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Chapter 4

Concordance between physiological arousal and emotion expression during fear in young children with Autism Spectrum Disorders

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Revised manuscript requested to resubmit with minor revisions.

Abstract

This study aimed to measure emotional expression and physiological arousal in response to fear in 21 children with Autism Spectrum Disorders (ASD; age range 43-75 months) and 45 typically developing children (TD; age range 41-81 months). Expressions of facial and bodily fear and heart rate arousal were simultaneously measured in response to a remote controlled robot (Lab-TAB; Goldsmith, Reilly, Lemery, Longley, & Prescott, 1999). Heart rate analyses revealed a main effect of task from baseline to fear ($p < .001$), no interaction effect and no effect for group. In other words, the pattern of arousal was similar for both groups. In addition, children with ASD showed intact facial and bodily expressions of fearful affect compared to TD children ($p = .82$). With regard to the relationship between expression and arousal, the results provided evidence for concordance between expression and arousal in TD children, but evidence for discordance in children with ASD. A possible missing link between arousal and expression in response to fear may significantly impact social functioning of children with ASD. In light of both early identification and treatment it is important to be aware that emotional expressions in children with ASD do not necessarily reflect their internal experience.

Introduction

Humans are biologically prepared to express and recognize emotional states, skills that form a crucial component in social behaviour (Cole & Moore, 2015; Darwin, Ekman, & Prodger, 1998; Shariff & Tracy, 2011). The development of children with Autism Spectrum Disorders (ASD) is partly characterized by persistent deficits in social communication and social interaction, such as social-emotional reciprocity, nonverbal communicative behaviors, and deficits in developing, maintaining, and understanding relationships (APA, 2013). The majority of studies in the field of autism research investigated social behaviour and related impairments in social cognition, such as how children with ASD attend to and interpret the emotions of others (Harms, Martin, & Wallace, 2010; Mazefsky, Pelphrey, & Dahl, 2012). Much less is understood about how children with ASD experience and express emotions themselves, which might serve as explanatory construct for behaviour problems frequently reported in ASD (Mazefsky et al., 2012; Samson, Hardan, Podell, Phillips, & Gross, 2015). This imbalance in research focus might in part be explained by the difficulties in measuring emotions and the large phenotypic heterogeneity in early ASD (Benevides & Lane, 2015; Kim, Macari, Koller, & Chawarska, 2016). This study aims to provide this by including both physiological and behavioural components of emotional responding, specifically in a fearful situation.

Emotion is a complex construct and requires the coordination between a physiological response, cognitive appraisal, expression, and a behavioural response (Hockenbury & Hockenbury, 2010), also described as *emotional concordance* (Hollenstein & Lanteigne, 2014). Expressing emotions (for example facial and bodily expression, tone of voice, and gestures) serves a communicatory function which is necessary for the environment to be able to rapidly understand what is needed (Blair, 2003). This may especially be relevant in early childhood, when children are more dependent on caregivers for their physical and emotional needs and when interaction patterns between the child and caregivers shape the developing brain. Emotional (facial) expressions have been the topic of research since Darwin's *The expression of the emotions in man and animals* (Darwin, 1872) and have been of specific interest with regard to the study of ASD. The nature and specificity of emotion expression difficulties however, do not seem universal for all individuals with ASD (Nuske, Vivanti, & Dissanayake, 2013). Jahromi and colleagues (2012) showed that children with ASD express no differences in facial and bodily negativity when confronted with frustrating situations. In a study by Stagg and colleagues (2013), adults blindly rated children with ASD as being less expressive than typically developing children. In addition, children with ASD were rated lower on friendship measures by typically developing peers, suggesting that facial expressivity might influence friendship appraisal (Stagg et al., 2013).

Because of this heterogeneity in emotion expression research, it is of interest to study the degree to which emotion expressions are related to internal states of emotional arousal as expressed in physiological parameters of the autonomic nervous system (ANS). Emotional

arousal is driven by the highly complex and interactive functioning of the ANS and the limbic system of which the amygdala is an important area (Yang et al., 2007). Heart rate is considered an indicator of ANS activity, either reflecting reduced parasympathetic activity and/ or increased sympathetic influence (Levenson, 2014). Studies on emotional arousal in typical development have shown arousal responses expressed in heart rate increase, however dependent on type of emotion, induction method, and duration of the physiological variable (for an extensive review see; Kreibig, 2010). For children with ASD there seems to be no evidence for autonomic differences during resting parasympathetic activity, however the literature does support different ANS patterns in *response* to a variety of tasks (Benevides & Lane, 2015). For example, the amount and direction of change during challenging tasks may be different in children with ASD. The majority of studies described in this review however included higher functioning and elementary/ middle school aged children with ASD, which limits generalizability to younger and lower functioning children with ASD.

The importance of the ANS as regulator, activator, coordinator, and communicator is reflected in the constant monitoring and adjusting of our functioning enabling the body to respond to internal and external demands (Levenson, 2014). The concordance between emotional expression and internal emotional states allows parents, caregivers, but also the social environment in general to be able to rely on the emotional expressions of children as a signal that may trigger the need for support, comfort, and help. An impediment in the concordance, or discordance, may disrupt caregiver responses which, over time, may amplify early vulnerability into a developmental trajectory of increasingly dysfunctional emotion regulation, social development, and early language skills (Kasari, Sigman, Mundy, & Yirmiya, 1990; Sullivan & Lewis, 2003; Wan et al., 2012). For example, being able to correctly label a situation as fearful (cognitive appraisal) would lead to an increase in arousal (enabling the body to reach a state of action), facial and/or bodily expression of affect (for the environment to *read* these emotions), and a behavioural response (for example to ask for help or to run away). Understanding the various components of the emotional response is important because specific impairments in emotion would lead to differential interventions to improve emotional behaviour. Simultaneously studying behaviourally expressed emotions and the internal psychophysiological state of arousal might provide us with more insight into the heterogeneous emotion problems reported in ASD as it is important that these channels are in tune with each other.

Even though theoretically assumed, empirical evidence regarding the relationship between internal arousal states and external behavioural expression is mixed due to for example the complexity of operationalizing emotions in research settings (Mauss & Robinson, 2009). Studies investigating these two components in young children with ASD are scarce. A study by Legiša and colleagues (2013) included eight children (8-14 years) with high-functioning ASD and matched controls. They examined the responses to odours using facial expression and arousal which showed relative intact expression and arousal compared to controls

(Legiša et al., 2013). A study by Stein and colleagues (2014) showed that physiological stress (as measured by non-specific skin conductance response frequency during routine dental cleanings) was significantly correlated with overt behavioral distress, indicating that as physiological stress increased so did behavioral distress in children with ASD (mean age 8.2 years). These results stress the importance of studying such sensitive and objective mechanisms of emotional behaviour at a young age during critical periods of development to identify and target the developmental trajectory of ASD (Kim et al., 2016; Zwaigenbaum et al., 2015). These early markers might serve as indicators for tailored treatment development (Dawson & Bernier, 2013; Landa, 2008).

This study aimed to measure emotional expressions and physiological arousal simultaneously in response to a fearful situation in children with ASD compared to typically developing children (TD). The fear paradigm was chosen because this experiment does not require social interaction with others, not to interfere with the social stress of emotion. A strong negative stressor was selected in order to exceed the threshold for an expression, however evidence regarding the relationship between emotion intensity and expression is ambiguous (Reisenzein, Studtmann, & Horstmann, 2013). We hypothesized that children with ASD would show relative intact facial and bodily expressions of negative affect compared to typically developing children based on previous research (e.g., Jahromi et al., 2012). Furthermore, based on the available literature (Benevides & Lane, 2015) we expected that children with ASD would show different ANS patterns (the amount and direction of change in heart rate) in response to the stressor. With regard to concordance, it was hypothesized that the relationship between external expression and internal arousal might be different for children with ASD compared to TD children.

Methods

Ethics statement

The current study is part of a larger longitudinal study which was approved by the Ethical Committee of the Leiden University Medical Center, Leiden, the Netherlands. A written informed consent, according to the declaration of Helsinki, was signed by the legal caretaker(s) of the participants.

Participants

In this study, 21 children with ASD (age range 43-75 months) and 45 TD children (age range 41-81 months) participated (Table 1). Children with ASD were recruited through the Dutch Autism Center (Rivierduinen), the Dutch Autism Association (NVA), and the Dutch Association for Developmental Disorders (Balans). TD children were recruited through day-care centres, elementary schools, and postings in public areas in the Netherlands. Inclusion criteria for all participants were that parents and/or children were Dutch or

English speaking, children had no neurological conditions, previous head injuries with loss of consciousness, and/or metabolic diseases.

Autism diagnosis

Autism diagnosis was provided in a multidisciplinary consensus meeting of child psychiatrists and child psychologists according to the DSM-IV-TR criteria (APA, 1994). Both the Autism Diagnostic Interview-Revised (ADI-R; Le Couteur, Lord, & Rutter, 2003) and the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000) were used to evaluate retrospective or current (depending on age) ASD symptoms. For the ADI, the diagnostic algorithm was used and for the ADOS, standardized severity scores were calculated (Gotham, Pickles, & Lord, 2009). All children exceeded the diagnostic threshold on both the ADI-R and the ADOS (Table 2). For the non-clinical control group, parental versions of the Social Responsiveness Scale (SRS; Constantino & Gruber, 2005) and the Childhood Behaviour Checklist (CBCL 1,5-5; Achenbach & Rescorla, 2000) showed normed sum scores below the clinical cut off in the non-clinical control group.

Table 1. Demographic characteristics of the ASD group and TD group.

	ASD (N= 21) Mean (SD)	TD (N= 45) Mean (SD)	Group differences
Age in months	60 (9.33)	56 (11.56)	$t(64) = -1.40, p = .17$
Gender	M= 20 , F= 1	M= 35, F= 10	$(\chi^2 (1)=3.14, p=.08)$
FSIQ	86.29 (21.64)	108.31 (14.20)	$t(28.32)= 4.26, p <.01^*$
SES †	2.26 (0.73)	2.60 (0.48)	$(\chi^2 (4)=8.11, p=.09)$

* Group difference significant at $p <.05$

† SES: 1= low, 2= medium, 3= high ($N_{ASD} = 19, N_{control} = 43$)

Table 2. ADI and ADOS scores for the ASD group (N=21).

Scale	Mean (SD)
ADI Social Communication (cut-off=10)	18.67 (6.10)
ADI Communication	Verbal (N=18, cut-off=8) 14.11 (3.88) Non-verbal (N=3, cut-off=7) 13.33 (1.16)
ADI Repetitive behaviour (cut-off=3)	6.10 (3.19)
ADI Developmental deviance (cut-off=1)	4.10 (1.22)
ADOS Severity score (N= 18)	7.94 (1.73)

Intellectual functioning

In the ASD group, intellectual functioning was assessed using the test that matched the child's verbal, motor, and developmental level. 13 children (62%) completed the Dutch Wechsler Nonverbal Scale of Ability (WNV-NL; Wechsler & Naglieri, 2006), two children (10%) the Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III-NL; Wechsler, 2006), three children (14%) the Snijders-Oomen Nonverbal Intelligence Test (SON-R 2.5-7; Tellegen, Winkel, Wijnberg-Williams, & Laros, 1998), and three children (14%) the Mullen Scales of Early Learning (MSEL; Mullen, 1995). Appropriate normative standard IQ scores were computed. In case raw scores were outside the standard range for deviation scores, a ratio IQ was computed by taking the average age equivalents across the subtests, divided by the chronological age in months, and multiplied by 100. IQ scores based on age equivalents are similar to the normative standard IQ scores (Bishop, Guthrie, Coffing, & Lord, 2011) (see Table 1). In the non-clinical control group, all children completed the WNV-NL.

Procedure of the experimental session

After confirmation of the ASD diagnosis and scores below the clinical cut-off for the TD children, participants were invited to the lab-visit. This visit aimed to assess both intellectual functioning and the fear experiment. Before the visit, participants were explicitly prepared with a visual information brochure and a copy set of the electrodes to adjust. Research took place in a carefully selected room with limited stimuli. Children were given time to familiarize before and after the electrodes were applied by playing an age appropriate game, whilst seated in an adapted car seat to have a stable position suited for physiological measurement.

Baseline

To measure baseline, children watched a 3-minute video of a fish tank, which has been shown to be an adequate measure of resting state (Benevides & Lane, 2015; Piferi, Kline, Younger, & Lawler, 2000). Heart rate over the course of the video was analysed in epochs of 30 sec each, to identify a series of two consecutive epochs (1 min) representing resting state. This was done on group level, for the nonclinical control group and the ASD group separately.

Laboratory Temperament Assessment Battery Fear Paradigm

Fear was induced using the unpredictable mechanical toy task of the Laboratory Temperament Assessment Battery (Lab-TAB; Goldsmith et al., 1999). For this particular study, a remote-controlled robot was used. Parents were asked to sit in the back of the room out of direct sight, filling out questionnaires. In case parents would judge the situation as too fearful for their child, they would be able to stop the experiment. This did not occur during the 90 seconds of the paradigm that were used for analyses. The procedure of the

task was executed according to the Lab-TAB manual, placing the robot 1.5 m away from the child. An experimenter entered the room in a white laboratory coat and protection glasses. The robot made three approaches of 30 seconds each, starting by walking towards the child, stopping 15 cm in front of the child moving its arms and emitting noise. Then, the robot walked backward pausing for 10 s before moving forward again, repeating this sequence three times in total. The unfamiliar experimenter left the room, leaving the robot behind. The procedure was videotaped from two angles. Because the current experiment is part of a larger longitudinal study, the task was counter balanced using two versions of the robot in order to account for familiarity effects; the robot in a white robe and a plain robot. A pilot study revealed that responses between the two versions were equal within the control group and results were therefore taken together in further analyses ($t(43) = 1.24, p .22$).

Observational coding

The 10 s epochs were coded according to Lab-TAB manual (Goldsmith et al., 1999) and the facial indicators of emotion as described in the Fear Action Scale (FACS; Ekman & Friesen, 1976). The peak intensity of the emotion was coded within this 10 s epoch to catch the burst of facial expression during these intervals. Facial and bodily fear were both scored on a four-point scale (0-3): neutral (0; no sign of facial or bodily fear), mild (1; one observable facial or bodily sign of fear), moderate (2; two observable signs of facial or bodily fear), and severe (3; more signs of facial or bodily fear). The scores within both categories were averaged across the epochs. A composite fear score was calculated derived by summing these averages, scores ranged between 0 and 6. Inter-rater reliability (IRR) was assessed using a two-way mixed, absolute agreement intra-class correlation model (Hallgren, 2012). The IRR was substantial for fear ($ICC = .71, p < .001$). Two trained independent coders scored all videos of which 29% were double coded. Interrater reliability was monitored continuously in regular consensus meetings. Discrepancies were discussed within the team to obtain a final consensus score. Distress vocalizations, as described in the Lab-TAB manual were not included in the analyses since our study focuses on the observable expression of emotion. Furthermore, considering the low level of language functioning in some of the ASD children in this sample, verbal capabilities were not taken into consideration.

Physiological arousal

Electrodes were attached, at the top centre of the chest, (10 centimetres below the suprasternal notch) the bottom left, and right of the ribs (10 centimetres above the bottom of the rib cage). Heart rate was recorded continuously during baseline and the robot approach with AcqKnowledge (Version 4.3.1. BIOPAC Systems Inc.). Recordings were acquired through an Electrocardiogram amplifier (ECG100C) and a BIOPAC data acquisition system (MP150 Windows) with a sampling rate of 200 Hz. In AcqKnowledge a 0.5 Hz highpass filter and a 50 Hz notch filter were applied to stabilize the ECG signal. Recorded physiological data

was further processed by inspecting the detected R peaks and valid interbeat intervals (IBI) in MATLAB Release 2012b (The MathWorks, Inc., Natick, Massachusetts, United States). Motion artefacts were visually identified and excluded from the data. Heart rate data were summarized in 30-second epochs, in concordance with the behavioural data.

Data analysis

No children were excluded after inspection for outliers and normality checks regarding the ECG and behavioural data. First, baseline levels between the TD group and the ASD group were analysed. Next, a GLM repeated measures analysis was performed with the between subject factor group (ASD, TD) and the within-subjects factor task (60 seconds baseline, three approaches of 30 seconds fear paradigm). To further analyse the heart rate pattern, paired samples *t*-tests were done. The expression of fear between groups was analysed using an independent samples *t*-test after which the dynamics between expression and arousal were analysed using Pearson correlations, Cohen's *q*, and Analyses of Variance (ANOVA) separately for both groups, with Expression as dependent variable and low versus high heart rate as factor. Effect sizes are reported as η_p^2 with .01 being a small, .06 medium, and .14 a large effect (Cohen, 1977). Cohen's *d* effect sizes were calculated for the paired samples *t*-tests with 0.2 being a small, 0.5 medium, and 0.8 a large effect.

Results

Intellectual functioning

Within both the ASD and the TD group, there was no significant relationship between IQ and heart rate (ASD $r = .10$, $p < .68$, TD $r = -.17$, $p = .26$) nor between IQ and emotional expression (ASD $r = .38$, $p < .09$, TD $r = .07$, $p = .66$). Therefore, IQ was not controlled for.

Psychophysiological arousal during baseline

Baseline heart rate levels did not differ between children with ASD ($M 97.37$, $SD 12.50$) compared to TD children ($M 92.27$, $SD 12.21$) ($t(64) = -1.57$, $p = .12$).

Psychophysiological arousal in response to fear

GLM repeated measure analysis with the between-subjects factor group (ASD, TD) and the within-subjects factor Task (one baseline epoch and three fear approaches) revealed a significant main effect of Task ($F(3,192) = 21.01$, $p < .001$, $\eta_p^2 = .25$), no significant effect of group ($F(1,64) = 3.01$, $p = .09$), and no interaction effect ($F(3,192) = .72$, $p = .54$). Within group comparisons showed a similar increase from baseline to each fear approach for both children with ASD and TD group. In other words, there was an increase in arousal in response to fear, and this increase was similar for both groups (Figure 1).

Table 3. Within group increases in arousal (heart rate) in response to fear

	ASD	TD
Baseline to approach 1	$t(20) = -2.99, p < .01^*, d 0.6$	$t(44) = -3.38, p < .01^*, d 0.3$
Baseline to approach 2	$t(20) = -2.94, p < .01^*, d 0.6$	$t(44) = -4.31, p < .01^*, d 0.5$
Baseline to approach 3	$t(20) = -3.83, p < .01^*, d 0.8$	$t(44) = -4.55, p < .01^*, d 0.6$

* Significant at $p < .05$. Effect sizes displayed in Cohens d .

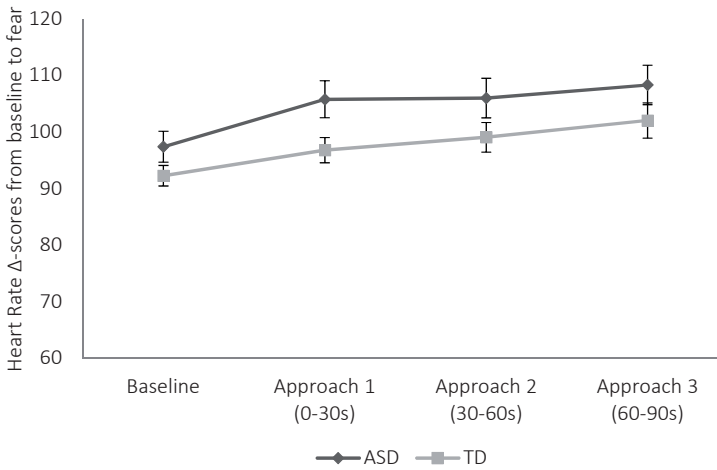


Figure 1. Heart rate at baseline and in response to fear. Error bars displaying SE .

Emotional expression of fear

Independent samples t -tests showed that children with ASD expressed equal levels of overall emotional fear compared to TD children as reflected in average expression across the total duration of the fear exposure ($t(64) = -.23, p = .82$). In other words, the expression of negative emotions in facial expression and body posture in response to fear was equal between children with ASD and TD children (see Figure 2).

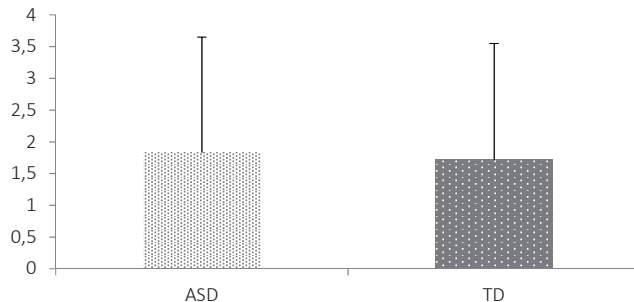


Figure 2. Absolute levels of expression in response to fear for ASD ($N=21$) and TD children ($N=45$)

Concordance between arousal and expression

To investigate the concordance between heart rate and the expression of fear, Pearson correlations were calculated within each group. Arousal response during the fear paradigm (as expressed in average heart rate during fear exposure) showed a positive correlation with expression of fear in the control group ($r = .45, n = 45, p < .01$), with 19.9% explained variance. In contrast to this result, there was no correlation between the arousal response and expression of fear in the ASD group ($r = .20, n = 21, p = .38$). To check whether any differences in sample sizes between the groups may account for the differences in significance of the correlations, we used the SPSS *select random sample of cases* function to randomly form subsamples of the TD group of equal size as the ASD group. Results showed that correlations remained significant in different subsamples (see Table 4).

To investigate the significance between the Pearson correlations in the TD group and ASD group, Cohen's q was calculated (Cohen, 1977). Correlations were transformed to Fisher's Z , after which the difference was calculated. Cohen's q was .32, indicating that group differences were of a medium effect size. In other words, the TD children who showed an increase in arousal were also the children who showed higher level of expression, in contrast to the ASD children.

Table 4. Multiple random sub-samples within the TD group ($N = 45$)

n	Pearson r	Significance
22	.41	.05*
22	.75	<.01*
22	.52	.01*

* Significant at $p < .05$.

To further investigate group differences in the concordance between arousal and expression, both the ASD group and the TD group were divided into a low- and high-arousal group using a median split. The median heart rate in the TD group was 96 BPM. The low-arousal group consisted of 22 children and the high arousal group of 23. The same was done for children in the ASD group, for which the median heart rate was 101 BPM, with 11 children in the low arousal group and 10 children in the high arousal group. ANOVA analyses, separate for the ASD and the TD group, with Expression as dependent variable and low versus high arousal as factor were conducted to determine if the level of facial and bodily expressions was different for groups with either high or low arousal. Results revealed that, within the TD group, the level of expression in response to fear was significantly different for high versus low aroused children ($F(1, 43) = 10.61, p < .01$), with significantly higher expression in high aroused children. Within the ASD group, expression in response to fear was not different between high versus low aroused children ($F(1, 19) = .52, p = .48$). This is illustrated in Figure 3.

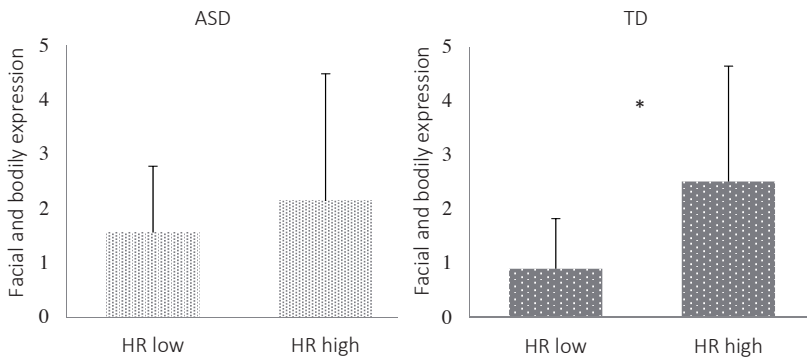


Figure 3. Facial and bodily expression of fear for low versus high arousal. ASD group left ($N= 21$) and TD group right ($N= 45$). Error bars displaying SD .

Discussion

The present study examined physiological arousal and emotional expression in response to fear in young children with Autism Spectrum Disorders (ASD) compared to typically developing children (TD). By simultaneously addressing external components of emotion as expressed in facial and bodily expressions and internal components of emotion as expressed in emotional arousal parameters of the Autonomic Nervous System (ANS), we aimed to provide more insight into possible differential deficits and concordance between these two layers of the complex construct of emotion.

Baseline measures of heart rate revealed no differences in resting state arousal between the ASD group and TD children, which is in line with previous studies (for a review see; Benevides & Lane, 2015). In response to fear, both heart rate patterns and levels of children with ASD revealed increased arousal to the same extent and pattern as TD children. This indicates that children with ASD have autonomic responses when their own emotions are triggered in a fearful situation, similar to TD children. These results provide evidence for an intact emotional arousal in response to fear in children with ASD. Previous studies, mainly concerning older individuals and dominantly high functioning, yielded mixed results regarding cardiac arousal in children with ASD possibly due to the varying nature of the measures that complicated comparisons (Benevides & Lane, 2015). The current study was designed to measure arousal without the possible interference of social interaction aspects, by introducing a stressor that did not require interaction from the children. Another study also reported significant changes in heart rate from baseline to a non-social stimulus (Stroop task) in children with ASD equal to TD children aged 8-15 years (Kushki et al., 2013). This might indicate that the emotional arousal response of children with ASD is intact when it comes to their own emotions, however deviant when it comes to responding to emotions of others. In support of this, a pilot study from Levine and colleagues (2012) showed equal ANS activity as measured with skin conductance between children (8-12 years old) with high

functioning autism and a comparison group in response to a social stressor (Trier Social Stress Test). However, cortisol measures during this task showed significant increases in cortisol response in the TD group, and a blunted or even decreased cortisol response in the ASD group. The authors are cautious with interpreting this result, since important factors such as age (8-12 years old), gender, and pubertal level could be of influence on the cortisol results and replication is needed. A recent eyetracking study in young children with ASD showed diminished arousal (heart rate) in response to the emotions of others in comparison to typically developing children (Zantinge, van Rijn, Stockmann, & Swaab, 2017). The either social- or non-social nature of the stimulus should be further investigated to better understand mixed results regarding arousal in children with ASD.

In addition to emotional arousal, we also examined facial and bodily expressions in response to fear. Structured behavioural observations revealed no differences in the intensity of fear expressions between the two groups. However, high amounts of individual variability were noted. Earlier studies also reported relatively intact expression of emotions in children with ASD in response to a negative stimulus, although both of these studies included children with high functioning ASD and the study by Legiša included older children (Jahromi et al., 2012; Legiša et al., 2013). With respect to expressivity, a study by Stagg and colleagues (2013) used videos of children with ASD being interviewed about their daily lives, families, and interests which were rated as being less expressive than TD children. These contradicting results regarding facial and bodily expressivity of children with ASD might be caused by the large individual variability and/or the intensity of the experienced emotion. It could be that subtle or ambiguous expressions are more impaired in children with ASD than unambiguous elicited negative emotions in response to a strong stressor. In daily life, especially in situations with a social nature, interactions are packed with subtle and ambiguous expressions that require the other to encode and respond in a prompt and appropriate manner. In sum, our study shows that with regard to own experienced emotions without social context, children with ASD do show appropriate levels of negative expressions that can clearly be observed on the outside.

Since average levels of arousal response and the accompanying expressions were intact in children with ASD, individual differences gave insight into the variance in both groups and the concordance between expression and arousal. These results revealed that emotional arousal and expression were significantly related within the TD group, but not within the ASD group. TD children who showed an increase in arousal were also the children with higher levels of fear expressions and vice versa. The results remained even after randomly reducing the sample size to match the sample size of ASD group to rule out that this was due to power. Further analyses revealed that the level of expression was different for TD children that were low versus high aroused. In contrast, children with ASD who were highly aroused, did not show increased intensity of expression compared to low aroused children with ASD. This indicates that children with ASD who experience an increase in

heart rate, do not necessarily show an increase in expressiveness and vice versa, in contrast to TD children. The difference concordance between children with ASD and TD children represented a medium effect size.

The importance of concordance is illustrated by a study from Mauss and colleagues (2011) who found that discordance was associated with later increased depressive symptoms, lower well-being, and over time undermined psychological functioning in undergraduate female students. Research also showed that discordance can confuse others about actual internal states and could lead to behaviour being interpreted as not trustworthy (Mauss et al., 2011). However, greater concordance does not automatically mean better emotional behaviour (Butler, Gross, & Barnard, 2014), so considering the context of the emotion involved is important. As some have suggested, different components of emotions in circumstances that have little emotional relevance are only loosely connected (Bulteel et al., 2014) and that the appearance of concordance is only associated to relatively strong emotions (Russell, 2003). Even though this last notion has also been debated (Reisenzein et al., 2013), our study provides evidence for the existence of concordance in a sample of TD children which stresses the need for future studies on this topic in young children since the majority of studies has been conducted in adults.

The seemingly absent relationship between expression and arousal in children with ASD might provide evidence that interacting systems are less dependent on each other or at least represent abnormalities in the automatic connections between brain systems responsible for generating behavioural *expression* and brain systems responsible for *experienced* arousal. Two main brain regions (among others) involved in the experience and expression of emotions are the amygdala and the insula. The amygdala plays a critical role in the processing emotional information and, as part of the social brain, has been linked to social-emotional problems in individuals with ASD (Baron-Cohen et al., 2000). In the normal population, amygdala activity has been related to heart rate in healthy adolescents (Yang et al., 2007) and to high intensity facial expressions compared to low intensity and neutral facial expressions (Lin et al., 2016). The insula is important for the sensory perception of emotions, but is also involved in generating emotional and communicative facial expression (Jezzini et al., 2015). In other words, the insula enables the translation of an observed or imitated facial emotional expression into its internally felt emotional significance (Carr, Iacoboni, Dubeau, Mazziotta, & Lenzi, 2003; Dapretto et al., 2006). A study by Giuliani and colleagues (2011), showed that anterior insula volume was positively correlated to expressive suppression (Giuliani et al., 2011). The anterior insula (together with motor as well as somato- and limbic-sensory processing) has also been related to the expression of pleasant facial affect (Hennenlotter et al., 2005). Down-regulation of negative emotions reflected in reduced activation of emotional arousal-related brain structures like the amygdala and the insula have also been found (Ochsner & Gross, 2005). There is evidence for abnormalities of the limbic system, in particular the amygdala and the insula, that may be responsible

for social-emotional characteristics of ASD (Gaigg & Bowler, 2007). The current study at least provides evidence for poor connectivity between emotional arousal and emotional expression, and might fit with the idea that ASD symptoms may arise as a consequence of disconnection between various functional brain systems, rather than impairments in one single area.

There are some limitations and related suggestions for future research in light of this study. The sample sizes did not allow for more complex statistics to consider patterns of changes in real-time in response to the task. For future studies it is recommended to look at these (individual) patterns of concordance in more detail (for recommendations see; Bulteel et al., 2014; Hollenstein & Lanteigne, 2014). The current sample included a limited amount of girls with ASD which limited comparability between genders. Also, our study included no other indices of arousal than heart rate and expressive behaviour in response to fear. It would be recommended for future studies to also include measures of emotion regulation, cognitive measures, self-report, a broader range of emotions, and other indices of ANS functioning to gain a more complete picture on the different levels of emotion. As heart rate is modulated by both sympathetic and parasympathetic branches of the ANS, measures of ANS activity should be explored further (Benevides & Lane, 2015). In line with this, the current results must be seen in light of a fear-specific response. Different responses might be observed in response to other emotions. Considering the limited amount of comparative studies on this topic in young children with ASD, there is need for caution as well as more research to further explore the results described in the current study. Finally, the results of the current study and the methods used could have important implications to the literature regarding anxiety in children with ASD. The prevalence of anxiety disorders in children with ASD is thought to be as high as 30% however the underlying mechanisms and risk factors for these comorbid problems remain unclear (Kim, Szatmari, Bryson, Streiner, & Wilson, 2000; Simonoff et al., 2008; White, Oswald, Ollendick, & Scahill, 2009).

A missing link between arousal and expression in response to emotions (in this case fear) may impact social functioning of children with ASD. The ability to interpret children's expressive signals is essential to promote mutually satisfying interactions (Sullivan & Lewis, 2003). Better understanding emotional responding in children with ASD is important, and awareness of discordance between expression and arousal may be meaningful in psychoeducation and intervention strategies. Considering the increasingly young age at which interventions are implemented (Kim et al., 2016), this could be achieved by raising awareness in the social environment of the children that emotional expressions in children with ASD do not necessarily reflect their internal experience. When children are (mentally) old enough, these interventions could include new techniques that enable children to practice expressive behaviours with techniques such as portable video modelling, which have yielded positive results, especially with regard to generalizability of the learned behaviours (Macpherson, Charlop, & Miltenberger, 2015).

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