

Leonard H.C., Bedford, R., Charman, T., Elsabbagh, M., Johnson, M.H., Hill E.L. & the Basis team (2014). Motor development in children at-risk of autism: A follow-up study of infant siblings. *Autism*, 18(3), 281-291. (Sage)  
DOI: 10.1177/1362361312470037

Motor development in children at-risk of autism: A follow-up study of infant siblings

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### **Keywords**

Autism Spectrum Disorder, motor development, infant siblings, face processing, Broader Autism Phenotype

### **Acknowledgements**

We are grateful for the generous contributions BASIS families have made towards this study. Special thanks go to Kim Davies, Kathy Filer, Louise Hood, Sarah Moore and Susannah Price for their contributions to testing during this project.

### **Abstract**

Recently, evidence of poor or atypical motor skills in Autism Spectrum Disorder (ASD) has led some to argue that motor impairment is a core feature of the condition. The current study uses a longitudinal prospective design to assess the development of motor skills of twenty children at increased risk of developing ASD, who were recruited and tested at 9 and 40 months old, on the basis of having an older sibling diagnosed with the condition. All children completed a range of motor, face processing, IQ and diagnostic assessments at a follow-up visit (aged 5-7 years), providing a detailed profile of development in this group from a number of standardised, parental report and experimental measures. A higher proportion of children than expected demonstrated motor difficulties at the follow-up visit, and those highlighted by parental report as having poor motor skills as infants and toddlers were also more likely to have lower face processing scores and elevated autism-related social symptoms at 5-7 years, despite having similar IQ levels. These data lend support to the argument that early motor difficulties may be a risk factor for later motor impairment as well as differences in social communication and cognition, traits that are related to ASD.

The development of motor skills in children with a diagnosis of Autism Spectrum Disorder (ASD) has received an increasing amount of attention in recent years, with some beginning to argue that motor impairments may be a core feature of ASD (e.g., Fournier et al., 2010; Hilton et al., 2011). Understanding motor

functioning in ASD is of great interest, as motor impairments can have adverse effects on school achievement (e.g., Alloway, 2007; Michel et al., 2011), language and social cognitive outcomes (e.g., Archibald & Alloway, 2008; Cummins et al., 2005), and a range of activities of daily living, including dressing or feeding oneself (e.g., Summers et al., 2008). Evidence for a relationship between motor development and language and social communication skills can also be seen in typical development through the tight coupling of motor and language milestones throughout infancy (Iverson, 2010). In addition, motor development can affect how infants interact with individuals around them, with improved object manipulation skills resulting in altered patterns of attention to others in the environment (Libertus & Needham, 2010). The onset of crawling and walking also produces more opportunities for joint attention (through gaze following) and social referencing (through interpreting facial expressions) by changing the type and number of interactions infants have with their caregivers (e.g., Campos et al., 2000; Tamis-LeMonda et al., 2008). It is therefore important to investigate motor difficulties in ASD, in which language and social communication difficulties are key diagnostic criteria, as these may be related to early motor delays or problems.

Previous research into motor difficulties in children diagnosed with ASD has identified widespread motor dysfunction (see Mari et al., 2003, for a review), with a high proportion of children with ASD displaying impairments in producing speeded movements (e.g., Jansiewicz et al., 2006), planning and learning motor sequences (e.g., Hughes, 1996; Mostofsky et al., 2000; Rinehart et al., 2001), executing skills such as throwing, catching or balancing (e.g., Green et al., 2009; Manjiviona & Prior, 1995; Whyatt & Craig, 2011) and on more general tests of gross and fine motor skills (e.g., Lloyd et al., 2011; Provost et al., 2007; Staples & Reid, 2010). Retrospective analyses of motor behaviour in home movies of children later diagnosed with ASD have reported mixed findings (Baranek, 1999; Ozonoff et al., 2008; Teitelbaum et al., 1998), but such videos may not be representative of the infant's motor functioning (Baranek, 1999). One way of addressing this problem is to assess motor behaviour *prospectively* in infants at greater risk of developing ASD due to heritability within families, and it is this method that has been utilised in the current study.

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 younger siblings is between 10-20% (Constantino et al., 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 (Rogers, 2009). Many of these prospective studies have not been designed with motor development in mind, but some have collected standardised motor data during infancy, particularly from the Vineland Adaptive Behavior Scales-II (VABS; Sparrow et al., 2005), and the Mullen Scales of Early Learning (MSEL; Mullen, 1995). These assess a range of abilities including Gross Motor and Fine Motor skills by parent report and standardised assessment, respectively. Gross and fine motor impairments in at-risk siblings have been reported after 14 months of age on the MSEL (Landa & Garrett-Mayer, 2006) and after 20 months on the VABS (Toth et al., 2007). Leonard et al. (under review) reported even earlier differences between at-risk and low-risk siblings on the VABS, with at-risk siblings performing more poorly on both gross and fine motor scales at 7 months.

One reason that results may differ between these research groups is due to the number of at-risk siblings in each of the samples who go on to develop ASD (Rogers, 2009). For example, Toth et al. (2007) only tested unaffected younger siblings, while both other studies included follow-up to ascertain which infants in the at-risk group later developed ASD. As motor functioning is reported to be a reliable predictor of later autism diagnosis (Brian et al., 2008), with those with intact motor skills more likely to have better outcomes or lose an early ASD diagnosis entirely (Sutera et al., 2007), it seems possible that those samples that do not include any affected siblings will find fewer and/or later differences in motor ability. This is supported by a recent paper that compared siblings concordant and discordant for ASD (Hilton et al., 2011). The authors reported poorer functioning in the affected siblings on a fine-grained standardised motor assessment, the Bruininks-Oseretsky Test (Bruininks & Bruininks, 2005), in children aged over 4 years, while unaffected siblings performed at the normative mean. This study did not, however, present any longitudinal data, so it is not clear whether poorer functioning in the affected siblings was evident at earlier ages or if motor skills were stable over development in both affected and unaffected siblings.

The first aim of the current exploratory study was to build a profile of functioning in at-risk siblings at 5-7 years across a range of age-appropriate standardised, parental report and experimental measures of motor and social skills, IQ and autism-related symptomatology. This would provide a more detailed understanding of the relative strengths and weaknesses in this group later in childhood than is usually reported. Hilton et al. (2011) reported significant correlations between motor functioning and scores on the Social Responsiveness Scale (SRS; Constantino & Gruber, 2005), a parent report measure of autistic social traits, with more severe symptoms predicting greater motor impairments in the affected siblings, and recent reviews have reported an association between motor impairment and overall symptom severity in ASD (Jeste, 2011; Maski et al., 2012). Other studies have reported face processing deficits or atypicalities in individuals with ASD (e.g., Adolphs et al., 2001; Dawson et al., 2005; Klin et al., 1999; Riby et al., 2007) and in at-risk siblings (Elsabbagh et al., 2012; Elsabbagh et al., 2009; McCleery et al., 2009). Given that ASD is often characterised by both poor motor functioning and atypical or impaired face processing, and that aspects of face processing and motor development may be inextricably linked (e.g., Campos et al., 2000), assessing these outcomes in the at-risk siblings was central to the current investigation.

The other objectives of the current study were related to the longitudinal data collected at 9 and 40 months from the children later assessed at 5-7 years. Specifically, the second aim was to investigate whether differences in motor skills reported by parents on the VABS at 9 and 40 months persisted to 5-7 years when assessed by a more fine-grained standardised measure, the Movement ABC-2 (MABC-2; Henderson et al., 2007), and whether differences would also be evident on the other outcome measures as a function of early motor scores. The third aim was to address an important question relating to the use of parental report and standardised assessment measures of motor functioning during infancy and childhood. In particular, the degree of correlation between the two types of measure at each age point is of interest to both researchers and practitioners, as parent reports can be a useful and efficient method for identifying infants and children who may be at increased risk of developing motor difficulties. Correlations between data collected from the VABS and MSEL at the earlier visits, and between the VABS and MABC-2 at the later visits, were therefore calculated to address this question.

## Method

### Participants

Participants were families taking part in an ongoing longitudinal research program: BASIS, a collaborative network facilitating research with infants at-risk for autism. Thirty infants were recruited in the first year of life as part of this larger study from a database of volunteers on the basis that they had an older sibling with a confirmed clinical diagnosis of ASD (18 males). These infants were assessed at 9 months of age on a range of standardised and experimental tasks and parental report questionnaires. As part of this larger study, follow-up assessments took place at 40 months ( $N = 28$ ), when characteristics of ASD are known to be clearer than at earlier ages, and in line with other prospective studies of at-risk siblings. Data were again collected from questionnaires and standardised and experimental tasks. Of the 28 participants from the second stage of testing, 20 were able to return for a follow-up visit at the age of 5-7 years (Mean age = 6 years, 2 months;  $SD = 5$  months; Males = 12) for more fine-grained motor assessments and additional tasks.

Recruitment, ethical approval, informed consent, as well as anonymised data collected from the first (pilot) cohort of BASIS were made available through BASIS for the current study. Some of the measures collected are anonymised and shared among scientists to maximise collaborative value and to minimise assessment burden on the families. A clinical advisory team of senior consultants works closely together with the research teams, and if necessary with the family's local health services, to ensure that any concerns about the child arising during the study are adequately addressed.

At the time of enrolment, none of the infants had been diagnosed with any medical or developmental condition. All had an older sibling with a community clinical diagnosis of an autism spectrum disorder (hereafter, proband), diagnosis of whom was confirmed by two expert clinicians involved in the research team, using the Development and Wellbeing Assessment (DAWBA; Goodman et al., 2000) and the parent report Social-Communication Questionnaire (SCQ; Rutter et al., 2003). Most probands (18/20) met criteria for ASD on both the DAWBA and SCQ, in addition to having been diagnosed with an ASD by a local clinician. Of the remaining two probands, one did not have the DAWBA completed but met criterion on the SCQ, and for the other neither DAWBA nor SCQ data were available. Parent-reported family medical histories were taken, with no exclusions made on the basis of significant medical conditions in the proband or immediate family members. At 5-7 years (hereafter, "follow-up visit"), only one child had an independent diagnosis of ASD from a qualified clinician, and this diagnosis was confirmed by scores on the DAWBA, SCQ and the Autism Diagnostic Observation Schedule (ADOS-G; Lord et al., 1989) conducted during the visit.

### Materials

Data were collected through a range of measures at each assessment. The measures of interest for the current report are outlined in Table 1 for each assessment point, and are split into 'diagnostic measures', 'motor measures', 'face processing measures' and 'IQ measures', corresponding to the aims presented above. Verbal and non-verbal IQ were assessed at the follow-up visit using the Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III UK; Wechsler, 2003). Participants completed the Information and Vocabulary subtests, and their scores were prorated to

produce Verbal IQ (VIQ); similarly, the Block Design and Matrix Reasoning scores were prorated to produce Non-verbal IQ (NVIQ). The rest of the measures are described in more detail below.

--Table 1 about here--

### *Diagnostic Measures*

Data from the SCQ (Rutter et al., 2003) and the ADOS (Lord et al., 1989) were used in the analyses for each of the participants. The SCQ is a parental report measure of ASD-related symptoms consisting of 40 questions relating to social communication and language, and the total score is used in the analyses. The ADOS is a semi-structured assessment of ASD-related symptoms in which the participant completes a number of tasks aimed to tap certain behaviours, such as joint attention, conversation and gesture. Different modules can be used depending on the participant's expressive language level and chronological age. Scores on items are summed to produce total scores for different domains, including social communication, repetitive behaviours and creativity. Only the total social communication score will be included in the current analyses.

### *Motor measures*

The VABS (Sparrow et al., 2005) was completed for participants at all three assessments. This instrument measures communication, daily living, socialisation and motor skills, as well as maladaptive behaviour. Only the motor skills domain from each test will be considered in this report and will be separated into Gross and Fine Motor subdomains. Parents and caregivers reported whether they had seen a particular behaviour on a scale of "Never", "Sometimes" or "Usually". They could also respond "Don't Know" or "No opportunity" to any of the items. A motor composite score, combining Gross and Fine Motor scores, was also used to assess the percentile rank of the scores from each of the participants, i.e., each participant's performance in comparison to a normal distribution of scores.

The MSEL (Mullen, 1995) is a standardised test of early cognitive and motor development between the ages of 0-68 months, consisting of measures of receptive and expressive language, visual reception and gross and fine motor skills, and was conducted at 9 and 40 months. The motor domain of the MSEL is made up of Gross Motor and Fine Motor subdomains, and 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, as demonstrated by correct use of that object when placed in front of the child (e.g., a spoon). The close connection of many of these 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 (Leonard et al., under review; Lloyd et al., 2011). In the current analysis we use the Visual Reception scale from the Mullen to account for general developmental differences at 9 months without confounding motor ability with development. The use of the Early Learning Composite (ELC) or 'ratio NVIQ' (Lloyd et al., 2011) is not appropriate for the current analyses, as these measures are calculated using Fine Motor scores.

The MABC-2 (Henderson et al., 2007) is a standardised test of motor skills, consisting of three subtests: manual dexterity (e.g., posting coins, threading beads, drawing), aiming and catching, and static and dynamic balance, each of which is comprised of a series of speeded and non-speeded motor tasks. This test was

conducted at the follow-up visit for a more fine-grained measure of motor ability in this group. Both raw scores and percentile scores are used in the current data analyses. Percentile ranks are used in the MABC-2 to highlight those with ‘significant’ or ‘borderline’ movement difficulties, based on those scoring below the 5<sup>th</sup> and the 15<sup>th</sup> percentiles, respectively, and these cut-offs are used in the current analyses.

### *Face processing*

An experimental face processing task was conducted at the follow-up visit, adapted from Bruce et al. (2000) to test recognition in the four main face processing domains: expression, gaze, speech sound (lip reading) and identity. As described earlier, both expression and gaze processing may develop with the onset of crawling and walking (e.g., Campos et al., 2000; Tamis-LeMonda et al., 2008), and it was of interest to assess whether lip reading and identity matching were also related to early motor activity. Grey scale images of children’s faces were presented and participants were asked to identify which image was showing a particular expression, gaze direction or speech sound, or which two images represented the same identity. The first three tests were therefore identification tasks, while the identity test involved matching, as it is not possible to choose an identity that corresponded to a category provided by the experimenter without extensive training (Bruce et al. 2000). Each test comprised twelve trials. The dependent variables were accuracy and Fractional Success Rate (FSR), i.e., the number of children that passed each test by scoring above chance (10/12 correct trials). This battery has been developed for children aged 4-10 years and is developmentally sensitive (Bruce et al., 2000). Examples of each test are presented in Figure 1, and further details can be found in Bruce et al. (2000).

--Figure 1 about here--

### Procedure

At the 9 month and 40 month visits, informed consent was obtained from the parent and the child took part in a number of standardised and experimental tasks, either sitting on the parent’s lap (9 months) or independently (40 months). Parents usually completed the questionnaires prior to the visit.

At the follow-up visit, the tasks were explained to both the parent and the child in appropriate language and pictures, and informed consent was obtained from both parties. Parents completed the questionnaires throughout the visit, while the participant completed the tasks. Tasks were varied in order depending on the time of the visit and the needs of each child, with breaks as necessary. No individual task took longer than 45 minutes. Participants received stickers throughout the testing session for motivation and reward, and were given a certificate at the end of the session for their participation.

### Results

As outlined in the introduction, three questions were addressed by the analyses: (i) What is the profile of motor and social functioning of the younger siblings of children with autism at 5-7 years? (ii) Do early differences in motor skills correspond to differences in outcomes at later ages? (iii) Are poor motor skills identified by parent report evident in standardised assessments? Each of these questions is addressed, in turn, below.

#### **(i) What is the profile of motor and social functioning at 5-7 years?**

The means and standard deviations of the data collected from the MABC-2, VABS, face processing battery, WPPSI and diagnostic outcome measures at the follow-up visit are presented in Table 2. To examine variability across the whole group of at-risk infants, scores on the SCQ and ADOS were used as continuous measures of social-communication functioning or autism-related symptomatology. Any data that were not normally distributed were either square-root transformed or analysed using non-parametric tests, as appropriate, and Bonferroni corrections were applied as necessary.

--- Table 2 about here ----

As shown in Table 2, there was a wide range of scores on the IQ test. Using an average IQ score across verbal and nonverbal domains, all children had an IQ above 70 except for the child with a confirmed diagnosis of autism. This child's data were not found to have undue influence on the analyses, as for several tasks it was not possible to calculate standardised or percentile scores from raw scores, or the child was unable to understand or complete the task (see individual analyses below). Those scores that could be used were included in the analyses, in order to reflect the range of functioning in the group. In addition, IQ was taken into account in the analyses as appropriate.

#### *Motor outcome measures*

Standardised scores were not available for two children on the VABS, as their raw scores fell outside of the norms provided, but analyses that were conducted on raw scores included data from these children. Standardised and percentile scores were also not available on the MABC-2 for one of these children for the same reason. Analyses of the longitudinal data were conducted without these participants' scores, and no significant differences were evident in the analyses, therefore these data were included in the analyses reported. On the MABC-2, two of the 19 scorable children were highlighted as having significant movement difficulties (scoring at or below the 5<sup>th</sup> percentile) and four children with borderline movement difficulties (scoring between the 6<sup>th</sup> and 15<sup>th</sup> percentiles) for the Total Percentile. A further four children fell within these groups on at least one subcomponent of the test. Although the VABS motor composite is not designed for classification of motor disorders, percentile points are provided. Using the same criteria for the VABS Motor Composite Percentile highlighted four children as having significant motor difficulties, and seven children with borderline movement difficulties.

#### *Face processing outcome measures*

Data for each of the subtests on the face recognition battery are presented in Table 2, and Figure 2 provides a comparison of the current results to those reported by Bruce et al. (2000) with a group of 5-6 year olds. One participant with a confirmed ASD diagnosis did not provide data for any of the subtests due to difficulty understanding the task demands, while data is missing from a further two participants for the speech sound subtest due to technical difficulties. The percentage of children scoring above chance on each test (at least 10 out of 12 correct answers) varied between tests. More children passed the expression identification subtest (95%) than the gaze identification subtest (79%), the speech sound subtest (82%), and the identity matching subtest (63%), which is a similar pattern to that reported by Bruce et al. (2000; see Figure 2).

---Figure 2 a and b about here---

**(ii) Do early differences in motor skills correspond to differences in outcomes at later ages?**

As the MABC-2 cannot be used with children younger than 3 years, it is useful to investigate whether the cut-offs used by this instrument to identify those with significant and borderline movement difficulties can be applied to other measures that can be administered during infancy. As the MSEL does not provide a composite motor score, we tested this question using the motor score provided by the VABS, applying the 15<sup>th</sup> percentile cut-off for motor difficulties from the MABC-2 to this instrument. This allowed children with any movement difficulty or delay in infancy (i.e., both ‘borderline’ and ‘significant’ movement difficulties, according to the MABC-2 criteria) to be compared to those children scoring above this critical range. The group was therefore split according to each child’s Motor Composite Percentile on the VABS at 9 months into those scoring below the 15<sup>th</sup> percentile (“poor motor group”) and those scoring above the 15<sup>th</sup> percentile (“typical motor group”). These data are shown in Figure 3.

Independent t-tests and appropriate non-parametric equivalent tests conducted on these groups revealed no significant difference on the MSEL Visual Reception score at the same age,  $U = 39.50, p = .96$ , or on IQ at the follow-up visit,  $t(17) = .06, p = .96$  (NVIQ),  $U = 38.50, p = .59$  (VIQ). There was, however, a significant difference in MABC-2 Total Percentile at the follow-up visit, with the poor motor group ( $N = 10$ ) scoring significantly worse on the MABC-2 than the typical motor group ( $N = 8$ ),  $t(16) = 2.33, p = .03$  (see Figure 3a). Closer inspection revealed that three of the children in the poor motor group scored below the 15<sup>th</sup> Total Percentile on the MABC-2 at the follow-up visit. A further four children in this group were in the low normal range at the follow-up visit (scoring in the 25<sup>th</sup> percentile on MABC-2 Total Percentile). Splitting the children into groups based on their VABS Motor Composite Percentile at 40 months ( $N$  poor motor = 5,  $N$  typical motor = 14) revealed a more marginal group difference on MABC-2 Total Percentile at the follow-up visit,  $t(12.24) = 2.22, p = .05$ , with the group with poorer motor scores at 40 months performing more poorly on the MABC-2 at follow-up.

---Figure 3 about here---

In order to test if splitting the groups on the basis of their motor ability at 9 months also produced differences in autism-related social communication traits, the poor motor and typical motor groups were compared on their scores on face processing and diagnostic measures. On the face processing battery, the groups differed on two of the subtests at the follow-up visit, namely Expression Identification,  $U = 14.00, p = .01$ , and Gaze Identification,  $U = 15.00, p = .01$ , with those children in the poor motor group at 9 months performing significantly worse on these (see Figure 3b). No differences between groups were found for Speech Sound Identification or Identity Matching ( $ps > .06$ ). In addition, no group differences were found based on VABS Motor Composite Percentile at 40 months ( $ps > .08$ ) on any of the face processing subtests. In terms of the scores on diagnostic measures, groups split at 9 months did not differ significantly on any of the measures (see Figure 3c). However, when split at 40 months, children in the poor motor group scored



significantly higher on the SCQ,  $t(18) = 2.79, p = .01$  than the typical motor group, although the difference was not significant for the ADOS ( $p = .5$ ).

**(iii) Are poor motor skills identified by parent report evident in standardised assessments?**

Early motor data from the VABS and MSEL at 9 and 40 months were analysed, along with the motor outcome data from the follow-up visit. At 40 months, only Fine Motor data were available from the MSEL. The VABS and MSEL data are presented in Table 3, along with the number of participants included for each measure at each age. At 9 months, scores on the VABS Gross Motor scale correlated significantly with both the MSEL Gross Motor,  $r = .83, p < .001$ , and Fine Motor scores,  $r = .62, p = .01$ . At 40 months, the two measures were again significantly correlated, with the MSEL Fine Motor scores correlated with both the VABS Gross Motor,  $r = .69, p = .001$ , and Fine Motor scores,  $r = .67, p < .01$ . These results remained significant even after controlling for IQ.

Correlation analyses were also conducted on raw data from the MABC-2 composites and the VABS motor scales and motor composite to test the relationships between the two measures at 5-7 years. Manual Dexterity scores on the MABC-2 were significantly correlated with both the VABS Gross Motor,  $r = .62, p < .01$ , and VABS Fine Motor scores,  $r = .57, p = .01$ , but the other MABC-2 components and VABS scales were not significantly correlated once the analyses were controlled for multiple correlations ( $ps > .04$ ). Once IQ was partialled out of these correlations, the results were no longer significant.

---Table 3 about here---

## Discussion

The current study aimed to (i) investigate the profile of motor and social skills in a group of children at increased risk of developing ASD, (ii) examine the relationships between motor development and outcomes associated with ASD in this group, and (iii) assess the degree of correlation between standardised and parental report measures of motor skill. In relation to the first question under investigation (i), only one child had an independent diagnosis of ASD, and others in the group showed a wide range of motor, social and cognitive skills throughout their early development. At 5-7 years of age, 6 out of 19 (31.6%) of the group showed motor difficulties, scoring below the 15<sup>th</sup> percentile on a standardised motor assessment, the Movement ABC-2. This is a higher rate than expected from a typical sample of children. There was a relatively wide range of IQ and social-communication scores, but face processing scores largely fell in line with those found by Bruce et al. (2000) with children of the same age.

Interestingly, in relation to the second question being investigated (ii), those children with poorer motor skills at 9 months (defined here as scoring below the 15<sup>th</sup> percentile on the VABS) performed significantly worse on the standardised motor assessment at 5-7 years than those performing above the cut-off, suggesting that motor difficulties in infancy can be highlighted effectively and used to predict the risk of later motor problems. In addition, differences in face processing and ASD-related traits at 5-7 years were evident in those children identified as having motor difficulties

at 9 months. These later differences at 5-7 years were evident in the 'poor motor group' despite the fact that there were no differences between groups on IQ at this point, or on Visual Reception score on the MSEL at 9 months. This MSEL scale was used as a proxy for general ability during infancy due to the close connection of many of the items to stages of cognitive development (Leonard et al., under review; Lloyd et al., 2011), without using the MSEL Early Learning Composite or 'ratio NVIQ' (Lloyd et al., 2011), which are calculated using Fine Motor scores. The fact that the differences between subgroups at 5-7 years decreased when groups were assigned by parent-reported motor score at 40 months is perhaps not surprising; by this age, the key motor milestones, including sitting and standing upright, crawling, walking, reaching and grasping, should have been achieved, and medical professionals would likely have flagged children that haven't reached these milestones as having an underlying neurological disorder.

In relation to the final question of this investigation (iii), the moderate to strong correlations between parent-report measures and standardised motor assessments at 9 and 40 months support previous findings (Hilton et al., 2011; Leonard et al., under review; Lloyd et al., 2011), suggesting that parent questionnaires can be an efficient method of identifying motor difficulties in early life, and can be followed up by appropriate clinical assessments. The correlations between the VABS and the MABC-2 at 5-7 years, on the other hand, were not as strong and were not significant once IQ was partialled out. This could be for a number of reasons. It is possible that the parent report is less reliable at this stage, as parents have fewer opportunities to observe the quality of skilled actions when their children attend school compared to when the infant is at home with the parent for most of the time and when each new skill is acknowledged and celebrated. The MABC-2 is also a much more fine-grained, detailed motor assessment than the VABS and the MSEL, which are more general measures of development across a number of different domains. It is possible, therefore, that a more age-appropriate parental report measure used at older ages would be better correlated with the MABC-2, or at least benefit from a broader variety of standardised/age appropriate and psychometrically sound measures (including the DCDQ'07, for example). In order to address each of these issues, future studies could use an additional age-appropriate motor report measure and ask both parents and teachers to complete it for each child.

It is difficult to compare the results we have obtained to previous studies that have used a different motor assessment (Hilton et al., 2011), or have reported mean standardised scores and not percentiles (Whyatt & Craig, 2011). However, the current study showed lower levels of impairment than those reported for the ASD group by Whyatt & Craig (2011), which might be expected considering that the majority of the current group do not have ASD diagnoses. In comparison to the 'unaffected siblings' reported by Hilton et al. (2011), the current group generally performs worse on motor tasks. These differences may be due to the different motor assessment used by Hilton et al. (2011), as there are mixed findings concerning the equivalence of the Movement ABC and the Bruininks-Oseretsky test in testing motor ability (e.g., Cairney et al., 2009; Spironello et al., 2010). Overall, the current study has taken a different approach to assessing motor difficulty in the sibling group. Specifically, we have conducted in-depth analyses of longitudinal motor data in one group rather than comparisons of motor outcomes at later stages between groups. Using the normative percentiles provided by the MABC-2, we have been able to assess motor difficulties in each participant, as well as compare mean scores on tasks for children that have scored above and below a particular cut-off at early ages. These longitudinal data

suggest motor difficulties identified at 9 months can persist into early childhood, and this has important implications for other aspects of cognitive and social development.

### **Conclusions and Future Directions**

The current study has added to the increasing amount of evidence of motor difficulties in infants and children at risk of developing ASD (e.g., Landa & Garrett-Mayer, 2006; Leonard et al., under review), with around a third of the group performing poorly on a standardised test of motor skill at 5-7 years. Perhaps more importantly, infants with poor motor skills who were identified by parent report as early as nine months showed poorer social-cognitive outcomes at later ages than children with typical motor skills. Most strikingly, the face processing domains that were poorer in children with early motor difficulties were gaze and expression processing, supporting the suggested link between motor skill and early social referencing and joint attention (e.g., Campos et al., 2000; Tamis LeMonda et al., 2008). Specifically, the ability to move around and explore the environment, manipulating objects and sharing them with others, provides more opportunities to engage in joint attention and changes the types of vocalisations and expressions the infant receives from the parent. As the onset of crawling and independent walking would be expected to occur by the age of 18 months (WHO, 2006), these interactions between early motor skill and face processing would seem to be relatively time-sensitive (within the fairly broad typical range), and could explain why differences in motor skill at 40 months did not affect later face processing outcomes. It is important to note that, while children with poorer early motor skill performed worse on these two face processing tasks, they still scored within the normal range on these tasks at 5-7 years. As all of the children who completed the face processing task did not have a diagnosis of ASD at this stage, this is perhaps not surprising. However, this result has implications for those at-risk siblings who do go on to develop ASD, providing a possible mechanism for the development of poor or atypical social-cognitive abilities through the cascading effects of early motor delay or difficulties.

Future research should continue to compare groups of children with and without ASD, along with their siblings, as well as conducting more in-depth analyses within these groups. It will also be important to compare these participants with children with other neurodevelopmental disorders, particularly those with Developmental Coordination Disorder who are diagnosed on the basis of a core motor impairment (Sugden & Chambers, 2005). Such a comparison will enable a better understanding of the nature and specificity of motor difficulties in ASD, assessing the degree to which elevated motor symptoms are related to ASD or whether they present a more general risk factor for poor outcomes that manifest themselves differently across disorders. Such longitudinal, cross-syndrome studies will be vital in developing interventions and ameliorating the effects that motor difficulties can have on daily life and educational achievement, and in raising the awareness of the research community, practitioners and the general public concerning the importance of motor development in typical cognitive and social functioning.

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**Table 1.** Measures included in the current paper from each data collection point, at 9 months, 40 months and 5-7 years (follow-up visit).

Measure	Age of Data Collection		
	9m	40m	5-7 yrs
<i>Diagnostic</i>	n/a	ADOS SCQ	ADOS SCQ
<i>Motor</i>	VABS MSEL	VABS MSEL (fine motor)	VABS MABC-2
<i>Face Processing</i>	n/a	n/a	Bruce et al. (2000) battery
<i>IQ</i>	MSEL (Visual Reception)	MSEL (Visual Reception)	WPPSI (short form)

*Note.* VABS = Vineland Adaptive Behavior Scales; MSEL = Mullen Scales of Early Learning; ADOS = Autism Diagnostic Observation Schedule; SCQ = Social Communication Questionnaire; MABC-2 = Movement ABC-2; WPPSI = Wechsler Preschool and Primary Scales of Intelligence

**Table 2.** Means, standard deviations and range of scores for each of the measures at the follow-up visit.

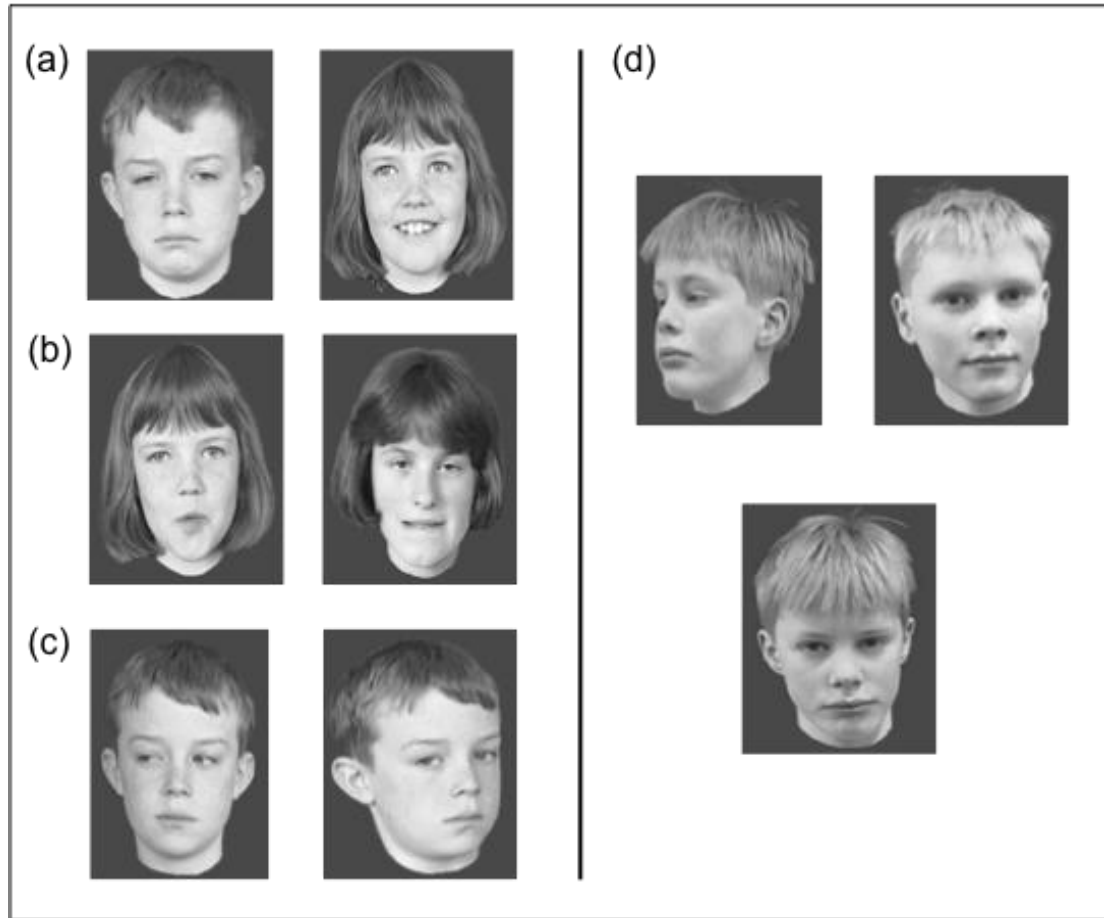
<b>Measure</b>	<b>Mean (SD)</b>	<b>Range</b>
<i>MABC-2 Composite Raw Scores</i>		
Manual Dexterity	24.25 (8.61)	3 – 34
Aiming and Catching	17.80 (6.02)	6 - 28
Balance	28.21 (6.36)	17 - 37
Total	72.00 (15.08)	39 - 94
<i>VABS</i>		
Gross Motor Raw Score	74.35 (6.85)	48 -80
Fine Motor Raw Score	55.35 (12.41)	13 - 68
Motor Composite Standardised Score	83.58 (13.99)	51 - 111
<i>Face Processing Battery Raw Scores</i>		
Expression Identification	91.18 (6.89)	75-100
Gaze Identification	85.78 (21.40)	42 - 100
Speech Sound Identification	89.71 (15.74)	50 - 100
Identity Matching	80.88 (24.61)	17 - 100
<i>WPPSI Composite Standardised Scores</i>		
Verbal IQ	96.55 (18.38)	53-127
Nonverbal IQ	89.80 (23.30)	45 - 131
<i>Diagnostic Measures Raw Scores</i>		
ADOS Total	7.35 (5.64)	0 - 20
SCQ Total	4.75 (5.96)	0 - 24

Note. Scores for the VABS motor composite and the WPPSI IQ composites can only be calculated after standardisation, and are therefore shown as standardised rather than raw scores.

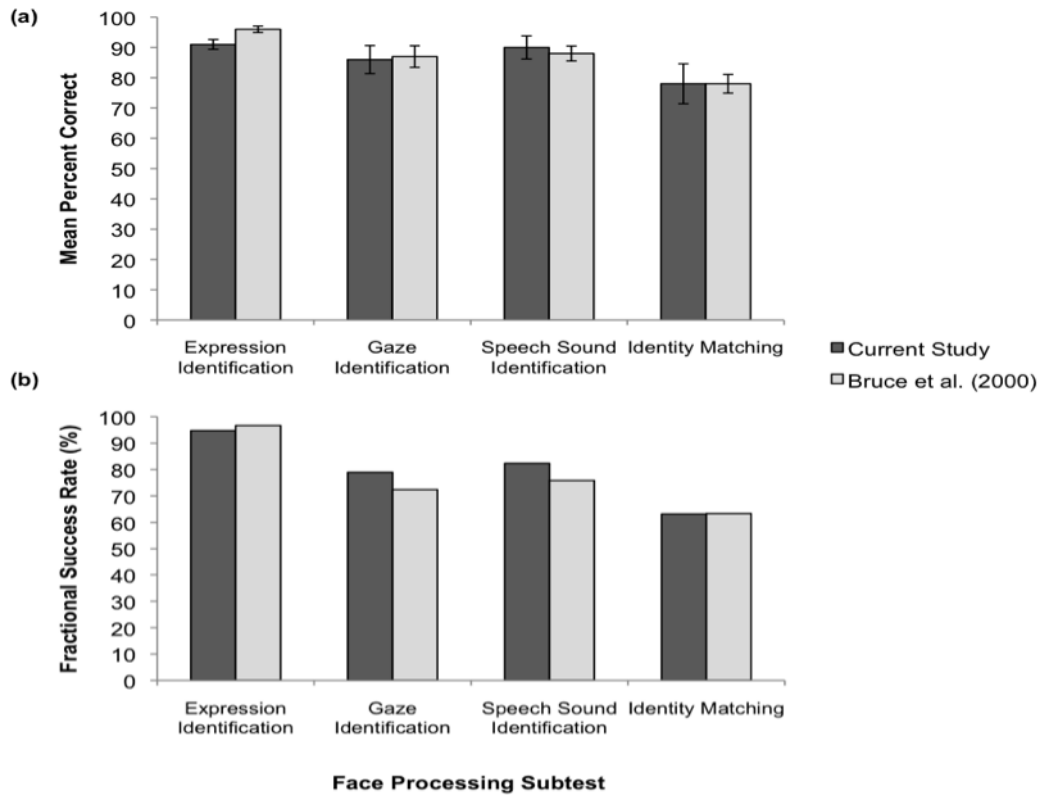
**Table 3.** Mean raw scores (standard deviations), range of scores and number of participants on the VABS and MSEL motor scales prior to the follow-up visit.

<b>Age</b>	<b>Measure</b>			
	<b>VABS</b>		<b>MSEL</b>	
	<i>Gross Motor</i>	<i>Fine Motor</i>	<i>Gross Motor</i>	<i>Fine Motor</i>
<i>9 months</i>	15.50 (8.62) Range: 6 - 43 N = 20	11.84 (2.69) Range: 6 - 16 N = 19	12.47 (1.87) Range: 9 - 16 N = 19	13.05 (2.17) Range: 10 - 19 N = 19
<i>40 months</i>	62.50 (9.16) Range: 42 - 78 N = 20	37.10 (9.30) Range: 25 - 55 N = 20	n/a	34.74 (5.97) Range: 22 - 46 N = 19

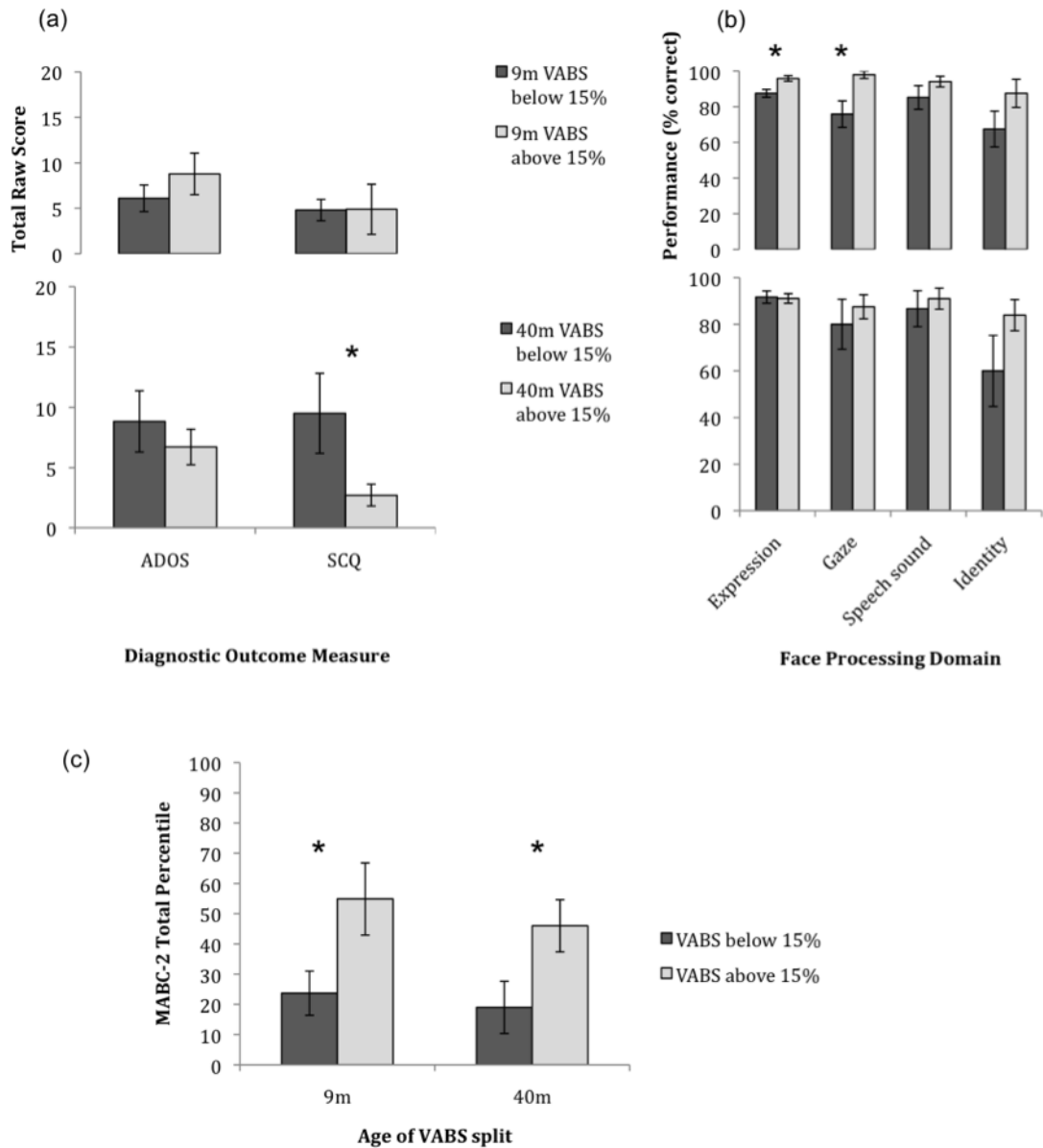




**Figure 1.** Examples of trials from each subtest in the face processing battery adapted from Bruce et al. (2000). (a) Expression Identification: identify which person is ‘sad’. (b) Speech Sound Identification: identify which person is saying ‘oo’. (c) Gaze Identification: identify which person is ‘looking at you’. (d) Identity Matching: identify which of the pictures at the top of the screen is the same person as the picture presented at the bottom of the screen.



**Figure 2.** Comparison of the current face processing outcomes and those of Bruce et al. (2000) using the same tasks. **(a)** Mean percent correct and standard error for each face processing subtest. **(b)** Fractional Success Rate (number of children scoring above chance) for each face processing subtest.



**Figure 3.** Scores of subgroups split on VABS Motor Composite scores at 9 months and 40 months for (a) diagnostic outcome measures, (b) face processing subtests and (c) MABC-2 total percentile. ‘VABS below 15%’ are classified as the ‘poor motor’ group, while ‘VABS above 15%’ are classified as the ‘typical motor group’. Error bars show standard error of the mean, and \* signifies a significant difference between groups ( $p < .05$ ).