

Knowledge and Attitudes Toward Tay-Sachs Disease Among a College Student Population

CHERYL F. AUSTEIN, M.P.H.,** MARGRETTA R. SEASHORE, M.D.,^b
AND STEPHEN S. MICK, Ph.D.^c

^a*Program Analyst, Department of Health and Human Services, Office of the Assistant Secretary for Planning and Evaluation Washington, D.C.*; ^b*Associate Professor of Human Genetics and Pediatrics, Department of Human Genetics, Yale University School of Medicine, New Haven, Connecticut*; ^c*Associate Professor, Department of Epidemiology and Public Health, Yale University School of Medicine, New Haven, Connecticut*

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To assess the feasibility of screening the single Jewish population for Tay-Sachs disease (TSD), a questionnaire examining the knowledge of and attitudes toward TSD and genetic screening was sent to 348 Yale University Jewish undergraduates. Of those students responding (63 percent), 78 percent were able to answer general genetic questions correctly while only 1.9 percent could answer specific Tay-Sachs questions correctly. A majority of the students (93.9 percent) indicated some concern about being a carrier for TSD, believed that carrier status would affect future social and reproductive behavior, and expressed an interest in having TS carrier status determined while still single (77.4 percent). Strong correlations were found between knowledge and attitudes, but no significant differences appeared between male and female respondents. In addition to leading to improvements in Tay-Sachs screening programs, the observations have led to suggestions that may be generalized to other genetic screening programs.

INTRODUCTION

Screening for heterozygotes for detrimental mutations is becoming a feasible means to avoid the birth of children with severe, often lethal, genetic disorders. Because this ability may be coupled with the potential for causing harm to those screened, careful evaluation of screening programs and critical attention to their design is essential. Screening programs to detect the Tay-Sachs carrier state form the most extensive experience available to study, looking for ways to improve this kind of health care.

Until recently, Tay-Sachs disease (TSD) was untreatable and unpreventable. Although there is still no effective treatment, TSD can now be prevented primarily because of three major factors: (1) TSD occurs in a specific, well-defined population, (2) TS carriers can be detected by means of a simple blood test, and (3) TS can be diagnosed in the fetus *in utero*. In addition, the screened population can be offered positive reproductive alternatives if they are found to be at risk for having a TS child [1].

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*Conducted while an Associate in Research, Yale University School of Medicine, Department of Human Genetics

Address reprint requests to: Margretta R. Seashore, M.D., Department of Human Genetics, Yale University School of Medicine, 333 Cedar Street, Box 333, New Haven, CT 06510

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Preventing a disease by detecting carriers for a genetic trait presents several problems: who should be screened, how should potential recipients of screening be reached, and how can social and emotional consequences of knowledge of carrier status be prevented. In the case of TSD, large screening programs have been established in conjunction with educational programs to inform Jewish couples about TSD and to encourage them to be screened [1,2,3]. Before screening programs have been undertaken, therefore, an attempt has been made to reach all potential screenees through some form of education about TSD specifically, as well as about some basic genetic principles. Participation of these individuals has been found to be significantly related to attitudes regarding screening and future reproductive behavior among these couples [1,3]. In addition, psychosocial examinations of those participating in these screenings have indicated a better understanding of the genetic risk of individual and/or spouse carrier status among those who willingly participate in both education and screening programs [3].

Although it is now agreed that a couple's TS carrier status should be determined before starting a family, it is still undetermined whether pre-childbearing individuals should be screened while single, before marriage, or just before starting a family. Until recently, most TS screening programs have been directed toward married couples, only [1,2,3,4].

A large-scale TS screening and education program was conducted among high school students in Montreal [5]. The authors found some initial anxiety among carriers but indicated a favorable attitude toward the screening and education by this population. No program has singled out the college age student as a potential recipient of genetic screening, however, because of this possibility of psychosocial damage from carrier status knowledge and its impact on mate selection and reproductive behavior [6].

The present study was designed to investigate the possibility of screening single people for TSD by examining the impact that carrier status would have on these individuals. The study attempted to evaluate students' knowledge about genetics, in general, and TSD, in particular, by examining a specific college age population. In addition, questions were asked about the students' attitudes regarding the possibility of being a carrier for a genetic trait and its effect on their future childbearing behavior and mate selection. The interaction between the students' knowledge and attitudes was examined to detect any relationships that might exist between these two factors and any bearing that this might have on the psychosocial impact of genetic screening among single people.

In order for TS screening to be most effective, carrier status should be determined before the onset of pregnancy. The majority of college age students are not married and have not yet had any children. The college campus, itself, could provide an ideal setting for mass screenings. Finally, it is felt that college students have generally been exposed to some courses in genetics and probability, either in high school, in college, or through the popular news media, and this could be taken into consideration when planning education and screening programs.

METHODS

The population studied was selected from a list provided by the rabbi from the Yale University Hillel (a Jewish student organization). The total number of undergraduate Jewish students was estimated to be approximately 1,200 and it was decided that the study population would consist of one-third of this group. Names were chosen from alphabetized file cards categorized by the year of graduation. A

systematic sample of 348 students was selected and consisted of 117 females and 231 males.

Each student was sent a cover letter, a stamped return envelope, and a questionnaire that had been pre-tested on another Jewish sub-population from a neighboring college campus. The individuals were asked to return the questionnaire within two weeks. A telephone follow-up was initiated to encourage non-respondents to return their questionnaires. A second questionnaire was sent to those who had not been reached through the first mailing.

THE QUESTIONNAIRE

The self-administered questionnaire was designed to elicit three major kinds of information: socioeconomic data, students' knowledge about genetics and TSD, and students' attitudes toward TSD genetic screening. The knowledge questions required two specific types of information in order to answer them correctly: two of the questions could be answered correctly without any prior knowledge of TSD and two other questions could only be answered correctly with some exposure to TS information. The remaining knowledge questions referred to related information including the definition of amniocentesis and the source of TS knowledge. The attitude questions dealt with choices for future reproductive behavior if an individual and/or his mate were found to be a TS carrier. These questions were also directed toward the students' feelings about when and if people should be screened for a genetic trait and if they, themselves, would be interested in participating in a TS carrier screening program.

RESULTS

Socioeconomic Data

At the completion of the data collection phase, 219 or 63 percent of the students had returned their questionnaires. Of the males, 65.8 percent responded, and 57.3 percent of the females responded. No factors other than sex and year in college were known about the non-respondents. There was a decreasing rate of return with increasing year, but class and age were not significantly related to other variables.

One bias that was known to be associated with this particular population was the high socioeconomic background from which the individuals had come. No significant relationships were found between income and genetic knowledge although a negative relationship (χ^2 sig = 0.006) was found between income and TS knowledge. There is no obvious explanation for this relationship but it may be that TS information is closely related to parents' ethnic identity more than to their income or education. In some populations, it often appears that religious identity decreases with increasing education and income. The modal parental income was more than \$30,000 and the mean educational level of the parents was one year past college.

Over 50 percent ($N = 110$) of the students were Jewish Reform, 27.9 percent ($N = 60$) were Conservative, and 3.3 percent ($N = 7$) were Jewish Orthodox. More than 80 percent of the respondents knew that they were of Ashkenazi heritage. A majority of the students (81.5 percent) indicated that they were single. The remaining students were either engaged (0.9 percent), going steady (10.6 percent), or living with mate (6.9 percent).

Knowledge

Eight knowledge questions were included in the questionnaire (Table 1). No sig-

TABLE 1
Percentage Distribution of Genetic Knowledge and TSD Information by Sex

Knowledge (% correct)	Total Sample	Males	Females
1. Is TSD hereditary?	83.6	84.2	83.3
2. If both parents are carriers, can the child have the disease?	81.3	81.6	81.8
3. If one child has TSD, will the chances for the second child having TSD increase, decrease, or remain the same?	60.7	62.5	57.6
4. Amniocentesis is:	54.8	52.6	60.6
5. If one parent is a carrier, can the child have TSD?	40.6	41.4	39.4
6. The most serious result of TSD is:	38.8	39.5	37.9
7. 4,000 people, how many TS carriers?	15.5	18.4	9.1
8. 4,000 people, how many TS babies born?	15.1	16.4	12.1

nificant correlations were found to exist between sex and knowledge, but unlike previous studies of married couples where women were found to be more interested and better informed [3,4], our data revealed little difference in knowledge between males and females, suggesting that they are equally informed about TSD and genetics.

Table 1 also shows sharp differences in the frequency of correct answers among the knowledge questions. Question 1 above can be easily deduced from the general tone of the questionnaire. Questions 2 and 3 can only be correctly answered with some prior knowledge of genetics and probability. Question 4 does not necessarily require prior understanding of TSD. The last four questions (5–8), however, represent basic information generally presented in TS educational materials including pamphlets, popular new journals, and basic genetic textbooks.

In order to detect any significant relationships between these eight questions, factor analysis was performed on all knowledge questions. Factor analysis [8] is a statistical concept used to determine an underlying pattern of relationships that might exist among several variables. The variables are "rearranged" to a smaller set of factors that may be responsible for relationships in the data. Factor analysis generally involves: (1) the preparation of a correlation matrix, (2) an attempt to reduce data into initial factors, and (3) the development of interpretable factors through the use of rotation. Factor analysis, when applied to these questions, revealed two independent factors, each consisting of two questions. Factor I consisted of questions 2 and 4 and Factor II consisted of questions 5 and 6. These two factors were named "genetics" and "informed," respectively. The remaining questions, although related somewhat to these factors, were not strongly associated with any particular grouping. Because of the interrelationships among the questions discussed above, Factors I and II only were used in the analysis. Primarily, "genetics" dealt with general genetic screening information that need not be related to knowledge about TSD, whereas "informed" consisted of questions that referred directly to information about TS birth and carrier frequencies. Both factors, "genetics" and "informed," were computed to be the number of correct answers on the two questions that contributed to each category. Scores on both, therefore, ranged from 0 to 2, 2 being a

perfect score. This classification into two distinct categories produced statistically significant differences in level of knowledge.

Table 2 shows that although this sample population scored well on questions measuring basic genetic principles, it tested poorly on measures of TSD knowledge: the carrier and birth frequencies of TSD. In this single Jewish population, therefore, the level of genetic and probability knowledge far exceeds the level of TS knowledge. Basic knowledge of genetics and probability, therefore, did not necessarily imply understanding of TSD.

Table 3 shows the highly variable sources from which the students learned about TSD. Although a large proportion of the students had never learned about TSD, the news media and the school were significant sources of information. Not only were the news media the most common source of TS information, they were also the most effective. One hundred percent of those students scoring perfectly (2) on "informed" named the news media as their source of TS information. Finally, a serious void in the role of the medical community in transmitting TS information was revealed through the small number of students who named their doctor as their source of knowledge.

Attitudes

In the attitude questions, the students were asked to predict their reactions to learning of their carrier status and the effect that carrier status might have on future reproductive behavior. Although this is far from being an ideal means of evaluating reactions to being a carrier, it is the most effective method given the study design. Some critics [3] of college age screening have stated that the psychological handicap of knowing that one is a carrier for a deleterious gene may be a major area of concern for future social behavior. To determine how this age group might act and to learn how they would feel about screening among their own age group, we asked them how much being a carrier would matter to them, what they would do if they and/or their mate were found to be TS carriers, when they would tell a prospective mate about their carrier status, what they would do if they or their spouse were found to be carrying a TS fetus, what they would do if they had a TS child, and if they would be interested in attending a TS carrier screening.

When asked how much being a carrier for TS would matter to them, 49.2 percent ($N = 97$) of the students answered "a great deal" and 44.7 percent ($N = 88$) answered "somewhat." Only 6.1 percent ($N = 12$) answered "hardly at all." In previous studies of married couples [1,3,4], it was found that females were more interested in being screened and generally showed more concern for their future reproductive behavior. In the age group studied here, however, these large differences did not appear. Several factors could have contributed to these feelings including the new "lib-

TABLE 2
Percentage Distribution of Scores for Questions Representing Knowledge of
General Genetic Information and TSD Information

No. Correct	"Informed" (%)	"Genetics" (%)
None (0)	71.2	13.2
One (1)	26.9	8.7
Two (2)	1.9	78.1
Total	100.0 ($N = 219$)	100.0 ($N = 219$)

TABLE 3
Percentage Distribution of the Source of Tay-Sachs Information

Source of Knowledge (%)	Respondents
In school	13.0
From a friend	4.7
From a doctor	0.9
News media	29.3
Other	14.9
Never learned	37.2
Total	100.0 ($N = 215$)

eration” of women, a greater sense of responsibility for reproductive behavior among men, and the high educational levels of both the males and females in this population.

In order to determine the students’ views on the most appropriate time to be screened for TSD, they were asked when they thought one should be tested for TS carrier status. In response, 32.0 percent ($N = 57$) thought that screening should be done in high school, 21.3 percent ($N = 38$) in college, 33.7 percent ($N = 60$) premarital, 8.4 percent ($N = 15$) pre-childbearing, and 4.5 percent ($N = 8$) at other times including “as a child.” In relation to this question, the students were asked when they would tell a prospective mate about their TS carrier status: 8.8 percent ($N = 17$) would tell a prospective mate “while dating,” 66.3 percent ($N = 128$) would tell “before getting engaged,” 23.8 percent ($N = 46$) would tell “between engagement and marriage,” 0.5 percent ($N = 1$) would tell “after marriage,” and 0.5 percent ($N = 1$) would never tell a prospective mate.

When asked what effect *one* mate being a carrier for TSD would have on their future reproductive behavior, 17.6 percent ($N = 35$) of the respondents said it “would have no effect,” 2.0 percent ($N = 4$) said they “would want no children,” and 80.4 percent ($N = 160$) said they “would consult a family doctor and/or a genetic counselor.” If *both* mates were found to be carriers for TSD, 2.5 percent ($N = 5$) felt it “would have no effect,” 10.0 percent ($N = 20$) felt that they “would want no children” and 87.5 percent ($N = 175$) would “consult a family doctor and/or a genetic counselor.” The significance of these two questions will be discussed in the next section.

In the case where they or their mate were found to be carrying a TS fetus, 89.2 percent ($N = 140$) of the students said they would abort the fetus. Only 10.8 percent ($N = 17$) said they would allow the child to be born. In addition, 71.2 percent ($N = 108$) of the students would consider having another child if their first had TSD while 28.8 percent ($N = 45$) would not have any additional children.

Finally, when asked if they would be interested in attending a TS carrier screening program, 77.4 percent ($N = 150$) of the respondents said they would be interested in participating and 20.0 percent ($N = 39$) said they would not be interested. Only 2.6 percent ($N = 6$) of the respondents had been tested before. For those responding “no” interest in being screened, the most common reasons given included: “no one in my family has ever had it”; “I want to know what it is first”; “no time”; and “don’t believe I’m a carrier.”

Relationship Between Knowledge and Attitudes

In determining the relationships between knowledge and attitudes, the distinction between “genetic” knowledge and TS “informed” became significant. The data show

that 100 percent of those knowing nothing about genetics knew nothing about TSD and 100 percent of those getting perfect scores on “informed” obtained perfect scores on “genetics.” It might be hypothesized that knowledge about genetics would coincide with or precede knowledge about TSD. Knowledge about genetics, however, does not necessarily constitute knowledge about TSD, as we have shown. A student, for example, may be quite well educated in genetics and know nothing about TSD. The reverse could also be hypothesized: a person may have specific knowledge about TS and have little, if any, understanding of broader genetic principles. This most likely would have come from direct exposure to a TS pamphlet, a magazine, or a TV program with no regard for previous genetic knowledge. The knowledge that an individual could acquire in this way, however, would be superficial with little understanding of the applications of these facts to their own lives.

Since both knowledge about genetics and TSD are important in interpreting the risks associated with TS carrier status, both factors may be related to attitudes (Table 4). The correlation that appears in Table 4 is also found in several of the other attitude questions: as the amount of information about genetics and TSD increases, the degree of negative reaction toward being a possible carrier decreases.

Table 5 illustrates the importance of understanding the underlying genetic principle that distinguishes between one parent being a carrier and both parents being carriers for the TS gene: as the amount of genetic knowledge increases, the difference in responses between one parent being a carrier and both parents being carriers increases.

Another indication of future reproductive behavior is the response to the question asking how much being a carrier would matter (Table 6). Table 6A illustrates that the more concerned a person is about being a carrier, the earlier he would want to tell a prospective mate of his carrier status. In Table 6B, it is evident that the more concerned a person is about being a carrier, the less likely he is to have children if his first has TSD. Finally, Table 6C indicates that although a person might be concerned about being a carrier, he would still be interested in being screened.

DISCUSSION

Published studies conducted with married couples [2,4] have shown that exposure

TABLE 4
Percentage Distribution of Attitudes Toward Carrier Status by Scores Representing Genetic Knowledge and Tay-Sachs Information

	Carrier Matter?			Total
	A Great Deal	Somewhat	Hardly	
	“Genetics” Score (%)			
None (0)	64.7	29.4	5.9	100.0
One (1)	62.5	31.3	6.3	100.0
Two (2)	46.3	47.6	6.1	100.0
$\chi^2 = 3.516$ sig = .4753 4 d.o.f.				<i>N</i> = 197
	“Informed” Score			
None (0)	50.7	43.4	5.9	100.0
One (1)	47.4	45.6	7.0	100.0
Two (2)	25.0	75.0	0.0	100.0
$\chi^2 = 1.798$ sig = .7727 4 d.o.f.				<i>N</i> = 197

TABLE 5
Percentage Distribution of Attitudes Toward Future Reproductive Behavior if One or Both
Mates are Tay-Sachs Carriers by Scores Representing Genetic Knowledge

	You OR Mate Carriers			You AND Mate Carriers		
	No Effect	No Children	Consult Doctor	No Effect	No Children	Consult Doctor
	"Genetics" Score (%)					
None (0)	12.5	0.0	87.5	6.3	0.0	93.8
One (1)	5.3	0.0	94.7	5.3	15.8	79.0
Two (2)	19.5	2.4	78.1	1.8	10.3	87.9

$\chi^2 = 8.484$ sig = 0.3877 8 d.o.f. $N = 199$ $\chi^2 = 12.32$ sig = 0.1371 8 d.o.f.
 $N = 200$

to TS education before participation in a screening program may be related to several key factors corresponding to health perceptions. Those individuals not well informed about all aspects of their genetic screening were found to react with much more intensity and had more misconceptions about the disease for which they were screened. In addition, pre-screening education provided reasons for positive future reproductive behavior whenever possible, rather than a complete restriction on child-bearing behavior.

Our study suggests that attitudes are directly correlated with knowledge, as has been found in married populations. Education, therefore, appears to be a key factor

TABLE 6
Percentage Distribution of Attitudes Toward Carrier Status by Future Social,
Reproductive, and Screening Behavior

Behavior	Carrier Matter?		
	A Great Deal	Somewhat	Hardly
A. Tell Prospective Mate			
While dating or before engagement	83.4	68.6	60.0
Between engagement and marriage	16.7	30.2	30.0
After marriage	0.0	1.2	0.0
Never	0.0	0.0	10.0
$\chi^2 = 25.830$ sig = 0.0011 8 d.o.f.			$N = 186$
B. Consider Having More Children			
Yes	55.2	85.1	100.0
No	44.8	14.9	0.0
$\chi^2 = 18.986$ sig = 0.0001 2 d.o.f.			$N = 149$
C. Interested in Attending Screening			
Yes	85.9	80.5	22.2
No	14.9	19.5	77.8
$\chi^2 = 20.715$ sig = 0.0000 2 d.o.f.			$N = 176$

in predicting emotional outcomes from screening rather than marital status. Educating the single person, however, might involve additional assurances for a positive reproductive future.

In order to improve the delivery of educational programs, our distinction between the two types of knowledge becomes a potentially important factor. One cannot generalize about educational backgrounds when planning a TS education program. In this sample that had never been formally approached with TS information, for example, only 37 percent of the students stated that they had never learned about TSD, yet less than 2 percent of the students scored perfectly on "informed." As shown, this population appeared to understand basic genetic principles, yet they were not well informed about TSD. One cannot assume, therefore, that the understanding of basic genetics directly leads to the understanding of TS birth and carrier frequencies. The data did show, however, that those who scored well on basic genetic principles tended to be better informed about TSD. One can only suppose that although more than 2 percent of the students had been exposed to TS information, only those who had a good understanding of genetics and were exposed to TS information retained the knowledge that was needed to complete the TS portion of the questionnaire. In planning an educational program, therefore, a careful examination of the backgrounds of potential screenees should include an evaluation of how much information they have in both of these categories.

Several other important problems were revealed from the data, however, including the low level of information that doctors are providing about TSD. Less than 2 percent of the students named their doctor as their source of TS information, yet the majority of them would seek their doctor's advice regarding carrier status and future reproductive behavior. Lowden [7] has reported from Canada that only 31 percent of physicians whose practices served a largely Jewish population advised Tay-Sachs screening, although 76 percent had been asked questions about such screening. Until this void in communication can be overcome, educational programs directed at the medical community as well as the general population must be undertaken to insure positive attitudes toward genetic screening among those being screened and their peers.

In conclusion, then, this study revealed the following correlations regarding knowledge and attitudes of college students toward TSD and genetic screening:

1. Although a group may be well educated about basic genetic principles, it is not necessarily well informed about simple TS birth and carrier frequencies as well as the other concepts associated with TS education.
2. Students show some anxiety about being carriers for TSD but they also tend to choose positive alternatives for possible reproductive behavior.
3. Students feel that screening for TS carriers should be done when an individual is still single and would choose to tell a prospective mate about their carrier status before marriage.
4. The response to a question concerning anxiety about being a carrier is a good predictive measure for other attitudes, particularly toward future reproductive behavior.
5. The amount of background information a person has concerning both TSD and genetics is positively correlated with attitudes and anxiety. As information increases, anxiety about carrier status and future reproductive behavior decreases. Some anxiety is necessary, however, as a motivating force for screening.
6. The medical community must be informed and encouraged to disperse effective information and guidance regarding genetic diseases.

Should single persons be screened for TS carrier status and other genetic conditions, then? The results from this study show that the group studied displayed an eagerness to participate, good educational backgrounds, and a strong relationship between TS information and attitudes among this age group. Although these students are concerned about learning of their carrier status and future reproductive behavior, the data show a marked decrease in concern with increasing information regarding carrier and birth frequencies of TSD, and it can be assumed that because of their high level of genetic knowledge, they could be more easily taught about TSD and its genetic implications. In addition, the majority of these students would desire screening while still single or directly before marrying, and indications were that carrier status would not influence mate selection. Finally, using the college campus as a screening site could also improve efficiency of delivery. The population is well defined, there is usually a Jewish student organization for assistance, and education and screening programs can be adapted for each college population.

The implications from this study can be applied to other screening programs, as well. First, the importance and complexities of knowledge should be considered when planning an educational program and the importance of screening as a part of preventive medicine must be stressed to both lay and professional populations. The development of health attitudes, in general, is dependent upon factors including the level of anxiety, knowledge and information, and partially related to socioeconomic backgrounds. When all these factors are considered, therefore, an effective and successful screening program can be designed for most populations at risk, including single people.

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