Concordance between physiological arousal and emotion expression during fear in young children with autism spectrum disorders

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Abstract

This study aimed to measure emotional expression and physiological arousal in response to fear in 21 children with autism spectrum disorders (43–75 months) and 45 typically developing children (41–81 months). Expressions of facial and bodily fear and heart rate arousal were simultaneously measured in response to a remote controlled robot (Laboratory Temperament Assessment Battery). Heart rate analyses revealed a main effect of task from baseline to fear ($p < 0.001$, $\eta_p^2 = 0.25$), no interaction effect and no effect for group. In addition, children with autism spectrum disorder showed intact facial and bodily expressions of fearful affect compared to typically developing children. With regard to the relationship between expression and arousal, the results provided evidence for concordance between expression and arousal in typically developing children ($r = 0.45$, $n = 45$, $p < 0.01$). For children with autism spectrum disorder, no significant correlation was found ($r = 0.20$, $n = 21$, $p = 0.38$). A moderation analysis revealed no significant interaction between expression and arousal for children with and without autism spectrum disorder ($F(1, 62) = 1.23$, $p = 0.27$, $\eta_p^2 = 0.02$), which might be the result of limited power. The current results give reason to further study concordance between expression and arousal in early autism spectrum disorder. Discordance might significantly impact social functioning and is an important topic in light of both early identification and treatment.

Keywords

arousal, autism, concordance, expression, fear

Introduction

Humans are biologically prepared to express and recognize emotional states, skills that form a crucial component in social behaviour (Cole and Moore, 2015; Darwin et al., 1998; Shariff and 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 behaviours 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 et al., 2010; Mazefsky et al., 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 et al., 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 and Lane, 2015; Kim et al., 2016). This study aims to provide this by including both physiological and behavioural components of emotional responding, specifically in a fearful situation.

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Emotion is a complex construct and requires the coordination between a physiological response, cognitive appraisal, expression and a behavioural response (Hockenbury and Hockenbury, 2010), also described as *emotional concordance* (Hollenstein and Lanteigne, 2014). Expressing emotions (e.g. 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, 1965 [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 et al., 2013). Jahromi et al. (2012) showed that children with ASD express no differences in facial and bodily negativity when confronted with frustrating situations. In a study by Stagg et al. (2013), adults blindly rated children with ASD as being less expressive than typically developing (TD) children. In addition, children with ASD were rated lower on friendship measures by TD 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 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 and 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 et al., 1990; Sullivan and 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 (e.g. 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.

Although 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 and Robinson, 2009). Studies investigating these two components in young children with ASD are scarce. A study by Legiša et al. (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 et al. (2014) showed that physiological stress (as measured by non-specific skin conductance response frequency during routine dental cleanings) was significantly correlated with overt behavioural distress, indicating that as physiological stress increased so did behavioural 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 and 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 TD children. The fear paradigm was chosen because this experiment

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

	ASD (N = 21)	TD (N = 45)	Group differences
	Mean (SD)	Mean (SD)	
Age in months	60 (9.33)	56 (11.56)	$t(64) = -1.40$, $p = 0.17$
Gender	M = 20, F = 1	M = 35, F = 10	(χ^2 (1) = 3.14, $p = 0.08$)
FSIQ	86.29 (21.64)	108.31 (14.20)	$t(28.32) = 4.26$, $p < 0.01*$
SES[a]	2.26 (0.73)	2.60 (0.48)	(χ^2 (4) = 8.11, $p = 0.09$)

ASD: autism spectrum disorder; TD: typically developing; SD: standard deviation; FSIQ: Full Scale Intelligence Quotient; SES: socio-economic status.
[a]SES: 1 = low, 2 = medium, 3 = high ($N_{ASD} = 19$, $N_{control} = 43$).
*Group difference significant at $p < 0.05$.

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 et al., 2013). We hypothesized that children with ASD would show relative intact facial and bodily expressions of negative affect compared to TD children based on previous research (e.g. Jahromi et al., 2012). Furthermore, based on the available literature (Benevides and 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

This 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 Centre (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 *Diagnostic and Statistical Manual of Mental Disorders* (4th ed., text rev.; DSM-IV-TR) criteria (APA, 1994). Both the Autism Diagnostic Interview–Revised (ADI-R; Le Couteur et al., 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-R, the diagnostic algorithm was used and for the ADOS, standardized severity scores were calculated (Gotham et al., 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 and Gruber, 2005) and the Childhood Behavior Checklist (CBCL 1.5–5; Achenbach and Rescorla, 2000) showed normed sum scores below the clinical cut-off in the non-clinical control group.

Intellectual functioning

In the ASD group, intellectual functioning was assessed using the test that matched the child's verbal, motor and developmental level. In all, 13 children (62%) completed the Dutch Wechsler Nonverbal Scale of Ability (WNV-NL; Wechsler and Naglieri, 2006), 2 children (10%) the Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III-NL; Wechsler, 2006), 3 children (14%) the Snijders-Oomen Nonverbal Intelligence Test (SON-R 2.5–7; Tellegen et al., 1998) and 3 children (14%) the Mullen Scales of Early Learning (MSEL; Mullen, 1995). Appropriate normative standard intelligence quotient (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 et al., 2011; see Table 1). In the non-clinical control group, all children completed the WNV-NL.

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

Scale		Mean (SD)
ADI-R social communication (cut-off = 10)		18.67 (6.10)
ADI-R communication	Verbal (*N* = 18, cut-off = 8)	14.11 (3.88)
	Non-verbal (*N* = 3, cut-off = 7)	13.33 (1.16)
ADI-R repetitive behaviour (cut-off = 3)		6.10 (3.19)
ADI-R developmental deviance (cut-off = 1)		4.10 (1.22)
ADOS severity score (*N* = 18)		7.94 (1.73)

ADI-R: Autism Diagnostic Interview–Revised; ADOS: Autism Diagnostic Observation Schedule; ASD: autism spectrum disorder; SD: standard deviation.

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 for a lab-visit. This visit aimed to assess both the 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 familiarize. 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, while seated in an adapted car seat to have a stable position suited for physiological measurement.

Baseline

To measure baseline, children watched a 3-min video of a fish tank, which has been shown to be an adequate measure of resting state (Benevides and Lane, 2015; Piferi et al., 2000). Heart rate over the course of the video was analysed in epochs of 30 s each, to identify a series of two consecutive epochs (1 min) representing resting state. This was done on group level, for the non-clinical 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 s of the paradigm that was 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 s 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 counterbalanced 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* = 0.22).

Observational coding

The videos were coded in 10-s epochs (with the sound on) according to Lab-TAB manual (Goldsmith et al., 1999) and the facial indicators of emotion as described in the Fear Action Scale (FACS; Ekman and 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 4-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 the 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 (intra-class correlation coefficient (ICC) = 0.71, *p* < 0.001). Two trained independent coders scored all videos of which 29% were double coded. IRR 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.

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

	ASD	TD
Baseline to approach 1	$t(20) = -2.99$, $p < 0.01$,* d 0.6	$t(44) = -3.38$, $p < 0.01$,* d 0.3
Baseline to approach 2	$t(20) = -2.94$, $p < 0.01$,* d 0.6	$t(44) = -4.31$, $p < 0.01$,* d 0.5
Baseline to approach 3	$t(20) = -3.83$, $p < 0.01$,* d 0.8	$t(44) = -4.55$, $p < 0.01$,* d 0.6

ASD: autism spectrum disorder; TD: typically developing.
Effect sizes displayed in Cohen's *d*.
*Significant at $p < 0.05$.

Physiological arousal

Electrodes were attached at the top centre of the chest (10 cm below the suprasternal notch), the bottom left and to the right of the ribs (10 cm 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 (ECG) amplifier (ECG100C) and a BIOPAC data acquisition system (MP150 Windows) with a sampling rate of 200 Hz. In AcqKnowledge, a 0.5-Hz high-pass 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, MA, USA). Motion artefacts were visually identified and excluded from the data. Heart rate data were summarized in 30-s 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 and ASD groups were analysed. Next, a generalized linear model (GLM) repeated-measures analysis was performed with the between-subject factor group (ASD, TD) and the within-subjects factor task (60 s baseline, three approaches of 30 s 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 concordance between expression and arousal was analysed using Pearson correlations, Cohen's *q* and analysis of variance (ANOVA) separately for both the groups, with Expression as dependent variable and low versus high heart rate as factor. Finally, a moderation analysis (2×2 ANOVA) was conducted with Expression as dependent, low versus high aroused as independent and both groups as fixed factors to explore whether there was an interaction effect. As this was an exploratory analysis (due to a lack of statistical power), results were interpreted with caution. Effect sizes are reported as η_p^2 with 0.01 being a small, 0.06 medium and 0.14 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, and 95% confidence intervals (CIs) are reported as 95% CI.

Results

Intellectual functioning

Within both the ASD and TD groups, there was no significant relationship between IQ and heart rate (ASD $r = 0.10$, $p < 0.68$, TD $r = -0.17$, $p < 0.26$) nor between IQ and emotional expression (ASD $r = 0.38$, $p < 0.09$, TD $r = 0.07$, $p < 0.66$). 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; $t(64) = -1.57$, $p = 0.12$).

Psychophysiological arousal in response to fear

GLM repeated-measures 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 ($F(3, 192) = 21.01$, $p < 0.001$, $\eta_p^2 = 0.25$), no significant effect of group ($F(1, 64) = 3.01$, $p = 0.09$) and no interaction effect ($F(3, 192) = 0.72$, $p = 0.54$). Within-group comparisons showed a similar increase from baseline to each fear approach for children both in the ASD and TD groups. In other words, there was an increase in arousal in response to fear and this increase was similar in both the groups (Table 3 and Figure 1).

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 ($t(64) = -0.23$, $p = 0.82$). In other words, the expression of negative emotions in facial expression and body posture in response to fear was equal in children in both the ASD and TD groups (see Figure 2).
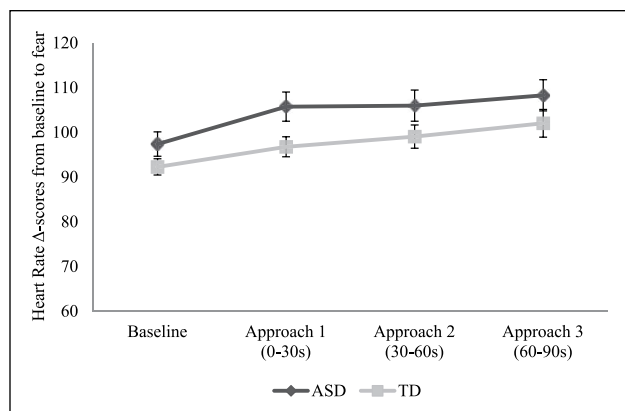
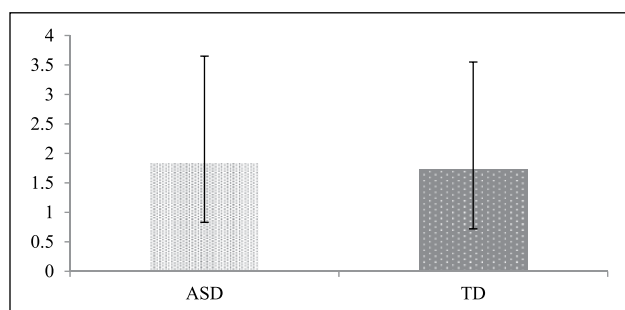**Figure 1.** Heart rate at baseline and in response to fear. Error bars display *SE*.

Figure 2. Absolute levels of expression in response to fear in 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 significant correlation with expression of fear in the control group ($r=0.45$, $n=45$, $p<0.01$), with 19.9% explained variance. There was no correlation between the arousal response and expression of fear in the ASD group ($r=0.20$, $n=21$, $p=0.38$). To check whether any differences in sample size 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 sub-samples of the TD group of equal size as the ASD group. Results showed that correlations remained significant in different TD sub-samples (see Table 4).

To test the significance between the Pearson correlations in the TD group and ASD group as expressed in an effect size, Cohen's *q* was calculated (Cohen, 1977). Correlations were transformed to Fisher's *Z*, after which the difference was calculated. Cohen's *q* was 0.28 (95% CI=−0.27 to 0.83), indicating that the difference (in strength of the correlation) between the TD group and

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

n	Pearson *r*	Significance
22	0.41	0.05*
22	0.75	<0.01*
22	0.52	0.01*

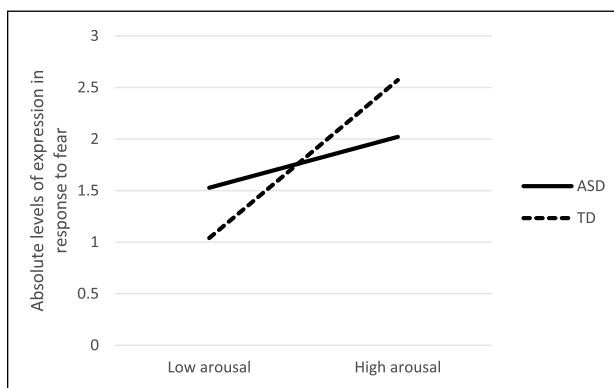TD: typically developing.
*Significant at $p<0.05$.

Figure 3. Levels of expression in response to fear in low-aroused and high-aroused children, stratified by group.

the ASD group was of medium effect size, which, however, needs to be interpreted with caution considering the 95% CI.

To further explore possible group differences in the concordance between arousal and expression, both groups were divided into a low- and high-arousal group using a median split. The median heart rate was 100 BPM. The low-arousal group consisted of 25 TD children and 8 ASD children. The high-arousal group consisted of 20 TD children and 13 children from the ASD 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 ($F(1, 43)=9.28$, $p<0.01$, Cohen's $d=0.9$, 95% CI=0.30–1.53 indicating a large effect size), with a 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 ($F(1, 19)=0.35$, $p=0.56$, Cohen's $d=0.3$, 95% CI=−0.62 to 1.15, indicating a medium effect size). Finally, a moderation analysis (see Figure 3) including both the TD and ASD groups showed no significant interaction effect between arousal and expression across groups on a multivariate level ($F(1, 62)=1.23$, $p=0.27$, $\eta_p^2 = 0.02$).

Discussion

This study examined physiological arousal and emotional expression in response to fear in young children with ASD compared to TD children. 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 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 and TD group children, which is in line with previous studies (for a review see; Benevides and 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 and Lane, 2015). This 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, it is deviant when it comes to responding to emotions of others. In support of this, a pilot study by Levine et al. (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 eye-tracking study in young children with ASD showed diminished arousal (heart rate) in response to the emotions of others in comparison to TD children (Zantinge et al., 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 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 et al. (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. For children with ASD, no significant relation between expression and arousal was found. If replicated, this could suggest that children with ASD, who are highly aroused, do not show increased intensity of expression compared to low aroused children with ASD. The difference in concordance between children with ASD and TD children represented a medium effect size; however, this result needs to be interpreted with caution. Finally, the moderation analysis did not reveal an interaction effect, possibly due to a limited sample size, calling for further research in larger groups that allows for such a multivariate analysis approach. Although no multivariate interaction was found, taken together, the current results do provide a reason to further investigate the concept of concordance in larger samples of children with ASD. The importance of concordance is illustrated in a study by

Mauss et al. (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 et al., 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). Although 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.

If studies with larger samples show that children with ASD are indeed characterized by reduced concordance between the expression and experience of emotion, this may provide important insights into interacting systems that are less dependent on each other or at least point to 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 et al., 2003; Dapretto et al., 2006). A study by Giuliani et al. (2011) showed that anterior insula volume was positively correlated to expressive suppression. 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 and Gross, 2005). There is evidence for abnormalities of the limbic system, in particular the amygdala and the insula, which may be

responsible for social-emotional characteristics of ASD (Gaigg and Bowler, 2007). This study at least provides some suggestion of 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 were small and prevented more thorough statistical analyses such as evaluation of more complex multivariate interaction effects and statistics to consider patterns of changes in real-time in response to the task. For future studies, it is recommended to repeat the moderation analysis with larger groups (this is supported by the medium effect size that was found) and look at (individual) patterns of concordance in more detail (for recommendations, see Bulteel et al., 2014; Hollenstein and 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 and 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 this study. Finally, the results in this 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 et al., 2000; Simonoff et al., 2008; White et al., 2009).

It may be very relevant to better understand the degree of concordance or discordance between arousal and expression in response to emotions of children with ASD, in order to gain insight in underlying mechanisms of social developmental problems. The ability to interpret children's expressive signals is essential to promote mutually satisfying interactions (Sullivan and Lewis, 2003). If replicated, awareness of any discordance could lead to a better understanding of emotional responding in children with ASD. Awareness of a possible discordance between expression and arousal may also 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 et al., 2015).

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