Papers and Originals

Autistic Conditions in Early Childhood: A Survey in Middlesex

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Kanner (1943, 1957) was the first to point out that one particular syndrome of abnormal behaviour could be separated from the previously undifferentiated group of childhood psychoses. Children with the syndrome (called early infantile autism) were distinguished by three outstanding characteristics: lack of responsiveness to other human beings—that is, "autism"—an insistence on the preservation of sameness in the environment, and an onset in the first two years of life. The children were attractive and alert in appearance, and Kanner believed they were not intellectually subnormal, though their functional abilities were grossly impaired. Their parents in most cases seemed vocationally successful and intelligent.

In retrospect the syndrome had already been brilliantly described by Itard, whose account of the "Wild Boy of Aveyron," written in the early years of the nineteenth century, has never been bettered (Humphrey, 1932). Itard did not agree with Pinel's diagnosis of severe subnormality and evolved an educational technique which is now being rediscovered by psychologists (Lovaas, 1966).

Subsequent writers have not been able to improve upon Kanner's clinical acumen. The best recent descriptions, unmarred by interpretations of what might or might not be going on in the child's mind, are by Rutter (1966a) and Wolff and Chess (1964, 1965). These writers, as well as Creak and Ini (1960), confirmed certain of Kanner's findings, but in children in whom onset was sometimes later than the age of 2 years. Like Kanner, Rutter and Wolff and Chess emphasize the importance of the speech disorder, which in many cases seems impossible to differentiate from the receptive type of developmental aphasia (Pronovost et al., 1966). In Creak's (1963) and in Rutter's (1966b) series, followed up into late adolescence or adulthood, there was no evidence of any development into schizophrenia. Kanner has also pointed out the lack of genetic relationship between schizophrenia and early infantile autism.

No epidemiological study has been made and the prevalence of the condition is unknown. The present survey was undertaken at the suggestion of Dr. Guy Wigley, then Medical Officer of Health for Middlesex and now Deputy Medical Adviser to the Greater London Council. Funds were generously provided by the Middlesex County Council.

Terminology

The term "autistic" can reasonably be applied, descriptively, to a much wider range of conditions than those included in the present series, just as this series contains some children without an onset in early infancy and some who had evidence of gross organic illness. No doubt, eventually, a more exact terminology will be possible which differentiates groups of

 Medical Research Council Social Psychiatry Research Unit, Institute of Psychiatry, London S.E.5. children. All we can do at this stage is to describe as precisely as possible the basis for selection of a series and to assign a name arbitrarily. The term "autistic condition of early childhood" is used in this article to emphasize that, in addition to those with Kanner's syndrome, some children have been included who showed characteristics which Kanner might have considered a basis for exclusion (notably, an onset between 2 and 5 years, and the presence of gross organic features). However, the other substantial group of conditions, also sometimes called "autistic," in which the main symptom is social withdrawal, in the absence of the central features described here, has been excluded from this series and our conclusions do not refer to them.

Aims and Method

The major aims of the survey were to estimate the prevalence of "autistic conditions in early childhood" in Middlesex and to investigate certain assumptions which had been made about the male: female ratio, birth order, parental occupation and intelligence, and the prevalence of mental disorder in close relatives.

The method has been described in detail by Lotter (1966a). Briefly, the whole population of Middlesex aged 8, 9, or 10 on 1 January 1964 (78,000) was screened by means of a specially developed behavioural questionary based on an amplified and modified version of the Creak Committee's "Nine Points" (Creak et al., 1961). This was completed by school-teachers, occupation centre supervisors, nurses, or parents. Only 1% of the population at risk could not be screened. Special attention was given to children in ordinary schools reported as showing any kind of deviant behaviour which might possibly be part of the syndrome, and to all children known to the local authority as handicapped in any way, physically or mentally.

The 135 children who were selected by the first screening process were then examined and given psychological tests, and further information on behaviour was obtained from a trained informant. As a result of this second screening process 61 children were retained in the series and a detailed social, medical, and developmental history was obtained from the parents, who were also tested on the Progressive Matrices and Mill Hill Vocabulary Scale.

Finally, all the available medical and social records were examined and the 54 children for whom sufficient information was available were divided into three groups according to a standard procedure. The "nuclear autistic group" all showed Kanner's two essential symptoms (but not necessarily the onset pattern) in marked degree—that is, lack of responsiveness to people and insistence on the preservation of sameness. The "non-nuclear autistic group" showed one or other of

these symptoms but not both: all showed many other characteristics of the syndrome as well. The remaining "non-autistic group" showed only minor fragments of the syndrome.

The screening procedures used at each stage were assessed for reliability in various ways and found to be satisfactory. Several details of the mothers' accounts (motor milestones, birth weight, and presence or absence of certain behaviours) could also be checked, and were found to be reasonably reliable.

Results

Children

Table I shows the prevalence of affected boys and girls aged 8, 9, and 10 on the census day. If the nuclear and non-nuclear groups are combined the overall prevalence is 4.5 per 10,000 children of this age group, about twice that for blindness. There was an excess of boys in all groups, the highest male: female ratio occurring in the nuclear group (2.75:1). This is much higher than the highest ratio reported for subnormal children—for example, 1.7:1 (Malzberg, 1954).

Table I.—Prevalence on 1 January 1964 Among Children Aged 8, 9, and 10 Residing in the County of Middlesex

		Boys	Girls	Both sexes	Rate per 10,000
Nuclear autistic group Non-nuclear autistic group	::	11 12	4 5	15 17	2·1 2·4
Total autistic children Non-autistic group	::	23 13	9	32 22	4·5 —

Of the 15 children in the nuclear group five had an I.Q. above 55; the rest were untestable or below 55. Five of the 17 non-nuclear autistic children also had I.Q.s above 55. All these 10 children were boys.

Considering the nuclear and non-nuclear groups together, 22 children had a gradual onset from an early age, while 10 had a definite and recognizable setback in development, eight of the latter having a low I.Q. or being untestable at the time of the survey. Delay in motor or speech development (15 out of 32 had marked delay in sitting or walking) was also associated with functional level at the time of the survey.

Nine of the 32 autistic children were mute and a further 10 had only a very limited use of speech: all these were functioning at a low level of intelligence. Five of the eight children whose speech was less severely affected and all the five whose speech was almost normal had intelligence quotients above 55.

There was no evidence of any special likelihood for an autistic child to be first-born (17 out of 32, compared with an expected 15.3) or to have any other specific birth rank. Most autistic children had normal siblings. These was no deviation from the expected frequencies of maternal age at the time of birth of the autistic children. There was no excess of children weighing less than 5½ lb. (2,500 g.) at birth in the autistic children or their siblings, but one-quarter of the children in the non-autistic group were immature according to this criterion. Their siblings had a similar median birth weight but no excess of immaturity.

Significant complications during pregnancy and delivery were reported in 6 out of 28 autistic children for whom medical records were available, compared with 3 out of their 44 siblings (Fisher's exact probability=0.073). Nine (28%) of the 32 autistic children had recorded evidence suggesting neurological abnormality: four had convulsions (three of them beginning in the first year), three had abnormal E.E.G.s, one was deaf (due to rubella), and one had congenital protrusion of one eye and a history of severe coal-gas poisoning. Eleven

of the 22 non-autistic children had an equivalent range of abnormalities.

Parents

The distribution of fathers' occupations is shown in Table II. (In the general Middlesex population there were 5.0% in class I, 18.5% in class II, 57.2% in class III, and 19.3% in classes IV and V.) The fathers of nuclear autistic children are particularly likely to have occupations in the upper two classes (9 out of 15).

TABLE II.—Occupation of Fathers Classified by Social Class

Social Class		Nuclear Autistic	Non-nuclear Autistic	Non- Autistic		
I II III IV, V	::		::	5 4 4 2	2 3 9 2	0 5 12 4
Total Not kn	 lown	::	::	15	16 1	21 1

The education of the parents differentiates the three groups of children even more clearly, as can be seen from Table III. The parents' scores on the Mill Hill Vocabulary Test confirm these results.

TABLE III.—Actual and Percentage Distribution of Parents (Mothers and Fathers Combined) According to Educational Level Reached, For the Three Survey Groups

					lear istic	Non-nuclear Autistic		Non- Autistic		
Educational level			No.	1) %	No.	2) %	No.	3) %		
School up to 15 years only					7	25	19	59	34	81
School after 15, or technical or other formal training				12	43	9	28	7	17	
Professional graduate	educ	ation o	r univ	ersity	9	32	4	13	1	2
Total		•••	•••		28	100	32	100	42	100

Data not known for two parents in each group.

The adoptive parents (father, graduate; mother, school after 15) were included for one child in the nuclear autistic group.

Significance (1) v. (2) v. (3): $z^4 = 24^4$.15, d.f. 4, P < 0.001.

Data on the mental health of parents were obtained for 28 of the 30 parents of nuclear autistic children (three admitted to psychiatric hospital), 32 of the 34 parents of non-nuclear autistic children (two admitted to hospital), and 42 of the 44 non-autistic children (one admitted to hospital). None of the parents of autistic children had suffered from schizophrenia, though affective disturbances were not uncommon. Information was also obtained about 283 siblings of 97 parents of children in the three groups, only one of whom was reported to have had an illness resembling schizophrenia (one of the mothers of a nuclear autistic child had a sister who had been admitted to an institution in the United States and treated by leucotomy).

Thus of 189 parents, uncles, and aunts of the autistic probands, 6 (3.2%) had been admitted to hospital for psychiatric illness, a figure very little different from that for the general population. In addition, out of 86 siblings of autistic children three showed behaviour disorder and an I.Q. in the E.S.N. range, though none had a clear-cut autistic disorder.

Services

The Middlesex County Council (like the Greater London Council, of which it now forms part) was outstanding in its efforts to provide special facilities for handicapped children. It is inevitable, however, that facilities for autistic conditions, where education and medical procedures are still experimental,

should lag behind those for children with other handicaps such as blindness or deafness.

In most cases the problem was picked up early enough, though eight children were not referred to specialists till after the age of five, when earlier referral might have been helpful. Roughly half the parents of autistic children were strongly critical of the advice they received about diagnosis, management, and placement when they saw a specialist (see Table IV).

TABLE IV.—Mothers' Opinions on the Advice Received About Diagnosis, Management, and Placement of Their Autistic Children

	Diag	nosis	Management		Placement	
	No.	%	No.	%	No.	%
Satisfied Dissatisfied but not strongly critical Strongly critical	6 9 14	21 31 48	5 11 13	17 38 45	9 4 16	31 14 55
Total	29	100	29	100	29	100

Three mothers could not be interviewed.

These criticisms were not casual or unfair—many mothers made explicit allowance for the special diagnostic and placement difficulties posed by their children. Inadequate guidance about placement attracted most comment—patients felt they were not given sufficient information about the alternatives available and the possible consequences of any decision they might make. Many mothers felt that they should have been given a better account of what was the matter with the child, and nearly half said that they were offered little or no helpful advice about how to manage their children at home. One mother, for example, said that she was told at a paediatric clinic about her 3-year-old son: "He's mentally defective; there is no hope for him whatsoever," and that thereafter she was left on her own, "like a leper, a young mother with a young baby, clueless about what to do. The only advice I ever got was 'let him play in the garden with a ball.'" This might have been an extreme example, but many of the mothers gave accounts which indicated that the medical and social work specialists they saw had not always been able to devote sufficient time to skilled counselling (Lotter, 1966b).

Only 16 of the 32 autistic children were receiving education (half of them in Steiner schools). The rest had not been given a trial in a special education unit; in fact, most had received no schooling at all. The decision to place these children in a non-educational setting cannot therefore have been based on adequate information about their response to skilled teaching methods in classes specially designed to contain and manage their difficult behaviour.

Discussion

The true incidence of autistic conditions in early childhood would be higher than the prevalence estimated from the results of this survey if some children with mild autistic disorders improved to such an extent that they would not have been picked up at the age of 8, 9, or 10 by the methods used. In addition, there may well be an excess mortality in very severely affected children. On the basis of the present results there are about 1,400 children aged 5-14 with the nuclear syndrome in England and Wales and another 1,600 autistic children with a rather less clear-cut clinical picture. Of these 3,000, at least 1,000 have a testable intelligence quotient above 55, and many of the others would undoubtedly benefit from remedial education. These figures compare with 1,400 blind children, and 2,100 partially sighted children, receiving special education, in January 1962, in schools approved by the Department of Education.

The high male-female ratio, the greater likelihood of paranatal complications, the high proportion suffering from physical abnormalities, together with the symptoms of the condition (particularly the speech disorder), suggest an organic aetiology. It is interesting in this connexion that Ingram (1960) also found a high male-female ratio and a substantial proportion of upper-middle-class parents in his series of children with developmental aphasia. The speech difficulties of children with receptive aphasia are closely similar to those found in psychotic children (Rutter, 1966a).

There is clearly no genetic or familial relationship to schizophrenia; a finding which has been paralleled in every well-documented series. Adults resident for long periods in a mental hospital tend to develop the syndrome of institutionalism (Barton, 1959; Wing, 1962) and thus acquire an artificial clinical homogeneity. Autistic children are particularly vulnerable to an understimulating environment (Rutter, 1965), so that if they grow up in an institution they may come to look like other institutionalized patients. These behavioural analogies have no diagnostic implications, and in any case do not hold for children brought up in their own families and given adequate education. None of the positive features of schizophrenia, such as delusions or hallucinations or specific catatonic phenomena, occur in autistic children, nor do they develop in later life (Rutter, 1966b). Adult schizophrenics do not have a history of Kanner's syndrome in childhood nor a raised incidence of autistic conditions in their relatives. There seems to be no reason why the two classes of disorder should be confused.

The unusual distribution of parental occupations requires explanation, but no plausible hypothesis is immediately evident. It has usually been assumed that this distribution is due to educated parents seeking advice more efficiently and refusing to accept a diagnosis of mental subnormality. There is no evidence for this in our data. Since there were 14 autistic children whose fathers had occupations in classes I and II, there would have to be 46 in classes III, IV, and V in order to achieve the same distribution as in the general population, whereas only 17 were found. It is extremely unlikely, in view of the very thorough search of local health and education authority records, that as many as 29 children in classes III–V could have been missed.

Rimland (1964) argued that the child with infantile autism might be a homozygote for high intelligence but carry a high vulnerability to various noxia during pregnancy and delivery. In the past, explanations have been based on an assumption that educated women are less likely to give their children expert and loving care during the first few months of life. There is little sociological evidence for this view, and in any case most educated women do not have autistic children; indeed, most parents of autistic children also have normal children. Finally, there is no evidence from any of the numerous series of children who were really deprived of maternal love during early infancy-for example, those raised in unsuitable institutions from birth—that such conditions produce a high incidence of childhood autism. The aetiology of the condition therefore remains obscure, though one report suggests that tryptophan metabolism may be abnormal (Heeley and Roberts, 1965).

Services leave a great deal to be desired, though most of the problems experienced by parents in the survey were also mentioned by the parents of children of severely subnormal intelligence (Tizard and Grad, 1961). The special nature of the disorder, however, gives some of the difficulties extra weight. The child may appear alert and attractive in spite of his odd behaviour. When the specialists seem puzzled and offer little helpful advice, and adequate educational and other services are not available, the parents may become profoundly isolated. The recent special units that have been set up in the London area (for example, Elgar, 1966) should help to relieve this problem, as will the growing experience of

paediatricians, psychiatrists, and social workers. The parents should, of course, always be put into touch with the appropriate voluntary organization (the National Society for Autistic Children).

Summary

A survey of children aged 8, 9, and 10 resident in the former County of Middlesex on 1 January 1964 showed that 4.5 per 10,000 had "autistic conditions of early childhood." This means that there are about twice as many autistic children in the country as there are blind children. There was a raised male-female ratio but no special birth order. Autistic children were more likely to have suffered from complications during pregnancy and delivery than their siblings, and one-half had marked delay in motor milestones. It was confirmed that the parents of autistic children (particularly those in the "nuclear group") were likely to be above average in educational attainment, occupational level, and intelligence. There was no evidence for a genetic or clinical relationship between early childhood autism and schizophrenia. Clinical and educational services still leave much to be desired.

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Evolution of the Ventilatory Capacity in Chronic Bronchitis

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The forced expiratory volume (F.E.V.) and its derivative the indirect maximum breathing capacity (M.B.C.) are used as indices of airways obstruction in patients with chronic obstructive airways disease. Prevalence studies of particular populations or groups of normal subjects have shown the F.E.V. to decline linearly with age. It is agreed that the ventilatory capacity declines further with progression of obstructive airways disease, but little information is yet available about the rate and mode of decline, particularly in relation to the phase of illness. The results of a long-term follow-up study are now reported in which the changes in the F.E.V._{0.75} have been carefully observed in a group of patients with established chronic obstructive airways disease attending an outpatient clinic. Particular attention has been paid to the long-term changes in individual patients and also to the effects of acute exacerbations of symptoms. It was hoped that the information might lead to a better understanding of the progress of airways obstruction, which is generally believed to be the chief cause of disability in chronic bronchitis.

Methods of Study

A study was made of 112 men and 13 women attending the clinic on account of frequent exacerbations of chest illness or breathlessness, usually the latter. Patients were included who had been followed regularly for at least two years, and this was the major criterion for admission to the study. In 1962 the patients answered the Medical Research Council (1960) short questionary on respiratory symptoms. All except two patients admitted to a productive cough, but a further 10 thought it occurred less often than on "most days for three months of the year" (grade 0). Of these 10 patients, five had attacks of bronchial asthma defined as sudden attacks of reversible severe

Department of Medicine, University of Sheffield, the Royal Hospital, Sheffield 1. Present address: Department of Environmental Medicine, Johns Hopkins University, 615 North Wolfe Street, Baltimore, Maryland 21205, U.S.A. Requests for reprints to Sheffield. wheezing and breathlessness not associated with cardiovascular disease, one had hay-fever in the summer months, and four said they experienced frequent attacks of chest illnesses but denied the persistent production of sputum between attacks. Twenty-four patients said they produced sputum only in the mornings (grade 1) and 89 said that it was raised both in the mornings and throughout the day (grade 2). Therefore 123 patients could be classified as cases of chronic bronchitis of some severity (Medical Research Council, 1965). One hundred complained of breathlessness which limited them to their own pace on the level or of more severe disability, and only four denied any breathlessness. Of the 125 patients, 106 had suffered at least one attack of chest illness in the previous three years, but most of them experienced more frequent exacerbations than this. Three patients have developed congestive cardiac failure (cor pulmonale) during the period of observation.

The patients usually attend the clinic at monthly intervals or less in the winter-time and every two or three months in the summer. In the four years up to the end of 1965 the F.E.V._{0.75} and the forced vital capacity (F.V.C.) were measured by means of a Poulton spirometer (McKerrow, McDermott, and Gilson, 1960) on each visit by one of three trained technicians. The F.E.V. was read directly off this instrument as the indirect M.B.C. in litres per minute. Five serial recordings were taken, the result being recorded as the mean of the best three readings. Before 1962 the same measurements were made annually with a Collins-Gaensler spirometer. From this machine the absolute F.E.V._{0.75} was measured, corrected for temperature, and then multiplied by 40 to derive the indirect M.B.C. In a comparison of the two instruments no significant difference was found between consecutive readings on the same patients (t=16.6, P<0.001).

Since the initiation of these studies the indirect M.B.C. has become a less popular method of recording this type of measurement. Thus in the analysis of the data the figures have been divided by 40 to convert them to the F.E.V._{0.75} as they were originally measured. Though formulae are available for