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Social Contract Theory and Just Decision Making: Lessons from Genetic Testing for the BRCA Mutations

ABSTRACT. Decisions about funding health services are crucial to controlling costs in health care insurance plans, yet they encounter serious challenges from intellectual property protection—e.g., patents—of health care services. Using Myriad Genetics' commercial genetic susceptibility test for hereditary breast cancer (BRCA testing) in the context of the Canadian health insurance system as a case study, this paper applies concepts from social contract theory to help develop more just and rational approaches to health care decision making. Specifically, Daniels's and Sabin's "accountability for reasonableness" is compared to broader notions of public consultation, demonstrating that expert assessments in specific decisions must be transparent and accountable and supplemented by public consultation.

The intersection of health insurance coverage and intellectual property arrangements such as patent protection is an international phenomenon. Every health care insurance system, whether private or public, spreads the cost of health care services across a population. The fact that intellectual property protection affects the costs and, therefore, the availability of health care services is relevant to all insurance systems. But unique aspects of culture, politics, and economics shape the responses of various components of a health care insurance system, and local responses are distinguishable from, although not independent of, national and global responses. Thus, the intersection of health care accessibility and intellectual property protection is best studied through examination of a local experience and tracking the relevant influences. The present analysis is based on the doctoral research of one of the authors (Williams-Jones), which entails a close study of one Canadian pro-

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vincial health insurance system's response to the Myriad Genetics patents and its implications for accessibility (Williams-Jones 2002a).

The majority of Canadians who access genetic services do so through the public health care system, but for those with the means, private purchase also is an option. The rapid expansion of the Internet and the creation of a "global marketplace" have made it possible for Canadians to purchase genetic testing through international sources (Williams-Jones 2003). With an agreement in 2000 between the U.S. biotechnology company Myriad Genetics and the Canadian diagnostic company MDS Laboratory Services, Canadians were able to purchase genetic testing locally for conditions such as hereditary breast or colorectal cancer (Myriad Genetics 2000a & b). This development is not without controversy, and there is ongoing public debate about whether the Canadian health care system should prohibit, permit, or encourage provision of and access to private health care services (Evans 2000; Mazankowski 2001; Romanow 2002).

In the context of genetic testing, the situation is further complicated by gene patents that confer to biotechnology companies controlling rights over genes, mutations, and susceptibility tests.[1] Gene patents can increase the cost of testing and restrict provision to particular licensees (Merz et al. 2002; Williams-Jones 2002b; Cho et al. 2003). Licensees that are private companies have an interest in marketing their products to patients to build demand for testing services (Hull and Prasad 2001; Gollust, Hull, and Wilfond 2002). If successful, marketing stimulates pressure to expand insurance or permit the growth of privately accessible services. In either case, a consequence of increased demand and use of genetic testing is an increased burden on publicly supported ancillary health care services, such as genetic counseling. Patents also may restrict downstream research into better, cheaper, or more accurate tests and therapeutics (Borger 1999; Knoppers, Hirtle, and Glass 1999; Heller and Eisenberg 1998). The potential costs raise serious concerns for continued expansion of public health care insurance to cover a growing number of genetic tests. The clear challenge to the ability of public health insurance to reduce or eliminate inequities based on access to health care services is a good reason to evaluate the benefits of strong intellectual property protection. It is critical to investigate the potential for restricting access to genetic services outside of the public system, evaluate which services are sufficiently beneficial to warrant coverage as part of public health care insurance, and explore the ethical role of public consultation and account-

ability in these evaluations. In using the case of BRCA testing in Canada, our intent is not to provide a definitive argument for or against the coverage of BRCA testing as part of public health care insurance, but instead to explore the elements necessary for such a decision-making process to be considered just.

JUSTICE IN ACCESS TO SERVICES AND PRIORITY SETTING

A common objection to access to private health care services is that it is unfair. Health, it is argued, is fundamental to the enjoyment of other important goods, such as the freedom to make life plans about one's career or education, thus it is the responsibility of a just society to ensure that all citizens have access to needed health care.[2] This view is a fundamental tenet of universal public health insurance schemes, namely that citizens should have equal access to needed health care services regardless of ability to pay. Such an egalitarian ethic is enshrined in Canadian legislation: According to the *Canada Health Act* (CHA), citizens are entitled to health insurance that is publicly administered, comprehensive, universal, portable, and accessible (Canada Health Act, R.S. 1985, c. C-6). Health care services covered under the CHA may not be made available for private purchase, and the federal government is empowered to withhold health transfer payments to provinces that permit such private services until those provinces comply with the CHA (Deber et al. 1998).[3] Provincial legislation in 6 of 10 provinces also prohibits private insurance for medically necessary hospital and physician services (Flood and Archibald 2001).

At first glance, federal and provincial legislation appear to guarantee public provision of all needed health care services and prohibition of private purchase. But these requirements apply only to an agreed upon set of "listed" hospital and physician services (Flood 1999). For "non-listed" services—such as dental care, many reproductive technologies, and most pharmaceuticals used outside hospitals[4] (Giacomini, Hurley, and Stoddart 2000)—commercial provision and private purchase is permitted. Despite the requirements for "comprehensiveness" and "universality" found in the CHA, some beneficial health care services continue not to be publicly insured and so are available only to people with sufficient wealth or private health insurance. In 2002, private spending on health care, including pharmaceuticals, complementary care, dental care, in Canada totaled C$32.9 billion (Canadian Institute for Health Information 2003).

Decision making about resource allocation in the context of public health insurance often occurs in a complex and interest-driven fashion,

influenced by a range of social, economic, and political factors (Flood 1999). The *ad hoc* nature of such decision making and the existing uneven distribution of benefits means that injustices occur—some people receive the medical services they need while others do not (Sherwin 1996; Caulfield et al. 2001). Just access to health care might be thought to entail equal access to every needed medical service, but taken literally and especially if "need" is construed broadly, this approach is unworkable. The current political reality is characterized by limited financial resources available for health care and the need to fund other important goods such as social services, education, and public works.

The cost of health care in Western countries continues to increase, in part as a result of pressure from a range of interests, including consumer groups, clinicians, and industry, to introduce new and often expensive medical diagnostics and treatments (Daniels 2001). In Canada, total public health care expenditures (in current dollars) increased from C$9 billion in 1975 to C$79 billion in 2002, corresponding to a rise in percent GDP from 7.3 percent in 1981 to 9.3 percent in 2001 (Canadian Institute for Health Information 2003). Recent budget surpluses and mounting public pressure to reform health care have been met by increased federal government cash transfers to the provinces and a new Health Reform Fund, totaling C$34.8 billion over five years (Department of Finance (Canada) 2003). But even this large investment may not be enough. For example, in the wake of the difficulties in containing the Severe Acute Respiratory Syndrome (SARS) outbreak in February 2003, the Canadian Medical Association (2003) has called for a further C$1.5 billion to address extraordinary health emergencies.

Whatever share of public funds governments spend on health care, the amount still will be insufficient to support all potentially beneficial services.[5] Decisions will be required to determine which services should be funded, and whether and how private purchase of services should be permitted (Evans et al. 2000; Deber et al. 1998; Caulfield et al. 2001). Before discussing how one might make such decisions, it is helpful to explore the issues that arise in the provision of a particular health care service, namely genetic susceptibility testing for hereditary breast and ovarian cancer.

PROVISION OF BRCA TESTING IN CANADA

The discovery and sequencing in the early 1990s of two genes associated with hereditary breast and ovarian cancer (BRCA1 and BRCA2) helped to make possible genetic susceptibility testing for patients to deter-

mine their risk status. This information may facilitate life planning, anxiety reduction, and access to specialized surveillance and prevention strategies. Treatments such as prophylactic surgery (Lynch, Lynch, and Rubinstein 2001; Hartmann et al. 2001) and tamoxifen (King et al. 2001) can significantly reduce (but not eliminate) the risk of developing cancer, and access to BRCA testing will be an important part of clinical management (Robson 2002).

In most provinces in Canada, BRCA testing is provided through public health care institutions—such as the Hereditary Cancer Program (HCP) at the B.C. Cancer Agency in Vancouver, British Columbia—as part of coordinated clinical oncology programs. These public programs have guidelines restricting access to patients with risk factors, such as a strong family history (multiple cases of breast or ovarian cancer), early age of onset (pre-menopausal), or membership in a specific ethnic group (e.g., Ashkenazi Jewish, French Canadian) (Carter 2001). Testing is not available to "patients off the street" with little or no family history of disease. But with the granting in 2000 and 2001 of four Canadian patents on the BRCA genes to Myriad Genetics, the continued public provision and control of genetic testing services has been jeopardized.

Myriad licensed MDS Laboratory Services—one of Canada's largest medical diagnostics companies—to be the exclusive Canadian provider of Myriad's patented *BRACAnalysis* test (Myriad Genetics 2000b), and began a campaign to convince Canadian health care institutions to comply with the patents and refer all tests to MDS or Myriad (Canadian Press 2001; Kent 2001a). This move generated strong professional and government opposition across Canada (Williams-Jones 2002b), and only British Columbia complied with Myriad's demands and ceased in-house testing. (Québec performs some mutation testing locally but sends index testing to Myriad for full analysis.)

In the spring of 2001, the B.C. Ministry of Health Services, on advice from legal counsel, informed HCP and the B.C. Cancer Agency that should they wish to continue providing BRCA testing, HCP would have to purchase testing from Myriad out of its existing operating budget. The cost of BRCA testing at HCP is approximately C$1,200 per test (Kent 2001a). The purchase of testing from Myriad at triple the cost—C$3,850 / US$2,400 for full sequencing—would quickly exhaust HCP's budget and undermine its ability and mandate to provide services to patients at risk for a variety of hereditary cancer syndromes. HCP halted BRCA testing in July 2001.

By complying with the Canadian BRCA patents, even when other provinces had rejected Myriad's patent claims and continued to provide in-house testing (Eggertson 2002), the B.C. Ministry of Health Services arguably took a stand in favor of protecting intellectual property rights. But given this position, to not then increase funding to HCP to cover the difference in cost of purchasing Myriad's test made the B.C. government complicit in the discontinuation of publicly provided BRCA testing in the province. In effect, the Ministry engaged in *de facto* priority setting, placing IP protection ahead of equitable access and establishing two categories of patients, those who could and those who could not afford the test. In February 2003, the B.C. Minister of Health Services authorized the resumption of in-house BRCA testing, in line with a reversal of the government's position on gene patenting. According to the Minister: "B.C. women and other future patients have a right to all the information they need to stay healthy. It is completely unethical to use patents based on genetic sequencing to block patients' access to their own genetic information, particularly when we already have the knowledge, ability and equipment to provide women with this information" (British Columbia Ministry of Health Services 2003). The Minister also called on the federal government to follow the lead of countries such as France and the Netherlands and oppose the Myriad patents, and to review the patenting of DNA more generally.

At the national level—and strictly speaking in contravention of the *Canada Health Act*—patients, for a period of two years, were treated differently depending on the province where they lived. Although this may be a somewhat inevitable feature of a provincially managed national health insurance system, the financial basis for the discrepancy and the establishment of two groups of patients within the province is an injustice. Since the Canada Health Act is the relevant social contract for fair distribution of health services, it is worth exploring how a social contract can be renegotiated or enforced.

SOCIAL CONTRACT THEORY

One approach to deliberating about fair allocation of health care resources is social contract theory, most notably elucidated in John Rawls's (1971) *A Theory of Justice*, in which society is organized in accordance with mutually beneficial principles of justice. We can adapt Rawls's approach to imagine how a group of rational, self-interested decision makers would develop principles for a just health care system. These decision

makers, in an ideal bargaining situation, do not know their current or future positions in society—e.g., social class, education, health status and needs—and can be trusted not to pursue their own interests at the expense of others. Of a range of principles that such decision makers might identify, Norman Daniels's version of the equality of opportunity principle has achieved some prominence in theorizing about just health care and priority setting (Daniels 1985, 2001).

Equality of Opportunity

The principle of equality of opportunity is rooted in a conception of justice that focuses on the creation of a "fair or level playing field" and requires not only the elimination of discrimination, but also efforts to ameliorate social factors that limit opportunity. Poverty and lack of education, as well as illness and disease, can have profound negative consequences for people's ability to pursue their life goals freely. One purpose of providing health care (or social assistance or public education) is to help people have a fair chance at pursuing their life goals and objectives so they can participate as full members of society (Daniels 1985; Sherwin 1996). A just society, then, should "remove the barriers to opportunity that are due to disease" (Buchanan et al. 2000, p. 16) through the provision of health care, so that people who are disadvantaged can become normally functioning members of society.

The principle of equality of opportunity provides a strong basis for an entitlement to health care, something that most Canadians support, but does not imply the unrealistic claim to all possible health care services. In using this principle as part of resource allocation decision-making processes about public health care services, one would support only those services that effectively help individuals maintain their health; other services could reasonably be denied public funding, although this would not necessarily restrict private purchase.

Daniels develops his notion of equality of opportunity in more detail by referring to: (1) Christopher Boorse's notion of "species normal functioning" as the goal of health care services (Boors 1975, 1976, 1977),[6] supplemented by (2) compensation for those with less than "normal functioning"—e.g., through symptomatic relief and support for chronic and disabling conditions. Daniels suggests that a sufficiently rich notion of equality of opportunity can include all the goals and services one reasonably would want in a health care system. Assessing the efficacy of genetic testing (or any health care service for that matter) for promoting (1) or (2)

depends largely on the social context—risks and benefits are different depending on existing treatments, family relationships, economic status, and confidentiality arrangements. An equality of opportunity perspective would ask whether a given genetic test or other health service is useful for (1) or (2) in general, then invoke clinical judgment and patient autonomy (or substituted judgment) for detailed evaluation of specific clinical applications. Conversely, if a particular type of surgery, medicine, or genetic test is not useful for (1) or (2), then equality of opportunity would not require that it be made available to clinicians or patients who might want it due to their unique circumstances.

Accountability for Reasonableness

Equality of opportunity may be a sufficiently robust principle for Daniels, but he also is concerned that in a pluralistic liberal democratic society, there will be differing views of what should constitute the substantive principles of justice for health care decision making. In collaboration with James Sabin, Daniels proposes a procedural approach to just decision making that is open to a wider range of principles and reasoning and is publicly accountable. Specifically, decision-making processes should be "accountable for reasonableness" and based on appeals to reasons that are "not only . . . publicly available, but [also] those that 'fair-minded' people can agree are relevant to pursuing appropriate patient care under necessary resource constraints" (Daniels and Sabin 1998, p. 51). Four conditions must be met for a process to be accountable for reasonableness: (1) Publicity: Rationales for coverage of new technologies must be transparent and publicly accessible; (2) Relevance: Rationales must be reasonable, that is based on appeals to evidence or principles that fair-minded parties accept as relevant; (3) Appeals: There must be mechanisms for challenges, ongoing review, and revision of decisions as new information develops or the context changes; and (4) Enforcement: Decision-making processes must be publicly regulated to ensure that the first three conditions are met (Daniels and Sabin 1998, p. 57).

By grounding decision-making processes on accountability for reasonableness, Daniels and Sabin explicitly invoke a rationalist, expert-driven model. Decision makers must be "fair minded" and willing to reason openly, seek mutually acceptable rules to narrow and resolve disagreements, and aim for the common good. The hope of this procedural justice approach to decision making is that even if a particular priority-setting decision does not please all stakeholders, such as particular groups of

patients, it still will have been arrived at in a manner that is open, transparent, and based on sound reasoning that is publicly accountable. Patients who are excluded from care need to know that their exclusion is reasonable and not the result of arbitrary cost-cutting decisions.

Daniels and Sabin hope that patients and clinicians will judge such a transparent and publicly accountable process to be fair and acceptable. However, they acknowledge that a decision-making process that is accountable for reasonableness—i.e., meets their four conditions—does not supplant the need for broader public democratic deliberation about the overall goals and objectives of public health care. Instead, decisions resulting from such a process should become part of larger democratic public deliberation. In the following section, we apply the concepts of equality of opportunity and accountability for reasonableness to the case of BRCA testing in Canada's national health insurance system and explore the types of information, reasons, and principles that are needed to evaluate whether this service should be covered by public health care insurance.

RATIONALES FOR COVERAGE OF BRCA TESTING

Prevalence & Test Accuracy

In 2003, an estimated 21,200 Canadian women were diagnosed with breast cancer—a cumulative lifetime risk of 1 in 9—with 5,300 women predicted to die from the disease (Canadian Cancer Society 2003). Of these women, only 5 to 10 percent are likely to have inherited an allele associated with increased risk of developing the disease (Szabo and King 1997; Carter 2001; Narod 2002). Mutations in the BRCA1 and BRCA2 genes have been strongly associated with hereditary breast cancer, conveying a cumulative lifetime risk of 40 to 85 percent for breast cancer, and 16 to 40 percent for ovarian cancer, depending on the mutation and one's family history (Carter 2001).[7] But even for very accurate testing methods, only 20 to 25 percent of patients with a strong family history—e.g., early age of onset or multiple affected family members—will have a positive BRCA mutation; a 2002 study by researchers at Myriad Genetics detected BRCA mutations in only 17.2 percent of 10,000 individuals (of whom 5,503 indicated a personal history of breast or ovarian cancer) analyzed over a 3-year period (Frank et al. 2002).

In other words, for 75 to 80 percent of breast cancer patients, the heritable component of their cancers remains unknown. There are almost certainly other, yet to be discovered, genes that affect breast cancer risk in

families negative for BRCA mutations. Some of these may be high penetrance genes that confer significantly increased risk, such as a putative BRCA3 gene (Thompson et al. 2002), and others will be low-penetrance and either confer moderately increased risk such as CHEK2 (CHEK2-Breast Cancer Consortium 2002) or effect tumor growth such as P53 (Rahko et al. 2003). Social and environmental factors clearly also influence the risk of developing breast cancer (Narod 2002). Interpretation of test results, thus, is closely tied to a detailed evaluation of the patient's family history, so BRCA testing is not a useful screening test for the general population. Most people without a family history of cancer will not have any detectable BRCA mutations. The test will provide clinically relevant information only to those people with a strong family history of breast cancer and where the family mutation is found (in an index case) and can then be used to determine occurrence of the mutation in other family members. However, for most people with a strong family history of the disease, no family mutation will be identified (the heritable component remains unknown) and thus the tests results will be "uninformative"—these people remain at high risk based on their family histories.

Benefits & Costs

Patients with a family history of hereditary breast cancer live with the anxiety of being at risk, as well as the objective risk of developing the disease. They may have to undergo regular high-risk screening, care for affected family members, and deal with the personal trauma of early disease onset as well as the death of family members from the disease. Living with this condition in one's family may seriously compromise an individual's (and family members') equality of opportunity. Studies of families at risk for hereditary cancers, such as breast, ovarian, or colorectal cancers, show that even in the absence of cures, access to genetic testing information can be extremely important for a variety of psychosocial reasons (Prospero et al. 2001; Hutson 2003). Genetic information may be used to facilitate life planning, to initiate family discussions of issues such as social and psychological support, guilt, and responsibility for other family members (Burgess and d'Agincourt-Canning 2002), or to help people make changes in career plans.

A strong family history by itself should be sufficient to initiate regular monitoring—as well as counseling and other support services—as part of high risk cancer screening clinics. A positive result from BRCA testing—i.e., a result that indicates a deleterious mutation—will confirm a patient's

putative high-risk status and may convince some physicians to monitor patients with a family history more aggressively (d'Agincourt-Canning 2003). This, in turn, may facilitate access to other health care services that can significantly reduce a patient's risk of developing the disease, such as preventative drug therapies or prophylactic measures (Lynch, Lynch, and Rubinstein 2001; Hartmann et al. 2001; King et al. 2001), and have a positive effect on risk-reduction behavior and earlier diagnosis of tumors (Scheuer et al. 2002). Although some patients who receive negative test results are not reassured, those patients found not to have the identified family mutation are considered to be no longer at high risk—i.e., they have the same background risk as the general population—and thus can avoid frequent, expensive, and unpleasant monitoring. BRCA testing thus can be an important means of helping at-risk families to have more normal lives and access to a fair range of opportunities.

However, although genetic testing for hereditary breast cancer appears to provide tangible clinical results, it also can have negative physical, social, and psychological sequelae. Drug treatments or prophylactic surgery are not cures and may be difficult to apply given insufficient knowledge concerning the optimum time to undergo surgery or the desirability or length of time for using tamoxifen in particular populations—e.g., childless women or women with early/late menarche (Narod 2002). Positive test results may lead to fatalistic attitudes about developing cancer or increase fear and anxiety. Genetic information presents patients with potentially difficult choices about whether and how to discuss their results with other family members and whether they should be advocates for promoting testing in the family (Hallowell et al. 2003). In the case of testing for specific mutations associated with increased prevalence in certain communities—e.g., the three "Ashkenazi Jewish" mutations—an individual choice may contribute to stigmatization of and discrimination against the larger community (Evans, Skrzynia, and Burke 2001; Koenig et al. 1998).

In addition to these more personal negative sequelae, the commercial nature of BRCA testing has cost implications for the public health care system (Sevilla et al. 2003). Patenting of the BRCA genes and the associated susceptibility test resulted in a tripling of the cost of service provision—C$1,200 for in-house testing compared with C$3,850 for testing purchased through Myriad or one of their licensees (Kent 2001a; Eggertson 2002). As previously discussed, this cost is simply unaffordable for many health care services and could result in termination of service provision,

as was the case for two years in B.C. Provinces, states, or nations that defy the Myriad patent and continue to provide in-house testing would avoid this cost increase but be faced with the legal costs associated with potential patent infringement suits and the payment of damages if they lose the case.

In fact, Myriad has not launched any patent infringement suits and actually is facing legal opposition itself, with a number of French laboratories challenging the legitimacy of the European patents (Benowitz 2002; Institut Curie 2001). Myriad's control of the BRCA testing market has been weakened further by the recent award to Cancer Research UK of the European Union patent for BRCA2, which will be licensed free of charge to public laboratories engaged in research and not-for-profit clinical genetic testing. Canadian provinces such as Ontario are openly defiant and daring Myriad to take them to court (Lindgren 2003; Eggertson 2002). The lengthy and costly nature of defending one's patent rights often means that the "winner" will be the one with the deepest pockets—and a nation or province will have much deeper pockets than a biotech company such as Myriad (Williams-Jones 2002b).

The availability of BRCA testing for private purchase introduces other long-term social costs. People may seek genetic testing even when medical professionals do not deem them to be at sufficient risk, because the information is valued for "non-clinical" uses, such as anxiety reduction or initiation of family dialogues (Burgess and Hayden 1996; Cox and McKellin 1999). Some patients also may opt for private purchase when the service is not provided through the public health care system. The reasons for using the technology and the way it is provided have begun to "drift" (Williams-Jones and Graham 2003), and these reasons may be manipulated by direct-to-consumer (DTC) advertising. Myriad is marketing its *BRACAnalysis* test both to physicians and to the general public through TV, print media, and the Internet (Myriad Genetics 2002). Critics argue that this advertising campaign exploits a climate of genetic determinism and public anxiety in an effort to convince members of the general public that they need and should either purchase or demand from their physicians an expensive genetic test that is unlikely to be clinically useful (Moreno 2002; Krasner 2003; Gollust, Hull, and Wilfond 2002).

Although DTC advertising is not permitted in Canada, the globalization of media (TV, radio, Internet) means that Canadians can access advertising about Myriad's genetic test. Since Myriad has an interest in selling its services to as large a market as possible, it should not be surprising

that the company's access criteria—i.e., that a physician determine that his/her patient would benefit from testing, e.g., has a single family member with breast cancer—are less restrictive than those in the public health care system (Birmingham 1997; Smith 1997). When patients need only convince their physicians that they should have the test—after all they are paying out-of-pocket—the public health care system loses its former ability to constrain utilization and to ensure that tests are only made available to those people for whom the tests will provide accurate and useful information (Carter 2001; Holtzman and Shapiro 1998). Direct purchase could increase costs to the health care system, as more people would require genetic counseling (usually to be told that they are not at risk), a service not included in the price of direct purchase. In addition, those few people found to have positive test results then become eligible for other health care services and monitoring, costs that are legitimately covered by public health care insurance.

The benefits and costs of BRCA testing present a complex picture of intangible benefits and difficult-to-assess risks and costs that seem less reasonable to assume when the cost of testing is tripled (Col 2003). The need to absorb the cost increase within a small, multi-focused hereditary cancer program budget led to an evaluation of "lost-opportunity" costs. The creation of two classes of patients—those who could pay for direct access and those who could not—highlighted the unfairness to the at-risk population, independent of the justification of the mezzo-allocation decision. Discrepancies between provincial programs highlighted different attitudes toward legal risks and support for IP protection, although the reasoning behind the different provincial policies can be assessed only speculatively. Given that Myriad's BRCA test is only the first of several genetic tests to receive strong patent protection, the associated advertising and development of a direct purchase market raise systemic issues for a public health care system: What are the goals of the system, how well are those goals served by patent protection, how should decisions be made about what to include in health insurance, and should direct purchase be restricted?

PUBLICLY ACCOUNTABLE PROCESSES

There is substantial public pessimism about the way government representatives and policy makers are involved in decision-making processes, as well as the influences that shape their decisions. The *ad hoc* nature and opacity of the decision making is not conducive to public trust or sup-

port—decisions are made in a "black box" and usually only the results are available for public inspection. For Daniels and Sabin, rational decisions about health care resource allocation, even if based on "reasonable" evidence or principles, will be insufficient if the decisions rendered *and* their rationales are not also publicized. Transparency—which Daniels and Sabin call condition 1: Publicity—is essential for public accountability.

In the case of access to BRCA testing in British Columbia, the B.C. Ministry of Health Services did not publicize its reasoning for complying with the Myriad patents in face of opposition to those patents by other provinces. The Ministry simply told the B.C. Cancer Agency and the Hereditary Cancer Program to comply with the patent and purchase testing from Myriad. HCP, however, did publicize its decision to terminate the provision of BRCA testing. Letters were sent to all patients enrolled in the program—and a note was posted on the B.C. Cancer Agency website—explaining that, due to the Ministry, HCP could no longer afford to provide BRCA testing to its patients because purchasing testing from Myriad would triple the cost of the services and undermine the program's ability to provide other services to patients with a diversity of hereditary cancers. HCP would continue to offer counseling support for patients and facilitate referrals to Myriad or MDS should patients wish to purchase testing themselves (Coldman 2001).

The lack of transparency on the part of the B.C. Ministry of Health Services in their decision to comply with the Myriad patents, and in the reversal of this decision two years later (British Columbia Ministry of Health Services 2003), makes an evaluation of the underlying rationales impossible. Without disclosure of the reasons for the decisions (transparency), it is impossible to evaluate whether they were based on a rational and careful consideration of the threat of a patent infringement suit from Myriad, the costs of a law suit balanced against the benefits of continued service provision, or the costs of increased utilization and lack of control that would result from forcing the service into the private sector (considerations of relevance). Loss of trust is a predictable consequence of making decisions that lead to a loss of benefits without providing justification. As has been demonstrated in Europe with respect to genetically modified foods, backroom decision making can have a significant negative effect on public trust (Millstone 2000). At a minimum, the provincial authorities could have provided their reasoning in the letter to HCP so that it could be quoted in the letters HCP sent to patients and families.

Transparency and relevance, however, are insufficient for public accountability (or accountability for reasonableness). Daniels and Sabin also require mechanisms for appeal, dispute resolution, as well as ongoing review and revision of decisions. This condition does not require public input into the initial decision—a decision-making process is fair and accountable for reasonableness if it is open to challenges and disputes of particular funding arrangements. Openness ensures the opportunity to revise decisions in light of new evidence and to be responsive to changing social and political realities of health care and technology development. Once again, in the absence of transparency, and the consequent inability to assess the relevance of the reasoning, it is difficult to determine the extent to which the B.C. Ministry of Health Services' policy reversal was the result of appeals. In any event, the official route for appeals to the Ministry is not well publicized, which leads to another discrepancy in that only well-informed individuals know how to register their concerns.

Enforcement is the final condition of accountability for reasonableness. Although public administrators and elected officials claim that the "public" interest is their primary focus, only a decision that is transparent, relevant, and open to appeal meets the conditions for accountability for reasonableness. The final condition of enforcement requires public regulation to ensure that these other conditions are met. Although the decisions in B.C. related to the support of the BRCA patents and subsequent restriction of in-house testing were made by public officials, there was no mechanism to hold them accountable to the first three conditions.

PUBLIC CONSULTATION

Daniels and Sabin see an accountability for reasonableness approach to decision making as consistent with democratic deliberation, but they do not endorse active public participation in specific decision-making processes, apart from the requisite appeals mechanism. Direct patient or consumer involvement is unnecessary because:

> . . . the conditions we advocate would by themselves establish when decisions are reasonable in the relevant sense. Consumer participation might improve deliberation about some matters, but it is unlikely that we could ever enlist active enough consumer participation to deliberation about limit setting. . . . Simply being accountable to a "board" containing consumer representatives would not ensure that the right sort of deliberation took place at appropriate levels in the plan. In addition, there is no realistic

> mechanism for making consumers who participate truly representative of the consumer population as a whole. (Daniels and Sabin 1998, p. 61)

The power of decision making, for Daniels and Sabin, is best placed in the hands of those people with the requisite policy, scientific, and medical expertise, as many of the issues under debate will be simply beyond the capacity or interest of most citizens (Lomas 1997). Technology assessment and evaluations of the accuracy, effectiveness, and usefulness of a particular health care service tend to be relatively technocratic—it is largely scientists, physicians, and policy analysts who gather relevant information and determine which services or technologies are safe and useful, and for which populations. In general, society benefits from having scientific and medical professionals make these evaluations and regulatory structures that control access to medical services and technologies that could be hazardous if used inappropriately—e.g., prescription access to certain pharmaceuticals.

Daniels and Sabin correctly note that representativeness is a significant problem for a more ambitious form of public consultation. But some public participation would improve representation, and some methods or combination of methods for participation are better than others. The objective of public consultation might be either the strong position that citizens should be involved directly in making actual policy decisions—e.g., through citizen advisories, citizen juries, or forms of deliberative polling—or a more qualified notion of trying to improve the range of perspectives brought to bear on issues.

It may be that the place for more active public involvement is within broader deliberative democratic processes. Citizen participation in democratic society should involve more than simply the election of government representatives; it should include deliberation about the general principles and values of a society. Without citizen participation and consultation in these areas, elected officials and bureaucrats are left to their own understandings, and are overly dependent on the media and lobby groups to translate the diverse perspectives and concerns of the population. For example, public deliberation about health care reform or resource allocation decisions informs policymakers about what citizens consider to be core services and are willing to pay for with their tax dollars. The hope is that public consultation on questions of policy development will result in polices that integrate a full range of values and concerns and garner widespread acceptance (Knoppers 2000), as well as constructively direct subsequent critique. Public consultation on these broader values, combined

with Daniels's and Sabin's accountability for reasonableness related to particular decisions, will improve public accountability for decisions on complex issues such as intellectual property protection and health care funding. As Ezekiel Emanuel (2000, p. 10) argues, members of managed care organizations (and by extension health insurance plans) should be ". . . given the opportunity to consent to the allocation of health care resources that will affect them," because "justice requires that those who have to live with the consequences of the allocation be afforded the opportunity to affirm that the allocation reflects their values."

In modern liberal democracies, there is clearly a place for elected regional and national government representatives to make decisions on behalf of the citizenry about law, policy, or regulations. The contemporary drive toward more inclusive processes usually is justified on the basis of increased sociocultural pluralism, a sense of powerlessness, and concern about the instrumental approach of expert advice neglecting other social values (Bloomfield et al. 1998). Daniels's and Sabin's four conditions fail to be adequately representative, and we maintain that a process that is insufficiently representative, even when transparent and reasoned, fails to be "accountable" to all the relevant perspectives. Accountability usually implies that the decision makers are held responsible for how they made their decisions and for the outcomes of the policy. Broader public consultations to identify the range of interests that are relevant to diverse populations is an essential element of decision making, no matter who the analysts are.

CONCLUSION

Adult genetic testing will have to meet high standards to be included among funded Canadian health care services when compared to other beneficial services such as hospital care, wheelchairs, or pharmaceuticals (Caulfield et al. 2001). The case of BRCA testing in Canada is useful for illustrating the difficulty of integrating technical or practical considerations with more theoretical discussions. We have not argued for or against the public provision of BRCA testing in light of the Myriad patents, but instead have sought to elucidate the relevant factors needed to make such a decision. Determining whether access to BRCA testing delivers sufficiently beneficial consequences and enhances equality of opportunity enough to merit public funding can be accomplished only with active participation from those stakeholders most directly involved, that is, patients, families, clinicians, and support groups. Such stakeholder and citizen involvement

nevertheless needs to involve more than the simple collection and integration of public opinion. The role and appropriate methods of public consultation must be considered carefully—the field of environmental policy and public consultation has an informative history and literature (cf. Renn, Webler, and Wiedemann 1995). Public input needs to build on the scientific and technical evaluations—e.g., is the technology safe, useful, minimally harmful, supportive of normal functioning, or cost-effective relative to other options?—but it also must move beyond a strictly technocratic view and integrate personal experiences of illness to better nuance and expand the basic medical definitions of benefit and utility (Wertz and Gregg 2000; Secretary's Advisory Committee on Genetic Testing 2000). As reflected in the literature on risk perception, "there is no such thing as an objective characterization of risk" (McDaniels 1998, p. 131), and identifying which risks and benefits are important requires public involvement.

It also is essential to analyze the larger social and political context in which the test was developed and marketed, and how it shapes and is shaped by patients, consumers, researchers, clinicians, government policy, and the commercial biotechnology industry, to name only a few of the relevant factors (Ontario Ministry of Health and Long Term Care 2002). There will be cost implications of any policy for the system as a whole—e.g., counseling for patients who have purchased testing privately, and diagnostic testing and monitoring of those who test positive for a mutation—that must also be taken into account (Col 2003; Sevilla et al. 2003). The Myriad case is but one example of a growing number of tests for genetic susceptibility that are coming under commercial control—see, e.g., the difficulties associated with restrictive licensing for testing for hereditary hemochromatosis (Merz et al. 2002). Assuming that the Canadian patents on the BRCA1 and BRCA2 genes stand (there may yet be challenges to the patents from provincial governments such as Ontario), a host of other patents on susceptibility genes are likely to follow that make it more difficult to justify on cost-benefit or cost-effectiveness analyses the public provision of genetic testing services. Economic policy that encourages innovation through strong patent protection increases costs of health care services and proliferates new services. Without sufficient understanding of the implications of patents and commercial genetic testing on costs and access to health care services, health policy related to access will be piecemeal.

There is no easy way to make decisions about what services to fund with a limited health care budget. At a minimum, a more systematic analysis is to be desired over the current *ad hoc* and manipulated decision-making environment. Large-scale public consultations are important to establish the range of interests public health insurance should serve, and at what costs. This is an important step toward representation and establishing accountability. In addition to scientific and clinical expertise, there is an important place for clear reasoning about substantive principles of justice such as equality of opportunity. Finally, employing procedural justice mechanisms to support decision-making processes that are accountable for reasonableness is not only good politics, it is a strong requirement of justice.

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NOTES

1. The protection of strong intellectual property rights (IPR) in new technologies increasingly is seen as fundamental to continued technological and economic growth. Without strong IPR, it is maintained, companies would not be able to recoup the costs of (or make a profit from) investments in research, thereby undermining the development of new and beneficial biotechnologies. Nevertheless, there is also substantial debate about whether patenting of genes and biological materials is ethically acceptable (see, e.g., McGee 1998; Caplan 1998; Eisenberg 2002). The ethical permissibility of human gene patents is beyond the scope of this paper, and we focus instead on analyzing the implications of gene patents for access to health care services. For a broader discussion, see (Nuffield Council on Bioethics 2002; Royal Society 2003).

2. Some advocates for universal health care insurance will make a stronger claim for equal access based on social solidarity (Bergmark 2000; Houtepen and ter Meulen 2000). Personal wealth should not permit some people to obtain better or preferential treatment, and if not all useful health care services can be made available to all citizens then only those services that can be made available to all should be permitted. Other commentators use a more nuanced—e.g., relational—view of justice that goes beyond mere distribution to include consideration of the broader costs of providing particular services, such as lost opportunity costs in not being able to fund other services (Sherwin 2001). Nevertheless, one should bear in mind that health care services are not as important to health, or at least population health, as broader social changes that address issues such as income disparity, employment and job stress, or diet (Evans, Barer, and Marmor 1994; Kaplan et al. 1996; Mechanic 1999).
3. During the last decade, federal government fiscal constraints have resulted in a steady reduction in transfer payments from the federal government to the provinces (Flood 1999), although this has to some extent been reversed by new federal funding initiatives (Department of Finance (Canada) 2003). The reduction in funding has significantly compromised the power of the federal government to constrain the privatization of health care. Provinces are less dependent on transfer payments that constitute ever smaller portions of provincial health care budgets (which are nevertheless growing steadily), and there is less money for the federal government to withhold as punishment. The result is a noticeable growth in patient-paid access to health care services across the country, such as private MRIs, PET scans, and laser eye clinics in British Columbia, Ontario and Québec (see Kent 2001b; Pinker 2000).
4. Some financial support from provincial governments is provided to subsidize the costs of obtaining prescription drugs—e.g., PharmaCare in British Columbia covers costs for anyone spending more than 3 percent of his/her income, with full coverage for the indigent and a 2 percent deductible for the elderly.
5. This is a controversial assertion, since one might argue that reorganization and better management of service delivery—e.g., discarding services with no proven benefit and eliminating expensive pharmaceuticals that are no more effective than cheaper competitors—might enable the public funding of all services of proven benefit. However, if medical technological and pharmaceutical development continues unabated and these products prove effective (but also costly), it will not be feasible to fund them all. This issue requires

substantial evidence-based research, and the economic limits to adoption of all efficacious health care may be inevitable.

6. Although it is clearly neither "objective" nor sufficient, the species-norm notion of health is important and useful if carefully used (Buchanan et al. 2000; Lewens 2003).
7. Much of the research on the incidence of breast cancer and penetrance of the BRCA mutations is drawn from studies of large families with many affected individuals. There is some evidence that risk figures may overestimate and not accurately reflect the levels of risk in families with less extreme incidences of cancer or in the general population (Robson 2002; King, Marks, and Mandell 2003).

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