TOWARDS COLLABORATIVE PARTNERSHIPS IN THE GOVERNANCE OF CANADIAN HEALTH RESEARCH ETHICS

by

KIMBERLY TAYLOR

B.A., The University of Victoria, 2004

A THESIS SUBMITTED IN PARTIAL FULFILLMENT OF THE REQUIREMENTS FOR THE DEGREE OF

MASTER OF ARTS

in

THE FACULTY OF GRADUATE STUDIES

(Philosophy)

THE UNIVERSITY OF BRITISH COLUMBIA

(Vancouver)

October 2009

© Kimberly Taylor, 2009
Abstract

The most valuable resources common to all human health research endeavors are those individuals who voluntarily subject themselves to potential risks for the hope of improving human health and advancing medical science. Although human subjects’ participation in health research is acknowledged as instrumental to our understanding of health and illness and thereby an interest to society, the role of human subjects in determining how research is conducted, monitored, and, if successful, translated into health care, has been minimal until fairly recently.

This paper argues from both moral and political grounds that meaningful and effective partnerships between human subjects and research communities ought to be mandated as a standard practice within Canadian policy guidelines for all health research initiatives that are conducive to collaborative engagements. By rendering decision-making a truly democratic endeavor, with the implementation of collaborative relationships between scientific and human subject communities with a similar framework to the Tri-Council Policy Statement and the Canadian Institute of Health Research’s guidelines on research involving Aboriginal People, a more transparent and accountable health research system will ensue. As a result, I argue, the health research enterprise in Canada as a whole will engender greater public trust.
Table of Contents

Abstract .......................................................................................................................................................... ii
Table of Contents ......................................................................................................................................... iii
Acknowledgement ........................................................................................................................................ iv
Dedication ..................................................................................................................................................... v

1. Introduction ............................................................................................................................................... 1

2. Human Subjects: From Objects to Agents of Science ................................................................. 5
   2.1 Pre-WWII: Subjects as Objects ......................................................................................................... 5
   2.1.1 The Clinician-Researchers’ Duty to Human Subjects ................................................................. 7
   2.1.2 The Growth of Health Research ................................................................................................. 9
   2.2 Post-WWII: Subject as Passive Agent ............................................................................................ 11
   2.2.1 Bureaucratization of Research: legislation & moral codes of conduct .................................... 17
   2.3 An Era of Activism: Subject as Active Agent ............................................................................... 21
   2.4 Conclusion ....................................................................................................................................... 25

3. Research Partnerships: The PXE International and Breast Cancer Foundation as Examples ........ 26
   3.1 Participatory Methods of Inquiry ..................................................................................................... 27
   3.2 Research Partnerships: PXE International .................................................................................... 30
   3.2.1 Founding PXE International ....................................................................................................... 31
   3.2.2 PXE International Blood and Tissue Bank ................................................................................ 34
   3.2.5 Biobanks and Research Ethics ................................................................................................... 35
   3.2.6 PIBTB and Research Ethics ......................................................................................................... 39
   3.3 Research Partnerships: Breast Cancer Advocacy ........................................................................... 42
   3.4 Conclusion ....................................................................................................................................... 46

4. Democratizing Science Research .................................................................................................... 48
   4.1 Post-positivist Theory and Democracy .......................................................................................... 49
   4.1.1 The Value of Lay Expertise ........................................................................................................ 50
   4.2 Deliberative Democratic Theory & Practice .................................................................................. 53
   4.2.1 Types of Deliberative Democratic Participation ........................................................................ 55
   4.3 Deliberative Democracy & Health Research ............................................................................... 57
   4.5 Conclusion ....................................................................................................................................... 59

5. Ethical Challenges and Recommendations .................................................................................... 61
   5.1 Challenges of Collaborative Research .......................................................................................... 63
   5.1.1 Reflections on Methodological and Ethical Challenges ............................................................ 68
   5.2 Assessing Collaborative Health Research ..................................................................................... 74
   5.3 Conclusion: Recommendations for the Canadian Context ............................................................ 77

BIBLIOGRAPHY ........................................................................................................................................ 82
Acknowledgments

I wish to acknowledge and thank my supervisor, Dr Michael McDonald, first and foremost for his guidance, patience, and encouragement throughout the development of this thesis paper. His vast knowledge and experience in the area of health research ethics have been an invaluable contribution to my Masters education in Applied Ethics, for which I am truly grateful. I also wish to acknowledge and thank my committee member, Dr Scott Anderson, for his insightful advice and comments upon review of my thesis paper. I also thank my Philosophy graduate advisor, Dr Alan Richardson, for his assistance in guiding me through the completion of my Masters of Arts degree.

Lastly, I would also like to thank my colleagues and mentors on the CIHR funded research project “Centring the Human Subject in Health Research: understanding the meaning and experience of research participation,” at UBC’s Centre for Applied Ethics– Dr Susan M. Cox, Dr Anne Townsend, Dr Darquise Lafrenière, and Nina, as well as Dr Patricia Kaufert, Dr Joseph Kaufert, and Lisa Labine at the University of Manitoba’s Department of Community Health Science, for allowing me the opportunity to learn from your wide range of expertise in Applied Ethics.
Dedication

I would like to dedicate this thesis paper to my friends and family that supported me throughout this rather lengthy, yet rewarding process. I would especially like to dedicate this paper to Matt Jameson, who I cannot begin to thank enough for his endless support and encouragement from beginning to end.
CHAPTER I
Introduction

Progress is by our choosing an acknowledged interest of society, in which we have a stake in various degrees; science is a necessary instrument of progress; research is a necessary instrument of science; and in medical science experimentation on human subjects is a necessary instrument of research. Therefore, human experimentation has come to be a social interest.
(Jonas, 1969)

The most valuable resources common to all human health research endeavors are those individuals who voluntarily subject themselves to potential risks for the hope of improving human health and advancing medical science.¹ Without human subjects² researchers could not have attained the wealth of knowledge that continues to drive the rapidly changing course of scientific research and, subsequently, our understandings of health and illness. Although human subjects have been instrumental to advancements in health research for centuries, by voluntarily (or involuntarily) contributing their persons to experiments that have the potential to significantly affect (whether negatively or positively) their physical, psychological, and/or social wellbeing, the role of human subjects in determining how research is conducted, monitored, and, if successful, translated into health care, has been minimal until fairly recently.

² The United States government defines a "human subject" as: "a living individual about whom an investigator… conducting [a systematic investigation designed to develop or contribute to generalizable knowledge] obtains (1) data through intervention or interaction with the individual, or (2) identifiable private information." 45 Code of Federal Regulations § 46.102(f).
The degree to which human subjects ought to be involved in decisions affecting the agenda and practice of health research initiatives has been a topic of much debate, one that extends well beyond the reaches of this paper. Although I cannot address all of the issues surrounding the suitable role for human subjects in all types of health research endeavors, I will argue that since human experimentation is an acknowledged interest of society, as Jonas eloquently states, meaningful and effective partnerships between human participants and health research communities ought to be an essential requirement of all health research initiatives that are conducive to collaborative engagements. I will argue this case from both moral and political grounds. From a moral perspective, research involving collaborative means of engagement is fundamental to the principle of respect for the autonomy of human subjects. From political grounds it is widely held in democratic theory that deliberations amongst a range of experts from diverse epistemological backgrounds will begin to shift the culture of health research towards that of a more democratic endeavor, thereby further legitimizing the health research institution and, subsequently, increasing public trust in the enterprise as a whole. As a body whose aims are to advance societal interests and, as noted above, since health research is an acknowledged interest of society, it follows that it is the state’s responsibility to ensure that meaningful and effective collaborations between research subjects and researcher communities are maintained. Thus, I shall conclude with the argument that Canadian policy directives ought to require that collaborative

---

engagement measures be implemented, in varying degrees, into all types of health research in order for ethics approval to be granted.4

To begin unpacking this argument I present a historical overview of the human subject’s role in health research in Chapter two. Through an examination of the place of human subjects across the history and development of health research ethics I discuss how the human subject has evolved from an “object” towards an active agent and “participant” of health research. Chapter three involves an examination of participatory methodologies and highlights two unique cases in which research subject have taken on successful partnerships with researcher communities. The first case is the non-profit group PXE International whose aim is to support and initiate research for the rare genetic disorder Pseudoxanthoma Elasticum, or PXE. The second discusses the historical case of breast cancer advocacy groups in ushering in a new wave of active patient-subjects, a group who remain a powerful force in the political arena of the health research enterprise today.

Chapter four shifts gears and discusses the progression in recent years towards the democratization of science. Through a discussion of the postpositivists’ theory of participatory democracy and experiential expertise, I argue that lay participants and, more specifically, human subjects add an invaluable dimension to scientists’ and health researchers’ expert knowledge on a given disorder. This chapter ends with an examination of deliberative democratic theory and practice, where I argue for more deliberative democratic engagements with human subjects in health research practices

4 C. Weijer, personal communication with M. McDonald.
in order to render a transparent and accountable decision-making processes and, as a result, the research institution as a whole may warrant greater public trust.

Chapter five concludes this paper with a reflection on some of the challenges of collaborative partnerships that may threaten the legitimacy and success of a health research initiative. Following this examination I offer some reflections on possible solutions to these methodological and ethical challenges. I then turn to look at some of the literature that has been published to date on the assessment of public participation in health research endeavors. Lastly, I conclude with some recommendations for moving forward towards a more collaborative approach to health research in the context of Canadian society.
CHAPTER II
Human Subjects: From Objects to Agents of Science

“In [1998] the NHS research and development programme recommended a ‘firm commitment to involving consumers in research—not as "subjects" of research, but as active participants in the process of deciding what research should take place, commissioning research, interpreting the results, and disseminating the findings.” (Boynton, P. 1999)

The purpose and aim of this chapter is to shed some light on the position of human subjects in the health research enterprise. I will discuss some of the key shifts in the role of human subjects in the context of a changing landscape of research ethics over the course of the past century. I begin by outlining some of the major developments in research ethics that have helped define the rights of human subjects and shape the professional and moral duties researchers and other stakeholders in health research owe to them. Beginning with the notion of the human subject as a material “good,” or commodity of research, I will discuss how the view of the human subject has begun to shift towards that of an active, autonomous agent and, more recently in the last two decades, has begun to take on a more prominent role as partners in certain sectors of the health research enterprise. As the above quote indicates, recognition of this shift in the position of the role of human subjects in research culture did not occur until fairly recently.

2.1 Pre-WWII: Subjects as Objects

Medical research did not begin to develop into the multi-billion dollar industry that drives innovations in preventative health, diagnostics, and treatment today until the later
half of the twentieth century. Prior to the Second World War health research initiatives were not considered a high priority on the political agenda for most countries (Moreno, 2001, p.11). Although medical experiments became increasingly more sophisticated throughout the nineteenth century, it was still for the most part a “cottage industry” in its early stages. In fact, the use of live animals was far more commonplace than human beings well up until the twentieth century (Rothman, 1999, p.20).

Long before HIV/AIDS activist groups began advocating for changes to standard operating procedures on research involving randomized clinical trials (RCT), animal activists united to demonstrate against medical experiments involving morally questionable research practices. Beginning in Britain in the nineteenth century, massive public demonstrations against controversial research practices that used live animals in non-therapeutic experiments ignited what became infamously known as the anti-vivisection movement (Brody, 1998, p.14). In support of such movements was British philosopher Jeremy Bentham whose position was grounded on an animal’s experience of physical pain rather than their rational capacity; as Bentham argued: “The question is not, can they reason? Nor, can they talk? But can they suffer?” (Brody, 1998, p.14). As a result of the anti-vivisection movement the British government passed the **Cruelty to Animals Act** in 1876, becoming the first national law to regulate animal research (Schuppli and McDonald, 2005, p.97). One derivative of legislating the use of animals in research, however, was an increased prevalence of human beings being recruited as “guinea pigs” for health research experiments.

Although the involvement of human subjects and the sheer number of scientific research endeavors increased exponentially throughout the nineteenth century with the
emergence of experimental medicine, medical research remained largely small-scale in scope. Typically, a single investigator would conduct their experiments on a small number of individuals, including the researchers themselves, their colleagues, families, and/or neighbors (Rothman, 1999, p.18-19). The experiments were primarily for therapeutic purposes and thereby morally justifiable on the grounds that the benefits of the research outcome would outweigh the risks to the patient-subjects involved. As such, regulation of the research enterprise remained in the hands of the clinical research community.

2.1.1 The Clinician-Researchers’ Duty to Human Subjects

Though it has been argued that human subjects were little more than tools for scientific pursuits, physician-researcher’s moral obligations to human subjects were considered as early as the thirteenth century. This is evident in the writings of influential Jewish physician and philosopher Maimonides. The Oath of Maimonides maintains that the physician-researcher ought to “always treat patients as ends in themselves, not as means for learning new truths” (Rothman,1999, p.19). Similarly, in physiologist Claude Bernard’s own teaching philosophy was an acknowledgement of the clinician-researcher’s moral responsibility: “Christian morals forbid only one thing, doing ill to one’s neighbor. So, among the experiments that may be tried in man, those that can only do harm are forbidden, those that are innocent are permissible, and those that may do good are obligatory” (Veatch, 1991, p.16). Nevertheless, regulations to ensure that the conduct of medical research was in accord with these moral principles were nonexistent. In fact, there were no substantive or procedural requirements of
investigators and institutions involved in health research to protect the interests of human subjects from potential harms ensuing from a research initiative. It was as Michael Polanyi refers to as a period of scientist self-governance where “the choice of subjects and the actual conduct of research is entirely the responsibility of the individual scientist, [and] the recognition of claims to discoveries is under the jurisdiction of scientific opinion expressed by scientists as a body” (Polanyi, 1951), p. 53. Thus, the moral integrity of health research was largely left to the discretion of the research community, which later led to the peer review system.

The therapeutic intent of much of the research that was being conducted in the early nineteenth century, as well as the paternalistic nature of the clinical-researcher’s relationship with his patient-subjects, granted researchers the authority to judge not only what sort of research was morally justifiable to pursue, but how protocols ought to be designed and, therefore, how research would affect the experience of human subjects involved. Most human subjects had little to no consultation with the clinical researcher about all of the potential risks and benefits participation in a particular research project may result and how the outcomes, if favorable, may be implemented into the delivery of their clinical care. The patient-subject was thought to be too vulnerable and incapable of making rational judgments regarding what was in their own best interests (Veatch, 1991, p.3). Questioning the authority of the medical professional and his competence in being able to adequately judge what was his patient-subject’s own best interest was against social norms (Rothman, 1999, p.19, 28-29). Factors including the power imbalance between the clinician-researcher and patient-subject, coupled with the assumed vulnerability and lack of competence on the part of the subject and an
inherent trust placed in the health researcher, all contributed to the medical community having full authoritative power concerning scientific research. To this end, the human subject’s role in the research endeavor was that of a passive “material” resource for the advancement of scientific knowledge and human health somewhat akin to chemical reagents in pharmaceutical research.

2.1.2 The Growth of Health Research

As research became increasingly sophisticated and clinician-researchers more educated in the art of scientific investigation involving human subjects, innovations in medicine and science rapidly advanced (Rothman, 1999, p.24). With new discoveries, such as germ theory in the 1890s, and advances in molecular genetics in the 1900s that led to breakthroughs in our understandings of inheritable diseases, came greater interest in the marketability of health research (Rothman, 1999). Science based medicine emerged in the early 1800s in the United States out of the Parisian empirical research school of thought, which encouraged large scale clinical and epidemiological studies, but it was the German government who made the greatest commitment to advancing health research (Callahan, 2003, p.13). By the end of the nineteenth century changes in the scale of medical research in the U.S. and Canada largely attributed to funding from private industry (Callahan, 2003, p.15). By the early twentieth century, American medicine had gained public prestige, increased professional competence, and a sound foundation in research, which led to an organized and effective campaign against disease and, in result, greater private industry and government funding allocated to health research (Callahan, 2003, p.17-18).
Greater stakes in health research as a highly profitable enterprise led to some scientists to “skirt the boundaries of ethical behavior in experimentation [by] elevating medical progress over the subject’s welfare.”\(^5\) In addition, a utilitarian line of argument was often used to defend morally questionable research, whether therapeutic or non-therapeutic, as being justifiable on the grounds that health research was essential for the greater good of society. The commonly held belief was that the risks associated with certain research protocols, which may cause injury or harm to a few individuals, would be insignificant in comparison to the potential benefits that cures and immunization from life threatening diseases would bring to mankind. This Utilitarian sentiment in support of large-scale health research initiatives became ever-increasingly widespread with wartime conditions in the early twentieth century and, as such, the “war on disease” ensued.

During the Second World War, wartime conditions on society created an urgent need for innovative research to help fight against some of the most fatal enemies: disease and illness (Rothman, 1999, p.32). Optimism that science-based medicine would alleviate human suffering and disease was an attitude held by both the medical and lay community. One physician went so far as to claim in an issue of the *Journal of the American Medical Association*: “In fifty years science will have practically eliminated all forms of disease” (Callahan, 2003, p.19). In times of war the balance between the individual and society’s interests tended to shift in favor of the public’s needs, thereby temporarily allowing for certain sacrifices to be made, whether voluntarily or forced, on

\(^5\) Rothman points out that there were instances in which the public opposed the blurring of ethical boundaries in health research. For example, the yellow fever experiments by American army surgeon Walter Reed who recruited his subjects without fully divulging all risks associated with the study, including the possibility of death. (Rothman, 1999, p.25).
certain populations for the sake of the greater good of society (Jonas, 1969, p.157).

Others argued against the prevailing ethos of the time that harbored strong Utilitarian sentiments. As Hans Jonas notes:

> Medical experimentation on human subjects falls somewhere between this overpowering case and the normal transactions of the social contract. On the one hand, no comparable extreme issue of social survival is (by and large) at stake. And no comparable extreme sacrifice or foreseeable risk is (by and large) asked. On the other hand, what is asked goes decidedly beyond, even runs counter to, what it is otherwise deemed fair to let the individual sign over of his person to the benefit of the “common good.” Indeed, our sensitivity to the kind of intrusion and use involved is such that only an end of transcendent value or overriding urgency can make it arguable and possibly acceptable in our eyes. (Jonas, 1969, p. 157)

To this end, interpreting the model of clinical research ethics during conditions of War must be seen through a lens that takes into account the culture of medical research in its given social context.

2.2 Post-WWII: Subject as Passive Agent

The most commonly cited turning point in the history of research ethics was the end of WWII and the increasing emergence of governance mechanisms to act on behalf of the interests of human subjects. Increased protectionist mechanisms for human subjects came in direct response to the devastating revelations of the Nazi medical experiments that were conducted involuntarily on vulnerable concentration camp prisoners. At the 1947 Nuremberg Military Tribunals, or United States v. Karl Brandt et al., official guidelines were drafted as a set of standards for judging Nazi physicians and scientists who had conducted biomedical experiments on concentration camp prisoners
The National Commission for the Protection of Human Subjects of Biomedical and Behavioral Research, 1979). The ensuing Nuremberg Code was comprised of ten recommendations for the governance of ethical clinical research practices. The Nuremberg Code has since become the framework for which international governance standards on health research involving human subjects has been founded (Emanuel, 2003).\(^6\) The creation of the Code was one of the most significant assertions on an international level that formally acknowledged human subjects as autonomous agents who required respect for their persons from the scientific community through such requirements as voluntary consent agreements aimed to fully inform the potential subjects of the study procedures and any possible risks and benefits resulting from participation in the research initiative.

Although the recommendations of the Nuremberg Code were known to the international scientific community and were made aware to the general public through news reports, they were not systematically implemented into practice (Moreno, 1996, p.30-33). The Nuremberg Code was largely ignored within the North American medical community, purportedly because it was regarded as a document that was required to restrain particular immoral actors, namely the totalitarian regime of Nazi Germany (Emanuel, 2003, p.3). The idea that the Nazi experiments had any personal implications

\(^6\) The code was comprised of the following ten maxims: 1- voluntary consent, 2- experiment should yield fruitful results for the good of society, 3- experiment should be designed and based on the results of animal experimentation and knowledge of the natural history of the disease, 4- it should be conducted to avoid all unnecessary physical and mental suffering, 5- No experiment should be conducted where there is an a priori reason to believe death or disabling injury will occur, 6- the degree of risk to be taken should never exceed that determined by the humanitarian importance of the problem to be solved by the experiment, 7- proper preparations should be made to protect the experimental subject against possibilities of injury, disability, or death, 8- the experiment should be conducted only by scientifically qualified persons, 9- the human subject should be at liberty to bring the experiment to an end if he has reached the physical or mental state where continuation seems to him to be impossible, 10- The scientist in charge must be prepared to terminate the experiment at any stage, if he has probable cause to believe, that a continuation of the experiment is likely to result in injury, disability or death to the experimental subject.
for the reputable medical community of the Allied Nations was unimaginable (Faden et al., 1996). Two decades later, the World Medical Association adopted the Declaration of Helsinki in 1964 whose principles were directly influenced by the Nuremberg Code. Two major developments occurred with the Declaration of Helsinki; firstly, it allowed some flexibility on the notions of voluntary and informed consent, thereby allowing room for some incompetent individuals to be included in research and, secondly, it clearly distinguished therapeutic from non-therapeutic research (Childress, 2000, p. 351). Despite these early efforts to formulate standard practices in the medical research community and respect the agency of human subjects, self-policing by the clinical research community continued to be the norm.

This is evidenced by an astonishing amount of cases in which North American clinical researchers took part in highly questionable experiments that were exposed within popular media and scientific journal articles during the 1960s and 70s. Most notably, in 1966 Henry K. Beecher’s paper “Ethics and Clinical Research” documented a series of studies performed in prestigious universities, hospitals, government institutions, and private industry that showed evidence of unethical treatment of vulnerable human subjects within non-therapeutic research experiments. Beecher states in his introduction:

Evidence is at hand that many of the patients in the examples to follow never had the risk satisfactorily explained to them, and it seems obvious that further hundreds have not known that they were the subjects of an experiment although grave consequences have been suffered as a direct result of experiments described here. There is a belief prevalent in some sophisticated circles that attention to these matters would ‘block progress.’ But, according to Pope Pius XII, ‘science is not the highest value to which all other orders of values should be… subordinated. (Beecher, 1966, p.1354)
In addition to Beecher’s shocking revelations, other high profile cases citing instances of abuse against human rights were widely reported, many of which were conducted on vulnerable populations including women, children, and the mentally ill. One of the most famously cited studies to make research ethics history is the Tuskegee Syphilis Study that took place in Alabama, USA. The study began in 1932 and recruited over 400 men, most of whom were illiterate sharecroppers, to assess the natural course of Syphilis. Human Subjects were not informed of the research procedures, what they would be injected with by the research workers, nor were they informed of any of the risks associated with this study. When Penicillin went on the market in the late 1940s, a known treatment for syphilis, participants were not administered any medication. In fact, the study lasted until press reports about the study reached the secretary of the Department of Health, Education and Welfare in 1972.

Another pertinent case was the Jewish Chronic Disease Hospital experiments, where physicians injected live cancer cells into hundreds of unknowing patients beginning in 1963, which led to the infamous Hyman C. Jewish Chronic Disease Hospital court case (Katz, 1972). Furthermore, in the final report of the Law-Medicine Research Institute (LMRI) of Boston University conference on the topic “Concept of Consent in Clinical Research” in 1961, some researchers openly reported never seeking consent from their healthy subjects prior to their participation in non-therapeutic research (President’s Advisory Committee on Human Radiation Experiments, 1996, p.79). One researcher who attended the conference reflected on his involvement in a study that had administered hallucinogens to healthy subjects without their full

---

knowledge or consent as follows: “It wasn’t that we were Nazis and said, ‘If we ask for consent we lose our subjects,’ it was just that we were so ethically insensitive that it never occurred to us that you ought to level with people that they were in an experiment” (President’s Committee on Human Radiation Experiments, 1996, p. 79).

Yet perhaps the most shocking revelation was the government sponsored Radiation Experiments that took place on unknowing human subjects during the Cold War.

President Clinton established the Advisory Committee on Human Radiation Experiments (ACHRE) in April 1994 to investigate allegations of abuses of human subjects involved in government-sponsored research involving radiation over a period of three decades. Evidence supporting these allegations was collected by the ACHRE and proved that from 1947 until 1974 the U.S. government funded numerous dangerous experiments on uninformed subjects to test their physiological reactions when exposed to radiation. In a similar vein to early nineteenth century wartime conditions, the hope was that through large-scale investigations on a few hundred human subjects, progress would be made towards understanding the effects varying levels of atomic radiation exposure has on military personnel during or following combat during the Cold War period when atomic warfare was a looming threat.

The human radiation experiments began in 1947 when the Manhattan Project researchers and officials began the expansion of the government’s support of biomedical radiation research under federal contract (President’s Advisory Committee on Human Radiation Experiments, 1996, p.46). The Armed Forces Medical Policy Council (AFMPC) called for policy to be established for the use of human volunteers in experimental research at Armed Forces facilities on January 13th 1953. This led to the
Wilson Memorandum, issued on February 26th, 1953, that would be the first policy document to “render the research subject to the principles and conditions laid down as a result of the Nuremberg trials” (President’s Advisory Committee on Human Radiation Experiments, 1996, p. 59). The Wilson memo stipulated that written, witnessed informed consent would be required from the research subjects, the consent must be voluntary, the subjects have the right to withdraw from the research at any point, and that the research risks ought not have greater weight than the potential benefits. Although formal policies documented the need for informed consent and principles of beneficence in the Wilson Memorandum, evidence shows that that such standards were largely not made available to or followed through by the investigators and therefore rarely realized in practical terms (President’s Advisory Committee on Human Radiation Experiments, 1996, p. 64).

ACHRE interviews with human subjects as well as researchers involved in the radiation experiments exposed highly unethical treatment of human subjects throughout the thirty years of its existence. Following the exposure of these countless cases in which explicit disregard of basic human rights and freedom took place at the hand of government and private institutions in North America and elsewhere, a social ethic attuned to concerns of human rights over collective interests began to develop (McDonald, 2001, p.37). It became apparent that self-policing mechanisms by the research community did not ensure adequate protective measure for human subjects, no matter the political affiliation or government regime of the nation.

The change towards a stringent protectionist system of governance led to what Rothman referred to as “strangers” outside the realm of medicine, which included
lawyers, philosophers, theologians and other bioethicists, attending at the bedside and issuing best practice guidelines for ethical research involving human subjects (Rothman, 2001). Although this shifted the control of setting standards of research ethics from the hands of clinical researchers to “experts” as defined by the multidisciplinary field of research ethics, decision-making on ethical research practices remained a top-down procedure; scientific, legal, and ethics experts laid down prescriptive research ethics guidelines in the absence of any consultation with those who were affected by its output. Although human subjects were no longer viewed as simply mere objects for scientific advancements, they remained passive agents in the research endeavor nonetheless. As a result, increased protection not only for the human subjects, but also for the research institution and its stakeholders, led to a bureaucratization of the medical research enterprise.

2.2.1 Bureaucratization of Research: legislation & moral codes of conduct

In direct response to public disclosure of the Tuskegee Syphilis Medical Experiments that took place over the course of four decades, between 1932 and 1972, in 1974 the National Research Act was enacted in the United States. This led to the creation of the National Commission for the Protection of Human Subjects of Biomedical and Behavioral Research, a federal regulatory body governing research involving human subjects (Rothman, 1999, p.5). This newly enacted Law required all government funded research to be overseen by an external Institutional Review Board (IRB) comprised of a range of experts, including lawyers, ethicists and clinical scientists.
In 1979, the National Commission for the Protection of Human Subjects of Biomedical and Behavioral Research issued the Belmont Report, setting forth ethical principles that continue to be the hallmark of regulatory documents concerning research involving human subjects (Dresser, 2001, p.13). The Belmont Report empowered human subjects, to a certain degree, by recognizing them as moral agents deserving of rights and freedoms through three fundamental principles:

1) **Respect for persons**—The human subject must be respected as an autonomous moral agent. As autonomous agents, they must be fully informed and given the choice to voluntarily participate or refrain from participation in research. Secondly, upon consenting to take part in the research, human subjects are free to withdraw from the study at any point.

2) **Beneficence**—Researchers have a duty to maximize potential benefits, while minimizing harms that may result from participating in the research.

3) **Distributive justice**—Researchers ought to ensure that the burdens and benefits of research are distributed equally amongst individuals and/or groups in society. That is, no one individual or group ought to bear the burden of participating in research, nor should any benefits of research be received disproportionately by one group/individual.

(Dresser, 2001, p.13)

Nevertheless, following the Belmont Report it is argued that some members of the research community continued to use their own paternalistic judgment in determining what was ‘good’ or ‘in the best interest’ for their research subject, and in some extreme cases continued to risk the welfare of the research subject for the ends of innovative health research (Moreno, 2001, p. 15). As noted in Jonathan Moreno’s *Goodbye to All That: The end of moderate protectionism in human subject research* (2001), the 1989 revised guidelines of the Declaration of Helsinki left a slippery slope for physicians.

---

8 The most sophisticated account of the principles of beneficence, autonomy, justice and non-maleficence was first laid out in Beauchamp and Childress’ 1979 book “Principles of Biomedical Ethics. New York: Oxford University Press.

9 It should be noted that Tom L. Beauchamp, co-author of “Principles of Biomedical Ethics” went on to play an active role in the drafting of the final Belmont Report. For an interview with Beauchamp on the history of the creation of the Report, please see http://www.hhs.gov/ohrp/docs/InterviewBeauchamp.doc.
involved in therapeutic research studies in being able to decide when to seek consent from their patients in instances that they deemed seeking consent would be problematic.\textsuperscript{10}

Regardless, the institutionalization of measures to ensure adequate protection of human subjects involved in health research created a shift in the clinical researcher-subject relationship. Increased protectionism gave the human subject a greater degree of power in the sense that the researcher himself became accountable to a legislative body outside from his own institution with the establishment of the Institutional Review Board (IRB). In the U.S., the principal means by which the state is able to exercise its governance over research practices is by restricting its Federal funding to those research initiatives that subscribe to Federal regulations. Regulations are set out by the Federal Policy for the Protection of Human Subjects under the U.S. Code of Federal Regulations known as the Common Rule (McDonald and Meslin, 2003, p. 7).\textsuperscript{11} Knowing that failure to meet ethics standards set out by the Common rule, such as acquiring free and informed consent from his subjects, could invoke the authority of the IRB to question the integrity of his work and even withdraw research funding, sought to promote greater protection of human subjects.

Similarly in Canada, the three major government funding agencies, the Medical Research Council (MRC), the Natural Sciences and Engineering Research Council (NSERC), and the Social Sciences and Humanities Research Council (SSHRC), jointly

\textsuperscript{10} Moreno defines ‘therapeutic research’ as “Medical Research Combined with Professional Care.” Jonathan Moreno, p. 15. The terminology of ‘therapeutic’ and ‘non-therapeutic’ has been raised as problematic, stemming as a result of the gaps left open by the Declaration of Helsinki. As Levine notes “‘therapeutic research’ is problematic because all clinical trials of therapeutic agents include some components that may be therapeutic and others that are clearly non-therapeutic. Levine, (1999) “The need to revise the Declaration of Helsinki.” The New England Journal of Medicine, 341(12), p. 532.

\textsuperscript{11} \textit{U.S. Code of Federal Regulations}, Title 45, Subpart A. Online version can be found at: http://www.hhs.gov/ohrp/humansubjects/guidance/45cfr46.htm
created the Tri-Council Policy Statement (TCPS): Ethical Conduct for Research Involving Humans. Founded on the principles of the Belmont Report, in 1998 the TCPS was the first uniform regulatory guidelines governing all research institutions that received funding from any of the three major federal funding agencies in Canada. Akin to the United States’ IRB system was Canada’s use of Research Ethics Boards (REB) as a regulatory mechanism for good ethical practices in health research involving human subjects. In order to act as an arms-length gatekeeper, the REB is established by the highest levels of the Institution where the research involving human subjects is carried out and consists of at minimum five members with multi-disciplinary backgrounds in health research, law, and ethics, and must include at least one community member (TCPS, 2004, Section 1.B). The responsibility of the REB is to assess the ethical acceptability of all research study protocols that involves human subjects, or tissue and/or DNA samples from human subjects, that fall under its Institution’s jurisdiction. The primary task for the REB is thus to protect the prospective human subject’s interests by way of assessing whether the study procedures meet the minimum ethics requirements set out by the TCPS, and weigh the level of risk(s) against the potential benefit(s) participation in the study may have on prospective human subjects. This is largely done through an examination of the researcher’s submission of an ethics application that details the study procedures, researcher affiliations and qualifications, any conflict of interest issues, and all related study documents, such as the informed consent document. Following this assessment and deliberation amongst the board members (with either a partial or full board

---

deliberation depending on the level of risk involved), the REB has the authority to either 1) approve the study protocol and grant the researchers the right to commence data collection, 2) request that modifications to the study protocol be made by the researchers before approval can be granted, or 3) terminate the proposed or ongoing research involving human subjects. With the disheartening list of historical cases of unethical exploitation of human subjects in medical research, increased external oversight mechanisms and regulatory frameworks to ensure the protection of subjects in health research on an international scale was well warranted. The definitive feature of increased protectionism reflects a movement away from the paternalistic model of research in part on account of the importance placed on the individual’s free and informed consent.13

2.3 An Era of Activism: Subject as Active Agent

The inclusion of human subjects into deliberations concerning the affairs of certain sectors of health research alongside scientific “experts” is in part attributed to the context of the post 1960’s era with the disabilities rights and self-help movement, as well as increased consumerism (Stockdale & Terry, 2002, p. 81). The inclusion of public members in developing effective public health practices within community mental health centers was mandated in the US during the nineteen-seventies with the Health Maintenance Organization Act in 1973, the National Health Planning and Resources Development Act of 1974, and the Health Revenue Sharing Act of 1975 (McCormick, 2004, p. 627).

13 Central to modern research ethics is the importance of the autonomous research subject’s right to voluntary informed consent. The relationship between the benevolent clinician-researcher and autonomous human subject thereby became a fiduciary, rather than paternal, relationship based on an inclusive decision-making process.
Promotion of lay public participation in health research initiatives did not begin until the late nineteen-eighties. This key turning point came with the HIV/AIDS activist movement. This movement generated the most significant transition from the view of subjects-as-object towards subjects as active agents engaging with the research community on decisions concerning the conduct of health research. The transition stemmed from the HIV/AIDS patient groups’ backlash against the standard scientific methodology of double blinded randomized control trials (RCT). RCT is a standard practice that involves a randomly chosen subset of the patient-subject group receiving a placebo in replacement of any study drug for their fatal disease (Dresser, 2001, p.23).

Advocacy groups formed by HIV/AIDS patients came to stand together against what they saw as immoral, unjust standard research practice, namely randomization that was considered by the clinical research community to be good science.

Many activists became highly educated in medical terminology and developed an understanding for the science behind the illness as a means to be seen as credible when standing up against industry experts. Patient groups revolted against what they perceived as unethical treatment and some individuals even began to exhibit noncompliant behaviour during study trials. The backlash against standard research practices ultimately led to the development of new clinical trial guidelines and, as Dresser notes, changed the way many members of the scientific community saw the role of consumers (Dresser, 2001, p.25). The values and interests of the consumer

---

began to be taken seriously and seen as a critical component in decisions relating to the development of health research initiatives.

HIV/AIDS organizations’ success prompted other patient groups to move beyond their role as fundraisers and began to engage with the research communities as consumers deserving of a voice. The most notable organization to follow in the HIV/AIDS activists’ footsteps was the Breast Cancer organizations. The role of patient-subjects in the research endeavor had previously remained largely on the periphery with the majority of their efforts geared towards raising funds for research initiatives, while having little influence over the direction and design of the projects they participated in, which was one of their central goals (Dresser, 2001; Batt, 1994). The most critical question that needed to be addressed from most cancer patients’ point of view was on preventative health measures women could take to prevent the occurrence of breast cancer at the outset. Political and economic interests continued to drive the research agenda, thereby forcing patient-groups, such as the breast cancer society, to continue advocating their right to voice their concerns and values in regards to the kinds of research initiatives that were of concern to them.

With the success of the HIV/AIDS, Breast Cancer patients, and other organized health consumer advocates initiation of collaborative partnerships began to emerge in the nineteen-nineties. In Canada, the House of Commons Sub-Committee on the Status of Women expressed concerns about “a type of ‘closed’ circle of researchers and concluded that “it is outdated, in 1992, to adopt an approach that suggests that only physicians and scientists are equipped and qualified to evaluate the efficacy of research proposals and to make policy decisions on the nature and direction of cancer research”
The committee’s recommendation was for lay people to participate on the board of directors and research proposal review committees of Health Canada, the National Cancer Institute of Canada, the Medical Research Council of Canada, and the National Health Research and Development Program. Similarly, the US National Breast Cancer Coalition took on partnership roles through the National Institute of Health (NIH) Specialized Program of Research Excellence, which allocated $210 million funds specifically for Breast Cancer research (Waller and Batt, 1995, p.831). In the UK, the Department of Health, Health Technology Assessment Programme, Medical Research Council and Mental Health Foundation, have all advocated for an increase in the number of lay members and human subjects taking on meaningful roles within the processes of health research (Boote, 2002, p.214). The increasing emergence of recommendations advocating for lay public involvement in health research endeavors highlights the importance that nations on an international level place on public trust in the research enterprise. What is perhaps even more urgent, however, is the need for the inclusion of those individuals who are directly affected by the research, the human subjects themselves, in meaningful and effective roles within the research enterprise. Though there is evidence that certain sectors of health research have begun moving towards establishing standard participatory research practices, as I shall discuss in the following Chapter, there is still a significant ways to go before the many cultures of research accept human subjects as true partners in the health research enterprise.
2.4 Conclusion

Though views towards the role of human subjects began to shift as the research institutions and governing bodies began to see them as central figures in the advancement of health research, the extent to which they became active ‘partners’ in the health research enterprise, sharing equal power with the research community, is highly contested (Corrigan and Tutton, 2006). It must also be stressed that the ideal of participatory approaches is still far from the norm. Nonetheless, others such as Merz et al. note that the foundation has been laid and “a new type of relationship is emerging as groups become key players in the promotion of studies of the causal role of genetics in diseases” (Merz et al., 2002, p.965). For the scientific community this meant receiving input on how to design research, yet also insight into recruitment and retention strategies and possibly even gain increased trust in return from the patient-subject communities.

In the following chapter I discuss the emergence of participatory approaches in the health research enterprise and argue that these practices ought to be encouraged by the Canadian TCPS guidelines on appropriate ethical conduct for all types of health research involving human subjects in health research whenever collaboration is possible. I present and discuss two cases of disease-based advocacy groups that have challenged the health research community into taking on partnerships with their respective patient-subjects. By examining these two unique cases of effective patient-group and research community partnerships, I will discuss the benefits and ethical challenges of consumer and research community partnerships.
CHAPTER III
Research Partnerships:
The PXE International and Breast Cancer Foundation as Examples

People concerned about research ethics should welcome the emergence of research advocacy. At the foundation of modern research ethics is the belief that scientists alone ought not decide research practice and policy. Rather, the values and preferences of the broader community should guide research. (Dresser, 2001)

As indicated in the previous chapter, the promotion of a collaborative approach to decision-making has been far from the norm in the health research enterprise. Only recently has participatory methods been applied within a few areas of health research. Most notably, health research investigators involved in research with highly visible and/or vulnerable communities have begun to employ standardized participatory frameworks into the design, development, and/or dissemination of their research initiatives in order to assure the community is shown respect and added protection for its members involved in the research. In order to support my central argument that collaborative relationships between human subject and research communities on a broader scale are essential for research practices being ethical, I will discuss two cases in which collaborative relationships between patient-consumer groups and research communities have adapted participatory frameworks to promote effective, meaningful, and mutually beneficial partnerships.

First, I begin this chapter with a brief discussion of the values inherent to participatory methods of inquiry. I will then present two cases in which consumer groups
have become active partners within the health research enterprise. The first case I will present is the non-profit organization PXE International, a foundation to raise awareness and promote research on the rare genetic disease *Pseudoxanthoma Elasticum*. The second case I will discuss is the Canadian Breast Cancer Foundation and their success in transitioning the role of human subjects into active consumers of health research. My aim in exploring these two cases is, firstly, to highlight examples of effective and successful research partnerships between human subject advocacy groups with their respective research community and, secondly, to discuss some of the key advantages a partnership between a small-scale consumer group in comparison with a large-scale and highly influential consumer group and the research community may garner.

### 3.1 Participatory Methods of Inquiry

The use of participatory methods of inquiry became part of community-based research practices in the 1970s. Participatory frameworks first developed within social science research that typically involved oppressed and/or vulnerable collectives, the most prevalent areas being research involving groups in developing countries, HIV/AIDS patients, and aboriginal communities (Khanlou and Peter, 2005, p.2334). In the latest draft of Canada’s Tri-Council Policy Statement it stipulates that any type of research, whether it be social science or biomedical, involving aboriginal community members requires a collaborative partnership between the research and aboriginal
communities before ethics approval will be granted.\textsuperscript{15} This requires that all decision-making pertaining to a research project be made in accordance with members of the community throughout each stage of the project’s development, unless otherwise agreed upon by both parties.\textsuperscript{16}

The extent to which human subjects in collaborative research endeavors have been involved in the research design, process, oversight and/or dissemination of research findings has varied across community groups and research initiatives. Hubbard et al. have developed a typology of the various levels of human subject involvement as follows: consultation, collaboration and user-controlled involvement (Hubbard et al., 2007, p.234). According to Hubbard et al., consultation reflects those human subjects who are asked about their views about a particular decision, yet these views may not necessarily be adopted, but they may have some influence on the outcome. Collaboration involves active, ongoing partnerships with members of the public, or patient-groups, and user-controlled involvement is when the power, initiative, and decision-making lie in the hands of the consumers first and foremost. Ideally, the extent to which human subjects are involved and the ways in which they are involved as “partners” or “consultants” concerning the research endeavor is a decision that is made based on a number of factors, such as: the type of research protocol, the human subjects’ health and ability (physical/emotional/cognitive) to become involved, the willingness and desire of the human subjects to be involved as active agents, the added

\textsuperscript{15} As indicated in the previous chapter, this requirement pertains solely to government-funded research. Private funded research initiatives are not bound to the TCPS guidelines. This is currently a requirement for all research sponsored by CIHR.

\textsuperscript{16} Reference to section 6.1 TCPS (report on aboriginal populations and research ethics). Specifically, the TCPS requires that researchers consult with the aboriginal community in decisions regarding all facets of the research project; including monitoring and ethics review of the study protocol, direction of the research question, design of the protocol, conduct of the research, and the dissemination of study results.
value that their involvement as participants or consultants would bring to the research endeavor, as well as the potential benefits collaboration would offer to the communities and individual subjects involved.

Human subjects may be involved, either in consultative, collaborative, or user-controlled capacities, during the initial stages of the research project by aiding in the identification and prioritization of the research question(s). They may be involved in decisions concerning the research procedures, such as the type of informed consent process that is most appropriate (e.g. individual or community consent), and the process by which study results are disseminated. They may also be more directly involved in the oversight of the research initiative, taking on the role of an advisory or research ethics board members. Whatever the role and degree of human subjects’ participation entails, the overarching aim of these methodologies is to move away from the traditional top-down model of the scientific experts single-handedly steering the research agenda and process, and towards a joint-venture between consumers and investigators joining together in various stages of the decision-making process on a given health research initiative.

Inherent in this participatory approach is the notion that research ought to advance the interests of the research community as well as the participants involved in the hopes of mutually benefiting both parties. In effect, the first principle in research ethics, to respect the autonomy of human subjects, is maintained with the use of participatory frameworks more effectively than with exclusively conventional measures, such as signing a consent document developed by the research investigator. As Wallworth notes, “the autonomy of each party is respected, in the sense that
participation in the joint undertaking is informed and consensual, and each contributes something essential to the project, such as complementary skills, knowledge, experiences and competencies" (2008, p.58). Supporters of a collaborative relationship argue that such an inclusive approach to decision-making, one that ensures all stakeholders’ voices are heard, addresses concerns around social justice issues and further protects vulnerable groups from exploitation (van der Riet, 2008, p.559). In addition, I argue that addressing social justice issues and promotion of a democratic dialogue amongst all stakeholders will legitimize the research outcome and, thus, render a more trustworthy research enterprise. This argument concerning the urgency of collaborative research endeavors stemming from a political perspective is directly addressed in Chapter 4.

3.2 Research Partnerships: PXE International

Pseudosanthoma Elasticum, or PXE, is a rare autosomal recessive genetic disorder that may lead to vision, skin, and arterial defects due to a mineralization of the elastic tissue. As a result, patients may experience loss of vision, loose and hanging skin on the neck, underarms, knees, problems with their cardiovascular and gastrointestinal systems, and in a remote few cases has resulted in death (Stockdale and Terry, 2002, p.91). PXE is caused by mutations in a single gene, ABCC6, a gene that is highly expressed in the liver and kidney (Plomp, et al., 2008 p.118). It is estimated that PXE affects approximately 1 in 100,000 to 1 in 25,000 individuals.\footnote{ According to statistics provided by the PXE International website: http://www.pxe.org/english/View.asp?x=1693}
3.2.1 Founding PXE International

The non-profit organization *PXE International* is a leading example for other advocacy organizations to become active collaborators with the scientific community in the industry of genetic research.\(^1\) The organization was founded by parents Sharon and Patrick Terry, a theologian and an engineer by training, after being informed that their two children aged 7 and 4 had inherited pseudoxanthoma elasticum (PXE). Upon their discovery in 1994 that their children had inherited this rare and little known genetic disorder, Sharon and Patrick Terry began educating themselves on anything they could find out about the biological and social implications of living with PXE. After having consulted the medical literature the Terry’s found that few research initiatives existed that sought to advance the knowledge of and treatments for the disorder and, as a result, no clear understanding existed in the scientific community about the progression of the genetic disease (Stockdale and Terry, 2002, p.92).

Central to being able to understand the natural history for any genetic disease is first being able to gather a large enough cohort of subjects who share the same phenotypic expression in order to have a significant enough statistical power in being able to begin studying the disease. For rare orphan diseases, such as PXE, this is especially challenging when the number of affected persons is inherently extremely

\(^1\) There has been some debate within the literature on whether the involvement of patient and/or advocacy groups into discussions on health research, on issues such as how to allocate research funding, the design of a specific research proposal, recruitment of subjects, and dissemination of research findings, may be considered an equal “partnership” between lay interest groups and the scientific community. As Corrigan & Tutton argue in *What’s in a name? Subjects, volunteers, participants, and activists in clinical research*: “While such initiatives may well involve ‘participants’ more actively in certain aspects of the research, the impetus for this kind of involvement may stem more from the needs of the researchers to ensure recruitment is made easier and retention rates are maintained than a genuine desire to encourage patients to be more actively involved in research as such.” P. 102
small (Terry and Boyd, 2004, p.178). In addition to a lack of a large enough subject pool to draw biological data from, is the impeding factor of limited research funding. The draw for funding agencies to allocate research funds to a disease that affects a very small percentage of the population, in some cases where there is an n of one, is virtually non-existent. The lack of awareness and drive to establish a network of individuals living with the genetic disorder that would promote and facilitate research, coupled with a highly uncoordinated research community whose work was not conducive to an open source concept of sharing research data, as well as limited interest in funding research involving such a rare genetic disorder motivated the Terry’s to formulate PXE International in 1995 (Terry and Boyd, 2004).

The objectives of the non-profit organization were to bridge the gap between patient communities and clinician researchers with the aim of achieving progress on the research and development of treatments for PXE. The organization first aimed to build a network within the patient community that would act as a support system and provide education for individuals and their families living with the condition, while simultaneously developing a network for clinician researchers working within the field of genetics (Stockdale, p.92). The following seven objects were the goals and mission statement of PXE International, as outlined by PXE International:

1. **Respect**: central and commanding respect for the lived experience of affected individuals
2. **Improved clinical services**: recognition of the urgent need for improved clinical services, health insurance issues, and so on, and action to alleviate some of these issues.
3. **Rights and access**: attention to disability rights issues and access to treatments, giving rise to activism designed to alleviate these problems
4. **Interdisciplinary**: an interdisciplinary approach, encouraging the interaction of many specialties, including service providers, clinicians,
and researchers, in the quest to meet the broad needs of the community and protect research participants

5. *Data sharing*: a broad base of affiliations with researchers, allowing for the creation of consortia to pool resources rather than confine them to one lab or another

6. *Promotion of research*: grounding in the philosophy that basic research on any disorder will lead to discoveries for others, an approach that encourages working diligently with the Genetic Alliance (alliance of orphan disease groups) and other coalitions


PXE International developed a strategic plan with the help of a professional advisory board and the organization Genetic Alliance (Terry and Boyd, 2004, p.180).¹⁹ This led to the establishment of a network in which individuals living with PXE could gain information about the disease, written in clear and plain language that was digestible to a non-scientific audience; the network provides a venue to form support groups to help cope with the physical and emotional implications of the condition. The organization was not only a vehicle to disseminate knowledge about the disease in a more effective and wide-reaching manner, but also acted as a means to recruit participants into research initiatives. Increasing the awareness of the need for individuals with PXE to donate their biological samples for the purposes of genetic research was a fundamental goal for PXE International, which later resulted in the establishment of the PXE International Blood and Tissue Bank (PIBTB) in 1996.

¹⁹ Genetic Alliance is a large international coalition of more than 600 disease and professional groups involved in genetics. [http://www.geneticalliance.org/](http://www.geneticalliance.org/)
3.2.2 PXE International Blood and Tissue Bank

The biobank\textsuperscript{20} collected fifteen hundred samples of blood, tissue and epidemiological data from affected individuals with the overarching aims being to uncover the genetic etiology of PXE, thereby hoping to develop effective treatments with the use of innovative therapeutic technologies and, ultimately, a cure for pseudoxanthoma elasticum. The biobank allowed researchers to gain access to a large pool of tissue samples from affected individuals, thereby facilitating an initiative within which to attract the genetic research community to focus research efforts and allocate funding on PXE. In addition to facilitating research, developing the biobank allowed control over decisions concerning who could gain access to its resources, and under what conditions such access would be given, to remain in the hands of the consumer group (Terry and Boyd, 2004, p.180).

With a partnership established between scientists conducting the research and the founders of PXE International acting as gatekeeper, many of the ethical issues that are common impediments to genetic research, particularly for large-scale multi-investigator research involving biobanks, were lessened, and in some instances avoided all together. Some of the common impediments to biobanking research involve informed consent, privacy, and ownership rights of participant’s genetic data.

\textsuperscript{20} According to Tansey and Burgess, a biobank is defined as: “a collection of genetic materials and health information for research related to human health. Such information might be used to develop personalized treatments, identify inherited risks for disease, or understand the role of genomic and environmental contributions to health in populations.” Tansey, J. and M. Burgess “The foundations, applications and ethical dimensions of biobanks.” Electronic Working Papers Series W. Maurice Young Centre for Applied Ethics, University of British Columbia at www.ethics.ubc.ca, page 1
3.2.5  Biobanks and Research Ethics

1. How will informed consent from prospective subjects be achieved?

A basic principle in research law and ethics is the notion of informed consent—all prospective human subjects must be competent, or have a competent authorized third party provide proxy consent, and fully informed about all known possible risks and benefits participation in a research study may ensue before voluntarily consenting to participate. Identifying foreseeable possible risks and benefits associated with health research and ensuring that the benefits outweigh the risks involved is fundamental to the primary task of a research ethics board review of any given protocol (Emanuel, 2000, p. 2706-2707). With the increasing prevalence of genetic research and biobanks, an assessment of possible risks and benefits associated with donating one’s personal health history and genetic data for use in unknown research purposes, renders an analysis of possible risks and benefits next to impossible. In addition, it further raises the question whether informed consent is even possible for research involving the use of biobanks, since potential subjects will not be informed of the risks and benefits involved in order to make a sound judgment whether or not to participate.21

Though there is no single solution to this major challenge to biobanking research, some scholars have proposed the use of blanket consent (where human subjects provide their consent for researchers to use their DNA and tissue samples linked with their personal health information for all types of health research) in place of traditional individual consent, while others prefer to move away from consent altogether (Sade, 2002) to “models that consider the authorization of samples for future uses as specified

21 For further discussions see Tansey and Burgess, 2004, p. 29-30
by participants and overseen by an arm’s length oversight body” (Secko et al., 2009, p. 782).

2. How will privacy of the subject’s data be maintained?

As noted in Section 8 of the Canadian governance documents for publicly funded research involving human subjects, the Tri-Council Policy Statement, special concern for the privacy of genetic information must be taken into careful consideration by researchers and REB members, particularly when it may have some implication for family and/or community members not involved as subjects in research:

*Consistent with the data confidentiality provisions of Section 3, Article 8.6 outlines the duty of researchers to address ethical issues raised by the banking of genetic material. In this context, although consensus has not been reached, a number of issues need to be considered by the researcher and clarified for the REB, particularly concerning privacy, confidentiality of records, and information derived from stored genetic material. A special concern arises when it is difficult to separate genetic information on an individual from information on his or her biological relatives or community. Access to genetic material and to the results of the research should be limited to the researcher, and if such limitation will not be the case, then the issue should be discussed with the research subject. Similarly, unauthorized access to stored genetic material or results by third parties should be prevented. Specifying whether banked genetic material will be anonymized, i.e., without identifiers, may help alleviate the concerns that other biological relatives may inadvertently be identified by linked data. (TCPS, Article 8.6 Banking of Genetic Material, Section F).*

Biobanks that must ensure a linkage between epidemiological and genetic data and identifying information and in some cases the requirement to re-contact individuals to consent to further research, renders the ability to keep the data fully anonymized extremely difficult and in some instances impossible, which thereby compromises the individual and/or family members’ protection of privacy (Andrews, 2005, p.25).
3. Who has ownership rights over the subject’s genetic data?

In lieu of the changing culture of research towards that of a commercial industry, questions of ownership rights over research data and how the benefits of biobank research will be distributed have garnered a great deal of attention in the bioethics literature (Einsiedel, 2003; Andrews & Lori, 2005; Tansey & Burgess, 2004). The American Medical Association’s Code of Ethics states that “potential commercial applications must be disclosed to the patient before a profit is realized on products developed from biological materials” and “human tissue and its products may not be used for commercial purposes without the informed consent of the patient who provided the original cellular material” (Andrews & Lori, 2005, p.25). Similarly in Canada, the TCPS stipulates:

Article 8.7 adds a specific obligation to the disclosure requirements for obtaining free and informed consent from those being subjected to genetic research: the potential for commercial use of genetic data. There is significant legal and moral controversy regarding ownership of genetic material or research data, and concepts of ownership may vary from one cultural group to another and between legal systems. It is unethical for a researcher to claim ownership of genetic material by claiming that the concept of private ownership did not exist in the community involved. Consistent with the free and informed consent provisions of Section 2, the researcher may have to seek further permission from the group. The fact of commercial sponsorship of genetic research should be revealed to the subject at the beginning of the project. Similarly, possible commercialization occurring after involvement in research should also be revealed at the outset if possible.
(TCPS, Article 8.7 Commercial Use of Genetic Data, Section G).

---

22 This was brought to the fore as a result of the case of Moore v. Regents of the University of California in 1990 when the court recognized that physician/researchers have a fiduciary duty to their patients to disclose their intent to use patient tissue samples for research or commercial purposes. (Moore v. Regents of University of California, 51 Cal. 3d 120, 132-133, 793 P. 2d 479 Cal. Rptr. 146, 153 (1990)).
Therefore, by fully disclosing the researcher’s intentions to generate commercial benefits from the research, most commonly through patenting genes of a genetic disorder, the researcher and/or research institution may gain legal control over monetary benefits of research. In the case of *Moore v. Regents of the University of California*, in which John Moore sued scientists when he discovered his genetic material was used to develop a cell line that later profited financially through patent rights over the cell line, the courts ruled in favor of the researchers. The reasons given were that Moore did not have any property rights over his own genetic material since he had voluntarily consented for their use in the genetic researchers’ study. Despite this and similar court rulings, concepts of “ownership” rights over control of one’s genetic material in the context of health care and research remain unclear (Charo, 2006, p.1517).

Although these remain pertinent challenges to research involving biobanks and their possible solutions varied and unclear, several scholars have proposed one way of mitigating these issues is with the use of deliberative democratic engagements involving a wide range of stakeholder groups (Cragg et al., 2000; People Science & Policy Ltd., 2002; Burgess et al., 2008). As Secko et al. argue in their 2009 paper *Informed consent in biobank research: A deliberative approach to the debate*: “based on the potential societal impact of biobanking, we argue that any resolution of the uncertainty around consent must consider the informed, deliberative input of a range of perspectives within the citizenry” (p.782-783). Through an informed and deliberative process, stakeholder groups that includes both members of the research and scientific community and prospective human subjects, meet face-to-face to learn from one another and
understand each others’ perspectives and values around issues such as appropriate privacy measures (whether or not to anonymize data and who has access to the data), informed consent procedures (e.g. individual or blanket consent), and ownership rights (the data will be on loan to the researchers, there will be a joint ownership and therefore joint sharing of research benefits, or it may be agreed that the researchers have full ownership rights over the genetic and epidemiological data). Through such deliberations, it is argued that whatever the resulting policy advice on the governance of biobanks, it will be an outcome based on a diverse representation and, thus, will be legitimate and accountable to all stakeholders’ interests.

3.2.6 PIBTB and Research Ethics

As stated earlier, with a consumer-run organization such as PXE International in control of the tissue and data repository, many of the above ethical issues involved with biobank research were lessened or avoided through collaborative relations between the consumer group and scientific community. According to leaders of PXE International, policies were developed to ensure that confidentiality was maintained in order to minimize the risk of discrimination to research participants. Facilitation of ethical research practices that are sensitive to the needs and interests of the participating subjects is at the forefront of PXE Internationals’ mission statement (Stockdale and Terry, 2002, p. 95).

As individuals faced with similar life experiences of living with a rare genetic disorder, or being the caretakers of such individuals, PXE International claims it is able to address some aspects of informed consent that are unique to rare disease research that researchers setting the agenda alone could not have achieved in the same way.
Whether their claims that participants are better informed and their privacy better protected follows through in practice and shows an improvement over existing government or industry-controlled biobanks is uncertain since there is no empirical evidence in the literature that has compared this patient-subject run biobank with government or private run biobanks. Hence, empirical research to support or dismiss this claim is well warranted. Nonetheless, partnerships do seem to address the problems some other biobank projects have encountered with their participants on the issue of property rights. PXE International controls the use of genetic and epidemiological data for research purposes and, as such, any patentable outcome of the genetic research belongs to PXE and all publications resulting from the research are co-authored by the founders and researcher team responsible for the new development. In effect, each party shares in the research efforts and rewards, thereby fulfilling the mutually beneficial relationship. As anthropologist Karen-Sue Taussig observes, PXE International “is creating a new set of social relations in which ‘ordinary people’ are not just the ‘containers of DNA’ but are also co-producers of scientific knowledge” (Tutton et al., 2004 p. 30).

The discovery of the PXE causing gene ABCC6 in 2000, as a result of stored genetic samples within the PIBTB, allowed scientists who were involved in identifying the gene and the founders of the non-profit organization to gain patent rights over identification of mutations in the ABCC6 gene. Contrary to many other researchers’ intention of attaining patent rights to gain a monopoly over the research, the founders of PXE International saw patenting the gene as a means to ensure useful and timely treatments and to make all tests and treatments accessible and affordable to the patient
community (Tutton et al., 2004). As such, it is claimed by PXE International that it “is able to act as steward of the gene—representing the interests of the PXE community in the process of moving from gene discovery to commercialization in the form of diagnostics or therapeutics” (Terry et al., 2007, p.161).23 The co-founders of the organization have sought an open access approach to research, continually partnering with research organizers and funding agencies to develop further research initiatives and technological innovations not only for PXE, but for other rare orphan diseases as well.

Their partnership with Transgenomic, a global biotechnology company, established three laboratories world wide (Jefferson Medical College in Philadelphia, the University of Gent in Belgium, and the University of Witwatersrand in South Africa) to genotype hundreds of samples, which has recently led to the development of genetic testing for the PXE causing gene ABCC6 along with pre and post-genetic counseling for patients (Terry et al, 2007, p.161).24 Partnerships with PXE international requires researchers to submit to a memoranda of understanding that ensures the development of research and treatment for PXE is done in a manner that facilitates sharing of data, co-publication of research findings, and clear delineation of the roles and responsibilities of each party. PXE International and its founders have now moved into a mentorship role for other rare genetic disorder groups. They have guided the establishment of the Genetic Alliance Biobank, established in 2003, which has

---

23 It must be pointed out that a clear drawback in the literature on PXE International is that all publications must be co-authored by its founders. In effect, little to no empirical data from an outside source is currently unavailable to objectively critique the operations of PXE International.

24 For information about this service see: http://www.pxe.org/english/view.asp?x=1686
developed a similar infrastructure as PXE International with partnerships between interest groups and research communities at its foundation (Terry et al., 2007).

### 3.3 Research Partnerships: Breast Cancer Advocacy

The first cancer research fundraiser began in the mid-1940s and was operated by the American Society for the Control of Cancer (ASCC) (Batt, 1994, p.295). Physicians, namely surgeons, governed the ASCC board. Advertisements for Breast Cancer awareness campaigns were sponsored by interest groups such as the American Medical Association, whose slogans were “A Message of Hope” rather than messages geared towards preventative measures against breast cancer. As Sharon Batt states, “[the goal was] to combat cancerphobia that kept the disease in the closet and to advance medical knowledge…. [Physicians] wanted to get patients into their offices. They wanted funds for research” (Batt, 1994, p.215).

In 1944 the ASCC became the American Cancer Society when Mary Lasker, a philanthropist and citizen lobbyist of New York City, took over control of the non-profit society and ushered in a campaign towards increased education and funding for Cancer research (Batt, 1994, p.217). Inspired by the efforts and success of Lasker’s Cancer Foundation in the US, breast cancer activist Nancy Paul set up the Canadian Breast Cancer Foundation in Toronto in 1986. The foundation sought not only to garner research funding, but also aimed to influence society’s awareness of the disease and promote standardized breast cancer screening programs.

By the late 1980s, breast cancer advocacy groups followed the lead of the HIV/AIDS activist movement in demanding a voice in the prioritization of research.
Beginning as a grassroots movement, breast cancer advocates learned from the HIV/AIDS activists that the best means of drawing attention and interest to your cause is by educating the citizenry and commercialization within the highly competitive research market (Dresser, p.6). The breast cancer advocacy movement first began to render influence in the Department of Defense (DOD) Program for Breast Cancer Research, by 1993 they had convinced Congress allocate $210 million in funding for breast cancer research (Dresser, 1994, p.25). Within the DOD program, research activists saw their opportunity to gain influence over the conduct of the research initiative in that the program was new and lacked any direction in determining how to direct research funds. This program was one of the first to establish within its review committee two seats reserved specifically for consumers, allowing them a role in voting for or against funding for proposed research protocols. By 1999 the first patient advocate was elected chair of the review committee.

The involvement of patients both in developing health services and in the conduct of research beginning in the early nineteen-nineties was an answer to frustrations that breast cancer advocates saw with the scientist-driven research agenda. As Batt explains, “Researchers own the enigma of breast cancer. As custodians of the intellectual conflict with the disease, they engage in the search for understanding that could eventually solve the puzzle. This struggle with the unknown, they define what questions are important” (Batt, 1994, p.292). The direction of research was highly politicized by the prospects that the new science in the era of genetics promised. In result, the focus of breast cancer research centered on identifying breast cancer-
causing genes, two of which were first discovered in 1990. Further down on the list of research priorities were areas that focused on prevention and psychosocial effects associated with having breast cancer—areas that were perhaps of greater interest to those affected by the disease, especially since breast cancer can be caused by an interplay of genetics and environmental factors.

Over the course of the past two decades breast cancer advocates have been able to reach their goal of taking part in discussions concerning the direction and prioritization of cancer research. The notion that breast cancer survivors and advocates must be present whenever decisions concerning the agenda and direction of breast cancer research are made has been increasingly acceptable and even mandated as a requirement for some institutions to receive research funding (Boote, 2002, p.217). At present in the United States, the leading advocacy organization with the most influence as a medical consumer lobby group is the National Breast Cancer Coalition (NBCC) (Platner et al., 2002, p. 102). Organizations such as NBCC have institutionalized themselves by educating their cancer advocates about current research, public policy, and legislation concerning cancer research, as well as cultivating effective advocacy skills in order for patient-subjects to take on active and effective roles in the research enterprise (Platner et al., 2002, p.103). In Canada, the Canadian Breast Cancer Research Initiative (CBCRI) established a partnership with the research bodies and major breast cancer advocacy groups in 1993 (Monahan & Stewart, 2003). The CBCRI was established as a direct result of breast cancer advocates insistence on rigorous efforts to fight against breast cancer. CBCRI instated that all research ought to include

\[25\] BRCA1 and BRCA2
the voice of those directly affected by the disease; “The role of the consumer, then, was elevated as a vehicle through which the scientific and governmental communities could be made more accountable to the public at large and discussion at the table of scientific and governmental decision-making could be injected with a unique societal, humanistic voice” (Monahan & Stewart, 2003).

The inclusion of those who were once left silent as passive recipients of treatment and subjects in cancer research has gone beyond merely having their interests heard. As discussed in the previous chapter, efforts to ensure an active partnership between breast cancer advocates and the research community has led to their involvement in a variety of research activities—from setting research priorities and allocating research funds, sitting on research ethics boards and grant review panels, planning the design and recruitment of cancer research proposals, analyzing research findings, and disseminating research results to their patient group and wider public. A collaborative model has been advocated within policy directive at the national level in Canada, the US, the UK, and Australia.

In 1998, the UK’s Secretary of State for Health called for a collaborative ‘partnership’ dynamic between the state, health professionals, and patients, which are reiterated in the Health and Social Care Act in 2001 (Wright et al., 2006, p.5). The National Breast Cancer Foundation of Australia has undertaken a national cancer research prioritization strategy through consultation with patients, members of the medical and scientific community and policy makers (Wright et al., 2006). The World Health Organization also endorses the involvement of consumers in health research and care in that such a perspective compliments the perspective of the clinician and the
biomedical researcher, thereby providing a more holistic interpretation of health (Boote et al., 2002, p.217). Whether through educational awareness campaigns, breast cancer patients advocating for changes to the agenda of Breast cancer research, or technological innovations, breast cancer rates have decreased exponentially in the US and Canada over the last two decades. In Canada the rate of Breast Cancer has declined by 25% since 1986, whereas in the US the incidence of Breast Cancer increased by 25% from the early 1980s to 1992-1993, but declined again by 18% from 1993 to 2005-2006 in Women aged >45 years.26

### 3.4 Conclusion

Through an exploration of collaborative relationships between two very unique cases, I have outlined some of the key advantages participatory approaches to research endeavors can offer. In sum, both the rare genetics disorder organization, PXE International, and the Breast Cancer Foundation were able to shift the research agenda from the hands of the research community into a direction that was conducive to shared decision-making between consumers and researchers on the prioritization of their respective health research initiatives. In doing so, the founders of PXE International were able to gain a monopoly over the commercial interests of their genetic data, thereby increasing funding for a disease that would have otherwise been under-funded and under-researched. Breast Cancer patient advocates, on the other

---

hand, were able to shift the direction of breast cancer research to topics that were relevant and important to them as a community. Involving cancer patients in the prioritization of research initiatives, rather than a researcher-driven agenda for monetary gain or public prestige, led to research on measures to prevent the occurrence of breast cancer, rather than exclusively on therapy and treatment.

In the following chapter I shall investigate the notion of participatory and inclusive decision-making with the participatory theory of democracy and mechanisms to advance effective and meaningful collaboration. First I will discuss the notion of “expert knowledge” and argue for the inclusion of a range of epistemologies, including the value of experiential knowledge, when determining health research agendas and practices. I will then discuss the use of deliberative democratic approaches in decision-making in the area of policy making and health research.
Chapter IV
Democratizing Science Research

If the principles of deliberative democracy were to be more fully realized in the practices of bioethics forums, the decisions the participants reach would be more morally legitimate, public-spirited, mutually respectful, and self-correcting. Deliberation-friendly forums could help reduce our deliberative deficit. By making democracy more deliberative, we stand a better chance of resolving some of our moral disagreements, and living with those that will inevitably persist, on terms that all can accept.
(Gutmann & Thompson, 1997)

The argument that Canadian policy on research ethics ought to require collaborative approaches of engaging human subjects, in varying degrees, within all health research sectors is supported by both moral and political claims. As I have previously indicated, promotion of collaborative methodologies in research is a moral imperative since human subjects ought to have the right to contribute to decisions that will directly or indirectly affect them and/or their communities. As discussed in Chapter three, contributions may be in the form of providing consultative advice, having a collaborative role and/or taking on a user-controlled role with researchers on the direction of the research question (as was the goal for some Breast Cancer advocates), questions around intellectual property rights (such as the case of founders of PXE International), and/or appropriate study methodology (as was the primary goal the HIV/AIDS patient-subjects achieved when they made claim to their right to voicing concerns over the ethical conduct of clinical trial research.) The focus of this chapter, however, will be to unpack the latter claim; that is, on political grounds and from a
procedural justice perspective, collaborative health research practices are imperative to the legitimacy and trustworthiness of the health research enterprise as a whole.

This Chapter first begins with an examination of the aims of a participatory democratic approach to science policy decision-making and the inherent value of lay public experiential knowledge. Secondly, I will discuss a methodology that has been used in contemporary society to bridge the gap in public policy discourse between scientists and non-scientist experts, the practice of deliberative democratic engagements. Lastly I will argue, on account of the first two sections of this chapter, that participatory democratic processes ought to be systematically implemented into health research practice where conflicting moral issues pertaining to research policy warrant public deliberations.

4.1 Post-positivist Theory and Democracy

The notion that an engaged and educated citizenry is imperative to the success of a modern society is a position that has been advanced for centuries in democratic societies. Key political theorists and critics of liberal democratic theory, such as John Dewey, Jürgen Habermas, and Michael Foucault, advocated for the involvement of both experts and citizens in deliberations on social issues with conflicting moral, economic, and/or political interests. The post-positivist movement, made famous by such influential theorists as Dewey, Habermas, and Foucault, to name a few, challenged traditional interpretations of science as an objective mode of inquiry, and instead pointed to the value-laden nature of its practice, with, as Fischer remarks, “its objects and relationships named and described by the scientists themselves. The activity of science
is thus seen to be a product of the very social world it seeks to explain” (Fischer, 1993, p.167). With a new definition of scientific inquiry, supporters of post-positivist theory encouraged a more participatory view of democracy in regards to science policy, one in which would embrace the joint partnership of scientific experts with lay citizens’ knowledge in order to come to a more holistic understanding of the world and a shared approach to science policy decision-making in practice.

In his work *Legitimation Crisis* (1973) Habermas denounced ‘scientistic practices’ as elitist, pointing to the top-down structure of science policy decision-making processes. In the same year, Michael Foucault published his highly influential work *The Order of Things* in which he highlights the power inequalities inherent to the structure of science policy and, Fischer notes, as such concludes that “social science [is] a discipline of social control rather than human advancement and democratic emancipation” (Fischer, 1993, p. 166). In his earlier work *Liberalism and Social Action* (1935) Dewey reflects on the notion of an inclusive democracy: “The method of democracy – inasfar as it is that of organized intelligence – is to bring conflicts out into the open where their special claims can be discussed and judged in the light of more inclusive interests than are represented by either of them separately.” It is criticisms by such postpositivists supporters as Habermas and Foucault, as well as Dewey’s conception of an ideal democratic order that lays at the foundation of arguments advocating for citizen engagement in discourses on science policy decision-making.

### 4.1.1 The Value of Lay Expertise

The promotion of engaging the lay public into science policy discourse of public concern, an area that has traditionally been reserved for policymakers and others with
relevant scientific expertise, has gained increasing popularity in the political practice of contemporary democratic societies. Reasons for its growing popularity have been attributed, along with the postpositivist movement, to an increasingly empowered public as well as the growing complexities in society, with such realities as globalization and scientification (Caron-Flinterman et al., 2007, p.340). It is also largely a result of the recognition that lay citizen’s knowledge is intrinsically valuable.

In such disciplines as Sociology of Science and Science & Technologies Studies, it has been widely argued that the lay public, as consumers of science, hold a unique type of knowledge that is invaluable to scientific research. As Lyall et al. (2004) describe, this group represents the “public downstream users,” or end-user groups whose daily experiences lend unique perspectives to understandings of science that can complement the scientists’ professional expertise and understandings of a particular research question (Lyall, 2004). This “experiential knowledge” brings a new and perhaps broader understanding to scientific investigations that are often times too narrowly focused on the pathology of the disease under study. For example, a patient-subject living with a particular condition may complement the scientist’s standard approach to their research practices by revealing strategies they use to cope with their disorder, thereby rendering more appropriate research practices that takes into account the individuals’ own experiences and, as a result, could lead to more accurate research results. As Caron-Flinterman et al. (2007) argue, “If one aims to enhance the moral and political legitimacy and the quality of biomedical research, patients and their knowledge need to be involved in decision-making processes” (p. 341). A combined body of knowledge generated by multiple sources of experiential knowledge on a given
condition has been termed as “experiential expertise” (Caron-Flinterman et al., 2005, p. 257). The success of the HIV/AIDS patient groups in the nineteen eighties in prohibiting certain researcher’s standardized approaches to randomized clinical drug trials and the use of placebos has been largely attributed, as Steven Epstein argues in his 1995 article “The Construction of Lay Expertise: AIDS Activism and the Forging of Credibility in the Reform of Clinical Trials”, to the collection of invaluable experiential expertise. Epstein notes “[the HIV/AIDS activism] case demonstrates that activist movements, through amassing different forms of credibility, can in certain circumstances become genuine participants in the construction of scientific knowledge– that they can (within definite limits) effect changes both in the epistemic practices of biomedical research and in the therapeutic techniques of medical care” (Epstein, 1995, p. 409).

There are many lines of thought, such as logical empiricism, that would denounce the notion of a lay citizen’s experiential expertise as a valid form of knowledge, and certainly there are those who would deny experiential knowledge as comparable to that of a professional scientists expertise. Indeed, the knowledge derived from professional expertise and that of lay citizens’ experiences do not hold the same weight in all circumstances, but from a pragmatist position, both can be acceptable forms of knowledge based on the context and the utility such knowledge brings to research. In a study conducted in 2005 by Caron-Flinterman et al., it was determined that the pragmatic utility of lay citizens’ experiential knowledge in influencing the practices of biomedical research studies did have potential value for biomedical research. Yet despite the studies’ acknowledgement of the influence of patient-subject’s experiential knowledge can have on the direction and practice of health research, the
study found that the actual involvement of patient-subjects is a rare occurrence in the biomedical research enterprise.

4.2 Deliberative Democratic Theory & Practice

The primary principles of participatory democratic theory, justice and equality, are the very foundation of deliberative democratic theory. One of the most prominent political theorists in recent times to apply deliberative theory into practice was James Fishkin and his introduction of the deliberative polls in 1988 (The Deliberative Democracy Handbook, p.7). Following in John Dewey and other postpositivist theorists idealistic view of a participatory democracy, Fishkin's notion of deliberative democratic engagements' aim was to engage diverse groups, involving lay citizens along with experts and professionals, in a democratic dialogue concerning contentious public policy issues.

All parties, citizens and experts alike, partake in a dialogue aimed to inform one another through a series of questions and reflections. After having listened to others' values and opinions and having rationalized one's own reasons to take a position on a given issue, the participants deliberate in order to collectively seek moral agreement when a consensus is possible, and maintain mutual respect when they cannot (Gutmann and Thompson, 1997). The goals of deliberative democratic processes are three-fold: (1) Legitimation: The process aims to generate inclusive, voluntary, reasoned, and equal dialogue amongst its participants, which will thereby render the outcome of such deliberations legitimate; (2) Justice: a reciprocal and informed deliberation will garner a just outcome in so far as participants engage in a cooperative
and shared dialogue, allowing the opportunity to give voice to all individuals involved in
the deliberation; and (3) Preference Formation: the process allows individuals to form
their preferences on a given issue after having been informed about the issue, been
given the opportunity to respectfully listens to others’ preferences and positions, and
been given the opportunity to voice their own values and beliefs (Gutmann and
Thompson, 1997). Therefore, a successful deliberative democratic event is one which is
1) well represented by all interested stakeholders, 2) all parties are able to exercise
their opinion and perspectives equally, 3) the participants are willing to listen, respect,
and try to understand others’ opinions and values, 4) participants are reflective on the
issues, and 5) participants are willing to change their initial preference during the
process of deliberation (Secko et al., 2008, p. 294). Conversely, some of the features
that may inhibit a deliberative event include instances where 1) adequate representation
of the community is not met (perhaps due to certain practical constraints, such as when
socioeconomic factors may deter certain communities of individuals to volunteer their
time to take part in the deliberation); 2) the participants have not been provided with
adequate level of information to be able to successfully deliberate on the issue (perhaps
the information provided is not at a level of understanding that is reasonably accessible
to the general community); 3) participants are not willing or able to participate on an
equal basis (perhaps due to power hierarchies, or certain voices overtaking the
deliberation, known as stakeholder capture); and/or 4) participants are unwilling to listen
and learn about other perspectives on the issue under deliberation.27

27 For a discussion of some of the failures of deliberative democratic events and proposed
solutions that may have mitigated these problems, please refer to: Fung, A., (2005) p. 404-412.
4.2.1 Types of Deliberative Democratic Participation

Over the course of the last thirty years, there has been an influx in the varying types of public engagement mechanisms that are premised on the ideals of deliberative democratic theory, namely citizens’ juries, planning cells, deliberative polling, consensus conferences, and citizens’ panels. Where deliberative polling and citizens’ panels more closely resemble traditional research methods such as survey and opinion polls, the citizens’ juries and planning cells have deliberation as their defining feature.\(^{28}\) Nonetheless, as Abelson et al. note “common to all is the deliberative component where participants are provided with information about the issue being considered, encouraged to discuss and challenge the information and consider each others’ views before making a final decision or recommendation for action” (Abelson et al., 2003, p.242.) Selecting the appropriate type of deliberative democratic event is dependent on the issue in question and the outcome that is being sought since, as Ryfe (2002) notes: “deliberation is inherently rooted in context, and different kinds of contexts demand different kinds of conversations” (p. 369). There are numerous examples of deliberative democratic engagement events that have been implemented for a variety of aims and to address a variety of issues. For example: Citizens Assemblies and other public engagement mechanisms have been used to address questions from appropriate electoral reform practices to meeting limited health care resource challenges.

In British Columbia, Canada, the B.C. Citizens Assembly was established with a mandate to look at how votes cast in provincial elections translate into seats in the

---

\(^{28}\) For a discussion on the methodological design and evaluation of the various types of public engagement events refer to Abelson, J. et al. (2003) “Deliberations about deliberative methods: issues in the design and evaluation of public participation processes.”
Legislature and make recommendations for a new propositional electoral system in B.C. The result was the final report, issued in 2005, which led to the proposal known as BC-STV, which provided BC residents to move towards the Single Transferable Voting System. Although the majority of BC citizens voted in favor of BC-STV, it did not reach the required 60% super-majority required in the legislation. Nevertheless, the BC Citizens Assembly was seen as a success in ushering in the first deliberative public engagement event of that size in B.C. In the UK and New Zealand, citizens’ juries became increasingly popular during the 1990’s as a means of gaining public input into decisions affecting health care rationing and priority setting (Abelson et al., p.243). Similarly, in the U.S. the Oregon Health Services Commission (HSC) created the Oregon Health Plan in 1991 with a prioritized list of health services in order to establish the range of health care benefits in Oregon’s Medicaid reform strategy (Garland, 1999, p.244). The HSC implemented a series of community meetings and public hearings in order to uncover the community values, combined with expert information, in order to shape the prioritization process. At the end of the participants’ deliberation, prevention and quality of life were ranked highest, followed by cost-effectiveness, ability to function and equity (Fung, p.407). Lowest on the list were values such as: mental health and chemical dependency, personal choice, community compassion, impact on society, length of life, and personal responsibility. Though the deliberation itself has been seen to as a successful deliberation, critics of the outcome have pointed to the bias in the groups’ representation as a reason for low ranking items being clearly linked to socioeconomic status; of the 1 003 participants, 77% were college graduates, 34% had

The BC Citizens Assembly website and final report can be accessed at: http://www.citizensassembly.bc.ca/public
a household income of over $50 000, and 70% were health care or mental health workers (Fung, p. 407). Measures, such as utilizing a random sampling recruitment strategy, would have helped to recruit a more equal representation of the Oregon community. In addition to political and health care issues, deliberative democratic engagement events have been useful in addressing complex ethical, political, and legal issues pertaining to conflicting policy options in health research endeavors.

4.3 Deliberative Democracy & Health Research

Increasing globalization of health research, such as the trend in recent years towards large scale, multi-site, international clinical trials, along with progressively sophisticated scientific and technological innovations, such as the advent of personalized genomic research, bring to bear even more complex questions concerning ethical health research practices. Many have argued that implementing citizen engagement mechanisms in policy debates on health research will aid in addressing a number of the issues that threaten the structure of governance; that is, deficits of knowledge, trust, and legitimacy (Horlick-Jones et al., 2007, p. 259). Citizen engagement mechanisms address these deficits by aiming to fully inform and properly educate the public on science and technology issues, thereby bringing an informed public into the debate on complex scientific research questions. As such, it follows that the public at large will be apt to place greater trust in such an inclusive and transparent system, which, in turn, will render a more legitimate system of governance, one that is accountable to all parties involved.
There have been a number of cases of deliberative democratic engagement events that centre on questions pertaining to health research worldwide. Yet, perhaps not surprisingly, one of the most common types of health research to employ public engagement mechanisms is the genetic/genomic, or biomedical research context and, most notably, research involving biobanking. For example, in 1998 the UK Biobank project was funded through the Medical Research Council, the Wellcome Trust and the UK Department of Health, developed the largest biobank worldwide. The project recruited 500,000 participants in order to explore the interaction of environment and genetic factors for common diseases (Avard et al., 2009, p. 8-9). The UK Biobank was established under the pretention that the wider public would be involved in decisions pertaining to appropriate governance and research ethics measures of the Biobank. Specifically, the public consultations involving deliberative forums aimed to address public attitudes towards consent, confidentiality, and security of data, commercialization, governance, recruitment and the communication of genetic information. In the People, Science, and Social Policy Report, it was noted that the consultation failed to sufficiently represent all of the social groups and that further consultation was warranted (Avard et al., 2009, p. 8).

Similarly, in Canada, the Providence of Quebec established the biobank project CARTaGENE in order to better the role genetics and the environment played in the health of the general population. In 2003, public participation mechanisms were employed in order to gain public trust by engaging with and listening to the public's views towards CARTaGENE policies and procedures similar to the aims of the UK

---

30 CARTaGENE project website is: http://www.cartagene.qc.ca/index.php?lang=english
Biobank, which have been received favorably in public participation feedback (Avard et al., 2009, p.7). Deliberative democratic events continue to be a popular mechanism in gaining public into complex ethical and methodological issues with the potential to have a huge impact on the greater good of society. Currently, public engagement events are being held and evaluated at the University of British Columbia’s Centre for Applied Ethics for the purposes of providing policy advice on the governance of the upcoming B.C. biobank, known as the BC biolibrary.\(^{31}\) This public engagement initiative will employ a similar structure and design as the pilot project that UBC’s Dr Michael Burgess initiated in 2005. The objective of this pilot project was to bring together a stratified random sample of the BC public to discuss a hypothetical biobank design in order to understand the public values that should shape the governance of a British Columbia biobank.\(^{32}\)

### 4.5 Conclusion

This chapter outlined the substantial evidence, supported by disciplines as diverse as political science, philosophy, and social science and technology studies, of the invaluable contribution lay knowledge has to science policy decision-making. This recognition should lead health researchers to acknowledge the inherent importance of human subjects’ own experiential expertise and how they may be able to contribute to the research enterprise as active partners. By adapting to the same guiding principles of

---

\(^{31}\) The BC biolibrary project website is: www.bcbiolibrary.icapture.ubc.ca

participatory democratic theory, principles based on equality and justice, and bringing together a range of epistemologies to derive more informed and inclusive decision-making practices, the health research enterprise, as a transparent and accountable system, will warrant greater public trust in medical research.

In the final chapter that follows, I shall investigate some of the ethical implications of involving consumers in decision-making concerning health research agendas, through the use of case examples provided in chapter three, as well as some of the empirical work that has been done to date on the effectiveness of human subject as partners in health research. Following my analysis I shall conclude with my recommendations for appropriate means of engaging consumers in partnerships with scientific communities, with reference to my analysis of deliberative democratic engagements in chapter four.
Chapter V
Ethical Challenges and Recommendations

In designing and conducting research, researchers should consider their relationship to participants as a form of collaboration, even in fields where participants do not (indeed cannot) contribute to the design of the research. The touchstone for the researcher should be to respect the welfare, autonomy, and equal moral status of all participants. That will engender trust, and the trust of individual participants, as well as public trust, is necessary for the research process. (TCPS Draft 2nd Edition, 2008)

Throughout this paper my aim has been to argue for a more accountable and inclusive governance system through the promotion of effective collaboration in all areas of health research, whenever possible, within the Canadian guidelines on ethical health research practice. I have argued this position by first reflecting in Chapter two on the changing landscape of research ethics and the transformative role of the human subject from passive object to that of an increasingly active agent in health research. The case examples in Chapter three bring to light instances in which effective engagement of human subjects as active agents with scientific communities led to successful research partnerships with mutually beneficial outcomes. Whereas Chapter four argues for deliberative democratic theory as the framework for collaborative research practice and takes deliberative democratic engagements as an example of ways in which lay public and/or human subject communities may be effectively and meaningfully included in decisions pertaining to the governance and priority setting of a particular health research endeavor.

Although there is evidence that the ideal of collaborative partnerships within health research is widely supported, as I have argued elsewhere, the actual active
engagement of research subjects into decisions that may affect the priorities, governance, and conduct of a health research initiative is rarely seen in practice. This is predominantly the case for publically funded health research in Canada, and most likely even less common for privately funded research initiatives. As the opening quote in this Chapter states, the newest draft of Canada’s Tri-Council Policy Statement emphasizes the value of research collaboration as a means of respecting all human subjects and garnering public trust. Although this is a step in the right direction, the TCPS guidelines do not provide any further insight on how researchers across the wide range of health research initiatives can aim to foster meaningful and effective collaborative relationships. The only section of the policy statement that provides any informative guidance on appropriate ethical and effective collaborative partnerships remains with research involving vulnerable populations in Chapter nine “Research Involving Aboriginal People.” In addition to the TCPS policy guidelines, the Canadian Institute of Health Research established the Aboriginal Ethics Working Group (AEWG) in March 2004 in order to set out another set of ethics guidelines for Aboriginal people for all research funded through CIHR. The final guidelines were issued in May 2007. As a more substantive set of guidelines for research involving Aboriginal people than the first edition of the TCPS, the CIHR guidelines have contributed to the 2nd Draft of the TCPS’ section on research involving Aboriginal people.

In light of the arguments I have made throughout this paper on the value and benefits of collaborative research relationships, I propose that the guidelines that have been developed for research involving Aboriginal People serve as a template to be applied more generally within the guidance framework for all areas of Canadian health
research initiatives that are conducive to such collaborative engagements. In doing so, Canadian policy directives will follow in the international trend that has developed over the course of the last two decades towards mandating the inclusion of public participants into the processes of health research (Boote et al., 2001, p.214). In order to further support this central thesis argument, the aim of this Chapter will first be to address the ethical challenges that collaborative research relationships may bring to bear on the conduct of health research. Following this discussion I shall review the, albeit limited, empirical work that has been done to assess the value, impact, and effectiveness of those collaborative research projects that have been implemented in practice. Lastly, I shall conclude this paper with reflections on recommendations for ethical, effective and meaningful collaborative approaches to research in the Canadian context.

5.1 Challenges of Collaborative Research

The benefits of collaborative research partnerships between research subjects and scientific communities, as I have stated elsewhere, can include: creating a more informed lay public in regards to the value and process of scientific research; providing subjects the opportunity to voice their opinions on the research priorities, helping frame the research question, and/or influence the research design; increasing accountability for all parties involved (subjects, researchers, and funding agencies); research results may be disseminated back to the research subject community more effectively; and increasing public trust in the research endeavor overall (Shea et al., 2005, p. 354). What are decisively more difficult issues to address are the numerous methodological,
practical, and ethical challenges that these very partnerships may afford the research endeavor.

Some of the key problems that have been identified in a paper by McCormick et al. (2004) on the problems arising in collaborative relationships involving breast cancer research, which I believe can be applied to health research more generally, are described in three distinct areas: (1) Relationship Issues, (2) Methodological Issues, and (3) Social, Political, and Cultural Problems. Firstly, Relationship issues are identified as those issues that pertain to a lack in interest from the patient-subject community in becoming active partners in research, as well as a lack of resources that are necessary to participate (e.g. resources that would allow patient-subjects to take the time out of their day to participate), a lack of trust (in the researcher themselves or research institution from the patient-subject perspective), inequitable power distribution amongst participants and scientists, and changes in the ideas and/or goals of the research project in question. Secondly, Methodological issues are identified as the problem that arises when non traditional methods are used to capture the values, opinions and concerns of the lay participant, thereby rendering the research approach susceptible to critiques towards its credibility as a sound scientific pursuit. And Thirdly, Social, Political and Cultural Problems include the possibility of receiving input from a certain sector of the patient-subject population, thereby missing out on other key perspectives, and the reality that expectations of the patient-subjects may be quite different from that of the scientific community (e.g. patient-subjects may expect to see quick results from the research project).
In support of the claims made by McCormick et al. in the identification of social, political and cultural problems that have the potential to impede the success of collaborative research engagements, is Tallon and co-authors’ results from their research study as reported in their 2000 paper “Relation between Agendas of the Research Community and the Research Consumer.” This study reported on the conflict that arises when scientist-researcher and human subject community’s research agendas are not in line with the other. In certain instances, one party’s research agenda may well warrant priority over the other, such as when it is widely held amongst a range of experts and publics, and there is evidence to support it, that a particular research agenda ought to be advanced ahead of a patient-interest groups. This may be the case during a National, or International pandemic, when research funding in the public sector may need to shift focus away from certain sectors, for the interest of public health. In other cases, it may be appropriate for agendas to merge, such as cases where research on direct care and basic science can be merged towards one research aim. As a recommendation Tallon et al. advocate for more democratic research funding processes, as well as a mandatory reporting of funding sources and the degree to which the research project enlists consumer involvement (Tallon et al., 2000, p.2040).

In addition to the methodological and practical concerns that have been identified, important ethical challenges arising from collaborative research relationships must be addressed. Firstly, one of the most troubling concerns raised is the threat of fair distribution of research funding. The concern is that if it becomes the norm that patient-subject communities or established advocacy organizations partner with the research communities to set research priorities and agendas there will be little chance for fair and
equal access to research funding. This would have significant implications for small-scale disease groups, such as rare genetic disorders like PXE. Drawing back to the case of PXE International and the Breast Cancer Advocacy organizations, larger cohorts of patient-subjects from the Breast Cancer advocacy groups would fare far better in receiving research funding over smaller scale disease groups with rare, and even ultra rare, orphan diseases as the larger organization would have access to more resources (including far more individuals willing and able to become active participants in health research), and, hence, garner greater attention towards their cause. This is an even greater concern in the Canadian research context where no government policy currently exists to draw special attention to and funding for rare orphan diseases.\footnote{In contrast, many other Western countries do offer government funded programs for rare orphane diseases. For example, in the USA the Rare Orphan Disease Act, aimed to provide incentives for drug developers to manufacture treatment specifically targeted for rare diseases was first enacted in 1983.}

A second ethical issue arising from collaborative research relationships is the tensions resulting from having to act in dual capacities as a human subject and consumer-collaborator. Much like some critiques that have been raised against the potential for bias in the clinician-researcher’s dual role and, hence, conflicting responsibilities towards their patients-subjects as care giver and scientist, the dual role of a human subject/consumer-partner may present its very own biases and conflicting responsibilities. Biases of the human subject/consumer-partner roles may arise in their ability to adequately represent the interests and opinions of the average human subject. For example, if a patient-subject is involved in a genetic research study where they must give blood and tissue samples, yet are also involved in the design of the research study and deliberations with the research team on best research practices on such
ethical concerns as privacy, with the intent on representing the interests of the larger pool of human subjects, their opinions may become skewed in light of these deliberations and insights from the research team. In effect, it may occur that over time they cannot represent the average patient-subject even though they share the same genetic condition since their understandings of science, policy, and research have been influenced in a way that the average human subject would not be. In addition to the potential for bias, this dual role may also present tensions if presented with conflicting responsibilities. Again, much like the dual role of the clinician-researcher, the human subject/consumer-collaborator has responsibilities to both to the interests of the larger pool of human subjects taking part in the research endeavor, as well as to the interests of the research project itself and members of the research team.

Tied to this last issue concerning the dual role of human subjects/consumer-collaborator is the challenge as to whether their understandings of risk can become compromised. Due to the potential for their dual responsibilities to conflict at times, it may become increasingly difficult to appropriately assess the research risks in the same way as they might have been capable had they not the same degree of invested interest in the outcome of the research. For human subjects alone, it is often times difficult to assess and weigh research risks and benefits, particularly so when that human subject has a particular disease condition and limited treatment options. But tack on another added layer of complexity, such as becoming personally and perhaps even emotionally invested in the design and outcome of the research project, and one’s ability to assess the research risks in any rational manner may become compromised.
5.1.1 Reflections on Methodological and Ethical Challenges

The many methodological, practical, and ethical challenges that have been raised and identified as potential roadblocks are well warranted concerns that must be addressed in order for effective collaborative research relationships to ensue. Nevertheless, none of the methodological and/or practical as well as ethical concerns gives ground for dismissing the inherent value of involving collaborative approaches to all health research projects that warrant and are capable of supporting collaborative endeavors. For this reason, I wish to address a number of the issues that have been raised with suggestions for possible solutions.

(1) Relationship Issues

A lack of interest in becoming an active partner in a given research endeavor as well as lack of resources are surely valid issues that may be a real deterrent for some health research projects to implement collaborative research practices. However, it must also be stressed that there are many varying degrees of public participation options that require little time, effort, and resources. For example, researchers may think about conducting a brief survey and/or interview (either face-to-face, on the phone, or via internet) at the end of the human subjects’ involvement in the research study in order to gain feedback from their subjects on how they felt about the research design, (e.g. how they felt about their approaches to consent process, or the number of times they had to come in for an appointment) and what this experience was like for them. In this regard, if the researcher’s aim was initially to receive human subjects’ input at the initial stages of the study’s design, but did not have the resources to train and/or there was a lack of volunteers from the human subject pool, it would still
be beneficial to gain human subjects’ perspectives on the research design elements after having completed the study, either to enhance the study if it is ongoing, or in preparation as lessons learned for the any subsequent research studies.

In regards to issues around a lack of trust and an inequality of power between human subjects and researchers, when such concerns are present it would be highly beneficial to borrow from participatory democratic models of thought and engage in deliberative democratic processes, such as the model of a citizens jury. The fundamental aim, as noted in Chapter Four, is to generate a dialogue where one is free to share their position and values on a given topic in a manner that is respectful and open to new and different modes of thought. As such, the aim is to generate trust through open, democratic, and informed deliberation.

(2) Methodological Issues

The second concern was raised over the legitimacy of non-traditional methods of inquiry. Although this may be the case in certain instances, this claim cannot hold without an assessment of the efficacy and utility of non-traditional methodologies against the efficacy and utility of traditional methods of inquiry. Therefore, all health research initiatives with a participatory framework ought to require that measures of assessment be established into its design in order to determine whether the benefits of these non-traditional methods of inquiry are as good as or an improvement over traditional methodology on the same or similar research topic. This may be achieved through the use of survey and/or interview techniques with the participants and scientific communities while the research is ongoing and/or after the research is complete, as
well as through a comparison of research study results with a collaborative component against similar research endeavors that did not involve a participatory framework.

(3) Social, Political, and Cultural Problems

The notion that involving some human subjects as consumer-collaborators, in cases when all of the human subjects cannot partake in a collaborative relationship with the research team, may present problems if key perspectives are missing. This is a valid concern, yet surely not a new one. This is indeed an issue common to all democratic societies seeking valid representation. Nevertheless, there are several means of getting around this issue when this is presented as a concern for a particular research endeavor. One is to ensure that all relevant perspectives have the chance to be heard by creating partnerships with research subjects on a rotational basis. This, for instance, would make sense for cases where a projects’ advisory board or research ethics oversight committee required one (or more) human subject to sit on the board as a consumer member. Another method, if rotational membership is not possible, would be to ensure that the human subject/ consumer-collaborator is systematically checking back in with the human subject pool at large on a regular basis to listen to their concerns and preferences (subject to continual reporting and meeting with the human subject community being, of course, both appropriate and practical).

In regards to the second concern raised under this category, that human subjects and research community may have conflicting research agendas, this reality must also be addressed in the same manner when faced with a lack of trust in research and/or the researcher. Deliberative democratic ideals aim to lay out all of the participants’ values,
opinions, reasons, as well as to inform participants of the realities of the research design. This method of engaging the lay public with the scientist community would be optimal in that its strategy would be to fully inform both parties of the aims and agendas so that, once these were understood, and even if they cannot come to a consensus, they may respectfully accept one another’s positions.

(4) Funding

The ethical challenge involving the prospect of having patient organizations or other organized community groups increasingly influence research priorities and funding decisions is a notable concern. For this reason, it would be wise to follow the suggestion raised by Rebecca Dresser in *Science as Salvation* (2001). That is, we ought to involve a deliberative democratic approach to funding allocation. As Dresser notes, “in the context of allocating research funds, distributive justice principles impose on the government a duty to divide resources in a way that gives individuals a fair opportunity to benefit from federally supported research,” and as such “this duty can be invoked to rule out extreme positions, such as directing all funds to research concentrated on a single disease, the health problems of one sex or ethnic group, or a single scientific topic” (Dresser, 2001, p. 101). Hence, potential ethical challenges such as that discussed earlier of having large scale, powerful advocacy organizations direct attention and funds towards well known diseases, such as Breast Cancer research, and away from much smaller scale disease groups, such as PXE, is avoided. In fact, deliberative democratic approaches may benefit those seldom heard from disease
groups in that in Canada there are few opportunities for individuals living with rare forms of a genetic disorder to be acknowledged by the research community at large.

(5) Conflicting Roles and Responsibilities

The second ethical issue that is identified above concerning the dual role of human subject/consumer-collaborators and the potential for conflicts of interest to arise is a concern not inherent to human subjects alone. As I’ve pointed out, similar critiques are made against clinicians who also act as research investigator, thereby risking a conflict of interest in relation to their responsibility towards their patient-subject. Where clinician-researchers ought to implement safeguards in their research practices, such as requiring third parties to recruit and obtain informed consent from their patient-subjects in order to avoid influencing their free choice, similar suggestions ought to follow for human subject/consumer-collaborators. Safeguards ought to be implemented in partnerships that may run the risk of creating bias and/or conflict in one’s responsibilities. For example, debriefing techniques may be required of human subject/consumer-collaborators after having participated in a research project, if the research results may risk being biased by the consumer-collaborator role. Such techniques aim to allow the participant opportunities of self-reflection concerning their role and responsibility, thereby rendering a more transparent research process.

(6) Risk Assessment

The final ethical issue that is discussed is, as I mentioned earlier, tied to the dual role of the human subject as partner. One’s ability to judge, from a rational and

---

unhindered point of view, the level of risk involved in a given research protocol is of real
cconcern for research partnerships, one that runs the risk of rendering human subjects
involved in such partnerships more vulnerable to harm, rather than its objective of
empowering subjects. In light of this challenge, it is imperative for all partnerships in
health research to ensure certain safeguards are met. For instance, informed consent
cannot end with the signing of a consent document, but rather it ought to be a process
that is implemented throughout the course of the research project. In this way,
researchers and research workers can check back with the human subjects at various
points in the study in order to assess their understandings of risk, motivations to
continue (or discontinue) with the study, and their level of understanding of the
implications of the research process and findings.

The above proposed solutions to some of the potential challenges that research
partnerships may face are only a few of the safeguards and mechanisms that may be
useful for some types of health research studies. Actual solutions to potential
challenges are context dependent, based on the type of research being studied, the
resources available, and the nature of the human subjects’ involvement in the health
research initiative. As Dresser further notes, “although opening the door to advocacy
participation carries risks, they are not so severe as to justify excluding advocates from
the process. Affected groups have a legitimate interest in having their values and
preferences represented” (p.101).
5.2 Assessing Collaborative Health Research

After having laid out the potential challenges that collaborative research partnerships may raise for health research and offering a few suggestions in order to avoid or significantly reduce their impact on health research, I now turn to reflections on empirical assessments of collaborative health research endeavors. A number of studies and literature reviews have been published on the recent prevalence of partnerships in health research, papers aimed to offer critiques of its obstacles, report on its benefits, and provide reflective recommendations for future participatory research endeavors (Oliver et al., 2008; Wright et al., 2006; McCormick et al., 2004; Caron-Flinterman et al., 2007; Stevens et al., 2003; Gray et al., 2000; Boote et al., 2002; Hubbard et al., 2008). All of the research into collaborative health research practices was in favor of using participatory methods of engaging human subjects in health research, with the exception of a few that called for more meaningful research partnerships and increased power for the human subjects involved (Stevens et al., 2003), equal balance in the research design objectives and clearer picture of the obstacles, in order to garner more realistic participatory methods (Gray et al., 2000), and more empirical work in the area in order to properly assess the value of participatory research methodologies in health research (Boote et al., 2002).

In addition, all but three of these published papers were from the United Kingdom and the majority focused on research involving breast cancer patients. This reflection may not be overly surprising when taking into consideration the literature review by Hubbard et al. (2007) on the activity of research involving public participation in cancer research. As Hubbard states “most articles [out of a total of 3607 documents] originated
from the USA or the UK, which suggests that the agenda of involvement is more advanced in these two countries or shows that there is more publication activity in these two countries than elsewhere,” and in addition “involvement activity is most prominent around women with breast cancer, which suggests that this group of patients to date made the most significant progress in advocating their involvement in research” (Hubbard et al., 2007, p. 236). Indeed, this statement is supported by other research that suggests the UK is the leader in instituting public participation methodologies in health research. As Boote et al. indicate, funding and regulatory bodies in the UK have instituted policy directives to involve patients and the public in decisions pertaining to research and development (p. 214). In 2001, the Health and Social Care Act was enacted with the aim of requiring UK health authorities, Primary Care Trusts and NHS Trusts to require the involvement and consultation of patients in the development of health services (Wright et al., 2006, p. 4). In addition, there is explicit commitment to patient involvement detailed in the UK’s ‘Research and Development for First Class Service: R&D Funding in the New NHS’ (Wright et al., 2006). Although efforts are most prominent in areas such as the UK and USA, public participation is being implemented in a variety of health research projects on an international level as well as across Canada. As I mentioned in Chapter four, public participation in biobanking research has been and is currently being implemented in several institutions across Canada (CARTsGEN project in Montreal and the BC Biolibrary in Vancouver, B.C.). In addition, consumer organizations such as the Cochrane Musculoskeletal Group (CMSG), also based in Canada, continues to work to increase consumer participation in determining

Some of the assessments on public engagement strategies in health research have included similar challenges to the efficacy of participatory methodology as I listed early. In particular, issues relating to difficulties in funding participatory methods of inquiry, and challenges to the legitimacy of research involving public engagement events were identified, as were issues around distrust, lack of patient-subject expertise, and unequal power dynamics were noted as obstacles to the success of participatory methodologies in health research (McCormick et al., 2004, p. 635-636). Nevertheless, the majority of empirical work showed that participatory methodologies provided a valuable contribution of health research. As Hubbard et al. note, as a result of participating as partners in breast cancer research “the women involved described feeling empowered and changed their feelings towards scientists from fear and anxiety to mutual respect. Similarly, some of the prejudices and preconceptions held by scientists about ‘hysterical’ women with breast cancer soon dissipated” (Hubbard et al., 2007, p. 239). Other findings saw participatory approaches as enhancing the appropriateness of research findings and methodologies that were used (Wright et al., 2006, p.11-12). Despite these findings that show meaningful and effective partnerships in health research, it was largely reported in the literature that collaborative research engagement continues to be far from the norm in health research practice. Even in the UK, where NHS have ushered in policy incentives to promote public participation practices, according to a report published in 2001, only 42% of NHS providers receiving
R&D Support Funding involved patients in their research activities (Wright et al., 2006, p. 4).

5.3 Conclusion: Recommendations for the Canadian Context

Through an examination of the movement towards more democratic and participatory methods of inquiry, this paper has argued on both moral and political grounds that there is an imperative for Canadian policy on the ethics of health research to mandate more meaningful and effective partnerships between the human subject and research community. I do not seek to attempt to argue the degree human subjects ought to be involved in all types of health research partnerships, since that is context dependent based on the type of health research initiative and protocol, and the preferences of the human subject and health researchers involved. Determining appropriate degrees of human subject participation requires an empirical investigation into the utility of subject involvement across the spectrum of health research studies and some experimentation with different forms of partnerships in a variety of social settings.

Rather, the aim of this paper has been to point to Canada's position as one of the most influential leaders in International standard setting for ethical conduct of research involving human subjects and, as such, advocate the need to incorporate collaborative mechanisms in similar modalities as other leading democratic countries as the U.S. and the UK. With Canada's Tri-Council Policy Statement's section on the ethical conduct of research engagements with aboriginal communities and the more detailed CIHR guidelines, the framework is already set. As such, I wish to make several
general recommendations over the ways in which the TCPS and CIHR guidelines may be applied more generally to other self-identifying groups or communities of human subjects with shared interests and/or histories.

Article 3 of the CIHR guidelines stipulates that Aboriginal communities ought to be given the option of a participatory-research approach, one in which “will result in shared power, equitable resourcing and mutual understanding, and will help the research proceed in a manner that is culturally sensitive, relevant, respectful, responsive, equitable and reciprocal with regard to the benefits shared between the research parties and the Aboriginal community” (CIHR, 2007, Article 3). This is certainly a requirement that ought to be applied to all health research endeavors that are conducive to and could benefit from employing participatory methodologies. In Article 3, the CIHR guideline promotes sentiments of equality, justice, and fairness akin to the ideals of a deliberative democratic endeavor. Recommendations such as having ongoing meaningful and active collaboration where “the parties establish a dialogue allowing them to find solutions in an atmosphere of mutual respect in good faith, with full and equitable participation. Consultation requires time and an effective system for communicating among those who hold an interest in the research” (CIHR, 2007, Article 3). Thus, extending these recommendations to research endeavors with human subjects more generally could be attained through consultation workshops and meetings between human subject and researcher communities based on principles of deliberative democracy in order to come to mutually beneficial agreements concerning the approach the participatory-research endeavor may take, which can then be written
up in the form of a “Memorandum of Understanding,” similarly to the approach outlined in Section 2.15 of CIHR’s Guidelines for Health Research involving Aboriginal People.

In addition to providing human subjects the option to implement a participatory-research approach in the design of the research study whenever possible, the CIHR guidelines also stipulate specific measures that the research community must take to ensure respect for the participating community. The guidelines stipulate that the researchers ought to respect the perspectives of aboriginal communities concerns over individual anonymity, privacy, and confidentiality issues (Article 5), intellectual property rights (Article 8), rights and proprietary interests of individual’s biological data (Article 12), and their involvement in the dissemination of research results (Articles 14 and 15), all of which must be addressed in a written research agreement. Although there are certainly historical reasons that Aboriginal communities are specifically owed special attention and sensitivity concerning these highly contentious ethical issues, these issues are concerns shared by many other human subject communities. For instance, these requirements are similar in the design and execution of the biobank owned and operated by the rare genetic disorders group PXE International, as well as Genetic Alliance. Thus, it is evident that similar guidelines outlined in the CIHR guidelines for Aboriginal People may be appropriate for a variety of other human subject groups. Taking the rare disease groups as an example again, individuals and family members with rare forms of a genetic disease are particularly at risk to being easily identifiable in a given research initiative due to the uniqueness of their condition and small population size. Therefore, these individuals as a community require added protection and understandings about their confidentiality and privacy concerns (which may include
concerns over their employment and or life insurance if identified) by the researcher community. This may be achieved by instituting stipulations such as the CIHR guidelines which state that in order for any third party to access biological data or samples, it can only be done with the consent of the researchers, individuals and community members (CIHR, 2007, Article 12).

In addition, the CIHR guidelines lay out a number of requirements that the research community owes to the Aboriginal communities. Article 9 states that the research question should be of interest to and of benefit for both the researchers and Aboriginal communities, Article 10 states that the researcher ought to support the education and training of Aboriginal members in research methods and ethics, and Article 11 stipulates that the researchers are obligated to learn about the culture of the Aboriginal community. Again, these requirements are reflective of the ideals of a successful deliberative democratic endeavor that has aims that are mutually beneficial to all parties involved, requires that parties are open to listening to and understanding other stakeholders’ perspectives and interests, and that all participants in the endeavor ought to be fully informed and educated about the issue at hand. Therefore, these requirements can and should also be easily made applicable more generally as a requirement for all health research endeavors in order to promote deliberative democratic ideals in the culture of health research.

In addition to the requirements outlined in the fifteen articles, the CIHR guidelines provides a detailed outline of the requirements that the researchers owe to the human subjects throughout all stages of the development of the research protocol and the research process (CIHR, 2007, Section 3.2). At each level of the research process,
from protocol design, to the process of consent, to the recruitment of subjects, to the
data collection, analysis, interpretation and dissemination of research results, it
stipulates that the Aboriginal community ought to be involved and/or informed, in some
capacity, at all stages, and provides a list of each element that researchers ought to
consider. Although this may not be applicable and practical for all types of health
research endeavors, the template that this document can offer to health research
initiatives in general is invaluable in providing precise guidance for health researchers
when meaningful and effective collaborative research initiatives are attainable.

As the first quote in this Chapter highlights, the TCPS guidelines acknowledge
the dependency of health research on public support and trust. Therefore, by ensuring
that the public, and human subjects more specifically, are included, in varying degrees,
in the development of Canadian policy concerning health research priority setting,
practice, and governance, through such means as I have recommended in the brief
outline above, a more transparent and accountable system will ensue and, hence, the
health research enterprise as a whole will engender public trust.
BIBLIOGRAPHY

45 Code of Federal Regulations S 46.102(f)


Faden, R. R., Ledere, S.E., & Moreno, J.D. (1996). U.S. Medical Researchers,
the Nuremberg Doctors Trial, and the Nuremberg Code: A Review of Findings of
the Advisory Committee on Human Radiation Experiments. JAMA, 276 (20),
1667-1671.

Expertise: From Theoretical Inquiry to Practical Cases. Policy Sciences, 26(3):
165-187.

York: Routledge.

Deliberative Democracy in an Unjust World. Political Theory, 33 (2), 397-419.

Policy. Public Understanding of Science, 8 (3), 241-254.

Strategies for Effective Civic Engagement in the Twenty-First Century. San

Incidence, 1980-2006: Combined Roles of Menopausal Hormone Therapy,
Screening Mammography, and Estrogen Receptor Status. Journal of the
National Cancer Institute, 99 (15), 1152-1161.

Research: Reflections on a Study with Breast Cancer Self-help Groups. Health
Expectations, 3 (4), 243-252.

Center Report, 27(3), 38-41.


as Information Systems: The Role of Knowledge and the Concept of Translation
Quality. Public Understanding of Science, 16 (3), 259-278.

Involving People Affected by Cancer in Research: a Review of the Literature.
European Journal of Cancer Care, 17 (3), 233-244.

Interagency Advisory Panel on Research Ethics: Canadian Institutes of Health
Research, Natural Sciences and Engineering Research Council of Canada,
Social Sciences and Humanities Research Council of Canada. (December


*Moore v. Regents of University of California, 51 Cal. 3d 120, 132-133, 793 P. 2d 479 Cal. Rptr. 146, 153 (1990)*


Sade, R. M. (2002). Research on Stored Biological Samples is Still Research. *Archives of Internal Medicine, 162*, 1439-1440.


