

Beyond Asperger Syndrome

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Introduction: The Childhood Picture

Among children referred to a child psychiatry department about 40%, more boys than girls (3.5:1), presented in middle childhood with common child psychiatric symptoms or educational difficulties but without the adverse circumstances that usually explain these. Over half the children were outgoing but some were withdrawn and uncommunicative, and occasionally they had had selective mutism. They often caused great concern to parents and teachers because they could not conform to ordinary social demands, reacting with weeping, rage or aggression if pressed to do so (Wolff, 1995). Their pre-school development was sometimes unusual but rarely worrying. Specific developmental delays were common; and parents complained of their children's difficulties in adapting to new circumstances; their obstinacy and ritualistic behaviour; emotional remoteness; and a "lack of feeling". School entry usually precipitated the clinic referral because the children could not relate to their peers or meet classroom demands for conformity.

At the time a diagnosis of "schizoid personality disorder" was made because the children resembled descriptions of this in the literature and in ICD-9 (World Health Organisation, 1978), the diagnostic scheme then in use. This label was applied to conditions now comprising the Type A personality disorders (schizotypal, paranoid and schizoid) of DSM-IV (American Psychiatric Association, 1994). We realised at once that our children were like those described by Asperger (1944; see also Frith, 1991), except that our group included girls (Wolff and Chick, 1980). The diagnoses we would now consider more appropriate for these children are mild Asperger or schizotypal personality disorder.

The following core features characterised the children:

- (i) solitariness
- (ii) lack of empathy and emotional detachment
- (iii) increased sensitivity at times with paranoid ideas
- (iv) rigidity of mental set, especially the single minded pursuit of special interests
- (v) unusual or odd styles of communication (such as over or under communicativeness, vagueness, and odd use of metaphor).

One further feature was later discovered, which Asperger too described: (vi) an unusual fantasy life. Like Asperger, we found many parents to have similar personality traits.

We operationalised the core features and for each child diagnosed schizoid, we identified as a control, another clinic attender, matched as well as possible for sex, age, occupational background, IQ, and year of referral. A case note analysis was done for 32 matched pairs of schizoid and control boys and for 33 matched pairs of girls (Wolff and McGuire, 1995).

The mean age at referral of the schizoid children was ten years and the mean maximum IQ was 109 for boys and 103 for girls (Wolff and McGuire, 1995). Among 32 schizoid boys, 16 presented with common child psychiatric symptoms, including conduct disorder, mixed conduct and emotional disorder, school refusal, soiling, and hyperkinesia; and six with educational failure. Only 10 presented with the features of schizoid personality itself.

Among the 33 schizoid girls comorbidity was even more common, 24 presenting with other child psychiatric disorders; 5 with educational failure. In contrast to the controls, pure emotional disorder was very rare, but conduct disorder was as common in schizoid as in control boys, and more common in schizoid girls than in their controls. Asperger too drew attention to the frequency of conduct disorders, even "maliciousness", in the children he described.

Of the core features, being a 'loner' and having 'unusual fantasies' significantly differentiated schizoid girls and boys from their controls. Having special interest patterns differentiated highly between schizoid and control boys, but not between the girls among whom this was rare. Impaired empathy, excessive sensitivity and odd styles of communication, all, as predicted, characteristic features of schizoid children in later life (see below), had rarely been recorded in childhood.

An important finding was that significantly more schizoid than control children had specific developmental delays of language, educational or motor functioning, serious or multiple in half the schizoid boys and a third of schizoid girls. Asperger too had found clumsiness and educational delays to be common in his children. In our groups, three schizoid boys but no schizoid girls had had earlier symptoms suggestive of autism, but never the full syndrome beginning under the age of three years. Four schizoid children had been selectively mute, and two had had serious developmental language delays. These two children, of normal intelligence, did not differ symptomatically from other schizoid children, and by the time of follow-up, their language had improved.

The Follow-Up Studies

These aimed firstly, to validate the syndrome, with the idea that, if the childhood syndrome was the beginning of a well known adult personality disorder, we should be able to recognise this and its core features when the children were grown-up; and second, to determine the adult life adjustment of "schizoid" children. We devised a focused interview designed to capture the postulated core features and enable an overall diagnosis of schizoid personality disorder to be made, as well as ratings of psychiatric morbidity, work adjustment, friendships, intimate relationships, and social integration. More recent instruments for assessing personality disorders congruent with current diagnostic classifications were not then available.

In the two follow-up studies, when the children were of mean ages 22 and 27 years (Wolff and Chick, 1980; Wolff, 1995), interviewers were "blind" to the childhood data and inter-rater reliabilities were adequate. On both occasions the overall diagnosis differentiated extremely well between schizoid and control young people, as did ratings of the five core features of the condition as well as one other symptom: an unusual fantasy life. We also found that the children grown-up were not excessively "introverted" on Eysenck's extra/introversion scales, and that a test for "psychological construing", that is the attribution of emotions and motivations to people in photographs, differentiated schizoid young men significantly from their controls and was related to interviewer ratings of "impaired empathy" (Chick et al, 1979), which we might now equate with impaired mentalizing abilities or "theory of mind".

The second follow-up interview incorporated a schedule for schizotypal personality (SSP) which showed 75% of schizoid men and women to fulfil the criteria for this (Wolff, 1995).

An important finding was that the schizoid children grown-up were far less impaired in psychosocial functioning than people currently given a diagnosis of Asperger syndrome (Tantam, 1991), thus resembling Asperger's original description. They had higher rates of treatment for psychiatric disorders compared with the controls, and their rate of working harmoniously at their expected level of occupation and, in boys, their rate of having had an intimate sexual relationship were significantly reduced. But their rates of independent living, of marriage, and stability of employment were not statistically different from those of the controls. Only one of 49 schizoid children followed-up was in residential care compared with over half the 60 patients with Asperger syndrome diagnosed by Tantam (1988a).

Two conclusions follow for the clinician: 1) there is a group of children, not nearly as impaired as children with autism or Asperger syndrome as currently defined (WHO, 1992 and 1993; APA, 1994), who need to be diagnosed because their more subtle, underlying difficulties are very long lasting, and schools and families need to accommodate to the children's special personality make-up; and 2) their overall outcome is reasonably good.

Schizoid Personality In Childhood And Later Psychiatric Morbidity: Is There A Link With Schizophrenia?

Because the features of our children resembled schizotypal personality disorder and because in later life most fulfilled the criteria for this, we wanted to find out whether they had an excess of schizophrenic illnesses. A records survey was undertaken of all psychiatric hospital admissions in Scotland of the total cohorts we had seen of 109 schizoid men and 32 schizoid women, then over 16 years, and of their matched controls of other psychiatrically referred children (Wolff, 1995).

Seven schizoid children but only one control had developed schizophrenia. Overall, 5.0% of schizoid young people and 0.7% of controls were affected at a mean age of 26.5 years, compared with an estimated population prevalence rate in the UK by 27 years of 0.31 - 0.49%. The numbers, though small, suggest that the risk for schizophrenia in schizoid children, while sufficiently low for a good prognosis to be given in childhood, is about ten times greater than that of other referred children and of the general population. This, together with their clinical features, support the notion that they have a schizophrenia spectrum disorder.

Two of the 32 schizoid boys followed-up, had transient delusional and hallucinatory states in childhood (consonant with the features of schizotypy) in response to stressful experiences. These symptoms responded well to psychotropic medication. While one of the boys subsequently had episodes of minor depression, neither developed a psychosis in later life.

The psychiatric records survey also showed that schizoid children made more use of psychiatric services in adult life than their controls. Our first follow-up had revealed an excess of depressive symptoms (Wolff and Chick, 1980), often in response to increasing self-awareness of social difficulties. The rate of suicide in our total cohorts of schizoid people was also increased: 4.0% compared with none among the controls and a population prevalence rate by the same age of 0.0026%.

Schizoid Personality And Later Delinquency

Because our children had often had co-morbid conduct disorders and because Asperger syndrome has been found associated with unusual delinquent acts in adult life, we undertook a Criminal Records search for the total cohorts of 109 schizoid boys and 32 schizoid girls then over the age of 16, and of their controls (Wolff, 1995). Not surprisingly, in all groups of these psychiatrically referred children grown-up, the percentage of people with convictions was greater than that expected for the general population of the same age. 32% of schizoid men and 34.5% of schizoid women had recorded convictions, compared with 34.5% and 15.5% of the controls. Comparable population norms were 22% for men and 5% for women. Schizoid women thus had exceptionally high rates of criminality.

There was no difference in the nature of offences between schizoid and control groups and none had committed an especially violent crime. But some years later, one of the schizoid men committed a seriously aggressive sexual assault, having adopted an alias and false identity (Wolff, 1995). He had been a charming but totally solitary child with serious specific developmental learning difficulties, always unpredictably aggressive, and frequently excluded from school.

In contrast to the controls, adult convictions in the schizoid group were strongly associated with aggressive behaviour in childhood. The one feature that appeared to protect against later delinquency, in both schizoid and control groups, was a childhood presentation with a pure emotional disorder. This occurred in 20 of the 65 controls but in only 5 of 65 schizoid children.

How Can Treatment Help?

In the absence of controlled treatment studies, this section is based on clinical experience alone.

The first important step is to recognise the condition and convey to child, parents, and teachers that the difficulties stem from the child's make up. If other members of the family have similar traits this can be reassuring because affected parents tend to manage their lives quite well and hope for an improved future adjustment can be more realistically based. It is important to make clear that neither parents nor ill will on the part of the child are to blame for the problem. It must be stressed that the child's basic personality features are not likely to change and that family and school will have to accommodate to his or her special needs.

On the other hand, treatment of associated symptoms: specific educational delays, aggressive outbursts, stealing, depression or, more rarely, hyperkinesia and attention deficit disorder, or delusional experiences, can be very effective. The treatment and interventions so clearly outlined by Klin and Volkmar (1997) for Asperger syndrome apply also to the children here described.

Special educational measures are often needed. Affected children may find the hurly burly of the playground intolerable and need to be allowed to seek refuge in a quiet place instead. Often too, they are helped by being excused from team games. Remedial teaching for associated specific educational impairments is indicated; as well as small group teaching when a noisy classroom is more than the child can manage. If he cannot be motivated for prescribed classwork, the curriculum needs to be built around his special interests. If there is severe educational retardation or if the behaviour at school is intolerable because of aggressive outbursts, eccentricities provoking to other children, or other symptoms such as oppositional behaviour, aggression, depression, stealing or school refusal special schooling has to be arranged.

Behavioural treatment approaches for aggressive outbursts can be very effective; and social skills training can improve peer relationships. Conduct and emotional disorders are usually a reaction to the child's inability to tolerate even mild pressures for conformity to ordinary school life, and in a less demanding setting, such behaviour often ceases. But a small minority of the children are aggressively delinquent whatever their environment, and may then be exposed to the juvenile justice system. When custody is demanded by a court, schizoid children fare better in a small residential school, community, or psychiatric unit, where not all the residents are delinquent so that the level of aggression is low; where the staff is well trained in the care of disturbed children; and the staff/child ratio is high. They do not do well in large, noisy institutions for young offenders, where bullying and violence predominate and there is little privacy.

When hyperkinesia is associated, medication is as helpful as for other hyperkinetic children; and psychotropic medication is indicated during transient delusional and/or hallucinatory states. Long term psychotropic medication has no place in the treatment of these children.

The primary tasks of psychiatrist or psychologist, however, are to provide long term, even if infrequent, support for the family as they negotiate the child's path through school into further education and a working life; and to act as the child's advocate in relation to the school and school psychological services. Here a diagnosis of "Asperger syndrome", now familiar and acceptable, may be more helpful in facilitating access to services than the rather vague label of "constitutional personality disorder", although the diagnostic criteria for Asperger syndrome may not be fulfilled.

It is also important to maintain an optimistic stance: both Asperger and we ourselves found that the children's adjustment improves with age once the pressures for conformity, always greatest during the school years, are at an end and they can find their own niche in life. The outlook is particularly good for schizoid children of high ability without comorbid aggressiveness.

Changing Diagnostic Concepts

The term "schizoid" was coined by Eugene Bleuler to describe shut-in, suspicious, sensitive people within the normal range of personality variation. Such features Bleuler found premonitory in half his patients with schizophrenia who, even as children, stood out because they could not play with others, followed their own ways instead, and were regarded as strange by other children because of their odd, intellectual characteristics (see Wolff, 1995). In 1926 Ssucharewa (see also Wolff, 1996) wrote the first account of six boys, clinically resembling Asperger's cases as well as our own, under the title "Schizoid personality disorder of childhood".

Recent diagnostic classifications substitute the term "schizotypal" for what was previously called "schizoid" personality disorder, and in 1986 Nagy and Szatmari described 20 "schizotypal" children, recognising their similarity to our own cases, as well as to Asperger's (1944) and Wing's (1981). Their features were social isolation, social anxiety, magical thinking, bizarre preoccupations, poor rapport and odd speech. The authors thought the disorder might be a mild form of autism or a variant of adult schizophrenia. Two of the children later developed schizophrenia. Szatmari subsequently dropped the term "schizotypal" in favour of Asperger syndrome.

We realised from the start that our children resembled Asperger's cases (Wolff and Chick, 1980). He stressed the children's giftedness, the association with maliciousness and unusual fantasy. He reported the social disability to decrease in adulthood when, despite continuing difficulties in intimate relationships, work adjustment was often excellent. This contrasts with more recent accounts of people with Asperger syndrome (Tantam, 1988a and b; Tantam, 1991; and Wing, 1992), who were rarely able to lead independent lives or maintain employment, and hardly ever married. Most of Tantam's patients had the triad of impairments typical of autism in early childhood, although not always beginning under the age of three, and most also scored highly on a measure for schizoid/schizotypal personality. This was thought to be secondary to the developmental disorder.

There are two reasons for preferring the schizoid/schizotypal label for the children we have studied, unless the category of Asperger syndrome is specifically modified.

First, the children do not meet the criteria for Asperger syndrome of ICD-10 (WHO, 1992; 1993) or DSM-IV (APA, 1994). They do not have the abnormalities of reciprocal social interaction, nor the restrictive, repetitive, stereotyped patterns of behaviour "as for autism". Our children's features resemble those of autism, but are not the same. ICD-10 criteria include the absence of clinically significant general delay in spoken or receptive language or cognitive development; and they include circumscribed interests or restricted, repetitive, and stereotyped behaviours. An exclusion criterion is schizotypal disorder, but the definition includes schizoid disorder of childhood and autistic psychopathy. DSM-IV criteria also exclude significant delay of language and cognitive development; indicate that the disturbance causes clinically significant impairment in social, occupational or other areas of functioning; and differentiates the disorder from schizoid personality.

A few of our children were of below normal intelligence and some had early language delays, occasionally severe. And it is now known that severe developmental language disorder can be associated both with social oddities and impaired intimate relationships as well as an increased risk of later paranoid psychosis (Mawhood, 1995).

In ICD-10, the criterion of circumscribed interest patterns or restricted and stereotyped behaviour does not really capture the, often sophisticated, special interests of our schizoid young people. In the most intelligent of the schizoid men, such interests formed the basis for a successful career choice: in astrophysics and graphic design. Only in a few of the less intellectually gifted, could their special interests be described as restricted and repetitive.

Finally, an unusual fantasy life, occasionally amounting to pathological lying and the adoption of aliases, was prominent in some of our cases, as indeed it was in Asperger's too, and should be mentioned as a diagnostic feature of Asperger syndrome, if the schizoid group here described is to be included within this diagnostic category.

As Klin and Volkmar (1997) indicate, the diagnosis of Asperger syndrome has been defined in varying ways, and according to current diagnostic criteria, it cannot unequivocally be differentiated from high functioning autism. Prior et al (1998), in a cluster analysis, also found no clear demarcation between high functioning children diagnosed as having autism, Asperger's syndrome or other pervasive developmental disorders, merely differences in social and cognitive impairments. In particular, early language delay or deviance did not differentiate between the groups of children. Yet none of our children had ever fulfilled the criteria for autism.

A second reason for not classifying our children within the pervasive developmental disorders is because different diagnostic labels should not be used for the same condition merely because it is recognised in childhood rather than later life. Yet researchers into schizotypal personality in adult life do not always read the child psychiatric literature. Olin et al (1997) for example, overlooking the work of ourselves and of Nagy and Szatmari (1986), in a study of teacher ratings of school behaviour in adolescents later diagnosed as having a schizotypal personality disorder, state that "no study" has yet reported on the early behaviour of people given this diagnosis. Their teacher ratings characterising these youngsters included being lonely; content with isolation; anxious with peers; having disturbed and inappropriate behaviour; and disciplinary problems.

In summary, the children we described could be classified either as having a schizoid/schizotypal personality disorder whose diagnostic criteria they fulfil, or as having Asperger's autistic psychopathy according to Asperger's original description. The current diagnostic category of Asperger syndrome is inappropriate unless its criteria both in DSM-IV and ICD-10 are modified to omit the exclusion of significant delays in speech and language and of schizoid and schizotypal disorders; to specify the less severe social impairments and more sophisticated all-absorbing interests in comparison with autism; and to include a criterion for unusual fantasy.

Could The Disorder We Called "Schizoid" Belong Both To The Autistic And The Schizophrenia Spectrum?

Links with the autistic spectrum

The individual features of schizoid personality in childhood resemble the symptoms of autism and Asperger syndrome but, as we have seen, they are not the same.

Yet there may be a genetic relationship between autism, Asperger syndrome and our "schizoid" or schizotypal group. Autism and Asperger syndrome have been found in members of the same families (Gillberg, 1991); parents of autistic children have an excess of mild schizoid personality traits (Wolff et al, 1988); both twin and family studies of autistic children have shown an excess of cognitive and social deficits, as well as repetitive, stereotyped behaviour, milder but similar to those of autism itself, in non-affected identical twins and first degree relatives of autistic people (Bailey et al, 1996; Bolton et al, 1994; Fombonne et al, 1997; LeCouteur et al, 1996). To these disorders the labels "lesser variant" or "broader phenotype" of autism have been applied, and language delay and reading and spelling difficulties are now recognised as constituting a part of this (Rutter et al, 1999a). Only one case of Asperger syndrome was found among these relatives, perhaps because associated language delays at present preclude this diagnosis. Yet it would be strange if the clinical features of individuals demonstrably sharing the genes of people with autism, were different from those of the clinically recognised autism spectrum disorders, such as Asperger syndrome or our "schizoid" group, which Rutter (1999) sees as either "mild autism" or "some different condition".

Links with the schizophrenia spectrum

We need to be clear that, while schizoid and schizotypal characteristics are found to excess premorbidly in patients, especially men, with schizophrenia (Foerster et al, 1991), very few children with such personality disorders will, as our study showed, go on to develop this illness. Ssucharewa too (Wolff, 1996) stressed that the schizoid disorder she described in childhood was the same as that seen premorbidly in schizophrenic patients, but that the children's condition was stable and did not deteriorate.

Our children shared many features described for children who later develop schizophrenia. In a case-control study of juvenile schizophrenia, Hollis (1995) found an excess of children with specific developmental language delays of expressive and receptive language, specific developmental motor impairments, and abnormalities of social development, in particular, poor peer relationships, shyness and social withdrawal. Jacobsen and Rapoport (1998) document premorbid developmental delays of speech and language and transient symptoms of pervasive developmental disorders in childhood onset schizophrenia. The British National birth cohort studies showed that children who developed schizophrenia as adults had specific developmental delays as well as solitariness and socially inappropriate behaviour (Jones et al, 1994; Done et al, 1994).

Thus the precursors of schizophrenia consist both of solitariness and impaired social skills as well as specific developmental delays, including language delays, apparently resembling the broader autism phenotype.

Schizotypal and other Type A personality disorders are found to excess also among biological relatives, especially men, of patients with schizophrenia (Kremen et al, 1998). The considerable discordance for schizophrenia between MZ twins one of whom is affected, is explained, as for the discordance between MZ twins one of whom has autism, on the basis that the schizophrenia genotype is not expressed unless released by some kind of, possibly prenatal, stressors (Gottesman, and Bertelsen, 1989).

To reconcile the possibility of a common genetic factor or factors for autism and schizophrenia, manifesting with variable expressivity as features of schizoid/schizotypal/Asperger disorders, with the fact that autism and schizophrenia do not co-aggregate in families, we would need to postulate that for autism as well as schizophrenia other genetic factors, different for each condition, as well as possible environmental stressors, are among the necessary causes.

Rutter (1999b) has recently suggested that genetic effects may be related to specific facets of a disorder or even to dimensional vulnerability traits, rather than to the psychiatric condition as a whole.

Conclusions

1. Current diagnostic criteria for Asperger syndrome identify only the more seriously impaired patients within the groups described by Asperger and ourselves. More mildly affected children and adults, some of whom are gifted, need to have the nature of their difficulties recognised as constitutionally determined, so that their symptoms are not erroneously attributed to faulty upbringing. The diagnosis is often obscured by co-morbidity.
2. Associated specific developmental disorders need special educational provisions; and associated conduct and other psychiatric disorders require realistic treatment approaches. The children's special interests and gifts need to be preserved and fostered; and psychiatric or psychological support for child and family should continue for many years. Although the children are not as impaired as children currently given an Asperger syndrome diagnosis, the problems they pose to their families and schools are often formidable.
3. It may be difficult to reconcile the fact that "schizoid personality disorder" appears to lie at one end of the autistic spectrum where it merges with normal personality variation, while there is also evidence linking it to the schizophrenia spectrum. Etiologically the problem is less serious. Both schizophrenia and autism are now regarded as neurodevelopmental disorders on a genetic basis, each involving several genes.

And both in autism and during schizophrenic episodes similar deficits in mentalizing ability and language have been found (Corcoran et al, 1997; Drury et al, 1998; Frith, 1992); Baltaxe and Simmons, 1992).

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