

CONTENTS / TABLE DES MATIÈRES

- 3 Editorial / *Éditorial*  
**Together We Move Forward**  
Mary Grossman
- 9 Discourse / *Discours*  
**Pain Assessment and Management in Canada:  
We've Come a Long Way But There Are Challenges  
on the Road Ahead**  
Michael H. McGillion, Judy Watt-Watson
- 17 **Attrition in Randomized and Preference Trials  
of Behavioural Treatments for Insomnia**  
*Taux d'abandon dans le cadre d'essais de thérapies comportementales contre  
l'insomnie avec répartition aléatoire et selon les préférences des participants*  
Souraya Sidani, Richard R. Bootzin,  
Dana R. Epstein, Joyal Miranda, Jennifer Cousins
- 35 **Method of Treatment Allocation: Does It Affect  
Adherence to Behavioural Therapy for Insomnia?**  
*Méthode d'attribution des traitements : a-t-elle une influence  
sur l'adhésion à une thérapie comportementale contre l'insomnie ?*  
Souraya Sidani, Richard R. Bootzin,  
Dana R. Epstein, Joyal Miranda, Jennifer Cousins
- 53 **Pilot Testing of a Psycho-educational  
Telephone Intervention for Women Receiving  
Uninformative BRCA1/2 Genetic Test Results**  
*Essai pilote d'une intervention psycho-éducative par téléphone pour les  
femmes ayant reçu des résultats non concluants après des tests de dépistage  
génétique concernant BRCA1 et BRCA2*  
Christine Maheu, Wendy S. Meschino, Weiming Hu, Joanne  
Honeyford, Ingrid Ambus, Meredith Kidd, Aronela Benea, Xin Gao,  
Madhis Azadbakhsh, Christian Rochefort, Mary Jane Esplen



- 72 **Honouring Stories: Mi'kmaq Women's Experiences  
With Pap Screening in Eastern Canada**  
*Histoires de dignité : comment le test de Pap est vécu  
par des Micmaques de l'Est du Canada*  
Catherine MacDonald, Ruth Martin-Misener,  
Audrey Steenbeek, Annette Browne
- 97 **Rethinking Case Study Methodology  
in Poststructural Research**  
*Repenser la méthode de l'étude de cas à partir du post-structuralisme*  
Shan Mohammed, Elizabeth Peter, Denise Gastaldo, Doris Howell



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## EDITORIAL

# Together We Move Forward

It is a privilege to assume the mantle of Editor-in-Chief of *CJNR*, a peer-reviewed general research journal that has played such a seminal role in the development of nursing scholarship throughout Canada and beyond for nearly five decades. The legacy of this journal is a source of great pride that can be shared by all nurse scientists, educators, clinicians, and administrators who have unwaveringly dedicated their professional lives to promoting academic nursing's rightful place in the learned decision-making hallways of health care.

It seems like only yesterday that I was invited to serve as the Associate Editor of *CJNR* under the inspiring leadership of Dr. Laurie Gottlieb. At that time (the early 1990s), doctoral programs in nursing were almost unheard of, the first fully funded Canadian nursing PhD program being established at the University of Alberta only in 1991. Since those early days, the commitment to excellence in nursing scholarship at the Journal has not wavered, nor has the belief that a journal should also reflect the values, concepts, and methodologies of its evolving discipline.

Taking stock is a normal exercise during a period of transition, and the process applies as much at *CJNR* as anywhere else. Turning to *CJNR*'s distinguished past for precious insights reveals a journal that has been fully implicated in the evolution of nursing scholarship in Canada and beyond. As a general research journal, *CJNR* may be said to embody the heart of Canadian academic nursing. Yet it also enjoys an international reach, offering an intellectually stimulating environment for exploring theoretical and methodological perspectives of common concern. By serving as a scholarly forum where ideas are proposed, challenged, tested, and integrated into the profession's growing scientific field of practice, *CJNR* highlights the unifying capabilities of a journal dedicated to developing nursing knowledge and clinical practice.

It seems obvious that *CJNR* will continue to serve academic nursing by honouring the discipline's epistemological underpinnings and the use of conventional and innovative methodologies to generate complementary forms of knowledge that enrich the profession's conceptual contexts, scientific knowledge, and scope of practice (Thorne & Sawatzky, 2014). The future holds the promise, however tentative, of narrowing the gap between research findings and clinical practice. Perhaps the same collective determination that distinguished the previous generation of nurse scholars may be called upon to ultimately enable the next generation of

clinical nurses to care for patients and families according to the full reach of the discipline.

A review of past *CJNR* issues offers insightful snapshots in the evolution of academic nursing. It seems to me that the development of the discipline over the last 45 years was nurtured by an obvious partnership between *CJNR* and nurse academics, who more often than not saw their shared aspirations for a fully actualized profession reflected, validated, and supported throughout the diverse columns of *CJNR*. When I reflect on the past, it becomes clear to me that the founding generation of nurse academics across the four arms of the discipline undertook their mission to develop nursing scholarship more in the name of the *nursing profession* than merely for the sake of their own *professional careers* — *the stakes for the future of the profession were that high*.

The accomplishments are impressive (Canadian Nurses Association & Canadian Association of Schools of Nursing, 2013):

- PhD-prepared faculty in tenure-track positions with successful research portfolios
- multiple funding resources at the federal, provincial, and university levels for nursing research
- entry to practice at the baccalaureate level, with the regrettable exceptions of Quebec, Nunavut, and the Northwest Territories
- nurse practitioner programs in 26 of Canada's 111 nursing schools

Yet the road ahead beckons with unfulfilled professional aspirations. How will nurse academics in the 21st century harness their tremendous intellect to shape and enhance the scientific scholarship of practising nurses across the clinical landscape? One inventive strategy has been the initiation of quality improvement studies in the clinical field. Yet these innovative studies are, for the most part, carried out within clinical environments framed by traditional hierarchical nursing administrative structures, medical paradigms, and standardized policies and procedures that together tend to strangle any incentive for generating tailored clinical interventions based on the integration of the scientific evidence filtered through aesthetic and personal ways of knowing (Thorne & Sawatzky, 2014).

In an era of growing medical and technological complexity, the question begs to be asked: What should constitute the evolving theory, scientific knowledge base, and clinical skills of nurses working in different roles and capacities across clinical environments, the majority in general hospital units? Should not evidenced-based knowledge through the prism of middle-range models increasingly serve as the principal driver of practice? Policy and procedures have their place, but within the larger epistemological context of the discipline. Moreover, what innovative

organizational structures might be introduced to enable clinical nurses to function more autonomously, according to the philosophical assumptions (values), epistemological foundations, and clinical skills of the discipline? For, in the final analysis, nursing as a profession is truly “known” only by the quality of the *evidence*-based care that is experienced by the public.

Not to be overlooked are the emerging scientific discoveries that may have important practice-based implications for the future of the nursing discipline within the field of health care. These scientific developments deserve our serious consideration, as they throw into relief the question of what new phenomena should be incorporated into the evolving scientific scope of our professional practice. For example, a core value of nursing is a focus on the “whole person.” But how should we effectively apply this value in clinical interventions? Scientific advances in the biological and molecular sciences (including genetics) would likely serve to deepen nursing’s knowledge of the complex and dynamic bio-psychosocial and spiritual dimensions of the whole that give rise to developmental and healing processes.

Another nursing value, “the whole is greater than the sum of all contributing parts,” is not always clear (Verhoef, Vandenheyden, & Fønnebo, 2006). One interpretation is that the components of any interventional strategy have multiple synergistic and distinct effects throughout the human organism. Does a multi-modal, multi-targeted approach aptly reflect this value in clinical practice? Should nursing research evaluate the effects of the components of a clinical intervention on biological (as well as psychosocial or behavioural) targets that have been shown to modulate psychological and behavioural endpoints, reflective of the health or overall resilience of the whole person (McEwen, Eiland, Hunter, & Miller, 2012)? Knowledge of nursing interventions that can induce a healing or restorative effect on an array of known biological targets would augment nurses’ clinical decision-making capabilities on behalf of their patients (Pavlov & Tracey, 2005). We need nurse scientists who will collaborate with geneticists and physiologists for the purpose of advancing our knowledge of the whole person, from the molecular to the spiritual, as conceptualized through the prism of nursing.

Interestingly, in the past decade the use of complementary therapies has seen an exponential rise among patients with chronic illnesses wishing to optimize their health. This trend has begun to attract scientific attention among researchers in medicine and psychology, yet only rarely among nurse researchers. Ironically, as early as 1969 the nursing regulatory body in Quebec published a policy statement recognizing mind-body modalities such as relaxation response (a form of breathing), massage, imagery, and reflexology (a form of touch) as clinical strategies consistent with the goals and objectives of the nursing profession.

A growing body of scientific evidence suggests that these therapies promote healing at the psychological, behavioural, and biological levels; other research has begun to yield the underlying biological mechanisms of action, including the switching on/off of specific genes associated with these complementary therapies in relation to the potential attenuation of many chronic illnesses (e.g., Dusek et al., 2008). Given that mind–body therapies are thought to lie within the purview of nursing and appear to induce an array of health benefits, particularly in distressed patients who are concomitantly under medical treatment, they deserve our scientific consideration as clinical interventions that might be integrated into a therapeutic approach to promote the health and healing of patients before, during, and after medical interventions.

“Tailoring nursing care” according to the preferences and beliefs of patients is also a well-known nursing value, and it has led to the study of new methodologies that challenge the underlying precepts of deductive models. Guided by the philosophical assumptions of the nursing discipline, Dr. Souraya Sidani and her colleagues have been courageous leaders in the quest to identify innovative research methods that are consistent with nursing values and that provide reliable, defensible scientific evidence that can be applied to nursing’s field of practice. *CJNR* is pleased to publish two such methodological articles in this issue. These articles and their ilk, though controversial, challenge our conventional thinking and call upon us to explore new methodological frontiers that can advance evidenced-based practice according to the values of our discipline (Thorne & Sawatzky, 2014; Verhoef et al., 2006).

As opportunities for research funding shrink, multidisciplinary collaboration on studies with shared yet distinct purposes is becoming a greater imperative, and one that nurse researchers will surely capitalize on, to the profession’s benefit. Another imperative for the profession is to address the growing gap between nursing science and the nature of clinical practice, and the related gap between academic preparation and clinical expectations in the workplace. These discussions are essential not only for advancing a unifying vision of practice over the next quarter century but for articulating a cohesive strategy for enabling the future practice of nursing to the full extent of its discipline. Whether such discussions lead to a meaningful re-integration of the four arms of practice may also determine whether the profession is able to fend off current and future politically and economically induced encroachments on our practice. One lesson we can learn from nursing’s past is the formidable potential of academic nurses working in unison to realize the goals of the discipline. Today’s challenges in the workplace have become more complex and the professional stakes are just as high.

So, who will constitute the nursing vanguard of the future? The obvious response is nurses from all four arms of practice, but in a science-valued society the vanguard that will likely be publicly recognized is the growing cohort of nurse scientists who can speak from a position of scientific credibility. Nurse researchers, however, represent a small fraction of the nursing profession — and their scientific achievements, while laudable, tend to have minimal impact on the quality and type of care experienced by the majority of patients and families. In the public's mind, it is still medicine more than nursing whose professional authority is derived ultimately from *evidenced-based* clinical interventions. We need to change that perception. If our scholarship, and particularly our clinical research, is not seen to be directly relevant to the quality of care of patients and families, then our discipline has a credibility problem. The methodological traditions associated with the qualitative and quantitative empirical findings that have informed nursing must include pilot studies and, especially, controlled trials that can evaluate the effectiveness of nursing interventions in the health and healing of patients and families.

Compounding the clinical challenges inherent in the paucity of nurse researchers in the health-care field is the growing proportion of nurse scientists publishing in specialty and medical journals, driven by the understandable contention that doing so will enhance their professional credibility and prestige. Nonetheless, this scholarly published work tends to remain buried in these scientific silos well beyond the collective reach of nurses in education, clinical practice, and administration. Yet all nurses should have easy access to this research so that the profession can advance as a cohesive whole. We are only as strong as our weakest link.

By publishing in a general research nursing journal, nurse scholars stand to expose their findings to a diverse academic nursing audience, fostering the conditions for cross-pollination in vital areas such as curriculum development and clinical practice. Without such academic forums for contributing directly to the four arms of the profession, the nurse scientist's relevance in the nursing profession may be weakened. We are inextricably connected, and the nurse scientist, I believe, can play a leadership role in advancing nursing practice in the 21st century — contingent upon producing nursing research that is seen to advance and inform all arms of practice.

As a general research journal, *CJNR* has consistently embodied the essence of the nursing discipline — exemplifying its core values, key concepts, and frameworks, while debating issues, airing concerns, and exploring possibilities emanating from the four interrelated arms of practice. It has enabled the kind of reflection that has engaged the whole profession. Indeed, *CJNR's* legacy suggests that we *all* have a collective responsibility to advance the nursing profession above and beyond pursuing our indi-

vidual career paths. To that end, the Journal remains an invaluable vehicle for promoting the nursing profession across the four arms of practice.

In keeping with *CJNR*'s distinguished past, the Journal is committed to publishing high-quality articles that address the study of theory, science, and practice in the field of health and illness, as well as scholarly discourses on diverse health-related topics of relevance to the profession and the public. In the future, we will offer articles on knowledge synthesis by leading researchers and on quality-improvement initiatives in the clinical workplace. From time to time we may offer a "focused cluster" of articles in a specific field of inquiry, in keeping with the Journal's mandate to cover the depth as well as the breadth of nursing's scientific knowledge. This March 2015 issue, Volume 47, Number 1, with its cluster of methodological articles, re-launches the Research Methods section as a regular feature.

Finally, as a general nursing research journal with global reach across the four arms of practice, *CJNR* will continue to serve as a unifying force for nursing scholarship and debate. We must all continue to grow together as a profession worldwide, firm in the belief that as a unified voice we can achieve the shared actualization of our profession.

**Mary Grossman**  
**Editor-in-Chief**

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## Discourse

# **Pain Assessment and Management in Canada: We've Come a Long Way But There Are Challenges on the Road Ahead**

**Michael H. McGillion, Judy Watt-Watson**

Canada has a rich history in the interprofessional study and treatment of pain. Seminal work by the pre-eminent Canadian scientists Melzack and Wall (1965) advanced our understanding of pain as a complex, subjective, and multidimensional experience. Their contributions laid the foundation for exceptional Canadian researchers and clinicians to engage in world-leading research and education, over the last four decades, on acute and persistent pain mechanisms and the implications for assessment and management. A major challenge that has driven the research and education agenda is the ubiquity of pain-related misbeliefs — that is, beliefs that persist despite evidence — which preclude effective pain assessment and management (Carr, 2009; Furstenberg et al., 1998; Lebovitz et al., 1997; Lin, Alfandre, & Moore, 2007; McGillion, Watt-Watson, Kim, & Graham, 2004; Strong, Tooth, & Unruh, 1999; Watt-Watson, 1992; Ying, Schulman-Green, Czaplinski, Harris, & McCorkle, 2007).

Despite evidence to the contrary, health-care professionals (HCPs) across health-care sectors routinely believe that pain is directly proportionate to the degree of tissue injury, that patients must demonstrate severe pain before receiving medication, that observable signs are the most reliable indicators of pain, and that patients should be encouraged to endure as much pain as possible before using an opioid analgesic (Carr, 2009; Furstenberg et al., 1998; Lebovitz et al., 1997; Lin et al., 2007; McGillion et al., 2004; Strong et al., 1999; Watt-Watson, 1992; Watt-Watson, Chung, Chan, & McGillion, 2004). Other misbeliefs, held by HCPs and patients alike, include the common notion that enduring pain is an expectation following surgery and that one pain-management strategy at a time is sufficient (Watt-Watson, 1992). These barriers to effective pain management are compounded by the fact that many

patients do not admit to having pain and are reluctant to ask for help (Watt-Watson, Clark, Finley, & Watson, 1999).

As a profession, nursing has played a major role in the efforts of the international pain community to institute change. In terms of pre-licensure education, the International Association for the Study of Pain (IASP) recognized the problem of pain misbeliefs over 20 years ago when it published its first core curriculum, laying the foundation for educational initiatives such as the University of Toronto Centre for the Study of Pain–Interfaculty Pain Curriculum (UTCSP–IPC), founded in 2002 (Hunter et al., 2008; Watt-Watson, Hunter, et al., 2004). The UTCSP–IPC continues to bring together students from six health sciences faculties (in 2014, a total of 947 students) to take part in a mandatory 20-hour curriculum targeting pain misbeliefs and optimal interprofessional pain assessment, care planning, and management practices. In 2012 the IASP approved an interprofessional pain curriculum based on the original tenets of its core curriculum and developed by a subgroup of its Education Initiatives Working Group (International Association for the Study of Pain, 2012). This curriculum now provides a means for health professions to learn a common language and to develop a basic understanding of pain mechanisms and biopsychosocial concepts fundamental to competent practice, grounded in the recognition of adequate pain relief as a fundamental human right.

In the practice arena, advances in the development of clinical practice guidelines (CPGs) have also been made. For example, the *Registered Nurses Association of Ontario Guidelines for Pain Assessment and Management*, now in its third edition, has led the way in providing interprofessional care teams with evidence-based recommendations for assessing and managing people with, or at risk for, any type of pain (Registered Nurses Association of Ontario, 2013). Promulgation of these CPGs, among others, has been made possible by a swell of intervention research in this country over the last four decades — much of it led by nurses. Because of the work of Canadian nurse leaders, we now have a strong foundation for understanding ways to optimize pain assessment and management for hospitalized infants and their families (Stevens et al., 2011; Stevens et al., 2014), an area of practice that was particularly lacking in the past. Concerted effort has also led to greater understanding of factors that confer vulnerability on the transition from acute to persistent pain following surgery (Choinière et al., 2014; McGillion et al., 2012), as well as to effective educational interventions that improve clinicians' knowledge and behaviour with regard to pain (McGillion et al., 2011; Parry et al., 2010; Watt-Watson, Carr, & McGillion, 2011; Watt-Watson, Stevens, et al., 2004). Cutting-edge work has advanced the delivery and international uptake of pain self-management education for both adolescent and adult

populations suffering from complex forms of persistent pain (LeFort, Gray-Donald, Rowat, & Jeans, 1998; McGillion et al., 2014; McGillion, LeFort, Stinson, 2008; McGillion, Watt-Watson, et al., 2008; Stinson et al., 2014).

Without question, the advances made by the international and national pain communities are to be celebrated, as are the contributions of nurse leaders who have helped to make such advances possible. Yet recognition of our achievements must be balanced with awareness of the new and ongoing challenges that we face. Our work is far from over.

We now possess the knowledge and the technology to manage pain effectively, yet alarming numbers of Canadians are still left in pain after surgery, even in our top hospitals. Evidence suggests that only 30% of analgesic medication ordered is actually administered in hospital and that up to 50% of patients report pain in the moderate-to-severe range following surgical procedures (Choinière et al., 2014; Watt-Watson, Stevens, et al., 2004). Left untreated, acute post-operative pain leads to persistent pain in 10% to 50% of patients who undergo common procedures, including inguinal hernia repair, mastectomy and wedge lumpectomy, cardiac and thoracic surgery, and major joint replacement (Kehlet, Jensen, & Woolf, 2006). Such pain challenges are not limited to the hospital setting. One in five Canadian adults report persistent pain that imposes significant activity limitation, difficulty at work, and financial hardship due to increased reliance on health services not covered by public health-insurance plans (Moulin, Clark, Speechley, & Morley-Forster, 2002; Schopflocher, Taenzer, & Jovey, 2011). The prevalence of persistent pain also increases with age, with rates as high as 65% among community-dwelling seniors and 80% among seniors living in long-term-care facilities (Hadjistavropoulos et al., 2009). The situation is no better among Canadian children and adolescents; like adults, one in five suffer persistent forms of pain, with an estimated 5% to 8% enduring pain that is severe enough to interfere with school work, social development, and physical activity (Huguet & Miró, 2008; Stanford, Chambers, Biesanz, & Chen, 2008).

Our way forward must be paved with the clear understanding that, despite the gains we have made, wide gaps persist between best evidence and clinical practice. Sound pain care in Canada is still threatened by pain-related misbeliefs and outdated management approaches (Lynch, 2011). Overlying these ongoing (and all too familiar) challenges is the contemporary controversy surrounding opioid analgesics (Lynch, 2013). Central to the debate is mounting public concern over prescription opioid analgesic (POA) use and misuse. As nurses with a vested interest in doing our utmost to ensure that our patients receive adequate pain

relief, we must continue to do what we have always done: turn to, and be clear on, the evidence.

There is no question that both the misuse and the non-medical use of POAs have resulted in unintended mortality (Lynch, 2013). However, the constant focus of the media — print, broadcast, and social — on opioid abuse, as opposed to the need for a balanced approach to the therapeutic use of POAs, has resulted in limited access to analgesics for those who legitimately need them. Recent research based on data from the Ontario Drug Benefit Plan indicates that 91.6% of deaths involving opioids also involve other substances, such as alcohol, benzodiazepines, or tricyclic antidepressants, suggesting that these agents are being used not necessarily for pain but for other reasons, such as addiction (Dhalla et al., 2009). Amidst the controversy, a major advance in promoting appropriate prescribing practices with safeguards has been the *Canadian Guideline for Safe and Effective Use of Opioids for Chronic Non-Cancer Pain* (Canada: National Opioid Use Guideline Group, 2010). These CPGs may have contributed to the recent decrease in non-medical use of POAs in Canada — the estimated prevalence of POA use to get “high” is now 0.4% (Lynch, 2013). Recent legislative changes in some provinces have extended the scope of practice for nurse practitioners to include prescription of controlled drugs and substances. It is paramount, therefore, that nurses continue to stay abreast of controversies as well as the latest developments in order to ensure safe prescribing practices.

As we continue to advocate for change, it is important to remember that the landscape of pain-related care and debate is changing rapidly. Traditionally, patients have tended to trust that clinicians are using evidence-based knowledge as a foundation for their clinical actions, even in cases where delivery of suboptimal pain relief has been the norm (Watt-Watson & Murinson, 2013). Today, patients and families are no longer passive recipients of health care; they bring their own information and beliefs to pain management (Watt-Watson & Murinson, 2013). The rise in uptake of social media and communication technologies, leading to a more engaged public, requires that HCPs also stay informed and continue to advocate for sound, evidence-informed practice. We remain accountable, to the public and ourselves, to lead and manage practice change.

New challenges, such as those born of the POA controversy (Lynch, 2013), give rise to new opportunities for research, such as the need to better understand the influence of media-related uptake on clinician and patient beliefs and their treatment-related preferences and values. Nurses can also be leaders in interprofessional pain curricula by promoting a focus on development of media-related competencies for best practice. Rising to such challenges is critical if we are to continue to be leaders in

pain research, education, advocacy, and practice. Considering what we have achieved over the last 40 years, there is no doubt that when it comes to the challenges that lie on the road ahead, we have what it takes to lead the way.

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Michael H. McGillion, Judy Watt-Watson

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## *Pain Assessment and Management in Canada*

Michael H. McGillion, Judy Watt-Watson

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Michael H. McGillion, Judy Watt-Watson

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# Attrition in Randomized and Preference Trials of Behavioural Treatments for Insomnia

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Dana R. Epstein, Joyal Miranda, Jennifer Cousins

Preferences for treatment contribute to attrition. Providing participants with their preferred treatment, as done in a partially randomized clinical or preference trial (PRCT), is a means to mitigate the influence of treatment preferences on attrition. This study examined attrition in an RCT and a PRCT. Persons with insomnia were randomly assigned ( $n = 150$ ) or allocated ( $n = 198$ ) to the preferred treatment. The number of dropouts at different time points in the study arms was documented and the influence of participant characteristics and treatment-related factors on attrition was examined. The overall attrition rate was higher in the RCT arm (46%) than in the PRCT arm (33%). In both arms, differences in sociodemographic and clinical characteristics were found between dropouts and completers. The type of treatment significantly predicted attrition (all  $p \leq .05$ ). The results provide some evidence of a lower attrition rate in the PRCT arm, supporting the benefit of accounting for preferences as a method of treatment allocation.

**Keywords:** treatment preferences, preference trial, attrition, methodology, intervention research

Résumé

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## Taux d'abandon dans le cadre d'essais de thérapies comportementales contre l'insomnie avec répartition aléatoire et selon les préférences des participants

Souraya Sidani, Richard R. Bootzin,  
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Les préférences en matière de traitement influent sur le taux d'abandon. Offrir aux participants le traitement qui correspond à leurs préférences, comme dans le cadre d'un essai clinique avec répartition partiellement aléatoire ou selon les préférences, constitue un moyen d'atténuer l'incidence de la préférence en matière de traitement sur le taux d'abandon. La présente étude examine les taux d'abandon observés lors d'un essai clinique avec répartition aléatoire et d'un essai clinique avec répartition partiellement aléatoire ou selon les préférences. Un groupe de personnes souffrant d'insomnie se sont vu attribuer une thérapie comportementale de façon aléatoire ( $n = 150$ ) et les membres d'un autre groupe selon leurs préférences ( $n = 198$ ). Le nombre d'abandons au sein de chacun des groupes a été consigné à différents moments de l'étude, puis une analyse des caractéristiques des participants et des facteurs liés à chaque thérapie a été effectuée afin de déterminer leur influence sur le taux d'abandon. Le taux d'abandon global s'est avéré plus élevé au sein du groupe avec attribution aléatoire de la thérapie (44 %) qu'au sein de l'autre groupe (33 %). Dans les deux groupes, des différences d'ordre sociodémographique et liées à des caractéristiques cliniques ont été observées entre les participants ayant abandonné et ceux qui ont terminé la thérapie. Le type de thérapie suivi permettait de prédire de façon notable s'il y aurait abandon (tout  $p \leq 0,05$ ). Les résultats montrent un taux d'abandon moins élevé parmi les participants qui se sont vu attribuer une thérapie selon leurs préférences, ce qui appuie l'hypothèse selon laquelle il y a un avantage à tenir compte des préférences dans la méthode d'attribution des traitements.

**Mots-clés :** préférences en matière de traitement, essai avec répartition selon la préférence, taux d'abandon, méthode, recherche

## **Introduction**

Attrition or withdrawal of eligible participants before, during, or after exposure to treatment presents a major threat to internal and external validity in randomized clinical trials (RCTs) (Valentine & McHugh, 2007). Attrition is often attributed to personal and clinical characteristics of participants (e.g., education, health status), features of the study (e.g., burdensome procedures), and attributes of the treatment (e.g., complex and inflexible protocols) (Ahern & LeBroque, 2005; Kemmler, Hummer, Widschwendter, & Fleishhacker, 2005). Preferences for treatment have been recently recognized as factors contributing to attrition in an RCT (Preference Collaborative Review Group [PCRG], 2009). Participants may have a preference for the experimental or the comparison treatment. With random assignment, participants are allocated to the preferred or non-preferred treatment. Those assigned to the non-preferred treatment may be disappointed at not receiving the treatment of choice and hence withdraw from the trial (Sidani, Miranda, Epstein, & Fox, 2009). Providing participants with the treatment of choice is a means to mitigate the influence of treatment preferences on attrition, as evidenced in the results of two meta-analyses (PCRG, 2009; Swift, Callahan, & Vollmer, 2011) showing a lower attrition rate for participants assigned to a treatment that is congruent with their preferences (i.e., matched), as compared to participants with mismatched treatment.

Behavioural therapies for managing insomnia have demonstrated efficacy, evidenced by large effect sizes in reducing sleep onset latency and in improving sleep quality and moderate effects sizes in decreasing the length of time awake after sleep onset and in increasing total sleep time (Irwin, Cole, & Nicassio, 2006; Morin et al., 2006). However, trials of behavioural therapies for insomnia are plagued with high attrition rates, estimated at 40% (Ong, Kuo, & Manber, 2008). Recent evidence (Epstein, Sidani, Bootzin, & Belyea, 2012; Hebert, Vincent, Lewycky, & Walsh, 2010) relates withdrawal to participants' characteristics (e.g., health status) and perceptions of the behavioural therapies (e.g., dislike of the treatment). The extent to which attrition is reduced by providing persons with insomnia the behavioural treatment of their choice is not known and was investigated in this two-arm trial.

The arms represented two designs commonly used to determine the influence of treatment preferences: the RCT and the PRCT (partially randomized clinical or preference trial). In the RCT, participants indicate their preference at baseline but are randomized to treatment. The influence of preference is usually examined by categorizing participants into the match subgroup (i.e., received treatment that is consistent with their choice) and the mismatch subgroup (i.e., received non-preferred treat-

ment) and comparing the subgroups on attrition (PCRG, 2009). In the PRCT arm, participants indicate their preference, which then guides their allocation to treatment. Those with no preference are randomly assigned to treatment, whereas those expressing a preference for a particular treatment are allocated to that treatment (for details, see Sidani, Miranda, et al., 2009).

The overall purpose of the trial was to examine attrition and predictors of attrition in the RCT and PRCT arms. The predictors were as follows: participants' demographic, sleep, and psychological characteristics; perceived acceptability of the treatments measured at pre-test; type of treatment to which the participants were assigned; and method of allocation to treatment. These predictors have been found to be associated with attrition in previous studies (e.g., Epstein et al., 2012; Hebert et al., 2010). The study's specific objectives were to (1) describe the attrition rate at different points in time (before, during, and after completion of treatment) and the overall attrition rate in the RCT and PRCT arms; (2) describe the overall attrition rate for the subgroups of participants assigned to each treatment randomly or on the basis of preference; (3) explore reasons for withdrawal reported by participants in the RCT and PRCT arms; (4) compare participants who withdraw and those who complete the study on demographic and clinical characteristics, measured at pre-test, within the RCT and PRCT arms; and (5) examine predictors of attrition in the RCT and PRCT arms of the study.

## **Materials and Methods**

### ***Design***

The two-arm trial was conducted at two sites that participated in a large methodological study to evaluate the utility of different research designs in enhancing the validity and clinical relevance of findings in intervention evaluation research (Sidani, Epstein, Bootzin, Mortiz, & Sechrest, 2007). The sites were located at research-intensive universities in large US cities. The application of the same participant eligibility criteria and recruitment strategies was intended to maintain the comparability on pre-test demographic and clinical characteristics of the samples accrued at the two sites. At both sites, the same behavioural treatments for insomnia were implemented by master's-prepared therapists who were given standardized training in the conceptualization and delivery of the treatments and who adhered to the protocol manual in delivering the treatments. Further, the same methods were applied to measure the outcomes at pre-test, post-test, and follow-up.

At both sites, eligible participants completed measures of demographic, psychological, and sleep characteristics at pre-test. After providing

these data, participants completed the Treatment Acceptability and Preference (TAP) scale (Sidani, Epstein, Bootzin, Moritz, & Sechrest, 2009). This scale provides a written description of the behavioural treatments under evaluation and contains items to rate the acceptability of each treatment and questions about treatment preferences. In the RCT arm, participants responded to the items assessing treatment acceptability but were not asked to indicate which treatment they preferred, in order to minimize the ethical consequences of ignoring participants' preferences (PCRG, 2009). Participants were then randomly assigned to treatment, using sequential opaque, sealed envelopes. In the PRCT arm, participants rated the treatments' acceptability and indicated their preferred treatment; those who had no preference were randomly assigned to treatment, whereas those who expressed a preference were allocated to the treatment of choice. A \$40 incentive was given to offset transportation costs associated with attending the data collection and treatment sessions at the study office.

The study protocol was approved by the Institutional Research Board at the participating academic institutions. All participants provided informed, written consent prior to enrolment.

### ***Setting and Sample***

Persons with chronic insomnia formed the target population. Inclusion criteria were as follows: (1) community-dwelling, non-institutionalized adult 21 years or older; (2) proficiency in English; (3) complaint of difficulty falling asleep and/or maintaining sleep as indicated by sleep onset latency and/or time awake after sleep onset of at least 30 minutes per night, for 3 or more nights per week, ascertained using a sleep diary kept for 14 days at pre-test, and at least 3 months' duration as reported by participants. Exclusion criteria were as follows: self-reported diagnosis of sleep apnea, cognitive impairment reflected in a score of less than 27 on the Mini-Mental State Exam (Folstein, Folstein, & McHugh, 1975), or psychological impairment evidenced by a Global Severity Index T score of over 50 on the Brief Symptom Inventory (Derogatis & Melisaratos, 1983).

Persons with insomnia were recruited through advertisements in local newspapers and newsletters and distribution of flyers and brochures to community health centres and clinics. Persons interested in the study were asked to contact the research office for more information.

### ***Treatment Options***

The behavioural treatments for insomnia included sleep education and hygiene (SEH) and multi-component intervention (MCI). SEH provided

information about sleep processes and functions and about general strategies to promote sleep, such as avoiding caffeine at night and nicotine around bedtime. The information was given in a booklet that participants could read at their convenience. SEH was found minimally effective in reducing the severity of insomnia and in improving sleep outcomes, such as sleep efficiency (Morin, Culbert, & Schwartz, 1994). The MCI consisted of stimulus control therapy (SCT) and sleep restriction therapy (SRT), in addition to SEH. The specific instructions making up the SCT focus on developing new sleep habits and rising at the same time every morning, with the goals of re-associating the bed and bedroom with sleep and forming a consistent sleep pattern (Bootzin & Epstein, 2011). SRT restricts the amount of time spent in bed to the person's sleep time identified through the sleep diary maintained at pre-test and developing a consistent sleep-wake schedule (Wohlgemuth & Edinger, 2000). The MCI was administered in four 90-minute group sessions followed by two telephone contacts over a 6-week period. It was found to be effective in reducing the severity of insomnia and improving sleep outcomes (Morin et al., 2006).

### ***Sample Size***

The sample size was estimated to detect differences in the sleep outcomes, of a moderate magnitude, between behavioural therapies and method of allocation to treatment (i.e., random and preference). Medium-sized effects (.4–.6) were anticipated on the basis of (1) results of systematic reviews indicating that SEH is a minimally effective treatment and the MCI had moderate-to-large effects on sleep outcomes (Morin et al., 1994), and (2) findings of a meta-analysis showing a low-moderate effect for treatment preferences on outcomes (Swift et al., 2011). Applying Cohen's (1992) criteria for a medium effect size for the treatment and method of treatment allocation comparisons, and setting alpha at .05 and beta at .80, the number of participants needed was 50 per group. The total sample size was 300, distributed as follows: 100 (50 x 2 treatment groups) for the RCT arm and 200 (50 x 2 treatment groups x 2 methods of treatment allocation) for the PRCT arm.

### ***Variables and Measures***

***Demographic variables.*** Age, sex, education level, ethnicity, and employment status were assessed using standard questions. Education level was represented by number of years of formal schooling. To balance the distribution across meaningful categories, ethnicity was dichotomized into white and non-white and employment status into employed and non-employed.

**Sleep variables.** The sleep outcomes included sleep parameters, perceived sleep severity, beliefs about sleep, and self-efficacy about sleep. The sleep parameters were assessed using the sleep diary, completed daily upon awakening and returned to a voice-mail service to minimize recall bias. The sleep diary demonstrated test-retest reliability ( $r = .69-.93$ ) and validity, evidenced by significant correlation between the values of the respective sleep parameters estimated with data reported in the sleep diary and recorded using actigraphy (Buysse, Ancoli-Israeli, Edinger, Lichstein, & Morin, 2006). The sleep parameters, computed from relevant diary data, were (1) sleep onset latency (SOL): length of time, in minutes, to fall asleep; (2) wake after sleep onset (WASO): length of time, in minutes, spent awake, over all awakenings; and (3) sleep efficiency (SE): the percentage of the total time in bed actually asleep. Perceived insomnia severity was measured using the Insomnia Severity Index (ISI) (Morin, 1993). The ISI contains seven items that have demonstrated internal consistency reliability and concurrent and construct validity (Morin, Belleville, Bélanger, & Ivers, 2011). Self-efficacy about sleep was measured using the nine-item Self-Efficacy Scale (Lacks, 1987). It inquires about confidence in carrying out sleep-related behaviours, such as feeling relaxed when lying in bed, and demonstrated acceptable reliability (Cronbach's  $\alpha = .82$ ) in this study.

**Psychological variables.** The psychological variables included depression and sleep-related anxiety. The Center for Epidemiologic Studies–Depression (CES–D) scale (Radloff, 1977) was used to assess depressive mood. It has established psychometric properties in different populations (Naughton, Shumaker, Anderson, & Czajkowski, 1996). Sleep-related anxiety was measured using the Sleep Anticipatory Anxiety Questionnaire (SAAQ) developed by Bootzin, Shoham, and Kuo (1994). The SAAQ captures pre-sleep cognitive and somatic arousal. It has been found to be reliable and valid (Kuo, Raccioppo, Bootzin, & Shoham, 1994).

**Treatment acceptability and preferences.** Acceptability and preferences for the SEH and MCI were assessed using the TAP measure, which has been shown to have acceptable psychometric properties (Sidani, Epstein, et al., 2009). The measure presents information on each treatment's goals, activities, mode of delivery, dose, effectiveness, and side effects, followed by items requesting participants to rate the acceptability of each treatment separately, using a five-point scale ranging from *not at all* acceptable (0) to *very* acceptable (4). Participants in the PRCT arm are then asked if they have preferences among the treatments they rate and which treatment they prefer to receive to manage their insomnia.

**Attrition.** A log was used to track participants' withdrawal from the study at different time points and to document the reasons they gave for dropping out. Attrition rate was computed as the percentage of participants who withdrew after being found eligible and providing written consent and baseline data. Attrition rates were calculated for (1) early withdrawal — that is, after completion of pre-test measures but before exposure to treatment; (2) withdrawal during the 6 weeks of treatment; (3) late withdrawal — that is, after completion of post-test measures but prior to the 3-month follow-up; and (4) overall withdrawal — that is, at any time during the study.

### ***Data Analysis***

The sleep parameters were computed from pertinent items of the sleep diary and averaged over the 14-day period at baseline. Total scores were computed for each sleep and psychological characteristic as well as for treatment acceptability. A factorial analysis of variance for continuous variables and a chi-square test for categorical variables were used to determine the comparability of participants' characteristics assigned to SEH and MCI, randomly or by preference, in the RCT and PRCT arms.

To address objective 1, the attrition rates were computed for the RCT and PRCT arms for each time point and across all time points (overall attrition). To address objective 2, the overall attrition rates were estimated for those assigned to the SEH and the MCI randomly or by preference. Chi-square test was used to examine differences in the number of participants who withdrew from the study by arm (RCT vs. PRCT), method of treatment allocation (random vs. preference), and type of treatment (SEH vs. MCI). To address objective 3, the reasons given by participants for dropping out were content analyzed and the number reporting the same reason was calculated. To address objective 4, independent sample *t* test was used to compare participants, within each arm of the study, who did and did not withdraw on pre-test variables. To address objective 5, logistic regression was applied to identify predictors of attrition in both arms of the study (i.e., data from all participants were pooled for this analysis). The predictors were entered into the regression model, using the forced entry method, in three blocks. The first block included treatment-related variables — that is, perceived acceptability, type of treatment, and method of treatment allocation. The second block consisted of sociodemographic characteristics. The third block contained the clinical (i.e., sleep and psychological) characteristics. The Wald test and its associated *p* value and the odds ratio (OR) indicated variables that significantly contributed to attrition in the RCT and PRCT arms.



## Results

### *Attrition Rates*

In the RCT arm, 183 persons showing interest in the study agreed to be screened; 33 did not meet all eligibility criteria and 150 were found eligible and provided written consent. The numbers of consenting individuals who withdrew from the trial before, during, and after exposure to treatment are reported in Table 1. The early withdrawal rate was 35.3%, the dropout rate during the treatment period was 2.0%, and the late withdrawal rate was 8.6%. The overall attrition rate was 46%. A total of 97 participants completed the pre-test measures. Of these, 45 (46.3%) were randomized to SEH and 52 (53.7%) to MCI. The percentage who withdrew from the study was 16% for those randomized to SEH and 12.8% for those randomized to MCI.

<b>Study Time Point</b>	<b>RCT Arm</b>	<b>PRCT Arm</b>
Number of participants	150	198
Number of withdrawals before treatment (early)	53	35
Number of withdrawals during treatment period	3	16
Number of withdrawals after treatment (late)	13	15
Total number of withdrawals from study	69	66

In the PRCT arm, 224 persons underwent screening; 26 were not eligible and 198 were eligible and consented to participate. The numbers of consenting individuals who withdrew at different time points are presented in Table 1. The early attrition rate was 17.6%, the dropout rate during the treatment period was 8.0%, and the late withdrawal rate was 7.5%. The overall attrition rate was 33.3%. A total of 163 participants completed the pre-test measures. Of these, 31 indicated no preference for the insomnia treatments and were randomly assigned to SEH ( $n = 15$ ) and MCI ( $n = 16$ ); the percentages of these participants who withdrew from the study were 20% and 0%, respectively. The remaining participants ( $n = 132$ ) expressed a preference and were allocated to the chosen treatment: 21 selected SEH and 111 MCI; the percentages of these participants who dropped out were 19.1% and 8.3%, respectively.

Results of the chi-square test comparing the total number of participants who withdrew from the study between the trial arms (RCT vs. PRCT), method of treatment assignment (random vs. preference), and

type of treatment (SEH vs. MCI) indicated no statistically significant differences (all  $p > .05$ ). However, the overall attrition rate was slightly higher in the RCT (46%) than the PRCT (33.3%); this difference is related to the higher early withdrawal rate observed in the RCT (35.3%) compared to the PRCT (17.6%).

***Reasons for Withdrawal***

In both study arms, most of the participants who dropped out of the study did not return the research assistant's phone call inquiring about their reasons for doing so (30% in the RCT, 43% in the PRCT). The reasons stated by the remaining participants were categorized into characteristics of participants, study, and treatment (Table 2). Some participants gave more than one reason; therefore, the percentages were computed for the total number of reasons provided within each arm. The most frequently stated reasons for withdrawal related to characteristics of participants, representing 37.6% and 46.4% of the reasons given in the RCT and PRCT arm, respectively. Characteristics of the study accounted for 18.1% of the reasons given in the RCT and 12.5% of those given in the PRCT arm. Characteristics of treatment were reported more commonly in the RCT arm (19.4%) than in the PRCT arm (7.1%).

<b>Reason</b>	<b>RCT Arm</b>	<b>PRCT Arm</b>
<b><i>Characteristics of participants</i></b>		
Too busy	13	10
No longer interested	10	
Relocation	2	7
Health condition	2	7
Improved sleep		2
Transportation	2	
<b><i>Characteristics of study</i></b>		
Scheduling conflict	10	5
Inadequate compensation		1
Dislike filling in sleep diary	3	1
<b><i>Characteristics of treatment</i></b>		
Demanding	4	
Dislike treatment	5	3
Getting treatment elsewhere	2	
Treatment did not work	4	1

**Table 3 Pre-test Characteristics by Type and Method of Allocation to Treatment Within RCT and PRCT Arms**

Characteristic	RCT			PRCT		
	Random			Random		
	SEH	MCI	Preference	SEH	MCI	Preference
Number of participants	45	52	111	15	16	21
Age (mean)**	52.4	55.2	49.6	45.3	39.8	42.0
Sex ( <i>n</i> women)	3	2	7	3	0	4
Marital status ( <i>n</i> married)	30	25	59	6	12	11
Education (mean years)	15.9	16.7	15.7	16.4	16.1	17.6
Ethnicity ( <i>n</i> white)	42	45	97	14	14	16
Employment ( <i>n</i> employed)	83	28	81	12	15	17
SOL (mean)	35.2	45.3	41.3	32.2	45.3	37.3
WASO (mean)*	48.9	56.9	49.7	48.1	52.9	36.5
SE (mean)**	72.4	69.1	71.2	74.9	69.6	74.9
ISI (mean)**	18.2	17.1	17.0	13.9	17.7	15.5
SES (mean)	2.4	2.3	2.4	2.6	2.2	2.5
CES-D (mean)	13.5	11.9	13.1	14.6	15.6	10.8
SAAQ (mean)	2.3	2.2	2.2	2.2	2.5	2.2
Acceptability of MCI (mean)**	2.4	2.6	2.6	2.1	2.6	1.9
Acceptability of SEH (mean)**	2.3	2.1	1.6	2.4	2.4	2.7

\*  $p \leq .05$ , \*\*  $p \leq .01$   
 Legend: SOL = sleep onset latency; WASO = wake after sleep onset; SE = sleep efficiency; ISI = Insomnia Severity Index; CES-D = Center for Epidemiologic Studies-Depression scale; SAAQ = Sleep Anticipatory Anxiety Questionnaire; MCI = multi-component intervention; SEH = sleep education and hygiene

### ***Predictors of Overall Attrition***

The predictors of overall attrition included the participants' socio-demographic, sleep, and psychological characteristics and perceived acceptability of the behavioural treatments, measured at pre-test, as well as the treatment assigned and the method of treatment allocation. The average values on these characteristics are presented in Table 3 for the subgroups, in the RCT and PRCT arms, allocated to the SEH and MCI randomly or by preference. Factorial analysis of variance and chi-square test comparing these characteristics by study arm, method of allocation, and type of treatment showed statistically significant differences on five characteristics.

**Age.** On average, participants in the RCT arm were older than those in the PRCT arm,  $F(1, 254) = 13.8, p < .01, \eta^2 = .05$ . Those assigned to the SEH randomly or by preference were younger than those allocated to the MCI using either method, while those in the PRCT randomized to the MCI were the youngest subgroup (i.e., method of treatment allocation  $\times$  type of treatment interaction effect),  $F(1, 254) = 4.2, p = .03, \eta^2 = .01$ .

**WASO.** The average number of minutes awake after sleep onset differed by type of treatment,  $F(1, 253) = 4.8, p = .02, \eta^2 = .01$ ; participants assigned to SEH randomly or by preference had a lower mean than those in the MCI.

**Sleep efficiency.** There was a statistically significant main effect for type of treatment,  $F(1, 253) = 6.6, p = .01, \eta^2 = .02$ . Participants allocated to the MCI randomly or by preference reported lower levels of sleep efficiency than those assigned to the SEH using either method.

**Perceived severity of insomnia.** A statistically significant study arm  $\times$  type of treatment interaction effect was found,  $F(1, 247) = 8.5, p = .004, \eta^2 = .03$ . In the PRCT arm, those allocated to the SEH randomly or by preference perceived lower levels of insomnia severity than those assigned to the MCI by either method.

**Treatment acceptability.** The perceived acceptability of SEH and MCI differed for participants allocated to these treatments, regardless of the method of treatment allocation and study arm,  $F(1, 256) = 20.5, p < .01, \eta^2 = .07$  for MCI;  $F(1, 256) = 18.6, p < .01, \eta^2 = .06$  for SEH. There was a tendency for participants to rate the assigned treatment as slightly more acceptable than the alternative one; the differences in rating were prominent among participants expressing preferences for the treatments under evaluation.

The extent to which these variables contributed to the overall attrition was examined using logistic regression analysis. The selected predictors were entered in three blocks. In the first block, consisting of treatment-

related variables, only the type of treatment had a statistically significant association with attrition (OR = .74, 95% confidence interval: .35 - .90, Wald test = 3.85,  $p = .049$ ), indicating that participants assigned to the SEH (regardless of method of allocation) were more likely to withdraw from the study. The second and third blocks, including sociodemographic and clinical characteristics, showed no statistically significant relationship with attrition (all  $p > .05$ ).

## **Discussion**

The findings of this study indicate (1) a slightly higher overall attrition rate in the RCT arm as compared to the PRCT arm, with the largest percentage of participants dropping out prior to exposure to the allocated treatment; (2) a slightly higher percentage of participants withdrawing from the RCT as compared to the PRCT and reporting treatment-related factors as reasons for doing so; (3) participants who withdrew differed from those who completed the study on two characteristics assessed at pre-test in the RCT and one characteristic in the PRCT; and (4) the type of allocated treatment was the only significant predictor of attrition. The method of assignment to treatment did not contribute significantly to attrition.

The results pertaining to the attrition rate and reasons for withdrawal are consistent with and extend the trends reported in the literature on attrition in general and on behavioural treatments for insomnia. The overall attrition rates observed in the RCT and PRCT arms are within the range (10–40%) reported for studies evaluating behavioural therapies for insomnia in clinical settings (Ong et al., 2008). However, the overall attrition rate in the RCT was higher by 12.7 percentage points than the rate in the PRCT arm. This difference is attributed to the higher rate of early attrition (i.e., before exposure to treatment) in the RCT as compared to the PRCT; the difference was 17.7 percentage points. The exact reason for early withdrawal from the RCT may be difficult to identify. A review of reasons for withdrawal provides some explanation. A larger number of participants in the RCT than in the PRCT mentioned treatment-related factors (treatment is demanding, dislike of treatment), loss of interest in the study, and scheduling conflict as reasons for withdrawal (Table 2). These reasons reflect unfavourable reactions to the trial, which may be related to disappointment with the randomized treatment. Disappointment and subsequent dissatisfaction with treatment represent the mechanism underlying attrition in an RCT (PCRG, 2009). Participants in the RCT arm viewed the MCI as slightly more acceptable than the SEH and may have been dismayed if they were randomized to the SEH. Dismayed participants may have dropped out early. This

point is supported in the larger percentage of participants in the RCT arm dropping out of the SEH (16%) than out of the MCI (12.8%). This finding also suggests that participants with preferences enrol in an RCT because they are aware that they have a 50% chance of being randomized to the preferred treatment; however, they withdraw from the trial early on to avoid exposure to the allocated treatment if it is incongruent with their choice (Bradley, 1993).

In contrast, participants in the PRCT arm were asked to indicate their preferences and were allocated to their chosen treatment. The “act of choosing” a treatment and the subsequent sense of control may explain participants’ decision to pursue treatment (Leykin et al., 2007), as implied in the lower early withdrawal rate relative to the RCT arm. However, a slightly larger percentage of participants in the PRCT than the RCT dropped out during treatment. The reasons for withdrawal given by these participants cannot account for this finding. Since many participants in the PRCT arm dropped out without stating a reason, it is not possible to rule out the following factors as contributing to attrition during the treatment period: (a) dissatisfaction with some aspects of treatment delivery, such as therapeutic alliance with the therapist, experienced benefits, or discomfort (i.e., side effects) associated with the allocated therapy (Ong et al., 2008); and (b) changing health condition or sleep pattern of participants. The role that these factors, particularly satisfaction with treatment, play in influencing attrition should be further investigated under the random and preference methods of treatment allocation.

Participants who withdrew and those who completed the study differed on a small number (1–2) of characteristics: age, ethnicity, and WASO. The results of these comparisons should be viewed with caution, particularly in the PRCT arm. The comparisons were done on a large number of variables in both arms, potentially leading to type I error. In the PRCT arm, the numbers of dropouts and completers were not balanced, which may have led to violation of the equality of variance assumption and potential incorrect conclusions. Nonetheless, the differences in sociodemographic and clinical characteristics of dropouts and completers observed in the RCT and PRCT arms have been reported in previous studies (Ahern & LeBroque, 2005). In general, this consistent finding suggests that differences in the profile of dropouts and completers are prevalent in intervention evaluation trials that use random or preference-based methods of assigning participants to treatments. The differences result in self-selection bias; the sample of participants who complete a study may not represent all subgroups of the target population, which limits the generalizability of results pertaining to the effects of the treatment and the contribution of preferences.

Of the sociodemographic, clinical, and treatment-related variables included in the regression analysis, only the type of treatment significantly predicted attrition. Method of treatment assignment was not associated with withdrawal. Participants allocated to the SEH, randomly or by preference, were more likely to withdraw from the trial. There are two explanations for this finding. First, significant differences were observed in the sociodemographic (age) and clinical (WASO, sleep efficiency, insomnia severity) characteristics and treatment acceptability for participants allocated to the SEH and the MCI randomly or by preference. These differences could have confounded the influence of treatment on attrition. Second, the SEH is considered minimally effective in managing insomnia (Morin & Benca, 2012). It is possible that participants who received this treatment, regardless of allocation method, did not experience improvement in their sleep. A few participants in the RCT and PRCT arms indicated that the treatment “did not work” (Table 1). Therefore, they may have lost interest in the study and thus withdrew. Perceived ineffectiveness of treatment has been reported as a reason for withdrawal (Ahern & LeBroque, 2005; Kemmler et al., 2005). This raises questions about the suitability or appropriateness of including minimally effective treatments in trials aimed at evaluating the influence of treatment preferences on attrition, adherence, and outcomes. Treatments with differential acceptability and effectiveness may contribute to differential attrition, as was found in this study, which represents a major threat to the validity of conclusions regarding the effects of treatment and/or preferences. Therefore, treatments of comparable acceptability and effectiveness should be selected in preference trials.

The implementation of the RCT and PRCT arms in this study points to some limitations of these designs in examining the contribution of treatment preferences in intervention evaluation research. In the RCT arm, the high rate of early withdrawal raises the possibility that participants with strong preferences declined to enrol in the study to avoid randomization to the non-preferred treatment. Thus, enrollees may hold weak or no preferences that do not affect their responses to the randomly assignment treatment; this, in turn, could lead to incorrect conclusions about the impact of preferences (Swift et al., 2011). Alternatively, with randomization participants may be incidentally allocated to their treatment of choice; when a large proportion of participants (e.g., > 75%) receive the preferred treatment, the distribution of those allocated to the treatments randomly or by preference is unbalanced, limiting the comparisons among the groups defined by type of treatment and method of allocation and the resulting conclusion regarding the influence of treatment preferences. A similar unbalanced distribution is highly likely in the

PRCT arm if most participants choose one treatment over the alternative treatments under evaluation, as was the case in this study.

## Conclusion

The results of this study provide evidence of lower early and overall attrition rates in the PRCT arm, which is consistent with the findings of two meta-analyses (PCRG, 2009; Swift et al., 2011). However, additional research is needed to determine the reproducibility of the findings when alternative treatments are active, equally effective behavioural therapies for insomnia and to elucidate the mechanism responsible for the low attrition among participants allocated to the preferred treatment.

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## *Preference and Attrition*

Souraya Sidani, Richard R. Bootzin, Dana R. Epstein, Joyal Miranda, Jennifer Cousins

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# Method of Treatment Allocation: Does It Affect Adherence to Behavioural Therapy for Insomnia?

Souraya Sidani, Richard R. Bootzin,  
Dana R. Epstein, Joyal Miranda, Jennifer Cousins

Adherence to treatment is critical in determining the effects of behavioural therapy and may be affected by participants' preference for treatment. The purpose of this study was to determine the extent to which method of allocation to treatment (random vs. preference-based) influences adherence (exposure and enactment) to behavioural therapy. Participants received behavioural therapy for the management of insomnia randomly or by preference. Exposure was assessed as attendance at the treatment sessions, enactment as self-reported application of treatment recommendations. Participants ( $N = 262$ ) attended a mean of 5.6 treatment sessions, applied the treatment recommendations frequently, and reported high levels of overall compliance. There was no difference between the random and preference groups in terms of exposure to and enactment of treatment. Randomization to the preferred treatment, dissatisfaction with the allocated treatment, and self-report bias could play a role in the findings and should be explored in future research.

**Keywords:** treatment preferences, adherence, enactment, exposure, methodology, intervention research

Résumé

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## **Méthode d'attribution des traitements : a-t-elle une influence sur l'adhésion à une thérapie comportementale contre l'insomnie ?**

**Souraya Sidani, Richard R. Bootzin,  
Dana R. Epstein, Joyal Miranda, Jennifer Cousins**

L'adhésion au traitement a un effet déterminant sur l'efficacité d'une thérapie comportementale et est susceptible d'être influencée par les préférences des participants en matière de traitement. L'objectif de la présente étude est de déterminer la mesure dans laquelle la méthode d'attribution des traitements (aléatoire ou fondée sur les préférences) exerce une influence sur l'adhésion (exposition ou mise en action) à une thérapie comportementale. Les participants à l'étude se sont vu attribuer une thérapie pour la gestion de l'insomnie selon une méthode aléatoire ou fondée sur leurs préférences. L'exposition a été définie et évaluée comme une présence aux séances de thérapie et la mise en action comme l'application des recommandations associées à la thérapie, selon les déclarations des participants eux-mêmes. Les participants ( $N = 262$ ) ont assisté en moyenne à 5,6 séances de thérapie, ont appliqué les recommandations associées à la thérapie de façon fréquente et ont indiqué avoir fait preuve d'un degré élevé de respect de la thérapie en général. Aucune différence n'a été observée entre les groupes avec attribution aléatoire et ceux avec attribution selon les préférences en ce qui concerne l'exposition à la thérapie et la mise en action de celle-ci. La répartition aléatoire des participants à la thérapie préférée, l'insatisfaction de participants quant à la thérapie attribuée et le caractère subjectif des déclarations des participants ont possiblement joué un rôle dans l'établissement des résultats et devraient être analysés plus en profondeur dans le cadre d'une prochaine étude.

**Mots-clés :** préférences en matière de traitement, adhésion, mise en action, exposition, méthode, recherche

## **Introduction**

Adherence to treatment is important in determining the effectiveness of behavioural therapy in producing the hypothesized improvement in outcomes. Less than optimal adherence to treatment has been reported in a meta-analysis; specifically, 40% of patients took the medications as prescribed but twice as many did not adhere to the recommended health behaviours related to diet, exercise, and smoking. Furthermore, the odds of a good outcome are three times higher in adherent compared to non-adherent patients (DiMatteo, Giordani, Lepper, & Croghan, 2002). Similarly, a meta-analysis of psychological interventions for pathological gambling (Pallesen et al., 2003) found that attendance at a large number of planned sessions was associated with improved outcomes.

Several factors have been identified as predictors of adherence to treatment. The factors are categorized into (1) characteristics of participants, such as age, beliefs, and lifestyle; (2) characteristics of the clinical problem experienced by participants, including its chronicity and perceived severity; and (3) characteristics of the treatment, such as invasiveness and complexity (Brawley & Culos-Reid, 2000; Martin, Bowen, Dunbar-Jacob, & Perri, 2000). Treatment preferences have been recognized as factors influencing adherence (TenHave, Coyne, Salzer, & Katz, 2003). Allocation of participants to their preferred treatment has been proposed as a way to mitigate this influence (Corrigan & Salzer, 2003; Rowe et al., 2005).

This methodological study investigated the extent to which assigning participants to their treatment of choice, as compared to randomizing them to treatment, enhances adherence to behavioural therapy in the context of an intervention evaluation trial.

### ***Treatment Adherence***

Generally, adherence refers to patients' involvement in treatment activities (Wilbur, Chandler, & Miller, 2001). Involvement in behavioural therapy encompasses exposure to and enactment of treatment (Borrelli et al., 2005; Burgio et al., 2001). *Exposure* relates to attendance at the planned treatment sessions during which the treatment recommendations or strategies to change the target behaviour are discussed and practised. *Enactment* is application of the treatment recommendations in the context of daily life between treatment sessions. Appropriate and consistent application of treatment recommendations contributes to behavioural change and subsequent outcome achievement. Deviations in participants' implementation of the treatment reduce the potency of the intervention and increase variability in post-treatment outcomes. This increased variability

translates into increased error variance and lowers the statistical power to detect significant treatment effects (Gibson, 2003).

### ***Treatment Preferences***

The phrase “treatment preferences” refers to participants’ choice of therapy — that is, the treatment they desire for the management of the presenting clinical problem (Sidani, Epstein, Bootzin, Moritz, & Miranda, 2009). Individuals enrolled in a trial may have preferences for the treatments under evaluation. The preferences are generated from previous knowledge and experience and from information about the treatments disclosed during the process of obtaining consent (Sidani & Braden, 2011).

In randomized trials, participants’ preferences are ignored when participants are randomly assigned to treatment. Randomization creates two subgroups. The first comprises those who, by chance, are assigned to their preferred treatment. These participants may develop enthusiasm for, engage in, and adhere to treatment. In contrast, the second subgroup consists of participants who receive the non-preferred treatment. They may be disappointed because they are deprived of their treatment of choice. They may lose their motivation for the treatment and may not initiate, engage in, and adhere to the allocated treatment. Ultimately, they achieve poor outcomes, which contributes to misleading conclusions about treatment effectiveness (Howard & Thornicroft, 2006; Huibers et al., 2004; Preference Collaborative Review Group, 2009).

Preference trials are intended to address treatment preferences that threaten the validity of conclusions in intervention research. In this design, participants are given information about the treatments, requested to indicate their treatment of choice, and allocated to their chosen treatment. Participants with no preference are randomized to treatment (Bradley, 1993). Provision of the preferred treatment is believed to enhance engagement in and adherence to treatment (Leykin et al., 2007).

### ***Influence of Treatment Preferences on Adherence***

A total of 10 studies evaluated the influence of allocating participants to the preferred treatment on adherence to treatment. The studies involved different treatments for the management of various clinical problems. Their results were inconsistent in supporting the utility of preference-based allocation in improving adherence to treatment. The same conclusion was reached in two systematic reviews of studies that examined the influence of preferences on adherence to treatment for depression (Gelhorn, Sexton, & Classi, 2011; Winter & Barber, 2013).

Of the 10 studies, the results of three showed no statistically significant difference in attendance at treatment sessions between participants

allocated to the preferred and non-preferred therapy for the management of diabetes (Hitchcock Noël et al., 1998) and depression (Dobscha, Corson, & Gerrity, 2007; Mergl et al., 2011). The remaining seven studies reported higher levels of adherence among participants receiving treatments that were congruent with their preferences. The treatments included pharmacological, educational, and behavioural therapies for depression (Bedi et al., 2000; Chilvers et al., 2001; Hunot, Horne, Leese, & Churchill, 2007; Kwan, Dimidjian, & Rizvi, 2010; Raue, Schulberg, Heo, Klimstra, & Bruce, 2009) and for the management of heart disease (Janevic et al., 2003) and mental health problems (Macias et al., 2005).

In the above studies, adherence was operationalized as exposure to treatment — that is, attendance at the planned sessions for non-pharmacological therapy. The studies reviewed did not examine the extent to which accounting for treatment preferences influences the enactment of therapy. Also, the evidence regarding the contribution of preference-based allocation to treatment enactment is limited.

Since enactment is another critical aspect of adherence, the overall purpose of this study was to determine the extent to which the method of allocation (random vs. preference) in a treatment evaluation trial influences adherence as operationalized by both exposure to and enactment of the behavioural intervention.

## **Methods**

### ***Design***

The data for the study were obtained from a large methodological trial examining the utility of different research designs in enhancing the validity and clinical relevance of findings related to the effectiveness of behavioural interventions (Sidani, Epstein, Bootzin, Moritz, & Sechrest, 2007). The data set pertained to those who were assigned to the same behavioural therapy for the management of chronic insomnia on the basis of chance or preference, the purpose being to control for possible variability in adherence to different treatments and hence to examine the unique influence of allocation method (random vs. preference) on adherence. Assignment to treatment took place after eligible consenting individuals provided pre-test data. One group of participants were randomly assigned to the behavioural therapy using opaque, sealed envelopes that were opened in their presence to reveal the allocated treatment. The other group were assigned to the preferred treatment, which was determined in a systematic process: the participants were provided information about the treatments under investigation, asked to rate the acceptability of these treatments, and asked to indicate their treatment of choice (for details, see Sidani et al., 2009). Differences between the two groups in terms of per-

sonal and clinical characteristics were examined and controlled for statistically, to minimize their potentially confounding influence on treatment adherence.

### **Sample**

Participants were eligible for the methodological trial if they were 21 years of age or older, were non-institutionalized, were English-speaking, and complained of chronic insomnia described as difficulty falling or staying asleep of at least 30 minutes per night for 3 or more nights per week ascertained by means of a 14-day sleep diary and at least 3 months' duration as reported by participants. Individuals were excluded if they had sleep apnea (self-reported) and were under treatment for sleep apnea, cognitive impairment (score < 27 on the Mini-Mental State Exam; Folstein, Folstein, & McHugh, 1975), or psychological impairment (Global Severity Index T score > 50 on the Brief Symptom Inventory; Derogatis & Melisaratos, 1983).

The study comprised 262 participants assigned to the same behavioural therapy. This sample size was adequate to detect small-moderate differences in adherence (exposure and enactment) between participants allocated by chance or preference, setting beta at .80 and  $p$  at  $\leq .05$  (Cohen, 1992).

### **Intervention**

The intervention was behavioural therapy for the management of chronic insomnia. It consisted of two components. The first provided information about sleep, factors contributing to insomnia, and recommendations for promoting sleep. The second offered support to participants in applying the recommendations by discussing barriers to their implementation in daily life and assistance with generating ways to overcome the barriers. Two categories of recommendation, general and specific, were presented. The general recommendations consisted of strategies to use during the day or evening to promote good sleep, such as engaging in physical activity during the day and avoiding caffeine and nicotine in the evening. The specific recommendations included instructions to go to bed only when sleepy, avoid any non-sleep-related activities in bed, get out of bed if unable to fall asleep or get back to sleep within 15 to 20 minutes, wake up at the same time every day, and avoid naps, or, if needed, take a nap, in bed, for no more than 30 minutes (Bootzin & Epstein, 2011). The intervention was given in four face-to-face group sessions of 60 to 90 minutes' duration and two individual telephone sessions that lasted 15 to 20 minutes. The six sessions were offered once a week over a 6-week period.



### ***Variables and Measures***

***Personal characteristics.*** Standard items were used to assess participants' age, gender, education level, race, marital status, and employment status. Education level was quantified with the number of years of formal schooling. Race and marital and employment status were represented in the following categories: white versus non-white, married versus non-married, and employed versus non-employed.

***Clinical characteristics.*** Participants indicated the types of insomnia they experienced: difficulty falling asleep and/or difficulty staying asleep and the length of time they had had insomnia. The severity of insomnia was assessed using the sleep diary and the Insomnia Severity Index (ISI). Participants completed the sleep diary for 14 days prior to receiving the therapy. They reported the daily values upon waking to a voice-mail service, to minimize recall bias. The sleep parameters, computed from relevant diary data, included (1) sleep onset latency, or the length of time, in minutes, to fall asleep; (2) wake after sleep onset, or length of time, in minutes, awake across all awakenings; and (3) sleep efficiency, or the percentage of the time in bed actually asleep. The sleep diary demonstrated test-retest reliability ( $r = .69-.93$ ) and validity, evidenced by significant correlation between the values of the respective sleep parameters estimated with data reported in the sleep diary and recorded using actigraphy (Buysse, Ancoli-Israeli, Edinger, Lichstein, & Morin, 2006). The ISI was administered at pre-test to assess participants' perceived severity of their sleep problem. The ISI contains seven items measuring the nature, severity, and impact of insomnia. It has excellent internal consistency reliability (Cronbach's  $\alpha \geq .90$ ) and construct validity (Morin, Belleville, Bélanger, & Ivers, 2011).

***Adherence.*** The two aspects of adherence, exposure to and enactment of treatment, were examined in this study. *Exposure* was assessed via attendance at group and telephone sessions. The therapists leading the sessions recorded the presence or absence of each participant at each scheduled treatment session and the total number of sessions attended was counted. *Enactment* of treatment recommendations was reflected in three ways. *First*, participants reported on their application of the following behavioural therapy recommendations: (1) using the bed for sleep only and not for any other activity, (2) getting out of bed when unable to fall asleep or fall back to sleep within 15 to 20 minutes, and (3) taking a nap in bed only if necessary. Three items related to the application of these recommendations were integrated into the sleep diary form, and therefore were completed by the participants daily throughout the 6 weeks of treatment. Enactment of these recommendations was quantified in two ways: the number of days, within each treatment week, that each recommendation

was applied; and the number of days that its application was not needed (e.g., the participant did not take a nap). *Second*, participants were requested to indicate whether or not, within 1 week following completion of treatment, they implemented the strategies to promote sleep that represented the sleep hygiene recommendations. The 13 recommendations were as follows: engaging in physical activity during the day, avoiding vigorous exercise around bedtime, reducing noise in the bedroom, reducing light in the bedroom, maintaining a comfortable temperature in the bedroom, avoiding a heavy evening meal, having a light bedtime snack as needed, avoiding alcohol before bed, avoiding caffeine before bed, avoiding nicotine before bed, putting the clock out of sight in the bedroom, avoiding long naps, and avoiding stressful thoughts when in bed. The total number of strategies applied was computed. *Third*, an additional item was used to measure overall compliance with the treatment within 1 week after treatment completion. Overall compliance was rated on a five-point scale ranging from *not at all* (0) to *very much* (4).

### ***Data Analysis***

Descriptive statistics (frequency, mean, standard deviation) were used to characterize the sample relative to personal and clinical characteristics. Independent samples *t* test for continuous variables and chi-square test for dichotomous variables were used to examine differences in these characteristics between the two groups of participants: those randomized (random group) and those allocated on the basis of preference (preference group) to the behavioural therapy. Baseline variables that showed differences between the random and preference groups were considered as covariates in subsequent analyses and were controlled for statistically. One-way analysis of covariance was used to compare the two groups on the number of sessions attended (exposure), number of sleep hygiene recommendations applied (enactment), and self-reported overall compliance with treatment (enactment), while controlling for the covariates. Repeated measures analysis of covariance was done to determine differences in the application (enactment) of the behavioural therapy recommendations between the groups across the 6 weeks of treatment, while controlling for baseline differences. Statistically significant main group effect and interaction (group x time) effects supported the influence of treatment allocation method on adherence.

## **Results**

### ***Personal Characteristics of Participants***

Participants ranged in age from 21 to 90 years with a mean of 56 ( $\pm$  16.1) years. The sample comprised slightly more women (59.5%) than

men (40.5%). Education level varied from 3 to 30 years of formal schooling with a mean of 15.7 years ( $\pm 3.5$ ). Approximately half of the participants were married (53.3%) and approximately half were employed (55.6%). The majority (90%) were white.

### ***Clinical Characteristics of Participants***

The majority of participants indicated that they experienced difficulty falling asleep (72.5%) and difficulty staying asleep (91.5%). On average, they had had insomnia for 11 ( $\pm 11$ ) years. The mean values of the sleep parameters obtained with the sleep diary were as follows: 42.9 minutes for sleep onset latency ( $\pm 30.7$ , range: 1.9 to 235.5); 54.4 minutes for wake after sleep onset ( $\pm 33.9$ , range: 0 to 201.5); and 69.9% for sleep efficiency ( $\pm 10.4$ , range: 27 to 90). The mean score for perceived insomnia severity was 17.6 ( $\pm 3.9$ , range: 8 to 28), indicating that, on average, participants experienced clinical insomnia of moderate severity.

### ***Method of Treatment Allocation***

Of the 262 participants, 164 (63.1) were randomized to the behavioural therapy and 96 (36.9%) were allocated to this treatment based on their preference.

### ***Group Comparison on Personal and Clinical Characteristics***

The average values on the personal and clinical characteristics for the random and preference groups are presented in Table 1. Statistically significant differences were observed for three characteristics: age,  $t(257) = 4.6, p = .001$ ; gender,  $\chi^2(1) = 9.2, p = .004$ ; and employment status,  $\chi^2(1) = 18.9, p = .001$ . Those in the preference group were young, employed women. Since age and employment status were related (older participants were not employed), only age and gender were considered covariates in subsequent group comparisons to determine the influence of method of allocation to treatment on adherence.

### ***Group Comparison on Adherence to Treatment***

After controlling for baseline differences in age and gender, there were no statistically significant differences in exposure and enactment between the random and the preference groups. In terms of exposure, the mean number of sessions attended was 5.7 ( $\pm .54$ ) in the random group and 5.6 ( $\pm .63$ ) in the preference group,  $F(1, 234) = 2.69, p = .10$ . The adjusted mean values for the enactment of the behavioural therapy recommendations over the 6 weeks of treatment are shown in Table 2. The number of days on which the first recommendation, using the bed for sleep only, was applied was comparable in the two groups,  $F(1, 232) = 1.05, p > .05$ , and over time,  $F(5, 229) = .61, p > .05$ . The number of

**Table 1 Mean (SD) Scores on Baseline Characteristics**

	<b>Random Group (n = 164)</b>	<b>Preference Group (n = 96)</b>
<b>Personal characteristics</b>		
Age (mean)	59.5 (15.4)	50.2 (15.7)
Gender (% female)	52.5	71.6
Education (mean)	15.7 (3.4)	15.6 (3.7)
Marital status (% married)	56.1	48.4
Employment status (% employed)	45.1	72.6
Race (% white)	90.2	89.4
<b>Clinical characteristics</b>		
Type of insomnia		
Difficulty falling asleep (%)	73.2	71.9
Difficulty staying asleep (%)	92.0	91.6
Duration of insomnia (mean)	10.3 (10.7)	12.3 (11.6)
Sleep onset latency (mean)	44.7 (29.8)	40.3 (22.3)
Wake after sleep onset (mean)	57.0 (36.8)	49.8 (27.1)
Sleep efficiency (mean)	69.1 (11.1)	71.2 (9.2)
Insomnia severity (mean)	17.8 (4.0)	17.2 (3.8)

days on which this recommendation was not needed was slightly higher in the preference group,  $F(1, 234) = 3.34, p = .069$ , throughout the treatment period,  $F(5, 230) = 1.35, p > .05$ . Participants in both groups applied the second recommendation, getting out of bed if cannot fall asleep or fall back to sleep within 15 to 20 minutes, with similar frequency,  $F(1, 234) = .65, p > .05$ , which decreased significantly over time,  $F(5, 230) = 5.32, p = .001$ . The number of days on which application of the second recommendation was not needed did not differ between groups,  $F(1, 234) = .85, p > .05$ , or over time,  $F(5, 230) = 1.67, p > .05$ ; however, the preference group reported a slightly higher number of days on which this recommendation was not needed than the random group in the last 4 weeks of treatment. For the third recommendation, taking a nap in bed, there was no statistically significant effect for group,  $F(1, 234) = 2.39, p > .05$ , or time,  $F(5, 230) = .64, p > .05$ , even though the mean values were consistently lower for the preference group as compared to the random group across the 6 weeks of treatment. The number of days on which the third recommendation was not needed did not differ between groups,  $F(1, 234) = .38, p > .05$ , or over time,  $F(5, 232) = .72, p > .05$ .

**Table 2 Between-Group Mean Scores on Enactment of Therapy Recommendations Across Treatment Period**

Recommendation	Week 1		Week 2		Week 3		Week 4		Week 5		Week 6	
	R	P	R	P	R	P	R	P	R	P	R	P
Use bed to sleep only: number of days applied	6.4	6.1	6.4	6.2	6.4	6.1	6.5	6.2	6.4	6.2	6.3	6.4
Use of bed to sleep only: number of days not applicable	.05	.23	.06	.14	.06	.20	.05	.23	.05	.19	.06	.15
Get out of bed: number of days applied	2.0	1.9	1.7	1.7	1.9	1.6	1.7	1.3	1.7	1.5	1.6	1.5
Get out of bed: number of days not applicable	2.3	2.3	2.7	2.8	2.6	3.0	2.9	3.2	2.8	3.3	2.8	3.1
Take nap in bed: number of days applied	1.0	0.7	0.9	0.6	0.9	0.6	0.8	0.6	0.8	0.5	0.7	0.4
Take nap in bed: number of days not applicable	4.9	5.0	4.7	5.0	4.7	4.8	5.0	5.3	5.0	5.1	5.2	5.2

*Legend:* R = Random group; P = Preference group

On average, participants reported applying approximately seven of the 13 sleep hygiene recommendations after treatment completion. The mean number of recommendations applied was 6.8 for the random group and 6.7 for the preference group,  $F(1,234) = .04, p > .05$ . In addition, there was no significant between-group difference in post-treatment self-reported compliance,  $F(1,205) = 1.85, p > .05$ . The mean score was 3.1 for the random group and 2.9 for the preference group, reflecting an above-average level of self-reported compliance with treatment.

## **Discussion**

This study extends previous research investigating the influence of participants' preferences for treatment on adherence to treatment by examining two aspects of adherence: exposure to and enactment of treatment. Exposure was assessed using the number of planned intervention sessions attended by participants, consistent with previous studies. Enactment was indicated by participants' reported application of specific treatment recommendations and overall compliance with treatment, which were not systematically evaluated in previous studies. Overall, exposure to and enactment of behavioural therapy for insomnia were comparable for those who were randomized and those who were allocated to the preferred treatment, even though the latter group showed a slightly higher degree of enactment of the therapy recommendations over the 6 weeks of treatment. The results are consistent with the findings of three studies (Dobscha et al., 2007; Hitchcock Noël et al., 1998; Mergl et al., 2011), indicating that accounting for treatment preferences is not associated with enhanced adherence to treatment. Taken together, these results contradict those found in seven studies (Bedi et al., 2000; Chilvers et al., 2001; Hunot et al., 2007; Janevic et al., 2003; Kwan et al., 2010; Macias et al., 2005; Raue et al., 2009) and imply that participants receiving their preferred treatment exhibit higher levels of adherence than those not allocated to their treatment of choice. There are four possible explanations for the inconsistency of findings related to the contribution of treatment preferences to adherence.

First, differences in the target population and the treatments under evaluation could account for the inconsistent findings. Persons who experience a pervasive and burdensome clinical problem that affects usual functioning and many domains of health, such as insomnia, for a long time may encounter challenges in initiating, engaging in, and adhering to treatment, particularly if the treatment is demanding. For instance, the participants in the present study reported clinical insomnia of moderate severity for an average of 11 years. Chronic insomnia is associated with negative consequences, including daytime fatigue, which impairs physical,

psychological, and social functioning (Bootzin & Epstein, 2011). Thus participants may have felt that they lacked the energy and motivation needed to attend all treatment sessions and apply all treatment recommendations. The implementation of behavioural therapy for insomnia demands much effort to modify the way one approaches sleep, the bedroom environment (e.g., light, noise), general daily habits (e.g., activity, caffeine intake), and sleep habits (e.g., getting out of bed if not asleep, consistent wake-up time) (Epstein & Bootzin, 2002). Participants who were not willing to initiate and maintain these changes, or who desired a “quick fix” for their sleep problem (Epstein, Babcock-Parziale, Haynes, & Herb, 2012), may have withdrawn from the study. Consequently, those who remained in the study and completed post-test measures may represent a biased sample comprising individuals in desperate need of treatment and willing to expend much effort to apply treatment recommendations in order to manage their insomnia. This is suggested in the observed mean values on the indicators of exposure to and enactment of treatment, implying moderate to high levels of attendance at the sessions and application of specific therapy recommendations for both groups (random and preference).

Second, it is possible that participants randomized to the behavioural therapy preferred this treatment. If so, this subgroup of participants received treatment congruent with their preference, which contributed to their initiation of and adherence to treatment. The size of this subgroup may have affected the magnitude of the difference in adherence levels between the random and preference groups (Sidani & Braden, 2011). If a large number of those in the random group received congruent treatment, then they were satisfied with the allocated treatment and were eager to apply and follow its recommendations. Their performance would not differ from that of participants allocated to their preferred treatment, thereby reducing the power to detect significant between-group (random and preference) differences in adherence. This explanation is highly plausible, because 82% of the sample were randomized to the behavioural therapy for which they expressed a preference. Conversely, it is possible that those allocated to behavioural therapy on the basis of their choice were dissatisfied with it. In other words, although members of this subgroup were given the treatment they desired, they may not have evaluated it favourably once exposed to it. For instance, they may have disliked its constituent activities (e.g., group discussion) or may have viewed its specific recommendations (e.g., maintaining a consistent wake-up time) as incongruent with their lifestyle. Accordingly, they would not have attended all treatment sessions or applied all treatment recommendations, as proposed by Huibers et al. (2004) and Kiesler and Auerbach (2006). The association of satisfaction with adherence to

treatment should be investigated in future research on the influence of treatment preferences.

Third, the data on enactment were obtained through self-report. This method of data collection is prone to bias. Specifically, participants may have indicated that they were following the treatment recommendations during the 6 weeks of treatment because they were aware that the researchers and therapists were reviewing their sleep diary, which was necessary to determine the total sleep time to prescribe. Therefore, they may have wanted to draw a positive image of themselves or to please the therapists, which may have resulted in social desirability bias and an overestimate of their adherence level. The participants' post-treatment self-report regarding application of the recommendations and overall compliance with treatment could be tainted by recall bias, in addition to social desirability bias. Other means of assessing enactment of treatment, such as report by objective measures and participants' significant other or bed partner, should be explored to improve the validity of adherence measurement. Further, measurement of enactment could be expanded to capture the extent to which the treatment recommendations are applied correctly, as suggested by Borrelli et al. (2005), and there is still a need to apply these throughout the treatment period, as was done in this study. The correct and consistent application of insomnia treatment recommendations, such as getting out of bed if one cannot fall asleep or fall back to sleep, yields improvement in the early weeks of treatment; this improvement, manifested in consolidated sleep, reduces the need to apply the treatment recommendation. This may explain the study findings indicating that participants who received the behavioural therapy on the basis of preference, as compared to those who were randomized, reported a slightly higher number of days on which the recommendations were not needed.

Last, the persons with insomnia who participated in the study could represent a selective subgroup of the target population. It is possible that non-adherent participants withdrew from the study for various reasons, including dislike of using the diary (capturing sleep and treatment enactment data). Therefore, participants in the study could be characterized as adherent.

The findings suggest that allocation of participants to the treatment of choice may not significantly enhance exposure to behavioural therapy for the management of chronic insomnia, but may contribute to slightly higher enactment of treatment recommendations early in the treatment period. Also, the results highlight areas for further investigation to elucidate the contribution of preferences to adherence. It is important to assess participants' evaluation of and satisfaction with the allocated treatment, in the random and preference groups, and to determine the extent to



which satisfaction influences adherence to treatment directly or mediates the relationship between treatment preferences and adherence, as suggested by Bradley (1993). The validity of the findings on adherence would be greatly enhanced if the concept's key aspects, exposure and enactment, were to be measured using a mix of self-report, reports of others, and objective measures. In addition, a comprehensive assessment of enactment should cover not only the application of the recommendations pertaining to each treatment component, but also the extent to which the recommendations were applied correctly.

### Conclusions

The results of this study show that accounting for preferences in allocating participants to treatment, in the context of a trial, does not significantly improve adherence to treatment. Conceptual and methodological factors may have contributed to the findings and should be further investigated to elucidate the mechanism through which preferences affect exposure to and enactment of treatment.

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## Adherence and Preference

Souraya Sidani, Richard R. Bootzin, Dana R. Epstein, Joyal Miranda, Jennifer Cousins

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## *Adherence and Preference*

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# Pilot Testing of a Psycho-educational Telephone Intervention for Women Receiving Uninformative BRCA1/2 Genetic Test Results

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Evidence suggests that women who receive uninformative results for breast and ovarian cancer (BRCA1/2) gene mutations may experience as much distress as women whose results indicate the presence of a gene mutation. No intervention to reduce distress after receipt of uninformative results has yet been tested. The purpose of this study was to test the feasibility and preliminary effects of a psycho-educational telephone (PET) intervention to reduce distress in women who receive uninformative BRCA1/2 results. A single group with repeated measures was used to assess the impact of the intervention on 72 such women. After receiving the results, most of the women continued to feel uncertain about their carrier genetic status. However, their distress significantly decreased between receipt of uninformative results and 3 months post-intervention ( $p = 0.01$ ). The preliminary findings suggest that a PET uncertainty intervention is clinically feasible and may reduce the distress of receiving uninformative results.

**Keywords:** breast cancer, BRCA1/2, genetic testing, uninformative results, pilot intervention study

Résumé

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**Essai pilote d'une intervention  
psycho-éducative par téléphone pour  
les femmes ayant reçu des résultats  
non concluants après des tests de dépistage  
génétique concernant BRCA1 et BRCA2**

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Les données permettent de croire que les femmes qui obtiennent des résultats non concluants à la suite de tests de dépistage d'une mutation des gènes liés aux cancers du sein et des ovaires (BRCA1 et BRCA2) sont susceptibles d'éprouver une détresse aussi importante que celles dont les résultats indiquent la présence d'une mutation génétique. Aucune intervention visant à atténuer le sentiment de détresse après réception de résultats non concluants n'a encore été mise à l'essai. L'objectif de la présente étude est d'évaluer la faisabilité et les effets préliminaires d'une intervention consistant en un appel psycho-éducatif destiné à réduire la détresse de femmes ayant reçu des résultats de dépistage génétique non concluants concernant les gènes BRCA1 et BRCA2. Un groupe unique sondé à plusieurs reprises a été étudié afin d'évaluer l'effet d'une telle intervention sur 72 femmes. Après la réception de leurs résultats, la plupart éprouvaient toujours de l'incertitude concernant leur statut de porteuse ou non d'une mutation génétique. Toutefois, une diminution considérable de leur détresse a été observée entre la réception des résultats non concluants et une période de trois mois après l'intervention par téléphone ( $p = 0,01$ ). Les constatations préliminaires donnent à penser qu'une intervention psycho-éducative par téléphone à propos de l'incertitude est réalisable et permet de réduire la détresse des femmes dont les résultats sont non concluants.

**Mots-clés :** cancer du sein, BRCA 1/2, dépistage génétique, résultats non concluants, essai pilote d'une intervention

## Introduction

Genetic testing is becoming increasingly more available for mutations of two genes, BRCA1 and BRCA2 (BRCA1/2), that place carriers at increased risk for breast and ovarian cancer. Members of families with a history of breast or ovarian cancer (i.e., affected individuals) who undergo testing for BRCA1/2 can receive four possible test results: (1) *BRCA positive*: carrying a pathogenic familial genetic mutation; (2) *BRCA negative/true negative*: not carrying a familial pathogenic mutation; (3) *BRCA variant of uncertain significance (VUS)*: identified gene mutation with an unknown effect; or (4) *BRCA uninformative*: absence of known familial mutation despite striking but unexplained personal and/or family history of cancer (Culver et al., 2013; Leblond et al., 2011). Most individuals who are tested (75% or more) receive uninformative results (Culver et al., 2013; Schwartz et al., 2004). Current evidence suggests that individuals who receive such results exhibit distress levels similar to those of mutation carriers who test positive (van Dijk et al., 2006). Further, it appears that the distress among individuals who receive uninformative results does not follow the descending trend observed in the recipients of true-negative test results (Schwartz et al., 2002).

Lack of closure and relief after receiving uninformative BRCA1/2 test results has been observed in studies with this population (Ardern-Jones, Kenen, Lynch, Doherty, & Eeles, 2010; Dorval et al., 2005; Maheu & Thorne, 2008) and may explain partially the continued distress experienced by these individuals. The ambiguous nature of their true cancer risk in light of their uninformative BRCA1/2 result may further generate this distress (Maheu & Thorne, 2008). There are also the added cancer risk perception and personal and family cancer experiences that are closely correlated with emotional distress (Esplen et al., 2000; Leblond et al., 2011). In spite of these findings, studies tend to suggest that most individuals undergoing genetic testing for BRCA1/2 experience little clinical distress regardless of test result outcomes (Leblond et al., 2011; Meiser, 2005; O'Neill et al., 2009). However, we need more long-term studies on the psychological and behavioural impact of BCRA1/2 genetic test results (O'Neill et al., 2009; van Dijk et al., 2006; van Oostrom et al., 2003).

In a previous study (Maheu & Thorne, 2008), 17 of 21 women from families at high risk for breast cancer and with a previous breast cancer diagnosis (affected) interpreted their uninformative results as having a mutation that genetic testing missed, leaving them feeling distressed and in a state of limbo. Given the overexpressed cancer patterns in their families, these women were doubtful about the validity of their test results. This finding suggests the need for an intervention to reduce uncertainty

by providing support from health professionals so that women can more accurately interpret their uninformative genetic test results and experience less distress.

We designed an intervention based on Mishel's (1988) illness uncertainty theory, according to which uncertainty and distress are decreased if a frame of reference is provided, along with relevant information and organized and accessible support (Gil et al., 2006). Other means of reducing uncertainty include effective coping strategies and formal and informal social support, such as counselling by health professionals (Gil et al., 2006). Earlier research suggests that events in the cancer experience, such as testing for a gene mutation, may threaten personal perceptions of control and illness uncertainty (Stiegelis et al., 2004). Providing a psycho-educational telephone (PET) intervention, in addition to standard genetic counselling, has been found to reduce the distress and anxiety of mutation carriers in the short term (Graves et al., 2010). No such intervention has been tested with affected women who receive uninformative BRCA1/2 test results. This article reports on a pilot intervention aimed at reducing distress among women with a personal and family history of breast or ovarian cancer who received uninformative BRCA1/2 test results. As no psycho-educational intervention has been evaluated for women who receive ambiguous test results, we considered a feasibility and acceptability pilot study with this design to be the most appropriate approach before proceeding to a full clinical trial (Feeley et al., 2009). The primary aim of the study was to test the feasibility, acceptability, and preliminary effects of a standardized PET intervention for this group of women. The intervention was standardized through the development of a detailed manual created for this study. We hypothesized that our PET intervention would reduce distress in women receiving uninformative BRCA1/2 results. Our secondary aim was to identify predictors of distress among the women sampled in this study.

## **Methods**

### ***Study Design***

The intervention and its evaluation took place in a hereditary cancer program (HCP) at North York General Hospital in Toronto, Canada. We used a single group with repeated measures whereby participants completed questionnaires at four time points: while waiting to receive the BRCA1/2 test results following their usual care genetic counselling session (T1), immediately after receiving uninformative results (T2), and at 3 months (T3) and 1 year (T4) post-intervention. All questionnaires, along with addressed, stamped return envelopes, were mailed to participants and completed by them at home.



Usual care at the HCP consists of initial genetic counselling for all individuals who qualify for BRCA1/2 testing, followed by a second counselling visit to discuss the results. The initial counselling includes general explanations of genetic inheritance and the implications of positive, negative, or uninformative results. In the study, the two-step PET intervention began following the receipt of T2 questionnaires, which were completed immediately after receipt of the BRCA1/2 results. In step 1, an information booklet and a relaxation compact disc (CD) were given to each woman at her second genetic counselling appointment, when she received her genetic test result. In step 2, 1 month after each woman received her result, telephone follow-up care was provided. The telephone follow-up care represented step 2 of the PET intervention and was delivered by one of the two genetic counsellors from the HCP, who also provided the usual genetic counselling care to the women enrolled in the study.

### ***Study Population***

Between 2007 and 2012, women scheduled to receive BRCA1/2 results were approached through the HCP. Those who met four inclusion criteria were given a package inviting them to enter the study: (1) a breast cancer (BC) diagnosis (affected women), (2) a significant family history of BC, (3) a scheduled appointment to receive their test result, and (4) ability to understand and read English. Excluded from the study were women who had an identified BRCA1/2 mutation in the family at the study's beginning or received notice of one during the course of the study. The final sample comprised 68 women.

### ***Measures: Outcome Variables***

The baseline questionnaire asked participants for basic demographic and lifestyle data (e.g., alcohol intake, smoking habits, and health/lifestyle behaviours, such as making changes to improve weight, diet, and exercise). Psychosocial functioning was assessed using measures of cancer-specific distress (T1–T4), genetic-testing distress (T2–T4), risk perception (T1–T4), and interpretation of uninformative results (T2–T4).

### ***Feasibility and Acceptability***

Participant retention, satisfaction with intervention (i.e., proportion who completed the PET intervention and who voiced satisfaction with the intervention), and completion of study measures were monitored as indicators of feasibility and acceptability of both the intervention and the study methods.

### ***Cancer-Specific Distress***

To assess our primary outcome of distress from undergoing genetic testing, the Impact of Event Scale (IES) was used (Horowitz, Wilner, & Alvarez, 1979). The IES has been extensively used to measure distress among women with BC (Appleton et al., 2004; Esplen et al., 2000), with good internal consistency for the total score and subscale scores (Cronbach's  $\alpha = 0.91, 0.88,$  and  $0.84$  for the total scale, intrusion and avoidance, respectively) (Thewes, Meiser, & Hickie, 2001). The IES is a 15-item questionnaire rated on a four-point Likert scale (0, 1, 3, 5), with two subscales that measure intrusive thoughts (7 questions; score: 0–35) and avoidance of certain thoughts, feelings, or situations (8 questions; score: 0–40). The total IES score combines the two subscales, for a possible score of 0 to 75. Although no clinical cut-off has been validated, total scores over 27 indicate risk of post-traumatic stress disorder (PTSD) and scores over 35 a probable diagnosis of PTSD (Reed, 2007). Participants' IES total scores were obtained at T1 to T4. Cronbach's alpha for the IES score at T1 was 0.89.

### ***Genetic-Testing Distress***

To measure the impact of result disclosure after genetic testing, the Multidimensional Impact of Cancer Risk Assessment (MICRA) questionnaire was used. MICRA comprises 25 items that incorporate three subscales measuring distress associated with genetic test results (6 items), uncertainty associated with test results and future plans (9 items), and positive experiences with genetic testing (4 items). A total score is built from these three subscales, with two additional global items and four conditional items, to produce a score ranging from 0 to 125. Acceptable reliability of the total score and the three subscales has been reported in a sample of women with BRCA1/2 test results (Cronbach's  $\alpha = .84$  [Graves et al., 2010]) for total score, .87 for distress, .84 for uncertainty, and .82 for positive experience (Halbert et al., 2011). Unlike other standardized measures of psychological distress, MICRA was specifically developed to measure distress associated with disclosure of genetic test results (Cella et al., 2002). Consequently, MICRA was administered following result disclosure such as at T2, T3, and T4. Cronbach's alpha for the MICRA total score at T2 was 0.90.

### ***Risk Perception***

To examine risk perception, we asked each woman to estimate her personal risk of BC compared with women of a similar age, using a five-point Likert scale (1 = *much less likely*; 5 = *much more likely*) at T1 to T4. We also asked each woman to rate her perceived risk of carrying an

inherited mutation on an eight-point Likert scale (1 = *non-existent*; 8 = *very high*) at T1 to T4. Cronbach's alpha for the scale was calculated from the four time points as the scale has only one item. Cronbach's alpha was greater than 0.83.

### ***Interpretation of Uninformative Results***

We asked participants how they interpreted their uninformative results at T2 to T4 using the following four lay-interpretation options (Maheu & Thorne, 2008): (1) I am certain that **I have** an inherited mutation; (2) I am certain that **I have** an inherited mutation, **but** the current testing procedures could not detect it; (3) I think that **I may or may not have** an inherited mutation, but the current testing procedures could not detect it; and (4) I am certain that **I do not have** an inherited mutation. The third option represents the proper medical interpretation of uninformative results, in that they do not exclude the possibility of an inherited mutation. This interpretation produces uncertainty for recipients of test results (Leblond et al., 2011; Maheu & Thorne, 2008).

### ***Psycho-educational Telephone Intervention***

The two-step PET intervention provided cognitive and behavioural coping strategies to help women understand and manage complex genetic information stemming from uninformative test results. The intervention was begun after participants completed the T2 questionnaire.

**Step 1** consisted of (1) a 33-page information booklet to address participants' need for knowledge about cancer genetics and to clarify information they received in their genetic counselling sessions, and (2) a relaxation CD to help them manage their anxiety. The booklet, modelled on Stiegelis et al.'s (2004) study, contained three levels of information. Level 1 consisted of facts about BC risk, genetic testing for BRCA1/2, and clarifying information to help women interpret their genetic test results. Level 2 comprised relaxation strategies and techniques based on cognitive-behavioural therapy, including guided imagery from the relaxation CD, calming self-talk phrases, and coping strategies drawn from an uncertainty self-management intervention (Gil et al., 2006). Level 3 contained stories, drawn from a previous study (Maheu & Thorne, 2008) co-led by the first author, about other women who received uninformative BRCA1/2 test results and had to make sense of them. According to uncertainty theory (Mishel, 1988), stories about others who went through a similar experience facilitates event congruency, thus reducing uncertainty. As with Stiegelis et al.'s (2004) study, only positive stories of receiving uninformative BRCA1/2 test results were included.

The level 2 information mirrored another uncertainty intervention study (Gil et al., 2006) by recommending the use of calming self-talk

phrases whenever uncertainty triggered negative thoughts; for example, “While I am scared, I can cope. I have a health care team who looks after me.” Use of this coping strategy can decrease anxiety by reframing intrusive negative thoughts with more comforting thoughts. Use of the relaxation CD was also recommended for when uncertainty triggered negative thoughts. The booklet recommended that participants play the CD at least once a day for 21 days.

**Step 2** consisted of a telephone follow-up care session lasting 5 to 15 minutes and taking place 1 month after the women received their uninformative test results. All the women were contacted by one of the two counsellors who provided usual care for the initial genetic counselling and issuing of results. The goal of step 2 was to answer any questions the women might have and to address any misinformation or confusion concerning their test results. According to illness uncertainty theory (Mishel, 1988), contact with a trusting, caring, credible authority can reduce overall uncertainty. Both of the genetic counsellors conducting the follow-up care session used a two-step guide: (1) review the woman’s understanding of the test results, the booklet, and the relaxation CD; and (2) ask whether she used the CD and the coping strategies suggested in the booklet. Two weeks before the session, a research assistant contacted the women to remind them to review the booklet and practise some of the relaxation exercises and also to schedule the session.

### **Sample Size**

To achieve 80% power to detect a same-group difference of 2.7, with a SD of 9, from pre- and post-intervention IES scores at alpha 0.05, we estimated a sample size of 68. This calculation was based on change in total IES observed in a previous randomized controlled trial (RCT) group intervention for women at high risk for BC (Lerman et al., 1996). However, we could have considered a smaller sample size, since the study on which we based our calculation was an RCT.

### **Analyses**

Descriptive analyses were carried out, including distributions, means, and standard deviations on all demographic, lifestyle, and outcome data using the software SAS 9.3 and using 0.05 as the criterion for statistical significance. Using multivariate generalized estimating equation (GEE) modelling, data collected at T1, T2, T3, and T4 were analyzed for changes in distress levels between the different time points. Correlation analyses were also conducted, to assess relationships between potential predictors and distress measures.

Predictors of distress (IES) and impact of receiving test results (MICRA) were assessed using the multivariate regression models under

the GEE framework that can accommodate correlations among repeated participant measures. Potential predictors entered into the regression model were perceived BC risk, perceived inherited risk, age, education, employment status, and interpretation of test results. In our analysis, the missing data occurred at various time points. In order to assess the missing mechanism, we performed Little's (1988) MCAR test using R package "BaylorEdPsych." The test yields an insignificant  $p$  value of 0.5755, which indicates that the missing data occurred completely at random. As the missing proportion was above 5%, we performed multiple imputations to obtain multiple completed data sets using R package "mi" (Su, Gelman, Hill, & Yajima, 2011). We generated three imputed complete data sets and ran GEE on each data set and pooled the three sets of analyses into a single analysis using multiple imputation principles (Rubin, 1987).

## **Results**

### ***Intervention Acceptability and Feasibility***

For the study, we approached 80 women, 75 of whom agreed to participate. Of these 75 women, 68 (91%) returned their T1 questionnaire. Three women tested positive and 72 received uninformative BRCA1/2 test results. Of the 72 who received uninformative results, 43 returned their T2 questionnaire. All 43 completed the intervention (received the booklet/CD and telephone follow-up), and 34 of those (79%) returned their T3 questionnaire. Finally, 33 of the 34 (97%) returned their T4 questionnaire. Overall, the retention rate at T2 was 60%; higher retention rates were observed at T3 and T4 (79%, or 34 out of 43, and 97%, or 33 out of 34, respectively).

Loss to follow-up was observed mainly at T2, when 40% of eligible women did not return their questionnaire package. The main reasons women gave for dropping out were not enough time to complete the questionnaire, feeling too ill, or changes in personal life. Overall, we recruited 75 women, meeting our sample-size requirement to obtain statistical power in this study. Dropouts occurred at different time points, leaving our sample smaller than the initial target of 68. However, even with the smaller sample, the study produced some significant results. This indicates that the actual effect size was larger than the speculated one in our preliminary sample calculation.

At the end of the telephone follow-up session, women who completed the intervention (i.e., booklet, relaxation CD, and telephone follow-up) were invited to comment on their overall acceptability of and satisfaction with the intervention, including use of the booklet and CD.

Of the 43 women who completed the intervention, 33 agreed to provide verbal feedback. Those who declined said they lacked time to extend the session beyond the telephone follow-up care review. Regarding the booklet, all 33 women had read it at least once and 10 had read it more than once. The women felt that the booklet's content was understandable and did not cover too much information but did cover their main areas of concern, such as how to interpret uninformative BRCA1/2 test results. Regarding coping skills such as calming self-talk and listening to the relaxation CD, although more than 80% of the women enjoyed learning and using the calming self-talk phrases to help them relax, they preferred the relaxation CD; however, they acknowledged that calming self-talk was more immediately accessible when they encountered anxiety triggers related to their state of uncertainty. While we recommended *daily* use of the relaxation CD, 80% of the 33 women said that they had used the relaxation CD at least once during the previous week and calming self-talk phrases at least three times during the previous week.

We asked the women to rate how much more relaxed they felt after using calming self-talk phrases and the relaxation CD on a scale from 0 to 10, with 0 being *not at all*. With the use of calming self-talk phrases, close to 73% of the women rated feeling more relaxed at 6 or above; with the use of the relaxation CD, close to 82% rated their increased relaxation at 7 or above. Overall, the women expressed gratitude for the additional opportunity to review their interpretation of the test results with a genetic counsellor via the telephone intervention.

### ***Psychosocial Variables***

We conducted independent sample *t* tests to compare participants who returned their questionnaire and those who did not at the four time points. The *t* tests did not reveal any differences in demographic characteristics or distress scores, as measured by total IES.

### ***Changes in Psychosocial Variables***

Over time, there were some significant decreases in distress following the intervention. Multivariate GEE modelling was conducted on the IES total (the study primary outcome) and the MICRA total score. Baseline scores for IES and MICRA were controlled in the GEE regression analysis. We found one significant effect between T2 and T3 (the intervention was administered after T2 and the first post-intervention measurement was taken at 3 months post-intervention, at T3) for IES total score ( $z$  value:  $-0.7094$ ;  $p = 0.01$ ), indicating a decrease in distress. Scores for IES were stable between T3 and T4 (12 months post-intervention), suggesting that any intervention effect was maintained ( $z$  value:  $-0.35$ ;  $p = 0.33$ ).

Mean IES total score for each time point measure were T1 = 24.08 (18); T2 = 21.74 (18); T3 = 16.39 (15); T4 = 19.25 (18). With the MICRA scale measuring impact of genetic testing, no significant differences were noted over time, including on the MICRA distress subscale.

From pre- to post-intervention (T1–T3), there was a statistically significant decrease in the percentage of women with a total IES score indicative of increased risk of PTSD ( $p = 0.005$ ). Specifically, 38% fewer women had a score of 27 or higher (T1,  $n = 25$ ; T3,  $n = 6$ ). Of these, 53% fewer had a score of 35 or higher (T1,  $n = 15$ ; T3,  $n = 5$ ). This evidence of reduced distress is encouraging, considering that previous descriptive studies found that distress among women affected by cancer tended to remain elevated over time when left untreated (Carlson et al., 2004).

### ***Personal Risk Estimates***

Paired  $t$  tests were conducted on women's personal ratings of their risk of developing BC and carrying a gene mutation. There was a statistically significant decrease in mean perceived mutation-carrying risk between T1 and T2 ( $p = 0.005$ ). This result suggests that women interpreted their uninformative results as not carrying an inherited breast and ovarian cancer gene mutation. However, no main effect over time was noted, which suggests that the intervention did not influence women's perceived risk of carrying a mutation. Following receipt of genetic test results, perceived risk of developing BC remained unchanged between T1 and T4.

### ***Participants' Interpretations of Their Test Results***

After receiving their test results (T2), most of the women (27 of 43) still felt ambiguous about their mutation-carrier status (options 2 and 3). Few (3 of 43) interpreted their results to mean that they were carriers (option 1), while the rest (13 of 43) felt that they were definitely not carriers (option 4). Between T2 and T4, the women's interpretations changed little. The proportion of women reporting each interpretation option is consistent with the results of previous studies (Cypowyj et al., 2009), except for option 4. In this study, a higher than average number of women interpreted their results as not carrying a pathogenic mutation with certainty.

One clinical concern with individuals interpreting uninformative BRCA1/2 test results as indicating certainty that they carry a pathogenic genetic mutation is that it could be associated with increased distress. However, we found that, although most women interpreted their results as ambiguous, there was no association between interpreting uninformative results as option 2 or option 3 and distress as measured by total IES score.

### ***Predictors of Distress***

Clinically, the ability to predict who is at risk for distress due to genetic testing is important in order to determine who may require additional emotional support and to offer timely interventions. We analyzed potential predictors of distress by examining IES total scores (distress associated with genetic testing). The scores were processed in a linear regression analysis with the following independent variables: baseline (T1) demographic and lifestyle data, perceived BC risk, perceived inherited-mutation risk, and interpretation of results. Significant Pearson correlations were observed and then multivariate GEE modelling was conducted on IES total scores. The result is described below.

### ***Predictors of Distress Associated With Undergoing Genetic Testing (IES Total Score)***

The main objective of our GEE analysis was to assess the efficacy of the intervention. The GEE result suggested significant decreases in distress at T3 (3 months after intervention completion) compared with T2 (immediately after receipt of uninformative results) (OR = 0.5678;  $p = 0.0215$ ), with T1 (baseline) average IES scores having a significant effect on IES level at all time points (log OR = 0.3109;  $p < 0.0182$ ). Through GEE, we found several other predictors of distress associated with undergoing genetic testing. As the subgroup analysis explores a large number of predictors, the multiplicity issue can inflate the overall familywise type I error rate. Multiplicity adjustment was therefore needed, and we performed Bonferroni's correction to control the familywise type I error rate. As there were 13 predictors in the model, we adjusted the  $p$  value threshold to  $0.05/13 = 0.0038$ . Other significant predictors of distress were not planning lifestyle changes to improve health (OR = 5.7221;  $p = 0.0006$ ) and greater time lapse between genetic testing and cancer diagnostic (OR = 1.0650;  $p = 0.0011$ ). Significant protective factors predicting lower levels of distress were having university education (OR = 0.2817;  $p = 0.0014$ ), having higher income (OR = 0.6947,  $p = 0.0001$ ), not employed (OR = 0.0692,  $p < 0.0001$ ), and a trend towards lower perceived risk of developing BC (OR = 0.2271;  $p < 0.0452$ ). These findings from subgroup analysis can be used to generate hypotheses for future validation.

## **Discussion**

The results of our pilot study suggest that a psycho-educational telephone intervention informed by illness uncertainty theory is feasible and acceptable in clinical practice. The intervention may be beneficial for women with BC and a family history of the disease who receive



uninformative BRCA1/2 test results and who are experiencing related distress. Of the women who took part in the interview at the end of the study, all provided strong support for the utility of the intervention. Overall, our preliminary results point to a marked decrease in distress between receipt of test results and 3 months post-intervention, as well as a sustained decrease in distress at 12 months post-intervention. Although this decreased distress may reflect a decrease in cancer-related distress rather than distress associated with receiving genetic test results, previous studies have shown that, when left untreated, general cancer distress tends to remain elevated (Carlson et al., 2004). Hence, considering that the IES scale used in the study was anchored on distress associated with undergoing genetic testing for BRCA1/2, it may be that the intervention did decrease genetic testing distress.

In contrast to the IES scores, the MICRA total score did not indicate a decrease in impact of genetic testing disclosure post-intervention. This may reflect a limitation of the scale, having only five items of distress to measure a multifactorial situation. For future studies, we recommend the use of a recently published validated tool, the Genetic Psychosocial Risk Instrument Scale (GPRS) (Esplen et al., 2013), which specifically measures genetic distress from a multifactorial angle. The GPRS also has the advantage of being able to screen for psychological risk before individuals receive their genetic test results, allowing for preventive action. This tool also contains a clinical cut-off score, while the MICRA scale does not.

Our results suggest that distress did not increase among participants between T1 (pre-testing) and T2 (following receipt of test results); therefore, uninformative results did not increase the women's distress. However, we did not find the substantial decrease in distress reported in previous studies among individuals who receive certain-negative results (Bish et al., 2002; van Dijk et al., 2006). Moreover, the psychological reactions of the participants receiving uninformative test results more closely resembled the reported reactions of those who receive positive results indicating the presence of a gene mutation.

The predictors of distress identified in this study are similar to those reported by other studies. Not uncommonly, distress decreased over time, and, consistent with the findings of previous studies, baseline (T1) distress predicted post-genetic-testing (T2) distress (den Heijer et al., 2013). Women who waited longer after their cancer diagnosis to obtain genetic testing and who were not exercising (Dorval et al., 2008) tended to experience more distress, while higher education, higher income, not being employed, and low perceived risk of developing BC seemed to provide a protective shield against high levels of distress.

Women who receive uninformative BRCA1/2 genetic test results remain a vulnerable and understudied group (Ardern-Jones et al., 2010).

The unresolved uncertainty about their BC risk and mutation-carrier status can impair their quality of life (Dorval et al., 2005). Similar to other researchers investigating the impact of uninformative results (Bish et al., 2002), we found that few women believed, after receiving uninformative results, that they were highly likely to carry an inherited BRCA1/2 mutation. Although most still interpreted their results as ambiguous, we conclude that, for the majority of participants, their perceived mutation risk and test-result interpretation did not result in increased distress. However, a small subset of women interpreted their results as indicating certainty of carrying an inherited mutation, and this interpretation predicted mutation-related distress.

Telephone support in clinical contexts such as the one tested in this study is not new. Studies demonstrating the efficacy of telephone support report that participants view this approach as more “normal” than returning to the hospital to receive in-person support and thus are more satisfied with telephone support (Beaver et al., 2009; Beaver et al., 2011). In our study, the telephone intervention was delivered by the genetic counsellors who had counselled the women before their BRCA1/2 testing. The genetic counsellors were guided by a fully developed and manualized intervention collaboratively designed by the interdisciplinary research team — a strength of the study. All participants reached by telephone expressed satisfaction with their care and relief at having some closure. The intervention manual can be used as a guide for other trained health-care providers in genetics, such as genetic nurses working in hereditary cancer centres, in using the PET intervention with their population.

### ***Limitations***

Although the results of this preliminary study demonstrate that the PET intervention has the potential to reduce distress, the study has some limitations that need to be taken into consideration. Because of the absence of a randomized-control design, combined with the absence of a control group, the possibility of reduced distress over time without the intervention cannot be ruled out. However, the findings of previous studies show that post-genetic-test distress is unlikely to decrease without specific psychotherapy interventions (Graves et al., 2010). Thus, our findings provide some information on the changes that may have occurred among the women who received uninformative test results following the PET intervention.

### ***Recommendations for Practice and Research***

These results have direct implications for the care and quality of life of BC survivors who receive uninformative and ambiguous BRCA1/2

genetic test results. Targeted interventions for this group are greatly needed, considering the large proportion of individuals likely to receive uninformative results. Future research should investigate whether the telephone counselling alone, or the full intervention, produced significant changes in the variables measured in this study. In summary, this is an important study because of the subject matter and its essential preliminary design in obtaining important information that will allow for a subsequent RCT. A pilot study based on an RCT design could make an important contribution to the nursing literature. These small-scale pilot RCTs can reveal significant trends that can stir clinical debate and even shape clinical practice.

### **Conclusion**

Preliminary evidence suggests that a psycho-educational telephone intervention, consisting of a psycho-educational information booklet, a relaxation CD, and a telephone follow-up care session, is clinically feasible and could minimize the potential negative psychological impact of receiving uninformative BRCA1/2 test results. Although a cause-and-effect relationship between the intervention and reduced distress could not be established in this pilot study, the results are promising and provide evidence of the need to evaluate the effectiveness of the intervention's components in an RCT format.

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*Pilot Intervention for Women Receiving Uninformative BRCA1/2 Test Results*

Christine Maheu et al.

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# Honouring Stories: Mi'kmaq Women's Experiences With Pap Screening in Eastern Canada

Catherine MacDonald, Ruth Martin-Misener,  
Audrey Steenbeek, Annette Browne

Mi'kmaq women are reported to have lower rates of Papanicolaou (Pap) screening and higher rates of cervical cancer than non-Aboriginal women. This qualitative participatory study used postcolonial feminist perspectives and Indigenous principles to explore Mi'kmaq women's experiences with Pap screening within the contexts that shaped their experiences. Community facilitators assisted with the research process. Talking circles and individual in-depth interviews were conducted with 16 Mi'kmaq women. Also, health-care providers were interviewed in 2 Mi'kmaq communities. The findings indicate that historical and social contexts are shaping Mi'kmaq women's screening experiences and that these experiences are diverse, as are their understandings about screening. Some women were accessing regular screening despite challenging personal circumstances. The results highlight the need for nurses and other health-care providers to understand the uniqueness of each woman's experiences with Pap screening. Improvements in screening rates depend on multifaceted nursing approaches developed in partnership with Mi'kmaq women.

**Keywords:** Aboriginal women, cervical cancer prevention, Pap screening, participatory action research, postcolonial feminist perspectives, health-care access



## **Histoires de dignité : comment le test de Pap est vécu par des Micmaques de l'Est du Canada**

**Catherine MacDonald, Ruth Martin-Misener,  
Audrey Steenbeek, Annette Browne**

Selon les données, les Micmaques subissent le test de Papanicolaou (Pap) en plus petite proportion que les femmes non autochtones et présentent un taux plus élevé de cancer du col de l'utérus. La présente étude qualitative et participative adopte une approche féministe postcoloniale et s'appuie sur les principes autochtones pour examiner la façon dont les Micmaques vivent le dépistage du cancer du col de l'utérus (test de Pap), et les différents contextes où leur expérience de ce dépistage prend forme. Des animateurs communautaires ont pris part au processus de recherche. Des cercles de discussion et des entrevues individuelles approfondies ont eu lieu auprès de 16 Micmaques. Des fournisseurs de soins de santé ont également fait l'objet d'entrevues dans deux communautés micmaques. Les constatations indiquent que les contextes social et historique contribuent à façonner l'expérience vécue par les Micmaques au moment du test de Pap et que cette expérience varie, de même que la compréhension qu'ont les femmes du dépistage. Certaines femmes participent à un dépistage régulier, malgré une situation personnelle difficile. Les résultats obtenus font ressortir la nécessité pour les infirmières et les autres fournisseurs de soins de santé de comprendre le caractère unique de l'expérience de dépistage vécue par chaque femme. L'amélioration des taux de dépistage est tributaire de la mise en place d'approches à multiples facettes des soins infirmiers élaborées en partenariat avec les Micmaques.

**Mots-clés :** femmes autochtones, dépistage du cancer du col de l'utérus, test de Pap, étude participative, approche féministe postcoloniale, soins de santé

The health of Aboriginal<sup>1</sup> women is foundational to the well-being of their families, communities, and Nations (Native Women's Association of Canada, 2007). Despite recent improvements in health status at the population level, and despite the collective efforts of Aboriginal women to foster strength and health within their families and communities, health and social inequities persist in this population in comparison with the general Canadian population (Halseth, 2013).

In this article we explore cervical cancer as one particular health issue that needs attention. Although Papanicolaou (Pap) screening has been effective in decreasing the morbidity and mortality rates of cervical cancer (Canadian Cancer Society, 2012), Aboriginal women continue to have lower rates of screening and higher rates of cervical cancer than other Canadian women (Brassard et al., 2012; Zehbe, Maar, Nahwegahbow, Berst, & Pintar, 2012).

Much of the health-care literature, particularly the epidemiological literature, reports that Aboriginal women have higher rates of cervical cancer than non-Aboriginal women. Often, these rates are reported along with risk factors for cervical cancer, such as certain high-risk behaviours, lower rates of screening, and high rates of sexually transmitted infections (Gerberding, 2004; Johnson, Boyd, & MacIsaac, 2004; Reeves, 2008; Sheets, 2002; UNAIDS & World Health Organization, 2006). When the high rates of cervical cancer among Aboriginal women are attributed to these risk factors, Aboriginal women are frequently labelled as "high risk in terms of their reproductive health" (Browne & Smye, 2002, p. 32). However, research indicates that multiple factors impact the health status of Aboriginal women and shape their access to health services and their health-care experiences. It is therefore imperative to contextualize health status in light of sociopolitical, historical, and economic factors affecting health (Adelson, 2005; Browne, 2007; Browne et al., 2011). It is equally important to recognize that women's health is shaped by various strengths and abilities, including knowledge of local languages and community and cultural practices and ceremonies (Kirmayer, Dandeneau, Marshall, Phillips, & Williamson, 2011).

In many cases the mainstream health-care system is poorly aligned with the health-care needs of Aboriginal people because it tends to conceptualize health and illness as stemming from "lifestyle" or cultural differences and to overlook the contextual factors that shape health and illness (Browne & Dion Stout, 2012). Health-care policies and strategies

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<sup>1</sup> The word Aboriginal refers to the original peoples of North America and their descendants. The Canadian Constitution recognizes three groups of Aboriginal people: First Nations, Métis, and Inuit. These are three distinct peoples with unique histories, languages, cultural practices, and spiritual beliefs (Aboriginal Affairs and Northern Development Canada, 2013).

aimed at helping Aboriginal people but based on Western models of health-care delivery and on individualistic, disease-based discourses can perpetuate racism, social exclusion, marginalization, and inequitable access to care (Barton, 2008; Martin, 2012). Nurses are in key positions to address inequities that affect access to Pap screening for Aboriginal women by incorporating the concept of cultural safety into screening practices, developing respectful relationships prior to screening, tailoring screening approaches to the specific needs of individual women, and adopting relational and collaborative approaches.

The purpose of this article is to report on findings from a community-based participatory study conducted in partnership with two Mi'kmaq communities in eastern Canada focused on cervical cancer screening. This qualitative study explored women's experiences with Pap screening in the two rural Mi'kmaq communities using a broader lens to conceptualize their health-care experiences. This allowed for women's participation in Pap screening to be considered with a fuller understanding of the historical, economic, and sociopolitical contexts that construct these experiences. Storied interviews were conducted with Aboriginal women and health-care providers. The research questions were as follows: 1. *What are Aboriginal women's experiences with Pap screening?* 2. *What are Aboriginal women's awareness of and knowledge about Pap screening?* 3. *What are the perceptions of Aboriginal women and health-care providers regarding the reasons why some Aboriginal women are not participating in Pap screening?* 4. *What are the sociopolitical, economic, and historical factors that shape Aboriginal women's participation in Pap screening?*

### **Theoretical and Methodological Perspectives**

The study was informed by postcolonial feminist theoretical perspectives (Anderson, 2004; Reimer-Kirkham & Anderson, 2002) and Indigenous principles in a two-eyed seeing approach. The term "two-eyed seeing" (Sesatu'k Etuaptmnl) refers to the need to learn from one eye the strengths of Aboriginal traditional knowledge and from the other eye the strengths of Western scientific knowledge (Iwama, Marshall, Marshall, & Bartlett, 2009). Postcolonial feminist theory provides an analytic lens for exploring how women's lives and health have been positioned and shaped by politics and history (Browne, Smye, & Varcoe, 2005, 2007). "Postcolonial" does not mean that colonialism is over or completed: "the post in postcolonial refers to a notion of both working against and beyond colonialism" (McConaghy, 2000, p. 268). The use of postcolonial feminist perspectives allowed for Mi'kmaq women to be viewed not as a gendered group but as individuals with distinct historical, socio-economic, and political experiences (Lewis & Mills, 2003). There was an

understanding of how power, privilege, socio-economics, and race have contributed to inequities in access.

Indigenous principles fundamental to conducting respectful research with Aboriginal people provided the foundation for the theoretical development of the study (Martin, 2012; Weber-Pillwax, 2004). Indigenous principles of relationality, respect, reciprocity, relevance, and responsibility were also incorporated into the framework (Loppie, 2007; MacDonald, 2012). Respectful relationships with community members were developed through frequent visits to the health centre and attendance at community events prior to initiating the study. This was vitally important for the acquisition of knowledge about the communities, community members, community norms, and community expectations with regard to the research.

## **The Study**

### ***Qualitative Participatory Action Research Design***

The research process was guided by a qualitative research design using participatory action research (PAR) and Indigenous principles. The qualitative nature of the study provided a means for analyzing and interpreting how the colonial past and the current sociopolitical and economic climate impact Aboriginal women's access to Pap screening. PAR principles of equity, social justice, democratic collective decision-making, and reciprocity (Loppie, 2007; MacDonald, 2012; Ortiz, 2003; Vollman, Anderson, & McFarlane, 2004; Vukic, Gregory, & Martin-Misener, 2012) were applied throughout the research. These principles were upheld by enabling the participation of community members, acknowledging that the viewpoints of all participants were valued, using collaborative decision-making regarding the research process and dissemination of findings, developing relationships, and fostering open communication. PAR is grounded in the practical problems and health issues of people in a given community. Thus, the research topic was determined with some community members by developing collaborative relationships while visiting the communities and the community health centre.

Women told stories from their contextualized social, economic, political, and historical realities and were considered co-investigators and active participants in different phases of the research. They provided input into the research questions, participant recruitment, data collection and analysis, modification of the interview guide, and dissemination of findings.

Ethical approval was obtained from the University Health Sciences Human Research Ethics Committee at Dalhousie University, Mi'kmaq Ethics Watch at Cape Breton University, and Mi'kmaq community leaders. The OCAP (ownership, control, access, and procession) principles

(1998) and the Canadian Institutes of Health Research (2007) guidelines for health research involving Aboriginal people were also followed. The OCAP principles were respected by informing the women about the entire study prior to their participation, discussing anonymity and confidentiality with them, ensuring that each informed consent form was signed and a copy given to the participant, and engaging participants in discussions about the findings and their dissemination.

**Role of community facilitator.** Community members, who were recommended (based on interest and willingness to participate) by health-care providers in each community, were invited and agreed to be the community facilitators. Their role was to organize the talking circles (described below), assist with recruitment, act as liaison between the community and the primary researcher, provide feedback regarding the interview guide and the research process, and review and comment on themes throughout the analysis. Community facilitators expressed an interest in being active throughout the research process and repeatedly affirmed the relevance of the research topic for their community. They communicated the information that there were some women in the community who did not go for Pap screening and that in each community they knew a woman who had a diagnosis of cervical cancer or who had died of cervical cancer.

**Recruitment of Mi'kmaq women.** Sixteen women were recruited using purposive and snowball sampling. Aboriginal women of diverse ages, socio-economic backgrounds, education, and Pap screening experiences were purposively recruited by community facilitators, who invited 20 to 30 Mi'kmaq women to participate in a talking circle with a health professional, a health centre director, and the primary researcher. The purpose of the talking circle was to facilitate collaboration and promote co-designing of the study.

The primary researcher used talking circles because traditionally a talking circle is structured to permit all voices to be heard (Tompkins, 2002). The talking circles for this study, one in each community, were premised on the PAR principles of respect, shared decision-making, reciprocity, and relationality, allowing for the mutual exchange of information in a comfortable environment with food. Women posed questions, made comments, told stories, and provided suggestions regarding the interview questions and the research process. The first talking circle comprised eight women and a nurse, while the second comprised 10 women, a nurse, and a health centre director. Those who participated in the talking circle informed other women in the community about the study, which resulted in the recruitment of a few other women.

To be included in the study, women had to (1) be a self-identified or status Aboriginal Mi'kmaq; (2) live in one of the two Mi'kmaq commu-

nities; (3) be between 21 and 75 years of age; (4) have had a least one Pap screening; (5) be able to provide informed consent; and (6) be able to read, understand, and speak English. A letter of introduction describing the study was distributed.

After consent for participation had been obtained, storytelling interviews using an in-depth guide, developed for the study with input from the community facilitators and women, were arranged at a time and location agreeable to each participant.

***In-depth interviews.*** The women had the choice of taking part in one or two in-depth interviews so that each would have an opportunity to tell her stories about beliefs, attitudes, and experiences with respect to Pap screening. The interviews lasted from 60 to 90 minutes. A total of 16 women participated in the first interview (13 attended the talking circles) and all but one agreed to take part in the follow-up interview. The second interview took place following transcription of the first interview and preliminary analysis of the findings. During the second interview, the women were asked to provide feedback on data interpretation, preliminary findings, and emerging themes. Five health-care providers volunteered to take part in an in-depth interview and four agreed to a follow-up interview. In total, there were 40 interviews.

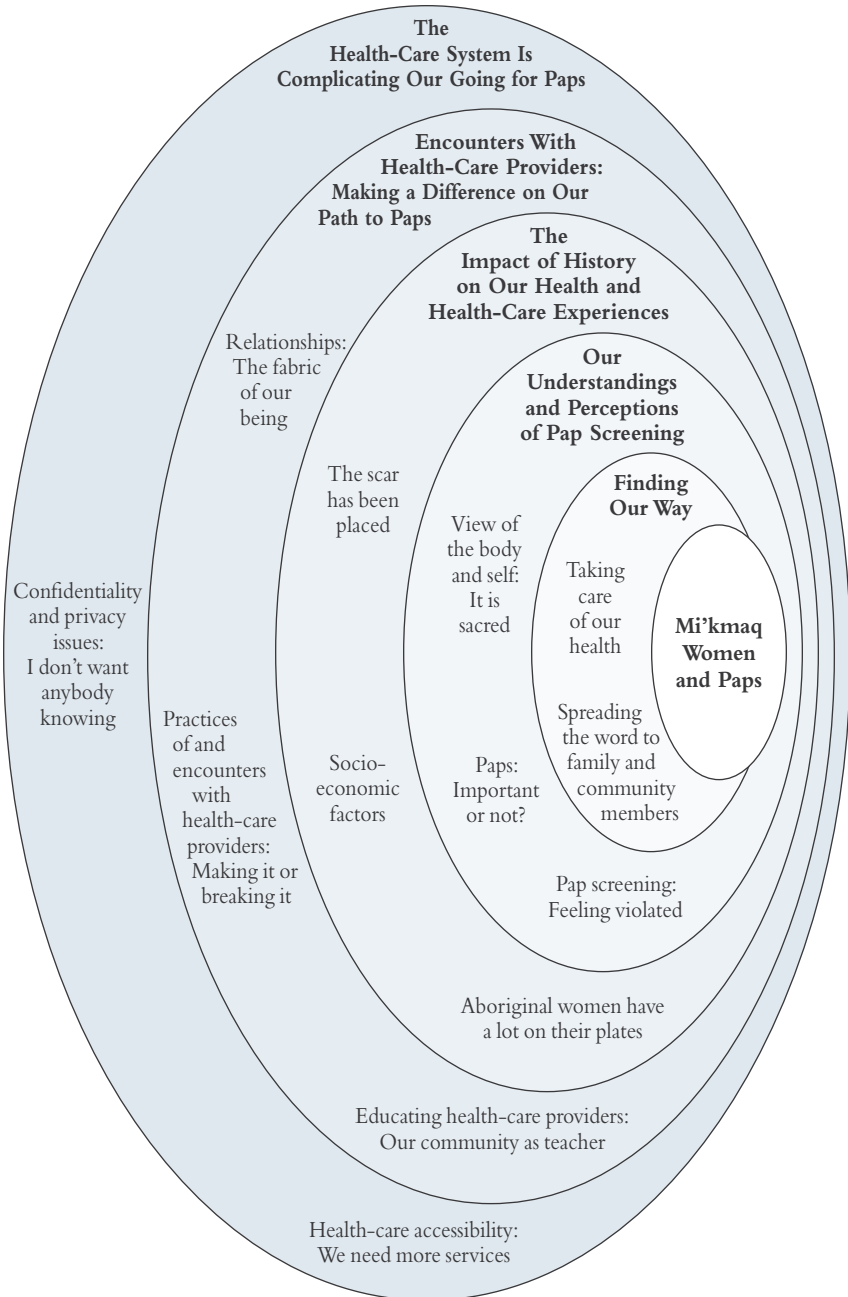
### ***Data Analysis***

After the audiotaped interviews were transcribed verbatim, data were imported into NVivo 8 qualitative software. Women had an opportunity to read and validate their transcribed interviews and provided input about preliminary themes and subthemes. Thematic analysis processes described by Sandelowski (1995) and O'Connor and Gibson (2003) were employed to identify themes elicited from the participants' stories. Although these themes were coded primarily by the researcher, women changed some of the titles of the themes and subthemes and offered explanations from an Indigenous perspective why the titles were or were not appropriate. Women clarified, altered, or confirmed that the data adequately reflected and were consistent with their stories and their experiences of Pap screening. Community facilitators also reviewed some of the coding processes to ensure validity of themes and subthemes. The trustworthiness of the research was enhanced through member checking, recording of field notes, forming of partnerships with and debriefing with community facilitators, peer review, and debriefing.

## **Findings and Discussion**

The five themes and subthemes (see Figure 1) identified in the Mi'kmaq women's stories will be presented by integrating excerpts from inter-

Figure 1 *Themes and Subthemes: Mi'kmaq Women's Experiences With Pap Screening*



views. The findings and their interpretation are interwoven with the literature in a relational manner that honours and represents Aboriginal women's stories in an attempt to minimize the fracturing of knowledge passed on in oral form (Brown & Strega, 2005; Smylie, 2001).

### ***Finding Our Way***

This theme and its two subthemes reveal how Aboriginal women are finding their way by paying attention to and taking care of their health and the health of their families and communities. Women described being committed to taking care of their health by seeking knowledge, becoming educated, and sharing knowledge with family and community members.

***Taking care of our health.*** Many women described the importance of taking care of their health by taking action to address some of the determinants of health, even though they were confronted with considerable disadvantages and adversity. Some talked about taking care of their health by becoming educated, being employed, and instituting early childhood learning. Others told stories about returning to cultural and traditional ways, revitalizing their language, and passing along parenting skills. For some women, seeking knowledge about health care was a way of taking care of their health.

A few women took care of their health by making sure follow-up appointments were made for their test results and treatments and keeping track of the dates for these appointments: "I know when my appointments [for Pap screening] are due. Usually I'll remember and I keep track."

These findings offer a counterbalance to much of the epidemiological literature implying that Aboriginal women do not take care of their health nor make personal efforts to do so. Much of the research has focused on women's low rates of Pap screening, high rates of cervical cancer, barriers to accessing health services, strategies to improve Pap screening rates, and reproductive health issues currently confronting Aboriginal women (Black, 2009; Johnson et al., 2004; O'Brien, Mill, & Wilson, 2009; Steven et al., 2004). Epidemiological statistics alone do not convey the current state of Aboriginal health (Graham & Stamler, 2010) and suggest that lifestyle choices are the reason for women not accessing Pap screening. According to Nelson (2012), the burden of responsibility for health and social problems is continually placed on Aboriginal people, who are denied the resources needed to adequately tackle these problems. Epidemiological research needs to be developed with other types of research that consider the contexts of Aboriginal people's lives.

***Spreading the word to family and community members.*** This subtheme concerns the importance that the women placed on sharing their knowl-



edge and wisdom about health and Pap screening with family and community members. While some women said that there has been a loss of some traditional ways and knowledge, they continued to use storytelling with family and community as a path to taking care of their health. The women also spoke of the knowledge and advice they could share with health-care providers and policy-makers to make Pap screening a more positive experience. A few viewed sharing words of wisdom about Pap screening to be their personal responsibility as women, mothers, aunts, grandmothers, and friends, as it was traditionally with other aspects of health: "We learned about it [Pap screening] through word of mouth from family."

Some women in the community had taken on more formal educational roles, such as appearing on posters or presenting at health conferences; this was seen as important for raising awareness and recruiting Aboriginal women for Pap screening. One woman spoke of an acquaintance who attended health conferences and appeared on posters to share her knowledge about the importance of Pap screening and prevention of cervical cancer: "She was going to a health conference and they asked her to participate [in it] because she's . . . on the posters and stuff . . . that's probably the first time I really heard of it as cancer prevention, and that would have been . . . 4 years ago."

The interview data show that Aboriginal women are often forced to seek information about health care from Western sources such as pamphlets and other print materials, which are not always representative or reflective of their realities. Several participants indicated that brochures and other educational materials that are reflective of Aboriginal women's lives positively influenced Aboriginal women to access Pap screening: "It would seem important for them to attend [screening], and they could relate to those women in the pamphlets." This demonstrates the importance of having information that is relevant and appropriate for Aboriginal women and that promotes "the empowerment of Aboriginal women to take control of their own health care needs" (Black, 2009, p. 174).

### ***Our Understanding and Perceptions of Pap Screening***

The women expressed a multitude of beliefs, experiences, and feelings about Pap screening that often involved cultural ways of thinking about the body and self.

***View of the body and self: It is sacred.*** Several participants explained that an Aboriginal view of the body is holistic and encompasses physical, emotional, mental, and spiritual domains that are not viewed as distinct or separate. Some women described their perceptions of the body and self as holistic, rooted in culture and traditions.

Some women described the perineum as a “sacred area.” One woman viewed that part of the body as sacred because “that’s where your baby’s life comes through, of course it’s sacred.” In traditional Aboriginal culture, the ability to give birth and raise children positions women in an esteemed, sacred, and respected role (Carroll & Benoit, 2004). Some women referred to the area of the body where Pap screening is conducted as a “private area.” A few said that they did not associate this part of the body with Aboriginal culture: “I don’t really feel like there’s a cultural thing — it’s a private area.”

Although some of the women expressed the opinion that Aboriginal and Western views of the body and self are different, others did not. One woman said, “Not all First Nations people view the body and self in the same way.”

A few women attributed the reluctance to discuss or seek Pap screening to the legacy of residential schooling and abuse. Traditionally in Aboriginal culture, teaching and knowledge acquisition about the body and sexuality were passed on from one generation to the next orally by way of narratives, storytelling, talking circles, and sharing circles (Barnes, Josefowitz, & Cole, 2006). However, due to residential schooling and the fracturing of families, many teachings passed down to children about the body have been lost. Women talked about the loss of children to residential schools and the resultant disruption in traditional parenting and teaching. Children were removed from their family, community, and home environments and placed in a foreign environment where they were abused and forbidden to speak of or follow any traditional teachings. Consequently, children were no longer educated about aspects of the body, health, sexual health, and historical teachings that previously had been passed down and had contributed to their well-being and the strengthening of family ties (Kinnon & Swanson, 2002).

***Paps: Important or not?*** This subtheme describes women’s understandings and perceptions regarding the importance of Pap screening and its impact on their accessing of screening services. Many women communicated similar yet at times diverse and conflicting perceptions of the role of Pap screening. Several believed that Pap screening, although uncomfortable and at times embarrassing, is important and continued to regularly access services: “I don’t like it, but I know it’s important to make sure, health-wise, everything is OK in your female area.”

Some of the women accessed Pap screening to remain healthy in order to care for their children, to be able to have children, and, when pregnant, to prevent cervical disease through early detection: “I don’t want to find out that I have cervical cancer. I want to make sure that I don’t end up with any kind of disease . . . and if so, [that] they can get it early.” Others, however, illustrated that not all women view Pap screening as

valuable, relevant, or necessary, due to a lack of knowledge and the impact of residential schooling, which is discussed under the next subtheme.

In a few other instances, fear of having cervical cancer was a reason given for *not* being screened. This finding is similar to what others (Black, 2009; Letendre, 2008; O'Brien et al., 2009) have found in studies with Aboriginal women. Fear of cervical cancer prompted some women in the study to access screening and prevented others from doing so.

***Pap screening: Feeling violated.*** This subtheme represents Aboriginal women's accounts of their negative experiences with Pap screening. Some women told explicit personal stories of Pap screening making them feel violated: "Sexual abuse could be a reason for our women [Aboriginal] not to go. It could bring up bad memories from the past."

Some of those who described feeling violated linked it to prior experiences of abuse and a filtering down from residential school experiences; others did not articulate this experience: "Abuse and everything that filters out of residential school . . . there [were] a lot of things that happened that make Paps uncomfortable."

For some women, not accessing Pap screening was a way to protect themselves against reliving experiences of sexual abuse and violation. Pap screening for Aboriginal women can be viewed as an extension of colonization and the pain and suffering from sexual abuse and acts of violence that occurred in residential schools. This abuse and violence has affected not only residential school survivors themselves but also their descendants. A potential additive of historical trauma and sexual abuse is having a Western health-care provider, particularly a male, perform Pap screening.

The literature highlights Aboriginal women's lack of knowledge and information about Pap screening and lack of understanding about what happens to them during the screening experience (Amankwah, Ngwakongnwi, & Quan, 2009; O'Brien et al., 2009; Steven et al., 2004). In the present study, women linked a lack of knowledge about and understanding of Pap screening with being traumatized or violated. A few even revealed that although they were informed about the procedure, they remained terrified and fearful of the results. Many of the women demonstrated courage in moving on from extremely violating experiences of sexual abuse and assault to engage in regular Pap screening, a procedure that itself can be a violating experience.

### ***The Impact of History on Our Health and Our Health-Care Experiences***

This theme comprises three subthemes wherein women discuss the historical, social, political, and economic factors that influenced their health, their health care, and particularly their accessing of Pap screening.

***The scar has been placed.*** This subtheme reflects the women's perceptions concerning the impact of historical trauma on their accessing of health services, including Pap screening. Almost every participant described in depth the impact of historical trauma, particularly with reference to residential schooling and/or colonization, on Aboriginal people's access to health services and Pap screening. Despite the historical trauma, they continued to thrive in less than adequate circumstances and struggled against negative forces that resulted from pain, suffering, and losses inflicted by residential schooling. Two of the women had been in residential schools themselves. Many others shared stories illuminating inequities in health and access to health care and the disempowering impact of residential schooling: "Issues with trusting health-care providers and the health-care system, I think, have been passed on from our history, from our ancestors that attended residential schools . . . They still have power over us, just like they did when we were in residential schools."

One woman spoke of being stereotyped as a First Nation person when accessing health care: "It's just like where you're from or what your background is or what your culture is — right away you're stereotyped just from what other people say." This perspective is supported by the findings of a qualitative study conducted by Tang and Browne (2008) into Aboriginal people's perception that they are treated differently by health professionals when accessing care due to their Aboriginal identity and low socio-economic status. Discrimination, stereotyping, or racism in encounters with health-care providers was influential in shaping their access to Pap screening. Historical trauma as a result of residential schooling has engendered distrust of non-Aboriginal people and affected Aboriginal women's accessing of health services (Dion Stout, 2012; Haskell & Randall, 2009; Waldram, Herring, & Young, 2006).

***Socio-economic factors.*** Socio-economics was a primary factor in the failure to access health services and Pap screening. Participants identified being poor, lacking money for transportation or child care, lacking education, being a young, single parent, and being economically dependent on the government as factors that influenced their accessing of Pap screening. Some alluded to gender and Aboriginal identity as contributing to their socio-economic status and access to care. The women were focused on daily survival and making ends meet. Reasons for not accessing Pap screening are summed up in the comment of one woman: "We never had a lot of money or education. We didn't know it [Pap screening] was important."

Kurtz, Nyberg, Van den Tillaart, and Mills (2008) report that economic and social disadvantage caused by poverty, lack of education, and unemployment are associated with poor health and health outcomes, especially among Aboriginal women. Similarly, Black, Yamada, and Mann

(2002) identify poverty, Aboriginal status, and lack of education as reasons for failure to access Pap screening. Essentially, one cannot expect Aboriginal women to access health care when their basic needs are not being met and poverty related to disconnection from lands, traditions, and families due to colonization persists (Dion Stout, 2012).

Several women reported that being pregnant ensured economic stability and being cared for by the community. Being pregnant is not linked to economic security in the literature. The women did not expand on why they were assured access to health services such as Pap screening only when they were pregnant or on why resources to access Pap screening when needed were not provided.

***Aboriginal women have a lot on their plates.*** This subtheme elucidated the issues and multiple and diverse roles and responsibilities of Aboriginal women within their families and their communities that impacted their access to health services and Pap screening. Some participants spoke of not accessing screening due to working outside the home, attending school, keeping house, or having community and child-care responsibilities: "Having a lot on their plates, they do not place themselves at the top of the list for care, particularly when they are single parents or have jobs." However, some made time for Pap screening. One married working mother said that she took time for health care and screening: "I've been really busy, hard to find time, but I go for Paps."

These findings are consistent with those reported in the literature. Women's family and caregiving responsibilities and roles directly affect their health, and in many Aboriginal communities other health-care issues take precedence over women's health (Barnett, White, & Horne, 2002; National Aboriginal Health Organization, 2006). For many participants, it was evident that mothering and family were the most prominent of all traditional roles and values. Some women had been forced to take low-paying jobs outside the home or even outside the community while continuing to be responsible for the care of the family and community, which at times took priority over Pap screening.

### ***Encounters With Health-Care Providers: Making a Difference on Our Path to Paps***

This theme described women's perceptions of how health-care providers are making a difference in their access to Pap screening.

***Relationships: The fabric of our being.*** This subtheme concerns the women's perceptions about the impact of relationships with health-care providers on their accessing of Pap screening. The majority detailed the importance of building meaningful, trusting, and respectful relationships with health-care providers, which positively influenced their access to Pap screening. One woman said that fostering relationships was "building

a start to increasing Pap testing.” Another said, “The way we’re treated and looked at by health-care providers plays a big part in how we see ourselves.”

Several participants suggested that visiting the community and attending cultural ceremonies and funerals were ways for health-care providers to build relationships in the community. Others expressed the view that not all health-care providers are trusted by community members and that this complicates relationship-building. These viewpoints reflect the diversity of perspectives within Aboriginal communities and among Aboriginal community members as a result of the differences in historical and social experiences and encounters with non-Aboriginal people, including health-care providers. For some of the participants the gender of the health-care provider also had an impact on their ability to form a relationship during Pap screening. A young woman explained: “A female health-care provider understands what it is like to be a woman and knows what it is like to experience Pap smear screening.” For a few others, the provider’s gender was not considered important for relationship-building during Pap screening. Building and maintaining relationships are considered cornerstones of life, health, and survival in Aboriginal communities (Henderson, 2000; Wilson, 2001). Aboriginal women want relationships with health-care providers that foster the creation of safe and ethical spaces where their voices and their concerns about health and health-care access can be heard (Kurtz et al., 2008). One woman expressed this eloquently: “Relationships are part of the fabric of our being.”

***Practices of and encounters with health-care providers: Making it or breaking it.*** The women’s perceptions about the practices of and encounters with health-care providers influenced their accessing of health services and Pap screening. Women gave examples of when providers’ practices either helped or hindered their accessing of health care. The stories were about their own health-care encounters or those of members of their family or their community. Some women indicated that mainstream health services were not aligned with their needs when accessing Pap screening. Others described negative experiences with providers and gave examples of being discriminated against and not receiving culturally safe and competent care: “They generalize [about] us too much. They think we’re drunks. You’re hung over or something like that. Or . . . pill poppers.”

In some health-care systems, women are receiving culturally unsafe care as a result of the devaluing of Aboriginal knowledge, traditions, and ways. Culturally unsafe nursing care and practices encompass situations in which an action “diminishes, demeans or disempowers the cultural identity and well-being of any individual” (Nursing Council of New

Zealand, 2011, p. 7). The literature reveals instances of Aboriginal women encountering health-care providers whose unsafe practices include stereotyping, discrimination, and racism (Browne et al., 2011; Browne et al., 2012; McGibbon & Etowa, 2009). In the present study, lack of cultural safety was evident in the few Aboriginal pamphlets or teaching tools available or offered in mainstream health-care settings. Not a single participant told of being provided with information about health or health care in her own language.

Although the women seldom used the term “cultural safety” in their stories, they provided examples and expressed a preference for culturally relevant services. A few related positive experiences and described receiving culturally safe care from health-care providers who explained information adequately. For the participants, a key requirement for positive and safe care was relationships built on trust and mutual respect, as well as health-care providers being educated with regard to Aboriginal people and communities.

***Educating health-care providers: Our community as teacher.*** Several participants indicated that health-care providers working in Aboriginal communities need specific knowledge about their culture, history, and language, which can be taught by community members. A few also conveyed the importance of health-care providers being able to talk with community members and listen to their stories as a way of becoming knowledgeable about the community and its members. Thus, health-care providers need to learn to understand nuanced, non-verbal communication and how to address an Elder. They also need to learn how to initiate conversations about sensitive topics like Pap screening and ways to convey information to Aboriginal women. Participants spoke of the need for health-care providers to be educated about the community where they are working, and not just by reading about the community and Aboriginal people: “Get educated and trusted in the community, and learn from the community as opposed to just [from] a book . . . I think that maybe if they [came to] study in here [community] for a bit, they would understand us more.” One woman spoke of the need for health-care providers to be educated not only about the history of Aboriginal people, but also about the family unit, which has changed over time: “I think they should know a little bit about the history. They should also know about family units, [about] how families survive.”

Also apparent was the importance of recognizing the diversity that exists among Aboriginal people and not “essentializing” all Aboriginal people or using a “one size fits all” approach to health care in Aboriginal communities. According to the National Aboriginal Health Organization (2010), health-care providers need to become acquainted with and understand cultural beliefs, attitudes, and practices in order to address

barriers that Aboriginal people confront when accessing Pap screening. Several women mentioned that Pap screening workshops in their community would be helpful in educating women about the procedure, the results, and the importance of regular screening. Some even suggested that these workshops could be enjoyable social events for women by featuring food, presents, and door prizes. Black (2009) also raises the idea of organizing a day or week dedicated to Pap screening in the community, such as Pap Week or Papalooza, with games and prizes as incentives.

### ***The Health-Care System Is Complicating Our Going for Paps***

The women perceived the health-care system as influencing and complicating their access to services such as Pap screening.

***Confidentiality and privacy issues: I don't want anybody knowing.*** Women described issues around confidentiality and privacy that they or others had experienced and also fears about lack of confidentiality around Pap screening. For most participants, confidentiality and privacy were major concerns that affected their comfort in accessing screening services. Confidentiality concerns included being seen by others at the health centre, security issues with records, sharing of information by community members employed at the health centre, and sharing of information by health-care providers who socialized with community members. In both communities, almost half of the women revealed that they did not go to their community health centre for Pap screening for fear of confidentiality and privacy breeches. Instead, they opted to leave their community to access Western health services. One woman stated: "I've never actually been there [community health centre] for a Pap test. . . . The reason is [that] I know my results will come back there. I know my file will be there, everything about me — anybody can look at my file. That's why I won't go there." This woman said that she was uncomfortable with other community members having access to her health file: the community was "so close knit" that everyone knew each other.

According to Bourke et al. (2004), social relations impact confidentiality in rural communities. The lack of anonymity has specific consequences for sensitive health issues such as sexual and reproductive health. Confidentiality is more difficult to maintain in rural and small communities, particularly if the receptionist, patient, and health-care provider have relationships prior to and separate from the health-care encounter (Bourke et al., 2004). In rural communities, people know many of the particulars of each other's lives. Also, community members may have more than one role because of their social position or occupation, which can result in the sharing of personal information about other community members (Bourke, 2001). In a study conducted in British Columbia, confidentiality and privacy concerns were cited as barriers for Aboriginal



women obtaining Pap screening (Black, 2009); women residing on reserve did not want to have Pap screening at their community health centre when outside health-care providers came to the reserve, for the reason that other community members would know their business.

A few participants also mentioned that community medical drivers could be related to them and want to know why they were going for a certain appointment. In order to be paid, the drivers had to submit medical forms stating why the person required medical transportation.

Yet not all women had concerns about confidentiality or privacy in their community health centre: "I have [had] Pap smears there [community health centre] and there were no issues around privacy or confidentiality."

***Health-care accessibility: We need more services.*** Some women identified issues within mainstream health-care systems that impact Aboriginal women's access to Pap screening. Several reported a lack of Pap screening services, timely appointments, transportation, and interpreters. Accessibility issues were compounded by the confidentiality concerns discussed above. Even when health services were available, they were not accessible to all Aboriginal women: "We need more services. I like the idea of women's clinics and women's health days that would be accessible for everyone."

For the few who spoke primarily Mi'kmaq, there were literacy issues and a lack of translators available for services provided outside the community. One Elder said, "Each hospital should have an interpreter." The women also viewed mainstream health-care systems as inflexible, unwelcoming, indifferent, and not always considerate of their wishes. These findings indicate that the needs and preferences of Aboriginal women are not always the same in terms of Pap screening. To respond to the differences, health services should be available both within the community and outside it. Barnett et al. (2002) propose that health-care scheduling be not only flexible and convenient for Aboriginal women but also responsive to their diverse needs so that the right services are being offered to meet their specific needs and life contexts.

### ***Strengths and Limitations of the Study***

The use of PAR approaches, Indigenous principles, talking circles, and in-depth interviews enabled respectful and trusting relationships to be developed between researcher and participants. This approach fostered open dialogue and the opportunity to share diverse and rich stories about Pap screening in the context of Mi'kmaq women's lives. Many of the participants said that they had not previously talked about their experiences with Pap screening. Thus, it is evident that this research provided women an opportunity to give voice to their experiences.

Although the sample represented a broad range of ages, having only two women over the age of 60 is a study limitation. Also, the inclusion requirement that women speak, read, and understand English may have been a deterrent to participation. Women under 21 years of age and women who had never had Pap screening were excluded from the study; therefore, the findings may not be transferable to their experiences or perspectives.

### ***Health-Care Practice and Policy Recommendations***

It is evident from the results that health-care providers need to take account of the social determinants of health and the contexts of Aboriginal women's lives when considering why they are or are not accessing Pap screening. It is also vital that providers appreciate the impact of historical trauma, interpersonal violence, and trauma-informed care for Aboriginal people while at the same time being aware that Aboriginal people have strengths to counter traumas and violence. A "one size fits all" approach to health care will not be effective; it is essential that Pap screening practices with Aboriginal women be individualized. Health-care providers must acknowledge the limitations of mainstream standards and best practice guidelines and the fact that these may not always apply to Aboriginal people. It is critical that time be invested in building relationships in communities prior to the initiation of screening. Health-care providers should enhance education and knowledge about Pap screening with Aboriginal women in ways that acknowledge the women's realities, needs, and requests and that include opportunities for them to share their stories, perceptions, and experiences with other women. This would raise awareness of the importance of screening and honour storytelling as an authentic method for sharing knowledge. Also, Pap screening should be offered consistently in communities, with extended clinic hours and personal reminders, in order to increase access for Aboriginal women.

We need policies to address the complex determinants of health that contribute to major disparities in access to health care and Pap screening for Aboriginal women. We need to formalize confidentiality and privacy policies with Aboriginal communities and to educate health-care providers and all workers in community health centres about patients' right to privacy and confidentiality. We also need to develop and implement confidentiality policies for medical drivers who transport Aboriginal women to health services, including Pap screening. In addition, we need policies that clarify jurisdictional responsibilities for funding and screening supplies in Aboriginal communities. Finally, Aboriginal women and community members should be consulted on policy development, implementation, and evaluation related to Pap screening.

## Conclusion

This qualitative study explored Mi'kmaq women's experiences with Pap screening in two First Nation communities in eastern Canada and considered the historical, economic, and sociopolitical contexts that shaped these experiences. It is important to recognize that some Aboriginal women are accessing Pap screening regularly in spite of challenging circumstances. In general, epidemiological data alone do not provide insight into women's experiences with Pap screening nor identify reasons why women are or are not accessing screening. There are multiple factors, such as history, politics, socio-economics, health-care providers, and health-care systems, that impact women's access. It is critical that nurses and other health-care providers be aware of these diverse factors and how they influence women's access to Pap screening. Health-care providers need to consider the social determinants of health and the contexts of women's lives when considering why they are or are not accessing screening and need to individualize care, while offering consistent and convenient screening services. Building relationships with communities, creating safe spaces for screening, educating women, and providing trauma-informed and culturally safe care are also vital in encouraging Aboriginal women to access Pap screening. Improving Pap screening services for Mi'kmaq women requires multifaceted, culturally safe nursing approaches that are developed in partnership with the women themselves and their communities.

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Catherine MacDonald, Ruth Martin-Misener, Audrey Steenbeek, Annette Browne

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*Mi'kmaq Women's Experiences With Pap Screening*

Catherine MacDonald, Ruth Martin-Misener, Audrey Steenbeek, Annette Browne

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# Rethinking Case Study Methodology in Poststructural Research

Shan Mohammed, Elizabeth Peter,  
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Little consideration has been given to how case study might be used in post-structural research to explore power relations that constitute a phenomenon. Many case study scholars, most notably Robert Yin, adopt a postpositivist perspective that assumes the “truth” can be accessed through applying prescriptive and rigid research techniques. Using a discussion of Michel Foucault’s key theoretical ideas and the insights gained through a Foucauldian case study of people with advanced cancer who continue to receive curative treatment, the authors argue for the expansion of case study in poststructural inquiry. They propose that the use of poststructuralist case study is valuable because of the flexibility and comprehensiveness of the methodology, which allows for the exploration of a deeper understanding of the broader discourses that shape a phenomenon, as well as how power/knowledge relations shape the behaviours and perceptions of people. They also introduce the reflexive implications of post-structural case study research.

**Keywords:** case study, qualitative research, advanced cancer, poststructuralism, Foucault, reflexivity

*Résumé*

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## **Repenser la méthode de l'étude de cas à partir du post-structuralisme**

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Peu d'attention a été portée à la façon dont la méthode de l'étude de cas peut être utilisée dans le cadre de l'approche post-structuraliste pour étudier les relations de pouvoir qui structurent un phénomène. De nombreuses études de cas universitaires, en particulier celles de Robert Yin, adoptent une perspective post-positiviste qui postule l'existence d'une « vérité » à laquelle il serait possible d'accéder par l'application de techniques de recherche normatives rigoureuses. À partir d'une présentation des principales théories de Michel Foucault et d'une réflexion tirée d'une étude de cas foucauldienne portant sur des personnes atteintes d'un cancer avancé qui ont continué de recevoir un traitement curatif, les auteurs de l'article développent une augmentation pour un plus grand recours aux études de cas réalisées dans un cadre post-structuraliste. Ils font valoir que la méthode post-structuraliste confère une grande valeur aux études de cas en raison de sa souplesse et de son caractère englobant, et qu'elle permet une analyse plus approfondie des discours généraux donnant forme à un phénomène et des relations de pouvoir et de connaissance qui façonnent les comportements et les perceptions. Les auteurs traitent également des implications réflexives de la réalisation d'études de cas dans le cadre de l'approche post-structuraliste.

**Mots-clés :** étude de cas, recherche, cancer avancé, post-structuralisme, Foucault, réflexivité

## **Introduction**

Case study is a methodological approach to empirical inquiry that explores a relatively bounded phenomenon in depth and examines the contexts under which this phenomenon occurs, particularly when the margins between context and subject are blurred (Yin, 2009). The study of cases is commonly used as a teaching technique in education, such as the training of health professionals (Stake, 2000). The examination of a patient case study, including exploration of the contexts that contribute to disease and surround the experience of illness, might be used to educate nurses about prevention and treatment. As a research tool, case study has an extensive history in both the social sciences and the health sciences (Sandelowski, 2011). Case study is often viewed as a methodology with broad research application since it is used in a variety of qualitative, quantitative, and mixed-method research (Flyvberg, 2006; Stake, 2000; Yin, 2009).

Within the field of qualitative research, case study has been described as a flexible methodology that has usability in different research paradigms (Luck, Jackson, & Usher, 2006). Case study has additionally been conceived of as a taken-for-granted methodology that is often invisible in qualitative abstracts and titles (Anthony & Jack, 2009). The lack of methodological guidance and the emphasis on postpositivist standardization in case study might discourage poststructural health researchers from employing this methodology. For qualitative researchers unfamiliar with case study, it may be challenging to conceptualize the ways that assembling a case can help to accomplish the aims of poststructural research — that is, to open up an understanding of power, knowledge, and discourse that constitute a phenomenon.

Much of the dominant methodological writings about case study tend to adopt a postpositivist perspective that assumes the “truth” can be accessed through applying prescriptive and rigid research techniques (Yin, 2009). Despite this trend, we call for the expansion of case study in poststructural research. We argue that the use of case study is a valuable methodological approach in poststructural research because it facilitates a deeper understanding of the social, political, and historical circumstances that shape a phenomenon and how power relations shape the actions and perceptions of people. To accomplish our aims, we first outline some of the key theoretical ideas of Michel Foucault, one of the most prominent thinkers in poststructural theory, and then show how case study aligns with a Foucauldian perspective. We then discuss insights gained through a poststructural case study of people with advanced cancer who continue to receive curative treatment. Our intention is not to outline a new approach for case study through suggesting systematic

methodological steps, but rather to explain how poststructural scholars can benefit from the utilization of case study. The reflexive implications of poststructural case study research are also discussed.

### **Case Study Within a Postpositivist Paradigm**

Postpositivism is a widely accepted research tradition that suggests that knowledge can be generated through the measurement of an objective reality that is yet to be uncovered by the researcher (Cresswell, 2013; Guba & Lincoln, 2005). In this research paradigm, knowledge is something that is viewed as the scientific or empirical “truth” until the emergence of new evidence proves otherwise (Guba & Lincoln, 2005). Since there are certain laws, theories, and hypotheses that govern the world in postpositivism, methodology in this worldview is focused on the manipulation of variables, quantitative measurement, and experimental designs (Creswell, 2013).

Robert Yin is the most prominent methodologist on case study, and is well cited across several academic disciplines, the business sector, and government-sponsored research. Yin’s (2009) understanding of case study methodology is strongly influenced by a postpositivist worldview, particularly his claim that case study can be used to access the empirical “truth” located in a phenomenon. Yin argues that the “core of scientific method” (p. vii) is not experimentation per se, but rather can be accessed through the analytical strategy called “plausible rival hypothesis” (p. vii). Since case study takes contextual factors into account, Yin views this approach as more empirically robust than experimental methods. The primary analytical strategy in case study is to eliminate competing rival explanations or other influences, such as threats to validity and investigator bias, in order to ensure that the main hypothesis is true; in this perspective, there is less room for multiple views of reality and less flexibility about what constitutes the “truth” in empirical inquiry (Yin, 2009).

Because of the dominance of Yin’s work in the methodological literature, qualitative researchers who work with poststructural theory and disagree with the assumptions of postpositivism may be discouraged from using case study methodology. We suggest otherwise and argue that case study approached from a poststructural orientation is a flexible and comprehensive methodology that yields an opportunity for in-depth exploration of the phenomenon under study. At the same time, we do not seek to fully discount the value of Yin’s work in this article. Yin offers helpful suggestions about key methodological decisions in the building of cases that have significant value across all forms of case study research.

## **A Poststructural Theoretical Perspective**

In this section we outline some of the assumptions of poststructuralism to provide a background for the reader who may be unfamiliar with this framework. Michel Foucault, the late-20th-century French philosopher, was influential in developing poststructuralist theory (also known as Foucauldian theory) as a challenge to the assumptions of structuralism. By viewing language and culture as a closed system of signs and other categories, structuralism is a worldview that explains human phenomena in terms of their underlying structures, or distinct social structures such as socio-economic class (Piaget, 1970). Opening up the rigidity of structuralism, Foucault (1966) is concerned with a genealogical understanding of the world, which examines the historical “conditions of possibility” that allow forms of knowledge to emerge or, conversely, to be suppressed. To Foucault (1984), a genealogy involves developing “histories of the present” with a focus on “the unstable ensemble of faults, fissures, and heterogeneous layers that threaten the fragile inheritor from within and from underneath” (p. 82). From this perspective, knowledge is not universal, essentialist, or inherent, but rather is viewed as something that is situational, discontinuous, and open to historical and political revisionism (Foucault, 1972; Guba & Lincoln, 2005). Poststructuralism posits that knowledge, objects, individuals, and relationships have multiple meanings that shift with various contexts or different historical locations.

Foucault (1972) defines discourse as a system of thought composed of different patterns of action, practices, ideas, beliefs, and attitudes that systematically construct the objects of which they speak. Discourses exist under what Foucault (1972) calls “positive conditions of a complex group of relations” (p. 45). Foucault (1972) uses the term “discursive relationship” (p. 46) to denote the group of relations that discourse must establish “in order to speak of this or that object, in order to deal with them, name them, analyse them, classify them, explain them” (p. 46). As opposed to the notion of universal “truth” in postpositivism, in poststructuralism each discursive situation has its own politics of truth that dictate what some consider to be true and false knowledge (Foucault, 1980). Certain systems of thinking or discourses (for example, scientific discourse) are seen as accepted and dominant ways of understanding the world, whereas other types of discourse are viewed as less credible (Packer, 2011). Discourses are not limited exclusively to systems of knowledge, but often shape people’s thoughts, perceptions, attitudes, and behaviours (Foucault, 1976). People often alter their actions in order to fit certain norms and behavioural expectations that they internalize to police themselves (Packer, 2011).

Foucault made significant theoretical contributions to the notions of power and knowledge. These ideas are so aligned in poststructuralism that they are collapsed into the single term *power/knowledge* (Foucault, 1976; Mansfield, 2000). To Foucault, knowledge is enmeshed in relations of power; it regulates the social conduct of individuals and their bodies through various practices. Rejecting the idea that power is enforced from above, Foucault (1976) suggests that power is relational; people exercise power from innumerable points and power is located everywhere because it “comes from everywhere” (p. 93). At the same time, Foucault (1976) suggests that “where there is power, there is resistance” (p. 95), and this resistance is never in a position of exteriority in relation to power. People exercise resistance at multiple points throughout the web of power relations in dispersed and nuanced ways that are difficult to locate (Foucault, 1976).

In place of an essentialist and universal identity, Foucault (1976) describes how the self is composed of multiple subjectivities that exist concomitantly within one individual and shift with changing social locations. Selfhood does not exist outside of being subjected; there is no self without being a subject and the self is always constituted by the production of discursive systems (Mansfield, 2000). Power/knowledge also constitutes our subjectivities. The self is socially constructed through the interplay of multiple forms of power/knowledge in multiple locations (Foucault, 1976). Both the individual subject and the elements that make up our individuality, such as our gestures and use of language, are effects of power (Mansfield, 2000).

The constitution of people’s subjectivities is not shaped just by external forces; Foucault (1988) uses the phrase “technologies of the self” (p. 18) to describe a form of self-constitution. These techniques permit individuals to “effect by their own means or with the help of others a certain number of operations on their own bodies and souls, thoughts, conduct, and way of being” (Foucault, 1988, p. 18). Individuals employ such techniques to transform themselves “in order to attain a certain state of happiness, purity, wisdom, perfection, or immortality” (Foucault, 1988, p. 18).

### **The Argument for Poststructural Case Study Methodology**

In this section, we discuss how case study is a helpful methodology for accomplishing some of the research aims of poststructuralism. Some have suggested that case study is not even a methodology because it has been described as a simple data-collection plan (Gerring, 2004) and because all forms of qualitative research eventually become the study of cases (Sandelowski, 2011). We take the position that case study is a methodol-

ogy because it is a qualitative research design with a well-established set of procedures (Creswell, 2009), which lends itself to poststructural inquiry. Moreover, there is a longstanding tradition between poststructural thought and the study of cases (Flyvberg, 2006). Foucault often looked to historical cases, such as his examination of the prison system in Western Europe or the psychiatric management of mental illness, as a way to develop his ideas about politics, power, and the body.

Although Yin (2009) does not fully define “context” as a conceptual idea, case study aligns with a poststructural approach because they are both concerned with the indistinct boundaries between the phenomenon and the contexts that constitute it. Whereas Yin (2009) might be concerned with the contexts that permit a researcher to test a rival hypothesis, poststructural researchers use case study to explore the discursive contexts that shape a phenomenon. Moreover, because the process of building a case allows and even encourages the collection of multiple sources of data (Stake, 1995; Yin, 2009), it allows poststructural researchers to select strategically what kind of data will be collected and who might be interviewed. One of the important aims of poststructural inquiry is to examine critically how people’s patterns of thinking and action are shaped by broader discourses. Cases can be collected that include viewpoints from multiple social actors and data sources from multiple levels, such as the local and personal, but also the institutional and social. Yin (2009) describes the importance of data triangulation, also described as the “convergence of evidence” (p. 117), so the overall proposition of the case study is supported by multiple forms of evidence. In comparison, poststructural researchers might use multiple sources of data to consider tensions between social actors or discourses.

Another important aim of poststructural inquiry is to consider how power/knowledge relations constitute and operate with a research phenomenon. Case study is focused on the examination of a relatively bounded phenomenon and a limited number of events and conditions and their relationships (Dooley, 2002). Since cases represent both positive and negative practices, the collection of a case might include the setting, the people involved, events, problems, and conflicts (Dooley, 2002). Case study facilitates the exploration of complex and diffuse types of relationships and patterns that are present in case-based data (Dooley, 2002; Stake, 1995). As opposed to a simple interview study, case study methodology facilitates the examination of multiple relationships among different types of participants from different social positions, documents, or observational data. Since case study is concerned with how participants might function and act within limited contexts (Stake, 1995), this methodological approach also lends itself well to examining the nuanced ways that people resist power relations.

## **A Poststructural Case Study of Advanced Cancer Treatment**

### *Assembling Cases and Collecting Case Data*

To illustrate how a poststructural case study is possible, we describe our experiences conducting a case study of people with advanced cancer who continue to receive curative treatment. As we have stated, our intention is not to outline a rigid or systematic scheme for using case study methodology in poststructural research, but rather to describe some of the steps that we undertook in the particular context of our study because the ontological and epistemological aims of poststructural inquiry resist a standardized methodological approach.

The research phenomenon was patients' search for life extension through the search for biomedical and potentially curative treatment, despite the diagnosis of advanced, life-limiting cancer. Curative treatments are defined as oncological therapies, such as second-line chemotherapy or experimental trials, that are intended to eliminate cancer but may not improve the prognostic outcome of metastatic cancers and may lead to harsh physical side effects. Our study was guided by two research questions. The first was *How do discourses constitute the search for life extension through biomedical treatments for those with advanced cancer?* Building on this first question, the second was *What kinds of subjectivities are produced by the discourses in operation when individuals with advanced cancer seek life extension through biomedical treatments?*

We classified the cases as radical or atypical (Baxter & Jack, 2008; Flyvberg, 2006; Stake, 2000) in the sense that the search for curative treatment is often viewed in clinical practice as a problematic activity because it may call attention to patients' discontentment with their care, disputes with professionals, and barriers to acknowledging the closeness of death. Because radical cases involve social actors that are not obvious to an outside observer and encompass exceptional ideas and practices, as well as the shared norms and common standards of practices being disrupted in the phenomenon under study (Flyvberg, 2011), they are more comprehensive than representative cases.

The bounded nature of case study methodology encouraged us to stay focused on our research topic and to generate rich information to answer our research questions (Sandelowski, 2011; Stake, 2000; Yin, 2009). Cases were thus focused on data related to the search for oncological treatment in the later stages of cancer treatment, as opposed to initial diagnosis or the overall experience of illness. Since the perspectives of people with advanced cancer were largely missing from the literature on this research topic, cases began with interviewing of participants with advanced cancer and then moved outwards to include interviews with other social actors, examination of documents, and field observations.



Using snowball sampling (Browne, 2005), we asked participants with cancer to identify which people they perceived to be key in their search for curative treatment. To obtain multiple perspectives on the search for curative treatment, we chose as our study participants seven patients, five family members, two oncologists, three palliative care physicians, two oncology nurses, and an unlicensed natural healer. Participants with cancer were college- or university-educated, in their mid-thirties to mid-seventies, and originally from a variety of countries such as Belgium, Canada, Iran, Korea, and Nepal.

Case study methodology is associated with collecting multiple types of data (Stake, 1995; Yin, 2009). As documents often record an important “technology of power” (Hodder, 2000, p. 703), the analysis of documents facilitated a deeper examination of the discourses and subjectivities in operation. We asked participants to identify what documents they used in their search for curative treatment. Documents included Web sites, self-help books, pamphlets, magazine articles, self-made graphs of medical records, and self-written summaries of illnesses. Because we wanted to consider some contextual and setting-specific influences (Dooley, 2002), we conducted approximately 5 hours of field observation. Although we asked them during interviews, participants with cancer did not give us permission to attend any formal appointments with health professionals. Most participants did not identify any observable events (for example, public lectures or information sessions) related to cancer treatment that they were planning to attend.

### *Analyzing Case Study Data From a Poststructural Perspective*

To analyze our findings we drew on both Foucauldian discourse analysis and analysis of case study in general. Yin (2009) recommends that theoretical propositions influence the analysis of the cases. In addition, Yin (2009) states that there are “few fixed formulas or cookbook recipes” (p. 127) to guide case study analysis. Paralleling this approach, we employed a flexible and iterative approach to our analysis that Frost et al. (2010) propose is needed to embody the “spirit of poststructural inquiry” (p. 444). While much has been written about discourse analysis (Yates & Hiles, 2010), less consideration has been given to how this analytical approach might be intertwined with case study methodology.

A primary concern of case study research is understanding the relationships, complexities, and problems within an individual case (Stake, 1995). As we progressed through the analytical process, we began to consider the conceptual differences and similarities of the data between individual cases. Rather than look for the central “truths” in our analysis, we considered the fluidity and inconsistency of meanings, which are characteristic of poststructural thinking. To locate discourses, we exam-

ined the different practices of participants in searching for curative treatment (Frost et al., 2010). These practices included various techniques to obtain knowledge and shape the self, along with a variety of negotiation strategies and assertive communication styles. To locate subjectivity in the data, we considered how participants enacted various discourses through practices of power, agency, and resistance (Hook, 2001). We considered the ways in which participants with cancer shaped their lives, identities, ways of conduct, and thoughts as expressions of subjectivity that were also associated with locating curative treatment (Yates & Hiles, 2010).

### ***A Representative Case***

To demonstrate the application of case study methodology to poststructural inquiry, we briefly present the data and study findings through discussion of a representative case. A pseudonym is used and the case details are obscured. Jean, a man in his late thirties, was diagnosed with cancer several years ago that progressed to advanced disease with metastases to multiple sites in his body. He completed several unsuccessful rounds of chemotherapy and radiation and was seeking experimental oncological treatment at the time of the interview, in addition to trying alternative treatments such as positive thinking, meditation, and mindfulness training. Well educated with a university degree, he previously worked in the information technology industry. He was married with no children, was currently not employed, and experienced pain and fatigue. Data collected in his case included two interviews with Jean, one interview with his spouse, one interview with his palliative care physician, and several books and articles that he referenced in his search for curative treatment.

Drawing on his occupational background, Jean often brought mainstream scientific journals and medical textbooks, mass media articles, and alternative medicine reports to consultations with oncologists in an attempt to open up a discussion about additional curative treatments. Oncologists often dismissed the types of information presented during the encounter, claiming that the studies were not rigorous or were conducted on a population that was not specific to Jean's cancer. Not having his self-initiated research acknowledged led to both frustration and suspicion:

*The reason why I do all this research is because doctors don't, quite often. They don't get together and they don't consider everything — all the drugs that you're taking. And if you don't educate yourself on it, you could be dead.*

Despite the social prestige of biomedicine, Jean often assertively questioned the authority of his oncologists by critiquing the merits of their

knowledge and skill in treating his advanced cancer. In this quote, he describes the ultimate consequence of not taking personal ownership of one's treatment through researching one's disease — that is, death. Later in his interview, Jean described multiple strategies he used when discussing curative treatment with oncologists, such as asking pointed questions, requesting information about their training, and even informing them that he would double-check their treatment suggestions by getting second opinions from different physicians.

Although we were unable to interview Jean's oncologist, his palliative care physician described the challenges of working with patients who bring information about curative treatment to the medical encounter:

*They read all this stuff and they don't know how to put it into context. So it's helpful that patients are proactive and go on the Internet and they ask questions. Um, but sometimes you do spend a lot more time re-educating them [laughter], you know, sort of redirecting that knowledge.*

This physician reported that an important part of the clinical role is to re-educate, redirect, and re-contextualize the information that patients bring forward. Later in the interview, the physician described the sophisticated formal training and advanced clinical skills needed to work in oncology. From this perspective, medical knowledge, a type of legitimized understanding, may be somewhat inaccessible to a lay audience and is often the dominant form of knowledge when determining whether or not a cancer treatment is provided.

The documents that Jean accessed in his search for curative treatment often supported the assertive questioning of health professionals. One of Jean's peer-run patient support groups operated a Web site with a feature article titled "Questions for Your Doctor," which included a suggested list of questions for patients and families to ask their oncologists. In addition to more obvious questions about side effects, the article included bolder questions probing the physician's clinical skills and professional expertise, such as "How many of these procedures have you done?" and "What is your success rate in terms of getting rid of the cancer and minimizing side effects?" From this perspective, the degree to which cancer responds to treatment might depend on the quality of the oncologist and people with cancer should have the right to hold oncologists accountable for their unique healing capabilities.

The overall results of our analysis suggest that the search for life extension through curative treatment is constituted by multiple discourses and that multiple subjectivities are produced by these discourses. For the purposes of this article, and only because we present the results from one case, we discuss one discourse and two subjectivities.

See Mohammed, Peter, Gastaldo, and Howell (in press) for a more complete report on our findings.

The results of our analysis suggest the emergence of the *discourse of self-care*. Originating in response to the dominance of biomedicine's claim on legitimized knowledge, this discourse shaped certain modes of conduct, attitudes, and everyday practices that participants took up in order to generate their own curative possibilities despite having late-stage disease. This discourse was defined by participants' active use of bio-scientific knowledge and their manipulation of how treatment is administered in the cancer-care system as emergent practices of this discourse. The discourse of self-care threatened to dislocate the traditional hierarchical arrangements of bio-scientific knowledge and other treatment practices set forth by biomedicine. The conceptualization of this discourse in our study was facilitated by case study methodology. Having divergent viewpoints from participants within a case, which were limited to the search for curative treatment as a research phenomenon, led to the emergence of different discursive patterns and tensions in our analysis.

The discourse of self-care produced two types of subjectivity that we discuss in this article: (1) *the cancer expert subject*, and (2) *the mistrusting subject* (Mohammed et al., in press). The rise of the emergent discourse of self-care, which was characterized by the practice of challenging the authority of physicians, also provided the conditions for individuals to take on more assertive knowledge roles such as Jean's role in carrying out extensive research on cancer treatment. The cancer subject moved beyond being merely an informed individual to assume an expert role with certain knowledge and therapeutic abilities that were not only on a par with those of physicians but, from the perspectives of some participants, often surpassed the expertise of physicians (Mohammed et al., in press). The cancer expert subject often made self-researched knowledge claims about treatments as a way to resist the authority of certain physicians.

The discourse of self-care also encouraged participants to invest in their own capacities to generate the possibilities of life extension, thereby downplaying the need to trust health-care providers in clinical encounters. The mistrusting subject focused on discussions and materials that questioned the dominance of biomedicine (Mohammed et al., in press). One key rationale of the mistrusting subject was that one should view biomedicine with caution and recognize that it may not have any more power over cancer than patients do, since it cannot effectively cure certain forms of metastatic cancer. Both types of subjectivity discussed in this article highlight the role of Foucault's (1988) technologies of the self in the formation of particular types of subject who are

strong enough to push for life extension assertively, despite the debilitating and terminal nature of their disease (Mohammed et al., in press).

### **Using Case Study to Examine Power/Knowledge Relations**

Case study methodology was helpful when considering the multiple ways that power/knowledge operated throughout the data. In particular, collecting multiple forms of data across the cases allowed us to consider the practices of patients and family members and then compare and contrast them to the perspectives of health professionals. Moreover, collecting documents that were located both within and outside the biomedical mainstream led to creative insights into the numerous ways that power/knowledge relations shape social discourses about curative treatments.

Our analyses of power/knowledge relations highlight Foucault's conceptual claim that people's resistance takes place in various densities and occurs in an irregular and sometimes unexpected fashion. For instance, before we collected the data we assumed that participants with advanced cancer would be too vulnerable or would lack sufficient knowledge to negotiate their treatment. After collecting and analyzing the data we were surprised by the intensity with which participants resisted the dominance of biomedical authority yet still wanted to receive oncological treatment (Mohammed et al., in press). Although information provided by health-care providers was perceived as essential, knowledge obtained from a wider assemblage of sources (for instance, one's own body, self-navigation of cancer research, informal social networks, and the mass media) was also given a certain level of importance. In their practice of resistance, participants did not necessarily view the knowledge obtained from health professionals as inherently more credible, but rather used self-obtained understandings to compare, confirm, and sometimes discredit the knowledge obtained in the clinical encounter (Mohammed et al., in press).

### **Reflexivity in Poststructural Case Study Research**

A poststructural theoretical framework often considers how the subjectivities of the researcher impact the research process, resulting in a concern with speaking for others and attention to the power relations inherent in knowledge production (Choi, 2006; Packer, 2011). As it deconstructs the authority of the researcher, reflexivity helps to disclose how power/knowledge relations and dominant ideologies operate through the research process (Choi, 2006). We worked from the poststructural position that the researcher is the primary instrument for data collection. The awareness of our own subjectivities was an important facet of promoting methodological rigour (Manias & Street, 2001).

In order to bring more openness and sensitivity to the research process, the first author (SM) had to consider his previous personal relationships with people who have advanced cancer within two contexts: his familial history and his clinical work as an oncology nurse. The first author conducted all the data collection in this study. Considering the effects of his positionality was the beginning of the reflexive process. For instance, he had to continually reflect on his previous positions as “nurse” and “caregiver” with the emerging positions as “researcher” and “producer of knowledge.” He had to consider reflexively how the boundaries between these positions became blurred and what this blurring meant to the study process. For example, when witnessing the suffering of participants, he had to consider carefully how his instinct as a clinician to intervene in a therapeutic sense affected the interview process. His concern with protecting and “saving” cancer patients, a role oncology nurses are often socialized to take up, occasionally led to hesitancy to probe deeper about certain emotionally laden topics (for example, funeral preparations), despite the importance of these topics to the study aims. Through continual reflection and debriefing with his co-investigators in the study, he developed a better awareness of his role as researcher, and this translated into a deeper but still respectful exploration during interviews.

As researchers who are all nurses using a critical perspective, we developed an awareness of our location within the very discourses (for example, biomedicine) that we aimed to investigate and problematize. McCabe and Holmes (2009) argue that reflexivity in poststructural inquiry is more than promoting research validity, but is also about acknowledging the “nature and function of power” (p. 1524) of participants and researchers. As a result, we were cognizant of how our authoritative expertise as clinicians and researchers vested us with a sense of power, which could have either liberating or repressive effects (McCabe & Holmes, 2009). The interview could become repressive in that participants could find themselves in a discursive field where they might see themselves as deviant or bad (McCabe & Holmes, 2009). To limit this possibility, the first author attempted to cultivate an open and respectful research environment where participants would feel free to disclose their perspectives without judgement. The research interview can be a liberating experience because it isolates the individual from the judgement of society (and the health-care team) and provides a space where one can explore different and often controversial perspectives (McCabe & Holmes, 2009).

In the interests of openness and transparency, the first author shared his identity as an oncology nurse with participants and sometimes discussed with them how this might have affected the research relationship.

Yet this raised concerns about whether participants modified the information they were willing to share. Some participants, concerned about upsetting the social arrangements that supported their ability to access treatment, may have been hesitant to bring up certain subjects because of his professional background. For instance, some interviewees avoided naming particular health professionals when critiquing their care. For other participants, his identity as a nurse did not stop them from expressing their frustrations and thoughts about challenging health professionals. According to the arguments of McCabe and Holmes (2009), these individuals were able to use the interview as “a vehicle for reflexive thought and action” (p. 1523) in order to examine their own stance against normative values. Through reflexive engagement with the researcher, these individuals were able to explore particular power structures and dominating discourses that greatly impacted their search for treatment (McCabe & Holmes, 2009).

In order to promote reflexivity, we had to continually review the multiple assumptions that we brought to the research process (Patton, 2002). In particular, we had to reflexively examine how our previous assumptions about health-care teams and the dominance of physicians, which were generated by our own clinical work, might have coloured our examination of the power dynamics between patients and physicians. When participants described their concerns about physicians’ inability to be attentive, we could empathize with the challenges of exercising power in a relationship with physicians. When we reflected back, we wondered whether we were reverting back to being resentful of the power of physicians, which nurses are sometimes socialized to do in the clinical world. For the first author, this tendency resurfaced during his interview with an oncologist. Because the oncologist seemed rushed and unwilling to engage in a deeper conversation about practice, the interview provoked a sense of annoyance in the first author and fed into his assumption that physicians are often dismissive, a claim supported by certain participants. Using reflexive discussions as a research team, we concluded that pejoratively assigning a normative view of physicians and rendering the patient and family powerless in the clinical encounter is antithetical to a poststructural approach. Through a reflexive process, we became more attuned to seeing how constrained physicians were in their clinical role, how participants exercised their own practices of resistance to counter biomedicine, and the discursive interrelationships that constitute the dynamic between these two groups.

Examining our personal assumptions about death due to cancer was crucial because it helped us to understand how we might have shaped our own selves through the research process, an important facet of reflex-

ivity (Patton, 2002). We occasionally found ourselves mesmerized by participants' confidence and dedication to life extension, which was likely a social performance of their faith in their own curative abilities. On occasion, we found ourselves caught up in the wider culturally endorsed assumption that one can transcend death by adopting certain subjectivities. Reflexively considering our assumptions about death helped us to better engage with the actual possibilities of dying in this work, which was a prevalent theme in the findings.

### **Limitations**

The major limitation of our discussion in this article, which may affect its applicability to future research, is our reporting of only one research study in one clinical area. Both poststructuralism as a theoretical framework and case study as a research methodology have many possibilities for qualitative researchers. Although we have discussed the suitability of this methodology for a poststructural case study in the context of advanced cancer, case study researchers need also to conceptualize and share, in future articles, how they have previously used this methodology. Through these developments, case study can perhaps emerge from its current status as a taken-for-granted methodology.

### **Conclusion**

In this article we have critically examined Yin's (2009) postpositivist orientation to case study methodology. We have suggested that the ontological and epistemological assumptions of this dominant view might deter qualitative researchers who use other theoretical perspectives, such as poststructuralism, from using case study methodology. As a counterpoint, we call for the expansion of case study methodology in poststructural research. After briefly mapping out Foucault's key theoretical assumptions, we have argued that the use of case study within a poststructural approach is valuable because of the flexibility and comprehensiveness of the methodology, which allows for exploration of the broader discourses that shape a phenomenon as well as the power/knowledge relations that shape people's behaviours and perceptions. By presenting an example of a case study of people with advanced cancer who continue to receive curative treatment, we have illustrated our use of poststructural case study. We have also explored the complexities of reflexivity to poststructural case study. Case study methodology has the potential to effectively support qualitative studies that examine the roles of power, knowledge, and discourse on health and disease.



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## *Rethinking Case Study Methodology in Poststructural Research*

Shan Mohammed, Elizabeth Peter, Denise Gastaldo, Doris Howell

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