

# A Survey To Evaluate Parental Consent As Public Policy for Neonatal Screening

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**Abstract:** Most states currently have laws which result in compulsory neonatal screening practices, despite a widespread consensus that participation in genetic services and programs should be voluntary. In 1976, Maryland adopted a regulation designed to respect parents' rights to refuse neonatal screening by imposing a parental consent requirement. The results of a study designed to evaluate the effects of this regulation are reviewed here.

Many health care providers were unaware of the parental consent regulation. However, hospitals were

generally in compliance with the technical stipulations of the regulations. There was little evidence that the regulation resulted in additional costs to the health care system, either in terms of hospital staff time or in terms of loss of efficiency in the number of infants screened. Mothers affected by the regulation were largely in favor of being informed about neonatal screening and learned a significant amount of new information from the disclosure process. They were almost evenly divided on whether they favored parental consent. (*Am J Public Health* 1982; 72:1347-1352.)

## Introduction

Mass newborn screening for phenylketonuria (PKU) and other hereditary disorders is a major public health program in the prevention of mental retardation. The cost-effectiveness of such in-hospital screening was established for PKU in the early 1970s,<sup>1-3</sup> and PKU screening is mandated by law in most states.\* Although several of these states provide that screening will not be performed if the parent

objects and many permit parental objection on religious grounds, the screening is nonetheless compulsory for all practical purposes.<sup>5</sup> That is, the exigencies of the situation are such that there is usually no opportunity for parents to voice objections.\*\* For example, the heel prick is routinely drawn in the nursery, not in the presence of the parents, and is done without the parents' awareness.

Despite this pervasive, routinely accepted practice, the overwhelming conviction in the literature on genetic policy is that compulsory genetic programs are inappropriate. For example, it is the position of the Research Group on Ethical, Social and Legal Issues in Genetic Counseling and Genetic

\*In the case of PKU, mental retardation can usually be prevented by placing afflicted infants on low phenylalanine diets, with best results achieved when the diet restriction begins as early in life as possible. Detection of all infants at risk requires biochemical screening of all newborn babies. The blood sample required for screening is obtained by a heel prick which most experts agree is as low risk a medical intervention as is currently available. However, osteomyelitis has been reported as a rare complication associated with repeated

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heel punctures: routine screening seldom involves more than one.<sup>4</sup> Nevertheless, parents in Maryland are now informed of this risk. To the knowledge of the Health Department, however, this complication has never occurred in the State. Thus it would be difficult to argue, particularly as osteomyelitis is treatable and should result in little or no permanent disability, that this risk is more than negligible in routine screening of healthy neonates.

\*\*At the time the National Academy of Sciences' Committee for the Study of Inborn Errors of Metabolism was preparing their report,<sup>5</sup> PKU screening was mandated by law in 43 states. Parental objection on any grounds was permitted in five of these states; on religious grounds in 30 states. However, this Committee found little evidence that parents had an opportunity to exercise their right of refusal regarding PKU screening. In its report, the Committee recommended that parents be made aware of their right to refuse testing at an early enough time to exercise that right. This report did note, however, that there would probably be no constitutional barriers to compulsory newborn screening (p. 93).

Engineering of the Hastings Center that "there is currently no public health justification for mandatory screening for the prevention of genetic disease."<sup>6\*\*\*</sup> This position is also the federal policy. The Genetic Diseases Title of Public Law 94-278, which provides assistance in the establishment and operation of genetic testing and counseling programs, requires that the "participation by an individual in any program or portion thereof under this part shall be wholly voluntary."<sup>7</sup>

In accord with this position, a regulation was issued in Maryland in 1976 requiring prior parental consent for newborn screening for PKU and other hereditary metabolic disorders:

"Before the administration of the test, the parent or guardian shall be informed fully of the reasons for the test and of his or her legal right to refuse to have the test performed on the child. An individual who has been provided and has signed a written explanation of the test approved and furnished by the Department, shall be considered fully informed."<sup>8</sup>

This regulation was promulgated by the Maryland Commission on Hereditary Disorders, an advisory body which was created by the Maryland legislature in 1973 to assure adequate public involvement in the development of genetic policies and programs.<sup>9</sup> Although there was opposition to the regulation, the Commission's response was to endorse the study reported in this paper before reconsidering the regulation. The study was organized around four important policy considerations: 1) the extent of health care providers' awareness of and hospitals' technical compliance with the regulation; 2) the reactions of providers; 3) costs of the regulation; and 4) the extent to which the regulation's objective—to promote meaningful parental choice—was achieved. The results reviewed here support the thesis that a parental consent requirement is a feasible institutional practice.

## Methods

The study was conducted in two phases. In Phase I, health care providers who were involved in implementing the parental consent regulation were interviewed. In Phase II, women from whom consent had been solicited under the regulation were interviewed.

### Phase I

All hospitals in the state of Maryland with active obstetrical units (N = 39) were included in the sample. The target population included chiefs of obstetrics and pediatrics, hospital administrators, administrative level nurses, and those individual staff nurses who were actually responsible for obtaining consent.

Hospital administrators, directors of nursing, and maternal-child health service chiefs were interviewed initially over the telephone in February and March of 1978 in order to

\*\*\*It should be noted that this research group did not focus specifically on newborn screening.

evaluate their awareness of the PKU parental consent regulation. Successfully completed telephone interviews were obtained from 92 per cent of pediatricians (N = 36) 90 per cent of obstetricians (N = 35), 80 per cent of nurse administrators (N = 34), and 90 per cent of hospital administrators (N = 35).

Within two months of the initial telephone interview, providers were interviewed again. This interview was administered over the telephone to physicians, and in person to hospital and nursing administrators. Whenever possible, those nursing staff actually administering the disclosure form to parents were also interviewed in person. If staff was not available, the requisite number of interviews in questionnaire form were left, along with stamped addressed envelopes, with the administrative level nurse. Sufficient questionnaires were left to allow participation of 100 per cent of the eligible staff at any given site. The second interviews were successfully completed for 74 per cent of obstetricians (N = 29), 95 per cent of pediatricians (N = 37), 92 per cent of hospital administrators (N = 36), 95 per cent of nursing administrators (N = 37), and 66 per cent of those staff nurses with actual responsibility for obtaining consent (N = 344). The interview assessed providers' attitudes toward the parental consent requirements, providers' perceptions of the cost to the institution of complying with the consent regulation, and a description of the process and procedures of obtaining consent.

### Phase II

In Phase II of the study, women were interviewed on obstetrical units in seven Maryland hospitals following delivery. The methodology of Phase II has been described in detail elsewhere and will only be summarized here.<sup>‡</sup>

Women delivering at each of the seven participating hospitals on any given day were randomly assigned to an experimental or control group at a ratio of two to one. Controls were interviewed before they received the disclosure form, experimentals were interviewed after receiving the form and giving consent to the test. A separate consent was required for participation in the study and all women approached agreed to be interviewed. A total of 210 women received predischarge interviews and 418 were interviewed after they gave consent.

There were no significant differences in the demographic characteristics of women assigned to the control and experimental conditions ( $p < .05$ ). The mean age of respondents was 25. Eighty-two per cent had completed 12th grade and 77 per cent were White.

‡See, for example, Final Report: DHEW Grant No. MC-R-240405-02-0, September, 1980; "Feasibility and effectiveness of obtaining consent for neonatal screening"; and Chwalow AJ, *et al*: "Informed consent: the impact of written vs. verbal consent on parents' attitudes and their decision to agree to neonatal screening" (in preparation). This latter paper reports the results of an experimental manipulation imbedded within this design in which women were randomly assigned to give oral consent or written consent. There were no significant differences between the oral and written groups on the variables reported here.

TABLE 1—Providers' Attitudes toward Parental Consent and Notification for PKU Screening: Relative Frequencies (Per Cent)

	Chiefs of Obstetrics	Chiefs of Pediatrics	Administrators	Adminis- trative Nurses	Floor Nurses	Total
Should parental consent be obtained for PKU screening?						
Yes (%)	31.3	21.6	40.0	45.9	35.4	129
No (%)	68.8	78.4	60.0	54.1	64.6	238
N	32	37	35	37	226	367
$X_4^2 = 7.63, P > .05$						
For respondents who reject parent consent, should parents be told about the PKU screening test before it is done?						
Yes (%)	90.9	85.7	71.4	80.0	65.7	167
No (%)	9.1	14.3	28.6	20.0	34.3	64
N	22	28	21	20	140	231
$X_4^2 = 11.58, P < .05$						

## Results

### Provider Awareness and Hospital Technical Compliance

The first question asked of providers in the initial telephone interview was "Are you aware of any state of Maryland regulation requiring parental informed consent to any procedures on newborns?" Sixty-nine per cent of chiefs of pediatrics and 53 per cent of nursing administrators indicated that they were aware of such a regulation and about two-thirds of these correctly recalled that PKU screening was the procedure implicated. Only 30 per cent of chiefs of obstetrics and 29 percent of hospital administrators reported being aware of any such regulation.††

A multiple regression analysis was conducted in an attempt to identify factors that might be associated with awareness of regulation; if chiefs of pediatrics were self-employed, they were significantly less likely to be aware of the regulation ( $p < .05$ ). For all respondents, no significant associations with demographic or hospital characteristics were identified.

Subsequent analyses, using data from the second interview, revealed that those who became aware of the regulation by receiving a letter sent by the Maryland Department of Health and Mental Hygiene (which was the official means of informing) were significantly more likely to be able to correctly explain the regulation during the telephone interview than those who attributed awareness to alternative channels.

Hospitals were generally in compliance with the technical stipulations of the regulation. Each of the 38 hospitals visited during Phase I was found to be using the Maryland State Department of Health and Mental Hygiene disclosure

††Study respondents had not as yet been informed of the specific focus of this research project on parental consent for PKU screening; not surprisingly, correct responses to this question were, therefore, low.

form. Moreover, based on reports of administrators and patient care staff, most hospitals were complying with the wording of the regulation that requires that the PKU test be performed "as close to discharge as possible."

### Reactions of Providers

The majority of providers disapproved of the state regulation requiring parental consent prior to PKU screening. Specifically, 69 per cent of chiefs of obstetrics, 78 per cent of chiefs of pediatrics, 64 per cent of hospital administrators, 51 per cent of floor nurses, and 35 per cent of directors of nursing indicated that they disapproved of the PKU regulation.

Providers' attitudes toward parental consent for PKU screening was assessed a second time in a question that was embedded in a series of items eliciting respondents' opinions concerning whether parental consent ought to be obtained for 16 newborn procedures. This item did not specifically mention the Maryland State regulation. Results were similar (Table 1).

Those providers who indicated that parental consent ought *not* to be obtained for PKU (238 of the original 367) were asked whether they thought that parents ought to be told about the test before the procedure is done. With the exception of staff nurses (who provided the information to patients), the overwhelming majority of respondents who rejected prior parental consent favored informing the parent that the PKU test will be done (Table 1). Although Light Margolin categorical analysis of variance revealed a statistically significant difference between staff nurses and other providers ( $X_{LM}^2 = 7.65, p < .05$ ), this difference accounted for only 3 per cent of the variance in responses.

Multiple regression analyses were performed in an attempt to identify factors predictive of providers' attitudes toward parental consent for PKU screening. There were no consistent patterns associating either hospital statistics, the demographic characteristics of the providers, or providers'

perceptions of their patients with their position on parental consent for PKU screening.

### Costs of the Regulation

*Staff Time*—Chiefs of pediatrics and staff nurses were asked their perceptions of the institutional costs involved in complying with the PKU regulation. In general, the costs of compliance in terms of staff time were negligible. Over 90 per cent of respondents indicated that their hospital had not had to make any adjustments in its staffing patterns in order to comply with the parental consent regulation. Most nurses (66 per cent) responded that obtaining consent or refusal took from one to five minutes. Another 22 per cent indicated that they spent between six and ten minutes obtaining consent.

*Numbers of Babies Screened*—Based on Health Department records for the period July 1, 1978–June 30, 1979, 0.05 per cent (27 of approximately 50,000 mothers) refused neonatal screening.

Although hospitals were required to inform the Health Department of refusals, it is possible that some failed to do so. Comparison of the percentage of infants screened in this period with the equivalent 12-month period in the year preceding the regulation failed to identify any reduction in the proportion of infants screened. Thus far, there have been no reported cases of untreated PKU attributable to parental refusal.

*Why Mothers Refuse*—An effort was made to interview the 27 women who had refused screening. Many of these women could not be located and only seven agreed to be interviewed. There is no way of determining how representative these seven are of the entire group of refusers.†††

The refusers were interviewed four to 14 months after they delivered (mean =  $7.3 \pm 3.6$  months). Each of the seven refusers remembered having refused and indicated that they preferred the current procedure to one in which screening would be performed routinely without provision for parental refusal.

The reasons for refusal given by five of the seven refusers indicated a poor understanding of screening. Two of these mothers did not speak English and their husbands responded to the interviewer. A third mother said that her husband did not want the test as his baby appeared healthier than many others. A fourth mother refused because her baby was already undergoing what she thought were overlapping diagnostic procedures, while the fifth mother felt her baby was all right and did not need the test.

The remaining two mothers refused because they were breastfeeding and had heard that the test would not be helpful if performed when the baby was just beginning to nurse. These two women said their babies were tested after

†††The mean age of the refusers was 28.9 ( $\pm 7.9$ ) years. The mean age of 14 controls matched (2:1) to the refusers for hospitalization for delivery was 26.9 years. Six of the seven refusers (85.7 per cent) and 13 of the 14 controls (92.9 per cent) completed the 12th grade, but only one of the refusers compared to nine of the controls had any college. The major household wage earner was professional or managerial in only one of the refusers (14.3 per cent), but in five of the controls.

discharge. One of the other seven refusers said her baby had been tested in the pediatrician's office after the doctor explained the procedure to her in her native language. A check by the Maryland Department of Health and Mental Hygiene of specimens received after discharge indicated that the infant of one other refuser was tested.

### Objective of the Regulation—Meaningful Parental Choice

*Mothers' Knowledge*—All of the hospitals in this study were obtaining consent through a standard disclosure form provided by the state. This form had been evaluated at a 10th grade reading level as measured by the Flesh scale of readability.<sup>10</sup> Women's comprehension of the information on the form has been reported in detail elsewhere.\* In general, we found strong evidence that women had learned substantial amounts of information from the form. Prior to receiving the disclosure form, only 53 per cent of control women reported that they had heard of a screening test for diseases which cause retardation, and only 52 per cent reported they had ever heard of PKU or related diseases. Of those who had heard about PKU screening, only 55 per cent knew without prompting that the test involved a heel prick and only 38 per cent knew that the major problem caused by PKU is mental retardation. By contrast, 77 per cent of the women interviewed after receiving the disclosure form knew without prompting that PKU screening was done by a heel prick; with prompting (through a multiple choice knowledge item) this figure jumped to 92 per cent. Sixty-five per cent of experimental women were able to spontaneously recall that the major problem caused by PKU is mental retardation, and 66 per cent spontaneously identified at least one of the diseases for which their babies would be screened.

*Mothers' Attitudes toward Parental Consent*—There were no significant differences in the attitudes of control and experimental women towards parental consent for PKU screening. Forty-six per cent of all women said they preferred to have the screening test done routinely without their consent. Reasons given by those favoring this option, tantamount to compulsory screening, are shown in Table 2. At least 80 per cent of women said that consent was not necessary because the test was simple and not dangerous. Of those women who preferred that their permission be sought, the overwhelming reason was that they wanted to know everything that would be done to their babies. Less than 5 per cent of answers reflected a desire for a right to refuse the test (Table 2).

Even if screening was compulsory, the vast majority, 82 per cent of controls and 78 per cent of experimentals, said that they would want to be told about the test before it was done. Only 3.5 per cent of women indicated that in the case of compulsory screening they would not want to be told about the test unless the results were positive.

\*See Holtzman NA, et al: "The effect of informed consent on mothers' knowledge of newborn screening and their attitude towards consent" (in preparation). This paper reports the results of our analysis of correlates of knowledge and attitudes, including hospital characteristics, maternal age and income, and timing of consent solicitation.

TABLE 2—Reasons for Favoring Mandatory Screening or Parental Consent

	Group	
	Pre-Disclosure	Post-Consent
Why would you prefer to have the second test done routinely without your permission being asked?		
N	90	170
Not necessary—Simple test (%)	82.2	81.7
So parents won't worry needlessly (%)	4.4	3.5
Provider's responsibility (%)	6.7	7.1
Other (%)	6.7	7.7
Why would you prefer that your permission be asked before screening?		
N	94	185
Want to know everything (%)	78.7	78.9
Tests need consent (%)	10.6	12.4
Want the right to refuse (%)	5.3	4.8
Other (%)	5.4	3.9

*Awareness of the Consent Process*—An attempt was made to determine whether experimental women understood the significance of their having been asked to consent to neonatal screening. Only seven of the 418 women held the false belief that the test would be done even if they refused permission. However, 12 per cent reported that they did not know they could have refused the screening test.

### Discussion

This project represents a relatively new approach to policy analysis. Although evaluation studies and technology assessments are common, empirical research is generally not directed toward the ethical issues raised by a public health policy.\*\* Empirical research, however, can make a significant contribution to moral analysis of health policies. It is common wisdom that good ethics demands good facts. It is often the case that moral disagreements about a proposed policy rest on important empirical assumptions about the consequences of the policy or about the nature of the relationships and obligations affected by the policy. Sometimes these moral disagreements can be resolved as data become available that bear on the contested empirical assumptions.

In the case of the Maryland parental consent regulation, initial opposition to the regulation rested largely on two questionable empirical assumptions: that the PKU program would cease to be cost-effective and that the objective of the regulation—meaningful parental choice—would not be achieved.

This study was specifically designed to test these assumptions. There was no evidence that the parental consent regulation had a negative effect on the public's health. The

chance of missing a PKU infant at the observed rate of parental refusal (0.05 per cent) is 100 times less than the chance of missing a PKU infant because of a false negative test result. There was also no evidence that the program had become less cost-effective because of increased costs to the health care system. Although health care providers continued to oppose the regulation, there was evidence of technical compliance without compromise of other required postpartum services. On the other hand, there was substantial evidence that the disclosure and consent process improved mothers' knowledge about screening and was, in general, well received by parents. Regardless of whether they had participated in the disclosure process, mothers were almost equally divided between those who favored consent before screening and those who did not. However, 79 per cent of all women wanted to be informed that their baby would be tested, even if screening was compulsory. When these data were presented to the Maryland Commission on Hereditary Disorders in 1980, the Commission determined to continue with the parental consent requirement. Although there are limits to the extent to which moral disputes about public policies can be resolved by empirical research,<sup>13</sup> in this case the Maryland Commission interpreted the data as eliminating the major moral and policy objections to the parental consent regulation.

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\*\*Empirical research about ethical and legal issues is increasing. Much of this research has focused on informed consent.<sup>11,12</sup>

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