

Classification of Pervasive Developmental Disorders: Some Concepts and Practical Considerations

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Classifications have to meet a variety of purposes. Clinical and research needs are different and there is much to be said for separate clinical and research schemes. Care is needed to ensure that classifications provide an appropriate medium for teaching about diagnosis and do not cause difficulties when used as a "passport" to resources. Principles of classification are considered in relation to the need to take course, as well as symptomatology, into account, and with respect to the neuropsychiatric interface. The value of a multiaxial approach is noted. The pros and cons of autism and pervasive developmental disorders (PDD) as an overall descriptive term, of lumping or splitting, and of different choices with respect to PDD subcategories are discussed.

SOME CONCEPTUAL ISSUES

As the papers in this special issue clearly indicate, there is considerable variability among clinicians and researchers in their approach to diagnostic and classification issues. Sometimes it is assumed that the differences can be fully resolved on the basis of empirical research findings. Of course, it is highly desirable that psychiatric classifications should have a firm scientific basis and it is good that decisions now are being made with reference to research data, rather than through armchair speculations or weight of authority. This marks a major, and wholly beneficial, departure from the practices that prevailed two or three decades ago. The conse-

quence has been a remarkable coming together of views, as people have been persuaded by the force of data. However, although this constitutes a most important advance, it is crucial to appreciate that not all classification issues can be resolved through science (Kendler, 1990). In part, this is because classifications have to serve a variety of purposes, some of which diverge in their requirements (Schopler & Rutter, 1978). Because DSM-IV is a classification produced by the American Psychiatric Association, in the interests of simplicity and ease of reference, we have referred to it and ICD-10 as *psychiatric classifications* throughout this paper. However, many other disciplines are involved in their use and development.

Clinical and Research Needs

ICD-10, the World Health Organization (WHO) classification scheme, recognized this divergence in its production of both clinical (WHO, 1990a) and research (WHO, 1990b) versions. The former expresses diagnostic distinctions in terms of concepts and criteria put in the form of clinical guidelines. They are more explicit and detailed than in its predecessor, ICD-9, and in this as well as other respects much closer to the DSM approach of the United States, but there are no rigid quantitative rules that a particular number of symptoms must be present. This is both because this approach is designed to approximate to high quality clinical practice and because clinical usage requires that a minimum proportion of cases should be left totally undiagnosed. By contrast, the research criteria are both operationalized so far as possible (that is to say that the means by which a criterion should be judged are made explicit), and quantified (that is, the number of criteria that must be met are specified). These rules have been introduced because it has been appreciated that, for research purposes, it is desirable to have as much direct comparability across studies as possible, and a reasonable degree of homogeneity within diagnostic groupings. A price has to be paid for this detailed specification and the main cost lies in the proportion of cases left undiagnosed. Inevitably, some of the cases that are left out would have been included if decisions had been up to the clinician. This is an unavoidable consequence of any system that involves categorical distinctions based on cutoff points, however well based the latter may be. There will always be cases where the overall clinical picture meets the general specifications, but there is one symptom too few, or an IQ one point above the cutoff, or a language level just short of the standard score requirement. It is important to realize that this issue is intrinsic to any categorical classification based on quantified rules and not

just those that concern psychiatric disorders. Thus, cardiologists face the same problem with blood pressure cutoffs for hypertension.

In our view, the ICD-10 approach of having separate (but closely related) clinical and research versions is preferable to the DSM approach of forcing one classification to meet both sets of needs. However, there are limitations to the ICD-10 scheme and there are more than two usages for a classification. We wish to highlight two further applications that are particularly important: (a) teaching and (b) a "passport" to resources. DSM-III (American Psychiatric Association [APA], 1980) and DSM-III-R (APA, 1987) have generated a plethora of books and videotapes designed to present the classification as a means of teaching trainees about diagnosis. For this reason above all others, we find ourselves in disagreement with Waterhouse, Wing, Spitzer, and Siegal (1992). They argue that neither etiology nor course are key elements in diagnosis, although they may be useful for "chart information." In our view, this does not represent the best clinical practice. Families have a right to know whether a progressive downhill course (as is usually the case with Rett syndrome) or continuing developmental advance (as with autism) is to be expected. This is not a matter to be relegated to administrative chart information; it constitutes the heart of clinically applicable diagnosis. Similarly, we reject the claim that etiology is not an essential part of diagnosis. That it is not *all* is obvious (see Rutter, 1965), but to exclude it entirely from diagnosis is unacceptable. For our part, we would not wish to consult a clinician who, for example, regarded the presence of either tuberous sclerosis or fragile X as outside diagnosis. In both cases, the identification of cause carries implications for the individual and for genetic counseling. This is especially so now that molecular genetic advances in the fragile X (Davies, 1991) allow carriers to be identified with a high degree of accuracy. In our view, it is clinically inappropriate to see this as a matter only for chart information. *How* etiological considerations should be dealt with in a psychiatric classification is another matter, an issue to which we return. Also, of course it is necessary in clinical practice to pay detailed attention to the reliability and validity of medical investigations (see, for example, the problems that arose in early studies of the fragile X phenomenon when very low rates of fragile X expression were accepted as adequate diagnostic indicators; Bolton et al., 1992). Similarly, it is crucial to appreciate the probabilistic nature of prognosis, individual variations in course and outcome, and the need for families to maintain hope. These considerations are part and parcel of good clinical practice, but etiology and course have to play a key role as well.

In making positive comment about the desirability of having separate clinical and research classifications, as in ICD-10, we noted that there were limitations to the research scheme provided by ICD-10. Two require par-

ticular mention. First, in many circumstances, dimensional approaches based upon functional considerations may work better than categorical distinctions. For example, in respiratory medicine, it was found that a continuous measure of pulmonary function was often more useful than the categorical diagnostic distinction between, say, chronic bronchitis and emphysema (Scadding, 1980, 1982). Similarly, with Pervasive Developmental Disorder (PDD), often, it may be more useful to assess children in terms of their nonverbal IQ and language level than with respect to whether or not they fulfill the criteria for autism or atypical autism. In other words, the ICD-10 research classification (like other research classifications) provides a useful starting point but it would be a grave error to regard it as a sufficient classificatory tool. It is desirable to retain the overall research categorization in order to ensure compatibility across studies but investigators have an obligation, with each study, to consider which further distinctions need to be made to fulfill the needs of that particular study.

The second limitation is that, very commonly, research has to test the utility of distinctions not included in the standard classifications. This need will arise both with advances in knowledge and with new concepts or hypotheses. For example, Ghaziuddin, Tsai, and Ghaziuddin (1992a) note the inconclusive nature of the evidence on whether clumsiness does, or does not, constitute an essential defining characteristic of Asperger syndrome. The issue is of considerable theoretical interest and it seems desirable to find out whether the presence or absence of clumsiness makes a difference in any clinically relevant respect such as family history or course. Because of the continuing uncertainty over the connections between autism and Asperger syndrome, probably it would be wise to extend such an investigation to autism, in order to determine whether a similar difference applies there as well. At present, such a diagnostic distinction is not built into the classification, and as Ghaziuddin et al. (1992a) point out, so far there is a lack of evidence to justify such a distinction. However, one purpose of research is to advance knowledge on diagnostic distinctions, and this means trying out new methods of differentiation.

Access to Resources

The second further usage of classification, namely, a passport to resources, raises more ticklish issues. There is no doubt that in some countries, perhaps especially the United States, this is a widespread practice. In our view, this is a clear *misuse*, or indeed abuse, of classification, for several different reasons. We highlight three. First, a single diagnosis does not carry with it uniform service requirements. Thus, it is known that autis-

tic individuals span an IQ range from profoundly retarded (say 20) to superior (say 120). It is manifestly absurd to suppose that their educational requirements will be the same. Those who are profoundly retarded are likely to require personal care and supervision for the rest of their lives and the great majority will never acquire even rudimentary spoken language. By sharp contrast, some of the intellectually advantaged will need to be prepared for college entry and some will go on to professional careers. It would not be a service to either group to force them into one all-encompassing educational facility for autistic individuals.

Second, as studies from the Isle of Wight survey (Rutter, Tizard, & Whitmore, 1970/1981) onwards have shown, service needs are greatest for those children with multiple handicaps or impairments. Most thought needs to be given to the provision for children, who, for example, are both autistic and deaf, or autistic and mentally retarded, or autistic and severely epileptic. It is not helpful to approach their needs from the perspective of having to decide which of two overlapping diagnoses is the prime diagnosis. Of course, psychiatric classifications allow for multiple diagnosis. That is not the problem. The difficulty stems from the fact that most therapeutic facilities do not reflect the reality of multiple diagnosis. Rather, a choice needs to be made between a facility designed to cater for autistic individuals, or one for the mentally handicapped, or one for the severely epileptic.

Third, it is extremely limited clinical practice to see service provision solely in terms of entry to some diagnostically defined service or facility. Rather, the good clinician needs to accept the responsibilities of looking at each child's needs across the board and preparing, as it were, a "prescription" for services that cover those needs whether they be educational, psychological, medical, or whatever. We regret the expectation that one diagnosis equals one therapeutic "package." Nevertheless, being pragmatic realists who have to work within existing structures, however much we consider them misguided, we accept the need to take these passport-to-resources considerations into account.

Neuropsychiatric Interface

Before turning to the specifics of classification issues with respect to autism and PDD, there are two other broader conceptual issues that need to be addressed. First, Waterhouse et al. (1992) claim that ICD-10 has used different bases to justify different diagnoses. Thus, they argue that Rett syndrome is justified on the basis of its being a genetically distinct entity, and disintegrative disorders on the basis of an associated neurological deficit. In fact, both claims are factually incorrect as reference to the

ICD-10 documents readily demonstrates. To the contrary, both are differentiated on the basis of their distinctive clinical course. That seems to us entirely reasonable. It would be absurdly restrictive to have to base diagnoses solely on a cross-sectional clinical snapshot. That is not what happens in the rest of medicine and there seems to be no reason to impose that straitjacket on psychiatry. Nevertheless, Waterhouse et al. (1992) seem to be suggesting that diagnoses should be based only on behavioral patterns, and that etiological considerations should never play any part in classification decisions. In our view, that is going too far and, certainly, it would make the approach in psychiatry quite different from that in the rest of medicine. Rather, what is needed is to use a wide range of criteria (including etiology and course, as well as symptomatology) to determine which disorders clearly need to be differentiated because they diverge in so many different ways, and those that might best be considered together because, in spite of some differences, they have so much in common (Rutter, 1965, 1978b; Rutter & Gould, 1985). The move from a theoretical to a phenomenological approach to classification arose because the prevailing theories lacked empirical support. The move was needed, and has been helpful, but that does not mean that the phenomenological groupings should ignore well-substantiated research findings.

The second issue concerns the "territories" of psychiatry and neurology. Tsai (1992) touches on this consideration in his thoughtful review of the research findings on Rett syndrome. He argues that there are good reasons for keeping the condition within a broad PDD grouping, but it may be useful to discuss the issues involved. Two seem to us crucial. First, there is the implicit (or sometimes explicit) assumption in some quarters that once a condition has been shown to have an organic basis, it should be removed from psychiatry, because psychiatry deals only with disorders of the mind, and not those of the brain. That is clearly an insupportable position for two rather separate reasons. First, there are numerous psychiatric disorders already known to have an organic basis and yet remain within psychiatry because the patients' clinical needs are best met that way. For example, that would apply to both the dementias and severe mental retardation. The criterion is *not* whether the cause lies in the brain but rather whether the disorder tends to present in terms of altered behavior (rather than, say, a limp or a paralysis).

Second, there is the dualistic view in which there can be workings of the mind that are unconnected with the brain. This is out of keeping with empirical findings. Thus, the phenomenon of imprinting has been found to involve consistent accompanying neural changes (Horn, 1990). Of course, imprinting is not due to brain damage. On the contrary, it constitutes a form of learning, but that is not to say that the learning takes place outside

of the brain and without associated neural alterations. All operations of the mind are likely to involve some changes in brain structure or function, sometimes temporary and sometimes lasting; the limitation lies simply in our power to demonstrate these changes. It should be added that most people now consider it highly likely that autism will prove to have a neurobiological basis; in that respect it is no different from, say, Rett syndrome. Rutter and Schopler's (1988) arguments concerned possible differences between different varieties of PDD in terms of *types* of neurobiological abnormality and not in terms of the presence or absence of such abnormality.

In short, we need to decide on whether particular conditions are best classified under neurology or psychiatry, *not* with respect to organic etiology but rather in terms of the ways in which the disorder presents and the types of service needs. In that connection, it requires emphasis that these are not mutually exclusive alternatives. Thus, in ICD-10, as in previous WHO classifications (which encompass the whole of medicine and not just psychiatry), Down syndrome is classified both in terms of the chromosomal abnormality that constitutes the cause and in terms of the mental retardation to which often it gives rise. Exactly the same applies with the organic brain syndromes arising in adult life.

The second broad issue concerns the assumption that the territorial decision must revolve around the choice of deciding diagnosis on the basis of cause or clinical picture. Waterhouse et al. (1992) raise the specter in terms of the presumed need to have a fragile X-PDD category, if Rett syndrome is allowed. Quite rightly, they point out that individuals with fragile X show a wide diversity of clinical pictures; some of these meet even narrow traditional criteria for autism whereas others can be classified as "pervasive developmental disorder not otherwise specified" (PDDNOS). They could have gone on to note that many do not fulfill the criteria for any sort of PDD but nevertheless, do exhibit clinically significant psychiatric problems of other kinds (Reiss, Feinstein, & Rosenbaum, 1986). For obvious reasons, it would not be reasonable to classify this heterogeneous group of disorders under a single heading simply because they all relate to the fragile X anomaly. That is not what we suggest, nor so far as we know is this approach advocated by anyone else. It would be contrary to sound previous practice in medical classification. Thus, tuberculosis meningitis is classified separately from pulmonary tubercle even though both have the same cause; they are differentiated because the clinical implications are quite different. Similarly, general paralysis of the insane and syphilitic gummas are classified separately for the same reason. This is an important principle because pleiotropy is very widespread in medicine; that is, one cause can give rise to a wide range of clinical manifestations (Plomin, 1991). It is, of course, clinically useful to be able to use a classi-

fication to pick out diverse types of disorders with a common cause. That requires a system that can deal with both causes and clinical pictures; we will return to the issue of how this need might best be met.

STRUCTURAL ORGANIZATION OF CLASSIFICATION SYSTEMS

With these conceptual and practical considerations in mind, we need to turn our attention to some of the specific issues that apply to PDD. The first of these concerns the overall structural organization of the system. The most obvious need is for a multiaxial format. The reasons why this is required were discussed at some length by Tarjan et al. (1972) but may be summarized succinctly here.

First, when a disorder may be conceptualized from several different perspectives, clinicians tend to differ on how they prioritize these facets. Thus, in one of the case history exercises for ICD-9, a mentally retarded autistic child with evidence of organic brain dysfunction received a diagnosis of autism from about a third of the participating clinicians, one of mental retardation from another third, and one of organic brain syndrome from a final third (Rutter et al., 1969). In discussion, it became clear there was virtually complete diagnostic agreement in spite of an almost complete classification *disagreement* based on the diversity regarding which facet was chosen as the basis for diagnosis. By organizing the classification on a multifactorial basis so that psychiatric syndrome, intellectual level, and medical condition were on separate axes, each and every one of which had to be coded, this artifactual unreliability could be eliminated. It is apparent, also, that this step automatically succeeds in meeting a second need, namely, the provision of a fuller set of clinically relevant information. Thus, follow-up studies have been consistent in showing that the long-term prognosis of autistic individuals is strongly dependent on their level of IQ. It is necessary to know whether the clinical picture is that of autism, and also whether there is associated mental retardation, and to what degree.

The third advantage of a multiaxial format is that it avoids the introduction of diagnostic biases deriving from variations in causal assumptions. Thus, it is clear that if diagnoses are a compound of etiology and symptom pattern, clinicians will vary in how they use the compound. For example, should an organic basis be inferred on the basis of a few neurological soft signs (such as clumsiness or mirror movements), or should traditional hard signs (such as a hemiplegia) be required? Of course, it is possible to make a psychiatric classification purely phenomenological but there is a deep and understandable resistance to such an extreme course. Thus, both ICD-10 and DSM-III-R, although avowedly atheoretical, have categories for or-

ganic disorders and for posttraumatic stress disorders. Also, field trials of multi-axial classifications have shown that psychiatrists appreciate being able to code medical and psychosocial features that may be causal (Rutter, Shaffer, & Shepherd, 1975). It is likely that such provision helps to ensure that phenomenological diagnoses are truly made on a phenomenological basis without regard to etiological assumptions because these are provided for elsewhere in the multi-axial classification.

For these reasons, we are persuaded of the desirability of adopting a multi-axial format for psychiatric classifications. However, if this is to be effective in fulfilling its purpose, it is crucial that it is organized so that it is mandatory that *all* axes are coded in all cases and, for this to be possible, there must be provision for a "no abnormality" code on each axis. DSM-III-R falls short in both these regards and it appears that, usually, it has not been implemented in multi-axial fashion. ICD-10 constitutes an interesting compromise in this connection. Because it was decided by WHO that a multi-axial format would not be suitable for all branches of medicine, neither the clinical nor research versions explicitly adopt a multi-axial framework. However, because it was recognized that such a framework did have advantages in psychiatry, the scheme was constructed in such a fashion that it could easily be put into a multi-axial framework and that has been done for child psychiatry.

Given the adoption of a multi-axial approach, it is necessary to take a further set of decisions on the specific axes to be provided and how each is to be constructed. It will readily be appreciated that there can be no one right way of doing this. Rather, it is a matter of which type of organization is most likely to meet the most needs in the most satisfactory fashion. A key issue in that connection is whether autism and other PDDs should be grouped with clinical psychiatric syndromes, or with specific developmental disorders (e.g., of language), or with mental retardation. It might be thought that the key deciding feature ought to be the nature of autism and of PDD. As it would be generally agreed that they constitute disorders of development, it might seem to follow that they should be on the same axis as either specific developmental disorders or mental retardation or both. However, that is not the most relevant consideration. Instead, it is necessary to ask which facets of diagnosis (other than autism) are most crucial to record in a uniform, systematic fashion in all cases. Thus, is it clinically more important to be sure to record whether there is mental retardation in addition to autism, or an anxiety disorder in association with autism? Put in this way, it is obvious that the greater need is to code the intellectual level and the presence and absence of a specific developmental language disorder. It could also be argued that autism is defined in terms of behavioral abnormalities and not in terms of cognitive level. On both

counts (but the former is much more important), the multi-axial version of ICD-10 includes PDD on the clinical psychiatric syndrome axis, there being separate axes for intellectual level and for specific developmental disorders. It could be argued that the latter two axes could be combined into one, but numerous clinicians have pointed out that there is only a weak association between the two. There are individuals with a very severe and persistent language disorder in spite of normal intelligence, and mentally retarded individuals who are surprisingly proficient in language. Accordingly, as there seemed to be something to be lost, and nothing to be gained, by combining specific and general cognitive disorders, ICD-10 has kept them on separate axes.

Following the previous multi-axial version (of ICD-9), and in keeping with clinical and research needs, further axes are provided for medical conditions and for abnormal psychosocial situations (the latter having been extensively revised in recent times; van Gooor-Lambo, Orley, Poustka, & Rutter, 1990). In this way, information can be routinely retrieved on whether, for example, autism is associated with the fragile X anomaly (or any other medical condition).

DSM-III and DSM-III-R also have an axis for level of adaptive functioning, an axis not included in the multi-axial version of ICD-9. As it has become clear that in child psychiatry as a whole, this adds very important information (Bird, Gould, Yager, Staghezza, & Camino, 1989; Weissman, Warner, & Fendrich, 1990), it was decided to include this axis in the multi-axial version of ICD-10. Its content is closely similar to that in DSM-III-R but differs from it in excluding symptoms from consideration (as these are dealt with on the psychiatric syndrome axis) and, hence, in focusing exclusively on the different ways in which adaptive functioning may be manifested.

In our view, there is much to be said for DSM-IV adopting a full-hearted multi-axial approach. It would have advantages for the whole of child psychiatry but it would be particularly helpful in the case of autism and other PDD. It is a natural next step as DSM-III was pioneering in being the first official psychiatric classification to adopt this approach; all that is needed now is to put it on a more systematic basis.

OVERALL TERM FOR CLASSIFICATION GROUPING

A surprising amount of heat (and not much light!) has been generated over what term should be applied to the overall group of disorders that includes autism. At one time, autism was considered as a type of schizophrenia or a variety of infantile psychosis. This grouping came increasingly

to be rejected as evidence accumulated on the numerous crucial differences between autism and schizophrenia (Rutter, 1972). Werry's (1992) review of the evidence on early onset schizophrenia reaffirms that schizophrenia, of a type broadly comparable to that seen in adulthood, does occur in childhood; that there is no evidence to justify differentiating a subvariety of schizophrenia according to childhood onset; and no reason to pool autism and schizophrenia. DSM-III took what seemed a step forward in its recognition that autism constituted a disorder of development, and not a psychosis. The term *pervasive developmental disorders* was established to cover autism and similar disorders. At the time this seemed to help clinically in underlining the fact that usually it was inappropriate to treat autistic individuals as if they had an emotional disorder, or were mentally ill. Nevertheless, most services continued to be organized under the diagnostic banner of autism, rather than PDD. Now there seems to be some pressure to revert to the original diagnostic term of autism, and to abandon PDD. Thus, a year or so ago, a group of researchers (Baird et al., 1990) urged this course of action on the grounds that it was known that autism constituted a specific developmental disorder, and *not* a pervasive one; that severe mental retardation *was* a pervasive developmental disorder; and that PDD was not an appropriate term to use with parents. From a scientific perspective, these arguments seem to us mistaken, as they did also to Volkmar and Cohen (1991). We presume that the specific psychological deficit suggestion refers to the finding that autistic individuals lack what has come to be called a "theory of mind" (Baron-Cohen, Leslie, & Frith, 1985). We agree that this was a most important scientific discovery, particularly as it promised to link the cognitive and social deficits characteristic of autism (Frith, 1989). However, it remains uncertain whether the cognitive deficit is as specific as first supposed; thus, Ozonoff and her colleagues (Ozonoff, Pennington, & Rogers, 1991; Ozonoff, Rogers, & Pennington, 1991) showed that executive planning deficits were more characteristic of autism. Also, some three quarters of autistic individuals are also mentally retarded—a developmental impairment that cannot be attributed to a "theory of mind" deficit. Moreover, follow-up studies show that even autistic individuals of normal nonverbal intelligence show widespread impairments in most domains of life, impairments substantially greater and broader than those found with even the most severe specific developmental language disorders (Rutter, Mawhood, & Howlin, 1992). Of course, the disorders are not all-pervasive in that nonverbal intelligence may be spared. However, the same consideration (the other way round) applies to severe mental retardation. In spite of their global mental handicap, many mentally retarded individuals show social and communicative functioning far superior to that found in autism, as numerous studies show.

The argument about what you say to parents, as put by Baird et al. (1991), also seems to us mistaken. There is, however, here a small but crucial difference between ICD-10 and DSM-III-R in that connection. In the former, PDD is a generic term, and not a diagnosis. As such, it is equivalent to other portmanteau headings such as "Behavioral Syndromes Associated with Physiological Disturbances and Physical Factors." It is not intended to be used as a diagnostic term and should not, therefore, constitute the basis of what is said to parents. DSM-III-R is slightly different in that, although, PDD is a generic term, PDDNOS is given as a diagnosis. Of course, it would be a foolish and nonthinking clinician who simply gave parents the term in the manual as a diagnosis. It is the responsibility of clinicians to explain to parents the nature of the disorder shown by their child, and a single diagnostic term (however well based) is rarely adequate for that purpose. Moreover, many medical diagnoses are scientifically accurate but quite meaningless to lay people unless explained.

Accordingly, we reject the arguments of Baird et al. (1991). Nevertheless, we accept that there is a problem in relation to services. We have already made the point that services should not be based on the "diagnosis equals predetermined package" principle, but we have also observed that all too often, some children may be denied access to services that they need, and from which they would benefit, on the grounds that they suffer from PDD, and not autism; clearly that is unacceptable. We suggest that two steps might help in that connection, without in any way giving rise to difficulties. First, that the overall generic term, PDD, be given a subheading indicating that an alternative term for exactly the same group of disorders is "autistic spectrum disorders"; second, that PDDNOS be given the alternative term of "atypical autism." So far as we are concerned, there are no particular scientific advantages in this slight amendment of terminology but there may be considerable resource access advantages, and that is just as important.

"Lumping" or "Splitting"

It is well known that clinicians tend to be either lumpers (other things being equal, they prefer to use broad diagnostic groupings) or splitters (they prefer finer diagnostic distinctions). On the face of it, it seems a very straightforward scientific matter to arbitrate between the two. The key question is whether the empirical evidence justifies one approach over the other. However, a little thought soon shows that the issue is more complicated than it seems. That is because, the answer to that seemingly straightforward question depends on the starting point adopted. Thus, the review

papers in this special issue ask whether there is evidence that Asperger syndrome is meaningfully different from autism (Ghaziuddin et al., 1992a, 1992b); and whether disintegrative disorders constitute a meaningfully different syndrome (Volkmar, 1992). The answer is a pretty straightforward "yes" in the case of Rett, a "probably" in the case of disintegrative disorders, and an uncertain "maybe" in the case of Asperger syndrome. Of course, all of this depends on how much research has been undertaken and it is obvious that it is very little so far. Should one pay more attention to the fact that Ozonoff and her colleagues (Ozonoff, Pennington, & Rogers, 1991; Ozonoff, Rogers, & Pennington, 1991) found important psychological differences between autism and Asperger syndrome, or the fact that this awaits replication? Given inadequate data (and clearly that is the state of affairs), the question is whether it is preferable to lean towards lumping or splitting.

That brings us to the alternative approach. There is a huge amount of research showing that autism is meaningfully different from schizophrenia, mental retardation, specific developmental language disorders, and indeed, any other condition with which it has been compared. It was the sheer weight and consistency of that evidence that led us to argue previously that "the evidence on its validity is stronger than for any other psychiatric condition in childhood" (Rutter & Schopler, 1988; p. 411). It is pertinent to ask which set of diagnostic criteria was used in all of these validating studies. Because standardized diagnostic approaches were less in use in the 1960s, 1970s, and early 1980s than they are today, it is not possible to give an unequivocal answer to that crucial query. However, in that many of the studies claimed to have used Kanner's (1943) criteria, or Rutter's (1978a) criteria, or DSM-III (APA, 1980), it may be inferred that it is likely that most tended towards narrower, rather than broader, diagnostic criteria. If so, the starting point has to be whether there is evidence to justify pooling the so-called "atypical" varieties of autism with the so-called "nuclear" type. The answer has to be that there is not much evidence, although, there is a little on clinical course. That is not because the studies have been undertaken and failed to show commonality, but rather because so few comparative studies have been performed.

The net result is that there is a lack of clear-cut guidance on whether a narrower or broader diagnostic approach is to be preferred and on just which groups should be included in the generic PDD group. In a sense, the latter question is both easier to answer and less open to dispute. That is because there is general agreement on the broad domains of abnormality that characterize PDD, namely, qualitative impairment or deviance in reciprocal social interaction, qualitative impairment or deviance in communication, and stereotyped or repetitive patterns of behavior. The

disagreements largely concern matters of subdivision or subcategorization, and the breadth or narrowness of the autism diagnosis.

With respect to the latter, several special issue papers are in agreement that DSM-III provides the narrowest of the standard approaches, and DSM-III-R the broadest, with ICD-10 intermediate (see also Hertzog, Snow, New, & Shapiro, 1990). With the possible exception of Van Bourgondien, Marcus, & Schopler, (1992) (who did not consider ICD-10 criteria), they are also agreed that ICD-10 most closely approximates to clinicians' preferences (Fombonne, 1992; Volkmar, Cicchetti, Bregman, & Cohen, 1992; Volkmar, Cicchetti, Cohen, & Bregman, 1992, Szatmari, 1992b).

In other words, most clinical preferences were for an approach that was somewhat broader than DSM-III but narrower than DSM-III-R. However, by contrast, Van Bourgondien et al. (1992) reported that the clinicians in their study, using the CARS, inferred a diagnostic system somewhat broader than DSM-III-R. This discrepancy illustrates the uncertainty of reliance on clinical judgment, or service needs, as the standard against which criteria are to be judged. Szatmari (1992a, 1992b) argues that this comparison constitutes validation, but we do not agree. All that it shows is clinical consensus. Validation requires some external criterion outside the symptomatology (Rutter, 1978b), in terms, for example, of cause, course, or response to intervention. The only study in the special issue to provide that is Fombonne's (1992), in which he shows good differentiation from mental retardation without autism using ICD-10 criteria. So far so good, but obviously, on its own that constitutes an insufficient basis for choosing between diagnostic approaches.

To make that choice, it is necessary to ask about the possible costs that may be involved if the approach chosen is wrong. This consideration brings out the important point that the scientific and clinical implications are somewhat different. Let us take each of the main classifications on offer to illustrate what this might mean in practice. ICD-10 provides the greatest subcategorization. If research shows that some or all of these diagnostic differentiations are unwarranted, the only scientific cost will be that too many subdivisions will have been provided and that some subgroups will need to be combined in the future. The scientific payoff, however, is that this could then be done with confidence on the basis of empirical research findings. From a clinical perspective, the possible costs probably need to be considered in terms of effects on development of, and access to, services. Data are lacking on whether the total set of PDD categories in ICD-10 includes more or fewer children than DSM-III or DSM-III-R. However, it seems highly likely that, if anything, they probably include more if only because the concept has been broadened in the three

directions of a milder, intellectually more able group (Asperger syndrome); of a more severe mentally retarded group (atypical autism); and of disorders involving a loss of skills (Rett syndrome and disintegrative disorders). The question is whether the subcategorization, if scientifically unwarranted, will help or hinder the development of appropriate services and children's access to them. Because the subcategorization emphasizes the diversity of needs (e.g., between the extremes of Asperger and Rett syndromes where the expected adult outcome is totally different), we consider that it is likely to prove helpful in service development. We need a diversity of provision and not just one standard model. Nevertheless, there has to be some concern lest services become too diagnostically restrictive and hence exclude children in need unnecessarily. However, in our view, the use of terms such as atypical autism, rather than PDDNOS, would make this substantially less likely.

If we turn now to DSM-III-R, although the relevant data are not available, it is likely that the overall coverage of the PDD group as a whole would not be very different from ICD-10. The main difference lies in the markedly diminished differentiation *within* PDD. Again, we have to consider the scientific and clinical costs if further research should show that greater differentiation is required. From a scientific perspective, the cost would be considerable because nothing would have been learned through the use of DSM-III-R. Indeed the only way for research to show that greater differentiation is needed is if some *other* classification scheme constitutes the basis for the research. The clinical costs would depend on whether the scientific differentiation had clinical implications. If Rett syndrome is anything to go by, the implications might well be serious. Thus, there are needs for physiotherapy for the muscle weakness that generally develops and for pediatric care with respect to the epilepsy that occurs in almost all cases. However, there are also educational costs, as a recently referred child with undiagnosed Rett syndrome illustrates. At the time of referral, the school and the parents were in conflict, with each blaming the other for the child's progressive deterioration, neither realizing that that was to be expected with the condition from which the child suffered.

The scenario with DSM-III is different yet again in that it is probable that the overall PDD category is *less* inclusive than with either DSM-III-R or ICD-10 because of the greater weighting towards severe social impairment as seen in very young children. However, it is possible that older, less handicapped, autistic individuals would be picked up by the "residual autism" category. The clinical and scientific costs, if DSM-III proved to include too few subdivisions, would be similar to those noted for DSM-III-R but with the additional bias noted above, plus the fact that one subdivision, childhood onset, seems to have very little to be said for it (see

Waterhouse et al., 1992) and, hence, probably constitutes one subdivision too many.

With all three classifications, it is necessary also to consider the possibility that the particular criteria used for subdivision might prove to be mistaken. To a degree, that is almost certain to be the case in that it is rather unlikely that the present criteria, in any of the systems, will exactly match up with the cause or causes of autism when they become known, or with distinctions in terms of course or response to intervention as may be demonstrated by future research. Accordingly, we reemphasize the point that we made earlier; that it is highly undesirable for research to be constrained by any one classification system. We hope that NIMH, and other research funding agencies, will appreciate that that is so, and be supportive of well thought-out, hypothesis-driven research to test alternative new forms of subclassification.

Our evaluation of these considerations leads us to conclude that it is important to ensure that the overall provision for PDD is broad (as in either DSM-III-R or ICD-10); that the terminology provides links with autism; and that the scientific needs, as well as discriminating service development, be met through an adequate subcategorization of PDD disorders. As this has been best worked out in ICD-10, that constitutes a convenient starting point; and it seems helpful that the DSM-IV Options Book (APA, 1991) suggests that the two classifications are likely to be reasonably comparable to each other.

CHOICE AND DEFINITION OF PDD SUBCATEGORIES

The evidence relevant to the case for the inclusion of specific PDD subcategories has been well summarized in the papers included in this special issue and the key points can be summarized briefly. The strongest case concerns Rett syndrome (Tsai, 1992). Already, in spite of its relatively short history, it has a very substantial research literature that is consistent in showing both a characteristic clinical picture and a characteristic course over time, as well as the striking finding that its occurrence appears to be virtually confined to girls. It could not sensibly be subsumed with autism because the clinical picture and course are both different, and to put it into the undifferentiated category of PDDNOS would result in a serious loss of information. Because ordinarily it presents first as a pervasive developmental disorder, rather than with neurological signs and symptoms, it is appropriate for it to be classified under PDD. The cause is not yet known. When it is identified, it may well be appropriate for the clinical syndrome to be grouped in PDD, with the specific medical cause (if such

a one is discovered) classified elsewhere, in the same fashion that already exists for Down syndrome.

Disintegrative disorders provide a less clear-cut case, but we agree with Volkmar (1992) that, on balance, there is sufficient evidence to warrant its inclusion in DSM-IV. As Volkmar points out, the category is *not* meant to cover the relatively high proportion of cases of autism in which there is a degree of regression in language at about the age of 2 years. So far as is known, this later onset autism does not differ in any important ways from cases of autism without regression. Moreover, it should not be assumed that the change at age 2 in later onset autism implies an acquired disorder. Because the brain areas subserving some psychological functions alter over the course of development, and because the maintenance of psychological functions varies with age in its input requirements, lesions present at birth may well have functional sequelae that vary with age (Goodman, 1991). The case for differentiating disintegrative disorders is different in two key respects; first, the period of prior normal development is substantially longer than is usually the case with autism; and second, the pattern of regression is different in that it generally includes the loss of skills outside communication and social relationships. As Volkmar and Cohen (1989) have shown, such cases differ strikingly from "later onset" autism; moreover the latter do not fulfill ICD-10 criteria for disintegrative disorder. Furthermore, both the course and clinical pattern of disintegrative disorders differ from Rett syndrome (Volkmar, 1992). Whether or not disintegrative disorders will turn out to be due to acquired or later onset neurological diseases of a kind that rarely apply to autism remains to be seen. This seems to be so in a minority of instances (Corbett, Harris, Taylor, & Trimble, 1977) but that is not the justification for including the category separately from autism.

The situation with respect to Asperger syndrome is more complicated, if only because different investigations have used rather different concepts and sets of criteria. In Wing's (1981) usage, it overlaps with autism and many cases probably constitute milder varieties of autism. On the other hand, the concept of "schizoid disorder of childhood" (Wolff & Barlow, 1979), which approximates to Asperger syndrome seems rather broader than that, and it is clear that many, probably most, would not meet the usually accepted criteria for autism. Moreover, Wolff and Barlow found psychological differences between autism and Asperger syndrome, as more recently did Ozonoff and her colleagues (Ozonoff, Pennington, & Rogers, 1991; Ozonoff, Rogers, & Pennington, 1991). It is quite possible that at least some of these differences could be a function of the insensitivity of the measures used, with the Asperger syndrome individuals less severely affected. The genetic data from both twin (Bailey et al., 1991) and family

studies (Bolton et al., 1991) strongly suggest that there are cognitive cum social disorders that do not fulfill the usual diagnostic criteria for autism and yet are part of the autism phenotype. It seems reasonable to suppose that some examples of Asperger syndrome (perhaps most) will ultimately prove to be a variant of autism but it is not at all clear that that will apply to all. As there is an obvious research need to compare autism with Asperger syndrome, we suggest that there is a need for a separate PDD category for Asperger syndrome in order to encourage and facilitate that research. However, for obvious reasons, it will be necessary to define the syndrome in such a way that there is no overlap with autism.

It is necessary to go on to ask whether there is a comparable need for other separate subcategories. ICD-10 has a coding for overactive disorder associated with mental retardation and stereotyped movements (which is not considered in any of the review papers in this special issue). This was introduced to cover severely retarded individuals who exhibit hyperactivity and stereotypies but who do not show the qualitative abnormalities in communication and in reciprocal social relationships that characterize autism. There is reason to separate this group from the more usual varieties of attention deficit disorders with hyperactivity, if only because it seems that stimulant medication often makes them worse and in particular aggravates their stereotyped behavior (Aman & Singh, 1982). However, it remains uncertain how much this ill-defined group of disorders has in common with autism. It might reasonably be described as a pervasive developmental disorder, but so far it lacks a research literature to justify its inclusion as a separate category. It is not clear where such cases should be included.

Another group of disorders that warrant consideration are the social deficits often associated with severe developmental disorders of receptive language (Rutter & Mawhood, 1991). There are now several follow-up studies that show that considerable problems in reciprocal social relationships are frequently evident and persist into adolescence and early adult life. They stand out as different from autism in many respects but equally they seem to reflect a persistent and pervasive social disorder that does not fit easily into any of the recognized psychiatric categories. Once again, it is not clear how this group of disorders should be defined and there is insufficient empirical evidence to justify a separate category in DSM-IV.

Yet another group of disorders that have to be encompassed in some way are those associated with the fragile X anomaly. They do not seem to present a homogeneous behavioral pattern but marked social anxiety seems very common and this is often associated with stereotyped behavior of various kinds (Cohen, Fisch, Sudhalter, Wolf-Schein, & Hanson, 1988). There is a growing body of evidence showing the differences from autism but it

remains quite uncertain how far there is a distinctive disorder that requires separate subclassification.

We have noted these several examples, *not* because we think they warrant separate categories on the basis of current knowledge but rather to emphasize the range of disorders outside autism but which involve some kind of pervasive persistent abnormality of social relationships, communication patterns, or stereotyped behavior that is present from early childhood. It seems clear that they fit more readily into PDD than into any other broad diagnostic grouping. Until there is better evidence on their characteristics, there is probably no alternative but to have some kind of PDDNOS category to cover this (presumably rather heterogeneous) group of disorders. In the past, DSM has sought to provide precise rules for all categories but it is necessary to recognize that in many instances we lack the data needed to formulate such rules and, from both an educational and clinical perspective, it is desirable to be quite explicit in making overt our relative ignorance.

That leaves the need to decide where to set the limits for the boundaries of the autism category. As we have indicated, there is no scientific evidence that provides an unequivocal answer. The advantages of the medium breadth approach (as in ICD-10) lie in its use for research and the advantages of the quite broad approach (as in DSM-III-R) in its clinical application. In that DSM-IV has to meet both needs, we support the medium breadth approach, provided that the remainder category (PDDNOS or its equivalent) is titled in a way that makes the links with autism and that is not open to the possible abuse of its operation to exclude them from services that seem likely to meet their needs. We are uncomfortably aware of the likely heterogeneity of this remainder category but accept that this is the price that has to be paid for trying to avoid the same problem with the main autism category. However, if the PDDNOS category (or some equivalent) is to serve its intended purpose in DSM-IV, it is necessary to provide some overarching conceptual description of the types of disorder the generic grouping of PDD is meant to include. This cannot be expressed in terms of operationalized rules as its purpose is to provide coverage of a group of disorders for which the research evidence is insufficient for the formulation of precise diagnostic criteria. Nevertheless, unless the concept of PDD is clearly formulated, there is a danger that it will cease to have any useful meaning. However, unless there is a remainder category there is a serious danger that disorders will be excluded from all categories with the consequence that children will be denied access to services they need and from which they should benefit. This dilemma is one of the inevitable consequences of having to make one classification scheme serve both clinical and research purposes.

FORMULATION OF DIAGNOSTIC CRITERIA

The final issue to which we need to pay attention concerns the formulation of the various specific diagnostic criteria. One of the important advances in DSM-III-R was the acceptance that the criteria must concern abnormalities that are deviant in relation to the person's mental age (Waterhouse et al., 1992) and it is important that that be carried through to DSM-IV. This is a necessary specification in order to avoid PDD being no more than an expression of severe developmental impairment. Of course, the specification leaves the problem of how to decide when a particular behavior is or is not discrepant with mental age, an issue also identified by Van Bourgondien et al. (1992). There is no straightforward psychometric method by which such discrepancies can be quantified in the absence of systematic data on the mental age correlations of each behavioral feature. The study reported in this issue by Volkmar, Cicchetti, Cohen, and Bregman (1992) makes an important start on this task, but it also illustrates the problems that are entailed. Both DSM-III-R and ICD-10 have sought to deal with the issue by three steps. First, so far as possible, diagnostic criteria are expressed in terms of *qualitative* features that would be abnormal at any age; however this is possible with only some criteria. Second, the criteria have been extended to include both those that are likely to be most discriminating with young severely retarded children and those that discriminate best with autistic adults of normal nonverbal intelligence. Third, the rules provide for the possibility of the criteria being met through different combinations of features (a necessary possibility if the developmental spread is to be encompassed). These three steps are clearly helpful but we need to recognize that there are still likely to be particular difficulties in the case of profoundly retarded young children (with the main danger that of false positives) and of adults of normal nonverbal intelligence (where there is the opposite main danger of false negatives). The field trials for DSM-IV should be helpful in coming to decisions on the most satisfactory cutoff points for criteria but it is crucial that the main arbiter should be *discriminative* validity between autism and PDD and other diagnostic groups (especially mental retardation unaccompanied by autism), and not just agreement with clinician diagnoses of autism.

CONCLUSION

The papers in this special issue demonstrate well the serious and thoughtful efforts to assemble the research findings that should serve as the empirical basis for decisions on the classification of PDD for DSM-IV.

They also provide a useful bringing together of some of the key conceptual issues that need to play a part in the decision making. Our goal in this overview essay has been to note the complexities that are inherent in the exercise, especially when a single classification scheme has to be used for clinical, research, and administrative purposes. It has become clear that there are some unavoidable dilemmas and trade-offs in the decision-making on classification issues. The solution that is most advantageous for one purpose may carry disadvantages for others. It is also obvious that nosological progress is dependent on hypothesis-driven further research. A range of research strategies including genetic, neurobiological, neuropsychological, and clinical-longitudinal approaches is needed to determine whether, for example, autism and Asperger syndrome represent different conditions or variants of one disorder or whether disintegrative disorders constitute a meaningful diagnostic entity. In expressing our own views on what might be most appropriate for DSM-IV, we have been mindful of the need to foster future progress as well as to represent the present state of knowledge. The increase in our understanding of autism since Kanner first described the syndrome in 1943 has been most gratifying, but it is equally apparent that there is a great deal still to be explained. It is important that DSM-IV be constructed in such a fashion that it will facilitate the research that is going to be needed in the years to come.

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