CAN VAGINISMUS BE DISCRIMINATED FROM DYSPAREUNIA?

A TEST OF THE PROPOSED DSM-5

GENITAL PAIN/PENETRATION DISORDER CRITERIA

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April 2012

A thesis submitted to McGill University in partial fulfillment of the requirements of the degree of Doctor of Philosophy

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ABSTRACT

Current empirical research does not support the DSM-IV-TR definition of vaginismus as well as its diagnostic distinction from dyspareunia. This has resulted in a DSM-5 proposal to redefine and collapse vaginismus and dyspareunia under one category named "Genito-Pelvic Pain/Penetration Disorder". Fear has, however, been proposed as a possible differentiator between vaginismus and dyspareunia/PVD. The primary goal of this thesis was, therefore, to examine how well fear could differentiate vaginismus from dyspareunia/PVD. In the first chapter of this thesis, a literature review is included to examine the prevalence, classification/diagnosis, etiological factors and treatment of vaginismus. This review reveals that: 1) vaginal spasm is not a valid or reliable diagnostic criterion for vaginismus; 2) genital pain is an important characteristic of most women suffering from vaginismus; 3) vaginismus cannot be easily differentiated from dyspareunia/PVD; 4) fear is an under investigated factor that appears to characterize women with vaginismus. In the second chapter entitled "Can Vaginismus be Discriminated from Dyspareunia? A Test of the Proposed DSM-5 Genital Pain/Penetration Disorder Proposal", fear, measured through self-report, behavioral and physiological indices, is examined in terms of how well it discriminates 50 women suffering from vaginismus, 50 women suffering from dyspareunia/PVD and 43 controls. Genital pain, vaginal muscle tension, sexual functioning and childhood sexual and physical abuse are also re-examined as possible factors differentiating vaginismus and dyspareunia/PVD. Fear, particularly behavioral measures of fear, and vaginal

muscle tension were found to discriminate the vaginismic group from the dyspareunia/PVD and control groups while genital pain discriminated well both clinical groups from controls. Despite significant statistical differences on fear and vaginal muscle tension between vaginismus and dyspareunia/PVD, a large overlap was observed which may explain the great difficulty health professionals have to reliably discriminate both conditions. Overall, this body of work provides evidence for fear, vaginal muscle tension and genital pain being important characteristics of vaginismus supporting the DSM-5 proposal of adding these characteristics in the definition of vaginismus. It further supports the importance of multidisciplinary assessment and treatment interventions for vaginismus including gynecologists, physiotherapist and sex therapists. Whether vaginismus and dyspareunia/PVD should be collapsed into one category as proposed for the DSM-5 is further discussed in terms of its diagnostic and treatment advantages and disadvantages.

RÉSUMÉ

La définition du vaginisme selon le DSM-IV-TR ainsi que sa distinction avec la dyspareunie ne sont pas appuyées par les recherches empiriques actuelles. Ceci a résulté en une proposition de combiner et de redéfinir le vaginisme et la dyspareunie sous une catégorie intitulée "Désordre de Douleur/Pénétration Génito-Pelvien". La peur a toutefois été proposée comme un facteur pouvant possiblement différencier le vaginisme de la dyspareunie/DVP. Le but principal de la présente dissertation était par conséquent d'examiner si la peur pouvait distinguer le vaginisme de la dyspareunie/DVP. Le premier chapitre de la présente dissertation comprend une revue de littérature dans le but d'examiner la prévalence, la classification/diagnostique, les facteurs étiologiques ainsi que les traitements du vaginisme. Cette revue de littérature démontre que: 1) le spasme vaginal n'est pas un critère valide et fiable pour le vaginisme; 2) la douleur génitale est une caractéristique importante de la majorité des femmes souffrant de vaginisme; 3) le vaginisme et la dyspareunia/DVP ne peuvent être facilement distingués; 4) la peur est un facteur sous-investigué qui semble caractériser les femmes souffrant de vaginisme. Le second chapitre s'intitule "Can Vaginismus be Discriminated from Dyspareunia? A Test of the Proposed DSM-5 Genital Pain/Penetration Disorder Proposal" et comprend une étude empirique examinant si la peur mesurée à l'aide d'auto-évaluation, de comportements et d'indices physiologiques peut distinguer 50 femmes souffrant de vaginisme, 50 femmes souffrant de dyspareunie/DVP et 43 contrôles. La douleur génitale, la tension musculaire vaginale, la fonction sexuelle ainsi que les expériences dans

l'enfance d'abus sexuel et/ou physique ont été réexaminées comme facteurs pouvant également différencier le vaginisme de la dyspareunie/DVP. Les résultats ont démontré que la peur, plus spécifiquement les comportements de peur, et la tension musculaire vaginale ont différencié significativement le groupe de femme souffrant de vaginisme, du groupe de femme souffrant de dyspareunie/DVP et du groupe contrôle. La douleur génitale a quant à elle distinguée clairement les deux groupes cliniques (vaginisme et dyspareunie/DVP) du groupe contrôle. Malgré les différences démontrées dans la présente étude entre le vaginisme et la dyspareunie/DVP, un chevauchement important a été observé ce qui peut venir expliquer la grande difficulté qu'éprouvent les professionnels de la santé à distinguer de manière fiable ces deux conditions. Dans l'ensemble, cet ouvrage fournit des preuves que la peur, la tension musculaire vaginale et la douleur génitale sont des caractéristiques importantes du vaginisme et par conséquent appuie en partie la proposition pour le DSM-5 d'inclure ces caractéristiques dans la définition du vaginisme. Cet ouvrage supporte également l'importance de traitement multidisciplinaire pour le vaginisme comprenant gynécologue, physiothérapeute et sexologue/psychologue. La proposition effectuée pour le DSM-5 de combiner le vaginisme et la dyspareunie/DVP sous une catégorie est davantage discutée en termes de ces avantages et de ces inconvénients au niveau du diagnostique et du traitement de ces conditions.

CONTRIBUTION OF AUTHORS

This thesis is comprised of two papers. The first consists of a literature review written in collaboration with Stephanie C. Boyer, Rhonda Amsel, Dr. Samir Khalifé, and Dr. Yitzchak M. Binik. The second paper is co-authored by myself, Stephanie C. Boyer, Rhonda Amsel, Dr. Samir Khalifé and Dr. Yitzchak M. Binik. The following is a statement regarding the contributions of the various authors to the two papers.

Most of the literature review was researched, written, and revised by me; Stephanie Boyer helped in the search of articles and co-authored the treatment section while Rhonda Amsel, Dr. Khalife and Dr. Binik served in an editorial capacity.

The second paper resulted from a study that I formulated, developed, conducted, analyzed and wrote in collaboration with Stephanie C. Boyer, Dr. Khalifé and Dr. Binik. Stephanie Boyer helped in the development and validation of the behavioral observation system, and in the data collection and analysis. Dr. Binik served as an advisor during the elaboration of research questions and development of the protocol, and in an editorial capacity during the writing of the manuscript. Dr. Khalifé performed the gynecological examination. Rhonda Amsel served as a statistical consultant and assisted in the methodological development of the study as well as in an editorial capacity.

STATEMENT OF ORIGINAL CONTRIBUTION

This dissertation is a manuscript-based thesis comprised of two chapters that provide original contribution to the field of vaginismus. The first chapter entitled "Vaginismus: A review of the literature on the classification/diagnosis, etiology and treatment" was published in 2010 in *Women's Health, 6(5), pp.705-719.* This manuscript provides a thorough and up-to-date review of the research evaluating prevalence, classification/diagnosis, etiology and treatment vaginismus published through 2009, in addition to proposing a future perspective. It further discusses the DSM-5 proposal of collapsing vaginismus and dyspareunia under one category named "Genito-Pelvic Pain/Penetration Disorder". The results from this paper indicate that: 1) The current definition of vaginismus is not supported by empirical evidence; 2) Genital pain is an important characteristic of vaginismus; 3) Vaginismus and dyspareunia are difficult to differentiate; 4) Fear is an under investigated factor that appears to characterize women with vaginismus.

The second chapter entitled "Can Vaginismus be Discriminated from Dyspareunia? An investigation of the Proposed DSM-5 Genital Pain/Penetration Disorder Proposal" was submitted to *Archives of Sexual Behavior.* This manuscript provides the first empirical investigation of fear using a variety of measurement methods including self-report, a blinded behavioral observation system, and physiological indices while women with vaginismus, dyspareunia/PVD and controls are undergoing a gynaecological examination. It is further the first manuscript to investigate whether the differences found

between vaginismus and dyspareunia/PVD on fear, vaginal muscle tension, and genital pain are dimensional or categorical. The results from this study provide evidence that: 1) Fear and vaginal muscle tension appear to characterize women with vaginismus and to distinguish them from women with dyspareunia/PVD and controls; 2) Although fear and vaginal muscle tension were found to statistically distinguish vaginismus from dyspareunia, a large overlap was observed between both conditions on fear, vaginal muscle tension, and genital pain; 3) Vaginismus, as currently diagnosed, is a multifactorial condition comprising of fear, genital pain, and vaginal muscle tension.

ACKNOWLEDGEMENTS

The present dissertation as well as my passage through graduate school would not have been possible without the incredible help, support and knowledge of many dedicated members, the first of whom is my supervisor, Dr. Yitzchak Binik. There are not enough words to thank my supervisor who has made available his support in numerous ways. His extraordinary patience, encouragement, empathy, and knowledge gave me the strength to pursue and complete my doctoral studies. I will forever be thankful. He has been one of my greatest source of support and understanding in my search for a balance between family and graduate school. Thank you for being so generous Irv.

In addition to my supervisor, I would like to show my sincere gratitude to Dr. Samir Khalifé, an exceptional obstetric gynecologist who demonstrated great empathy and professionalism to the participants volunteering in my doctoral research project. I wish to thank Monique Khalifé, Francis Shaugnessy and all the staff at Dr. Khalifé's clinic who were very helpful and welcoming and made my testing days so much more pleasant. I wish to thank dearly Rhonda Amsel for her incredible patience, statistical guidance, emotional support, and encouragement. She has truly been an amazing source of support and advice during the writing process of my thesis. Rhonda, I am really grateful for all of your help in this extraordinary statistical journey.

To my dearest lab mates, Seth Davis, Alina Kao, Dr. Tuuli Kukkonen, Dr. Melissa Farmer, Dr. Nicole Flory, Laurel Paterson, Dr. Kimberley Payne, Sabina

Sarin, thank you for your extremely helpful feedback, advice, friendship, understanding, and generosity. You were all important sources of motivation which allowed me to complete my doctoral studies. I was surrounded by incredible labmates who made my journey in graduate school so much more enjoyable and fun. I wish to offer my deepest gratitude to Dr. Caroline Pukall for her supervisory assistance, moral support and guidance.

My doctoral research project would not have been possible without several generous and brilliant assistants who worked numerous hours including Alessandra Asousa, Natalie Cartwright, Jackie Huberman, Anita Kapuscinski, Caroline Maykut, Katherine Muldoon, Olga Pinkhasov, Anton van Hamel, and Christina Yager. Jackie, thank you so much for having been so helpful in the writing process of my thesis. A special thank you to Stephanie Boyer for her dedication to this work; she was extremely helpful at various stages of my research project and made the work in this thesis possible.

I wish to express my sincere appreciation for the grants and scholarships received in support of the project from the McGill University Health Consortium, and le Fonds de la Recherche en Santé du Quebec.

To all of my friends, especially Mélanie Therrien, Émilie Clec'h, Julie Grenier, Caroline Ménard, Marie-Frédérique Montel, Florence Marcil-Denault, and Brigitte Marsh, I wish to thank you for believing in me at moments when I doubted myself. Thank you for your emotional support, encouragement and

motivation. Your words of encouragement and your understanding gave me the strength to pursue until the end.

This thesis is dedicated to my beloved family for their unconditional love and support. Thank you for having always believed in me and for having offered me the best in life. To my parents, you are a source of inspiration for me, thank you for loving me the way you do and for being so present in my life, ‘’I love you’’. Finally, to my dearest husband, the love of my life, Maxime Frappier, I am forever grateful for your understanding, encouragement, incredible patience and optimism. Without you I would not have had the energy and motivation to undergo and complete my doctoral studies. We are forming a truly amazing team; I consider myself extremely fortunate to have met you. To my two adored and beautiful sons who were born during my doctoral studies, you are my greatest gifts and successes in life. Every time I think of you, I get a boost of energy and love. I wish to thank you for your love, your smiles, and your affection which always gave me strength and courage.

THESIS INTRODUCTION

Vaginismus and dyspareunia are listed as the two main "sexual pain disorders" in the DSM-IV-TR. Vaginismus is defined as "involuntary muscle spasms of the outer third of the vagina" while dyspareunia is defined as "genital pain associated with sexual intercourse" (APA, 2000). There is a recent debate as to whether vaginismus and dyspareunia would be better classified under one category or remain separate conditions (see *Binik 2010 in Archives of Sexual Behavior*). The first manuscript included in this dissertation entitled "Vaginismus: a review of the literature on the classification/diagnosis, etiology and treatment", published in *Women's Health, 6(5), pp.705-719* addresses this debate by reviewing the research evaluating the classification/diagnosis, etiology and treatment of vaginismus published through 2009. The review raises important questions regarding the role of fear in the diagnosis of vaginismus and in its ability to distinguish vaginismus from dyspareunia.

The second chapter included in this dissertation is entitled "Can Vaginismus be Discriminated from Dyspareunia? An investigation of the Proposed DSM-5 Genital Pain/Penetration Disorder Proposal". This chapter presents findings from a clinical research study which examines whether measures of fear can distinguish vaginismus from dyspareunia/PVD (provoked vestibulodynia, the most common form of superficial pre-menopausal dyspareunia). The study also examines whether genital pain, vaginal muscle tension, sexual and physical abuse, and sexual functioning can distinguish vaginismus from dyspareunia/PVD.

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Lahaie, M.-A., Boyer, S. C., Binik, Y. M., Amsel, R., & Khaflifé, S. (2010). Vaginismus: a literature review. *Women's Health*, *6*, 705-719.

VAGINISMUS: A REVIEW OF THE LITERATURE ON THE CLASSIFICATION/DIAGNOSIS, ETIOLOGY AND TREATMENT

Published in Women's Health

Reference:

Lahaie, M-A., Boyer, S.C., Amsel, R., Khalife, S., & Binik, Y.M. (2010). Vaginismus: a review of the literature on the classification/diagnosis, etiology and treatment. *Women's Health, 6*(5), 705-719

Abstract

Vaginismus is currently defined as an involuntary vaginal muscle spasm interfering with sexual intercourse that is relatively easy to diagnose and treat. As a result, there has been a lack of research interest with very few well-controlled diagnostic, etiological or treatment outcome studies. Interestingly, the few empirical studies that have been conducted on vaginismus do not support the view that it is easily diagnosed or treated and have shed little light on potential etiology. A review of the literature on the classification/diagnosis, etiology and treatment of vaginismus will be presented with a focus on the latest empirical findings. This review suggests that vaginismus cannot be easily differentiated from dyspareunia and should be treated from a multidisciplinary point of view.

Keywords:

Vaginal Muscle Spasm, Vulvar Pain, Dyspareunia, Provoked Vestibulodynia, Fear of Vaginal Penetration, Sexual Abuse, Psychological Treatments, Pelvic Floor Physiotherapy, Pharmacotherapy

Introduction

Vaginismus is described as an involuntary vaginal muscle spasm interfering with sexual intercourse (APA, 2000). Since the term was first coined in the 19th century, vaginismus has been conceptualized as a relatively infrequent but well-understood and easily treatable female sexual dysfunction. In 1859, gynecologist J. Marion Sims wrote the following: "From personal experience, I can confidently assert that I know of no disease capable of producing so much unhappiness to both parties of the marriage contract, and I am happy to state that I know of no serious trouble that can be cured so easily, so safely, and so certainly" (p. 361). This conceptualization was perpetuated by Masters and Johnson who reported a treatment outcome success rate of 100% (Masters & Johson, 1970). It seems likely that this presumed high cure rate and lack of diagnostic controversy deterred new research. In fact, Beck described vaginismus as "an interesting illustration of scientific neglect" (p.381) (Beck, 1993).

Since Reissing et al's review of the vaginismus literature, a few important empirical studies on the diagnosis and treatment of vaginismus have been published (Reissing, Binik,& Khalife, 1999). Interestingly, their results challenge the validity of the current definition of vaginismus as well as the notion that it is an easily diagnosable and treatable condition. The current review will examine the literature on the classification/diagnosis, etiology and treatment of vaginismus with a focus on the latest empirical findings.

Prevalence

There are no epidemiological studies examining the population prevalence of vaginismus. This may be true since such a study would probably require a stressful gynecological examination that sufferers might often prefer to avoid. As a result, there have been dramatically varying estimates regarding the prevalence of this problem. Some like Masters and Johnson claim that it is a relatively rare condition (e.g., Masters & Johnson, 1970, Schmidt & Arentewicz, 1982) while others suggest that it is one of the most common female psychosexual dysfunctions (e.g., Simons & Carey, 2001; Crowley, Richardson,& Goldmeier, 2006; Kabakçi & Batur, 2003; McGuire & Hawton, 2001). Although the population prevalence remains unknown, the prevalence rates in clinical settings have been reported to range between 5-17% (Spector & Carey, 1990).

In a British study, Ogden and Ward examined the help-seeking behaviours of women suffering from vaginismus and found that the professional most frequently consulted was the general practitioner (Ward & Ogden, 1994). Unfortunately, their respondents reported that general practitioners were the least helpful health professional they consulted. Overall, there was general dissatisfaction with available help which may reinforce many vaginismic women's pre-existing avoidance in seeking help. This is consistent with Shifren et al's findings in the US that only one third of women with "any distressing sexual problem" consult (Shifren et al., 2009). According to their sample, the barriers for receiving professional help were poor self-perceived health and embarrassment in discussing sexual problems.

Classification and Diagnosis

Vaginal Muscle Spasm

In her 1547 treatise on "The Diseases of Women", Trotula de Salerno is thought to have provided the earliest description of what we today call vaginismus: "a tightening of the vulva so that even a woman who has been seduced may appear a virgin" (Trotula of Salerno, 1940). Much later, Huguier gave the first medical description of the syndrome; however, it appears that Sims first coined the term "vaginismus" in 1862 while addressing the Obstetrical Society of London (Huguier, 1834). Sims described vaginismus as "an involuntary spasmodic closure of the mouth of the vagina, attended with such excessive supersensitiveness as to form a complete barrier to coition" (p.362) (Sims, 1861). To date, the involuntary muscle spasm remains the core element of the definition of vaginismus suggested by the American College of Obstetrics and Gynecology (ACOG) and by the Diagnostic and Statistical Manual of Mental Disorders-IV-TR (DSM-IV-TR) (ACOG, 1995; APA, 2000). The International Classification of Diseases-10 (ICD-10) categorizes vaginismus either as a "pain disorder" or as a "sexual dysfunction comprised of a spasm of the pelvic floor muscles that surround the vagina, causing the occlusion of the vaginal opening with penile entry being either impossible or painful" (WHO, 1992).

This 150-year consensus concerning the definition of vaginismus is striking given the lack of empirical findings validating the vaginal muscle spasm criterion (Reissing et al., 1999). In fact, Reissing et al (N= 87) found that although vaginismic women demonstrated a greater frequency of vaginal muscle spasm

while undergoing a gynecological examination than did age, relationship and parity matched healthy controls or women suffering from dyspareunia associated with provoked vestibulodynia (PVD), only 28% of the vaginismus group actually displayed a vaginal muscle spasm. Moreover, only 24% reported experiencing spasms with attempted intercourse. Even more puzzling was the finding that two independent gynecologists agreed only 4% of the time on the diagnosis of vaginismus (Reissing, Binik, Khalife, Cohen,& Amsel, 2004). These findings call into question the primary diagnostic criterion of vaginismus.

Another method of evaluating the validity of the vaginal muscle spasm criterion is via the electrical recording of muscle activity which can be done through surface (sEMG) or needle electromyography. Recent sEMG and needle EMG studies have investigated the activity of the pelvic floor muscles in women diagnosed with vaginismus. Reissing et al found that women with vaginismus displayed lower pelvic floor muscle strength and greater vaginal/pelvic muscle tone compared to matched controls but no significant differences at all between the vaginismus and PVD group (Reissing et al., 2004; Reissing, Brown, Lord, Binik,& Khalife, 2005). Shafik and El-Sibai (N= 14) also demonstrated through needle EMG a higher EMG activity at rest and on induction of the vaginismus reflex in the levator ani, puborectalis and bulbocavernosus muscles in women with vaginismus compared to age matched controls (Shafik and El-Sibai, 2002). Consistent with the findings above, Frasson et al (N=30) found significant needle EMG basal and reactive hyperexcitability in primary lifelong vaginismus and in women with PVD accompanied by vaginismus as compared to controls (Frasson

et al., 2009). On the other hand, three well-controlled sEMG (Ns ranging from 29 to 224) studies did not confirm a significant difference in ability to contract and relax the pelvic floor muscles between women with and without vaginismus (Van der Velde & Everaerd, 2001; Van der Velde, Laan,& Everaerd, 2001; Engman, Lindehammar,& Wijma, 2004).

These contradictory results may be partially explained by the lack of an operationalized definition of the term muscle spasm as well as the lack of consensus regarding which muscles are involved in vaginismus. Some authors refer to broad groups of muscles such as the muscles of the outer third of the vagina, the pelvic muscles or the circumvaginal and perivaginal muscles (e.g., Abrahams, 1977; Van Lankveld, Brewaeys, Ter Kuile,& Weijenborg, 1995; Fertel, 1977; Van de Wiel, 1990; Poinsard, 1968; Lamont, 1994), while others refer to more specific ones such as the bulbocavernosus, the levator ani, and puboccoccygeus (e.g., Steege, 1984; Binik, 2010). None of these studies indicate how they concluded which muscles are involved (Reissing et al., 1999). The term spasm itself is also controversial as there is no agreement on whether spasm refers to an involuntary muscle cramp, a defensive mechanism or a hypertonicity of the pelvic floor muscles.

In addition to the lack of agreement regarding the term muscle spasm and the muscles involved in vaginismus, there is no empirically standardized diagnostic protocol for vaginal muscle spasm. Although Masters and Johnson claimed that a pelvic exam was necessary to diagnose vaginismus, researchers and clinicians have frequently relied on self-report of difficulties with vaginal

penetration (Sims, 1861; IASP, 1994). The lack of a standardized diagnostic protocol is not a trivial problem since studies concerning vaginismus may well include highly diverse samples. The fact that studies using the vaginal muscle spasm DSM-IV-TR definition of vaginismus failed to find a vaginal spasm suggests that vaginal muscle spasm is not a reliable diagnosis and as a result diverse patient populations might have been included (Frasson et al., 2009; Van der Velde & Everaerd, 2001; Van der Velde et al., 2001; Engman et al., 2004).

Pain

Even though vaginismus is classified as a sexual pain disorder in the DSM-IV-TR, pain is not mentioned in the diagnostic criteria. Other definitions of vaginismus such as those published by the ACOG (ACOG, 1995), the International Association for the Study of Pain (IASP), the World Health Organization and Lamont do mention pain in their definitions (WHO, 1992; IASP, 1994; Lamont, 1978). However, no description of the pain characteristics, such as location, quality, intensity, and duration are provided (Binik, 2010). There is also a lack of information regarding whether the pain is a cause or consequence of the vaginal muscle spasm (Binik, 2010). While most clinical reports and research concerning vaginismus do not make reference to the pain element in vaginismus (Har-Toov, Militscher, Lessing, Abramov,& Chen, 2001), some authors believe that pain is one of its core components (McGuire et al., 2001; Spector & Carey, 1990; Ogden & Ward, 1995; Shifren et al., 2009; Trotula of Salerno, 1940; Huguier, 1834; ACOG, 1995; WHO, 1992; Reissing et al., 2004; Har-Toov et al., 2001; Kaneko, 2001; Payne, Bergeron, Khalife,& Binik, 2005). In

fact, several studies have found that a large percentage of women suffering from vaginismus experience pain with attempted vaginal penetration (Reissing et al., 2004, Abrahams, 1977; Engman, 2001; Kaneko, 2001; Ter Kuile, Van Lankveld, Vlieland, Wilekes,& Weijenborg, 2005; Basson, 1996; De Kruiff, Ter Kuile, Weijenborg,& Van Lankveld, 2000; Engman, Wijma,& Wijma, 2008). The pain experienced by women with vaginismus has been found to be very similar to the pain reported by women with PVD (see below for definition) (Reissing et al., 2004; Ter Kuile et al., 2005; De Kruiff et al., 2000).

According to the DSM-IV-TR, vaginismus can be classified as either lifelong (primary) or acquired (secondary). It has often been suggested that PVD may result in acquired vaginismus (Steege, 1984; Lamont, 1978; Fordney, 1978). Although lifelong and acquired vaginismus are generally considered to differ in their etiology and response to treatment, there are no empirical data validating these claims.

Differential Diagnosis of Vaginismus from Dyspareunia

According to the DSM-IV-TR, there are two mutually exclusive sexual pain disorders: vaginismus and dyspareunia. Dyspareunia is defined as "recurrent genital pain associated with sexual intercourse" (p. 556, APA, 2000). PVD is reported to be the most frequent subtype of dyspareunia in pre-menopausal women with a prevalence of 7% in the general population (Meana, Binik, Khalife,Cohen, 1997; Harlow, Wise,& Stewart, 2001). Women with PVD typically experience a severe, sharp, burning pain upon vestibular touch or attempted vaginal entry (Meana et al., 1997; Friedrich, 1987; Bergeron, Binik, Khalife,

Pagidas,& Glazer, 2001). It is diagnosed through the cotton-swab test, which consists of the application of a cotton-swab to various areas of the vulvar vestibule and surrounding tissue (Friedrich, 1987).

Despite the fact that vaginismus and dyspareunia associated with PVD have been portrayed as two distinct clinical entities, they have many overlapping characteristics such as the elevated vulvar pain and vaginal/pelvic muscle tone (Reissing et al., 2004, De Kruiff et al., 2000). In fact, a number of studies have demonstrated that a large percentage (range between 42 to 100%) of women with vaginismus also meet the criteria for PVD (Reissing et al., 2004; Engman et al., 2004; Basson, 1996; De Kruiff et al., 2000). This may in part explain why health practitioners (i.e., gynecologists, physical therapists, and psychologists) show significant difficulties reliably differentiating vaginismus from PVD (Reissing et al., 2004). It should be noted, however, that PVD is characterized as superficial dyspareunia. The pain of deeper dyspareunia is usually easily differentiable from that associated with vaginismus. Women with vaginismus, however, were found to display significantly higher levels of emotional distress while undergoing a gynecological examination and to avoid significantly more sexual and non-sexual vaginal penetration attempts as compared to women with PVD (Reissing et al., 2004; Kaneko, 2001; De Kruiff et al., 2000).

Fear

Clinical reports have long suggested that fear plays an important role in vaginismus (e.g., Masters & Johnson, 1970; ACOG, 1995; Friedrich, 1987; Bergeron et al., 2001; Kaplan, 1974; Tugrul & Kabakçi, 1997). Only a few studies

have investigated this further (Tugrul & Kabakçi, 1997; Silverstein, 1989; Kennedy, Doherty,& Barnes, 1995). For example, fear of pain was the primary reason reported by women with vaginismus for their abstinence as well as the core motive underlying their avoidance of sexual intercourse (Reissing et al., 2004; Ward & Ogden, 1994). Moreover, a large percentage (range between 74% to 88%) of women with vaginismus report significant fear of pain during coitus (Tugrul & Kabakçi, 1997; Ward & Ogden, 1994). Women suffering from vaginismus share a number of characteristics with individuals suffering from a "specific phobia." Specific phobias are defined as "marked and persistent fear that is excessive or unreasonable, cued by the presence or anticipation of a specific object or situation" (APA, 2000). Individuals with a specific phobia will experience feelings of anxiety, fear, or panic upon encountering the feared object or situation. As a result, they will tend to actively avoid direct contact with the phobic stimulus (APA, 2000). Women with vaginismus report fear of vaginal penetration and associated pain and display high levels of emotional distress during vaginal penetration situations, such as during gynecological examinations (Reissing et al., 2004; Tugrul & Kabakçi, 1997). Women with vaginismus also tend to avoid situations involving vaginal penetration (i.e., gynecological examination, tampon insertion, and sexual intercourse) (Reissing et al., 2004).

It still remains unknown, however, whether vaginismic women avoid these particular situations in order to diminish their anxiety level like individuals suffering from a specific phobia or in response to their pain experience or both. Nonetheless, the avoidance of vaginal penetration cannot be solely explained by

the experience of pain since women with dyspareunia, who also experience severe pain during vaginal penetration, have not been shown to avoid vaginal penetration situations as much as women suffering from vaginismus (Reissing et al., 2004; De Kruiff et al., 2000). Although fear appears to be a promising factor that characterizes women with vaginismus, the existing empirical studies lack appropriate control groups, standardized instruments to measure fear, and appropriate statistical analysis (Tugrul & Kabakçi, 1997; Silverstein, 1989; Kennedy et al., 1995; War & Ogden, 1994).

Summary

The current definition of vaginismus is problematic. First, the vaginal muscle spasm criterion has never been empirically validated and it appears that vulvar pain and the fear of pain or of vaginal penetration characterizes most women currently diagnosed with vaginismus. Moreover, vaginismus cannot be reliably differentiated from superficial dyspareunia. A recent consensus definition reflects these conclusions and defines vaginismus as: "persistent or recurrent difficulties of the woman to allow vaginal entry of a penis, finger, and/or any object, despite her expressed wish to do so. There is variable (phobic) avoidance, involuntary pelvic muscle contraction, and anticipation /fear/experience of pain. Structural or other physical abnormalities must be ruled out or addressed" (Weijmar Schultz et al., 2005). Binik has also recently proposed a new conceptualization that combines vaginismus and dyspareunia into a single genito-pelvic pain/penetration disorder characterized by persistent or

recurrent difficulties for 6 months or more with at least one of the following: 1. inability to have vaginal intercourse/penetration on at least 50% of attempts; 2. marked genito-pelvic pain during at least 50% of vaginal intercourse/penetration attempts; 3. marked fear of vaginal intercourse/penetration or of genito-pelvic pain during intercourse/penetration on at least 50% of vaginal intercourse/penetration attempts; 4. marked tensing or tightening of the pelvic floor muscles during attempted vaginal intercourse/penetration on at least 50% of occasions (Binik, 2010).

Etiological Factors

Psychological Factors

Although the definition, diagnosis and treatment of vaginismus have focused largely on the organic symptom of vaginal muscle spasm, the proposed etiological factors have primarily been psychogenic. The most frequently proposed include negative sexual attitudes, psychological and/or physical trauma and relationship difficulties.

Negative Sexual Attitudes and Lack of Sexual Education

The associations between negative sexual attitudes, sexual ignorance and vaginismus have been frequently mentioned in the vaginismus literature (e.g., APA, 2000; Silverstein, 1989; Audibert & Kahn-Nathan, 1980). For example, Ellison claimed that vaginismus primarily resulted from: a lack of sexual knowledge and the presence of sexual guilt both leading to a fear of engaging in intercourse (Ellison, 1968; Ellison, 1972). These are consistent with, Silverstein,

Ward et al and Basson's conclusion that women suffering from vaginismus hold negative views about sexuality in general and about sex before marriage (Basson, 1996; Silverstein, 1989; Ward & Ogden, 1994). However, all these studies suffer from a number of important methodological limitations such as small sample sizes (Ns = 22-89), lack of appropriate statistical analyses and control groups, as well as absence of standardized measurement instruments, and a standardized protocol to diagnose vaginismus (Basson, 1996; Silverstein, 1989; Ward & Ogden, 1994; Ellison, 1969; Ellison, 1972). There are only two etiological studies of vaginismus which have included a standard statistical analysis or a control group (Dubble, 1977; Reissing, Binik, Khalife, Cohen,& Amsel, 2003) and only one that used a standardized measurement instrument (Biswas & Ratnam, 1995); their results do not support the notions that women with vaginismus hold negative sexual attitudes and/or have lower levels of sexual knowledge and education.

Relationship Factors

Vaginismus has frequently been reported to result from a dysfunctional couple relationship (Biswas & Ratnam, 1995; Van de Wiel, 1990). The available empirical evidence is controversial. For example, Tugrul and Kabakci's (N=40) uncontrolled study demonstrated that 85% of vaginismic women who applied for the treatment of vaginismus and 90% of their husbands evaluated their marriages as satisfactory (Tugrul & Kabakçi, 1997). Hawton and Catalan (N=30) found that couples suffering from vaginismus have a significantly better relationship and communication when compared to 76 couples presenting other

types of female sexual dysfunctions (Hawton & Catalan, 1990). Although relationship factors have not been empirically demonstrated to play a significant role in the etiology of vaginismus, women who suffer from vaginismus do have fewer sexual relations and avoid more sexual contact when compared to healthy controls (Tugrul & Kabakçi, 1997; Reissing et al., 2003). It remains unclear, however, whether these are causes or consequences of vaginismus.

Partners of women with vaginismus have been reported in clinical reports to suffer from sexual dysfunction as well as to display passive and unassertive personalities (Masters & Johnson, 1970; Van Lankveld et al., 1995; Silverstein, Ellison, 1972; Dawkins & Taylor, 1967; Friedman, 1962; O'Sullivan, 1979). Controlled empirical findings using standardized instruments evaluating type of personalities and male sexual dysfunction, however, have not supported this view (Van Lankveld et al., 1995; Kennedy et al., 1995; Duddle, 1977). For example, when the personality characteristics of male partners of women with vaginismus are compared to controls or norms, no differences were demonstrated. Moreover, the few studies that investigated the chronology of sexual dysfunction in partners of women with vaginismus concluded that sexual dysfunction such as erectile and premature ejaculation are generally the result rather than the cause of vaginismus (Lamont, 1994; Friedman, 1962; Barnes, 1986; Harrison, 1996).

Sexual and/or Physical abuse

Although the experience of sexual and/or physical abuse is generally considered an important etiological factor in vaginismus, the empirical evidence

is less conclusive (APA, 2000; Biswas & Ratnam, 1995; Leiblum, 2000). Five out of six studies (Hawton & Catalan, 1990; O'Sullivan, 1979; Barnes, 1986; Van Lankveld, Brewaeys, Ter Kuile,& Weijenborg, 1995; Van Lankveld et al., 2006) found no evidence of a higher prevalence of sexual and physical abuse. The sixth study found only weak evidence since women with vaginismus were twice as likely to report a history of childhood sexual interference (attempts at sexual abuse and sexual abuse involving touching) as compared to a "no pain" group (Reissing et al., 2003). Larger studies with matched control groups and well validated definitions of abuse are required to resolve this issue.

Biological Factors

Organic Pathology

A number of organic pathologies (e.g., hymeneal and congenital abnormalities, infections, vestibulodynia, trauma associated with genital surgery or radiotherapy, vaginal atrophy, pelvic congestion, endometriosis, vaginal lesions and tumors, scars in the vagina from injury, childbirth, or surgery, and irritation caused by douches, spermicides, or latex in condoms) resulting in painful/difficult/impossible vaginal penetration have been suggested as etiological factors (Reissing et al., 1999; Crowley et al., 2006; ACOG, 1995; Leiblum, 2000; Abramov, Wolman,& Higgins, 1994). There have been no controlled studies evaluating this possibility.

Pelvic Floor Dysfunction

Pelvic floor muscle dysfunction (e.g., hypertonicity, reduced muscle control) has been suggested as a predisposing factor in the development of vaginismus (Rosenbaum, 2005; Meana et al., 1997). Barnes, Bowman,& Cullen's (1984) uncontrolled study (N=5) argued that vaginismic women had difficulty evaluating vaginal muscle tone and as a result experienced problems distinguishing between a relaxed state and a spasm. It remains unclear, however, whether pelvic floor dysfunction is a predisposing factor or the defining symptom. To date, no controlled longitudinal studies have investigated the role of pelvic floor muscle dysfunction in the etiology of vaginismus.

Summary

Although a long list of psychological factors have been proposed as playing a role in the etiology of vaginismus, very few have been supported by empirical research. In addition, no biological factors hypothesized to be involved in the development of vaginismus have been adequately investigated.

Treatment

There has been much controversy over the treatment of choice for vaginismus. Sims recommended a surgical intervention which consisted of the removal of the hymen, the incision of the vaginal orifice, and subsequent dilatation (Sims, 1861). Soon thereafter, the need for a surgical procedure was questioned given that dilatation alone appeared to result in favorable outcomes

(Reissing et al., 1999; Von Scanzoni, 1867; Thorburn, 1885). Walthard, who conceptualized vaginismus as a phobic reaction to an excessive fear of pain, was one of the first to recommend psychotherapy (Walthard, 1909). Throughout the early 20th century, psychoanalysis was often prescribed following the notion that vaginismus was a hysterical or conversion symptom (Fenichel, 1945; Musaph & Haspels, 1976). In the 1970's, Masters and Johnson greatly I nfluenced the treatment of sexual dysfunction, in general, and reported that vaginismus could be easily treated with behaviorally-oriented sex therapy which included vaginal dilatation (Masters & Johnson, 1970). The success rates for the various treatments, ranging from vaginal dilatation to psychoanalysis to behaviorally-oriented sex therapy were always reported to be excellent. Current treatments for vaginismus can be divided into four main categories: pelvic floor physiotherapy, pharmacological treatments, general psychotherapy and sex/cognitive behavioral therapy. Table 1 summarizes the treatment outcome studies of vaginismus.

Pelvic Floor Physiotherapy

The rationale for the use of pelvic floor physiotherapy in the treatment of vaginismus is that it will aid in developing awareness and control of the vaginal musculature as well as restore function, improve mobility, relieve pain and overcome vaginal penetration anxiety (Rosenbaum, 2005; Barnes et al., 1984; Rosenbaum, 2008). Physical therapists use a variety of techniques to achieve these goals such as breathing and relaxation, local tissue desensitization, vaginal dilators, pelvic floor biofeedback, and manual therapy techniques (Rosenbaum, 2005; Rosenbaum, 2005; Barnes et al., 1984; Rosenbaum, 2008). To date, there

are two studies with 100 % success rates that have investigated the efficacy of biofeedback in the treatment of vaginismus (Barnes et al., 1984; Seo, Choe, Lee,& Kim, 2005). Unfortunately, they have very small sample sizes (Ns less than 12) and lack appropriate control groups (Barnes et al., 1984; Seo et al., 2005). In addition, one study had only 6 month follow-up with the success rate dropping to 60 % (Barnes, 1986; Barnes et al., 1984). Considering the importance accorded to the vaginal muscle spasm component in vaginismus, it is surprising that pelvic floor physiotherapy has not been investigated more extensively.

Pharmacological treatment

Three main types of pharmacological treatment have been proposed for vaginismus: local anesthetics (e.g., lidocaine), muscle relaxants (e.g., nitroglycerin ointment, botulinum toxin), and anxiolytic medication (Hassel, 1997; Peleg, Press,& Ben-Zion, 2001; Mikhail, 1976; Plaut & RachBeisel, 1997; Brin &Vapnek, 1997; Ghazizdeh & Nikzad, 2004; Shafik & El-Sibai, 2000; Bertolasi et al., 2009). Local anesthetics such as lidocaine gel have been proposed based on the rationale that vaginismic muscle spasms are due to repeated pain experienced with vaginal penetration, and, hence, the use of a topical anesthetic aimed at reducing the pain is hypothesized to resolve the spasm (Hassel, 1997). Its efficacy has been reported only in a case study in which a 5% lidocaine gel was applied on the hyperesthetic areas of the vaginal introitus of a 17 year old women suffering from primary vaginismus. A topical nitroglycerin ointment, hypothesized to treat the muscle spasm by relaxing the vaginal muscles, was

also discussed only in a case study (Peleg et al., 2001). A Muslim Bedouin couple presenting with primary vaginismus were able to engage in a satisfactory sexual relationship following the application of a topical nitroglycerine ointment (Peleg et al., 2001). Given that all the available information is in the form of case studies, no firm conclusion can be reached.

Botulinum toxin, a temporary muscle paralytic has been recommended in the treatment of vaginismus with the aim of decreasing the hypertonicity of the pelvic floor muscles (Brin & Vapnek, 1997). In Shafik and El-Sibai's (2000) treatment study (N=13), women with vaginismus who received an injection of botulinum toxin were able to engage in "satisfactory intercourse" as compared to no improvement in a control group receiving saline injections. The successful outcome persisted for an average follow-up of 10.2 months. Nonetheless, there are a number of limitations to this promising study such as the small sample size, lack of information on how vaginismus was diagnosed and lack of independent determination of treatment outcome. A recent treatment outcome study (N=39) demonstrated that women with vaginismus secondary to PVD who received repeated injections of botulinum neurotoxin type A into the levator ani displayed improvements on standardized measurements of sexual activity (i.e., the Female Sexual Functioning Index), on possibility of having sexual intercourse, on levator ani EMG hyperactivity and on bowel-bladder symptoms (Bertolasi et al., 2009). After a 39 month follow-up, 63.2% of their participants had completely recovered from vaginismus and PVD, 15.4% still needed some injections, 15.4% had dropped out and the remaining had not completed the treatment protocol.

Another pharmacological treatment which has been proposed is the use of anxiolytics such as diazepam in conjunction with psychotherapy based on the hypothesis that vaginismus is a psychosomatic condition resulting from past trauma and thus, anxiety-reducing medication will resolve the symptoms. Mikhail's uncontrolled study found that the administration of intravenous diazepam during psychological interviews in 4 women with vaginismus resulted in successful intercourse (Mikhail, 1976). Unfortunately, conclusions concerning the pharmacological treatment of vaginismus are limited because most studies lack appropriate placebo control groups and do not randomly assign patients to treatment, are based on small samples or do not use standardized outcome instruments.

General psychotherapy

A variety of psychological treatments for vaginismus have been investigated including marital, interactional, existential-experiential, relationship enhancement and hypnosis (Kennedy et al., 1995; Elkins, Johnson, Ling,& Stovall, 1986; Gottesfeld, 1978; Harman, Waldo,& Johnson, 1994; Kleinplatz, 1998; Rosen & Leiblum, 1995; Pridal & LoPicollo, 1993; Ben-Zion, Rothschild, Chudakov,& Aloni, 2007; Delmonte, 1988). The psychological treatments are often based on the notion that vaginismus results from marital problems, negative sexual experiences in childhood or a lack of sexual education. The therapy can be conducted in an individual or couple format. Generally, in individual therapy, the treatment is to identify and resolve underlying psychological problems that could

be causing the disorder. In couple's therapy, vaginismus is conceptualized as a problem for the couple and the treatment tends to focus on the couple's sexual history and any other problems that may be occurring in the relationship. Although the reported success rates are high (78-100%), all except two are case studies with poorly designed and described treatment interventions as well as a lack of information on how vaginismus was diagnosed. The two reports which are not case studies lack appropriate control groups and have no follow-up data (Kennedy et al., 1995; Ben-Zion et al., 2007).

Sex / Cognitive-Behavioral Therapy

In the 1970's, Masters and Johnson reported that vaginismus could be easily treated with behaviorally-oriented sex therapy that included vaginal dilatation (Masters & Johson, 1970). The first step of their treatment consists of the physical demonstration of the vaginal muscle spasm to the patient (and her partner) during a gynecological examination. The couple is then instructed to insert a series of dilators of graduated sizes at home guided by both the patient and her partner with the aim of desensitizing the patient to vaginal penetration. Masters and Johnson's treatment regimen also emphasized the importance of education regarding sexual function and the development and maintenance of vaginismus in order to relieve the psychological impact of the condition. As a result of the influence of Masters and Johnson, several studies were conducted on the efficacy of sex therapy in the treatment of vaginismus with excellent success rates reported resulting in continued utilization of this treatment for

vaginismus (Hawton & Catalan, 1990; Chakrabarti & Sinha, 2002; Grillo & Grillo, 1980; Jeng, Wang, Chou, Shen,& Tzeng, 2006; O'Sullivan & Barnes, 1978; Oystragh, 1988; Ng, 1993; Fuchs, 1980; Wijma & Wijma, 1997; Wijma, Janson, Nilson, Halbook,& Wijma, 2000; Schnyder, Schnyder-Lüthi, Ballinari,& Blaser, 1998; Biswas & Ratnam, 1995; Scholl, 1988). These studies were, however, uncontrolled (Hawton & Catalan, 1990; Grillo & Grillo, 1980; Jeng et al., 2006; O'Sulliva & Barnes, 1978; Fuchs, 1980; Biswas & Ratnam, 1995; Ter Kuile, et al., 2009) or case studies (Chakrabarti & Sinha, 2002; Oystragh, 1988; Ng, 1993; Wijma & Wijma, 1997; Wijma et al., 2000) and all presented important methodological flaws such as lack of waiting list control group and of standardized measurements to evaluate treatment outcome as well as elevated or unreported drop-out rates**.**

The first ever randomized controlled therapy outcome study for vaginismus was recently published. This study investigated a cognitive-behavioral sex therapy for the treatment of vaginismus (Van Lankveld et al.,2006). The treatment included the sexual education and vaginal dilatation technique as in Masters and Johnson's treatment protocol. It was also comprised of cognitive therapy, relaxation, and sensate focus exercises. Participants received the treatment for three months either in group therapy or in bibliotherapy format. At post-treatment, 18% (14% group therapy; 9% bibliotherapy) of participants in the treatment group reported successful attempted penile-vaginal intercourse while none of the women in the waiting list control group reported having had successful intercourse. Interestingly, there was no significant

difference in efficacy between the group therapy and bibliotherapy treatment format. At three month and one-year follow-ups, 19% of the participants in the cognitive behavioral sex therapy group and 18% in the bibliotherapy group had achieved intercourse.

Although the rate of successful outcome was far below what was expected based on previous non randomized controlled treatment outcome studies, internal analyses of the data suggested that successful outcome was mediated by changes in fear of coitus and avoidance behavior, Van Lankveld group reformulated their conceptualization of vaginismus from a sexual disorder to a vaginal penetration phobia (Van Lankveld et al., 2006; Ter Kuile et al., 2009). A recent study carried out by the same group investigated a treatment for vaginismus focusing more explicitly and systematically on the fear of coitus through the use of prolonged and therapist aided exposure therapy (Ter Kuile et al., 2009). The treatment was comprised of education on the fear and avoidance model of vaginal penetration as well as of a maximum of three 2 hour sessions of in vivo exposure to the stimuli feared during vaginal penetration. A replicated (N=10) randomized single-case A-B-phase design was used. The results showed that 9 out of 10 participants were able to engage in intercourse following treatment and these findings persisted at a 1-year follow-up. In addition, the exposure treatment was successful in decreasing fear and negative penetration beliefs.

Evaluation of Treatment Research

Vaginismus has traditionally been considered as an easily treatable sexual dysfunction. The elevated success rates reported in the literature must, however, be considered in light of uncontrolled designs, small sample sizes, elevated or unreported drop-out rates which are not evaluated with intent to treat statistics, as well as a lack of long-term follow-up data. In fact, the only randomized controlled treatment trial does not support the notion that vaginismus is an easily treatable condition (Van Lankveld et al., 2006).

A basic issue in treatment evaluation is how a successful treatment outcome is defined. The great majority of studies has defined success as the ability to achieve vaginal penetration through sexual intercourse. While successful penetration is clearly a crucial first step, if it is not accompanied by pleasurable feelings, then treatment success is questionable. For instance, Schnyder et al. (1998) found that although 98% of the women in their sample were able to have intercourse by the end of treatment with vaginal dilators, 50% were still experiencing pain during penetration. Similarly, although 9 out of 10 participants in the Ter Kuile et al. (2009) fear reduction study were able to experience penetration, none of the measures of sexual enjoyment or pleasure significantly improved. While it appears that high success rates in vaginal penetration may soon be achievable, the therapeutic challenge of increasing vaginismic women's pleasure has not even been approximated.

Conclusion

Although most research concerning vaginismus presents significant methodological limitations, certain conclusions can be made from the few well-controlled studies. First, vaginal muscle spasm is not a valid or reliable diagnostic criterion for vaginismus. Second, vulvar pain is an important characteristic of most women suffering from vaginismus and should be always evaluated. Third, although vaginismus and dyspareunia are presently considered two mutually exclusive disorders, they share many characteristics and are very difficult to differentiate using our current clinical tools. Fourth, fear and avoidance of vaginal penetration situations have been mentioned to be an integral part of vaginismus; interestingly, there are no controlled published studies examining its role. Finally, the present conceptualization of vaginismus as an easily treatable sexual dysfunction has not been supported by empirical research. Unfortunately, it is very difficult to conduct research when inherent problems exist with the definition of vaginismus.

Future Perspective

Unlike the current DSM-IV-TR definition of vaginismus, Binik's new conceptualization of vaginismus as a "genito-pelvic pain/penetration disorder" takes into consideration existing empirical findings as it incorporates pain, muscle tension, and fear. Binik's diagnostic criteria are easily translatable into dimensional terms and do not categorically separate vaginismus from provoked vestibulodynia. This new conceptualization also has significant diagnostic and

therapeutic implications in that it suggests that a multidisciplinary approach taking into account muscle tension, genital pain, and fear will be necessary to attain a high success rate. It is unlikely that a lone professional will be able to provide such a treatment. A multidisciplinary team including a gynecologist, physical therapist and psychologist/sex therapist should be involved in the assessment and treatment of vaginismus to address its different dimensions.

Executive Summary

Introduction

- Vaginismus continues to be perceived by clinicians as a well-understood and easily treatable female sexual dysfunction despite the lack of research supporting these claims.

Prevalence

- Although the population prevalence of vaginismus remains unknown, it has been reported to range between 5-17% in clinical settings.

Classification and Diagnosis

- There has been a 150-year consensus concerning the definition of vaginismus as an involuntary vaginal muscle spasm despite the lack of research supporting the vaginal muscle spasm criterion.
- Women with vaginismus may demonstrate high pelvic floor muscle tension, and/or experience genital pain, and/or report fearing vaginal penetration or pain.

- Vaginismus and dyspareunia are currently considered two mutually exclusive disorders despite empirical findings demonstrating that health practitioners have great difficulty reliably differentiating both conditions.
- Recently, new definitions of vaginismus integrating pelvic floor muscle tension, genital pain and fear have been proposed.

Etiology

- Most psychological factors that have been proposed to play a role in the etiology of vaginismus (i.e., abuse, relationship factors, negative sexual attitudes and lack of sexual education) have not received empirical support.
- Although organic pathologies and pelvic floor dysfunction have often been implicated in the development of vaginismus, they have not been empirical investigated.

Treatment

- Current treatment options for vaginismus include pelvic floor physiotherapy, pharmacological treatments, general psychotherapy and sex/cognitive-behavioral therapy.
- The success rates for the various treatments have generally been reported to be excellent despite the lack of randomized controlled treatment outcome studies validating this claim.
- To date the only randomized controlled treatment outcome study that investigated the efficacy of cognitive behavioral sex therapy for

vaginismus does not support the notion that vaginismus is an easily treatable condition.

- A recent exposure treatment focusing more extensively on the fear component of vaginismus has shown promising results.

Future Perspective

- A new conceptualization of vaginismus as a "genito-pelvic pain/penetration disorder" characterized by inability to have vaginal intercourse/penetration, genito-pelvic pain, fear of vaginal intercourse/penetration, and tension of the pelvic floor muscles has recently been proposed.
- A multidisciplinary diagnostic and adequate treatment approach for vaginismus addressing the fear, genital pain, pelvic floor muscle tension, and sexual pleasure is recommended. This set of skills is not easily accomplished by individual practitioners and should probably be addressed by a multidisciplinary team.

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Table 1. Review of treatment outcome studies for vaginismus

	Study Type	Diagnostic method	Sample	Treatment	Definition of Success	Drop-out rate	Result	Follow-up (FU)
Pelvic Floor Physiotherapy								
Barnes et al, 1984	Uncontrolled clinical study	a) Pelvic exam b) Self-report	N = 5	Biofeedback, psychotherapy & dilators	Intercourse	Not reported	100% success	6 month FU: success ↓60%
Seo et al, 2005	Uncontrolled clinical study	a) Pelvic exam b) Self-report	N = 12	Functional electrical stimulation-biofeedback & CBT	"Satisfactory intercourse"	Not reported	100% success	Not reported
Pharmacological Treatment								
Hassel, 1997	Case study	Not stated	N= 1	5% Lignocaine gel	a) Ability to undergo a pelvic exam b) Intercourse	NA	Success	Not reported
Peleg et al, 2001	Case study	Pelvic exam	N=1	Nitroglycerin ointment	"Satisfactory intercourse"	NA	Success	12.5 month FU: success maintained
Mikhail, 1976	Uncontrolled clinical study	Referral with diagnosis	N = 4	IV Diazepam & marital & psychotherapy	Disappearance of symptoms	Not Reported	100% success	2 to 6 month FU: success maintained (ongoing psychotherapy)
Plaut et al , 1997	Case study	Referral by gynecologist	N=1	Anxiolytic medication & psychotherapy	Intercourse	NA	Success	FU (time unspecified): Success maintained
Brin et al, 1997	Case study	Pelvic exam	N=1	Botulinum toxin injections	Intercourse	NA	Success	2 year FU: success maintained

Ghazizadeh et al, 2004	Uncontrolled clinical study	Referral with diagnosis	N = 24	Botulinum toxin injections	a) Painless pelvic exam b) "Satisfactory intercourse"	Not reported	a) 96% no symptoms during pelvic exam b)75% satisfactory intercourse; 17% mild pain	2 to 24 month FU: success maintained
Shafik et al, 2000	Uncontrolled clinical study	Not reported	N = 13	1) N=8 Botulinum toxin 2) N=5 saline	"Satisfactory intercourse"	Not reported	1) 100% success 2) No improvement	8 to 14 months FU: success maintained
Bertolasi et al., 2009	Uncontrolled clinical study	EMG recordings	N=39	Botulinum toxin type A injections	a) Intercourse b) EMG c) Psychometrics	15,4%	63.2% success	Not reported
Psychological Treatments								
General Psychotherapy								
Barnes, 1986	Uncontrolled clinical study	a) Pelvic exam b) Self-report	N = 55	N=50 Brief psychotherapy, education & dilators N=5 Biofeedback	Intercourse	Not reported	84% success	6 month (4 couples lost at FU) 3 couples no longer having intercourse
Kennedy et al, 1997	Uncontrolled clinical study	a) Pelvic exam b) Self-report c) No local pathology	N = 18	Individual psychotherapy for both partners, education, *in vivo* desensitization & retraining of sexual behavior	Intercourse	Not reported	78% success	Not reported

Elkins et al, 1986	Case study	Unconsummated marriage	N=1	Interactional therapy	Intercourse	NA	Success	12 and 15 month FU: Children born, but no info on sexual function
Gottesfeld, 1978	Case study	a) Unconsummated marriage b) Self-report	N=1	3 years of psychotherapy with hypnosis	Intercourse with orgasm	NA	Success	2 years FU: success maintained
Harman et al, 1994	Case study	Diagnosed by physician	N=1	Relationship enhancement therapy & sexual education	"Improvement of relationship"	NA	Success	6 weeks FU: success maintained
Kleinplatz, 1998	Case study	Referral by gynecologist	N=1	Psychotherapy (existential-experiential)	a) Disappearance of vaginismus b) "Sexual well-being"	NA	Success	6 month FU: success maintained
Pridal et al, 1993	Case study	Referral	N=1	Brief psychotherapy, relaxation, Kegels & dilators	"Satisfactory intercourse"	NA	Success	3 month FU: success maintained
Ben-Zion et al, 2007	Controlled clinical trial	DSM-IV criteria	N = 32	1) N=16 couple therapy & other treatments 2)N=16 surrogate therapy & other treatments	Intercourse	Not reported	1) 100 % success 2)75% success	Not reported
Delmonte, 1988	Case study	Referral	N=1	Psychotherapy, marital therapy, relaxation-hypnosis	Painless intercourse	NA	Success	6 month FU: success maintained
Sex / Cognitive-Behavioral Therapy								
Hawton et al, 1990	Controlled clinical trial	Referral	1) N=30 vaginismus 2) N=76	Sex therapy & Kegels	Vaginismus resolved or largely	10% in vaginismus group	1) 80% success	3 month FU: success rate ↓76, 67%

			other female sexual dysfunctions		resolved		2) 51% success	
Van Lankveld et al, 2006	Randomized controlled treatment outcome study	a)Pelvic exam b)Self-report	N = 117	1) N=43 CBT group therapy 2) N=38 CBT bibliotherapy 3) N=36 Wait-list control	Intercourse	21%	1) 9% success 2)18% success 3) no success	3 month & 1 year FU: 1) success rate ↑ 21% 2) success rate ↓15%
Chakrabarti et al, 2002	Case study	Self-report	N=1	Sex education & psychotherapy	Intercourse	NA	Success	Maintained (time unspecified)
Grillo et al, 1980	Uncontrolled clinical study	a) Pelvic exam b) Painful hymenal rings/rigid remnants	N = 17 Dyspareunia with comorbid vaginismus	Surgical removal of hymenal remnants & sex therapy & dilators & Kegel	a) Intercourse with orgasm b) Painless pelvic exam	Not reported	100% success	Not reported
Jeng et al, 2006	Retrospective study	a) Pelvic exam b) Self-report	N = 120	Sex therapy & Xylocaine & oral analgesics & relaxation & dilation	Intercourse	Not reported	93% success	3 month and 1 year FU: 83% intercourse with orgasm
O'Sullivan, 1978	Uncontrolled clinical study	Pelvic exam	N = 46	Short-term sex therapy & dilators	"Normal sexual function"	48%	52% success	Not reported
Oystragh, 1988	Case study	Unconsummated marriage	N=1	Sex therapy & hypnosis & dilators	Painless intercourse	NA	Success	Maintained (time unspecified)
Ng, 1993	Case study	Unconsummated marriage	N=1	Mien-Ling dilators	"Pleasurable intercourse"	NA	Success	2 month FU: success maintained
Fuchs, 1980	Uncontrolled clinical study	Not stated	N = 71	Systematic desensitization: 1) N=18 *in vitro*	Intercourse	2% (*in vivo* group)	1) 89% success 2) 98%	2 to 5 year FU for 65 patients: "Normal

				2) N=54 *in vivo*			success	sexual adjustment" maintained
Wijma et al, 1997	Case study	Self-report		CBT following *in vivo* systematic desensitization & phobia counter-conditioning	a) Intercourse free of pain/fear b) No recurrence of vaginismus	NA	Success	6 month & 1.5 year FU: success maintained
Wijma, et al, 2000	Case study	VVS diagnosis & penetration not possible	N=1	Systematic desensitization	Disappearance of burning pain	NA	Success	Not reported
Schnyder et al, 1998	Randomized clinical study	DSM-III-R criteria	N = 44	1) N=21 *in vivo* dilation & relaxation & education 2) N=23 *in vitro* dilation	a) Painless intercourse b) Increased sexual desire & orgasm capacity	5%	1) 98% success 2) 50% still having intercourse pain	6-22 month FU: (8 lost) 50% disappearance, 47.7% improvement
Biswas et al, 1995	Uncontrolled clinical study	Not stated	N = 19	Rapid desensitization under anesthesia	"Satisfying intercourse"	Not reported	100% success	Not reported
Scholl, 1988	Uncontrolled clinical study	a) Pelvic exam b) Self-report	N = 23	Sex therapy & dilators & Kegel exercises	Intercourse	13%	87% success	1- 4 years FU: 95% continue intercourse
Ter Kuile et al, 2009	Replicated Single-Case Design	a)Pelvic exam b)Self-report	N = 10	Exposure therapy	a)Intercourse b) Psychometrics	0%	90% success and signif. less fear	3 months and 1 year FU: success maintained

TRANSITIONAL TEXT

This review suggests that the DSM-IV-TR spasm based definition of vaginismus is flawed (APA, 2000). It also suggests that the notion that vaginismus and dyspareunia/PVD are mutually exclusive disorders is not consistent with the available research and clinical experience. In line with these findings, the review discusses a DSM-5 proposal to redefine and collapse vaginismus and dyspareunia under one category named "Genito-Pelvic Pain/Penetration Disorder". However, the review highlights that fear appears to be a promising factor that characterizes women suffering from vaginismus and that may distinguish them from women suffering from dyspareunia/PVD. The following empirical paper explores whether fear measured through self-report, behavioral and physiological indices while women undergo a gynecological examination can discriminate vaginismus from dyspareunia/PVD. The roles of genital pain, vaginal muscle tension, sexual functioning and sexual and physical abuse in vaginismus are also re-examined.

CAN VAGINISMUS BE DISCRIMINATED FROM DYSPAREUNIA/PVD?

A TEST OF THE PROPOSED DSM-5

GENITAL PAIN/PENETRATION DISORDER CRITERIA

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Article submitted to Archives of Sexual Behavior on April 1st, 2012

Abstract

Current empirical research does not support the DMS-IV-TR diagnostic distinction between vaginismus and dyspareunia. This has led to a DSM-5 proposal to collapse vaginismus and dyspareunia into one diagnostic category called genito-pelvic pain/penetration disorder (Binik, 2010). Fear, however, has been suggested as a possible differentiator between these two disorders but this has not yet been empirically examined. The primary purpose of this study was therefore to investigate whether fear as evaluated by subjective, behavioral, and psychophysiological measures could differentiate women with vaginismus from those with dyspareunia/PVD and controls. A second aim was to re-examine whether genital pain, vaginal muscle tension, sexual functioning, and childhood sexual and physical abuse differed between vaginismus and dyspareunia/PVD sufferers. 50 vaginismic women, 50 women with dyspareunia/PVD and 43 controls participated in an experimental session comprising a structured interview, pain sensitivity testing, a filmed gynecological examination and several self-report measures. Results demonstrated that fear and vaginal muscle tension were significantly greater in the vaginismic group as compared to the dyspareunia/PVD and no-pain control groups. Moreover, behavioral measures of fear and vaginal muscle tension were found to discriminate the vaginismic group from the dyspareunia/PVD and no-pain control groups. Preliminary taxometric analyses suggested that this difference may have been due to a small subgroup of women in the vaginismus group. Genital pain, sexual functioning, and sexual and physical abuse did not differ significantly between the vaginismus and

dyspareunia/PVD groups. However, genital pain was found to discriminate both clinical groups from controls. Despite significant statistical differences on fear and vaginal muscle tension variables between women suffering from vaginismus and dyspareunia/PVD, a large overlap was observed between these conditions which may explain the great difficulty health professionals experience in attempting to reliably differentiate vaginismus from dyspareunia/PVD. Whether vaginismus and dyspareunia/PVD should be collapsed into one disorder or remain two mutually exclusive conditions in the DSM-5 is further discussed.

Keywords: Vaginismus, Dyspareunia, Provoked Vestibulodynia, Fear, Self-report, Behavioral measures of fear, Psychophysiological measures of fear, DSM-5, Psychiatric Classification, Vaginal Muscle Tension, Genital Pain, Sexual Functioning, Sexual and Physical Abuse

Introduction

Dyspareunia and vaginismus are classified in the DSM-IV-TR as distinct and mutually exclusive sexual pain disorders. Vaginismus is defined as "the involuntary spasm of the musculature of the outer third of the vagina that interferes with intercourse", while dyspareunia is defined as "genital pain associated with sexual intercourse" (APA, 2000). Most health professionals and classificatory systems appear to accept this notion of two distinct conditions (e.g., ICD-10, 1992; American College of Obstetrics and Gynecology, 1995; Merskey & Bogduk, 1994; Basson et al., 2004; Binik, 2010; Lahaie, Boyer, Amsel, Khalifé,& Binik, 2010).

This diagnostic differentiation between dyspareunia and vaginismus is reflected in the development of different treatment approaches for these disorders. The standard treatment approach for vaginismus has focused on eliminating vaginal muscle spasm in addition to the provision of some form of systematic desensitization, progressive vaginal dilatation, and sexual education (e.g., Masters & Johnson, 1970; Sims, 1861; Kaplan, 1974; Beck, 1993). On the other hand, treatments for dyspareunia have traditionally concentrated on treating the presumed underlying medical (e.g., infection, inflammation) or psychological (e.g., sexual abuse, marital problems, lack of sexual arousal) factors that are causing the pain. Such treatments have ranged from medication and surgery to sex and couple therapy.

Unfortunately, the original classificatory decision to separate vaginismus and dyspareunia was not based on empirical research (Binik, 2010). In fact,

subsequently collected data has challenged this longstanding decision. First, several studies have shown that the differential diagnosis of vaginismus from dyspareunia is not reliable (Basson, 1996; de Kruiff, ter Kuile, Weijenborg,& van Lankveld, 2000; Engman, Lindehammar,& Wijma, 2004; Reissing, Binik, Khalifé, Cohen,& Amsel, 2004; Engman, 2007; Engman, Wijma, &Wijma, 2007; Engman, Wijma,& Wijma, 2008). In particular, attempts to differentiate provoked vestibulodynia (PVD), the most common form of superficial dyspareunia, from vaginismus using measures of pelvic floor muscle spasm or genital pain have failed (de Kruiff et al., 2000; Reissing et al., 2004). This has led many to argue that vaginal muscle spasm, the central defining characteristic of vaginismus, may not be a valid or diagnostically reliable marker of the disorder (Engman et al., 2004; van der Velde, 1999; Shafik & El-Sibai, 2002; Frasson et al., 2009; van der Velde, Laan,& Everaerd, 2001; Reissing et al., 2004). Such findings have led to the new DSM-5 proposal to collapse vaginismus and dyspareunia into one category to be called "Genito-Pelvic Pain/Penetration Disorder" (Binik, 2010). This disorder would be defined as marked difficulty with at least one of the following: 1. vaginal intercourse/penetration; 2. genito-pelvic pain; 3. fear of vaginal intercourse/penetration; 4. heightened pelvic floor muscle tension during attempted penetration (http://www.dsm5.org/ProposedRevision/Pages/proposedrevision.aspx?rid=435).

Despite the fact that it has not been possible to reliably discriminate vaginismus from dyspareunia/PVD either on the basis of pelvic floor muscle spasm or genital pain, it has been suggested that fear maybe a possible

differentiator. As early as 1909, Walthard suggested that vaginismus was a phobic reaction to an excessive fear of pain. This idea was also discussed by Kaplan (1974) and supported by data collected by Ward and Ogden (1994). Unfortunately, this notion was not systematically pursued until Reissing et al. (2004) demonstrated that vaginismic women undergoing a gynecological examination displayed a significantly higher number of defensive/avoidant behaviors than matched controls or women suffering from dyspareunia/PVD. Reissing et al. (2004) proposed that vaginismus might be better conceptualized as a specific phobia characterized by an excessive fear and avoidance of vaginal penetration situations. Although the behavioral measures of fear in the Reissing et al. (2004) study clearly differentiated vaginismus from dyspareunia/PVD, there were some significant methodological limitations to this research. For example, those rating fear were not blind to participants' diagnosis. In addition, the characterization of fear was solely based on behavior and did not include the assessment of subjective or physiological indicators of fear. Finally, there was no assessment of the reliability of the behavioral rating system for fear.

Therefore, the primary goal of the present study was to investigate whether the degree of fear displayed during a vaginal penetration situation such as a gynecological examination could discriminate women suffering from vaginismus from those with dyspareunia/PVD and controls. A variety of methods were used to measure fear, including self-report, blinded behavioral assessments, and psychophysiological indicators such as heart rate, skin conductance, and non-genital muscle tension. A secondary aim was to re-

examine whether vaginismus could be distinguished from dyspareunia/PVD by pelvic floor muscle tension and genital pain. Finally, two additional issues were investigated: a) are sexual and physical abuse related to vaginismus, and b) is vaginismus like dyspareunia/PVD associated with disruption in general sexual functioning.

Our first hypothesis was that fear measured through self-report, behavioral, and psychophysiological indices would distinguish women in the vaginismus group from women in the dyspareunia/PVD and control groups. More specifically, women in the vaginismus group would display significantly greater fear as compared with women in the dyspareunia/PVD and control groups. We further hypothesized that women in the dyspareunia/PVD group would also demonstrate greater fear than controls.

Our second hypothesis was that pelvic floor muscle tension rather than spasm would distinguish women in the vaginismus group from women in the dyspareunia/PVD and control groups. More specifically, women in the vaginismus group would display the highest degree of pelvic floor muscle tension during the gynecological examination followed by women in the dyspareunia/PVD group and controls. Finally, our third hypothesis was that genital pain would discriminate the clinical groups (vaginismus and dyspareunia/PVD) from controls, but would not distinguish vaginismus from dyspareunia/PVD.

Materials and Methods

The present study was reviewed and approved by the McGill University Faculty of Medicine Institutional Review Board; written informed consent was obtained from all study participants.

Participants

The participants included 50 vaginismic women, 50 women suffering from dyspareunia/PVD, and 43 controls. The participants were primarily young (M_{age} = 25, range 18-41), well-educated (76% had an undergraduate degree or more), born in North America (73% North America, 10% Asia, 9% Europe, 8% other) and unmarried (27% single, 56% dating, 14% married, and 3% other). No significant differences between study groups were found on age, level of education, relationship status, birthplace, or religion. Significant differences between groups were found, however, on primary language, χ^2 (2, N = 143) = 12.17, $p < .05$, and cultural identity, χ^2 (2, N = 143) = 7.15, $p < .05$. A significantly higher percentage of women in the vaginismus group reported having a primary language other than French or English as compared with women in the dyspareunia/PVD and control groups. In addition, a significantly higher percentage of women in the vaginismus and control groups reported a cultural identity other than "Canadian" or "Québecoise" as compared to women in the dyspareunia/PVD group. No significant group differences were found, however, on any of the dependent variables between: a) women with vaginismus who reported "French" as their primary language compared to women with vaginismus who reported "English" or "other language" as their primary

language, b) women with vaginismus reporting a "Canadian" or "Québecoise" culture compared to those reporting "other" as their cultural identity, or c) controls reporting a "Canadian" and "Québecoise" culture compared to those reporting “other” as their cultural identity.

Measures

Psychophysiological monitoring

An ambulatory monitor (TEL 100C, Harvard Apparatus Canada) was used for psychophysiological monitoring (ECG: electrocardiograms, EDR: electrodermal activity, and EMG: electromyography) with data recorded on an MP100 system (Biopac Systems Inc. AcqKnowledge). ECG recordings were accomplished via electrodes (100/PK, EL 503) placed on the right lower abdominal region and below the left collarbone. EMG-recording electrodes (100/PK, EL 503) were placed on the right trapezius muscle. EDR-recording electrodes (100/PK, EL 507) were positioned with an electrode paste (Gel 101) on the distal phalanges of the left middle and ring fingers.

Self-Report Measures

A semi-structured interview adapted from Reissing et al.’s (2004) study was administered to collect information on socio-demographic background and medical, gynecological, and relationship history. The Specific Phobia section of the Structured Clinical Interview (SCID-I; First, Spitzer, Gibbon,& Williams, 1997) was added to the interview with the aim of evaluating whether participants met the DMS-IV-TR diagnostic criteria for a specific phobia of vaginal penetration (APA, 2000). Some of the questions were modified by adding the words "fear of

vaginal penetration" at appropriate places. The SCID-I is a semi-structured diagnostic interview designed to assist clinicians, researchers, and trainees in making reliable DSM-IV-TR psychiatric diagnoses.

The following standardized questionnaires were administered to measure pain, fear/anxiety, sexual functioning, and the occurrence of childhood sexual/physical abuse. All of these questionnaires are standardized psychometric instruments with demonstrated reliability and validity. The McGill Pain Questionnaire (MPQ; Melzack & Katz, 1992) was administered to measure the sensory and affective dimensions of the pain that participants experienced during the gynecological examination. The Pain Catastrophizing Scale (PCS; Sullivan, Bishop,& Pivik, 1995) was administered twice to examine the cognitive and emotional characteristics of participants' non-genital and genital pain. This scale is a measure of pain magnification, rumination, and helplessness (Sullivan et al., 1995). The Fear Survey Schedule-II (FSS-II; Geer, 1966) was administered to measure the degree of fear for various objects and situations. The Fear of Pain Questionnaire-III (FPQ-III; McNeil & Rainwater, 1998) was administered to assess fear of three broad categories of pain: Severe, Minor, and Medical Pain. The Trait and State subscales of the State Trait Anxiety Inventory (STAI; Spielberger, Gorsuch,& Lushene, 1970) were used to evaluate the presence of general and situational anxiety. The Female Sexual Functioning Index (FSFI; Rosen et al., 2000) was administered to measure general female sexual functioning; it is a brief self-report measure of female sexual dysfunction composed of 6 subscales: desire, arousal, lubrication, orgasm, satisfaction, and

pain. The Childhood Experience of Abuse Questionnaire (CEAQ; Bifulco, Brown,& Harris, 1994) was administered to investigate whether participants had experienced any physical and/or sexual abuse in their childhood. Finally, two questionnaires, the Vaginal Penetration Survey (VPS) and the Sexual Disgust Sensitivity Index (SDSI), developed by the principal authors, were used to assess the degree of fear and disgust participants experienced with imagined or attempted vaginal penetration situations. The VPS was based on the FSS-II (and is comprised of 21 items referring to imagined or attempted vaginal penetration situations that may cause fear or unpleasant feelings (e.g., "When I imagine or try to engage in activities involving vaginal penetration such as intercourse or tampon insertion, I fear it will be painful"; "When I imagine or try to engage in activities involving vaginal penetration such as intercourse or tampon insertion, I fear that I will be unable to find the vaginal opening"). Participants were asked to evaluate on a 5-point scale how much they were disturbed by each item nowadays, with response options ranging from 0 (*not at all*) to 4 (*very much*)[1]. The SDSI was based on the Disgust Sensitivity Scale (Haidt, McCauley,& Rozin, 1994), which measures disgust sensitivity across seven domains: animals, body products, death, envelope violations (injuries, wounds, etc.), food, hygiene, and sex. The SDSI consists of 22 items referring to sexual objects, practices, and experiences that may cause disgust (e.g., sight and smell of vaginal secretion; smell of semen; performing oral sex). Participants were asked to evaluate on a 5-point scale how much they were disgusted by each item, with response options ranging from 0 (*not at all*) to 4 (*very much*)[1].

Pain Sensitivity testing

Pain sensitivity testing was carried out with a vulvalgesiometer to measure deltoid and vulvar pain thresholds (Pukall, Binik,& Khalifé, 2004). Each participant was first presented with tactile and pain stimuli on the deltoid muscle of the right arm. Testing started with the lowest pressure exerted by the vulvalgesiometer (3 grams) and consecutively higher pressures were applied after an inter-stimulus interval of 10 seconds. Non-painful and painful intensity, unpleasantness, and emotional distress ratings were recorded with each application. Testing stopped once the participant reported a minimal level of pain, defined as a self-report pain intensity rating of 2 on 10. The same protocol as described above was carried out at the 9 o'clock position and at the base of the hymeneal ring on the vulvar vestibule (e.g., entrance of the vagina).

Gynecological examination

A standardized pelvic examination used in previous research (Bergeron et al., 2001; Meana, Binik, Khalifé,& Cohen, 2007) was carried out by the participating gynecologist. The first author and a female research assistant were present during the examination. The protocol consisted of the following: a) visual and digital examination of the vulva; b) assessment of the degree of difficulty inserting into the vagina a cotton-swab, one finger, and two fingers; c) internal digital examination of the vagina and reproductive organs; c) examination of muscle tension and presence of vaginal muscle spasm; d) a cotton swab test (Friedrich, 1987) at three vestibular sites (3, 6, and 9 o'clock, the sequence of testing was randomized). The cotton-swab test is the generally accepted

gynecological examination for diagnosing provoked vestibulodynia and consists of the application of a cotton-swab to different areas of the vulvar vestibule (Friedrich, 1987). During each step of the gynecological examination, participants were asked to rate if any pain or anxiety was experienced, and if so to rate the intensity on a verbal analogue scale from 0 (no pain; no anxiety) to 10 (worst pain ever experienced; worst anxiety ever experienced). The gynecologist also separately rated the degree of difficulty following the insertion of a cotton-swab then following the insertion of one finger and two fingers on a 4 point-scale from 0 (*no problem*) to 3 (*impossibility*). Vaginal/pelvic muscle tone was evaluated using Lamont's 6-point rating scale from 0 (*normal tone*) to 5 (*perineal and levator ani contractions*; Lamont, 1978). The gynecologist also globally rated degree of pelvic floor muscle tension displayed by participants during the gynecological examination on an 11-point rating scale from 0 (*no tension*) to 10 (*strong tension*).

Behavioral Measures

During the gynecological examination, two behavioral measures indicative of fear were used. First, the gynecologist separately rated the level of defensive/avoidant reactions following the insertion of a cotton-swab, following the insertion of one finger, and following the insertion of two fingers. Defensive/avoidant reactions were defined by Reissing et al. (2004) as behaviors interfering with, delaying, or terminating the examination and were rated on a 5-point scale from 0 (*no problematic reaction during the exam*) to 4 (*the participant terminated the exam*). Second, the participant's face and body were filmed during

the gynecological examination with a Canon Elura 80 Digital Camcorder in order to evaluate the frequency and intensity of displayed behavioral reactions of fear and pain. The recording of each participant's exam was uploaded onto a computer and edited using iMovie software**,** before being burnt onto a DVD. Two trained research assistants who were blind to group membership independently viewed recordings on a MacBook laptop using QuickTime software and were asked to code the videos by following a checklist of eight categories of behaviors: 1) neck arching, 2) facial grimacing, 3) participant closing legs, 4) gynecologist having to open legs of participants, 5) pelvic withdrawal, 6) participants' placing one or both hands on head, 7) verbal expression (e.g., ahh, ouch, stop), 8) paraverbal vocalizations (e.g., sigh or gasp). The above behaviors were separated in two main categories: protective behaviors and communicative behaviors. Protective behaviors included closing legs, pelvic withdrawal, and placing one or both hands on head while communicative behaviors included neck arching, facial grimacing, and verbal and paraverbal behaviors. Each behavior's occurrence was coded in terms of its frequency and intensity on a 3-point scale: mild, moderate, or severe. This behavioral observation system was based on previously developed coding systems for laboratory pain and affective behavior studies (Prkachin, Hughes, Schultz, Joy,& Hunt, 2002; Keefe & Block, 1982; Sullivan, Martel, Tripp, Savard,& Crombez, 2006; Sullivan, Adams,& Sullivan, 2004; Enkman & Friesen, 2007). This system was adapted to the gynecological examination by viewing a sample of 80 women including women with

vaginismus, dyspareunia/PVD, and controls undergoing a gynecological examination to record the behavioral reactions of fear and pain displayed.

The training of the raters started with a 3-hour session during which the pain behavior definitions and coding procedure were reviewed and video clips were shown to illustrate the different categories of behaviors including the varying intensities. Raters began coding 5 hours per week in addition to weekly/biweekly training sessions to increase inter-rater reliability. During the training sessions, non-eligible participant videos were watched with the researchers to discuss coding decisions with regards to the different behavior frequencies and intensities. Using a sample of 44 women meeting the criteria outlined above for vaginismus (n = 15), dyspareunia/PVD (n = 15), and controls (n = 14), the validity and reliability of the behavioral observation system was investigated. It was found to have high inter-rater reliability, good internal consistency, and good construct and discriminant validity. Inter-rater reliability was measured through the correlation of rater 1 and rater 2's scores for the 44 participants, which were the total frequency and average intensity ratings for each behavior. The correlations regarding the frequency of behaviors were all over .8, ranging from r = .85 for verbal vocalizations (p < .001) to r = .99 for opening legs (p < .001). With regards to intensity ratings, the correlations were all above .7, with the exception of paraverbal vocalizations (r = .58, p < .001). Paired samples t-tests were conducted to evaluate whether raters significantly differed in their coding of particular behaviors. No significant differences were found for 11 out of the 15 frequency and intensity ratings. Rater 2 scored significantly more

behaviors than rater 1 for neck arching frequency (t (43) = 2.12, p < .05), closing legs frequency (t (43) = 2.17, p < .05) and pelvic withdrawal frequency (t (43) = 5.37, p < .01), whereas rater 1 scored significantly more instances of hands on head than rater 2 (t (43) = 2.71, p = .01).

Internal consistency of the behavioral observation system was investigated through the correlation of each category of behavior with the total frequency of behavior scores (i.e., the co-investigator average of the overall number of pain behaviors for each participant). All correlations were significant (p < .002), except for hands on head frequency (r = .21, p >.05) and intensity ratings (r = .29, p >.05). Discriminant validity was assessed through a one-way ANOVA examining whether the gynecologist's diagnosis differed based on the amount of behaviors shown by each participant. Participants diagnosed with a sexual pain disorder (i.e., vaginismus or dyspareunia/PVD) demonstrated significantly more behaviors (M = 43.98, SD = 30.77) than controls (M = 9.88, SD = 8.07), F (1, 43) = 15.33, p < .001.

Procedure

Our sample was recruited via local media announcements, advertisements, and health professionals' referral. Advertisements were aimed at women who were either experiencing "difficulties with vaginal penetration", "pain with vaginal intercourse", or "no pain with intercourse". A telephone screening interview was conducted with potential participants to insure their eligibility and to explain the study procedures. During the screening interview, potential participants described whether difficulties and/or pain were experienced with

different vaginal penetration situations (tampon insertion, gynecological examination, and vaginal intercourse). If participants met the criteria for either the vaginismus, dyspareunia/PVD, or control group an appointment was scheduled.

The inclusion criteria for vaginismus were based on those of Reissing et al.'s (2004) study and focused on the inability to achieve and avoidance of vaginal penetration: (1) never having been able to experience vaginal penetration (i.e., penile-vaginal intercourse or gynecological examination or tampon insertion), despite attempts on at least 10 separate occasions; (2) never having been able to experience vaginal penetration despite attempts on at least two separate occasions and demonstration of "active avoidance" of vaginal penetration, or (3) current inability to experience vaginal penetration AND "active avoidance" of vaginal penetration for at least 1 year, although vaginal penetration was experienced at least once before this period. Active avoidance was defined as an average of less than 1 attempt at vaginal intercourse every 2 months over the past year and meeting one of the following two criteria: 1) never successfully completing a gynecological examination, 2) never having used tampons.

The inclusion criteria for dyspareunia/PVD were based on those of Bergeron et al. (2001): (1) pain occurring during intercourse on at least 60% of all episodes, and (2) pain, at the entrance of the vagina, elicited by direct touch or pressure at the vestibule which has a burning or cutting quality; and (3) pain that is personally distressing and has been present for at least 6 months.

The inclusion criteria for the control group were: (1) current ability to experience vaginal penetration without difficulty and/or pain, and (2) no history of

vulvar/vaginal/pelvic pain or penetration difficulties during intercourse, gynecological examination, or tampon insertion.

The exclusion criteria for all three groups were: (1) current pregnancy or breast-feeding, (2) post-menopausal status, (3) major medical conditions (e.g., endometriosis, cancer) or treatments (e.g., radiation therapy) which may affect the genital/pelvic area; or (4) current major psychiatric conditions (e.g., schizophrenia, bipolar disorder).

The experimental session was carried out in a gynecologist's office and lasted approximately 3-4 hours. The study procedures were re-explained at the start of the session and written informed consent was obtained. Participants were informed that they could withdraw from the study at any time, including during the procedure, without any prejudice to their treatment or to themselves. Following the experimental session, participants were provided with diagnostic information and possible treatment interventions, and were referred appropriately if necessary. All participants received $75 as compensation for their participation. Throughout the study, previous diagnoses and group membership were not disclosed to the gynecologist who performed the gynecological examination.

Statistical Analysis

Differences between groups on completion of questionnaires and procedures as well as on socio-demographic, general health, sexual functioning, and childhood sexual and physical abuse variables were analyzed using Chi-square analyses for discrete variables and ANOVAs for continuous ones. Tukey HSD post-hoc tests were used to evaluate significant ANOVA results.

Given the large number of dependent variables in the present study, fear, vaginal muscle tension, and genital pain total scores were computed. The fear total score was computed by taking the mean of the 43 standardized self-report, psychophysiological, and behavioral fear variables. The vaginal muscle tension total score was computed by taking the mean of the five standardized vaginal muscle tension variables, while the genital pain total score was computed by taking the mean of the 18 standardized genital pain variables. Group differences on these total scores were analyzed using ANOVAs followed by Tukey HSD post-hoc tests.

Following the results of the above statistical analyses and to avoid redundancy, five separate principal components analyses with varimax rotation were performed on the following continuous measures: 1) 14 self-report fear measures (self-reported anxiety following each gynecological procedure; scores on the PCS, FPQ-III, FSS-II, VPS, SDSI, STAI-S, and STAI-T); 2) 8 psychophysiological measures of fear (heart rate, heart rate variability, mean EMG and EDR peaks taken during the pain sensitivity testing and gynecological examination); 3) 19 behavioral measures of fear (gynecologist's rating of level of defensive/avoidant reactions during the insertion of a cotton-swab, of one finger, and of two fingers; frequency and intensity of the eight categories of behavior from the behavioral observation system; frequency and intensity of protective and communicative behaviors); 4) 18 pain variables (self-reported pain intensities following each gynecological procedure, Present Pain Index and Pain Rating Index of the MPQ, deltoid and vulvar vestibule pain threshold); 5) 5 vaginal

muscle tension variables (scores on the Lamont scale; gynecologist's rating of degree of muscle tension on an 11-point-rating scale; gynecologist's rating of degree of difficulty inserting a cotton-swab, one finger, and two fingers). The variables were standardized within groups to eliminate the influence of mean differences on the correlations. Missing data were replaced with the group mean. The criterion used to extract the factors (or components) from the principal component analysis was having an eigenvalue greater than one. Component scores were calculated by totaling the variables, standardized across groups, which loaded highest on that particular component.

The components extracted from the principal component analysis were entered into a stepwise discriminant function analysis to determine the most parsimonious set of components that could significantly separate the groups. The first analysis was conducted using all components and the three groups (vaginismus, dyspareunia/PVD, and controls). To further investigate our hypothesis that fear alone may differentiate vaginismus from dyspareunia/PVD and controls, a second discriminant analysis was conducted using the fear components and the three groups (vaginismus, dyspareunia/PVD, and controls). To examine which fear, vaginal muscle tension, and/or genital pain components could best distinguish vaginismus from dyspareunia/PVD, a third discriminant analysis was conducted using all components and the vaginismus and dyspareunia/PVD groups.

Finally, a taxometric analytic method, MAXCOV-HITMAX (Meehl, 1995), was used to explore whether differences between vaginismus and

dyspareunia/PVD on fear, genital pain, and vaginal muscle tension were categorical (i.e., taxonic or discrete groups) or dimensional (i.e., continuous along a scale where the two poles represent the most extreme group differences) in nature. MAXCOV-HITMAX is a taxometric method that examines the maximum covariation between two variables as a function of a third. The sample is subdivided into a sequence of ordered subsamples based on their scores on the third variable. The covariance between the other two variables is then calculated for each of these subsamples. If the differences are dimensional in nature, the covariances will be randomly distributed around a single value resulting in a flat profile when plotted. If a categorical difference (i.e., taxonic) exists, the plot will be convex (an inverted U) where the covariances between two variables or more will vary as a function of a third. MAXCOV-HITMAX is generally computed on large samples (*N*> 300) since it involves dividing the sample into a set of ordered subsamples, each for its own analysis (Meehl, 1995; Cole, 2004). We, therefore, consider the results of our analyses exploratory. Cole (2004) suggested the use of a sliding window for the selection of subgroups with one subgroup overlapping an adjacent subgroup, when the sample sizes are small. Following Cole's suggestion, the sample was divided into ten overlapping categories for fear, ten overlapping categories for genital pain, and five for pelvic floor muscle and covariances were calculated between the remaining two variables for each category. Cole's suggestion was also attempted on twenty overlapping categories; however, the resulting graph did not help to clarify the taxonic structure.

Results

Differences between groups on completion of experimental procedures

No significant differences were found between groups on unwillingness to undergo or finish the pain sensitivity testing nor on completion of the following questionnaires: PCS with reference to general pain, VPS, SDIS, FPQ-III, FSS-II, STAI-T, STAI-S, and FSFI. Significant differences between groups were found on unwillingness to undergo or finish the gynecological examination, χ^2 (2, N = 143) = 36.86, p< .01 with a significantly higher number of women in the vaginismus group (n=24) discontinuing compared to women in the dyspareunia/PVD (n = 3) and control groups (n = 1). Significant differences between groups were found on completion of the MPQ, χ^2 (2, N = 143) = 8.66, p< .05; and PCS with reference to pain experienced during the gynecological examination, χ^2 (2, N = 143) = 9.77, p<.01 with a significantly higher number of women in the vaginismus group (n=10) not completing the MPQ due to their unwillingness to undergo the gynecological examination compared to dyspareunia/PVD (n=1) and controls (n=4). A higher number of women in the vaginismus (n=9) and control groups (n=8) did not complete the PCS with reference to the pain experienced during the gynecological examination as compared to women in the dyspareunia/PVD group (n=0). Controls who did not experience any pain during the gynecological examination were not asked to complete the PCS with reference to the pain experienced during the gynecological examination.

Differences between groups on fear, genital pain, and vaginal muscle tension total scores

Fear total score

One-way ANOVA revealed significant differences between groups on the fear total score, F (2, 140) = 87.63, p < .001 (see Figure 1). Tukey HSD post-hoc comparisons revealed that women in the vaginismus group had significantly higher mean scores than women in the dyspareunia and control groups (all p values < .001). Women in the dyspareunia/PVD group also showed significantly higher mean scores on the fear total score than controls (p < .001).

Genital Pain total score

One-way ANOVA revealed significant group differences on the genital pain total score, F (2, 140) = 62.96, p < .001 (see Figure 1). Tukey HSD post-hoc comparisons revealed that women in the vaginismus and dyspareunia/PVD groups had significantly higher mean scores than women in the control group (all p values < .001). No significant group differences were found on the genital pain total score between the vaginismus and dyspareunia/PVD groups.

Vaginal Muscle Tension total score

One-way ANOVA revealed significant group differences on the vaginal muscle tension total score, F (2, 140) = 27.47, p < .001 (see Figure 1). Tukey HSD post-hoc comparisons revealed that women in the vaginismus group had significantly higher mean scores than women in the dyspareunia/PVD and control groups (all p values < .001). No significant group differences were found on the

vaginal muscle tension total score between the dyspareunia/PVD and control groups.

Figure 1 displays the differences between groups on the fear, genital pain, and vaginal muscle tension total scores. As seen in Figure 1, women in the vaginismus group scored higher on fear, vaginal muscle tension, and genital pain followed by women in the dyspareunia/PVD group who also score higher than controls on all three measures. To further examine the structure of between group differences on fear, genital pain, and vaginal muscle tension, each participant was plotted according to their score on the combinations of the three factors taken two at a time (see Figures 2, 3 and 4). The plots from Figures 2, 3 and 4 suggest a large overlap between the vaginismus and dyspareunia/PVD groups on the fear and genital pain total scores. The plots from Figures 3 and 4 further suggest that a subgroup of women in the vaginismus group scored highly on the vaginal muscle tension total score and differed from the remaining sample.

Data reduction

The five principal component analyses (PCA) extracted three components for self-report measures of fear, three components for psychophysiological measures of fear, six components for behavioral measures of fear, five components for genital pain, and two components for vaginal muscle tension (see Table I).

Discrimination between the vaginismus, dyspareunia/PVD, and control groups

A discriminant function analysis performed on all 19 components found that the vaginismus, dyspareunia/PVD, and control groups could be significantly discriminated by two standardized discriminant functions (Function 1: genital pain, and Function 2: vaginal muscle tension and protective behaviors; see Table II). Using these functions, 70% of women with vaginismus, 78% of women with dyspareunia/PVD, and 93% of women in the control group were correctly classified. Twenty-four percent of women in the vaginismus group were misclassified as dyspareunia/PVD, while 6% were misclassified as controls. Eighteen percent of women in the dyspareunia/PVD group were misclassified as vaginismus and 4% were misclassified as controls. Just over 2% of controls were misclassified as vaginismus and 4.7% as dyspareunia/PVD.

Genital pain (component 4, table I) had the highest loading (0.894) on function 1. Genital pain component 4 comprised of scores on the MPQ and self-reported pain intensities with the cotton-swab test. Vaginal muscle tension (component 1, Table I) had the highest loading (.586) on function 2. Vaginal muscle tension component 1 consisted of the gynecologist's rating of degree of pelvic floor muscle tension, the Lamont's scale, and the degree of difficulty the gynecologist experienced inserting two fingers. Behavioral measures of fear (component 1, table I) also loaded highly (.510) on function 2 and were comprised of the total number of protective behaviors.

As can be seen in Figure 5, function 1 (genital pain) discriminates well between women in the control group and women in the vaginismus and dyspareunia/PVD groups. Function 2 (vaginal muscle tension and protective behaviors) does not discriminate as well between women in the vaginismus, dyspareunia/PVD, and control groups. A large overlap can be observed between the vaginismus and dyspareunia/PVD groups on function 1 (genital pain) as well as between all three groups on function 2 (vaginal muscle tension and protective behaviors).

Discrimination between vaginismus, dyspareunia/PVD, and controls based on fear measures only

The discriminant analysis based on the fear variables included the three self-report measures of fear components, the six behavioral measures of fear components, and the three psychophysiological measures of fear components. This analysis found that the vaginismus, dyspareunia/PVD, and control groups could be significantly discriminated by two standardized discriminant functions (Function 1: behavioral and self-report measures of fear, and Function 2: psychophysiological measures of fear; see table III). Using these functions, 72% of women with vaginismus, 72% of women with dyspareunia/PVD and 86% of women in the control groups were correctly classified. Fourteen percent of women in the vaginismus group were misclassified as dyspareunia/PVD and 14% misclassified as controls. Twenty four percent of women in the dyspareunia/PVD group were misclassified as vaginismus and 4% misclassified

as controls. Regarding controls, 2.3% were misclassified as vaginismus and 11.6% as dyspareunia/PVD.

Behavioral measures of fear (component 1 and 5, table I) and self-report measures of fear (component 2, table I) had the highest loadings (0.679, 0.616, 0.600) on function 1 (behavioral and self-report measures of fear). Behavioral measures of fear component 1 were composed of protective behaviors while behavioral measures of fear component 5 comprised of verbal and grimacing intensity scores. Self-report measures of fear component 2 was composed of scores on the SDSI, VPS, FSS-II, FPQ-III, PCS (with reference to the gynecological examination) and the gynecologist's rating of the participant's degree of fear during the gynecological examination. Psychophysiological measures of fear (component 1 and 2, table I) had the highest loadings (-.596, .577) on function 2 (psychophysiological measures of fear) and comprised of heart rate and EMG taken during the gynecological examination and sensory testing.

As can be seen in Figure 6, function 1 (behavioral and self-report measures of fear) discriminates well between women in the control group and women in the vaginismus and dyspareunia/PVD groups. Function 2 (psychophysiological measures of fear) does not discriminate as well between women in the vaginismus, dyspareunia/PVD, and control groups. Again, a large overlap can be observed between the vaginismus and dyspareunia/PVD groups on function 1 (behavioral and self-report measures of fear) as well as between all three groups on function 2 (psychophysiological measures of fear).

Discrimination between vaginismus and dyspareunia/PVD

In an attempt to clarify the distinction between the two clinical groups, a discriminant analysis was performed on the vaginismus and dyspareunia/PVD groups without including the controls using all nineteen components. The discriminant analysis was significant (see table IV). Using this function, 72% of women in the vaginismus group as were 82% of women in the dyspareunia/PVD group were correctly classified. Behavioral measures of fear (component 1, table I) and vaginal muscle tension (component 1, table I; 0.658, 0.599) had the highest loadings on the discriminant function and referred to the degree of difficulty the gynecologist experienced inserting two fingers, vaginal muscle tension rated by the gynecologist, and protective behaviors displayed during the gynecological examination. Women in the vaginismus group scored higher on the behavioral measures of fear component 1 and on the vaginal muscle tension component 1 than women in the dyspareunia/PVD group.

Dimensional vs. categorical differences between vaginismus and dyspareunia/PVD

The results of our exploratory taxometric analyses using the MAXCOV-HITMAX method on fear, vaginal muscle tension, and genital pain are illustrated in the three plots depicted in Figure 7. As can be observed from the plotted covariations for both fear and vaginal muscle tension, the curves suggest a taxonic structure as a clearly defined peak can be observed on the right hand side of the graph. A similar but less clearly defined peak is observed for genital pain. More specifically, the plots appear to depict a group of women scoring in

the upper 20th percentile of fear and vaginal muscle tension that is distinct from the remaining 80% of women. This group comprised mainly of women in the vaginismus group.

On the other hand, a discriminant analysis was performed after removing women scoring in the upper 20th percentile of fear and vaginal muscle tension and was still capable of distinguishing the vaginismus group from the dyspareunia/PVD group suggesting a dimensional structure where vaginismus and dyspareunia/PVD represent the poles on a continuum. Using this function, 65% of women in the vaginismus group as were 84% of women in the dyspareunia/PVD group were correctly classified. Physiological measures of fear (component 2, 0.580) had the highest loading on the discriminant function and referred to EMG taken during the gynecological examination and the sensory testing.

Differences between groups on the SCID, childhood sexual and physical abuse, and sexual functioning

SCID (specific phobia)

Significant differences between groups were found on the number of women meeting the diagnostic criteria for a specific phobia of vaginal penetration, χ^2 (2, N = 143) = 19.661 $p \leq 0.01$, with a significantly higher number of women in the vaginismus (n = 19) and dyspareunia/PVD (n = 13) groups meeting the diagnosis compared to women in the control group (n = 0).

Childhood experience of sexual and physical abuse

Chi-square analyses found no significant group differences on experienced sexual and physical abuse in childhood.

Sexual Functioning

One-way ANOVAs revealed significant differences between groups on the FSFI total score, F (2, 135) = 15.2, $p < .001$; desire score, F (2, 138) = 4.77, $p < .05$; FSFI arousal score, F (2, 138) = 5.4, $p < .05$; lubrication score, F (2, 137) = 6.42, $p < .05$; satisfaction score, F (2, 138) = 8.44, $p < .001$; and pain with intercourse score, F (2, 136) = 65.26, $p < .001$. No significant differences between groups were found with regards to the FSFI orgasm score, F (2, 138) = 1.99, $p = .140$. Tukey's HSD post-hoc comparisons revealed that women in the vaginismus and dyspareunia/PVD groups had lower mean scores on the FSFI total score, arousal score, lubrication score, satisfaction score, and pain with intercourse score compared to women in the control group. No significant differences were found between the vaginismus and dyspareunia/PVD groups. However, Tukey's HSD post-hoc test revealed that women in the dyspareunia/PVD group had significantly lower mean scores on desire compared to the control group while women in the vaginismus group did not differ from either women in the control group or women in the dyspareunia/PVD group on desire score.

Discussion

Overall, the current findings support all proposed hypotheses, and have important implications for the current DSM-5 proposal to collapse vaginismus and

dyspareunia under one diagnosis. Consistent with our first hypothesis, fear as measured by self-report, physiological, and behavioral measures was significantly greater in women suffering from vaginismus as compared with women suffering from dyspareunia/PVD, all of whom displayed significantly more fear than controls. The discriminant analyses demonstrated that behavioral measures of fear distinguished women suffering from vaginismus from those with dyspareunia/PVD and controls. Not only did vaginismic women display greater fear during the gynecological examination, a significantly higher percentage of these women (46%) were unwilling to finish or undergo the gynecological examination compared to only 6% of women in the dyspareunia/PVD group and 2% of controls. These data support and extend Reissing et al.'s (2004) results by having measured fear on multiple dimensions as well as by using blinded raters and a standardized observational system to measure behavior during the gynecological examination. These findings also suggest that women suffering from vaginismus are either more fearful of vaginal penetration than women suffering from dyspareunia/PVD or use more avoidant coping strategies in response to attempted penetration or both. Recent findings suggest that women suffering from vaginismus not only fear vaginal penetration situations, but also may have a general heightened fear/anxiety susceptibility (Nasab & Farnoosh, 2003; Watts & Nettle, 2010; Borg et al., 2012). Why this heightened susceptibility becomes focused on vaginal penetration remains unclear.

Consistent with our second hypothesis, vaginal muscle tension significantly distinguished women suffering from vaginismus from those with

dyspareunia/PVD and from controls. This finding is consistent with Reissing et al.'s (2004) study using digital palpation to evaluate pelvic floor muscle tension. Results from studies using surface electromyography (sEMG) to measure vaginal muscle tension as the major measurement method are, however, inconsistent (Shafik & El-Sibai, 2002; Frasson et al., 2009; Reissing et al., 2004; Van der Velde et al., 1999; Engman et al., 2004). These divergent findings may be secondary to limitations with sEMG methodology including placement variability, crosstalk, noise, and movement artifact (Gentilcore-Saulnier, McLead, Goldfinger, Pukall,& Chamberlain, 2009). Although results using digital palpation appear more consistent across studies, it should be emphasized that it remains a subjective assessment technique and may be influenced by the patient's affective reaction such as fear and pain (Reissing et al., 2004). For instance, in the present study, women suffering from dyspareunia/PVD were not found to differ from controls on vaginal muscle tension; this result is inconsistent with several other studies (Reissing et al., 2004; Engman et al., 2004; Gentilcore-Saulnier et al., 2009). Morin, Bergeron, Khalifé, Binik,& Ouellet (2010) developed a pelvic floor muscle evaluation instrument (ultrasonography) to overcome some of these shortcomings. It is likely that in the future ultrasonography will become an important method of assessing the pelvic floor since it does not require the insertion of a probe into the vaginal canal (Majida et al., 2009), making it an ideal method to assess whether pelvic floor muscle dysfunction is present in women suffering from vaginismus. This method has already been found useful in detecting pelvic floor muscle dysfunction in women suffering from

dyspareunia/PVD as well as in men with chronic pelvic pain (Morin et al., 2010; Davis, Morin, Binik, Khalife,& Carrier, 2011).

What appears most consistent across studies is the lack of evidence for vaginal muscle spasm as the defining characteristic of vaginismus. It is possible that increased pelvic floor muscle tension is, however, consistent with fear inducing such tension or with the possibility that vaginismus and dyspareunia/PVD may be part of a more "general defense reaction" (Van der Velde et al., 2001). It is not known whether pre-existing elevated levels of pelvic floor muscle tension increase susceptibility to vaginismus and dyspareunia/PVD or are the result of elevated fear/anxiety or both.

Our third hypothesis that women suffering from vaginismus like those suffering from dyspareunia/PVD experience significantly greater genital pain during attempted vaginal penetration than controls was also supported. This is consistent with several other studies demonstrating that a large percentage of women suffering from vaginismus also experience vulvar pain with attempted vaginal penetration and that this pain does not differ significantly in intensity, quality,or location from women with dyspareunia/PVD (Reissing et al., 2004; TerKuile, Van Lankveld, Vlieland, Willekes,& Weijenborg, 2005; Basson, 1996; De Kruiff et al., 2000; Engman et al., 2008). These findings suggest that the DSM-IV-TR diagnostic criteria for vaginismus are incomplete since the experience of genital pain is not mentioned. This further supports the notion that the differential diagnosis of dyspareunia/PVD from vaginismus based on the experience of genital pain may not be reliable.

The findings from this study also provide information concerning other important issues related to distinguishing vaginismus from dyspareunia. The literature presents inconsistent information concerning whether vaginismic women suffer from a higher incidence of sexual and physical abuse (e.g., APA, 2000, Biswas & Ratnam, 1995; Leiblum, 2000; Dupree Jones, Lehr,& Hewell, 1997, Reissing, Binik, Khalifé, Cohen,&Amsel, 2003; Watts et al., 2009; Barnes, 1986; Hawton & Catalan, 1990; O'Sullivan, 1979; van Lankveld, Brewaeys, Ter Kuile,& Weijenborg, 1995; Van Lankveld et al., 2006). Our findings suggest that they do not. The lack of a well-validated definition of sexual abuse may in part explain the divergent results across studies. Vaginismic women's tendencies to avoid and fear vaginal penetration may have led clinicians to suspect sexual abuse or traumatic sexual experiences. Despite these inconsistent findings on sexual abuse, the DSM-IV TR includes sexual abuse as an "associated feature" of vaginismus.

The DSM-IV-TR (APA, 2000) further mentions that sexual response is generally not impaired in vaginismic women. Our findings do not support this notion and suggest a lowered level of general sexual functioning in women suffering from vaginismus and dyspareunia/PVD replicating Reissing et al.'s (2004) findings. Repeated experiences of fear and pain during attempted vaginal penetration may affect a women's desire to have sex and her ability to become sexually aroused and lubricated. Sexual situations may have become threatening or stressful rather than pleasurable for these women.

Reissing et al. (2004) suggested that vaginismus could be conceptualized as a "vaginal penetration phobia." Specific phobias are characterized by "a marked and persistent fear that is excessive or unreasonable cued by the presence or anticipation of a specific object or situation. Exposure to the phobic stimulus almost invariably provokes an immediate anxiety response, which may take the form of a situationally bound or situationally predisposed panic attack. The person recognizes that the fear is excessive or unreasonable. The phobic situation(s) is avoided or else endured with intense anxiety or distress" (APA, 2000, p. 449).Self-report, behavioral, and psychophysiological measures demonstrated significant fear and avoidance in women with vaginismus during the gynecological examination suggesting that vaginismic women have several characteristics in common with individuals suffering from a specific phobia. One problem with characterizing vaginismic women as phobic is that over a third in our sample did not believe that their "fear was excessive or unreasonable" and therefore did not meet the diagnostic criteria for a specific phobia of vaginal penetration. If the current DSM-5 proposal is accepted then this criterion of excessive or unreasonable fear will be replaced with "fear and anxiety that is out of proportion to the actual danger posed by the specific object and situation" and would be assessed by the clinician rather than the individual (http://www.dsm5.org/ProposedRevision/Pages/proposedrevision.aspx?rid=162). Using this newly proposed criterion, almost all of the women excluded by the DSM-IV "excessive and unreasonable" criteria would now receive a phobic diagnosis. Categorizing vaginismus as a specific phobia may be useful

therapeutically as indicated by a recent study using flooding as the primary treatment (Ter Kuile et al., 2009).

This characterization, however, may have some disadvantages. For example, defining vaginismus as a specific phobia may lead to ignoring other potentially important symptoms such as genital pain and vaginal muscle tension. In addition, differences in fear of vaginal penetration between women suffering from vaginismus and those with dyspareunia/PVD may not only be related to the degree of fear but also to the use of different coping styles. Future studies should further investigate whether differences exist between women suffering from vaginismus and dyspareunia/PVD on their primary appraisal of vaginal penetration situations as well as on their coping response to stressful events in general.

Overall, results from this study do not fully support Binik's (2010) DSM-5 proposal to collapse vaginismus and dyspareunia into one category named "Genito-Pelvic Pain Penetration Disorder." Our findings demonstrate that fear and vaginal muscle tension can statistically distinguish women suffering from vaginismus from those with dyspareunia/PVD. The preliminary taxometric analyses also suggest that a small subgroup of women suffering from vaginismus appear to be categorically different on fear and vaginal muscle tension from the remaining women with vaginismus as well as from women suffering from dyspareunia/PVD. However, these analyses also suggest that the structure of the differences on fear, vaginal muscle tension, and genital pain appears to be dimensional for the majority of women suffering from vaginismus and

dyspareunia/PVD. Although complex statistical procedures are capable of distinguishing vaginismus from dyspareunia/PVD based on fear and vaginal muscle tension variables, this task may be more arduous for health professionals in a clinical setting. In fact, clinical research has confirmed that health professionals do not succeed in reliably distinguishing these conditions (Reissing et al., 2004; ter Kuile et al., 2005; Engman et al., 2007; Engman et al., 2008).This is not surprising given the large overlap observed in Figures 5 and 6 between both conditions on several dimensions. Collapsing vaginismus and dyspareunia into one category as proposed by Binik (2010) has certain advantages, however, such as increasing diagnostic reliability and forcing clinicians to carefully assess all the relevant dimensions of vaginismus and dyspareunia/PVD (i.e., vaginal penetration, genital pain, fear, and pelvic floor muscle dysfunction). The new category also does not rely on invalid criteria such as vaginal spasm. The use of Binik's (2010) proposed category would hopefully motivate a multidisciplinary team including gynaecologists, sex therapists, and pelvic floor muscle physiotherapists to be involved in the assessment and treatment of vaginismus and dyspareunia/PVD. On the other hand, this new category may blur the search for possible differences and treatments specific to one of the four diagnostic dimensions.

The present study has several limitations. First, the sample size (43-50/group) for the three groups was relatively small and may have resulted in inadequate power to detect important differences or to adequately exploit taxometric methodology. Second, 46% of women in the vaginismus group

discontinued the gynecological examination. The missing data from these women was replaced by the means of the group in which the participant was categorized. This method of dealing with missing values can be considered a very conservative strategy as it is highly likely that women who discontinued the gynecological examination would have displayed greater fear and vaginal muscle tension during the gynecological examination. Third, the participants were unlikely to have been a representative sample of women suffering from vaginismus in the general population. Those fearing and avoiding vaginal penetration situations the most would be the least likely to participate in a study involving a gynecological examination. This, however, is a problem common to all research and clinical studies of vaginismus since a pelvic examination is required to make a DSM-IV-TR diagnosis. Fourth, our analyses were based primarily on results from an attempted gynecological examination. Difficulties with or the inability to experience a gynecological examination are highly correlated with difficulties with or inability to experience intercourse but they are not identical. Finally, the dyspareunia group included only women suffering from PVD. A more heterogeneous sample of women suffering from dyspareunia may have resulted in different results and greater discrimination between the vaginismus and dyspareunia groups.

In conclusion, although measures of fear and vaginal muscle tension were able to discriminate vaginismus from dyspareunia/PVD, our findings also suggest a large overlap between both conditions on these dimensions and on the dimension of genital pain. The unwillingness to experience/attempt vaginal

penetration appears to be the single best differentiator of vaginismus from dyspareunia/PVD, but this may be a function of fear and associated coping styles. Translating these findings into reliable and valid diagnostic criteria for clinicians still remains a challenge.

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Table I. Components extracted from the principal component analysis

	Components	
Self-report measures of fear		
	Component 1	Self-reported anxiety during palpation of labia minora, pelvic floor muscles, uterus and adnexae; self reported anxiety during insertion of a cotton-swab, one finger, and two fingers; Self-reported anxiety during the cotton-swab test
	Component 2	Total scores on the SDSI, VPS, FSS-II, FPQ, PCS with reference to pain experienced during the gynecological examination; gynecologist's ratings of participants' degree of fear during the gynecological examination
	Component 3	Total scores on the STAI-S, on the STAI-T and on the PCS with reference to non-genital pain
Psychophysiological measures of fear		
	Component 1	Heart beat per minute and heart rate variability (low frequency/high frequency ratio) to during pain sensitivity testing and gynecological examination
	Component 2	EMG during pain sensitivity testing and gynecological examination
	Component 3	EDR during pain sensitivity testing and gynecological examination
Behavioral measures of fear		
	Component 1	Total number of protective behaviors; pelvic withdrawal frequency and intensity, opening legs frequency, closing legs frequency and intensity
	Component 2	Gynecologist's rating of defensive/avoidant behaviors during palpation of labia minora, during insertion of a cotton swab, during insertion of one finger, and during insertion of two fingers

Component 3	Total number of communicative behaviors; verbal frequency, grimacing frequency, neck arching frequency
Component 4	Frequency and intensity of placing one or both hands on head
Component 5	Verbal intensity, grimacing intensity
Component 6	Neck arching intensity

Genital pain

Component 1:	Self-reported pain intensities during palpation of pelvic muscles at sites 9, 3h, and 6 o'clock; self-reported pain intensities with insertion of a cotton-swab, 1 finger, and 2 fingers
Component 2:	Self-reported pain intensities during palpation of uterus and adnexae
Component 3:	Self-reported pain intensities during palpation of labia minora at 9 and 3 o'clock
Component 4:	McGill Pain Questionnaire PPI and PRI scores; self-reported pain intensities with cotton-swab test at 3, 6, and 9 o'clock
Component 5:	Deltoid and vulvar pain thresholds

Vaginal muscle tension

Component 1:	Degree of muscle tension according to Lamont's scale; gynecologist's rating of degree of muscle tension on an 11 point scale; gynecologist's ratings of difficulty inserting 2 fingers
Component 2	Gynecologist's ratings of difficulty inserting a cotton-swab and inserting one finger

Table II. Correlations between discriminating variables and standardized canonical discriminant functions (vaginismus, dyspareunia/PVD, control)

Discriminating variables	Function 1	Function 2
Genital pain component 4	.894*	.007
Self-report measures of fear component 2[a]	.474*	.169
Self-report measures of fear component 1 [a]	.444*	.196
Behavioral measures of fear component 3 [a]	.434*	.352
Genital pain component 1[a]	.397*	.272
Behavioral measures of fear component 5 [a]	.365*	.197
Behavioral measures of fear component 2 [a]	.347*	.340
Behavioral measures of fear component 6 [a]	.337*	.041
Genital pain component 3 [a]	.214*	-.062
Genital pain component 5 [a]	-.183*	-.070
Self-report measures of fear component 3 [a]	.158*	-.018
Behavioral measures of fear component 4 [a]	.125*	.097
Vaginal muscle tension component 1	.265	.586*
Behavioral measures of fear component 1	.505	.510*
Psychophysiological measures of fear component 2	-.178	.441*
Psychophysiological measures of fear component 1	.255	-.439*
Vaginal muscle tension component 2 [a]	.142	.246*
Genital pain component 2 [a]	.205	.245*
Psychophysiological measures of fear component 3 [a]	-.023	-.070*

*Largest absolute correlation between each variable and any discriminant function

[a] This variable was not used in the analysis

Table III. Correlations between discriminating fear variables and standardized canonical discriminant functions (vaginismus, dyspareunia/PVD, controls)

Discriminating variables	Function 1	Function 2
Behavioral measures of fear component 1	.679*	.472
Behavioral measures of fear component 5	.616*	.100
Self-report measures of fear component 2	.600*	.139
Behavioral measures of fear component 3 [a]	.583*	.298
Self-reported measures of fear component 1 [a]	.419*	.168
Behavioral measures of fear component 6 [a]	.306*	.007
Self-report measures of fear component 3 [a]	.218*	.078
Psychophysiological measures of fear component 1	.315*	-.596*
Psychophysiological measures of fear component 2	-.214	.577*
Behavioral measures of fear component 4 [a]	.142	.161*
Psychophysiological measures of fear Component 3 [a]	.021	-.104*

*Largest absolute correlation between each variable and any discriminant function

[a] This variable was not used in the analysis

Table IV. Correlations between discriminating variables and standardized canonical discriminant functions (vaginismus and dyspareunia/PVD)

Discriminating variables	Function 1
Behavioral measures of fear component 1	.658
Vaginal muscle tension component 1	.599
Behavioral measures of fear component 3 [a]	.419
Psychophysiological measures of fear component 2	.387
Behavioral measures of fear component 2 [a]	.361
Behavioral measures of fear component 5 [a]	.308
Self-report measures of fear component 2[a]	.287
Self-report measures of fear component 1 [a]	.278
Psychophysiological measures of fear component 3	.276
Vaginal muscle tension component 2 [a]	.252
Genital pain component 1 [a]	.235
Genital pain component 5 [a]	-.222
Genital pain component 2 [a]	.177
Genital pain component 4 [a]	.162
Behavioral measures of fear component 4 [a]	.153
Behavioral measures of fear component 6 [a]	.125
Self-report measures of fear component 3 [a]	.097
Genital pain component 3 [a]	-.013
Psychophysiological measures of fear component 1 [a]	.008

[a] This variable was not used in the analysis

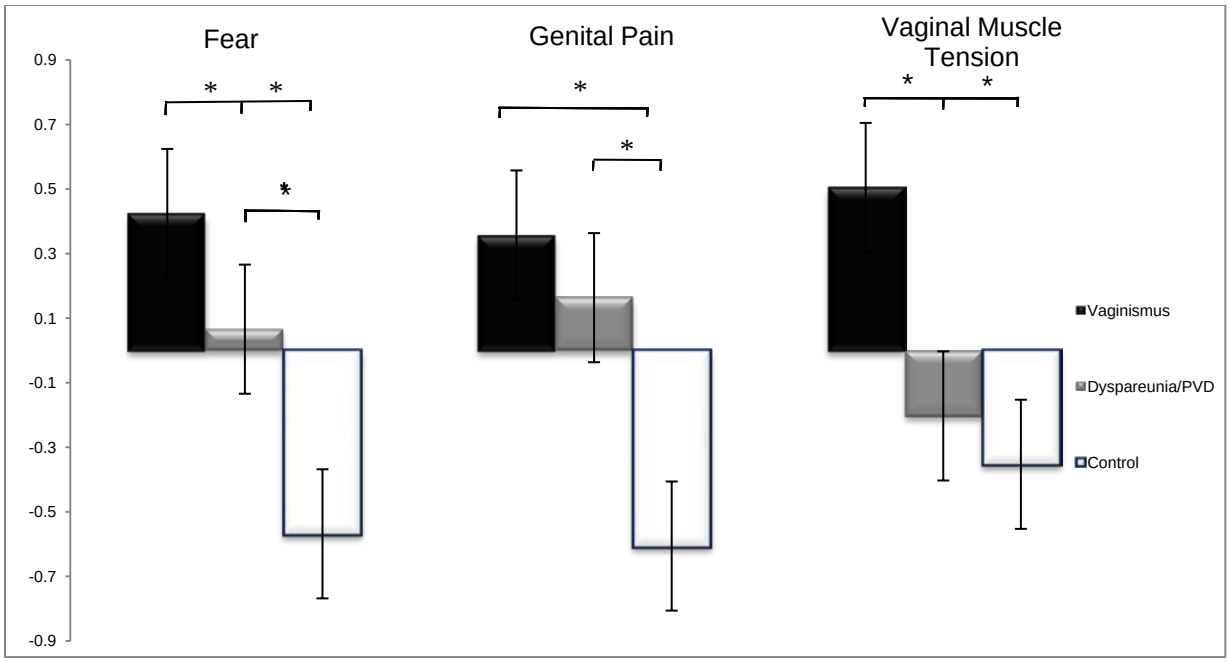

Figure 1 Differences between groups on the mean standardized total scores for fear, genital pain and vaginal muscle tension with the standard deviations.

* $p < .001$

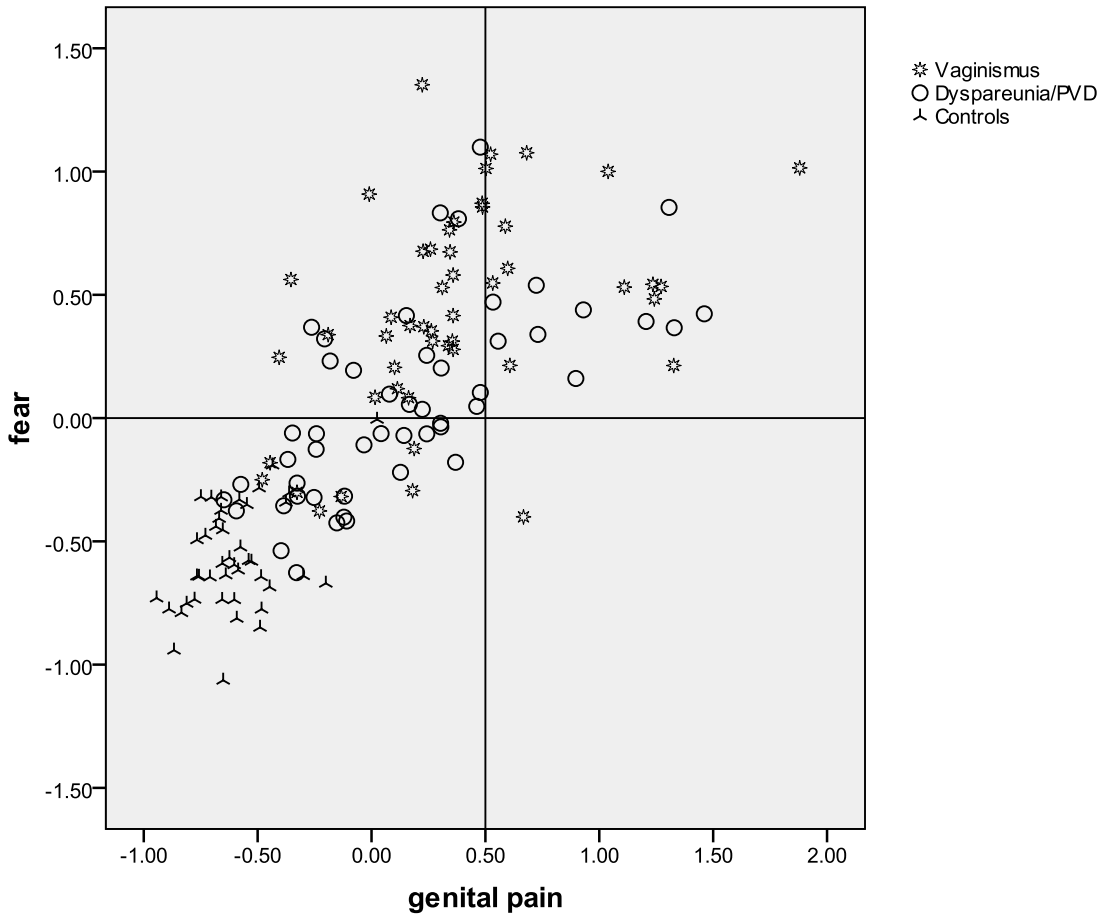

Figure 2. Scatter plots displaying each participant's scores on the genital pain and fear total scores.

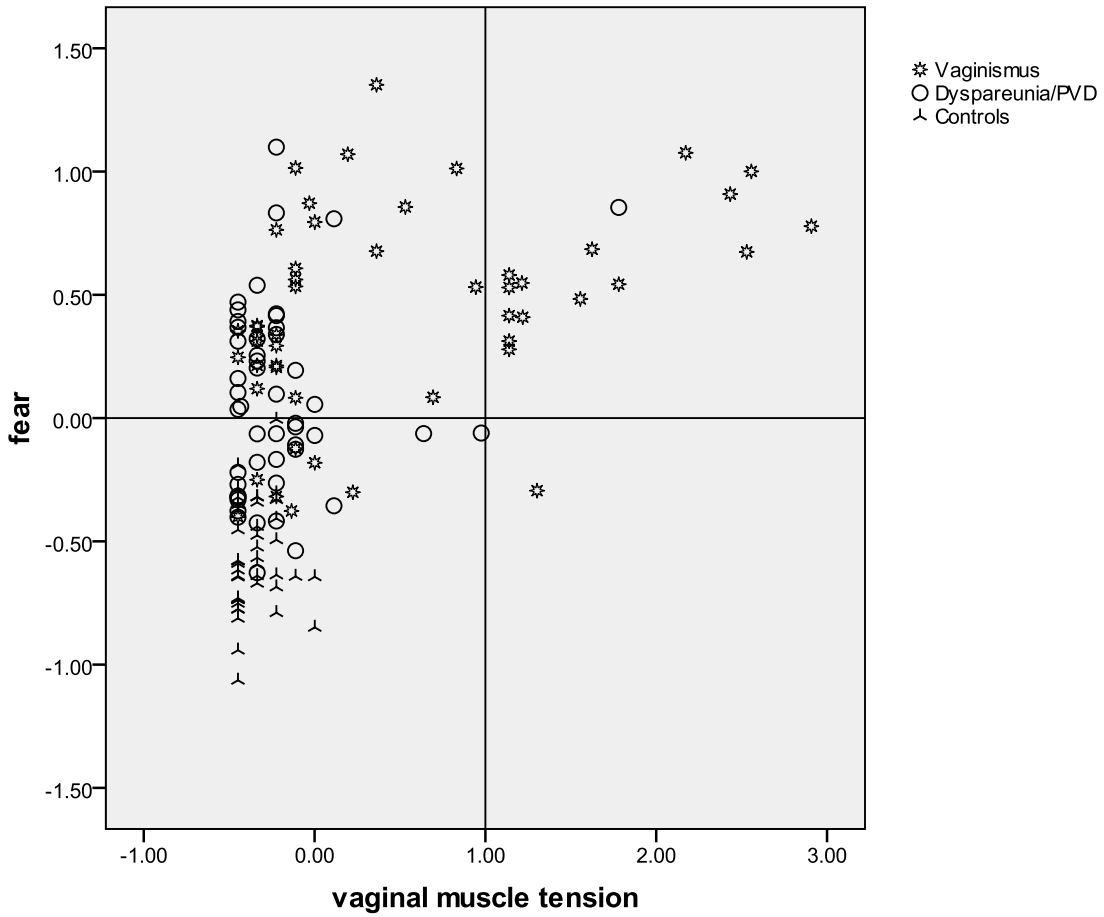

Figure 3. Scatter plots displaying each participant's scores on the vaginal muscle tension and fear pain total scores.

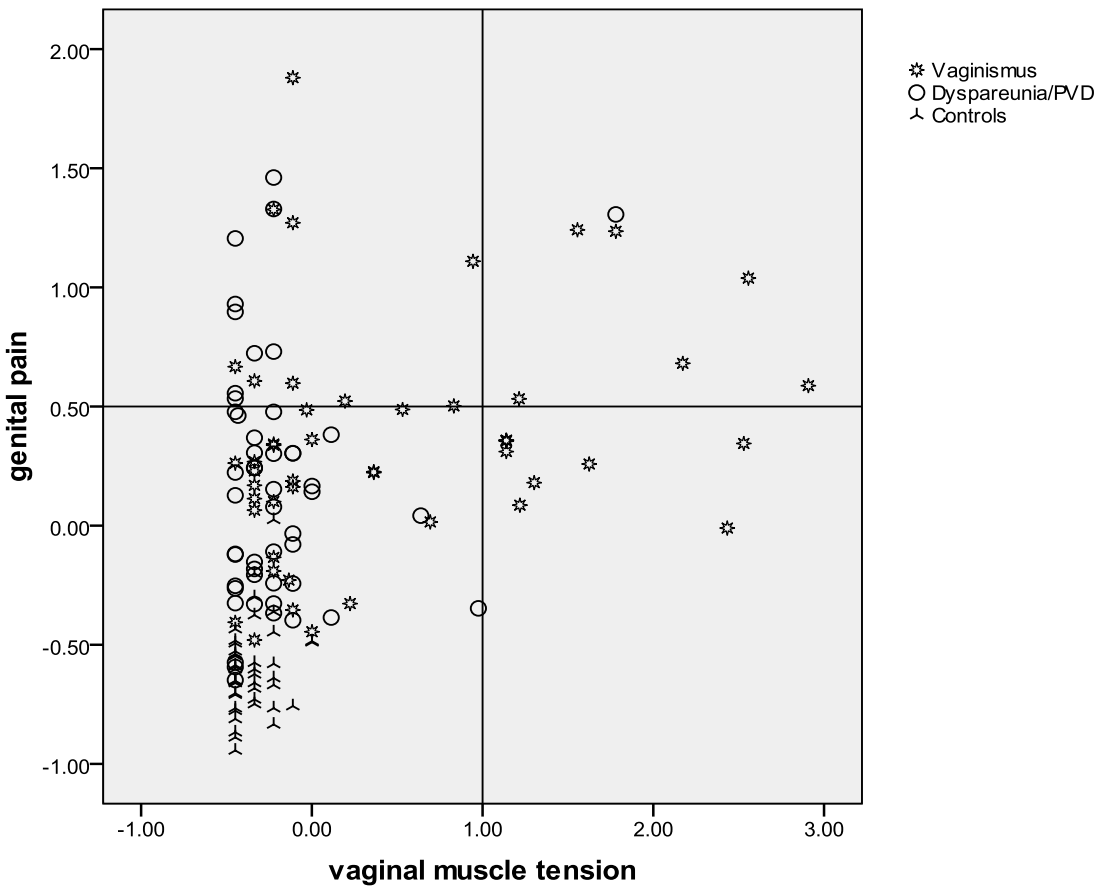

Figure 4. Scatter plots displaying each participant's scores on the vaginal muscle tension and genital pain total scores.

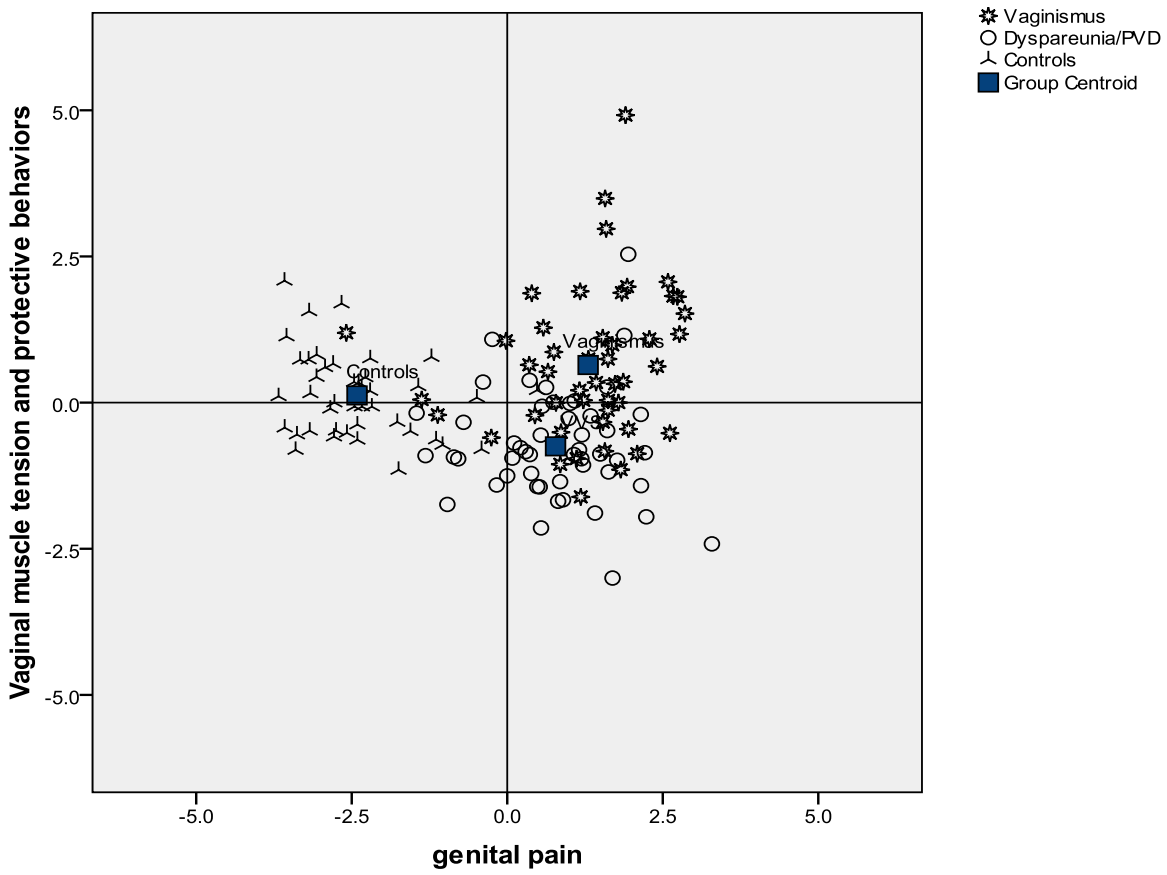

Figure 5. Canonical discriminant coefficient functions discriminating the vaginismus, dyspareunia/PVD and control groups

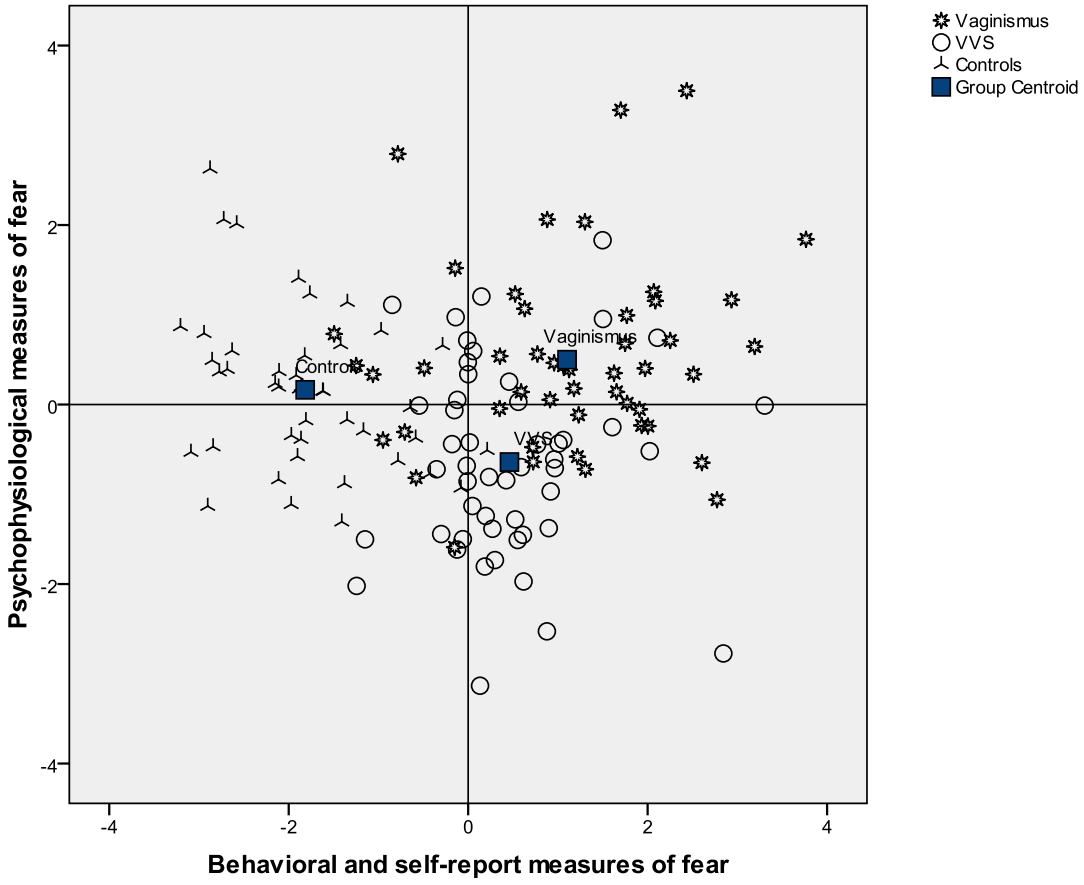

Figure 6. Canonical discriminant coefficient functions on fear components discriminating the vaginismus, dyspareunia/PVD and control groups

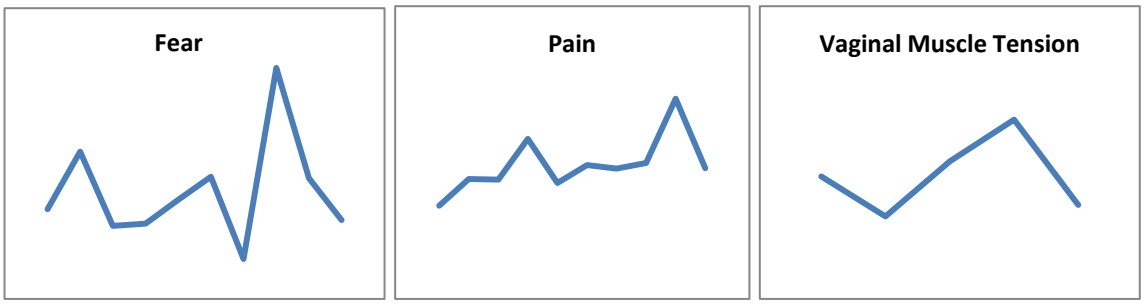

Figure 7. MAXCOV-HITMAX curves based on the plotted covariances along ordered subsamples of fear, genital pain and vaginal muscle tension.

Literature Review Update

In order to update the literature review presented as the first chapter of this thesis (Lahaie, Boyer, Amsel, Khalifé,& Binik, 2010), I conducted MEDLINE, PsychINFO and EMBASE searches using "vaginismus" as a key word. These searches resulted in 11 new publications on the following topics: 1) prevalence (N=1; Christensen et al., 2011); 2) classification/diagnosis (N=1; Basson, Wierman, van Lankveld & Brotto, 2010); 3) etiology (N=6; Huijding, Borg, Weijmar-Shultz,& de Jong, 2011; Borg, de Jong,& Weijmar Shultz, 2010; Borg, de Jong & Weijmar-Schultz, 2011; Watts & Nettle, 2010; Borg, Peters, Weijmar Schultz,& de Jong, 2012; Reissing, 2012); 4) treatment (N=3; Fageeh, 2011; Pacik, 2009; Jindal & Jindal, 2010); 5) health-seeking behaviors (N=1, Reissing, 2012).

1. Prevalence

Christensen et al. (2011) conducted a population-based epidemiologic study with the aim of updating prevalence estimates for sexual dysfunctions and sexual difficulties in Denmark and of identifying sociodemographic factors associated with sexual dysfunctions. Vaginismus was defined as "vaginal cramps that precluded penetration" and was considered to be a sexual dysfunction in the epidemiologic survey if it occurred frequently and was regarded as a problem by the respondent. Using these criteria, .4% of Danish women reported vaginismus as a sexual dysfunction. An additional 4% of women reported "vaginal cramps that precluded penetration" as a sexual difficulty (i.e., occurring "rarely" or "sometimes" but considered a problem, or occurring "often" or "everytime" but not

considered a problem). Vaginismus was most common in women under the age of 30. Two earlier population-based prevalence studies, one conducted in Denmark and one in Sweden, investigated the prevalence of vaginismus and their findings are consistent with those of Christensen et al. (Ventegodt, 1998; Fugl-Meyer, Sjogren Fugl-Meyer, 1999). There is converging evidence that vaginismus is a relatively rare sexual dysfunction in the Scandinavian population. Whether this translates to other populations remains unknown though there are suggestions that vaginismus may be more frequent in other cultural contexts (e.g., Hiller, 2000; Ng, 1999; Ng, 2000; Tugrul & Kabakçi, 1997). Conducting cross cultural research using similar methods and definitions of vaginismus would be an important next step.

2. Classification/Diagnosis

Basson et al. (2010) discuss a new definition of vaginismus which was proposed in an earlier article published in 2003 (Basson et al., 2003). The proposed definition of vaginismus is "persistent or recurrent difficulties for the woman to allow vaginal entry of a penis, a finger, and/or object, despite the woman's expressed wish to do so. There is often (phobic) avoidance, involuntary pelvic muscle contraction and anticipation/fear/experience of pain." Basson et al. (2003) were the first to provide a new definition of vaginismus that includes genital pain and fear. In line with their conceptualization of vaginismus as a multifactorial condition, Basson et al. (2010) recommend a biopsychosocial approach for the assessment and treatment of vaginismus. These new recommendations are consistent with the conclusions of our literature review that

important limitations exist with the current definition of vaginismus and that a multidisciplinary diagnostic and treatment approach is suggested to address the different dimensions of vaginismus. These recommendations also concur with Binik's (2010) recommendation that fear and pain are important defining characteristics of women suffering from vaginismus.

3. Etiology

Disgust

Most of the new etiological studies have focused on the role disgust and fear/anxiety play in vaginismus. In one study, women suffering from vaginismus were found to evaluate sexual penetration stimuli more negatively than neutral stimuli but this was not the case for women with dyspareunia (Huijding et al., 2011). In a second study by the same research group, sexual stimuli were found to elicit "automatic disgust" for both women with vaginismus and dyspareunia (Borg et al., 2010). Automatic reflexive disgust was investigated by measuring reaction time to sexual penetration pictures and by recording facial electromyography of the levator labii muscle region, a physiological marker of disgust. The automatic reflexive disgust associations were found to persist longer in vaginismic women as compared to women with dyspareunia. Moreover, women with vaginismus showed enhanced subjective disgust (i.e., reflective, explicit attitudes) to sexual stimuli as compared with women in the dyspareunia and control groups. The authors concluded that autonomic/reflexive disgust associations to sexual stimuli appear to be involved in both vaginismus and dyspareunia which may explain their shared difficulties with vaginal penetration.

However, women with dyspareunia did not display enhanced subjective disgust to sex stimuli. The authors report that unlike women with vaginismus, women with dyspareunia may overcome this automatic/reflexive association between sex and disgust which may explain why penetration is still possible in dyspareunia but not in vaginismus.

The same research group examined whether strong adherence to conservative moral principles, defined as a tendency to limit actions and impulses or a general difficulty with transgression, and less adherence to liberal ones could be involved in the negative affective responding toward particular sexual behaviors in vaginismus and dyspareunia (Borg et al., 2011). They found that vaginismic women scored higher on conservative moral values and lower on liberal ones, and showed greater resistance to engage in particular sex-related behaviors (e.g. "touch and carefully examine sex aids"; "visit a sex shop", "join a swinger group") compared to controls. Women with dyspareunia scored between those with vaginismus and controls not differing significantly from either group on conservative moral values, liberal values, or resistance to engage in particular sexual behaviors. The authors concluded that highly conservative values in addition to resistance to engage in particular sex-related behaviors may be a pathway towards the development and maintenance of vaginismus.

Results from these investigations suggest that disgust is implicated in both vaginismus and dyspareunia and that differences between these conditions may lie more in the severity of the disgust association supporting a dimensional distinction between vaginismus and dyspareunia. Moreover, the findings from

one study (Borg et al., 2011) suggest that one pathway toward developing this negative association may be the adherence to high conservative values and to strict sexual standards. Although this is an interesting theory, the fact that these studies are cross-sectional does not allow for etiology to be determined. It is also possible that disgust and adherence to conservative moral values are consequences of experiencing difficulties with vaginal penetration rather than causes.

Anxiety/fear

Two recent studies investigated the role of fear/anxiety in women suffering from vaginismus. One study found that women with vaginismus score significantly higher on state and trait anxiety as compared with controls (Watts & Nettle, 2010). The trait anxiety that women with vaginismus present may affect their general perception of situations as dangerous or threatening. Why the anxiety becomes focused on vaginal penetration remains unknown. However, one possibility is related to recent findings of heightened levels of catastrophic pain cognitions and harm avoidance in women suffering from vaginismus as compared with women with dyspareunia and women with no sexual complaints (Borg et al., 2012). Given that genital pain appears to be an important characteristic of women suffering from vaginismus (e.g., Reissing, Binik, Khalife, Cohen,& Amsel, 2004; Ter Kuile, Van Lankveld, Vlieland, Wilekes,& Weijenborg, 2005; Basson, 1996; De Kruiff, Ter Kuile, Weijenborg,& Van Lankveld, 2000), vaginismic women may be more inclined to react to the anticipated or experienced genital pain with anxiety and emotional distress resulting in greater

avoidance of vaginal penetration situations. These findings concur with the conclusions from our literature review suggesting that fear/anxiety appears to characterize women suffering from vaginismus.

In line with these findings, Reissing (2012) recently conducted a survey investigating vaginismic women's beliefs about the causes of their condition with "fear of pain based on previous painful attempts" being the most frequently reported. Women with lifelong vaginismus also indicated causal beliefs related to "*expectation*

that intercourse is painful", "fear of injury", fear of losing control" as well as disgust-based attributions. These results concur with Ward and Ogden's (1994) previous survey findings and provide further support to the role of fear and pain in vaginismus. These results also suggest that women with lifelong vaginismus may differ from women with acquired vaginismus on the intensity and quality of their affective reactions to sexual stimuli.

Results from these three new investigations support the notion that anxiety/fear characterize women with vaginismus. Moreover, these findings suggest that the anxiety/fear may not be specific to vaginal penetration only. The format of these studies, however, does not allow conclusions to be made regarding etiology.

4. Treatment

Botox

Two uncontrolled retrospective studies investigated the use of Botox in women suffering from vaginismus and reported excellent success rates. Fageeh

(2011) reported a 100% success rate with the use of Botox in six vaginismic women who had not benefitted from "conventional behavioral therapy". Pacik (2009) described that out of 20 vaginismic patients treated with intra-vaginal Botox injections, 16 were able to achieve intercourse within 3 months; three were still under treatment, and one was considered a failure.

Sensate Focus

One uncontrolled clinical study examined the effectiveness of a modified sensate focus technique which included counseling and education, pelvic floor muscle relaxation and contraction exercises, gradual desensitization, counseling of male partner and sensate focus exercises aimed at improving communication and the relationship (Jindal & Jindal, 2010). Out of the 63 women suffering from vaginismus who participated in this study, a complete resolution was reported in 60 women. Complete resolution was defined as having resolved the vaginismus by having achieved sexual intercourse and pregnancy. Twenty five women achieved pregnancy through sexual intercourse, five through intrauterine insemination, and three through in vitro fertilization. For women who achieved pregnancy through intrauterine and in vitro fertilization, complete resolution was considered only if they had successfully achieved sexual intercourse. Unfortunately, similar to most treatment outcome studies reported in our literature review, these three studies present many methodological limitations including lack of a control or placebo group, small sample sizes, and lack of standardized measurement instruments.

5. Health-seeking behaviors

Using an online survey, Reissing (2012) examined the health-seeking behaviors of women suffering from lifelong and acquired vaginismus and found that the health professionals most frequently consulted were gynecologists and family doctors with only gynecologists being rated as helpful. These findings concur with Ogden & Ward's (1995) survey results. Reissing (2012) also examined which interventions vaginismic women rated as most helpful. Women suffering from vaginismus reported that educational gynecological examinations, talking about the meaning of the penetration problem with a health professional, vaginal dilatation, and sex education were the most helpful interventions. Although physiotherapists were reported to be less commonly consulted by vaginismic women, they were considered to be the most helpful health professionals. These findings are in line with the recommendations made in our literature review that a multidisciplinary team including a gynecologist, physical therapist and psychologist/sex therapist should be involved in the assessment and treatment of vaginismus.

Conclusion

Since our literature review, there has been continued interest in the classification/diagnosis, prevalence, etiology and treatment of vaginismus. The finding that vaginismus is a relatively rare sexual dysfunction in the Scandinavian population is an important contribution and would be important to replicate cross culturally. Most of the new etiological studies have focused on the role of disgust and fear/anxiety in vaginismus. These etiological studies unlike the vast majority of previous ones used control groups, standardized measurement instruments

and appropriate statistical methods. The role of genital pain and vaginal muscle tension in vaginismus has received less empirical interest since our literature review. This contrasts with the treatment studies as two out of the three new treatment outcome studies for vaginismus focused on pharmacological interventions aimed at eliminating the vaginal muscle spasm component of vaginismus. Similar to most treatment studies for vaginismus to date, they report high success rates and present many methodological limitations.

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GENERAL CONCLUSIONS AND DIRECTIONS FOR FUTURE RESEARCH

The literature review and empirical investigation included in this dissertation examine the role of fear in vaginismus and the ability of this variable to distinguish vaginismus from dyspareunia/PVD. The first chapter reviews the research evaluating the classification/diagnosis, etiology and treatment of vaginismus and discusses the DSM-5 proposal to collapse vaginismus and dyspareunia under one category (Binik, 2010). The review reveals that: 1) The DSM-IV-TR (APA, 2000) definition of vaginismus is not supported by current empirical investigations and clinical experience; 2) Genital pain is an important characteristic of most women suffering from vaginismus; 3) Vaginismus and dyspareunia/PVD are difficult to differentiate; 4) Fear is an under investigated factor that appears to characterize women suffering from vaginismus.

The second chapter presents the results from our empirical investigation which examined whether the degree of fear displayed during a gynecological examination could discriminate women suffering from vaginismus from those with dyspareunia/PVD and controls. Genital pain, vaginal muscle tension, sexual functioning and childhood sexual and physical abuse were also re-examined.

The most important conclusions that can be drawn from this study are that: 1) Fear and vaginal muscle tension appear to characterize women with vaginismus and to distinguish them from women with dyspareunia/PVD and controls; 2) Although fear and vaginal muscle tension were found to statistically distinguish vaginismus from dyspareunia, a large overlap was observed between both conditions on fear, vaginal muscle tension, and genital pain; 3) Vaginismus,

as currently diagnosed, is a multifactorial condition comprising of fear, genital pain, and vaginal muscle tension.

Behavioral measures of fear were found to be one of the most important factors distinguishing vaginismus from dyspareunia/PVD; women in the dyspareunia/PVD group also displayed greater fear than controls. In addition, a high percentage of women in the vaginismus group were unwilling to terminate or undergo the gynecological examination compared to women in the dyspareunia/PVD and control groups. However, whether women with vaginismus avoid more vaginal penetration situations than women with dyspareunia/PVD as a result of greater fear or as a result of using more avoidant coping strategies or both remains unclear because coping has never been investigated. Recent findings support the greater fear hypothesis and demonstrate that vaginismic women display heightened levels of catastrophic pain cognitions and of harm avoidance compared to women with dyspareunia and women with no sexual complaints (Borg, Peters, Weijmar Schultz,& de Jong, 2012). The authors concluded that in line with the fear-avoidance model, the heightened pain catastrophizing found in women with vaginismus may promote hypervigilance to potential pain stimuli and result in avoidance (Leeuw, Goossens, Linton, Crombez, Boersma,& Vlaeyen, 2007). Although this is an interesting possibility, women suffering from dyspareunia/PVD avoid significantly less vaginal penetration situations despite high levels of hypervigilance to pain, fear of pain and catastrophic cognitions to sexual pain (Payne, Binik, Amsel,& Khalifé, 2005; Pukall, Binik, Khalifé, Amsel,& Abbott, 2002). Another possibility is that women

suffering from vaginismus use more avoidant coping strategies in response to painful stimuli and/or in response to stressful events in general than women suffering from dyspareunia/PVD. This hypothesis could be tested by assessing coping styles of women with vaginismus and dyspareunia/PVD through validated coping questionnaires. Avoidance coping could also be compared between vaginismic and dyspareunia/PVD women while being exposed to laboratory induced non-genital pain conditions and to non-painful stressful conditions. There are, to date, no such studies.

Heightened vaginal muscle tension was also found in our study to characterize women suffering from vaginismus and to distinguish them from those with dyspareunia/PVD. There is, however, conflicting evidence as to whether women with vaginismus present hypertonic pelvic floor muscles. This conflicting evidence may be a result of methodological limitations with the current measurement techniques used to investigate pelvic floor muscle function in vaginismic women (i.e., digital palpation and surface electromyography; Gentilcore-Saulnier, McLean, Goldfinger, Pukall,& Chamberlain, 2009). Digital palpation and sEMG are invasive techniques requiring the insertion of one finger or a probe into the vaginal canal. The most fearful and avoidant vaginismic women would be the least likely to participate in such studies. A relatively new, non-invasive, and reliable pelvic floor muscle evaluation method using ultrasonography overcomes many of the current limitations with digital palpation and sEMG, in addition to not requiring any vaginal insertion (Majida et al., 2009; Majida, Braekken, Bo, Benth,& Engh, 2010). Using ultrasonography may allow

for a more objective assessment of pelvic floor muscle function in women suffering from vaginismus and dyspareunia/PVD. It could also provide valuable information regarding the interplay between fear, genital pain, and vaginal muscle tension. For instance, ultrasonography could be used to investigate Van der Velde, Laan,& Everaerd's (2001) general defense reaction hypothesis which claims that the involuntary contractions of the pelvic floor muscles in women with vaginismus are a result of automatic defensive reactions. This could be done by examining the activity of the pelvic floor muscles when vaginal penetration is attempted. Such study would provide important new information regarding the role of pelvic floor muscle in vaginismus as well as help to disentangle the relationship between fear, genital pain, and vaginal muscle tension.

Although our findings demonstrate that fear and vaginal muscle tension are capable of statistically distinguishing vaginismus from dyspareunia/PVD, a large overlap was observed between both conditions on several dimensions. Preliminary taxometric analysis suggested that a group of women scoring in the upper 20^{th} percentile of fear and vaginal muscle tension was distinct from the remaining $80^{\%}$. This group comprised mainly of women from the original vaginismus group. Our relatively small sample size (43-50/group), however, did not allow to adequately exploit taxometric methodology. A replication of our study using a larger sample of women with vaginismus and dyspareunia/PVD may allow for a better use of taxometric analyses. A larger study using taxometric analyses may also provide additional information regarding the small group of vaginismic women which was found to be categorically different from the

remaining women on fear and vaginal muscle tension. Whether this small group of women represent a different disorder in itself would be interesting to further investigate.

Our findings support the notion that vaginismus is a multifactorial disorder including fear, genital pain, and vaginal muscle tension symptomatology. Our results also support the DSM-5 proposal of defining vaginismus using all of the above dimensions. We therefore believe that these women would benefit from a multidisciplinary diagnostic and intervention approach including gynecologist, physiotherapist and sex therapist. There are, however, to date no treatment outcome study investigating the efficacy of such multidisciplinary assessment and interventions in vaginismus. Most treatments for vaginismus to date have focused on eliminating the vaginal muscle spasm through either pharmacological, psychological or pelvic floor physiotherapeutic means. Ter Kuile et al. (2009), using a prolonged and therapist-aided exposure therapy, investigated a treatment for vaginismus focusing explicitly and systematically on the fear of coitus. Nine out of ten participants were able to engage in intercourse following only on average 126 minutes of treatment exposure. Their exposure treatment was also successful in decreasing fear and negative penetration beliefs. Although these results are very encouraging, none of the measures of sexual enjoyment or pleasure significantly improved. The lack of improvement on sexual function may be a result of not having addressed sexual function, genital pain, and vaginal muscle tension. A future direction would be to investigate the effectiveness of interventions for vaginismus addressing all dimensions of this

condition including fear, genital pain, pelvic floor muscle function, and sexual functioning.

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Appendix-1 Reprint of Publication

Vaginismus: a review of the literature on the classification/diagnosis, etiology and treatment

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Vaginismus is currently defined as an involuntary vaginal muscle spasm interfering with sexual intercourse that is relatively easy to diagnose and treat. As a result, there has been a lack of research interest with very few well-controlled diagnostic, etiological or treatment outcome studies. Interestingly, the few empirical studies that have been conducted on vaginismus do not support the view that it is easily diagnosed or treated and have shed little light on potential etiology. A review of the literature on the classification/diagnosis, etiology and treatment of vaginismus will be presented with a focus on the latest empirical findings. This article suggests that vaginismus cannot be easily differentiated from dyspareunia and should be treated from a multidisciplinary point of view.

Vaginismus is described as an involuntary vaginal muscle spasm interfering with sexual intercourse [1]. Since the term was first coined in the 19th Century, vaginismus has been conceptualized as a relatively infrequent but well understood and easily treatable female sexual dysfunction. In 1859, gynecologist Sims wrote that 'from personal experience, I can confidently assert that I know of no disease capable of producing so much unhappiness to both parties of the marriage contract, and I am happy to state that I know of no serious trouble that can be cured so easily, so safely and so certainly' [2]. This conceptualization was perpetuated by Masters and Johnson who reported a treatment outcome success rate of 100% [3]. It seems likely that this presumed high cure rate and lack of diagnostic controversy deterred new research. In fact, Beck described vaginismus as 'an interesting illustration of scientific neglect' [4].

Since Reissing *et al.*'s review of the vaginismus literature, a few important empirical studies on the diagnosis and treatment of vaginismus have been published [5]. Interestingly, their results challenge the validity of the current definition of vaginismus as well as the notion that it is an easily diagnosable and treatable condition. The current article will examine the literature on the classification/diagnosis, etiology and treatment of vaginismus with a focus on the latest empirical findings.

Prevalence

There are no epidemiological studies examining the population prevalence of vaginismus. This may be true since such a study would probably require a stressful gynecological examination that sufferers might often prefer to avoid. As a result, there have been dramatically varying estimates regarding the prevalence of this problem. Some such as Masters and Johnson claim that it is a relatively rare condition [3,6], while others suggest that it is one of the most common female psychosexual dysfunctions [7–10]. Although the population prevalence remains unknown, the prevalence rates in clinical settings have been reported to range between 5–17% [11].

In a British study, Ogden and Ward examined the help-seeking behaviours of women suffering from vaginismus and found that the professional most frequently consulted was the general practitioner [12]. Unfortunately, their respondents reported that general practitioners were the least helpful health professional they consulted. Overall, there was general dissatisfaction with available help, which may reinforce many vaginismic women's pre-existing avoidance in seeking help. This is consistent with Shifren *et al.*'s findings in the USA that only a third of women with 'any distressing sexual problem' consult [13]. According to their sample, the barriers for receiving professional help were poor self perceived health and embarrassment in discussing sexual problems.

Classification & diagnosis

Vaginal muscle spasm

In her 1547 treatise on 'The Diseases of Women', Trotula of Salerno is thought to have provided the earliest description of what we today call vaginismus: 'a tightening of the vulva so that

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Keywords

• dyspareunia • fear of vaginal penetration • pelvic floor physiotherapy • pharmacotherapy • provoked vestibulodynia • psychological treatments • sexual abuse • vaginal muscle spasm • vulvar pain

even a woman who has been seduced may appear a virgin' [14]. Much later, Huguier gave the first medical description of the syndrome; however, it appears that Sims first coined the term 'vaginismus' in 1862 while addressing the Obstetrical Society of London [15]. Sims described vaginismus as 'an involuntary spasmodic closure of the mouth of the vagina, attended with such excessive supersensitiveness as to form a complete barrier to coition' [2]. To date, the involuntary muscle spasm remains the core element of the definition of vaginismus suggested by the American College of Obstetrics and Gynecology (ACOG) and by the Diagnostic and Statistical Manual of Mental Disorders-IV-TR (DSM-IV-TR) [1,16]. The International Classification of Diseases (ICD)-10 categorizes vaginismus either as a 'pain disorder' or as a 'sexual dysfunction comprised of a spasm of the pelvic floor muscles that surround the vagina, causing the occlusion of the vaginal opening with penile entry being either impossible or painful' [17].

This 150-year consensus concerning the definition of vaginismus is striking given the lack of empirical findings validating the vaginal muscle spasm criterion [5]. In fact, Reissing *et al.* (n = 87) found that although vaginismic women demonstrated a greater frequency of vaginal muscle spasm while undergoing a gynecological examination than did age, relationship and parity matched healthy controls or women suffering from dyspareunia associated with provoked vestibulodynia (PVD), only 28% of the vaginismus group actually displayed a vaginal muscle spasm. Moreover, only 24% reported experiencing spasms with attempted intercourse. Even more puzzling was the finding that two independent gynecologists agreed only 4% of the time on the diagnosis of vaginismus [18]. These findings call into question the primary diagnostic criterion of vaginismus.

Another method of evaluating the validity of the vaginal muscle spasm criterion is via the electrical recording of muscle activity, which can be done through surface electromyography (sEMG) or needle electromyography. Recent sEMG and needle EMG studies have investigated the activity of the pelvic floor muscles in women diagnosed with vaginismus. Reissing *et al.* found that women with vaginismus displayed lower pelvic floor muscle strength and greater vaginal/pelvic muscle tone compared with matched controls but no significant differences at all between the vaginismus and PVD group [18,19]. Shafik and El-Sibai (n = 14) also demonstrated through needle EMG, a higher EMG activity at rest and on induction of the vaginismus reflex in the levator ani, puborectalis and bulbocavernosus muscles in women with vaginismus compared with age-matched controls [20]. Consistent with the findings above, Frasson *et al.* (n = 30) found significant needle EMG basal and reactive hyperexcitability in primary lifelong vaginismus and in women with PVD accompanied by vaginismus as compared with controls [21]. On the other hand, three well-controlled sEMG (ranging from 29 to 224) studies did not confirm a significant difference in ability to contract and relax the pelvic floor muscles between women with and without vaginismus [22–24].

These contradictory results may be partially explained by the lack of an operationalized definition of the term 'muscle spasm' as well as the lack of consensus regarding which muscles are involved in vaginismus. Some authors refer to broad groups of muscles such as the muscles of the outer third of the vagina, the pelvic muscles or the circumvaginal and perivaginal muscles [25–29], while others refer to more specific ones, such as the bulbocavernosus, the levator ani and puboccoccygeus [30,31]. None of these studies indicate how they concluded which muscles are involved [5]. The term spasm itself is also controversial as there is no agreement on whether spasm refers to an involuntary muscle cramp, a defensive mechanism or a hypertonicity of the pelvic floor muscles.

In addition to the lack of agreement regarding the term muscle spasm and the muscles involved in vaginismus, there is no empirically standardized diagnostic protocol for vaginal muscle spasm. Although Masters and Johnson claimed that a pelvic exam was necessary to diagnose vaginismus, researchers and clinicians have frequently relied on self report of difficulties with vaginal penetration [2,32]. The lack of a standardized diagnostic protocol is not a trivial problem since studies concerning vaginismus may well include highly diverse samples. The fact that studies using the vaginal muscle spasm DSM-IV-TR definition of vaginismus failed to find a vaginal spasm suggests that vaginal muscle spasm is not a reliable diagnosis and as a result diverse patient populations might have been included [21–24].

Pain

Even though vaginismus is classified as a sexual pain disorder in the DSM-IV-TR, pain is not mentioned in the diagnostic criteria. Other definitions of vaginismus such as those published

by the ACOG [16], the International Association for the Study of Pain (IASP), the WHO and Lamont do mention pain in their definitions [17,33,34]. However, no description of the pain characteristics, such as location, quality, intensity and duration are provided [32]. There is also a lack of information regarding whether the pain is a cause or consequence of the vaginal muscle spasm [32]. While most clinical reports and research concerning vaginismus do not make reference to the pain element in vaginismus [35], some authors believe that pain is one of its core components [10,18,36–40]. In fact, several studies have found that a large percentage of women suffering from vaginismus experience pain with attempted vaginal penetration [18,25,35,37,40–43]. The pain experienced by women with vaginismus has been found to be very similar to the pain reported by women with PVD [18,40,42].

According to the DSM-IV-TR, vaginismus can be classified as either lifelong (primary) or acquired (secondary). It has often been suggested that PVD may result in acquired vaginismus [31,34,44]. Although lifelong and acquired vaginismus are generally considered to differ in their etiology and response to treatment, there are no empirical data validating these claims.

Differential diagnosis of vaginismus from dyspareunia

According to the DSM-IV-TR, there are two mutually exclusive sexual pain disorders: vaginismus and dyspareunia. Dyspareunia is defined as 'recurrent genital pain associated with sexual intercourse' [1]. PVD is reported to be the most frequent subtype of dyspareunia in premenopausal women with a prevalence of 7% in the general population [45,46]. Women with PVD typically experience a severe, sharp, burning pain upon vestibular touch or attempted vaginal entry [45,47,48]. It is diagnosed through the cotton-swab test, which consists of the application of a cotton swab to various areas of the vulvar vestibule and surrounding tissue [47].

Despite the fact that vaginismus and dyspareunia associated with PVD have been portrayed as two distinct clinical entities, they have many overlapping characteristics, such as the elevated vulvar pain and vaginal/pelvic muscle tone [18,42]. In fact, a number of studies have demonstrated that a large percentage (range between 42 and 100%) of women with vaginismus also meet the criteria for PVD [18,24,41,42]. This may explain, in part, why health practitioners (i.e., gynecologists, physical therapists and psychologists) show significant difficulties reliably differentiating vaginismus from PVD [18]. It should be noted, however, that PVD is characterized superficial dyspareunia. The pain of deeper dyspareunia is usually easily differentiable from that associated with vaginismus. Women with vaginismus, however, were found to display significantly higher levels of emotional distress while undergoing a gynecological examination and to avoid significantly more sexual and nonsexual vaginal penetration attempts as compared with women with PVD [18,37,42].

Fear

Clinical reports have long suggested that fear plays an important role in vaginismus [3,16,47–50]. Only a few studies have investigated this further [50–53]. For example, fear of pain was the primary reason reported by women with vaginismus for their abstinence as well as the core motive underlying their avoidance of sexual intercourse [18,53]. Moreover, a large percentage (range between 74 and 88%) of women with vaginismus report significant fear of pain during coitus [50,53]. Women suffering from vaginismus share a number of characteristics with individuals suffering from a 'specific phobia'. Specific phobias are defined as 'marked and persistent fear that is excessive or unreasonable, cued by the presence or anticipation of a specific object or situation' [1]. Individuals with a specific phobia will experience feelings of anxiety, fear or panic upon encountering the feared object or situation. As a result, they will tend to actively avoid direct contact with the phobic stimulus [1]. Women with vaginismus report fear of vaginal penetration and associated pain and display high levels of emotional distress during vaginal penetration situations, such as during gynecological examinations [18,50]. Women with vaginismus also tend to avoid situations involving vaginal penetration (i.e., gynecological examination, tampon insertion and sexual intercourse) [18].

It still remains unknown, however, whether vaginismic women avoid these particular situations in order to diminish their anxiety level similar to individuals suffering from a specific phobia, or in response to their pain experience, or both. Nonetheless, the avoidance of vaginal penetration cannot be solely explained by the experience of pain since women with dyspareunia, who also experience severe pain during vaginal penetration, have not been shown to avoid vaginal penetration situations as much as women suffering from vaginismus [18,42].

Although fear appears to be a promising factor that characterizes women with vaginismus, the existing empirical studies lack appropriate control groups, standardized instruments to measure fear and appropriate statistical analysis [50–53].

Summary

The current definition of vaginismus is problematic. First, the vaginal muscle spasm criterion has never been empirically validated and it appears that vulvar pain and the fear of pain or of vaginal penetration characterizes most women currently diagnosed with vaginismus. Moreover, vaginismus cannot be reliably differentiated from superficial dyspareunia. A recent consensus definition reflects these conclusions and defines vaginismus as: 'persistent or recurrent difficulties of the woman to allow vaginal entry of a penis, finger and/or any object, despite her expressed wish to do so. There is variable (phobic) avoidance, involuntary pelvic muscle contraction and anticipation/fear/experience of pain. Structural or other physical abnormalities must be ruled out or addressed' [54]. Binik has also recently proposed a new conceptualization that combines vaginismus and dyspareunia into a single genito–pelvic pain/penetration disorder characterized by persistent or recurrent difficulties for 6 months or more with at least one of the following [32]:

- Inability to have vaginal intercourse/penetration on at least 50% of attempts;
- Marked genito–pelvic pain during at least 50% of vaginal intercourse/penetration attempts;
- Marked fear of vaginal intercourse/penetration or of genito–pelvic pain during intercourse/penetration on at least 50% of vaginal intercourse/penetration attempts;
- Marked tensing or tightening of the pelvic floor muscles during attempted vaginal intercourse/penetration on at least 50% of occasions.

Etiological factors

Psychological factors

Although the definition, diagnosis and treatment of vaginismus have focused largely on the organic symptom of vaginal muscle spasm, the proposed etiological factors have primarily been psychogenic. The most frequently proposed include negative sexual attitudes, psychological and/or physical trauma, and relationship difficulties.

Negative sexual attitudes & lack of sexual education

The associations between negative sexual attitudes, sexual ignorance and vaginismus have been frequently mentioned in the vaginismus literature [1,51,55]. For example, Ellison claimed that vaginismus primarily resulted from: a lack of sexual knowledge and the presence of sexual guilt both leading to a fear of engaging in intercourse [56,57]. These are consistent with Silverstein, Ward *et al.* and Basson's conclusion that women suffering from vaginismus hold negative views about sexuality in general and about sex before marriage [41,51,53]. However, all these studies suffer from a number of important methodological limitations such as small sample sizes (n = 22–89), lack of appropriate statistical analyses and control groups, as well as absence of standardized measurement instruments and a standardized protocol to diagnose vaginismus [41,51,53,56,57]. There are only two etiological studies of vaginismus that have included a standard statistical analysis or a control group [58,59] and only one that used a standardized measurement instrument [60]; their results do not support the notions that women with vaginismus hold negative sexual attitudes and/or have lower levels of sexual knowledge and education.

Relationship factors

Vaginismus has frequently been reported to result from a dysfunctional couple relationship [60,61]. The available empirical evidence is controversial. For example, Tugrul and Kabakçi's (n = 40) uncontrolled study demonstrated that 85% of vaginismic women who applied for the treatment of vaginismus and 90% of their husbands evaluated their marriages as satisfactory [50]. Hawton and Catalan (n = 30) found that couples suffering from vaginismus have a significantly better relationship and communication when compared with 76 couples presenting other types of female sexual dysfunctions [62]. Although relationship factors have not been empirically demonstrated to play a significant role in the etiology of vaginismus, women who suffer from vaginismus do have fewer sexual relations and avoid more sexual contact when compared with healthy controls [50,59]. It remains unclear, however, whether these are causes or consequences of vaginismus.

Partners of women with vaginismus have been reported in clinical reports to suffer from sexual dysfunction as well as to display passive and unassertive personalities [3,26,51,57,63–65].

Controlled empirical findings using standardized instruments evaluating type of personalities and male sexual dysfunction, however, have not supported this view [26,52,58]. For example, when the personality characteristics of male partners of women with vaginismus are compared with controls or norms, no differences were demonstrated. Moreover, the few studies that investigated the chronology of sexual dysfunction in partners of women with vaginismus concluded that sexual dysfunction such as erectile and premature ejaculation are generally the result rather than the cause of vaginismus [30,64,66,67].

Sexual &/or physical abuse

Although the experience of sexual and/or physical abuse is generally considered an important etiological factor in vaginismus, the empirical evidence is less conclusive [1,60,68]. Five out of six studies [62,65,66,69,70] found no evidence of a higher prevalence of sexual and physical abuse. The sixth study found only weak evidence since women with vaginismus were twice as likely to report a history of childhood sexual interference (attempts at sexual abuse and sexual abuse involving touching) as compared with a 'no pain' group [59]. Larger studies with matched control groups and well-validated definitions of abuse are required to resolve this issue.

Biological factors

Organic pathology

A number of organic pathologies (e.g., hymeneal and congenital abnormalities, infections, vestibulodynia, trauma associated with genital surgery or radiotherapy, vaginal atrophy, pelvic congestion, endometriosis, vaginal lesions and tumors, scars in the vagina from injury, childbirth or surgery, and irritation caused by douches, spermicides or latex in condoms) resulting in painful/difficult/impossible vaginal penetration have been suggested as etiological factors [5,8,16,68,71]. There have been no controlled studies evaluating this possibility.

Pelvic floor dysfunction

Pelvic floor muscle dysfunction (e.g., hypertonicity and reduced muscle control) has been suggested as a predisposing factor in the development of vaginismus [39,45]. Barnes *et al.*'s uncontrolled study (n = 5) argued that vaginismic women had difficulty evaluating vaginal muscle tone and as a result experienced problems distinguishing between a relaxed state and a spasm [72]. It remains unclear, however, whether pelvic floor dysfunction is a predisposing factor or the defining symptom. To date, no controlled longitudinal studies have investigated the role of pelvic floor muscle dysfunction in the etiology of vaginismus.

Summary

Although a long list of psychological factors have been proposed as playing a role in the etiology of vaginismus, very few have been supported by empirical research. In addition, no biological factors hypothesized to be involved in the development of vaginismus have been adequately investigated.

Treatment

There has been much controversy over the treatment of choice for vaginismus. Sims recommended a surgical intervention that consisted of the removal of the hymen, the incision of the vaginal orifice and subsequent dilatation [2]. Soon thereafter, the need for a surgical procedure was questioned given that dilatation alone appeared to result in favorable outcomes [5,73,74]. Walthard, who conceptualized vaginismus as a phobic reaction to an excessive fear of pain, was one of the first to recommend psychotherapy [75]. Throughout the early 20th century, psychoanalysis was often prescribed following the notion that vaginismus was a hysterical or conversion symptom [76,77]. In the 1970s, Masters and Johnson greatly influenced the treatment of sexual dysfunction, in general, and reported that vaginismus could be easily treated with behaviorally oriented sex therapy, which included vaginal dilatation [2]. The success rates for the various treatments, ranging from vaginal dilatation to psychoanalysis to behaviorally oriented sex therapy were always reported to be excellent. Current treatments for vaginismus can be divided into four main categories: pelvic floor physiotherapy, pharmacological treatments, general psychotherapy and sex/cognitive behavioral therapy. Table 1 summarizes the treatment outcome studies of vaginismus.

Pelvic floor physiotherapy

The rationale for the use of pelvic floor physiotherapy in the treatment of vaginismus is that it will aid in developing awareness and control of the vaginal musculature as well as restore function, improve mobility, relieve pain and overcome vaginal penetration anxiety [39,72,78]. Physical therapists use a variety of techniques to achieve these goals, such as breathing and relaxation, local tissue desensitization, vaginal

Table 1. Review of treatment outcome studies for vaginismus.

Author (year)	Study type	Diagnostic method	Sample (n)	Treatment (n)	Definition of success	Drop-out rate	Result	Follow-up	Ref.
Pelvic floor physiotherapy									
Barnes *et al.* (1984)	Uncontrolled clinical study	Pelvic exam Self report	5	Biofeedback, psychotherapy and dilators	Intercourse	Not reported	100% success	6-month FU: success <60%	[72]
Seo *et al.* (2005)	Uncontrolled clinical study	Pelvic exam Self report	12	Functional electrical stimulation-biofeedback and CBT	'Satisfactory intercourse'	Not reported	100% success	Not reported	[79]
Pharmacological treatment									
Hassel (1997)	Case study	Not stated	1	5% lignocaine gel	Ability to undergo a pelvic exam Intercourse	NA	Success	Not reported	[80]
Peleg *et al.* (2001)	Case study	Pelvic exam	1	Nitroglycerin ointment	'Satisfactory intercourse'	NA	Success	12.5-month FU: success maintained	[81]
Mikhail (1976)	Uncontrolled clinical study	Referral with diagnosis	4	iv. diazepam and marital and psychotherapy	Disappearance of symptoms	Not reported	100% success	2- to 6-month FU: success maintained (ongoing psychotherapy)	[82]
Plaut *et al.* (1997)	Case study	Referral by gynecologist	1	Anxiolytic medication and psychotherapy	Intercourse	NA	Success	FU (time unspecified): success maintained	[83]
Brin *et al.* (1997)	Case study	Pelvic exam	1	Botulinum toxin injections	Intercourse	NA	Success	2-year FU: success maintained	[84]
Ghazizadeh *et al.* (2004)	Uncontrolled clinical study	Referral with diagnosis	24	Botulinum toxin injections	Painless pelvic exam 'Satisfactory intercourse'	Not reported	96% no symptoms during pelvic exam 75% satisfactory intercourse; 17% mild pain	2- to 24-month FU: success maintained	[85]
Shafik *et al.* (2000)	Uncontrolled clinical study	Not reported	13	Botulinum toxin (8) Saline (5)	'Satisfactory intercourse'	Not reported	100% success No improvement	8- to 14-month FU: success maintained	[86]
Bertolasi *et al.* (2009)	Uncontrolled clinical study	EMG recordings	39	Botulinum toxin type A injections	Intercourse EMG Psychometrics	15.4%	63.2% success	Not reported	[87]
Psychological treatments: general psychotherapy									
Barnes (1986)	Uncontrolled clinical study	Pelvic exam Self-report	55	Brief psychotherapy, education and dilators (50) Biofeedback (5)	Intercourse	Not reported	84% success	6-month (four couples lost at FU), three couples no longer having intercourse	[66]

CBT: Cognitive behavioral therapy; EMG: Electromyography; FU: Follow-up; iv.: Intravenous; NA: Not applicable; VVS: Vulvar vestibulitis syndrome.

Table 1. Review of treatment outcome studies for vaginismus (cont.).

Author (year)	Study type	Diagnostic method	Sample (n)	Treatment (n)	Definition of success	Drop-out rate	Result	Follow-up	Ref.
Psychological treatments: general psychotherapy (cont.)									
Kennedy *et al.* (1995)	Uncontrolled clinical study	Pelvic exam Self-report No local pathology	18	Individual psychotherapy for both partners, education, *in vivo* desensitization and retraining of sexual behavior	Intercourse	Not reported	78% success	Not reported	[52]
Elkins *et al.* (1986)	Case study	Unconsummated marriage	1	Interactional therapy	Intercourse	NA	Success	12- and 15-month FU: children born, but no information on sexual function	[88]
Gottesfeld (1978)	Case study	Unconsummated marriage Self-report	1	3 years of psychotherapy with hypnosis	Intercourse with orgasm	NA	Success	2-year FU: success maintained	[89]
Harman *et al.* (1994)	Case study	Diagnosed by physician	1	Relationship enhancement therapy and sexual education	'Improvement of relationship'	NA	Success	6-week FU: success maintained	[90]
Kleinplatz (1998)	Case study	Referral by gynecologist	1	Psychotherapy (existential–experiential)	Disappearance of vaginismus 'Sexual well-being'	NA	Success	6-month FU: success maintained	[91]
Pridal *et al.* (1993)	Case study	Referral	1	Brief psychotherapy, relaxation, Kegels and dilators	'Satisfactory intercourse'	NA	Success	3-month FU: success maintained	[93]
Ben-Zion *et al.* (2007)	Controlled clinical trial	DSM-IV criteria	32	Couple therapy and other treatments (16) Surrogate therapy and other treatments (16)	Intercourse	Not reported	100% success 75% success	Not reported	[94]
Delmonte (1988)	Case study	Referral	1	Psychotherapy, marital therapy, relaxation–hypnosis	Painless intercourse	NA	Success	6-month FU: success maintained	[95]
Sex/cognitive-behavioral therapy									
Hawton *et al.* (1990)	Controlled clinical trial	Referral	Vaginismus (30) Other female sexual dysfunctions (76)	Sex therapy and Kegels	Vaginismus resolved or largely resolved	10% in vaginismus group	80% success 51% success	3-month FU: success rate <76 (67%)	[62]

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Table 1. Review of treatment outcome studies for vaginismus (cont.).

Author (year)	Study type	Diagnostic method	Sample (n)	Treatment (n)	Definition of success	Drop-out rate	Result	Follow-up	Ref.
Sex/cognitive-behavioral therapy (cont.)									
Van Lankveld *et al.* (2006)	Randomized controlled treatment outcome study	Pelvic exam Self-report	117	CBT group therapy (43)	Intercourse	21%	9% success	3-month and 1-year FU: success rate >21%	[70]
				CBT bibliotherapy (38)			18% success	3-month and 1-year FU: success rate <15%	
				Wait-list control (36)			No success		
Chakrabarti *et al.* (2002)	Case study	Self-report	1	Sex education and psychotherapy	Intercourse	NA	Success	Maintained (time unspecified)	[96]
Grillo *et al.* (1980)	Uncontrolled clinical study	Pelvic exam Painful hymenal rings/rigid remnants	17 Dyspareunia with comorbid vaginismus	Surgical removal of hymenal remnants and sex therapy and dilators and Kegel	Intercourse with orgasm Painless pelvic exam	Not reported	100% success	Not reported	[97]
Jeng *et al.* (2006)	Retrospective study	Pelvic exam Self-report	120	Sex therapy and xylocaine and oral analgesics and relaxation and dilation	Intercourse	Not reported	93% success	3-month and 1-year FU: 83% intercourse with orgasm	[98]
O'Sullivan *et al.* (1978)	Uncontrolled clinical study	Pelvic exam	46	Short-term sex therapy and dilators	'Normal sexual function'	48%	52% success	Not reported	[99]
Oystragh (1988)	Case study	Unconsummated marriage	1	Sex therapy and hypnosis and dilators	Painless intercourse	NA	Success	Maintained (time unspecified)	[100]
Ng (1993)	Case study	Unconsummated marriage	1	Mien-Ling dilators	'Pleasurable intercourse'	NA	Success	2-month FU: success maintained	[101]
Fuchs (1980)	Uncontrolled clinical study	Not stated	71	Systematic desensitization *in vitro* (18)	Intercourse	(*in vivo* 2% group)	89% success	2- to 5-year FU for 65 patients: 'normal sexual adjustment' maintained	[102]
				Systematic desensitization *in vivo* (54)			98% success		
Wijma *et al.* (1997)	Case study	Self-report		CBT following *in vivo* systematic desensitization and phobia counter-conditioning	Intercourse free of pain/fear No recurrence of vaginismus	NA	Success	6-month and 1.5-year FU: success maintained	[103]
Wijma *et al.* (2000)	Case study	VVS diagnosis and penetration not possible	1	Systematic desensitization	Disappearance of burning pain	NA	Success	Not reported	[104]

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Table 1. Review of treatment outcome studies for vaginismus (cont.).

Author (year)	Study type	Diagnostic method	Sample (n)	Treatment (n)	Definition of success	Drop-out rate	Result	Follow-up	Ref.
Sex/cognitive-behavioral therapy (cont.)									
Schnyder *et al.* (1998)	Randomized clinical study	DSM-III-R criteria	44	*In vivo* dilation and relaxation and education (25) *In vitro* dilation (23)	Painless intercourse Increased sexual desire and orgasm capacity	5%	98% success 50% still having intercourse pain	6- to 22-month FU: (8 lost) 50% disappearance, 47.7% improvement	[105]
Biswas *et al.* (1995)	Uncontrolled clinical study	Not stated	19	Rapid desensitization under anesthesia	'Satisfying intercourse'	Not reported	100% success	Not reported	[106]
Scholl (1998)	Uncontrolled clinical study	Pelvic exam Self-report	23	Sex therapy and dilators and Kegel exercises	Intercourse	13%	87% success	1- to 4-year FU: 95% continue intercourse	[107]
Ter Kuile *et al.* (2009)	Replicated single-case design	Pelvic exam Self-report	10	Exposure therapy	Intercourse Psychometrics	0%	90% success and significantly less fear	3-month and 1-year FU: success maintained	[108]

CBT: Cognitive behavioral therapy; EMG: Electromyography; FU: Follow-up; iv.: Intravenous; NA: Not applicable; VVS: Vulvar vestibulitis syndrome.

dilators, pelvic floor biofeedback and manual therapy techniques [39,72,78]. To date, there are two studies with 100% success rates that have investigated the efficacy of biofeedback in the treatment of vaginismus [72,79]. Unfortunately, they have very small sample sizes (<12) and lack appropriate control groups [72,79]. In addition, one study had only 6-month follow-up with the success rate dropping to 60% [66,72]. Considering the importance accorded to the vaginal muscle spasm component in vaginismus, it is surprising that pelvic floor physiotherapy has not been investigated more extensively.

Pharmacological treatment

Three main types of pharmacological treatment have been proposed for vaginismus: local anesthetics (e.g., lidocaine), muscle relaxants (e.g., nitroglycerin ointment and botulinum toxin) and anxiolytic medication [80–87]. Local anesthetics, such as lidocaine gel, have been proposed based on the rationale that vaginismic muscle spasms are due to repeated pain experienced with vaginal penetration and, hence, the use of a topical anesthetic aimed at reducing the pain is hypothesized to resolve the spasm [80]. Its efficacy has only been reported in a case study in which a 5% lidocaine gel was applied on the hyperesthetic areas of the vaginal introitus of a 17-year-old women suffering from primary vaginismus. A topical nitroglycerin ointment, hypothesized to treat the muscle spasm by relaxing the vaginal muscles, was also discussed only in a case study [81]. A Muslim Bedouin couple presenting with primary vaginismus were able to engage in a satisfactory sexual relationship following the application of a topical nitroglycerine ointment [81]. Given that all the available information is in the form of case studies, no firm conclusion can be reached.

Botulinum toxin, a temporary muscle paralytic, has been recommended in the treatment of vaginismus with the aim of decreasing the hypertonicity of the pelvic floor muscles [84]. In Shafik and El-Sibai's treatment study (n = 13), women with vaginismus who received an injection of botulinum toxin were able to engage in 'satisfactory intercourse' as compared with no improvement in a control group receiving saline injections [86]. The successful outcome persisted for an average follow-up of 10.2 months. Nonetheless, there are a number of limitations to this promising study, such as the small sample size, lack of information on how vaginismus was diagnosed and lack of independent determination of treatment outcome. A recent treatment outcome study (n = 39) demonstrated that women with vaginismus secondary to PVD, who received repeated injections of botulinum neurotoxin type A into the levator ani, displayed improvements on standardized measurements of sexual activity (i.e., the Female Sexual Functioning Index), on possibility of having sexual intercourse, on levator ani EMG hyperactivity and on bowel–bladder symptoms [87]. After a 39 month follow-up, 63.2% of their participants had completely recovered from vaginismus and PVD, 15.4% still needed some injections, 15.4% had dropped out and the remaining had not completed the treatment protocol. Another pharmacological treatment that has been proposed is the use of anxiolytics, such as diazepam, in conjunction with psychotherapy based on the hypothesis that vaginismus is a psychosomatic condition resulting from past trauma and, thus, anxiety-reducing medication will resolve the symptoms. Mikhail's uncontrolled study found that the administration of intravenous diazepam during psychological interviews in four women with vaginismus resulted in successful intercourse [82]. Unfortunately, conclusions concerning the pharmacological treatment of vaginismus are limited because most studies lack appropriate placebo control groups and do not randomly assign patients to treatment, are based on small samples or do not use standardized outcome instruments.

General psychotherapy

A variety of psychological treatments for vaginismus have been investigated, including marital, interactional, existential–experiential, relationship enhancement and hypnosis [52,88–95]. The psychological treatments are often based on the notion that vaginismus results from marital problems, negative sexual experiences in childhood or a lack of sexual education. The therapy can be conducted in an individual or couple format. Generally, in individual therapy, the treatment is to identify and resolve underlying psychological problems that could be causing the disorder. In couples therapy, vaginismus is conceptualized as a problem for the couple and the treatment tends to focus on the couple's sexual history and any other problems that may be occurring in the relationship. Although the reported success rates are high (78–100%), all except two are case studies with poorly designed and described treatment interventions as well as a lack of information on how vaginismus was diagnosed. The two reports that are not case studies lack appropriate control groups and have no follow-up data [52,94]

Sex/cognitive behavioral therapy

In the 1970s, Masters and Johnson reported that vaginismus could be easily treated with behaviorally oriented sex therapy that included vaginal dilatation [3]. The first step of their treatment consists of the physical demonstration of the vaginal muscle spasm to the patient (and her partner) during a gynecological examination. The couple is then instructed to insert a series of dilators of graduated sizes at home guided by both the patient and her partner with the aim of desensitizing the patient to vaginal penetration. Masters and Johnson's treatment regimen also emphasized the importance of education regarding sexual function and the development and maintenance of vaginismus in order to relieve the psychological impact of the condition. As a result of the influence of Masters and Johnson, several studies were conducted on the efficacy of sex therapy in the treatment of vaginismus with excellent success rates reported resulting in continued utilization of this treatment for vaginismus [62,96–107]. These studies were, however, uncontrolled [62,97–99,102,106,108] or case studies [96,100,101,103,104] and all presented important methodological flaws, such as the lack of a waiting list control group and of standardized measurements to evaluate treatment outcome as well as elevated or unreported drop-out rates.

The first ever randomized controlled therapy outcome study for vaginismus was recently published. This study investigated a cognitive-behavioral sex therapy for the treatment of vaginismus [70]. The treatment included the sexual education and vaginal dilatation technique as in Masters and Johnson's treatment protocol. It was also comprised of cognitive therapy, relaxation and sensate focus exercises. Participants received the treatment for 3 months either in group therapy or in bibliotherapy format. At post-treatment, 18% (14% group therapy; 9% bibliotherapy) of participants in the treatment group reported successful attempted penile–vaginal intercourse while none of the women in the waiting list control group reported having had successful intercourse. Interestingly, there was no significant difference in efficacy between the group therapy and bibliotherapy treatment format. At 3 month and 1-year follow-ups, 19% of the participants in the cognitive behavioral sex therapy group and 18% in the bibliotherapy group had achieved intercourse.

Although the rate of successful outcome was far below what was expected based on previous nonrandomized controlled treatment outcome studies, internal analyses of the data suggested that successful outcome was mediated by changes in fear of coitus and avoidance behavior. Van Lankveld's group reformulated their conceptualization of vaginismus from a sexual disorder to a vaginal penetration phobia [70,108]. A recent study carried out by the same group investigated a treatment for vaginismus focusing more explicitly and systematically on the fear of coitus through the use of prolonged, therapist-aided exposure therapy [108]. The treatment was comprised of education on the fear and avoidance model of vaginal penetration as well as of a maximum of three 2 h sessions of *in vivo* exposure to the stimuli feared during vaginal penetration. A replicated (n = 10) randomized single-case A–B phase design was used. The results showed that nine out of ten participants were able to engage in intercourse following treatment and these findings persisted at a 1-year follow-up. In addition, the exposure treatment was successful in decreasing fear and negative penetration beliefs.

Evaluation of treatment research

Vaginismus has traditionally been considered as an easily treatable sexual dysfunction. The elevated success rates, reported in the literature must, however, be considered in light of uncontrolled designs, small sample sizes, elevated or unreported drop-out rates, which are not evaluated with intent-to-treat statistics, as well as a lack of long-term follow-up data. In fact, the only randomized controlled treatment trial does not support the notion that vaginismus is an easily treatable condition [70].

A basic issue in treatment evaluation is how a successful treatment outcome is defined. The great majority of studies have defined success as the ability to achieve vaginal penetration through sexual intercourse. While successful penetration is clearly a crucial first step, if it is not accompanied by pleasurable feelings, then treatment success is questionable. For instance, Schnyder *et al.* found that although 98% of the women in their sample were able to have intercourse by the end of treatment with vaginal dilators, 50% were still experiencing pain during penetration [105]. Similarly, although nine out of ten participants in the Ter Kuile *et al.* fear reduction study were able to experience penetration, none of the measures of sexual enjoyment or pleasure significantly improved. While it appears that high success rates in vaginal penetration may soon be achievable, the therapeutic challenge of increasing vaginismic women's pleasure has not even been approximated [108].

Conclusion

Although most research concerning vaginismus presents significant methodological limitations, certain conclusions can be made from the few well-controlled studies. First, vaginal muscle spasm is not a valid or reliable diagnostic criterion for vaginismus. Second, vulvar pain is an important characteristic of most women suffering from vaginismus and should be always evaluated. Third, although vaginismus and dyspareunia are presently considered two mutually exclusive disorders, they share many characteristics and are very difficult to differentiate using our current clinical tools. Fourth, fear and avoidance of vaginal penetration situations have been mentioned to be an integral part of vaginismus; interestingly, there are no controlled published studies examining its role. Finally, the present conceptualization of vaginismus as an easily treatable sexual dysfunction has not been supported by empirical research. Unfortunately, it is very difficult to conduct research when inherent problems exist with the definition of vaginismus.

Future perspective

Unlike the current DSM-IV-TR definition of vaginismus, Binik's new conceptualization of vaginismus as a genito–pelvic pain/penetration disorder takes into consideration existing empirical findings as it incorporates pain, muscle tension and fear. Binik's diagnostic criteria are easily translatable into dimensional terms and do not categorically separate vaginismus from provoked vestibulodynia. This new conceptualization also has significant diagnostic and therapeutic implications in that it suggests that a multidisciplinary approach taking

Executive summary

- Vaginismus continues to be perceived by clinicians as a well-understood and easily treatable female sexual dysfunction despite the lack of research supporting these claims.

Prevalence

- Although the population prevalence of vaginismus remains unknown, it has been reported to range between 5 and 17% in clinical settings.

Classification & diagnosis

- There has been a 150-year consensus concerning the definition of vaginismus as an involuntary vaginal muscle spasm despite the lack of research supporting the vaginal muscle spasm criterion.
- Women with vaginismus may demonstrate high pelvic floor muscle tension and/or experience genital pain and/or report fearing vaginal penetration or pain.
- Vaginismus and dyspareunia are currently considered two mutually exclusive disorders despite empirical findings demonstrating that health practitioners have a great difficulty reliably differentiating both conditions.
- Recently, new definitions of vaginismus integrating pelvic floor muscle tension, genital pain and fear have been proposed.

Etiology

- Most psychological factors that have been proposed to play a role in the etiology of vaginismus (i.e., abuse, relationship factors, negative sexual attitudes and lack of sexual education) have not received empirical support.
- Although organic pathologies and pelvic floor dysfunction have often been implicated in the development of vaginismus, they have not been empirically investigated.

Treatment

- Current treatment options for vaginismus include pelvic floor physiotherapy, pharmacological treatments, general psychotherapy and sex/cognitive behavioral therapy.
- The success rates for the various treatments have generally been reported to be excellent despite the lack of randomized controlled treatment outcome studies validating this claim.
- To date the only randomized controlled treatment outcome study that investigated the efficacy of cognitive behavioral sex therapy for vaginismus does not support the notion that vaginismus is an easily treatable condition.
- A recent exposure treatment focusing more extensively on the fear component of vaginismus has demonstrated promising results.

Future perspective

- A new conceptualization of vaginismus as a 'genito–pelvic pain/penetration disorder', characterized by the inability to have vaginal intercourse/penetration, genito–pelvic pain, fear of vaginal intercourse/penetration, and tension of the pelvic floor muscles, has recently been proposed.
- A multidisciplinary diagnostic and adequate treatment approach for vaginismus addressing fear, genital pain, pelvic floor muscle tension and sexual pleasure is recommended. This set of skills is not easily accomplished by individual practitioners and should probably be addressed by a multidisciplinary team.

into account muscle tension, genital pain and fear will be necessary to attain a high success rate. It is unlikely that a lone professional will be able to provide such a treatment. A multidisciplinary team, including a gynecologist, physical therapist and psychologist/sex therapist, should be involved in the assessment and treatment of vaginismus to address its different dimensions.

Financial & competing interests disclosure

The authors have no relevant affiliations or financial involvement with any organization or entity with a financial interest in or financial conflict with the subject matter or materials discussed in the manuscript. This includes employment, consultancies, honoraria, stock ownership or options, expert testimony, grants or patents received or pending, or royalties. No writing assistance was utilized in the production of this manuscript.

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