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Temperament in the First 2 Years of Life in Infants at High-Risk for Autism Spectrum Disorders

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Abstract The current study investigated early temperament in 54 infants at familial high-risk of ASD and 50 controls. Parental report of temperament was assessed around 7, 14 and 24 months of age and diagnostic assessment was conducted at 3 years. The high-risk group showed reduced *Surgency* at 7 and 14 months and reduced *Effortful Control* at 14 and 24 months, compared to controls. High-risk infants later diagnosed with ASD were distinguished from controls by a temperament profile marked by increased *Perceptual Sensitivity* from the first year of life, and increased *Negative Affect* and reduced *Cuddliness* in the second year of life. Temperament may be an important construct for understanding the early infant development of ASD.

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Introduction

ASDs are neurodevelopmental disorders resulting from genetic and other organic etiological factors which affect brain development very early in life (Levy et al. 2009). While symptoms emerge during infancy, diagnosis is not typically made until 3–4 years of age (Mandell et al. 2005; Yirmiya and Charman 2010). Attention has recently turned to the study of infant siblings of children with ASD (Elsabbagh and Johnson 2010; Rogers 2009; Zwaigenbaum et al. 2007), given highly heritability of the condition (i.e., later-born siblings of diagnosed children are at substantially higher risk for developing ASD than are those in the general population; Bolton et al. 1998; Ozonoff et al. 2011). Whilst facilitating the discovery of early signs of ASD, such studies also demonstrate that, as a group, high-risk infants like other undiagnosed relatives of individuals with ASD, often share some features of the condition; referred to as the Broader Autism Phenotype (BAP; e.g., Bailey et al. 1998). Despite advances in the understanding and early identification of ASD, diagnosis remains behaviourally-based, and heterogeneity in the presentation of ASD is apparent in terms of symptom onset, developmental course, and in associated levels of cognitive and adaptive functioning (Bryson et al. 2007).

Adopting an individual-differences approach can assist in addressing the heterogeneity observed within ASD, and has provided impetus for investigation of the construct of *temperament* in early ASD. Temperament has been defined as a ‘behavioural style’ (Thomas and Chess 1977), and a constitutionally-based pattern of individual differences in reactivity and self-regulation, emphasising emotional, attentional, and activity-related characteristics of the child (Rothbart and Derryberry 1981). Like the behavioural presentation of ASD, temperament too is considered to *emerge* across early development (Rothbart et al. 2000), and may provide a key to understanding symptom emergence on the course toward ASD outcome. Mundy et al. (2007) propose a 'modifier model' of autism, stating that modifier processes influence symptom expression, contributing to the observed variability. Temperament is proposed as one such modifier, alongside factors such as socialisation and cognitive style. Modifier processes need not be syndrome-specific, and may also include genetic influences which are independent of those carrying risk for the condition in question, but which influence its phenotypic expression (Mundy et al. 2007). Rothbart et al. (1995) argue that temperament can increase or buffer the risk of psychopathology, the course of a disorder, and response to intervention. As such, studying individual-difference factors, including temperament, in the context of ASD, affords the potential for earlier detection of emerging symptoms ASD, as well as for the appropriate later selection of therapeutic interventions.

Temperament in Typical Development

The study of temperament in typical infants has been comprehensive and facilitated by the well-validated battery of parent-report measures developed and revised by Rothbart and colleagues. Age-specific instruments capture overt behavioural manifestations of core underlying temperament factors, which present relatively consistently across different periods of early life, and correspond to important later aspects of personality, social interaction skill, and psychopathology (see Rothbart et al. 2001). Across the Infant Behaviour Questionnaire—Revised (IBQ-R; Gartstein and Rothbart 2003, for infants aged up to 18-months), the Early Childhood Behavior Questionnaire (ECBQ; Putnam et al. 2006, for toddlers aged to 3-years), and the Child Behavior Questionnaire (CBQ; Rothbart et al. 2001, for children aged to 7-years), temperament traits appear to cluster robustly around three broad factors; Surgency, Negative Affectivity and Effortful Control.

Surgency largely comprises approach behaviours, demonstrated from around 2- to 3-months through smiling, laughter, and other body movements (Rothbart 2007), and predictive of later approach and positive affective behaviours within trait extraversion (Rothbart et al. 2000). Negative Affectivity behaviours of anger and frustration also emerge around 2- to 3-months, followed by fear from around 7- to 10-months (Rothbart 1988). Rothbart et al. (2000) found these traits to remain stable until 7-years. Furthermore, infant fear reactions were predictive of later internalizing characteristics and negatively related to externalizing characteristics, while infant frustration and anger (particularly around 10-months) were related to both internalizing and externalizing. Effortful control—the ability to regulate attention, emotions, and behaviour to achieve goals—has recently been argued to emerge around the end of the first year of life, but remaining unstable until late toddlerhood (Kochanska et al. 2000). Rothbart and Bates (1998), however, claimed that this factor first develops in later infancy and toddlerhood, and may continue to develop right through adolescence.

Temperament in Early ASD

Children with ASD have been rated by their parents as less rhythmic, adaptable and persistent, and as more withdrawn and negative, than children with other delays (Bailey et al. 2000). Furthermore, Kasari and Sigman (1997) found that more temperamentally 'difficult' children were less engaged and responsive when interacting with a parent or an experimenter, suggesting an association between temperamental and social skills variation. To date, three studies (only one of which has been published) have assessed temperamental differences in children with ASD (aged 3–10 years) and controls using the measures devised by Rothbart and colleagues. Lower Effortful Control was consistently found to differentiate children with ASD from controls—particularly difficulties focussing and shifting attention, and attaining inhibitory control (Janes 2001; Konstantareas and Stewart 2006; Landry 1998). Parents have also reported children with ASD to have greater Negative Affect, displaying more discomfort and presenting as less sootheable (Konstantareas and Stewart 2006), and as more angry and perceptually sensitive (Landry 1998) than their peers. While no group differences have been reported regarding the overall factor of Surgency, Landry (1998) reported higher activity levels and less positive affect and approach for children with ASD, than controls.

Existing research on infant temperament comes mostly from retrospective studies alongside a small number of recent prospective studies. Retrospective analysis of home-video footage points to very early differences in the attention and affect of infants developing ASD (Baranek 1999; Clifford and Dissanayake 2008; Werner et al. 2000), while retrospective parent reports have suggested that these infants often show a lack of positive affect (Clifford and Dissanayake 2008; Young et al. 2003) and increased negative affect (Clifford and Dissanayake 2008; Watson et al. 2007) compared to controls, alongside greater detachment, hypersensitivity, impulsivity, and self-regulatory impairments (Gomez and Baird 2005). Within a prospective case series, Bryson et al. (2007) found each of nine high-risk infants who later developed ASD to present early temperament profiles characterised by irritability, intolerance of intrusion, proneness to distress, and regulatory difficulties. In a larger, prospective study of 65 high-risk infants, Zwaigenbaum et al. (2005) found that those who given an ASD ‘classification’ at 24-months had demonstrated trait passivity at 6-months, followed by decreased positive affect, extreme distress reactions, and inability to easily disengage visual attention at 12-months, all of which play an important role in the development of later self-regulation ability (Rothbart et al. 1992). In a later publication, Zwaigenbaum and colleagues reported 24-month temperament data collected from the same high-risk group, now followed-up to 36-months (Garon et al. 2009). ASD diagnosis was confirmed in 34 individuals (20.8 % of the group), with these toddlers distinguishable from other high-risk toddlers and controls on the basis of presenting a profile of ‘Effortful Emotion Regulation’, comprising low positive affect, high negative affect, and difficulty controlling attention and behaviour. Finally, in a large, prospective population study, Bolton et al. (2012) found few temperamental differences between 6-month-old infants who went on to receive an ASD diagnosis and those who did not. By 24-months, however, clear group differences had emerged, including increased activity and reduced distractibility in the former.

Existing results provide evidence of the potential benefits to be gained by adopting an individual-differences approach to the investigation of ASD, presenting a fairly consistent picture of an early temperament profile characteristic of infants later diagnosed with ASD. A profile featuring poorly regulated attentional and behavioural control and mood is apparent from within the first year of life, and comparison with studies of the temperament profiles of older children with diagnosed ASD suggests continuity from infancy across childhood. Our developing understanding of the interconnected emergence of temperament and ASD symptoms across infancy and toddlerhood remains in its early days, however, and prospective high-risk studies such as that reported here afford the framework within which we may come to understand how early temperament contributes to the development of ASD as well as within the BAP.

Current Aims and Hypotheses

The current study aimed to examine the way in which infant temperament, assessed longitudinally across infancy, through the use of validated parent-report measures, might present within a sample of infants at familial high-risk for ASD. Infants who were later-born siblings of a child with an established ASD diagnosis were compared to control infant with no such family history of ASD, with the aim of observing individual differences in the first years of life which would inform the behavioural manifestation of ASD symptoms and of the BAP. From previous findings, we expected that a temperament profile of high negative affect and low positive affect and social approach (i.e., components of Surgency), along with low Effortful Control (i.e., ability to control and regulate attention, emotions and behaviour) would distinguish those high-risk infants who developed ASD from those who developed more typically. Given current conceptualisation of the BAP, we sought also to explore whether the temperament profile of high-risk infants who did not develop ASD would differ from that of control infants with no such family history.

Methods

Data for this study were made available through the British Autism Study of Infant Siblings (BASIS, www.basisnetwork.org; NHS NRES London REC 08/H0718/76). Parents gave informed consent.

Participants

Participants were 104 infants followed longitudinally from around 7-months of age to around the third birthday. Of these, 54 were deemed to be at high-risk for ASD (hereafter, HR group) on the basis of having an older sibling (hereafter, *proband*) with a community clinical diagnosis of ASD. In four cases, probands were half-siblings, while in three cases, infants had two probands, and 45 of all probands were male while nine were female. Proband diagnosis was confirmed by two expert clinicians (TC, PB) based on information from the Development and Wellbeing Assessment (DAWBA; Goodman et al. 2000) and the Social Communication Questionnaire (SCQ; Rutter et al. 2003). Most probands met criteria for ASD on both the DAWBA and SCQ ($n = 44$). While a small number scored below threshold on the SCQ ($n = 4$) no exclusions were made due to attainment of the DAWBA threshold and expert opinion. For two probands, data were only available for *either* the DAWBA or the SCQ, while for four probands, neither measure was available (although parents confirmed local clinical ASD diagnosis at intake). Parent-reported family medical histories were examined for significant medical conditions in the proband or extended families members, with no exclusions made on this basis.

The remaining 50 infants were classified as low-risk controls (hereafter, LR group). LR infants were recruited from a volunteer database at the Birkbeck Centre for Brain and Cognitive Development. These infants were full-term (with one exception), had normal birth weight, and there was no ASD within first-degree family members (as confirmed through parent interview regarding family medical history). All had at least one older-sibling (in three cases, only half-sibling/s), 28 of whom were male and 22 female. Screening for possible ASD in these older siblings was undertaken using the SCQ (Rutter et al. 2003), with no child scoring above instrument ASD cut-off; $n = 1$ missing).

Procedure and Measures

Participant infants attended up to four visits as part of their longitudinal participation in the BASIS programme of research. Initial visit scheduling was intended to occur roughly around 6-months of age, with subsequent visits around each of the first, second, and third birthdays. Mean ages of HR and LR infants were well-matched at each of the four visits: Visit 1 mean age = 7.2 months, $SD = 1.1$, $t(99) = 0.12$, $p = .902$; Visit 2 $M = 13.7$ months, $SD = 1.5$, $t(96) = 0.32$, $p = .751$; Visit 3 $M = 23.7$ months, $SD = 1.0$, $t(91) = 0.88$, $p = .379$; Visit 4 $M = 37.8$ months, $SD = 3.8$, $t(99) = 1.80$, $p = .858$.

Parent-Reported Temperament

Parents completed the IBQ-R (Gartstein and Rothbart 2003) at each of Visits 1 and 2 (hereafter, IBQ1 and IBQ2, respectively) and the ECBQ (Putnam et al. 2006) at Visit 3. The IBQ-R assesses the original IBQ items—Activity Level, Smiling and Laughter, Fear, Distress to Limitations, Duration of Orienting, Soothability, and Vocal Reactivity—as well as nine additional scales which are downward extensions of the CBQ (Rothbart et al. 2001a, b)—Positive Anticipation (Approach), Falling Reactivity, High and Low Intensity Pleasure, Perceptual Sensitivity, Sadness, Cuddliness, Social Fear, and Attentional Shifting. The eighteen scales of the ECBQ (Putnam et al. 2006), designed to target the 18-month to 3-year range, are a downward extension of the CBQ and an upward extension of the IBQ-R (see "Appendix" Table 6).The broad factors, Surgency, Negative Affectivity and Effortful Control, and their constituent scales, are outlined in "Appendix" Table 6.

Outcome Characterization

Alongside the standard measures of cognitive (Mullen Scales of Early Learning; MSEL; Mullen 1995) and adaptive development (Vineland Adaptive Behavior Scales; VABS; Sparrow et al. 2005) taken at each visit for all children, HR toddlers were assessed at each of Visits 3 and 4 using the Autism Diagnostic Observation Schedule—Generic (ADOS-G; Lord et al. 2000), a semi-structured play assessment used to evaluate autism-related social and communication behavioural characteristics. Observations obtained from this assessment were augmented, at Visit 4, with parent-report information from the Autism Diagnostic Interview—Revised (ADI-R; Lord et al. 1994).

ASD diagnosis was made for HR toddlers based on consensus ICD-10 (World Health Organisation 1993) criteria (including childhood autism, atypical autism, and other pervasive developmental disorder; PDD), by experienced researchers (TC, KH, SCh, GP) who considered all available information gained across Visits 3 and 4. Of the 53 HR toddlers retained to Visit 4, 17 (representing 31.5 % of the group; 11 boys and six girls) met criteria for an ASD diagnosis (hereafter, HR-ASD subgroup). Given the children's young age, and in line with proposed changes to the upcoming DSM-V, no attempt was made to assign specific diagnostic sub-categories. Another subgroup of HR toddlers was considered to present other developmental concerns. These 12 toddlers (hereafter HR-Atypical subgroup, representing 22.6 % of the sample; three boys and nine girls) did not meet ICD-10 criteria for an ASD. However, each individual scored above ASD threshold on *one* of the ADOS-G ($n = 10$) *or* ADI-R ($n = 1$), and/or scored $>1.5SD$ below MSEL population mean ($n = 2$). The remaining 24 HR toddlers were classified as typically developing (hereafter, HR-Typical subgroup). Table 1 characterises the 53 HR and 48 LR toddlers retained to Visit 4, with the former group separated by diagnostic outcome.

Statistical Analyses

In order to analyse group differences in early temperament, multivariate analysis of variance (MANOVA) was undertaken, including each of the three temperament factors (*Surgency*, *Negative Affect* and *Effortful Control*) in separate MANOVAs for IBQ1, IBQ2 and ECBQ. Follow-up univariate ANOVAs were conducted to ascertain where group differences lay. Follow-up MANOVAs and univariate ANOVAs were conducted to determine which temperament dimensions accounted for the differences observed across the overall factors found to be important in the initial series of MANOVAs. This approach was undertaken in order to reduce the number of overall comparisons conducted, given the high number of temperament dimensions assessed at each visit. There were two instances where the overall MANOVA for the factors approached, but did not reach, significance (IBQ1 *Surgency* and ECBQ *Effortful Control*, $ps = 0.06$). Follow-up tests were nevertheless undertaken and are reported here given that (a) the necessarily restricted sample size of the current prospective investigation meant a very conservative approach might miss detecting important effects due to low statistical power (Type II errors), and (b) these tests were theoretically relevant given the literature. All follow-up

Table 1 Sample characterisation (means and standard deviations) for low-risk toddlers and subgroups of high-risk toddlers (separated by diagnostic outcome) at final assessment around the third birthday

	LR	HR-Typical	HR-Atypical	HR-ASD
N	48	24	12	17
Boys:Girls	20:28	7:17	3:9	11:6
MSEL[1]	115.8 (16.23)^A	113.5 (13.28)^A	103.42 (9.08)	94.42 (23.31)^B
ADOS[2] SA[3]	4.9 (3.48)^A	3.67 (1.76)^A	9.75 (3.52)^B	9.47 (4.45)^B
ADOS RRB[4]	1.08 (1.5)^A	1.08 (1.28)^A	1.42 (1.24)^A	3.18 (1.94)^B
ADOS SA + RRB	5.98 (3.83)^A	4.75 (2.00)^A	11.17 (4.09)^B	12.65 (5.61)^B
ADI-R[5] social	–	1.63 (1.66)^A	3.42 (4.94)^A	9.75 (5.54)^B
ADI-R communication	–	2.21 (1.84)^A	3.58 (5.45)^A	8.44 (5.14)^B
ADI-R SBRI	–	0.50 (0.89)^A	1.08 (1.31)^A	3.63 (2.22)^B

Superscripts where the letters differ indicate significant differences ($p < .05$) across groups for the given measure. Means with no superscript do not differ significantly from any other group

[1] Mullen Scales of Early Learning (Mullen 1995) Early Learning Composite Standard Score

[2] Autism Diagnostic Observation Schedule—Generic (ADOS-G; Lord et al. 2000)

[3] ADOS-G revised algorithm score: social affect subtotal (Gotham et al. 2007)

[4] ADOS-G revised algorithm score: restricted repetitive behavior subtotal (Gotham et al. 2007)

[5] Autism Diagnostic Interview—revised (Lord et al. 1994)

univariate tests were conducted using Bonferroni correction to control for multiple comparisons.

Results

Between Group Analyses

For the temperament data reported by parents at each visit, comparisons were first undertaken across LR and HR groups. HR data were then divided on the basis of diagnostic outcome subgroup—HR-Typical, HR-Atypical, and HR-ASD—with comparisons made between these groups and the LR group. Data were missing for a small number of cases at each visit: three LR cases (1 IBQ1, 1 IBQ2, 2 ECBQ), four HR-Typical cases (1 IBQ1, 1 IBQ2, 2 ECBQ), no HR-Atypical cases, and three HR-ASD cases (all ECBQ).

Infant Temperament Around 7-Months (IBQ1)

A MANOVA to assess group differences between LR and HR groups on the three IBQ1 temperament factors revealed a main effect of group, $F(3, 97) = 3.15$, $p = .028$, $\eta^2 = 0.089$. Group membership was observed to have a significant effect on the *Surgency* factor, $F(1, 99) = 5.23$, $p = .024$, $\eta^2 = 0.050$, with the LR group demonstrating higher levels of *Surgency* than the HR group. The groups did not differ on *Negative Affect* or *Effortful Control*. Follow-up MANOVAs were conducted to determine which temperament dimensions within the overall *Surgency* factor drove these group differences. The LR group were reported to demonstrate significantly greater *High Intensity Pleasure* behaviours, $F(1, 99) = 6.37, p = .013, \eta^2 = 0.060$ and also to *Approach* more frequently, $F(1, 99) = 8.71$, $p = .004$, $\eta^2 = 0.081$, than were the HR group.

A MANOVA to assess differences for the four subgroups (LR, HR-Typical, HR-Atypical and HR-ASD) on the three temperament factors revealed a significant main effect of subgroup, $F(9, 224) = 2.54$, $p = .009$, $\eta^2 = 0.076$. Subgroup membership was seen to have a significant effect on the *Surgency* factor, $F(3, 94) = 5.04$, $p = .003$, $\eta^2 = 0.139$. The subgroups did not differ on *Negative Affect* or *Effortful Control* (*p*s > .05). Follow-up ANOVAs revealed that both LR and HR-ASD subgroups scored more highly than the HR-Typical subgroup on levels of *Surgency* ($p = .006$). Table 2 presents relevant means and standard deviations.

Follow-up MANOVAs on the temperament dimensions within the overall *Surgency* factor revealed subgroup differences for *Approach*, $F(3, 94) = 5.66$, $p = .001$, $\eta^2 = 0.153$ and *Perceptual Sensitivity* $F(3, 94) = 3.57$, $p = .017$, $\eta^2 = 0.102$, such that the LR group were reported to display higher levels of *Approach* than both the HR-Typical ($p = .013$) and HR-Atypical subgroups ($p = .015$), and the HR-ASD subgroup were reported to show higher levels of *Perceptual Sensitivity* than the HR-Typical subgroup ($p = .011$).

Infant Temperament Around 14-Months (IBQ2)

A MANOVA to assess group differences between the LR and HR groups on the three IBQ2 temperament factors

Table 2 Means (and standard deviations) for infant temperament factor and dimension scores at 7-month IBQ assessment, across low-risk and high-risk groups, and separating high-risk diagnostic subgroups

	LR group	HR group	HR diagnostic subgroups		
			Typical	Atypical	ASD
Factors					
Surgency	**4.82 (5.60)**^{A}	**4.54 (0.64)**	4.31 (0.63)^{B}	4.43 (0.55)	4.84 (0.56)^{A}
Negative affect	3.10 (0.59)	3.30 (0.75)	3.17 (0.69)	3.52 (0.34)	3.31 (0.95)
Effortful control	4.93 (0.47)	4.74 (0.56)	4.75 (0.56)	4.62 (0.74)	4.69 (0.47)
Dimensions					
High-intensity pleasure	**6.07 (0.61)**	**5.75 (0.66)**	5.67 (0.47)	5.85 (0.51)	5.84 (0.62)
Approach	**5.45 (0.81)**^{A}	**4.90 (1.00)**	4.75 (0.90)^{B}	4.58 (1.12)^{B}	5.40 (0.77)
Perceptual sensitivity	3.99 (1.44)	3.97 (1.38)	3.45 (1.26)^{A}	3.77 (1.02)	4.71 (1.46)^{B}

Bold typeface indicates significant difference between LR group and overall HR groups, while superscript letters indicate significant difference across LR and/or HR subgroups (different superscript letters indicate significant difference at $p < .05$, while means with no associated superscript indicate no difference from any other subgroup)

revealed a significant main effect of group, $F(3, 94) = 3.50$, $p = .018$, $\eta^2 = 0.101$. Group membership was again observed to have a significant effect on the *Surgency* temperament factor, $F(1, 96) = 5.23$, $p = .024$, $\eta^2 = 0.052$, and also the *Effortful Control* factor, $F(1, 96) = 8.42$, $p = .005$, $\eta^2 = 0.081$, with the LR group demonstrating higher levels of both than the HR group. Follow-up MANOVAs revealed that parents reported their LR infants to *Smile and Laugh* more than was true for their HR infants, $F(1, 96) = 7.42$, $p = .008$, $\eta^2 = 0.072$, and also to *Approach* more frequently, $F(1, 96) = 7.41$, $p = .008$, $\eta^2 = 0.072$. In regards to the *Effortful Control* factor, HR infants were rated as less *Cuddly* than their LR counterparts, $F(1, 96) = 8.44$, $p = .005$, $\eta^2 = 0.081$.

The MANOVA assessing differences across the four subgroups (LR, HR-Typical, HR-Atypical, HR-ASD) on the three IBQ2 temperament factors revealed a significant main effect of subgroup, $F(9, 219) = 3.78$, $p < .001$, $\eta^2 = 0.110$. Between-subjects effects revealed subgroup membership to have a statistically significant effect on the *Surgency* temperament factor, $F(3, 92) = 3.38$, $p = .022$, $\eta^2 = 0.099$, and also on the *Effortful Control* factor, $F(3, 92) = 6.97$, $p < .001$, $\eta^2 = 0.185$. With regards to the *Surgency* factor, follow-up univariate ANOVAs revealed LR infants to score more highly than HR-Typical infants ($p = .025$). The HR-ASD group was reported to demonstrate less *Effortful Control* than both the LR group ($p < .001$) and HR-Atypical subgroup ($p = .013$). Table 3 presents relevant means and standard deviations.

Follow-up MANOVAs revealed that, within the *Surgency factor*, subgroup differences emerged for *Smiling and Laughter*, $F(3, 92) = 5.33$, $p = .002$, $\eta^2 = 0.148$, such that the HR-ASD subgroup were reported to show lower rates of these behaviours than the LR group ($p = .001$) and for *Perceptual Sensitivity*, $F(3, 92) = 3.56$, $p = .017$, $\eta^2 = 0.104$, such that the HR-ASD subgroup was rated as more sensitive than the HR-Typical subgroup ($p = .047$). The same was also observed for *Approach*, $F(3, 92) = 3.56$, $p = .017$, $\eta^2 = 0.104$. However, when Bonferroni correction was applied to post hoc tests, none of the subgroups differed significantly from one another.

Within the *Effortful Control factor*, subgroup differences emerged for *Low Intensity Pleasure*, $F(3, 92) = 2.87$, $p = .041$, $\eta^2 = 0.086$, and *Cuddliness*, $F(3, 92) = 7.90$, $p < .001$, $\eta^2 = 0.205$. Again, however, when Bonferroni correction was applied to post hoc tests, none of the subgroups differed significantly on *Low Intensity Pleasure*. In regards to *Cuddliness*, the HR-ASD subgroup was rated as less cuddly than all other groups and subgroups (LR, $p = .000$; HR-Typical, $p = .008$; HR-Atypical, $p = .021$). These data are also presented in Table 4.

Infant Temperament Around 24-Months (ECBQ)

The MANOVA assessing differences between LR and HR groups on the three ECBQ temperament factors revealed a significant main effect of group, $F(3, 89) = 3.02$, $p = .034$, $\eta^2 = 0.092$. Between-subjects tests revealed that the LR group were reported to have higher levels of *Effortful Control* than the HR group, $F(1, 91) = 7.70$, $p = .007$, $\eta^2 = 0.078$. A follow-up MANOVA on the temperament dimensions within the *Effortful Control* factor revealed that the LR group was rated as having higher levels of *Cuddliness*, $F(1, 91) = 4.61$, $p = .035$, $\eta^2 = 0.048$, and *Inhibitory Control*, $F(1, 91) = 5.77$, $p = .018$, $\eta^2 = 0.060$, than the HR group.

The MANOVA assessing group differences on the three ECBQ temperament factors for the four groups revealed a significant main effect of group, $F(9, 211) = 2.44$, $p = .012$, $\eta^2 = 0.077$. Between-subjects tests revealed that group

Table 3 Means (and standard deviations) for infants' temperament factor and dimension scores at 14-month IBQ assessment, across low- and high-risk groups and separating high-risk diagnostic subgroups

	LR group	HR group	HR diagnostic subgroups		
			Typical	Atypical	ASD
Factors					
Surgency	**4.99 (0.40)**^{A}	**4.76 (0.52)**	4.65 (0.54)^{B}	4.72 (0.40)	4.90 (0.44)
Negative affect	3.40 (0.55)	3.60 (0.88)	3.47 (0.08)	3.57 (0.65)	3.73 (1.20)
Effortful control	**4.82 (0.45)**^{A}	**4.51 (0.53)**	4.6 (0.49)	4.79 (0.47)^{A}	4.24 (0.45)^{B}
Dimensions					
Smiling and laughter	4.74 (0.70)^{A}	4.22 (1.03)	4.33 (0.91)	4.51 (0.57)	3.83 (1.03)^{B}
Approach	**5.76 (0.59)**	**5.39 (0.74)**	5.41 (0.65)	5.30 (0.80)	5.57 (0.58)
Perceptual sensitivity	4.21 (0.95)	3.90 (1.39)	3.42 (1.09)^{A}	3.64 (0.87)	4.42 (1.64)^{B}
Low-intensity pleasure	4.81 (0.92)	4.5 (1.1)	4.53 (1.04)	5.00 (0.75)	4.17 (0.67)
Cuddliness	**5.69 (0.70)**^{A}	**5.23 (0.80)**	5.51 (0.61)^{A}	5.58 (0.52)^{A}	4.82 (0.57)^{B}

Bold typeface indicates significant difference between LR group and overall HR groups, while superscript letters indicate significant difference across LR and/or HR subgroups (different superscript letters indicate significant difference at $p < .05$, while means with no associated superscript indicate no difference from any other subgroup)

Table 4 Means (and standard deviations) for infants' temperament factor and dimension scores at 24-month ECBQ assessment, across low- and high-risk groups and separating high-risk subgroups

	LR group	HR group	HR diagnostic subgroups		
			Typical	Atypical	ASD
Factors					
Surgency	4.99 (0.60)	4.90 (0.61)	5.02 (0.44)	4.72 (0.66)	4.93 (0.76)
Negative affect	2.54 (0.37)^{A}	2.67 (0.49)	2.63 (0.47)	2.58 (0.58)	3.02 (0.79)^{B}
Effortful control	**4.80 (0.46)**^{A}	**4.52 (0.50)**	4.59 (0.48)	4.74 (0.46)	4.23 (0.36)^{B}
Dimensions					
Sadness	2.40 (0.63)^{A}	2.83 (0.83)	2.76 (0.77)	2.81 (0.86)	3.11 (1.23)^{B}
Shyness	2.82 (0.79)^{A}	3.37 (0.95)	3.12 (0.73)^{A}	2.90 (0.68)^{A}	4.13 (1.17)^{B}
Soothability	5.68 (0.45)^{A}	5.36 (0.079)	5.51 (0.43)^{A}	5.59 (0.74)^{A}	4.48 (1.40)^{B}
Low-intensity pleasure	5.08 (0.57)^{A}	4.84 (0.62)	5.09 (0.50)^{A}	4.96 (0.45)	4.53 (0.50)^{B}
Cuddliness	**5.49 (0.74)**^{A}	**5.04 (1.17)**	5.30 (0.99)	5.37 (1.10)	4.40 (1.38)^{B}
Inhibitory control	**4.03 (1.02)**	**3.54 (0.97)**	3.58 (0.75)	3.62 (1.22)	3.40 (1.10)

Bold typeface indicates significant difference between LR group and overall HR groups, while superscript letters indicate significant difference across LR and/or HR subgroups (different superscript letters indicate significant difference at $p < .05$, while means with no associated superscript indicate no difference from any other subgroup)

membership had a statistically significant effect on the *Negative Affect* temperament factor, $F(3, 89) = 3.39$, $p = .022$[4], $\eta^2 = 0.103$, and also on the *Effortful Control* factor, $F(3, 99) = 4.87, p = .004, \eta^2 = 0.141$. With regards to the *Negative Affect* factor, follow-up univariate ANOVAs revealed that the HR-ASD subgroup scored more highly than the LR group ($p = .013$). Further, the HR-ASD subgroup was reported to demonstrate less *Effortful Control* than the LR group ($p = .002$). Table 4 presents group and subgroup means and standard deviations.

Follow-up MANOVAs revealed that within the *Negative Affect* factor, group differences emerged for *Sadness*, $F(3, 89) = 3.31$, $p = .024$, $\eta^2 = 0.100$, such that the HR-ASD subgroup were reported to show higher rates of sadness than the LR group ($p = .029$). This was also true for *Shyness*, $F(3, 89) = 9.11$, $p = .000$, $\eta^2 = 0.235$, and *Soothability*, $F(3, 89) = 5.99$, $p = .001$, $\eta^2 = 0.168$. The HR-ASD subgroup was rated as more shy than all other groups and subgroups (LR, $p = .000$; HR-Typical, $p = .005$; HR-Atypical, $p = .002$); and as less soothable than the all others (LR, $p < .001$; HR-Typical, $p = .022$; HR-Atypical, $p = .025$). Within the *Effortful Control factor,* group differences emerged for *Low Intensity Pleasure*, $F(3, 89) = 4.26$, $p = .007$, $\eta^2 = 0.126$, with the HR-ASD

Table 5 Summary of key temperament differences across 7-, 14- and 24-month assessments

7-Month IBQ	14-Month IBQ	24-Month ECBQ
HR versus LR comparison		
Surgency: LR > HR	**Surgency**: LR > HR	
Approach: LR > HR	*Approach*: LR > HR	
High-intensity pleas.: LR > HR	**Effortful control**: LR > HR	**Effortful control**: LR > HR
	Cuddliness: LR > HR	*Cuddliness*: LR > HR
		Inhibitory control: LR > HR
Subgroup comparisons		
Surgency: LR and HR-ASD > HR-Typical	**Surgency**: LR > HR-Typical	
Approach: LR > HR-Typ. and HR-Atypical	*Smiling and laughter*: LR > HR-ASD	
Percept. sensitivity: HR-ASD > HR-Typical	*Percept. sensitivity*: HR-ASD > HR-Typical	
	Effortful control: LR and HR-Atypical > HR-ASD	**Effortful control**: LR > HR-ASD
	Cuddliness: LR, HR-Typ and Atyp > HR-ASD	*Cuddliness*: LR > HR-ASD
		Low intensity pleasure: LR and HR-Typ > HR-ASD
		Negative affect: HR-ASD > LR
		Sadness: HR-ASD > LR
		Shyness: HR-ASD > LR, HR-Typ and Atyp
		Soothability: LR, HR-Typ and Atyp > HR-ASD

Factors shown in bold typeface, dimensions shown in italics

subgroup scoring below the LR group ($p = .006$) and HR-Typical subgroup ($p = .018$). The same was true for the *Cuddliness* dimension, $F(3,\ 92) = 4.73$, $p = .004$, $\eta^2 = 0.137$, with the HR-ASD group rated as less cuddly than the LR group ($p = .002$), as well as the HR-Typical and HR-Atypical subgroups, although these latter group differences only approached significance ($p = .064$ and $p = .073$, respectively).

In order to compare the pattern of group differences across broad factors and individual dimensions for the IBQ1, IBQ2 and ECBQ measures taken across three visits, Table 5 summarises significant findings.

Discussion

This study aimed to examine, for the first time, early temperament across three longitudinal time points in infancy, within a group of infants at high-risk for ASD with whom formal diagnostic assessment processes were undertaken around 3 years of age. The main question was whether high-risk infants later diagnosed with ASD could be distinguished from their counterparts without ASD diagnostic outcome and from low-risk controls by particular temperament characteristics around 7, 14 and 24 months. A secondary and exploratory aim was to understand more about the broader autism phenotype, through comparison of high- and low-risk profiles and through the inclusion of a subgroup of high-risk siblings with diagnostic outcomes other than ASD (i.e., other atypicality and typical outcome).

Surgency

No specific hypotheses were made regarding the overall *Surgency* factor, given the mixed findings in the literature on older children with ASD and given the multiple dimensions contributing to this factor (which also differ across the ages sampled). Infants at high-risk for ASD were reported by their parents to show lower rates of *Surgency* from as early as the second half of the first year of life, than was true for low-risk infants, yet those who went onto receive a diagnosis of ASD were initially reported to have *higher* levels of *Surgency* than their counterparts considered to be typically-developing at outcome. However, by the beginning of the second year of life, only the low-risk group was rated more highly on *Surgency* than the high-risk infants developing toward typical outcome.

When assessing the dimensions contributing to *Surgency*, infants later diagnosed with ASD were rated by their parents as being more perceptually sensitive to environmental stimuli than were those at similar high-risk but who went on to typical outcome. This was true as early as the first year of life, but also maintained during the second year. This is most likely to explain the higher levels of *Surgency* factor seen in the subgroup of infants developing ASD, overall, and inconsistent with some findings in the literature on Surgency in older children with ASD (Landry 1998; Schwartz et al. 2009). However, there is variability regarding what Surgency comprises across studies. Schwartz et al. for example, find that fear and approach motivation within their Surgency factor drove the observed group differences. Thus, perhaps a dimension (or even item level) comparison is more appropriate. In this respect, the finding that infants later diagnosed with ASD were more perceptually sensitive is in keeping with Landry's (1998) finding for 6- to 7-year-olds, although this feature forms part of the negative affect factor in their study. Children with ASD are known to be more sensitive to sensations such as textures, sounds and smells (Ornitz et al. 1977; Volkmar et al. 1986). In Zwaigenbaum et al.'s (2005) study, it was found that infants as young as 12 months who were later classified as having ASD displayed intense responses to sensory input. The current findings extend what is currently known to suggest that even infants aged around 7 months who go onto develop ASD display more sensory sensitivity than do high-risk counterparts who go on to typically-developing outcome. Additionally, using an experimental methodology, Guiraud et al. (2011) have found reduced sensory habituation to auditory stimuli at 9-months in high-risk infants compared to low-risk controls, although this cohort has yet to be followed-up to establish diagnostic outcome.

Also within the *Surgency* factor, at around 7 months, low-risk controls were rated as engaging in higher levels of approach behaviour (i.e., toward pleasurable activities such as new toys) than were high-risk infants, and also compared specifically to those who went on to typically-developing or other atypical (but not ASD) outcome. At around 14 months, while low risk infants were seen to approach more often than high-risk infants, this was not the case when high-risk infants were separated by outcome subgroup. Thus, overall, infants later diagnosed with ASD were not reported to have lower levels of approach and excitement for toys, objects and activities. It is interesting that high-risk infants without ASD outcomes (who may represent the BAP), but not those with ASD outcome, had lower levels of approach behaviour compared to low-risk controls, and it is likely that this is contributing to the lower rates of *Surgency* in the high-risk infants with typical outcome. It is possible that children with the BAP do indeed show less approach and excitement for pleasant objects and toys, either because they are less rewarded by these stimuli, or because their parents are rating their temperament in comparison to an older sibling with ASD who may have had higher levels of approach to these situations. Why the ASD outcome subgroup had higher levels of approach that was comparable to that of low-risk controls is unclear, but it is possible that this could be explained by the notion that children with ASD tend to find non-social stimuli more rewarding than social stimuli (Dawson et al. 1998; Leekam et al. 2000) and are thus generally more object focussed—a characteristic that may not be as salient within the BAP. However, this explanation must be interpreted with caution, especially given that this finding is in direct contrast to that of Garon et al. (2009) who found *higher* levels of approach in high-risk infants without ASD outcome, than in low-risk controls.

High-risk infants were reported to smile and laugh less frequently than low-risk infants and, as hypothesised, parents rating their infants who were later diagnosed with ASD reported less smiling and laughing in comparison to the ratings made by parents of low-risk controls. This finding is consistent with many retrospective studies on the early presentation of infants later diagnosed with ASD (e.g., Clifford and Dissanayake 2008; Werner et al. 2000), and with the two prospective studies of infants at high-risk for ASD which reported positive affect from 12 months in infants later classified with ASD (i.e., Garon et al. 2009; Zwaigenbaum et al. 2005). It is also consistent with studies of older children with ASD (aged 6–7 years; i.e., Landry 1998).

Effortful Control

As expected, all infants at high-risk for ASD, and specifically those who went onto develop ASD, were rated as demonstrating lower levels of *Effortful Control* than were low-risk controls, throughout the second year of life. This finding is consistent with those of previous studies on children with ASD (Janes 2001; Konstantareas and Stewart 2006; Landry 1998), and with studies on traits of effortful control seen in infancy (Garon et al. 2009; Zwaigenbaum et al. 2005). In particular, Zwaigenbaum et al. found that an inability to disengage visual attention from particular objects at 12-months distinguished high-risk infants later classified with ASD from low-risk controls, and Garon et al. found that infants diagnosed with ASD at 36-months could be differentiated from other high- and low-risk infants in terms of difficulty controlling attention and behaviour at 24-months. Thus, the current findings extend from these recent studies to suggest that lower rates of the factor of *Effortful Control,* early in the second year of life, differentiate children later diagnosed with ASD from other high- and low-risk infants. The implications of these findings are important given that difficulties in regulation have been found to leave children at risk for the development of anxiety (Rothbart and Bates 2006) and problem behaviour in ASD (Adamek et al. 2011).

However, unlike Zwaigenbaum et al. (2005) and Garon et al. (2009), it was not the items of attention shifting and duration of orientation that differentiated the current sample of infants developing ASD from the other subgroups. Rather, it was the *Cuddliness* dimension which drove the overall difference in *Effortful Control*. Infants at high-risk for ASD were rated as less cuddly than low-risk controls and, as predicted, those individuals who went onto develop ASD were rated as less cuddly than were other infants at high- and low-risk, throughout the second year of life. These results fit the well-known parental reports of infants who are later diagnosed with ASD (e.g., Vostanis et al. 1998). Relatedly, infants who went on to ASD were reported to engage in less low intensity pleasure at around 24 months, which consists of many quiet social activities such as being sung to, being rocked, and listening to a story.

Negative Affect

In terms of the *Negative Affect* factor, while no differences emerged across high- and low-risk groups, at the finer level of analyses, those infants later diagnosed with ASD were reported to have higher levels of overall negative affect than all other subgroups of infants, at 24-months. Similar results were reported by Garon et al. (2009), who found that infants later diagnosed with ASD displayed higher negative affect than other high-risk infants and compared to low-risk controls. Zwaigenbaum et al. (2005) found that distress reactions were common in high-risk infants later classified as having ASD and Bryson et al. (2007) found that irritability and a proneness to distress/negative affect characterised later-diagnosed infants. Considering the dimensions comprising the *Negative Affect* factor, infants developing ASD were rated by their parents as displaying greater levels of sadness and shyness, and as being more difficult to soothe. This finding is in keeping with the two patterns commonly reported by parents of infants with ASD—either that they are 'easy' or 'perfect', or that they are very difficult to soothe and settle (Wing 1980). Children with ASD have also been rated by their parents as being less soothable and showing more discomfort (Konstantareas and Stewart 2006). What is interesting here is that these findings were specific to the subgroup of infants developing toward ASD diagnostic outcome. That is, there was a propensity for greater *Negative Affect* in general in those children at high-risk for ASD who went on to develop the ASD.

In terms of the BAP, it was evident that approach behaviours were less likely in the high-risk infants who did not go on to ASD, contributing to lower rates of *Surgency*. Observation of the subgroup means suggested that there did not seem to be a 'spectrum' profile to most temperament traits (i.e., whereby characteristics increased in strength from the HR-Typical group, to the HR-Atypical group and further still to the HR-ASD group). This was with the exception of *Negative Affect* from the beginning of the second year of life, and for *Sadness* at 24-months of age.

Limitations and Future Directions

The current findings should be interpreted in light of some limitations. First, this study was unable to improve upon previous research designs by using a multi-method, multi-modal assessment of temperament. Relying on the parent as the sole informant might also result in potential bias, and the effect to which having an older child with an ASD might alter parental reporting behaviours and younger sibling temperament traits remains unknown. This latter point would be an interesting avenue for future research across high-risk sibling studies as a whole, and the addition of an observational measure of temperament in infancy would be beneficial in allowing comparisons between parent-reported and directly-observed temperament. On a related point, infants who have an older sibling with ASD and who themselves go onto develop ASD may not be representative of all children with ASD, and it is not known to what extent parental expectations and concerns influence the expression of ASD, particularly in relation to temperament in infancy. It may be that parents interact with their later-born infant differently as a result of already having a child with ASD, which may well affect the temperament of that later-born.

Due to the small sample size and the large number of analyses that would be required, we were not able to conduct a finer grained analysis of the individual items within the temperament dimensions. It would be informative to ascertain whether the combinations of items within the dimensions might be masking some findings in relation to early developing toward ASD. For example, questions around excitement over a toy or object are combined with questions around excitement over relatives visiting (i.e., social motivations) for the approach dimension, which may explain why the infants developing ASD did not differ from the other high-risk infants who approached less often than low-risk controls. It would be interesting for future research to assess which items within the dimensions were distinguishing the groups.

Finally, we did not include children at risk of other developmental disabilities, thus we cannot comment on those dimensions of temperament which may be specific to ASD. Research indicates that, for other disorders, characteristic temperament profiles can be observed; e.g., lower reported rates of *Effortful Control* and higher rates of *Negative Affect*, but similar levels of *Surgency* in Attention Deficit/Hyperactivity Disorder. These have been found in children aged 6–14 years (De Pauw and Mervielde 2011), and shows the same profile as that seen for infants developing ASD at 24-months of age. Future work is needed to understand which of the above-mentioned temperament traits and profiles are specific to ASD in infancy.

Conclusions

This study offers the first assessment of temperament traits across the 7- to 24-month period in a sample of children at familial high-risk for ASD, who were followed through and formally diagnosed with ASD around 3-years, and with comparison afforded to other high-risk infants who did not receive such a diagnosis, and also to low-risk controls. The findings indicate that, in the first year of life, higher levels of *Surgency* tend to differentiate those who later receive a diagnosis of ASD from other high-risk infants. In the second year of life, lower levels of *Effortful Control* tend to characterise these children, while by around the second birthday, both lower *Effortful Control* and higher *Negative Affect* appear to characterise a later diagnosis of ASD. The analysis of group differences at a domain level also revealed an important overall pattern: by the second half of the first year of life and continuing into the second year of life, high-risk children later diagnosed with ASD showed increased perceptual sensitivity. Throughout the second year, they showed lower rates of smiling, laughing, and cuddling, and around the second birthday, higher rates of negative affect including sadness, shyness and low soothability, were evident. This pattern suggests that the co-occurring behavioural traits evident in the first year for infants who go on to have ASD are 'lower level' features, such as increased perceptual sensitivity, but that those emerging in the second year of life are 'higher level' and more inherently social characteristics, such as reduced cuddliness and affective behaviours (e.g. reduced smiling and soothability).

It is interesting that, by 24 months, the primary group differences were those that distinguished the high-risk infants who went on to an ASD outcome from both the low-risk controls and other high-risk outcome subgroups (with other atypicality and typically-developed outcomes). This could mean that, by 24-months, temperament is an important construct that has clearly emerged, is noted by parents, and is playing a part in the expression of ASD. This may fit with the 'modifier' model of temperament (Mundy et al. 2007), in which it is suggested that non-syndrome-specific modifiers, such as temperament, may interact with etiological processes, to influence the outcome of children with ASD.

These early patterns of temperament that distinguish ASD infants from non-ASD infants have implications for earlier identification within the community, and could possibly be added into current checklists employed at maternal and child health centres. Furthermore, Rothbart et al. (1995) proposed that temperament characteristics can alter response to interventions, and it is suggested here that knowledge of temperament traits and profiles may aid in realising the goal of selecting individualised interventions for young children with ASD. For example, children known to have lower levels of effortful control (e.g., difficulty focusing, regulating and shifting attention, and poor inhibitory control) might benefit more from an environment structured to suit these needs. Overall, it is hoped that further understanding of the factors that modify the expression of ASD and response to intervention, such as temperament, will lead to more positive outcomes for those affected by ASD.

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Conflict of interest None.

Appendix

See Table 6.

Table 6 Temperament Scales, factor loadings (positive and negative) and definitions

CBQ Factors and scales	IBQ-R	ECBQ	Definition
Surgency/Extrversion (SE)			
Activity level	+	+	Level (rate and intensity) of gross motor activity including rate and extent of locomotion
High-intensity pleasure	+	+	Pleasure derived from activities involving high intensity, rate, complexity, novelty, and incongruity

Table 6 continued

CBQ Factors and scales	IBQ-R	ECBQ	Definition
Impulsivity		+	Speed of response initiation
Approach/+anticipation	+	+	Positive excitement and anticipation for and rapid approach toward expected pleasurable activities
Sociability		+	Seeking and taking pleasure in interactions with others
Perceptual sensitivity	+		Detection or perceptual awareness of slight, low-intensity stimulation from the external environment
Smiling and laughter	+		Smiling or laughing during general caretaking/play
Vocal reactivity	+		Amount of vocalisation exhibited in daily activities
Negative affectivity (NA)			
Frustration/anger		+	Negative affect related to interruption of tasks/goal blocking
Fear	+	+	Negative affect related to anticipation of distress, threats, pain
Discomfort		+	Negative affect related to sensory qualities of stimulation: intensity, rate, or complexity of light, movement, sound, texture
Sadness	+	+	Negative affect (e.g., tearfulness) and lowered mood and energy related to suffering, disappointment, and object loss
Soothability		−	Rate of recovery from peak distress, excitement, general arousal
Falling reactivity	−		Rate of recovery from peak distress/excitement/general arousal; ease of falling asleep
Motor Activation		+	Repetitive, small motor movements; fidgeting
Perceptual sensitivity		+	Detection or perceptual awareness of slight, low-intensity stimulation from the external environment
Shyness		+	Slow or inhibited approach and/or discomfort in social situations involving novelty and uncertainty
Distress to limitations	+		Fussing, crying, showing distress when confined, in caretaking activities, or when unable to perform desired action
Effortful Control (EC) Orienting/Regulation			
Attentional focus/duration of orienting[3]	+	+	The capacity to focus attention as well as to shift attention when desired; attention for extended periods of time
Inhibitory control		+	The capacity to plan future action and to suppress inappropriate responses, moderate and refrain from a behaviour
Low-intensity pleasure	+	+	Pleasure derived from activities or stimuli involving low intensity, rate, complexity, novelty, and incongruity
Soothability	+		Rate of recovery from peak distress, excitement, general arousal
Cuddliness	+	+	Desire for warmth and closeness with others/enjoyment in and moulding of the body to caregiver
Attention shifting		+	Ease/ability of transferring focus from one activity/task to other

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