## ORIGINAL PAPER

# Sex-typical Play: Masculinization/Defeminization in Girls with an Autism Spectrum Condition

Rebecca C. Knickmeyer · Sally Wheelwright · Simon B. Baron-Cohen

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**Abstract** We tested the hypothesis that prenatal masculinization of the brain by androgens increases risk of developing an autism spectrum condition (ASC). Sex-typical play was measured in n = 66 children diagnosed with an ASC and n = 55 typically developing age-matched controls. Consistent with the hypothesis, girls with autism did not show the female-typical play preferences, though this was only seen on non-pretence items. Boys with autism showed a preference for male play on non-pretence items, in keeping with their sex. Girls with autism engaged in more pretend play than boys with autism, suggesting that pretence is relatively more protected in females with autism. We conclude that play preference studies in ASC provide partial support for the fetal androgen theory.

**Keywords** Autism · Sex differences · Play · Fetal testosterone

## Introduction

The 'extreme male brain' (EMB) states that autism is an exaggeration of typical sex differences in empathizing and

R. C. Knickmeyer · S. Wheelwright · S. B. Baron-Cohen Department of Psychiatry, Autism Research Center, University of Cambridge, Douglas House, Cambridge, UK

R. C. Knickmeyer (⋈)

Department of Psychiatry, University of North Carolina-Chapel Hill, CB #7160, 7023 Neurosciences Hospital, Chapel Hill, NC 27599–7160, USA

e-mail: rebecca\_knickmeyer@med.unc.edu

Present Address:
R. C. Knickmeyer
Neurodevelopmental Disorders Research Center, University of
North Carolina, Chapel Hill, NC, USA

systemizing (Baron-Cohen 2002). The EMB originally defined the 'male' and 'female' brain purely psychometrically, but it has since been suggested that specific aspects of autistic neuroanatomy, such as increased brain volume (Hazlett et al. 2005) and accelerated growth of the amygdala during childhood (Hazlett et al. 2005; Sparks et al. 2002) may also be extremes of typical male neuroanatomy (Baron-Cohen et al. 2005).

The EMB does not specify which biological mechanisms shape the sex differences in cognition and neuroanatomy that may be altered in autism. In contrast, the fetal androgen theory specifically hypothesizes that increased levels of prenatal androgens produce excessive masculinization of the brain and thereby increase the risk for autism spectrum conditions (Baron-Cohen et al. 2004). Experiments in animals leave no doubt that androgens, especially testosterone produced in fetal and neonatal life, act on the brain to produce sex differences in neural structure and function (De Vries and Simerly 2002; Simerly 2002).

Evidence in support of the prenatal androgen hypothesis comes primarily from studies of typical children. These studies relate amniotic testosterone levels to behaviors such as eye contact, vocabulary development, social interaction, and empathy that in the extreme are diagnostic features of ASC (Lutchmaya et al. 2002a, b; Knickmeyer et al. 2005; Chapman et al. 2006). Further evidence comes from studies exploring physical indications of high testosterone exposure, such as the 2D:4D ratio, delayed menarche, and polycystic ovarian syndrome in females with ASC (Knickmeyer et al. 2006; Ingudomnukul et al. 2007; Lutchmaya et al. 2004). A final source of evidence comes from studies of autistic traits in rare medical syndromes such as congenital adrenal hyperplasia in which fetal testosterone levels are elevated (Knickmeyer et al. 2006; see Baron-Cohen et al. 2005; and Knickmeyer and Baron-Cohen 2006 for reviews).



An alternative method for testing whether individuals with autism show evidence of high testosterone exposure is to examine sex-typical behaviors known to be related to prenatal testosterone. Such sex-typical behaviors have not been extensively examined in people with an ASC. One study reported higher levels of tomboyism in girls with an ASC (Ingudomnukul et al. 2007). Sex-typical play represents one of the largest and best replicated sex differences studied in typical populations. Boys engage in more rough play and athletic games and prefer construction and transportation toys, while girls engage in more play-parenting and prefer play with dolls and kitchen supplies (DiPietro 1981; Humphreys and Smith 1984; Pellegrini and Smith 1998). Females exposed prenatally to increased adrenal androgens as a consequence of congenital adrenal hyperplasia or to exogenous androgenic progestins show increased male-typical play (Berenbaum and Hines 1992; Berenbaum and Snyder 1995; Dittman et al. 1990; Nordenstrom et al. 2002; Servin et al. 2003; Zucker et al. 1996). Males exposed to abnormally low levels of testosterone as a result of exposure to environmental compounds (such as polychlorinated biphenyls) that interfere with the production and action of testosterone show reduced maletypical play (Vreugdenhil et al. 2002).

In the present paper we report the first study examining sex-typical play in children with autism spectrum conditions. Our study extends previously studied relationships between sexually dimorphic human behavior and autism to include sex-typical play and tests the hypothesis that prenatal masculinization of the brain is a risk factor for ASC. Our primary interest was in females with ASC, whom we predicted to show either an absence or reversal of the female-typical preferences in terms of their games. We included males with autism for comparison, but results must be treated with caution for the following two reasons: First, whilst there is abundant evidence that high fetal testosterone exposure masculinizes play preferences in females, it is not clear that high fetal testosterone exposure in males hypermasculinizes play. The effect of fetal testosterone on play in males may thus be non-monotonic. That is, increasing exposure may produce increasing masculinization up to a certain dose, but increase beyond this dose may cause a reversal toward the original state (demasculinization). Hines and Kaufman (1994) reported that boys with congenital adrenal hyperplasia showed decreased rough and tumble play compared to control males, possibly demonstrating a non-monotonic effect. Secondly, the preference for sex-typical games is very strong (d = 3.45 and d = 3.49for boys and girls respectively, on the Children's Play Questionnaire, the questionnaire used in this study). This creates a great deal of scope for seeing defeminization/ masculinization effects in females, but a more limited scope for seeing hyper-masculinization in males.



#### Methods

**Participants** 

We tested two groups of children

ASC Group. n = 66 children (20 female, 46 male) diagnosed with an ASC participated in the study. All families were members of the Cambridge Autism Research Centre Volunteer database. Families join the database through the Centre's website (http://www.autismresearchcentre.com). Individuals are directed to the website by charities such as the National Autistic Society (UK), specialist clinics carrying out diagnostic assessments, and adverts in newsletters/ web-pages for people with autism. To qualify as a volunteer a diagnosis must have been made by a suitably qualified professional, such as a psychiatrist or psychologist, using established ICD-10 or DSM-IV criteria for ASC (APA 1994; ICD-10 1994). As our primary interest was in females (for reasons explained earlier), we began by contacting all families in the database who had a daughter, between the ages of 5 and 14, diagnosed with an ASC. The families of 76 girls with an ASC were initially contacted, and 20 families returned the questionnaire described below (a response rate of 26%). Although low, this response rate is within the expected range for postal surveys. We then contacted families of 102 parents of sons with an ASC who were similar to the participating girls in age range and diagnosis. 46 families with a diagnosed boy returned the questionnaire (a response rate of 46%). Of the females, specific diagnoses were available for 19. Seven were diagnosed with Asperger Syndrome (AS), 8 with autism, 1 with high-functioning autism (HFA), 1 with atypical autism, and 1 with pervasive developmental disorder not otherwise specified (PPD-NOS). Of the males, specific diagnoses were available for 41. Twelve were diagnosed with AS, 27 with autism, 1 with HFA, and 1 with PDD-NOS. Rates of specific diagnoses did not differ between responders and non-responders.

Control Group. n = 55 typically developing children (24) female, 31 male), whose mothers had previously completed the CPQ as part of a study of fetal testosterone and sextypical play (Knickmeyer et al. 2005) acted as controls. These children were also enrolled in a longitudinal study of amniotic testosterone and development. All their mothers had decided/been advised to have an amniocentesis due to late maternal age or other factors that increase the risk of fetal abnormality. Children who have had amniocentesis show no evidence of decreased well-being or impaired brain development (Finegan et al. 1996) and any child whose medical records indicated ill health at birth, for example requiring long stays in the SCBU (Special Care Baby Unit), was excluded from the study. Maternal age could affect the degree to which mothers encourage and expect children to show sex-typical play, but maternal age was not a significant predictor of sex-typical play in the control sample (Knickmeyer et al. 2005).

# Matching

Participants ranged in age from 4.58 to 14.17 decimal years (mean (SD) = 7.92 (3.00) for the entire group). Mean (SD) ages were 9.92 (2.06) for girls with ASC, 5.16 (0.28) for control girls, 10.42 (2.12) for boys with ASC, and 5.08 (0.28) for control boys. Children with ASC were significantly older than the control group, t (67) = -19.57, p < 0.001, however the CPQ specifically asks parents to report their child's preferences at 5 years of age. In this sense both groups were matched for the age at which parents described their child on the CPQ. Both groups were matched for sex, parental occupation (with a similar range of professional and manual occupations), handedness, parental educational level (years beyond high-school education, and proportion of parents with a university degree), and ethnicity.

#### Procedure

The families were sent invitation letters along with consent forms and a copy of the Children's Play Questionnaire (CPQ) (shown in Appendix) (Knickmeyer et al. 2005), which is an updated and shortened version of the Child Game Participation Questionnaire (Bates and Bentler 1973). Parents were told that this was a study of play, but they were not told that it was a study of sex-typical play. The CPQ includes 10 masculine items, 10 feminine items, and 8 neutral items. The sex-neutral items are included to prevent biased answering which might occur if parents knew the test was designed to assess sex-typical play. For each game, mothers indicate their child's interest (at age 5) on a Likert scale (1 = not at all interested to 5 = very interested).

# Scoring

A female-typical score is calculated by adding together a child's scores over all female-typical items (items 1,4,8,11,13,17,21,24,26,28). For each item, a response of 1 is scored as 0, a response of 2 is scored as 1, a response of 3 is scored as 2 and so forth. A male-typical score is calculated by adding together a child's scores over all male-typical items in the same way (items 2,5,6,9,12,14,16,19,22). The male- and female-typical scores have a possible range from 0 to 40. Both scales show extremely large sex differences in typical children (d = 3.2 and d = 3.6 for female- and male-typical scores respectively). Both boys

and girls show a strong preference for sex-typical items (d = 3.45 and d = 3.49 for boys and girls respectively). All items classified as female- and male-typical show strong sex differences in the predicted direction, correcting for multiple comparisons (Knickmeyer et al. 2005).

A potential concern with any measure of sex-typical play applied to children with ASC is that many such games require pretence (e.g., pretending to be a school teacher or a cowboy). Children with ASC participate in less pretend play than typical children (Baron-Cohen 1987; Jarrold et al. 1993). This creates the possibility that a real change in preference for sex-typical play might be concealed by an overall drop in scores due to reduced participation in games that require pretence. Therefore, we classified all sex-typical items based on whether they required pretence. The following items were classed as requiring pretence (1,2,4,5,6,11,12,17,19,26,28); neutral items were not included in the analysis; all other items were classed as non-pretence.

## Statistical Analyses

A repeated measures analysis was run with sex-typicality of item (male, female) and pretence of item (pretence, nonpretence) as within subjects factors and child's sex (male, female) and diagnosis (ASC, typical control) as between subjects factors. The primary outcome of interest is the pattern of preference, not absolute scores. Consider the following hypothetical case involving 2 boys. The first boy has a score of 40 on male-typical items and 20 on femaletypical items; the second boy has a score of 20 on maletypical items and 0 on female-typical items. Although the first boy has a higher absolute score on male-typical items, it would not be appropriate to label him as more maletypical than the second boy, given that each shows the same degree of preference for male- as opposed to femaletypical items. In addition, because children with an ASC have difficulty with social interaction and a higher level of repetitive behavior and restricted interests, their opportunities to engage in a range of games may be reduced. We therefore expected that children with autism would have reduced scores on all subscales. This is supported by our finding that the children with autism had slightly lower scores on neutral items when compared to typical controls, t(113) = 8.10, p < 0.001.

Our primary prediction, based on the EMB theory, was that females with an ASC would show a reduced preference for female-typical items over male items on games that did not require pretence. This could be manifested as an elimination of any preference for female-typical versus male-typical items, or a reversal of preference. We also predicted that for games that did not require pretence,



males with an ASC would show a preference for male-typical items over female-typical items, in line with their sex. We did not necessarily predict a hyper-masculinization of preference for the reasons discussed above. We also predicted that for games that did require pretence, both males and females with an ASC would score extremely low, regardless of sex-typicality of item. This would resemble an elimination of sex-typical preference in both males and females with an ASC, but such an interpretation would be inappropriate if pretend play was essentially absent in our children with an ASC.

#### Results

Table 1 shows the mean, standard deviation, and range for male-typical pretence items, male-typical non-pretence items, female-typical pretence items, and female-typical

**Table 1** Mean, standard deviation, and range for male-typical pretence items, male-typical non-pretence items, female-typical pretence items, and female-typical non-pretence items

	Mean	SD	Range	N
Male-typical prete	ence			,
Controls				
Boys	13.8	4.95	4-20	31
Girls	2.82	3.07	0–9	22
Diagnosed				
Boys	1.89	3.83	0–14	46
Girls	1.20	2.22	0–9	20
Male-typical non-	pretence			
Controls				
Boys	14.4	3.02	10-20	31
Girls	4.77	3.61	0–12	22
Diagnosed				
Boys	8.93	4.54	0–18	46
Girls	5.30	4.87	0–17	20
Female-typical pr	etence			
Controls				
Boys	5.03	3.02	0–11	31
Girls	15.6	4.67	4–20	22
Diagnosed				
Boys	1.07	1.85	0–8	46
Girls	6.96	5.42	0–17	20
Female-typical no	on-pretence			
Controls				
Boys	5.77	3.17	1–15	31
Girls	14.9	3.82	5-20	22
Diagnosed				
Boys	1.77	2.64	0–9	46
Girls	7.44	6.26	0–20	20

**Table 2** Direction and effect size of preference for male- and female-typical games in children with and without ASC

		Direction of preference	Effect size (Cohen's d)
Non-pretence	Control boys	M > F	2.7
	Control girls	F > M	2.7
	Diagnosed boys	M > F	1.9
	Diagnosed girls	M = F	0.38
Pretence	Control boys	M > F	2.1
	Control girls	F > M	3.2
	Diagnosed boys	M = F	0.27
	Diagnosed girls	F > M	1.4

non-pretence items for each group (control males, control females, diagnosed males, and diagnosed females). The repeated measures analysis showed a significant 4-way interaction between pretence of item, sex-typicality of item, diagnosis, and sex of child, F (1,116) = 6.01, p = 0.02. Further analysis concentrated on exploring this interaction rather than focusing on the main effects and lower level interactions. Table 2 shows whether each group showed a significant preference for male- versus female-typical items (with pretence and non-pretence items displayed separately) and the effect size of this difference. t-tests were used to explore relationships in greater detail.

As expected, controls showed a significant preference for sex-typical games; t(60) = 13.55, p < 0.001 and t(42) = 11.58, p < 0.001, for boys and girls respectively. In keeping with our primary prediction, girls with ASC did not show a preference for female-typical items on non-pretence items, t (19) = -1.30, p = 0.21. Although girls with ASC did not show a strong preference for male-typical items, the lack of any preference for female-typical items suggests that girls with ASC are masculinized/defeminized compared to typical girls (who show an extremely strong preference for female items) in games where pretence is not involved.

Boys with autism showed a strong preference for maletypical non-pretence items as opposed to female-typical non-pretence items, in line with their biological sex, t (45) = 11.8, p < 0.001. Also in keeping with predictions, individuals with ASC (boys and girls combined) had a strong preference for items that did not involve pretence (male and female items combined), t (65) = -9.71, p < 0.001. Boys with an ASC had an average score of less than 2 on both male-typical pretence and female-typical pretence items. Although boys with an ASC did not show a preference for male-typical pretence items over female-typical pretence items, it seems more appropriate to interpret this as a floor effect on games requiring pretence than as evidence for demasculinization or feminization in boys with an ASC.



An unexpected finding was that girls with an ASC had higher scores on games involving pretence than boys with an ASC, t(64) = 4.06, p < 0.001, suggesting that pretence is to some extent relatively 'preserved' in girls with ASC. In addition, girls with ASC showed a significant preference for female-typical pretence items, in comparison to male-typical pretence items, t(19) = -4.96, p < 0.001, in keeping with their biological sex, a finding not predicted by the EMB.

## Discussion

The current study examined whether sex-typical play showed masculinization/defeminization in children with ASC. Studies of individuals exposed to abnormally high or low levels of fetal testosterone as a result of genetic disorders (notably congenital adrenal hyperplasia) and exposure to synthetic progestins have indicated a role for fetal testosterone in the development of sex typical play. Thus, the current study also indirectly tests the hypothesis that masculinization of the brain as a result of elevated fetal testosterone exposure increases the risk of ASC.

For games that did not require pretence, girls with ASC did not show a preference for female-typical items as would be predicted by their biological sex. Girls exposed prenatally to increased endogenous adrenal androgens (Berenbaum and Hines 1992; Berenbaum and Snyder 1995; Hines and Kaufman 1994) or to exogenous androgenic progestins (Ehrhardt et al. 1968) also exhibit a reduced preference for sex-typical play. The similarity of our findings to those reported for girls with known exposure to elevated fetal testosterone levels is consistent with the hypothesis that greater exposure to or sensitivity to fetal testosterone alters the brain is such a way as to contribute to the development of ASC.

An alternative interpretation is that children with an ASC may be less sensitive to societal factors that encourage sex-typical play and discourage sex-atypical play. Parents do encourage their children to use sex-appropriate toys (Lytton and Romney 1991) and typical children recognize 'appropriate' toys and roles at an early age and emulate the behavior of same-sex models in preference to opposite-sex ones (Greif 1976). However, if this were the explanation for our finding in females with an ASC we might expect males with autism to show no preference for male typical non-pretence items over female-typical non-pretence items. This was not the case in our study; males with an ASC showed the expected preference for male non-pretence items as opposed to female non-pretence items.

Although our primary interest at the outset was in games that did not require pretence (as we expected both males and females with autism to score so low on the pretence items that a meaningful examination of sex-typical preferences was not possible), examination of items requiring pretence also revealed an interesting pattern. While males with ASC showed almost no interest in games requiring pretence, as predicted, girls with ASC actually had higher scores on games involving pretence than males with ASC. Girls with ASC showed a significant preference for female pretence items as opposed to male pretence items, in keeping with their biological sex. This finding was not predicted by the fetal androgen theory.

An alternative theory explaining the sex differences seen in ASC (and compatible with the EMB) has been proposed by Skuse and colleagues. Inspired by their work on Turner syndrome (X monosomy), they suggest there is an imprinted locus on the short arm of the paternally derived X-chromosome that is responsible for the female advantage in socio-cognitive abilities and protects them from developmental conditions such as autism (Creswell and Skuse 1999; Skuse 2000; Skuse et al. 1997; Thomas et al. 1999). If sex differences in play are an index of fetal testosterone exposure but the features of autism that the EMB seeks to explain are related to the imprinted locus, one would hypothesize that the preference for sex-typical play would be relatively unaffected by autism. Whilst this could explain our results for pretence items in females, it does not explain those we observed for non-pretence items.

It is also interesting to speculate why males were more severely affected than females by pretence. One possibility is that the boys in our ASC group had more severe forms of ASC. No quantitative measure of severity or IO was available. However, we did have specific diagnoses for many of the children in the sample. There are 4 recognised subgroups on the autistic spectrum: classic autism, highfunctioning autism (HFA), Asperger Syndrome (AS), and pervasive developmental disorders not otherwise specified (PDD-NOS). These subgroups vary in severity of symptoms, with classic autism being more severe than AS, HFA and PDD-NOS, if severity is measured in terms of conspicuousness. Approximately half of the girls in our sample were diagnosed with classic autism, while the other half were diagnosed with AS, HFA, and PDD-NOS. Within the boys, relatively more children were diagnosed with classic autism than with AS, HFA, and PDD-NOS. However, the difference was not significant when tested with a Fisher's exact test, p = 0.16.

Alternatively, pretence could be some-how 'protected' or relatively more preserved in females with ASC. This protection could result from the social environment. Parent-daughter dyads, particularly mother-daughter dyads, are more likely to engage in pretence play than parent—son dyads (Lindsey and Mize 2001; Lindsey et al. 1997; Tamis-LeMonda and Bronstein 1991). The directionality of this relationship is not clear, but if parents of children with



ASC do attempt to engage daughters in pretend play more often than they do sons with ASC, this could provide a richer environment in which girls with ASC could learn to engage in pretend play. Protection could also result from biological factors, including hormonal and direct genetic effects. This warrants further study.

When interpreting the results of this study, it is important to keep in mind its limitations. First, this is a postal study from a volunteer database. While this allowed us to collect a fairly large sample size (especially of girls with ASC, who are much rarer than males with ASC), it meant we could not confirm IQ or language ability in this group. Second, the study includes children with comorbid diagnoses. As discussed above, we did attempt to include similar numbers of males and females with autism and less severe diagnoses such as AS, but the sample size was not sufficient to test if the pattern of results differed as a function of diagnostic subgroups. Third, the control group was significantly younger than the ASC group. Although all mothers were asked to report their child's behavior at age 5, and in this sense their child's age described on the questionnaire is directly comparable, mothers of children with ASC had to recall their child's earlier behavior, while parents of controls reported present behavior. This could have affected parental recall accuracy, a factor that could be checked in future studies.

# Conclusion

The current study explored the association between sextypical play behavior and ASC. Our analysis provides some support for the fetal androgen hypothesis, in that girls with ASC did not show a preference for female-typical play on games that did not require pretence. These results mirror the finding that among women with ASC there are increased rates of 'tomboyism' by self-report about their childhood interests and activities (Ingudomnukul et al. 2007). Boys with ASC showed the expected male preference for male-typical play when games did not require

pretence. In contrast, girls with ASC did show a sex-typical preference for female games when games did involve pretence, a finding not predicted by the androgen theory. Further examination of other sex-typical behaviors (both those linked to androgen and those linked to sex chromosome effects) in ASC is needed. Finally, our results raise the intriguing possibility that pretence is relatively more 'protected' in girls with ASC than in boys with ASC. Whether this simply reflects better imitation of typical play behavior by girls with ASC, in an effort to try to appear more 'normal', or reflects a more intact capacity to pretend, are possibilities that also require further experimental study. We conclude that the study of play preferences in ASC provides partial support for the fetal androgen theory, which warrants direct testing via amniocentesis in sufficiently large samples than has hitherto been possible. Such a study is underway in our lab. Such studies will ultimately need to address if fetal testosterone levels are elevated in ASC, and/or if androgen receptor sensitivity is increased. Such studies have the potential to explain the developmental neurobiology of ASC and why more males than females develop ASC.

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## Appendix: The children's play questionnaire

Child's name —			——Your name———		
Date	of	birth	Today's	date	

In this section you will find a list of games or activities that children may play. Read through the list and indicate how interested your child would be in each game by ticking the right box. 1 means not at all interested, 3 is somewhat interested, and 5 is very interested

1 not	2	3 some-	4	5 very
at all		what		interested
interested		interested		

Playing with Barbie-type dolls\*
Pretending to be a soldier\*\*
Looking at picture books
Role-playing domestic activities (eg cooking, cleaning, bathing)\*
Pretending to be a superhero\*\*
Playing with toy guns or other weapons\*\*
Playing with stuffed animals



#### Table a continued

1 not	2	3 some-	4	5 very
at all		what		interested
interested		interested		

Playing dress up (fashion/jewelry)\*

Playing with toy cars and trucks\*\*

Riding on tricycles/bicycles

Role-playing family relationships (eg parenting/marriage)\*

Pretending to be an astronaut (space-man) or explorer\*\*

Skipping rope or skipping\*

Playing with toy tools\*\*

Swimming

Playing with electric trains\*\*

Playing school (pretending to be a teacher)\*

Playing on swings

Playing Cowboys and Indians or similar\*\*

Playing on see-saws

Dancing\*

Playing with toy airplanes\*\*

Play fighting/wrestling\*\*

Playing with hair (e.g., brushing someone else's hair)\*

Watching cartoons

Playing tea-parties\*

Playing board-games (e.g.,Ludo, Snakes and Ladders)

Playing with baby dolls\*

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<sup>\*</sup> Female-typical games

<sup>\*\*</sup> Male-typical games

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