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Infant vocalizations and the early diagnosis of severe hearing impairment

Rebecca E. Eilers, PhD, and D. Kimbrough Oller, PhD

From the Mailman Center for Child Development and the University of Miami Ear Institute, Departments of Psychology, Pediatrics, and Otolaryngology, University of Miami, Miami, Florida

To determine whether late onset of canonical babbling could be used as a criterion to determine risk of hearing impairment, we obtained vocalization samples longitudinally from 94 infants with normal hearing and 37 infants with severe to profound hearing impairment. Parents were instructed to report the onset of canonical babbling (the production of well-formed syllables such as “da,” “na,” “bee,” “yaya”). Verification that the infants were producing canonical syllables was collected in laboratory audio recordings. Infants with normal hearing produced canonical vocalizations before 11 months of age (range, 3 to 10 months; mode, 7 months); infants who were deaf failed to produce canonical syllables until 11 months of age or older, often well into the third year of life (range, 11 to 49 months; mode, 24 months). The correlation between age at onset of the canonical stage and age at auditory amplification was 0.68, indicating that early identification and fitting of hearing aids is of significant benefit to infants learning language. The fact that there is no overlap in the distribution of the onset of canonical babbling between infants with normal hearing and infants with hearing impairment means that the failure of otherwise healthy infants to produce canonical syllables before 11 months of age should be considered a serious risk factor for hearing impairment and, when observed, should result in immediate referral for audiologic evaluation. (J Pediatr 1994;124:199-203)

Undiagnosed hearing loss in infancy is not uncommon; the average age of diagnosis in the United States approaches 3 years.\(^\text{1}\) Referral by pediatricians for audiologic evaluation sometimes occurs only after repeated requests by parents, or after recognition of late onset of meaningful speech during the second or third year of life. Consequently, infants often are not provided with hearing aids or other habilitative devices during a key period in which language is naturally acquired. It is possible, however, to identify a small group of infants at very high risk for severe hearing impairment during the first year of life. Infants with severe to profound hearing losses fail to produce canonical (well-formed) babbling before 11 months of age, whereas infants with normal hearing almost always babble canonically before 11 months of age.\(^\text{2}\) The absence of this easily recognized vocal behavior can serve as a diagnostic marker of hearing impairment years before the failure of the appearance of normal speech.

Infants with normal hearing produce vocalizations in stages of development that have been described by Stark\(^\text{3}\) and Oller.\(^\text{4,5}\) These stages reveal step-by-step progress in the vocal capacities required for the production of speech-like syllables. Four clear stages can be discerned, the last of which, the canonical stage, marks the onset of mature (well-formed) syllables that appear commonly between 5 and 8 months of age. Once the infant reaches the canonical stage, he or she typically produces strings of syllables that are so speechlike that parents often assert that the infant is

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Reprint requests: Rebecca E. Eilers, PhD, Mailman Center for Child Development, PO Box 016820, Miami, FL 33101.

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talking. These syllables are not used referentially (i.e., in association with word meaning) or instrumentally during the middle of the first year of life. However, the syllables do meet the phonetic requirements of well-formed speech sounds in any of the world’s spoken languages.

Early claims that auditory experience has little effect on early vocal behavior of infants\(^6\) have been replaced by evidence that auditory experience appears to be critical to the development of mature syllables. Infants who are deaf have significant delay in the onset of canonical stage vocalizations.\(^3\)\(^,\)\(^7\)\(^-\)\(^10\) Furthermore, there appear to be consistent differences in precanonical vocalizations of deaf infants and hearing infants,\(^11\) as well as different sound production preferences once the canonical stage is reached.\(^10\)\(^,\)\(^12\)

The differences observed between the vocalizations of hearing infants and deaf infants are not subtle; rather, they are salient and striking. The onset of the canonical stage virtually always occurs by 10 months of age in normally developing hearing infants; no deaf infant has yet been found to begin the canonical stage before 11 months, and often the onset is much later.\(^2\) Other infants who have near-normal audition but pervasive developmental delay (e.g., infants with Down syndrome) have a slight delay in the onset of the canonical stage but overlap considerably with normally developing infants in age at onset.\(^13\) Healthy infants born prematurely, as well as infants in families of low socioeconomic status, have no delay in onset of the canonical stage.\(^14\)

Despite the striking differences between normally developing and hearing-impaired infants, few deaf infants have been studied extensively and relatively small numbers of hearing infants have been evaluated longitudinally for comparison purposes. In our earlier work we reported a longitudinal evaluation of 21 hearing infants and 7 deaf infants who were followed during the first 2 years of life, providing enough detail to determine accurately the age at onset of the canonical stage.\(^2\) Since that time we have enlarged our sample considerably to include additional deaf infants and hearing infants, many of whom were born prematurely and others of whom were from families of low socioeconomic status. We hypothesized that the onset of canonical babbling is robust, appearing reliably before 11 months of age in hearing infants, regardless of gestational age at birth or socioeconomic status. The specific purpose of the study was to determine whether late onset of canonical babbling could be used as a criterion to determine risk status for hearing impairment during routine well-baby visits during the first year of life.

**METHODS**

**Subjects**

*Normally hearing subjects.* Seventy-three infants were followed longitudinally from the age of 2 months. Infants were recruited by mail solicitation to participate in a study of vocal development. Only infants with unremarkable perinatal and postnatal histories were studied. Twenty-one infants were preterm. Selection criteria for the preterm infants included birth weight between 1400 and 2100 gm and absence of either congenital defects or perinatal or postnatal complications. Infants were excluded if they had evidence of intraventricular hemorrhage, hypoxia, respiratory distress syndrome, severe hyperbilirubinemia, or other medical complications requiring significant medical intervention, or if their 5-minute Apgar score was less than 7. All ages for preterm infants were corrected for duration of gestation. Gestational age estimates were based on postnatal examinations\(^15\)\(^,\)\(^16\) and yielded a mean age at birth of 33 weeks (range, 28.5 to 35 weeks). The mean birth weight of the preterm infants was 1820 gm (range, 1446 to 2070 gm).

*Subjects with hearing impairments.* Thirty infants with varying levels of hearing impairment ranging from profound (>100 db better-ear pure-tone average) to severe (80 to 90 db better-ear pure-tone average) were recruited from an infant-parent educational program for the hearing impaired. Of the 30 infants, 27 had been identified at birth through a screening program for infant hearing\(^17\) (Infant Hearing Impairment Program, Children’s Medical Service, State of Florida, administered through the University of Miami and Jackson Memorial Hospital). For the youngest children, hearing loss estimates were based on auditory brain-stem response testing alone, with absence of a response at 95 dB nHL (normal hearing level) assumed to indicate a loss in the profound range.

**Additional subjects.** In addition to the subjects previously described, seven infants with hearing impairments and 21 normally developing infants from our previous study\(^2\) were included in the analyses. Data from these infants were collected in a fashion similar to data from the new group of infants. Because of the rarity of detailed records of deaf and hearing infants’ vocalizations, it seemed important to describe the results for as many infants as possible. (A table listing the cause, age at identification of hearing impairment, age at fitting with hearing aid, and level of hearing loss of both groups of hearing-impaired subjects is available from the authors.)

**Sampling and recording procedures.** The term infants with normal hearing were seen every other week, and the preterm infants were seen monthly by project staff members. In addition, each infant had laboratory evaluation on 5 consecutive days after the parent-designated onset of the canonical stage to ensure accuracy of age at onset assigned to each infant.\(^3\)\(^,\)\(^18\)\(^,\)\(^19\) Parents had been taught to recognize canonical babbling and had been instructed to call the project staff as soon as it was noted. Canonical syllables were described as instances of infants' “talking” but with-
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Fig. 1. Onset of canonical babbling in severely to profoundly hearing-impaired infants and in hearing infants. Data on preterm infants are shown at corrected ages.

out meaning, using well-formed syllables or sequences of syllables. Examples of “dada,” “meme,” “baba,” “ma,” and “ya” were given. After notification by a parent or identification of canonical babbling during a monthly visit, the infant’s vocalizations were recorded for stage verification. Each recording session was 20 to 30 minutes in duration and consisted of the infant’s interacting with project staff and accompanying family members in a sound-treated room. An attempt was made to obtain a minimum of 50 utterances per recording. If the 50-utterance minimum was not reached, or if the parent reported that the vocalizations were atypical of the infant, the session was rescheduled. Infants were designated to be in the canonical stage if, and only if, repeated occurrences of canonical syllables were observed during 5 consecutive days after onset. In all but two cases, parental reports were verified in the laboratory evaluations. In two cases, canonical babbling was documented by project staff at a later time than at the initial report. Parents proved to be reliable and timely informants concerning their infants’ use of canonical syllables.

The vocalizations of infants with hearing impairments were also recorded monthly by the same procedures as previously outlined. In addition, most of the infants with hearing impairments were enrolled in either a parent-infant educational program for the hearing impaired or a full-day educational program. In either case, the infants were seen either weekly or daily by a speech therapist. Vocalization samples were collected in the context of these programs, and the speech therapists reported the onset of canonical syllables as soon as they occurred. The specification of the onset of the canonical stage was more difficult for the infants with hearing impairments than for the infants with normal hearing. Five of the infants with hearing impairments had not reached the canonical stage by the time of this report, although they were older than 12 months of age. Some infants with hearing impairments had an erratic onset of the canonical stage, producing canonical syllables on some days but failing to produce them in subsequent samples, or producing them with lower frequency than infants with normal hearing in the canonical stage. Nevertheless, for the purposes of this study, once canonical syllables occurred, even erratically, infants with hearing impairments were said to be in the canonical stage. Thus the criterion for stage assignment was more stringent for the infants with normal hearing than for the infants with hearing impairment.

RESULTS

Figure 1 illustrates that each of the 94 preterm or term infants with normal hearing began canonical babbling before 11 months of age. The onset of canonical babbling for infants with normal hearing ranged from 3 to 10 months (mode, 7 months). In contrast, none of the 37 infants with hearing impairment had an age at onset of the canonical stage before 11 months of age. The onset of canonical babbling for infants with hearing impairment ranged from 11 to 49 months (mode, 24 months). Thus there was no overlap in the distribution of onset of canonical babbling for deaf infants and hearing infants, despite the fact that the crite-
The key results of this study are as follows: (1) infants with severe to profound hearing impairment are significantly delayed in the onset of canonical babbling, a precursor to the development of meaningful speech; (2) there is no overlap in onset distribution of canonical babbling between hearing-impaired and normally hearing infants, and the failure of canonical babbling to start may be taken as a marker of extreme risk of severe to profound hearing impairment; (3) parents are good informants regarding their infants’ ability to produce canonical syllables; and (4) the attainment of the canonical stage in infants with hearing impairment is correlated with age at amplification and intervention, suggesting that early identification may be critical to successful language acquisition.

Although universal newborn hearing screening has been recommended by the National Institutes of Health Consensus Conference,3 hearing screening is still limited, and typically only high-risk infants are screened. Fully half of all infants with hearing impairments do not have neonatal or historical risk factors at the time of screening and are thus missed.1 The results of this study suggest that infant vocalizations may play an important role in the assessment of infant hearing during the first year of life, because a large number of infants receive no hearing screening, and the pediatrician may use the lack of canonical vocalizations as an indicator to refer infants with suspected severe to profound hearing loss by the time they reach 11 months of age. Infants who do not, by parental report, produce canonical syllables by 11 months should be referred for audioligic evaluation. Such referral may result in identification of hearing impairment nearly 2 years earlier than the average age at diagnosis in the United States.1

Pediatricians may delay referrals for audioligic assessment even when given parental reports of an infant’s failure to develop early language (first words and primitive sentences). The pediatrician’s hesitation is often based on knowledge of the large individual differences in language development in normally developing infants and young children. Language delays may, in fact, be based on a wide variety of factors in addition to hearing loss. To our knowl-

**Fig. 2.** Relation between age at onset of canonical babbling and age at auditory amplification in hearing-impaired infants and young children.
edge, delays in the onset of canonical babbling beyond 11 months of age, however, rarely occur in otherwise normally developing infants and are of sufficient concern to warrant immediate audiologic evaluation by means of conditioned behavioral techniques such as visual reinforcement audiometry\(^{20,21}\) and, if necessary, auditory brain-stem response audiometry.\(^{20,21}\) Prompt identification by the first birthday will allow the child to benefit from early amplification and intervention, and will maximize the child's potential for oral language development by aiding the child during the period in which language is normally learned.

Although early infant vocal behavior often provides a window from which to observe severe to profound hearing loss, it is not yet clear how less severe losses might affect canonical babbling. It is probable that less severe losses are less disruptive of normal vocal development; thus the infant with moderate hearing impairment may not appear to have delayed onset of babbling and yet may have significant delays in speech and language development.\(^1\) Similarly, infants with progressive, slow-onset losses (such as those often associated with cytomegalovirus infection) or late-occurring losses (such as those associated with meningitis) may not be identified if only canonical babbling is used to assess risk. Consequently, it is important for all infants to be screened for hearing loss early in the second year of life—or sooner if risk factors associated with hearing loss have been identified.

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