatic and the asymptomatic group, when we repeated the analyses without parous women and without women above 30 years old in the symptomatic group. There were no significant differences between the three study groups (PaV, PaV+ VVS, and asymptomatic) when we repeated the analyses without women investigated with the smaller vaginal sensor.

sEMG of the pelvic floor muscles in asymptomatic women had good test-retest reliability (correlations coefficients from 0.33 to 0.90, mean 0.72) between the first and second examination.

Itch and burning pain in women with partial vaginismus with or without vulvar vestibulitis (I, III, IV)

Summary: In Study III, itch was reported by 43% of women with partial vaginismus during a standardized penetration situation compared to 0% in the asymptomatic group. Sensations of burning pain preceded in 77% of cases sensations of itch. In Study IV, nearly one third of the women reported itch.

In the case report (Study I), the woman reported itch during the last two minutes of the pelvic examination, and the itching lasted for five minutes after the exposure was over (Figure 16).

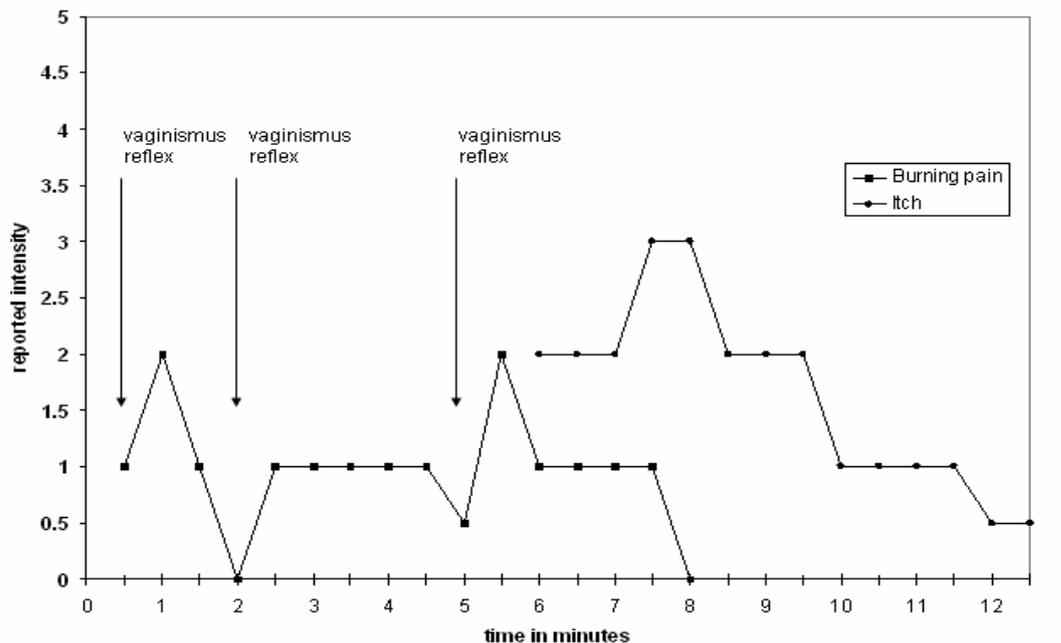


Figure 16. Reported intensity of burning pain and itch (from 0-10) in a woman with vaginismus during pelvic examination. The examiner's two fingers are withdrawn from the vagina/introitus after eight minutes of examination (Study I). Reprinted with permission from the Journal of Reproductive Medicine.

In Study III, 43 women (81.1%) in the target group of women with partial vaginismus with or without vulvar vestibulitis experienced burning pain and 23 (43.4%) itch during the standardized penetration situation, compared to 0% of the asymptomatic women. Twenty-two women in the target group (41.5%) reported both complaints.

In 17 of 22 women (77.3%), burning pain preceded the appearance of itch and in four cases the two complaints coincided. The median time from the moment when burning pain started until itch appeared was 150 sec. Burning pain and itch reactions in relation to moment of registration are presented in Figure 17.

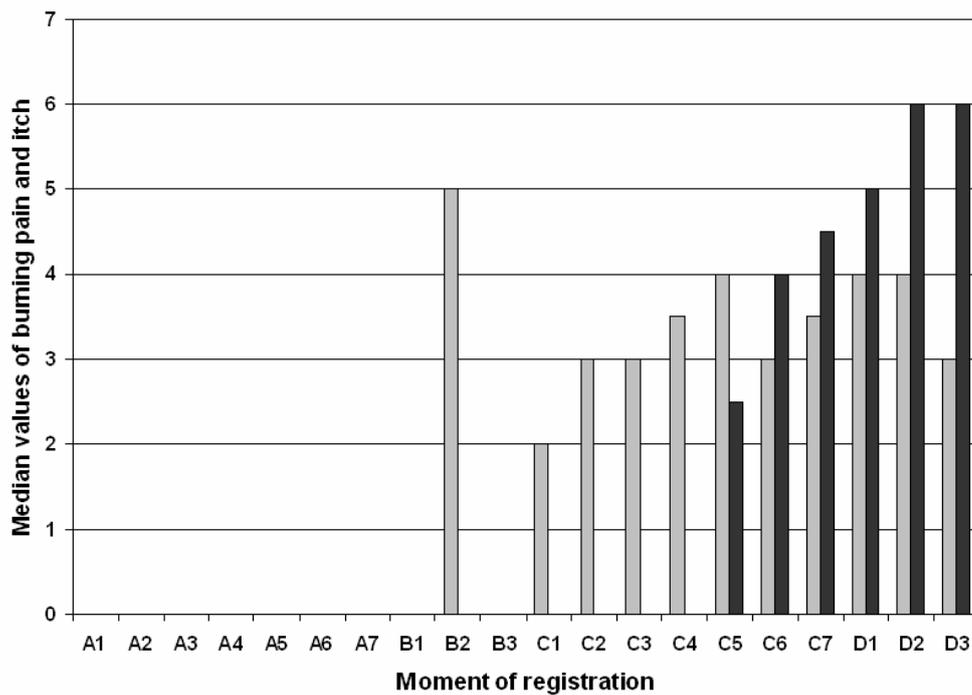


Figure 17. Median values (0–10) of burning pain (*grey bars*) and itch (*black bars*) in relation to time during assessments A1–D3 in a standardized penetration situation in 22 women with partial vaginismus who reported both symptoms (Study III). Reprinted with permission from Taylor and Francis Group.

There was only one significant difference between women with partial vaginismus solely and women with partial vaginismus and vulvar vestibulitis. Women with partial vaginismus and vulvar vestibulitis reacted more often with burning pain during the insertion of the sensor than women with partial vaginismus only ($p=0.045$).

In the pain development study (Study IV), itch in unprovoked situations, and/or during and after intercourse was reported by 29.2% of the women at onset of the problem, and by 27.7% when the women ceased having intercourse.

Pain development (I, IV)

Summary (Study IV): When the pain problem started in women with partial vaginismus with or without vulvar vestibulitis, pain *after* intercourse was significantly more common than pain during penetration. Pain intensity during penetration increased substantially from onset of the problem until the women ceased having intercourse. Burning pain during micturition after intercourse was a common finding throughout the course of the problem. Pain during penetration was described as short and sharp/incisive, while pain after intercourse was described as burning/smartering and long lasting.

Pain development in the woman in the case report.

The description of development of coital pain in the case report (Study I), corresponds well to the findings in Study IV.

Development of pain during penetration and after intercourse

In a sample of 24 women with partial vaginismus of whom 19 also had vulvar vestibulitis, pain after intercourse was a more common symptom than pain during penetration at onset of the problem. Twenty-three of 24 women (95.8%) had pain ≥ 1 after intercourse, while 12 of 24 (50%) had pain ≥ 1 during penetration ($p=0.001$) at onset of the problem (Figure 18). All 12 women with pain during penetration had also pain after intercourse.

All 19 women with partial vaginismus and vulvar vestibulitis had pain after intercourse when the problem first started and 10 had pain during penetration

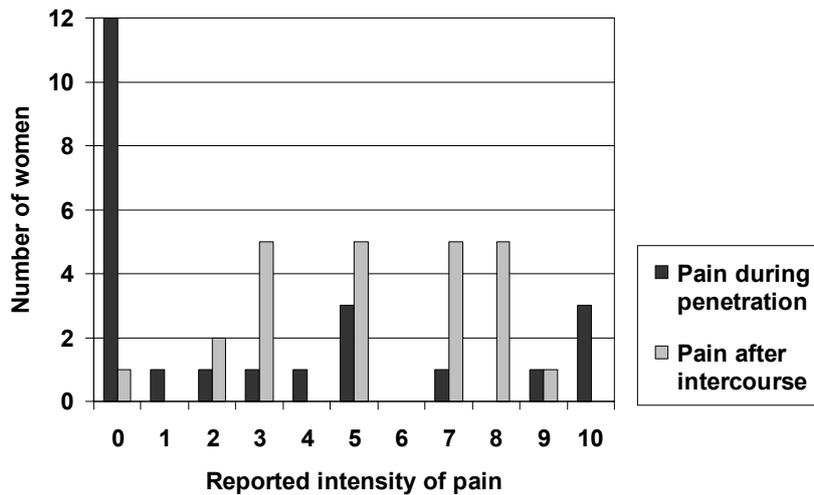


Figure 18. At onset of the problem: distribution of women according to reported pain intensity (0–10) at penetration and after intercourse (n = 24).

Eighteen women had ceased having intercourse; 15 of these 18 women were diagnosed as having both partial vaginismus and vulvar vestibulitis.

When the women ceased having intercourse, pain after intercourse and pain during penetration were equally common: Sixteen of 18 women (88.9%) had pain ≥ 5 after intercourse, while 15/18 (83.3%) had pain ≥ 5 during penetration ($p=1.0$) (Figure 19).

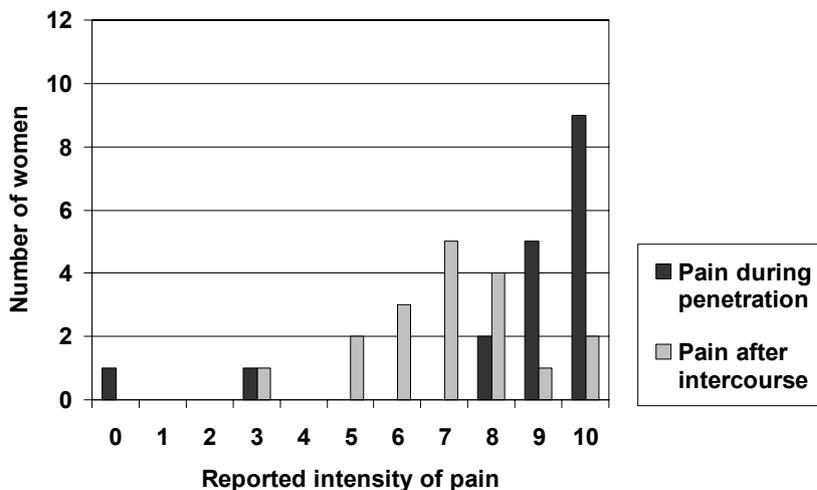


Figure 19. When the women ceased having intercourse: distribution of women according to reported pain intensity (0–10) at penetration and after intercourse (n=18).

From onset of the problem to the moment when the women ceased having intercourse, there was a marked increase of the pain intensity, especially for the pain during penetration (Figure 18 and 19). Median intensity of *pain during penetration* was 0.5 (P25=0.0, and P75=5.0) at onset of the problem, and 9.0 (P25=8.0, and P75=10.0) when the women ceased having intercourse, ($p=0.003$). Median intensity of *pain after intercourse* was 5.0 (P25=3.0 and P75=7.4) at onset of the problem, and 7.0 (P25=5.9 and P75=8.0) when the women ceased having intercourse, ($p=0.06$).

Pain during intercourse among those women who continued to have intercourse

At the time of the interview in Study IV, six women had intercourse, and of those, four women estimated their pain during penetration and after intercourse to ≥ 5 . For those four women the pain problem had lasted in median 66 months.

In all, 16/24 women had continued to have intercourse despite pain for more than one year.

Reason for the decision to cease having intercourse

Pain after intercourse was stated as a reason for the decision to cease having intercourse to almost the same extent as pain during penetration (Table IV).

Table IV. Reasons given by 18 women with partial vaginismus with or without vulvar vestibulitis for their decision to cease having intercourse

	n=18*
Pain during penetration is too strong	12
Pain after intercourse is too strong	10
Pain during intercourse is too strong	8
We had decided not to have intercourse because of the pain	7
Pain at micturition after intercourse is too strong	5
Fear of intercourse	5
My partner does not want to have intercourse because of my pain	4
Impossible, my partner cannot penetrate	3
I have no partner	1
Other reasons	3

*several answers possible

Pain quality and pain duration

Twenty-three of 24 women chose by themselves to use the words burning and/or smarting pain to describe the *pain after intercourse*. Sixteen women used both words. Twenty-one of 24 women did not use burning and/or smarting pain to describe the *pain during penetration* ($p=0.001$).

Pain during penetration lasted for one minute while pain after intercourse had duration of two hours (medians).

Vulvar pain during everyday life

Twenty-two percent of the women described symptoms of vulvar pain during everyday life (generalized unprovoked vulvodynia) when the problem first started, and 33% when they ceased having intercourse.

Burning pain during micturition after intercourse

A majority of the women reported burning pain during micturition after intercourse; 69.6% when the problem first started and 77.8% when they ceased having intercourse.

All women with burning pain at micturition after intercourse also had pain after intercourse.

Long-term follow-up results (V)

Summary: Long-term follow-up results in women with vaginismus treated with CBT show good treatment outcome in our study. The long-standing results, however, were highly associated to the definition of treatment outcome and had a variation of 75% (from 81% who could have intercourse, to 6% who had totally pain-free intercourse and were able to enjoy intercourse). A positive treatment result of therapy was maintained from end of therapy to follow-up. Every tenth woman with vaginismus in the study healed spontaneously after thorough assessment.

Results related to intercourse or enjoyment of intercourse

Outcome measures of *intercourse* and *enjoyment during intercourse* at follow-up were calculated for 36/44 of index cases (women not having a partner (n=7), and women not having had intercourse due to childbirth (n=1) during the previous two months were excluded).

The proportion of women with “positive treatment outcome” had a range of 75% depending on the definition used (from 81% (29/36) who could have intercourse, to 6% (2/36) who had totally pain-free intercourse and were able to enjoy intercourse). Sixty-one percent (22/36) of index cases reported an ability ≥ 6 to have intercourse without pain at follow-up. About one third (11/36) reported an ability ≥ 9 to have intercourse without pain.

Sixty-one percent (22/36) of index cases reported an ability ≥ 6 to enjoy intercourse. Half of the index cases reported an ability ≥ 6 both to have intercourse without pain and to enjoy intercourse at follow-up.

Median value for ability to have intercourse without pain was 8 (P25=5.5, and P75=9), and for ability to enjoy intercourse 7 (P25=5.5, and P75=9) (Figure 20 and 21).

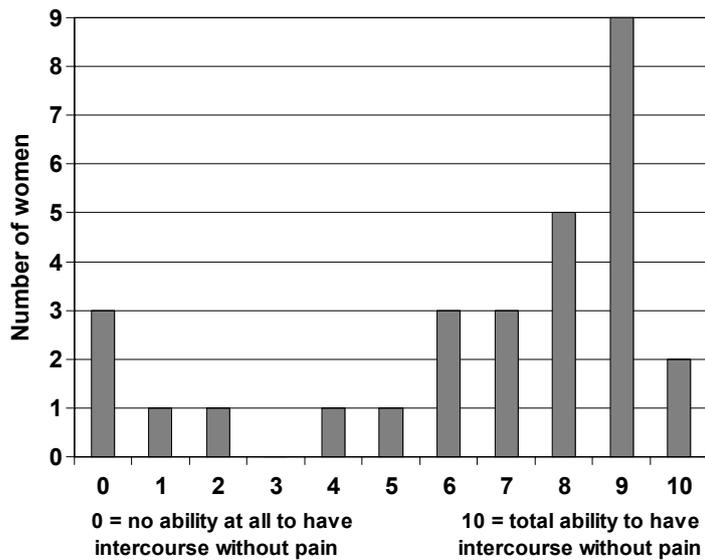


Figure 20. Reported ability to have intercourse without pain at follow-up among index cases with ability to have intercourse, n=29.

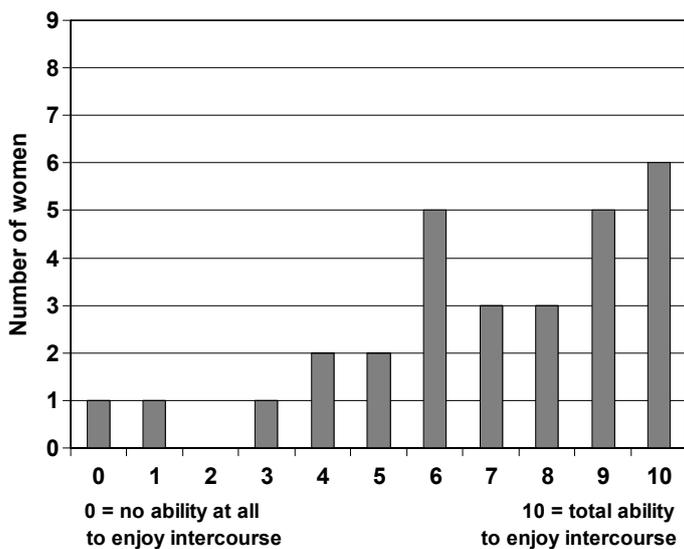


Figure 21. Reported ability to enjoy intercourse at follow-up among index cases with ability to have intercourse, n=29.

Comparison of treatment results at end of therapy and at follow-up

Twenty-three of 36 index cases had been able to have technical intercourse at end of therapy. Of those, 21/23 women (91%) were able to have intercourse, and 17 (74%) reported an ability ≥ 6 to have intercourse without pain at follow-up.

Twenty-four of 43 index cases (one case missing, 56%) reported a decrease of their problems of vaginismus and superficial coital pain from end of therapy to follow-up, 12 women (28%) reported unchanged problems, and seven women (16%) an increase of problems.

Results related to individual treatment goals

At follow-up, a high proportion of index cases (n=44) had fulfilled their treatment goals (reported goal fulfilment ≥ 6 on a scale from 0-10), “intercourse without pain” (71%), “intercourse without fear” (80%), “ability to enjoy intercourse” (69%), and “possibility to get pregnant” (77%). For fulfillment of the treatment goal “intercourse without pain” the median value was 8 (P25=7, and P75=10), for “intercourse without fear” 8 (P25=5, and P75=9), and for “ability to enjoy intercourse” 8 (P25=3, and P75=10).

Twenty-two of 44 women (50%) stated “the possibility to get pregnant” as a treatment goal, but for none of them this was their only goal.

Reports of self-worth before treatment and at follow-up

At follow-up, index cases (n=44) reported an increase in self-worth as sex partner from assessment to follow-up from 2 to 8 (median, $p < 0.001$), as a woman from 3 to 9 (median, $p < 0.001$) and as a human being from 6 to 10 (median, $p < 0.001$).

DISCUSSION

Discussion of the results

Diagnostic criteria

In our studies we tried not to make two different diagnostic groups of women with vaginismus and with vulvar vestibulitis. Instead, we decided to find out how many women could be diagnosed as having vaginismus, both total and partial, and/or vulvar vestibulitis when we used the diagnostic criteria for vaginismus in DSM-IV-TR and Friedrich's first and second criteria of vulvar vestibulitis. Only tenderness to pressure within the vulvar vestibule has been found to differentiate dyspareunia patients with and without vulvar vestibulitis (Bergeron et al. 2001a). In our studies only women with superficial coital pain with severe tenderness to pressure by a cotton swab test within the vestibule (≥ 5 on a scale from 0-10), were diagnosed as having vulvar vestibulitis. By this model, we found that 57-79% of women with vaginismus also could be diagnosed as having vulvar vestibulitis in our studies. These findings are in accordance with the findings of de Kruiff et al. (2000) and ter Kuile et al. (2005) who reported pain on touch in the vestibulum (defined as pain or no pain) in 69% of women with vaginismus. We found no woman with vulvar vestibulitis who did not also have vaginismus. This finding is also in accordance with recent findings. Reissing et al. (2005) reported 90% clear pathology of the pelvic floor muscles (vaginal hypertonicity, lack of vaginal muscle strength, and restriction of the vaginal opening) in women with vulvar vestibulitis, and significantly more pathology of the pelvic floor muscles than in women with "no pain with intercourse". The women in Reissing's study study, had an average pain rating during cotton swab test of 6, on a scale from 0-10.

Yet, healthy women also report pain on vestibular touch. In a group of gynaecologic patients without coital pain, 18% of the women had a positive swab test in the vestibulum (Goetsch 1991). In the same study, women with vulvar vestibulitis had more pain-positive sites and reported higher pain intensity than women who had a positive swab test but no pain during intercourse. Van Beurden et al. (1997) found a positive touch test in 53% of healthy women with vestibular erythema but without pain during intercourse. Twenty-three percent had both vestibular erythema and localised tenderness confined to the vulvar vestibule. The findings of a positive swab test in women without coital pain make it uncertain to set a diagnosis of vulvar vestibulitis merely by a positive swab test. These findings may support the use of severe tenderness (≥ 5 on a scale from 0-10) to pressure by a cotton swab test within the vestibule for a clinical diagnosis of vulvar vestibulitis.

In our study of pain development (Study IV) all 19 women diagnosed as having partial vaginismus and vulvar vestibulitis described pain after intercourse, but only 10 of them had pain during penetration at onset of symptoms. When women in our study ceased having intercourse, severe pain during penetration and after intercourse were equally common. Despite these findings, and despite earlier descriptions of postcoital pain in women with vulvar vestibulitis (Michlewitz et al. 1989; Furlonge et al. 1991; Mann et al. 1992; Schover et al. 1992; Secor and Fertitta 1992; de Jong et al. 1995; Reid et al. 1995; Meana et al. 1997; Pukall et al. 2000; Danielsson et al. 2001; Driver 2002; Danielsson et al. 2003; Reed et al. 2003; Reissing et al. 2005), postcoital pain is *not* included in the two most commonly used definitions of vulvar vestibulitis to describe the coital pain. In Friedrich's definition (Friedrich 1987), only severe pain on attempted vaginal entry is included. In ISSVD's new classification of vulvodynia (Moyal-Barracco and Lynch 2004), where vulvar vestibulitis is reclassified as provoked vestibulodynia, only discomfort on intromission is used in the definition. This means that nearly half of the women diagnosed as having partial vaginismus and vulvar vestibulitis in our study would not have been diagnosed with vulvar vestibulitis at onset of

symptoms according to the existing diagnostic systems. According to our theories and definitions, however, all women could have been diagnosed as having partial vaginismus, when these problem started.

Earlier studies have stated that the intercourse-related pain described by women with vaginismus is very difficult to distinguish clinically from the pain described by women with dyspareunia/vulvar vestibulitis (van Lankveld et al. 1995; de Kruiff et al. 2000; Reissing et al. 2004). In none of these studies is pain after intercourse or other early symptoms included, and we do not know if an addition of these aspects would have made a differentiation possible.

In addition, the existing diagnostic criteria for vulvar vestibulitis/provoked vestibulodynia make it difficult to distinguish between this condition and vaginismus. When we scrutinized the existing diagnostic systems, we found a correspondence between all our subgroups of vaginismus and Friedrich's first and second criteria of vulvar vestibulitis and all ISSVD's criteria of provoked vestibulodynia.

In DSM-IV-TR, dyspareunia and vaginismus are the only pain conditions classified outside pain disorders, and vaginismus is the only pain condition in this classification system that does not require the experience of pain (American Psychiatric Association 2000). The DSM's classifications of dyspareunia and vaginismus have been criticised by Payne et al. (2005). In their article, the authors criticise the limitation of pain in dyspareunia to sexual activity. According to DSM-IV-TR, women with pain during tampon insertion and/or gynaecological examinations could not be diagnosed as dyspareunia. Vaginismus in DSM-IV-TR is indistinct as to what type of vaginal penetration is included in the definition. In the summary of the diagnostic criteria it is written: Criterion A. "Recurrent or persistent spasm of the musculature of the outer third of the vagina that interferes with sexual intercourse". In the "Diagnostic features" it is written: "The essential feature of vaginismus is the recurrent or persistent involuntary contraction of the perineal muscles surrounding the outer third of the vagina when vaginal penetration with penis, finger, tampon, or speculum is attempted (Criterion A)", i.e. not only penetration with a penis are included. A diagnosis of partial vaginismus, in our studies, does not have this limitation to sexual activity. Payne et al. (2005) writes that "pain and chronic pelvic floor tension are present in women with both vaginismus and dyspareunia, and the difference may simply be one of severity rather than kind". They suggest that behavioural avoidance and distress upon exposure might be differentiating characteristics between women suffering from vaginismus and dyspareunia. This might be true for women with total vaginismus, but still this suggestion does not render a diagnosis in women with non-coital penetration pain.

The existing diagnostic systems of sexual dysfunctions have also been criticised by Basson et al. (2003). This group defines dyspareunia as "persistent or recurrent pain with attempted or complete vaginal entry and/or penile vaginal intercourse". A diagnosis of vaginismus is in their system not limited to intercourse, and vaginismus is defined as "persistent difficulties to allow vaginal entry of a penis, a finger, and/or any object, despite the woman's expressed wish to do so". The group states that in cases of vaginismus, discomforting or painful vaginal entry may occur, which makes the differentiation between vaginismus and dyspareunia uncertain also in this system.

In conclusion, we regard partial vaginismus and vulvar vestibulitis not as two mutually exclusive diagnoses; instead, they represent two diagnoses which may occur at the same time. We have found the study definitions of partial vaginismus and vulvar vestibulitis used in Study II, II, IV and V of value for clinical work. According to this, women with partial vaginismus and a slightly positive swab test (< 5 on a scale from 0-10) in the vestibulum are diagnosed as having solely partial vaginismus, and when there is a positive swab test ≥ 5 an

additional diagnosis of vulvar vestibulitis is added. The clinical value of setting both diagnoses is that therapeutic approaches for both vaginismus and, if necessary, for vulvar vestibulitis (after the vaginismus has been treated) might be used.

Symptoms of both partial vaginismus and vulvar vestibulitis

An increasing number of studies has shown that there is an association between vaginismus and dyspareunia/vulvar vestibulitis, and that women with vulvar vestibulitis have pelvic floor pathology. Many authors regard this pelvic floor pathology as secondary to vulvar vestibulitis. Our theory is that in many women the vaginismus reflex comes first, and repeated penetrations or attempts to penetrate in an unaroused state with a partly closed vagina lead to the findings and symptoms seen in women with vulvar vestibulitis. This theory is supported by findings in our studies.

In the study of burning pain and itch reactions during standardized penetration (III), women with partial vaginismus and vulvar vestibulitis reacted more often with burning pain during the insertion of the sensor than women with partial vaginismus only. According to our theory, the vaginismus reflex itself triggers the burning pain response during penetration and the sensation in these cases is intensified if the women have an additional vulvar vestibulitis.

The finding in four of our studies (II, III, IV, and V) of women with partial vaginismus without vulvar vestibulitis, but no women with vulvar vestibulitis without partial vaginismus, could also support the assumption that partial vaginismus comes first. In some women, vulvar vestibulitis later appears.

Women with total vaginismus report concomitant vulvar vestibulitis to a lesser extent than women with partial vaginismus in our studies (Study II and V). This is a reasonable finding. Women with total vaginismus are not able to have penetrations and often totally avoid penetration situations, and accordingly symptoms of vulvar vestibulitis do not develop.

In Study IV, according to our theory and diagnostic criteria of partial vaginismus, all women could be diagnosed as having partial vaginismus when their problem started. Nine of 19 women diagnosed as having partial vaginismus and vulvar vestibulitis at the time of the interview reported burning pain after intercourse as their only symptom when their problem started, i.e. none of these women could have been diagnosed as having vulvar vestibulitis at this moment. If vulvar vestibulitis should be regarded as the primary pain process in women with partial vaginismus and vulvar vestibulitis, a re-evaluation of the existing diagnostic criteria of vulvar vestibulitis may be necessary.

Statements from other researchers might also support our theory. Zolnuon et al. (2006) state that it requires an explanatory model in which pelvic floor muscle dysfunction is regarded as a primary pathologic process to explain the observed clinical efficacy of biofeedback in women with vulvar vestibulitis (Glazer et al. 1995; Bergeron et al. 2001b; Danielsson et al. 2006).

None of the findings in our studies affirmed or questioned the theory that partial vaginismus is secondary to vulvar vestibulitis.

From a clinical point of view, the important matter is not whether the vaginismus reflex is primary or secondary to vulvar vestibulitis. The crucial thing is to diagnose an existing partial vaginismus in women with vulvar vestibulitis, and to pay attention to vaginismus as a maintaining factor for the symptoms of superficial coital pain. If women with vulvar vestibulitis are treated only with pain-coping strategies for vulvar vestibulitis, or even with surgery, the increased tension and the vaginismus reflex of the pelvic floor muscles will not disappear. This increases the risk of treatment failure or relapse of symptoms after

“successful” treatment. If women with partial vaginismus and vulvar vestibulitis are treated with cognitive behaviour therapy for vaginismus, according to our clinical experience findings of pain on touch in the vestibulum might disappear in most cases without specific treatment. In accordance with this clinical finding, ter Kuile and Weijnenborg (2006) reported that a cognitive behavioural group program for women with vulvar vestibulitis (similar to the treatment program presented by their research group for women with vaginismus) affects vestibular pain, vaginal muscle control, pain control and sexuality.

Partial vaginismus and generalized unprovoked vulvodynia

One third of women with partial vaginismus in Study IV reported generalized unprovoked vulvodynia, a symptom that could be explained by theories of conditioning. Harlow et al. (2001) found that reported pain at the time of first use of tampons was associated with an increased risk (OR 2.4) of chronic lower genital tract discomfort later in life. “The data support the theory that vulvar trauma in early life may influence or serve as a marker for risk of subsequent chronic vulvar disorders” (Harlow et al. 2001). According to our theory, pain during tampon insertion in this group of women acts as unconditioned stimulus and elicits a vaginistic reflex with burning pain (unconditioned response). Thoughts of the problem (e.g. “I am too narrow”, “will I be able to have intercourse?”) act as conditioned stimuli and elicit a conditioned response, a persistent vaginistic reflex and consequently an increased tone in the pelvic floor muscles and generalized unprovoked vulvodynia. In this situation the vaginismus reflex may clinically appear as hypertonicity of the pelvic floor muscles, which is referred to as persistent involuntary spasm in DSM-IV-TR (American Psychiatric Association 2000).

Reed et al. (2003) have suggested that provoked vestibulodynia (vulvar vestibulitis) and generalized vulvodynia are two presentations of the same problem on a continuum of severity rather than two distinct entities. Similar findings are also reported in other studies (Dalton et al. 2002; Giesecke et al. 2004).

Burning pain in a standardized penetration situation and during pelvic examination

In Study III, burning pain was triggered in most of the cases during the insertion of the vaginal sensor, i.e. “penetration”. Mere contraction and relaxation of the pelvic floor muscles, without any inserted sensor, did not provoke any burning pain. Clinically, women with partial vaginismus report sensations of burning pain during pelvic examination and insertion/extraction of one or two fingers into the vagina, when the examiner simultaneously can feel a partial vaginismus reflex around his/her finger or fingers. When the vaginismus reflex is already triggered, voluntary contractions of the pelvic floor muscles around the examiner’s fingers and/or movement of the examiner’s fingers often provoke even more burning pain, and itch, in a woman with untreated partial vaginismus. We assume that a similar reaction occurred during the standardized penetration situation in women with partial vaginismus, but not in the comparison group

Itch in a standardized penetration situation

In Study III, we used the “standardized penetration situation”, and found that sensations of burning pain in most cases preceded sensations of itch. This result may be explained in two ways, according to a theoretical model described by Schmelz (2001). (1) Itch and pain were triggered at the same time, and the itch sensation was at first suppressed by the burning pain sensation. This assumption is not supported by the finding that itch in most of the cases in our

study occurred while burning pain was still present. (2) There was a mechanical stimulus, which after sensitization of the “itch neurons” by histamine, was perceived as itch. The findings that itch in most cases appeared during the vaginal measurement when the woman contracted and relaxed her pelvic floor muscles around the sensor, i.e. existence of a mechanical component, could support this assumption. In patients with chronic pruritus it has been found that painful stimuli, including mechanical stimulation, evoke itch (Ikoma et al. 2004). The same phenomenon could also be induced in healthy subjects by conditioning by histamine (Ikoma et al. 2004). The second part of the theory could also be supported by findings of a higher number of degranulated mast cells in the vestibular mucosa in women with vulvar vestibulitis than in women in a control group (Bornstein et al. 2004). This theoretical model by Schmeltz could explain the reports of itch during and after intercourse in women with partial vaginismus. Here, reflex contraction of the vagina and movements of the penis in the vagina acts as mechanical stimulus, perceived as itch.

Pain during penetration and after intercourse

In our study of pain development (IV) in women with partial vaginismus, pain after intercourse was more common than pain during penetration at onset of the problem, and it seemed also to be a powerful symptom when the women ceased having intercourse. Pain after intercourse was stated as a reason for the decision to cease having intercourse almost to the same extent as pain during penetration.

There was a great increase in the reported intensity of pain during penetration from the moment when the problem started to the moment when the women ceased having intercourse. The time lapse from first appearance of symptom to refraining from intercourse was nine months (median), and at this latter moment, the reported median pain intensity of pain during penetration was 9, and 7 after intercourse. This means that the women ceased having intercourse only when the pain was extremely severe.

In earlier studies of women with superficial coital pain, focus has been on the penetration-related pain and pain during intercourse; and on descriptions of pain at the time of the study (Meana et al. 1997). Danielsson et al. (2003) calculated by means of answers in an interview with women with severe dyspareunia the predictive value (1 indicates a high predictive value) of pain during penetration (0.37) and after intercourse (0.26) for a diagnosis of vulvar vestibulitis. The authors did not further comment on the findings, but according to their results both pain during penetration and after intercourse have a low predictive value for a diagnosis of vulvar vestibulitis. The low predictive value of pain during penetration in that study questions the existing diagnostic criteria of vulvar vestibulitis.

Both pain during penetration and after intercourse can be explained when regarding the partial vaginismus reflex as a conditioned response (Wijma and Wijma 1997). The penetration situation, associated with pain or fear, has become a conditioned stimulus, and elicits the conditioned responses, a vaginismus reflex and a feeling of burning pain. As pain in itself is a stimulus for the vaginismus reflex, a vicious circle is easily created. Not only during penetration but also after intercourse, women with partial vaginismus describe a contraction of the pelvic floor muscles, simultaneously accompanied by a feeling of burning pain. This tendency to contract the pelvic floor muscles after penetration has to be focused separately during the treatment of vaginismus. By this approach, pain after intercourse could be eliminated.

Zolnuon et al. (2006) discuss the possibility of vulvar vestibulitis as a primary muscle dysfunction and compares this disorder with findings in patients with temporomandibular

disorder. In this, primary abnormalities in muscle function are intimately associated with the development of pain (Dworkin and LeResche 1992). Zolunom et al. (2006) found a concomitant diagnosis of temporomandibular disorder in 78% of women with vulvar vestibulitis.

Postcoital burning pain at micturition was a common symptom in our pain development study (IV), present in 70% of the women from the early start of the problem, and the prevalence did not change when the problem got worse. The muscles of the pelvic floor involved in the partial vaginismus reflex also surround the urethra. We therefore interpret postcoital pain at micturition as provoked by a partial vaginismus reflex as well, and as a subgroup of postcoital pain. Support of this theory comes from the finding that all women with burning pain at micturition after intercourse also had pain after intercourse.

It is not possible to answer the question if postcoital burning pain and pain during micturition are early symptoms of partial vaginismus with or without vulvar vestibulitis from the findings in Study IV. To answer this question a new study has to be carried out where women with postcoital burning pain are diagnosed as early as possible for both partial vaginismus and vulvar vestibulitis, and then followed and reevaluated.

Quality and duration of pain

In the pain development study (IV) we found that pain during penetration lasted for one minute, and was most often described as sharp/incisive/bursting, while pain after intercourse had a duration of two hours and was described by words as burning and/or smarting. One earlier study has described that women with vulvar vestibulitis use both thermal and incisive words to express their sensations of pain during intercourse (Bergeron et al. 2001a). The pain descriptions in that study were not divided into different time periods of intercourse (Bergeron et al. 2001a). The duration of pain related to penetration has not, to our knowledge, been measured before, but in other studies of women with vulvar vestibulitis, postcoital pain has been described as long lasting (Furlonge et al. 1991; Schover et al. 1992; Bergeron et al. 1997; Reissing et al. 2005). In women with vaginismus, pain duration in relation to intercourse has not been studied.

The women's use of words to describe the pain in Study IV indicates that the pain during penetration and after intercourse is triggered from the mucosa rather than from the musculature. Muscle pain in general is described as aching and cramping with diffuse and referred localization (Graven-Nielsen & Arendt-Nielsen, 2002), and no women in our study used aching and cramping to describe the pain in relation to intercourse. If the pain during penetration and after intercourse is triggered from the mucosa, both types of pain could be mediated by different types of nerve fibres, according to the descriptions of pain. The short and sharp/incisive/bursting pain during penetration could be mediated by A δ -fibres, while the longlasting and burning and/or smarting pain after intercourse could be mediated by C-fibres. There may also exist an interaction between pain from the mucosa and pain from the musculature, as "pain fibres (C-fibres and A δ -fibres) that are sensitized by mucosal inflammation initiate sensitization and contraction of the underlying musculature. Further sensitization of the muscle pain receptors may in turn, through a process of central sensitization, reduce sensory pain thresholds, resulting in a 'vicious cycle' of inflammation and additional muscle contraction" (Graven-Nielsen and Arendt-Nielsen 2002; Svensson, Graven-Nielsen and Arendt-Nielsen 1998) cited in (Zolnoun et al. 2006). Vulvar vestibulitis is regarded by some authors as a neurogenic inflammation (Bornstein et al. 2004; Graziottin and Brotto 2004). This neurogenic inflammation in the mucosa of the vulvar vestibule in women with vulvar vestibulitis may, according to the theory above and to our theory,

contribute to the reinforcement of the vaginismus reflex. Also the secondary changes (primary and secondary hyperalgesia and allodynia) described in women with vulvar vestibulitis (Bohm-Stärke et al. 2001b; Pukall et al. 2002) might lead to reinforcement of the vaginismus reflex in women with partial vaginismus and vulvar vestibulitis.

It is not known if the smooth muscle fibres described in the vaginal wall (Smouth, Jacoby and Lillie 1969) take part in the vaginismus reflex and contribute to the pain in women with superficial coital pain.

Existing methods for measurement of vestibular/introital pain

Bohm-Stärke et al. (2001b) have reported that all women with vulvar vestibulitis (n=19) interrupted distension of the vaginal introitus (by use of a small, soft, inflatable rubber balloon) due to pain, whereas only two control-group subjects (n=20) reported pain. The mean pressure in the balloon at which the distension was interrupted in women with vulvar vestibulitis was lower than in control subjects. These findings are not commented on or discussed in the article. Maybe the researcher by this method provoked a pain similar to the penetration related pain described by women in our study. If so, this could be developed to a method for indirect measuring of penetration pain in women with superficial coital pain. Today, the most common way of measuring pain in women with vulvar vestibulitis is the women's report of pain intensity when the vestibulum is touched with a cotton swab or when an vulvar-algesiometer is used (Pukall et al. 2004; Danielsson et al. 2006). None of these methods, however, include any distension of the vaginal introitus, which is why they do not measure the functionally relevant quality of the penetration pain. Nor do we, with any of these three methods, know if this provoked and measured introital/vestibular pain reflects the pain these women experience during intercourse. In a recent treatment study of women with vulvar vestibulitis, the pressure at which the women reported pain in the vestibulum, when the vulvar-algesiometer was used, did not correlate to the women's own reports of whether they were cured or not from their coital pain (Danielsson et al. 2006).

Why do women with coital pain continue to have intercourse despite pain?

In the pain development study (IV), 16/24 women with partial vaginismus continued to have intercourse for more than one year despite pain. The women ceased having intercourse only when the pain during penetration and after intercourse was extremely severe. At the time of the interview in Study IV, six women had intercourse, and of these, four women had severe pain during penetration and after intercourse. These four women had had their problem for more than five years. These findings raise the question why women continue to have intercourse despite pain. This question has been studied in a qualitative study (constant comparative method, from grounded theory) at our department. Twelve women (21-49 years) with secondary vaginismus coming for consultation to the Psychosocial Division of Obstetrics and Gynaecology, Linköping, were interviewed. All women had previous experiences of intercourse without pain, and were included in Studies IV and V of this thesis. The author found a core category of striving to experience entirety including the wish to feel like a normal woman. Categories found were unfulfilled expectations, sexual pleasure, integrity and hope (Örtoft 2006). Even if the informants in this study stated that they had intercourse despite pain to fulfill their own needs, both the partner and the environment probably influenced their understanding of what it is to be a normal woman.

Roberts et al. (1995) showed in their study that women often have their sexual focus on the partner instead of on themselves. The women undervalued the importance/value of their own

sexual pleasure compared with the value associated with the pleasure the partner experienced, and the women's enjoyment of sexual intercourse was regarded both by men and women as proof of the man's technique/work.

Reissing et al. (2004) have suggested that women with total vaginismus (unclear if both primary and secondary total vaginismus are included in the assumption) may differ from women with dyspareunia/vulvar vestibulitis in that they are more fearful of pain and/or more unwilling to bear the pain associated with penetration. In cases of total primary vaginismus, this is a reasonable assumption, but women with partial vaginismus who had ceased having intercourse also seem to be unwilling to bear the pain associated with penetration at the moment when they ceased to have intercourse. Both "pain during penetration is too strong" and "pain during and after intercourse is too strong" were the most frequent reasons for these women's decision to cease having intercourse, and more frequent reasons than fear of intercourse. We do not know if "fear of intercourse" also includes "fear of pain during intercourse", as we did not ask for fear of pain during intercourse and fear of intercourse (not including fear of pain) separately in our study.

There is a need for further research in order to understand this complicated interplay. The knowledge of why women with superficial coital pain continue or cease to have intercourse is important for understanding and treating these women. Continuing to have intercourse with fear or pain reinforces the vaginistic reflex.

sEMG measurement of pelvic floor muscles

It is a convincing clinical finding that women with partial vaginismus with or without vulvar vestibulitis have an increased tone in the pelvic floor muscles, and high muscle tone has been found by palpation in women with vaginismus and dyspareunia/vulvar vestibulitis (de Kruiff et al. 2000; Reissing et al. 2004). Yet, our sEMG measurements of pelvic floor muscles in women with partial vaginismus with or without vulvar vestibulitis did not reveal any differences in resting tone or ability to contract or relax the pelvic floor muscles when compared to the findings in an asymptomatic group. The findings in the present study raise the question whether this increased tone is of other origin than electrogenic.

When we apply the theories of Simons and Mense (1998) to our findings, two possible explanations emerge. (1) The increased tone we observe clinically ought to be of a different origin than electrogenic contraction, i.e. changed viscoelastic properties of the soft tissues associated with the muscle and/or endogenous contracture within the muscle fibre. (2) sEMG of pelvic floor muscles was in our model of investigation an unreliable method to measure electrogenic contractions which did exist (β -error). The sizes of our samples were small. With a test significance level of 0.05 and a power of 80%, more than 1800 participants per group would be needed in order to demonstrate a difference in pre-test baseline between the symptomatic and asymptomatic groups in our study. This need of very large groups to show a difference makes our model of sEMG of the pelvic floor muscles useless to establish the diagnoses of partial vaginismus/vulvar vestibulitis.

In two other studies sEMG has not revealed any differences between women with and without vaginistic reactions in ability to contract and relax the pelvic floor muscles (van der Velde and Everaerd 1999; van der Velde et al. 2001).

Shafik and El-Sibai (2002) used needle EMG for measurements of EMG activity at rest (see Introduction, page 10). In this study, we do not know to what extent the pain evoked by the needle insertion into the pelvic floor muscles acted as conditioned stimulus in women with vaginismus and provoked a vaginismus reflex. If so, the increased EMG activity measured at

rest may in fact be due to a vaginistic reflex. In the case report in Study I the mere thought of penetration evoked burning pain and a supposed vaginismus reflex. If needle insertion provokes a vaginistic reflex, needle EMG becomes inapplicable for the studies of muscle activity at rest in women with vaginismus.

The second main question is to what extent the investigation in a pelvic examination chair influenced the results. If we compare our data in the asymptomatic group with data from Glazer et al. (1998) (who used the same type of equipment in a supine position in a resting chair), our data correspond well except for one variable. Our mean value of the pre-test resting baseline level is more than double, 4.35 vs. 1.96 μ V. The reason for this is unknown. Glazer has stated that the difference is due to the different examination positions (personal communication). Reissing et al. (2004) used the same equipment as we did, but in a supine position (as in Glaser's study) and with adequate leg support. Nor in this study were there any significant group differences between women with vaginismus and dyspareunia/vulvar vestibulitis and a no pain group, on measures of pelvic muscle tone (pre-test resting baseline). This finding and an earlier finding of Wennergren et al. (1991) that optimal relaxation of the pelvic floor muscles can be achieved by adequate leg support, question the importance of position for measurement of pelvic floor muscle tone at rest.

In conclusion, sEMG was not of any diagnostic value in our study for distinguishing between women with partial vaginismus with or without vulvar vestibulitis and asymptomatic women. A possibly existing increased tone in pelvic floor muscles in these two groups of symptomatic women may be of other origin than electrogenic contractions.

Long-term follow-up study

Long-term follow up results in women with vaginismus (for nearly four years) treated with CBT during 16 sessions show good treatment outcome.

Eighty-one percent of index cases had intercourse at long-term follow-up, and a majority of them (61%) reported an ability ≥ 6 both to have intercourse without pain and enjoyable intercourse. Some women were able to enjoy intercourse despite considerable pain during intercourse. In Study V, ability to have intercourse at end of therapy was in general maintained, as reported at follow-up. In some cases, there was even an improvement in ability to have intercourse after treatment. More than one third of women who could not have technical intercourse at end of therapy managed to have intercourse without much pain at follow-up. This finding is in agreement with the women's own impression of improvement or not after therapy. More than half of index cases reported a decrease of their problems of superficial coital pain and vaginismus from end of treatment to follow-up.

Long-term follow-up results of treatment outcome were, however, highly associated in Study V to definition of treatment outcome, with a variance of 75% depending on the definition used. This choice is thus a delicate issue, which may be influenced by the researchers' values.

When questions of goal fulfilment were used as outcome measures, we found that index cases had a high degree of fulfilment of their goals, intercourse without pain and intercourse without fear. Our cognitive behaviour model for treatment of women with vaginismus may explain these findings. In this model, much attention is paid to the women's learning to have vaginal penetration without pain and fear. Forty-two of 44 index cases had intercourse without pain as a goal and 35 intercourse without fear. This implies that among index cases reduction of pain was as important as a treatment goal as reduction of fear. In a study by ter Kuile et al. (2006) it was found that CBT for lifelong vaginismus modified coital fear, but unfortunately questions of coital pain were not included in that study. The inclusion or not of

the pain component in women with vaginismus seems to play an important role also in the design and evaluation of treatment studies.

We also found that half of the index cases stated ability to get pregnant as treatment goal (but no one as a single treatment goal), which is in accordance with findings in a previous study (Drenth et al. 1996). A British special interest group for sexual dysfunction has in a recent review stated recommendations for the management of vaginismus (Crowley et al. 2006), in which they recommend that “vaginal penetration by a penis should not be assumed to be the desired outcome of women presenting with vaginismus”.

Index cases reported higher self-worth as sex partners, women, and human beings at follow-up than before treatment. We could not say from the findings in our study that this is due to the effect of CBT. Neither do we know if the women’s ability to have intercourse affected their self-worth as women and human beings, as no such study exists.

In Study V, 12% in our total sample of women assessed for superficial coital pain and diagnosed as having vaginismus, and 23% of those assessed but not eligible for the CBT program had healed without treatment at follow-up. In a randomized treatment study of women with lifelong vaginismus (mean duration of problems 11 years) (van Lankveld et al. 2006), none of the women in the waiting-list group had intercourse at the post-study assessment after three months, which probably is too short a period for self-healing to occur in women with a long duration of vaginismus. Women in the waiting-list group in that study were not educated about the problem at the assessment session, as they were in our study. In our treatment study (V) the duration of problems was 3.8 years (median). Our study included women with both total and partial, primary and secondary vaginismus, but women with partial vaginismus was the dominant group. Van Lankveld’s study (2006) included women with lifelong/primary vaginismus (TPV). It is reasonable to assume that women with partial secondary vaginismus have better ability for self-healing than women with total primary vaginismus, who have never been able to have intercourse.

It seems important to identify women with ability for self-healing, make a thorough assessment, give practical advice and education about the problem, and allow time for those women who have the capacity to heal without treatment. In cases of self-healing, the women achieved control on their own, of an important part of their body over which they earlier lacked control. This might be empowering for women with vaginismus. In some published studies, only one or two sessions were needed to treat some of the women with vaginismus who were included in the study (Lamont 1978; Jeng et al. 2006). Should these results be regarded as treatment success or as self-healing? Is it an ethical and good way of using resources to treat women with vaginismus who have a capacity for self-healing?

Methodological concerns

Participants

The groups studied have in all our studies been small, which increases the risk of Type II (β -) errors and makes the generalisation of the results difficult.

In Study III and IV significant differences were found despite small samples. There is, however, in both of those studies a need for replication in a larger study group, to see which findings can be confirmed.

We had no control over how the asymptomatic women in Study II and III selected themselves to the study as we made contact with them through advertisements. It is reasonable to believe that these women had a greater interest in pelvic floor function than ordinary young females

and by this a better ability to contract and relax those muscles. A possible difference between the symptomatic and the asymptomatic group in Study II was therefore not masked by the method of selection.

We based the exclusion criteria for women in the asymptomatic group in Study II and III on our assumptions of what the symptomatic group would look like. Based on literature we expected to find nulliparous women < 30 years old in the symptomatic group, but that was not the case. Maybe this is due to a selection by referral of complicated cases. Repeated calculations without women > 30 years old and without parous women in the symptomatic groups did not influence the results.

In Study V, a higher proportion of non-respondents than respondents reported ability to have technical intercourse at end of therapy. As results achieved at end of therapy seem to be maintained, it is not likely that loss of respondents gave falsely positive treatment outcome at follow-up. Some women in the non-respondent group have told us that after end of therapy they lived a life “without thoughts of their previous problem”, which might have resulted in unwillingness to answer a questionnaire.

Use of the small vaginal sensor

In both Study II and III we used the small vaginal sensor in part of the symptomatic sample. We found it unethical to use the larger vaginal sensor in this group of women who had earlier reported pronounced pain reactions during vaginal palpation. The size of the bud of the sensor is in both these studies of minor interest for the measurements. In Study II the stalk of the sensor (where the measurement plates are mounted) is of more interest and the difference in the diameters (2.5 mm) of the stalks of the two types of sensors is probably too small to influence the results of the sEMG recordings. In Study III the central point was whether the sensor was big enough to trigger some pain and itch, but not unbearable sensations of pain and itch in the groups studied. It is reasonable to assume that the decision to use the smaller vaginal sensor in some women did not lead to falsely increased differences between the symptomatic and the asymptomatic group.

sEMG of pelvic floor muscles

During the sEMG recordings all our participants were investigated in a lithotomy position, as this was the position we used for our clinical examinations during which we by digital examination had palpated a vaginismus reflex. A possible source of error could be that the lithotomy position created an increased tone in the pelvic floor muscles in all women, both symptomatic and asymptomatic; see above discussion on “sEMG measurement of pelvic floor muscles”, page 57.

sEMG measures depolarisations from several muscle units and we do not know to what extent the distance from the pelvic floor muscle to the sensor affects the measurements. It is difficult to conclude if the absence of statistically significant differences is due to methodological shortcomings of our study, as long as a “gold standard” of sEMG measurements in women with superficial coital pain does not exist.

Itch and burning pain

The measurements of burning pain and itch were performed in an unblinded way. We do not think it would have been possible to perform this study totally blinded. Women with pain

during vaginal penetration often behave in a different way than women without pain, even before a pelvic examination, and often react with withdrawal movements of the body during the investigation and elevation of the fourchette towards the clitoris. For a researcher who also works clinically with treatment of women with vaginismus, these findings are almost impossible not to register. At the moment of the study, the researcher (ME) was the only one in Sweden trained to use the sEMG equipment in this group of women.

For practical reasons we decided to use the Numerical Rating Scale (NRS) instead of the Visual Analog Scale (VAS) for the women's report of itch and burning pain. The validity of the NRSs has been well documented. They demonstrate positive and significant correlations with other measurements of pain intensity (Jensen and Karoly 2001).

In Study III we used clinical examination and wet smear but not scheduled vaginal cultures to exclude ongoing Candida infection. For clinical use, it is recommended that vaginal culture for Candidiasis is used if findings in wet smear are inconclusive (Sjoberg and Rylander 2003). In at least 20% of asymptomatic women a positive culture for Candidiasis is found (Drake and Maibach 1973; Sobel 1993; Sjoberg 1996), and "positive vaginal cultures may reflect colonization only and should not be used as the sole basis for diagnosis" (Sobel 1993). This means that if a vaginal culture had been taken, one fifth of our included women would theoretically have been excluded though they did not report symptoms and had no clinical findings of Candida infection. Women in the symptomatic group were examined several times, as they all took part in our treatment program. This ought to have diminished the risk for not diagnosing a subclinical Candida infection in this group.

Women in Study III used less incisive words for pain during penetration than women in Study IV. We do not know if the standardized penetration situation provoked less incisive sensations during penetration than intercourse had done.

Telephone interview of pain development history

Study IV is a retrospective self-report study, as no better method was found to study reports of pain symptoms at onset of the problem. There are several shortcomings with such a design (Jensen and Karoly 2001). Yet, pain intensity is a fairly homogeneous dimension, and one that is relatively easy for adults to identify and gauge (Jensen and Karoly 2001). But, if a person has more pain when she/he is asked to recall previous pain, she/he tends to rate her/his previous pain as more severe than if she/he presently has slight or no pain (Jensen et al. 1996; Jensen and Karoly 2001). According to these findings, the women in our study who were already treated or were in therapy might have reported less prominent pain symptoms from the early parts of their history. This might e.g. have resulted in a falsely low prevalence of reported pain after intercourse or at penetration among treated women.

Half of the participants had earlier met the interviewer during a consultation or during treatment for superficial coital pain. This might have influenced the women to answer the questions as they believed the interviewer wanted them to answer (desirability bias) (Saunders 1991). In all the interviews, there was also a risk of the interviewer interpreting the answers in the way she expected or wanted them to be. The solution to these problems would have been an independent and blinded interviewer. The disadvantage with such a solution is the risk of getting less detailed information, or high internal dropout rates due to the sensitive issues that are discussed.

Study IV included no reference group. The aim of the study was only to report on findings of different types of pain during the course of the problem of superficial coital pain in women with partial vaginismus with or without vulvar vestibulitis.

Long-term follow-up

Questions concerning self-worth and goal fulfilment are retrospective, and we do not know to what extent the women's present estimates of themselves influence their memories of self-worth before treatment.

In Study V, we did not use a Visual Analogue Scale (VAS) (Scott and Huskisson 1976; Jensen and Karoly 2001) to measure pain during intercourse at follow up; instead we decided to measure the women's reports of their ability to have intercourse without pain. This decision is in agreement with the intent of the CBT model used, where focus is on abilities rather than on remaining problems. If a woman with vaginismus is asked only for degree of pain in relation to intercourse we do not know if the pain is experienced merely during penetration and/or throughout the intercourse, and/or after intercourse. Moreover, we do not know if the pain is experienced during all or only during some of the intercourses. By use of "ability to have intercourse without pain", the woman is able to report in more terms general her own apprehension of this rather complex ability.

We also used the women's grading of fulfilment of their individual goals of therapy as measurement of treatment outcome in Study V. By this method several treatment outcome measurements biases could be reduced, e.g. goal of therapy defined by therapists; effect of therapy measured by therapists; effect of therapy measured as male performance (penetration and ejaculation); effect of therapy not measured in accordance with the women's own goals of therapy, e.g. ability to get pregnant.

Clinical implications

We suggest an increased attention to symptoms of postcoital burning pain in women with superficial coital pain. We do not yet know if postcoital burning pain is an early indicator of partial vaginismus with or without vulvar vestibulitis. If so, there might be a possibility for earlier treatment interventions than today.

There is a clinical value in regarding partial vaginismus and vulvar vestibulitis not as two mutually exclusive diagnoses but as two diagnoses, which often occur at the same time. By this approach, therapeutic approaches for both vaginismus and, if necessary, for vulvar vestibulitis (after the vaginismus has been treated) might be used.

In women with diagnostically unclear vulvar itch, the pelvic floor muscles have to be evaluated for the existence, or not, of increased tone and/or a partial vaginismus reflex.

Future perspectives

Further studies are needed:

- To study the relationship between partial vaginismus and vulvar vestibulitis.
- To understand the aetiology, pathophysiology, and maintaining factors of the increased muscle tone in women with partial vaginismus with or without vulvar vestibulitis.
- To understand the pathophysiology of pain and itch sensations related to the vaginismus reflex and to the increased muscle tone.
- To evaluate in a larger study where the retrospective period is made as short as possible, if postcoital burning pain has any value as an early indicator of partial vaginismus with or without vulvar vestibulitis.
- To understand why women with coital pain continue to have intercourse/attempted intercourse despite the pain.

There is also a need for well-designed randomised treatment studies of women with both total and partial vaginismus.

Future research of women with vaginismus requires detailed information of which diagnostic criteria for vaginismus are used in the studies.

In future studies of treatment of women with vaginismus, vaginal penetration cannot be assumed to be the desired outcome in all cases.

CONCLUSIONS

- Partial vaginismus was more common in our samples of women with superficial coital pain than total vaginismus.
- There was a great overlap of the clinical diagnoses of partial vaginismus and vulvar vestibulitis in our samples.
- sEMG of the pelvic floor muscles was of no value in our study in distinguishing women with partial vaginismus with or without vulvar vestibulitis from each other or from an asymptomatic group.
- sEMG of the pelvic floor muscles was a method with good test-retest reliability.
- Women with partial vaginismus with or without vulvar vestibulitis reported not only burning pain but also itch during a standardized penetration situation, while asymptomatic women did not.
- In women with partial vaginismus with or without vulvar vestibulitis who reported both burning pain and itch, the sensations of burning pain preceded the sensations of itch in most of the cases.
- During the course of developing painful coitus, women in our sample with partial vaginismus with or without vulvar vestibulitis more often had pain after intercourse than pain during penetration at the onset of the problem.
- Pain during penetration increased dramatically from onset of the problem to when the women ceased having intercourse in our sample of women with partial vaginismus with or without vulvar vestibulitis.
- Pain after intercourse in women with partial vaginismus with or without vulvar vestibulitis in our sample lasted for several hours and was described with words like burning and/or smarting, while pain during penetration was described as brief and with words like sharp/incisive/bursting.
- Pain during everyday life was described by up to one third of women in our sample with partial vaginismus with or without vulvar vestibulitis.
- Burning pain during micturition after intercourse was described by a majority of women in our sample with partial vaginismus with or without vulvar vestibulitis.
- Long-term follow-up results among women with superficial coital pain and vaginismus treated with a CBT program for vaginismus show in our study good treatment outcome.
- An ability to have intercourse at end of therapy was maintained at follow-up in our study among women with superficial coital pain and vaginismus treated with CBT for vaginismus.
- Fulfilment of treatment goals was reported by a majority of women in our study with superficial coital pain and vaginismus treated with a CBT program for vaginismus.
- Women with superficial coital pain and vaginismus treated with a CBT program for vaginismus reported an increase in self-worth as sex partner, woman and human being from assessment to follow-up.
- Every tenth women in our sample with superficial coital pain and vaginismus healed spontaneously after thorough assessment and while on waiting list.

SUMMARY IN SWEDISH

Svensk sammanfattning

Ytlig samlagssmärta är i dagsläget ett vanligt problem. En studie har visat att 13% av unga kvinnor i Sverige har haft svår samlagssmärta i mer än sex månader. Samlagssmärta delas vanligen upp i två huvudgrupper: samlagssmärta utan vidare definition och vaginism/slidkramp.

Vaginism finns i två huvudformer, total vaginism (TV) när slidan är helt stängd och partiell vaginism (PaV) när slidan bara är delvis stängd och samlag åtminstone till en början går att genomföra till pris av brännande smärta. Vaginism kan finnas från första samlaget (primär) eller komma efter en period av smärtfria samlag (sekundär). Nästan all tidigare forskning om vaginism har behandlat total primär vaginism (TPV), som oftast har sin grund i en rädsla för penetration och/eller smärta, medan bilden vid den mycket mindre studerade partiella vaginismen domineras av samlagssmärta. Vulva vestibulit (VVS) är den i dag vanligaste diagnosen hos unga kvinnor med ytlig samlagssmärta. Diagnoskriterierna för VVS är smärta vid penetration och punktömheter i vestibulum/förgården. Kvinnor med PaV och kvinnor med VVS, beskriver oftast sin samlagssmärta på ett liknande sätt, vilket gör gränsdragningen mellan dessa tillstånd oklar.

Studie I

Denna fallbeskrivning utgör en introduktion till själva avhandlingsarbetet.

I artikeln beskrivs symptomdebuten med brännande smärta efter samlag och smärtutvecklingen hos en kvinna som i tidigt skede fått diagnosen VVS. När kvinnan sökte oss hade hon anamnes och symptom typiska för en PaV som utvecklats till en TV. Kvinnan beskrev också symptom på en kontinuerlig brännande smärta och klåda i förgården. Utredning och behandling med kognitiv beteendeterapi beskrivs detaljerat. Under behandlingens gång får kvinnan också besvär med brännande smärta och klåda efter övningarna. Den brännande smärtan och klådan i dessa situationer tolkas som betingade responser till penetrationssituationen på samma sätt som slidkrampsreflexen i sig. Artikeln belyser diagnossvårigheterna inom området VVS och vaginism.

Studie II

Syftet med denna studie var att undersöka i vilken utsträckning kvinnor med ytlig samlagssmärta kunde diagnostiseras lida av både PaV och VVS, och att undersöka i vilken utsträckning yt-elektromyografi (ytEMG) av bäckenbottenmuskulaturen kunde särskilja kvinnor med bara PaV, kvinnor med PaV och VVS och kvinnor utan symptom på ytlig samlagssmärta.

Bland 224 kvinnor med ytlig samlagssmärta fann vi att 102 av 224 kunde diagnostiseras ha både PaV och VVS. Alla kvinnor med VVS hade slidkramp, medan 42 av 224 kvinnor hade PaV men inte VVS. I artikeln förs en diskussion om sambandet mellan PaV och VVS.

Vi undersökte 47 kvinnor med PaV varav 33 också hade VVS och 27 symptomfria kvinnor med ytEMG av bäckenbottenmuskulaturen. Vi fann ingen signifikant skillnad mellan de tre undersökta grupperna (PaV enbart, PaV+VVS och asymptomatiska kvinnor).

YtEMG av bäckenbottenmuskulaturen var i denna studie av ringa värde för att skilja grupperna åt och den ökade vilotonus i bäckenbottenmuskulaturen som man kan palpera hos kvinnor med vaginism skulle därför kunna vara av annat ursprung än elektrogen utlösta kontraktioner.

Studie III

I denna studie ville vi mäta de symptom på brännande smärta och klåda som vi kliniskt funnit hos många kvinnor med vaginism och som vi beskrivit i fallbeskrivningen i Studie I.

Femtiofyra kvinnor med PaV+/-VVS och 27 asymptomatiska kvinnor deltog i undersökningen. Vi använde oss även i denna studie av ytEMG av bäckenbottenmuskulaturen men den vaginala ytEMG sensorn användes endast som "standardiserad penetrator" och kvinnorna fick knipa och slappna av i bäckenbottenmuskulaturen enligt ett standardiserat program. De skattade också graden av brännande smärta och klåda vid 20 förutbestämda tillfällen under den standardiserade penetrationssituation.

Åttio procent (43/53) av kvinnorna med PaV+/-VVS beskrev en brännande smärta i underlivet och 43% (23/53) klåda under den standardiserade penetrationssituationen jämfört med 0% av kvinnorna i den asymptomatiska gruppen. Tjugotvå kvinnor med PaV+/-VVS beskrev båda symptomen. Hos 17 av dessa 22 kvinnor uppkom den brännande smärtan före klådan och i fyra fall uppkom symptomen samtidigt. Tidsskillnaden mellan uppkomsten av brännande smärta och klåda var 150s (median).

Både brännande smärta och klåda var i denna studie vanligt förekommande hos kvinnor med PaV. Klådreaktioner är vad vi känner till inte studerade tidigare i denna grupp och inte heller finns den tidsmässiga förskjutningen mellan uppkomsten av brännande smärta och klåda beskriven tidigare.

Studie IV

Syftet med denna studie var att utförligt beskriva smärtan vid penetration och efter samlag hos kvinnor med PaV; dels tidiga symptom och dels symptom när kvinnorna slutar att ha samlag. Vi använde oss av en semi-strukturerad telefonintervju med 24 kvinnor med PaV+/-VVS.

När problemet började var smärta efter samlag vanligare förekommande än smärta vid penetration. När kvinnorna slutade att ha samlag var båda symptomen lika vanliga. Intensiteten på smärtan vid penetration ökade kraftigt (från 0,5-9 på en skala från 0-10), från tidpunkten när problemen började till dess kvinnorna slutade att ha samlag. Intensiteten på smärtan efter samlag var hög redan från början (5) och ökade till sju till dess kvinnorna slutade att ha samlag. Smärtan vid penetration varade en minut och beskrevs som skarp/skärande/bristande, medan smärtan efter samlag beskrevs som brännande/svidande och varade i två timmar. Sjuttio procent av kvinnorna beskrev en brännande smärta när de kissade efter samlag, 30% beskrev klåda efter samlag eller i vardagslag. Frekvensen av båda dessa besvär var lika när symptomen började som när kvinnorna slutade att ha samlag. Brännande smärta i underlivet i vardagslag beskrevs av 22% av kvinnorna när besvären började och av 33% när de slutade att ha samlag.

De flesta tidigare studier har bara beskrivit smärtan vid penetration, särskilt hos kvinnor med VVS, men denna studie tyder på att brännande smärta *efter* samlag är ett vanligare symptom än penetrationssmärta när besvären börjar hos kvinnor med PaV+/-VVS.

Studie V

Syftet med denna studie var att presentera långtidsuppföljningsresultat av kognitiv beteendeterapi (KBT) i en serie av kvinnor med vaginism.

Från en grupp med 124 konsekutiva kvinnor med ytlig samlagssmärta och vaginism valde vi alla kvinnor som behandlats med KBT för vaginism (n=59) och skickade till dem ett frågeformulär angående långtidsuppföljningsresultat. Svarsfrekvensen var 44/59 (74,6%) och uppföljningstiden i medeltal 39 månader. Kvinnorna hade haft sina problem i nästan fyra år innan utredningen/behandlingen började, och hade behandlats under 16 sessioner (median).

Vid långtidsuppföljningen kunde 81% av de kvinnor som svarade på enkäten (indexfallen) ha samlag och 61% rapporterade en förmåga ≥ 6 (på en skala från 0-10) att ha samlag utan smärta och att njuta av samlag. Medianvärdet för förmåga att ha samlag utan smärta var åtta och för förmåga att njuta av samlag sju. Frekvensen av kvinnor med positiva behandlingsresultat varierade med 75%, från 81% som kunde ha samlag till 6% som kunde njuta av samlag och ha helt smärtfria samlag. En stor andel av indexfallen hade vid långtidsuppföljningen till stor del (≥ 6 på en skala från 0-10) uppfyllt sina individuella behandlingsmål, förmåga till samlag utan smärta (71%), förmåga till samlag utan rädsla (80%), förmåga att njuta av samlag (69%) och möjlighet att bli gravid (77%). Indexfallen värderade sig själva signifikant högre som sexpartners, kvinnor och människor vid långtidsuppföljningstillfället än vid tiden före första undersökningen på vår klinik.

Tolv procent av kvinnorna i hela materialet med vaginism läkte utan behandling efter en noggrann undersökning och information om problemet.

Vi fann i denna studie att kvinnor med ytlig samlagssmärta och vaginism som fått behandling med KBT (16 sessioner) uppvisade goda behandlingsresultat vid en långtidsuppföljning. Resultaten var emellertid i hög grad associerade till definitionen av behandlingsresultatet. Två tredjedelar av kvinnorna hade till stor del uppfyllt sina mål med behandlingen.

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