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Evaluating Loss to Follow-up in Newborn Hearing Screening in Massachusetts

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ABSTRACT

OBJECTIVE. The purpose of this work was to examine loss to follow-up on the use of diagnostic or intervention services for Massachusetts infants and children screened or diagnosed with hearing loss and risk factors for becoming lost to follow-up.

METHODS. We used data from the Massachusetts Childhood Hearing Data System and Early Intervention Information System. We calculated the percent use of audiologic evaluation for Massachusetts infants born in 2002–2003 who did not pass hearing screening and Early Intervention services for those with hearing loss. We generated crude and adjusted relative risks, as well as confidence intervals, to estimate associations of maternal and infant factors with the use of audiologic evaluation and early intervention services. Factors evaluated included child's birth weight and hearing screening or diagnostic results and maternal age, race or ethnicity, marital status, smoking status during pregnancy, educational attainment, health insurance, and residence region.

RESULTS. In 2002–2003, 11% of Massachusetts children who did not pass hearing screening became lost to follow-up on the audiologic evaluation, and 25% of those with hearing loss did not receive early intervention services. Children were at higher risk of becoming lost to follow-up on audiologic evaluation if their mothers were nonwhite, covered by public insurance, smokers during pregnancy, or residing in western, northeastern, or southeastern Massachusetts compared with those in the Boston region. Of children with hearing loss, those with a unilateral or mild or moderate degree of hearing loss, normal birth weight, or living in the southeastern or Boston region were more likely to go without early intervention services.

CONCLUSIONS. Massachusetts has excellent follow-up rates overall. Our analyses allow the program to prioritize limited resources to subgroups of infants who are at high risk of becoming lost to follow-up.

EVERY YEAR, ~200 infants and young children are diagnosed with a permanent hearing loss in Massachusetts.¹ Without appropriate intervention, children with hearing loss can experience delays in cognitive, verbal, behavioral, and emotional development, which may later affect academic achievement and employment success.^{2–4} These delays are apparent for children with severe and profound hearing loss, as well as for those whose losses fall within mild and moderate ranges.^{2,5}

To ensure the optimal development of children with hearing loss, Massachusetts passed a universal newborn hearing screening law in 1998.⁶ The law mandates that birth facilities screen all newborns for hearing loss and refer those who do not pass a screening for an audiologic diagnostic evaluation. Birth facilities are required to refer newborns who do not pass a hearing screening to 1 of the Massachusetts Department of Public Health (MDPH)–approved audiologic diagnostic centers (ADCs) before discharge. MDPH-approved ADCs must have an annual pediatric caseload of $\geq 10\%$ to ensure the quality of diagnostic services.

In Massachusetts, all infants and children with hearing loss, of any type and degree, who are < 3 years of age, are eligible for Early Intervention (EI) services. Families are informed about EI services when the diagnosis of hearing loss

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Key Words

early hearing detection and intervention, lost to follow-up, early intervention

Abbreviations

MDPH—Massachusetts Department of Public Health
 ADC—audiologic diagnostic center
 EI—Early Intervention
 IFSP—Individualized Family Service Plan
 UNHSP—Universal Newborn Hearing Screening Program
 EHDI—Early Hearing Detection and Intervention
 CHDS—Childhood Hearing Data System
 EIS—Early Intervention Information System
 EBC—electronic birth certificate
 AE—audiologic evaluation report
 FIR—family intake record
 ABR—auditory brainstem response
 OAE—otoacoustic emission
 aRR—adjusted relative risk
 CI—confidence interval

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is made, although the actual referral may only occur if families opt for the services. On referral, Massachusetts EI programs provide initial counseling and work with families to develop Individualized Family Service Plans (IFSPs), which include ongoing audiologic monitoring. Families of infants and children who miss or do not pass hearing screening or who are diagnosed with hearing loss are followed by the Universal Newborn Hearing Screening Program (UNHSP) to ensure the use of outpatient screening, diagnostic evaluation, EI, and other services.

National Early Hearing Detection and Intervention (EHDI) goals and objectives include: (1) all newborns will be screened for hearing loss before 1 month of age; (2) all infants who do not pass a hearing screening will have an audiologic diagnostic evaluation before 3 months of age; and (3) all infants identified with a hearing loss will receive appropriate EI services before 6 months of age.⁷ Thus far, in accordance with national EHDI goals, Massachusetts has achieved a 99% screening rate, with 29 MDPH-approved ADCs providing diagnostic evaluations and 62 EI programs providing services to children with hearing loss.

Recently, there has been national emphasis on ensuring that appropriate follow-up, through diagnostic and intervention services, occurs among children who do not pass a hearing screening or who are diagnosed with hearing loss.⁸ In particular, disparities in diagnostic and intervention services among some socioeconomic groups, including racial and ethnic minorities, teenaged or less than high school-educated mothers, or families with public insurance, are of concern. Experiences from other public programs have suggested that these children may be at a high risk of becoming lost to follow-up.⁹⁻¹¹ Furthermore, families in rural areas may face additional barriers to service access.⁸ Other factors, including the severity of a child's condition, may also impact the use of services.¹² To address these concerns, this study evaluated loss to follow-up on the use of diagnostic or intervention services for Massachusetts infants and children screened or diagnosed with hearing loss and examined the risk factors for becoming lost to follow-up.

METHODS

Data Sources and Study Population

This study linked 2 databases, the Massachusetts Childhood Hearing Data System (CHDS) and Early Intervention Information System (EIIS), to obtain information on demographic and medical characteristics, hearing screening results, audiologic evaluation results, and EI enrollment for Massachusetts children born in 2002–2003.

The CHDS is a statewide surveillance and tracking data system created in 2001 to assist the UNHSP in implementing and evaluating its newborn hearing program activities. The CHDS is composed of 3 components: electronic birth certificate (EBC), audiologic evaluation report (AE), and family intake record (FIR) from the newborn hearing program. The EBC provides hearing

screening results and demographic and medical information for all children born or residing in Massachusetts. Hearing screening is performed using auditory brainstem response (ABR), otoacoustic emissions (OAEs), or a combination of ABR and OAE. Information on specific screening technology is collected at the facility level but not at the individual screening level. Only the results of the final hearing screening are reported, regardless of whether it occurs before hospital discharge or as an outpatient rescreen. The AE collects audiologic evaluation data from MDPH-approved ADCs for children with parental consent. It also collects data on risk factors for hearing loss (eg, a history of meningitis) and intervention information given during the appointment. All of the infants who come in for audiologic evaluation receive objective auditory brainstem tests (eg, ABR, OAE, or tympanometry). In addition, behavioral audiometric tests (eg, visual reinforcement audiometry) are given to infants and toddlers >6 months of age to confirm diagnosis. Criteria are the same for a bilateral and unilateral hearing loss. In cases where there are discrepancies in diagnoses between auditory brainstem tests and behavioral audiometric tests, audiologists in the ADCs make professional judgments. At the time of this analysis, all of the infants with hearing loss or who were at risk for progressive or late-onset hearing loss received ongoing audiologic monitoring every 6 months until 3 years of age. In addition, the ADCs were required to submit AE reports to the MDPH for the first visit and when there is a change on the type, degree, or risk factor of hearing loss. For the purpose of this study, results from the most recent AE reports were used. The FIR collects service use data from families and records staff interactions with families during outreach contacts, including a decision to decline follow-up. From the CHDS, our study obtained information on a child's birth characteristics (infant birth weight, maternal age, race or ethnicity, marital status, smoking status during pregnancy, educational attainment, health insurance coverage, and region of residence), hearing screening result, audiologic evaluation result, and reason for not using audiologic evaluation or EI services.

The EIIS collects information on EI referral, evaluation, IFSP, and discharge through a Web-based application. Massachusetts EI programs have an open referral process. Referrals can be made by any interested individual, including hospital personnel, health care providers, parents, social services agencies, and day care providers who believe the child may be eligible for the program. When a diagnosis of hearing loss is made, ADCs are required to discuss EI services with families and make referrals if families choose to use the services. Within 45 days of the referral, a trained evaluator conducts the eligibility evaluation at the child's home or other natural setting. An IFSP that tailors EI services to meet the unique needs of the child and family is developed for eligible children. The UNHSP works with families and EI programs to ensure that the evaluation visits and development of IFSPs occur. For UNHSP evaluation purposes, we used the first date of a referral in the EIIS

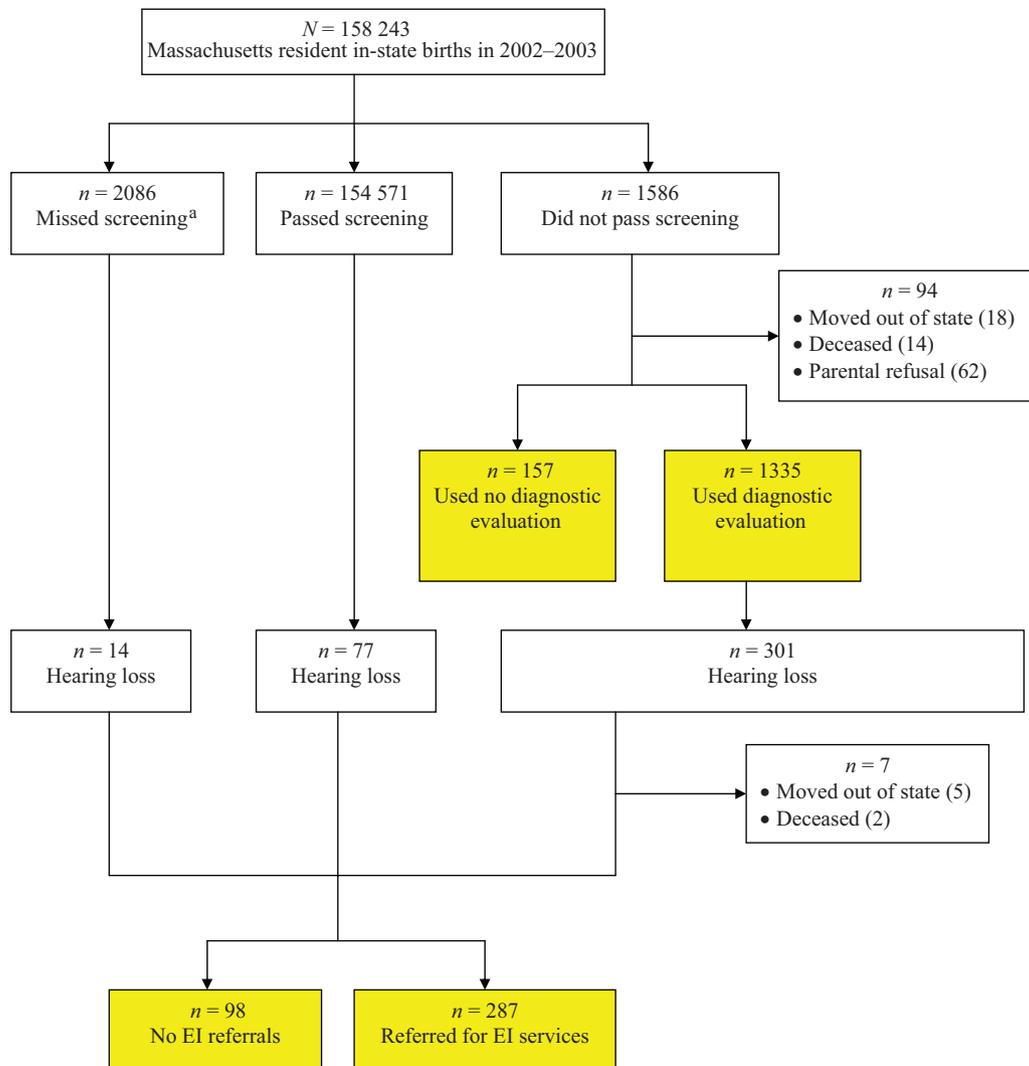


FIGURE 1 Study population. ^a Includes infants who died shortly after birth, whose parents refused screening, or whose hearing-screening data were missing.

as an indicator of the provision of EI services to children with hearing loss.

All of the Massachusetts residents born in the state between January 1, 2002, and December 31, 2003, were included in our study population (Fig 1). For assessing the use of audiologic evaluation services (analysis 1), analyses were limited to those who did not pass hearing screening at birth. When examining the use of EI services, analyses focused on those who were diagnosed with hearing loss at an MDPH-approved ADC by December 31, 2004 (analysis 2). Of 158 243 Massachusetts infants born during this period, 1586 (1%) did not pass a hearing screening and were referred for audiologic evaluation. Ninety-four children who did not pass a hearing screening were excluded from analysis, including those who moved out of state shortly after birth ($n = 18$), died ($n = 14$), or whose parents declined to be followed up by MDPH based on the FIR ($n = 62$). Overall, the analytic sample size for analysis 1 was 1492 children. Except for higher proportions of children born

weighing <2500 g or living in the Boston region, children excluded from analysis 1 were no different in other characteristics from those included in analysis 1. Of 301 children born in 2002–2003 who did not pass their hearing screening and were diagnosed with hearing loss, 7 were excluded from analysis, including those who, shortly after diagnosis, either moved out of state ($n = 5$) or died ($n = 2$). Fourteen children who missed the screening and 77 who passed the screening were diagnosed with hearing loss. Our overall analytic sample size for analysis 2 was 385 children.

Measures of Variables

Outcome Variables

Two outcome variables were evaluated: loss to follow-up on the use of audiologic evaluation services and loss to follow-up for EI referrals. Children were considered lost to follow-up on the use of audiologic evaluation services if they did not pass their newborn hearing screening and

did not receive an audiologic evaluation at an MDPH-approved ADC or if their parents declined consent to release information to MDPH and were unable to be reached by outreach staff (for whom we did not have FIR data to exclude them from analysis). Children were considered lost to follow-up for EI referrals if MDPH was informed of their diagnosis of hearing loss by 1 of the MDPH-approved ADCs but their record could not be located in the EIIS as of September 30, 2005.

Covariates

We assessed the predicting values of child factors (birth weight and hearing screening or diagnostic results) and maternal factors (age, race or ethnicity, marital status, smoking status during pregnancy, educational attainment, health insurance, and residence region) on the outcomes. These factors were selected because of their empiric associations with participation in public programs.^{8–12} A child had low birth weight if he or she was born weighing <2500 g. Children were given a bilateral hearing referral if both ears did not pass a newborn hearing screening or a unilateral hearing referral if either ear did not pass a newborn hearing screening. Children were defined as having a hearing loss if either a type or degree of hearing loss was reported. The CHDS collects data on 4 types of hearing loss: conductive, sensorineural, auditory dys-synchrony (auditory neuropathy), and mixed. Children who were unable to respond to sound of >71 dB in either ear were defined as having a severe or profound degree of hearing loss. Children who were unable to respond to sound of 21 to 70 dB, who had high frequency loss (>2000 Hz) only, or whose degree of hearing loss was indeterminable were classified as having mild or moderate hearing loss. Hearing loss was further divided into 2 categories: unilateral (1 ear affected) and bilateral (both ears affected).

Women who self-reported their race as white non-Hispanic on the EBC were classified as white, and all of the other races or Hispanic ethnicity were grouped as nonwhite. Women were classified as unmarried if they were single, divorced, or widowed. Maternal educational attainment was divided into 2 categories: at or more than high school if they completed ≥ 12 years of elementary or secondary education and less than high school if they completed <12 years of elementary or secondary education. Health insurance information was based on the source of payment for the delivery as recorded on the EBC. Mothers had public coverage if expenses for delivery were paid through government programs, including Medicaid and the free care pool. Mothers had private coverage if a third party other than the government paid for their delivery expenses. Mothers with no private or public coverage were classified as being uninsured (including self-pay and unknown coverage).

Data Analysis

We calculated the percentage use of audiologic evaluation services for infants who did not pass a hearing screening and EI services for children with hearing loss.

We generated crude and adjusted relative risks (aRRs), as well as confidence intervals (CIs), to estimate associations of maternal and infant factors with the use of audiologic evaluation and EI services. aRRs were estimated using a Poisson regression model with a robust error variance.^{13,14} In all of the regression models, we included a child's birth weight and the mother's age, race or ethnicity, marital status, smoking status during pregnancy, educational attainment, health insurance for delivery, and residence region. Laterality of hearing screening results was added to the models of the use of audiologic evaluation services, whereas laterality and degree of hearing loss were added to the models of the use of EI services. Because of collinearity between maternal age and educational attainment and between maternal age and marital status, the effect of maternal age was evaluated in the models where maternal educational attainment and marital status were excluded. In addition, the effects of maternal educational attainment and marital status were evaluated in the models where maternal age was excluded. Because of the small sample size in the models of the use of EI services, we dichotomized residence regions based on the lost-to-follow-up results and regrouped health insurance (private versus public or no insurance) to reduce the number of variables in the model.

We used χ^2 statistics to compare the characteristics between our study sample and the Massachusetts birth population and to evaluate the associations among covariates. We performed collinearity diagnostics and examined potential interactions among covariates. All analyses were done using SAS 8.01 (SAS Institute, Inc, Cary, NC).¹⁵ Unless noted otherwise, all of the differences discussed in the text and tables are significant at the .05 level.

RESULTS

Characteristics of Study Population

Most of the 1492 Massachusetts children born in 2002–2003 who did not pass hearing screening were of normal birth weight (87%; Table 1), unilateral referrals for diagnostic evaluation (71%), born to mothers who were aged ≥ 20 years (92%), white (62%), married (65%), nonsmokers during pregnancy (92%), completed at least a high school education (83%), or were privately insured (60%). More infants who did not pass hearing screening lived in the metro west (24%) or southeastern (23%) regions, whereas fewer lived in western (5%) Massachusetts.

These characteristics were also found among most of the 385 Massachusetts children born in 2002–2003 who were diagnosed with hearing loss. Furthermore, 64% of these children had bilateral hearing loss, and 71% of them had mild or moderate hearing loss. Higher proportions of them lived in the northeastern (22%), metro west (22%), or southeastern (20%) regions of Massachusetts.

Characteristics of all of Massachusetts's births in 2002–2003 were included in Table 1 as a reference. Infants who did not pass hearing screenings or who were

TABLE 1 Characteristics of Study Population: Massachusetts 2002–2003

Variable	Did Not Pass Hearing Screening		Diagnosed With Hearing Loss		Characteristics of Massachusetts Births, %
	N	%	N	%	
Total	1492	100	385	100	
Infant factors					
Birth weight					
≥2500 g	1298	87 ^a	307	80 ^a	92
<2500 g	194	13 ^a	78	20 ^a	8
Laterality of hearing referral or loss					
Bilateral	429	29	247	64	NA
Unilateral	1063	71	138	36	NA
Degree of hearing loss					
Severe or profound	NA	NA	112	29	NA
Mild or moderate	NA	NA	273	71	NA
Maternal factors					
Age					
≥20 y	1374	92 ^a	352	91 ^a	94
<20 y	118	8 ^a	33	9 ^a	6
Race or ethnicity					
White	929	62 ^a	256	66 ^a	72
Nonwhite	563	38 ^a	129	34 ^a	28
Marital status					
Married	980	65 ^a	255	66 ^a	73
Not married	512	34 ^a	130	34 ^a	27
Smoked during pregnancy					
No	1374	92	360	94	92
Yes	118	8	25	6	8
Education					
At least high school	1249	83 ^a	319	83 ^a	87
Less than high school	243	16 ^a	66	17 ^a	13
Source of delivery payment					
Private insurance	898	60 ^a	264	69 ^a	76
Public insurance	472	32 ^a	114	30 ^a	22
No insurance	122	8	7	2	2
Residential regions					
Boston	270	18 ^a	55	14	13
Western	70	5 ^a	36	9	11
Central	203	14	49	13	14
Northeastern	257	17 ^a	84	22	21
Metro west	354	24	85	22	23
Southeastern	338	23 ^a	76	20	18

NA indicates not applicable.

^aData were significantly different from Massachusetts birth population on the basis of χ^2 statistics.

diagnosed with hearing loss were different from the Massachusetts birth population in birth weights and most maternal factors evaluated in this study (age, race or ethnicity, marital status, educational attainment, and source of delivery payment). Furthermore, infants born in Boston or southeastern Massachusetts were less likely to pass hearing screening, whereas those living in western Massachusetts were more likely to pass hearing screening.

Loss to Follow-up on the Use of Audiologic Evaluation Services
Eleven percent ($n = 157$) of Massachusetts children born in 2002–2003 who did not pass hearing screening were

TABLE 2 Associations of Maternal and Infant Factors With Loss to Follow-up on the Use of Audiologic Diagnostic Evaluation Among Infants Who Did Not Pass Hearing Screening: Massachusetts 2002–2003 (N = 1492)

Variable	Receiving No Diagnostic Services		Relative Risk (95% CI)	
	n	%	Crude	Adjusted ^a
Total	157	11		
Infant factors				
Birth weight				
≥2500 g	133	10	Reference	Reference
<2500 g	24	12	1.21 (0.80–1.82)	0.98 (0.66–1.44)
Laterality of hearing referral				
Bilateral	52	12	Reference	Reference
Unilateral	105	10	0.81 (0.60–1.11)	0.76 (0.56–1.05)
Maternal factors				
Age ^b				
≥20 y	134	10	Reference	Reference
<20 y	23	19	2.00 (1.34–2.98)	1.08 (0.73–1.60)
Race or ethnicity				
White	75	8	Reference	Reference
Nonwhite	82	15	1.80 (1.34–2.42)	1.54 (1.11–2.21)
Marital status ^c				
Married	70	7	Reference	Reference
Not married	87	17	2.38 (1.77–3.20)	1.18 (0.78–1.79)
Smoked during pregnancy				
No	129	9	Reference	Reference
Yes	28	24	2.53 (1.76–3.63)	1.49 (1.01–2.18)
Education ^c				
At least high school	105	8	Reference	Reference
Less than high school	52	21	2.55 (1.88–3.45)	1.36 (0.97–1.93)
Source of delivery payment				
Private insurance	57	6	Reference	Reference
Public insurance	90	19	3.00 (2.20–4.11)	1.82 (1.19–2.77)
No insurance	10	8	1.29 (0.68–2.46)	1.09 (0.58–2.03)
Residential regions				
Boston	19	7	Reference	Reference
Western	18	26	3.65 (2.03–6.58)	3.75 (2.01–7.01)
Central	11	5	0.77 (0.37–1.58)	1.09 (0.52–2.27)
Northeastern	31	12	1.71 (0.99–2.96)	2.24 (1.30–3.85)
Metro west	21	6	0.84 (0.46–1.54)	1.40 (0.76–2.60)
Southeastern	57	17	2.40 (1.46–3.93)	2.93 (1.75–4.86)

^aData were adjusted for variables in the table.

^bData were analyzed when maternal education and marital status were excluded in the model.

^cData were analyzed when maternal age was excluded in the model.

not reported to have used audiologic evaluation services by any of the MDPH-approved ADCs (Table 2). Among the 1335 children who used audiologic evaluation services, 91% ($n = 1209$) received services by 3 months of age. The median time between the date of not passing a hearing screening and the first diagnostic evaluation was 28 days (range: 1–1036 days; data not shown).

In the bivariate analyses, we found that infants were at higher risk of becoming lost to follow-up on the audiologic evaluation if they were born to mothers who were teenagers compared with mothers aged ≥20 years, nonwhite compared with white mothers, unmarried compared with married mothers, smokers compared with nonsmoking mothers, mothers with less than a

high school education compared with mothers with at least a high school education, publicly insured compared with the privately insured mothers, or those living in the western or southeastern Massachusetts compared with mothers in the Boston region. After adjusting for the potential confounding effects, these disparities remained among infants those born to women who were racial or ethnic minorities, smokers during pregnancy, or covered by public insurance. Specifically, nonwhite infants were 1.5 times more likely to become lost to follow-up on audiologic evaluation than their white counterparts (95% CI: 1.1–2.2). A similar risk was observed among infants born to women who smoked during pregnancy when compared with those whose mothers did not smoke (aRR: 1.5; 95% CI: 1.0–2.2). Infants born to women with public insurance were almost twice as likely as those born to the privately insured to become lost to follow-up on audiologic evaluation (aRR: 1.8; 95% CI: 1.2–2.8).

Furthermore, geographic variations in the use of audiologic evaluation services were also observed for infants who did not pass their hearing screening. After adjusting for the covariates, infants from western, northeastern, or southeastern Massachusetts were 2 to ~4 times more likely to become lost to follow-up on audiologic evaluation than those living in the Boston region (western aRR: 3.8, 95% CI: 2.0–7.0; northeastern aRR: 2.2, 95% CI: 1.3–3.9; southeastern aRR: 2.9, 95% CI: 1.8–4.9).

Loss to Follow-up for EI Referrals

Of the 385 Massachusetts children born in 2002–2003 who were diagnosed with hearing loss, 294 (76%) had not passed their hearing screening (20% had passed and 4% missed screening or had missing data; Fig 1). The median age of diagnosis of hearing loss was 1.2 months for those who had not passed hearing screening, 7.7 months for those who had passed screening, and 8.7 months for those who missed screening or whose screening data were missing.

Twenty-five percent ($n = 98$) of Massachusetts children with hearing loss did not receive EI referrals (Table 3). Infants born with normal birth weight were 2.1 times more likely to use no EI services on the diagnosis of hearing loss than those born with low birth weights (95% CI: 1.2–3.8). Furthermore, infants with milder forms of hearing loss had increased risk of becoming lost to follow-up on EI services compared with their counterparts who have more severe forms of hearing loss. Compared with those with bilateral hearing loss, infants with unilateral hearing loss were 2.5 times more likely to receive no EI referrals (95% CI: 1.8–3.5). Those with milder degrees of hearing loss (mild, moderate, high frequency, or degree not determined) were 1.9 times more likely to become lost to follow-up on EI referrals than infants with severe or profound hearing loss (95% CI: 1.2–3.0). The use of EI referrals for infants with hearing loss also varied by residence regions in Massachusetts. Children living in the southeastern or Boston region were 1.8 times more likely to go without EI

TABLE 3 Associations of Maternal and Infant Factors With Loss to Follow-up on the Use of EI Services Among Infants Diagnosed With Hearing Loss: Massachusetts 2002–2003 (N = 385)

Variable	Receiving No EI Referrals		Relative Risk (95% CI)	
	N	%	Crude	Adjusted ^a
Total	98	25		
Infant's factors				
Birth weight				
≥2500 g	10	13	Reference	Reference
<2500 g	88	29	2.24 (1.22–4.09)	2.08 (1.16–3.76)
Laterality of hearing loss				
Bilateral	39	16	Reference	Reference
Unilateral	59	43	2.71 (1.91–3.83)	2.47 (1.75–3.50)
Degree of hearing loss				
Severe or profound	15	13	Reference	Reference
Mild or moderate ^b	83	30	2.27 (1.37–3.76)	1.90 (1.20–3.03)
Maternal factors				
Age ^c				
≥20 y	90	26	Reference	Reference
<20 y	8	24	0.95 (0.51–1.78)	0.80 (0.41–1.57)
Race or ethnicity				
White	58	23	Reference	Reference
Nonwhite	40	31	1.37 (0.97–1.93)	1.19 (0.84–1.69)
Marital status ^c				
Married	65	25	Reference	Reference
Not married	33	25	1.00 (0.69–1.43)	0.85 (0.56–1.28)
Smoked during pregnancy				
No	93	26	Reference	Reference
Yes	5	20	0.77 (0.35–1.73)	0.85 (0.40–1.81)
Education ^d				
At least high school	78	24	Reference	Reference
Less than high school	20	30	1.24 (0.82–1.87)	1.12 (0.74–1.70)
Source of delivery payment				
Private insurance	64	24	Reference	Reference
Public insurance	34	28	1.16 (0.81–1.65)	0.95 (0.64–1.39)
No insurance				
Residential regions				
Boston or southeastern	50	38	2.02 (1.44–2.82)	1.82 (1.30–2.55)
Other regions	48	19	Reference	Reference

^a Data were adjusted for variables in the table.

^b Data include mild, moderate, and high frequency and degree not determined.

^c Data were analyzed when maternal education and marital status were excluded in the model.

^d Data were analyzed when maternal age was excluded in the model.

referrals than infants living in other Massachusetts regions (95% CI: 1.3–2.6).

Of those 287 infants with hearing loss who used EI referral services, 73% were referred for EI services by 6 months of age, with the median age at referrals being 3.2 months old (range: 0.0–31.9 months; data not shown). A total of 110 children were referred to EI programs for reasons other than confirmed hearing loss (therefore, the EI referral dates occurred before the diagnosis of hearing loss). Of those 177 children whose EI referral occurred after the diagnosis, the median time between the dates of diagnosis of hearing loss and EI referral was 2.4 months (range: 0.0–29.9 months).

No interactions were found in the models. When collinearity diagnostics were performed, all of the vari-

ables had small SEs in the models, and all of the variance inflation factors were <2 , indicating that collinearity is not a problem in our models.

DISCUSSION

Appropriate follow-up with families from screening to definitive diagnosis and intervention is the most important element to ensure success of UNHSPs. Our study reveals that Massachusetts has made great progress in achieving the national EHDI goals, yet there is room for improvement. Approximately 89% of Massachusetts infants who did not pass their newborn hearing screening received an audiologic evaluation compared with the national average of 55%.¹⁶ The use of audiologic evaluation was associated with sociodemographic factors. Infants born to women who were racial or ethnic minorities, had public insurance, or smoked during pregnancy were at higher risk of becoming lost to follow-up on the use of audiologic evaluation services. Furthermore, 75% of Massachusetts infants and children diagnosed with hearing loss receive EI referrals (comparable national data were not available). Instead of social factors, lack of use of EI services was related to a child's health status, including laterality and degree of hearing loss and birth weight.

One of the strengths of our study was the use of population-based data to identify specific factors associated with loss to follow-up. The finding of higher rates of loss to follow-up on the audiologic evaluation among infants born to women covered by public programs emphasizes the importance of collaboration across state programs to reach out to families with the greatest needs. Furthermore, developing strategies to ensure equal access for racial and ethnic or linguistic minorities is necessary for Massachusetts' families to complete the recommended diagnostic follow-up. This includes developing culturally and linguistically sensitive outreach and educational tools to help parents from these groups understand the EHDI process and providing services in languages other than English. Moreover, although it is possible that families in southeastern and western Massachusetts may seek audiologic care from Rhode Island and New York states, geographic variations in the use of both diagnostic and EI services suggest a need to improve the physical accessibility of services. This may include an increase in the number of MDPH-approved ADCs in some regions and/or a provision of transportation services to families residing in areas where no public transportation is available. Although mothers in western Massachusetts whose infants did not pass hearing screening were more likely to have characteristics associated with lack of use of audiologic evaluation services (ie, teenagers, less than a high school education, unmarried, smokers during pregnancy, or publicly insured), geographic effects on the use of audiologic services remained significant after adjusting for these factors. Qualitative analyses of data from focus groups may also be necessary to understand the reasons that prevent families in different geographic regions from using services.

Although expected, the finding of higher rates of loss to follow-up on the EI referrals among children with

milder degrees or unilateral hearing loss indicates a need to reach out to this particular subgroup. It is possible that families that do not choose regular EI services receive developmental monitoring through other venues, such as well-child visits. For the purpose of UNHSP, the MDPH does not consider such monitoring as EI follow-through. Currently, ~30% to 40% of Massachusetts children with mild-to-moderate or unilateral hearing loss do not use EI services. Despite what may be perceived to be a minor problem, children with mild-to-moderate or unilateral hearing loss can have significant deficits in auditory and psycholinguistic skills and school performance without intervention.^{5,17,18} These issues are not unique to Massachusetts. In July 2005, the Centers for Disease Control and Prevention called for a national meeting to raise awareness about mild and unilateral hearing loss among state EHDI programs.¹⁹ Continuously educating providers and parents on the importance of EI services on the development of children with milder or unilateral hearing loss and monitoring their children's program participation are warranted.

One of the potential explanations for the association of certain variables with service use is that provision of services or communication of information may not be the same for infants with and without these characteristics. It is also possible that the failure to follow up may be related to the existence of other serious health issues in addition to hearing loss. However, available data do not allow us to further explore these possibilities. Future studies to examine these mechanisms are necessary.

Our findings were consistent with a previous study examining the 1998 Hawaii UNHSP data, in which maternal age was not a significant predictor of failing to complete the newborn screening and follow-up process.²⁰ In our study, the variation in the use of audiologic evaluation services between teenage and adult mothers is explained by other factors, including being racial and ethnic minorities, being unmarried, smoking during pregnancy, completing less than a high school education, and being covered by public insurance programs (correlation matrices for covariates are available from Dr Liu on request). On the other hand, we did not find the associations between the completion of the follow-up process and low birth weight as reported in the Hawaii study. Differences in defining "loss to follow-up" may explain the discrepancies. Forty-seven percent of the children who failed to complete the follow-up process in the Hawaii study were among the 2% who missed the initial hospital hearing screening (were transferred, had initial inconclusive findings, etc). Given that ~1% of infants missed the initial hearing screening in Massachusetts, our focus for this study was only on those who were referred but failed to go for the audiologic evaluation.

Including multiple variables in the analyses allowed this study to investigate the variance explained by the individual variable alone. We were unable to examine the overlapping variance of covariates. In the first analysis, 3 variables (maternal age, marital status, and educational attainment) lost statistical significance in the adjusted models. In addition to being correlated with

each other, mothers who were teenagers, unmarried, or completing less than a high school education in the first analysis were also more likely to be racial or ethnic minorities, smoking during pregnancy, covered by public insurance programs, or living in western Massachusetts. In the second analysis, all of the variables that were statistically significant in the bivariate analyses retained their significance in the multivariate model, suggesting that the unexplained variance overlapped among covariates is a lesser concern.

We presented results from both bivariate and multivariate analyses to facilitate programs in developing interventions. Although the predictive value on the audiologic evaluation is explained by other factors, as a group, teenaged mothers (a proxy for other factors) may be easier to target for intervention than to identify a group of mothers with a particular characteristic (ie, smoking during pregnancy) that was significant in the multivariate models. MDPH plans to use adjusted estimates to flag high-risk families for priority follow-up and crude estimates for population outreach. Effects such a practice have on improving loss to follow-up will be evaluated.

Our analysis revealed that 20% of the children with diagnosed hearing loss were reported to have passed the screen. The UNHSP conducts systematic data quality assurance reporting with all of the birth hospitals in Massachusetts. For example, facilities are requested to verify screening results for children reported to have passed the screen but who received a diagnostic evaluation. Verifying screening results by chart review is not conducted because of the size of the birth population in Massachusetts. The UNHSP strives to correct as many of the data mistakes as possible, but there remains a possibility of data error. In 2002–2003, >76% of children with hearing loss who passed hearing screening had mild hearing loss, and ~80% were bilaterally affected.

One limitation of this study is our inability to collect audiologic data from facilities other than the MDPH-approved ADCs, such as local otolaryngology (ear, nose, and throat) offices. As a result, we may have overestimated the loss-to-follow-up figure on audiologic evaluation. Furthermore, based on the FIR, $\geq 4\%$ ($n = 62$) of parents of infants who do not pass hearing screening refuse to share audiologic information with the MDPH. Although we could assume that these children have received their initial follow-up audiologic evaluation, we excluded them from the analyses because of the uncertainty. The estimates remained virtually unchanged if they were included in the analyses. To improve the data reporting, the UNHSP staff continues to work with birth hospitals to ensure referrals to the MDPH-approved ADCs and with the ADCs and families to increase the parental consent rates. In addition, the MDPH continues to seek venues to recruit more ADCs that provide audiologic evaluation services to pediatric populations. Limitations are also applicable in accessing data among residents who were born or moved out of state. Massachusetts continues to work with other New England states and the Centers for Disease Control and Prevention to overcome legal barriers for EHDI data sharing among states.

CONCLUSIONS

The Massachusetts UNHSP has excellent follow-up rates overall. The use of population-based data ensures equitable follow-up for all infants at risk for hearing loss. Results from our analyses allow the program to target its efforts and limited resources to the subgroups of infants who are at high risk of becoming lost to follow-up. Recommendations include modification of the data system to flag high-risk families for priority contact, collaboration with other state programs, development of culturally and linguistically sensitive outreach and educational materials, and improvement in transportation and in the number and location of ADCs. Because EI services are available to all Massachusetts infants and children with hearing loss, educating providers to make referrals and parents to consent for services may considerably improve service use. Future efforts should also include qualitative analysis to identify issues and barriers that families may face in receiving EHDI services.

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