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Executive function in parents of children with autism

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ABSTRACT

Background. Previous studies have shown that individuals with autism show impaired performance on tests of executive function (Ozonoff *et al*. 1991, 1993; Hughes & Russell, 1993; Hughes *et al*. 1994). There is also strong evidence for genetic involvement in autism (see Rutter, 1991 for review). If executive dysfunction is a core impairment in autism, then similar impairments are hypothesized to exist in a subtler form among the parents of autistic children.

Methods. Forty parents of autistic children were compared with 40 parents of learning disabled children and 36 adults from unaffected families on three computerized tests of executive function. These tasks tapped attentional-shifting skills, visuospatial planning and working memory. Participants also received a computerized control test of spatial memory-span. In addition, the interviewer's initial impressions of family members were coded using a new 33-item questionnaire.

Results. A significant proportion of parents of autistic children (especially fathers) showed impaired executive function. By contrast, parents did as well as both comparison groups on a control test of spatial span, and on other 'non-executive' measures from the tasks, indicating that the autism group were as able and motivated as comparison groups. Interestingly, impairment of executive function was significantly correlated with the interviewer's pre-test impression of social abnormality among parents of autistic children.

Conclusions. The hypothesis that a significant proportion of parents of autistic children show impaired executive function was supported. Parents showed good memory ability, but relatively poor planning skills and attentional flexibility. The extent to which this is an inherent trait in family members, rather than a reflection of the difficulties involved in caring for an autistic child, remains to be examined.

INTRODUCTION

Autism is a rare but severe developmental disorder, first described by Kanner (1943) and characterized by a 'triad of impairments' including lack of social relatedness, poor communicative skills, and an absence of imaginative play coupled with repetitive stereotypic behaviour (Wing & Gould, 1979). Although much of Kanner's initial account is still considered remarkably accurate, some parts (such as the assumed normal intelligence of most children with autism) have been disproved, while other aspects remain the subject of controversy. One such area, of particular relevance to the current paper, is Kanner's (1943) description of the parents of autistic children as 'limited in genuine interest in people ... and showing a great deal of obsessiveness' (p. 250). Although other clinicians have also reported social abnormalities among parents of autistic children (Eisenberg, 1957; Creak & Ini, 1960; Wolff *et al*. 1988; Landa *et al*. 1992), several other researchers have reported that such findings are *not* more frequent in this group than among controls (summarized in

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McAdoo & DeMyer (1978); see also Gillberg *et al*. 1992).

Contradictory findings such as these raise two issues for debate. First, what is the relation between the peculiarities reported for parents of autistic children and autism itself? Early accounts that attributed a direct causal significance to social abnormalities in parents (most notoriously, Bettelheim's (1963) concept of the 'refrigerator mother') have since been robustly disproved (Cox *et al*. 1975; Cantwell *et al*. 1976). Indeed, in many recent studies, disturbance in parental behaviour is now recognized as a response to the stress of caring for an autistic child (e.g. Wolf *et al*. 1989). At the same time, the evidence for genetic involvement in autism is extremely strong (see Rutter, 1991 for a review). It, therefore, seems likely that while some of the unusual features reported for parents of autistic children will reflect the difficulties of raising a severely handicapped child, other features may represent a more subtle manifestation of autistic traits.

The second question concerns the extent to which the conflicting conclusions drawn in the above studies are artefacts of differences in methodology, and in particular, differences in the control groups and informants used (Piven *et al*. 1994). This possibility highlights the importance of systematic, objective and reliable assessments. Studies which meet these criteria have found differences among the close relatives of autistic children. In particular, several researchers have reported an elevated incidence of social abnormalities (Wolff *et al*. 1988; MacDonald *et al*. 1989; Landa *et al*. 1991, 1992) and language or reading problems (Bartak *et al*. 1975; August *et al*. 1981; Minton *et al*. 1982; MacDonald *et al*. 1989; Piven *et al*. 1990).

Further, the abnormalities found in the above studies appear conceptually similar to the difficulties experienced by individuals with autism. It is worth noting that all of these studies have focused on sociability and communication, the first two of the autistic triad of impairments. The third area that includes poor imaginative activity and repetitive, stereotyped behaviour has so far received very little attention. Recently however, interest in this area has been sparked by findings of marked deficits in people with autism in a closely associated domain, that of executive function.

'Executive function' is an umbrella term, typically associated with frontal-lobe functioning, and used to encompass the processes that underlie goal-directed behaviour, such as planning, working memory, inhibition of prepotent responses, and cognitive flexibility. Individuals with autism (of all ages and abilities) have been found to be impaired in each of these skills, performing poorly on tests of planning and attentional set-shifting (Ozonoff *et al*. 1991; McEvoy *et al*. 1993; Hughes *et al*. 1994; Hughes, 1996*a*), as well as on tasks that require the inhibition of perceptually triggered maladaptive responses (Hughes & Russell, 1993; Ozonoff *et al*. 1993; Hughes, 1996*b*) or self-monitoring and adaptive responses to external feedback (Prior & Hoffman, 1990). The predicted consequences of executive dysfunction include a marked difficulty in novel or ambiguous situations, but intact performance in routine or well-learned situations. These predictions fit well with many observed characteristics of autistic behaviour. In this paper, we shall consider whether similar impairments in executive function are also apparent among the parents of children with autism, and if so whether these deficits are associated with abnormalities in everyday interactional skills. As a caveat, executive dysfunction has been reported for a variety of developmental disorders (see Hughes *et al*. 1994), and this may tempt the sceptic to argue that any kind of brain dysfunction can produce impairments of executive control. It is therefore important to elucidate the exact nature of executive dysfunction in autism. For this reason, an information processing approach was adopted, using a set of computerized tasks to facilitate fine-level analysis of individual executive function performance.

METHOD

Participants

Recruitment of parents of autistic children

Families who had approached the Robert Debré hospital in the previous 2 years, and whose children met the selection criteria, were invited by letter to participate in the study. Criteria for inclusion were as follows: full diagnosis of autism in the proband child, by both DSM-III-R (American Psychiatric Association, 1987) and ICD-10 (World Health Organization, 1992) criteria; absence of concomitant medical con-

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Table 1. *Characteristics of the target children on which the two groups of parents were matched*

	Autistic children	Learning disabled children
Number of cases	24	27
Sex ratio M:F	18:6	18:9
Mean age in years (& range)	9·7 (4–18)	10·1 (4–16)
Mean IQ (S.D.)	56·5 (18·0)	54·7 (9·1)
Number of mothers seen	20	22
Number of fathers seen	20	18
Mean age for fathers (range)	41·5 (33–57)	42·9 (35–60)
Mean age for mothers (range)	40·0 (33–55)	41·0 (31–54)
Father's years of post-11 education (S.D.)	13·4 (2·9)	12·9 (2·0)
Father's occupational category		
I	7	6
II	16	20
III	10	8
IV	7	6

ditions; child living with both natural parents; parents native speakers of either French or English living within 2 hours of travel by public transport from Paris (though to supplement numbers, five families were contacted through an autism centre in Lyon). Forty-four families were contacted; eight could not be traced, six refused to participate, and in three families the proband child had since been institutionalized. After screening, the group comprised of the parents of 24 autistic probands. Although every effort was made to solicit the participation of both parents, for a few families one parent was repeatedly unavailable and so the final target group (hereafter referred to as the autism group) consisted of 20 mothers and 20 fathers.

Recruitment of parents of children with learning disabilities and normal controls

As for the autism group, all families contacted had approached the Robert Debré hospital in the previous 2 years. Most referrals were from educational authorities, and unlike the autistic group, this initial cohort was predominantly of low socio-economic status. Families were therefore selected on the basis of parental occupation and age to be as similar as possible to families of autistic probands. The following exclusion criteria were applied: divorce, records of disadvantaged living conditions, and known organic pathology. Sixty families were contacted, 11 refused to participate, 17 could not be traced and five were excluded because the target child was not in regular contact with his or her natural father. After screening the comparison group consisted of the parents of 27 children with moderate to severe learning disabilities. The final comparison group (hereafter referred to as the learning-disability group) included 22 mothers and 18 fathers. In addition, a second control group of adults with no incidence of psychiatric history or learning disability in their immediate families was recruited from staff and friends at the Robert Debré Hospital in Paris. This group included 18 men and 15 women.

Diagnosis and assessment of intellectual ability of autistic probands

Diagnosis was made by M.L. using the Autism Diagnostic Interview (Le Couteur *et al.* 1989; French translation by Leboyer, 1994). Intellectual ability was assessed using the WPPSI and WISC-R for more able probands (Centre de Psychologie Appliquée, French translation, 1974; 1981), and the McCarthy Scales (McCarthy, 1972) and the Psychoeducational Profile (Schopler & Reichler, 1979) for younger, less able probands.

Matching of probands and parents

Every effort was made to match probands for IQ, age and sex (see Table 1). The two groups were well matched for proband IQ ($t = -0{\cdot}41$, df = 50, NS) and proband age ($t = 0{\cdot}45$, df = 50, NS). There was no significant difference in the sex ratios in the two proband groups ($\chi^2 = 0{\cdot}42$, df = 1, NS). As shown below, families were also matched for parental age, education, and occupational level, as well as for maternal age at birth of proband.

Mean ages of participants

In the autism group, the mean age was 41·5 years for fathers and 40·0 years for mothers. In the learning disability group, the mean age was 42·9 years for fathers and 41·0 years for mothers. In the normal control group, the mean age was 41·7 years for fathers and 36·4 years for mothers. The mean maternal age at birth of the proband was 30·3 years in the autism group and 30·0 years in the learning disability group. Kruskal–Wallis tests showed no significant group

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difference in age for either mothers or fathers, and no difference in maternal age at birth of proband.

Occupation and educational level of participants

All except two of the fathers in the two clinical groups were in full-time employment, while only three mothers were in paid employment. Adult normal controls were recruited from the staff at the Robert Debré Hospital. Each group was relatively heterogeneous with respect to socio-economic status, coded on the *Catégorisation Internationale de Type de Profession* (CIST, 1988). A chi-square test showed no significant difference in the distribution of occupational categories ($\chi^2 = 5{\cdot}95$, df = 6, NS). Each participant's educational level was also recorded (see Table 1). A Kruskal–Wallis test showed no significant between-group difference in the years of education for participants.

Materials and procedure

Participants were presented with four multi-stage tasks from the Cambridge Neuro-psychological Test Automated Battery (CANTAB), a set of computerized paradigms run on an IBM compatible computer with a high-resolution colour monitor and touch-sensitive screen. These tasks included the 'IDED' set-shifting test of attentional flexibility, the Tower-of-London test of planning ability, a visual search test of spatial working-memory, and a control-test of spatial span that was not expected to tap executive function. Subjects were seated at a comfortable distance in front of the computer, and it was explained that they would have to respond to stimuli by touching the screen. Before the first test, subjects were given a simple 'motor screening task' in which they were asked to touch a flashing cross as it appeared on the screen. On satisfactory completion of this task, subjects were given the four tests described below.

Attentional flexibility task

The attentional flexibility task used was a computerized multistage set-shifting task, akin to the widely used Wisconsin Card Sorting Test (Grant & Berg, 1948), but with novel shapes introduced at each shifting stage. This 'total-change' paradigm enables transfer of learning within a dimension (intradimensional shifting) to be distinguished from transfer to a new dimension (extradimensional shifting). Hence the task is known as the intradimensional–extradimensional (ID/ED) shift task. Full procedural details can be found in Hughes *et al.* (1994).

The display panel for the IDED task showed four boxes, symmetrically positioned at the top, bottom, left and right of the screen. The two test stimuli were presented in two boxes, their position varying from trial to trial. Subjects were told: '*Can you see the two patterns? One of these patterns is the right one, and one of the patterns is wrong. Have a guess at which pattern is correct. If you choose the right one the computer will show the word "Correct" in green. If you get it wrong the computer will show the word "Wrong" in red. Keep choosing the pattern you think is correct. There is always a rule you can follow. Occasionally that rule will change and you have to be prepared for this. You can start now.*' The task was presented in nine stages. At each stage, the criterion for success was a run of six correct choices in 50 trials. if this criterion was not achieved the test was discontinued.

The first stage involved simple discrimination (sd) between a pair of pink shapes. Stage 2 was a simple reversal (sr) using the same pink shapes. At stage 3 ('compound discrimination-separate' (c/p-d) stage), a pair of white line patterns was introduced, so that the display consisted of two boxes with one pink shape and one white line and two empty boxes as before. The subject had simply to continue responding to the same pink shape as before. At stage 4, the rule remained unchanged, but for this stage and all subsequent stages, the white lines were *superimposed* on the pink shapes (cp-d), to prevent locational learning. At stage 5 (cp-r) the rule was reversed. Stage 6 (intra-dimensional shift (id-s)) was marked by a set of four new exemplars: success at this stage still depended on choosing one of the pink shapes. The rule was reversed at stage 7 (id-r). New exemplars were introduced to mark stage 8 (extra-dimensional shift (ed-s)), for which success depended on choosing the previously irrelevant white lines. At stage 9 (ed-r), the contingencies for the newly relevant dimension were reversed. Performances were rated by 'survival rate', trials-to-criterion, and errors-to-criterion.

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Planning task

This task is a variation on one developed by Shallice (1982), based on the 'Tower of Hanoi' problem. Full procedural details can be found in Hughes *et al*. (1994). Two sets of three coloured balls were presented, one in the top half of the screen and one in the bottom half. In each half of the screen there were three pockets, one that could hold three balls, one that could hold two balls, and one that was filled by just one ball. On each trial a red ball, a blue ball and a green ball were placed in predetermined positions in the pockets of each of the two displays. The subject was asked to rearrange the balls in the bottom display, in order to copy the goal arrangement in the top display. A ball could be 'moved' by first touching it and then touching an empty space in one of the other pockets. 'Illegal' moves, such as trying to place a ball high in a pocket when there was no other ball beneath it, or trying to remove a ball from under another, were carefully explained to the subject, and if attempted, evoked no response from the computer.

Subjects were instructed to examine the position of the balls at the beginning of each problem and attempt to solve it in the minimum number of moves. They were encouraged not to make the first move until they were confident that they could execute the full solution. The programme recorded the number of moves required by the subject, and measured the selection and movement latencies for both the first and subsequent moves. After six practice trials, the subject was given a set of 2-, 3-, 4-, and 5-move problems that corresponded exactly to those used in the original Tower of London test (Shallice, 1982).

For each test problem, a 'yoked control' provided baseline measures of motor initiation and execution times. These baselines could be subtracted from the corresponding latencies on the test-problems to estimate subject thinking times, both before initiating their first moves, and averaged over all subsequent moves on each problem.

Spatial working memory task

In this task, subjects were required to 'search through' a set of boxes on the screen by touching each one such that it 'opened up' revealing what was inside. The object was to collect 'blue tokens' hidden inside the boxes. Subjects were told that at any one time there would be a single token hidden inside one of the boxes. Their task was to search until they found it, at which point the next token would be hidden. The key instruction was that *once a blue token had been found within a particular box*, *then that box would never be used again to hide a token*. On each trial, the number of blue tokens to be found equalled the number of boxes on the screen. After four practice trials with two boxes, there were four test trials with each of four, six, and eight boxes. Performance was scored by the number of between and within search errors.

Spatial short-term memory task

This computerized version of the Corsi Block Tapping task (Milner, 1971) was chosen as a control task. Spatial short-term memory span was determined using a pseudo-random arrangement of nine white squares. Subjects were asked to watch carefully as a sequence of boxes changed colour for 3 s, and to remember the sequential order of the boxes which changed. Following one demonstration trial by the experimenter, the task began with a simple two-box sequence. After each successful trial, the next sequence was increased in length by one to a maximum of nine. During each trial, a number in the bottom left-hand corner of the screen indicated the length of the current sequence. After an incorrect attempt, another sequence of the same length was presented. The test stopped after the subject had failed three consecutive trials at any given level. The spatial short-term memory span was calculated as the final level at which the subject had successfully recalled at least one sequence of boxes.

Interviewer's impression of parents

After each initial contact visit to the families' homes, the interviewer wrote a short account of her impression of family members. These reports (written before the participants had performed the executive function tasks) were later recoded using a 33-item questionnaire developed for use with relatives by the MRC Child Psychiatry Unit (A. Bailey & M. Rutter, personal communication). The questionnaire had a three-point scale for each item, and included items such as 'abnormal eye-contact', and

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'peculiarities in volume, rate, or intonation of speech'. Since the interviewer was not blind to the diagnostic status of the proband child, these rating scales were not used to compare parents in the two clinical groups, but did provide a useful means of relating performance on the tasks to everyday interactional behaviour.

RESULTS

Attentional flexibility task

Success rates on each stage of the attentional set-shifting task are shown for each group in Fig. 1. There was no difference in group performance for the first seven stages of the task, indicating that all three groups were equally attentive and motivated. However, at the critical 'extradimensional shift' (ed-s) stage, only 20/40 parents in the autism group (nine mothers and 11 fathers), as compared with 37/40 parents in the learning disability group (20 mothers and 17 fathers), and 29/33 control subjects (14 mothers and 15 fathers) were successful. Likelihood ratio analysis showed a highly significant difference between the autism group and the learning disability group, $\chi^2(1, 79) = 19{\cdot}2$, $P < 0.0001$, as well as between the autism group and the control group, $\chi^2(1, 72) = 12{\cdot}6$, $P < 0.0005$. There was no difference in success rate between the learning disability group and the control group, $\chi^2(1, 79) = 0{\cdot}44$.

A three-way ANOVA (group × sex × shift) for trials-to-criterion showed significant main effects of shift (ed-shift more difficult, $F(1, 112) = 114{\cdot}4$, $P < 0.0001$); group (autism group worse, $F(2, 112) = 6{\cdot}5$, $P < 0{\cdot}005$) and sex (males more successful than females, $F(1, 112) = 5{\cdot}7$, $P < 0{\cdot}05$). However, Tukey's *post-hoc* comparisons showed no 'honest significant difference' in trials-to-criterion for any two groups. On the more sensitive measure of 'errors-to-criterion', the same three-way ANOVA showed significant main effects of shift ($F(1, 112) = 130{\cdot}2$, $P < 0{\cdot}0001$), group ($F(2, 112) = 6{\cdot}3$, $P < 0{\cdot}005$) but not sex ($F(1, 112) = 3{\cdot}6$). Here, *post-hoc* comparisons with Tukey's test showed that the autism group made significantly more errors than normal controls.

Mean response latencies at each shift were log-transformed for ANOVA and analysed in the same way as the error/trial to criterion scores. There was no significant effect of either group or stage. These results are not reported due to lack of space.

Planning task

As in Hughes *et al*. (1994), problems were divided into two levels: 'easy' (2- and 3-move) and 'difficult' (4- and 5-move) problems. A good measure of performance on this task is the proportion of 'perfect' solutions (problems solved in the minimum number of moves). Nearly all subjects obtained perfect solutions for all the 2- and 3-move problems. On the 4- and 5-move problems, the mean percentage of perfect solutions (and standard errors) was 49·7% (2·7) for the autism group, 55·3% (3·1) for the learning disability group, and 65·5% (3·2) for the control group. A three-way ANOVA (group × level × sex) on this measure showed strong effects of both group ($F(2, 450) = 10{\cdot}6$, $P < 0{\cdot}001$) and level ($F(1, 450) = 503{\cdot}6$, $P < 0{\cdot}001$), but no effect of sex ($F(1, 450) = 0{\cdot}0$). As for the 'number of extra moves' measure, the interaction between group and level was also significant ($F(2, 450) = 3{\cdot}3$, $P < 0{\cdot}05$). Tukey's test showed that for both mothers and fathers the autism group obtained perfect solutions to significantly fewer 4- and 5-move problems than the control group.

Response latencies and thinking times for the three groups at each level were log-transformed to reduce skewness before ANOVA tests were made. Two-way ANOVAs (group × level) showed a significant effect of level ($F(1, 226) = 71{\cdot}5$, $P < 0{\cdot}001$), but no effect of group ($F(2,226) = 0{\cdot}17$, NS) on initial motor times. Similar results were obtained for initial thinking times: effect of level ($F(1, 226) = 71{\cdot}6$, $P < 0{\cdot}001$); effect of group ($F(2,226) = 0{\cdot}5$, NS). Subsequent movement and thinking times were averaged over the number of moves taken by participants on each problem, before summing over problem level. For subsequent motor times, a two-way ANOVA indicated an effect of level ($F(1,226) = 406{\cdot}3$, $P < 0{\cdot}001$) and of group ($F(2,226) = 4{\cdot}0$, $P < 0{\cdot}025$). For subsequent thinking times the effect of level was significant ($F(1,226) = 37{\cdot}0$, $P < 0{\cdot}001$) but the effect of group was not ($F(2,226) = 2{\cdot}4$). For all of these ANOVA tests, there were no significant interaction terms between any main effects.

Finally, although the above measures are useful in assessing performance at a group level

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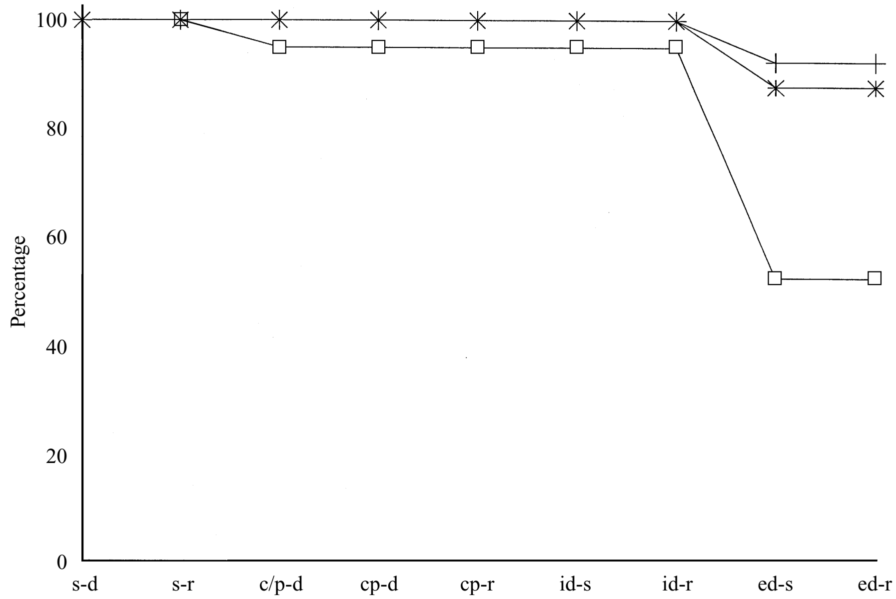

FIG. 1. The percentage of parents in each group (□, autism; +, learning disability; ✳, control) passing each stage of the attentional shifting task.

Table 2. *Summary of group performances on each task* (*means and standard deviations*)

Task	Group…	Autism (A) Mean	Autism (A) (S.D.)	Learning disability (LD) Mean	Learning disability (LD) (S.D.)	Normal control (NC) Mean	Normal control (NC) (S.D.)	Test statistic *F*-value	Test statistic *P*	Group difference*
IDED (ED-s)	Trials to criterion	32·45	(18·40)	21·35	(13·11)	18·52	(14·10)	8·40	0·001	A > LD = NC
	Errors to criterion	14·29	(9·73)	8·75	(7·30)	7·82	(8·43)	6·20	0·005	A > LD = NC
Spatial span	Max. spatial span	5·63	(1·23)	5·25	(1·19)	6·21	(1·39)	5·25	0·01	NC > LD
Spatial working, memory	Between search, errors	34·45	(20·32)	29·18	(17·86)	20·24	(2·60)	5·68	0·005	A > NC
Tower of London (4 and 5 moves)	Number extra moves	1·78	(0·18)	1·71	(0·16)	1·24	(0·81)	3·04	0·05	—
	Solutions correct (%)	49·08	(16·82)	55·31	(19·38)	65·53	(18·23)	6·86	0·005	A = LD > NC

* Assessed by Tukey's test for 'honestly significant difference', $P < 0{\cdot}05$.

(Table 2), they do not reveal how individuals within a given group are performing. To address this issue, two criteria for success on the task were adopted: a lenient measure (used in previous studies with autistic children) of perfect solutions on half or more of the 4- and 5-move problems; and a more stringent measure (more appropriate to the adult populations in this study) of perfect solutions on more than half of the 4- and 5-move problems. The lenient criterion was achieved by 70% of mothers and 65% of fathers in the autism group; 68% of mothers and 83% of fathers in the learning disability group; 80% of mothers and 94% of fathers the control group. There was no difference between mothers, but the difference between fathers did approach significance (for mothers: $\chi^2 = 0{\cdot}67$, df = 2, $N = 57$, NS; for fathers $\chi^2 = 5{\cdot}35$, df = 2, $N = 56$, $P < 0{\cdot}07$).

The second and more stringent criterion was achieved by 45% of mothers and 25% of fathers in the autism group; 41% of mothers and 61% of fathers in the learning disability group; and 73% of mothers and 78% of fathers in the control group (see Fig. 2). Again, mothers in the three groups did not differ on this measure

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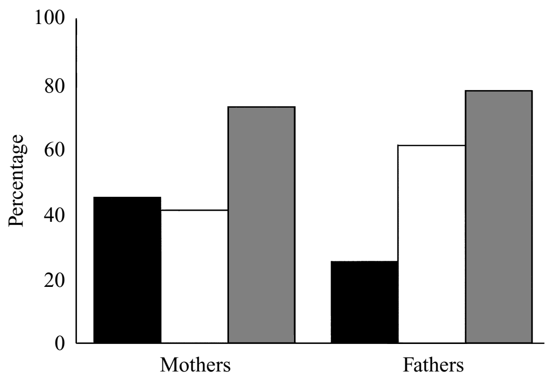

FIG. 2. The percentage of mothers and fathers in each group (■, autism; □, learning disability; ▨, control) solving more than half of the 4- and 5-move planning problems in the minimum number of moves.

($\chi^2 = 3{\cdot}38$, df = 2, $N = 57$, NS). However the difference between the number of successful fathers in each group was highly significant ($\chi^2 = 10{\cdot}9$, df = 2, $N = 56$, $P < 0{\cdot}005$). Fewer fathers in the autism group achieved criterion than in the learning disability group ($\chi^2 = 4{\cdot}4$, df = 1, $N = 38$, $P < 0{\cdot}05$) or the normal control group ($\chi^2 = 10{\cdot}6$, df = 1, $N = 38$, $P < 0{\cdot}001$).

Working memory task

The three groups were compared on two types of error count for this task. The first category of 'between search errors' refers to the frequency with which subjects returned to open a box in which a token had already been found. The second category of 'within search errors' refers to the incidence of subjects returning to a box already opened and shown to be empty in the same search sequence. For fathers, the mean values (and standard errors) of total between search errors were as follows: 33·4 (4·7) for the autism group, 28·2 (4·0) for the learning disability group and 17·9 (3·3) for the control group. The corresponding values for mothers were: 35·5 (4·7) for the autism group, 30·1 (4·0) for the learning disability group and 23·1 (4·1) for the control group. A two-way ANOVA (group × sex) of total between errors showed a significant main effect of group only ($F(2,112) = 5{\cdot}3$, $P < 0{\cdot}005$). *Post hoc* comparison of mothers and fathers pooled using Tukey's test for 'honestly significant difference' showed that this group difference was significant between the autistic and normal groups only, and was attributable to a difference between the performance of fathers but not mothers. A two-way ANOVA (group × sex) on within-search errors showed no significant effect of either group ($F(2,112) = 0{\cdot}1$) or sex ($F(1,112) = 0{\cdot}1$).

Spatial short-term memory task

For fathers, mean spatial span values (and corresponding standard errors) were 5·8 (0·3) in the autism group; 5·3 (0·3) in the learning disability group; and 6·2 (0·2) in the control group. For mothers, mean spatial span values

Table 3. *Correlations between task T-scores for each group*

Task/group	Spatial span	Working memory	Attentional flexibility	Planning
Spatial span				
Autism	—	0·251	0·386*	0·332*
Learning disability	—	0·389**	0·137	0·132
Control	—	0·466**	0·142	0·191
Working memory				
Autism	0·251	—	0·492***	0·279
Learning disability	0·389**	—	0·101	0·214
Control	0·446**	—	0·184	0·349*
Attentional flexibility				
Autism	0·386*	0·492***	—	0·290
Learning disability	0·137	0·101	—	0·228
Control	0·142	0·184	—	0·182
Planning				
Autism	0·332*	0·279	0·290	—
Learning disability	0·132	0·214	0·228	—
Control	0·191	0·349*	0·182	—

* $P < 0{\cdot}05$; ** $P < 0{\cdot}01$; *** $P < 0{\cdot}005$; **** $P < 0{\cdot}001$.

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(and corresponding standard errors) were 5·5 (0·2) in the autism group; 5·2 (0·3) in the learning disability group; and 6·1 (0·3) in the control group. A two-way ANOVA (group × sex) on the spatial span measure showed a significant main effect of group only ($F(2,112) = 5{\cdot}0$, $P < 0{\cdot}01$). *Post-hoc* comparison with Tukey's test for 'honestly significant difference' showed that although the normal group showed significantly better spatial spans than the learning disability group, there was no significant difference between the autism group and either of the other two groups. (Table 3). When mothers and fathers were considered separately, Kruskal–Wallis tests showed no significant difference between fathers ($N = 57$, df = 2, $\chi^2 = 0{\cdot}2$), and a non-significant trend for mothers in the learning disability group to do more poorly than mothers in the other two groups ($N = 56$, df = 2, $\chi^2 = 5{\cdot}1$, $P < 0{\cdot}08$).

DISCUSSION

Since the commencement of this study, Ozonoff *et al.* (1995) have reported executive dysfunction among siblings of individuals with autism. To our knowledge, however, this is the first study in which parents of autistic children have been given tests of executive function. In summary, parents of autistic children were compared with both parents of learning disabled children and a control group of adults from unaffected families, on four computerized tasks. Three tasks tapped distinct aspects of executive function (attentional flexibility, planning and working memory), while the fourth was a spatial-span control task. The results confirmed the hypothesis that parents of autistic children show impaired executive function. Compared with normal controls, the autism group performed poorly on all three executive function tasks. When compared with parents of learning disabled children, both mothers and fathers of autistic children showed clear difficulty on the critical 'extra-dimensional' shift stage of the IDED task, despite their success on all the previous stages. The processes involved in these earlier stages (discrimination learning, rule-reversal and transfer of learning) can therefore be eliminated from accounts of the performance deficits of this group. Fathers were also significantly impaired on the planning task (using both lenient and stringent criteria for planning efficiency). The autism group also performed worse than the learning disability group on the working memory task, but here the differences were non-significant.

Before discussing these findings, there are two points worth emphasizing. First, parents in the autism group did not appear to be less attentive or motivated than the other two groups. This can be seen from their good performance on the attentionally demanding spatial-span task, as well as from several control measures in the other tasks (e.g. within-search errors on the working memory task, and pre-transfer stages of the IDED task). Secondly, none of these tasks required verbal or pragmatic skills, since each task was fully computerized, and contained a set of practice trials to familiarize the participants with the rules and objectives of that task. As noted at the start of this paper, previous studies have focused upon communicative impairments among parents of autistic children. The impaired executive function performance of this group is therefore all the more striking in view of the non-verbal, non-social nature of the tasks used in this study.

In previous work with relatives of autistic children, the notion of a 'lesser variant' has proved useful. This term, first coined by Folstein & Rutter (1977), is used to describe peculiarities that are subtle in manifestation, but similar in kind to autism. Hitherto, the focus has been very much upon language-based cognitive impairments (e.g. reading disorders, poor spelling, delayed speech). Recently however, Bolton *et al.* (1944) have argued that the boundaries of this proposed lesser variant should be extended to encompass social peculiarities and stereotypic behaviours. The results of the present study, as well as those of Ozonoff *et al.*'s (1995) study with siblings, support this proposal, and suggest that the differences among relatives of autistic children can be observed in all three domains of the autistic 'triad of impairments'. That said, since this is a family study, it is not possible to establish whether such differences have a genetic, environmental or transactional basis.

One question that can however be addressed is: how widespread were problems of executive function among the autism group? An exploratory chi-square 'goodness-of-fit' test for the composite scores showed no significant difference ($\chi^2 = 1{\cdot}25$, df = 1, $N = 80$) between

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the autism group and the learning disability group, suggesting that poor executive function is not universal, but rather characteristic of a subgroup of parents of autistic children. In order to examine this possibility further, all those participants whose T-scores[1] fell below the grand mean for all three executive function tasks were identified. This group of poorly performing participants included 10 parents in the autism group (six fathers and four mothers); one mother from the learning disability group and two mothers in the normal control group. That is, pervasive problems of executive control were observed in only 25% of the autism group, and yet were 5–10 × more frequent in this group than in the two control groups. Furthermore, the group difference was more evident for fathers than for mothers. Although based on small numbers, these findings are consistent with the evidence from both biological and epidemiological studies (reviewed in Wing, 1988), which suggest that there are multiple aetiologies for autism, and that males are more frequently affected than females.

A second question prompted by the findings of this study concerns the complex nature of the tasks used, since this suggests that success or failure on a given task may have several potential causes. Did the three groups differ in the strategies they adopted to solve the tasks? To answer this question directly, individual performances were compared across each task (see Table 2). It was predicted that high spatial-span should be associated with good performance on the working memory task. Interestingly, this predicted correlation was found for both control groups, but not for the autism group. Instead, for the autism group alone, spatial span was correlated with both attentional flexibility and planning performance. While planning was more strongly correlated with working memory than with spatial-span for both control groups, the opposite pattern was true for the autism group. That is, the performance of the autism group was not only quantitatively but also qualitatively distinct from that of controls, suggesting that the autism group may have relied on unusual strategies to solve these tasks.

[1] T-scores are computed from standard z-scores: [$z_y = (Y - \bar{Y})/\text{s.d. of } Y$], z-scores have a mean of zero and S.D. of one. T-scores have a mean of 50 and S.D. of 10. Thus, $T = 10z + 50$, rounded up to the nearest integer.

Another important question that arises from this study is whether poor performance on these tests of executive function was related to other aspects of participants' functioning in the real world. In particular, was task performance related to clinical impressions of autism-related abnormalities in the individual? To address this issue, composite T-scores for executive function were compared with pre-test interviewer total ratings of social abnormalities in each participant. A Spearman's rank correlation test demonstrated a modest, but significant, relation for the autism group (rho $= 0{\cdot}329$, df $= 1$, $N = 40$, $P < 0{\cdot}05$). That is, not only did parents of autistic children perform less well as a group than parents of learning disabled children on tests of executive function, but also, at an individual level, this performance deficit was associated with abnormalities in social function generally considered to be peculiar to autism. In contrast, composite executive function scores were not correlated with social behaviour ratings for parents of learning disabled children (rho $= 0{\cdot}179$, df $= 1$, $N = 40$). The association between executive dysfunction and social abnormalities for parents of autistic children is particularly interesting, in that several authors have suggested that executive dysfunction might contribute to the social and imaginative deficits among individuals with autism (Ozonoff *et al.* 1991; Harris, 1993; Hughes & Russell, 1993; Jarrold *et al.* 1993).

Empirical support for a link between executive function and social behaviour can be found in several studies of autism. An interesting connection between these two domains was first suggested by Wing & Gould's (1979) finding that, in an epidemiological sample of socially impaired children, restricted interests and repetitive actions were significantly associated with the degree and type of social impairment. Since that time, set-shifting performance (a standard measure of executive function) has been shown to be closely correlated with social behaviour in young autistic children (McEvoy *et al.* 1993) and to predict social outcome in longitudinal studies of high functioning autistic children (Berger *et al.* 1993) and adolescents (Szatmari *et al.* 1989). In addition, intervention studies with autistic children (Clark & Rutter, 1981; Volkmar *et al.* 1985; Dadds *et al.* 1988) all converge on the conclusion that placing autistic children in highly

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structured situations (in which the demands for executive function are reduced) facilitates the development of social behaviour.

Of course, much of the above is based on correlational data, and so does not speak to the issue of causality. One alternative and plausible explanation for the association between executive function and social behaviour is that both depend upon neuroanatomically proximal substrates. However, the predicted consequences of executive dysfunction (impaired performance in novel, changing or ambiguous situations) offer *a priori* grounds for the argument that deficits in executive control should be especially marked in social interactions, since these typically involve all three of the factors above. From this viewpoint, it may be possible to reconceptualize some of the earlier findings from parents of autistic children. For example, both the 'verbal disinhibition', described by Wolff *et al.* (1988) as discriminating parents of autistic children from controls, and the narrative difficulties for this group reported by Landa *et al.* (1992) are consistent with executive dysfunction. Thus, it is possible that executive dysfunction plays a significant part, not only in the problem of autistic stereotypes, but also for the whole of the behavioural phenotype of autism.

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