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## Sudden Infant Death Syndrome: Cry characteristics

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## ABSTRACT

**Objective:** To acoustically evaluate the cries of SIDS infants and compare these cry features to a group of healthy term (HT) infants, as well as previously published results for SIDS infants.

**Methods:** Pain-induced crying episodes were collected from four infants during the first weeks of life that later died of SIDS. Temporal and spectral features of each crying episode were characterized based on measures of cry duration, cry fundamental frequency ( $F_0$ ), and cry formant frequencies ( $F_1$  and  $F_2$ ).

**Results:** The SIDS infants were found to produce cries with longer duration compared to HT infants. The cries of SIDS infants also differed from HT infants in regard to the absolute difference in  $F_2 - F_1$  frequency.

**Conclusions:** The acoustic features considered in the present study support the contention that the cries of SIDS infants are reflective of atypical respiratory-laryngeal control. Although research of this nature is rare, there is evidence to suggest an acoustic profile of crying that is specific to SIDS.

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## 1. Introduction

Since the mid-1990s, the occurrence of Sudden Infant Death Syndrome (SIDS) has decreased dramatically in the United States from a rate of 7000 babies per year to fewer than 2500 per year. The marked reduction in annual SIDS cases in the United States has been attributed, in part, to the baby's sleeping position. The American Academy of Pediatrics [1] established the "Back to Sleep" campaign, which stressed that infants should be placed for sleep in a wholly supine position. Despite this welcomed reduction in death rate, SIDS continues to be the number three cause of infant death after congenital malformations and disorders related to short gestation [2,3]. As such, the need to further reduce the risk of SIDS remains of paramount importance.

Ongoing SIDS research has led to discovery of a number of environmental, developmental and physical risk factors associated with these deaths [4], although no single factor has been identified as increasing the risk of SIDS. As the search for a definitive cause continues, a list of prognostic indicators has been developed. Perhaps most unique among the list of these indicators is crying behavior. Because cry represents a combination of respiratory, laryngeal and vocal tract functions, any unusual or deviant cry patterns are likely to be a reflection of poor organization in either parasympathetic or sympathetic strands of the nervous system [5].

Inferences about the anatomical and physiological behaviors subserving infant cry are possible through the use of acoustic analysis. The most common acoustic features of infant crying that correspond to respiratory, laryngeal and vocal tract functions are cry duration, fundamental frequency ( $F_0$ ) and formant frequencies ( $F_1$  and  $F_2$ ), respectively. The publications by Barr et al. [5] and Soltis [6] provide a comprehensive overview of acoustic cry research.

There are few studies examining the acoustic crying behavior of SIDS infants [7-10]. The low number of acoustic studies is not surprising when considering the unique circumstances surrounding collection of cry samples. However the examination of cries in SIDS infants could provide potentially valuable information about the neurobehavioural integrity demonstrated by infants prior to their sudden and unexpected death. Such information may serve as a predictive risk marker for the syndrome. A survey of these past studies suggests that an acoustic profile of crying behavior in SIDS has yet to be established. For example, Stark and Nathanson [7] obtained cry samples from a 4-day-old infant who later died suddenly at 6-months of age. These researchers found the infant's cries to be of shorter duration and having a higher  $F_0$  than healthy term (HT) infants, indicating the respiratory and laryngeal systems regulating cry function differed between HT and SIDS infants.<sup>1</sup>

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Q2 <sup>1</sup> The infant examined by Stark and Nathanson [8] did not appear to die in the classic fashion reported in other SIDS cases. Rather, the infant died "suddenly" as a result of choking on a plastic toy item. Past acoustic cry studies of SIDS infants have considered the results of Stark and Nathanson as a possible comparative data set. However, it is questionable whether the child exhibited the symptoms often associated with SIDS.

Colton and Steinschneider [8] evaluated the cries in one SIDS infant and observed the cries to be of (1) long duration, (2) with a low  $F_0$ , and (3) low  $F_1$  frequency. These findings indicate that all three subsystems regulating cry are aberrant in SIDS infants compared to HT infants but the direction and magnitude of the differences is contradictory to the Stark and Nathanson findings. Golub and Corwin [9] evaluated two SIDS infants and found cry patterns that did not fully support either of the earlier two studies. Specifically, cry duration and cry  $F_0$  did not differ between SIDS and HT infants; however, the SIDS infants were found to show unusual vocal tract constriction; which presumably was a result of a high  $F_1$  frequency. These findings were supported in a later study by Corwin et al. [10] of 12 SIDS infants but the results are difficult to compare to earlier SIDS studies because specific acoustic values for various acoustic parameters were never reported. Rather, their analysis involved establishing confidence (CI) intervals for the various acoustic features and identifying instances when SIDS cries fell outside the CI for non-SIDS cries.

In addition to the varied findings obtained from acoustic analyses, the qualitative descriptions of crying behavior in SIDS are also mixed. There are reports of SIDS cries being weak [11] or reflective of a floppy mandible [8]. Still, other reports characterize SIDS cries as tense and reflective of vocal tract constriction [7,10]. One likely reason for the differences across studies is that there may be multiple etiologies associated with the sudden unexplained death of an infant [4]. At present, the question as to whether there is an acoustic pattern of crying in SIDS that can be differentiated from the cries of normal HT infants remains unanswered.

Obviously, a major drawback in this area of research is the limited number of cry studies, and the challenges involved in collecting a sample of participants. The present study offers further insight as to the acoustic pattern of crying in SIDS. We were interested in determining whether the inclusion of these new, additional data would provide clarity in regard to the characteristics of crying in SIDS. Therefore, the results were compared to a group of non-SIDS health term (HT) infants, as well as past results reported for SIDS infants.

## 2. Methods

### 2.1. Participants

The SIDS infants examined in this study comprised a portion of the database from the Collaborative Home Infant Monitoring Evaluation (CHIME) Study. The CHIME Study was a multi-center project supported by the National Institute of Child Health and Human Development [12]. Five sleep respiratory physiology (i.e., polysomnography) laboratories from across the US were involved in the CHIME Study, including Chicago, Cleveland, Honolulu, Los Angeles, and Toledo. A primary objective of the CHIME Study was to assess cardio-respiratory events in infants at high epidemiologic risk for SIDS. The Institutional Review Board at each of these sites provided ethical approval for the study. From the extant database, the cries of four infants were selected for analysis (1 female, 3 males). This group comprised the total number of SIDS babies across the five clinical sites for which audio samples of their cries were available. The four cases were reviewed by a medical examiner at each CHIME site, and autopsies were performed to confirm a diagnosis of SIDS. The gestational age for these infants ranged from 28 to 39 weeks with an average age of 32 weeks. Birth weights ranged from 825 to 3690 g with a mean birth weight of 1937 g. Three of the infants had a risk group classification of "preterm" and were small for their gestational age. One infant had a risk group classification of "sibling of SIDS." One infant was recorded at the Toledo site, one from the Los Angeles site, and two were from the Cleveland site.

In addition to the four SIDS infants, a group of 15 HT infants (7 females, 8 males) were examined in the present study and served as a comparison group. The mean gestational age for the HT infants was 39 weeks with a mean birth weight of 3000 g. A portion of the cry data collected for the HT infants has been previously reported in Robb et al. [13,14] and is included in the present study to serve as a comparison to the SIDS infants.

### 2.2. Data collection

The data collection protocol involved audio collection of cry samples from each infant at various times within a 3-month period following birth. Cry recordings took place in the laboratory. Cries were elicited from each infant by applying a painful stimulus to the sole of the infant's right foot. The collection of pain cries was selected over spontaneous (i.e., non-pain) cries for two reasons. First, the collection of pain cries allowed for a standard procedure that could be implemented across all data collection sites (see below). Secondly, there is research to indicate that the neural circuitry differs between pain cries and spontaneous cries [15,16]. An infant's response to pain is one of extreme stress-arousal and is a form of involuntary expression; whereas spontaneous cries reflect a form of voluntary expression [17].<sup>2</sup> Thus, collection of pain cries affords constancy in phonatory behavior reflecting a high state of arousal.

All infants were lying supine and in a quiet, restful state prior to cry recording. The pain stimulus was delivered by a two-inch stainless steel wire attached via a spring to a heel block. The sole of the infant's right foot was placed in the block and the steel wire was cocked and released, providing the pain stimulus to the infant's heel. The same device was used across the five clinical sites so as to ensure a standard form of cry elicitation. An episode of crying commenced with the first audible cry following administration of a pain stimulus. A crying episode was defined as the total period of continuous crying activity [16]. The completion of a crying episode was noted as the last audible cry that was followed by a minimum of 15 s of silence. All cries were audio recorded using a condenser microphone (Realistic 33-1052) situated at a constant 15 cm from the infant's mouth.

### 2.3. Acoustic analysis and measurements

The first three cries (i.e., utterances) comprising each infants overall crying episode were measured. These early occurring cries are assumed to reflect an automatic or "reflexive" response by the infant [18]. Cries occurring during later periods of the crying episode represent a leveling-off of the infant's behavioral response to the cry-eliciting stimulus [19,20]. Accordingly, early occurring cries allow for a better inter-individual comparison of crying behavior. Acoustic analysis of the cry samples was performed by the first author (MPR) using an acoustic analysis software package (Praat ver 5008). The cry samples were digitized at 11 kHz and examined as a dual display on a computer monitor. The displays consisted of an amplitude-by-time waveform and a corresponding wideband spectrogram. The specific measurements made were as follows:

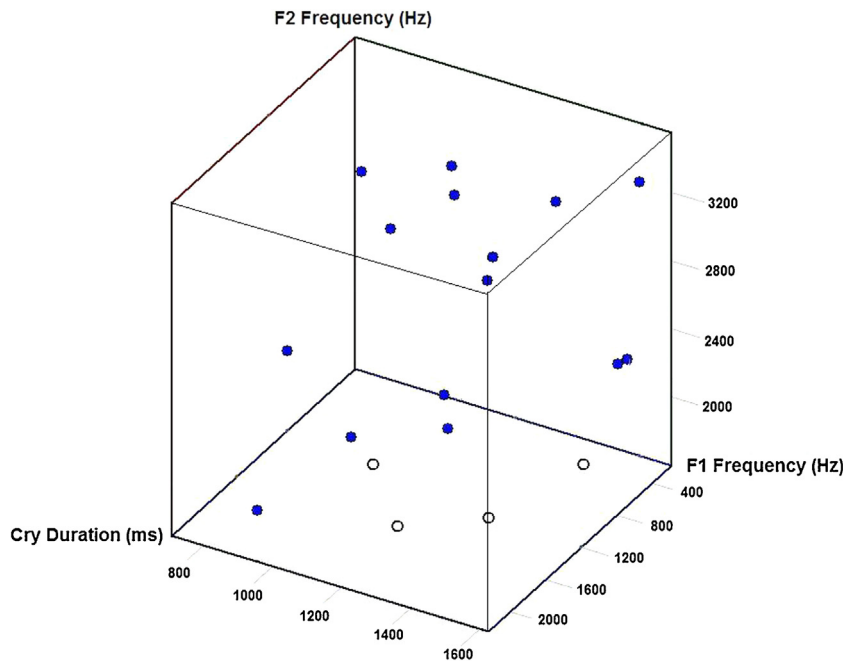
#### 2.3.1. Cry utterance duration

The duration of a cry utterance was measured as the time span separating the amplitude onset and offset points of a perceivable cry occurring on expiratory airflow. A cry utterance was at least

<sup>2</sup> There is a growing body of acoustic cry research that centers on an infant's production of spontaneous (non-pain) cries [17]. The non-distress nature of these cries is thought to provide information regarding the emotional (rather than health) status of the infant, particularly beyond 2 months of age.







**Fig. 1.** Three-dimensional display of the results for analysis of cry duration,  $F_1$  frequency and  $F_2$  frequency for the SIDS infants (open circles) and HT infants (closed circles). Note the general clustering of the SIDS infants.

mechanoreceptors are assumed to be vital for control of a rhythmic breathing pattern. In the case of SIDS, the neural receptors are impaired, whereby the “switch” from end-phase expiration to inspiration is prolonged [22]. While it is important to recognize that the present data set is small, we are intrigued by the possibility that the unusually long cry utterance durations exhibited by the SIDS infants may have been reflective of a disordered breathing pattern.

Previous reports of the cry  $F_0$  surrounding SIDS have ranged from findings of no difference between SIDS and HT infants [9,10], to a significantly low  $F_0$  [8] or a very high  $F_0$  [7]. The present study found no significant difference between infant groups in their cry  $F_0$ . Physiologically,  $F_0$  is reflective of the mass, length, and stiffness of the vocal folds. Colton and Steinschneider’s [8] past report of both a low  $F_0$  and long cry duration in their SIDS case is perhaps not surprising because vocal  $F_0$  is found to decrease naturally as a function of increasing utterance length [23,24]. In the present study, the SIDS infants were found to have a slightly lower  $F_0$  compared to HT infants. These same groups differed in regard to cry duration. So while it seems the same duration- $F_0$  relationship was evident between the present SIDS infants and those of Colton and Steinschneider, we hesitate to conclude that the  $F_0$  of cries produced by the SIDS are uniquely different from those of HT infants. Research suggests there is no increasing or decreasing trend in cry  $F_0$  among normal infants during the first three months of life [25]. Any alterations in  $F_0$  would appear to be linked to the unusually long cry utterance durations (i.e., respiratory behavior) rather than abnormalities in vocal fold vibratory behavior. In addition, it is necessary to mention that the present analysis of  $F_0$  (as well as  $F_1$  and  $F_2$ ) was confined to measurement of the midpoint location of each cry utterance. It is possible that measurement of the entire cry utterance, or other locations within each utterance would have yielded different results. However, this approach was taken to ensure that a noise-free portion of the cry utterance was sampled.

Cry resonance is dictated by the size and shape of the vocal tract. Accordingly, physiological differences in vocal tracts will result in variability of  $F_1$  and  $F_2$  frequency [18]. Previously, Colton and Steinschneider [8] reported a low  $F_1$  and high  $F_2$  frequency in

their SIDS case; while Corwin et al. [10] found a strong pattern of a high  $F_1$  frequency in their SIDS cases. Although there was no significant difference between the present group of SIDS and HT infants according to the median  $F_1$  and  $F_2$  frequencies, there was a clear pattern of a higher  $F_1$  and lower  $F_2$  among the SIDS group.  $F_1$  frequency is correlated with tongue height which affects pharyngeal space [18]. A large mouth opening during cry production would result in a high  $F_1$  frequency. In addition,  $F_2$  frequency is correlated with changes within the oral cavity, primarily tongue retraction [18]. A more retracted tongue during cry production would result in a lower  $F_2$  frequency. The combined effects of  $F_1$  and  $F_2$  frequency were examined in the present study by calculating the absolute (Hz) difference. The results of this analysis identified a significantly smaller  $F_2 - F_1$  difference among the SIDS infants compared to HT infants. This finding could be inferred to indicate a more compact tongue positioning in the vocal tract [16]. The collective results of the formant analyses would appear to support the contention by Corwin et al. that cries of SIDS infants are reflective of atypical vocal tract resonance.

Interestingly, there is a small body of research that has examined infant facial expression during moments of crying [26,27]. In particular, the facial expression exhibited during pain cry has been interpreted in regard to the regulation of negative emotion [28]. The extent of horizontal stretching and opening of the mouth has been linked to the intensity of pain experienced by the infant [26]. In the present study, it is conceivable that a high  $F_1$  and low  $F_2$  exhibited by the infants was a result of facial expression in response to acute pain. Unfortunately, the cries collected for the present group of SIDS and HT infants were based on audio-only recordings, so it was not possible to evaluate facial expression. The relationship between facial expression and acoustic features of infant cry would seem to be an overlooked area of research, particularly in regard to the search for risk markers.

The pattern of crying observed in the present group of SIDS infants appeared to differ from HT infants for features of cry duration and cry resonance, with no apparent differences in vocal fold vibratory behavior. The results tend to parallel those previously reported by Corwin et al. [10] and thus provide more clarity in regard to the acoustic profile of crying in SIDS. While it

remains unlikely that examination of crying will serve as a stand-alone predictor of SIDS, we are encouraged by the likelihood that some specific features of crying may serve as unique prognostic indicators of unexplained infant death.

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