Published in final edited form as:

Res Dev Disabil. 2013 September ; 34(9): 2717–2724. doi:10.1016/j.ridd.2013.05.036.

Componential Deconstruction of Infant Distress Vocalizations via Tree-Based Models: A Study of Cry in Autism Spectrum Disorder and Typical Development

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Abstract

Understanding early episodes of cry is essential to improve caregiver-child interaction and child well-being. Caregiver perceptions of cry are based on interpretations of different acoustic characteristics of the cry, including the length of the pauses, the number of utterances, and the fundamental frequency. In this study, we used tree-based models to establish a hierarchy of effect in terms of how these acoustic characteristics influence perceptions of cries of children with autism compared to cries of typically developing children. In two studies, one in Italy and the other in Japan, we found that cries of children with autism are perceived more negatively. We also found that the length of the pauses, more than the number of utterances or fundamental frequency, determines listeners' negative perceptions. Implications for early caregiver-child interactions are discussed.

Keywords

cry; autism; autistic disorder; perception of cry in Japan; tree-based models; perception

Introduction

Autism or Autistic Disorder (AD) is a developmental atypicality characterized by impaired communication, disrupted social skills, and restricted and repetitive behavior (Kanner, 1943). AD in the definition of the DSM-IV-TR (APA, 2004) is one of three recognized Autism Spectrum Disorders (ASDs); the other two are Asperger syndrome and Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), which is diagnosed when the full set of criteria for autism or Asperger syndrome are not met. In the U.S.A., the prevalence of ASD is about 11.3 per 1,000 (one in 88), and about 45% of ASD cases are AD (CDCP, 2012). Incidence of AD is rising worldwide in both the developed (CDCP, 2012)

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and developing world (Bakare et al., 2012). Increasing incidence of AD has also been described in Japan (Honda et al., 2005).

The newly proposed definition of Autism in the DSM-V (APA, 2013) sees revisions to specific criteria needed for a diagnosis of Autism Spectrum Disorder. For the new criteria, the Communication and Social Interaction domains are combined into “Social/Communication Deficits.” This change highlights the significance of the social communication domain in autism, and so it is increasingly important to learn about Social/Communication Deficits in AD in early infancy. Here we studied crying in early autism because cries represent one of the first social behaviors with clear communicative intent and valence (Barr, Konner, Bakeman, & Adamson, 1991; Bieberich & Morgan, 2004; De Pisapia, Bornstein, Rigo, Esposito, De Falco, & Venuti, 2013). Deviations between cries of children with autism and typically developing children are expected considering the connection between crying and functions of the brainstem and limbic system, both of which appear to be compromised in children with autism (Amaral, Schumann, & Nordahl, 2008; Schulkin, 2007). Specifically, the limbic system, which is the central neurological core of socio-emotional life, is linked to the lower part of the brainstem, which controls the muscles of the larynx. The larynx is the major source of sound during crying, and it generates sound through the vibration of the vocal folds. In some children with AD, anomalies of functioning of the brainstem and/or limbic system contribute to cries with atypical features.

Cries vary in several acoustic parameters, notably pause length (also called duration of inspiration), number of utterances, and fundamental frequency (Lester & LaGasse, 2008). Studies that have focused on duration of inspiration, the time between cry utterances (interutterance interval), show that during pauses babies usually hold their breath. This variable is useful for assessing neural control of the respiratory tract (Lester & LaGasse, 2008). The number of utterances represents the frequency of cry sounds per unit time across an episode of crying (LaGasse et al., 2005). Utterances are important for assessing neural control of the respiratory tract, short utterances are an index of unstable respiratory control (LaGasse et al. 2005). Historically, fundamental frequency (f_0), which represents the base frequency during vocal fold vibration heard as the pitch of the cry, has been considered the most telling acoustic characteristic of a cry (Lester & LaGasse, 2008). High or low f_0 s are typically associated with neurological deficits (see LaGasse et al., 2005, for a review).

In previous studies, we found that the cries of children later diagnosed with autism had shorter pauses and higher f_0 s than the cries of developmentally delayed or typically developing children. Sheinkopf and colleagues (2000), Esposito and Venuti (2010a, b), and Venuti and colleagues (Venuti, Caria, Esposito, de Pisapia, Bornstein, & de Falco, 2012), employing behavioral and fMRI experiments, reported that adults in Western and Eastern settings (Italy and Japan) perceive the cries of children with autism as more distressed and distressing than the cries of typically developing children (Esposito et al., 2012). These findings agree with other reports on how communicative and non-communicative sounds with fewer pauses and higher pitch are perceived as more aversive and distressed (Haas & Edworthy, 1996). It has been supposed that higher f_0 and (perhaps) shorter pauses and fewer utterances are responsible for perceptions of distress (Esposito, Venuti, & Bornstein, 2011; Zeskand, Klein, & Marshall, 1992). However, what is not yet clear is which of these acoustic variables influences negative perceptions of cry of children with autism.

The main purpose of this analysis is to follow up our previous investigation of how AD and TD episodes of crying are perceived in different cultures through deconstruction of the acoustic parameters of the cry. Specifically, we discovered that pause length, number of utterances, and fundamental frequency all play roles in the perception of distress in cry. In the present study, we attempt to establish a hierarchy among these acoustic characteristics in

terms of how each influences perceptions of distress of crying of children with AD compared to those of typically developing (TD) children. To investigate this issue, cries of children with AD and TD were presented to two similar groups of Italian and Japanese adults who judged the level of distress cries expressed (*how distressed the infant sounded*) and the level of distress felt while listening to the same cries (*how distressed the participant felt after listening to the cry*). Distress expressed and distress felt to AD and TD cries were compared. Subsequently, judgements of the cry stimuli were submitted to recursive partitioning (also known as tree-based models; Costello et al., 2003) to analyse the respective roles the three primary acoustic features (length of pauses, number of utterances, and fundamental frequency) play in these perceptions of distress. Given a set of independent variables (here acoustic features) and a dependent variable (here levels of distress related to cry stimuli), tree-based models provide unique information about the (1) hierarchy of contribution of independent variables in explaining the distribution of the dependent variable and (2) which value of the independent variable subdivides the dependent variable into groups that differ statistically. To evaluate whether the results were broadly general or culturally specific, we studied adults in two contrasting cultures, Italy and Japan.

Our study was intended to shed light on how and why some characteristics of AD cries appear to be highly distressing to adult listeners and potentially render parental responses inadequate (Trevvarthen et al., 1998) and advance our understanding of the effect of different cry characteristics and their potential impact on the parent-child relationship.

General Method

Materials

Cry stimuli were extracted from 20 home videos of unedited cry bouts of 20 Caucasian firstborn 13-month-olds who belonged to two groups: Autistic Disorder (AD; $n = 10$, 5 boys/5 girls) and Typically Developing (TD; $n = 10$, 5 girls/5 boys). The home videos belonged to a database collected during a longitudinal study of early markers of autism. Children with AD received a clinical diagnosis between 36 and 40 months of age from a child psychiatrist according to DSM-IV-TR criteria, confirmed by both Autism Diagnostic Interview-Revised (ADI-R; ADI-R is a semistructured, investigator-based interview for caregivers of individuals who may AD; ADI-R psychometrics characteristics were excellent: both specificity and sensitivity are higher than 90%; Lord, Rutter, & Couteur, 1994) and ADOS-G (Autism Diagnostic Observation Schedule-Generic; ADOS-G is a semistructured assessment of social interaction, communication, play, and imaginative use of materials for individuals who may have AD; ADOS psychometrics in differentiating AD vs non AD are excellent: specificity = 97% and sensitivity = 96.7%; Lord, Risi, Lambrecht, Cook, Leventhal, DiLavore, et al., 2000). To eliminate cases of secondary autism, children with AD were free from other medical conditions (e.g., seizures, Fragile X syndrome) and had no visual or hearing impairments. Their cognitive level was evaluated as average at the age of 4 years using the Griffiths Mental Development Scales (reliability of the administration is ~ 90%; Griffiths, 1996; Hanson, 1982; Luiz, Barnard, Knoesen, & Kotras, 2004) and was in the typical range. Children with TD were part of a longitudinal research project and did not present any cognitive concerns as confirmed by their Wechsler Preschool and Primary Scale of Intelligence-II (WPPSI-II; the reliability coefficients for the WPPSI-II U.S. composite scales range from .89 to .95; Wechsler, 1989) scores at age 4 years. A research assistant, who was unaware of the purposes of the study and blind to children's group membership, gleaned video records of the two groups of children for appropriate stimuli. An exclusion criterion was that the audio recordings of cries contained background noises that would interfere with acoustic analysis (e.g., adult talk, sounds from toys, or other environmental noises).

After independently screening the home videos, to optimize and equate for sound quality and volume, the assistant selected the first episode of cry that occurred after a feeding. From this video the first 15 sec were used. To ensure that the stimulus cries were representative of the typical range of cry sounds for AD and TD groups, cry sounds were digitized and analysed using Praat acoustic analysis software (Boersma & Weenink, 2005). Episodes of crying in the two groups differed in the duration of pauses (a silence longer than 250ms within the episode of crying) in sec (AD: $M = 1.81$, $SD = 2.05$; and TD: $M = 4.75$, $SD = 1.29$), $F(1,19) = 3.43$, $p = .01$, and in the number of utterances (expressed vocalization of distress between two pauses; AD: $M = 4.98$, $SD = 2.39$, and TD: $M = 7.51$, $SD = 2.33$), $F(1,19) = 2.11$, $p = .05$, within episodes of crying. Next, long-term average spectrum (LTAS) was employed to provide spectral information for each cry episode. LTAS is helpful in discriminating cry characteristics of different categories of children (Lin & Green, 2007). For all cries, the First Spectral Peak (FSP) of the LTAS was obtained. FSP is the frequency value (in Hz) of the first amplitude peak across the LTAS. It is an estimate of the average f_0 of an episode of crying (Lin & Green, 2007). FSPs (in Hz) of cries were higher for AD ($M = 535.12$, $SD = 28.88$) than for TD children ($M = 465.71$, $SD = 32.56$); $F(1,19) = 3.16$, $p = .03$. These results are consonant with previous findings that have indicated that cries of children with autism usually have different waveform modulations in terms of shorter pauses and fewer utterances as well as higher fundamental frequency (Esposito et al. 2004; Esposito & Venuti, 2010a,b; Esposito et al. 2012).

Stimulus presentations

After stimulus preparation, 20 audio files were presented randomly to participants (recorded at 44,100 Hz with a stereo resolution of 32 bit) using a personal computer and a headset. Participants were asked to rate the level of distress expressed in the cries (by which we mean perceived distress of the child) and to rate the level of distress they felt (by which we mean distress felt by the adult observer) while listening to the cries. Participants used a 7-point Likert-type scale for both ratings where 1 = *the lowest level of distress* and 7 = *the highest level of distress*.

Analytic plan

To compare AD and TD cries, a general linear model (GLM) with repeated measures and two levels (AD and TD) was employed. To determine which feature(s) of a cry—pause length, number of utterances, or fundamental frequency--were operative, we employed recursive partitioning, specifically regression tree models (Costello et al., 2003). Recursive partitioning explores the structure of a dataset while developing easy-to-visualize decision rules for predicting a continuous (regression tree) outcome. The regression tree or tree-based model consists of two main steps: growing (exploring relations among variables) and pruning (minimizing overfitting the data). Tree-based models provide information about the (1) hierarchy of the importance of independent variables in explaining the distribution of the datapoints of the dependent variable and (2) which value (defined as the “node”) of the independent variable subdivides (defined as the “split”) the dependent variable in two groups that differ statistically. Using the rpart package for the statistical software R (ver. 3.1-52; Therneau & Atkinson, 2012), the set of parameters used for the splits was: (1) a node must have at least 30 data-point pairs to be considered for a split, (2) at least 20 data-point pairs were required for each terminal node, and (3) surrogate splits were permitted to allow for missingness. The complexity parameter was set equal to zero to allow each tree to grow to its full size, and the tree was then pruned (resulting in the optimal tree) to remove branches containing nodes with a t value greater than 1.645 ($\alpha = .05$).

Preliminary analyses

Prior to data analysis, correlations among the independent variables of pause length, number of utterances, and fundamental frequency were assessed. Pause length and number of utterances were as expected correlated, $r(18) = .53$, $p < .05$; however, these two variables shared only 28% of their common variance, and so we treated them independently. Fundamental frequency was uncorrelated with pause length, $r(18) = .03$, ns , and number of utterances, $r(18) = .02$, ns . Then, univariate and multivariate distributions of dependent variable scores (Expressed Distress and Felt Distress) for the AD and TD cries were examined for normality, homogeneity of variance, outliers, and influential cases (Fox, 1997) for both the Italian and Japanese samples. Distress scores were normally distributed. The distance of each case to the centroid was evaluated to screen for multidimensional outliers (Fox, 1997). No significant differences emerged in either study between sample subgroups (parents and non-parents, females and males), and so single group analyses were conducted. As expected, statistically significant correlations between expressed distress and felt distress scores were found in both the Italian, $r(78) = .61$, $p < .001$, and Japanese, $r(78) = .69$, $p < .001$, samples. Because we were theoretically interested in the different dependent variables, and the two dependent variables shared only 37% and 48% of their common variance, respectively, we treated them separately.

Study 1: Italian Participants

Participants

A total of 80 adults recruited in urban Trento (North of Italy), 40 parents of typically developing children (20 fathers M age: = 33.3, SD = 4.3; 20 mothers M age: = 32.1, SD = 3.8) and 40 non-parents (20 males M age: = 29.2, SD = 4.8; 20 females M age: = 27.9, SD = 3.5), participated. Participants were recruited through a database of volunteers available at the University of Trento. The study was conducted in accordance with ethical principles and approved by the Ethical Board of the Department of Psychology and Cognitive Science and Education of the University of Trento. Informed consent was obtained from all the participants.

Results

Expressed distress—Participants rated the expressed distress level for AD cries (M = 4.72, SD = .29) greater than for TD cries (M = 2.99, SD = .15), $F(1,79) = 143.6$, $p < .001$, $\eta_p^2 = .09$; see Figure 1A. Figure 1B shows the optimal tree. For both AD and TD cries, the variable that explained the distribution of expressed distress best was length of the pauses. Specifically, cries with pauses shorter than 1.5 sec generated higher levels of expressed distress than those with longer pauses. Next to the length of the pauses, f0s higher than 608Hz resulted in higher levels of expressed distress. The effect of number of utterances was not statistically significant in this model.

Felt distress—Participants rated felt distress for AD cries (M = 4.44, SD = .23) greater than for TD cries (M = 2.96, SD = .19), $F(1,79) = 154.5$, $p < .001$, $\eta_p^2 = .08$. Figure 1C shows the optimal tree. The variable that explained the distribution of felt distress best was again length of the pauses. Specifically, cries with pauses shorter than 1.5 sec generated higher levels of felt distress than those with longer pauses. Next to the length of the pauses, f0s higher than 618Hz resulted in higher levels of felt distress. The effect of number of utterances was not statistically significant in this model.

Study 2: Japanese Participants

Participants

A total of 80 adults recruited in urban Chiba, 40 parents of typically developing children (20 fathers $M_{age} = 30.8$, $SD = 4.9$; 20 mothers $M_{age} = 31.2$, $SD = 3.8$) and 40 non-parents (20 males $M_{age} = 29.2$, $SD = 4.9$; 20 females $M_{age} = 28.1$, $SD = 3.1$) participated. Participants were recruited through a database of volunteers available at Chiba University. The study was conducted in accordance with ethical principles and approved by the Ethical Board of the Faculty of Education at Chiba University. Informed consent was obtained from all the participants.

Results

Expressed distress—Participants rated the expressed distress level for AD cries ($M = 4.80$, $SD = .16$) greater than for TD cries ($M = 3.64$, $SD = .14$), $F(1,79) = 178.2$, $p < .001$, $\eta_p^2 = .11$; see Figure 2A. Figure 2B shows the optimal tree. For both AD and TD cries, the variable that explained the distribution of expressed distress best was again the length of the pauses. Specifically, cries with pauses shorter than 2 sec generated higher levels of expressed distress than those with longer pauses. Next to the length of the pauses, the other parameters explained the distribution of expressed distress less but equally: Episodes of cries with more than 7 utterances resulted in lower levels of expressed distress, and those with f_0 s higher than 515Hz resulted in higher levels of expressed distress.

Felt distress—Participants rated felt distress for AD cries ($M = 3.57$, $SD = .23$) greater than for TD cries ($M = 2.68$, $SD = .14$), $F(1,79) = 145.1$, $p < .001$, $\eta_p^2 = .09$. Figure 2C shows the optimal tree. The variable that explained the distribution of felt distress best was again the length of the pauses. Specifically, cries with pauses shorter than 1.5 sec generated higher levels of felt distress than those with longer pauses. Next to the length of the pauses, episodes of cries with more than 7 utterances resulted in lower levels of felt distress, and those with f_0 s higher than 518Hz resulted in higher levels of felt distress.

General Discussion

Cries of children with autism are perceived as more distressed and elicit more distress in listeners. It has been suggested that higher fundamental frequency as well as shorter pauses and fewer utterances are responsible for these perceptions of distress. In the present study, we analysed which of these acoustic parameters negatively influences perceptions of crying of children with autism and in typically developing children. Specifically, the present study deconstructs ratings of distress in cries of children with AD or TD to discern the relative importance of various characteristics of the cry to expressed and felt distress. Although many previous studies have pointed to fundamental frequency as the principal negative influence in perceptions of crying, we found that pause length has the strongest impact on perceptions of distress compared to fundamental frequency and the number of utterances. It is likely that previous research has missed the importance of pauses because vocalizations have mainly been manipulated in terms of f_0 rather than the absence of vocalizations (i.e., pauses). Our findings also confirm this result in two contrasting cultural groups. It would appear necessary to attend to pause length as well as f_0 and number of utterances in future studies that analyse cry in different populations or utilize cries as stimuli in eliciting (and measuring) adults' behavioral or brain responses.

Episodes of crying of children with autism elicited higher levels of both *expressed* and *felt* distress. This result confirms previous studies conducted both in Western (Esposito & Venuti, 2008) and Eastern (Esposito et al., 2012) samples. This finding seems informative

from both clinical and parental care perspectives. Clinically, this study advances our understanding of early social interaction in autism (that is not fully studied because of the difficulties associated with making a reliable diagnosis of AD before the age of 18 to 24 months). From a parental care perspective, it is possible that biases in evaluating levels of distress expressed in cries of children with special needs interfere with the adequacy of caregiver responses. Indeed, a caregiver's adequate behavioral response, which ideally met the infant's needs, is a necessary first step in establishing wholesome future relationships, but can be compromised when children express atypical emotions (for example, when children express their distress through crying with shorter pauses, fewer utterances, and/or higher fundamental frequency). Overestimating the level of distress expressed in an episode of crying may jeopardize caregiver responsiveness, whereas "successful recognition and evaluation of children's vocalizations can be critical for bonding mechanisms and for offspring well-being and survival" (Seifritz et al., 2003, p. 1367).

The fact that episodes of cries of children with autism have shorter pauses and fewer utterances has some specific clinical implications. Trevarthen and colleagues (1998) speculated that deficits in the early development of turn-taking may be key to the emotional and cognitive development of children with autism. Crying episodes with shorter and fewer pauses reduce the possibility for adult listeners to interact with the child and at the same time diminish the possibility for children learning the basis of turn-taking. Additional study might yield greater understanding of mechanisms underlying this early social deficit.

The novelty of our findings, in particular the fact that f_0 was not determined to be the most important acoustic parameter in the perception of expressed or felt distress, calls for careful consideration. To determine the generality of our initial findings (Study 1, Italian sample), we undertook the same experiment in a different cultural context (Study 2, Japanese sample). Again, f_0 was not the most important parameter to perceptions of expressed or felt distress lending our findings external validity. Between the two studies, we found only one difference in the pattern of the results: the number of utterances was not statistically significant in the Italian model, but was in the Japanese model. Although we do not have strong data to explain this difference, we believe that for some cultural and/or linguist differences, utterances are more important for the perception of cry in Japan than in Italy. Specifically, in Japanese the onomatopoeic sound for a baby cry is "aan-aan (アーンアーン)," and it stresses repetition of more than one utterance (Takei & Tamori, 1993; Waida, 1984). By contrast, in Italy the onomatopoeic sound of a baby cry is "nguee", which stresses the sound more than repetition of more than a single utterance. Thus, it may be that Japanese participants felt as more typical cries that had more repetitions of the same or similar instances (more utterances) because cries with more utterances match their psychological prototype of a baby's cry. It will be interesting to develop this cultural aspect in future studies.

A contribution of the current study to the literature of the field of early social deficit in autism and their implications for parenting lies in our applying tree-based models to the analysis of cries. We found, contrary to expectations, that the length of pauses in cries is the most important variable in explaining both expressed and felt distress in listeners. The majority of previous studies have pointed to fundamental frequency as perhaps the most important variable in perceptions of cry atypicality (see, for example, LaGasse et al. 2005). Moreover, previous studies have not focused on cries of children with autism compared with TD cries.

We found no differences among parents and non-parents in judging expressed and felt distress to episodes of child cry. This result accords with previous studies of perceptions of cry conducted in Japan. Adachi, Murai, Okada, and Nihei (1985) reported differences in the

perception of distress expressed among parents versus non-parents. However, cries of high-risk infants (which are similar to the cries of children with autism) were judged similarly by parents and non-parents alike. Moreover recent evidence using fMRI reported no difference between parents and non-parents in brain processing of the cries of children with autism and typically developing matched controls (Venuti et al., 2012). Of course, additional testing is needed to verify whether or not differences between parents and non-parents agree in their perceptions of distress of crying episodes.

Some limitations of these experiments point to future directions of study. In these studies we only analyzed audible features of episodes of crying. It may be that non-audible features (i.e., ultrasound emissions) increase the level of distress perceived or felt by directly influencing the autonomic nervous system. For this reason, it would be informative to replicate the present study using cries recorded with a large range of sounds (from the lower limit of the human hearing range to the low level of ultrasounds, ~30kHz). Another limitation of the study is the fact that we assessed parents of typically developing children. A later study might assess parents of children with ASD. This would be important to consider because parents of children with ASD may be more used to cries of children with ASD and not consider it as distressing as those of typically developing children. Our results also underscore the desirability of future research on brain responses of adults to infant crying. Future studies might use neuroimaging techniques to identify brain neural networks that render the perception of atypical cries (i.e., with shorter pauses as well as fewer utterances and higher fundamental frequencies) as more distressed. These studies may also aim to provide fresh insights into early caregiver–child interaction in typical and atypical circumstances.

Acknowledgments

All participants in this study are gratefully acknowledged. This research was supported by the ST fellowship from Japan Society for the Promotion of Science (JSPS PE09064), Grant-in-aid for Scientific Research from Japan Society for the Promotion of Science (Projects #22530694 and # 24730563), the FPR Program (RIKEN Brain Science Institute), the Intramural Research Program of the DiSCoF, University of Trento (Italy), and the Intramural Research Program of the NIH, NICHD (USA).

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Highlights

- Understanding early episodes of cry is essential to improve caregiver-child interaction
- Cries of children with autism are perceived more negatively than those of typically developing children
- Among the acoustic characteristics of a cry, the length of the pauses, more than the number of utterances or the fundamental frequency, determines listeners' negative perceptions

Figure 1A.

Italian sample. Mean levels ($\pm$ SEM) of judged expressed distress and of felt distress hearing cries of children with AD or TD. *** $p < .001$. Figure 1B. The optimal tree that predicts expressed distress. The regression tree or tree-based model provides information about (1) the hierarchy of the importance of independent variables in explaining the distribution of the dependent variable and (2) which value of the independent variable divides the dependent variable in two groups that differ statistically. The bottom rectangle shows the distribution of expressed distress from lower (left) to higher (right). The values in the oval leaves of the tree refer to the condition of the independent variable that statistically divides the distribution of the dependent variable. Below each oval leaf, the indications “yes” or “no” refer to whether or not the condition is met. Each leaf is divided in two sub-leaves. The terminal leaves (quadrangles) represent subgroups that cannot be further subdivided. The n value in the terminal leaves represents the size of the group, and M is the mean value of the group for the dependent variable. Figure 1C. The optimal tree for felt distress.

Figure 2A. Japanese sample. Mean levels ($\pm$ SEM) of judged expressed distress and of felt distress while hearing cries of children with AD or TD. *** $p < .001$. Figure 2B. The optimal tree that predicts expressed distress. Figure 2C. The optimal tree for felt distress.