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Tay-Sachs Screening: Motives for Participating and Knowledge of Genetics and Probability

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Tay-Sachs screening represents a new form of medical practice, novel both to the public and to the medical profession. To suggest to a person that he has some chance of possessing genes injurious to himself or his children, and then to persuade him to do something which may make the probability a certainty, is a departure from traditional medical practice in which the physician is consulted only when there is need. For this reason, the initiator of a genetic screening program should weigh its potential psychological and social impact.

To test this impact, a mass screening program of couples at risk for Tay-Sachs disease was evaluated. The aims of the study were: (1) to discover what attracted the participants to the screening program and how they decided to be tested; (2) to test the participants' knowledge of the odds for having a child with Tay-Sachs disease, for being a carrier, and, to a lesser extent, of genetics in general; and (3) to evaluate the impact of the program on the personal lives of the subjects and to test their satisfaction with it. The results of the study are reported in two papers of which this is the first.

MATERIALS AND METHODS

Because of the drawbacks of retrospective studies in which answers to questions about attitudes may be colored by subsequent experiences, two populations were studied; one was composed of people tested 1–3 years before being interviewed (retrospective), and the other, of people tested and interviewed during 1973–1974 at the Johns Hopkins Hospital (prospective).

The Retrospective Group

This population was drawn from a mass screening program organized by M. M. Kaback. During 1971 and 1972 sera from approximately 7,000 persons living in the Baltimore-Washington area were tested for hexosaminidase activity. The characteristics of this population, the organizational details of the screening sessions, and the methods used for

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testing and assigning carrier or noncarrier status have all been reported previously [1, 2]. For this paper it is necessary only to add that public education prior to screening was offered by means of pamphlets and fliers, newspapers, television, meetings at synagogues, and even sessions with physicians in the hope that they might alert their patients. In addition, the physicians were offered informative pamphlets to hand out in their offices.

From this group of 7,000 persons, a sample of 940 individuals was selected which included: (1) all carriers and their spouses from either Baltimore or Washington; (2) all persons from the Baltimore area whose first test results were inconclusive and their spouses; (3) a group of men living in the Baltimore area whose wives were not tested because they were pregnant at the time; and (4) every tenth person shown to be a noncarrier from the Baltimore area and his or her spouse. Each person was asked by letter to submit to a questionnaire to be administered at home by a trained interviewer. Those who failed to answer the letter were telephoned repeatedly until an answer was obtained. The composition of the final sample of 635 persons interviewed and the reasons for the failure of the remaining 305 persons to participate are shown in table 1. The heavy loss of carriers was mainly due to individuals from Washington who answered neither letter nor telephone.

TABLE 1

COMPOSITION OF POPULATION SAMPLED

	No. Sampled	No. Interviewed	Refused	Separated	Dead	Moved	Could Not Be Reached
Retrospective group:							
Carriers and spouses	428	256	30	10	2	38	92
Inconclusives and spouses	142	104	16	10	...	4	8
Noncarriers	304	218	28	14	...	8	36
Husbands of pregnant wives	66	57	1	2	...	2	4
Prospective group	104	104	...	...	...	...	...
Total	1,044	739	75	36	2	52	140

The Prospective Group

For persons unable or unwilling to attend mass screening sessions, testing facilities were available at the Johns Hopkins Hospital. At the hospital, the subjects filled out a questionnaire giving demographic information followed by a half-hour session with a physician during which their questions were answered, information on the frequency of Tay-Sachs disease and the carrier state was given, and a blood sample was taken; within 10–30 min after talking with the physician, the subjects were interviewed. They were then asked to submit to a second interview after the results of the test were known. Data obtained from 104 persons (52 couples) are included (table 1). Three of these were carriers. Twenty-nine of those who submitted to the first part of the interview were not available for the second, reducing the number available for parts of the study to 75.

The Interview

The interview consisted of questions appropriate for all participants plus a few which were related to specific categories; for example, questions for carriers might be inappro-

priate for noncarriers or for persons with inconclusive results. A few questions were presented to the prospective subjects only.

The interview was mainly closed-ended and contained 60–70 questions covering the following: (1) data which define the populations; (2) how the subjects found out about the screening program, what attracted them to it, and how they decided to be tested; (3) their knowledge of genetics and probabilities; and (4) the impact of the testing on their personal lives, their satisfaction with the program, and their attitudes toward screening in general. For the retrospective group, 95% of the interviews were administered at home and the remaining 5% in the subject's office. For the prospective group, the initial interview was done in the hospital; the second interviews were all at home. Apart from the husbands of pregnant wives, only couples were included, and each spouse was interviewed separately.

RESULTS

Characteristics of the Population

Age, occupation, education, and income. The mean age of the retrospective participants was 29.7 years and of the prospectives, 27.2 years. Most of the men were engaged in professional or managerial work, while their wives were primarily involved with household duties. Forty-five percent had some graduate training, 75% were college graduates, and only 6% had no college experience. Approximately one-half claimed an annual income of more than $20,000.

Family plans. Fifty-five percent of the retrospective and 91% of the prospective subjects planned to have more children. Among the retrospective couples, 11 women (4%) and among the prospectives, 21 women (40%) were pregnant. If we count the 57 husbands of pregnant, but untested, wives as couples, then about 25% of all couples were in the process of having a baby.

Attitudes to reproductive control. The principal reason for being tested given by the pre-screening educational material was the elimination of the risk of Tay-Sachs disease by means of antenatal diagnosis and abortion of an affected fetus. It was not surprising, therefore, to find that only five participants opposed birth control (16 were indifferent and five refused to answer), and only 15 were categorically opposed to abortion. When asked the conditions under which they favored abortion, the participants gave such answers as "if the fetus is defective" or "if the parents want it" or both; the former outnumbered the latter 2:1. Only 6% of the couples disagreed about favoring abortion, but 26% differed with regard to acceptable reasons for abortion.

Reaching the Decision to be Tested

Discovering the screening program. The sources through which the participants learned of the screening program are given in table 2. Although both members of a couple were likely to acknowledge the same source, one-third indicated that each had learned of the program independently. Two-thirds of the women stated that they heard about the program before their husbands, a statement with which two-thirds of the husbands agreed. About 10% of both sexes revealed that they had heard about the program simultaneously, and a further 10% did not know. The

TABLE 2

DISTRIBUTION OF PRINCIPAL MEANS OF DISCOVERY ABOUT TAY-SACHS SCREENING

	Synagogue	Rabbi	Doctor	Newspaper	Radio/TV	Relatives	Friends	Other	Multiple Sources	Do Not Know	Total
Retrospectives:											
Carriers	41	...	5	49	12	57	28	13	45	6	256
Noncarriers	24	5	4	45	7	34	26	17	51	5	218
Inconclusives	10	...	...	23	7	20	15	12	14	3	104
Husbands of pregnant wives	4	...	1	18	4	9	9	3	5	4	57
Prospectives	4	2	12	12	11	16	13	14	19	1	104

carrier couples were significantly more likely to have heard about screening from relatives ($P > .001$).

Deciding to be screened. After learning about the program, the participants discussed it among themselves and with others, 35% with friends and 28% with relatives. Only 2.5% discussed the issue with a physician, despite the fact that 90 women were pregnant at the time of the screening, and less than 1% discussed the matter with a rabbi, despite the ethical issues involved in abortion.

Although both partners of a couple usually agreed immediately to be screened, 47 (8%) said that one of them had been reluctant; males were four or five times more likely to be disinclined because of lack of interest, insignificant probability of having a baby with the disease, no time, or fear of venipuncture.

Motivation for Testing

Reasons given for being tested. Most (72%) of the participants gave health-related reasons for being tested (i.e., to have a healthy child, to discover what one's chance of having a Tay-Sachs child might be, or the realization that one is a Jew and vulnerable); thirty-four (4.6%) mentioned a relative or an acquaintance who had a child with Tay-Sachs disease as a reason, 7% cited pregnancy, 4% (mostly males) cited social pressures, and 12% did not know. Two-thirds of the couples agreed on the reason they cited, but in 36%, the spouses gave a different reason.

Prior knowledge of Tay-Sachs disease. Just under one-half (43.8%) of the participants said they had heard of Tay-Sachs disease before they knew about the existence of the screening program. There were no significant interclass differences. They learned about the existence of the disease from newspapers, television, and radio (33%), friends and relatives (28%), school (15%), and medical sources (6.5%).

Twenty-two percent of the participants knew or had heard of a family with a child with Tay-Sachs disease prior to the screening test. Carriers were significantly more likely to have a relative with the disease ($P < .001$).

Knowledge of Genetics of Tay-Sachs Disease

Some questions were asked to probe the participants' knowledge of genetics and Tay-Sachs disease, while others tested what the participants knew about the probabilities for having the disease or for being a carrier. It should be emphasized that the questions within each category are related, so that if one understands one question, he should be able to answer all in that category; however, a single respondent might perform well in one but indifferently in the other. The methods for grading the answers will be given as each question is considered.

Qualitative knowledge. The questions which tested qualitative knowledge are listed in table 3. Scoring was lenient; good answers were those which a person with a nodding acquaintance with biology would give. Fair answers showed some glimmering of understanding, however faint. The third category was "none." The response was mediocre; few respondents gave answers which indicated understanding. They did best on the question, "What is a Tay-Sachs carrier?"

TABLE 3

LEVELS OF QUALITATIVE KNOWLEDGE

	Good	Fair	None	Total
What is a Tay-Sachs carrier?				
Retrospective	133 (20.9)	364 (57.4)	138 (21.7)	635 (100.0)
Prospective	16 (15.4)	65 (62.5)	23 (22.1)	104 (100.0)
What is a Tay-Sachs gene?				
Retrospective	34 (5.4)	187 (29.4)	414 (65.2)	635 (100.0)
Prospective	0 (0.0)	31 (29.8)	73 (70.2)	104 (100.0)
What do genes do?				
Retrospective	21 (3.3)	503 (79.3)	110 (17.4)	635 (100.0)
Prospective	6 (5.8)	83 (79.8)	15 (14.4)	104 (100.0)

NOTE.—Nos. in parentheses = percentages.

Quantitative knowledge. The questions listed in tables 4 and 5 tested quantitative knowledge. All of the questions were directed to the participant's own experience rather than to abstract knowledge. For example, the question of the probability for having a Tay-Sachs child if both parents were carriers was not asked since there were no couples in which both were carriers, despite the fact that such abstract knowledge might have motivated the participants to come for the screening. The first three of these questions are not dependent upon the respondent's knowledge of his genotype or upon the composition of his family, but the remaining questions do depend on such information. Performance was generally poor, but that of the prospective respondents, who had been recently informed, was significantly superior to that of the retrospective group. An analysis of the responses to the quantitative questions in table 4 follows.

TABLE 4

LEVELS OF QUANTITATIVE KNOWLEDGE

	Good	Fair	None	Total
Disease frequency in Jews				
Retrospective	24 (3.8)	6 (0.9)	605 (95.3)	635 (100.0)
Prospective	20 (19.2)	17 (16.3)	67 (64.5)	104 (100.0)
Carrier frequency in Jews				
Retrospective	67 (10.6)	63 (9.9)	505 (79.5)	635 (100.0)
Prospective	59 (56.7)	5 (4.8)	40 (38.5)	104 (100.0)
Chance child will marry carrier				
Retrospective	59 (9.3)	65 (10.2)	511 (80.5)	635 (100.0)
Prospective	36 (34.6)	4 (3.8)	64 (61.6)	104 (100.0)
Chance sib is a carrier				
Retrospective	53 (11.7)	...	401 (88.3)	454 (100.0)
Prospective	12 (18.5)	...	53 (82.5)	65 (100.0)

NOTE.—Nos. in parentheses = percentages.

1. What is the frequency of Tay-Sachs disease in Jews? Only a minority of either group knew the odds which ought to be a factor in prompting preventive action. The question was graded as follows: 1 in 3,000–1 in 5,000 was "good"; any incidence bracketing the above and ranging between 1 in 1,000 and 1 in 100,000 was "fair"; and anything else was "none."

2. What is the carrier frequency in Jews? An answer of 1 in 20–1 in 50 was "good," any frequency bracketing the above and ranging between 1 in 10 and 1 in 200 was "fair," and anything else was "none." Carrier couples scored significantly better than noncarrier couples ($P < .02$), while the scores of the inconclusives and husbands of pregnant wives lay between these two groups.

3. What is the chance that your child might marry a carrier? This is a catch question intended to see if those who knew the carrier frequency could apply that knowledge. The carriers were most likely to know the answer, the inconclusives and husbands of pregnant women were next, and the noncarriers were the least likely to know. The difference between carriers and noncarriers was significant ($P < .02$). Apparently the identity of the two questions was evident to some respondents since among 137 persons who gave a numerical answer, whether right or wrong, 79.6% gave the same to both questions.

4. What is the chance of your sib being a carrier? This question was designed to test the way in which an individual would apply information about his genotype. Only subjects who had sibs answered, and responses were graded right or wrong. Among the retrospective couples, two subjects had step-sibs; there were others with inconclusive results whose genotypes were, in their minds, unresolved. In addition, 54 had sibs who had already been tested. For carriers, .5 was correct, for noncarriers the correct answer was 1 in 20–1 in 50. Of the 103 carriers who answered, 43 (41.7%) answered correctly, but only 10 (2.4%) of the remaining retrospective respondents gave correct answers. For the latter this is one-fourth to one-fifth the number who responded correctly to essentially the same question when it was posed in other words. There were no significant differences between noncarrier couples, "inconclusives," and husbands of pregnant women, although all of these differed from the carriers ($P < .001$).

Table 5 is concerned with questions directed to couples who knew the genotypes of both partners.

1. Can your child be a carrier? Answers were graded right or wrong. Those who perceived the issue posed in this question most clearly were the noncarriers who knew that they could not have a carrier child. The most confused were the spouses of the carriers who differed from the latter in their response ($P < .01$) as well as from all of the others ($P < .001$). The number of "inconclusives" who gave correct answers also fell between those of the noncarriers and the carrier spouses, differing significantly from both ($P < .001$ for both).

2. What is the chance your child will be a carrier? In answer to this question, eight of the "inconclusives" professed not to know their genotypes. Grading was as follows: .5 was correct for both members of carrier couples, and none was the correct answer for noncarrier couples. Other answers were graded as wrong. Again

TABLE 5

ANSWERS TO QUESTIONS INVOLVING KNOWLEDGE OF GENOTYPE

	CAN YOUR CHILD BE A CARRIER?			PROBABILITY YOUR CHILD CAN BE A CARRIER		
	Right	Wrong	Total	Right	Wrong	Total
Carriers	94 (74.0)	33 (26.0)	127*	55 (43.0)	73 (57.0)	128
Spouses of carriers	74 (58.3)	53 (41.7)	127	33 (25.8)	95 (74.2)	128
Noncarriers	197 (90.4)	21 (9.6)	218	118 (54.1)	100 (45.9)	218
Inconclusives	82 (78.8)	22 (21.2)	104	41 (39.4)	55 (60.6)	96
Prospectives	50 (66.7)	25 (33.3)	75	20 (36.3)	55 (63.7)	75
Total	497 (76.3)	154 (23.7)	651 (100.0)	267 (41.4)	378 (58.6)	645 (100.0)

NOTE.—Nos. in parentheses = percentages.
* One carrier couple did not answer.

the noncarriers performed best and the carrier spouses worst with the others in between.

A final question was, Is there any chance your child might have Tay-Sachs disease? Six hundred eighty (94.4%) of the respondents answered this correctly; four answered incorrectly; and 26 did not know. Of the 30 who gave incorrect answers or did not know, 18 were members of carrier couples. This means that about 7% of the carriers or their spouses have a lurking uncertainty about the outcome of a pregnancy.

Analysis of Quantitative and Qualitative Knowledge

The respondents' performance in response to the quantitative questions in table 4 was unrelated to age, education, income, number of present or desired children, knowledge of the chance of a sib to be a carrier, and belief in having learned more about genetics. Good performance was marginally related to believing one had learned more about Tay-Sachs disease ($P < .02$).

The correlation of quantitative knowledge between husband and wife is shown in table 6. Excluding the 185 couples both members of which knew nothing, the table shows a good deal of scatter suggesting that whatever else these couples discuss, they do not instruct one another in genetics.

Combined scores for quantitative and qualitative knowledge are shown in table 7. Scores for three of the quantitative questions, disease frequency, carrier frequency, and chance of child marrying a carrier, were combined to give each respondent a single score by giving a numerical equivalent to good, fair, or none for each question and then calculating a combined score for each participant. These scores were then divided into good, fair, poor, and none. A similar system was applied to the qualitative questions. To make statistical comparisons, the good and

TABLE 6

WITHIN COUPLE CORRELATION OF KNOWLEDGE

		HUSBAND			
		Good	Fair to Poor	None	Total
WIFE	Good	6	3	1	10
	Fair to poor	9	16	35	60
	None	12	56	185	253
	Total	27	75	221	323

fair scores have been combined into a category labeled "something" and the poor and none into "very little."

Table 8 illustrates the relationship between quantitative and qualitative knowledge. When the levels of qualitative and quantitative knowledge for each individual are correlated, the random scatter of performances shown in the table reveals that these two kinds of information are unrelated in the minds of these people.

The respondents were asked whether since being tested, they had learned more about Tay-Sachs disease and genetics. Since we are unaware of what the retro-

TABLE 7

COMBINED SCORES FOR QUANTITATIVE AND QUALITATIVE KNOWLEDGE

	Something	Very Little	Total
Retrospectives:			
Carriers	31 (24.1)	97 (75.8)	128
Spouses of carriers	24 (18.8)	104 (81.2)	128
Noncarriers	22 (10.1)	196 (89.9)	218
Inconclusives	18 (17.3)	86 (82.7)	104
Husbands	11 (19.3)	46 (80.7)	57
Prospectives	52 (50.0)	52 (50.0)	104
Total	158	581	739
		χ^2	P
Carriers vs. Spouses		0.8	>.05
Carriers vs. inconclusives + husbands		3.2	>.05
Carrier couples vs. noncarriers		13.8	<.001
Inconclusives + husbands vs. noncarriers		6.4	<.02, >.01
Carrier couples + inconclusives + husbands vs. noncarriers		12.8	<.001
Prospectives vs. all others		14.7	<.001

NOTE.—Nos. in parentheses = percentages.

TABLE 8

QUANTITATIVE VS. QUALITATIVE KNOWLEDGE

		QUANTITATIVE				
		None	Poor	Fair	Good	Total
QUALITATIVE	None	30 (4.7)	6 (0.9)	0 (0.0)	0 (0.0)	36
	Poor	242 (38.1)	67 (10.6)	31 (4.9)	2 (0.3)	342
	Fair	167 (26.2)	47 (7.4)	15 (2.4)	6 (0.9)	235
	Good	15 (2.4)	5 (0.8)	1 (0.2)	1 (0.2)	22
	Total ...	454 (71.4)	125 (19.7)	47 (7.5)	9 (1.4)	635

NOTE.—Nos. in parentheses = percentages.

spective subjects knew at the time of screening, we can learn only whether or not they *thought* they had increased their knowledge. About one-fourth of the retrospective respondents said they had learned something new about Tay-Sachs disease and about one-eighth about genetics. When asked about the sources of the information learned, most cited the media, meetings, and literature, with medical sources lagging here as elsewhere, especially as a source of learning in genetics (table 9). The response of the prospective subjects was similar; 14.6% said they had learned more about the disease and 17.3%, about genetics.

DISCUSSION

Both the retrospective and prospective groups consisted of well-educated young people who in general wanted more children, regularly used birth control, accepted modern indications for abortion, and knew that antenatal diagnosis represented a solution to the problem of Tay-Sachs disease.

The participants learned of Tay-Sachs screening by word of mouth, newspapers, or television. Only rarely did they learn about it from a physician, even though some pregnant women were receiving obstetrical care. Furthermore, they discussed

TABLE 9

SOURCES OF INFORMATION ABOUT TAY-SACHS DISEASE AND GENETICS AFTER SCREENING

Source	About Tay-Sachs Disease	About Genetics
Medical	38 (22.9)	31 (13.3)
Literature, meeings, school	43 (25.9)	40 (40.8)
Media	56 (33.8)	25 (35.5)
Contact with patient's families	11 (6.6)	3 (3.1)
Other	18 (10.8)	17 (17.3)
Total	166 (100.0)	98 (100.0)

NOTE.—Nos. in parentheses = percentages.

the issue principally with family and friends rather than physicians or rabbis, despite the medical and moral issues involved.

For the most part it was the women who heard about the screening first and were responsible for the subsequent testing. This is compatible with the conventional wisdom which allocates the supervision of family health to the wife. The participants recognized that the risk could be minimized as a consequence of screening, whichever way it turned out, but some came as a result of social pressures rather than any sense of risk or benefit.

These observations support the idea that people will usually accept preventive measures if they perceive the seriousness of the disorder and their susceptibility and that there are benefits to be had in accepting the proffered opportunity [3]. A test of this hypothesis was carried out by Becker et al. [4] who studied a different sample of the population from which this one was drawn. They showed that, in contrast to those who came forward to be tested, those who did not were less educated, older, and less likely to want more children. The latter also perceived themselves to be less susceptible and had a greater fear of the consequences of screening. An important question is how to offer this service in such a way as to make those perceptions closer to reality and the rejection of the test, if that option is preferred, more rational. One suggestion given by Becker consists of emphasis in the pre-screening education on the following: (1) the high probability among Jews of being a carrier; (2) the innocuousness of that condition; and (3) explanations of the measures which can assure carrier parents that subsequent pregnancies can result in unaffected infants.

Another answer to this question is implicit in the data presented here and elsewhere [5]. The participants learned of the opportunity and decided to be tested without the approval, or often even the knowledge, of any family doctor or other physician to whom one might turn naturally for counsel in regard to matters of health. Perhaps more people would avail themselves of this service if physicians were to advocate it.

Why have physicians stood aloof? There is evidence that physicians, especially graduates of more than 10 years, do not know much genetics and do not perceive its relevance to their work [6]. In addition, the low status given preventive medicine in medical school may help to give genetic screening a low priority. Finally, it has been shown that physicians interpret risk figures variably and that their actions are related directly to their own perception of the risk, not to its numerical value [6].

On the other hand, to recommend that a test be done to prevent a serious disease is not outside ordinary medical practice, so it is a plausible hypothesis that if screening for Tay-Sachs disease were actively championed by practicing physicians, more people would avail themselves of the service.

Physician advocacy also has the virtue of reducing the chance that a couple could be overlooked for lack of awareness. Some people do not read newspapers, watch television, participate in social activities at the synagogue, or discuss matters of this sort even with friends or relatives. But if screening for Tay-Sachs disease

were a part of primary medical care, the message would be more generally available. This suggests that the promotion of screening for Tay-Sachs disease, as well as for other conditions, must involve bringing the thinking of physicians into consonance with the aims of preventive medicine.

The participants were clearly aware of the dread nature of Tay-Sachs disease and of the vulnerability of Ashkenazi Jews in general, but their imperfect knowledge of the genetics of the disease and of their individual susceptibility deprived most of them of information which should be useful in deciding whether or not to be tested. These highly educated and aware people are capable of grasping anything they recognize as important, but evidently the value of genetic knowledge was not perceived. We are not suggesting that they ought to have had such perceptions, merely that they did not.

At least two reasons can be advanced to account for this lack. First, genetics is, to most people, out of context. If the respondents had been older, this would have been the result of lack of exposure since it is only in the last 20 years that genetics has become part of high school biology courses. Most people graduating from high school since the mid-1950s, however, had a course in biology in which the concepts of genetics were read or discussed, but genetics learned in high school may not be perceived as being related to life, and may be promptly forgotten.

Second, specific numerical odds are not often employed in the decisions of daily life. Rather there is a tendency to minimize high risks and to maximize low risks, except where personal experience or temperament lead one to feel lucky or pessimistic [7]. The perception of risks is also influenced by the gravity of the outcome; for example, a low risk will be taken more seriously if the outcome is particularly burdensome [8, 9]. But the principal element is the ability to discover meaning in the symbolism of odds; among human beings this is variable [7]. Indeed, it is a developmental quality; children are usually unable to deal with questions of odds until after 6 or 7 years of age [10]. Unfortunately, the degree to which adults differ in this property and the origins of those differences are not known, but variation does exist, so it is unrealistic to expect everyone to respond equally to educational information dealing with risks.

There is an additional inference to be drawn from the observed differences in knowledge between the categories of respondents. Among the retrospective couples, the carriers knew the most followed by the inconclusives and husbands of pregnant women; the noncarriers knew the least. Since the study is retrospective, this difference may be due to selective retention or learning after the fact. Indeed there is evidence for the latter, since significantly more carriers and spouses said that they had learned more about Tay-Sachs disease. Apparently a sense of personal involvement has been a spur to retaining, or to obtaining, knowledge. Some of the carriers who have discovered a disturbing aspect of their makeup are most inclined to do so, while the noncarriers, who have made no such discovery about themselves, see no reason for further concern. Some of the respondents with inconclusive results and some of the husbands of pregnant women also appear to have had an unsettling experience which stimulated them to remember or to learn more.

What, if anything, should be done? If the aim is to persuade all Jews to accept

screening, additional efforts to inform the community about the disease and its prevention might help to bring in more subjects. Eventually, in any case, if physicians become more generally involved, screening for Tay-Sachs is likely to become the rule rather than the exception. In that event, knowledge of genetics and probability would be only marginally helpful. But if the aim is an informed public capable of comprehending susceptibility and of making informed choices about preventive measures, then such knowledge is of paramount importance. The means of achieving this desirable state are not now at hand. We need first an informed medical profession to catalyze learning about preventive medicine in general and genetic screening in particular. Secondly, educators must accept responsibility for teaching science and biology in such a way that its relation to health is apparent to every child, and in this, medical educators should assume some leadership.

SUMMARY

A highly-educated, socially aware group of persons presented themselves for Tay-Sachs screening having learned about it mainly from friends, newspapers, radio, and television but not from physicians or rabbis. After learning that screening was possible and deciding that it is in principle a good idea, and after discussing it with relatives and friends but not with physicians and rabbis, they presented themselves for the test. Although the participants knew that Tay-Sachs is a serious disease and that Jews are vulnerable, few of them knew much about the genetics of the disease, its frequency, or the incidence of the carrier state.

This experience of screening for Tay-Sachs carriers suggests the need for physicians to learn the relation of genetics to preventive medicine, and for the public to learn more about the biology of man.

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