Genetic Susceptibility Testing for Beryllium: Worker Knowledge, Beliefs, and Attitudes

Ken Silver, DSc,[1]* Gary Kukulka, PhD,[2] John Gorniewicz, BA,[2] Kathleen Rayman, PhD,[3] and Richard Sharp, PhD[4]

Background *We sought to gain insight into workers' knowledge, beliefs, and attitudes on the subject of testing for genetic susceptibility to beryllium.*
Methods *Five focus groups were held with 30 current and former beryllium workers and nine family members. Audio recordings were transcribed and assessed by hierarchical coding using an inductive approach.*
Results *Some workers were unclear about the distinction between genotoxicity and heritability. A key finding is that they perceived the benefits of a positive test result to be related to enhanced autonomous decision-making. The major concern cited by participants was potential abuse of genetic information by employers. Complete financial separation of a prospective testing entity from the employer was seen as crucial.*
Conclusions *A window of opportunity exists to create regional partnerships for translational research on genetic susceptibility testing. Such partnerships would involve labor, management, public health scientists, primary care professionals, and other stakeholders. They would be critical to identifying testing strategies that maximize worker autonomy along with the public health advantages of genetic testing.* Am. J. Ind. Med. 54:521–532, 2011. © *2011 Wiley-Liss, Inc.*

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INTRODUCTION

Genetic analysis in epidemiologic studies of worker populations has revived the prospect of testing for susceptibility to illnesses caused by workplace toxicants, long a topic of debate in occupational health policy.

Early controversies focused on employer mandates and issues of worker privacy and other rights [Severo, 1980]. Much debated was the potential for genetic testing to justify the removal of ''susceptibles,'' while undercutting support for traditional industrial hygiene controls intended to make the workplace safe for all [Draper, 1991]. This debate unfolded decades before any genetic tests were available which actually met scientific standards for use in screening [Khoury et al., 1985]. Despite the long period of anticipation, it is unclear whether effective policies will be established in time to prevent disruptive social effects of genetic testing [Samuels, 1996, 1998; Rothstein, 2008].

Beryllium is the first industrial toxicant for which a valid genetic susceptibility test may soon be available [Silver and Sharp, 2006]. Initially, a glutamic acid substitution for lysine or arginine in position 69 of the HLA-DPB1-0201 allele of the major histocompatibility complex

[1]Department of Environmental Health, East Tennessee State University, Johnson City, Tennessee
[2]Department of Family Medicine, East Tennessee State University, Johnson City, Tennessee
[3]Graduate Programs, College of Nursing, East Tennessee State University, Johnson City, Tennessee
[4]Department of Bioethics, The Cleveland Clinic, Cleveland, Ohio
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*Correspondence to: Dr. Ken Silver, DSc, Department of Environmental Health, East Tennessee State University, P.O. Box 70682, Johnson City, TN 37614.
E-mail: silver@etsu.edu

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(''Glu69'') was found to be associated with elevated risk of chronic beryllium disease (CBD) [Saltini, 1995]. This marker has moderate to high sensitivity; it is present in 72–92% of workers with CBD. But it has low to moderate specificity: the marker is present in 30–45% of exposed workers unaffected by beryllium sensitivity (BeS) or CBD.

The gold standard for a screening test is the positive predictive value (PPV), defined as the proportion that will develop the disease among all who test positive [Ashford et al., 1990]. For the Glu69 marker it ranges from about 12% to 43% under reasonable sets of assumptions for disease prevalence, relative risk, and allelic frequency [Weston et al., 2002]. More recently, certain rare alleles have been identified with PPVs ranging up to 100% [Weston, 2005]. These PPVs are, in fact, estimates derived from a compelling mechanistic model which integrates molecular epidemiologic findings with the biophysical chemistry of beryllium's binding to variant protein products of the HLA-DPB1 gene [Snyder et al., 2008]. While these estimates could change with refinements to the model, the presence of an allele with a PPV of 100% in a beryllium-exposed worker would predict to a high degree of certainty that the worker will develop CBD.

In addition to causing CBD, a non-malignant condition which affects the lungs and other internal organs, beryllium is a recognized lung carcinogen [International Agency for Research on Cancer, 1993]. However, in contrast to the research on CBD, there has been little explicit discussion in the beryllium carcinogenesis literature about mechanistic or epidemiologic evidence of an association with genetic markers of susceptibility [Deubner and Roth, 2009].

The emerging markers of genetic predisposition must be distinguished from the beryllium lymphocyte proliferation test (BeLPT) which for the last decade has been widely administered to nuclear and weapons site workers to detect sensitization for the purposes of compensation [U.S. Department of Labor, 2005] and worker protection [U.S. Department of Energy, 2006]. The BeLPT is not a genetic test; it measures acquired sensitization to beryllium.

This investigation sought to gain insight into workers' knowledge, beliefs, and attitudes on the subject of genetic susceptibility testing for beryllium. The aim was to clarify the consequences of genetic testing and how it is understood and considered by employees and their family members at Department of Energy (DOE) facilities. In a break from the older—and quite voluminous—literature on workplace genetic testing, this study does not rely on surrogates such as labor-oriented analysts, advocates, academicians, or agencies to assess workers' interests. To our knowledge, it is the first formal academic study of this topic in which the results were educed from qualitative data in the voices and vernacular of workers for whom genetic susceptibility testing is a proximate reality.

METHODS

A qualitative inductive approach is well-suited for collecting and interpreting data grounded in the reality of subjects' lives, free of prior theoretical constraints. Here, focus groups [Morgan, 1993; Sandelowski, 2000; Xu et al., 2006; Alanen et al., 2009] were used to investigate beryllium workers' and their family members' understanding and judgments about genetic susceptibility testing.

Five focus groups were held with 30 current and former workers and nine of their family members between June 2007 and April 2008. The principal investigator (K.S.) recruited focus group participants from Los Alamos National Laboratory (LANL) and Sandia National Laboratory (SNL) (New Mexico) and Oak Ridge National Laboratory (ORNL) and the Y-12 National Security Complex (Tennessee) through union representatives, worker advocacy organizations, and a support group. Credentialed in environmental and occupational health with over a decade's involvement in public studies in the DOE complex, the principal investigator was granted time on the agendas of the union and advocacy organizations to describe the study and distribute recruitment packets. Leaders of the support group distributed recruitment packets and asked interested individuals to participate. Potential participants were current or former workers who were exposed to beryllium in the workplace, diagnosed with CBD or tested positive for BeS, or their first degree relatives. Individuals were included in the focus groups according to the order in which they contacted the principal investigator in advance, per instructions in the recruitment materials. No effort was made to select individuals on the basis of specific affiliations or previously expressed views. The resulting ''snowball'' sample was thus appropriate for the aims of the study.

A standardized procedure was used to kick-off each focus group. Light refreshments were available as participants arrived. They were welcomed by the facility host and principal investigator. Following a formal greeting and acknowledgments, the study and the informed consent process were explained; written consent forms were signed and collected. A sociodemographic questionnaire was also completed by participants. To maintain privacy, unique numbers were assigned to link each participant's questionnaire with their verbatim comments on the focus group transcripts. Participants were compensated with $150 stipends for time and travel expenses. A complimentary lunch was also provided. The principal investigator kept field notes to track his thinking and impressions as the study proceeded.

The guiding questions used by the facilitator are shown in Table I. The facilitator (G.K.) was a PhD health educator with over 20 years' experience in medical, nursing, and public health researcher who, incidentally, had worked in factories before attending college.

Probing questions were used to clarify responses (Table I). With the consent of participants the facilitator made audio recordings while the principal investigator functioned only as a non-participant observer, documenting group dynamics and proceedings. Because an aim of the study was to ascertain participants' prior knowledge of beryllium, the research team did not disseminate educational literature or present information about genetic testing in advance of the focus groups. Consistent with the study's aim of drawing out a baseline of workers' knowledge, beliefs, and attitudes, the facilitator did not pose detailed scenarios to study worker decision-making under specific policy options.

Focus groups lasted approximately 120 min. Following the transcription of each audio recording, full transcripts were reviewed and verified, and all transcripts were coded by the principal investigator and entered into NVivo 7 software. A research team member independently coded the three New Mexico transcripts directly in NVivo 7. Verbatim statements of focus group participants for each concept in the hierarchy were printed and bound into three-ring binders. This facilitated the analysis of responses to the guiding questions and the selection of exemplar remarks quoted in this report. To resolve discrepancies and inconsistencies three of the authors (K.S., G.K., J.G.) iteratively reviewed the text and coding themes. The themes were then developed into a coherent rendering of participants' attitudes and beliefs about genetic susceptibility testing.

A Certificate of Confidentiality (COC) was obtained from the US Centers for Disease Control and Prevention [Earley and Strong, 1995; Lutz et al., 2000] The COC provides researchers with added legal protection to refuse to disclose information on participants that could have adverse consequences for research subjects, such as damage to their financial standing, employability, insurability, or reputation. Obtaining the COC was also intended to add a level of trust that would allow participants to more openly discuss their personal experiences. In addition, all methods and materials were approved by the East Tennessee State University/James H. Quillen Veterans Affairs Medical Center Institutional Review Board (ETSU/VAIRB).

RESULTS

Sociodemographic characteristics for 37 of the 39 participants are shown in Table II. (Two participants declined to complete the questionnaire.) Two-thirds (66%) reported their first exposure to beryllium occurred 15–25 years earlier. Among those who disclosed their sensitivity or disease status, ten indicated either BeS (n = 5) or CBD (n = 5) (Table II). Notably, nearly half of the participants (18/37) provided no response about their BeS and CBD status. One of the participants was both a family member of beryllium workers and a DOE contractor employee, but had no known exposure at work.

The themes, which emerged from the data are listed in Table III and discussed below, with direct quotations from participants in italics.

TABLE I. Guiding and Probing Questions Used by the Focus Group Facilitator

1. Should workplace testing be offered to all workers or just some workers? Should it be optional or mandatory?
Do you know what the genetic test actually entails?
Have you heard much about it prior to us convening this session?
When did you first hear of it?
Have you ever been offered genetic testing?
What did you learn about DNA, genetic testing or susceptibility that you hadn't heard prior to coming today?
2. How would individuals benefit from taking a genetic susceptibility test? What burdens would they face as a result of taking the test?
Does anyone have a sense that it's not a good thing?
What's the disadvantage of having the test? Anything negative to you, your family, the other workers?
We're going to have testing. We've got some policies in place. You're on a committee. We've got the right groups involved. We're feeling trust in whoever is doing the testing.
You're going to feel safe, secure. What's the benefit for you or your family member getting tested? Are there any benefits?
3. What factors would increase or decrease your likelihood of pursuing testing?
Does genetic testing make sense to you?
If testing began at your workplace what would be your concerns?
Do you trust your employer to do it?
Who should do this program? What would it look like? What are your recommendations for policies?
4. How would you respond to a positive test result? To a negative test result?

TABLE II. Sociodemographic Characteristics of Focus Group Participants

Characteristics	Percent (n = 37)
Age	
25–44	5.4 (2)
45–90	91.9 (34)
No answer	2.7 (1)
Gender	
Male	78.3 (29)
Female	21.6 (8)
Race ethnicity	
Caucasian	48.6 (18)
Hispanic	40.5 (15)
African American	10.8 (4)
Education	
High school diploma	78.3 (29)
College or higher	21.6 (8)
Diagnosis	
BeS	13.5 (5)
CBD	13.5 (5)
Neither	24.3 (9)
No answer	48.6 (18)
Years since first exposure (workers only)	
5–14	34.5 (10)
15–24	24.1 (7)
25+	41.3 (12)
Workers and family members	
Beryllium workers	30
First degree family members	9

Knowledge and Understanding of Genetic Testing

A common understanding of exposures and operations emerged between focus groups in New Mexico and Tennessee. Despite notable geographical and cultural differences, the DOE facilities included in this study are among the largest employers located in rural states which afford limited alternative job opportunities. Still, a striking difference emerged between Los Alamos, New Mexico, and Y-12/ORNL in Tennessee in the resources locally available for workers to become knowledgeable about beryllium. In Tennessee, it was clear that workers engaged in a beryllium support group had gained sophisticated insights and information about BeS and genetic testing, which they have discussed and disseminated to their peers. For example, an active participant in the support group explained:

> You can be exposed just one little bitty time and then be super susceptible to it. They show

TABLE III. Hierarchy of Key Coding Themes and Sub-Themes Established From Focus Group Interviews and Transcripts

Key theme or topic	Sub-theme or topics
Understanding of basics beryllium issues	Source of information
	BeS-CBD connection
	Inter-individual variability
	CBD prognosis
	Medical removal protection benefits
Data handling	Court-ordered disclosure
	Data privacy
	Inclusion in medical record
	Proprietary interest in DNA
Clinical encounters	DOE former worker program
	DOL surveillance
	Local medical community
	Medical surveillance employer
Risk numbers	For laypersons
	For scientific authorities
	PPV
Gene testing program	Accuracy of testing
	Conflict of interest
	Cost of testing and sponsorship
	Interpretation of test results
	Manda-voluntariness of testing
	Testing venue
	Timing of test pre-/post-employment
	Trust
Implications of testing	Generic abuse by employers
	Alternative employment
	The common good
Benefits of testing	Autonomy
	Health decisions
	Variability in co-workers explained
Burdens of testing	Discrimination
	Exposure controls de-emphasized
	Insurability
	Long-term unemployment
	Superman syndrome
	Removal reassignment based on test
	Retiree perspective
	Retrospective remorse
	Self-non-disclosure of test results
Social Context	Company town
	Environmental contamination
	Ethnic communities
	Gossip
	National security secrecy
	Paycheck vulnerability
	Restricted duty

> symptoms within three months. And then other people . . . I've been around it for 27 years. And I might not show it for another 15 or 20 years, if I ever do. So that's one thing about genetics. If they get it [genetic testing] right, they can tell who would be super susceptible to it.

At LANL, where there is no support group, participants' knowledge was rooted in personal experiences with health outcomes and exposure scenarios. The LANL focus groups' level of knowledge appeared to be comparable to that of healthy, active workers who are beryllium-exposed but have not yet been called upon to make presentations to their peers on the subject, as some of the Tennessee support group members had done, nor studied the fact sheets provided at medical screenings. Demand for more information and knowledge in New Mexico became clear as participants posed a litany of substantive questions to the principal investigator at the beginning of one of the focus groups. Responses to these questions were only provided after the focus groups.

Some questions raised in the New Mexico focus groups, in the workers' own vernacular, are the same fundamental questions that lie at the core of molecular epidemiologic studies underway at research institutes and universities. *If there is a group of people working together with beryllium only maybe four are sensitized to it . . . So is there an explanation for that?*

Some misconceptions exemplify the educational challenges, which would confront a genetic testing program. The methods by which the genetic test is performed were not immediately obvious: *We don't know if they take blood, or your blood pressure. Or if they take a shot in your eye. Your urine?* Another critical area is the distinction between heritability and genotoxicity. Several participants expressed the mistaken belief that a ''genetic test'' measures genotoxicity to subsequent generations: *I'm thinking of a child that hasn't been born yet.* And*: If it alters your genetic material, that would alter your future generations.* Yet at least one other New Mexico participant did not share this misconception. He understood: *My children have the same genes as I do. So they're probably going to be sensitive* [sic] *to it if I was*.

Benefits and Burdens of Testing

Key to evaluating workers' beliefs and attitudes toward the benefits of genetic testing is the distinction between negative and positive test results. A negative test result would *ease the mind* and give individuals the *satisfaction of knowing* they lack the susceptibility marker. But, overwhelmingly, the perceived benefits of a positive test result were related to the principle of autonomy. Former workers who are beryllium sensitized expressed remorse that a genetic susceptibility test was not available to them when they began their careers. They would use the knowledge of their own susceptibility to steer their children away from careers with beryllium exposure.

We wanted to learn whether workers would value the added information provided by a genetic susceptibility test. For the sake of simplicity, we asked focus group participants to assume that in the absence of a genetic test the risk of disease is about 5% in their population of similarly exposed co-workers [Welch et al., 2004]. Does a positive genetic test which revises the risk number to 14% provide valuable information? Does it tell you something you don't already know?

Only one participant accepted the premise of the question and provided an unambiguous answer: *If right now 5% of the people could be susceptible to it and they did the genetic studies and found out that it's a lot more, it goes up to 15%, double or triple . . . That's so important*. Interestingly, this union member went on to refocus on environmental controls: *And what's even more important is how the control levels change after they find out the increase*.

Several other participants rejected the question's premise of a 5% risk of disease. Some placed the current known risk at the level of 25–50%, based on their life experience. A local leader on beryllium issues seemed to reject the underlying assumption that risks should be discussed in quantitative terms: *One percent is too much. If you are in the 1% group, adios*.

The major burdens cited by participants concerned the potential abuse of genetic information by employers. Passage of the Genetic Information Nondiscrimination Act (GINA), which prohibits employers from collecting or using genetic information, did not occur until the data-gathering phase of this project was nearly complete in April 2008. DOE workers will be skeptical about their employers' likelihood of complying with GINA regulations [Equal Employment Opportunity Commission, 2009]. Not even a hypothetical voluntary program with up-front promises of confidentiality was sufficient to allay privacy concerns fully.

> They do something beyond the law. So what? ''We get caught. We get a $5,000 fine. We get smacked. We promise not to do it again.'' But hell they're going to do it again anyway.

The overwhelming majority of comments concerned the potential for adverse job removal actions. Yet one retired construction worker had a more sanguine view of the employer's willingness and ability to provide alternative employment for workers who seek to be removed from beryllium jobs on the basis of a genetic susceptibility test: *They've got jobs all over the place. So if you can't fit*

into beryllium, they've always got a job for you. A measure of credence can be imputed to this statement by virtue of the fact that none of the focus groups revealed instances of frank discrimination against LPT-positive individuals.

Participants in New Mexico and Tennessee described DOE's ''Human Reliability Program,'' whereby employees in jobs considered of ''high risk'' to national security consent to give their employer far-reaching access to their personal information, including medical and prescription drug records as well as banking and financial data [U.S. Department of Energy, 2008]. It was felt that some enrollees would feel compelled to construe a genetic test result—even one obtained voluntarily with assurances of confidentiality—to be subjected to the self-reporting ethos of the program. An aura of inevitability surrounded the employer's ability to find out a worker's susceptibility status: *As long as you work for the government . . . they're going to find out*, an enrollee said.

Effects on co-workers were also cited. A union official asserted: *You can have "Superman Syndrome*," whereby a worker who is negative for the genetic marker becomes careless in observing measures to reduce exposure to beryllium, endangering co-workers. Inversely, voluntary self-removal from jobs with beryllium exposure, based on a positive test result, raised a two-fold problem. First, *Everybody knows . . . because he's not working with beryllium. So how do you really protect that person's privacy*? Second, drawing upon prior experience with light duty restrictions for physical injuries, a union officer predicted: *Well a lot of the other co-workers will give him s—t*. That is, policies for temporarily accommodating workers with physical limitations are met with resistance by construction workers. Machismo attitudes were cited.

Concern about exploitation of biological materials was expressed strongly in a focus group in Española, New Mexico, perhaps owing to the decades-long influence of organizations concerned with protecting the collective genetic resources of indigenous communities [Dobson and Williamson, 1999; Tsosie, 2007]. A worker commented on his actual experience with the informed consent process carried out by a university's researchers during periodic medical surveillance exams: *They take blood—like four, five, six vials of blood. And you never know what they did with it or anything*.

Perspectives on Test Administration: Voluntariness, Sponsorship, and Venue

The five focus groups in this study took place before passage of GINA. A question elicited workers' views on whether genetic testing should be voluntary or mandatory. The few expressing support for mandatory testing would limit it to those with known exposure to airborne beryllium.

The majority of responses were in favor of voluntary testing. Concern for *civil liberties* was cited. A union steward would predicate any offering of the test on participation in a mandatory educational program. *After education you have a choice to take or refuse the test*. A union construction worker suggested that employers pay release time to facilitate worker participation in a voluntary testing program.

The facilitator followed up the guiding question ''What factors would increase or decrease your likelihood of pursuing testing?'' by probing for specifics about who should sponsor a voluntary testing program and where testing should take place. Participants were asked to think of themselves as a ''committee'' charged with establishing policies for a testing program for their work peers.

Overwhelmingly, participants' comments focused on ensuring financial and political independence of the testing entity from the employer (Table IV). Specific suggestions were made for who should lead an outside testing entity. Involvement of representatives of the Centers for Disease Control, watchdog groups, and *an environmentalist . . . someone who can't be bought* were suggested. A hybrid structure, which would involve both labor and management was suggested by a retired construction worker:

> The companies oughta set up a safety board. And put the workers in charge . . . Choose a guy that's a B.A. [union business agent]. And have him in charge. That way the workers can say what they feel and get the information they need.

Avoiding a sole source contracting arrangement by involving more than one testing facility was also proposed. This would reduce the perceived risk of one testing lab, which becomes overly dependent upon the government or DOE contractor for funding, bending to the will of management in making clandestine disclosures of individuals' test results.

At Y-12/ORNL it was suggested that a worker's private physician could draw blood, receive the test results, and help the worker's family interpret them. *I trust my doctor to be honest. I don't trust them [the employer] to be honest at all*. Similar sentiments were voiced in New Mexico, but with two caveats. First, the Los Alamos medical community, where some of the workers' personal physicians practice, was seen as entwined with the employer. Second, in light of past errors in interpreting other clinical test results, personal physicians were not viewed as being equipped to handle all aspects of a beryllium genetic test. Responsibility for an educational program might need to rest with some other entity.

TABLE IV. Burdens of Testing: Potential Abuse by Employers

Exemplar worker statements about employer abuse
Private sector and generic employers
They'll get rid of you
My concern is how business is using things for their benefit. It could be used in an improper way. It's rough times out there, especially in the private sector
It's like a Big Brother thing. The corporations are owned by the insurance companies. That might keep me from getting the test
Contractors at government-owned nuclear facilities
They're ruthless. It's just beyond anyone's wildest dreams
We can have the best DNA test in the world. But if there's no accountability we've got nothing. Nothing
I've worked for the government. I'm very skeptical as to whether it will be done the way it should be done
We don't really trust the government
Maybe I've got Big Brother syndrome . . . because I don't trust anything anybody in the higher echelon [of] government, corporations or what anybody says
[an executive] from Brush-Wellman . . . stated [in 1994] if we could take that small percentage and spring them out, we could eliminate the disease. Eliminate the marketing problem that comes with having a toxic material. And we could begin doing away with some of those very expensive control measures
I don't think anybody . . . would trust the company not to misuse it. Or withhold the findings
It's an ethics issue. And they've been unethical for so long
The younger kids don't realize when they go there to work . . . that Medical is there just to protect the company. It is not there to protect you
Los Alamos is a company town. They all talk to one another when something like this comes up

Family Issues

Two separate focus groups were held for family members of beryllium workers. As noted earlier, some retired workers who are LPT positive would use knowledge of their own genetic susceptibility status to steer a child away from careers associated with beryllium. Some workers implied they would impute knowledge of their own susceptibility status to their children, while others explicitly stated they would like to see their work age children get tested.

Several additional burdens of genetic susceptibility testing were articulated. Often cited was psychological stress and worry likely to be experienced by family members of workers who continue to work around beryllium after receiving a positive test result.

A favorable view of the prospect of a genetic susceptibility test was expressed by a current employee whose father, spouse, and sibling are, respectively, dead from CBD, currently exposed, and LPT-positive: *Considering the history I have in my family, this would be just a perfect fit*. A retired union steward viewed the prospect of a voluntary genetic testing program in the wider context of the role of unions in families and communities: *You've got to plan your future and your needs, your relatives, the people you're around. And you've got to take care of your membership*.

Voluntary Self-Disclosure of Test Results

Family members figured prominently as key figures to whom workers would disclose their test results. But some would refrain from doing so to avoid worried reactions about their safety as a breadwinner. At Y-12/ORNL a key distinction was drawn:

> Having the disease and telling your family is different than having a genetic marker and telling your family. Now I wouldn't tell my family that I had a genetic marker. I would hold that to myself, I think.

One worker would disclose his genetic test result to a child for a different reason: *She's studying to be an* [expert] in a relevant field, and therefore could assist him in critically evaluating the implications of his test result.

With probing beyond the guiding question ''How would you respond to a positive [or negative] test result?'' it was revealed that disclosure to one's personal physician, while favored by many participants, was complicated by the modern reality that workers in health plans or those suffering from chronic illnesses may have two or three doctors. Which one should get the results?

In New Mexico, a greater proportion of workers would disclose a positive test result to their employer than would workers at Y-12/ORNL. One union construction worker, savvy about the internal decision-making structure of his employer's organization, would tell *Somebody higher up than my immediate supervisor that I'm not gonna be working around that [beryllium] no more—and hope they'd leave it at that*. Another would disclose his positive test result to *My fellow worker. Hey my test came*

out positive, dude. So you've probably got it—by way of encouraging his co-worker to avail himself of a voluntary testing program. Yet another participant expected a more guarded response:

> The employment in northern New Mexico is mostly based on the Laboratory. A lot of people would not want to lose their employment—not tell—on the basis of being discriminated against.

Parallels to the LPT and a Diagnosis of CBD

Participants drew frequent comparisons between the vexatious issues of a voluntary genetic testing program with problems already encountered under DOE's beryllium standard when employees have positive LPT results. Total earnings, seniority, and other job rights and benefits must be maintained for 2 years, while the sensitized employee is reassigned to beryllium-free work areas with air concentrations below the action level [U.S. Department of Energy, 1999]. But a participant stated:

> There are several of us here that had to retire because they were still exposing us to beryllium, and still even up to my day of retirement. Even though the company already knew it . . . But they were still exposing me even though they knew that my body's immune system was positive.

In New Mexico, a single case raised the question of what constitutes a work environment without beryllium exposure under the medical removal protection benefit provisions of DOE's standard. A sensitized worker reported:

> See, I'm beryllium sensitized, too. My employer had to know so . . . that I couldn't go into areas like that no more. In turn, I kept my job. Now I work in offices. And I don't go into [beryllium areas] no more.

No terminations of LPT-positive workers at any of the DOE sites were alleged. But according to focus group participants, some younger workers at Y-12/ORNL are eschewing the LPT. They are waiting until they have built up some retirement security before opting to take the LPT, perhaps in their fifties. Their aim is to avoid potential stigmatization and prolonged joblessness during the decades that typically elapse from initial sensitization to the onset of symptoms.

Participants' experiences with the LPT also informed their views on threats posed by a genetic test to individuals' insurability. A union officer reported:

> It becomes a pre-existing issue. And if they were to ever switch employment, it may affect their receiving insurance from their future employer. And that's why, still to this day, a lot of people will not go and take the LPT test that work in beryllium areas.

Among the perceived threats of a positive LPT or genetic test were insurance premiums going *through the ceiling where you couldn't afford it* and *They will block you off*. Even worse added another: *He's got it. Let's cancel his insurance*.

A retiree who has been diagnosed with CBD confirmed this threat:

> One thing that I have experienced after being diagnosed with CBD that could also be applied to genetic markers: I can't get mortgage insurance now because of my condition; our union offered $10,000 free. Life insurance, supplemental insurance. I can't even get that.

DISCUSSION AND CONCLUSIONS

Analysis of the transcripts of the five focus groups conducted in New Mexico and Tennessee reveals what current and former beryllium workers and their family members know about genetic testing and the beliefs they have formulated about its potential benefits and burdens. Our most important finding is that, overwhelmingly, the perceived benefits of genetic testing were related to autonomous decision-making by workers with respect to career choices for themselves and their children. Some would presume that their own test result applies to their children; others explicitly wished testing would be available to their children before entering a trade or facility with beryllium exposure. Of the many issues discussed in the focus groups, it is this desire to gain knowledge for future decision-making that provides the principal basis upon which workers would seek testing.

The free choice of a voluntary program leads to a diversity of outcomes as to whether—and to whom—individuals would voluntarily disclose their test result. The range of responses we report here could be helpful in developing guidelines to be used in a voluntary genetic testing program, laying out the implications of self-disclosing to various actors [Silver and Sharp, 2006].

Elsewhere, among beryllium workers enrolled in NIOSH studies of genetic markers, only a small

percentage (6.2%) requested their individual results [McCanlies et al., 2004]. Workers enrolled in a genetic study at LANL in the late 1990s may have been provided with information about their susceptibility status, along with genetic counseling [Lomax, 2002]. A labor-based program which assessed the attitudes of workers emphasized the importance of administering tests in surroundings and under conditions of the workers' own design. Issues of access to records, reliable information sources, certainty in payments for follow-up care, and holistic involvement by unions were identified [Samuels, 2000].

In our study, as expected, the burdens of testing which provoked the most discussion involved misuse of genetic information by employers. Not even a hypothetical voluntary program, in which test results are disclosed only to the worker, was immune to these concerns. Those enrolled in DOE's Human Reliability Program may feel especially compelled to disclose their genetic test result to their employer. Another crucial finding was the emphatic recommendation of complete financial separation of the testing entity from the employer. Involving more than one testing facility was advocated as a way to reduce the potential for untoward interference by a DOE contractor or the government.

An additional burden, perhaps counterintuitive to scientists, was cited: ''Superman syndrome''; a worker who tests negative no longer feels obligated to follow precautions to reduce exposure, putting themselves and coworkers at peril. This social dynamic could cause a workplace susceptibility-testing program to backfire, especially if: (a) the genetic marker is present in a minority of the work force; and (b) the test is not highly sensitive. These conditions are nearly satisfied by the Glu69 marker: its allelic frequency is about 30–40% among ethnic groups common to the United States, and it is absent from up to 28% of workers with CBD [Silver and Sharp, 2006]. So even voluntary removal of 100% of the individuals deemed ''susceptible'' would not create a safe environment for those with continued exposure.

In the private sector economy, control of health hazards adds significantly to the final cost of beryllium products [U.S. Department of the Interior, 1995] and environmental costs are rising faster than revenues, constraining the market potential of beryllium [U.S. Department of Commerce, 1993]. The beryllium industry's leading supplier (Brush–Wellman Co.), [Lang, 1994] and other industries, [Severo, 1980; Draper, 1991] have long shown interest in minimizing control costs for toxic substances by placing a greater emphasis on genetic testing. Combined with Superman syndrome, the net effect on employees could be to undermine collective action for control measures, historically one of the most effective forces for primary prevention [Weil, 1991, 1999; Boal, 2009].

Our study had several limitations. First, Congress passed the Genetic Information Nondiscrimination Act (GINA, Public Law 110-233) just as we were conducting the last two focus groups. Participants were unaware that GINA narrows the field of actors who may ''request, require, or purchase'' genetic information. Employers are frankly prohibited from doing so; there is no exemption for national security. GINA also proscribes involvement by union and joint-labor management entities [Equal Employment Opportunity Commission, 2009]. Despite this law, our data show that DOE workers will remain skeptical that their employers' compliance will be any better than with other federal health, safety and environmental laws. Second, to recruit participants we did not draw a random sample from a well-defined population of beryllium workers. Rather, by means of outreach through a support group and labor and community organizations, we recruited a snowball sample of hourly employees. The range and diversity of knowledge, beliefs, and attitudes captured by the study on many key issues weighs against systematic bias. However, an analogous study of salaried managers' knowledge, beliefs, and attitudes might prove very interesting indeed. In addition, the focus group approach, wherein individuals' concerns are voiced in a group setting, may have engendered a degree of personal reticence about sensitive health and employment concerns. So our findings may understate the degree of worker concern about genetic testing.

This is the first formal study to frame these issues in the voices and vernacular of directly affected workers. Policy makers may gain a better appreciation of the full range of issues related to genetic susceptibility testing in the workplace. We make no pretense of reporting consensus positions; no consensus was achieved or even sought. Nor do we present a synthesis of the individual views expressed. Rather, the comments presented are representative of the discussion which occurred in the focus groups (i.e., $N = 5$), capturing the range of knowledge, beliefs, and attitudes among beryllium workers at three federal nuclear weapons facilities who self-selected to participate after being presented with study recruitment materials by a local organization. Our findings were heavily influenced by unique features of work in the nuclear weapons complex, so generalizing beyond the workforce of beryllium-exposed employees at DOE facilities may not be warranted.

Our results do suggest a framework for implementing a genetic testing program in a way that has the potential to address many of the worker concerns identified. In our view, the most viable venue for genetic testing is medical surveillance programs that have been funded by DOE extramurally since the mid-1990s [Podonsky, 2009]. These programs have administered the LPT test to tens of thousands of current and former workers. Many of these

teams include occupational physicians well-versed in beryllium. By virtue of their university affiliations, these programs can keep abreast of the science of genetic markers, along with strategies for tailoring genetic counseling protocols. DOE former worker programs have advantages with respect to maintaining the privacy of genetic test results. The files of participants in medical surveillance projects are smaller than patient records in a private practice. So a genetic test result is less likely to be inadvertently transmitted to anyone other than the worker. Also, multiple layers of human subjects review are performed by institutional review boards, with a keen eye to the protection of participants' privacy. Teams led by university-affiliated physicians are also well-situated to provide leadership to family doctors on a regional basis.

Apart from research on molecular mechanisms of disease, a bigger role for former worker programs in applying markers of genetic susceptibility could be useful in informing individuals of their subsequent risk of disease and in earlier detection of disease. But the findings of our study stress that genetic testing as a preplacement screening tool should be approached with heightened caution.

Some DOE former worker programs are managed by labor unions with many years' experience representing nuclear workers on job safety and health and collective bargaining issues; university-based programs generally include union representatives on their advisory boards. These affiliations will be crucial to ensuring that any future efforts to make genetic testing available will first establish a full suite of protections for the rights and remedies noted in this study. One complication is that GINA's prohibition on requesting genetic information applies to unions as well as employers [Slaughter, 2008; Equal Employment Opportunity Commission, 2009]. Special entities for research and service that are supported, but not controlled, by unions may be a sensible vehicle for exploring the issue of genetic testing programs. Alternatively, some unions may warmly embrace GINA's prohibition as a bright-line codification of their longstanding opposition to the very idea of genetic testing [Draper, 1991].

A phased approach could make the genetic susceptibility test available to *former* workers initially—as a proving ground. Former workers could thereby be cultivated as leaders of later efforts to offer the test on a voluntary, confidential basis to *current* and *prospective* beryllium workers. Informal peer counseling might improve the quality of genetic counseling. A retiree's suggestion for a ''safety board'' established by employers with ''workers in charge,'' to administer a voluntary testing program, may have been loosely inspired by the Advisory Board on Radiation Worker Health. Involvement of regional primary care providers, pulmonologists, genetic counselors, labor, and advocacy organizations in shaping these programs could align resources for follow-up. In 1992, Holtzman forecasted that genetic tests with low acceptance, low PPVs, and high potential for insurance discrimination would not be used in population screening programs; such tests would only find use in the ''default destination'' of family-centered testing [Holtzman, 1992]. This prediction is borne out here by the preference of focus group participants for their personal physician.

No doubt, each such effort would wrestle anew with the ethical and social issues addressed in this paper. Their answers may differ by region of the country [Nelson et al., 2009]. Obtaining funding from an agency other than DOE will be important. Workers in New Mexico and Tennessee felt that complete financial separation from the employer is essential. Also, it would be necessary to avoid any boilerplate provisions, common in federal grants and contracts, which entitle the agency to take custody of project records. However seldom these powers have been utilized in the past, the critical thinkers we met in our focus groups who are distrustful of government would likely consider the removal of such provisions to be non-negotiable.

Facile comparisons are problematic. Yet an analogous body of social research and public debate focuses on genetic markers of breast cancer risk. Although not as closely tied to job exposures, genetic testing for susceptibility to breast cancer has sparked controversy around workplace discrimination [Greenhouse, 2010]. More broadly, focus groups and interviews have identified a similar constellation of issues: autonomy, insurance discrimination, family participation in decision-making, risk perception, and psychosocial effects (i.e., worry and depressive symptoms) [Bluman et al., 2003; Hallowell et al., 2005; Bakos et al., 2008; Henneman et al., 2010]. For the development of worker-centered testing programs, alternatives to conventional client-centered genetic counseling protocols are highly germane; these would place a greater emphasis on problem-solving training [McInerney-Leo et al., 2004]. One study found the highest level of interest in genetic testing for breast cancer susceptibility to be for a hypothetical test that combines a high PPV with non-invasive therapy [Press et al., 2001]. Yet worldwide data show that about one-fifth to one-third of women without breast cancer will opt for prophylactic mastectomy, and half will choose risk-reducing oophorectomy, on the basis of a positive BRCA test, the PPV of which can approach 90%. Among the nine countries studied, the United States had the highest rates of such surgeries [Metcalfe et al., 2008]. Obviously, leaving a beryllium job voluntarily is incommensurate with invasive surgery. Macroeconomic conditions, individuals' skill sets, and a multitude of private considerations will determine how radical or invasive the elective choice feels to the worker.

Our findings indicate that, the test's PPV of 14% is sufficient to provide information that some workers would

see as ''so important,'' in the words of a union member. With research in molecular epidemiology and modeling ongoing, the test's PPV may increase—and with it, workers' interest in genetic testing. Should the PPV ever exceed 50%, there won't be many winning arguments standing in the way of making the test available to interested workers. A positive result would mean ''more likely than not'' the person will develop disease. The test has not yet reached the stage of commercialization, despite an April 2000 public affairs newsletter at LANL forecasting ''transfer to industry'' in a year [Anon, 2000]. But once commercial testing interests enter the picture [Burke, 2005; Eng and Sharp, 2010; McGuire et al., 2010] there may be insufficient time and turf to execute a sound, phased strategy of needs assessment, intervention, and longitudinal follow-up of outcomes.

This study provides insight into workers' perspectives. But their direct involvement in the design and implementation of testing programs is likely to be far more effective in ensuring that if genetic testing is used, it will be used to the benefit, not the detriment, of workers and their families. Those concerned with protecting workers' rights, along with advocates for the preventive application of genetic science, currently have a window of opportunity to establish partnerships among key stakeholders—including labor, management, public health scientists, and primary care professionals—to control this technology.

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