

# Results of Newborn Screening for Hearing Loss

## *Effects on the Family in the First 2 Years of Life*

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**Objective:** To determine whether there was increased stress and impact on the family for mothers of infants whose screening results and subsequent diagnostic findings indicated hearing loss (HL) and mothers of infants with a positive screening result who subsequently pass the rescreening (false-positive group), compared with mothers of infants who pass the initial screening (control group), when their children were aged 6 to 10, 12 to 16, and 18 to 24 months.

**Design:** Matched cohort analytic study.

**Setting:** Home visits.

**Patients/Participants:** Mothers of 33 infants with confirmed HL, 42 infants with a false-positive screening result, and 70 infants in the control group.

**Interventions:** Screening for HL.

**Outcome Measures:** Scores on the Parenting Stress Index and the Impact on Family–Adapted Version G.

**Results:** Mothers of infants in the false-positive group did not report increased stress or impact. Mothers of infants with HL reported greater financial impact, total impact, and caretaker burden compared with mothers of infants in the control group. In multivariate analysis of the total cohort, the presence of HL was associated with increased total impact on the family; a neonatal intensive care unit stay was associated with increased stress and total impact on the family; and older maternal age and greater family resources were associated with decreased stress and total impact on the family.

**Conclusions:** Although a false-positive result or a pass of the screening for HL was not associated with increased stress or impact, identification of HL was independently associated with greater total impact on the family when the child was 18 to 24 months of age.

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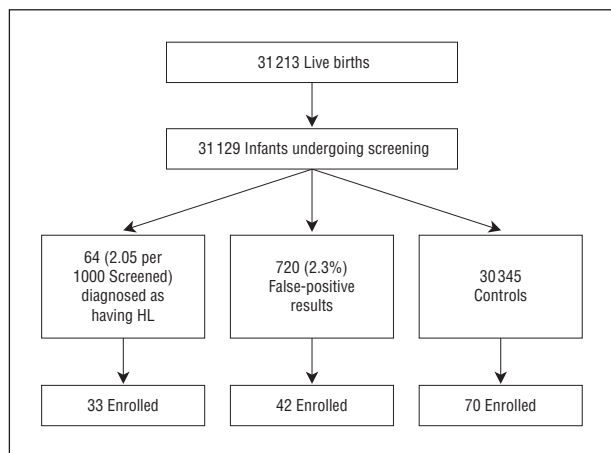
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**A**T PRESENT, APPROXIMATELY 92% of the 4 million infants born in the United States undergo screening for hearing loss (HL).<sup>1</sup> The incidence of congenital permanent HL ranges from 2 cases per 1000 births to 3 cases per 1000 births per year,<sup>2-5</sup> which extrapolates to 8000 to 12 000 infants annually in the United States. Rates of positive hospital screening results for HL of 2% to 3%, however, indicate that there are a significant number of false-positive screening results that may contribute to increased parenting stress.<sup>2,4,6-11</sup> In addition, a diagnosis of a permanent HL has been shown to be associated with increased stress.<sup>11</sup>

It has been reported that mothers of infants referred for additional testing after the neonatal screening experience increased stress at the time of the referral,<sup>12-15</sup> and it has been proposed that a

false-positive result of a neonatal HL screening may have a prolonged impact on families due to parental anxiety, distraction, disrupted family function, and unnecessary testing.<sup>16</sup> Families of infants diagnosed as having a permanent HL are likely to experience increased stress, particularly at the time of diagnosis.<sup>17-20</sup> The changes in stress and impact in the first 6 to 24 months of life for infants participating in a universal screening program for HL are unknown.

The primary objective of this study was to evaluate the long-term impact of HL screening results for mothers of infants with positive findings and who were diagnosed as having permanent congenital HL (HL group), mothers of infants with positive screening results who subsequently passed the rescreening (false-positive group), and mothers of infants who passed the initial screening (control group) when the child was aged 6 to 10,



**Figure.** Diagram of Rhode Island recruitment pool from October 15, 2002, to January 31, 2005. HL indicates hearing loss.

12 to 16, and 18 to 24 months. We hypothesized that (1) mothers of infants with HL will report the highest levels of stress and impact on the family; (2) mothers of infants with a false-positive screening result will have stress and impact levels similar to those of mothers of infants who pass the hearing screen; and (3) mothers in all study groups with stronger support systems and better resources will report decreased stress and impact on the family.

## METHODS

This was a prospective longitudinal study of a matched cohort of infants born and undergoing screening for HL in Rhode Island from October 15, 2002, through January 31, 2005. Infants underwent screening with a 2-step protocol that included otoacoustic emissions followed by automated auditory evoked response and an outpatient rescreening for those with positive findings. All patients with positive findings were referred for diagnostic testing. The Rhode Island rate of referral for newborns with positive screening results during this period ranged from 2.1% to 2.9%.

Inclusion criteria for the HL group included a positive result of a neonatal screening at a Rhode Island hospital and subsequent diagnosis of permanent congenital HL. Eligible families of infants with HL were identified first in the Rhode Island Tracking Database and recruited through Family Guidance, a statewide specialty early intervention program servicing families of infants and toddlers with HL. Families of infants with HL who did not participate in Family Guidance were recruited by mail.

Demographic information, neonatal data, and results of screening and diagnostic tests were collected from the Rhode Island Tracking Database and a Rhode Island Department of Health database to identify a pool of matched subjects in the false-positive and control groups. Attempts were made to match at least 1 infant in the false-positive group and 1 to 2 infants in the control group for each child in the HL group. Hierarchical matching criteria were sex, date of birth ( $\pm 30$ -90 days), hospital of birth, a well-infant nursery vs a neonatal intensive care unit (NICU) stay, maternal education, race/ethnicity, and health insurance. All infants with screening results were eligible, including infants with comorbidities and those from non-English-speaking families. All mailed letters and response forms were in English or Spanish. The **Figure** shows the eligible children for the study period and the number enrolled.

The following standardized, well-validated tools were administered to the primary caregiver during home visits at 6 to 10, ( $n=101$ ), 12 to 16 ( $n=126$ ), and 18 to 24 ( $n=123$ ) months of age. Scores of socioeconomic status were calculated with the Hollingshead Four Factor Index of Social Status<sup>21</sup> as a weighted combination of parental education and occupation. Widely used in the literature, the Hollingshead socioeconomic status score was validated against the US 1970 Census and National Opinion Research Center surveys.

The Family Support Scale<sup>22</sup> measures the helpfulness of sources of support to families rearing a young child and contains 18 items rated on a 5-point scale. The Family Support Scale has been used in a number of studies examining the effects of social support.<sup>23-28</sup> The reliability and validity were established in a study of 139 parents of preschool children with multiple disabilities.<sup>23,29</sup>

The Family Resource Scale<sup>30</sup> measures maternal perception of the adequacy of resources, including time and money in the household. The Family Resource Scale includes 31 items rated on a 5-point scale. The hierarchy is derived from a conceptual framework that predicts the inadequacy of resources necessary to meet individually identified need. The reliability and validity of the Family Resource Scale were established in mothers of preschool children who were developmentally disabled or at risk and participating in early intervention.<sup>23,30</sup>

The objective of the Impact on Family-Adapted Version G scale<sup>31</sup> is to measure the effect of the child's condition in producing change in the family. The following 6 dimensions of impact are scored: total impact, financial impact, family burden, caretaker burden, disruption of planning, and coping. Total impact is the unweighted sum of financial impact, family burden, caretaker burden, and disruption of planning. Items are reflective of the impact of the child's condition. The 24-item scale has been shown to elicit variability in response and to be internally consistent.<sup>31</sup>

The Parenting Stress Index<sup>32</sup> was designed to identify stressful areas in parent-child interactions. The short form consists of 36 items with 5-point Likert responses resulting in a total stress score and the following 3 subscales: parental distress, parent-child dysfunctional interaction, and difficult child. The short form relates highly to the full form ( $r=0.92$ ) and was validated among children aged 1 month to 12 years.<sup>32,33</sup>

The study was approved by institutional review boards at Women & Infants' Hospital; the Rhode Island Department of Health, Providence; and the Centers for Disease Control and Prevention, Atlanta, Georgia. Informed consent was obtained.

The analytic groups were defined by the results of screening for HL. Between-group analyses were performed using analysis of variance on continuous variables and the  $\chi^2$  test on categorical variables. Multiple regression models were constructed for the 2 outcomes of stress and impact, with hearing status and mediating variables as predictors. Independent variables that have been shown to mediate stress and impact were tested.<sup>2,4,6-23,29,30,34-37</sup>

The following 3 categories of independent variables were entered into the models: child factors (HL vs pass; false-positive results vs pass; and NICU stay vs well-infant nursery), maternal factors (married vs single and maternal age at the time of the visit), and environmental factors (Hollingshead socioeconomic status score and total Family Support and Family Resources scale scores). Dependent variables were total stress and total impact scores. Separate models were also constructed for infants with stays in a NICU and a well-infant nursery.

Before recruitment, estimates of power for between-group comparisons of mean scores for total stress were made. These were calculated using the normative values for the Parenting Stress Index, and a 1:2 ratio of infants with HL to control infants. For the largest HL group size used in this estimate ( $n=25$ ), we estimated that we would be able to detect a difference of 10.3 points

**Table 1. Maternal and Infant Characteristics Across the 3 Study Groups<sup>a</sup>**

Characteristic	Study Group			P Value
	HL	False-Positive	Control	
<b>Mothers</b>				
No. of mothers	30	39	58	
Maternal age, mean (SD), y	32 (6)	30 (6)	33 (6)	.17
Married	25 (83)	33 (85)	48 (83)	.97
Educational level				.41
< High school	2 (7)	2 (5)	5 (9)	
Finished high school	4 (13)	7 (18)	2 (3)	
Partial college	12 (40)	14 (36)	23 (40)	
Finished college	12 (40)	16 (41)	28 (48)	
White race	27 (90)	33 (85)	55 (95)	.38
Health insurance type				.73
Private	24 (80)	29 (74)	46 (79)	
Medicaid/HMO	4 (13)	8 (21)	10 (17)	
Medicaid/public assistance	2 (7)	1 (3)	2 (3)	
Self/no insurance	0	1 (3)	0	
Hollingshead SES score, mean (SD)	40.1 (14.6)	41.0 (11.8)	45.4 (11.9)	.11
<b>Infants</b>				
No. of infants	33	42	70	
Female sex	12 (36)	14 (33)	29 (41)	.68
Gestational age < 37 wk	22 (67)	21 (50)	32 (46)	.13
Birthweight < 1500 g	17 (52)	13 (31)	12 (17)	.002
NICU stay	21 (64)	18 (43)	43 (61)	.10
Participating in EI	31 (94)	15 (36)	17 (24)	.001

Abbreviations: EI, early intervention; HL, hearing loss; HMO, health maintenance organization; NICU, neonatal intensive care unit; SES, socioeconomic status.

<sup>a</sup>Mothers of multiple infants were counted once for maternal characteristics and assigned to the most critical group based on hearing screen status. The study groups are described in the "Introduction" section of the text. Unless otherwise indicated, data are expressed as number (percentage) of study subjects.

(approximately 0.67 SD), with 80% power at a significance level of  $\alpha = .05$ . Because the HL recruitment was 33 infants, the detectable difference decreased to about 9 points.

## RESULTS

We enrolled 33 of 64 families of infants with HL (Figure). Family Guidance enrolled 29 of 52 families (56%), and an additional 4 of 12 families (33%) were successfully recruited by mail. Of the 31 families with HL who refused to participate, 7 (23%) were nonnative English speakers and 7 (23%) reported experiencing situations precluding enrollment. All of the false-positive and control group matches (of 960 matches, 112 were enrolled) were recruited by mail.

The cohort consisted of 127 mothers of 145 infants, including 16 sets of twins and 1 set of triplets. Mothers in the 3 groups were well matched (**Table 1**). Infant groups were similar for sex, prematurity rate, and NICU status. More infants with HL had a birth weight of less than 1500 g. In addition, 31 infants with HL (94%) were participating in early intervention compared with 15 (36%) in the false-positive group and 17 (24%) in the control group. Infants in the false-positive group underwent rescreening at a mean age of 44 days, and infants were diagnosed as having HL at a mean (SD) age of 108 (98) days.

Total scores for the Family Support and Family Resource scales did not differ among the 3 study groups at the 3 study visits (**Table 2**). Of the subscale scores,

only the support score for general professional services at 18 to 24 months of age was higher in the HL group compared with the false-positive and control groups.

Maternal report of stress and impact is shown in **Table 3**. Total stress and subscale scores (not shown) were similar among the study groups at each study assessment. There was a trend for mothers of infants with HL to be less likely to have high stress scores (>85th percentile) than mothers in the 2 other study groups, which was significant at 18 to 24 months. There were no differences in the total impact scores or subscale scores among the study groups at 6 to 10 months of age. The financial impact score was significantly higher for the mothers of infants with HL at 12 to 16 and 18 to 24 months of age compared with the control group, and the total impact and caretaker burden scores were significantly higher at 18 to 24 months of age compared with the control and false-positive groups.

Regression analysis for the total cohort at the 3 visits indicated that false-positive results were not associated with outcomes, but a NICU stay was associated with increased stress and impact, and greater family resources were associated with decreased stress and impact (**Table 4**). At 18 to 24 months of age, HL was associated with an increased total impact on the family. Older maternal age was also associated with decreased stress at 6 to 10 and 12 to 16 months of age and decreased impact at 6 to 10 months of age.

Because of the strong influence of a NICU stay in the models, separate models were run for those with NICU

**Table 2. Family Support and Resources at 6, 12, and 18 Months of Age Across the 3 Study Groups<sup>a</sup>**

Outcome	Study Group			P Value
	HL	False-Positive	Control	
<b>Ages 6-10 mo</b>				
No. of mothers	29	27	45	
Total Family Support Scale score	38.6 (7.9)	35.7 (8.4)	36.2 (9.6)	.41
Total Family Resources Scale score	120.4 (15.7)	120.3 (13.8)	123.5 (16.0)	.60
<b>Ages 12-16 mo</b>				
No. of mothers	30	32	64	
Total Family Support Scale score	36.8 (9.6)	34.8 (9.9)	35.4 (11.4)	.75
Total Family Resources Scale score	123.6 (9.5)	123.8 (13.4)	125.4 (13.6)	.75
<b>Ages 18-24 mo</b>				
No. of mothers	31	31	61	
Total Family Support Scale score	37.7 (10.6)	35.8 (9.0)	35.1 (9.7)	.48
General professional service score	5.7 (2.6) <sup>b</sup>	5.0 (1.7)	4.5 (1.6)	.02
Total Family Resources Scale score	120.6 (12.7)	122.1 (14.9)	125.5 (13.7)	.23

Abbreviation: HL, hearing loss.

<sup>a</sup>Study groups are described in the "Introduction" section. Unless otherwise indicated, data are expressed as mean (SD).

<sup>b</sup>Compared with the control group.

**Table 3. Stress and Impact on the Family at 6, 12, and 18 Months of Age Across the 3 Study Groups<sup>a</sup>**

Outcome	Study Group			P Value
	HL	False-Positive	Control	
<b>Ages 6-10 mo</b>				
No. of mothers	29	27	45	
Total Parenting Stress Index	66.4 (15.2)	68.3 (18.7)	67.4 (15.6)	.91
Score > 85th percentile, No. (%)	4 (14)	4 (15)	8 (18)	.89
Total Impact on Family score	26.5 (5.0)	25.6 (3.8)	25.7 (3.1)	.62
<b>Ages 12-16 mo</b>				
No. of mothers	30	32	64	
Total Parenting Stress Index	65.9 (12.6)	66.6 (17.2)	68.6 (16.4)	.68
Score > 85th percentile, No. (%)	1 (3)	4 (13)	9 (14)	.29
Total Impact on Family score	26.1 (4.6)	25.2 (4.0)	24.8 (3.5)	.32
Financial impact	4.1 (0.8) <sup>b</sup>	3.8 (0.6)	3.6 (0.7)	.007
Family burden	7.3 (1.7)	7.0 (1.2)	6.9 (1.2)	.32
Caretaker burden	5.6 (1.3)	5.3 (1.0)	5.4 (0.9)	.40
Disruption of planning	9.1 (1.6)	9.2 (1.7)	9.0 (1.4)	.80
Coping	6.5 (1.7)	6.2 (1.2)	6.8 (1.7)	.19
<b>Ages 18-24 mo</b>				
No. of mothers	31	31	61	
Total Parenting Stress Index	66.7 (10.1)	67.5 (17.8)	68.4 (16.4)	.89
Score > 85th percentile, No. (%)	0	6 (19)	11 (18)	.04
Total Impact on Family score	26.6 (5.6) <sup>b,c</sup>	24.0 (4.5)	23.9 (4.3)	.02
Financial impact	4.1 (1.2) <sup>b</sup>	3.7 (0.8)	3.5 (1.0)	.03
Family burden	7.3 (1.5)	6.6 (1.5)	6.6 (1.4)	.07
Caretaker burden	6.0 (1.4) <sup>b,c</sup>	5.1 (1.1)	5.3 (0.9)	.004
Disruption of planning	9.3 (1.6)	8.6 (1.7)	8.5 (1.4)	.07
Coping	6.5 (1.9)	6.8 (2.0)	6.7 (1.9)	.75

Abbreviation: HL, hearing loss.

<sup>a</sup>Study groups are described in the "Introduction" section. Unless otherwise indicated, data are expressed as mean (SD).

<sup>b</sup>Compared with the control group.

<sup>c</sup>Compared with the false-positive group.

and well-infant nursery stays (**Table 5**). Neither false-positive status nor permanent HL contributed independently to stress for mothers of NICU infants. Increased maternal age was associated with decreased stress at 6 to 10 and 12 to 16 months of age, and greater resources were associated with lower stress levels at all 3 visits. The

only independent predictor of stress in the well-infant nursery models was family resources.

In the models for mothers of NICU infants, false-positive status did not contribute independently to impact. Hearing loss contributed to a greater impact at 12 to 16 months and 18 to 24 months of age, and more family

**Table 4. Regression Models for Total Stress and Impact Scores for Total Cohort**

Independent Variable	Stress Outcome, Age at Evaluation						Impact Outcome, Age at Evaluation					
	6-10 mo (n=101)		12-16 mo (n=126)		18-24 mo (n=123)		6-10 mo (n=101)		12-16 mo (n=126)		18-24 mo (n=123)	
	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value
Child NICU stay	9.09	.002	6.32	.02	5.35	.04	1.48	.04	1.23	.05	2.59	<.001
Child HL	-2.80	.39	-3.02	.35	-3.32	.30	0.23	.78	1.07	.17	2.24	.008
Child false-positive finding	0.27	.94	-2.59	.41	-1.30	.68	-0.44	.61	0.53	.48	0.31	.70
Marital status	0.15	.97	1.21	.78	-6.61	.13	0.15	.88	-2.10	.045	-1.47	.20
Maternal age at visit	-0.68	.004	-0.54	.03	-0.18	.45	-0.14	.03	0.04	.52	0.09	.14
SES score	0.21	.08	0.18	.14	0.19	.10	0.04	.23	0.04	.12	0.03	.36
Total Family Support Scale score	-0.10	.55	-0.19	.14	-0.07	.59	0.02	.65	0.01	.68	-0.05	.19
Total Family Resources Scale score	-0.52	<.001	-0.36	.002	-0.45	<.001	-0.13	<.001	-0.13	<.001	-0.15	<.001
Model $R^2$ , P value	.41	<.001	.23	<.001	.25	<.001	.33	<.001	.29	<.001	.41	<.001

Abbreviations:  $\beta$ , unstandardized regression coefficient; HL, hearing loss; NICU, neonatal intensive care unit; SES, socioeconomic status.

**Table 5. Regression Models for Total Stress Scores by Nursery Type**

Independent Variable	NICU Stay, Age at Evaluation						Well-Infant Nursery Stay, Age at Evaluation					
	6-10 mo (n=59)		12-16 mo (n=72)		18-24 mo (n=70)		6-10 mo (n=42)		12-16 mo (n=54)		18-24 mo (n=53)	
	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value
Child HL	-1.99	.66	-7.70	.07	-2.71	.53	-4.71	.33	3.46	.49	-3.17	.52
Child false-positive finding	1.85	.73	-3.46	.44	-0.04	.99	1.61	.70	-2.54	.57	-3.74	.40
Maternal age at visit	-0.88	.007	-0.89	.006	-0.50	.14	-0.22	.52	0.05	.91	0.10	.80
SES score	0.31	.056	0.16	.28	0.16	.30	-0.04	.83	0.03	.87	0.12	.53
Total Family Support Scale score	-0.11	.69	-0.11	.52	-0.04	.82	-0.15	.47	-0.32	.09	-0.21	.33
Total Family Resources Scale score	-0.54	<.001	-0.42	.003	-0.47	.001	-0.46	.002	-0.26	.16	-0.44	.003
Model $R^2$ , P value	.39	<.001	.30	<.001	.22	.01	.31	.03	.14	.31	.22	.06

Abbreviations: See Table 4.

**Table 6. Regression Models for Total Impact Scores by Nursery Type**

Independent Variable	NICU Stay, Age at Evaluation						Well-Infant Nursery Stay, Age at Evaluation					
	6-10 mo (n=59)		12-16 mo (n=72)		18-24 mo (n=70)		6-10 mo (n=42)		12-16 mo (n=54)		18-24 mo (n=53)	
	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value	$\beta$	P Value
Child HL	-0.19	.85	1.80	.053	3.30	.002	0.40	.78	0.33	.81	0.85	.54
Child false-positive finding	0.59	.62	0.63	.52	0.86	.44	-1.51	.23	0.15	.91	-0.75	.56
Maternal age at visit	-0.13	.07	-0.04	.52	0.05	.54	-0.09	.36	0.15	.17	0.13	.26
SES score	0.08	.02	0.08	.02	0.03	.46	-0.06	.26	-0.06	.31	0.01	.83
Total Family Support Scale score	0.04	.48	0.02	.56	-0.03	.54	-0.03	.60	-0.03	.53	-0.10	.11
Total Family Resources Scale score	-0.13	<.001	-0.13	<.001	-0.15	<.001	-0.11	.009	-0.15	.007	0.14	<.001
Model $R^2$ , P Value	.36	<.001	.33	<.001	.37	<.001	.35	.01	.23	.04	.32	.005

Abbreviations: See Table 4.

resources were associated with a smaller impact at each visit (**Table 6**). The only independent predictor of impact in the well-infant nursery models was family resources.

**COMMENT**

We identified similar levels of reported parenting stress for mothers in all our study groups at all 3 time points. We

speculate that there are windows of acute stress that occur in a temporal relationship to screening and diagnosis, and that stress diminishes for most families with the onset of intervention services and within the context of other family and environmental factors that mediate stress. We identified evidence of greater total impact on the family as the child with HL increased in age that appears to be greatly influenced by the availability of resources.

Our second hypothesis, however, that mothers of infants with a false-positive result of screening for HL will have stress and impact levels similar to those of mothers of infants in the control group was confirmed, suggesting that, even if there is stress at the time of the rescreening, these effects do not persist.

Most studies reporting stress associated with a false-positive screening result have been performed in the first months of life or close to the time of the screening or rescreening.<sup>11</sup> Similarly, Abdala de Uzcategui and Yoshinaga-Itano<sup>12</sup> found that 37% of mothers in mailed interviews reported feeling depressed at the time of their infant's referral for audiologic testing.

Stuart et al<sup>37</sup> reported similar maternal stress findings for mothers of infants who passed the screening and mothers of infants referred for further testing 1 month after discharge. We speculate that because programs that screen for HL have expanded in the United States, parents and health care providers are better informed about newborn screening. In addition, the mothers of children with false-positive screening results in the present study knew that their infant had passed the rescreening at the time of enrollment.

It has been suggested that there is a continuum of mounting stress for hearing families whose infants are identified as having HL that increases as they progress through the screening, rescreening, and diagnostic process.<sup>19</sup> There may be windows of acute stress linked to specific events, such as receiving the news of the screening result or the diagnosis. Stress related to disability or fear of disability can have a significant impact on family and child outcomes.<sup>6,20,34-36</sup> Meadow-Orlans<sup>20</sup> reported that mothers of 9-month-old infants who were deaf or hard of hearing perceive increased life stress (such as divorce), but that total parenting stress scores related to parent-child relationships were similar for mothers of infants with HL and mothers of infants with normal hearing.<sup>20</sup> In addition, some of the mothers of infants with HL reported extremely low stress scores, which were interpreted as denial. In our cohort, mothers of infants with HL were less likely to report high (>85th percentile) stress levels. We speculate that participation in early intervention and maternal report of higher general professional services may contribute to this finding.

The total Impact on the Family scores of the HL group began to diverge from those of the other study groups at 12 to 16 months of age. The higher total impact, financial impact, and caretaker burden scores of mothers in the HL group at 18 to 24 months of age may be a reflection of the parental time and costs associated with HL with increasing age and parallel the child's developing language and the mother's adjustment to new communicative needs.

Our third hypothesis was that mothers with stronger support and better resources will report decreased impact on the family and decreased stress. Only greater resources provided an independent contribution to decreased stress and impact. However, the strength of the contribution of a NICU stay in our models is impressive. Infants in the HL group with a NICU stay are more likely to have other medical and neurodevelopmental morbidities, placing additional demands on the family. Ap-

proximately 40% of children with permanent HL may have other disabilities.<sup>38,39</sup> The association of older maternal age with decreased stress could be attributed in part to more parenting experience.

The strengths of our study include that the maternal groups were well-matched, the mothers were interviewed at 3 time points, and the sample includes infants with NICU and well-infant nursery stays. Limitations are that we did not evaluate stress in the newborn period, and, because all families of children with permanent HL did not participate, there may be bias imposed by interest in the study. In summary, whereas a false-positive screening result does not have lasting effects on the family, HL has significant impact on the family, which is mediated by a spectrum of child, maternal, and environmental indicators.

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