Interrupted Infantile Apnea: Impact on Early Development, Temperament, and Maternal Stress

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Abstract:
The early cognitive and motor development and temperament characteristics of 25 infants with interrupted infantile apnea and the degree and source of stress experienced by their mothers were compared to the same dimensions in a sample of 25 control infants and their mothers. No significant differences in development or temperament were found between apneic and control infants with the following exceptions. Contrary to previous research, apneic infants were perceived as more active during sleep. In addition, apneic infants were perceived by their mothers as less "acceptable" than were control infants, contributing to increased stress. There were no significant differences on maternal characteristics contributing to stress with the exception of the high degree of social isolation experienced by the mothers of apneic infants. The findings suggest that, although there were apparently no present developmental ramifications of the apnea, these infants are at risk for being perceived more negatively. In addition, continued support services are needed to alleviate the social isolation experienced by the families of these infants.

Key words: apnea, cognitive development, maternal stress, temperament

Article:
The infant with interrupted apnea of unknown etiology, often labeled *interrupted infantile apnea* (IIA), *near-miss* for Sudden Infant Death Syndrome (SIDS), or *high risk* for SIDS, presents a complex medical and psychological challenge to the multidisciplinary health care team. Prolonged apnea is defined as cessation of breathing for at least 20 sec or a briefer episode of apnea associated with bradycardia, cyanosis, or pallor (American Academy of Pediatrics Task Force on Prolonged Infantile Apnea, 1985). The emotional impact on the parent(s) of discovering their limp, pale, or cyanotic infant, who may be revived only after vigorous manual or auditory stimulation and/or mouth-to-mouth resuscitation, is great (Bendell, Shelton, Krous, & Shirley, 1984). Furthermore, the resultant hospitalization and medical evaluation of the child and the subsequent adjustment to any prescribed treatment or preventative regimen (e.g., cardiopulmonary resuscitation or CPR, home monitoring) puts the family at increased risk for stress. As the apneic population has been identified only recently, questions remain regarding the ramifications of apnea and possible effects of subsequent oxygen desaturation on the infant's development, as well as concerns regarding the family's adjustment to this medical problem. Thus, the purpose of our investigation was to examine dimensions of the apneic infant's development and temperament style, aspects of the mother-child interaction, and maternal adjustment to monitoring.

Infant Development
As infants with IIA may have repeated hypoxic episodes, it is possible that these episodes may have an adverse effect on cognitive and motor development. Unfortunately, there is little research available addressing this question. Black, Steinschneider, and Sheehe (1979) reported on the development of a group of 28 infants 9 months old who were on home apnea monitors. Each infant was evaluated on the Bayley Scales of Infant

Development. None of the infants had significant delays in cognitive or motor development. However, infants with increased respiratory instability during the first week of life averaged significantly lower on both the mental and motor scales relative to those infants without respiratory instability. Although there are no longitudinal data available, Guilleminault (personal communication, October 1982) posits that apneic children might, as a group, be more highly represented in future learning-disabled populations. Thus, it is the purpose of our investigation to examine the possible ramifications of apnea on infant development. It was hypothesized that infants diagnosed as having infantile apnea would evidence greater developmental delays than would infants without apnea.

Temperament

A second area of interest involved the temperament style of the infants and their mothers. Because apneic infants present unusual problems for their families, we examined whether these infants would be seen as displaying a characteristic temperament pattern, in particular, a "difficult" pattern. Previous research in this area has provided somewhat contradictory findings. A study by McCaffree, Bendell, Shelton, and Mattice (1983) indicated that families with an apneic infant on a home-monitoring system perceived the infant as more passive than their other children. Similar observations were made by Naeye, Messmer, Specht, and Merritt (1976), who described the temperament of infants who later died of SIDS as lethargic (i.e., less intense and less active) in comparison with their siblings. Given the possible link (yet to be supported) between SIDS and apnea and the questionable validity of retrospective behavioral data, however, these findings do not provide clarification regarding the temperament of apneic infants. In an attempt to obviate these difficulties, Weissbluth, Brouillette, Liu, and Hunt (1982) prospectively investigated temperament characteristics of apneic infants, asymptomatic siblings of SIDS victims, and normal infants. In contrast to previous studies, no significant differences were found among the three groups. Within the apneic group, however, ratings of low activity and low intensity were correlated significantly with increased apnea and increased frequency of periodic breathing during sleep. Thus, we sought to provide additional data regarding the perceived temperament characteristics of these infants. Based on this available research, it was predicted that apneic infants may be rated as less active and less intense than controls.

In addition to the influence of the infant temperament on the resultant parent-child interaction, Thomas and Chess (1977) and others provided data demonstrating the importance of the match or "goodness-of-fit" between the infant's temperament and contextual demands (e.g., type of home environment, school demands, parents' temperament). In one study examining the importance of this match for later child adjustment, Scholom, Zucker, and Stollak (1979) reported that temperament similarity between parents and daughters was related to later adjustment in preschool girls, whereas father-son temperament dissimilarity was significantly related to adjustment in boys. Therefore, in addition to the data on temperament characteristics in general, the goodness-of-fit between mother-infant pairs was examined. We predict closer goodness-of-fit for control dyads than for apneic dyads.

Family Adjustment to Monitoring

Although there is a large body of literature focusing on the impact of the monitor (Black, Hersher, & Steinschneider, 1978; Cain, Kelly, & Shannon, 1980; McCaffree et al., 1983), few studies have examined other areas of stress and familial adjustment. In addition to the stress engendered by the infant's medical condition and the potential interferences in family life by the monitor, the differences of opinion within the medical community regarding the use of home monitors (American Academy of Pediatrics, 1985; Krous & Bendell, 1982) provide yet another potential source of stress. Furthermore, all physicians are advised to inform parents that even if their child is monitored, this cannot guarantee their child against SIDS (Duffty & Bryan, 1982). Because of the myriad of potential sources of stress, we sought to evaluate the impact of the apneic infant on maternal stress. More specifically, we hypothesized that mothers of apneic infants would perceive greater total stress than mothers of control infants.

Method

Subjects

Twenty-five infants with IIA, between 4 and 8 months chronological age, were selected on the following basis. Infants who had suffered a clinical HA episode were evaluated using an overnight sleep study in which the number and duration of apneic episodes were documented with 12-hr overnight polysomnographic techniques. Infants were placed in the Oklahoma Children's Memorial Hospital (OCMH) home-monitoring program if the results of their sleep study by 24-hr pneumogram were greater than norms of age-matched controls for duration and rate of central, mixed, and obstructive apneic episodes (Guilleminault et al., 1979). No families of infants selected for the study refused participation.

Assessment of recordings of respiratory efforts in this population revealed that central apneic rates greater than 6 sec occurred at a mean rate of 9.41 times per hour. Obstructive apneic rates occurred at a rate of 5.17 times per hour. Five of the infants (20%) were also identified as a subsequent sibling of SIDS with 10 infants (40%) being born prematurely. Eleven infants had respiratory difficulty at birth, and 3 had to be resuscitated using CPR subsequent to the identifying HA episode(s). Infants were monitored for an average of 8.9 months, with duration of monitoring at the time of the study at 3.9 months. All infants with medical problems in addition to the apnea (e.g., seizure disorder, surgically corrected gastroesophageal reflux) were excluded from the study. Twenty-five control infants, matched for gestational age, sex, socioeconomic status of the parents, and educational level of mothers, were obtained through the Pediatric Practice Model Clinic at OCMH. Three families selected for the control group refused participation in the study. Informed consent was obtained from all parents prior to enrollment in the study.

Measures and Procedures

The Bayley Scales of Infant Development, including the Mental scale, Motor scale, and Infant Behavior Record (Bayley, 1969), were administered to infants in a clinic examining room with the mother present. For the premature infants, performance on the Bayley scales was adjusted according to gestational age.

Infant and maternal temperament were assessed using the Dimensions of Temperament Survey (DOTS) for infants (Windle, Lerner, & Belsky, 1983) and for adults (Lerner, Palermo, Spiro, & Nesselroade, 1982). Derived from the theoretical and empirical bases provided by Thomas and Chess (1977), the 34-item questionnaire yields five statistically independent factors found to be highly congruent across age groups: Activity, Attention Span/Distractibility, Adaptability/Approach/ Withdrawal, Rhythmicity, and Irritability. These measures were chosen to assess temperament due to the similarity between the adult and infant versions, thereby allowing a more direct assessment of goodness-of-fit. Internal consistency, as estimated by Cronbach's alpha for the five factors on the adult version, ranged from .37 to .93 (M = .73) and from .53 to .96 (M = .75) for infants. Test-retest reliabilities are high for adults (.80s) and moderate for infants (.40s).

Finally, the 151-item Parenting Stress Index (PSI; Abidin, 1979, 1981) was administered. This self-report inventory assesses various dimensions of stress within the mother-child dyadic interaction pattern. Concurrent and construct validity range from .78 to .84. Reliability of the various subscales ranges from .62 to .80. Based on the accumulated research relating to child development, parenting, and stress, this instrument provides information about sources of stress within three major domains: child characteristics, mother characteristics, and situational/demographic characteristics. Within the three major domains are several sub-scales providing additional information on the source of stress. The PSI is designed to identify parent-child systems that are at risk for developing dysfunctional parent and/or child behaviors.

All self-report measures were read to the parents following the administration of the Bayley scales.

Results

Comparison of demographic data for the apneic and control groups revealed no significant differences in family income level, maternal and paternal educational level, or maternal and paternal employment status. In the apneic group, 46% were first born and 52% second born; in the control group, 42% were first born and 28% second born. Within the apneic sample, the length of time the infant was monitored was not significantly related to the apneic rates obtained on pneumograms ($ps > .05$).

Table 1. *Mean Scores of Apneic and Control Infants on the Bayley Scales*

	Apneic	Control	*t*
Mental Scale			
M	108.52 (16.21)	115.40 (14.33)	−1.59
Range	9.68 (6.34)	8.68 (6.34)	0.55
Motor Scale			
M	110.60 (18.73)	111.92 (14.57)	−0.27
Range	3.84 (4.36)	2.04 (2.94)	1.17

Notes: For both the apneic and control groups, $n = 25$. Standard deviations are in parentheses.

Cognitive and Motor Development

There were no significant differences between the apneic and control groups on the mean mental and motor development indices of the Bayley scales (see Table 1). However, scores for two of the apneic infants fell within the borderline range, between 1 SD and 2 SD below average, on both Mental and Motor scales. Neither of these infants' sleep studies deviated from the rest of the sample. In contrast, none of the control infants scored below the average range.

When the range of items passed was calculated, there was slightly greater variability on the Motor scale within the apneic group. The range score was determined for each scale by subtracting the basal item (i.e., the item prior to the first error) from the ceiling item (i.e., the last item passed). The apneic group, in general, demonstrated more splintering of skills on the Motor scale than did controls, although the difference was not statistically significant.

Temperament

On the infant DOTS, there were no significant differences between apneic and control infants on four of the five temperament dimensions (see Table 2). However, contrary to previous findings (Naeye et al., 1976), apneic infants were perceived by their mothers as significantly more active than control infants. However, both groups were within 1 SD of the means reported by Windle et al. (1983).

To determine the relationship between mothers' ratings of their own and their infants' temperament, each mother-infant pair was compared along each DOTS dimension (see Table 3) using the Pearson product-moment correlation coefficient. Mothers of apneic infants rated themselves and their infant similarly on the Adaptability and Rhythmicity factors, whereas control mothers viewed themselves and their infant as similar on the Activity Level and Rhythmicity factors.

Table 2. *DOTS: Apneic Versus Control Infants*

DOTS Factor	Apneic	Control	*t*
Activity Level	1.96	1.08	2.34*
Attention Span/Distractibility	3.32	3.68	−0.55
Adaptability	4.68	4.92	−0.60
Rhythmicity	5.56	5.48	0.14
Reactivity	3.96	3.80	0.39

Note: For both the apneic and control groups, $n = 25$.
*$p \leq .05$.

Table 3. *Temperament "Goodness of Fit" (r) Between Infants and Their Mothers on the DOTS*

DOTS Factor	Apneic	Control
Activity Level	.08	.42*
Attention Span/Distractibility	.15	.36
Adaptability	.39*	−.09
Rhythmicity	.43*	.48**
Reactivity	.08	.11

Note: For both the apneic and control groups, $n = 25$.
*$p \leq .05$. **$p \leq .01$.

Maternal Stress

The group means for the Child and Maternal subscales on the PSI are presented in Tables 4 and 5.

Table 4. *Child Characteristics on the PSI: Apneic Versus Control Groups*

	Apneic			Control		
Stress Measure	***M***	***SD***	**Range**	***M***	***SD***	**Range**
Adaptability	27.59	5.2	16–36	23.36	6.0	11–39
Acceptability	13.59	4.1	7–26	10.88	2.8	7–17
Demandingness	20.66	4.4	13–29	17.00	4.5	12–26
Mood	9.74	3.5	5–19	8.88	3.0	5–18
Distractibility	25.44	5.6	9–34	25.23	4.4	17–35
Reinforcement	9.89	3.4	6–18	8.76	2.5	6–15
Stress	106.89	18.7	60–138	94.12	19.0	60–136

Note: For both the apneic and control groups, $n = 25$.

Table 5. *Mother Characteristics on the PSI: Apneic Versus Control Groups*

	Apneic			Control		
Stress Measure	***M***	***SD***	**Range**	***M***	***SD***	**Range**
Depression	21.26	4.7	15–32	20.84	5.8	9–38
Attachment	12.07	4.2	7–23	11.88	3.3	6–20
Restriction	21.59	5.8	13–33	20.72	5.5	7–31
Competency	31.63	6.0	22–47	29.48	6.3	19–40
Isolation	16.33	4.5	10–25	11.84	4.2	6–20
Spouse	18.26	5.7	9–33	16.80	6.0	7–28
Health	14.59	4.0	10–26	13.92	3.0	10–21
Stress	135.29	24.5	98–196	124.92	25.0	80–188

Note: For both the apneic and control groups, $n = 25$.

The degree of stress of the two groups was evaluated using discriminant analysis. The analysis produced standardized discriminant weights, which can be interpreted as standardized regression coefficients (Kerlinger & Pedhazur, 1973, pp. 341- 345). Based on this analysis, mothers of apneic and control infants differed significantly on the amount of perceived stress, Wilks's lambda = .72, $F(7, 44) = 2.40$, $p < .03$. The only variable on the Child subscale making a significant contribution to the discriminant function was acceptability, $F(1, 49) = 5.80$, $p < .02$. High scores for the acceptability variable occur when the infants' characteristics fail to match parental expectations and hopes. The only variable on the maternal subscale making a significant contribution to the discriminant function was social isolation, $F(1, 44) = 8.11$, $p < .01$. In other words, mothers of apneic infants did not rate themselves as more stressed than mothers of controls, with the exception of the social isolation experienced in the care of their vulnerable infant.

The total PSI rating was significantly higher, 1(1, 24) = 2.59,p < .01, for the mothers of apneic infants than for the controls, indicating a greater level of stress in the apneic group in general (see Table 6). Although the length of time on the monitor has been shown to be correlated with severity and frequency of apneic episodes, (Wasserman, 1984), in our sample the length of time monitored and the apneic rates on the pneumogram were not significantly related.

Discussion

We examined the development and temperament characteristics of a group of apneic infants and the perceived maternal stress related to having an IIA infant. The absence of significant developmental differences between apneic and control infants suggests that serious delays may be ruled out in the IIA group, and this will provide helpful reassurance to parents; but the results may be utilized more appropriately as a baseline measure for comparison with future developmental studies. Longitudinal follow-up will be necessary to determine if more subtle cognitive deficits or learning disabilities will emerge in the apneic population. In addition, future research should examine the relationship between relatively lower developmental scores and such factors as increased frequency or greater severity of apneic episodes.

Regarding the temperament of these infants, previous research has suggested that apneic infants have been perceived by their caregivers as lethargic and, generally, less active than other infants (McCaffree et al., 1983). In contrast to these findings, our data revealed that, on the DOTS, mothers of apneic infants perceived their infants as more active than did the mothers of control infants. Yet there were no significant differences on the PSI Hyperactivity subscale. An analysis of the content of specific items on the DOTS and PSI may provide some information that would explain these differences.

Table 6. *Situational Characteristics on the PSI: Apneic Versus Control Groups*

	Apneic		Control		
Stress Measure	***M***	***SD***	***M***	***SD***	***t***
Situational Stress	30.68	6.16	26.96	5.29	2.29*
Life Events	17.04	12.15	12.64	7.15	1.56
Total Situational Domain	47.72	15.53	39.56	9.51	2.24*
Total PSI	339.76	48.23	303.08	51.77	2.59**

Note: For both the apneic and control groups, $n = 25$.
*$p \leq .05$. **$p \leq .01$.

Mothers' ratings of activity on the DOTS items referred to their infant's level of activity during sleep, whereas on the PSI, mother's ratings of distractibility/hyperactivity were based on more general statements about their infant's activity level during play, feeding, or bathing. Thus, the significant finding on the DOTS may be attributed to the mothers' attention being drawn particularly to their infants' activity level during sleep. Weissbluth et al. (1982) found significantly less night sleep and lower total sleep duration for IIA infants when compared with normal infants and asymptomatic siblings of SIDS infants. It is possible that the use of a monitor may create a feedback loop such that the child's movement and activity, which may trigger the alarm, are more readily, monitored by the parents.

Perhaps more important than the observation of increased activity level in the apneic group is the goodness-of-fit between the temperament of the parents and the actual behavior or, temperament traits of the infant. In our study, mothers' ratings of their own activity level on the DOTS were unrelated to their ratings of their infant's activity. This lack of a relationship may arise from the fact that the mother is particularly focused on and concerned about her infant's sleep activity level. This activity level may be perceived as related to the monitor and frequent nighttime arousals, which may not correlate with the parents' normal sleeping patterns. The apneic mother-infant pairs were very similar in adaptability and rhythmicity as measured by DOTS. Although the ratings on these factors were moderate, they can be interpreted as reflecting the maternal capacity for adapting to new situations, as well as the assumed predictability or orderliness of daily routines.

As noted earlier, there were no differences between the two groups in stress stemming from child characteristics, with the exception that apneic infants were perceived as less acceptable than the control infants. Previous research with the PSI (Abidin, 1983) indicated that parents, including those with children having handicaps and developmental delay, report significant stress stemming from child characteristics. Thus, at least during the 4- to 8-month period of infancy, the apneic infant was seen as less of a stressor to the mother than might have been predicted from previous research. This data can underscore recommendations from the Task Force on Prolonged Infantile Apnea that professionals emphasize normal development so that an infant can be nurtured and enjoyed (American Academy of Pediatrics, 1985). This research encourages the alleviation of negative perceptions of apneic infants. It is possible that the mother's expectation that her child will outgrow the apnea, as opposed to the expectations of parents who have a chronically handicapped child, has positive implications for her ability to separate her "infant" from the infant's "disorder."

One of the most significant findings on parenting stress was the lack of significant differences in stress related to mother characteristics, with the exception of social isolation. Anecdotal evidence indicated that mothers of apneic infants sometimes imposed the isolation upon themselves as they expressed reluctance to leave their infants in the care of even close relatives. Other mothers experienced difficulty securing baby-sitters who were competent in CPR and in the care of an apneic infant on a monitor. Numerous studies have documented the effects of the lack of, or loss of, social network support in increasing the risk for dysparenting, deleterious

health effects, and other undesirable conditions. Assisting parents in securing appropriate baby-sitting services or respite care continues to be problematic in apnea programs.

Focus for Intervention and Future Research

Professional intervention by clinical child psychologists with families of apneic infants needs to focus on counteracting the effects of social isolation. This intervention may include cultivating the formation of parent support groups and the provision of specially trained baby-sitters (usually trained health care professionals) who are familiar with the use of a monitor. Encouraging two-parent families to share the responsibility, as well as having other family members or friends trained in CPR techniques, would serve to lessen the burden of the care. For other families, identification of appropriate respite care facilities would be beneficial. The provision of a home visitor (e.g., public health nurse) could also be of value in bringing the social network into the home, as well as in evaluating the coping mechanisms of families who tend toward greater social isolation. Perhaps the fact that some parents view the monitoring period as temporary may preclude them from utilizing available support and coping mechanisms. Parents must be encouraged to avail themselves of support.

In addition to counteracting the effects of social isolation, professionals need to screen for parenting stress throughout and following the monitoring period. Future research is needed to examine whether the stress factors identified in this study are transient or persistent over time and how they relate to continued apneic episodes. If indeed the stress factors and negative maternal perceptions are related to the apnea and not the infant, one would expect these problems to resolve over time. If not, then apneic infants may be seen as more vulnerable than other children or may continue to be perceived as less acceptable than other children. This would place apneic infants and their families at greater risk for later difficulties in adjustment and interaction. Furthermore, additional research needs to be directed toward early identification and intervention for those families whose degree of anxiety prohibits discontinuation of the monitor once the medical need has resolved (American Academy of Pediatrics, 1985).

Our study was limited to examining maternal perceptions of the infant and the resultant stress factors. The perceptions of fathers and siblings also need to be examined before professionals can adequately understand the effects of apnea on the family system and provide appropriate interventions. In addition, this investigation provided initial baseline data on the development and temperament of apneic infants. Although it is encouraging that few significant differences between the apneic and control infants were found, additional longitudinal studies are needed to predict more accurately the potential outcome for apneic infants in terms of their development and familial adjustment.

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