Early and persistent motor difficulties in infants at-risk of developing autism spectrum disorder: A prospective study

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Early and persistent motor difficulties in infants at-risk of developing autism spectrum disorder: A prospective study

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Analyses were conducted in order to investigate motor development in younger siblings of children diagnosed with autism spectrum disorder (ASD). Infants at familial risk and low risk of developing ASD were tested longitudinally between the ages of 7 and 36 months. Data were analysed from motor scales on the Mullen Scales of Early Learning and the Vineland Adaptive Behaviour Scales at each age point. Significantly lower motor scores in at-risk infants were evident from the age of 7 months compared to the low-risk group. Infants who were later diagnosed with ASD demonstrated significantly poorer Fine Motor skills at 36 months than at-risk infants without any developmental difficulties. In addition, Gross Motor scores were highly correlated across the two measures for low-risk infants and infants who later developed ASD. Early motor difficulties may be an early indicator of a number of neurodevelopmental disorders, including ASD.

Keywords: Motor development; Autism spectrum disorders; Infancy.

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Autism spectrum disorders (ASD) are a group of pervasive neurodevelopmental disorders that affect around 1% of the UK population (Baird et al., 2006), and are diagnosed on the basis of a triad of impairments, including the delayed or atypical development of social interaction and communication and markedly restricted activities and interests (American Psychiatric Association, 2000). Although the diagnostic criteria require symptoms to be present before the age of three, and despite parents often reporting the recognition of symptoms in infants younger than 18 months (Chawarska et al., 2007), diagnosis before a child is 2 years old is rare (Charman & Baird, 2002).

In recent years, however, a better understanding of the heritability and genetic underpinnings of ASD (Abrahams & Geschwind, 2008; Autism Genome Project, 2007; Hallmayer et al., 2011) has led to a focus on the family members of individuals already diagnosed with ASD, who may show a number of subclinical characteristics of ASD (termed the “Broader Autism Phenotype”; Bolton et al., 1994; Pickles et al., 2000). In particular, a number of prospective studies with infants who have an older sibling with a diagnosis of ASD, and who are therefore more likely to develop ASD themselves, are currently being conducted. The most recent estimates of the recurrence rate in siblings is between 10–20% (Constantino, Zhang, Frazier, Abbacchi, & Law, 2010; Ozonoff et al., 2011), highlighting the importance of investigating early markers in this group to allow earlier identification and intervention for those most at risk. Those younger siblings who do not go on to develop ASD may also be at an increased risk of other difficulties, such as language delay, or may have subclinical characteristics of ASD (see Elsabbagh & Johnson, 2007, 2010; Rogers, 2009, for recent reviews).

These ongoing studies are finding subtle differences between at-risk and low-risk infants on a range of behavioural and neuroimaging methods early in childhood. While atypicalities or impairments have been found in the core diagnostic areas of social communication and language (Landa, Holman, & Garrett-Mayer, 2007; Yirmiya, Gamliel, Shaked, & Sigman, 2007; Zwaigenbaum et al., 2005) and repetitive behaviours (Iverson & Wozniak, 2007), other areas of cognition and behaviour, such as visual attention (Elsabbagh et al., 2009; Zwaigenbaum et al., 2005), sensory-related behaviours (Zwaigenbaum et al., 2005) and motor development (Iverson & Wozniak, 2007; Landa & Garrett-Mayer, 2006; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007), which have previously been considered “secondary symptoms” (Rogers, 2009, p. 133), have also been highlighted as possible key features of early development in ASD. The current report will focus on the last of these possible risk markers, considering differences between at-risk and low-risk infants in developing motor skills between the ages of 6 and 36 months.

Studies of motor skills in school-age children with ASD have repeatedly reported motor dysfunction in their participants, including difficulties with manual dexterity, ball skills and balance (Green et al., 2009; Manjiviona & Prior,
Research with younger children already diagnosed with ASD (Provost, Lopez, & Heimerl, 2007), retrospective studies of motor behaviour in infancy through video analysis (Baranek, 1999; Gernsbacher, Sauer, Gey, Schweigert, & Goldsmith, 2008; Ozonoff et al., 2008; Teitelbaum, Teitelbaum, Nye, Fryman, & Maurer, 1998) and prospective studies of at-risk infants (Iverson & Wozniak, 2007; Landa & Garrett-Mayer, 2006; Toth et al., 2007; see Bhat, Landa, & Galloway, 2011, for a review), have documented further atypicalities, ranging from subtle discrepancies in early motor skills to more severe difficulties. Although there are not always significant differences in the mean age of achieving key motor milestones, such as independent sitting, crawling and walking (Iverson & Wozniak, 2007; Ozonoff et al., 2008), the age range during which infants began walking, for example, was both wider and later for at-risk infants (10–18 months) compared to low-risk infants (9–14 months), and a higher proportion of children in the at-risk group were delayed in gross motor skills (Iverson & Wozniak, 2007). Understanding these early motor symptoms is not only useful in improving later motor functioning, but may also prevent knock-on effects on other domains, including those associated with the core deficits in ASD (Iverson, 2010; Rogers, 2009). Indeed, there is increasing evidence for a link between motor development and the development of social interaction skills. For example, improved object manipulation skills and reaching can change the patterns of attention to others in the environment at the age of 3 months (Libertus & Needham, 2010, 2011). Similarly, the onset of crawling and walking produces more opportunities for joint attention and social referencing by changing the type of interactions between infants and caregivers (e.g., Campos et al., 2000; Tamis-LeMonda et al., 2008). Early identification of motor symptoms in infancy could therefore have important implications for intervention and outcomes in individuals with ASD in later life.

The purpose of the current report was to compare motor development in a prospective at-risk sample with typically developing controls on broad motor measures, assessed by the Gross and Fine Motor scales on the Mullen Scales of Early Learning (MSEL; Mullen, 1995) and the Vineland Adaptive Behaviour Scales-II (VABS-II; Sparrow, Cicchetti, & Balla, 2005). These measures are used widely in prospective samples of at-risk infants that have not been designed with motor development in mind, and can provide useful and reliable information on gross and fine motor skills across this age range. However, while previous studies have reported data from the MSEL (Landa & Garret-Mayer, 2006) or on both the MSEL and VABS-II (Toth et al., 2007) in these prospective samples, this is the first prospective study to measure the correlation between standardized and parental report measures of motor development in infants at risk of developing ASD compared to low-risk infants as early as 7 months, providing vital insights into the importance of the two types of assessment in effectively highlighting early motor delay. Following at-risk and low-risk samples longitudinally from the age of 7 months also allows investigation of differences between groups at
any of the four age points, which is important as different skills typically develop within each of the age bands, e.g., gross motor skills such as independent sitting and standing will typically develop up to the age of 18 months, while fine motor skills important for activities such as handwriting and tying shoelaces will continue to develop much later in childhood. The earlier development of fine motor skills is generally less well understood than the achievement of gross motor milestones. However, Gernsbacher et al. (2008) suggested that early development of fine motor skills was related to later speech fluency in individuals with ASD, through retrospective reports of motor development. The current prospective design will allow a closer investigation of the development of fine motor skills in those infants who do and do not go on to develop ASD.

Based on previous research with prospective samples (Landa & Garrett-Mayer, 2006; Toth et al., 2007), it was predicted that infants in the at-risk group would have significantly poorer motor skills than those in the low-risk group, and that any differences on the MSEL would be present after the age of 12 months (Landa & Garret-Mayer, 2006). Lower motor scores on the VABS-II in the at-risk group should be evident before the age of 24 months (Toth et al., 2007), although as this is the first study to compare groups on the VABS at earlier ages, it is not clear how early these differences may emerge. Finally, early motor development was considered in relation to later diagnosis by splitting the at-risk sample into three subgroups based on outcomes (diagnosed ASD, typically developing, other developmental concerns). If motor difficulties are a core feature of ASD, it would be predicted that infants in the at-risk group who go on to be diagnosed with ASD at 36 months would have poorer motor skills than the other subgroups earlier in infancy. However, if motor difficulties are a more general risk factor or marker for atypical development, then the infants who later develop ASD should not differ from the other at-risk siblings in their motor skills.

**METHOD**

**Participants**

Participants were families taking part in an ongoing longitudinal research programme: The British Autism Study of Infant Siblings (BASIS; www.basisnetwork.org), a UK collaborative network facilitating research with infants at risk for autism. Ethical approval was given by the NHS NRES London REC 08/H0718/76. Fifty-four at-risk infants (with an older sibling with a confirmed clinical diagnosis of ASD) and 50 low-risk infants (with an older sibling without a diagnosis of ASD or related conditions, and no family history of ASD) were recruited through a database of volunteers. This cohort has been reported on extensively in other publications (see, e.g., Elsabbagh et al., in press; Elsabbagh et al., 2012). Infants were assessed at 6–10 (“7 months”), 12–15 (“14 months”), 23–25 (“24 months”) and 35–37 months (“36 months”) and were matched for
TABLE 1
Participant characteristics for at-risk and low-risk infants at the four visits

<table>
<thead>
<tr>
<th>Visit</th>
<th>Group</th>
<th>N</th>
<th>Mean (SD)</th>
<th>N males</th>
</tr>
</thead>
<tbody>
<tr>
<td>7 months</td>
<td>Low-risk</td>
<td>50</td>
<td>7.4 (1.2)</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>At-risk</td>
<td>54</td>
<td>7.3 (1.2)</td>
<td>22</td>
</tr>
<tr>
<td>14 months</td>
<td>Low-risk</td>
<td>48</td>
<td>13.9 (1.3)</td>
<td>19</td>
</tr>
<tr>
<td></td>
<td>At-risk</td>
<td>54</td>
<td>13.7 (1.6)</td>
<td>22</td>
</tr>
<tr>
<td>24 months</td>
<td>Low-risk</td>
<td>49</td>
<td>23.9 (0.7)</td>
<td>19</td>
</tr>
<tr>
<td></td>
<td>At-risk</td>
<td>52</td>
<td>23.9 (1.1)</td>
<td>21</td>
</tr>
<tr>
<td>36 months</td>
<td>Low-risk</td>
<td>48</td>
<td>38.2 (3.0)</td>
<td>20</td>
</tr>
<tr>
<td></td>
<td>At-risk</td>
<td>53</td>
<td>37.7 (3.0)</td>
<td>21</td>
</tr>
</tbody>
</table>

gender and chronological age (see Table 1 for participant information). At the
time of enrolment, none of the infants had been diagnosed with any medical or
developmental condition. Infants at-risk all had an older sibling (“proband”) with
a community clinical diagnosis of ASD (four=half-sibling). Of the probands,
45 were male, 9 were female, and their ages ranged from 1 year 10 months to
14 years 3 months at the younger sibling’s birth ($M_{\text{age}} = 6 \text{ years 3 months}$,
$SD = 2 \text{ years, 9 months}$). Proband diagnosis was confirmed by two expert
clinicians using the Development and Wellbeing Assessment (DAWBA; Goodman,
Ford, Richards, Gatward, & Meltzer, 2000) and the parent-report Social
Communication Questionnaire (SCQ-Lifetime; Rutter, Bailey, & Lord, 2003)
one probands were within the age range required for these questionnaires
to be valid. 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. Inclusion criteria for low-risk infants required
no ASD diagnosis within first-degree family members. All low-risk infants had at
least one older-sibling (three = half-siblings), none of whom scored above the
ASD cut-off on the SCQ (one score was missing).

Participants from both low-risk and at-risk groups were assessed at 36 months
using the Autism Diagnostic Observation Schedule – Generic (ADOS-G; Lord,
Rutter, DiLavore, & Risi, 2000), with parents in the at-risk group completing the
Autism Diagnostic Interview – Revised (ADI-R; Lord, Rutter, & LeCouteur,
1994). For this group, consensus ICD-10 (World Health Organization, 1993)
ASD diagnoses were achieved using all available information from all visits by
experienced researchers. Toddlers from the at-risk group were considered
typically developing (TD-sibs) at 36 months if they: (1) did not meet ICD-
10 criteria for an ASD; (2) did not score above the cut-off on the ADOS-G or
ADI-R; (3) scored within 1.5 $SD$s of the population mean on the MSEL Early
Learning Composite (ELC) standard score ($> 77.5$), Receptive Language (RL)
and Expressive Language (EL) subscale $T$ scores ($> 35$). Finally, toddlers from
the at-risk group were considered to have other developmental concerns (AT-sibs) if they did not fall into either of the above groups, i.e., they either scored above theADOS-G or ADI-R cut-off or scored < 1.5 SDs below average on the MSEL ELC or RL and EL subscales. From the 53 at-risk infants in the current study seen for diagnostic assessment at 36 months, 17 (11 boys) met criteria for an ASD diagnosis (32.1%), 24 (7 boys) were in the TD-sibs group (45.3%) and 12 (3 boys) were in the AT-sibs group (22.6%). The number of infants receiving an ASD diagnosis in the current study is therefore higher than that reported in the large consortium paper recently published by Ozonoff et al. (2011) of 18.7%, but is similar to other studies with moderate-sized samples (e.g., 28 %, Landa et al., 2007; 29 %, Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011).

Materials

**Mullen Scales of Early Learning (MSEL; Mullen, 1995).** This is a standardized test of early cognitive and motor development between the ages of 0 and 68 months, consisting of measures of receptive and expressive language, visual reception and gross and fine motor skills, and it was conducted at all four visits. The motor domain of the MSEL is made up of the Gross Motor subdomain, including items such as the ability to roll over or walk, and the Fine Motor subdomain, assessing abilities such as grasping small objects. Items are scored as “present” or “absent”. The Visual Reception scale measures visual perceptual ability using items such as visual tracking of different stimuli and the identification of an object. The close connection of many of the items to general stages of cognitive development make this useful for assessing the role of any general developmental delay on the infant’s motor abilities. In the current analysis we used the Visual Reception scale from the MSEL as a proxy for general developmental differences. Raw scores are transformed into T-scale scores, mean = 50 (SD = 10). Internal consistency reliability for the five MSEL scales ranged from median values of .75 to .83, with test–retest reliability for the Gross Motor scale .96 and for the other four scales ranging from .82 to .85 (Mullen, 1995). Inter-scorer reliability ranged from .91 to .99. To assess validity, the MSEL scores were compared with other measures of development, including the Bayley Scales of Infant Development (BSID; Bayley, 1969). The correlation between the MSEL Gross Motor scale and the BSID Psychomotor Development Index was .76. The Early Learning Composite also correlated with the BSID Mental Development Index (.70; see Mullen, 1995, for further details).

**Vineland Adaptive Behaviour Scales – II (VABS-II; Sparrow et al., 2005).** This was completed for infants at all four visits. This instrument measures communication, daily living, socialization, motor skills and maladaptive behaviour. Only the Gross and Fine Motor scales are reported here and contain
similar types of items to those making up the MSEL. Parents/caregivers reported whether they had seen a particular behaviour on a scale of “Never”, “Sometimes” or “Usually”, with “Don’t Know” or “No opportunity” responses also possible. Raw scores were transformed into v-scale scores ($M = 15, SD = 3$). The VABS-II v-scale and MSEL T-scale scores will hereafter be referred to as “standardized scores”. Internal consistency reliability for the eleven VABS-II scales ranged from .72 to .96 between birth and 3 years, with Gross Motor coefficients between .81 and .92, and Fine Motor between .84 and .87 in this time period (Sparrow et al., 2005). Test–retest reliability for the Gross and Fine Motor scales were .81 and .83, respectively, at 0–2 years, and .80 and .89 at 3–6 years. Inter-scorer reliability was .72 (Gross Motor) and .82 (Fine Motor) for ages 0–6 years. Motor scores on the VABS-II correlated with scores on the first edition of the VABS (Sparrow, Balla, & Cicchetti, 1984), with correlations ranging from .91 at 0–2 years to .86 from 3–6 years (see Sparrow et al., 2005, for further details).

Procedure

The standardized assessment was conducted during testing visits consisting of a range of tasks. The administration of the entire testing protocol was flexible and child-led, since not every child was able to complete all tasks at all age points due to fatigue or timing issues (although missing data due to this on the MSEL and VABS-II were minimal). In terms of the VABS-II, this questionnaire was usually completed at home by the parents prior to the 7- and 14-month testing visits. Any missing items on the questionnaire were checked at the beginning of the sessions, before testing had commenced. At 24 and 36 months, the interview version of the questionnaire was conducted by a researcher during the visit. These versions differ only in their administration, and were conducted as described due to the change in time constraints in the testing sessions and the questionnaire load on the parents in the larger study from which the current data are drawn.

RESULTS

The main focus of the analyses was the motor scores from the MSEL and VABS-II. In order to take account of the relatively wide age range for each visit, inferential analyses were conducted on the standardized scores for each visit (7, 14, 24, 36 months). Standardized scores are presented in Table 2, including clinically relevant information provided by the Early Learning Composite (ELC). This is a standardized score calculated from the visual reception, fine motor, receptive and expressive language subscales ($M = 100, SD = 15$). However, while the ELC is often used as a measure of general developmental level, the inclusion of fine motor scores in its calculation introduces a confound in the comparisons reported in the current study. (Likewise, using a “nonverbal ratio IQ” score, e.g., Lloyd, MacDonald, & Lord, 2011, is not appropriate for the
TABLE 2
Mean (SD) of MSEL T-scores and VABS-II v-scores across age and groups, including standard scores for MSEL Visual Reception and Early Learning Composite

<table>
<thead>
<tr>
<th>Subscales</th>
<th>Low-risk</th>
<th>Combined</th>
<th>ASD-sibs</th>
<th>AT-sibs</th>
<th>TD-sibs</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>7 months</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>VABS GM</td>
<td>14.60 (2.62)</td>
<td>12.66 (3.03)</td>
<td>12.82 (3.47)</td>
<td>11.67 (1.56)</td>
<td>13.04 (3.31)</td>
</tr>
<tr>
<td>VABS FM</td>
<td>15.53 (2.56)</td>
<td>13.77 (2.69)</td>
<td>13.53 (2.13)</td>
<td>13.08 (1.93)</td>
<td>14.26 (3.36)</td>
</tr>
<tr>
<td>MSEL GM</td>
<td>50.17 (8.98)</td>
<td>45.40 (9.99)</td>
<td>46.06 (12.58)</td>
<td>45.67 (10.08)</td>
<td>45.17 (8.39)</td>
</tr>
<tr>
<td>MSEL FM</td>
<td>57.79 (9.49)</td>
<td>52.45 (10.47)</td>
<td>49.81 (11.08)</td>
<td>53.92 (8.76)</td>
<td>53.54 (11.11)</td>
</tr>
<tr>
<td>MSEL VR</td>
<td>54.73 (8.63)</td>
<td>50.53 (8.96)</td>
<td>50.00 (11.49)</td>
<td>48.58 (6.90)</td>
<td>51.71 (8.28)</td>
</tr>
<tr>
<td>MSEL ELC</td>
<td>104.79 (11.39)</td>
<td>94.00 (12.88)</td>
<td>92.13 (17.30)</td>
<td>92.83 (8.13)</td>
<td>96.13 (11.77)</td>
</tr>
<tr>
<td><strong>14 months</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>VABS GM</td>
<td>15.26 (2.63)</td>
<td>13.98 (2.58)</td>
<td>13.88 (2.91)</td>
<td>13.50 (3.00)</td>
<td>14.33 (2.15)</td>
</tr>
<tr>
<td>VABS FM</td>
<td>17.27 (2.18)</td>
<td>15.24 (2.54)</td>
<td>15.71 (3.06)</td>
<td>14.58 (2.64)</td>
<td>15.24 (2.10)</td>
</tr>
<tr>
<td>MSEL GM</td>
<td>51.04 (16.25)</td>
<td>46.26 (16.55)</td>
<td>45.53 (18.44)</td>
<td>43.92 (16.36)</td>
<td>48.83 (15.53)</td>
</tr>
<tr>
<td>MSEL FM</td>
<td>61.28 (9.23)</td>
<td>54.94 (12.44)</td>
<td>51.18 (12.91)</td>
<td>58.25 (10.64)</td>
<td>56.74 (12.45)</td>
</tr>
<tr>
<td>MSEL VR</td>
<td>55.85 (9.44)</td>
<td>51.19 (10.55)</td>
<td>47.71 (10.11)</td>
<td>55.17 (5.51)</td>
<td>52.35 (11.98)</td>
</tr>
<tr>
<td>MSEL ELC</td>
<td>106.41 (15.78)</td>
<td>97.40 (17.91)</td>
<td>89.18 (18.30)</td>
<td>99.75 (11.35)</td>
<td>103.35 (18.12)</td>
</tr>
<tr>
<td><strong>24 months</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>VABS GM</td>
<td>15.40 (1.65)</td>
<td>14.71 (1.85)</td>
<td>15.13 (1.89)</td>
<td>14.25 (1.66)</td>
<td>14.67 (1.93)</td>
</tr>
<tr>
<td>VABS FM</td>
<td>16.32 (2.38)</td>
<td>16.12 (2.10)</td>
<td>16.13 (2.31)</td>
<td>16.08 (2.19)</td>
<td>16.13 (2.01)</td>
</tr>
<tr>
<td>MSEL GM*</td>
<td>59.89 (8.08)</td>
<td>45.19 (11.24)</td>
<td>44.00 (13.21)</td>
<td>48.25 (8.17)</td>
<td>44.59 (11.35)</td>
</tr>
<tr>
<td>MSEL FM</td>
<td>54.33 (8.75)</td>
<td>49.94 (9.37)</td>
<td>47.13 (10.91)</td>
<td>51.17 (8.85)</td>
<td>51.21 (8.46)</td>
</tr>
<tr>
<td>MSEL VR</td>
<td>58.93 (12.42)</td>
<td>53.17 (12.92)</td>
<td>51.63 (14.35)</td>
<td>49.17 (14.99)</td>
<td>56.21 (10.48)</td>
</tr>
<tr>
<td>MSEL ELC</td>
<td>116.05 (14.02)</td>
<td>102.25 (19.77)</td>
<td>97.75 (24.74)</td>
<td>102.00 (16.84)</td>
<td>105.38 (17.53)</td>
</tr>
<tr>
<td><strong>36 months</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>VABS GM</td>
<td>14.41 (1.76)</td>
<td>13.62 (1.82)</td>
<td>13.12 (2.40)</td>
<td>13.67 (7.8)</td>
<td>13.96 (1.71)</td>
</tr>
<tr>
<td>VABS FM</td>
<td>15.85 (2.97)</td>
<td>14.85 (2.92)</td>
<td>14.00 (2.69)</td>
<td>14.50 (2.39)</td>
<td>15.62 (3.20)</td>
</tr>
<tr>
<td>MSEL GM</td>
<td>N/A</td>
<td>N/A</td>
<td>N/A</td>
<td>N/A</td>
<td>N/A</td>
</tr>
<tr>
<td>MSEL FM</td>
<td>56.58 (13.93)</td>
<td>48.58 (15.49)</td>
<td>39.47 (15.05)</td>
<td>50.67 (14.70)</td>
<td>54.00 (13.70)</td>
</tr>
<tr>
<td>MSEL VR</td>
<td>59.00 (10.92)</td>
<td>56.04 (13.87)</td>
<td>50.12 (18.66)</td>
<td>55.67 (12.41)</td>
<td>60.17 (9.13)</td>
</tr>
<tr>
<td>MSEL ELC</td>
<td>115.77 (16.25)</td>
<td>105.38 (21.52)</td>
<td>94.75 (28.51)</td>
<td>103.42 (18.98)</td>
<td>113.46 (13.26)</td>
</tr>
</tbody>
</table>

* MSEL GM scores at 24 months are based on 37 participants in each of the low-risk and at-risk groups. Data were not collected from the first participants in each group in the original study for methodological reasons, and not due to specific participant characteristics.

Current analyses.) Thus, Visual Reception scores were compared between groups as a proxy for developmental level as independently as possible of motor skill. These standardized scores are also presented in Table 2.

As the motor scales on the MSEL and VABS were not always completed for every child at all visits, cross-sectional analyses were conducted on the whole dataset. Differences between groups on the gross and fine motor scales were first compared within each visit (except for MSEL Gross Motor scores at 36 months,
for which data were not available). Planned contrasts between groups on gross and fine motor scales were corrected for multiple comparisons ($p = .01$). Next, correlations between the gross motor scales on the MSEL and VABS, and between the fine motor scales on these two measures, were conducted. All analyses were first conducted on the low-risk and at-risk groups. The at-risk group was then split into three subgroups, based on their diagnosis, and the analyses repeated between these three subgroups. As some data were not normally distributed, both parametric and non-parametric analyses were conducted on the data. In the cross-sectional analyses, results were almost identical using these methods, and so parametric tests are reported for all comparisons (in the only case when the significance of a result changed between parametric and non-parametric tests, both are reported). In the correlation analyses, a number of differences were found between parametric and non-parametric tests, so only non-parametric tests are reported, for clarity.

Cross-sectional analyses

At 7 months, at-risk infants had significantly lower VABS Gross and Fine Motor scores than the low-risk group, $t(100) = 3.57, p = .001, r = .34$ (Gross), $t(100) = 3.41, p = .001, r = .32$ (Fine). On the MSEL, groups also differed on both Gross Motor scores, $t(101) = 2.65, p = .01, r = .26$, and on Fine Motor scores, $t(101) = 2.82, p = .01, r = .27$. At 14 months, Fine Motor scores were significantly lower in the at-risk than the low-risk group on both the MSEL, $t(98) = 2.85, p = .01, r = .28$, and the VABS, $t(95) = 4.04, p < .001, r = .38$. Gross Motor scores on the two instruments were not significantly different between the groups (all $ts < 2.5, p > .02$). By 24 months, differences between groups were only found in Gross Motor skills, and reached our criterion for significance on the MSEL, $t(65.38) = 6.46, p < .001, r = .61$, but not the VABS, $t(97) = 1.96, p = .05, r = .19$. Fine Motor skills did not differ significantly on either measure (all $ts < 2.3, p > .02$). At 36 months, group differences did not reach our criterion for significance on the VABS, $t(97) = 2.19, p = .03, r = .22$ (Gross), and $t(97) = 1.68, p = .10, r = .17$ (Fine), but the at-risk group scored significantly lower on the MSEL Fine Motor scale than the low-risk group, $t(99) = 2.72, p = .01, r = .26$. Non-parametric tests were conducted on any data violating the assumption of normality. These tests did not change the results presented here, except that a significant difference was found between groups on the VABS Gross Motor scale at 14 months, $U = 839.0, p = .01, r = .26$.

In order to clarify whether differences in motor abilities between groups were simply signs of overall developmental delay, scores on the MSEL Visual Reception scale were compared. Group differences did not reach our criterion for significance between at-risk and low-risk groups, at 7 months, $t(101) = 2.18, p = .03, r = .21$, suggesting that any between-group differences in motor abilities found by this age were not the result of a more general delay in the at-risk...
group at the recruitment stage. Furthermore, the at-risk group did not differ significantly from the low-risk group on Visual Reception scores at any of the later visits (all \( t < 2.3, p > 0.03 \)).

Data from the at-risk group were also split into the three diagnostic subgroups (TD-sibs, AT-sibs, ASD-sibs; see Table 2), and comparisons between these subgroups at each visit were conducted. No significant differences were found between these subgroups at any age on any of the scales (all \( F_s < 2.7, p > .1 \)), except for differences in MSEL Fine Motor scores at 36 months \( F(2, 50) = 5.25, p = .01, \eta^2_p = .17 \). Post hoc comparisons with Bonferroni corrections revealed that this difference was driven by significantly higher Fine Motor scores in the TD-sibs subgroup than in the ASD-sibs subgroup (\( p = .01 \)), with no significant differences between the AT-sibs subgroup and either of the other two subgroups. Figure 1 presents the MSEL Gross and Fine Motor scores for the three subgroups at each visit, compared to the average standardized score of 50 (the low-risk group performed at or above this level). As this Figure depicts, the three subgroups all tended to perform slightly below average on the Gross Motor scale, while Fine Motor scores were generally at average or above for their ages in the TD-sibs and AT-sibs subgroup. However, those children who later developed ASD showed a decline in their Fine Motor skills compared to other children their age at the later visits, resulting in Fine Motor scores that were significantly lower than those of the TD-sibs by 36 months, and well below the standardized score of 50.

Correlations between measures on gross and fine motor scales

The degree of agreement between parental report and standardized assessment of motor skills was of interest: Bivariate correlations were conducted between the MSEL and VABS Gross and, separately, Fine Motor standardized scores at each visit (see Table 3). Spearman’s rho statistics are reported for all correlations to account for any data in one or more groups violating the assumption of a normal distribution. Gross Motor scores on the two measures were highly correlated at each visit in the low-risk group (all \( r > .5, p < .001 \)), and in the ASD-sibs subgroup (all \( r > .8, p < .001 \)). Fine Motor scores did not tend to correlate across instruments until 24 months of age, and then only in the low-risk group (24 months: \( r > .41, p < .01 \) and in the at-risk group as a whole (24 months: \( r > .35, p < .01 \); 36 months: \( r > .56, p < .001 \)). Agreement between the standardized test and parent reports for those children who developed atypically but did not go on to develop ASD was particularly poor (all \( r < .6, p > .03 \), except for Gross Motor scores at 14 months).

DISCUSSION

The current report aimed to measure motor development over infancy in those at-risk of developing ASD and those at low-risk, and was the first to directly
Figure 1. Mean standardized scores (and standard errors) for the three at-risk subgroups on (a) MSEL Gross Motor and (b) MSEL Fine Motor scales at each visit. Note: The MSEL Gross Motor scale was not administered at 36 months. The dotted line represents the average T-score of 50.

compare a parental report (VABS) and a standardized assessment (MSEL) of gross and fine motor skills between the two groups at 7 months. Both assessments revealed differences between low-risk and at-risk groups in motor skills as early as 7 months, consistent with past findings but identifying differences even earlier in development. Interestingly, those children in the at-risk group who were later diagnosed with ASD did not differ significantly from other at-risk children at early ages, but did have significantly lower Fine Motor scores than the TD-sibs.
subgroup at 36 months. Within-group analyses revealed that scores on gross motor scales were highly correlated between the two measures in both the low-risk group and the ASD-sibs subgroup, although correlations between fine motor scales were more dependent on age at testing.

The difference in the strength of correlations between gross and fine motor skills on the two measures is noteworthy. In particular, the strong correlation between gross motor scales could be due to the easier detection of changes in these abilities compared to fine motor skills, with important developmental milestones such as sitting unsupported and crawling being easily observable by both parents and researchers over the study timescale. In this case, it is much easier for a researcher to identify the behaviour in a controlled environment in which that motor act is the main focus. It is interesting to note that the gross motor scores are very strongly correlated in the ASD-sibs subgroup, as this suggests that scores on parent reports for these children may be even more reliable than for children without a later ASD diagnosis across the at-risk and low-risk groups, as these parents may have had more opportunities to observe motor assessments as part of diagnostic or therapeutic processes. It is also possible that the children in the ASD-sibs subgroup show more apparent atypicalities or differences in motor skill that are easily identified by both parents and researchers.

Both the VABS and the MSEL revealed differences between groups on gross and fine motor skills, with lower scores in the at-risk group than the low-risk group as early as 7 months. This is earlier than in previous research comparing samples on a range of gross motor skills (e.g., Landa & Garrett-Mayer, 2006), but in line with more recent research revealing head-lag in more at-risk than low-risk infants at 6 months (Flanagan, Landa, Bhat, & Bauman, 2012). Crucially, Visual

### TABLE 3

Bivariate correlations (Spearman's rho) between standardized scores on the gross motor scales of the MSEL and VABS, and between fine motor scales of the MSEL and VABS tests, across ages and groups

<table>
<thead>
<tr>
<th>Group</th>
<th>GM</th>
<th>FM</th>
<th>GM</th>
<th>FM</th>
<th>GM</th>
<th>FM</th>
<th>GM</th>
<th>FM</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>7</td>
<td>14</td>
<td>24</td>
<td>36</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low-risk</td>
<td>.79**</td>
<td>.38</td>
<td>.87**</td>
<td>.07</td>
<td>.53**</td>
<td>.41*</td>
<td>N/A</td>
<td>.17</td>
</tr>
<tr>
<td>At-risk (combined)</td>
<td>.65**</td>
<td>.15</td>
<td>.37*</td>
<td>.16</td>
<td>.19</td>
<td>.35*</td>
<td>N/A</td>
<td>.56**</td>
</tr>
<tr>
<td>ASD-sibs</td>
<td>.85**</td>
<td>.21</td>
<td>.96**</td>
<td>.35</td>
<td>.85**</td>
<td>.37</td>
<td>N/A</td>
<td>.49</td>
</tr>
<tr>
<td>AT-sibs</td>
<td>.61</td>
<td>–.15</td>
<td>.82**</td>
<td>.19</td>
<td>.13</td>
<td>.27</td>
<td>N/A</td>
<td>.56</td>
</tr>
<tr>
<td>TD-sibs</td>
<td>.59*</td>
<td>.30</td>
<td>.78**</td>
<td>.40</td>
<td>.36</td>
<td>.38</td>
<td>N/A</td>
<td>.57*</td>
</tr>
</tbody>
</table>

**Notes:** The number of participants in each group that completed both measures differs between ages. GM = Gross Motor; FM = Fine Motor. *p < .01; **p < .001.
Reception scores (which are assessed somewhat independently of motor function) did not differ between groups at this age, suggesting that an overall developmental delay is not responsible for motor differences. These significant differences at 7 months in fact result from infants in the at-risk group performing below average for their age, with the low-risk group performing at the expected level. It is important to note, however, that differences between groups on fine motor skills at later ages may be driven by scores well above the mean in the low-risk group, with the at-risk group showing no delay but performing at the expected level. This is particularly the case at 14 months, when low-risk infants have very high fine motor scores. It is possible that between 7 and 14 months, infants taking part in the current study in the low-risk group have more opportunities to practice fine motor skills due to other factors in the family environment, resulting in these particularly high scores, and therefore in the significant difference between groups. Nevertheless, in the gross motor domain, the at-risk group do perform consistently below the mean, suggesting that it is these skills that are most delayed in the at-risk group and that could have an effect on the way in which these infants move around and interact with their environment. Any differences in early exploration of the environment could also impact on the development of fine motor skills, which might explain why those children who go on to develop ASD have significantly poorer fine motor scores at 36 months compared to typically developing children in the at-risk group.

Considering further the division of the at-risk group into three at 36 months, the lack of significant differences early on between the three subgroups is an important finding, suggesting that poor motor skills may not be ASD-specific. Rather, they may be a more general indicator of atypical development as well as, in some cases, showing an association with the social and communication difficulties that are core to ASD. This view is supported by the number of children who presented other developmental concerns (AT-sib subgroup). However, while gross motor skills were generally similar between subgroups (slightly below average across visits), those infants who later developed ASD had increasing difficulties with Fine Motor skills over developmental time. This can be related to retrospective reports of poor fine motor skills in children with ASD and minimally fluent speech compared to those with fluent speech (Gernsbacher et al., 2008). That study used a composite of fine motor skills across a wide age range (between 6 and 24 months), whereas the current prospective design suggests that it is more complex fine motor skills that emerge later in development that might be associated with ASD. As more demands begin to be made on the fine motor system as children enter the school years, this gap could continue to widen. More research is required to understand the developmental progression of fine motor skills and the windows of achievement for fine motor milestones, as are available for gross motor milestones (World Health Organization, 2006). This could help to identify difficulties and to reduce the cascading effect of fine motor delays in the ASD-sibs subgroup.
The fact that the infants in the at-risk group had significantly lower motor scores than the typically developing low-risk group at 7 months is a novel finding which is of great interest. One possible explanation is that the socioeconomic status (SES) of the at-risk group was lower than that of the low-risk control group, as low SES has been related to poorer developmental outcomes in previous research (Bradley & Corwyn, 2002). However, a recent study specifically comparing 6- to 9-month-old infants from low and high SES backgrounds found no difference between motor scores on the Bayley Scales of Infant Development (Bayley, 1993), despite significantly lower scores on the Mental Development scale on this measure (Tomalski et al., accepted for publication). In our own data, significant differences in motor scores were still found between low-risk and at-risk groups when comparing only infants from high SES backgrounds, suggesting that any differences in SES would not explain the pattern of results presented in the current paper. More studies designed to investigate the relationship between SES and motor development in infancy would, however, be beneficial to our understanding of development in these at-risk groups.

Limitations

One difficulty with the preliminary data reported here is the fact that longitudinal relationships between motor skills could not be assessed effectively due to missing data, particularly from the MSEL Gross Motor scale from 24 months. Using a more fine-grained motor assessment from 36 months into early childhood will enable future studies to assess these developmental trajectories in more detail and assess the relationships with other variables over time. In addition, further studies could collect data between 7 and 14 months in order to document rapid motor changes within this age band, providing greater scope for understanding individual differences in later motor abilities. Understanding the changes in fine motor skills in this timeframe could be particularly useful for understanding the development of those infants who go on to develop ASD. Combining this measure with an examination of parent–child interaction during this focused time period would help to identify any general differences in families of at-risk versus low-risk infants that could contribute to the lower reported motor scores in this at-risk group. It will also be important to continue to assess these prospective samples into the school years to better understand the relationships between early motor development, later school achievement and ASD-related behaviours.

CONCLUSION

The current study has replicated previous evidence of motor delay in at-risk infants (Flanagan et al., 2012; Landa & Garrett-Mayer, 2006; Toth et al., 2007), adding new knowledge by testing infants at earlier ages and considering the differences between parent report and standardized measures over developmental
time. Due to the potential importance of early motor skills on the development of other cognitive domains (Iverson, 2010), it is vital that more research focuses on the motor profiles of infants, particularly those at risk of developing ASD, as atypical motor development may contribute to differences seen in other areas that make up its diagnostic criteria. It is important to note that, even if infants in the at-risk group do not go on to develop ASD, poorer motor development than that seen in low-risk infants as early as 7 months could have a negative impact on their language, social and cognitive development, affecting how infants interact with the environment and others around them (e.g., Campos et al., 2000; Iverson, 2010; Libertus & Needham, 2010; Tamis-LeMonda et al., 2008). In those infants that do develop ASD, fine motor skills seem to be a particular difficulty, and this could have important implications for the development of triadic interactions involving the manipulation and sharing of objects, which are often atypical in individuals with ASD (e.g., Charman, 2003). Early identification and interventions in which specific motor skills are taught or in which the child’s natural motor behaviour is stimulated, and which are suggested to be the most effective with infants at-risk of poor motor development (Blauw-Hospers & Hadders-Algra, 2005), could therefore be highly beneficial to these at-risk siblings. These interventions are important not only in improving motor capabilities, but also in ameliorating the effects of poor early motor skills on other areas of development. Based on the current data, while poor gross motor development may be a risk factor for a range of developmental difficulties, the effect that this may have on fine motor abilities could be of particular significance for those infants who develop ASD.

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