

Quasi-autistic Patterns Following Severe Early Global Privation

Michael Rutter, Lucie Andersen-Wood, Celia Beckett, Diana Bredekamp†, Jenny Castle, Christine Groothues, Jana Kreppner, Lisa Keaveney, Catherine Lord, Thomas G. O'Connor, and the English and Romanian Adoptees (ERA) Study Team

MRC Child Psychiatry Unit and Social Genetic and Developmental Psychiatry Research Centre, Institute of Psychiatry, London, U.K.

Six per cent of child in a sample of 111 children who were adopted into U.K. families from Romania, and who were systematically assessed at the ages of 4 and 6 years, showed autistic-like patterns of behaviour. A further 6% showed milder (usually isolated) autistic features. Such autistic characteristics were not found in a similarly studied sample of 52 children adopted in the first 6 months of life within the U.K. The children from Romania with autistic patterns showed clinical features closely similar to “ordinary” autism at 4 years but they differed with respect to the improvement seen by age 6 years, to an equal sex ratio, and to a normal head circumference. The children from Romania with autistic features tended to differ from the other Romanian adoptees with respect to a greater degree of cognitive impairment and a longer duration of severe psychological privation.

Keywords: Autism, institutional care, psychological privation, cognitive impairment, preschool children, social deficits, repetitive behaviour, circumscribed interests, sensory preoccupations.

Abbreviations: ADI-R: Autism Diagnostic Interview; ASQ: Autism Screening Questionnaire; ERA: English and Romanian Adoptees; GCI: General Cognitive Index; ROC: receiver operating characteristics.

Introduction

Surprisingly little is known about the long-term social sequelae of profound deprivation in early life. The initial studies of institution-reared children emphasised so-called “affectionless psychopathy” as the characteristic outcome (Bowlby, 1946)—a pattern of failure to form intimate committed relationships associated with anti-social behaviour (Rutter, 1981). Those early studies had many methodological limitations and it is evident that both the frequency of this outcome and its irreversibility were overstated. Nevertheless, Harlow’s studies of profound social isolation in rhesus monkeys showed devastating effects on their later social relationships (Harlow & Harlow, 1972), even though these were to some extent reversible by adaptive later social experiences (Novak, 1979). Hodges and Tizard’s (1989a, b) follow-up of children who spent their first few years in British residential nurseries of good quality noted that, despite later adoption or return to their biological families, many

showed a relative paucity of selective committed confiding relationships with other people in adolescence, although they seemed to have developed appropriate selective attachments with their parents. Chisholm, Carter, Ames, and Morison (1995), in their follow-up study of children from Romanian orphanages adopted by Canadian families, found that many showed a pattern of indiscriminate friendliness; our own study of Romanian adoptees similarly indicated effects on a range of social relationships (Keaveney & O’Connor, 1997; Kreppner, O’Connor, Dunn, Andersen-Wood, & the ERA Study Team, in press; O’Connor, Bredekamp, & Rutter, in press); and Kaler and Freeman (1994), in their pilot study of children within a Romanian orphanage, noted positive, but rather indiscriminate, peer relationships.

None of these studies of groups of deprived, institution-reared children has commented on the occurrence of autistic patterns. The same applies to most individual case studies of children reared in extremely adverse conditions (Skuse, 1984b). There are only two possible exceptions. First, Genie, a girl rescued from extreme privation only in early adolescence (Curtiss, 1977; Rymer, 1993), was described as showing pervasive social deficits, although these were not discussed explicitly in terms of autism. Second, there is one child in the family of deprived children described by Skuse (1984a), although the autism in that case was not primarily attributed to the psychological privation. Indeed, in terms of what is known about the organic basis of autism and the very

Requests for reprints to: Professor Michael Rutter, MRC Child Psychiatry Unit, Institute of Psychiatry, De Crespigny Park, Denmark Hill, London SE5 8AF, U.K.

The ERA Study Team comprised Judy Dunn, Kathryn Ehrich, Alexandra Harborne, Dale Hay, Jessica Jewett, Julie Messer, David Quinton, and Adele White in addition to the listed authors (apart from Catherine Lord).

† (deceased).

strong genetic component that is involved (Bailey, Phillips, & Rutter, 1996), autism would not be expected to arise as a result of severe privation. Nevertheless, shortly after the wave of Romanian children adopted by English families following the fall of the Ceausescu regime, two adoptees from Romania were referred to MR with the possible diagnosis of autism. As this coincided with the start of a systematic follow-up of 165 Romanian adoptees (see Rutter & the ERA Study Team, 1998), special attention was paid to possible autistic features to check whether this was an occasional sequel to profound early privation or whether the two cases represented no more than a coincidental association. The present paper reports the findings on 11 children suspected by parents or professionals of showing possible autistic features.

Sample and Methods

A randomly selected, age-stratified sample of 165 Romanian children, adopted into English families, and who came to England before the age of 42 months are being studied systematically (Rutter & the ERA Study Team, 1998). Of these, 111 were assessed at age 4 years, the remaining 54 being already too old to be seen at that age. These 54 have been assessed at 6 years of age and the 111 seen at age 4 have been re-examined at age 6. The group is being compared with a sample of 52 nondeprived U.K.-born children adopted by U.K. parents and placed before the age of 6 months.

The children's social, behavioural, and emotional functioning is being assessed by a combination of parental interview, questionnaires completed by parents and teachers (Elander & Rutter, 1995), and a video-taped play session that included a separation from, and reunion with, the mother (Cassidy & Marvin, 1992). Cognitive functioning is being evaluated through parental completion of the Denver Scales (Frankenburg, van Doorninck, Liddell, & Dick, 1986) and individual testing of the children using the McCarthy Scales (McCarthy, 1972). For the few children below the floor of the latter, the Merrill-Palmer Scales (Stutsman, 1931a, b) were employed.

In order to assess social deficits, communicative abnormalities, and stereotyped behaviour patterns that might possibly reflect autistic-like features, a questionnaire (Autism Screening Questionnaire: ASQ) based on the Autism Diagnostic Interview (ADI-R; Le Couteur et al., 1989; Lord, Rutter, & Le Couteur, 1994) was developed by Catherine Lord and Michael Rutter (Berument, Rutter, Bailey, Pickles, & Lord, 1998). It comprises 40 questions tapping social relationships (e.g. Does he/she seem unusually interested in the sight, feel, sound, taste or smell of things or people?), unusual communication (e.g. Has he/she ever used odd phrases or said the same thing over and over in almost exactly the same way?) and stereotyped behaviours (e.g. Are there things that he/she seems to have to do in a very particular way or order, or rituals that he/she has to have you do?). It correlates well with the ADI-R and shows good discriminative validity between pervasive developmental disorders and both mental retardation and other nonautistic psychiatric conditions. The best discrimination, based on receiver operating characteristics (ROC) analyses, is provided by a score of 14 or more. The ASQ was administered both at 4 years and 6 years.

A selection (numbering 26 so far) of the 165 children have been seen by MR for a more detailed clinical evaluation. This subgroup seen clinically included a range but it was weighted towards those where the parents wanted advice, or the children were undergoing educational statementing, or where the possibility of autistic features had been raised by someone. The

present report is concerned primarily with the subgroup of 11 children where the last was the case. In four cases a firm diagnosis of autism had been made by a consultant at some other clinical centre; in another four cases a diagnosis of "probable autism" or "autistic features" had similarly been made; and in three cases autistic features had been noted by either the parents or by members of the research team (or both). These 11 children came entirely from the group of Romanian adoptees; no queries regarding autism were raised with respect to any of the 52 within-England adoptees.

The ADI-R was used to obtain a detailed parental report on the children's development and current behaviour with particular reference to features associated with autism and other pervasive developmental disorders. The interview ratings (made by the investigator according to operationalised criteria applied to the informant descriptions) can be used to obtain an algorithm score for social deficits, communicative deficits, and for stereotyped repetitive behaviours. These scores, in combination, have been shown to have good reliability and high discriminative validity in differentiating autism from the non-autistic behaviours associated with mental retardation and other developmental disorders. In order to contrast the clinical features presented by the 11 Romanian adoptees showing possible autism with those found in more usual samples of children with autism, a comparison was made with 14 children from Catherine Lord's (Lord, Shulman, Pickles, & DiLore, 1997) longitudinal study of children with early diagnosed autism. They were selected from her larger prospectively studied sample on the basis of having a Wechsler Performance IQ of 50 or greater (mean = 78.3, *SD* = 18.9), and having an ADI-R interview assessment shortly before age 4 years (range 40 to 48 months) and a further ADI-R assessment after the age of 5 years (range 60 to 70 months).

Results

As reported more fully in a separate paper (Rutter & the ERA Study Team, 1998), the group of children adopted from Romania were very severely deprived at the time of coming to England. Half had a weight below the third percentile, a similar proportion showed developmental functioning in the retarded range and many had gastrointestinal, respiratory, and skin disorders. The conditions of care in the Romanian institutions that they had experienced varied from poor to appalling. Typically they remained in cots all day, had few if any toys or playthings, and were fed gruel through bottles with large teats (often left propped up for self-feeding); there was no personalised caregiving and very little talk or interaction with caregivers.

Despite these dreadful circumstances of rearing, the group, as a whole, showed remarkable catch-up. By the age of 4 years, only 2% of those who entered the U.K. before the age of 2 years had a weight below the third percentile, their mean score on the overall General Cognitive Index on the McCarthy scales was 100 compared with 109 in the comparison group, and their overall social functioning was well within the normal range.

Because the findings of our study showed that autistic-like features were most marked at 4 years of age and tended to diminish greatly over the next 2 years, the results of quantitative analyses focus on the 111 children who were assessed at both 4 and 6 years of age.

By the time the 6-year-old assessments on the 111 children were undertaken, the research team were aware

of both the possible importance of quasi-autistic features and of the forms they might take. Extensive interview and observational data were available at two ages for them to decide on the need for more detailed individual assessment by MR. The ASQ was not used in the selection process (in order to keep the measures independent). There was only one child for whom the team considered referral but who was not seen. Particularly because this allowed two opportunities to pick out children whose behaviour warranted a more detailed standardised clinical assessment of autistic features, it is likely that very few children with autistic features will have been missed; hence this group provides the best estimate of prevalence in those without severe cognitive impairment. The clinical descriptions, however, are presented for the entire sample of 11 children out of 165 considered to show a quasi-autistic pattern.

The 11 Children with Possible Autism

The 11 children picked out by professionals and/or parents because of possible autistic features varied in the behaviours thought to be autistic. In all instances, difficulties in social relationships and in communication, corresponding with key features of prevailing concepts of autism, were found. Thus, concerns were expressed about difficulties in forming selective friendships, in social reciprocity, in empathy for other people, and in the use of eye-to-eye gaze and gesture in social approaches and responses. Similarly, attention was drawn to impaired language development and to a lack of reciprocal conversation and social chat.

Preoccupations with sensations and intense circumscribed interests also constituted a prominent feature in a majority of the 11 children. These were particularly striking both because they did not take the form of the stereotyped behaviours (such as rocking and other stereotypies) seen in deprived environments, especially in mentally handicapped individuals (Berkson, Gutermuth, & Baranek, 1995; Clarke & Clarke, 1976; Ridley, 1994), and because often they came to a peak well *after* coming to England and were not evident at the time the children came to the U.K.

Two of the children were fascinated by watches and would take them off strangers (one researcher realised part-way through the testing session that, without her being aware of it, the child had succeeded in acquiring her watch!). One of these children had quite a collection of watches, both real and toy. Another child's fascination with vacuum cleaners was so all-encompassing that when family and friends were being visited, they had to be warned to hide away their vacuum cleaner. In another child, the interest focused on plumbing systems, toilets, and the disposal of sewage, about which he had quite an extensive knowledge. His usual first step in visiting a new house was to investigate the lavatory. One of the children who was fascinated with watches also had an intense interest in new £10 notes—not £5 or £20 ones and not crumpled £10 ones.

In seven instances, the children also showed a preoccupation with sensations—particularly those involving touch or smell. Sometimes these were combined, as in one

child who smelled objects, felt their texture and then put them to his lips. Often the focus on sensations involved specific objects or classes of objects—such as shiny clothes or the aforementioned £10 notes, which would be kissed. Another child liked the feel of men's moustaches, and another put vibrating objects to his head to feel the vibrations.

In no case was there any possibility that the children had been admitted to an institution because a handicapping condition had been diagnosed. Only 1 of the 11 children had been reared by a biological parent for as long as 2 weeks and this family care lasted only 4 months; another child was cared for during the first 12 months of life by a grandparent in bad health who left the child untended all day in poor conditions. All the other nine children were admitted to institutions during the first month of life, in seven cases this being from the time of birth onwards.

The behavioural patterns shown by the 11 children were not all the same and 3 groups seemed recognisable. First, there was one child who came to the U.K. under the age of 6 months and who, having progressed well, lost language at the age of about 20 months, became socially unresponsive, and developed some stereotyped behaviours. Within the next year language returned, she regained normal social behaviour and, when seen after starting school, appeared normal in all respects. The nature and origins of this autistic-like phase remain obscure; it was not obviously related to her experiences in Romania, and there is no evidence of autism now. She seems to have little in common with the remaining 10 children. Her behaviour is described in case history 1 in the Appendix but she is not considered further here.

The remaining 10 children divided into the 3 whose autistic pattern was associated with severe cognitive impairment and the 7 whose McCarthy General Cognitive Index (GCI) score exceeded 50. These two subgroups will be considered separately.

Autism Associated with Severe Cognitive Impairment

The three children with cognitive scores in the severely retarded range all had ADI-R scores that met the accepted algorithm criteria, all had received firm diagnoses of autism by professionals, and all had little or no spoken language (see cases 2–4 in the Appendix). The three children were among those adoptees who came to the U.K. at an older age (21, 30, and 39 months); each had received all, or almost all, of their upbringing in extremely poor institutional conditions, and one stood out in being exceptionally severely malnourished at the time of coming to the U.K. In addition all three had something further that was unusual in their background. One was reported to have been born at 26 weeks gestation with a birthweight of 2 lbs and a period of intensive care after birth; one had a major high tone hearing loss and came from an orphanage of unusually poor quality in which a large number of children are known to have died for reasons unknown in the 6 months prior to the child coming to the U.K.; and one had been kept isolated in a single room apparently because of racial discrimination

Table 1
Autism Diagnostic Interview Scores of Romanian Adoptees and Autism Sample Children^a

	Romanian adoptees				Autism sample			
	48–54 months (<i>N</i> = 6)		72–78 months (<i>N</i> = 6)		41–48 months (<i>N</i> = 14)		61–70 months (<i>N</i> = 14)	
	Mean	(<i>SD</i>)	Mean	(<i>SD</i>)	Mean	(<i>SD</i>)	Mean	(<i>SD</i>)
Social domain	13.3	(6.9)	5.8	(5.0)	14.4	(4.7)	19.9	(4.2)
Communication domain	10.8	(4.5)	7.2	(3.4)	9.7	(3.5)	12.3	(3.7)
Stereotyped repetitive behaviour	2.7	(1.9)	2.7	(1.5)	4.4	(1.7)	6.4	(1.7)
Total algorithm item score	26.8	(8.8)	15.7	(7.9)	28.6	(8.1)	38.6	(7.5)

^a Children with an IQ of 50 or more only.

and illegal status. Two of the three also had a head circumference still below the third percentile at 6 years.

Although, in many respects, these three children seemed to show a syndrome that closely approximated to autism, and although all showed serious cognitive impairment with indications of probable organic impairment, the pattern was atypical in several respects. Thus, all had learned makaton sign language. Their sign vocabulary was limited but each made substantial spontaneous use of makaton (as reported by parents and as observed by MR); this would be unusual in children with the ordinary type of autism. Two of the children made considerable social approaches (albeit deviant in quality), again an atypical element, and one had improved greatly between 4 and 6 years, as reflected in the halving of his ADI algorithm score. It is notable, too, that their head circumference was below normal, instead of increased, as found in many children with “ordinary” autism (Woodhouse et al., 1996).

Prevalence of Quasi-autistic Pattern at 4 to 6 Years

Because the importance, and high rate, of quasi-autistic patterns in the children adopted from Romania only became apparent well after the study began, their prevalence may best be assessed within the subgroup seen at both 4 and 6 years. Out of 111 children seen at both ages, there was 1 child with autism associated with severe cognitive impairment and 6 with quasi-autistic patterns not accompanied by severe retardation—an overall rate of 7 out of 111, or 6.3%. The quantitative analyses that follow are based on these 6 children.

Quasi-autistic Patterns in Children without Severe Retardation

Two rather different questions need to be addressed with respect to the subgroup of 6 children (out of the 111 present for both the 4- and 6-year assessments) with quasi-autistic patterns of behaviour but no associated severe cognitive impairment. First, there is the issue of the extent to which their behaviour parallels that found in “ordinary” autism, and second, there is the question of the ways in which they differ from the remaining children in the sample of Romanian adoptees. Table 1 summarises

the ADI-R findings as they compare with those in Lord et al.’s (1997) longitudinal study sample and Table 2 provides the statistical results. Lord’s group of autistic children all had contemporaneous ADI-R assessments at both age periods (circa 4 years and circa 5½ years). The same applied to four of the six adoptees from Romania. In the remaining two cases, the 4-year scores had to be derived from retrospective reporting from later conducted ADI-R interviews (at about 6 years of age). However, in these two cases, the findings were corroborated by contemporaneous detailed interviews at 4 years conducted as part of the overall research battery. As the findings do not vary according to the method of assessment, they are pooled here.

Three points stand out. First, at age 4 years the Romanian children and the children with autism in Lord’s sample had closely comparable ADI-R scores. Neither on the total algorithm item score, nor on the subscores on each of the three key diagnostic domains, were there any statistically significant differences. It may be concluded that, at that age, the possibility of autism had been raised quite reasonably and the gross ADI-R pattern of scores provided no particular grounds for querying the diagnosis. Second, the situation of age 6 years was quite different. At that age, all ADI-R scores were significantly lower in the Romanian sample group. Third, as follows from the first two findings, the pattern of change over time in the two groups was quite different, as shown by the statistically significant group by time interaction. The children in Lord’s sample had become progressively *more* autistic in their behaviour whereas the Romanian adoptees had become progressively less so.

With a sample as small as six, quantitative comparisons on individual ADI-R items would not be appropriate. Nevertheless, the details of the children’s behaviour did stand out as atypical in several crucial respects. This was apparent to some extent at age 4 years but it was most obvious at age 6. Thus, the one child who used makaton made some spontaneous use of signing to communicate and another child stood out in terms of her spontaneous flexible use of language. For example, when seen at 4 years she wanted her mother to retrieve a toy from behind the settee where she had dropped it. Her mother, who was engaged in conversation with MR, did not immediately respond and the child repeated the request in two rather

Table 2
Results of Repeated Measures Analysis for Data in Table 1: F value

	“Dataset”	Year	Interaction
Social domain	14.42***	0.55	23.19***
Communication domain	1.82	0.27	8.74**
Stereotypical repetitive behaviour	15.90**	4.58*	4.58*
Total algorithm item score	13.70**	0.08	28.23***

* $p < .05$; ** $p < .01$; *** $p < .001$. All other results nonsignificant.

different ways—a flexibility that would be very unusual in autism. Several showed much social approach, two were obviously friendly (although abnormally so) in their social style, and two others showed a pattern of indiscriminately friendly behaviour closely similar to that already described in nonautistic children adopted from Romania. For example, one child greeted MR with an immediate flow of questions, comments, and chat; asked about the contents of his briefcase and took MR off to her room, where she showed off her collections of belongings (including those involved in her circumscribed interests). Another child was emotionally animated in a fashion that would be unusual in autism and, by the age of 6 years, one child had selective friendships involving shared play and talk. Also, several showed social sharing at 6 years (bringing things they had done to show off to their parents) although they had not done so when younger. The reason why, despite these atypical features, four out of the six children met the social criteria for autism was that they tended to lack social reciprocity, showed markedly limited social awareness, lacked an appreciation of normal social boundaries, were seriously constrained in their ability to maintain (as distinct from initiate) social interaction, showed limited empathy, and sometimes made little use of their parents for security and comfort. It was the same lack of reciprocity and social usage of language that led all of them to meet the algorithm criteria for communication deviance. Some went through a phase of repetitive language, there were some mild indications of unusual uses of language, and one child had quite a range of idiosyncratic made-up words. Four of the six, at 4 years, also met the algorithm criteria for repetitive, stereotyped behaviour, with the sensory preoccupations and intense circumscribed interests the most frequent manifestation. Motor stereotypies were not prominent although they were present in some children.

The marked tendency for the autistic-like behaviours to improve over time was to some extent accompanied by a tendency for IQ scores to rise. This could not be compared with Lord's sample because the cognitive assessment in that study occurred at only one age and also used a different test. It was possible, however, to compare the cognitive gains in the six children with a quasi-autistic pattern seen at both 4 and 6 years with those seen in the remainder of the sample of Romanian adoptees at the same ages, excluding those who did not receive institutional care as there were no children with that background among the six (see Table 3). The 6 children showed a mean rise of 20 points (from 57 to 77) on the McCarthy GCI compared with a rise of only 7 points in the remainder of the sample, a difference that was statistically

significant (F values for group, age, and the interaction between the two were 35.28, 23.76, and 6.87 respectively, with the first two significant at the 1% level and the last at the 5% level). Nevertheless, despite this rise, the children showing a quasi-autistic pattern stood out from the rest of the Romanian adoptees with respect to their much lower cognitive level—a difference of 45 points at age 4 years and of 32 points at age 6.

As already noted, all of the six children with a quasi-autistic pattern had received an institutional upbringing, in all but two cases for virtually the whole of their rearing. This applied to most of the remainder of the sample but there were 18 out of 100 (i.e. the total sample of 111 seen at age 4 years minus the 11 children with a quasi-autistic pattern) reared by their biological families who did not receive institutional care. Table 3 compares the six with the remainder of the Romanian sample who had received an institutional upbringing and who were assessed at age 4 years ($N = 86$). Few differences were found except that none of the 6 children showing quasi-autistic behaviour had entered the U.K. before the age of 12 months compared with 59 of the remaining 86, a statistically significant difference (Fisher's Exact Test; $p = .002$). The between-group comparison on the mean age of entry to the U.K. showed a significant difference in the same direction (a mean of 17 months vs. 9 months). Otherwise, the subgroup of six was unremarkable. Their initial Denver score was somewhat lower (38.5 vs. 57.5) but the difference fell well short of statistical significance. As judged by their weight at U.K. entry they were no more malnourished than the remainder of the sample (indeed, their weights were marginally greater).

Extensive descriptions of the children's behaviour at the time of entering the U.K. were obtained from the parents when the children were age 4 years. Interviewer ratings of these descriptions were compared between the six children with the quasi-autistic pattern and the remainder of the sample (excluding those with overt mental handicap) who had a comparable age of entry (i.e. over the age of 12 months). Virtually all of the children in both groups showed major impairments in language and play, most were socially unresponsive, over half rocked and/or showed stereotyped behaviours but only one child (not in the quasi-autistic group) showed obsessions/abnormal preoccupations. In short, at least as observed by parents, there was nothing particularly distinctive about the behaviour of the children who later showed quasi-autistic behaviour. At that time they did not exhibit the marked circumscribed interests that were so striking later. What was distinctive was the pattern of behaviour (as described above) that developed between the time of coming to the U.K. and 4 years of age.

Table 3
Cognitive Level and Anthropometric Data at 4 and 6 Years on Romanian Adoptees with and without Quasi-autistic Patterns

	Quasi-autistic ^a (<i>N</i> = 6) ^c		Remainder ^b (<i>N</i> = 86) ^c		<i>F</i> ratio
	Mean	(<i>SD</i>)	Mean	(<i>SD</i>)	
McCarthy GCI at 4 years (<i>N</i>)	57.0 (6)	(15.5)	101.8 (84)	(15.4)	47.41***
McCarthy GCI at 6 years (<i>N</i>)	76.5 (6)	(17.4)	108.7 (80)	(17.6)	18.73***
Age at entry (months) (<i>N</i>)	16.8 (6)	(3.5)	8.6 (86)	(6.0)	10.97**
Denver at entry (<i>N</i>)	38.5 (5)	(9.7)	57.5 (78)	(37.0)	1.29
Denver at 4 years (<i>N</i>)	70.0 (5)	(13.1)	107.1 (75)	(23.7)	11.97***
Weight at entry (<i>N</i>)	-1.85 (6)	(0.95)	-2.42 (84)	(1.7)	0.71
Weight at 4 years (<i>N</i>)	0.21 (6)	(0.46)	-0.04 (83)	(0.90)	0.42
Head circumference at entry (<i>N</i>)	-2.48 (6)	(1.14)	-2.19 (47)	(1.9)	0.14
Head circumference at 4 years (<i>N</i>)	-1.55 (4)	(1.0)	-1.29 (74)	(0.99)	0.26

^a Children with quasi-autistic pattern.

^b Remainder of sample who experienced institutionalisation (excluding those with severe retardation).

^c Numbers vary slightly as a result of missing data.

* $p < .05$; ** $p < .01$; *** $p < .001$. All other results nonsignificant.

Milder Quasi-autistic Features

A further question arises as to whether these children showed a categorical pattern, quantitatively different from that seen in the rest of the sample, or rather an extreme of features seen in lesser degrees in the sample as a whole. To consider that question, attention needs to return to the whole sample of 165, asking whether there were further children (beyond the 11 described here) who might be considered to show milder quasi-autistic features. The research team was asked to review all interview questionnaire and observational data for this purpose. There were an additional 10 children (5 of whom were seen at both 4 and 6 years of age) who showed some milder, usually isolated, quasi-autistic features (see cases 12 to 21 in the Appendix). Three of these (cases 15, 16, and 21) were seen by MR and were not thought to show sufficient quasi-autistic features to be included. In three others (cases 12, 13, and 14) the possibility of autism had been raised by others. If they had been seen, it is possible that they might have been regarded as showing a quasi-autistic pattern, although that is far from certain. All three were in the group of older children seen only at 6 years, and two showed intellectual impairment.

If attention is focused on the 5 children out of the 111 seen at both 4 and 6 years who showed milder autistic features, 4 features stand out. First, the autistic features mainly involved stereotyped, repetitive behaviours (rather than severe social or communicative deficits). Thus, the behaviours included marked interests in sensations, motor mannerisms, and unusual preoccupations (see case histories 12 to 21 in the Appendix). Second,

although social withdrawal was not characteristic, a lack of social boundaries was frequent. Third, unlike the diagnosed subgroup, they had not entered the U.K. at an older age (see Table 4). Fourth, again in contrast with the diagnosed subgroup, they did not show cognitive impairment.

Finally, in view of the importance of comparing the autistic features in this group of severely deprived children with those seen in autism as found in other groups, the sex ratio warrants mention. Taking the total sample of 21 children with some autistic features (both marked and mild), 12 were female. The approximately equal sex ratio contrasts sharply with the marked male preponderance ordinarily found with autism (Bailey, Phillips, & Rutter, 1996).

Autism Questionnaire Findings in the Sample as a Whole

The question of whether there were milder autistic-like features in a larger proportion of children adopted from Romania beyond those outside the category of quasi-autistic behaviour could also be tackled by considering the total group of 111 Romanian adoptees assessed at age 4 with respect to the findings on the ASQ. The mean total score at age 4 years of the Romanian adoptees (6.0) was nonsignificantly higher than that in the comparison group (5.2) but the subscore for communicative impairment (3.0, $SD = 1.8$) was significantly higher than that for the within-U.K. adoptees (2.3, $SD = 1.4$). The overall dis-

Table 4
Cognitive Level and Anthropometric Data at 4 and 6 Years on Romanian Adoptees with and without Isolated Autistic Features

	Autistic features ^a (<i>N</i> = 5) ^c		Remainder ^b (<i>N</i> = 81) ^c		<i>F</i> ratio ^d
	Mean	(<i>SD</i>)	Mean	(<i>SD</i>)	
McCarthy GCI at 4 years (<i>N</i>)	97.4 (5)	(8.3)	102.0 (70)	(15.7)	0.42
McCarthy GCI at 6 years (<i>N</i>)	114.4 (5)	(10.4)	108.3 (75)	(17.9)	0.56
Age at entry (months) (<i>N</i>)	10.2 (5)	(5.9)	8.5 (81)	(6.0)	1.68
Denver at entry (<i>N</i>)	34.3 (4)	(2.5)	58.7 (74)	(37.6)	1.68
Denver at 4 years (<i>N</i>)	93.9 (5)	(27.6)	108.1 (70)	(23.3)	1.68
Weight at entry (<i>N</i>)	-2.42 (5)	(1.6)	-2.43 (79)	(1.7)	0.00
Weight at 4 years (<i>N</i>)	0.63 (5)	(1.2)	-0.08 (78)	(0.87)	2.99
Head circumference at entry (<i>N</i>)	-2.59 (3)	(1.79)	-2.16 (44)	(1.9)	0.14
Head circumference at 4 years (<i>N</i>)	-0.84 (4)	(0.50)	-1.32 (74)	(1.0)	0.86

^a Children with isolated autistic features.

^b Remainder of sample (excluding those with quasi-autistic patterns, with severe retardation, or with no experience of institutionalisation).

^c Numbers vary slightly as a result of missing data.

^d No results significant.

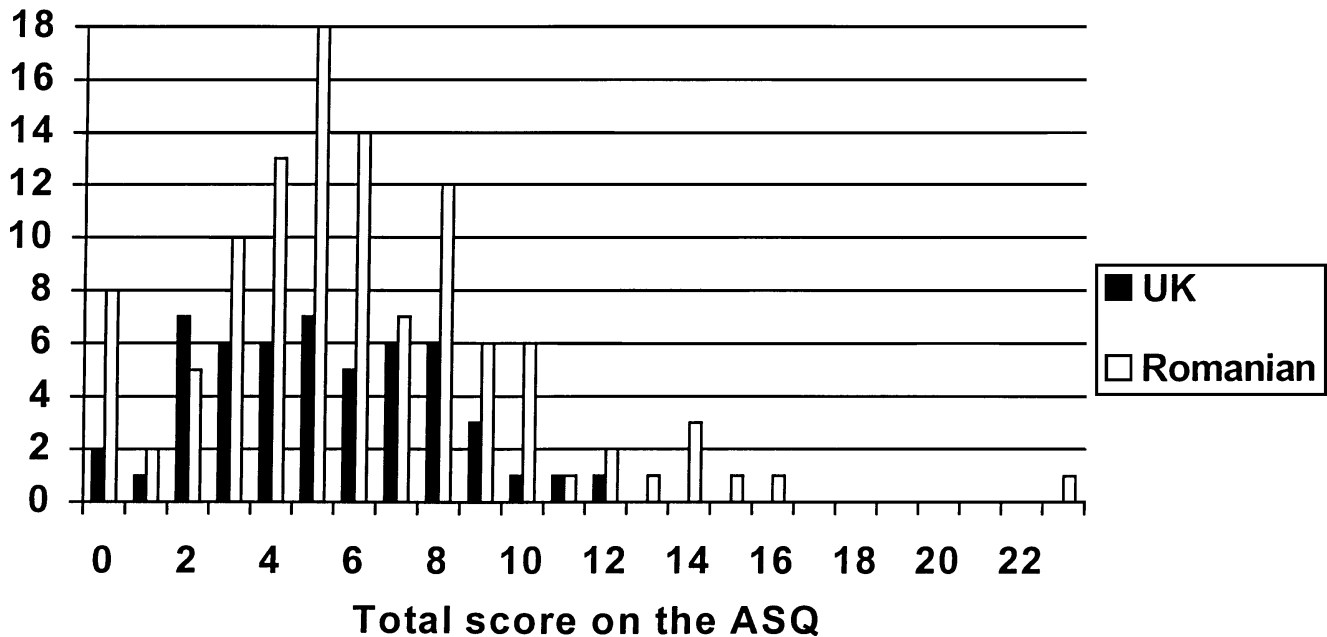


Figure 1. Scores on Autism Screening Questionnaire—Romanian and within-U.K. adoptees. Higher scores indicate greater prevalence of autistic features.

tribution of scores in the two groups (See Fig. 1) was broadly comparable, except that the Romanian sample included a small subgroup with high (deviant) scores not found in the within-U.K. adoptees sample. These occurred most frequently in the 11 children seen because of quasi-autistic features; of the 7 seen at age 4 years the mean score was 11.3 (*SD* = 6.4) compared with 5.7 (*SD*

= 3.3) in the remainder of the Romanian adoptees, and 5.2 (*SD* = 2.8) in the comparison sample. The first difference fell just short of statistical significance and the second was significant at the 5% level ($t = 2.28, p < .07$, and $t = 2.46, p < .05$, respectively).

Altogether, 6 of the 111 Romanian adoptees had a total score on the ASQ of 14 or more (suggesting a possible

pervasive developmental disorder) compared with none of the within-U.K. adoptees (exact test, $p = .10$). As Fig. 1 shows, in this particular sample, the point of no overlap between the groups came at a score of 13 or more, with 7 out of the 111 Romanian adoptees having that score or greater compared with none of the within-U.K. adoptees ($p = .06$). Of the six children with ASQ scores of 14 or more, all but one was seen by MR, and three were regarded as showing an autistic pattern. The remaining three were all regarded as showing milder autistic features.

The same questions were addressed with the ASQ findings at age 6 years. In both groups, the mean score was slightly lower at 6 years than it had been at 4 years, but the means were virtually identical in the two groups (4.3 vs. 4.2). As at 4 years, however, the ASQ scores of the children in the quasi-autistic group (including the milder patterns) who had not been assessed at age 4 stood out as higher. Of the six, three had scores of 14 or greater. Of the three children who showed autism associated with severe cognitive impairment (all being in the later age of entry subgroup), two had scores of 14 or more.

Thus, the ASQ findings are consistent with the reality of a subgroup of Romanian adoptees with a categorical pattern of quasi-autistic behaviour, a pattern not seen in the within-U.K. adoptees, but provide no indication that lesser degrees of the pattern are more widely distributed in the Romanian sample as a whole.

Discussion

The finding that some 6% of a group of 111 children from Romania adopted into English families showed a pattern of behaviour that closely resembled childhood autism is remarkable. The resemblance to autism was clearly demonstrated through the use of a well-validated, reliable, standardised diagnostic interview employed by an investigator very well experienced in its use and in the clinical manifestations of autism. It is evident, too, that the number of cases is far too high to be dismissed as a coincidence. The first question to be addressed, then, is whether the syndrome seen in these children constitutes childhood autism or, rather, a different clinical disorder that just happened to mimic it. That is not an easy question to answer because of the heterogeneity of the clinical manifestations of autism and the uncertainty over the extent to which the genetically influenced phenotype extends beyond the handicapping disorder of autism as traditionally diagnosed (Bailey et al., 1996).

It is tackled most appropriately by considering separately the three children who showed the autism associated with severe cognitive impairment and the seven children without severe retardation who showed quasi-autistic patterns. There is no strong reason not to regard the clinical picture in the first group as autism. All three children exhibited behaviour that met the full ADI-R algorithm criteria for autism at both 4 and 6 years of age, and all clearly had a handicapping pervasive developmental disorder. Because all these children had been admitted to institutional care in the neonatal period, the possibility that they were placed in institutions because of a previously recognised handicapping condition can be firmly ruled out. Similarly, it is implausible that the

presence of 3 such cases in a sample of 165 was a chance occurrence given the population base rate of autism somewhere in the range of 0.04 to 0.2% (Bryson, 1996; Fombonne, 1998). The question that follows, therefore, is what may have caused the autism. The presence of severe retardation together (in two cases) with a head circumference at follow-up that was still below the third percentile (despite weight and height within the normal range) provides circumstantial evidence suggesting organic brain dysfunction. It is known that autism, often with atypical features and associated severe mental retardation, can sometimes occur on the basis of non-genetic brain damage (Gillberg & Coleman, 1992; Rutter, Bailey, Bolton, & Le Couteur, 1994). Although there were some possibly important atypical features, the clinical impression, too, was of the syndrome of autism.

Did the syndrome arise, therefore, on the basis of some form of organic brain dysfunction or did the very adverse rearing conditions play a role? In all three cases the period of institutional care was prolonged (21, 30, and 39 months) and in two instances it was exceptionally poor. One child had been kept isolated in a single room and another was in an orphanage in which a large number of children are known to have died for reasons unknown in the 6 months prior to the child's coming to the U.K. The latter child also had a major high tone hearing loss and, at the time of U.K. entry, was extremely malnourished, weighing a mere 7.4 kg at the age of 30 months (6 *SD* below the U.K. mean). The third child was also unusual in terms of the report that he had been born at 26 weeks gestation with a birthweight of 2 lbs and had required intensive care in the neonatal period (details unknown). The risk of brain damage given those circumstances is quite high, particularly if the quality of paediatric care is suboptimal (Grether, Nelson, Stanley Emery, & Cummins, 1996).

It is not possible to do more than speculate on the aspects of privation that created the risk. Exceptionally severe malnutrition in the one case may have been important but, although both the other two children had low weights and heights at the time of U.K. entry, neither was below the third percentile and they were far from extreme within the standards of the adoptees from Romania. Animal studies have suggested that extreme stress leading to prolonged hypercortisolaemia can lead to neuronal, particularly hippocampal, damage and it is possible (although as yet unproved) that the same could apply to humans (see O'Brien, 1997). Severe sensory privation in early life can impede normal brain development (Blakemore, 1991; Greenough, Black, & Wallace, 1987) and social privation might also impair developmental programming for aspects of psychological functioning relevant to the genesis of autism (see Goodman, 1994). The unrecognised high tone hearing loss in one child was probably also a contributor, as suggested by the improvement that accompanied correction of the hearing deficit.

The further question that arises with these three children is whether their extremely deprived early rearing, together with the other unusual risk features, means that their prognosis is different from that which ordinarily applies to autism. Although no firm conclusions are possible at this point, the presence of some atypical

clinical features that mirrored those in the children showing what we have called a “quasi-autistic” pattern, together with the considerable (albeit limited) improvement shown by one of the three children, suggests that the outlook for progress may possibly be somewhat better than usual despite the degree of cognitive impairment at 6 years of age. Nevertheless, further follow-up is needed to determine whether or not that is actually the case.

The situation with respect to the children without severe mental retardation who showed an autistic-like pattern of behaviour is rather different. At the age of 4 years, their mean ADI algorithm scores did not differ from those in a similar-aged sample of autistic children studied by Lord et al. (1997), and the possibility of a diagnosis of autism had been raised very reasonably by the professionals involved in the care of four of them. Even at that age there were some atypical features and only two out of the six fully met the ADI algorithm criteria for autism across all three of the key domains of symptomatology. The atypicality of the autistic pattern, however, became much more strongly evident between the ages of 4 and 6 years. In sharp contrast to Lord et al.'s (1997) sample of autistic children, their autistic features lessened markedly over this 2-year span. By 6 years, only one of the six met the full algorithm criteria for autism, although all of this subgroup still stood out (from the remainder of the Romanian sample, from the within-U.K. adoptees, and from the general population) as showing unusual autistic-like features, albeit to a much milder degree than when younger. In comparison with the remainder of the group of adoptees from Romania, the six children with an autistic-like pattern also differed in showing a greater rise in their overall cognitive level.

The question of whether the syndrome shown by these children should be termed autism needs to be considered. The atypical course, with marked improvement between 4 and 6 years, is sufficient to cast doubt on the appropriateness of this diagnosis. Even with a sample as small as six, the differences from Lord et al.'s (1997) autistic group were statistically significant. The number of children is too low for any firm conclusions on the details of behaviour that signal atypicality. The features that stand out clinically, however, were the relatively high level of social approach (within the context of poor reciprocity, a deviant style, poor appreciation of social cues, and a lack of normal social boundaries) and the elements of indiscriminate friendliness observed in other children reared in institutions. The term “indiscriminate friendliness” is, however, perhaps a bit misleading both because most of the children *did* show discrimination in their responses to people, and because “friendliness” doesn't quite pick up the special quality of a lack of awareness of social boundaries and a poor appreciation of social cues. The communicative abnormalities stand out in a somewhat similar fashion. That is, although their communication was indubitably abnormal in style (in amount of reciprocal interchange and in lack of conversational flow), the extent of spontaneous usage, especially social usage, of both spoken language and sign language was somewhat unusual in relation to the typical picture in autism. The abnormalities were striking when the children were compared with normal children but the areas of positive functioning were probably clinically

important when the comparison was with children with autism.

The repetitive stereotyped behaviours were, in some respects, the most typically autistic of the features shown. Intense circumscribed interests and/or abnormal pre-occupations were characteristic. By contrast, motor stereotypies were less evident (despite the fact that, as with many of the children from Romanian institutions, rocking was a feature when they first came to the U.K.).

For all these reasons, the syndrome is better described as “quasi-autistic” in order to highlight the atypicality. A further question arises as to whether this pattern occurs in a present/absent categorical fashion or whether the features are found in most of the children from Romanian orphanages to a greater or lesser degree. The latter possibility seems unlikely on the basis of the distribution of ADI questionnaire scores, on the interview data for the sample as a whole, and on the findings on the other 18 children seen by MR. On the other hand, the former is not quite the case either. There were a further 10 children out of the 165 for whom either the ADI questionnaire scores or the interview data from the research interviews suggested milder, usually isolated, autistic features. Three of these children were seen by MR and in no case were the autistic-like features sufficiently marked to justify inclusion in the quasi-autistic behaviour group. Nevertheless, the *quality* did seem similar even though the degree was very much milder. On the other hand, the isolated autistic features usually involved stereotyped repetitive behaviour or circumscribed interests of some kind (rather than social and communicative impairment), the children had not come to the U.K. at an older age, and they did not show cognitive impairment (in all these aspects they differed from the group with quasi-autistic patterns). It may be concluded that the pattern of quasi-autistic behaviour applies only to a minority of the children, about 1 in 16. There is a further 1 in 16 with milder autistic features but it remains uncertain whether their behaviour is a milder version of the same thing or rather something a bit different. The ASQ findings, however, suggest that autistic-like features are *not* a feature of most of the children adopted from Romania. The only qualification that needs to be added is that the ASQ provides a much stronger measure of social and communicative abnormalities than it does of repetitive behaviours. Also, the ASQ provides only a limited coverage of circumscribed interests (Berument et al., 1998).

It remains to consider the origins of these quasi-autistic patterns. Because they were not found in any of the within-U.K. adoptees, and because the proportion of children from Romania with this pattern was as high as it was, some sort of causal connection with the children's adverse experiences in early life must be inferred. Unlike the situation with the three children showing severe cognitive impairment in addition to their autism, there are no strong grounds for inferring brain damage. The malnutrition experienced by the majority of the children from Romanian institutions may have played a contributory predisposing role but the subgroup of seven showing a quasi-autistic pattern were *not* more malnourished than other children from Romania. It seems unlikely that malnutrition constituted the main causal

factor. Also, although not systematically studied, autistic-like patterns have not been noted in any of the many previous investigations of malnourished children.

Some other aspect of the children's early experiences seems likely to be responsible. It is perhaps noteworthy that all of the children with this quasi-autistic pattern had experienced an institutional upbringing. Although that was true for the majority of the rest of the sample from Romania, there were 18 out of 100 children where that was not the case. The six differed from the remainder in being significantly older at the time of leaving institutional care to come to the U.K., and all were at least 12 months of age when they left institutions.

In considering the possible causes of the quasi-autistic pattern, three possible leads are available. First, the impaired social relationships that constituted part of the pattern had something in common with the so-called "indiscriminate friendliness" noted in other children from Romanian institutions both by us (Keaveney & O'Connor, 1997; O'Connor et al., in press) and by others (Chisholm et al., 1995). In both instances, impaired social reciprocity, poor appreciation of social cues, and a lack of awareness of social boundaries were striking. A few of these nonautistic indiscriminately friendly children from Romania also showed either a preoccupation with sensations or circumscribed interests, although their behaviour was not otherwise stereotyped or repetitive. The possibility that the pattern constitutes some form of deviant development of attachment relationships needs to be considered. Insofar as that may be the case, the marked degree of the children's lack of play, conversation, or even personalised caregiving from adults in the institutions may be invoked as likely to be relevant.

Second, the children with the quasi-autistic pattern stood out from the remainder of the adoptees from Romania with respect to their much greater degree of cognitive impairment—some 32 points on the McCarthy GCI at age 6 years. This suggests a somewhat different set of causal features. The cognitive impairments found in institution-reared children showing poor social relationships have generally been quite minor (Hodges & Tizard, 1989a, b) and the environmental factors associated with intellectual deficits tend to be somewhat different (Rutter, 1985). It may be inferred that the extreme lack of active experiences, social and nonsocial, as reflected in the confinement to cots, the lack of toys, and the infrequency of talk from adults, may have been influential.

The third lead is provided by the possible parallels with the autistic-like features described in congenitally blind children by Brown, Hobson, and Lee (1997). They found that autistic symptoms were much more common than in sighted children and that this was particularly so in those with associated mental retardation, but that, although the autistic qualities were sometimes marked, they were also often somewhat atypical. They postulated that the autistic-like features arose as a result of a failure in psychological perspective-taking deriving from a lack of visual experience. It is noteworthy, however, that the combination of blindness and cognitive impairment seemed to be important. It could be that the profound lack of social and perceptual experiences in the Romanian institutions played a comparable role.

In summary, the rate of autistic features was markedly

raised in this sample of severely deprived children from Romania; about 6% showed an overall quasi-autistic pattern and a similar proportion showed isolated autistic features. The characteristics of these children with autistic features, although phenomenologically similar in some respects to those found in "ordinary" autism, differed sharply in the marked improvement evident between 4 and 6 years of age and in the degree of social interest. It is also noteworthy that, in contrast with "ordinary" autism, the sex ratio was approximately equal and the head circumference was not raised (see Woodhouse et al., 1996). The quasi-autistic pattern seemed to be associated with a prolonged experience of perceptual and experiential privation, with a lack of opportunity to develop attachment relationships, and with cognitive impairment. Associated severe cognitive impairment was present in only a few instances—usually when there were additional biological risk factors. Although the rate of autistic features was clearly greatly raised in relation to general population norms, it is noteworthy that the great majority of the children who experienced severe psychological and nutritional privation did *not* show autistic features.

Acknowledgements—The study was supported by the Medical Research Council and by a grant from the Department of Health. We are very grateful to the families for their great cooperation during all phases of the study, and to the many other people who have provided us with information. We are very indebted to Karen Langridge, the study administrator, for help in ways too numerous to detail.

References

- Bailey, A., Phillips, W., & Rutter, M. (1996). Autism: Towards an integration of clinical, genetic, neuropsychological, and neurobiological perspectives. *Journal of Child Psychology and Psychiatry Annual Research Review*, 37, 89–126.
- Berkson, G., Gutermuth, L., & Baranek, G. (1995). Relative prevalence and relations among stereotyped and similar behaviors. *American Journal on Mental Retardation*, 100, 137–145.
- Berument, S., Rutter, M., Bailey, A. J., Pickles, A., & Lord, C. (1998). *Autism Screening Questionnaire: Diagnostic validity study*. Manuscript under review.
- Blakemore, C. (1991). Sensitive and vulnerable periods in the development of the visual system. In G. R. Bock & J. Whelan (Eds.), *The childhood environment and adult disease*. Ciba Symposium no. 156 (pp. 129–154). Chichester, U.K.: Wiley.
- Bowlby, J. (1946). *Forty-four juvenile thieves: Their characteristics and home life*. London: Bailliere, Tindall & Cox.
- Brown, R., Hobson, R. P., & Lee, A. (1997). Are there autistic-like features in congenitally blind children. *Journal of Child Psychology and Psychiatry*, 38, 693–703.
- Bryson, S. E. (1996). Brief report: Epidemiology of autism. *Journal of Autism and Developmental Disorders*, 26, 165–167.
- Cassidy, J., & Marvin, R. S. (1992). *Attachment organization in the preschool child: Procedure and coding manual*. Unpublished manuscript.
- Chisholm, K., Carter, M. C., Ames, E. W., & Morison, S. J. (1995). Attachment security and indiscriminately friendly behavior in children adopted from Romanian orphanages. *Development and Psychopathology*, 7, 283–294.
- Clarke, A. M., & Clarke, A. D. B. (Eds.). (1976). *Early experience: Myth and evidence*. London: Open Books.
- Curtiss, S. (1977). *Genie: A psycholinguistic study of a modern-day "wild child"*. London: Academic Press.
- Elander, J., & Rutter, M. (1995). Use and development of the

- Rutter Parents' and Teachers' Scales. *International Journal of Methods in Psychiatric Research*, 5, 1–16.
- Fombonne, E. (1998). Epidemiological surveys of infantile autism. In F. Volkmar (Ed.), *Autism and pervasive developmental disorders. Monographs in Child Psychiatry No. 2.* (pp. 32–62). Cambridge: Cambridge University Press.
- Frankenburg, W. K., van Doorninck, W. J., Liddell, T. N., & Dick, N. P. (1986). *Revised Denver Prescreening Developmental Questionnaire (R-PDQ)*. High Wycombe, U.K.: DDM Incorporated/The Test Agency.
- Gillberg, C., & Coleman, M. (1992). *The biology of the autistic syndromes* (2nd ed.). London: Mac Keith Press.
- Goodman, R. (1994). Brain development. In M. Rutter & D. Hay (Eds.), *Development through life: A handbook for clinicians* (pp. 49–78). Oxford: Blackwell Scientific.
- Greenough, W. T., Black, J. E., & Wallace, C. S. (1987). Experience and brain development. *Child Development*, 58, 539–559.
- Grether, J. K., Nelson, K. B., Stanley Emery, E., & Cummins, S. K. (1996). Prenatal and perinatal factors and cerebral palsy in very low birthweight infants. *The Journal of Pediatrics*, 128, 407–414.
- Harlow, H. F., & Harlow, H. K. (1972). The affectional systems. In A. Schrier, H. F. Harlow, & F. Stollnitz (Eds.), *Behaviour of non-human primates, Vol 2*. New York: Academic Press.
- Hodges, J., & Tizard, B. (1989a). IQ and behavioural adjustment of ex-institutional adolescents. *Journal of Child Psychology and Psychiatry*, 30, 53–75.
- Hodges, J., & Tizard, B. (1989b). Social and family relationships of ex-institutional adolescents. *Journal of Child Psychology and Psychiatry*, 30, 77–97.
- Kaler, S. R., & Freeman, B. J. (1994). Analysis of environmental deprivation: Cognitive and social development in Romanian orphans. *Journal of Child Psychology and Psychiatry*, 35, 769–781.
- Keaveney, L., & O'Connor, T. (1997). *The network of social relations of adoptees exposed to severe early deprivation*. Paper presented at the Society for Research in Child Development Biennial Conference, Washington, DC, April.
- Kreppler, J., O'Connor, T., Dunn, J., Anderson-Wood, L., & the English and Romanian Adoptees (ERA) Study Team. (in press). The pretend and social role play of children exposed to early severe deprivation. *British Journal of Developmental Psychiatry*.
- Le Couteur, A., Rutter, M., Lord, C., Rios, P., Robertson, S., Holdgrafer, M., & McLennan, J. (1989). Autism Diagnostic Interview: A standardized investigator-based instrument. *Journal of Autism and Developmental Disorders*, 19, 363–387.
- Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview-Revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders*, 24, 659–685.
- Lord, C., Shulman, C., Pickles, A., & DiLavore, P. C. (1997). *Learning and not learning to speak: Examples from a longitudinal study of preschool children with autism-spectrum disorder*. Paper presented at the Society for Research in Child Development Biennial Conference, Washington, DC, April.
- McCarthy, D. (1972). *The McCarthy Scales of Children's Abilities*. New York: The Psychological Corporation/Harcourt Brace Jovanovich.
- Novak, M. A. (1979). Social recovery of monkeys isolated for the first year of life: In long-term assessment. *Developmental Psychology*, 15, 50–61.
- O'Brien, J. T. (1997). The "glucocorticoid cascade" hypothesis in man. Prolonged stress may cause permanent brain damage. *British Journal of Psychiatry*, 170, 199–201.
- O'Connor, T. G., Bredenkamp, D., & Rutter, M. (in press). Attachment disturbances and disorders in children exposed to early severe deprivation. *Infant Mental Health Journal*.
- Ridley, R. M. (1994). The psychology of perseverative and stereotyped behaviour. *Progress in Neurobiology*, 44, 221–231.
- Rutter, M. (1981). *Maternal deprivation reassessed* (2nd ed.). Harmondsworth, U.K.: Penguin.
- Rutter, M. (1985). Family and school influences on cognitive development. *Journal of Child Psychology and Psychiatry*, 26, 683–704.
- Rutter, M., Bailey, A., Bolton, P., & Le Couteur, A. (1994). Autism and known medical conditions: Myth and substance. *Journal of Child Psychology and Psychiatry*, 35, 311–322.
- Rutter, M., & the English and Romanian Adoptees (ERA) Study Team. (1998). Developmental catch-up, and deficit, following adoption after severe early deprivation. *Journal of Child Psychology and Psychiatry*, 39, 465–476.
- Rymer, R. (1993). *Genie: An abused child's fight for silence*. New York: Harper/Collins Publishers.
- Skuse, D. (1984a). Extreme deprivation in early childhood-I. Diverse outcomes for three siblings from an extraordinary family. *Journal of Child Psychology and Psychiatry*, 25, 523–541.
- Skuse, D. (1984b). Extreme deprivation in early childhood-II. Theoretical issues and a comparative review. *Journal of Child Psychology and Psychiatry*, 25, 543–572.
- Stutsman, R. (1931a). Guide for administering the Merrill-Palmer Scale of Mental Tests. In L. M. Terman (Ed.), *Mental measurement of preschool children*. New York: Harcourt, Brace & World.
- Stutsman, R. (1931b). *Mental measurement of preschool children*. New York: World Books.
- Woodhouse, W., Bailey, A., Rutter, M., Bolton, P., Baird, G., & Le Couteur, A. (1996). Head circumference and other pervasive developmental disorders. *Journal of Child Psychology and Psychiatry*, 37, 665–671.

Manuscript accepted 2 September 1998

Appendix

Case 1: First words with meaning at 24 months, phrases by 30 months. Some tendency to line up objects and very little pretend play. Lost language after using phrases for several months and remained without language for about 6 months. Screaming and pointing (with appropriate use of eye-to-eye gaze), increase in aggression, very poor concentration, became very fearful of baths, did not smile, increased tendency to line up objects with walking around the lines in repetitive fashion. No loss of nonlanguage skills and continued to follow mother around the house. Attended speech therapist at about 3 years, quickly

learned makaton and began to use spoken language again, developed imaginative play and progressed well in socio-emotional development generally. When learning language, tended to use formula phrases for a while but then progressed to normal conversational chattiness. By age 5 years appeared psychologically normal with normal emotional expressiveness and social interaction. Normal intellectual level.

Case 2: When in orphanage at 16 months unable to sit up. When came to U.K. 4 months later sitting in unbalanced way.

Very apathetic. Started to walk at 30 months but walking and running still abnormal at 6 years; splints worn to prevent persistent toe walking (because of concern re possible tendon shortening). Attends school for children with severe learning difficulties. Still without speech at 6 years but spontaneous use of some 12 makaton signs accompanied by direct gaze and clear communicative intent, pats person's hand or makes noise to indicate that wants to sign. Tends to accost strangers looking intently and showing affection, but otherwise poor eye contact. No play with other children. Much hand flapping and finger mannerisms, grimacing, and turning self around. Repetitive stereotyped throwing of objects at faces; repetitive pushing of doors and switching lights on and off. Persistent smelling of objects. Mostly unresponsive to spoken voice; puts hands over ears with certain noises. Doesn't greet; doesn't come for cuddle if hurt; limited range of emotional expression but smiles, laughs, and claps hands to show pleasure, some appropriate social overtures but very impaired social reciprocity. Severe cognitive impairment.

Case 3: When first came to U.K. after third birthday, walking only in unsteady fashion; had to be spoon fed with liquidised foods but could drink from a cup; much rocking and head banging; frequent touching of walls and running fingers around tiles; very quiet and scarcely ever cried. Walking rapidly improved over 2–3 months. At about 5 years presented with characteristic picture of autism associated with severe cognitive impairment. Recurrent screaming. Over the next 18 months substantial progress in cognitive and social-communicative functioning but still severe developmental retardation. At 6½ years using both words and signs to indicate needs by very limited vocabulary and communications strictly need-oriented. Increase in social interaction with family, marked reduction in screaming; mood preponderantly happy, and gain in self-help skills. Much social approach to parents; lies on their laps; touches father's moustache but poorly modulated and integrated social interactions; no greeting; no bringing of objects to people; no constructive play; generally compliant. Very attached to beads and string and fiddles with them. Very agile.

Case 4: When came to U.K. at 2½ years, markedly underweight; crawling and just beginning to walk; no language; no eye-to-eye gaze. During next 6 months severe difficulties in feeding; much rocking; screamed when presented with toys; severe intestinal infection. Play gradually improved, did jigsaws with encouragement; preoccupied with Hoovers, wires, plugs, and exhaust pipes; teeth grinding; little social reciprocity. Hearing impairment discovered and hearing aid fitted about age 4. Marked improvement over next 2 years as shown by increasing spontaneous use of makaton signs, increasing social interaction, the beginning of pretend play, and reduction in stereotyped interests. Retesting of hearing has shown that the hearing loss was high tone and change of hearing aid was accompanied by a much greater willingness to wear it. At 7 years still has marked difficulties in peer relationships and limited social reciprocity but much more social approach and interest with some appropriate use of social signals. Makes sounds to accompany signs but although vocalisations clearly communicative in intent, very difficult to understand. Habit of loud repetitive noises; still preoccupied with Hoovers. Substantial cognitive gains but remains intellectually impaired.

Case 5: When came to U.K. just before age 2 years, only just able to sit and severe global developmental retardation. No speech or vocalisations, lack of facial expression. Would only take milk from bottle, and normal eating not achieved until some 18 months later. Initially would only sit on settee and any attempt to move the child led to panic. At 3½ years could not be got to leave house unless carried, any new situation or experience tended to provoke extreme anxiety and avoidance, but at 4 years beginning to respond to mother's reassurance.

Flapping of hands, spinning, flicking of fingers near eyes, wrist biting, staring at lights and ceiling. Walking by age 3 years but very unsteady. Indicated needs by taking someone else's hand and using it as an extension of own, without use of eye-to-eye gaze. Started to learn sign language at 4 years, and after 9 months beginning to put signs together. At 4 years no greeting or social smiling, no bringing of objects to parents. Intense interest in spectacles. Over next 2 years marked progress but with substantial setback associated with a change of school and by 6½ years vocabulary of some 40 to 50 words used in conjunction with makaton. Constructive play. Social approach with some teasing and playfulness but socially disinhibited (touching people's faces and spectacles); also much more impaired in social interactions with people outside the family than with parents. Mild learning difficulties.

Case 6: When came to U.K. at 18 months, socially detached and unresponsive, unable to walk, without language. First words at about 33 months, with a vocabulary of some 30 words by 3 years and the beginning of phrase speech by 43 months. At 4½ years spontaneous chat with flexible use of words, much social approach but unusual style of interaction, didn't bring objects to show to parents, didn't share, didn't join in peer group, lacked pretend play and didn't pick up social cues but affectionate with parents. Unusual preoccupation with characters in stories. Hand flapping, teeth grinding and toe walking. Very clumsy gait. By 6½ years spontaneously talkative and wanting to communicate but tendency to intrude into conversations and talk lacks reciprocity. Still lacks appreciation of social situations and social boundaries, poor at picking up social cues and doesn't joint in peer group but greets parents and likes to do things with them. Severe learning difficulties at 4 but only mild difficulties at 6 years.

Case 7: Came to U.K. just before third birthday, then well below the third percentile for weight, and only just beginning to walk very unsteadily. Would hold out arms to anyone but seemed to derive no comfort from being picked up. Fascination with lights; various hand and finger mannerisms; held objects close to eyes examining them minutely; rocking; facial tics. First words at 40 months; first phrases at 46 months. Period of pronominal reversal when learning to talk. No pointing to express interest. Put hands over ears with loud noises. Didn't smile and lacked eye-to-eye gaze. Was greeting parents by age 5, didn't come for comfort at that age but did so later. Gradual improvement in social interactions and in pretend play. At 7½ years intense circumscribed interests in planets and skeletons. Feels new objects with mouth or face. Full of spontaneous chat with social disinhibition and much social approach. Interested in other children but doesn't seem to know how to interact with them. Intelligence within normal range.

Case 8: Came to U.K. shortly after first birthday. Then sombre and stiff to hold. Some looking at faces but this lacked social engagement. Sitting and making noises but no babble. Phase of screaming before second birthday. First words at 30 months, phrases at 3 years. Tendency to use stock phrases and ask inappropriate questions; many made-up words; good mimic, lack of communicative reciprocity but much improved by 6½ years. Range of intense and unusual interests (including watches, spectacle frames, shiny clothes, and new £10 notes); preoccupied with sensations and will touch people without inhibition. Social relationships a complicated mix of confidence and dependency. No group play when younger but joining in with other children by age 6½ years. Lacks awareness of social boundaries. Speech fluent but with repetitive elements and limited conversational reciprocity. Mild learning difficulties.

Case 9: Came to U.K. at 18 months; sitting but not crawling; rocked; unresponsive to people; terrified of noisy equipment (such as lawn mower); very resistant to solid foods. During next

year showed hand flapping and twirling; preoccupation with light switches; and lack of social responsiveness and eye-to-eye gaze. First words at 33 months; first phrases at 42 months. Stereotyped phrases at age 4 years. By 5 years enjoying interactions with other children although lacked to and fro conversations with them. Much more socially responsive with parents and shares interests with them. Unusually intense circumscribed interest in plumbing and toilets. Marked improvement over next 18 months in that more communicative and more social but still a bit of a loner. Plenty of social chat, appropriate (although still somewhat limited) use of eye-to-eye gaze, affectionate; pretend play with peers; but still some difficulty picking up social cues. Circumscribed interests in plumbing, electricity and bodily systems still present at 6½ years but more varied and more socially integrated. Normal intelligence (although mild learning difficulties at 4 years).

Case 10: Came to U.K. at 16 months; then lacked eye contact, was socially unresponsive and resisted physical contact. First words at 30 months; first phrases at 4 years. Language still poor and unable to express self properly when started school at 5. No social chat and seemed oblivious of other children, although better with adults. During next year became indiscriminately friendly with adults, shouted out of the car to strangers and invited the postmen into the house. Talked on own topics, made inappropriate statements, lacked reciprocal conversation and had repetitive phrases. By 6½ years normal greeting, appropriate sharing, interested in other children but doesn't know how to interact with them, developed pretend play.

Case 11: Came to U.K. at about age 1 year. Responded indiscriminately to people; cuddly; fascinated by texture of materials; no communicative babble; watched objects but didn't play with them. First words at 20 months; first phrases at 26 months. Much echoing between 2 and 3 years. Little eye-to-eye gaze. At 4 years very limited communicative reciprocity; pronominal reversal; greeted parents and shared with them; affectionate; lacked social inhibitions and intruded into conversations; interested in other children but didn't know how to play with them. Preoccupations with clocks and watches; subsequently replaced by a period of fascination with doors and gates, opening and closing them repetitively and obsessively, hard to divert. Lined up cars and looked at them in unusual way. Variety of fears. Language much improved by 6½ years, although conversation still lacking in reciprocity. Also tendency to switch topics without warning. Puts things in unusual ways, goes off at tangents, and preoccupied with dates and ages. Socially improved but still apart from peers, shows pride in accomplishments. No shyness with strangers. Calendrical calculator and advanced mechanical reading skills. Occasional hand flapping. Poor concentration on school tasks but readily absorbed in special interest of computers. Rather clumsy. Normal intelligence.

Case 12: Intellectually functioning with mildly retarded range. Concerns re possible autism first raised in Romania; also later in U.K. by a special needs teacher. Used to walk in a rigid fashion with hands held in front. Obsessive about colours; had set of beads that had to be in specific order. Obsessed with a particular video that the child watches repetitively. Very attached to mother but tends to be detached with other people. Used to rock and head bang when first came to U.K.

Case 13: Intellectual functioning at borderline between normal and mild retardation. Possibility of autism queried by a

professional. Language very slow to develop but sudden spurt after age 5 years. Initially fascinated by lights, although not obsessively so. No interest in soft or cuddly toys. Used to roll and spin objects inappropriately. Rocks at night. Social difficulties in preschool years but by age 6 much improved, although still tendency to stand apart. Impaired reciprocal conversation. Some stereotypical hand movements.

Case 14: Average intellectual functioning. Possibility of autism queried by parents and by a professional. Markedly slow in language development, with only a few single words at 4 years. At about that age habit of examining hands, flicking fingers, rolling objects, repetitive use of toys, unusual sensory interests and rocking. Flicked other children's hair. Habit of jumping up and down. Much improved by age 6 but style of social interaction still unusual and motor mannerisms still present and some unusual ways of speaking.

Case 15: Averaging intellectual functioning. Unusual interest in curly hair and prominent foreheads; when younger associated with socially inappropriate touching of other people. Stereotyped routine of running or walking up and down with associated hand mannerisms only mildly evident by age 6. When younger, fear of plastic surfaces; also tendency to be overfriendly with strangers. Some concentration difficulties.

Case 16: Intellectual functioning at borderline between normal and mild retardation. Disinhibited socially with reduced awareness of social boundaries. Difficulty in concentration and in responding to group tasks. Especially when younger, idiosyncratic negative response to babies crying, tendency to injure self, and hand/finger mannerisms. Afraid of dogs and loud sounds. Difficulties in peer relationships. Interested in tactile sensations of hair and of fur or silk. Tendency when younger to touch people's faces. Also when younger, habit of tapping electric plugs, people's spectacles or teeth. Tendency to rock.

Case 17: Normal intelligence but experiencing difficulties at school. Used to rock but no longer did so at 6 years. Query of autism raised by nursery nurse because of jerky hand flapping mannerisms. Lack of normal social wariness. Chatty and friendly but socially unusual with impression of remoteness.

Case 18: Average intellectual functioning. Rather obsessive about ordering of shapes and colours. Preoccupied with particular video and cartoon character. Attributes human emotions to inanimate objects. Some repetition of phrases. Friendly and sociable at age 6.

Case 19: Average intellectual functioning. Language rather repetitive and slightly odd at age 4. Slow and precise speech at age 6. Habit of loud screeching noises. Finds social demands of school difficult and problem in making friends. Tendency for communication to be influenced by habit of associating objects/concepts.

Case 20: Average intellectual functioning. Fascinated by videos and photographs. Hand and finger mannerisms and unusual sensory interest.

Case 21: Average intellectual functioning but difficulties with both reading and arithmetic. Initially, after first coming to the U.K. at about 1 year of age, socially unresponsive, not using eye-to-eye gaze, not greeting, and not reacting when left. Severe tantrums, aggression, and defiance from second year. Wrist biting and head banging at about age 2½ years; still occasionally occurs at 6½ years. Several intense interests (e.g. a particular make of car), endless questions about size and speed; fascinated by feel of peoples' ears (also dogs' ears). Poor appreciation of social cues and boundaries; markedly socially disinhibited, social "show-off", and indiscriminately friendly.