Published in final edited form as:

Res Autism Spectr Disord. 2011 ; 5(4): 1510–1516. doi:10.1016/j.rasd.2011.02.013.

ASSESSMENT OF DISTRESS IN YOUNG CHILDREN: A COMPARISON OF AUTISTIC DISORDER, DEVELOPMENTAL DELAY, AND TYPICAL DEVELOPMENT

G. Esposito^{1,2}, P. Venuti¹, and M.H. Bornstein³

¹ Observation and Functional Diagnosis Lab- DiSCoF, University of Trento (Italy)

² Kuroda Research Unit for Affiliative Social Behavior, RIKEN Brain Science Institute (Japan)

³ Eunice Kennedy Shriver National Institute of Child Health and Human Development, NIH, (USA)

Abstract

Distress emotions in very young children are manifest in vocal, facial, and bodily cues. Moreover, children with different developmental conditions (i.e. Autistic Disorder- AD, Developmental Delay- DD, Typically Developing- TD) appear to manifest their distress emotions via different channels. To decompose channel of emotional distress display by group, we conducted a study in which video clips of crying of 18 children 18 months of age belonging to three groups (AD, DD, TD) were modified to isolate vocal, facial, or bodily cues, and 42 female adults were asked to judge the distress and typicality (expected normality) of the different stimuli. We find variation in adult judgements of distress and typicality by child group (AD, DD, TD) and by isolated cues (vocal, facial, or body). Although there is some overlap between responses to episodes of crying of children with AD and those with DD, the different cues of crying of children with AD tend to be considered more atypical and distressed than those of the other two groups (DD and TD). Early assessment of different cues of the expression of distress, and more generally of emotional expressivity in a child, may provide useful information for pediatricians and practitioners who are in contact with young children and must make clinical screening decisions. The findings also alert parents of children with AD to important aspects of their cries.

Keywords

Autism; Expression of Distress; Cry

Expressing distress represents one of first communication of human children to convey their needs and affect their environment (Irwin, 2003; Gingras, Mitchell & Grattan, 2005). In turn, caregiver responsiveness to children's distress plays an important role in the development of the child's personality, temperament, cognition, and language. Thus, expression of distress constitutes a biosocial phenomenon that reflects the status of the nervous system and indirectly mediates child development through parental intervention (Lester, 1984; Lester, Boukydis & Garcia Coll, 1995, LaGasse, Neal & Lester, 2005).

The vast majority of the studies of expressions of distress in very young children have focused on cry and its morphology. In general, an episode of crying elicits physiological

reactions in adults, such as increases in heart rate (Huffman, Bryan, del Carmen, Pedersen, Doussard-Roosevelt & Porges, 1998) and endocrine responses (Fleming, Corter, Stallings & Steiner, 2005). Such physiological reactions activate those listening to cries to take measures to eliminate their cause (Bowlby, 1969; Gustafson, Wood & Green, 2000; LaGasse et al., 2005). Studies of morphological characteristics of children's cry, such as its frequency and duration, have shown how these characteristics moderate adult responses (Gustafson & Green, 1989; Ziefman, 2003). For example, high-frequency cries are perceived as more aversive and distressed than low-frequency cries. A particular type of cry, characterized by low frequency, and typical of children with developmental delay (specifically Down syndrome), is perceived as less urgent than high-frequency cries (encountered in oxygen-deprived infants suffering brain damage; Frodi & Senchak, 1990).

Distress in very young children is expressed through different channels (i.e., vocal, facial, bodily cues). Information about children's state of distress is available not only through voice, but through facial and bodily cues as well. Irwin (2003) found that perceiver ratings of comfort and agitation differed for high- and low-distress cries on the basis of facial and vocal cues, but not bodily cues. In general, to respond appropriately to the level of distress a child expresses in everyday situations caregivers normally attend to the sound (in particular the intensity and pitch), evaluate the facial expression and the body movements of the child, and take the general circumstance into account (such as the hour of the day, time since the previous meal, and so forth). In the current study, we aim to investigate adult judgments of distress conveyed by different cues (vocal, facial, or bodily) in children with different developmental conditions (i.e., Autistic Disorder- AD, Developmental Delay- DD, Typically Developing- TD).

Expressions of distress vary with children's psychopathological status. For example, the fundamental frequency (f0) of cry is especially sensitive to neurological insult with both higher levels overall and greater variability. Autistic Disorder affects emotional expression, communication, and social skills to varying degrees. Furthermore, considering the connection between crying and the functioning of the brainstem and limbic system (both areas are compromised in AD children; Amaral, Schumann, & Nordahl, 2008), it seems reasonable to expect anomalies in the expression of distress of AD children (Bieberich & Morgan, 1998). The analysis of acoustic features of expression of distress has revealed a number of differences among children with AD and matched TD controls. In particular, episodes of cry in children with AD are shorter in duration, show less waveform modulation, and consist of greater dysphonation (Esposito & Venuti, 2008). Children who suffer a developmental delay (DD) often vary in the development of their communication skills and emotional expression, tending to rely on unconventional and idiosyncratic nonverbal behaviors (e.g., Meadan, Halle, Ostrosky, & DeStefano, 2008).

Because emotional expression, especially for distress, among children with AD or DD and TD children may show different profiles, it may be important to analyze their early manifestation. Indeed, differences in the expression of distress may influence parenting. A number of studies have investigated how parents perceive crying in children with AD and DD compared to TD children (Esposito & Venuti, 2008, 2009, 2010a, b). Esposito and Venuti (2008) interviewed parents of TD children and those of children with AD. Qualitative analysis revealed that parents of AD children more often mentioned negative patterns of feelings with respect to crying episodes (e.g., AD cries were considered unexpected for parents who could not identify causal factors). The authors also designed a Listen-and-Response experiment to test whether the atypical structure of crying episodes characteristic of children with AD biases adult perceptions. Twelve episodes of crying of children of two ages (13 and 20 months), garnered from home videos of children with AD and from matching control-group TD children, were randomly presented to participants who

were asked to estimate the age of the child who was crying, give reasons why the child was crying, and describe how they felt while hearing the cries. Adults reported feeling more negative mental states (i.e., anxiety) when listening to AD cries. In contrast, tender mental states were reported when listening to crying episodes of typically developing and intellectually challenged children.

Studies of cry in children with developmental conditions have mainly focused on acoustic components. Some studies have investigated the role of non acoustical components (e.g., facial expression, motor behaviors, etc.) in typically developing children (e.g., Irwin, 2003) but there are no studies in the current literature that have specifically investigated non acoustical components of distress in children with AD or DD. This circumstance is unfortunate because in atypical development, especially in AD, children tend to display less facial expressivity and specific motor dysfunctions (i.e., stereotypies). We hypothesize that these anomalies may influence the way adults perceive distress in these children.

In sum, distress emotions in very young children can manifest in vocal, facial, and bodily cues (Irwin, 2003). Moreover, children with different developmental conditions (AD, DD, TD) may manifest distress emotions via different channels (Bieberich & Morgan, 1998). Although previous work has focused on different cues that influence the perception of distress emotions in young children (e.g., Irwin, 2003) and different developmental conditions (e.g., Bieberich & Morgan, 1998; Iverson & Wozniak, 2007), no previous study has analyzed the specific effects of different cues of distress in different developmental conditions. To decompose channel of emotional distress display by group, we conducted a study in which we asked adults to judge the distress and typicality of isolated voices, faces, and body movements of crying AD, DD, and TD toddlers. In this way, this study promises to test the hypothesis that typically and atypically developing children express distress through different cues and to quantify which cues were most prominently associated with which atypical condition. Guided by the existing literature, we expected that: (i) vocal features of atypical crying will be perceived as more distressed and atypical; (ii) because of their poor facial expressivity, the faces of children with AD during episodes of crying will elicit lower levels of distress and more atypicality compared to children with DD or TD; and (iii) because of the presence of motor stereotypies, motor cues in episodes of crying in children with AD will elicit higher levels of perceived distress and more atypicality compared to children with DD or TD.

Method

Participants

Forty-two female nullipara ($M = 30.67$ years; $SD = 8.91$) participated in the study. Because the study aimed to investigate adult judgments of distress in children with different developmental conditions, to exclude any bias or expertise in listening to cries of typical or atypical children only adult non mothers were selected. Participants were recruited from an urban area in the north east part of Italy through advertisements placed in local public and commercial establishments. From all the people who responded, non mothers in an age range from 25 to 40 were selected to participate. All participants were Italian citizens with Italian background and had medium to high SES ($M = 48.67$ $SD = 13.98$) calculated using the Hollingshead Four-Factor Index of Social Status (1975).

The research study was conducted in accordance with ethical principles, and it was approved by the Ethical Committee of the Department of Cognitive Sciences of the University of Trento. Furthermore, informed consent was obtained from all participants.

Stimuli

Three types of stimuli were extracted from 18 retrospective home videos of unedited cry bouts of 18 firstborn 18-month-olds who belonged to one of three groups: Autistic Disorders (AD; $n = 6$, 3 boys/3 girls), Developmental Delay (DD; $n = 6$, 3 boys/3 girls), and Typically Developing (TD; $n = 6$, 3 girls/3 boys). Children with AD received a clinical diagnosis of autistic disorders at the age of 3, from a child psychiatrist according to DSM-IV-R criteria, confirmed by ADI-R, ADOS-G. Infants with a diagnosis of Pervasive Developmental Disorder not otherwise specified (PDD-Nos) or Asperger Syndrome were not included in the study. To prevent the presence in the sample of cases of secondary autism, children with AD were free from other medical conditions (e.g., seizures, Fragile X syndrome, etc.) and had no visual or hearing impairments. The DD group provided both a chronological and developmental age matching for the AD group. All DD children had normal vision and hearing, unimpaired hand use, and full mobility. No DD children had a current or previous diagnosis of AD. The DD group was etiologically heterogeneous, but all DD children showing facial or body features suggesting a developmental delay (e.g., Down syndrome) were excluded. Both AD and DD children were diagnosed in later childhood; their cognitive level was assessed after the age of 36 months, and their mean global development quotients (GDQ) using the Griffiths Mental Development Scales were not different (AD $M = 62$; $SD = 3$; DD $M = 58$; $SD = 5$, $t(11) = 1.21$, ns). Children with TD were part of a longitudinal research project on typical development and did not present any significant medical or developmental concerns as confirmed by Child Behaviour Check-list (CBCL) normal scores and the Wechsler Preschool and Primary Scale of Intelligence- II (WPPSI-II) at the age of 4 ($M = 102$; $SD = 5$). A research assistant who was unaware of the purposes of the study and blind to children's group membership gleaned video/audio records of the three groups of children from home videos that had been made when they were 18 months of age. The home videos included segments from family play situations or special events, which did not vary significantly across groups.

The three stimulus types consisted of (1) audio versions of a cry (voice only), (2) silent videos of the isolated face (face only), and (3) silent videos of the isolated body (body only). The 54 video/audio sequences each lasted 15 sec. The face-only and body-only stimuli were constructed using Adobe Premiere Pro 1.5 by changing pixel resolutions to hide the body or face, respectively. To ensure the homogeneity of these two conditions we selected only scenes with only the child visible, with the full body visible, and with the child facing the camera. During the voice-only stimuli, the video screen was a uniform light blue. To ensure that the cries in this study were representative of the typical range of cry sounds for the three groups (TD, DD, and AD), the cry sounds were digitized and analyzed using the Praat acoustic analysis software (Boersma & Weenink, 2005). A long-term average spectrum (LTAS) was employed to provide spectral information for the crying episode. The LTAS has been helpful in discriminating the cry characteristics of different categories of children (Lin & Green, 2007). For all episodes of crying 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 the episodes of crying (Lin & Green, 2007). The FSP of our cry episodes were for AD: $M = 528.38$, $SD = 41.25$; for DD: $M = 465.03$, $SD = 35.14$; and for TD: $M = 459.2$, $SD = 21.4$. Significant differences emerged among the groups, $F(2,18) = 2.62$, $p < .05$, $\eta^2_p = .03$. This result is consonant with previous findings (e.g., Esposito & Venuti, 2010a, b) that have highlighted how episodes of cries of children with AD at 18 months of age usually have higher f_0 .

Procedures

Video presentation used the full 19" screen of a DELL 1908 FP, and sound was played through a DELL 501 speaker system. Participants, blind to the nature and source (AD, DD,

TD) of the stimuli, were instructed to attend to each stimulus and, afterward, to rate each on two 7-point Likert-type scales. For vocal, facial, and bodily cues, one scale asked for ratings of *distress* level of the child (1 = *lowest level of distress* and 7 = *highest level of distress*), and the other scale asked for ratings of *typicality* (meaning expected normality) of the child (1 = *less typical* and 7 = *more typical*). Each trial began with an alerting signal, consisting of a short tone, followed by presentation of the video/audio stimulus separated by a 3-sec ISI. Presentation of the 54 stimuli was blocked by stimulus type, and the order of trials within each stimulus type was random.

Results

Preliminary Analyses

Prior to data analysis, univariate and multivariate distributions of distress and typicality ratings were examined for normalcy, homogeneity of variance, outliers, and influential cases (Fox, 1997). Scores were normally distributed. The distance of each case to the centroid was evaluated to screen for multidimensional outliers (Fox, 1997). Ratings of stimulus types (voice only, face only, body only) were uncorrelated.

Distress

A general linear model (GLM) with repeated measures for 3 stimulus types (Voice Only, Face Only, Body Only) and 3 stimulus groups (AD, DD, TD) was employed. Distress ratings of the stimuli appear in Figure 1. A significant interaction of Stimulus type by Stimulus group emerged, $F(1,2260) = 29.56, p < .01, \eta^2_p = .12$. Significant main effects both for stimulus type, $F(2,2258) = 38.11, p < .01, \eta^2_p = .19$, and stimulus group, $F(2,2258) = 26.96, p < .01, \eta^2_p = .14$, also emerged. Turkey HSD post-hoc tests indicated that participants rated Voice Only TD episodes of cry as expressing less distress than AD and DD cries (respectively: M difference = $-.56$, $SE = .11, p \leq .05$, and M difference = $-.51$, $SE = .11, p \leq .05$). No difference emerged between AD and DD Voice Only cries. For the Face Only stimuli, participants rated AD as less distressed than TD and DD (respectively: M difference = $-.62$, $SE = .12, p \leq .05$, and M difference = $-.83$, $SE = .15, p \leq .05$). No difference emerged for Face Only stimuli between TD and DD groups. For Body Only stimuli, participants rated AD more distressed compared to TD and DD (respectively: M difference = $.79$, $SE = .15, p \leq .05$, and M difference = $-.88$, $SE = .15, p \leq .05$). No difference emerged between TD and DD toddlers in the Body Only stimuli.

Typicality (expected normality)

A general linear model (GLM) with repeated measures for 3 stimulus types (Voice Only, Face Only, Body Only) and 3 stimulus groups (AD, DD, TD) was again employed. Typicality (expected normality) ratings of the stimuli appear in Figure 2. A significant interaction of Stimulus type by Stimulus group emerged, $F(1,2260) = 9.01, p < .01, \eta^2_p = .04$. A significant main effect for stimulus group also emerged, $F(2,2258) = 78.63, p < .01, \eta^2_p = .19$. Tukey HSD post-hoc tests indicated that for Voice Only stimuli participants rated AD episodes of cries as less typical than TD and DD cries (respectively: M difference = -1.06 , $SE = .12, p \leq .05$, and M difference = -1.85 , $SE = .21, p \leq .05$), and a significant difference also emerged between TD (more typical) and DD (less typical) cries (M difference = $.85$, $SE = .14, p \leq .05$). For Face Only stimuli participants rated AD less typical compared to TD and DD (respectively: M difference = -1.44 , $SE = .25, p \leq .05$, and M difference = -1.38 , $SE = .25, p \leq .05$); no difference emerged between TD and DD. For Body Only stimuli participants rated AD less typical compared to TD and DD (respectively: M difference = 1.19 , $SE = .12, p \leq .05$, and M difference = -1.01 , $SE = .11, p \leq .05$); no difference emerged between TD and DD stimuli.

Discussion

To decompose channel of emotional distress display by group, we conducted a study in which cry episodes of 18 toddlers 18 months of age belonging to one of three groups, children with autistic disorder, children with developmental delay, and typically developing children, were modified to isolate vocal, facial, or bodily cues, and 42 female adult participants were asked to judge the distress and typicality (expected normality) of the different stimuli. We found variation in adult judgements of distress and typicality by child group (AD, DD, TD) and by isolated cues (vocal, facial, or body). Guided by the existing literature, we expected that: (i) vocal features of atypical crying would be perceived as more distressed and atypical; (ii) because of their poor facial expressivity, the faces of children with AD during episodes of cry would elicit lower levels of distress and more atypicality compared to children with DD or TD; and (iii) because of their tendency to motor stereotypies, motor cues in episodes of crying in children with AD would elicit higher levels of distress and more atypicality compared to children with DD or TD.

- i. As expected, the vocal cries of children later diagnosed with autism were judged to be similar to those of children with developmental delay but more distressed than those of typically developing children. In respect to the level of typicality, vocal cries alone were judged to be more atypical in children with atypical development (both autism and developmental delay) than in typically developing children. Vocal cries alone of children with autism were perceived as even more atypical than vocal cries alone of children with a developmental delay.

A number of studies that examined vocal expression of distress of children with specific neurological damage has revealed an association between crying and developmental delay. In particular, acoustic qualities of a cry appear to be sensitive to neurological insult with both higher levels of overall and greater variability of the f0 (e.g., Wasz-Hocker et al., 1968; LaGasse et al., 2005). Our finding of differences in perceived distress of crying of children with developmental problems (either autism or developmental delay) can be interpreted in view of the results in previous studies of acoustical characteristics of these episodes of crying compared to those of typically developing children. Although there is some similarity between responses to episodes of crying of children with developmental delay and those with autism, the different cues of crying of children with autism tend to be considered even more atypical and distressed than those of developmentally delayed children.

- ii. Faces were seen as less distressed in children with autism than in typically developing or developmentally delayed children. The faces of children with developmental delay and typical development were perceived to be equivalently distressed. The faces of children with autism were also perceived to be less typical than those of developmentally delayed or typically developing children. The facial expression of children with developmental delay and typically developing children were rated as equivalently typical. One of the core symptoms of autism is the lack of emotional expressivity through their facial cues. For this reason, the facial expressions of crying AD children s were rated as less distressed than those of children of the other groups but at the same time they were rated as less typical.
- iii. The bodily movements of children with autism were perceived as more distressed than those of developmentally delayed or typically developing children, who were equivalent. The bodily actions of children with autism were also perceived to be less typical than those of developmentally delayed or typically developing children. The bodily movements of children with developmental delay and typically developing children were rated as equivalent. Qualitative analysis of the body

stimuli of children with autism used in our study suggests that these children appear rigid and suffer restricted tonicities relative to the other children. This interpretation agrees with previous reports of early motor anomalies in children with autism that have shown greater instability in the first years of life compared to typically developing children (e.g. Iverson & Wozniak, 2007).

The spectrum of Autistic Disorder children's expression of distress has a higher fundamental frequency contributing to the lower level of rated typicality, and the discrepancy between the face of a crying child and lack of emotion expressed in the face presumably contributes to lower typicality judgments for face only and body only cues in children with autism. Children with autism are perceived to be the least typical in terms of their vocal, facial, or bodily expression of distress. Cries of atypically developing children (such as pre-term infants) are usually of higher frequency, and for that reason are considered more urgent than moderate frequency cries (Rautava, Lempinen, Ojala, Parkkola, Rikainen, Lapinleimu et al., 2007). The cries of children with visceral colic have higher frequency and are perceived as more distressed (Huffman et al., 1998). The cries of children with autism have higher f_0 both at 5 and at 18 months of age (Esposito & Venuti, 2010). Because higher-frequency cries are often perceived as more aversive and distressed than lower-frequency cries (Huffman et al., 1998), the acoustic characteristics of episodes of crying in children with autism, namely their higher fundamental frequency, may account for adults' bias in interpreting and their evoking mental states of uneasiness. This bias may in turn jeopardize adequate parental responsiveness. Indeed, "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).

Implications

Anomalies in expressions of distress (namely cry) appear to be related to neurological compromise. What happens when parents cannot adequately understand their children's cries? An instructive example occurs when a parent interacts with a child who suffers from a neurological deficit, but before the deficit is diagnosed. This situation often happens in the case of children with AD, for whom diagnoses are usually provided only after 24 months of age. These findings suggest that previously undetected neurological damage might be exposed by analyzing cries, even when no other symptoms are present. On this argument, atypical cries may constitute an early indicator of developmental risk. Identifying the kind of cry and reactions to it could contribute to the early identification of Autism.

Other important practical implications follow from these results. For example, the finding that cries of children with AD were perceived as more distressed than TD children by vocal and bodily cues, but were perceived as the least distressed by facial cues, may have important implications for parenting that should be further investigated in successive studies. Our results have clinical implication as well. In particular, early assessment of the expression of distress, and more generally of emotional expressivity in a child, will inform pediatricians and practitioners who are in contact with young children and must make clinical diagnoses. This form of assessment should also prevent the relationship between caregivers and their children with AD or DD from falling prey to a vicious cycle of negative interactions. Indeed, as our results and other studies show, children with Autism and with Developmental Delay (Bieberich & Morgan, 1998; LaGasse et al., 2005; Rautava et al., 2007) experience problems in expressing their negative emotions through cry. These difficulties might be linked to brain anomalies and compromise various acoustic qualities of the cry, so that the cry may not be easily understood by caregivers. Problems in understanding their child's cries can create distress and make caregivers uneasy. In turn, distress leads caregivers to provide their children with inadequate feedback, which is ineffective in addressing the cause of crying. As a result, caregivers do not receive adequate

response from their children and so they may start to feel inadequate and unable to foster in their children a sense of well-being. Sensing something amiss, caregivers modify their parenting skills. For their part, children who cannot adequately communicate with their caregivers may engage in compensatory behaviors such as self-isolation, stereotyped behavior, or hyper- or hypokinesia. Thus, something related to a neurological disorder (in this example, crying) becomes a starting point for problems in interaction and sets the tone for an overall distressed relationship between the caregiver and child with autism or other developmental delay.

Limitations

One limitation of this study is the fact that we only recruited nullipara females ratings of child distress. To have a more general view of adult assessments of children's crying, follow-up studies should consider other populations, such as mothers, fathers, and non-fathers. A further limitation of this study is the use of retrospective home videos to extract cry stimuli. A shortcoming of retrospective studies of children with autism (or with other psychopathologies) is the impossibility of clearly describing the developmental level of the child at the time of the video. However, retrospective video analysis has proved an excellent option for accessing early periods in development, months or years before a child is formally diagnosed with autism (Baranek, 1999; Werner, Dawson, Osterling, & Dinno, 2000).

Acknowledgments

All the participants in this study are gratefully acknowledged as are all the families that made available the home videos from which the stimuli were extracted. The authors also gratefully acknowledge Nicola Chisté (University of Trento) for technical assistance. This research was partially supported by the Intramural Research Program of the NIH, NICHD (USA).

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Figure 1. Means (SEMs) for judged levels of *distress* of voices, faces, and bodies of typically developing (TD) children, children with Autistic Disorder (AD), and children with developmental disability (DD). Bars with different subscripts were significantly different in Tukey HSD post-hoc tests.

Figure 2. Means (SEMs) for judged levels of *typicality* (expected normality) of voices, faces, and bodies of typically developing (TD) children, children with Autistic Disorder (AD), and children with developmental disability (DD). Bars with different subscripts were significantly different in Tukey HSD post-hoc tests.