GENERAL

CHAPTER

General introduction
Maaike, 23 years: “Intercourse has always been painful to me. Every time we have intercourse, it hurts really bad. I mentioned it to the physician once, but he couldn't find anything wrong on the exam. He advised us to use some lubricant, but this didn't seem to help. After a while, I found myself avoiding to have sex with my boyfriend. A few weeks ago, we tried it again, but it was impossible to take him inside me. I feel like a failure. I know my boyfriend is very patient and never insists to have intercourse, but I don't want to disappoint him over and over again. Am I destined to be this way forever?”

Sandra, 34 years: “I used to enjoy sex, but about three years ago, intercourse really began to hurt. It burns and stings when we try to have intercourse. It also hurts when I insert tampons. Over time, I became more and more anxious that intercourse would hurt again and it usually did. Now it seems that my body just tightens up. The pleasure has gone, and instead, there is only the prospect of discomfort and frustration. Many nights I cry when we are done. What else can I do then gritting my teeth and bear the pain? I feel very inadequate and insecure. Our relationship is suffering, and I'm afraid this is going to tear us apart.”

The stories of Maaike and Sandra are quite representative for women who suffer from painful intercourse. When intercourse hurts persistently and there is no apparent physical pathology explaining the pain, the term dyspareunia is used to describe this condition. Dyspareunia represents a common health problem in women. As illustrated above, for many affected women intercourse may become a troublesome, frustrating or even aversive activity. Apart from the distressing interference of pain with sexuality, dyspareunia may also have repercussions on the woman's individual and relational well-being. To date, little is known about the underlying mechanisms of dyspareunia and, therefore, treatment is challenging.

The purpose of the present thesis was to gain more insight into the factors involved in the onset and/or maintenance of this poorly understood condition. This may not only be of theoretical interest, but may also provide new clues for improving available treatment modalities.
Below, a brief overview on dyspareunia will be presented, followed by an introduction of the prevailing cognitive-behavioral model on dyspareunia. It will be presented in the light of current general models on sexual (dys)function. Finally, an outline of the present thesis is provided.

**CLINICAL DESCRIPTION AND DIAGNOSIS**

Dyspareunia means, literally translated from ancient Greek, bed partners not fitting together. Nowadays, dyspareunia is defined as recurrent or persistent acute pain associated with penile-vaginal intercourse (Basson et al., 2003). In the Diagnostic and Statistical Manual of Mental Disorders, fourth edition text revision (DSM-IV-TR), dyspareunia is classified as a sexual dysfunction (American Psychiatric Association, 2000). There is currently considerable controversy about whether dyspareunia should be reconceptualised as a pain disorder rather than a sexual dysfunction (e.g., Binik, 2005). Dyspareunia can afflict men and women, but is less common in men. This thesis will deal with dyspareunia in women.

Dyspareunia is a diagnosis of exclusion arrived at only when physical illness or abnormalities that cause pain on (attempted) vaginal penetration, such as vaginal infections, cystitis, atrophia, adhesions, dermatologic diseases, scar tissue, and allergic reactions to hygiene products have not been found (see for an overview, Weijmar Schultz, Basson, Binik, Eschenbach, Wesselmann, & van Lankveld, 2005).

Two types of dyspareunia are distinguished: pain felt deep inside the pelvis during penile thrusting (deep dyspareunia), and pain experienced at the vaginal introitus (i.e., superficial dyspareunia). As the vast majority of symptomatic women reports superficial dyspareunia, this form was chosen as the dyspareunia subtype to be investigated in this thesis.

Vulvar vestibulitis syndrome (VVS) is believed to be the most common form of superficial dyspareunia in premenopausal women (Meana, Binik, Khalifé, & Cohen, 1997; Harlow, Wise, & Stewart, 2001). Women with VVS experience a sharp/burning pain at the entrance of the vagina in response to contact or pressure to the vulvar vestibule. This pain can be provoked by sexual and nonsexual activities (e.g., tampon insertion, sitting, cycling or tight clothing). Its etiology is unknown and there are no generally accepted associated physical findings (Lotery, McClure,
The diagnosis of VVS has been established according to the following criteria: 1) severe pain on vestibular touch or attempted vaginal entry; 2) tenderness in response to pressure on the vulvar vestibule, and 3) physical findings limited to vulvar erythema (i.e., redness of the skin) (Friedrich, 1987). However, as vulvar erythema is a normal finding in women with and without vulvar pain conditions (Bergeron, Binik, Khalifé, Pagidas, & Glazer, 2001a), the International Society for the Study of Vulvovaginal Diseases (ISSVD) has recently proposed to remove this criterion (Edwards, 2004; Haefner, 2007). Furthermore, the ISSVD has suggested to delete “vestibulitis” because the presence of inflammation, as implied by this term, has been debated (Lotery et al., 2004), and, instead, to rename VVS as "provoked vestibulodynia." During the period in which the present research project was conducted, the term VVS and Friedrich’s diagnostic criteria were universally accepted, and therefore, they have been used consistently throughout this thesis. It should be noted that there is still considerable confusion in the literature regarding the nomenclature and classification of dyspareunia. As a consequence, studies differ in their selection criteria for symptomatic women to be included, whereas other studies do not fully specify these criteria. For this reason, it is to date impossible to meaningfully distinguish between subtypes when describing the state of the art regarding prevalence, treatment and etiology of dyspareunia.

PREVALENCE AND INCIDENCE

Estimates of the prevalence vary considerably, depending on the differences in the definitions of dyspareunia that are used, subtypes of dyspareunia included in the study, the survey methods, and the population that was studied (e.g., with or without VVS). International studies have reported prevalence rates between 3 and 18% in the general population (e.g., Simons & Carey, 2001; Harlow et al., 2001), and between 15 and 20% in gynaecological practice (Goetsch, 1991). Twenty-six percent of female patients at sexology outpatient clinics in the Netherlands receive treatment for dyspareunia, which makes it the most prevalent problem at Dutch sexology outpatient clinics (Kedde, 2007).

While women of all ages report dyspareunia, it has a peak incidence in nulliparous
women in their twenties and thirties (Simons & Carey, 2001). In the present thesis exclusively premenopausal women were investigated.

**TREATMENT**

A variety of medical and non-medical treatment modalities are applied in attempts to relieve the genital pain (see for an overview, Farage & Galask, 2005; Meana & Binik, 1994; Bergeron, Binik, Khalifé, & Pagidas, 1997; Weijmar Schultz et al., 2005). Medical interventions include local use of topical anaesthetics or corticosteroid ointments, and surgery in the case of VVS (e.g., laser therapy or vestibulotomy [removal of painful vestibular tissue]). Non-medical treatments include physical therapy (e.g., electromyographic biofeedback) and cognitive-behavioral therapy (CBT). Physical therapy aims mainly at control over and relaxation of the pelvic floor. The primary foci of CBT-programs are on pain management and improvement of sexual functioning (especially enhancement of sexual arousal).

Knowledge about the effectiveness of these treatments is scarce. Most treatments claim success (Bergeron et al., 1997), although these claims are based on methodologically flawed research. Until now, there are only a handful of randomized trials (Bornstein, Livnat, Stolar, & Abramovici, 2000; Bergeron et al., 2001b; Danielsson, Torstensson, Brodda-Jansen, & Bohm-Starke, 2006; Nyirjesy, Sobel, Weitz, Leaman, Small & Gelone, 2001). Two of these studies compared the effectiveness of drug treatment (fluconazole and cromolyn cream, respectively) versus placebo and found that women who received drug treatment were not significantly more improved than women who received placebo (Bornstein et al., 2000; Nyirjesy et al., 2001). In another study, a treatment of electromyographic biofeedback was compared to topical lidocaine (Danielsson et al., 2006). No differences in outcome for both treatments were obtained, with an average of 11% of women considering themselves completely cured. In the randomised controlled study of Bergeron and colleagues (2001b) the effects of vestibulotomy, biofeedback, and CBT were compared. Success rates (in terms of partial or complete pain relief) for biofeedback, CBT, and vestibulotomy were 34.6%, 39.3% and 68.2%, respectively. Two-and-a-half year later, vestibulotomy and CBT appeared to be similarly effective in terms of self-reported pain during intercourse (Bergeron, Khalifé, Glazer, & Binik, 2008),
suggesting that women with dyspareunia/VVS can benefit from both medical and behavioural interventions. However, the above success rates also indicate that there are still many patients whose symptoms are not relieved after treatment. These moderate success rates may well be explained by the fact that the currently available treatment modalities rely on suspected, but largely unsubstantiated, etiological factors. Hence, in order to optimize health care to patients, it is of paramount importance to identify the factors involved in the onset and maintenance of dyspareunia.

**ETIOLOGY**

Despite the wide-ranging list of factors that has been proposed to explain the etiology of dyspareunia, the mechanisms underlying dyspareunia are still largely unknown. For a long time, research was guided by either a biomedical or psychological approach. The majority of studies has focused primarily on biomedical factors, which can be roughly divided into one of the following categories: vestibular pathology, genetic vulnerability, and central pain modulatory dysfunction (see for an overview Farage & Galask, 2005; Weijmar Schultz et al., 2005). Psychological causes that have been proposed are, for instance, negative feelings about sexuality, fear of sex, sexual traumata, relational conflicts, general psychopathology, and specific personality dimensions. Many of these biomedical and psychological factors are derived from clinical case reports, or lack a theoretical framework. Furthermore, research into the etiological candidates has been plagued by methodological flaws (e.g., lack of control groups, unvalidated instruments, lack of objective tests of physical conditions, vague inclusion criteria, or inclusion of heterogeneous samples) and was cross-sectional in nature. All these aspects may have obscured a clear understanding of the factors playing a role in the onset and maintenance of dyspareunia.

Apart from the abovementioned factors, there is almost universal consensus among theoreticians and clinicians that insufficient lubrication due to lack of sexual arousal and pelvic floor hypertonicity may be critically involved in the development of dyspareunia (e.g., Bancroft, 1989; Binik, 2005; de Jong, van Lunsen, Robertson, Stam, & Lammes, 1995; Kaplan, 1974; Meana & Binik, 1994; van Lunsen &
Ramakers, 2002; Spano & Lamont, 1975; ter Kuile & Weijenborg, 2006; Weijmar Schultz et al., 2005). Correspondingly, physical therapy and CBT aim at reducing pelvic floor muscle tension and enhancing sexual arousal. Despite the general acceptance of these etiological candidates, there is, surprisingly, a paucity of controlled, objective assessments of (genital) sexual arousal and pelvic floor function in women with dyspareunia compared to asymptomatic women. To gain more insight into the causal and maintaining factors of dyspareunia, a good theory or model that would enable specific and testable predictions is indispensable. This brings us to the presentation of the prevailing CBT-model on dyspareunia, in which lack of sexual arousal and pelvic floor muscle tension are incorporated.

EXPLANATORY MODEL OF DYSPAREUNIA

Already more than thirty years ago, Spano and Lamont (1975) introduced a circular model of dyspareunia in which it was assumed that pain during penetration, or memories of that pain, lead to fear of pain in new intercourse situations, which, in turn, results in decreased sexual arousal during sexual activity and therefore results in vaginal dryness and/or increased pelvic floor muscle tone (as a protective reaction to anticipated or actual pain). The combination of vaginal dryness and increased pelvic floor muscle tone, causing friction between the penis and vulvar skin, may result in pain and even tissue damage (VVS). This damage may in itself result in pain or may further increase already existing pain. According to this model, diminished sexual response and/or increased pelvic floor muscle tone can be a cause, as well as a consequence of the genital pain complaint (see Figure 1).

Spano and Lamont’s model (1975) has considerably influenced the contemporary CBT-programs for dyspareunia (see e.g., ter Kuile & Weijenborg, 2006). In conjunction with this model, CBT-programs focus on pain, fear of pain, and their hypothesized adverse impact on sexual desire, arousal and vaginal lubrication on the one hand, and pelvic floor muscle function on the other.

Empirical evidence for this model, however, is scarce. The few studies that have examined pelvic floor muscle function in women with dyspareunia/VVS point to pelvic floor pathology (Glazer, Jantos, Hartmann, & Swencionis, 1998; Reissing, Binik, Khalifé, Cohen, & Amsel, 2004; Reissing, Brown, Lord, Binik, & Khalifé, 2005;
Additional evidence for Spano & Lamont's hypothesis that fear leads to enhanced pelvic floor muscle activity stems from two experimental studies in which vaginal surface EMG was assessed in women with vaginismus and sexually functional women (van der Velde, & Everaerd, 2001; van der Velde, Laan, & Everaerd, 2001). These studies showed that exposure to threatening film clips (either with or without a sexual content) resulted in both groups of women in significantly increased pelvic floor muscle activity as compared to a sexual (without threat) or a neutral film clip. Thus far, it has not been investigated whether these data can be extended to women with dyspareunia.

A few observational studies investigated the relationship between dyspareunia and sexual arousal problems and showed that women with dyspareunia consistently reported lower sexual arousal (e.g., Meana and Binik, 1994; Reissing, Binik, Khalifé, Cohen, & Amsel, 2003; Wouda, Hartman, Bakker, Bakker, van de Wiel, & Weijmar

**Figure 1.** An adapted version of Spano & Lamonts (1975) circular model of dyspareunia (ter Kuile, Bolle, & Weijenborg, 2004).
Schultz, 1998) and lubrication than controls (Nunns & Mandall, 1997; van Lankveld, Weijenborg, & ter Kuile, 1996; Masheb, Lozano-Blanco, Kohorn, Minkin, & Kerns, 2004). Moreover, an inverse relationship between genital pain and lubrication has been documented (Wiegel, Meston, & Rosen, 2005). As such, these data suggest an important role for sexual arousal problems in the etiology and/or maintenance of superficial dyspareunia. However, self-assessment of sexual arousal indices through interviews or questionnaires may be distorted by inaccurate perceptions, or, particularly in sexually dysfunctional individuals, may be overly pessimistic (e.g., Rowland, 1999). Furthermore, these studies do not allow for definitive conclusions about the causal status of these associations.

Only one study directly investigated sexual arousal in women with dyspareunia by means of an objective measure (vaginal photoplethysmography). Wouda et al. (1998) observed similar levels of genital arousal during exposure to non-coital film fragments in women with and without dyspareunia, but reduced levels of genital arousal in symptomatic women during exposure to an intercourse film fragment. The authors argued that exposure to a depiction of coitus had an inhibiting effect on genital response because intercourse had become negatively associated with pain. These findings support Spano and Lamont’s hypothesis that fear of pain results in diminished genital arousal.

The specific model of dyspareunia (Spano & Lamont, 1975) bears striking resemblance with Barlow’s eminent cognitive-behavioral model on sexual functioning (1986), as both models assume that fear1 (or anxiety in terms of Barlow’s model) impairs sexual arousal (Barlow, 1986; Sbrocco & Barlow, 1996). Although some other critical components of Barlow’s model have received considerable empirical support, the impact of anxiety on sexual arousal in women remains unclear. Whereas some studies reported facilitative effects on genital responding (Hoon, Wincze, & Hoon, 1977; Palace & Gorzalka, 1990), but not on subjective sexual arousal (Palace & Gorzalka, 1990), other studies demonstrated a decrease in

1The distinction between fear and anxiety is theoretically based on the focus of threat. Fear is the emotional reaction to a specific, identifiable and immediate threat, such as a dangerous animal or an injury (Rachman, 1998). Anxiety, in contrast to fear, is a future-oriented affective state and the source of threat is more elusive without a clear focus. Clinically, the distinction between fear and anxiety is less evident, and these terms are frequently used interchangeably.
genital responding and subjective sexual arousal (Beggs, Calhoun, & Wolchik, 1987; Both, Everaerd, & Laan, 2003; Laan, Everaerd, & Evers, 1995) and still other studies did not found any effect at all (Elliott & O’Donohue, 1997; Sipski, Rosen, Alexander, & Gomez-Marin, 2004).

Also, in the information processing model of sexual arousal as proposed by Janssen, Everaerd, Spiering, & Janssen (2000), fear has been hypothesized to adversely influence sexual arousal. In short, in this model it is assumed that fearful associations with sex in sexually dysfunctional individuals are already present at an automatic (unconscious) level. Hence, for these individuals the confrontation with a sexual stimulus may lead to a negative appraisal of that stimulus, which, in turn, may impede both genital and subjective sexual arousal responses. Yet, the existence of automatic fear-related associations in women with dyspareunia or other sexual dysfunctions has not been empirically tested.

In sum, there is a strong emphasis on the role of fear and diminished sexual arousal in the onset and maintenance of dyspareunia in particular (Spano & Lamont, 1975) and sexual dysfunctions in general (Barlow, 1986; Janssen et al., 2000). Offering an attractive theoretical framework, Spano & Lamont’s circular model of dyspareunia (1975) functioned as the major guide for the present research project. The main research question of this thesis was: are fear of pain and diminished sexual arousal key components in dyspareunia? To address this question, five studies were designed. These studies and our hypotheses will be briefly presented below.

**OUTLINE OF THE PRESENT THESIS**

This thesis can be divided into three sections, based on the methodology used to investigate the role of fear and sexual arousal in dyspareunia. In all studies, premenopausal women with complaints of superficial dyspareunia (either with or without a concomitant diagnosis of VVS) were compared to women without sexual complaints.

**Psychophysiological research**

Chapter 2, 3 and 4 present experimental studies that included psychophysiological measurements of sexual arousal in response to exposure to visual sexual stimuli. To
measure genital arousal, vaginal photoplethysmography was used. The vaginal photoplethysmography, originally developed by Sintchak and Geer (1975), monitors changes in vasocongestion of the vagina, one of the earliest and most reliable correlates of genital arousal in women (Masters & Johnson, 1966). Vaginal lubrication is presumed to be the result of the passage of blood plasma in the capillaries through the vaginal epithelium, due to the increased pressure inside the capillaries during vasocongestion (Levin, 1992). This instrument has proven to provide a valid measure of genital response in women and is the most widely used physiological method for assessing female genital arousal responses. It is easy to use and has been demonstrated to be both sensitive and specific to sexual arousal (e.g., Laan, Everaerd, & Evers, 1995). Reports on genital sensations and affect in response to the sexual stimuli were obtained as indices of subjective sexual arousal, the second component of sexual arousal. Both components of sexual arousal were assessed because there is little agreement between genital arousal and reported feelings of sexual arousal in women (e.g., Chivers, Seto, Lalumière, Laan, & Grimbos, subm.; Laan & Everaerd, 1995; Laan & Janssen, 2007).

Chapter 2 reports on a study that was conducted analogous to the study by Wouda et al. (1998). That is, genital arousal and subjective responses to different visual sexual stimuli (noncoital and coital scenes) were compared. Exposing participants to both noncoital and coital film excerpts enabled us to explore the following questions: are women with dyspareunia characterized by a generally impaired genital responsiveness such that sexual arousal responses are lower to both kinds of sexual stimuli? Or do women with dyspareunia exhibit a conditioned fear response such that sexual arousal responses are lower only to stimuli that may induce fear of pain (i.e., coitus)? In line with Wouda et al.’s findings (1998), we hypothesized that women with dyspareunia would show equal levels of genital arousal as women without sexual complaints to the erotic stimuli depicting non-coital sex. When presented with a coitus scene, however, we expected women with dyspareunia to react with decreased genital response, whereas controls were expected to display a further increase in genital response. Women with dyspareunia were predicted to report overall less positive and more negative affect than the control women. In addition, the dyspareunia group was hypothesized to report more fear in exposure to the coitus stimulus than to the non-coital stimulus.
Chapter 3 presents a study that directly manipulated fear of pain to investigate its effects on genital arousal and subjective reports. To induce fear of pain, participants were told that they had a 60% chance of receiving painful stimuli while being exposed to one of two erotic film clips. It was hypothesized that fear of pain would adversely affect genital arousal and subjective reports in both women with and without dyspareunia. Based on research pointing to hypervigilance to pain in women with dyspareunia with VVS (Payne, Binik, Amsel, & Khalifé, 2005), we expected fear of pain to result in a more pronounced decline in genital arousal in symptomatic women. Furthermore, it was expected that the dyspareunia group would report overall less positive and more negative affect than controls. Finally, it was examined whether affected women with or without VVS differed on the dependent variables.

In Chapter 4 a study is described that investigated whether the appraisal of one and the same sexual stimulus can be successfully manipulated by providing discrepant information regarding that stimulus prior to viewing it. The information had either a focus on genital pain or sexual enjoyment. The focus on genital pain was chosen as an analogue of the expectation of experiencing pain during coitus as assumed to exist in women with dyspareunia, whereas the sexual enjoyment instruction was used as an analogue to the expectation of experiencing enjoyment during coitus in sexually functional women. A neutral instruction served as a control condition. In line with the information processing model on sexual arousal (Janssen et al., 2000), it was hypothesized that information with a focus on genital pain would result in lower levels of genital arousal and subjective reports as compared to information with a focus on sexual enjoyment.

**Indirect measure**

The study described in Chapter 5 tested the hypothesis that automatic (in the sense of fast, unintentional) fear-related associations with sexual stimuli are involved in superficial dyspareunia. According to the information processing model of sexual arousal (Janssen et al., 2000), automatically elicited associations are assumed to be related to genital responses (e.g., lubrication) whereas deliberate, controlled associations are linked to the experience of sexual excitement and further strategic/planned behaviour. As it has been argued that automatic fear responses may be best predicted by indirect measures (e.g., Egloff & Schmuckle, 2002), the
purpose of this experiment was to design and implement an indirect measure. Indirect measures have already been successfully used to assess disorder-relevant automatic associations in a range of psychological complaints, including pain (Vancleef, Peters, Gilissen, & de Jong, 2007) and specific fears (e.g., Teachman, Gregg, & Woody, 2001).

We preferred the Affective Simon Task (AST) (De Houwer & Eelen, 1998) above other implicit tasks because the AST does not, in contrast to other related tasks, require a contrast category relative to which the results should be interpreted. Furthermore, contrary to other tasks, the AST can be used to examine associations involving subcomponents of a concept, here “sex”. This enabled us to select both noncoital and coital stimuli in order to investigate whether negative sex-related associations in women with dyspareunia would be restricted to penetration stimuli or whether they would exist for sexual stimuli in general. In addition to automatic sex-related associations, self-report ratings of the stimuli were assessed as an index of deliberate sex-related associations.

**Self-report measures**

In Chapter 6 an observational study is presented, which was designed to systematically collect information regarding cognitive-affective evaluations of sexual stimuli, sexual functioning, and the experience of pain by means of validated questionnaires. Furthermore, this study was conducted to determine which of the discriminating variables would best predict group membership (dyspareunia or control). Finally, this study directly compared dyspareunia subgroups with or without VVS to find out whether these different subgroups are associated with different psychological profiles.

**Summary and Discussion**

Chapter 7 provides a summary of the main findings followed by a discussion of the findings in relation to theory and other relevant findings in this field. After considering some methodological limitations, implications of the findings for future research and clinical practice will be presented.