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IMPACT OF FALSE-POSITIVE NEWBORN METABOLIC SCREENING RESULTS ON EARLY HEALTH CARE UTILIZATION

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Abstract

Purpose—To analyze the association between false-positive newborn screening results and health care utilization.

Methods—We surveyed parents regarding their children's health care utilization. Parents of children who received false-positive newborn screening results were primarily enrolled by a screening laboratory in Pennsylvania. Parents of children with normal results were recruited through the Massachusetts birth registry. We used bivariate tests and multivariate regression to assess the association between newborn screening results and primary care utilization, emergency room use, and hospitalization by age six months.

Results—Our sample included 200 children with false-positive results and 137 with normal results. Variation in recruitment strategies led to sample children with false-positive results being more likely to be non-white, have unmarried parents and be of lower socioeconomic status. After adjusting for significant covariates, such as age, race and socioeconomic status, there were no significant associations between newborn screening results and child health care utilization.

Conclusions—Despite the reported negative psychosocial effects of false-positive results, our study found no impact on early health care utilization. These results may assist in economic analyses of newborn screening as they suggest that medical costs associated with false-positive results are limited to the cost of diagnostic testing and follow-up.

Keywords

health services utilization; neonatal screening; false positive results; metabolic diseases; genetic diseases

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INTRODUCTION

Over the past decade, newborn screening programs for metabolic disorders have grown rapidly, in large part due to increasing use of tandem mass spectrometry (MS/MS). Between 1995 and 2005 the average state added 19 disorders to its newborn screening program.[1] Despite the high sensitivity and specificity of screening with MS/MS,[2] expanded screening, such as that recommended by the American College of Medical Genetics Newborn Screening Expert Group,[3] will lead to an increased number of infants with false-positive results due to the low positive predictive values inherent when screening for multiple independent diseases of low prevalence. Estimates for the annual number of false-positive results in the United States range from 2500 to greater than 51,000.[1] Economic analyses of newborn screening find expanded screening with MS/MS to be cost-effective, in part because, once a program owns a tandem mass spectrometer, screening for additional diseases using MS/MS adds very little incremental cost.[4–7] However, these studies generally assume the cost of a false-positive result to be only the cost of confirmatory testing and care during the confirmatory period.

The true impact of false-positive newborn screening tests is just beginning to be well described. Early screening programs for phenylketonuria (PKU) showed poor parental understanding of false-positive results and a tendency for parents of such children to perceive their children as medically vulnerable.[8] More recent research has demonstrated associations between false-positive results and increased parental stress, mothers' perceptions that their children with false-positive results require increased parental care, and a trend towards increased hospitalization.[9,10] Long-lasting psychological stress has been seen in families of children with false-positive screening tests.[11,12] Studies such as these have led to speculation that false-positive results may lead to increased perceptions of child vulnerability,[1,13,14] which have been associated with increased health care utilization in other childhood conditions.[15–18] Indeed, early work suggested increased hospitalization is seen among children with false-positive newborn screen results;[10] however the associations between false-positive newborn screening results and health care utilization have not been well described. In this study we hypothesized that a false-positive result would be associated with increased outpatient and inpatient health care utilization in early childhood.

MATERIALS AND METHODS

Sample

Parents of children with false-positive newborn screen results, defined as the initial result being abnormal or inconclusive for any of 20 biochemical disorders, were invited to participate after a referral was made for additional confirmatory testing. The authors planned to recruit all subjects from within New England but were unable to access information for New England children with false-positive results who did not present for evaluation at a metabolic center. Therefore, we contracted with a private laboratory which conducts newborn screening for more than 99% of birthing hospitals in Pennsylvania, a state demographically similar to Massachusetts,[19] to recruit and interview parents of children with false-positive newborn screen results. Participants were enrolled by this laboratory and by clinical metabolic centers in New England, between 1999 and 2005. In Pennsylvania, recruitment materials were sent to the families of all known cases of false-positive newborn screen results. In New England, recruitment materials were sent to families who sought follow-up testing at one of the clinical metabolic centers. Parents were sent a letter explaining the study ≥6 months after the diagnosis of a metabolic disorder had been ruled out. Those who did not opt-out by returning a response card were called to participate in a telephone interview.

The children with normal test results, confirmed by parent self-report, were 6- to 12-month-old children selected randomly from the Commonwealth of Massachusetts Department of

Public Health Birth Registry of Vital Records and Statistics for live births between February 1999 and September 2005. These birth registry data include only children born to married couples.[10] Families were recruited by letter, followed by a telephone call if an opt-out card was not returned.

All participants were sent a small baby gift, worth $7.00, after completing the interview.

Exclusion criteria for both groups were children who had died and newborns whose birth weight was less than 2500 grams. The latter criterion avoided recruitment of significantly premature infants who frequently experience transient newborn screening abnormalities. Although both parents were invited to participate in the survey, for this study we utilized only data collected from mothers since when there were data from only one parent it was the mother in almost all cases. The institutional review boards of Children's Hospital Boston and The Children's Hospital of Philadelphia approved the study.

Data Collection

We conducted a one-time 30-minute telephone survey. Complete details of data collection have been reported previously.[9,10,20] We asked mothers "in the past year, how many times did you visit a primary care physician for your child's care" and analyzed these data as a count of the number of visits. Mothers were also asked "whether [they] had ever taken [their] child to the emergency room" and the dates of such visits. These data were dichotomized as children who had or had not been to the emergency room. Mothers provided the dates of all overnight hospitalizations, which allowed us to classify children according to whether or not they were hospitalized in the first 6 months of life. Finally, we asked for the number of times the child had visited a specialist, exclusive of any visits associated with confirming the false-positive screening result.

We collected data on sociodemographic characteristics including parental education and occupation (which makeup the Hollingshead Index of Social Position[21]), and marital status, which we dichotomized as married or unmarried. Parents were also asked if their "child has any health problems" and to name the type of problem. Additionally, participants completed the Parental Stress Index (PSI), a 36-item questionnaire from which we utilized the total stress score.[9,10,22]

Data Analysis

We initially conducted bivariate analyses to compare health care utilization in children with false-positive newborn screen results versus normal screen results. We used non-parametric tests (Kolmogorov-Smirnov test[23]) to assess the associations between newborn screen result and both primary care and specialist use. We employed chi-square tests to assess the proportions who were hospitalized in the first 6 months and who had ever been to the emergency room. We used SAS 9.1 (SAS Institute Inc., Cary, NC) for all statistical analyses.

We then developed regression models for each dependent variable, using confounding covariates, defined as those variables (such as child age, gender and race, Hollingshead Social Position or maternal marital status) which showed a bivariate relationship with that dependent variable and with screening result at $p \leq 0.2$. We did not include maternal education in any model because the Hollingshead Social Position incorporates maternal education. Additionally, because we could not verify the reported health problems, this variable was used only in sensitivity analyses, so as not to control for parent perceptions of vulnerability that may influence reporting of both child health problems and child health care utilization. Poisson regression was used for the association between newborn screen results and primary care

utilization. We used multivariate logistic regression models to assess the relationship between newborn screen results and emergency room use or hospital admission.

Children who were missing a variable used in an analysis were excluded from that particular analysis. The most frequently missing variable was Hollingshead Index of Social Position, missing for 4 (2%) children with false-positive results and 0 children with normal newborn screening results.

We performed three different sensitivity analyses. In the first we assumed that all children who were not yet 6 months at the time of interview were hospitalized prior to their 6 month birthday. In the second we eliminated all unmarried mothers from the false-positive cohort and reassessed all associations between the dependent variables and newborn screening results. We conducted these first two sensitivity analyses to help determine the influence of the variation in our recruitment strategy between the children with normal and the children with false-positive results. Finally, we re-analyzed all multivariate regressions with an additional covariate indicating whether or not the child's mother reported the child had any health problems, in order to account for any influence true disease may have had on health care utilization.

RESULTS

Participants

The final sample included 200 children with a false-positive newborn screen and 137 with a normal newborn screen. The response rates were 75% among parents of children with false-positive results and 63% among parents of children with normal results. 87.9% of participants with false-positive results were from Pennsylvania (Table 1).

The children in the false-positive group had a mean age of 12.9 months compared to 6.4 months for the normal screen group. Children in our sample with false-positive screens were more likely than children with normal screens to be non-white, have unmarried parents, come from families of lower socioeconomic status and have reported health problems. Additionally, mothers whose child had a false-positive result scored higher on the Parental Stress Index (Table 1).

Health Care Utilization

Children with false-positive screens had a median of 8 (interquartile range 6–12) primary care visits, compared to a median of 6 (interquartile range 5–8) for their normal screen counterparts (p<0.0001). After adjusting for children's age, which was the only significant covariate for primary care utilization, the adjusted rate ratio was 1.10 (95% CI: 0.98–1.22; p=0.09) (Table 2a).

Four (2.0%) children with false-positive screens had been to a specialist, each with no more than one visit. In contrast, 14 (10.5%) children with normal newborn screens had been to a specialist with the number of visits varying from 1–15 (mean 3.5; mode 1). These differences in specialist utilization were not significant (p=0.63).

In unadjusted analyses, children with false-positive screens were more likely to have ever visited the emergency room (28.6% versus 11.7%; p<0.001). After adjusting for the children's age, the odds ratio (OR) for children with a false-positive result having ever received care in an emergency room compared to children with a normal screening result was 1.31 (95%CI: 0.57–3.03; p=0.53). Further adjustment for significant sociodemographic variables did not significantly change the results (p=0.88), although parent marital status trended toward statistical significance (Table 2b).

15.4% of the children with false-positive newborn screens had been hospitalized by 6 months of age compared to 8.8% of the children with normal screens. The odds ratio for children with a false-positive screen being hospitalized in the first 6 months, compared to children with normal screens, was not significant in the unadjusted analysis (OR 1.89; 95% CI: 0.93–3.85; p=0.08). After adjusting for significant sociodemographic differences, specifically children's gender, race/ethnicity and family social position, the effect of newborn screening result remained nonsignificant (OR 1.51; 95%CI: 0.70–3.22; p= 0.29). Child gender was significantly related to odds of hospitalization (Table 2c).

Sensitivity Analyses

Among the children who had not been hospitalized, there were 13 normal screen children and 1 false-positive child who were just under 6 months of age at the time of interview. Conservatively assuming that all 14 of these children were hospitalized prior to 6 months of age, our results remained nonsignificant.

Because the children with normal screens all had married parents, we assessed whether this difference between the groups may have masked any association between newborn screen results and health care utilization. After excluding all children with unmarried mothers (from the false-positive group), the results for all utilization outcomes (primary care, specialists, emergency room, and hospitalization before 6 months) remained nonsignificant.

Finally, the addition of a covariate indicating that the child had a history of health problems did not change the significance of any results.

DISCUSSION

As newborn screening programs expand to include more disorders, many children will experience false-positive test results.[1] In our study, such results were not associated with increased health care utilization early in life. Although false-positive tests have been associated with higher parental stress,[9] this increased stress does not appear to lead to more health care utilization.

This information has particular relevance for cost-effectiveness evaluations of newborn screening. The majority of such evaluations of expanded newborn screening have limited the costs of a false-positive test to the costs that occur prior to diagnostic confirmation.[4–7] These studies assume that after a positive test result has been proven to be a false-positive no further costs are associated with that outcome. Our study suggests that, particularly for outpatient costs, this assumption is appropriate.

Early studies of population-based pediatric screening programs showed increased health care utilization following false-positive results[8] and parental misunderstanding of results leading to unnecessary restriction of children's activities.[24] The differences between these studies and the results reported here could suggest that, as newborn screening has expanded, parental understanding of such screening and parental interpretation of its results have improved. One could hypothesize that today's physicians and nurses more effectively communicate test results to families than did their predecessors. Studies of communication around newborn screening results, however, do not support this hypothesis. Researchers have consistently shown providers' ability to communicate about newborn screening is poor in both training and primary care settings.[25–29] It may be that physicians do not communicate the potentially serious nature of the initial positive screening test to families, thus parents have no reason for concern. This idea is supported by the fact that, as detailed elsewhere, in our study sample many parents of children with false-positive results did not know the correct reason for their child's follow-up testing.[9]

Alternatively, it may be that the association between false-positive newborn screening results and health care utilization varies by disorder, with positive tests for more life-threatening disorders leading to greater utilization. In such a situation, if our sample was biased towards less serious disorders we may have underestimated the impact of false-positive results on early health care utilization. Similarly, if our sample of children with normal results had particularly high rates of health care utilization, these levels could mask any increased utilization among our false-positive sample. However, both groups had lower than average rates of specialist use, [30] making it unlikely that the normal screen group had unusually high utilization rates.

Earlier studies, using a subset of our study sample, demonstrated increased parental stress among parents of children with false-positive newborn screening results.[9,10] Other authors have also found an association between false-positive newborn screen results and negative psychosocial effects.[31,32] We hypothesized that such psychosocial effects could lead to parents perceiving children with false-positive test results as vulnerable which in turn may lead to increased health care utilization.[16,17] Our results contradict this idea and show that the increased parental stress associated with false-positive results in other studies,[9] may not be associated with perceptions of child vulnerability that would be expected to lead to increased health care utilization. These results are consistent with a recent study, using a portion of our sample, which, using economic valuation methods, demonstrated that parents who have experienced a false-positive result have high tolerance for such results.[20]

This study was limited by its short time horizon. Parents were interviewed when their children were quite young, thus any long term differences in health care utilization would not have been seen. However, previous work has shown that perceptions of vulnerability decrease over time. [33] This suggests that differences in utilization due to vulnerability should have been captured in the time frame we used. Furthermore, as in any survey, parents may have been biased in their recollection of health care utilization, health problems or, in the case of the families assumed to have normal screens, newborn screening results. However, if the false-positive screens resulted in persistent parental concerns we would expect any differential response bias to favor both increased utilization among children with false-positive screens and an increased likelihood of parents correctly recalling the child's test results. Similarly, we chose not to control for the reported presence of health problems because of concern that such reports may be related to parental concerns, potentially triggered by newborn screening experiences, rather than children's actual health. We acknowledge that some, if not all, of the reported health problems may reflect actual disease and these children would be expected to utilize more health care. However, sensitivity analyses adjusting for such health problems found no associations between newborn screening result and health care utilization.

Additionally, children with false-positive results were significantly older than children with normal screens. We recognize that older children have greater opportunity for health care utilization, but also that younger children have more frequent scheduled primary care use. For this reason we attempted to compensate for differences in age through the inclusion of the child's age as a covariate in all regression analyses, except hospitalization in the first six months of life. In addition to differences in mean age, there were significant sociodemographic differences between our two samples. We believe these differences reflect our varied recruitment strategies, rather than true population differences between children who have normal and false-positive newborn screening results. Adjusting for these sociodemographic differences through regression analyses did not change our results, but may not fully address any differential access to care that may influence utilization. Moreover, in sensitivity analyses our results were robust to the exclusion of unmarried mothers from the false-positive group. Finally, we did not collect information regarding the specific false-positive result received. Therefore we can not assess whether a specific subset of false-positive results may be associated with increased health care utilization.

Adjusting for possible confounders reduced the association between a false-positive newborn screening result and health care utilization; this suggests that there may be sociodemographic disparities in health care utilization after a false-positive newborn screening. Further research in this area should specifically address whether such disparities exist and possible mechanisms, such as that some families may not receive adequate counseling and reassurance after a false-positive newborn screening result and hence may be more likely to seek additional medical care.

This is the largest study to date of children with false-positive newborn screening results. The sample afforded us with adequate power to detect a rate ratio as small as 1.17, one to two extra visits annually, for outpatient care. On the other hand, we were underpowered to detect differences in ER and inpatient utilization, events which are much less common than primary care visits. A relatively small study such as this one provides point estimates that can be used to determine sample sizes for future studies. For a study to have 80% power to detect a 50% difference in the odds of hospitalization in the first 6 months, or an odds ratio of 1.5, would require more than 2200 subjects. Similarly, more than 3400 would be required to detect a 50% difference in the odds of ER use. To detect a smaller difference in utilization, yet one that may be significant on a population basis, would require an even larger sample size. Additional research in this area should include follow-up studies with larger sample sizes and longer time frames.

Conclusion

The advancement of technologies such as tandem mass spectrometry and DNA microarrays is likely to lead to continued expansion of newborn screening.[34] This expansion should direct more attention to the impact of false-positive results. Our results suggest that false-positive results have little impact on early health care utilization and may therefore have little economic impact. However, it is unclear if this reflects parental understanding or misunderstanding of false-positive results. As education materials are developed for future screening programs a better understanding of why the false-positive result has little impact on health care utilization may help physicians counsel families with such results.

Acknowledgments

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Table 1

Characteristics According to Newborn Screening Result (unadjusted)

	False Positive (n=200)	Normal Screen (n=137)	p-value
Sociodemographic Characteristics			
Age at Time of Interview (months)	12.9	6.4	<0.0001
Female, %	47.5	46.7	0.88
Race/ethnicity, %			0.0001
Non-Hispanic White	79.0	92.7	
Non-Hispanic Black	12.5	0.7	
Hispanic	5.5	1.5	
Other	3.0	5.2	
Health Problems, %			0.01
Overall	33.5	20.7	
Prematurity	6.5	1.5	
Jaundice	9.0	5.9	
Respiratory Problems	4.5	1.5	
Ear Problems	4.5	0.7	
Gastrointestinal Problems	3.0	4.4	
Other	6.0	6.7	
Place of Residence, %			<0.0001
New England	12.1	100	
Pennsylvania	87.9	0	
Hollingshead Social Class[a], %			<0.0001
1, 2 or 3	63.3	91.2	
4 or 5	27.7	8.8	
Parents married, %	79.0	100	<0.0001
Maternal education, %			<0.0001
High School or less	37.5	9.5	
Greater than High School	62.5	90.5	
Mother's Parental Stress Index Score[b]	70.3	59.2	<0.0001
Health Care Utilization			
Number of outpatient visits, median	8	6	<0.0001
Ever visited a specialist, %	2	10.5	0.63
Ever visited an emergency room, %	28.6	11.7	<0.001
Hopitalized prior to 6 months of age, %	15.4	8.8	0.07

[a]Incorporates level of education and occupational status. Lower numbers indicate higher socioeconomic status.

[b]Higher number indicates greater stress.

Tables 2

Associations between false-positive newborn screening result and early health care utilization adjusted for significant sociodemographic confounders

a. Adjusted rate ratios (RR)for outpatient utilization in the previous year

	Outpatient Visits Adjusted Rate Ratio	95% Confidence Interval for Rate Ratio	p-value
False Positive Newborn Screen Result	1.10	0.98–1.22	0.09
One Month Increase in Child's Age	1.05	1.04–1.06	<0.001

b. Adjusted odds ratios (OR) for ever using the emergency room

	Ever Been to Emergency Room Adjusted Odds Ratio	95% Confidence Interval for Odds Ratio	p-value
False Positive Newborn Screen Result	1.19	0.50–2.81	0.88
One Month Increase in Child's Age	1.13	1.04–1.22	<0.01
Hollingshead Social Position <4[a]	1.07	0.55–2.07	0.85
Parents Married	0.46	0.20–1.01	0.05

c. Adjusted odds ratios (OR) for hospitalization prior to 6 months of age

	Hospitalized Prior to 6 months of Age Adjusted Odds Ratio	95% Confidence Interval for Odds Ratio	p-value
False Positive Newborn Screen Result	1.51	0.70–3.22	0.29
Hollingshead Social Position <4[a]	0.59	0.28–1.22	0.15
Female Sex	2.24	1.13–4.42	0.02
White Race	0.56	0.24–1.29	0.17

[a]Incorporates level of parent education and occupational status. Lower numbers indicate higher socioeconomic status