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The pathogenesis of autism: insights from congenital blindness

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There is substantial heterogeneity in the aetiology and clinical presentation of autism. So how do we account for homogeneity in the syndrome? The answer to this question will be critical for any attempt to trace the links between brain pathology and the psychological disabilities that characterize autism. One possibility is that the source of homogeneity in autism is not to be found 'in the child', but rather in dysfunction of the system constituted by child-in-relation-to-other. We have been exploring this hypothesis through the study of congenitally blind children, among whom features of autism, and the syndrome of autism itself, are strikingly common. To justify such an approach, one needs to establish that the clinical features in blind children have qualities that are indeed 'autistic-like'. We conducted systematic observations of the social interactions of two matched groups of congenitally blind children who do *not* have autism, rating their social engagement, emotional tone, play and language during three sessions of free play in the school playground. The qualities of social impairment in the more disabled children were similar to those in sighted children with autism. Additional evidence came from independent ratings of the children in a different play setting: on the childhood autism rating scale (CARS), the socially impaired children had 'autistic-like' abnormalities in both social and non-social domains. If we can determine the way in which congenital blindness predisposes to features of autism, we shall be in a better position to trace the developmental pathways that lead to the syndrome in sighted children.

Keywords: autism; blindness; social relations; intersubjectivity

1. INTRODUCTION

In this paper, we attempt to do three things. Our first aim is theoretical: we shall propose that to determine what makes autism a syndrome, it may be necessary to consider it as an interpersonal disorder. Second, we shall consider how research with congenitally blind children bears upon this thesis. Finally, we shall describe a formal exploratory study with non-autistic congenitally blind children that provides evidence for this account.

It is one of the striking things about autism, that it is both a relatively homogeneous and clinically valid constellation of clinical features, and a syndrome that has diverse aetiology and marked individual differences in clinical presentation. On a clinical-descriptive level, for example, Kanner's view was that each of his 11 cases were characterized by an 'inability to form the usual, biologically provided affective contact with people' (Kanner 1943, p. 250), and among a range of other characteristic abnormalities, those in the pragmatic aspects of language are almost universal (Tager-Flusberg 2000). On an epidemiological level, classic studies by Wing & Gould (1979) demonstrated that 'autism' really does exist as a triad of social impairments. However, despite evidence that genetic or other identifiable physical factors are important in a substantial number of children with autism, the goal of defining a common underlying physical substrate has proved elusive. On a psychological level, too, attempts to capture a dysfunction or set of dysfunctions that is universal to individual children, of early onset, and responsible for the characteristic pattern of clinical features, have met with only partial success. Even in those respects that have been most productive—and here, theory of mind approaches top the list (Frith 1989; Hobson 1993; Happe 1995; Baron-Cohen *et al.* 2000)—it remains unclear how far the children's limitations in understanding people's minds are the cause or the result of their abnormalities in non-verbal communication, or the cause or the result (or neither) of their ritualistic behaviour and relatively inflexible thinking.

There are two main alternatives to the idea that we should seek a single and specific underlying 'cause' for autism, whether on a physical or psychological level. The first is to reject the notion that there is a final common pathway to autism, and to suppose instead that the syndrome is the manifestation of several distinct areas of disability (e.g. Wing & Wing 1971; Goodman 1989). The second, equally radical alternative is to hold that there may be a final common pathway of psychological disorder to the syndrome, but to locate this essential factor in what happens or fails to happen *between* people. According to this hypothesis—which is emphatically not a return to the damaging psychogenic theories of earlier decades—there may be several different psychological abnormalities (as well as different neurological abnormalities and different underlying aetiological factors) in individual children with

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autism, but that whatever those abnormalities are, they interact with what the environment provides to result in a special kind of breakdown in social engagement between the affected child and others. It is this breakdown and its development sequelae that become manifest in the special 'autistic' quality of social and communicative impairment.

The claim here is that without taking into account the interpersonal quality and level of disorder, one will never arrive at a satisfactory theory of why the particular clinical features of autism co-occur in the way that they do. The claim is not that the interpersonal level underpins all the phenomena of autism. On the contrary, there will be 'lower-level' psychological abnormalities in most if not all cases, because there must be reasons why the disruption in social engagement is happening, and these abnormalities will have additional manifestations that may or may not be universal to autism. Perhaps the most obvious case in point is that brain pathology is often manifest in a degree of 'general' mental retardation, and one does not need to claim either that the general mental retardation is totally irrelevant in causing the autism (which it may or may not be, in any given case), nor that it results from social impairment (although this may exacerbate the cognitive impairment). What *is* being claimed is that the social impairment itself is a necessary and probably sufficient condition for the characteristic constellation of clinical features to develop over the early period of a child's life. There are central features of autism that are explicable in terms of 'lower-level' impairments only insofar as these operate through disrupting interpersonal engagement and interaction.

This kind of interpersonal account faces two immediate challenges. First, we need a more detailed specification of which aspects of interpersonal engagement are deficient in children with autism, and how these then give rise to at least some of the essential features of the syndrome. Here, the suggestion is that a young child needs emotional engagement and identification with the attitudes of other people not only to derive concepts of mind and to employ language with flexibility and context-sensitivity, but also to disembed from a one-track perspective *on the world* and to acquire the ability to symbolize in characteristically human ways; and that such emotional engagement and identification is seriously impaired in children with autism (see, for example, Hobson 1989, 2002; Hobson & Lee 1998, 1999). Second, we need to know just how much this account is meant to explain: how many of the characteristic abnormalities are supposed to be the developmental outcome of disorder that occurs in interpersonal transactions, and how many are spin-off deficits that arise from lower-order impairments that do not implicate this social level of explanation.

If one adopts the approach of developmental psychopathology, one is prompted not only to compare typical and atypical development (in the present case, 'normal' development and autism), but also to compare developmental processes and outcomes that are 'typically atypical' (in this case, classically autistic) with those that are 'atypically atypical'. If there are atypical forms of autism, their very unusualness may draw one's attention to otherwise neglected causal processes and psychological mechanisms in the pathogenesis of the syndrome. For example, autism may be observed in circumstances that (arguably) implicate relevant kinds of disruption in the system of child-in-relation-to-other, and restrict the critical kinds of childhood social experience. Two potential cases in point are children who early in life suffered terrible deprivation and privation in the orphanages of Romania (Rutter *et al.* 1999), and children who are congenitally blind.

There are special hazards in following this line of explanation. If one is drawing comparisons between features of typical and atypical autism, how similar is similar enough to justify such a comparison? Is it even permissible to think in terms of autism in this context, or should we confine ourselves to noting 'autistic-like' clinical features in atypical cases? The danger of the latter approach is that it seems to presuppose that there is a clear boundary in phenomenology and pathogenesis between 'typical' and 'less typical' instances of autism. If a child meets formal diagnostic criteria for the syndrome, then we should accept that child has the syndrome of autism *in certain important respects*. It is a subsidiary matter to tease out the ways in which the syndrome is atypical, for example with respect to particular clinical features or to natural history. Only in this way shall we recognize previously unrecognized diversity in more typical cases, and appreciate how there may be different routes to the syndrome and potentially at least, different routes by which the syndrome may evolve (and even partly remit) subsequently.

2. THE CASE OF CONGENITAL BLINDNESS

First, to state the obvious: even total congenital blindness is not sufficient to cause autism. The fact is that there are congenitally blind individuals who do not manifest features of autism (as illustrated later in this paper). However, there have been many clinical reports of autism or autistic-like conditions in children with congenital blindness (see, for example, Keeler 1958; Wing 1969; Chess 1971; Fraiberg & Adelson 1977; Rogers & Newhart-Larson 1989), and recent systematic investigations of relatively large groups of congenitally blind children reveal that a surprisingly high number—almost half the sample of 24 children between the ages of 3 and 9 years studied in special schools by Brown *et al.* (1997)—meet the formal diagnostic criteria for autism. Moreover, when Hobson *et al.* (1999) made close comparisons between a subgroup of the congenitally blind children with autism, and an age- and IQ-matched group of sighted children with autism, there were marked similarities and only suggestive evidence of group differences (especially in the less markedly 'autistic' quality of the blind children's social impairment). When it came to focus on the congenitally blind children without autism, systematic observations by Brown *et al.* (1997) revealed that they displayed a significantly greater number of 'autistic features' than matched sighted children; and in a separate study on different groups of non-autistic congenitally blind and matched sighted children, the blind children were significantly impaired on 'theory of mind' tasks (Minter *et al.* 1998; and see Hobson *et al.* (1997) for an overview of these studies).

The possibility arises that the 'effective environment' of congenitally blind children—that is, the environment as experienced by the children—may have conjoined with other factors in causing features of autism to develop in a substantial number of cases. However, we need to be criti-

cal in exploring this possibility. As Baron-Cohen (2002, p. 792) has recently remarked, '…might this be no more than a surface similarity? We should be careful not to assume that just because two church bells are ringing simultaneously they are causally connected by the same rope'. In addressing this challenge, one avenue of research is to explore the nature and neurofunctional basis of blind children's autistic-like psychological difficulties (e.g. O'Connor & Hermelin 1978). Another is to examine in more detail whether in congenitally blind children, there is coherence between an 'autistic-like' quality of social impairment—something beyond the kinds of difficulty in social relatedness one might expect in all blind children—and other clinical features of autism. Such study may enable us to discern whether there is an intrinsic link between the children's abnormal social relations and experience, and their other deficits.

3. THE PRESENT STUDY

There has been surprisingly little study of social interactions among children with congenital blindness. Apart from in-depth studies of the interactions between blind infants and their mothers (see, for example, Urwin 1983; Rowland 1983; Rogers & Puchalski 1984; Preisler 1991; Troster & Brambring 1992), most accounts of the social relations of young blind children have been contained in clinical-descriptive studies. In a report of young blind children in nursery school, Preisler (1993) (also Curson 1979; Sandler & Hobson 2001) described how the blind children seldom participated in sighted children's play or initiated contact with the other children, and there was little exchange of ideas or meanings. The play of blind children has also been described as impoverished and 'primitive', more often directed at adults than other children (Burlingham 1961; Wills 1968; Tait 1972*a*,*b*; Schneekloth 1989; Troster & Brambring 1994; Ferguson & Buultjens 1995; Skellenger *et al.* 1997). Not only do blind children rarely imitate others, except in the special case of vocalizations (Sandler & Wills 1965; Fraiberg 1977), but also they often appear muted in their affective expression (Burlingham 1961; Fraiberg 1968; Wills 1970, 1981) or reciprocal positive feelings to others (e.g. Kekelis 1992). Kekelis (1992) describes how the children may be preoccupied with their own thoughts and actions, abruptly shift topics of conversation, and pay little attention to other people's points of view, interests, language or other behaviour (see also Chernus-Mansfield *et al.* 1985; Andersen & Kekelis 1986; Skellenger *et al.* 1992).

In the extreme case, as we have seen, congenitally blind children may present with 'autistic-like' clinical features or with a more or less full picture of autism. But it may be argued that in those blind children with the syndrome of autism, the social impairment is simply a reflection of coincidental autism: there need be no intrinsic connection with the lack of visual input. This argument is less persuasive because one finds a spectrum of severity of 'autistic features' in blind children. Therefore special interest is attached to the clinical presentation of socially impaired blind children who are *not* classically autistic. Is there evidence that in these children, the social impairment is (i) like that of sighted children with autism and (ii) associated with other features of autism? If so, then perhaps there is some intrinsic connection between blind children's social impairment and their 'autistic-like' clinical features: in this case, the connection may also have a bearing on the pathogenesis of the full syndrome when it occurs in blind children; and if this is so, there may be lessons to be learnt for what leads to autism in sighted children.

In our study of these issues, we needed to establish that the qualities of the social and other impairments under review were not simply a reflection of behavioural strategies common to all children who are congenitally blind, nor a reflection of low IQ in the context of blindness. Therefore we constituted two IQ- and age-matched groups of congenitally blind children according to teachers' reports of their abilities to engage with others. The MS blind children served as a control group for those who were socially impaired (LS children). This allowed us to explore a matter that has not been addressed previously: within the population of congenitally blind children who do not have autism, is there a specific association between autistic-like social impairments and autistic-like non-social abnormalities when the children's age and IQ are taken into account?

We adopted two approaches to evaluating the children's social impairments. The first approach was to observe the children in free play in the school playground. Our observational technique and rating procedures drew on the approaches of several earlier workers such as Rubin *et al.* (1976), Connolly & Doyle (1984) and Guralnick & Groom (1987). Our interest focused on the quality and emotional tone of the children's social engagement, the types and sociability of their play, and the social and pragmatic aspects of their language use. Our predictions were that the LS children would contrast with the MS children in having more periods in which they were isolated and relatively unexpressive ('placid') emotionally, and in which they would fail to show play and more specifically, fail to engage in reciprocal play. On ratings of language use, we predicted that the LS group would show fewer periods of language directed towards other children, and make fewer utterances to others involving comments on things or events.

Our second approach was to invite an independent judge who was unaware of group constitution to rate videotapes of the children engaged in play with someone. This rater employed the CARS of Schopler *et al.* (1988) to assess the degree to which children displayed both social *and* non-social abnormalities that were 'autistic-like' in quality.

(a) *Participants*

Participants were 18 congenitally blind children selected on the basis that they were between 4 and 8 years of age (inclusive), they did *not* satisfy DSM-IV criteria for autism, they were not exhibiting high degrees of repetitive mannerisms which might have prevented interactions in the free-play settings, they had an IQ above that of severe learning disability (an IQ of 55), and finally, they fell into the appropriate subgroups according to teacher ratings of social ability. Nearly all of the children had been totally blind from birth; the exceptions were two of the MS children and two of the LS children, each of whom had light perception only. None of the children had been in their present nursery school for less than a year, so it was

Table 1. Participant characteristics.

child	CA (months)	IQ	MA (months)	diagnosis	teachers' 'social' ratings (max. = 5)
more social group					
1	107	57	61.0	optic atrophy hydrocephalus	5
2	76	96	73.0	retinopathy of prematurity	5
3	72	104	74.9	retinopathy of prematurity	5
4	93	87	80.9	retinopathy of prematurity	4.5
5	75	109	81.7	microphthalmia[a] (prostheses)	5
6	98	85	83.3	retinopathy of prematurity	4
7	96	101	97.0	uncertain: optic pathway disorder	4
8	90	115	103.5	retinopathy of prematurity	4.5
9	101	117	118.2	retinal aplasia	5
mean	89.8	96.8	85.9		4.7
s.d.	12.6	18.6	17.4		0.4
less social group					
1	76	65	49.4	retinopathy of prematurity	2
2	63	89	56.1	retinopathy of prematurity	2
3	102	62	63.2	congenital optic nerve hypoplasia	1.5
4	96	72	69.1	retinopathy of prematurity	3
5	76	106	80.6	Leber's amaurosis	2.5
6	85	96	81.6	Leber's amaurosis	2.5
7	109	85	92.6	Leber's amaurosis	3
8	104	100	104	Leber's amaurosis	3
9	113	112	126.6	Norries disease	3
mean	91.6	87.4	80.4		2.5
s.d.	17.3	17.9	24.5		0.6

[a] Isolated condition: not part of a wider syndrome or association.

unlikely that their behaviour reflected adjustment to a new school.

For teacher ratings, two qualified class teachers who knew each of 25 children were asked to fill in a questionnaire which included the question: 'on a scale of 1–5, how would you rate this child's behaviour in the ability to relate to adults and peers (rated separately), establishing normal mutual interpersonal contact with them?' The threshold at which children qualified for the socially impaired (LS) group was set at a mean score across adult and peer ratings of equal to or less than 3, with neither of the teachers' ratings higher than 3 for the child's relations with either adults or peers. Nine children met these criteria. We selected a corresponding group of nine MS children on the basis that they were similar in age and achieved the highest scores (4 or more) on the scale.

Children were tested on the verbal subtests of the Wechsler Preschool and Primary Scale of Intelligence (Wechsler 1967), or for the older children, the Wechsler Intelligence Scale for Children: Revised (Wechsler 1976). It should be noted from table 1 that although the two groups were closely similar in CA and MA, there was a modest discrepancy in the mean IQ scores. Across the whole sample of children, there was not a significant correlation between the scores for interpersonal relations on the teacher questionnaire, and CA, MA nor IQ.

(b) *Procedure*

(i) *Playground observations*

Children were observed for three sessions in their school playground during regular free play periods, in nearly all cases on three different days. There was at least one class of pupils in the playground at any one time, supervised by an adult. One of us (M.B.) acted as the observer. He followed a given child for *ca*. 5 min in any given session, and made judgements on a total of five 20 s observation periods. Each observation session was begun when a child was within 1.8 m of at least one other child, without an adult in the immediate vicinity. This established a common starting point for all children. After a period of 20 s of undistracted observation, the observer would spend *ca*. 40 s recording what he had observed by ticking off items on a prepared scoring schedule (described below). Once the scoring had been completed (minimum 40 s), the next 20 s observation period would commence. Overall, therefore, each child was observed for fifteen 20 s observation periods (see Appendix A for examples).

(ii) *Rating schedule for social interactions*

The rating schedule followed the format of tables 2 and 3, except that there were blank boxes to check off instead of the results presented. In addition, for each observation a rating was made of the child's proximity to another child (distant, within 1.8 m; within 0.9 m; or touching). With one exception the items within each category were constructed so that they were mandatory to complete and mutually exclusive, and the observer simply ticked the item that best characterized the child's behaviour for each category during the 20 s observation period. Thus, for example, after an observation period the observer would begin by judging the typical degree of proximity, and would then move to rate social engagement (choosing one

Table 2. Mean number of observation periods for which each item of behaviour was most characteristic.

	MS group ($n = 9$)		LS group ($n = 9$)	
	mean	range	mean	range
social engagement				
cooperative	12.28	5–15	4.83	0–11
conflictual	0.89	0–2	1.94	0–7
isolated	1.83	0–8	8.22	3–15
emotional tone				
placid	6.22	0–12	8.67	5–12
pleasure	7.89	2–12	3.39	0–7
distress	0.89	0–3	2.94	0–6
type of play				
absence of play	3.33	0–9	9.83	3–15
rough and tumble	7.17	0–15	1.39	0–7
functional/exploratory	1.22	0–6	1.33	0–7
symbolic with props	1.78	0–5	1.33	0–12
symbolic verbal	1.50	0–6	0.78	0–4
other	0	0	0.33	0–2
sociability of play				
(absence of play)	3.33	0–9	9.83	3–15
alone	1.22	0–7	1.78	0–6
parallel	1.44	0–5	1.06	0–3
reciprocal (equivocal)	2.22	0–4	1.89	0–6
reciprocal (definite)	6.78	2–10	0.44	0–2

of cooperative, conflictual or isolated), and so on. If he was in doubt about which of two items captured the most frequent behaviour in a particular 20 s, he would select the most social/affective. Because a child was scored for 15 observation periods, the maximum score for any given item was 15; and the total score across the potential items for any category (for example, the total of cooperative, conflictual and isolated ratings in the category of social engagement) was 15.

The one set of items that were not mutually exclusive, and that were not always rated because they required that a child spoke (which did not always happen), concerned pragmatic language use. Here, a given child could score positively for any or all items if he/she made *any* instruction, request or comment during a given rating period.

(iii) *Ratings on the CARS*

The children were assessed on the CARS of Schopler *et al.* (1988), within *ca.* 12 months of the playground observations. This was possible because for a separate investigation, we made half-hour videotapes of the children engaging with an adult in play, and an independent clinician (blind to the MS and LS group membership) was able to complete the CARS by reviewing these videotapes. The setting was that the child was invited to play with several toys, and then an investigator would model a theme and invite the child to continue. The CARS involves ratings on 15 items (see figure 1), each of which is scored from one (for age-appropriate behaviour) to four (for severely abnormal autistic-like behaviour). Children with scores lower than 30 are considered non-autistic, although it should be noted that the omission of item VII on visual responsiveness reduces by four the maximum achievable score.

4. RESULTS

(a) *Playground ratings*

(i) *Reliability of ratings*

The ratings were made by one of the investigators (M.B.) who was aware of the group of each child. To locate children with profound visual impairment but no other diagnosed neurological or other handicap, he visited several English regional schools for children with visual impairment. Therefore it was not possible to employ multiple raters for most observations. To check the reliability of his ratings, a second person who was unaware of the hypotheses underlying the study accompanied him to one school and conducted independent ratings of one observation session each with four randomly selected children. On the ratings for each category, the weighted kappa coefficients of agreement (with the percentages of exact agreement in brackets) for each category of behaviour were as follows: for emotional tone, kappa = 0.60 (85%); for social engagement, kappa = 0.88 (85%); for type of play, kappa = 1.0 (100%); for sociability of play, kappa = 0.98 (90%); and for social language, kappa = 0.87 (75%). According to the criteria of Landis & Koch (1977), kappa values of 0.61 and above represent 'substantial' agreement, and 0.81 and above 'almost perfect' agreement.

(ii) *Observations*

For most observation periods, the children of both groups remained within 0.6 m of a peer (in 82% of the observations of MS children, and 66% of those of LS children), but in 8% of periods for MS children and 24% of periods for LS children, the children were more distant than 1.8 m from others. Across all observations, only one child in the MS group and two children in the LS group spent more than half their time at a distance greater than

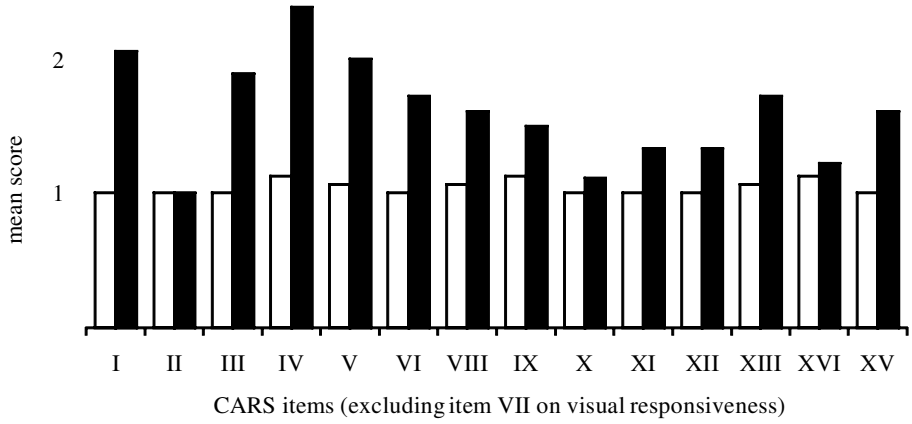

Figure 1. CARS group profiles: MS (open bars) versus LS (black bars) groups. (I, relating to people; II, imitation; III, emotional response; IV, body use; V, object use; VI, adaptation to change; VIII, listening response; IX, taste, smell and touch response and use; X, fear or nervousness; XI, verbal communication; XII, non-verbal communication; XIII, activity level; XIV, level and consistency of intellectual response; XV, general impressions.) Note that scores above unity indicate abnormality.

0.6 m from a peer. These results indicate that group differences in the remaining ratings were not simply a reflection of the LS children moving away from their peers.

Ratings of social engagement, emotional tone, and type and sociability of play

The results from these ratings are presented in table 2. We have presented mean rather than median scores out of 15 on each item for clarity of exposition. Within each category, the mean item scores add up to a total of 15.

In relation to the within-category items that exemplified our predictions most closely, one-tailed Mann–Whitney *p*-values for group differences (with the LS children showing the LS forms of behaviour) were as follows: for social engagement, the item of isolation ($U = 4$, $p < 0.001$); for emotional tone, the item of placidity turned out to yield a non-significant group difference, but on a two-tailed test the LS children showed significantly less pleasure ($U = 8$, $p < 0.005$); for type of play, the absence of play ($U = 8$, $p < 0.005$); and for the presence of equivocal or definite reciprocal play ($U = 4.5$, $p < 0.001$). Only two LS children showed more than three observation periods that included either definite or equivocal reciprocal play, and three showed no reciprocal play at all; by contrast, all but one MS child showed six or more periods involving reciprocal play, and four of the children showed 10 or more.

Ratings on use of language

The results from the ratings of social language and pragmatic language use are presented in table 3. We repeat that the ratings of pragmatic language use differ from the other ratings because the items of instruction, request and comment were not mutually exclusive. A child might be scored positively for each of these types of utterance, if he or she made at least one such utterance during a given observation period.

In keeping with our prediction, the LS children showed fewer periods in which they directed language towards other children (summing the items of 'direct to other', equivocally reciprocal and definitely reciprocal in table 3, Mann–Whitney $U = 11$, $p < 0.005$, one-tailed). Only one out of nine MS children but seven of the nine LS children had more ratings of non-reciprocal speech than speech that was equivocally or definitely reciprocal (Fisher's exact test, $p < 0.01$, one-tailed).

With regard to our second prediction, it was the case that the LS children showed fewer periods in which they offered comments to their peers (Mann–Whitney $U = 6.5$, $p < 0.001$, one-tailed). Six of the nine MS children made comments in at least 10 of the 15 observation periods, whereas none of the LS children did so. However, the dearth of comments was not absolute: four of the LS children were observed to make comments in more than five of the 15 periods, and although comments were rare among the remaining five children, all but one of them made comments on at least three occasions.

(b) *Ratings on the CARS*

The results on the CARS are presented in figure 1. Among those children who were socially engaged (MS), there was one child unavailable for testing on the CARS; otherwise in this group, for only one child and only on one item (level and consistency of intellectual response) was an item scored elevated by more than 0.5, and the highest overall score for a child was 15.5 (where 14 is the minimum score). Three children showed no abnormalities at all, three showed minor elevation of scores on a single item (body use, activity level, and level and consistency of intellectual response), and two showed abnormalities in smell and touch responses along with those in body and/or object use and/or listening response. These results indicate that in cases with little social impairment, congenital blindness *per se* is not necessarily associated with 'autistic-like' features.

These results may be compared with those from the socially impaired group (LS), in whom the range of individual scores was 17.5–27.5 (mean = 22.3, s.d. = 3.6). In figure 1 it can be seen that minor but significant abnormalities were present across most of the items of the CARS. This pattern is representative of individual children. For example, if one takes the criterion of an item score of at least two for 'autistic-like' abnormality, the numbers of children (out of nine) rated abnormal were as follows: six for relating to people, five for emotional response, seven for body use, six for object use, three for adaptation to change, five for activity level and four for

Table 3. (*a*) Mean number of observation periods for which each item of social language was characteristic. (*b*) Number of observation periods featuring each pragmatic use of language (not mutually exclusive).

	MS group ($n = 9$)		LS group ($n = 9$)	
	mean	range	mean	range
(*a*)				
none	2.50	0–8	5.39	1–9
self-directed	0.22	0–2	0.56	0–3
non-specifically outward	0.94	0–2	2.39	0–4
directed to other	0.72	0–2	2.78	0–9
reciprocal (equivocal)	3.44	0–6	2.50	0–5
reciprocal (definite)	7.17	2–11	1.39	0–4
(*b*)				
request	2.44	0–6	1.61	0–4.5
instruction	5.06	1.5–9	3.78	0–7
comment	9.39	5–13.5	4.78	1–9

'general impressions' of autism (an item on which only two children showed no abnormality). There were three individuals who scored *above* two for four items, two who did so for two items, and one for one item. Thus, there was evidence both that the social impairment had some 'autistic-like' quality, and that the range of abnormalities spread across the range of clinical features characteristic of autism.

5. DISCUSSION

The aim of this exploratory study was to examine the 'autistic-like' quality and breadth of abnormalities in socially impaired but not autistic congenitally blind children. The study was unusual in that it involved congenitally blind children *both* in index *and* control groups. The rationale was to control for the effects of blindness in shaping children's social relations, so that one could discern what is special about the social *and* non-social abnormalities that occur in those children with severe impairments in personal relatedness. The results indicated that in comparison with their MS blind peers, those whom teachers judged to be socially impaired were observed in the playground to be more socially isolated, less likely to express pleasure, and less likely to play or be involved in reciprocal play. The results highlight the nature and severity of the relative lack of reciprocal interpersonal engagement seen in some socially impaired blind children. Further observations pointed to additional parallels with deficits that are typical of sighted children with autism, for example in the children's relative dearth of comments on things and events.

In independent CARS ratings for 'autistic-like' abnormalities in a different play setting, a substantial majority of the socially impaired group were given elevated scores both for the autistic-like quality of their relating to people, and for 'general impressions' of autism. Moreover, the socially impaired but not the highly social children were also given moderately elevated scores for additional, relatively non-social clinical features characteristic of sighted children with autism, such as body and object use. The group differences occurred despite the fact that the two groups were closely similar in chronological and mental age (albeit not exactly matched for IQ, with the mean IQ of the LS group approximately nine points lower than that of the MS group).

A limitation of the study was that inter-rater reliabilities of the playground observations were established on a relatively small sample of the ratings. It might also be objected that there is a circularity in the methodology we have adopted, as we constituted the two groups of blind children according to teachers' ratings of sociability, and then proceeded to demonstrate that indeed one group was more social than the other. However, one aim of our study was to demonstrate something about the *qualities* of the social impairments of the more disabled group of children. For example, it is not simply that they tend to avoid other people, because even when they are close by their peers there are limitations to how they interact; it is not simply that they are clumsy in their social interactions, because they are less engaged with others in reciprocal interactions, whether emotionally or in language or in play. These observations highlight how there are wide-ranging individual differences in blind children's capacity for reciprocal engagement with others, and that such differences are not simply a reflection of intellectual ability. Both playground observations and separate CARS ratings indicated that the social impairments were of a kind reminiscent of autism. The second major finding was that additional, relatively non-social 'autistic-like' abnormalities were present almost exclusively in the socially impaired group.

The present study was not designed to address whether severe social impairments among blind children are associated with particular disorders. Although there have been suggestions that children with conditions such as Leber's amaurosis might have a special predisposition to autistic-like clinical features (Rogers & Newhart-Larson 1989), there is also evidence from our own previous research that such features may be associated with a range of medical conditions (Brown *et al.* 1997). In the present study, it was the case that all four children with the diagnosis of Leber's amaurosis were in our LS group, whereas the eight children with the diagnosis of retinopathy of prematurity were spread across the two groups.

To explain the association between the different kinds of 'autistic-like' abnormality in socially impaired blind children, there are several theoretical options. One might

argue that there is something special about the physical constitution of some blind children: perhaps some form of minimal brain damage associated with the conditions that led to blindness (Cass *et al.* 1994) that predisposes both to the social disabilities of these children and to their 'autistic-like' clinical features. Or one might consider that there are several sources of social impairment in blind children, including both physical and environmental factors, and that when potentiated by the children's lack of vision, these result in specific forms of impoverishment in interpersonal experiences that have developmental consequences which include several autistic-like features. We would stress that the socially impaired blind children of our study demonstrated a limited *reciprocal engagement with others*. Such engagement is pivotal for drawing a typically developing child into a flexible and creative engagement with other people's relatedness to the world, and prompting the child to grasp alternative meanings in reality and play.

Central to this thesis is that the syndrome of autism, whether in blind *or* sighted children, is the developmental outcome of profound disruption in the usual patterns of intersubjective coordination between the affected individual and others. The present results reveal how there are blind children who do *not* satisfy the diagnostic criteria for autism, but who nevertheless have marked impairments in interpersonal engagement. These are the very same children who also manifest several additional 'autistic features'. Our own preferred explanation is that vision has a special role in linking children with other people and with others' attitudes towards a shared world. Whether or not this proves to be correct, the findings indicate that there might be a variety of functional abnormalities—and correspondingly, a variety of conditions in the brains and/or perceptual systems and/or the environments of children—that can predispose to autism. And, however we explain the pathogenesis of autism, our explanation needs to encompass the phenomena of autism and autistic-like features in congenitally blind children.

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APPENDIX A

The following observation sessions concern two of the LS children. Each observation consists of five successive 20 s periods (labelled (i) to (v)), separated by periods of *ca*. 40 s while ratings were recorded.

A 6-year-old girl in the playground at lunchtime:

(i) she moved from an initially close position to become distant from the other children, involved with no-one, seemingly distressed and isolated; in using direct language, called out loud for a particular teacher; not involved in any play;
(ii) still distant, and distressed and isolated; showing no language or play;
(iii) moved within 0.6 m of both an adult and another child; still distressed and isolated; gave an undirected outward scream; no play;
(iv) still within 0.6 m of both an adult and child; distressed and in conflict; she suddenly called out, with a non-specific instruction—'don't do that!'; showing no play;
(v) within 0.6 m of an adult and now two other children; placid yet cooperative; engaged in an equivocally reciprocal exchange, making a verbal request, 'When we go in, can I hear your beautiful voice this afternoon?'; no play.

A 9-year-old boy during lunchtime outside: he was sitting on some steps while others were playing a game, calling out letters to each other.

(i) He was within touching distance of three other children who were playing the letter game; showing a placid emotional tone yet cooperative in social engagement; though no language or play.
(ii) He was led away by the hand by a girl classmate, reacting placidly yet cooperatively to this, though without showing any speech or play.
(iii) Being pulled around; distressed and conflicted, calling out to the other child to stop leading him around; no play.
(iv) Still being led by the other child, though now placid and cooperative again; no language or play.
(v) Still being led; distressed and isolated; giving an instruction to her, but not talking reciprocally; no play.

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GLOSSARY

CA: chronological age
CARS: childhood autism rating scale
IQ: intelligence quotient
LS: less social
MA: mental age
MS: more social