



Sexual and Relationship Therapy

Publication details, including instructions for authors and subscription information:

<http://www.tandfonline.com/loi/csmt20>

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Sophie Bergeron ^a, Mélanie Morin ^{a b} & Marie-Josée Lord ^c

^a Department of Psychology, Université de Montréal, Montréal, Canada

^b Université de Sherbrooke, Montréal, Canada

^c Kinatex West Island, Kirkland, Canada

Published online: 03 Aug 2010.

To cite this article: Sophie Bergeron, Mélanie Morin & Marie-Josée Lord (2010) Integrating pelvic floor rehabilitation and cognitive-behavioural therapy for sexual pain: what have we learned and where do we go from here?, *Sexual and Relationship Therapy*, 25:3, 289-298, DOI: [10.1080/14681994.2010.486398](http://dx.doi.org/10.1080/14681994.2010.486398)

To link to this article: <http://dx.doi.org/10.1080/14681994.2010.486398>

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Integrating pelvic floor rehabilitation and cognitive-behavioural therapy for sexual pain: what have we learned and where do we go from here?

Sophie Bergeron^{a*}, Mélanie Morin^{a,b} and Marie-Josée Lord^c

^a*Department of Psychology, Université de Montréal, Montréal, Canada;* ^b*Université de Sherbrooke, Montréal, Canada;* ^c*Kinatex West Island, Kirkland, Canada*

(Received 1 April 2010; final version received 13 April 2010)

The sexual pain disorders dyspareunia and vaginismus are highly prevalent yet misunderstood women's sexual health problems. We have proposed the adoption of a treatment approach integrating pelvic floor rehabilitation and cognitive-behavioral therapy in order to target the multidimensional aspects of these complex conditions. Looking back on the work that has been published in the area of sexual pain since we introduced this model in 2003, the present paper focuses on the progress that has been achieved since then, with an emphasis on the pelvic floor musculature and psychological factors. Specifically, the continuing debate about the classification of sexual pain is briefly summarized. Findings from treatment outcome research are reported. Growing evidence indicates that pelvic floor rehabilitation and cognitive-behavioral therapy lead to significant improvements in pain and sexual functioning, although there are still only a handful of published randomized controlled trials and only one study focusing on the integration of these two modalities. Recent advances concerning the role of the pelvic floor as well as cognitive and affective variables in the etiology of sexual pain are reviewed, with results showing that higher levels of anxiety, fear of pain, hypervigilance and catastrophizing, in addition to lower levels of self-efficacy, may contribute to the exacerbation of pain and associated sexual dysfunction. In terms of avenues for future research, two new measurement instruments for assessing the pelvic floor musculature are described, namely the dynamometric speculum and transperineal ultrasound. Ongoing challenges involved in the adoption of an integrated treatment approach are discussed.

Keywords: sexual pain; dyspareunia; vaginismus; physical therapy; cognitive-behavioral therapy; provoked vestibulodynia

Introduction

There have been a number of notable changes in the area of sexual pain since the publication of our Leading Comment in 2003 (Bergeron & Lord, 2003). First and foremost, the necessity of approaching treatment from a multidimensional, multidisciplinary perspective has now been espoused by most experts in the field, as exemplified by the recent recommendations of the Third International Consultation on Sexual Medicine relating to women's sexual pain disorders (van Lankveld et al., 2010). Despite this positive shift, empirical evidence to support the therapeutic efficacy of such an approach still lags behind. This limitation

*Corresponding author. Email: sophie.bergeron.2@umontreal.ca

notwithstanding, novel and scientifically sound knowledge has been generated concerning the role of the pelvic floor in dyspareunia and vaginismus, in addition to that of cognitive and affective variables in the experience of pain and associated sexual difficulties. Stimulating avenues for future research are being sketched out, as are new measurement instruments that constitute unforeseen windows into the mechanisms underlying the development and maintenance of sexual pain. Nevertheless, there remain ongoing challenges to be faced by scientists and clinicians alike, hopefully working hand in hand. One of the most enduring ones has to do with issues of classification.

What is sexual pain? The continuing debate

Binik and colleagues suggested that there is nothing sexual about sexual pain – an umbrella term encompassing a wide array of heterogeneous conditions – and that it should in fact be classified as a pain disorder rather than as a sexual dysfunction (e.g. Binik, 2005). Gynecologists also claim that dyspareunia and vestibulodynia are end points of multiple disease states (e.g. Goldstein, 2009). Others vehemently disagree with this position, arguing that sexual pain is the only valid sexual dysfunction (e.g. Tiefer, 2005). The current state of affairs regarding the status of sexual pain in the upcoming DSM-V is as follows: dyspareunia and vaginismus would be collapsed into a single diagnostic entity called genito-pelvic pain/penetration disorder. Five dimensions would be the focus of assessment: percentage success of vaginal penetration; pain with vaginal penetration; fear of vaginal penetration or of genito-pelvic pain during vaginal penetration; pelvic floor muscle dysfunction; and medical co-morbidity (Binik, 2010). Although a critique of this proposal is beyond the scope of the present paper, it is of interest to note that there is now an emphasis on fear of pain and pelvic floor muscle dysfunction, rendering all the more relevant an intervention model that targets cognitive-affective factors and the pelvic floor simultaneously.

Are we more advanced concerning treatment outcome research?

We view with great enthusiasm the inclusion of a chapter focusing entirely on pelvic floor rehabilitation in a textbook devoted to sex therapy (Rosenbaum, 2007), as well as chapters on psychological and pelvic floor approaches in a mostly biomedical textbook (e.g. Bergeron, Landry, & Leclerc, 2009). These clearly represent more signs that professionals in the field are beginning to think in an integrative fashion. Nonetheless, we were able to find only one published study since 2003 evaluating a multimodal approach to the treatment of dyspareunia, which consisted of psychosexual counselling provided by a social worker and pelvic floor rehabilitation performed by a midwife (Backman, Widenbrant, Bohm-Starke, & Dahlof, 2008). Although it did not include standardized measures of pain and sexual function, results of this prospective research showed that 67% of 24 participants reported occasional or mild pain at six-month follow-up, while 63% reported a major improvement in their sexual functioning. These findings are encouraging and suggest the need for more empirical work aimed at validating an integrated approach to care.

In parallel, other methodologically sound studies assessing cognitive-behavioral therapy (CBT) have been conducted. Ter Kuile and Weijnen (2006) prospectively evaluated the efficacy of 12 two-hour sessions of group CBT with 76 vestibulodynia

patients. At post-treatment, the women reported significant reductions in pain during intercourse and vaginal muscle tension, in addition to significant improvements in sexual satisfaction.

In a recently completed study (Bergeron, 2008), 97 women with vestibulodynia were randomly assigned to a 13-week trial of twice daily corticosteroid applications or group CBT. Although both groups showed significant pain reduction at six-month follow-up, women in the CBT arm reported significantly less pain than those in the topical treatment arm. In addition, the CBT intervention yielded significantly more improvements in sexual functioning and treatment satisfaction, while also resulting in reduced pain catastrophizing.

Masheb, Kerns, Lozano, Minkin and Richman, (2009) conducted a randomized trial comparing 10-week individual CBT and supportive therapy in a mixed group of 50 women with vulvodynia. At a one-year follow-up, participants had significant decreases in pain, although those assigned to CBT had greater improvements in pain during gynaecological examination, better sexual functioning and greater treatment satisfaction.

In a randomized trial in which 117 women diagnosed with lifelong vaginismus were assigned to cognitive-behavioral sex therapy, cognitive-behavioral bibliotherapy or a wait-list control condition, only 21% reported successful intercourse at the one-year follow-up, highlighting the limits of this 'talk therapy' approach (van Lankveld et al., 2006). However, a recent case series has shown that intensive exposure therapy in women with vaginismus can lead to 90% of them being able to achieve intercourse after only three two-hour exposure sessions over the course of one week (ter Kuile et al., 2009). Participants also displayed significant decreases in fear and negative penetration beliefs. It will be important for future research to investigate the extent to which this novel approach leads to pain reduction and improved sexual functioning in women with vaginismus.

Many studies have been published since 2003 supporting the effectiveness of different modalities targeting pelvic floor muscle dysfunctions. Components such as education, biofeedback, manual techniques and guided insertion techniques, as well as electro-therapeutic methods may be included in physical therapy intervention. In an effort to better describe such modalities, case reports have been published (Downey & Frederick, 2006; Fisher, 2007; Holland, 2003). Moreover, manual techniques have been argued to be a key component in physical therapy treatment. Even if there is a lack of supporting evidence, stretching and myofascial techniques have been reported to facilitate muscle relaxation, normalise tone, improve blood circulation and mobility in the pelvi-perineal region, adjust postural imbalances, as well as increase the vaginal opening and desensitize the area (Fitzgerald & Kotarinos, 2003; Rosenbaum & Owens, 2008). Recently, in a prospective study involving 11 women with provoked vestibulodynia, Gentilcore-Saulnier, McLean, Goldfinger, Pukall and Chamberlain (2010) demonstrated that a physical therapy program allowed the normalisation of pelvic floor dysfunctions such as lower pelvic floor response to pain, lower pelvic floor tone, improved vaginal flexibility and improved pelvic floor relaxation capacity. A significant reduction in pain during vaginal palpation was shown, although the outcomes regarding pain during intercourse or sexual function were not reported.

As for biofeedback, Bergeron, Khalifé, Glazer, and Binik (2008) published a 2.5-year follow-up study of women suffering from provoked vestibulodynia who had been randomized to vestibulectomy, biofeedback or CBT. Results showed that the

treatment effects were maintained and the pain intensity was even further reduced at follow-up. In another randomized trial, Danielsson, Torstensson, Brodda-Jansen and Bohm-Starke (2006) compared biofeedback to lidocaine. A significant improvement in 12/18 (66%) of the participants was demonstrated at a 12-month follow-up. Lastly, the efficacy of self-insertion techniques/dilators was evaluated by Murina, Bernorio and Palmiotto (2008) in a prospective study including fifteen women. It was shown that the use of dilators potentiates the beneficial effects of the following treatments: transcutaneous electrical nerve stimulation, infiltration, biofeedback, amitriptyline or pregabalin.

Studies were also carried out to investigate the effects of various electrotherapeutic modalities for the treatment of dyspareunia. In a prospective study involving 29 participants, Nappi et al. (2003) used electrical stimulation and showed improvements in pelvic floor contractility and muscle relaxation, as well as a reduction of pain. Similarly, Fitzwater, Kuehl and Schrier (2003) performed a retrospective study including 66 women with chronic pelvic pain and levator ani spasm. A reduction of muscle tension and pain after electrical stimulation treatment was demonstrated. Finally, in a randomized controlled trial involving 40 women with vestibulodynia, Murina et al. (2008) found a reduction of pain and an improvement in sexual function following transcutaneous electrical nerve stimulation treatment.

There is still a dire need for studies aiming to identify predictors of treatment outcome for either CBT, pelvic floor rehabilitation or a combination of both. In our work on CBT, we have found that higher levels of pre-treatment pain predicted a poorer pain outcome at a 2.5-year follow-up (Bergeron et al., 2008) and that higher pre-treatment levels of pain catastrophizing and lower levels of self-efficacy were associated with greater pain intensity at a six-month follow-up (Desrochers, Bergeron, Khalifé, Dupuis, & Jodoin, 2010). It remains difficult to assess which patients will benefit the most from an integrated approach involving CBT and pelvic floor rehabilitation and whether there are some that may not benefit at all.

Are there new data about the etiology of sexual pain that can inform an integrated approach?

As of yet, no psychological factors linked to the development and persistence of sexual pain have been identified due to the absence of experimental and/or prospective designs of published papers. The majority of controlled studies focusing on the cognitive and affective correlates of sexual pain – those that are targeted in CBT – have appeared only in the last decade, with most involving samples of women with vestibulodynia. This research has shown that compared to no-pain controls, women with vestibulodynia report more anxiety (Granot & Lavee, 2005; Payne et al., 2005), a greater tendency to avoid threat (Granot, 2005; Lundqvist & Bergdahl, 2005), as well as more pain catastrophizing (Payne et al., 2007), hypervigilance toward pain (Payne, Binik, Amsel, & Khalife, 2005) and fear of pain (Payne et al., 2007). Furthermore, it has been shown that higher levels of hypervigilance, fear of pain and pain catastrophizing are significant predictors of increased pain intensity in women with dyspareunia, whereas greater anxiety and avoidance are associated with poorer sexual function (Desrochers, Bergeron, Khalifé, Dupuis, & Jodoin, 2009). Findings of this study also indicated that lower levels of self-efficacy were related to both worse pain intensity and sexual function (Desrochers et al., 2009). However,

because this area of research is still in its infancy, the majority of studies published since 2003 have been descriptive and cross-sectional, which prevents reaching any conclusion regarding the etiologic implication of the above variables. Importantly, such designs do not distinguish the degree to which pain intensity may drive patients to fear and avoid threat, whether fear and anxiety are factors that exacerbate pain or whether both phenomena are occurring. This highlights the necessity of pursuing the exploration of psychosocial factors via prospective designs geared toward establishing their contribution to the exacerbation and maintenance of pain and associated sexual impairment.

Many studies have suggested that pelvic floor muscle dysfunctions are associated with sexual pain (Glazer et al., 1998; Reissing et al., 2004, 2005; White, Jantos, & Glazer, 1997). It has been hypothesized that pelvic floor dysfunctions may precipitate the onset of dyspareunia namely by increasing mucosal sensitivity (Zolnoun et al., 2006) or by closing the vaginal hiatus and preventing penetration in the case of vaginismus (Basson et al., 2000). Another hypothesis is that vulvo-vaginal pain may trigger pelvic floor dysfunctions (Glazer et al., 1998; Reissing, Binik, Khalife, Cohen, & Amsel, 2004; Reissing, Brown, Lord, Binik, & Khalife, 2005). Although the exact mechanisms remain poorly understood, it is plausible that dyspareunia is associated with a vicious cycle that involves pain/inflammation and further muscle dysfunctions (Graven-Nielsen & Arendt-Nielsen, 2002; Svensson, Graven-Nielsen, & Arendt-Nielsen, 1998). Several studies have been carried out to better understand such dysfunctions.

Higher pelvic floor muscle tone assessed by palpation (Gentilcore-Saulnier et al., 2010; Reissing et al., 2004, 2005) as well as superior resting electromyographic activity (Frasson et al., 2009) was found in women suffering from dyspareunia. Gentilcore-Saulnier et al. (2010) showed that women with vestibulodynia had lower vaginal flexibility and capacity to relax the pelvic floor muscle compared to controls. However, Reissing et al. (2004) and Engman, Lindehammar and Wijma, (2004) found a non-significant difference in resting electromyography (EMG) between the two groups. Methodological pitfalls in the current pelvic floor assessment techniques may explain such controversies. In fact, many confounders may seriously bias the inter-subjects comparisons when assessing amplitude of electromyographic signal, at rest for instance, such that these results must be interpreted with caution. Recently, Morin, Bergeron, Khalifé, Binik and Ouellet (2010) developed the dynamometric speculum for measuring objectively and directly the pelvic floor resting forces. Using this new instrument, an increase in pelvic floor muscle passive properties was confirmed in women with vestibulodynia compared to age-matched controls.

A protective-like defence consisting of an over-contraction of the pelvic floor muscles during intercourse has also been associated with dyspareunia (Reissing et al., 2004, 2005). Shafik and El-Sibai (2002) demonstrated an increase in EMG resting activity in women with vaginismus during dilator insertion compared to asymptomatic women. Gentilcore-Saulnier et al. (2010) suggested that the reaction of the superficial muscles of the pelvic floor might be more significant than the deeper ones.

Women with dyspareunia also show impairments in the pelvic floor contractile capacities. Lower strength and endurance were found in women with dyspareunia compared to controls using palpation (Reissing et al., 2004, 2005), EMG (Glazer et al., 1998; Reissing et al., 2004) and dynamometry (Morin et al., 2010). Moreover, Morin et al. showed a reduction in the pelvic floor rapidity of contraction in addition

to a reduced capacity to contract and relax the pelvic floor quickly, reflecting poor muscle control.

Similarly to the issues raised concerning research about the cognitive and affective components of sexual pain, studies examining the pelvic floor musculature to date do not answer the question of whether sexual pain causes pelvic floor problems or pelvic floor problems lead to the development of sexual pain. Only prospective and experimental designs will allow researchers to clarify the contribution of pelvic floor variables to pain and sexual impairment in women with sexual pain.

Future avenues for research – innovative methodologies for assessing the pelvic floor musculature

The current measurement instruments for assessing pelvic floor muscle function have been criticized for their methodological flaws. Digital palpation is argued to be a subjective tool (Bo & Finckenhagen, 2001). Reissing et al. (2005) explained that digital assessment may be influenced by the reactions of the patient (i.e. fear, pain) during the assessment. Regarding EMG, confounding factors such as the contact between the electrodes and the mucosa, degree of vaginal lubrication and thickness of the vaginal tissue can all greatly affect signal detection (Merletti & Hermens, 2004). These factors may compromise comparison between subjects, especially when assessing the amplitude of the signal.

To overcome these limitations, the dynamometric speculum was developed to objectively assess pelvic floor muscle function. The reliability and the validity of this methodology have been confirmed (Dumoulin, Gravel, Bourbonnais, & Lemieux, 2007; Morin Gravel, Ouellet, Dumoulin, & Bourbonnais, 2006). After having reduced the size of the speculum branches, this measure was recently used in the assessment of women with provoked vestibulodynia (Morin et al., 2010), showing several muscle impairments (higher passive forces, lower strength, lower endurance, lower speed of contraction and poorer muscle control).

There is a growing interest in the use of ultrasound in rehabilitation for assessing muscle morphology and function (Teyhen, 2006). Transperineal ultrasound demonstrated good reliability for assessing pelvic floor muscle morphology at rest and during contraction (Braekken, Majida, Engh, & Bo, 2009). For example, the advent of 3D and 4D ultrasound allows imaging and measurement of the urogenital hiatus area at rest and during contraction. Therefore, the constricting role of the pelvic floor can be assessed. This methodology is highly promising for assessing women with sexual pain since introduction in the vaginal canal is not needed.

Ongoing challenges involved in the adoption of an integrated treatment approach

Unfortunately, clinics where physical therapists and psychologists work together side by side and have weekly rounds to discuss patients are still the exception rather than the rule. Communication between psychologists and physical therapists remains difficult due to this organizational issue. Despite its high prevalence in women of childbearing age, health professionals trained in the treatment of sexual pain are a rarity in some large cities in Western countries, let alone in rural areas and in other parts of the world. Of those who are qualified, some know little about what the other health professionals are doing and are estranged from their colleagues' perspective,

which may discourage patients and affect their compliance with treatment. Lastly, as opposed to the quick fix offered by a surgical procedure, women with sexual pain find it cumbersome at times to practice the home exercises that are part and parcel of an approach integrating pelvic floor rehabilitation and cognitive-behavioral therapy and may become discouraged. We must be creative in our ways of explaining the rationale underlying our interventions and challenge women's assumptions about their pain being entirely physical or psychological. Thinking multidimensionally requires openness, flexibility and humility on the part of both patients and health professionals, yet this integrative, collaborative approach is beginning to bear fruit, to the benefit of women afflicted with sexual pain.

Acknowledgements

This work was supported by Canadian Institutes of Health Research (CIHR) and Social Sciences and Humanities Research Council grants awarded to Sophie Bergeron and by a CIHR postdoctoral fellowship awarded to Mélanie Morin.

Notes on contributors

Sophie Bergeron, PhD, is an Associate Professor of Psychology at Université de Montréal and a Clinical Psychologist at the Sex and Couple Therapy Service of the McGill University Health Centre (Royal Victoria Hospital). She received her PhD in Clinical Psychology from McGill University in 1999 under the supervision of Dr. Irv Binik. The author and co-author of several articles, chapters and conference papers on the topics of dyspareunia and vulvodynia, Dr. Bergeron's research focuses on the treatment outcome of dyspareunia as well as on the role of psychosocial variables in the experience of sexual pain. She has served on the executive committee of the Society for Sex Therapy and Research, is a consulting editor for the journal *Archives of Sexual Behavior* and is an advisor to the DSM-V Work Group on Sexual and Gender Identity Disorders.

Mélanie Morin, PhD, PT, completed a Masters (2003) and a PhD (2008) in Biomedical Sciences at the Université de Montréal. During her graduate work she developed pelvic floor dynamometry, a new methodology for assessing the pelvic floor muscles. Moreover, she studied the involvement of the pelvic floor in the pathophysiology of postpartum and postmenopausal stress urinary incontinence. She is currently completing her postdoctoral fellowship in the Departments of Psychology at the Université de Montréal and McGill University focusing on the dynamometric and ultrasound assessment of the pelvic floor musculature in women with and without vestibulodynia. She is also an adjunct professor at the School of Rehabilitation in the Faculty of Medicine of the Université de Sherbrooke and will begin a tenure-track position at that institution in the fall of 2010.

Marie-Josée Lord, BSc (PT), received her bachelor's degree in Physical Therapy from McGill University in 1984. She is a Canadian pioneer and leader in the field of pelvic floor physiotherapy, a much needed and under-served area of women's health. She has been instrumental in making physiotherapy treatment for pelvic floor dysfunction a more mainstream focus and has contributed to the education of hundreds of physiotherapists across Canada. She inspires her students to expand their knowledge and expertise and instils professional pride in her colleagues, encouraging them to be leaders in their fields. Her specialty, over the past 20 years, has been as a clinical physiotherapist treating a variety of pelvic floor dysfunction, mainly focusing on dyspareunia. She is the past newsletter editor of the Women's Health Division of the Canadian Physiotherapy Association.

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