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Software and Equipment

A. SuperCalc, Computer Associates International, Inc., 2195 Fortune Dr., San Jose, CA 95131.
B. Lotus 1-2-3, Lotus Development Corporation, Cambridge, MA.
C. Toshiba America Inc., 2441 Michelle Dr., Tustin, CA 92680.
D. Hewlett-Packard Personal Computer Group, 10520 Ridgeview Ct., Cupertino, CA 95014.
E. Diconix Inc., 3100 Research Blvd., Dayton, Ohio 45420.
F. Survey Mate, Henry Elkins and Associates, Inc., 15 Willow Circle, Bronxville, NY 10708.

An International Survey of Attitudes of Medical Geneticists Toward Mass Screening and Access to Results

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Synopsis

A survey on mass screening was sent to 1,053 medical geneticists in 18 nations, of whom 677 responded. Three theoretical screening situations were proposed, screening in the workplace for genetic susceptibility to work-related disease, carrier screening for cystic fibrosis, and presymptomatic testing for Huntington disease.

Of the respondents, 72 percent thought screening in the workplace should be voluntary, and 81 percent said employers should have no access without the worker's consent, including 22 percent who believed that employers should have no access at all. There was strong consensus in all but one nation that insurance companies should have no access to test results without the worker's consent, and strong consensus in two countries that they should have no access at all.

Most (82 percent) believed that screening for cystic fibrosis should be applied to the entire population, but 18 percent believed that it should be applied primarily to Caucasians.

In all, 66 percent of respondents believed that individuals at risk for Huntington disease should be told their test results only if they say that they wish to know, recognizing a "right not to know" whether they will develop the disease in later life. Twelve percent thought that spouses should have access to test results if they asked, and 26 percent thought that spouses should be informed of results even if they did not ask.

Geneticists in all nations were vividly aware of the potential damage from third party access to results, especially access by insurance companies. They had little sympathy with insurers' needs to assess actuarially accurate premiums.

WITHIN the next 10 years, genetic screening, either in the workplace or for common diseases, may affect the majority of people in developed countries (*1, 2*).

As the possibilities for mass screening increase, so will the attendant ethical problems. Ethical problems include balancing the prevention of harm from genetic disease against the possibilities of

social stigmatization of carriers or discrimination in employment, balancing public health concerns against the rights of individuals, and balancing the rights of third parties, such as employers, insurance companies, or relatives at genetic risk, against the individual's right to privacy. The history of screening for sickle cell carrier status highlights these as yet unresolved dilemmas (*3–5*). Clearly, the views of medical geneticists will carry weight, as scientists, lawmakers, and the public try to establish regulations for the conduct of mass screening and access to test results. Individual geneticists have ethical views (*6–10*), and government and scientific commissions have issued statements (*1, 11–13*), but there has been no systematic study of the actual approaches of medical geneticists to ethical problems in mass screening.

Fletcher and coworkers proposed that medical geneticists around the world would benefit from collective reflection on their preferred approaches (*14*). In order to gather information for future discussions, we studied the views of 677 medical geneticists from 18 nations when they were presented with ethical dilemmas concerning screening situations and access to results (*15*). Our purpose in surveying was to initiate discussion, not to make statistical comparisons among nations, many of which have a small number of geneticists. Ethical problems in screening are not limited by national boundaries. Professionals, policymakers, and the public may learn from approaches used in other nations.

Cross-national Survey

The rationale for choosing countries for study was (*a*) 10 or more practicing geneticists, (*b*) geographical and cultural distribution, and (*c*) the presence of a medical geneticist willing to distribute and collect questionnaires and to coauthor, with a specialist in medical ethics, a chapter in a future monograph, "Ethics and Human Genetics" (*16*).

We developed questionnaires after field studies at genetics centers in 12 nations. We asked respondents what they would do, from a fixed list of possible responses, and why, in their own words, they had chosen this particular course of action, with regard to three screening situations and access to test results. Respondents were asked to rank the ethical import of 10 future priorities, as they believed the priorities ought to be seen by geneticists. The list included carrier screening for common genetic disorders, screening for susceptibility to cancer and heart disease, and screening in the workplace for susceptibility to work-related disease.

We selected geneticists who held an MD, PhD, or equivalent degree, and who were engaged in delivering or administering genetic services (testing, counseling, prenatal diagnosis, or laboratory work). Although in some countries (notably the United States) counseling is sometimes done by specially trained persons who do not hold a doctorate, we decided to omit these persons to control for consistency of training across the entire sample.

In each country, including the United States, our contact geneticists tried to include all qualified medical geneticists in the survey. Lists were compiled from certifying boards, genetics centers, and the National Foundation - March of Dimes "International Directory of Genetic Services."

Of the 1,053 geneticists asked to participate, 677 (64 percent) returned completed questionnaires by the close of the study in February 1987 (table 1). Ninety-two percent answered all questions dealing with screening and 68 percent gave reasons for their actions. Eighty-one percent held MD degrees, 16 percent held PhD degrees, and 3 percent held other degrees. The geneticists had a median of 14 years in the practice of genetics; 82 percent were members of their national genetics society, and 84 percent were board certified in countries where certification in genetics was possible (Canada, the United States, and Hungary). Respondents spent an average of 45 hours a week in genetics. Sixty-five percent were male, and 82 percent were married with a median of 1.5 children. Religious backgrounds were 40 percent Protestant, 18 percent Catholic, 17 percent Jewish, 12 percent none, 5 percent Buddhist, 4 percent Hindu, and 4 percent other. Overall, they were nonpracticing, attending a median of one religious observance a year. Forty-nine percent characterized themselves as politically liberal, 15 percent as conservative, and 36 percent as both. In the United States, a comparison between 274 respondents and 208 nonrespondents listed in the 1986 combined "*Membership Directory*" of the Genetics Society of America, American Society of Human Genetics, and American Board of Medical Genetics revealed no statistically significant differences between respondents and nonrespondents in type of degree, gender, geographic area, or subspecialty.

The three screening situations presented were screening in the workplace for genetic susceptibility to work-related disease, carrier screening for cystic fibrosis, and presymptomatic testing for Huntington disease. At present, none of these is technologically feasible or accurate on a population-wide

basis. Respondents were asked to assume that reliable tests were available. In phrasing the questions, we did not make specific statements about sensitivity or specificity. The text of each question appears subsequently.

For each question, respondents were given a range of choices, and asked, "why did you choose this course of action?" No structure was provided for the "why" responses. Responses were entered into a Statistical Package for the Social Sciences (SPSS-X) program. The open-ended responses were coded according to a list of the 92 reasons most frequently given. In coding, we also noted whether or not a response included mention of specific consequences. Examples of specific consequences are discrimination in the workplace, loss of insurance coverage, or improved health care. We coded the first two reasons given and the mention of consequences.

Our criteria for consensus were those sometimes used in legislative processes, such as the ratification of amendments to the U.S. Constitution, in the absence of an accepted scientific criterion. We used a "3/4s rule" (3/4s of the respondents in each of 3/4s of the countries) to define a "strong consensus." This method allows representation for each country. If we had used percentages of the total number of responses, the United States, with 44 percent of all respondents, would have been disproportionately represented.

Screening in the Workplace

Although no test for genetic susceptibility to occupationally related disease has yet met U.S. Office of Technology Assessment criteria for actually predicting disease, an OTA survey of 336 major U.S. corporations in 1982 showed that 17 had already used genetic tests and 59 expected to use them within 5 years (*17*). Our question on screening concerned a test already used by some companies.

"Assume that an accurate, simple, and reliable mass screening test has been developed for alpha-1-antitrypsin deficiency. This raises the possibility that factory workers who will be exposed to dust and smoke could be screened. Assume that you are a member of an advisory group that will develop guidelines for mass screening of workers in your country. Do you believe that mass genetic screening of workers and prospective employees in potentially dangerous industries should be: (1) mandatory for all who would be occupationally exposed, or (2) voluntary?"

Serum alpha-1-antitrypsin (SAT) deficiency is an important biological factor predisposing the occurrence of emphysema. Approximately 80 percent of homozygous individuals will develop emphysema. The homozygous state occurs in approximately 1 in 4,000 persons in the U.S. population. Heterozygous individuals have an SAT level about 50 percent of normal and may be at increased risk for emphysema if they smoke or work in dusty environments. They comprise about 3 percent of the U.S. population. SAT testing is the second most frequently used form of genetic screening in U.S. industries, after screening of blacks for sickle-cell trait. Pilot studies have not been conducted, and benefit has not been proven (*17, 18*). At present, the value of SAT tests for predicting the development of emphysema in heterozygous individuals is unknown. Although the prior question is whether such screening should be conducted at all, we were interested in international views about mandatory or voluntary screening, if and when tests became sufficiently proven. In the absence of tests with proven benefits to workers, we used SAT for purposes of eliciting responses on mandatory or voluntary screening.

Geneticists strongly preferred voluntary over mandatory screening, by a 72 percent majority (table 2). In 10 nations (Australia, Denmark, Federal Republic of Germany, Greece, Italy, Norway, Sweden, Switzerland, United Kingdom, and United States) there was a strong (> 75 percent) consensus that screening should be voluntary. In one nation, German Democratic Republic (GDR), there was a

Table 1. Response to the international survey of geneticists

Nation	Number asked to participate	Number of persons responding	Response rate (percent)
Australia	14	12	86
Brazil	51	32	63
Canada	73	47	64
Denmark	28	15	58
Federal Republic of Germany	55	47	85
France	35	17	49
German Democratic Republic	25	21	80
Greece	11	7	64
Hungary	18	15	83
India	40	27	68
Israel	17	15	88
Italy	26	11	42
Japan	74	51	69
Norway	10	6	60
Sweden	26	21	81
Switzerland	10	5	50
United Kingdom	50	33	66
United States	490	295	60
Total	1,053	677	64

Table 2. Choices of geneticists responding to questions about a proposed screening for genetic susceptibility to work-related disease, and whether to provide specific groups with access to test results, by country

Country	Percent favoring screening to be voluntary	Percent who would not allow access without worker's consent, including percent choosing no access at all, by subject group, and percent of the total choosing no access at all							
		Employer		Worker's physician		Life, health, compensation insurers		Government health departments	
		No access without consent	No access	No access without consent	No access	No access without consent	No access	No access without consent	No access
Australia	[1] 92	[1] 100	...	67	...	[1] 94	47	67	42
Brazil	34	68	10	[2] 19	3	[1] 77	27	39	10
Canada	73	[1] 91	21	64	2	[1] 96	40	[1] 81	26
Denmark	[1] 86	[1] 100	47	60	7	[1] 100	59	[1] 86	29
Federal Republic of Germany	[1] 94	[1] 96	64	68	4	[1] 95	71	72	44
France	53	[1] 100	[1] 77	29	...	[1] 88	56	50	13
German Democratic Republic	[2] 16	[1] 81	50	[2] 22	...	[1] 90	[1] 78	38	25
Greece	[1] 83	50	...	[2] 17	...	[1] 83	17	[2] 17	...
Hungary	27	[1] 79	29	[2] 7	...	[1] 91	29	29	7
India	26	41	14	[2] 17	4	65	30	32	9
Israel	73	[1] 100	7	43	...	[1] 100	24	71	21
Italy	[1] 82	[1] 100	27	27	9	[1] 87	55	36	9
Japan	63	[1] 77	23	35	5	[1] 90	54	44	20
Norway	[1] 84	[1] 100	[1] 83	[1] 100	...	[1] 100	[1] 83	67	33
Sweden	[1] 91	[1] 100	35	[1] 91	...	[1] 98	38	[1] 79	26
Switzerland	[1] 100	[1] 100	60	40	...	[1] 100	73	[1] 100	60
United Kingdom	[1] 87	[1] 84	10	42	...	[1] 88	19	[1] 81	19
United States	[1] 77	[1] 76	12	63	2	[1] 88	34	[1] 78	27
Total	72	[1] 81	22	53	2	[1] 89	40	68	25

[1] Indicates strong (≥ 75 percent) consensus in favor of voluntary screening or no access.

[2] Indicates strong (≥ 75 percent) consensus in favor of mandatory screening or automatic access to results.

strong (> 75 percent) consensus that screening should be mandatory, and in three additional nations (Brazil, Hungary, and India) the majority agreed that it should be mandatory.

In all, 567 (84 percent) gave reasons for their choices. Those who thought screening should be voluntary cited the worker's autonomy or right to decide (74 percent), the danger of stigmatization, discrimination in employment, or misuse of information by institutional third parties (41 percent). Advocates of mandatory screening cited protecting the individual worker's health (64 percent), protecting public health (51 percent), and efficiency or cost-benefit arguments (22 percent). Nine percent of those who advocated voluntary screening and 12 percent of those who advocated mandatory screening based their choices in part upon concern for the economic interests of employers. Those who believed that screening should be mandatory were more likely than the rest to cite personal or social consequences of screening; 56 percent mentioned some specific consequence, as opposed to 40 percent of those who thought that screening should be voluntary.

The two groups differed on whose welfare they placed foremost; 97 percent who advocated voluntary screening and 58 percent who advocated mandatory screening ranked the worker's welfare as most important. Three percent who advocated voluntary screening and 37 percent who believed in mandatory screening placed "society's" welfare first. Only 1 percent placed the employer's welfare first. Believers in voluntary screening were more likely than others to describe a conflict of interest between worker and employer; 34 percent described such conflicts, as opposed to 13 percent who advocated mandatory screening. A majority of both groups, however, described no conflicts.

Access to Test Results

Respondents were asked, "Who should have access to the results of genetic screening for occupational susceptibility?" For each of eight persons or institutions, they were asked to choose one of the following: (1) yes, access; (2) yes, access, but only if worker approves; (3) no access. Ninety-eight percent said that the worker should have access; this included 86 percent who said that the worker should be told the results even if he or she did not ask for them. This view contrasts markedly with the history of concealment of asbestosis test

results from workers at some major U.S. corporations (*19*).

When asked whether the employer should have access to results of tests, 81 percent said that employers should have no access without the worker's consent, including 22 percent who believed that employers should have no access at all (table 2). There was strong consensus for no access without consent in all nations except Brazil, Greece, and India, and strong consensus for no access at all in France and Norway.

In all, 523 (77 percent) gave reasons for their choices about access. Thirty percent believed that it would be to the worker's benefit if the employer had some form of access; employers could shift susceptible workers to less dangerous jobs, though only 6 percent thought that working conditions in general would be improved. Nineteen percent described potential economic discrimination, stigmatization, or other misuse of test results by employers. Ten percent based their responses on the economic interests of the employer.

There was little consensus about whether the worker's physician should have access. There was strong (> 75 percent) consensus in two countries (Norway and Sweden) that the physician should not have access without the worker's consent and strong consensus in five countries (Brazil, GDR, Greece, Hungary, and India) that physicians should have automatic access. In all, 61 percent said that it would be to the worker's benefit for the physician to know; 26 percent, however, said that the worker had the right to decide. In general, respondents trusted physicians far more than they trusted institutional third parties; only 2 percent said that the physician should have no access at all.

There was strong consensus in all nations except India that insurance companies should have no access to test results without the worker's consent, and strong consensus in two countries (GDR and Norway) that they should have no access at all (table 2). Although we asked separate questions about four types of insurers (worker's life insurer, worker's health insurer, employer's worker compensation insurer, and employer's health insurer), the responses were so similar that we have averaged them. The distrust of insurance companies was such that 40 percent of respondents thought that they should have no access at all, even with the worker's consent. Very few (4 percent) thought that workers would benefit in any way from an insurance company knowing their test results. However, 30 percent thought that the information would be misused, to the worker's detriment. Many respondents pointed out that "access with consent" placed the worker in a no-win situation—if the worker denies access, he or she will probably be denied insurance. Some pointed to the need for regulations that would prevent workers from in effect being coerced into giving "voluntary" consent.

'Individual geneticists have ethical views, and government and scientific commissions have issued statements, but there has been no systematic study of the actual approaches of medical geneticists to ethical problems in mass screening.'

When asked whether government health departments should have access, 68 percent said that there should be no access without worker consent. There was a strong consensus to this effect in six nations (Canada, Denmark, Sweden, Switzerland, United Kingdom and United States). There was strong consensus in one nation (Greece) that the government should have automatic access. Fifty percent thought that it would benefit society if the health department had access, in terms of improved public health, working conditions for all workers, or social planning. Only 10 percent (5 percent in the United States) feared possible misuse of information by the government. One-fourth of respondents, however, believed that governments should have no access to results at all. In general, those who believed that screening should be mandatory were more likely than others to believe that third parties of all kinds should have access without consent.

Cystic Fibrosis Carrier Screening

The question was phrased, "Assume that a cheap and accurate test, reliable at all ages, has been developed for cystic fibrosis. It diagnoses both carriers and affected individuals, distinguishes between them, and also separates each of them from non-carriers. The test is now ready for application on a population-wide basis. Assume that a gradual introduction of screening has been proposed in your country. Also assume that accurate *prenatal diagnosis* has become available for cystic fibrosis, to whom and at what age should the carrier test

Table 3. Choices of geneticists responding to questions about which groups should receive a proposed mass carrier screening for cystic fibrosis, by country

Country	Percent choosing newborns, screening required	Percent choosing newborns, screening by consent only	Percent choosing children [1], screening by consent only	Percent choosing adults [2], screening by consent only	Percent choosing children [1] or adults [2], screening required
Australia	0	59	8	25	8
Brazil	25	16	0	53	6
Canada	18	27	11	42	2
Denmark	27	20	20	33	0
Federal Republic of Germany	11	30	9	47	3
France	35	12	6	35	12
German Democratic Republic	67	0	5	10	18
Greece	0	0	0	[3] 86	14
Hungary	67	7	0	7	19
India	39	31	4	23	3
Israel	20	7	7	53	13
Italy	18	0	27	46	9
Japan	0	51	15	30	4
Norway	33	33	0	34	0
Sweden	5	29	0	66	0
Switzerland	0	[3] 80	0	20	0
United Kingdom	0	30	27	37	6
United States	23	22	17	35	3
Total	20	25	13	37	5

[1] Children younger than 18 years. [2] Adults 18 years and older. [3] Indicates a strong consensus (≥ 75 percent).

first be given?" The choice of replies was "(1) to all newborns, required by law; (2) available to all newborns, but only with parents' consent; (3) available to newborns of Caucasian descent, with parents' consent; (4) available to all children younger than 12 years, required by law; (5) available to all children younger than 12, with parents' consent; (6) available to children younger than 12 of Caucasian descent with parents' consent; (7) available to all adolescents 13–17, required by law; (8) available to all adolescents 13–17, with parents' consent; (9) available to adolescents of Caucasian descent with parents' consent; (10) should *only* be given to persons over 18 years, but should be required by law; (11) should be available to all persons over 18 years; and (12) available to persons over 18 years of Caucasian descent who request it."

At present, no such test is available on a population-wide basis. Carrier testing and prenatal diagnosis are possible only on a family-specific basis, using DNA probes that require testing of genetic material from a member of the family who has cystic fibrosis, usually a child (*20–24*). Geneticists anticipate that within a few years tests suitable for mass screening will be developed. At present, however, our question is theoretical. Cystic fibrosis is the most common autosomal recessive disorder among whites, with an incidence of 1 in 1,600 and a carrier rate of 1 in 20. The disorder is relatively rare among nonwhites. Although treatment is available, it places a high daily burden on the family, and the child undergoes frequent hospitalization. Half the children die before the age of 20, and most of the rest before the age of 30. In each subsequent pregnancy, the parents face a one-in-four risk of having another affected child.

Respondents believed that the objectives of mass screening should be (*a*) informed reproductive planning for carriers; (*b*) efficiency; (*c*) preserving patients' (or parents') autonomy; (*d*) identifying and treating affected children as early in life as possible; and (*e*) preventing births of additional children with cystic fibrosis. There was 75 percent consensus among total respondents, and a > 75 percent consensus in each of eight nations (Australia, Canada, Federal Republic of Germany, Greece, Japan, Sweden, Switzerland, and United Kingdom) that cystic fibrosis screening, at whatever age, should be voluntary (table 3). Those who believed that workplace screening should be voluntary were more likely to believe that cystic fibrosis screening should be voluntary. There was no consensus, however, about the optimal age for initial screening in order to achieve program objectives. In all, 20 percent advocated screening newborns by law; 25 percent advocated screening newborns with parental consent; 13 percent would choose screening of children or adolescents younger than 18, with parental consent; 37 percent advocated screening adults older than 18 by consent; and 5 percent indicated other ages, by law. The only countries

with a strong (> 75 percent) consensus about age were Greece (adults) and Switzerland (newborns).

In all, 558 (82 percent) gave reasons for their choices. Those who advocated screening newborns by law gave as their reasons the benefits of early treatment (55 percent), program efficiency (31 percent), informed reproductive planning for the child's parents (30 percent), and preventing the births of additional children with cystic fibrosis (22 percent). Those who preferred newborn screening by consent described the benefits of early treatment (47 percent), preventing the births of children with cystic fibrosis (43 percent), the parents' right to decide (42 percent), informed reproductive planning (35 percent), and efficiency (19 percent). Most advocates of adolescent screening chose the ages 13–17 years, arguing that because reproductive activities may begin early, this is the most effective age at which to identify and counsel carriers with regard to family planning. Those who would prefer to screen adults by consent mentioned reproductive planning (74 percent), program efficiency (46 percent), and the individual's right to decide (40 percent). In all, only 4 percent mentioned the possibility of carrier stigmatization, a figure that seems low in view of past experiences with sickle cell and Tay Sachs screening (*2, 25*).

Most (82 percent) believed that screening for cystic fibrosis should be applied to the entire population, but 18 percent believed that it should be applied primarily to Caucasians. In five nations more than 20 percent said that screening programs should focus on Caucasians. These were Japan (55 percent), Italy (46 percent), Canada (27 percent), Brazil (22 percent), and Switzerland (20 percent). Many geneticists in the United States and other nations that favored screening for all said that few persons in their countries were without some Caucasian blood; to restrict screening on the basis of race would be discriminatory.

A total of 78 percent gave priority to the welfare of the person being screened, 10 percent to the welfare of society, and 9 percent to the health of future generations. Most (91 percent) envisaged no conflicts of interest in screening programs for cystic fibrosis. As in the previous question, advocates of mandatory screening were more likely than others to list and describe the consequences of screening programs.

Presymptomatic Test for Huntington Disease

Our third screening situation asked: "When a 99 percent accurate pre-symptomatic test for Huntington disease is developed that applies to all families, who should have access to the results of the test?" For each of seven persons or institutions, respondents were asked to choose one of the following: "(1) access if requested; (2) should be informed; (3) access if patient approves; and (4) no access."

Ranking of Future Priorities

1. Increased demand for genetic services
2. Allocation of limited resources
3. Carrier screening for common genetic disorders
4. Environmental damage to the unborn
5. New treatments for common genetic disorders
6. Screening for susceptibility to cancer, heart disease
7. Genetic screening in the workplace
8. Research on the human embryo, zygote, and fetus
9. Long-range eugenic concerns
10. Sex preselection for sex desired by parents

Such tests are not currently available on a population-wide basis, although they have been developed for specific families. Huntington disease is a fatal, nontreatable, nonreversible neurological disorder that first strikes in middle age and leads to progressive mental and motor deterioration during 10 to 15 years, culminating in death. It is an autosomal dominant disorder, meaning that each of the victim's children has a 50 percent chance of developing the disease, without prior warning, in later life. Until then, they live under a cloud of uncertainty. A presymptomatic test, given early enough, would permit potential victims to plan both their lives and their families. Early warning, however, will not lead to better prognosis, and will almost certainly produce depression, stigmatization, and loss of economic benefits for some. In the United States, there is a high probability that the question of access will be discussed with patients before they agree to be tested. Many of those at risk are themselves uncertain about whether or not they would wish to know the results of presymptomatic tests (*28, 29*).

The question is not whether patients should have access to test results if they ask (98 percent of our respondents believed that they should), but whether they should be informed of these results even if they do not wish to know. In all, 66 percent of respondents believed that individuals at risk should be told their test results only if they say that they wish to know (table 4). In other words, persons at risk for Huntington disease should have a "right

Table 4. Choices of geneticists responding to questions about whether to provide specific groups with access to results of a theoretical presymptomatic test for Huntington disease, by country: percent who would not allow access to results without the patient's consent, including percent of the total allowing no access at all for the specified group, and percent of the total choosing no access at all by the specified group

Country	Patient [1]		Spouse		Relatives at risk		Employer		Life and medical insurers	
	No access without consent	No access at all	No access without consent	No access at all	No access without consent	No access at all	No access without consent	No access at all	No access without consent	No access at all
Australia	50	...	[2] 75	8	67	...	[2] 100	67	[2] 100	58
Brazil	56	...	44	6	[3] 16	...	[2] 94	31	[2] 91	28
Canada	67	...	56	7	65	10	[2] 100	54	[2] 100	46
Denmark	73	...	53	7	27	...	[2] 100	60	[2] 100	67
Federal Republic of Germany	[2] 84	4	[2] 82	7	69	4	[2] 100	[2] 91	[2] 95	[2] 84
France	71	6	[2] 77	15	46	8	[2] 100	[2] 83	[2] 100	67
German Democratic Republic	[2] 75	...	[2] 79	16	65	20	[2] 100	[2] 89	[2] 90	[2] 85
Greece	33	...	[3] 17	...	50	...	[2] 100	17	[2] 83	17
Hungary	53	13	67	20	27	7	[2] 93	60	[2] 87	60
India	30	4	[3] 24	4	27	4	70	35	60	40
Israel	57	...	50	...	46	8	[2] 100	50	[2] 100	21
Italy	67	...	50	10	50	10	[2] 100	60	[2] 100	50
Japan	57	8	55	17	37	14	[2] 85	67	[2] 88	70
Norway	50	...	67	...	50	...	[2] 100	[2] 83	[2] 100	[2] 83
Sweden	71	...	[2] 79	...	57	...	[2] 100	62	[2] 100	48
Switzerland	[2] 80	...	60	...	[2] 80	...	[2] 100	[2] 80	[2] 100	60
United Kingdom	[2] 94	...	[2] 77	...	71	...	[2] 97	16	[2] 100	19
United States	67	...	65	1	57	2	[2] 98	34	[2] 94	36
Total	66	2	62	5	52	4	[2] 96	46	[2] 93	45

[1] If patients ask to be informed, as opposed to informing patients who do not want to be informed.

[2] Strong consensus (≥ 75 percent) in favor of restricted access.
[3] Strong consensus (≥ 75 percent) against restricted access.

not to know" whether they will develop the disease in later life. There was strong (> 75 percent) consensus about this right in four nations (Federal Republic of Germany, German Democratic Republic, Switzerland, and United Kingdom).

Access for the individual's spouse, who will face serious emotional and financial burdens if the person is affected, and for relatives at risk of developing Huntington disease, presents serious ethical dilemmas. If the tested individual does not give consent, the geneticist is faced with a conflict between the duty to preserve patient confidentiality and the duty to warn third parties of harm. In all, 62 percent thought that spouses should have no access to test results without patient's consent. There was strong (> 75 percent) consensus to this effect in six nations (Australia, Federal Republic of Germany, France, German Democratic Republic, Sweden, and United Kingdom). Twelve percent, both of the total and in the United States, thought that spouses should have access to test results if they asked, and 26 percent thought that spouses should be informed of results even if they did not ask.

In all, 459 (68 percent) gave reasons for their choices about access. Of those who would inform the spouse, 45 percent cited reproductive planning, 24 percent cited preparation for the future, and 19 percent said that the spouse had a right to know. In the two nations (Greece and India) where there was strong consensus that spouses should have access without patient consent, Huntington disease is rare. Many respondents had never seen a case.

There was no consensus about access for relatives at risk of developing Huntington disease. In all, 52 percent thought that relatives should have no access without the patient's consent, 24 percent thought that they should have access without the patient's consent if they wanted the test results, and 24 percent thought that they should be informed of the results, without the patient's consent, even if they did not ask. Reasons included the patient's right to privacy (17 percent), relatives' right to know (25 percent), and providing information for relatives' reproductive or future plans (27 percent).

When we asked about access for institutional third parties, however, there was overwhelming consensus almost everywhere that employers, schools, life insurers, and health insurers should not have access without consent. Geneticists' misgivings about these third parties was such that almost half said that they should have no access of any kind, even if the individual gave consent. Few

(5 percent) saw any benefit for individuals in allowing institutional third parties to know their test results, and 28 percent believed that institutions would misuse the information. Only 4 percent expressed concern for the economic interests of third parties. A question about access for school officials (not reported in table 4) produced responses similar to those for other institutions. Most geneticists thought that schools had no need to know that a student would develop a disorder in middle age.

Future Priorities

When asked to rank-order a list of priorities for the next 5 years, geneticists around the world placed carrier screening third out of 10, behind increased demand and allocation of limited resources. Geneticists in three countries (Greece, Hungary, and Italy) ranked carrier screening first, and geneticists in six countries (Federal Republic of Germany, Italy, Norway, Switzerland, United Kingdom, and United States) ranked it second. Screening for susceptibility to cancer and heart disease ranked sixth, and screening for susceptibility to work-related disease ranked seventh, above research on the human embryo, long-range eugenic concerns, and sex selection. Those who believed that screening in the workplace or carrier screening should be mandatory ranked these on average one step higher in priority than did those who advocated voluntary screening.

Conclusions

When confronted with ethical problems of screening, geneticists favored voluntary rather than mandatory screening. Exceptions were socialist nations where workers enjoy strong legal protection of their jobs. Geneticists in these nations considered it unlikely that screening would result in discrimination or economic hardship. In some developing nations where working conditions are hazardous and workers' movements are relatively weak, respondents regarding mandatory screening as a means of protecting the most vulnerable.

Geneticists in all nations were vividly aware of the potential damage from third party access to results, especially access by insurance companies. They had little sympathy with insurers' needs to assess actuarially accurate premiums. Requiring that institutional third parties obtain the individual's consent is not sufficient protection, for institutions have the economic power to force

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consent. Nations will need to develop regulations, before mass screening becomes a reality, that will (*a*) set standards for minimum predictive values, below which tests cannot be used; and (*b*) prohibit institutional third parties from access to the results, even with consent, unless such access will benefit or protect the individual's health.

Predictive value (true positives divided by true positives plus false positives) is a major policy and ethical issue in population-based testing. A test for cystic fibrosis homozygotes (incidence is 1 in 3,000) with 99 percent sensitivity and specificity would yield 30 false positives for every true positive. For common genetic predispositions, estimated predictive values (patients with positive results who will develop the disease) are diabetes, 0.12; coronary artery disease, 0.16; and lung cancer, 0.14 (*28*). For this reason it is imperative that screening be voluntary and that confidentiality be strictly protected. It is also necessary that positive benefits be demonstrated, such as a reduction in disability, before tests are widely used.

Preserving patient confidentiality will be difficult, especially if employers conduct the testing, even on a voluntary basis. In nations where private insurance plays a major role and where insurers will likely deny insurance to those who refuse access to test results, government-subsidized risk pools may be necessary to guarantee affordable coverage to all.

There is an analogy between genetic screening and testing for human immunodeficiency virus (HIV). It is likely that because of the urgency of the AIDS question, society will deal first with the issue of confidentiality and access to insurance raised by individuals signing waivers to insurers, a risky proposition at best. In nations without national health insurance, governments must decide how to prevent discrimination by insurers and other third parties on the basis of genetic information. Access for family members, especially those at genetic risk, presents complex ethical problems

that may be resolved differently in different cultural settings, but should be the subject of international discussion.

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