Behavioral Medicine with the Developmentally Disabled

Edited by Dennis C. Russo
and Jurgen H. Kedesdy

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Plenum Press • New York and London

Library of Congress Cataloging in Publication Data

Behavioral medicine with the developmentally disabled.

Includes bibliographies and index.
1. Developmentally disabled children—Medical care. 2. Developmentally disabled children—Rehabilitation. 3. Medicine and psychology. I. Russo, Dennis C., 1950–
II. Kedesdy, Jurgen Horst. [DNLM: 1. Behavioral Medicine. 2. Child Developmental Disorders—therapy. 3. Handicapped. 4. Rehabilitation. WS 350.6 B4187]
RJ135.B45 1988 618.92'89142 88-17953
ISBN-13:978-1-4612-8285-3 e-ISBN-13:978-1-4613-0999-4
DOI: 10.1007/978-1-4613-0999-4

A Division of Plenum Publishing Corporation
233 Spring Street, New York, N.Y. 10013

To my son, Nicholas Charles,
and my wife, Debbie
DCR

To Barbara
JHK

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Preface

Since the late 1960s, the behavioral treatment of mentally retarded, autistic, and other developmentally disabled persons has grown progressively more sophisticated. The literature on behavioral treatment has produced effective and clinically significant programs for the reduction of maladaptive behaviors such as tantrums, aggression, and self-injury; skills deficits have been remediated through improved programs for language and life skills acquisition; and new environments have been opened in the creation of mainstream educational opportunities.

In spite of these advances, it strikes us that this almost exclusive focus on behavior problems and skills remediation has been somewhat myopic and that much of the potential for application of behavioral science to solving problems of the developmentally disabled is as yet untapped.

In the 1980s, an important revolution has taken place: the development of the field of behavioral medicine. This field, in merging disease treatment and management with learning and behavior, has already made impressive progress toward a reconceptualization of health care that acknowledges the centrality of behavior in disease expression. Although there has, as yet, been only a preliminary application of this reconceptualization to the field of developmental disabilities, we are convinced that further extension has great potential.

This volume is therefore dedicated to exploring the potential and utility of an integration of the fields of behavioral medicine and developmental disabilities. To accomplish this, we have invited individuals whose research and clinical practice are representative of this integration to review existing experimental work, speculate on future applications, and provide guidance and stimulation to new arrivals to the field. The reader may find that the contents of this book do not cover all possible areas of interface between behavioral medicine and developmental disabilities. Rather, we have selected topics in which a sufficient body of

empirical knowledge or clinical expertise has accrued to allow for meaningful review and speculation.

We have subsumed the contents of this volume under three major headings: symptom management, assessment, and educative interventions. Because of the emerging character of the field, each specific area of application (e.g., prevention, feeding disorders, assessment of pharmacotherapy) is represented by both a primary contribution and one or more briefer, supplementary chapters, designed to provide additional perspective. We feel that this format of counterpoint and dialogue well reflects the evolving nature of behavioral medicine.

We wish to thank, in particular, Judy Favell and Richard Foxx, former coeditors of *Analysis and Intervention in Developmental Disabilities*, with whose support the initial idea for this book was conceived. Our deepest gratitude goes as well to Lynda Ferguson, who coordinated its assembly. We are hopeful that the reader will find these chapters instructive and a rich basis for future work in this evolving area.

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CHAPTER 1

Behavioral Medicine with the Developmentally Disabled

Major Issues and Challenges

Jurgen H. Kedesdy and Dennis C. Russo

The application of behavioral medicine to the developmentally disabled has a short history, presently characterized by underachievement, and (so like an underachiever!) a brilliant future. We will briefly review here the origins, current status, and the future of this important and growing field.

Origins

As terms that reference organized fields of study and coherent areas of practice, both *behavioral medicine* and *developmental disabilities* have remarkably brief histories. The 1977 Yale Conference on Behavioral Medicine (Schwartz & Weiss, 1978) marked the first major public recognition of behavioral medicine as a focus of common concerns, research interests, and procedures in an otherwise diverse group of professionals from the behavioral and health sciences. The definition of behavioral medicine

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that emerged from that conference, although not unanimously embraced by all behavioral medicine practitioners, provides a suitable point of embarcation for our consideration of the field:

> Behavioral medicine is the field concerned with the development of behavioral science knowledge and techniques relevant to understanding physical health and illness and the application of this knowledge and these techniques to prevention, diagnosis, treatment, and rehabilitation. (Schwartz & Weiss, 1978, p. 7)

The key elements of this definition identify a domain constructed around the intersection of the terms *behavioral science* and *physical disorder*. Emphasizing these terms establishes a discrimination that implicitly excludes from consideration other, often longer, histories of related concerns implied by the terms *psychosomatic medicine* (not always behavioral) on the one hand, and *behavior modification* (not usually concerned with physical disorders) on the other (cf. Russo & Varni, 1982).

Although many of the techniques employed in behavioral medicine have longer histories, as even more obviously do the physical disorders of the developmentally disabled to which these techniques are applied, it is difficult to find studies, or records of practice, earlier than the 1970s in which the clinical and methodological techniques of the behavioral sciences are applied to the analysis, amelioration, or prevention of medical problems in the developmentally disabled.

The emergence of behavioral medicine as a unified field depended upon at least three prior developments: (a) a growing body of scientific knowledge demonstrating the importance of environmental and behavioral factors in somatic functioning, (b) the successful application of behavioral techniques in the arena of health care, and (c) changes in the practice of medicine itself.

Increasing demonstration of the effects of environmental and psychological factors on, for example, cardiovascular (e.g. Engel, 1977), gastrointestinal (e.g. Weiss, 1984), and immunological (e.g. Jemmott & Locke, 1984) systems led to growing optimism about behavioral treatment options suggested by these findings (see Miller, 1983, and Russo, Tarbell, & Follansbee, in press, for general reviews of these issues).

Demonstrations of the efficacy of behavioral approaches occurred during a time of ongoing changes in the practice of medicine, chiefly a shift in emphasis from acute illness and infectious disease to chronic illness and preventative health care strategies (Russo & Varni, 1982). As medicine confronted the problems of chronic illness, the social and psychological contexts of health and illness were increasingly recognized, and psychosocial approaches that addressed the patient's well-being in addition to the physical basis of illness were increasingly valued (Mishler

et al., 1981; Russo, 1986). These historical forces proved a fertile ground for multifactorial models of health and illness addressed from a multidisciplinary perspective.

The field of "developmental disabilities," on the other hand, was fashioned as much from advocacy, social policy, legislation (e.g., the Developmental Disabilities Services and Facilities Construction Act of 1970) and litigation (e.g., Katz-Garris & Garris, 1983; Rothman & Rothman & Rothman, 1985) as from advances in science or clinical acumen. This is certainly not to say that important scientific advances (especially in genetic research and medical technology) have not helped to shape the field; only that these advances depended upon and were energized by ideology and moral conviction. It will be important to remember that the field we are addressing in this volume rests upon both a scientific and an ethical foundation; indeed, the growth and vitality of the field will depend upon a sustained moral conviction that developmentally disabled persons have a right to the same range and sophistication of services as the rest of the population. The continued extension of behavioral medicine techniques to the developmentally disabled may depend as much on advocacy as a demonstration of efficacy.

Current Applications

The contributions of behavioral medicine to clinical practice can be partitioned into four general domains: (a) the behavioral assessment of medical interventions, (b) behavioral interventions for symptom management, (c) interventions to increase compliance with medical regimens, and (d) behavioral techniques aimed at the prevention of illness and disease. Extensions to clinical practice with developmentally disabled persons have occurred within each of these domains, although not to an equal degree.

Assessment

The use of behavioral assessment technology, including a commitment to empiricism, observability, and functional analysis, to assess medical interventions with the developmentally disabled is an area in which considerable progress has already been made. For example, straightforward applications of observational recording instruments and single-case research designs in the evaluation of psychotropic medication effects in mentally retarded patients, although not yet as routine

as one would wish, are increasingly viewed as a necessary adjunct to responsible practice (see Schroeder, Chapter 9). Quite often the use of behaviorally defined, data-based decision-making has served to indicate an absence of therapeutic effect for major psychotropic medications (Aman, White, Vaithianathan, & Teehan, 1986; Singh & Millichamp, 1985) and had the ethically beneficial effect of removing individuals from unefficacious medication regimens. As Schroeder points out, psychoactive medication effects in the mentally retarded are often quite idiosyncratic. When the effects of powerful medications cannot be confidently predicted for specific individuals, it is especially important for assessment to be objective, reliable, and *idiographic*. When, in addition, the patient's ability to report the effects of medication is compromised by impaired or undeveloped cognitive and verbal functioning, observational recording and other behavioral assessment tools are especially appropriate.

Behavioral assessment technology has also been extended to the evaluation of medical interventions for seizure disorders, a condition with an estimated prevalence of between 23% and 50% in the mentally retarded population (Corbett, Harris, & Robinson, 1975). Behavioral assessment, coupled with radio-telemetered EEG, has been useful both in establishing an accurate diagnosis by discriminating between pseudoseizures and true seizures (Holmes, McKeever, & Russman, 1983; Neill & Alvarez, 1986) and by assessing the effects of seizures on cognitive and adaptive functioning (Neil & Alvarez, Chapter 5).

Finally, behavioral assessment has played an important role in identifying and measuring iatrogenic effects of both neuroleptic and anticonvulsant medications. Tardive dyskinesia, a movement disorder that may emerge secondary to the long-term use of antipsychotic drugs, is an increasing recognized problem in the mentally retarded (Gualtieri & Hawk, 1980) and several behavioral scales have been developed to assess the emergence of drug-induced movement disorders (Campbell, Green, Perry, & Bennett, 1983). Behavioral test batteries are also under development to assess behavioral toxicity (e.g., cognitive obtunding) associated with anticonvulsant medications (Novelly, Schwartz, Mattson, & Cramer, 1986). This represents an important development, because it appears that anticonvulsant drug toxicity in the mentally retarded is not uncommon, cannot usually be self-reported by cognitively impaired persons, and may be difficult for caregivers to discriminate (Aman, Paxton, Field, & Foote, 1986).

Symptom Management

The area of behavioral medicine research and practice that holds perhaps the most promise and presents us with some of the greatest

challenges is symptom management. Behavioral approaches to symptom management in developmentally disabled persons have involved three different types of interventions, either alone or in combination: (a) contingency management, (b) arousal modification, and (c) specific application of biofeedback. Contingency management has been, by far, the most common symptom management approach.

Contingency Management

Illustrative examples of behavioral medicine interventions of this type are behavioral protocols developed for the treatment of nonorganic feeding disorders, such as food refusal/selectivity and failure to thrive (FTT) (Handen, Mandell, & Russo, 1986; Riordan, Iwata, Finney, Wohl, & Stanley, 1984). In these cases, treatment consists of a fairly straightforward arrangement of differential consequences for appropriate and inappropriate food-related behavior (e.g., positive attention for food consumption coupled with ignoring and brief time-out for food refusal as well as shaping, fading, and other stimulus control manipulations), use of preferred foods as a consequence for consumption of nonpreferred foods (Riordan, Iwata, Wohl, & Finney, 1980). It is important to note that although behavioral interventions assume that learning has played a major role in the etiology of these feeding disorders, the interventions have been shown to be successful with children with very complicated medical presentations. Indeed, as Ginsberg (Chapter 2) points out, behavioral treatments have been very useful for the increasing incidence of iatrogenically induced feeding disorders that develop indirectly from the use of medical procedures (e.g., parenteral feeding) used to treat very ill children.

Additional applications of contingency management to feeding and eating disorders may be found in weight-reduction programs for the mentally retarded (see Burkhardt *et al.*, Chapter 17), weight management and food stealing in persons with Prader-Willi syndrome (see Page, Chapter 18), and the treatment of psychogenic vomiting and rumination (Marholin, Luiselli, Robinson, & Lott, 1980; Mulick, Schroeder, & Rojahn, 1980).

In many cases, contingency management of symptoms is the treatment of first choice for disorders that come to the attention of medical professionals. Incontinence is a case in point. Both urinary and fecal incontinence are only rarely organic in origin, with the prevalence of urinary incontinence of known and specific organic origin between 1% and 10% (Sorotzkin, 1984). When there is no specific physical basis for the disorder, contingency management may be considered a more benign intervention than the alternative, for example, treatment with im-

ipramine (see Moe, Chapter 7). Contingency management may also be the most reasonable treatment when there is obvious organic involvement, as will often be the case with developmentally disabled individuals. For example, Jeffries, Killam, and Varni (1982) have described the application of a differential reinforcement program to establish fecal continence in a 9-year-old with sacral-level myelomeningocele and reduced anal sensation.

Arousal Modification

Arousal modification, relaxation training, and stress management techniques have been widely applied to symptom management for both adults and children. Although applications of these techniques to the developmentally disabled are still preliminary and not entirely without controversy (cf. Luiselli, 1980), the outlook is generally quite promising. Several studies have reported the use of some variations of progressive muscle relaxation (Jacobson, 1938) with developmentally disabled children and adults, and a manual describing the extension of these techniques to children with special needs is available (Cautela & Groden, 1978). Wells, Turner, Bellack, and Hersen (1978), for example, taught the use of cue-controlled progressive muscle relaxation to a 22-year-old female with a diagnosis of borderline mental retardation and a psychomotor seizure disorder. This technique served to reduce the incidence of both self-reported seizures and anxiety.

Arousal modification in the developmentally disabled has also been approached using biofeedback-assisted self-regulation techniques. A number of studies (e.g., Cataldo, Bird, & Cunningham, 1978; Finley, Niman, Standley, & Ender, 1976) have shown beneficial effects of biofeedback-assisted reduction of EMG in children and adults with athetoid cerebral palsy. There have also been interesting applications of EMG feedback arousal modification in the treatment of severe behavior disorders (e.g. Hughes & Davis, 1980; Schroeder, Peterson, Solomon, & Artley, 1977).

Specific Biofeedback Applications

Although biofeedback is often used to assist in the achievement of general reduction of arousal, specific applications of biofeedback are those in which the immediate target response is also the response of clinical concern. For example, Whitehead, Parker, Masek, Cataldo, and Freeman (1981) have described a biofeedback treatment of fecal incontinence in children with myelomeningocele that involved teaching pa-

tients external anal sphincter contraction in response to rectal distension. The technique used in this and similar studies consists largely in making information from rectosphincteric manometry continuously available to patients and shaping voluntary external sphincter contraction.

Compliance with Medical Regimens

Many courses of medical treatment, especially for chronic disorders, such as asthma, diabetes, or rheumatoid arthritis, require the active cooperation and participation of the patient and/or the patient's caregiver. Adherence to therapeutic regimens and techniques to promote it have attracted considerable recent interest (Haynes, Taylor, & Sackett, 1979; Masek & Jankel, 1982). Medical noncompliance is an issue of special importance in the developmentally disabled population, but it has not, as yet, received much systematic attention.

Some areas that merit attention are studies of the effects of behavioral contingencies in promoting self-administration and compliance with medication regimens in adult mentally retarded individuals, behavioral interventions to increase compliance with dietary requirements in children with PKU, and systematic application of behavioral principles to increase compliance with medical exams. An interesting example of the latter is a recent report of the use of a systematic desensitization to enhance cooperation with gynecological examination in an autistic adolescent (Dahlquist, Gil, Kalfus, Blount, & Boyd, 1984).

In many cases, it is the parents and caregivers of developmentally disabled children and adults that implement medical and habilitative regimens. Lewis and Drabman (Chapter 15) review research on training parents to implement physical and occupational therapy programs for their developmentally disabled children. Factors that would promote optimal compliance with home-implemented programs have yet to receive systematic study. Lewis & Drabman and Budd (Chapter 16) suggest that viewing the problem of compliance from a multicausal, systemic perspective that includes an appreciation of the emotional stress associated with chronic care as well as a focus on building general problem-solving skills in families may be useful.

Prevention

Prevention in developmental disabilities has at least two important dimensions: the prevention of developmental disability itself and the primary prevention of illness and disease in developmentally disabled

populations. Behavioral contributions to both areas are preliminary but promising.

Finney and Friman (Chapter 13) review behavioral approaches to the prevention of developmental disabilities that accrue from fetal alcohol syndrome, phenylketonuria, lead poisoning, and motor vehicle trauma. Although the contribution of health education to the prevention of developmental disability is reasonably well appreciated, the health education model assumes that information alone is a sufficient condition for behavior change. This is often not the case, especially when the consumers of information are impaired (e.g., alcoholic mothers) or when the behavior relevant to prevention must be sustained over long periods of time (e.g., dietary regimens in PKU). In such cases, compliance-enhancement technologies and behavior management can also play important roles. The reduction of mouthing and pica in children at risk for lead ingestion (Finney, Russo, & Cataldo, 1982) is a good example of the use of a well-developed behavioral technology in the service of prevention.

Health promotion as a strategy to reduce morbidity and mortality in developmentally disabled persons has not received the attention it deserves. Probably the best developed area of health promotion is obesity reduction in mentally retarded children and adults (see Burkhart *et al.*, Chapter 17). The health risks associated with obesity, including coronary artery disease, pulmonary, renal, and liver problems, are well documented (Van Itallie, 1979), and developmentally disabled persons appear to be at least as likely to become obese as are persons in the normal population. There are, in addition, a number of syndromes in which obesity is characteristic, including Down's syndrome, Cohen syndrome, and Prader-Willi syndrome.

Behavioral treatment of obesity ordinarily includes multiple components such as reinforcement of weight loss, self-monitoring, stimulus control, modification of eating rate, reinforcement of energy expenditure, and nutritional education. Although the relative contribution of each of these components is not always clear, the use of multicomponent treatments may simply reflect the multifactorial nature of the disorder. In this sense, the behavioral treatment of obesity illustrates the multifactorial conceptualizations that often characterize the field of behavioral medicine.

Theoretical/Contextual Issues

As a field concerned with the interdisciplinary study of biobehavioral relationships relevant to health and disease, behavioral medicine

is consistent with the biopsychosocial model of medicine proposed by Engel (1977). The biopsychosocial model, in contrast to the traditional biomedical model, assumes that health and disease cannot be fully understood outside their psychosocial contexts. We will discuss some of the biobehavioral relationships that seem especially relevant to the practice of behavioral medicine with the developmentally disabled.

Learning and Illness

One of the most central assumptions of a behavioral medicine approach is that learning and illness (or health) are related in normal and potentially predictable ways. One way to view this particular biobehavioral relationship is to conceptualize illness as having potential for entry into the traditional three-term contingency of operant conditioning at each of the three available points.

Thus, illness may function as a discriminative stimulus (and/or as a setting event), as a response, or as a consequence. For example, illness sets the occasion for unique behavioral repertoires (e.g., taking medication, following rehabilitation programs) that may be functional only during illness. In addition, illness may function as a setting event to alter the probability of repertoires established under other conditions. For example, illness may increase the likelihood of a response class of "dependency," which may have been established during an earlier developmental period, thereby producing the impression of "regression."

The extent to which individuals learn from being ill is, of course, magnified during chronic illness. As Russo (1986) has observed: "Chronicity teaches." This is an especially pertinent consideration for the developmentally disabled in whom specific chronic medical conditions, such as seizure disorders, are highly prevalent, and who, by definition, have a chronic primary disability.

Second, illness may itself be learned and, even when the original basis for illness is largely organic, the behaviors that express illness may have a major operant component. For example, behaviors that express pain (e.g., grimaces, guarded posture, complaints) may be strengthened by sympathy and work avoidance (Fordyce, 1976). It is not surprising, therefore, that chronic pain disorders often respond incompletely to pharmacological and surgical procedures and that chronic pain treatment programs are increasingly based on the biopsychosocial model.

Finally, illness and health may be seen as the natural, albeit relatively long-term, consequences of behavioral practices, as, for example, coronary heart disease is associated with certain life-styles (Glass, 1977). The burgeoning field of health promotion, which carries with it the hope

of reducing the costs of medical care, is built upon the assumption that the probability of illness and disease can be substantially altered by changing the probability of behavior (Matarazzo, Weiss, Herd, Miller, & Weiss, 1984).

This three-term conceptualization of behavior and illness is by no means exhaustive, nor is it intended to suggest that all important bio-behavioral relationships are operant (Russo & Budd, 1987), but it does provide a useful starting point for the preliminary analysis of any case from a behavioral medicine perspective. It encourages us to ask, in each individual case the following questions:

1. What is (or was) learned by the patient during this illness or disability?
2. To what extent is the illness (or disability) learned?
3. How have learned behaviors contributed to this state of illness or disability?

As a clinical heuristic, this approach has the advantage of urging us to view the often maladaptive behaviors related to illness from the perspective of *normalcy* (Russo, 1986). Because there is often a tendency to assume that disability or pathology accounts for most of the variance in the behavior of developmentally disabled persons, an emphasis on the normal operation of learning principles can provide a beneficial counterpoise.

Medical and Behavioral Disorders

The interaction between behavior and illness in developmentally disabled persons is often especially convoluted and sometimes quite deceptive. As Gourash (1986) has noted, behavior can "masquerade" as a medical problem (e.g., pseudoseizures), and symptoms of organic disorder can masquerade as behavior disorders, as when, for example, gastroesophageal reflux may be mistaken for psychogenic vomiting. Quite often medical and behavioral disorders exist concurrently and are often expressed in similar, even identical ways. For example, Neil and Alvarez (1986) found that roughly half of their institutionalized, mentally retarded patients with a conclusive diagnosis of epilepsy also exhibited pseudoseizures and that there was no intrasubject basis for distinguishing between the topography of true and pseudoseizures.

The complex interaction of behavioral and medical factors creates an enormous potential for error, ranging from misdiagnosis to malpractice, and raises important professional issues of interdisciplinary communication, collaboration, and competence (Russo & Tarbell, 1984).

There is widespread recognition that appropriate diagnosis and treatment planning for developmentally disabled children and adults require an interdisciplinary process (Johnston & Magrab, 1976), but behavioral medicine requires an unusual amount of communication and reciprocal education among behavioral and medical practitioners in a process that is better described as *collaborative* than interdisciplinary. Furthermore, behavioral practitioners, especially, should be keenly aware of the boundaries of their competence and understand that they are vulnerable to charges of practicing medicine without a license (Knapp & Vandecreek, 1981).

The possibility of misidentifying behavioral and medical factors is especially high in the most severely impaired individuals, in whom the likelihood of both medical and behavioral disorder is proportionately higher, because diagnosis cannot depend upon self-reported symptoms.

Effects of Cognitive Dysfunction on Medical Care

From the perspective of the traditional biomedical model, in which illness is reduced to disordered biochemical and neurophysiological processes, medical care, addressed as it is to the amelioration of those processes, would not be expected to vary significantly as a function of the cognitive capacities of the patient. It is, of course, doubtful that any medical practitioner would seriously maintain such a position today, but very little research or systematic clinical attention has been directed to the understanding of how medicine is practiced with mentally retarded patients.

One important question is how much medical diagnosis is compromised in cases where self-report of the patient's symptoms is unavailable. Headache, for example, is a highly prevalent disorder in both pediatric and adult populations (Leviton, 1978; Sillanpaa, 1983). The diagnosis and classification (e.g., migraine versus muscle contraction) of headache depends heavily on descriptions of location (e.g., frontal versus temporal) and type of pain (e.g., pulsating versus steady). How is a definitive diagnosis of headache made in a patient unable to describe pain?

Nor is this problem restricted to the diagnosis of headache. Pain and discomfort are often thought to be antecedent or contributing conditions to the expression of behavior disorder. Self-injury, for example, sometimes is thought to express or ameliorate pain (Cataldo & Harris, 1982), and some studies have shown that self-injurious children suffer a relatively higher incidence of otitis media (DeLissovoy, 1963). The accurate determination that maladaptive behavior expresses pain de-

pends upon an *independent* assessment of pain—a task made more difficult in nonverbal patients.

In this connection, children and adults in pain are often treated differently. Schechter, Allen, and Hanson (1986), for example, showed that fewer postoperative analgesics are prescribed for children than adults. It would be important to know whether a similar bias obtains in analgesic prescription for the developmentally delayed. If this were so, underprescription of analgesics might be shown to play an important role in the etiology of behavior disorders.

Medical diagnosis is also heavily dependent on the patient's cooperation with medical tests. Many tests that are crucial to accurate diagnosis of medical disorders with high prevalence in the developmentally disabled, for example, radio-telemetered EEG, are difficult to obtain with patients who are aggressive, noncompliant, or otherwise behaviorally disordered. It is reasonable to suppose, therefore, that the concurrent presence of medical and behavioral disorder may compromise medical treatment.

Future Applications

Assessment

Behavioral assessment can and will continue to play an important role in the provision of quality health care for developmentally disabled persons. There is an ongoing need to objectively evaluate medications, especially the more experimental applications of psychopharmacology practiced with severely impaired individuals (e.g., Szymanski, Kedesdy, Sulkes, Cutler, & Stevens-Our, 1987). As Schroeder (Chapter 9) advises, closer collaboration between biomedically and behaviorally trained researchers can result in interventions that are both driven by biologically sophisticated theories and subjected to rigorous empirical tests.

Behavioral and psychosocial assessment can also make contributions to our understanding of the effects of other types of medical interventions. A fertile area of application may be surgical procedures, especially those such as fundoplication (Wilkinson, Dudgeon, & Sondheimer, 1981) or craniofacial surgery (Pertschuck & Whitaker, 1982), that are performed with relatively greater frequency in developmentally disabled persons.

Two issues should be stressed in this connection. First, it seems important, from an ethical point of view, to evaluate the psychosocial effects of procedures, especially elective procedures, when those pro-

cedures have not been chosen by the patient and when the patient cannot fully anticipate the personal consequences of those procedures. Second, because many of the traditional psychosocial measures that have been employed to assess the effects of medical procedures in adults and older children have limited utility with cognitively impaired patients, the observation-based technologies of behavioral assessment may be of special value.

Finally, many of the most challenging problems of assessment and treatment occur with "low-prevalence/high-severity" (see Schroeder, Chapter 9) disorders, for example, self-injury or Prader-Willi syndrome. These disorders raise significant questions of replicability of treatment, adequate intensity of resource allocation, and sufficiency of professional expertise. There is, therefore, an evident need for specialized behavioral medicine clinics dedicated to the assessment and treatment of low-prevalence/high-severity disorders.

Symptom Management

An area of symptom management that seems especially promising to extend is that of arousal modification and stress management. There is considerable room for both new applications and increased methodological rigor. Although limited data on the presence of stress-related disorders exist for this population, there seems little doubt that developmentally disabled persons are at high risk. For example, Chaney, Eyman, Givens, and Valdes (1985) found the prevalence of peptic ulcers among mentally retarded residents of a large residential facility to be higher than would be expected in the general population by a factor of about 3.

There are a number of reasons developmentally disabled persons may be underserved in this respect. This may, in part, be due to a general tendency to underestimate emotional difficulties in mentally retarded persons (Reiss, Levitan, & McNally, 1982) and in part to a belief that stress management and relaxation procedures may have limited utility for cognitively impaired persons. The latter question has not received sufficient empirical attention, but a recent study by Calamari, Geist, and Shahbazian (1987) is suggestive. These authors found that a multicomponent relaxation procedure, using progresive muscle relaxation, EMG biofeedback, modeling, and reinforcement, reduced EMG and activity in a group of 32 developmentally disabled persons who functioned in the profound to mildly retarded range of cognitive ability. Notably, level of intelligence and adaptive behavior functioning did *not* predict relaxation training success in these clients. Clearly, the relationship between

cognitive functioning and response to arousal modification procedures merits further study.

Compliance

As noted before, adherance to therapeutic regimens is a major issue in this population. Finney and Friman (Chapter 13) have described multiple compliance issues that occur in the prevention of developmental disability. Children with PKU, for example, must comply for years with a restrictive diet, medication schedules, and regular monitoring of phenalynine levels. Noncompliance with part or all of this regimen can reach crisis proportions, sometimes requiring hospitalization of the child or of the phenylketonuric mother during pregnancy. Similar issues occur in children with other inborn errors of metabolism, such as galactosemia.

In spite of considerable clinical consciousness of these issues, they have not received systematic study. It seems clear that the most common approach to these problems, patient education, might well be supplemented by more structured behavioral analysis and intervention for compliance enhancement.

Prevention

It has been estimated that nearly half of the mortalities from the 10 leading causes of death in this country can be attributed to unhealthy life-styles (Califano, 1979). The modification of health-related behavior can therefore have a major impact on death and disability. In spite of considerable awareness of these issues in the general population, little is known about health-related behavior in mentally retarded persons and, with the exception of obesity (see Burkhart *et al.*, Chapter 17), there have been few systematic attempts to study or modify health-related behavior in developmentally disabled persons.

A reasonable way to begin to remedy this shortcoming is to extend the techniques and philosophy of health enhancement into areas like smoking cessation that have been extensively promoted in the normal population but barely considered in disabled populations. Indeed, as Burgio (1987) observes in a recent study, in spite of the well-documented health hazards of smoking, cigarettes are routinely used as reinforcers in behavioral programming with mentally retarded persons. Burgio's study demonstrated the efficacy of using self-management procedures to reduce cigarette smoking (and caffeine consumption) in a mildly retarded adult male. Reduction of caffeine consumption had the secondary benefit of eliminating intractable nocturnal enuresis in this client.

It has also recently been pointed out that health education is an area in which developmentally disabled adults are apparently underserved (Brooks-Bertram, 1986). We would agree that this is an important area for future research.

Conclusions

Health care for developmentally disabled persons often raises a complex set of treatment and research issues, many of which occur at the intersection of the behavioral and medical sciences. There are at least two reasons why we should take special notice of these issues. First, although many behavioral medicine issues are generic with respect to patient population, others are not. We have, for example, raised the question of how medical diagnosis and subsequent treatment may be influenced by the cognitive impairment of the patient and have drawn attention to the special challenges to understanding and care posed by low-prevalence/high-severity disorders. Second, although the need for behavioral medicine services for developmentally disabled persons is as urgent as for any other population, if not more so, it is not clear that these services are routinely available for most mentally retarded or otherwise handicapped people. Traditionally, biomedical and behavioral research and treatment have been conducted independently, and clear structural lines continue to exist in many settings where the developmentally disabled are served that maintain the dichotomy between biomedical and behavioral services and that may actively discourage collaborative effort.

Behavioral medicine promises to make important contributions to improved health care for developmentally disabled persons. We believe this promise can be realized by building on the empirical and conceptual foundation provided in this volume and that the commitment to building upon this foundation raises, perhaps one notch higher, our "standards of compassion and dignity" (Kennedy, 1963).

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Part I

Symptom Management

CHAPTER 2

Feeding Disorders in the Developmentally Disabled Population

Amy J. Ginsberg

Consistent with the growing focus on health promotion, eating and other food-related problems have received increased attention in the behavioral medicine literature. Experimental investigations have spanned a continuum of eating problems from eating excesses, such as obesity and bulimia, to eating deficits, such as anorexia nervosa and nonorganic failure-to-thrive. More recently, researchers have attended to the treatment of organic feeding problems that may be associated with a chronic medical illness (such as short gut syndrome) or produced iatrogenically secondary to medical treatment (e.g., Ginsberg & Klonoff, 1985).

Feeding problems are particularly salient in developmentally disabled populations, occurring in as much as 6% to 33% of handicapped individuals (Coffey & Crawford, 1971; Palmer & Horn, 1978). Treatment of feeding problems in this population may be particularly challenging as the treatment must be designed to correspond to the appropriate developmental level. In addition, developmental disabilities appear to put the individual at increased risk for, or are at least highly correlated with, certain food-related disorders that are generally unique to this

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Table 1. A Classification of Feeding Problems in Delayed and Nondelayed Populations

Feeding problem	Populations	
	Nondelayed	Delayed
Insufficient food consumption	Anorexia nervosa Organic failure-to-thrive	Selective food refusal Psychosocial failure-to-thrive
Excessive food consumption	Obesity Bulimia (bingeing)	Obesity Pica
Inappropriate interactions with consumed food	Bulimia (purging)	Rumination Psychogenic vomiting

population and that are often resistant to treatment. For example, ruminative vomiting is reported much more frequently in delayed populations.

As reflected in Table 1, feeding problems in both delayed and nondelayed populations can generally be classified into three major areas: (a) insufficient consumption in terms of amounts or varieties of food, (b) excessive or inappropriate consumption, and (c) failure to maintain appropriate interactions with previously consumed food. However, the specific feeding disorders that are incorporated by this classification system may vary across delayed and nondelayed populations. For example, insufficient consumption in nondelayed populations often occurs in the form of anorexia nervosa, whereas in delayed populations, it may be manifested as selective food refusal. Similarly, in nondelayed populations, purging techniques associated with bulimia represent one type of inappropriate interactions with previously consumed food. In delayed populations, bulimia is infrequently reported, whereas rumination and psychogenic vomiting are more common examples of a failure to interact appropriately with consumed food.

This chapter will further explore this feeding classification via a selective review of the literature with an emphasis upon the adaptations necessary to treat feeding problems in the developmentally disabled population. In addition, future directions for research and clinical activity in this area will be addressed. Throughout the paper, particular emphasis will be placed on the interface between behavioral medicine and developmental disabilities. Thus, the extensive literature on teaching self-feeding skills and appropriate table manners to developmentally disabled populations will not be included in this review as it falls more within the realm of behavior modification than within the behavioral medicine domain; the reader is referred elsewhere for this literature

(e.g., Azrin & Armstrong, 1973; Baker, Brightman, Heifetz, & Murphy, 1976; Barton, Guess, Garcia, & Baer, 1970; Iwata, Riordan, Wohl, & Finney, 1982; O'Brien & Azrin, 1972).

Consumption of Inappropriate Amounts and Varieties of Food

Consumption of inappropriate amounts and varieties of food can be manifested either in the form of behavioral deficits or behavioral excesses. Thus obesity resulting from consumption of excessive amounts of food may be conceptualized to be as inappropriate as selective food refusal (refusal to accept a wide variety of tastes and textures) and failure-to-thrive. When taken to the extreme, inappropriate consumption can pose a serious health hazard; that is, both morbid obesity and total food refusal may result in compromised medical status.

Insufficient Consumption

With few exceptions (e.g., Szymanski & Biederman, 1984), anorexia nervosa is described exclusively in nondelayed populations. In contrast, selective food refusal, a disorder similarly characterized by restrictive intake, is more often described in developmentally disabled and autistic populations. Selective food refusal consists of insufficient consumption of a normal array of tastes and textures; thus it may be exhibited as multiple food dislikes or as prolonged subsistence on soft or pureed foods. The causes of this selective food refusal may vary, with one study of 52 cases reporting that 79% of the feeding problems were attributable to neuromotor dysfunction such as dysphagia, 4% were attributable to mechanical obstructions, and 17% were due primarily to environmental factors (Palmer, Thompson, & Linscheid, 1975). It is unclear to what extent inappropriate tongue movements, seen at higher rates in developmentally disabled children than in nondelayed children (Gisel, Lange, & Niman, 1984), may contribute to food refusal. At any rate, in the case of neuromotor dysfunction, techniques for appropriate positioning and for the adaptation of utensils (Kosowski & Sopczyk, 1985), as well as for controlling spastic tongue movements (Thompson, Iwata, & Poynter, 1979), have been developed to increase the probability of successful food acceptance. In the nonhandicapped population, neuromotor dysfunction does not appear to be implicated in feeding problems as often, although these individuals may also suffer from mechanical obstructions and other organically based difficulties.

Although a large percentage of feeding problems in developmentally disabled populations are attributable to nonpsychosocial factors, the majority of interventions have targeted environmentally based feeding problems. For example, gradual shaping and fading procedures, praise, attention, and positive reinforcement in the form of preferred pureed foods have been found to increase consumption of solid food when delivered contingently to children with nonorganically based solid food refusal (Bernal, 1972; Hatcher, 1979; Palmer *et al.*, 1975; Thompson & Palmer, 1974). Riordan, Iwata, Wohl, & Finney (1980) described a similar treatment program, employing preferred foods as reinforcers, for developmentally delayed children who consumed some solid foods prior to treatment but of insufficient variety. In addition, gradually thickening preferred pureed food with less preferred food has reportedly been effective in increasing subjects' tolerance for thicker, textured food (Clancy, Entsch, & Rendle-Short, 1969).

Many of the treatment approaches emphasizing principles of positive reinforcement also capitalize on deprivation effects. That is, if children are deprived of preferred foods until a less preferred food is consumed, the resulting increase in hunger might be a setting event or a "motivational" stimulus that facilitates reinforcement effects. Thus food deprivation may increase the probability that eating appropriately will come under contingency control. In some cases however, parents may be less likely to comply with a treatment program that is partially based on deprivation; in addition, such programs contain inherent risks of anemia and dehydration (Bernal, 1972). In response to this, Bernal (1972) recommended that a predetermined amount of water as well as vitamins be required components of such a program. She further suggested that, for cases in which weight loss is too stressful, one meal of strained or preferred food per day could be provided freely.

Total food refusal may be classified as an extreme version of selective food refusal. The causes may range from organic to learned etiologies, although, unlike anorexia nervosa, a psychiatric diagnosis is rarely entertained. Failure to accept even minimal food is generally more serious than selective food refusal, and the sequelae of developmental delays associated with total food refusal have been well documented (e.g., Beratis, Kolb, Sperling, & Stein, 1981). Like its counterpart of anorexia nervosa, the severity of total food refusal has been thought to justify the use of relatively forceful and aversive procedures, such as forced feeding techniques, despite the potential harm (such as injury and increased risk of aspiration) associated with physical prompts and forced feeding (Perske, Clifton, McClean, & Stein, 1977). Ives, Harris, & Wolchik (1978) described the use of a multicomponent forced feeding procedure with

an autistic-like child who accepted only instant breakfast drink. The authors noted that the procedure, used as a last-resort measure, may be necessary when shaping techniques cannot be employed due to the child's failure to sample new foods. However, alternative techniques warrant development as forced feeding procedures may be particularly difficult to implement at home due to parents' reluctance and to realistic fears of aspiration and choking.

Parenteral feedings, such as nasogastric or gastric tube feedings, may similarly be employed as "last resorts" when the child fails to consume sufficient calories to maintain health and promote growth. As with forced feedings, tube and intravenous feedings pose inherent risks as, in addition to the risks associated with surgery, they may cause ulcerations, sepsis, infection, and necrotizing enterocolitis (Holder, Leape, & Ashcraft, 1972; Schwartz & Maeda, 1985; Walsh & Kliegman, 1986). No clear guidelines appear to exist for choosing between forced feedings and tube feedings, assuming that less invasive approaches have been exhausted. In cases where the probability of success is high, such as with children who have a previous history of some appropriate food consumption, an attempt at forced feeding may be highly justified. Some physicians will implement tube feedings only after a child has failed all attempts at forced feeding. At other times, forced feeding may be medically contraindicated due to respiratory or gastrointestinal distress (e.g., Ginsberg & Klonoff, 1985), and parenteral feeds may therefore be required to sustain life. It is also likely that pediatricians and physicians differ in terms of their preferences for forced or tube feedings; physicians with less psychological sensitivity might more readily rely on medical or surgical procedures than on referrals to behavioral psychologists. It is likely that, in general, when a psychologist or learning specialist is involved in the treatment decision, tube feedings are utilized only when absolutely essential to sustain health.

A related feeding problem entailing eating deficits is psychosocial failure-to-thrive (FTT), a disorder generally diagnosed by weight falling below the third percentile on standard growth charts or by a flat growth curve for 2 to 3 months not associated with any known organic etiology. Children presenting with FTT at an early age display a high incidence of mental retardation (e.g., Drotar, Malone, & Negray, 1980), although cause and effect have not clearly been determined. As environmental neglect has been repeatedly implicated in FTT, many studies in the pediatric psychology literature have focused on family assessment and have involved the provision of maternal counseling and support services (e.g., Drotar, Malone, & Negray, 1979). These studies generally lack experimental rigor and provide insufficient data to allow for easy rep-

lication. Unfortunately, the behavioral literature on feeding problems has not generally addressed this population, and only a few operant treatment studies on nonorganic FTT exist (e.g., Larson & Ayllon, 1985). Finney and Christopherson (1983) note, however, that assessment procedures to evaluate both food acceptance and feeder–child interactions have been developed for other populations and would likely be readily applicable to the FTT population. Behavioral treatment for parents who are neglectful has been proven effective for nonfood problems (Wolfe, Kaufman, Aragona, & Sandler, 1981) and should readily generalize to eating delays. In addition, it has been suggested that tantrums, vomiting, and extreme selectivity occur in 25% of children with nonorganic growth failure and may be causally implicated (Hannaway, 1970). It is thus likely that behavioral techniques, aimed at decelerating those problem behaviors, would be effective in remediating at least some cases of nonorganic failure-to-thrive.

Excessive Consumption

In both delayed and nondelayed populations, excessive consumption is most often manifested as obesity. Binge episodes associated with bulimia, more common in nondelayed populations, are also characterized by excessive food consumption. In contrast, delayed populations exhibit higher rates of pica, a disorder that might be conceptualized as excessive oral consumption in that it consists of ingesting inappropriate and inedible items.

Pica, which can assume various forms, has been treated by a variety of operant procedures, including brief restraint (Bucher, Reykdal, & Albin, 1976), oral overcorrection procedures (Foxx & Martin, 1975), and environmental enrichment via increasing adult interaction and access to toys (Madden, Russo, & Cataldo, 1980b). A particularly hazardous form of pica involves the ingestion of items that contain lead. Pica, generally in the form of ingestion of paint chips, is the most frequently cited cause of severe lead poisoning (Lin Fu, 1973), a condition that has been linked to numerous neurological impairments including hyperactivity and developmental disability (e.g., Baloh, Sturm, Green, & Gleser, 1975). Recent studies utilizing simulated paint chips have demonstrated that behavioral techniques, including discrimination training, differential reinforcement procedures, and overcorrection can effectively decrease this form of pica (Finney, Russo, & Cataldo, 1982; Madden, Russo, & Cataldo, 1980a). These studies do not directly assess the etiology of pica but postulated hypotheses regarding causal factors include mater-

nal-child interaction, environmental deprivation, and poor nutrition (Madden et al., 1980a).

Obesity is a prevalent condition in the retarded population (Fox & Rotatori, 1982), although a recent investigation determined that obese retarded adults did not greatly differ from nonobese retarded adults in eating behavior (Fox, Burkhart, & Rotatori, 1983). Although the variables accounting for the maintenance of obesity in retarded adults are not well understood, it is known that certain forms of developmental disabilities may be associated with increased weight secondary to low energy requirements. For example, children with Prader-Willi syndrome, Down's syndrome, or spastic cerebral palsy may have lower energy needs in comparison to normal children and may therefore benefit from early modification of eating in the form of reduced caloric intake (Rice, 1981). However, behavioral treatments for obesity do not appear to differ greatly for delayed and nondelayed populations, and treatment manuals applicable to both groups are widely available (e.g., Stuart & Davis, 1972; Rotatori & Fox, 1981). The reader is referred elsewhere for a more extensive review of the obesity literature (see Chapter 17 by Burkhart, Fox, & Rotatori in this volume; Foreyt, 1977).

Inappropriate Interactions with Previously Consumed Food

In nondelayed populations, purging in the form of self-induced vomiting is a common component of bulimia. Nonorganically based vomiting is also often observed among the mentally retarded although generally in the form of ruminative vomiting rather than bulimia. Rumination poses particular treatment difficulty as the behavior may occur over a period of time without discrete onset and offset (Kohlenberg, 1970). Psychogenic or ruminative vomiting may seriously endanger physical health and, in such cases, brief applications of electric shock, contingent upon vomiting, have proven effective (Cunningham & Linscheid, 1976; Kohlenberg, 1970; Lang & Melamed, 1969). Various methods for the measurement of vomiting behavior have been described in the literature. Whereas Lang and Melamed (1969) administered shock contingent upon reverse peristalsis indicated by electromyograph (EMG) recordings, Kohlenberg (1970) and others (Cunningham & Linscheid, 1976) demonstrated that direct observations provide as valid a predictor of vomiting as do EMG data. Regardless of the measurement procedures employed, however, it has been suggested that vomiting may reoccur as much as 1 year after application of a punishment procedure, thereby indicating that a maintenance program is essential (Kohlenberg, 1970).

A review of treatment approaches for vomiting and rumination is provided elsewhere (Pazulinec & Sajwaj, 1983) and will not be discussed in detail here. The literature describes a variety of punishment techniques effective at decreasing ruminative vomiting, including applications of lemon juice (Sajwaj, Libet, & Agras, 1974; O'Neil, White, King, & Carek, 1979), Listerine mouthwash (Foxx, Snyder, & Schroeder, 1979) and Tabasco sauce (Murray, Keele, & McCarver, 1977) in conjunction with other procedures such as differential reinforcement for nonvomiting behavior. More recent attempts to decelerate vomiting have utilized the technique of food satiation (Foxx *et al.*, 1979; Rast, Johnston, & Drum, 1984) first described by Jackson, Johnston, Ackron, and Crowley (1975). This involves allowing free access to food rather than providing controlled meals. Data suggest that a functional relation, specifically an inverse relationship, exists between the amount of food ingested and rumination. Analytical studies are currently being conducted (Rast, Johnston, Ellinger-Allen, & Drum, in press) to clarify the variables controlling this phenomenon.

Future Directions

The preponderance of the literature described here involves treatment procedures derived directly from reinforcement and punishment theory. As a result, the literature is replete with techniques, derived from operant conditioning, which have also been applied to a host of other problem behaviors. If the field is to progress, additional studies of the controlling variables of feeding problems are as vital as the continuation of treatment studies. For example, Rast *et al.* (1984) note that it is unclear whether ruminative vomiting is an operant, respondent, or adjunctive behavior; it is therefore difficult to determine whether to manipulate social, dietary, or other unidentified variables. Although it has been demonstrated that satiation effectively decreases rumination, the theoretical rationale for this remains unclear as the variables controlling rumination have not been determined. Several hypotheses have been entertained, including the possibility that rumination is adjunctive in nature (Rast, Johnston, Drum, & Conrin, 1981) and that the oropharyngeal stimulation associated with rumination functions as a sensory reinforcer to maintain it (Ball, Hendricksen, & Clayton, 1974). Satiation may therefore be effective because it provides an alternative means of sensory stimulation to the oropharyngeal area. Similarly, it is not clear whether selective food refusal develops as a function of biochemical, social, olfactory, gustatory, or other yet unidentified variables.

For children with organic feeding problems, additional data are needed to explore the nature of the interaction between specific illnesses or medical procedures and specific feeding delays. Ginsberg and Klonoff (1985), for example, distinguished organic feeding problems that are associated with a chronic medical illness from feeding problems that are iatrogenically induced. As the treatment for these two categories of feeding problems may differ (Ginsberg & Klonoff, 1985), an understanding of the controlling variables may be crucial. The former category includes medical conditions that, by their very nature, directly affect one's ability to interact with food in a completely normal manner. For example, gastrointestinal disorders, such as short gut syndrome and malabsorption syndromes, often directly affect the ability to process food normally and may result in feeding difficulties. The actual process or results of digestion might be so aversive as to ultimately lead to increased food avoidance. The latter category, iatrogenically induced feeding problems, includes feeding problems that develop indirectly from medical procedures used to treat a disease. For example, although lung disease may not necessarily directly affect eating, feeding problems often develop secondary to ventilatory and respiratory support techniques.

The learning mechanisms underlying these iatrogenically induced feeding disorders are unclear, but several hypotheses may be considered. Feeding and swallowing may at times be incompatible with breathing for patients receiving mechanical respiratory support, and the patient's subsequent food avoidance often maintains, in the absence of mechanical support, due to conditioned fear of discomfort or lack of food experience. In addition, it has been suggested that a critical period exists in infancy for the development of appropriate eating (Illingsworth & Lister, 1964), and young infants who require sustained parenteral feeds for life support may exhibit feeding delays because they have missed crucial early learning experiences with food. As a result, these children often lack the skills to coordinate sucking, chewing, and the like. Parenteral feeds may also adversely affect the potential success of oral feeding because the patient may learn that the nonoral feeds are easier and require less effort. Both positive and negative reinforcement principles may be operative in the maintenance of oral food refusal and this vicious, coercive cycle may be difficult to break. For example, the behavior of parents and/or medical personnel "giving in," by supplying parenteral feeds to a child who refuses food orally, may be maintained (negatively reinforced) by the termination of an aversive stimulus such as the child's crying while the child may consequently learn to "hold out" since oral food refusal is positively reinforced by delivery of the easier, parenteral feed. Also, prolonged use of parenteral feeds via na-

sogastric or gastrostomy tubes may result in internal damage, such as ulcerations, causing increased discomfort when feeding orally and thereby contributing to food avoidance. Conditioned food aversions which may develop as a result are often very resistant to treatment. Finally, the transition to oral feeds may involve inherent risk; medical complications such as aspiration may negatively affect learning to accept oral feeds, and, in rare cases, death has been reported (e.g., Blackman & Nelson, 1985).

Given the need for increased accountability in medical settings, future research should also focus on issues of efficacy and clinical utility. Four major specific issues will be discussed here. First, consistent with the current focus on health promotion and illness prevention, the importance of prevention and early identification of feeding problems will be considered. A second and related issue concerns the need for further investigation of the variables contributing to length of treatment and subsequent follow-up. Third, problems with maintenance and generalization of treatment gains will be explored. Relapse prevention may also be relevant to this issue. Finally, methodological concerns will be addressed. Factors contributing to the difficulty in establishing adequate experimental designs and selecting appropriate dependent variables will be described.

Prevention and Early Identification

As more premature and chronically ill infants survive due to new developments in medical technology, the need for behavioral consultation regarding feeding should increase due to the frequent need to parenterally feed these neonates. A large percentage of infants on neonatal intensive care units present with low birth weight and corresponding developmental delay. As such, it is conceivable that the number of young developmentally disabled children with conditioned food aversions and dependence on tube feedings will increase. Routine behavioral consultation might lead to the development of methods of preventing feeding delays and dependencies on tube feedings and should also result in earlier identification of children at risk for organically based feeding problems (Ginsberg & Klonoff, 1987).

Relatively few studies have clearly described behavioral techniques to increase the oral acceptance of food in children maintained by parenteral feeds (Ginsberg & Klonoff, 1985, 1987; Handen, Mandell, & Russo, 1986; Linscheid, 1985; Page & Iwata, 1985). Generally, these techniques are employed with children ages 3 to 5 years despite the fact that a conditioned aversion might be strongly developed by that time. It

would seem worthwhile to direct future efforts toward the treatment of infants and children during the first 2 years of life to evaluate the potential of such early intervention procedures in preventing more chronic feeding problems requiring psychological or medical intervention. For children who will not sample food, alternatives to forced feeding techniques must be developed. Similarly, behavioral treatment of neuromotor and physiological dysfunctions adversely affecting eating (such as poor tongue control and organic vomiting) have been insufficiently explored despite data cited earlier that such dysfunctions are frequently implicated in feeding difficulties.

The literature on feeding problems in developmentally delayed populations generally emphasizes presenting problems that are nonorganic in nature, and ongoing medical consultation and collaboration are infrequently described. With respect to nondelayed populations, however, the behavioral medicine literature contains several treatment reports of feeding problems associated with medical conditions such as short gut syndrome, cancer, or recurrent reflux (e.g., Ginsberg & Klonoff, 1985; Harden *et al.*, 1986; Page & Iwata, 1985). Future research is warranted to determine whether special modifications are required to treat organically based feeding difficulties in developmentally delayed children. This should lead to increased collaboration between medical personnel and behavioral scientists specializing in the treatment of developmentally delayed individuals. For example, behavioral psychologists might work closely with gastroenterologists to develop treatment and identification procedures for individuals with gastrointestinal problems that put them at risk for feeding difficulties.

Length of Treatment

One of the most significant findings in a review of the treatment literature for feeding problems is the great variation in length of treatment. In an age of skyrocketing health care costs and stringent policies regarding third-party reimbursement, length of treatment is one of the most crucial economic variables in computing efficacy data. The average number of days in treatment, inpatient and outpatient, is provided in Table 2 for a sample of the studies reviewed here. Table 2 also includes the presenting problem and the dependent and independent variables of each study in an effort to provide data that might be relevant for an analysis of some of the controlling factors determining length of treatment.

Inpatient or outpatient status, in and of itself, does not seem to be a predictor of treatment length. Although institutionalized retarded pop-

Table 2. Dependent and Independent Variables across Feeding Studies

Presenting problem	Treatment	Dependent variables	Length of treatment	Authors
Selective food refusal	Preferred food reinforcement + praise	Weight gains	13 weeks	Hatcher, 1979
Selective food refusal	Withholding food + fading + contingent play	?[a]	4 weeks	Clancy, Entsch, & Rendle-Short, 1969
Selective food refusal	Preferred food reinforcement + praise	Number of bites/sips; % of food expulsion, tongue protrusion and hands in mouth; % of disruptive behaviors; number of grams consumed; weight gain	60–94 sessions (30 days?)	Riordan, Iwata, Wohl, & Finney, 1980
Ruminative vomiting	Electric shock	Amount of vomitus emitted; number of stomach tensions	3 days	Kohlenberg, 1970
Ruminative vomiting	Electric shock	EMG activity (under chin, upper chest & throat muscles); weight	5 days?[a]	Lang & Melamed, 1969
Ruminative vomiting	Electric shock	Weight gain	14 days	Cunningham & Linscheid, 1976
Ruminative vomiting	Food satiation	Number of vomiting responses	27–48 sessions (approximately 9–12 days)	Jackson, Johnson, Ackron, & Crowley, 1975
Ruminative vomiting	Food satiation	Ruminative responses per minute	171–264 days?[a]	Rast, Johnson, & Drum, 1984

Ruminative vomiting	Food satiation + Listerine	Percentage of ruminations	57 days	Foxx, Snyder, & Schroeder, 1979
Ruminative vomiting	DRO[b] + Tabasco sauce	Volume of vomit; weight gain	11 days	Murray, Keele, & McCarver, 1977
Ruminative vomiting	DRO[b] + lemon juice	Rumination frequency	39 days	O'Neil, White, King, & Carek, 1979
Conditioned food aversion	Contingency management; shaping	Amount of food (ml) consumed; number of bites	2–3 months	Handen, Mandell, & Russo, 1986
Conditioned food aversion	Contingency management; shaping; desensitization	kcal consumed; weight gain; amount of vomitus	30 days	Ginsberg & Klonoff, 1985
Crying; tantrums; food refusal	Food reinforcement + time-out	Frequency of crying; frequency of teaspoons consumed	6 sessions	Thompson & Palmer, 1974
Selective food refusal	Forced feeding + positive reinforcement	Percentage of food accepted	90 days	Ives, Harris, & Wolchik, 1978[c]
Selective food refusal	Shaping; social reinforcement + preferred foods	Number of meals eaten daily + cumulative frequency of new foods consumed	12–32 weeks	Bernal, 1972
Selective food refusal	Preferred food reinforcement; praise + ignoring	Grams of food consumed	13 sessions (3 weeks)	Palmer, Thompson, & Linscheid, 1975[c]

[a] Data not clearly determinable.
[b] Differential reinforcement of other behavior.
[c] Day school setting.

ulations could potentially serve as a captive audience for long-term treatment, the treatment duration in some inpatient studies was quite brief. Perhaps an inpatient setting allows for more intensive treatment than is feasible on an outpatient basis. On the other hand, some outpatient studies targeting selective food refusal reveal shorter treatment durations than similar inpatient studies.

As inpatient-versus-outpatient status does not appear to be a discriminating factor in length of treatment, it might be worthwhile, from an economics perspective, to consider developing variants of inpatient-based treatments for outpatient use. Although this may not be feasible for populations at medical risk, it seems reasonable to attempt to keep inpatient stays as short as possible, perhaps by conducting the baseline evaluation and latter portions of treatment on an outpatient basis. This method has been successfully implemented in one study that consistently reported an average of 4-week inpatient stays for children with iatogenically induced feeding problems secondary to ventilatory support or medical procedures (Ginsberg & Klonoff, 1985). Data are lacking, however, as to whether a developmental disability, in and of itself, complicates or interacts with a feeding problem in such a way as to necessarily inflate length of treatment. In addition, without an adequate classification system of feeding problems, it is difficult to determine to what extent differing lengths of treatment may be related to functionally dissimilar presenting problems.

Maintenance and Generalization

The issues of maintenance and generalization also deserve additional future investigation. Although maintenance of treatment gains has been documented for at least several months after treatment, longer term data, at times, reveal relapse (e.g., Kohlenberg, 1970). To promote maintenance, additional research is needed to explore techniques that more readily bring the behavior under natural control (Stokes & Baer, 1977). For example, in order to decrease reliance on parenteral feeds, it might be helpful to increase food deprivation in order to capitalize on "hunger."

At times, relapse may not be preventable as the controlling variables may be difficult to anticipate. Preliminary data suggest that nonhandicapped children with organically based feeding problems may experience frequent but temporary setbacks or "relapses" as a result of any change in physical status (e.g., contacting the flu or common cold) or change in the environment (e.g., inconsistencies in the handling of the feeding program or introducing a new feeder) (Ginsberg & Klonoff,

1985). Such setbacks generally require brief "booster" sessions. One might suspect that the developmentally disabled child would be equally, if not more, prone to such setbacks, although it is not known whether previously attained levels of food acceptance can be as easily regained. Follow-up studies should also assess collateral effects. For example, there has been some speculation that an increase in appropriate eating may be associated with increased pleasure both with respect to eating and to overall social interactions (Beratis *et al.*, 1981); additional empirical data in this area would be worthwhile.

In addition, only a few studies reported data on the generalization of newly acquired skills and appropriate eating behavior to other settings (e.g., Cunningham & Linscheid, 1976). This is an important area for investigation particularly given the current push toward deinstitutionalization and outpatient treatment programs. Response generalization has also not been adequately addressed; that is, at what point will learning to accept specific textures generalize to other, untrained textured foods?

Methodological Concerns

A wide variety of dependent variables have been utilized in the feeding literature, and future research might compare the utility of these measures. Dependent variables have included percentage of responses (Riordan *et al.*, 1980; Foxx *et al.*, 1979), number or rate of bites (Riordan *et al.*, 1980), grams consumed (Thompson, Palmer, & Linscheid, 1977), and weight gained (Hatcher, 1979). The choice of dependent variable may depend on the nature of the presenting problem. For example, rate of rumination may be an insufficiently revealing measure as episodes are generally difficult to operationalize. Amount or volume is thus easier to compute, although duration measures have not been sufficiently explored. When failure-to-thrive is the presenting problem, increases in weight, reflected on a growth curve, may be a more relevant dependent variable than amount of food consumed. However, amount consumed is clearly a useful variable when the focus is on increasing food acceptance; this can be measured in terms of number of bites accepted or in terms of overall grams consumed. These measures may raise questions of reliability, however, as bites are not easily quantifiable in terms of amount of food, and it could be argued that 3 grams of ice cream, 3 cc of liquid, and 3 grams of meat are not functionally equivalent. Relatively few of the studies reviewed incorporated time as a feature of the measurement procedure. This is disappointing because normal eating generally consists not only of consuming appropriate amounts and varieties

of food but also completing a meal within a reasonable period of time. Thus the time allotted for eating during training sessions should either be kept constant, or measures based on rate should be considered.

Utilizing several concurrent measures may prove most beneficial. Measuring amount or rate of food consumed in addition to changes in weight may provide diagnostic information. For example, increased food consumption without a corresponding increase in weight should prompt further assessment of the patient's medical status, monitoring of potential vomiting, and additional assessment of environmental conditions. A study by Riordan *et al.* (1980) was particularly noteworthy in that several dependent measures were recorded including number of bites per meal, percentage of disruptive behavior, grams consumed, and body weights. This allowed for a more fine-grained analysis of the controlling variables involved in increased food acceptance. Interestingly, the authors found that increases in food consumption did not consistently result in increased weight, and there was a lack of correspondence between bites and grams of food eaten that the authors attributed to variations in menu. As noted earlier, bites may be an unreliable measure, and increased food consumption may not always result in increased weight; the caloric value rather than amount of food should be calculated. In addition, an undiagnosed malabsorption problem may result in similar findings. At any rate, future research is needed to address the extent to which these dependent measures provide valid and reliable indicators of feeding progress.

Developing an adequate experimental design is particularly difficult in research involving the treatment of feeding disorders. As learning may lead to generalization of skills, in terms of consumption of a wider variety of tastes and textures, it is difficult to establish treatment controls. Once acquired via contingency control, food acceptance may not return to baseline levels when the contingency is removed as natural reinforcers may then maintain eating. Thus, reversal (ABA) designs might not adequately reflect experimental control. Alternatively, life-threatening conditions may ethically preclude reimposing baseline conditions once an effective contingency is developed. Similarly, ethical questions could be raised with respect to withholding treatment, especially if the presenting problem involves food refusal, for the sake of establishing a no-treatment control group. One effective alternative to reversal designs might be the use of a multiple baseline design across subjects. As skills may generalize across environmental settings once natural reinforcers take over, some researchers have suggested multiple baseline designs across foods rather than settings (Iwata *et al.*, 1982; Linscheid, 1985).

Feeding problems also pose a unique theoretical issue as, by defi-

nition, failure to accept food implies a disruption in the potency of at least one primary reinforcer. For some children with feeding disorders, other primary reinforcers may also be disrupted (Ginsberg & Klonoff, 1985). Treatment might thus necessitate increased reliance on potentially weaker conditioned reinforcers that may be even more difficult to establish in the absence of primary reinforcers. If treatment is effective, food would theoretically become reestablished as a primary reinforcer, and eating would no longer be under operant control. That is, food would become inherently reinforcing, thus precluding an experimental return to baseline or pretreatment levels. By definition, primary reinforcers are generally not learned, and few, if any, skill acquisition studies for other disorders focus on the acquisition of susceptibility to primary reinforcement. Given this, it is unclear whether a failure to return to pretreatment levels in terms of eating is theoretically different than a similar failiure with a non-food-related skill. In sum, the theoretical implications of feeding disorders should provide "food for thought."

Conclusions

A wide variety of feeding disorders in the developmentally disabled population has been reviewed. In evaluating the literature as a whole, it is clear that some disorders, such as ruminative vomiting, are more prevalent in the developmentally disabled group. Other disorders, such as anorexia nervosa and bulimia, appear to be reported much more frequently in the nondevelopmentally delayed population. Still other eating-related problems, such as selective food refusal, seem to cut across both disabled and nondisabled populations.

Although significant clinical and research activity has been directed at increasing food acceptance in developmentally delayed populations with nonorganic selective food refusal, much less attention has been paid to the treatment of feeding problems that are associated with a medical condition or are produced iatrogenically. In addition, the potential impact of a developmental disability on treatments that have already been empirically validated in nondisabled populations has not been clearly determined. In other words, to what degree do established treatments have to be modified, in terms of content or length, to allow for the disabled's participation? For individuals with delayed language or oral comprehension skills, programs that utilize verbal statements of contingencies would require modification. It is conceivable that language, although not essential, might facilitate progress. In addition, it

might be more difficult to solicit the developmentally disabled individual's cooperation with treatment, thereby potentially increasing the length of treatment. Depending on the nature of the disability, it may be more difficult for a delayed child to communicate information regarding a conditioned food aversion; for example, he or she may be less able to explain that the food avoidance is in part maintained by fear of swallowing and choking. Finally, given both potential impairments in cognition and increased neuromotor dysfunctions associated with some developmental disabilities, it is possible that a developmental disability would increase the probability that, as a last resort, an aversive procedure would need to be employed.

Feeding disorders have been managed independently by a variety of specialists including psychologists, occupational therapists, and physicians. Greater collaborative efforts across health-related professions should prove worthwhile. In particular, treatment of organically based feeding problems should result in increased collaboration between behavioral psychologists and medical specialists. As such, organic feeding disorders provide an appropriate target for increased professional attention from a behavioral medicine perspective. As feeding difficulties appear to be an inherent part of some medical specialties, such as gastroenterology and neonatology, future collaborative efforts should focus on employing a behavioral psychologist regularly as part of the treatment team. Such collaboration would likely lead to earlier identification and perhaps even prevention of feeding disorders.

ACKNOWLEDGMENT. Sincere appreciation is extended to Elizabeth A. Klonoff for helpful editorial comments on an earlier draft.

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CHAPTER 3

The Role of Development and Learning in Feeding Disorders

Thomas R. Linscheid

Ginsberg's review points out the wide range of feeding problems in the developmentally disabled population and attests to the success of behavioral management procedures in addressing these problems. In discussing this area, I will review the normal development of feeding patterns and behaviors, discuss feeding behavior as learned behavior in which both operant and respondent conditioning factors play a role, discuss the specific ramifications of cognitive and motor impairment in the development of feeding problems in the developmentally disabled, and finally, address future directions and the need for interdisciplinary collaboration in the management of these problems.

The Development of Normal Feeding Behaviors

Individuals with mental retardation or developmental disabilities, by definition, exhibit disordered or delayed development and therefore, to understand abnormalities, a working knowledge of normal development is essential. In the normal infant, early intake of food is exclusively liquid. Solid food in the form of cereals is usually introduced after the third month and around the sixth month of life. The infant's diet

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progresses to more textured foods and is composed primarily of table foods by 12 months of age. Illingworth and Lister (1964) postulated a critical period for the introduction of solid foods between 7 and 10 months of age and suggested that failure to introduce solid foods during this critical period results in difficulty accepting these foods later. Self-feeding, in the form of finger feeding, usually occurs in the second half of the first year of life with the ability for complete self-feeding using spoon and fork intact by 15 months of age in the normal infant (Christopherson & Hall, 1978).

Although appetite and food preferences remain rather constant through the first 12 months of life, after this time and for the next several years, toddlers and preschoolers may show rather dramatic variations in appetite and food preference. This time period is often the source of feeding problems in normal children as parents have difficulty managing either the introduction of solid foods or the variable appetite and food preferences that naturally occur.

In the developmentally disabled child, it is very difficult to know, given the complexity of cognitive and motor delays, when it is appropriate to introduce solid foods or when to expect the development of self-feeding skills. Oral motor problems that frequently accompany developmental delays make it difficult to ascertain whether refusal of certain textures is behaviorally or organically based. Motor impairments may also enter into the developmentally disabled child's self-feeding skills.

In the normally developing child, after the acquisition of self-feeding skillls, the child's mealtime behaviors are usually controlled by social interaction variables such as parental or peer approval. The child who allows food to fall from his or her mouth or eats with his or her fingers when the food is more appropriately eaten with a fork may be scolded by a parent or teased by peers, and this may serve to modify these behaviors toward a more socially acceptable mode. Social approval may be a much weaker or totally ineffective reinforcer in severely and profoundly developmentally delayed individuals. More direct and tangible reinforcers may be needed to control mealtime behaviors in this population.

Normally, during the elementary-school years, the appetite generally stabilizes and mealtime behaviors evolve to socially acceptable patterns. In the adolescent years, hormonal changes may result in changes of appetite and fluctuations in weight. In the developmentally delayed population, these developmental patterns may be extended past the chronological ages during which they are usually observed or may coincide very closely to normal physical developmental stages. In the

severely and profoundly handicapped individual, accompanying medical conditions may also interfere through disordered development of appetite regulatory mechanisms and motor skills.

One area that has seemed to receive little attention in the literature on feeding problems in the developmentally disabled is the effect of medication on appetite, growth, and gastrointestinal functioning. Reviews suggest that from 51% to 68% of institutionalized individuals are on psychotropic or antiepileptic medication (Aman & Singh, 1983). Nearly all of the commonly used medications reviewed by Aman and Singh (1983) have either behavioral side effects or direct effects on appetite and on endocrinological or gastrointestinal functioning. Further research is needed to better understand the interaction between feeding problems and current or past medications in the developmentally disabled population.

Learning Mechanisms in the Etiology and Maintenance of Feeding Disorders

The success of behavioral or learning-based treatments for feeding problems in both normal and developmentally disabled populations suggests the rather prominent influence of conditioning principles in the development of the eating problems. Most readers will have no difficulty in seeing the feeding situation in a classic operant conditioning format. Assuming a food-deprived organism, the visual and aromatic presence of food can be seen as the antecedent condition, the behavior involved in consuming the food (i.e., reaching for the food, placing it in the mouth, and swallowing) can be seen as the targeted behavior and the reinforcement derives from several possible factors such as oral stimulation, taste, and appetite reduction. In this model, presence of food as an antecedent condition becomes a stimulus for feeding behaviors. Indeed, this discriminative and signaling property of food forms the basis of many behavioral approaches to weight reduction (Stuart & Davis, 1972).

In the feeding situation, the most obvious reinforcer that maintains the operant behavior of eating *per se* is the food itself. Indeed, many successful reports of feeding treatments have described procedures in which acceptance of new foods or textures has been reinforced with access to preferred foods (cf. Palmer, Thompson, & Linscheid, 1975; Bernal, 1972). Interestingly, it is frequently our experience that following the introduction of a new food by reinforcement with a preferred food,

the new food can become reinforcing and may be used as the primary reinforcer later in treatment (Thompson, Palmer, & Linscheid, 1975).

Several factors in the developmentally disabled population may make difficult the use of access to food itself as a reinforcer. First, many state institutions currently operate under regulations designed to protect the rights of the retarded, and this is extremely necessary and appropriate. However, many times these guidelines require that the client be fed at predetermined intervals. In the client who must be fed and is refusing new foods or textured foods, it may not be possible to establish sufficient hunger to motivate the client to either accept or to try the new foods. In other words, in the absence of hunger, new foods may have less reinforcing value. Second, for individuals more severely and profoundly handicapped, it is unclear what role their accompanying medical condition and current medications may have in present feeding problems and appetite regulation.

Perhaps one area that has been overlooked in the analysis of feeding disorders has been the role of respondent conditioning. The presence of food, if followed by forced feeding or other unpleasant feeding practices, may come to illicit fear and/or avoidant responses in a two-factor learning model. An anecdotal but rather dramatic example of this involves my experience with a 44-year-old moderately retarded woman who was referred with a recent history of dramatic weight loss subsequent to total food refusal. The woman had a long history of obesity related to overconsumption of high calorie and high fat content foods. Several months prior to referral, she began to refuse foods, stating that she was not hungry. Referral for inpatient psychiatric evaluation had resulted in recommendations that the woman be transferred from her then-present living arrangement with her elderly parents to live in a group home with peers. The psychiatric evaluation concluded that the patient was depressed and needed greater peer interaction. Upon medical workup after referral to us, it was discovered that the woman was suffering from gallstones, and successful surgery was performed. In retrospect, the most parsimonious explanation for the food refusal was that the ingestion of her normal, high fat content food resulted in intense stomach pains within a very short period. Through a respondent conditioning process, anxiety or fear of these pains was conditioned to the visual presence of food or prompts to actually consume food. After surgery, despite explanations that the food would no longer cause discomfort or pain, the elicited anxiety remained, and it was necessary to conduct a highly structured desensitization and reinforcement-based treatment program.

This author and colleagues (Linscheid, Tarnowski, Rasnake, &

Brams, 1987; Palmer, Thompson, & Linscheid, 1975) have described the presence of a "food phobia" in children who have missed the critical period for the introduction of solid food. Anxiety and fear of the eating situation itself, the presence of food in the mouth, and tentative procedures used by feeders can result in rather dramatic anxiety-based responses, and this needs to be taken into account for successful treatment planning. Deconditioning of a food fear or phobia may require a slow and gradual approach. Interestingly, past reports of feeding treatments for oral intake refusal (Blackman & Nelson, 1985; Iwata, Roidan, Wohl, & Finney, 1982) have described forced feeding or modified forced feeding procedures. Conceptually, these could be thought of as "implosive" procedures similar to those frequently used in the treatment of phobias. If these techniques are to be used, they must be done consistently until the "phobia" is overcome. This is often difficult in the institutionalized developmentally disabled population as staff turnover, shift changes, and the like can result in much inconsistency. There is also a slight chance that food may be aspirated if the patient is emotionally distressed and physically resisting.

New Directions

Ginsberg correctly identifies an emerging area of feeding disorders related to the increased survival rate of medically compromised or premature infants. Frequently, these infants must be maintained via artificial feedings or respirators (Handen, Mandell, & Russo, 1986). The need for these measures precludes the accomplishment of normal feeding practices and behaviors. This is an area in which prevention and treatment techniques need to be developed. In my experience, professionals from disciplines that can impact on the problems are not consulted until the problems have already developed. Early intervention is the key to preventing severe problems, and a mechanism needs to be found in the acute care setting to include a wider range of disciplines earlier. The reason this involvement does not occur is probably due to the importance of medical survival as the primary concern. Medical personnel who are responsible for maintaining the infant's life (surgeons, neonatologists) are understandably concerned with immediately necessary measures (e.g., adequate nutrition versus age-appropriate methods of intake). When the medical crisis passes, the care of the infant or young child is turned over to other professional who may have more concern for the appropriate development of the child as well as his or

her medical condition. To convince the medical personnel who are in charge during the acute care phase, two things must happen. First, follow-up data suggesting the extent and nature of feeding problems (as well as other developmental problems) must be obtained and made available. Second, easily implemented and effective procedures that do not interfere with medical care need to be developed and tested.

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CHAPTER 4

Behavioral Medicine and Neurological Disorders

Jerry A. Martin

Behavioral medicine as a field rapidly developed during the 1970s and 1980s. It is not only difficult to identify its specific beginnings (Russo & Varni, 1982), but there is also considerable disagreement as to its exact definition. Although some (Pomerleau, 1979) have proposed a definition based on the extension of experimental analysis of behavior to health-related behaviors, others (Schwartz & Weiss, 1978) have focused on the interdisciplinary integration of behavioral and medical sciences with provisions for theoretical, basic, and applied research. As of this date, there is no consensus definition of behavioral medicine; perhaps this is as it should be in a developing field where, in the end, the field will be defined by its accomplishments. It should be noted, however, that a broad-based ecobehavior-analysis approach (Rogers-Warren & Warren, 1977) to health problems may not be all that different from the definitions of those who eschew such behaviorally based definitions. Although it is widely claimed that its integrative approach of many disciplines/professions is one thing that makes behavioral medicine unique, is such an approach all that different from what one *should* find in other areas such as mental health, special education, or community psychology?

Within the field of behavioral medicine, one major area of focus has been that of neurological disorders. Unfortunately, there has not been

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Table 1. Neurological Disorders

Central nervous system defects or damage
Developmental disabilities
Cerebral palsy
Epilepsy
Neural tube defects
Traumata or accidents
Cerebral vascular accidents
Spinal cord injury
Closed head injuries
Genetic diseases
Dystonia musculorum deformans
Osteogenesis imperfecta
Friedreich's ataxia
Muscular dystrophies
Myasthenia gravis
Neuromuscular diseases
Guillain-Barre'
Poliomyelitis
Multiple sclerosis

Note: Adapted from Martin, 1983.

agreement as to which medical disorders fall within this general category (Melin, Sjoden, & James, 1983). In the broadest sense, it might include everything from spinal cord injury to epilepsy to cerebral palsy. Such medical problems as chronic pain and insomnia may often stem from a neurological cause. A listing of some common types of neurological disorders as outlined by Martin (1983) can be found in Table 1. Although this listing is not all inclusive, it does give one an impression of the breadth of medical problems that are included. All have the common feature that the brain or its extension, the spinal cord, are, for some cause, functioning abnormally. A discussion of the complexities of these disorders is beyond the scope of this review. For a broader look at neurological disorders in the nondevelopmentally disabled population, excellent reviews can be found elsewhere (Baum & Singer, 1982; Ferguson & Taylor, 1980; Gentry, 1984; Ince, 1976; Karoly, Steffen, & O'Grady, 1982; Krasnegor, Arasteh, & Cataldo, 1986; Lutzker, Martin, & Rice, 1981; Pinkerton, Hughes, & Wenrich, 1982; Pomerleau & Brady, 1979; Russo & Varni, 1982; Tuma, 1982; Varni, 1983; Williams & Gentry, 1977).

For the purposes of examining behavioral medicine approaches to the neurological disorders of the developmentally disabled and behavioral medicine, it is appropriate to focus on the problems of spina bifida and cerebral palsy. Both can be considered developmental disabilities

as well as neurological disorders (Martin, 1983). They are exemplars of a behavioral medicine approach to neurological disorders in the developmentally disabled because the vast majority of the published applications of behavioral medicine to the developmentally disabled has been with these two groups.

Cerebral Palsy

Cerebral palsy is a nonprogressive disorder caused by damage to the brain before, at, or after birth (to the age of 22). It is nonprogressive in the sense that the damage occurs over a brief period of time rather than being prolonged. Defects in motor activity include incoordination, paralysis, and weakness are characteristic. The causes of cerebral palsy include prenatal events such as exposure to toxins and infections, anoxia at birth, and a variety of postnatal events such as head trauma, brain tumors, and meningitis (Martin, 1976, 1983). Because not all cerebral palsy is congenital, it is extremely difficult to pinpoint the incidence of this neurological disorder. Estimates have ranged from 1 to 3 per 1,000 children (Smith & Neisworth, 1975).

Movement disorders of cerebral palsy may be classified in a number of ways. The most typical is with regard to movement and to limb involvement as indicated below (adapted from Martin, 1983):

1. Movement
   Spasticity—involuntary contracture of muscles when they are stretched
   Athetosis—involuntary jerky movements
   Ataxia—awkward movements in response to a lack of balance
   mixed—two or more of the preceding in combination
2. Limb involved
   Monoplegia—one
   Diplegia—two
   Triplegia—three
   Quadriplegia—four
   Hemiplegia—both on one side
   Paraplegia—lower

The most common of these types are spasticity and hemiplegia. Movement of the extremities is often extremely difficult and inaccurate due to increased muscle tone with spasticity. The limb paralysis in hemiplegia occurs on the site of the body opposite the side of the brain injury (Martin, 1983).

Because cerebral palsy is caused by injury to the brain, a number of other problems are frequently associated with this disorder. These include an increased incidence of hearing and visual impairments, seizure disorders, and cognitive impairments. Some degree of mental retardation has been estimated to be present in up to 70% of those with cerebral palsy (Hohlman, 1953). One should be very cautious in interpreting such figures because the validity of most intelligence tests must be questioned when applied to those with moderate or severe movement disorders.

Medical approaches to the problems associated with cerebral palsy are numerous and may require the input of a team of health care professionals including neurologists, orthopedists, physiatrists, physical therapists, occupational therapists, speech pathologists, nurses, social workers, and psychologists. Although this is readily recognized, it occurs all too rarely, most often in major medical centers. Early and ongoing treatment are absolutely necessary in order to prevent the development of severe postural difficulties. Results of a lack of treatment through the years can be found in many state institutions for the mentally retarded. In such places, needed treatment for individuals with cerebral palsy was often absent due to insufficient resources and lack of treatment knowledge. The twisted, contorted bodies that resulted led to numerous health problems that may have contributed to premature deaths. Today's aggressive medical treatment of cerebral palsy may include bracing, splinting, surgery, medications as well as frequent rehabilitative (physical, occupational, speech) therapy. Although the problems of cerebral palsy are lifelong, with proper treatment their devastating effects can be often ameliorated.

Hill (1985) has provided a behavior analysis of cerebral palsy. Two sets of problems exist. First, the opportunity to practice movements is restricted because of motoric difficulties. Decreased mobility will limit access to stimuli, and objects that can cue motor movements and will limit the accuracy, strength, and timing of responses. Because movements are infrequently exhibited, they will be infrequently reinforced. Simple motor behaviors that are prerequisites to more complex motor tasks may never have the opportunity to be developed. Additionally, some motor behaviors may have a punishing effect on individuals with cerebral palsy. Such movements can elicit painful spasms, elicit abnormal reflexes, or a combination of both. These movements are painful because muscles have atrophied and tendons tightened because they have not been used. Obviously, if certain movements are painful, they will be avoided. This first set of problems obviously can lead to a limited motor repertoire. Hill (1985) also described a second set of problems that

can lead to deviant patterns of motor behavior. First, deviant patterns of motor behavior may result from a lack of control. Drooling and dyskinesias are examples of this. Second, deviant or unusual patterns of movement may become commonplace when abnormal motor behaviors are reinforced rather than more normal patterns of movement. The abnormal gait patterns of cerebral-palsied individuals who are able to walk are perfect examples of this. Despite inefficiency, these patterns are maintained because they serve to get an individual from one place to another.

Such a behavior analysis leads one to an understanding of the complex motor problems of cerebral palsy and should aid researchers and practitioners in rearranging antecedents and consequences of motor behaviors so that normal motor patterns are reinforced, aversive consequences for normal motor behaviors are avoided, and abnormal motor patterns go unreinforced.

Rather than describe the large number of research studies that have examined behavioral medicine approaches to cerebral palsy, a listing of representative studies can be found in Table 2. Although an attempt was made to include most published studies, some may have been inadvertently overlooked. However, those included will certainly give one a flavor of the scope of the research to this point in time. Frequently targeted behaviors include head control, fine and gross motor control, and locomotion. Positive reinforcement, including biofeedback techniques, has been used to almost the exclusion of aversive control. More specific issues are discussed in the conclusion (p. 56).

Spina Bifida

Spina bifida is a neural tube defect that occurs during fetal development when the spinal column fails to close. This defect may occur anywhere between the head and the lower end of the spinal column (Martin, 1983). When there is an opening in the spinal column, nerve fibers can protrude, and damage can result in a lack of motor function and/or paralysis below the defect (Hallahan & Kauffman, 1982). Reports of the incidence of neural tube defects have varied from 0.1 to 4.13 live births in every 1,000 (Bleck, 1982). In spina bifida occulta, no physical handicap or neurological disorder exists because the spinal cord does not protrude. Sometimes a small clump of hair is found growing from the skin in the area of the spinal defect. On the other hand, with meningocele, a protruding sac similar to a tumor is formed along the spinal

Table 2. Behavioral Medicine and Cerebral Palsy

Reference	Target behavior(s)	Technique
Asoto, Twiggs, & Ellison (1981)	Wrist & finger extension	EMG biofeedback
Ball & McCrady (1975)	Finger movements	Positive reinforcement
Ball, McCrady, & Hart (1975)	Head control	Positive reinforcement
Barton, Leigh, & Myrvang (1978)	Drooling	Positive reinforcement, punishment
Bragg, Houser, & Schumaker (1975)	Sitting	Positive reinforcement
Brudny, Korein, Levidow, Grynbaum, Lieberman, & Friedman (1974)	Prehension	EMG biofeedback
Cataldo, Bird, & Cunningham (1978)	Muscle control	EMG biofeedback
Conrad & Bleck (1980)	Gait	EMG biofeedback
Finley, Niman, Standley, & Ender (1976)	Fine & gross motor skills	EMG biofeedback
Finley, Niman, Standley, & Wansley (1977)	Fine & gross motor skills	EMG biofeedback
Fuller (1949)	Arm movement	Positive reinforcement
Grove & Dalke (1976)	[illegible]elchair propulsion	Positive reinforcement
Grove, Dalke, Fredericks & Crowley (1975)	Hea[illegible] control	Positive reinforcement
Halpern, Kottke, Burrill, Fiterman, Popp, & Palmer (1970)	Head control	Negative reinforcement
Hardiman, Goetz, Reuter, & LeBlanc (1975)	Gross motor skills	Positive reinforcement
Harris, Spelman, & Hymer (1974)	Head control & limb position	Positive reinforcement via sensory feedback
Inman (1979)	Spasticity	EMG biofeedback
Jacobson, Bernal, & Lopez (1973)	Pointing	Positive reinforcement
Jones, Favell, Lattimore, & Risley (1984)	Fine & gross motor skills	Environmental engineering
LaGreca & Ottinger (1979)	Spasticity	EMG biofeedback
Lee, Mahler, & Westling (1985)	Asymmetrical tonic neck reflex	Environmental engineering
Ortega (1978)	Spasticity	Relaxation
Parker, Cataldo, Bourland, Emurian, Corbin, & Page (1984)	Orofacial dysfunction	Positive reinforcement
Rapp (1980)	Drooling	Positive reinforcement, stimulus control
Rapp & Bowers (1979)	Drooling	Positive reinforcement, stimulus control
Reid & Hurlburt (1977)	Pointing	Positive reinforcement

Table 2. (Cont.)

Reference	Target behavior(s)	Technique
Rice, McDaniel, & Denney (1968)	Arm movement	Positive reinforcement
Rice, McDaniel, Stallings, & Gatz (1967)	Arm movement	Positive reinforcement
Rugel, Mattingly, Eichinger, & May (1971)	Weight bearing	Positive reinforcement
Sachs & Mayhall (1971)	Spasm control	Punishment
Sachs, Martin, & Fitch (1972)	Finger exercises	Positive reinforcement
Seeger, Caudrey, & Scholes (1981)	Gait pattern	EMG biofeedback
Spearing & Poppen (1974)	Foot dragging	Electronic feedback
Skrotzky, Gallenstein, & Osternig (1978)	Range of motion	EMG biofeedback
Thompson, Iwata, & Poynter (1979)	Tongue thrust	Positive reinforcement
Wolfe (1980)	Head control	Positive reinforcement
Wolpert & Woolridge (1975)	Head & shoulder control	EMG biofeedback

cord. Because there is only cerebral spinal fluid within this sac rather than nerve tissue, neurological handicaps will not occur. The final type of neural tube defect—myelomeningocele—is what is commonly referred to as spina bifida. Here, neurological damage does take place because the sac does contain part of the spinal cord with nerve fibers (Hallahan & Kauffman, 1982). Improved medical and surgical techniques have led to a dramatic increase in survival rates (Killam, Jeffries, & Varni, 1985). The closure of the defect in the spinal cord is usually done through surgery during infancy. The level of the defect will determine the exact muscles that are likely to be effected. In addition to paralysis or muscle weakness, children with spina bifida can have a loss of skin sensation (touch, pain, temperature), loss of bowel and bladder control, and hydrocephalus (Bleck, 1982).

It should be obvious that spina bifida is a devastating neurological disorder that, like cerebral palsy, demands the attention of a large number of health care professionals if proper management is to take place. Obviously, the input of additional physicians such as neurosurgeons and urologists is necessary. Hydrocephalus, if present, must be treated surgically with the insertion of a shunt as soon as feasible, or there is a risk of severe brain damage or death (Mandell & Ficus, 1981). Minimally, with any hydrocephalus, there are likely to be cognitive sequela.

Although the behavioral medicine literature on spina bifida is not

Table 3. Behavioral Medicine and Spina Bifida

Reference	Target behavior(s)	Technique
Dowrick & Dove (1980)	Swimming performance	Self-modeling
Feldman, Manella, & Varni (1983)	Self-care	Parent training
Feldman, Manella, Apodaca, & Varni (1982)	Self-care	Parent training
Horner (1971)	Crutch walking	Positive reinforcement
Jeffries, Killam, & Varni (1982)	Fecal incontinence	Positive reinforcement
Killam, Jeffries, & Varni (1985)	Urinary incontinence	Urodynamic biofeedback
Manella & Varni (1981)	Gait training	Positive reinforcement
Manella & Varni (1984)	Ambulation	Positive reinforcement
Rapport & Bailey (1985)	Fine & gross motor skills	Positive reinforcement
Wald (1981)	Fecal incontinence	Rectosphincteric biofeedback
Whitehead, Parker, Masek, Cataldo, & Freeman (1981)	Fecal incontinence	Rectosphincteric biofeedback

as extensive as that on cerebral palsy, it is growing rapidly. The complexity of the disorder offers a considerable challenge to the behavioral medicine practitioner. Table 3 outlines a list of representative research on the application of behavioral medicine to spina bifida. An attempt was made to be as comprehensive as possible, but some published research may have been overlooked. Again, the flavor of the behavioral medicine research can certainly be found through familiarity with this list. Targeted behaviors include locomotion, incontinence, and self-care skills. Positive reinforcement, including biofeedback techniques, has been predominant. The training of parents as therapists (Feldman, Manella, Apodaca, & Varni, 1982; Feldman, Manella, & Varni, 1983) is noteworthy as is the interdisciplinary approach. More specific issues are discussed below.

Conclusions

Perhaps the first application of behavioral medicine to the developmentally disabled was to a motor or neurological disorder. Fuller (1949) successfully used warm milk to reinforce arm movements in a "vegetative human organism" who likely suffered from cerebral palsy. Since that time, numerous investigators, as indicated in Tables 2 and 3, have used a variety of behavioral medicine techniques to alter the motor

functioning of the developmentally disabled. Although the focus in this chapter has been on two specific neuromuscular disorders in the developmentally disabled, the conclusions one can draw from the treatment research cited in Tables 2 and 3 are consistent with conclusions one could draw from the larger class of neurological disorders.

As previously noted, the literature has lacked well-controlled outcome studies (Martin, 1983). At this point in time, the problem still exists when one carefully examines the studies in Tables 2 and 3. One should not be led to believe that the situation is any different for other types of chronic disorders. As Hovanitz, Gerwell, and Russo (1984) have pointed out, isolated case studies on chronic illness appear, but rarely are they followed by a more formal investigation of several children. They suggest that this may be due to the clinical rather than research orientation usually found within medical settings. Although radical behaviorists may eschew classification and labeling, it may be necessary for behavioral medicine researchers to embrace a taxonomy for communication purposes and if a true impact is to be made upon the health care professions. A threefold categorical system consisting of medical diagnosis, behavioral topography, and developmental level has been suggested (Hovanitz, Gerwell, & Russo, 1984). Such an approach would certainly facilitate behavioral medicine research with the developmentally disabled. Although the publication of case studies is expedient, and it is extremely difficult to find homogeneous subjects and presenting problems, the time has come for research to shift to large-scale outcome studies. There is a long overdue need for between-group research designs for the purposes of what has been referred to as technique testing (as opposed to technique building) and the asking of actuarial questions (Hersen & Barlow, 1976).

Although the lack of controlled outcome studies may limit the widespread clinical use of behavioral medicine approaches to the neurological problems of the developmentally disabled, other factors may also contribute. Again, these are not factors unique to either the clinical problem or the population. For example, Hobbs, Beck, and Wansley (1984) have pointed out that relatively few behavioral pediatric studies have involved the collaborative efforts of behavior therapists and physicians. Likewise, Cataldo (1986) has noted that only rarely have studies of child behavior and developmental problems been conducted in the context of a pediatric practice. If one examines the literature found in Tables 2 and 3, in most instances one will not find collaborative efforts with other health care professionals, and seldom will the research have been conducted within a pediatric practice. Only a few of the papers have been published in journals that might be expected to be familiar to the medical

profession. It is time for behavioral medicine researchers to start practicing what they preach and to involve a wide variety of health care professionals in active research. The first to be involved should be physicians. Many benefits can be gained from such physician involvement, including a more thorough behavioral analysis, a more complete understanding of biobehaviors, more widespread acceptance of outcomes, and an opportunity to demonstrate the most powerful tool behavioral medicine practitioners have—precise measurement. Such collaboration should most certainly recognize that there may be limits to any biobehavioral intervention. Although behavioral medicine can have influence on the symptoms of many medically determined problems (such as spasticity in cerebral palsy or incontinence in spina bifida), one must always acknowledge that there may be points beyond which our interventions will not succeed. We should not promise too much too soon or generalize beyond the limits of our data.

A third area of concern about the research on the neurological problems of the developmentally disabled has to do with a lack of generality and long-term follow-ups. If we are ever to convince our health care colleagues of the utility of behavioral medicine, we must begin to deal with this issue. Reputable scientific journals should stop publishing behavioral medicine research that has not addressed the problems of generality and that fail to include long-term follow-up data. Obviously, both generality and follow-up issues plague much of behavioral medicine as well as behavior therapy and applied behavior analysis (Lutzker & Martin, 1981). As Baer (1986) has pointed out, targeted behaviors may be changed in an amount to be socially and personally useful; however, this change does not always generalize to other examples of the same class of behaviors or to other settings or maintain itself after intervention is terminated.

The clinical significance of behavioral medicine research will continue to be limited until the previously mentioned problems have been addressed. Case studies must be replaced by large outcome studies. Research must become collaborative, and it must have application to the environments in which physicians and other health care professionals work on a daily basis. Although a refocus of our research efforts is one way to impact upon clinical practice, another is to redouble our efforts to educate the health care profession in the "ways" of behavioral medicine. A major effort to influence the future behavior of health-care-professionals-in-training may afford more potential impact upon the future health care delivery system than dozens of published research projects (Lutzker *et al.*, 1981).

Although these issues must be dealt with before one can expect to

see a widespread use of behavioral medicine techniques applied to the neurological problems of the developmentally disabled, there is much reason for optimism. Specific training in behavioral medicine is becoming much more common in medical schools and in schools of allied health professionals. In institutional and community settings, a true interdisciplinary approach to the problems of the developmentally disabled is becoming the norm. In each instance, there is an opportunity to affect the practice of a wide variety of profesionals and to engage them in collaborative research and treatment. It is hoped that this will not be a one-way street, for there is much for us to learn from individuals from other professions. It is an humbling experience to learn that your advocacy for the use of reinforcers laden with sugar may be undermining a drool-control program instituted by a speech pathologist. Likewise, your attempts to use positive reinforcement during physical therapy sessions may produce heightened motor arousal, when the physical therapist has been diligently striving for inhibition of motor activity. Further, have we really considered the probability that certain of our procedures (such as an overcorrection procedure like forced arm exercises) may be extremely aversive to a person with spastic cerebral palsy? Until we ourselves get a better base of training in health care, we can only get along with the help of our friends (from other professions).

Obviously, behavioral medicine is not going to solve the many neurological problems of the developmentally disabled. Nevertheless, it is an approach that holds much promise. Along with other recent developments, such as bioengineering (Martin, 1983) and socioecological programming (Jones, Favell and Risley, 1984; Jones, Favell, Lattimore, & Risley, 1984), the future of the developmentally disabled with motor disorders should be much better than the past.

ACKNOWLEDGMENT. The assistance and comments of Deborah C. Parsons are gratefully acknowledged. Requests for reprints should be sent to Jerry A. Martin, Developmental Services Program, Florida Department of Health and Rehabilitative Services, P.O. Box 8420, Pensacola, FL 32505.

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CHAPTER 5

Neurobehavioral Analysis of Epilepsy in Developmentally Disabled Individuals

John C. Neill and Norberto Alvarez

Behavior analysis requires a deterministic viewpoint. By refining the conditions under which an individual is studied, the noncapricious functional relations between the environment and behavior can be obtained. For example, Pavlov (1927) and Skinner (1938) refined their experiments to the point where a single manipulation of an independent variable was sufficient to reveal a behavioral function. The occurrence of epileptiform neurophysiological activity can be analyzed from the same deterministic viewpoint. The functional relations of overt behavior, neurophysiological behavior, and the environment can be empirically determined by concurrent EEG and behavior monitoring under controlled environmental conditions. This approach is called *neurobehavioral analysis* because it represents the specialized practice of behavior analysis in the neurophysiology laboratory. This chapter will review three areas of the neurobehavioral analysis of epilepsy in developmentally disabled individuals: the diagnosis of pseudoepilepsy, the effects of the environment on epilepsy, and the effects of epileptiform discharge on stimulus control of behavior.

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Differential Diagnosis of Epilepsy versus Pseudoepilepsy

An epileptic seizure is any paroxysmal change in behavior that recurs intermittently and that is caused by an epileptiform discharge in the central nervous system. The individual who has epileptic seizures receives the diagnosis of epilepsy. Many developmentally disabled individuals display an array of sudden, brief, recurrent behaviors that are not associated with epileptiform discharge in the central nervous system. These behaviors are called pseudoseizures, and the individual who displays pseudoseizures may be said to have pseudoepilepsy. It is very difficult for observers, including trained medical staff, caretaker personnel, family, and teachers, to discriminate such behaviors from bone fide epileptic seizures.

In order to obtain a confirmed diagnosis of epilepsy, the behavior in question must be observed during an EEG examination (Daly, 1979). If epileptiform EEG events are reliably recorded simultaneously with the target behavior, then that behavior is classified as an epileptic seizure and the individual receives a diagnosis of confirmed epilepsy. If there is no epileptiform EEG observed concurrently with the target behavior, then the behavior is classified as a pseudoseizure, and the individual receives a diagnosis of pseudoepilepsy. If the individual displays both epileptic plus pseudoepileptic behaviors, then each behavior is classified accordingly, and the individual receives a diagnosis of epilepsy plus pseudoepilepsy. If the target behaviors are not observed, either because of lack of cooperation or because the behavior simply did not occur, then the evaluation is inconclusive, and the procedure may be repeated. In order to assure that the behavior in question does occur in the laboratory, the antecedents of the behavior may be replicated there (Fariello, Booker, Chun, & Orrison, 1983).

Telemetered EEG and Video Recording (TEEG-VR)

Standard EEG procedures require that the patient stay still because movement produces electrical artifacts that vitiate the EEG recording. Radio telemetry EEG methods allow the patient to move freely. The patient wears a combination preamplifier-transmitter that reduces electrical artifact. This method is combined with video recording in Simultaneous Radio-Telemetered EEG and Video Recording (TEEG--VR) (Porter, Penry, & Wolfe, 1976; Porter, 1980; Stålberg, 1976). The individual's behavior and EEG are simultaneously recorded on videotape, so that the relations between environmental events, behavior, and EEG events

Table 1. Number (%) of Patients Classified into Each Diagnostic Category[a]

Diagnosis	*n*	(%)
Epileptic	20	(16.1)
Epileptic plus pseudoepileptic	11	(8.9)
Pseudoepileptic	50	(40.3)
Inconclusive	43	(34.7)
Total *N*	124	

[a] From "Differential Diagnosis of Epileptic versus Pseudoepileptic Seizures in Developmentally Disabled Persons" by J. C. Neill and N. Alvarez, 1986, *Applied Research in Mental Retardation*, 7, pp. 285–298. Copyright 1986 by Pergamon. Reprinted by permission.

can be determined. This technique has provided a differential diagnosis of each seizurelike behavior in nonmentally retarded children (Holmes, Sackellares, McKiernan, Ragland, & Driefus, 1980); in nonmentally retarded adults (King *et al.* 1982; Luther, McNamara, Carwile, Miller, & Hope, 1982; Sutula, Sackellares, Miller, & Dreifuss, 1981); and in developmentally disabled individuals (Holmes, McKeever, & Russman, 1983; Neill & Alvarez, 1986; Neill, Alvarez, & Harrison, in press).

Results of TEEG-VR in Developmentally Disabled Individuals

Neill and Alvarez (1986) evaluated 124 mentally retarded individuals with TEEG-VR who were referred for evaluation of seizurelike behaviors. Table 1 shows the distribution of diagnosis. Of 81 patients in whom Neill and Alvarez obtained a good recording, 75% (61/81) exhibited at least one behavior that was nonepileptic, although the staff had been recording them as epileptic behaviors.

In addition, the topographies of the most frequent seizurelike behavior were not significantly related to diagnosis. These behaviors were myoclonus, eye blink, head drop, cessation of ongoing activity, and hand–arm automatisms. These results are in agreement with the results obtained by Holmes *et al.* (1983) who evaluated 38 mentally retarded individuals with TEEG-VR and also found that the clinical description of the behavior was not significantly related to the diagnosis obtained with TEEG-VR.

The Effects of the Environment on Epileptic Events

It has been shown repeatedly that individuals with epilepsy generally have less epileptic seizures when they are awake and are engaged

in challenging, interesting tasks than when they are asleep, bored, or inactive (e.g., Gotze, Kupicki, Munter, & Teichman, 1967; Guey, Bureau, Dravet, & Roger, 1969; Jung, 1939; Jus & Jus, 1962; Lennox, Gibbs, & Gibbs, 1936; Li, Jasper, & Henderson, 1952; Sato, Penry, & Dreifuss, 1976; Schwab, 1941; Shimazono, Hirai, Okuma, Fukuda, & Yamamasu, 1953; Tizard & Margerison, 1963a; Vidart & Geier, 1967). Rest and sleep are now used as standard procedure during EEGs to activate epileptiform discharges (Bickford, 1979).

The Effects of the Everyday Environment on Epilepsy in Developmentally Disabled Individuals

Neill and Alvarez (1987) intensively monitored three developmentally disabled individuals who had generalized epilepsy with TEEG-VR while the patients engaged in everyday activities, including peg board tasks, puzzles, talking, and resting. Familiar teachers and therapists gave the patients reinforcers and prompts to increase the frequency of adaptive behavior, without respect to epileptic discharge or seizures, which frequently overlapped with the programmed stimuli. In other words, the therapists and teachers were essentially naive in regard to the epileptic activity. The patients had many subtle generalized seizures, characterized by small jerks in the face or hands. The frequency and duration of epileptiform discharge was suppressed during prompts and reinforcement for on-task behavior. Figure 1 shows the data of one individual.

Teachers and therapists believed that this severely mentally retarded adult should be excluded from challenging activities because she did not appear to progress. However, the results indicated that this individual, and others like her, did benefit from a full schedule of challenging activities in which they were prompted and reinforced for operating on the environment. Such schedules may act as a beneficial environmental adjunct to antiepileptic drug (AED) treatment in patients with refractory forms of epilepsy (Aird, 1983). Of course, the specific approach to each behavior plan should be made on the basis of an empirical neurobehavioral analysis of epilepsy in each individual; some forms of epilepsy may be exacerbated by certain forms of environmental events (Feldman, Ricks, & Orren, 1983; Mostofsky & Balaschak, 1977).

The Question of Contingency Management of Seizurelike Behaviors

A number of investigators have reported that contingency-management procedures are effective in suppressing the frequency of sei-

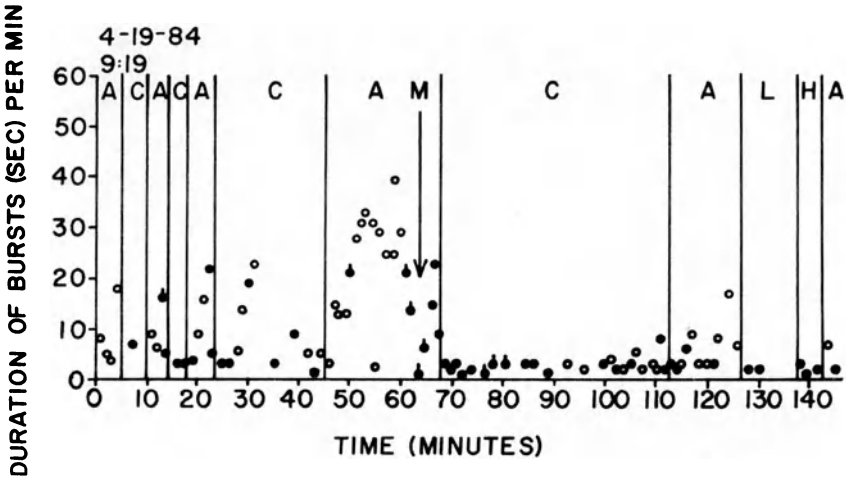

Figure 1. Time distribution of epileptic activity across environmental conditions for one individual. Each circle represents the cumulative duration of epileptiform EEG bursts (sec) for each 1-min interval. Epileptiform EEG bursts of less than 1 second are not plotted. Symbols: open circles = seizures were observed during at least one of the discharges during that 1-min interval; closed circles = no seizures were observed during any of the discharges during that 1-min interval; closed circles with lines = epileptiform discharges occurred while the subject's behavior was not clearly visible. Conditions: A = sitting quietly, no programmed activity; C = prevocational activities (pegboard & puzzles); M = white noise (90 dB, SPL); L = physical exercise; H = coffee and conversation with speech therapist. (From Neill & Alvarez, 1987. Reprinted by permission.)

zurelike behaviors (e.g., Burgio, Tice, & Brown, 1985; Iwata & Lorentzson, 1976; Rappaport, Sonis, Fialkov, Matson, Kazdin, 1983). The seizures were generally viewed as genuine epileptic seizures, which were exaggerated due to adventitious reinforcement (Dahl, Melin, Brorson, & Schollin, 1985). In view of the fact that prompts and reinforcement by naive teachers can suppress the rate of generalized seizures, it may be that bona fide epileptic seizures are decreased by nonassociative factors involved in contingency management, such as the increase in physical activity levels (Gotze *et al.*, 1967) or the changes in blood pH levels (Jung, 1939) that accompany active behavior (Aird, 1983). Thus suppression of genuine epileptic seizures by "contingency management" may not be directly due to contingent reinforcement of seizures. The suppression of epileptic seizures may be a result of pseudoconditioning, a change in behavior that is merely an artifact of the reinforcements and prompts (Rachlin, 1976).

Most of the contingency management studies did not record the

EEG concurrently with the seizurelike behaviors, so the true nature of the target behaviors was not conclusively shown to be epileptic. In some studies, the subjects were responsible for recording the occurrence of their own seizures (e.g., Dahl, Melin, & Lund, 1987). In view of the high incidence of pseudoepileptic behaviors and the existence of epilepsy plus pseudoepilepsy in a single individual, some of the seizurelike behaviors that have been successfully treated with contingency management may have been pseudoepilepsy (Holmes, *et al.*, 1983; Neill & Alvarez, 1986).

Cataldo, Russo, and Freeman (1979) did confirm that their subject's seizurelike behavior was epileptic during an EEG recording, which is obviously an important step in view of the pseudoepilepsy problem. This study is also noteworthy because it demonstrated a reduction in confirmed epileptic seizures with a contingent rest procedure. Although this result appears to disagree with the notion that generalized discharges increase during rest, the particular manner in which the contingent rest procedure was carried out may have acted to raise the general activity level of the patient. Research is needed to determine if epileptic seizures are, in fact, directly decreased by contingent reinforcement or by pseudoconditioning. In the meantime, it is important to evaluate the effects of each procedure empirically in each individual and to avoid a cookbook approach to treatment.

Evaluation of the Functional Effects of Subclinical Seizures and AEDs in Developmentally Disabled Individuals

AEDs can cause impairments in behavior (Gay, 1984; Gibbs, Gibbs, Gibbs, Gibbs, Dikman, & Hermann, 1982; Novelly, Schwartz, Mattson, & Cramer, 1986; Poling, 1986; Stores, 1975). In addition, generalized epileptiform discharges can have a decremental effect on an individual's response ability (e.g., Brown, Penry, Porter, & Dreifus, 1974; Courtourtois, Ingvar, & Jasper, 1953; Davidoff & Johnson, 1964; Erba & Cavazzuti, 1977; Goode, Penry, & Dreifus, 1970; Jung, 1939; Lombroso & Erba, 1982; Mirsky & Van Buren, 1965; Porter, Penry, & Dreifus, 1973; Schwab, 1941; Tizard & Margerison, 1963b). These two major effects must be differentiated. By simultaneously monitoring AED blood levels, epileptiform EEG discharges, and the ability of the individual to respond to significant events in the environment, one can determine whether AEDs or discharges are associated with impairment of behavioral function. We now turn to two procedures that have recently been useful for

demonstrating the impairment of function by subclinical seizures. In a sense, these procedures make the functional effects of subclinical seizures (and of AEDs) "clinical" by providing measures of behavior that are sufficiently sensitive to reveal the moment-to-moment changes in discrimination performance.

A Visual Discrimination Procedure

We have developed a procedure called the Operant Continuous Performance Task (OCPT) that measured the impact of AEDs and epileptiform discharge on visual stimulus control (Neill & Alvarez, 1983; Neill *et al.*, in press). Each patient was trained to press a transparent panel when a discriminative stimulus was briefly projected on it and not to press when a repetitive, brief delta stimulus was projected on it. After several sessions of pretraining, the patients were tested during TEEG-VR, and the behavioral-EEG data were used as a baseline for measuring the effects of future AED therapy changes. Figure 2 shows the results of testing in one patient.

Responding was not systematically interrupted by discharge in the first test session. When a number of changes in the patient's AED regime were made, there was a clear increase in latencies, and discharge was associated with either no responding, or late responding, to the S+ stimulus (see Figure 2 caption for details).

Further changes in AED treatment were effective in eliminating discharges altogether during laboratory testing, and response latencies to the discriminative stimulus became shorter, indicating that neither the discharge nor the new AEDs were impairing the patient's behavioral function.

An Auditory Discrimination Procedure

We have also developed an auditory discrimination procedure for the precise evaluation of epilepsy in developmentally disabled patients under controlled conditions (Neill, Alvarez, & Harrison, 1985; Neill, Alvarez, & Harrison, 1986; Neill *et al.*, in press). This procedure incorporates naturalistic features that produce rapid acquisition of auditory discriminations in nonhuman mammals: stimulus novelty, adjacency of response site and sound source, and complex acoustic content (Harrison, 1983a, b, 1984; Neill & Harrison, 1987). Presumably, an inability to acquire an auditory discrimination under conditions that included these naturalistic features would have significant generality to the individual's

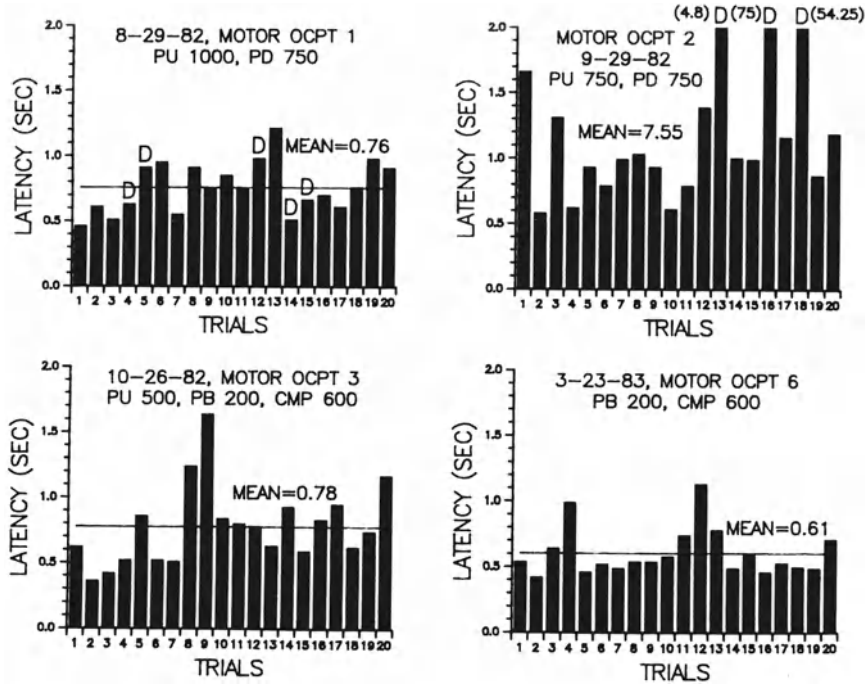

Figure 2. Response latencies of one patient during sessions 1, 2, 3, and 6 of motor OCPT. Symbols: D = discharge within 5 sec before or 5 sec after the S+ onset; PU = phenurone; PD = primidone; PB = phenobarbital; CMP = carbamazepine. Latencies which were greater than 2 sec are shown in parentheses in OCPT 2. (From Neill, Alvarez, & Harrison, in press, *Transitions in Mental Retardation*, Vol. 4. Copyright 1988 by Ablex. Reprinted by permission.)

behavioral adaptation in the everyday environment, where such features of the physical environment are common.

The patients sit before a stimulus response console, which moves to allow patients in wheelchairs access to the console. Two plexiglass panels, each containing a speaker, are mounted in the console. Toys are dropped automatically by a dispenser into a receptacle in the console, and an adjacent light flashes when the toy drops. A house light signals when the session is in progress. Each patient is trained to pick up a toy and place it on the console when it is delivered. At the end of the session, the toys are exchanged for effective primary reinforcers. Responding on both panels is reinforced in an alternating order. When each patient is responding on both panels on a variable interval schedule of reinforce-

ment, discrete trials of sound stimuli are introduced. A "correct" response on the panel adjacent to the sound source turns off the sound, is reinforced, and starts the ITI (intertrial interval). An "incorrect" response on the nonadjacent panel turns off the sound and starts the ITI.

The following example illustrates how the auditory stimulus control procedure was used to determine the effects of generalized epileptiform discharge on discrimination behavior in a severely mentally retarded adult woman who was being treated with ethosuximide, 1,250 mg per day, and phenobarbital, 150 mg per day. The discharges caused subtle clinical manifestations. The patient had been referred for evaluation of subclinical seizures.

Stimulus control of responding by the location of sound source developed slowly; by the twelfth session, the patient had acquired the discrimination above 80% correct. In session 12, trials were delivered in the presence and absence of discharge, in order to determine the effects of discharge on stimulus control.

The epileptiform discharges had a completely disruptive effect on auditory behavior. A trial-by-trial analysis of session 12 revealed the following critical facts: The subject had discharge on 22 trials, and her responding during these trials was 63.6% correct (14/22), which was within the range of baseline (no sound) responding. In other words, discrimination behavior occurred within chance levels if a discharge had occurred. Responding during the same session, when discharges did not occur during sound trials, was 100% correct (18/18). Thus, discharges were producing an impairment in stimulus control of auditory behavior, a functional deficit that presumably interferes with the patient's acquisition and performance of many adaptive behaviors.

Our experience with these visual and auditory stimulus control procedures suggests that developmentally disabled patients with generalized "subclinical" seizures may benefit from aggressive AED treatment and that stimulus control procedures with concurrent TEEG-VR may be useful for empirically evaluating these treatments. The relations between epileptic seizures and the everyday environments also suggests that a dense schedule of reinforcement may serve as a useful adjunct in the suppression of epileptiform events. The TEEG-VR technique has shown that many seizurelike behaviors of developmentally disabled individuals cannot be accurately diagnosed on the basis of casual observation and medical history. Taken as a whole, the neurobehavioral analysis of epilepsy provides a powerful approach toward precise diagnosis and treatment of epilepsy in developmentally disabled individuals.

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CHAPTER 6

Assessment and Treatment of Neuromuscular Disorders

Lynn H. Parker

Extensive research on the efficacy of behavioral interventions in neuromuscular disorders has shown that behavioral techniques are effective in improving neuromuscular control across a wide variety of disorders. However, it has been the trend in research with neuromuscular disorders to apply the same or similar procedures across diagnoses, age levels, and cognitive levels. Less attention has been paid to the differential effectiveness of treatment as a function of patient characteristics. Treatments have typically been developed for use with adult subjects of normal intelligence with few reported cases of the application of these procedures to the developmentally disabled.

In a recent review of neuromuscular disorders, Cataldo, Parker, Bird, & Emurian, (in press) reported that over 1,000 patients with a variety of neuromuscular disorders have been treated with some form of behavioral intervention since the early 1960s. Less than 200 of these subjects have had some type of developmental disability, typically cerebral palsy or spina bifida. Accompanying mental retardation was described in only 10% to 15% of the developmentally disabled, although the percentages may be underestimations due to under reporting of subjects' cognitive level. The applicability of results of these behavioral studies to the developmentally disabled is further qualified by the fact

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that almost 50% of the 200 developmentally disabled subjects were spina bifida patients who were specifically treated for neurogenic bowel dysfunction. Thus there has not been a large number of developmentally disabled subjects studied, and existing studies have focused on a limited range of target behaviors. As a result, there are significant gaps in our knowledge regarding the usefulness of existing procedures with the developmentally disabled. This chapter addresses adaptations of treatment procedures for the developmentally disabled and mentally retarded, the relative merits of EMG biofeedback and contingency management procedures in these populations, and the need for social validation and health outcome measures.

Procedural Adaptations for Developmentally Disabled and Mentally Retarded

Procedural adaptations that consider a subject's intellectual level are needed to extend the applicability of standard procedures to the developmentally disabled. It is wise for the clinician to consider the principles of programmed instruction offered by B. F. Skinner and associates (1968) when modifying procedures. It is also important to keep in mind that biofeedback, as a technique, is more effective when applied by a therapist who has the understanding of the basic functional relations of behavior and their application to the control of human behavior. Consequence manipulation is the critical variable in the control of operant behavior. However, the importance of stimulus control, reinforcement schedules, shaping, and stimulus fading should not be underestimated (Michael, 1970). Pragmatically, this means breaking the treatment goal into its components and into sufficiently small steps to insure that gradually changing reinforcement contingencies will not result in long periods of trial-and-error strategy on the part of the subject (Skinner, 1968). Shaping involves reinforcing the individual initially for a response that he or she can already perform at a reasonable rate. Many biofeedback software packages do not allow the clinician the flexibility to meet this requirement. Professional assistance in altering programs or creative programming may be necessary.

Choosing a reinforcer for the mentally retarded patient may be a challenge. Again, it is important to recall that a reinforcer is defined by the effect it has on behavior. Many training procedures that have been devised for adults with normal intellectual functioning assume that signs of accomplishment will function as a reinforcer. Additionally, many bio-

feedback procedures in which appropriate muscular or motor responses are followed by visual or auditory stimuli assume these stimuli to act as reinforcers. These stimuli are conditioned reinforcers and cannot be assumed to be effective for all individuals, especially for individuals with cognitive deficits. Use of contingency management procedures alone or in combination with traditional biofeedback procedures are likely to be more effective with the developmentally disabled.

Cataldo *et al.* (in press) discussed the need to test an instructional system before applying it to a particular neuromuscular problem. For example, the effectiveness of a stimulus as a reinforcer can readily be tested by making the stimulus contingent upon the performance of a response that is considered to be clearly under the individual's control. It should not be a response that is impacted by the individual's neuromuscular disorder. This step allows the clinician to know that treatment failure in conditioning a neuromuscular response is not due to the choice of the reward. Inattention to the adequacy of the reinforcement procedure will surely diminish treatment results.

Consideration of what has been called the learning style of the multihandicapped patient is also important. For example, studies report that children with cerebral palsy and spina bifida experience visual-perceptual, attentional and memory problems (Cull & Wyke, 1984; Horn, Lorch, Lorch, & Culatta, 1985; Morozas & May, 1985; Shaffer, Fredrich, Shurtleff, & Wolf, 1985). If biofeedback is part of the training procedure, the use of auditory rather than visual stimuli or the combination of both may be advisable. Also, developmentally disabled individuals may profit from frequent, shorter training trials that are massed rather than spaced.

Finally, attention to controlling stimuli in the training environment should facilitate stimulus generalization. Training should begin with the accompaniment of obvious cues with subsequent, gradual fading, leaving behavior primarily under the control of cues that will be present in the patient's environment.

EMG Biofeedback versus Contingency Management

Behavioral interventions with the developmentally disabled have included operant conditioning techniques, progressive relaxation, and an array of high technology interventions including device feedback (e.g., limb load monitors and head position trainers) and EMG biofeedback. Prior to the advent of biofeedback techniques, early studies primarily evaluated the effectiveness of contingency-management proce-

dures in changing undesirable motor patterns of cerebral-palsied patients. With the introduction of EMG biofeedback procedures with the nondevelopmentally disabled population, researchers began to apply these techniques to the developmentally disabled with the apparent abandonment of former contingency-management approaches. Despite the successful use of both types of procedures, these approaches have rarely been compared, used in combination, or discussed regarding their relative merits. The following discussion highlights the difference as well as complementary features of EMG biofeedback and contingency management.

Electromyographic-biofeedback procedures are designed to decrease or increase muscle activity with the assumption that these changes will enhance functional motor performance. The development of EMG biofeedback equipment was exciting to clinicians and researchers because it allowed muscular responses to be targeted and measured for the first time. Thus shaping could occur with the first responses being unobservable to the naked eye even when the final goal is an observable motor movement. The advantage of biofeedback procedures is the provision of immediate and precise consequences for molecular behaviors. In addition, biofeedback procedures may improve the quality of motor control by directly training muscle groups. For example, many individuals with cerebral palsy will have better upper extremity control if they contract all muscle groups in the arms while attempting to perform motor tasks. Although this may be an effective compensation strategy, it may be executed at great effort, causing the individual to become fatigued. If execution of this same response was possible with greater muscle control or muscle relaxation, the response cost to the individual could be minimized. In such a case, EMG biofeedback may be helpful by teaching inhibition or facilitation of involved muscle groups.

Contingency-management procedures when used without biofeedback equipment reinforce observable motor responses with no effort to directly train muscle control. Contingency-management procedures also have their reported advantages over the use of biofeedback procedures alone. Catanese and Sandford (1984) compared the efficacy of biofeedback versus biofeedback plus contingency management in the treatment of head position in patients with cerebral palsy. Four subjects received auditory feedback for head tilting. Social reinforcement of appropriate head position was subsequently introduced for two of the four patients. The two patients who received a combination of auditory feedback and social reinforcement showed better maintenance of treatment gains in the classroom setting. Authors argue that contingency-management procedures are more portable, can be used more readily across settings,

and thus enhance stimulus generalization. Eventual generalization of treatment gains presents a seemingly difficult problem for all biofeedback procedures. As discussed earlier, it is important to fade controlling stimuli in the training setting. Unfortunately, the presence of biofeedback equipment is an artificial and obvious stimulus that cannot be gradually faded during training. The use of portable trainers that can be worn in other settings may help to remedy this difficulty.

Contingency-management procedures may also prove to be more effective in shaping complex motor responses. Even the simplest upper extremity task requires refined and well-integrated control from multiple muscle sites. Some biofeedback procedures have provided subjects with pairs of EMG signals that they attempt to simultaneously control. However, it is highly unlikely that EMG signals from more than two muscle sights can be provided to subjects or that this information, if provided, can be effectively used by the subject. The relative merits of biofeedback and contingency-management procedures seem to advocate for a sequential training process in which EMG biofeedback is first used to shape simple muscular responses and is then followed by contingency-management procedures to enhance generalization and further shape more complex motor behavior.

Few studies have directly compared biofeedback and contingency-management procedures. The sequential use of biofeedback and contingency management has been reported in two studies (Catanese & Sandford, 1984; Whitehead, *et al.*, 1986). Whitehead *et al.* (1986) compared biofeedback and behavior modification in the treatment of fecal incontinence in children with spina bifida. Biofeedback involved visual feedback plus rewards for external sphincter contraction during training sessions. Behavior modification involved reinforcement for appropriate bowel movements in the toilet and enemas or suppositories following unsuccessful attempts to have a bowel movement for two consecutive days. In the first of two experiments, eight children were provided with biofeedback alone. Biofeedback was found to be insufficient in reducing the frequency of incontinence. In experiment 2, some children received behavior modification alone, whereas others received behavior modification plus biofeedback. Generally, results indicated that patients who received behavior modification alone showed as much improvement as patients who received a combination of treatments. However, the authors noted that there was a subgroup of patients for whom biofeedback did provide additional benefit. The authors conclude that the value of biofeedback in this population may have been overestimated but that certain individuals may require the combination of biofeedback and behavior modification in order to receive maximal benefit. These results

lend support to the notion of differential benefit from biofeedback and contingency management approaches.

Social Validation and Health Outcome Measures

Martin, in Chapter 4, thoroughly enumerates the current problems with the existing literature in behavioral treatment of neuromuscular disorders with the developmentally disabled. Among these are the lack of generalization to functional responses and lack of long-term follow-up data. Related to generalization and equally important is the social validation that treatment changes are significant and give the patient more adaptive skills. Social validation measures have routinely been obtained by professionals who have been involved in the treatment of the patient. As professionals, we could learn a considerable amount by obtaining these measures from other developmentally disabled patients who may have a different perspective on the importance of selected target behaviors and their usefulness in daily living. The choice of a target behavior should be optimally done through a discussion between the patient and therapist, although parties may not always agree. For example, the therapist may feel that approximations to a more normal motor pattern are desirable, whereas the patient prefers the development of a functional motor pattern that is not necessarily asthetically pleasing to the nonhandicapped observer.

Even when social validation is present, the therapist must consider the cost/benefit ratio to the patient. The multihandicapped individual has many needs and is frequently scheduled for various therapies that consume an extraordinary amount of time, often leaving little time for socializing and leisure activities. It is the ethical responsibility of the clinician and/or researcher to ascertain which patients will most profit from intervention and the cost in both hours and money of this intervention. When safe, less expensive, and more effective treatments are available from other disciplines, the patient should be made aware of these alternatives. Unfortunately, the existing data base is not sufficient to answer these questions and provide appropriate information to patients. One reason this knowledge is not currently available is that we rarely ask questions regarding the effects of our interventions on the general quality of life of treated patients.

As behaviorists, we like to select outcome measures whose change can be directly attributed to change in trained behaviors. Thus we either evaluate the trained behavior across nontrained settings or untrained

behaviors for which the trained behavior is considered a prerequisite or component. For example, if a patient is taught to relax a particular group of muscles, we evaluate the ability to relax other muscles, to execute a more complex response that is facilitated by relaxation, or to relax in a new stimulus setting. Rarely do we obtain more global or subjective measures, perhaps because changes in these measures are hard to attribute to treatment success. However, it is these very measures that will allow us to evaluate the impact of our treatment on the quality of life of patients. In addition to standardly obtained outcome measures, health outcome measures such as the cost, length, and duration of treatment, change in medications, decrease in physician visits, change in measures of adaptive living skills, and measures of school or job functioning should be obtained. it is through these measures that we will begin to modify our own behavior in order to develop more effective treatment procedures or to more accurately describe to our patients the limitations of our current interventions.

Conclusions

In summary, there has been much research on behavioral interventions with neuromuscular disorders. We have learned that environmental contingencies can shape and alter the motor behavior of neurologically impaired individuals. Unfortunately, the developmentally disabled have been an understudied population, and little is still known about the application of biofeedback and behavior modification procedures to this population. This commentary provides suggestions for altering procedures by incorporating basic and well-known principles of operant conditioning. Biofeedback and contingency management have both been reported successful in the treatment of neuromuscular disorders. It is time to evaluate the differential merits of these procedures and to consider the relative potency of the combination of approaches versus the use of either alone. Finally, no treatment lasts long without the ultimate demonstration of the clinical significance of treatment changes. Measures of social validation, cost/benefit ratio, and health outcome will be important to physicians and patients alike. These data will be critical in establishing the credibility of behavioral scientists as vital team members in the treatment of neuromuscular disorders.

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CHAPTER 7

Urinary and Fecal Incontinence in the Developmentally Disabled

Karl O. Moe

Urinary and fecal incontinence are rarely uniquely medical problems, and often the expertise of other health care professionals is needed to find solutions. Behavioral medicine specialists are one such group that offer treatment for many kinds of incontinence problems. This chapter reviews behavioral medicine treatment procedures for urinary and fecal incontinence, focusing on their application with the developmentally disabled. Organic causes of incontinence and medical treatments are discussed within this context.

A variety of definitions of incontinence could be offered. However, a simple but entirely usable definition is the involuntary loss of urine or feces at inappropriate times and in inappropriate places. Beyond that, most researchers make several distinctions when discussing incontinence. An incontinence problem is referred to as primary if toilet training has never been successfully completed. Secondary incontinence is defined as incontinence occurring after successful toilet training. Primary problems are often thought to be more strongly associated with underlying physical etiologies than are secondary problems. Doleys (1977),

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however, has questioned the prognostic utility of this distinction. In part, this is because an inadequate training program could lead to primary incontinence that is totally unrelated to the trainee's physical status and ability to learn.

Another distinction often made is between nighttime bed-wetting, or enuresis, and daytime wetting, diurnal enuresis, or simply incontinence. Daytime wetting is also thought to more strongly raise questions about physical etiologies. Noting the type of incontinence (bladder and/or bowel) and the time of day (night and/or day) is helpful in determining a child's developmental level. Development usually allows first for nighttime bowel continence followed, in order, by daytime bowel continence, daytime bladder continence and, finally, nighttime bladder continence. These distinctions are important because they help with accurate evaluation and diagnosis.

Incontinence deserves careful evaluation and treatment. Although most incontinence problems are relatively benign, incontinence may indicate the presence of an underlying physical disorder. Further, it is important to recognize that incontinence may limit a person's opportunities and affect self-esteem. Whitman, Scibak, and Reid (1983) suggest that, of all the self-care problems a severely or profoundly retarded individual (or any individual for that matter) can have, incontinence is probably the most problematic and limiting. Persistent incontinence can effectively exclude many incontinent individuals from educational and recreational programs until they are appropriately toilet-trained. Additionally, staff inferences about the general ability of people who are incontinent may be unduly affected by persistent incontinence.

General Incidence

Incidence estimates for incontinence in the general population vary. Starfield (1978) states that enuresis is the most common chronic condition in general pediatric practice. Pierce (1980) suggests that 92% of 1 year olds wet at night and 12% of the 4½ year olds still wet. He goes on to note that, including those who regress, 16% of the 5 year olds wet, but the percentage drops to 7% by age 7½. Doleys's (1977) summary suggests that bed-wetting continues to drop to about 2% of all 12 to 14 year olds, but that there is no way to predict which children will become spontaneously dry, or when. He also notes that enuresis occurs about twice as often in boys as in girls. The incidence of fecal incontinence is somewhat lower than that of urinary incontinence. Groves

(1982) reports incidence estimates from 1.5% to 3%. The ratio of boys to girls is estimated at between 3.5:1 (Groves, 1982) and 6:1 (Levine, 1982). The presence of some neurological disorders substantially increases the likelihood of fecal incontinence.

At the other end of the age spectrum, the prevalence of urinary incontinence in people over 65 is 5% to 15% if they are living at home, whereas the rate ranges to as high as 50% of those in hospitals and is higher still for elderly patients who permanently reside in institutions. The costs are dramatic. Care required due to incontinence accounts for up to 25% of the nursing time in nursing homes (Berkow, 1982). DeLeon (1986) cites a U.S. Public Health Service estimate that 13 billion dollars could be saved annually if the problem of geriatric incontinence were successfully addressed.

Incidence of Incontinence in Developmentally Disabled Populations

A few statistics will demonstrate the magnitude of the problem among developmentally delayed people. The incidence of enuresis among severely retarded residents of institutions is estimated to be about 70% (Azrin, Sneed, & Foxx, 1973; Sugaya, 1967). Conroy and Derr (1971) estimated in a survey for the then-Department of Health, Education and Welfare that 22.5% of the mentally retarded persons in the United States had a partial (10.2%) or severe (12.3%) handicap due to incontinence. Of the 243 children, 37% enrolled in the Rubella Birth Defect Evaluation Project had difficulties with elimination—bowel, bladder, or both (Chess, Korn & Fernandez, 1971). Lorber (1971) reports that 40% of children with myelomeningocele are fecally incontinent. Clearly, incontinence in this population deserves attention.

Urinary Incontinence

A variety of authors (Berkow, 1982; Ellis, 1963; Giles & Wolf, 1966) have suggested a number of organic and behavioral causes of urinary incontinence that is discussed later. However, at this point it would be helpful to review the physiology and function of the urinary tract, the various organic causes of incontinence, and current medical procedures for treatment.

Review of Urinary System Functioning

The urinary system consists of the kidneys that are drained by ureters into the bladder. The bladder is emptied through the bladder neck

into the urethra that empties to the outside. This complex system eliminates excess water as well as eliminating the products of protein metabolism from the body. Additionally, the kidneys help to regulate the volume of blood in the body, the acid-base balance, and the concentration of electrolytes. Normal adult kidneys filter approximately 50 gallons of plasma each day and produce about a quart of urine in the process. The bladder is essentially a sack made up of three layers of smooth muscle called the detrusor. The normal adult bladder holds 250 to 400 ml (8 to 13 ounces) of urine. The bladder capacity of a child is often estimated as age plus 2 ounces. Hjalmas (1976) has produced a set of regression equations that can estimate a child's bladder capacity based on his or her chronological age, weight, or height.

Two sphincter muscles normally work together to prevent unwanted draining of the bladder. The internal sphincter is an extention of the same smooth muscle group that makes up the detrusor. The internal sphincter is typically contracted and keeps the collected urine from draining out. When the bladder is close to full, the detrusor reflexively contracts. Simultaneously, the internal sphincter relaxes and the bladder empties unless the external sphincter and perineal muscle (abdominal floor) are contracted. These latter two muscles are striated muscles and, in the continent person, are under voluntary control. If these muscles prevent micturation, the detrusor will relax for a while and allow the bladder to collect more urine. Throughout this process, the ureters should collapse, preventing reflux or backward flow of urine into the kidneys that could cause disease such as hydronephrosis.

The neurophysiology of the urine system is incompletely understood. Perhaps most important is that detrusor contraction is apparently mediated by cholinergic activity of the parasympathetic nervous system. At the same time, the contraction of the bladder neck is apparently mediated by adrenergic activity. Yeates (1982) provides a clear description of the neurodevelopmental process that leads to continence. He notes that this process is often delayed in patients with other developmental delays.

Causes of Urinary Incontinence

Ellis (1963), as well as Giles and Wolf (1966), has suggested that the most likely causes of urinary incontinence among the mentally retarded are lack of appropriate role models, absence of sufficiently effective training programs, and lack of learning ability that limits response to pro-

grams not well adapted for mentally retarded persons. Berkow (1982) suggests that urinary incontinence may be associated with passive-aggressiveness, dependency, sleepwalking, antisocial behavior, sibling rivalry, and speech disorders. Nevertheless, it is clear that incontinence can be the symptom of organic pathologies or a central nervous system dysfunction that either limits sphincter strength and control or affects cortical inhibition of micturation or defecation.

There are a variety of causes for urinary incontinence. These include developmental delay, disturbance in the learning process, emotional problems, including those due to sexual abuse, specific pathological conditions, and toxic side effects of various drugs. Campbell (1951) estimated that only 5% of the incontinence cases in the general population have clear neurological or urological abnormalities. Similarly, Sorotzkin (1984) suggests that the incidence of organically caused incontinence is between 1% and 10% of the cases, depending on the sample and diagnostic differentiation. The percentage of incontinence cases with a clear physiological abnormality in a developmentally delayed population is probably somewhat higher. Children with demonstrated neurological or urological abnormalities have a higher incidence of incontinence. Lister, Zachary, and Brereton (1977) reported that only 21% of the myelomeningocele (spina bifida) patients in their sample were continent by age 6 as compared to the usual 85% to 90% of the general population, and only 25% were trained by age 10 as compared to the usual 95% to 97%. Lorber (1971) reported that only 17% of 134 children in a series of myelomeningocele patients had normal sphincter control.

Organic Problems

Although it is most likely that urinary incontinence does not involve a medically remediable problem, it is important to rule these possibilities out. The most common anatomic causes of incontinence are phimosis (narrow opening of the foreskin), meatal stricture and meatitis, urethral stricture, posterior urethral valves (boys), distal urethral stenosis (girls), and contracture of the bladder neck (Berkow, 1982). In these cases, normal urinary flow is obstructed, and the bladder becomes hypertrophied, trabeculated, and irritated. Uninhibited contractions are more likely to occur during sleep. Nonobstructive uropathies such as urethritis, trigonitis, and cystitis (bladder inflammations), juvenile diabetes, pinworms, epilepsy, and spina bifida may also cause enuresis. Finally, there are some other structural problems that may lead to incontinence. Idiopathic small bladders account for some cases of enuresis. An ectopic (out of place) ureter usually presents as a chronic history of day and

night leakage despite a normal pattern of voiding throughout the day. This is a problem only in females and requires surgical repair. A fistula or hole between the vagina and the urethra, bladder, or ureter will lead to incontinence. The leakage may or may not be constant. Surgical or accidental trauma or neoplasms can cause this kind of urinary fistula.

Stress incontinence (sometimes referred to as giggle incontinence) is due to weak urinary sphincters. It results in small amounts of leakage when intraabdominal pressure increases from lifting, running, or laughing. This may occur in men following prostrate surgery or damage to the lower urinary tract. It is the most common cause of incontinence in women. In women, incontinence with mild physical stress is commonly associated with a cystocele (bladder hernia). Apparent stress incontinence should be evaluated by a physician. Mild cases are often well treated by Kegel (stream interruption) exercises that strengthen the sphincters. More serious cases may be treated with medications or may require surgical repair.

Total incompetence of the urinary sphincter results in day and night leakage of urine, regardless of bladder distention. This may be a congenital condition or may be the result of trauma (e.g., due to prostrate surgery) and is a problem that requires medical evaluation but may be helped by a behavioral intervention.

Urge incontinence refers to loss of urine following a particularly strong and often sudden urge to void. Some degree of stress incontinence may be present. There are a variety of physical etiologies that present with urge incontinence. Some are quite serious (neoplasms), and all deserve careful medical evaluation.

Urinary retention with overflow incontinence is the result of overdistention of the bladder due to an obstruction or a problem with contraction of the detrusor. Either way, the patient has trouble starting a stream and completely emptying the bladder. This can be caused by a wide range of problems, including myelomeningocele, spina bifida occulta, cancerous growth, spinal cord injury, vesical neck contractures, urethral strictures or valves, urinary calculi, and meatal stenosis as well as conscious blocking of the urethra for some functional reason. Clearly, these possibilities deserve medical evaluation.

Neurogenic bladder is a vesical dysfunction. The bladder may be either too flaccid (hypotonic) or too contracted (spastic). A neurogenic bladder is the result of a neural problem at the level of the brain, spinal cord, or local nerve supply for the bladder. The problem may be congenital myelomeningocele, filum terminale syndrome) or acquired (spinal cord injury, diabetes mellitus, demyelinating or degenerative diseases). A neurogenic bladder may produce a variety of symptoms,

including frequent urination, incontinence, or urinary retention. Immediate treatment will probably include continuous or intermittent catheterization to prevent overdistention of the bladder. A variety of surgical procedures may be helpful.

Urologists have identified an acquired disorder that, although it resembles the neurogenic bladder in presentation, is not associated with demonstrable organic pathology (Allen, 1977; Hinman & Baumann, 1973). This disorder is variously referred to as a nonneurogenic neurogenic bladder, a neuropathic bladder, subclinical or occult neurogenic bladder, and "lazy" bladder syndrome. The typical presentation is a heavily trabeculated, large-capacity bladder. Residual urine is left in the bladder following urination that can lead to infection. Ureteral reflux is believed to develop due to the high pressures that develop in the bladder, and kidney damage may result. This disorder may be functional or may involve a lack of coordination betweeen the detrusor and external sphincter. It may be functional if, for example, the patient avoids urinating on a reasonably normal schedule to avoid school bathrooms. Allen (1977) advocates intermittent catheterization to manage the problem acutely and a behavioral retraining program to return the urinary system to normal function. Russo (1978) reports one behavioral program used to treat this sort of problem.

Urinary tract infections, which are associated with incontinence, are more common in female enuretics than nonenuretic females on the order of 5.5% versus 1.5% (Dodge, West, Bridgeforth, & Travis, 1970). However, it is most likely that the infections are due to the enuresis or some third cause (e.g., incomplete bladder emptying) that produces both problems. Whatever the case, symptomatic patients need medical evaluation and treatment concurrent with toilet training if not before.

Developmental Problems

A different set of physical problems stemming from developmental delay can result in incontinence. Small bladder capacity and slowed neurological development affecting coordination and/or tone of sphincters are the most common examples of this. Enuretics typically have smaller bladders and void smaller amounts than nonenuretics. Small bladders are more commonly a problem in boys than in girls (Herskowitz & Rosman, 1982). There is no definitive explanation as to why smaller bladders occur in some children and not in others. Sorotzkin (1984) reviews a variety of possible etiologies, including too frequent toileting by parents that prevents the child's bladder from being stretched to increase capacity, genetic determination, allergic reactions, and anxiety

leading to increased bladder irritability so that lower volumes set off uncontrollable contractions. Sorotzkin also cites research suggesting that a small bladder may be the result of enuresis rather than the other way around. Typically, a 3 to 4 year old has sufficient bladder capacity for effective toilet training.

Smith and Wong (1981) report interesting results on increasing the small bladder capacities of mentally handicapped children as they proceed through toilet training. After using a curve smoothing procedure, they plotted bladder capacity (i.e., amount voided) against day of training. The results for seven of nine children indicated that a curvilinear model provided the best fit to the data. Specifically, apparent capacity first rose, then fell, then rose again as they reached the end of training. Similarly, they found a curvilinear pattern (rise/fall/rise/fall) when plotting frequency of passing continent urine against day of training. One way to account for these data is that the results have more to do with cognition than bladder capacity. The results may represent increasing awareness of bladder function—both when there is urine present to void and how to void with less than a full bladder (the last step in Bettison's, 1979, developmental sequence). Additionally, these results can be partially accounted for by increasing awareness of the positive and negative reinforcement contingencies of the toilet training program.

Medical Treatment for Urinary Incontinence

Medication

Although a whole range of drugs have been tried as treatment for enuresis, only the tricyclic antidepressants have helped. Typically, imipramine is used. Three possible explanations exist for the helpfulness of imipramine: decreasing depression, changes in sleep pattern and quality, and the peripheral blocking of norepinephrine reabsorption that may increase alpha adrenergic activity in the bladder neck and urethral outlet. The most likely explanation appears to be the last (Caine, 1983). Blackwell and Currah's (1973) review of the pharmacologic literature on enuresis concludes that:

> . . . benefit, when it occurs, usually commences within the first week of treatment, but total remission is seen in under half the patients. Relapse tends to occur immediately following withdrawal after short periods of treatment, and . . . total remission occurs in only a minority of patients. . . . In general, the results obtained with drug treatment are less promising than either the short- or long-term effects of the "bell-and-pad." (p. 253)

Imipramine, while sometimes effective, is a potentially lethal drug. Further, the relatively common side effects, which include irritability, restlessness, and difficulty with concentration, make imipramine less than a desirable drug for a generally harmless condition. Its use in treatment of an essentially benign disorder should be limited to cases that have not been responsive to behavioral intervention. It should never be the first choice of treatment.

Surgical Interventions

In the small number of cases where an obstruction can be surgically corrected, the results can be dramatic. Berkow (1982) suggests that surgical correction of demonstrated obstructive uropathies can result in complete alleviation of bed-wetting in a few weeks or months.

Medical Referral

In general, all patients who are seen for incontinence problems should be thoroughly evaluated by a physician. Failure to do so could lead to a long but unsuccessful course of treatment in a person who has a serious and possibly treatable underlying physical condition. Thus failure to refer could be construed as unethical and possibly as malpractice. Further, some medications could be involved in the etiology of an incontinence problem. A referral that asks about this specifically could lead to a change in medication that would help solve the incontinence problem. Even when a physician has made the initial referral for toilet training, it is a good idea to consider the possibility of an underlying medical disorder. Table 1 suggests questions to ask that will help identify the need for further medical evaluation.

Comments on Behavioral Training Procedures

Behavioral treatment of urinary incontinence emphasizes the learned nature of continence and attributes a persistent lack of continence to poor or inadequately developed habits, poor learning experiences and environments, and insufficient reinforcement for developing a new habit (Doleys, 1977).

Training Using Alarms

The most consistently successful toilet training programs have used an alarm. Although the early success rates of 90% have rarely been

Table 1. Questions That May Suggest the Need for a Medical Referral[a]

General
Is incontinence nighttime only?
Does the incontinence occur daily?
Does the incontinence include both bowel and bladder?
Is incontinence associated with laughing or straining?
Does the child have trouble telling if he or she has a full bladder/bowel?
Is there a known seizure disorder?
Are there any indications of nighttime seizures (waking with unexplained muscle soreness, waking with bitten tongue, excessive morning sleepiness)?
Does the child have symptoms that could indicate diabetes (drink or eat excessively plus abdominal pain or weight loss or polyuria)?
Is there a history of hydrocephalus, spinal cysts, tumors or spina bifida?
Does the child seem to have a gait disturbance?
Has an appropriately designed program that was followed for at least a month failed to improve the pattern of incontinence?
Does leakage seem to be more or less constant?
For enuresis specifically
Are there symptoms of a urinary tract infection (urgency, high frequency, burning)?
Is there a family history of enuresis?
Is there a problem with dribbling after urination?
Is bladder capacity smaller than expected?[b]
Is the stream of urine small (suggesting stenosis or stricture)?
Is the child taking any medications such as theophylline, aminophylline, Mellaril, or Haldol regularly or occasionally?
For encopresis specifically
Does the child complain of pain from defecation?
Is the child taking any medications that are anticholinergic (e.g., imipramine), adrenergic (e.g., ritalin or noradrenaline), sedating (codeine, barbituates) or antipsychotic (Haldol, Thorazine)?

[a] A "yes" answer to any of these questions may suggest the need to consult with a physician.
[b] This can be estimated by the following equations (Hjalmas, 1976): Males—1.12 + 7.81 × weight in kg or 31.6 + 24.8 × age in years or −207 + 3.47 × height in cm. Females—28.6 + 7.06 × weight in kg or 37.4 + 22.6 × age in years or −188 + 3.26 × height in cm.

duplicated, Doleys's (1977) review of 12 behavioral studies found a very respectable 75% of 628 subjects met the researcher's criteria for success. The primary cause of failing to achieve initial success in toilet training programs is lack of persistence at applying the program (Doleys, 1977) that further suggests the potential efficacy of this approach. Doleys (1977) identified four behavioral theories and corresponding approaches to toilet training using an alarm. Mowrer and Mowrer (1938) account for their success with the bell and pad in terms of classical conditioning. Lovibond (1972) preferred accounting for the data in terms of conditioned avoidance training with the noise of the alarm and waking up

being aversive stimuli that the child tries to avoid. Turner, Young, and Rachman (1970) suggest that the same results are better understood in terms of a punishment paradigm: The alarm is aversive and serves to decrease the frequency of the immediately preceding response: micturation. Finally, Azrin *et al.* (1973) have looked at toilet training more from a sociomotivational point of view.

The most difficult issue in comparing different treatment programs is relapse rates. This seems to be where the most differences are. Again, early reports of very low relapse rates have not been duplicated. Relapse by 40% within 6 months seems to be far more common. Happily, retreatment appears to be generally successful. Doleys's (1977) review found that 68% of those who relapsed were successfully treated on the second try. Bollard (1982) reports that nearly 100% are successfully retreated.

Two approaches have been suggested for decreasing relapse rates. Finley, Rainwater, and Johnson (1982) have suggested that use of an alarm is best viewed in terms of an operant model and, therefore, continued success can be best encouraged by use of an intermittent schedule of reinforcement. Their results suggest that a schedule that activates the alarm 70% to 79% of the time led to substantially lower relapse rates than those experienced by the control group that received a standard training package.

Morgan and Young (Morgan, 1978; Young & Morgan, 1972 a,b) suggest that the most effective way to cut relapse rates is to use an overlearning procedure. To do this, they follow the usual 14 nights of dryness by having the child drink up to a quart of liquid at bedtime. Finley and colleagues (Finley *et al.*, 1982) suggest that it probably makes sense to combine both intermittent schedules and the overlearning approaches. They believe that it makes most sense to start with the increased fluid intake at the beginning of training.

Increasing Bladder Capacity (or Retention Training Control)

Small functional bladder capacity may contribute to enuresis. This theory has led to bladder-stretching exercises as a treatment for incontinence (Kimmel & Kimmel, 1970). Generally, the idea is to encourage the person to delay urinating for increasingly longer periods of time. This should help the detrusor muscle adapt to increasingly larger volumes. However, research to date does not suggest that this treatment by itself is entirely effective—especially as compared to the results ob-

tained with an alarm (Sorotzkin, 1984). Reinforcement for increased latency to micturation and for voiding increased amounts has not been consistently helpful. However, Doleys (1977) suggests that tangible reinforcers contingent on these increases may be more helpful with younger children and presumably with developmentally delayed children.

A report by Thompson and Hanna (1983) raises some questions about the safety of using increased fluid intake as part of a training program, especially with mentally retarded people. They note that overhydration can lead to seizures, coma, and death. They suggest screening to insure good kidney function and monitoring of electrolytes in order to prevent problems.

Other Behavioral Procedures

Smith and Wong (1981) note that there are two approaches to toilet training: (a) regular "potting" or sitting on the toilet at regular intervals regardless of other variables such as past history of times of accidents and (b) placing the trainee on the toilet at times suggested by baseline data at times when he or she frequently has an accident. Due to the tremendous variance in their data, they suggest that sitting at regular intervals makes the most sense and involves less data keeping and analysis, which are unlikely to provide useful information. They also suggest that fluid intake should be increased during training rather than restricted. Van Wagenen, Meyerson, Kerr, and Mahoney (1969) suggested forward moving as another solution to the question of how to schedule toilet training. Using an alarm, the trainee is trained to inhibit voiding in inappropriate places and to move quickly to the toilet when voiding is imminent. Thus, neither a regular potting approach nor use of questionable baseline data is needed, and each training trial is a close approximation of the target behavior.

Punishment has occasionally been tried (Doleys, 1977) but is generally seen as a poor choice and is seldom necessary or recommended. The only significant exception to this is the Azrin *et al.* (1973) Dry Bed Training program. This program includes an overcorrection procedure labeled positive practice that requires repeatedly pretending to be asleep in bed and then getting up to go to the bathroom following each instance of incontinence. It also includes cleanliness training that requires that the child immediately change pajamas and sheets following each instance of incontinence. These procedures are undoubtedly experienced as somewhat punishing by all children and most parents. Nevertheless, it is seen as both appropriate and helpful by most professionals.

Biofeedback procedures may prove to be especially useful for patients who have incontinence problems due to weakness or incoordination of the sphincter muscles. Although it is impossible to give direct EMG feedback from only the sphincter muscles without using needle electrodes and is somewhat inconvenient to place even surface electrodes, the sphincters are quite close to the surface, and satisfactory EMG readings can be obtained to give meaningful feedback. Some clinical work is being done that suggests this may be an effective training procedure (Masek, personal communication, April 1986).

Conclusions and Recommendations for Urinary Incontinence Remediation

The preponderance of the evidence indicates an alarm should be used if one is available. Although Azrin *et al.*'s (1973) Dry Bed Training puts a significant demand on both parents and children, it provides the best single framework for treatment and can be easily combined with an alarm. Overlearning and/or use of intermittent alarms should be strongly considered to help decrease risk of relapse. Finally, if the child's apparent bladder capacity is below what should be expected for his or her age and/or if the frequency of urination is high, Retention Control Training should be included as part of the program.

Treatment for enuresis should be based on whether "the presence of enuresis is interrupting the normal sequence of social, emotional, cognitive, or motor development" (Cohen, 1975, p. 558) rather than on the child's age. This certainly holds true for developmentally delayed people. Incontinence can clearly place limitations on what a person is allowed to do and thus deserves attention, but the treatment should not involve more risk than benefit.

Comments Specific to Developmentally Delayed Populations

Various approaches to toilet training for the developmentally delayed have been suggested in the scientific literature. One of the first was described by Ellis (1963). His was based on a simple stimulus-response analysis. Since then, a number of others have developed, tried, and reported using a number of tactics or components in different combinations and with varying experimental designs (usually insufficient for proving very much). The most commonly used and suggested com-

ponents are use of alarms (Azrin, Bugle & O'Brien, 1971; Hanson, 1983; Mahoney, Van Wagenen, & Meyerson, 1971; Mowrer & Mowrer, 1938; Van Wagenen & Murdock, 1966), an overcorrection procedure usually called cleanliness training (Azrin *et al.*, 1973), forward moving (Van Wagenen, *et al.*, 1969) and positive practice (Azrin *et al.*, 1973).

A review of behavioral approaches by Whitman, Scibak, and Reid (1983) suggests that none of the behavioral programs developed has been satisfactorily replicated and validated. However, they do note that Van Wagenen's (Van Wagenen *et al.*, 1969) forward-moving approach and the Azrin and Foxx (1971, 1974) dry bed training approach both offer some very attractive and promising features. It probably makes the most sense to combine the elements of both these programs that are most applicable and practical for the setting in which they are used. Whitman *et al.* suggest one example of how this might be done.

Fecal Incontinence

Fecal incontinence refers to involuntary elimination of fecal material or elimination in an inappropriate place (e.g., pants). Not all fecal incontinence is encopresis, which refers to incontinence of feces or fecal soiling by a person who is developmentally mature enough to be continent and who has no clear physiological problem (Parker & Whitehead, 1982). Herskowitz and Rosman (1982) suggest that bowel training is typically accomplished by age 3; failure of bowel training in a 4 year old is uncommon; and failure in a 5 year old is abnormal. An isolated accident certainly does not constitute encopresis. Additionally, staining due to diarrhea or too much mineral oil does not constitute encopresis. Disagreement exists among clinicians and researchers regarding what age to select as too old to be incontinent, how soiled a person must be, how frequently an accident must occur, and what etiologies qualify for a diagnosis. However, Parker and Whitehead (1982) suggest a reasonable working definition: incontinence at least once per week for at least 1 month by a child 4 years old or older. Herskowitz and Rosman (1982) report that the form of encopresis encountered most commonly is secondary and due to severe constipation in a child 5 years old or older. In contrast to enuresis where the majority of the cases are considered primary, 50% to 60% of the encopresis cases are secondary (Fritz & Armbrust, 1982).

A number of distinctions are normally made when discussing fecal incontinence. As with urinary incontinence, a distinction is made be-

tween primary and secondary incontinence. Groves (1982) suggests that a distinction between incontinence associated with retention and incontinence not associated with retention can also be helpful. *Retentive* incontinence refers to incontinence associated with constipation and/or impaction, whereas *nonretentive* refers to incontinence associated with passage of stool of normal form and consistency. Neither term is intended to suggest or not suggest psychogenic involvement. Finally, a distinction is typically made between organic and functional factors contributing to fecal incontinence. There is substantial overlap with urinary incontinence in the psychosocial problems that are suggested as causes for encopresis (passive-aggressiveness, dependency, sibling rivalry). As with urinary incontinence, fecal incontinence can also be the symptom of an organic pathology, which may be quite serious. The various organic pathologies as well as normal bowel function will be discussed later.

Fritz and Armbrust (1982) state that a small number of encopretics are mentally retarded. Encopresis has also been associated with distractibility, short attention span, low frustration tolerance, hyperactivity, and poor coordination, all of which may be present in mentally retarded people. To this list, Groves (1982) adds learning and language disorders, neuromuscular problems, dietary patterns, medications, and environmental or family changes and stresses. Fritz and Armbrust (1982) also note that encopretic children are five times more likely to be chronically constipated than other children. Additionally, 25% will also be enuretic; however, only 8 percent of the enuretic children also have trouble with encopresis. Generally, though, fecally incontinent children are developmentally and neurologically normal. However, as with urinary incontinence, fecal incontinence can be a symptom of a serious underlying medical problem.

Physiology of Defecation

Continence involves a complex interaction of muscles and associated afferent and efferent nerves. As Silverberg (1983) explains, the rectum is generally kept closed off from the anal canal by the internal sphincter, which is normally contracted, and other smooth muscles of the pelvic diaphragm. The levitor ani keeps the anorectal angle elevated sufficiently to prevent unwanted elimination. The lining of the rectum includes stretch receptors sensitive to being stretched by feces or gas. Sufficient stretching of the rectum leads to relaxation of the internal sphincter. Feces or gas then pass into the anal canal. The lining of the

anal canal is sensitive to touch, temperature, and pressure. It is able to differentiate between gas and feces. Defecation may be stopped by contraction of the external anal sphincter, which partially surrounds the internal sphincter. Additionally, the anorectal angle can be further increased by additional contraction of the levitor ani. This helps narrow the anal canal. The gluteal muscles can also be contracted to help maintain continence. In the continent individual, elimination will occur when the levitor ani is relaxed and a Valsalva maneuver, which increases intraabdominal pressure, is performed. This leads to relaxation of the external sphincter and helps move the contents from the rectum into the anal canal. Most of these mechanisms are operational by age 2 in the normal child. Parenthetically, Silverberg (1983) points out that toilet training has more to do with training the child to be aware of the sensations and knowing what to do than it has to do with organizing the neuromuscular physiology.

A number of factors affect bowel function. One is the peristaltic motion of the colon that in turn is affected by a number of factors. One of these is food entering the large intestine. This initiates the gastrocolic reflex that can produce powerful, propulsive movements of the bowel wall. Physical exercise and emotional state also both affect the function of the colon (Walker-Smith, Hamilton, & Walker, 1983). Physical exercise increases the muscle activity of the colon, especially right after meals. Anger and hostility also seem to increase the muscle activity, whereas depression seems to decrease activity. Dietary patterns, neurological dysfunction, and constipating medicines can all contribute to the development and maintenance of incontinence.

Causes of Fecal Incontinence

There are a variety of organic etiologies for fecal incontinence. Hersov (1977) argues that fecal incontinence is best conceptualized as a disorder with a multifactorial etiology. Medical problems include obstructions and neurologically based problems, including Hirschsprung's disease, which is common among Down's syndrome patients. Groves (1982) lists other specific diagnoses that are associated with hard stools, including dehydration, diabetes insipidus, idiopathic hypercalcemia, redundant rectosigmoid colon, and infantile renal acidosis. Hard stools can lead to avoidance of defecation as a way of avoiding pain. This subsequently leads to disrupted bowel function. Groves also lists a number of diagnoses commonly associated with constipation, including anal

fissure, stenosis, amyotonia congenita, cerebral palsy, hypothyroidism, congenital absence of the abdominal musculature, lead poisoning, and infectious polyneuritis. Additionally, there are other possible causes for fecal incontinence, including improper diet and seizure disorders. Finally, a number of medications can affect bowel function (Walker-Smith *et al.*, 1983). Cholinergic agents (e.g., acetylcholine) can increase activity. Adrenergic agents (e.g., Ritalin, noradrenaline), anticholinergic agents (e.g., imipramine, atropine), and sedatives (e.g., opiates, barbituates) all relax the colon and may have a constipating effect. It is important to identify medication with constipation as a possible side effect because a small change in medication type or dose may lead to an easy cure for constipation and incontinence. A number of conditions can affect sphincter strength, coordination, and conscious control. Developmental delay is typically thought to lead to incontinence by affecting sphincter strength and coordination. These problems typically involve nervous system deficiencies such as spina bifida, spinal cord tumors or lesions, and nervous sytem degenerative disorders (e.g., multiple sclerosis).

Even if there are clear organic reasons for incontinence, psychosocial factors still need to be considered. Important factors include the method of toilet training, the age at which training is attempted, and presence of a physiological predisposition to a particular style of bowel functioning. Other factors may include general parent–child relationships, the family system, and the presence of environmental stressors including history of abuse.

There are also purely behavioral etiologies of incontinence, for example, using an inadequate bowel training program. Failure to become continent may be the result of lack of skills on the part of the parents. Significant psychological problems in either the patient or parents need to be identified and treated because they can contribute to encopresis. Reaction to a specific stress (e.g., moves, sexual abuse, birth of a sibling) can result in delays in toilet training or regression. Encopresis can also serve a functional purpose such as gaining attention (Sluckin, 1975), avoidance of pain, or perhaps, as part of a power struggle (Garrard & Richmond, 1952; Hersov, 1977).

Generally, development of obstipation (severe constipation) is the process that leads to fecal incontinence and, usually, there is no clear physiological cause for the constipation. However, there is a typical course of events that occurs (Groves, 1982; Levine, 1982; Walker-Smith *et al.* 1983). Stool retention starts for any of a variety of reasons (school rules, fear of public/school bathrooms, medication side effects, attempts at independence or protest, inattention to cues, poor diet, desire for attention, not wanting to interrupt play, etc.). The rectum dilates due

to excess accumulation, and the defecation reflexes become duller. Consequently, stools become larger and harder. At this point, a vicious cycle can begin: Defecation becomes painful, evokes fear, and/or gains attention, which further encourage retention. When the person finally becomes impacted, the pressure causes maximum dilation of the rectum and substantial blunting of the sense of a need to defecate. The internal sphincter loses its tone and becomes patulous. Liquid stool moves through the colon and anal canal producing very little sensation, leaks out, and produces staining and soiling. Silverberg (1983) suggests that this sort of overflow incontinence occurs in more than 95% of encopretic patients. When this is the case, the colon will need to be cleaned out for toilet training to progress.

It is worth noting that this pattern describes how Hirschsprung's disease (also referred to as aganglionic megacolon) leads to incontinence. In Hirschsprung's, a portion of the colon is not ennervated sufficiently for it to contract. The feces are not moved through the colon and consequently create an obstruction that leads to constipation. Alternatively, the pattern described before can result in and be maintained by an acquired megacolon (also referred to as idiopathic megacolon) where the stretching of the bowel occurs despite normal ennervation. When badly stretched, the bowel is unable to maintain normal peristalsis or produce reliable, strong propulsive movements. Sensation eventually becomes dulled, which complicates the problem of training. The result is constipation and soiling. Although similar in effect, the two conditions can be differentiated on the basis of rectal biopsy and, perhaps, manometry.

Medical Treatment for Fecal Incontinence

Hersov (1977) notes that most children will simply outgrow encopresis by age 16. However, because few people are willing to put up with the problem for that long, a variety of interventions have been developed. Clearly, some underlying causes such as lead poisoning or dehydration must be dealt with first.

Medication Interventions

Herskowitz and Rosman (1982) suggest that the most common cause for encopresis is leakage of feces after severe and chronic constipation has developed. When this is the case, they advocate a three-phase approach to medical management of encopresis. The first phase consists

of removal of impacted feces through use of mineral oil and, in some cases, phosphate enemas. Further medical workup may be needed if this phase cannot be completed. The second phase consists of training. Training should include bowel training (coordination of the muscles involved, awareness of sensations involved) the trainee's behavior (going to the bathroom, undressing, cleaning, etc.), and the parent's or trainer's behavior (how to teach, reward, avoid struggles, etc.). This phase includes a schedule of regular sitting. Sitting right after meals is suggested because it takes advantage of the gastrocolic reflex (Brazelton, 1962). Use of mineral oil, stool softeners, and suppositories may be continued. The third phase consists of follow-up and support. Levine (1982) also offers a comprehensive medical approach including diagnosis, education, initial bowel cleanout with a regimen of enemas and suppositories, management with mineral oil, suppositories, and regular sitting, and, finally, a relatively long follow-up period.

Increases in dietary fiber, frequent sitzbaths, and topical anaesthetics for anal fissures are also normal medical interventions used when appropriate (Walker-Smith *et al.*, 1983).

Surgical Interventions

Surgical treatment for fecal incontinence includes repair of sphincters, plication procedures, creating a functional sling around the anus that is attached to the gluteal muscles, muscle transplant, and colostomies (Castro & Pittman, 1978). Hirschsprung's disease can be treated surgically by removing aganglionic sections of colon. Normal sections are then joined (Berkow, 1982).

Medical Referral

It is worth noting again that a medical evaluation should be completed as part of the initial workup for toilet training. Table 1 includes specific questions to ask about fecally incontinent patients that can help determine the need for referral.

Behavioral Medicine Procedures

There have been two behavioral approaches to fecal incontinence described in the literature. The first sort of intervention has simply involved reinforcement for improvement and eventually for complete con-

tinence (Ayllon, Simon, & Wildman, 1975; Bach & Moylan, 1975; Baker, Brightman, Heifetz, & Murphy, 1977; Edelman, 1971), extinction (Conger, 1970), or punishment for incontinence (Edelman, 1971; Gelber & Meyer, 1964). The second sort of intervention has added other procedures to the usual learning paradigms. Most typically, enemas and/or suppositories are used (Christophersen & Rainey, 1976; Groves, 1982). Azrin and Foxx (1971) as well as others (Doleys & Arnold, 1975) have added "full cleanliness training" that includes expressions of displeasure over accidents and correction of the problem (cleaning clothes and self). A schedule of regular sitting is often added as well (Groves, 1982). Young (1973) tried to take advantage of the gastrocolic reflex by having the sitting scheduled after a meal and backed it up with suppository administration if there was no bowel movement.

Jeffries, Killam, and Varni (1982) have presented a case study involving a 9-year-old male with a sacral-level myelomeningocele who had never achieved bowel or bladder continence. The patient had reduced sensation in the anal area but was described as otherwise normal. A straightforward positive reinforcement program for continent eliminations and absence of incontinence was established and helped the boy reduce instances of fecal incontinence from 7.7 per week during baseline to 1.4 per week at the end of treatment. After 9 months of follow-up fecal incontinence had been further reduced to 0.16 instances per week. An additional side benefit may have been the reduction in instances of bed-wetting from two to three times per week to never at follow-up. Of at least equal importance, his peer relationships and a variety of other behaviors improved. The change in the patient's behavior produced good results that eventually also reinforced his increased attention to maintaining continence. It is important to note that a strictly behavioral intervention was extremely helpful even in this case of obvious neurological impairment. This same point is made by a number of other researchers (Killam, Jeffries, & Varnie, 1985, Whitehead, Parker, Masek, Cataldo, & Freeman, 1981).

Several points should be made about behavioral interventions. First, regardless of the assumed etiology, a careful description of the problem behavior needs to be obtained so that a functional analysis of the problem can be done. This becomes all the more important for individuals with secondary incontinence and for those who show most of the developmental signs of readiness for toilet training. Among other things, this reduces the chances of initiating an inappropriate training program based on a premise of psychogenic causation (Groves, 1982). Further, it is important to differentiate those who are incontinent simply due to an inadequate training program and those who are incontinent for other

primary reasons (e.g., chronic constipation). In cases where regression occurs—especially if a number of behaviors are involved—it is appropriate to start with a thorough medical evaluation, but behavioral evaluation should be done simultaneously. This should include a look at family, school/work, and social functioning. Finally, it is important to gear a training program to the trainee's cognitive, emotional, and neuromuscular developmental level.

Biofeedback has been tried in some cases of fecal incontinence (Engel, Nikoomanesh, & Schuster, 1974; Goldenberg, Hodges, Hersh, & Jinich, 1980; Orne, 1979; Whitehead *et al.*, 1981). For the most, part the biofeedback system used has been the same as or similar to the balloon apparatus used for manometric studies of bowel function. This is generally expensive equipment and not as readily available as the more standard biofeedback equipment. These studies demonstrate that biofeedback can be helpful and that it makes sense to try to develop the required skills even in neurologically compromised patients. More recent clinical efforts have shown success with EMG training of sphincters using an insertable device (Masek, personal communication, April, 1986). This approach is more likely to be used in most settings.

Comments Specific to Developmentally Delayed Populations

Although developmental delay and mental retardation are commonly thought to be related to fecal incontinence, there is little research on this specific relationship. For example, there are no published guidelines for identifying when incontinence should, in fact, be attributed to developmental delay (Groves, 1982). However, it is clear that developmentally delayed youngsters are at greater risk for social stress than others due the rejection they are bound to receive and the greater trouble they have with keeping pace with peers in a broad range of activities. This increased stress alone can make toilet training more difficult.

Groves (1982) notes that generalization of any learned skill, including toilet training, to new settings can be especially difficult for developmentally delayed and mentally retarded persons. Thus hospitalization, changes in teachers or classrooms, vacations, or any other significant environmental change can be a precipitant of incontinence. It is most helpful to think of these situations as a generalization problem rather than regression or failure.

Finally, dietary patterns (e.g., preference for soft, low-bulk foods high in sugar), neurological dysfunction, and constipating medicines

(commonly used medications are listed in Table 1) deserve careful attention in a developmentally delayed population (Groves, 1982).

It is important to be aware that there is a paucity of research clearly relating incontinence to developmental delays and retardation. However, even the early research that is available (Baumeister & Klosowski, 1965; Giles & Wolf, 1966; Levine & Elliot, 1970) as well as more recent work that has been done (Butler, 1976; Sadler & Merkert, 1977; Song, Song & Grant, 1976) suggests that progress can be made using simple programs with profoundly retarded individuals. Further, because it is becoming increasingly clear that patients with demonstrable lesions can achieve or improve continence, developmental delay and retardation should not be used as an excuse for failing to implement a research-based toilet training program for even the most severely impaired (see Wagner & Paul, 1970).

General Comments about Behavioral Medicine Intervention for Incontinence

It is becoming increasingly clear that behavioral medicine has much to offer most patients with incontinence problems. At the same time, it is inappropriate to automatically consider all instances of incontinence as behavioral or psychological phenomena. Responsible care and professional ethics require that the possibility of a medical disorder be considered and ruled out. A good intervention program will start with a thorough functional analysis and appropriate medical evaluation. The program needs to take into account the multifactored nature of the problem. Consideration needs to be given to physiological development, psychological status, cultural and environmental factors, and the possible interactions of these factors. Specific developmental issues include whether the trainee can sit on the toilet, whether retention is possible until sitting on the toilet, ability to communicate a need to go to the bathroom, and perhaps some appreciation for the fact that others are interested in continence (Schaefer, 1979).

Symptom substitution is a concern of many clinicians as well as many parents. Psychodynamic formulations of encopresis, as well as other behavioral problems, have sensitized many practitioners to the likelihood of symptom substitution. However, work by Levine, Mazonson, and Barkow (1980) does not support this concern, at least when the initial problem was encopresis.

What To Do When It Doesn't Work

When a well-designed and executed program has been carried out with little or no improvement, an additional medical evaluation by a urologist or gastroenterologist may be helpful if the initial evaluation was not done by a specialist. Initial medical evaluations made prior to referral for a behavioral medicine intervention have occasionally been known to be somewhat cursory because the incidence of positive findings is relatively infrequent. Second, another functional analysis should be done. Adherence needs to be assessed, because, in cases that do not involve organic problems, the most common reason training programs appear to fail is that they are not adequately implemented. With this in mind, it is easy to appreciate the importance of continuing support and encouragement and long-term follow-up. Further, there is no reason to suspect that failure to follow a program would not occur in organically caused cases. In fact, it may be more likely to occur. Adherence is important to assess not only to insure that the established program is adequate but also because failure to follow a program can indicate significant depression, the presence of more pressing behavioral problems, and/or serious family or environmental problems. This can include understandable but maladaptive reaction to the developmental disorder. Finally Levine (1982) notes that nearly 20% of the patients referred to him for encopresis are relatively resistant to treatment and although remission rates are high, so are relapse rates. He suggests it can take the involved muscles a very long time (up to years) to respond and readjust as desired.

Wakefield, Woodbridge, Steward, and Croke (1984) give a description of a comprehensive, interdisciplinary approach to fecal incontinence. Their program includes a medical evaluation, a behavioral program for regular sitting and continent bowel movements, appropriate diet, physiotherapy (abdominal massage, anticonstipation yoga exercises, and hydrotherapy), medication, active communication with everyone involved in the child's care and education, and regular clinic visits. They see their program as aiming to reduce anxiety, restore confidence, provide hope, empty the bowel, and restore a regular toileting regime. They report that 20 of 31 patients were totally continent at the 12-month follow-up. This is a good example of how the interdisciplinary approach advocated by behavioral medicine can be effectively used.

Summary, Conclusions, and Directions for the Future

Although behavioral techniques can be applied successfully to cases where there is a clear organic problem, behavior modifiers need to be

aware of what the various medical problems can be and the need to make referrals for medical evaluation when appropriate. It is clear that the vast majority of incontinence cases will not be helped by surgery or drugs. Nevertheless, the cases that will be should be referred expeditiously with the offer to complete the behavioral training when the patient is ready. It needs to be recognized by the professionals who work with developmentally disabled individuals that maturational delay and mental retardation do not account for all the incontinence cases they see and medical intervention will be helpful in appropriately diagnosed individuals. Finally, incontinence is a good example of the overlap between behavioral medicine and developmental disabilities. There clearly are others as this edited volume illustrates. More cross-talk between the two fields will be helpful for both fields.

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CHAPTER 8

Use of Biofeedback in the Treatment of Incontinence

Bruce J. Masek

Biofeedback training for fecal and urinary incontinence is based on the assumption that patients with an absent or weak external anal or urethral sphincter response to rectal or bladder distention can learn to produce stronger sphincter contractions in response to distention. Learning is facilitated by providing the patient with immediate visual information about small variations in the activity of the sphincters through the use of electronic instrumentation. Gradually, over the course of biofeedback training, the patient learns to contract the external sphincters with sufficient magnitude and duration to oppose the forces of elimination.

Fecal Incontinence

Biofeedback of rectosphincteric reflexes in the treatment of fecal incontinence was first described by Engel, Nikoomanesh, and Schuster (1974). Their training method involved manometric recording of sphincteric responses using a three-balloon rectal tube pressure transducer connected via polyethylene tubing to a polygraph. The device allows for pressure measurements to be made from the proximal rectum and internal and external anal sphincters. The rectal balloon is also used to

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transiently distend the rectum. During biofeedback training, the patients viewed the polygraph tracings of their sphincteric responses when the rectum was distended. The visual feedback allowed patients to develop control of the external sphincter contraction when the rectum was stimulated and the internal sphincter reflexively relaxed. Patients also learned to contract the external sphincter in response to gradually decreasing volumes of rectal distention. Verbal reinforcement was provided by the experimenters for normal-appearing responses at first and then gradually faded out as patients demonstrated reliable sphincter contractions. Training trials were intermittently conducted without the benefit of feedback to promote generalization. Patients were instructed to practice sphincter contractions at home between training sessions. One to four biofeedback sessions, each lasting about 2 hours, were administered to the six adults and one child in the study.

Results indicated that four patients were fully continent after training with follow-up ranging from 6 months to 5 years. Included in this group was a child whose incontinence was secondary to a congenital neural-tube defect—myelomeningocele. Subsequently, other researchers have found these training procedures to be effective treatment for fecal incontinence in larger samples of children with myelomeningocele. (Wald, 1981, 1983; Whitehead, Parker, Masek, Cataldo, & Freeman, 1981; Whitehead *et al.*, 1986).

EMG biofeedback for fecal incontinence is a recent development that overcomes some of the problems identified with the three-balloon rectal tube method, namely movement artifact, distracting information contained in the polygraphic feedback display, and the lack of commercially available three-balloon rectal tube sensors. The technique involves insertion of an acrylic probe embedded with three metal electrode strips at the surface. The probe provides information on EMG activity of the external anal sphincter and is safe and painless for the patient. Computer-generated graphic display of the biofeedback signal, quantification of sphincter muscle EMG activity, and multiple-channel EMG recording to detect counterproductive abdominal and gluteal muscle activity are enhancements available at relatively low cost (Perry & Hullett, 1986).[1] Unfortunately, only one investigator (MacLeod, 1979, 1983) has published data on the use of the anal probe to treat fecal incontinence. Patients in these studies were 42 adults with incontinence to gas and liquid feces and, in some cases, solid stool as well. Results indicated

[1] Anal probe EMG sensors (Electronic Perineometer™) are available from Biotechnologies, Inc., 242 Old Eagle School Road, Strafford, PA 19089.

that 72% of the patients achieved at least a 90% improvement that was maintained in all cases for a minimum of 1 year.

Urinary Incontinence

Kegel (1948) is generally credited with developing the first biofeedback device to treat stress incontinence, which he named the perineometer. The device consisted of an intravaginal balloon the shape of a finger, a pressure gauge, and a hand pump. Inflation of the balloon allowed patients to monitor the strength of pelvic floor muscle contractions. Kegel (1956) reported 90% improvement among 455 patients treated with mild to moderate incontinence. Burgio, Whitehead, and Engel (1985) employed the three-balloon rectal tube method to treat 39 elderly patients with stress, urge, or detrusor motor instability incontinence. This was possible because, as the authors noted, the external anal and urethral sphincters are innervated by branches of the pudendal nerve, and activity in one reflects activity in the other, provided there is no damage to the sphincters. A catheter passed into the bladder was used to infuse sterile saline to monitor bladder pressure. Results indicated that the training procedures reduced incontinent episodes an average of 82%, 94%, and 85% for the three types of incontinence, respectively. Total continence was achieved by 13 patients.

Bladder–sphincter dyssynergia is a condition in which the external urethral sphincter does not fully relax during urination, resulting in incomplete bladder emptying, incontinence, and a propensity for urinary infections. Biofeedback training involves simultaneous surface EMG recording of external urethral sphincter activity and flowmeter recording of urinary flow rate during voiding. While viewing the polygraph tracings of these responses, patients attempt to relax the sphincter during voiding and maintain normal urinary flow. Sugar and Firlit (1982) reported that this method resulted in complete abatement of symptoms in 8 of 10 children between 6 and 16 years of age. Treatment was conducted during a 2-day hospitalization, and follow-up ranged from 6 to 19 months after discharge.

Conclusions

Biofeedback training for disorders of fecal and urinary continence is a recent development that is gaining acceptance as an alternative to

surgical procedures and pharmacologic therapy. However, because of the limited nature of the evaluative studies done to date, the efficacy of biofeedback treatment of incontinence is difficult to assess. More controlled studies that carefully define the patient groups who will most likely benefit from biofeedback training are needed.

Biofeedback training should not be considered unless there is demonstrable organic pathology or physiological abnormality resulting in incontinence. Manometric and urodynamic evaluation of fecal and urinary continence mechanisms provide essential information in this regard. Patients often require continued medical therapy during the early stages of training but then can be weaned from therapy as they progress in treatment. This decision needs to be made in consultation with the referring physician. Complications such as urinary infection, diarrhea, medication reaction, rectal fissure, or rash require prompt medical evaluation and can be contraindications for further training. In consideration of the preceeding, it is strongly recommended that clinicians doing biofeedback training for fecal and urinary incontinence accept referrals only from qualified medical specialists and work closely with them.

Biofeedback training for fecal and urinary incontinence requires specialized equipment and clinical expertise. As is the case with many therapies for medical disorders, patient cooperation and motivation to get better are important considerations. In this regard, children under the age of 6 have consistently demonstrated the poorest outcomes in previous research. Impaired cognitive functioning (IQ below 70) may also present an obstacle because of inability to concentrate on the training task (Whitehead, Burgio, & Engel, 1984).

The appeal of biofeedback training for fecal and urinary incontinence lies in its comparative safety, brevity of training, and the absence of a good alternative for many patients with incontinence due to sensory or motor impairment. Biofeedback training procedures for patients with severe congnitive impairment are lacking, as are procedures for other types of incontinence. Clearly this is a promising area of clinical investigation that has received minimal research support.

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Part II

Assessment Issues

CHAPTER 9

Behavioral Assessment Technology for Pharmacotherapy in Developmental Disabilities

Stephen R. Schroeder

Introduction

The burgeoning field of behavioral assessment in behavioral medicine and in developmental disabilities requires that, for a meaningful discussion within a brief chapter, the topic must be restricted even further. We have chosen behavioral assessment of pharmacotherapy with persons who have developmental disabilities because it is currently a very visible and controversial topic, with a rich research literature. The plan of this paper is to review the necessary behavioral and pharmacological issues that need to be considered when evaluating or conducting research in this area. It is not an exhaustive review. More complete treatment of our approach to behavioral assessment in mental retardation can be found in Rojahn and Schroeder (1983) and to behavioral pharmacotherapy with developmental disabilities in Schroeder, Lewis, and Lipton (1983). We will recommend several other current topical reviews as the various topics are treated. Five textbooks are recommended: Gadow and Poling (1988), Gadow (1986), Gualtieri, Schroeder, & Keppel (1987); Hersen (1986), and Werry (1978). The proceedings of three NIMH workshops, *ADD-H and Mental Retardation, Pharmacotherapy and*

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Mental Retardation, and *Rating Scales and Assessment Instruments for Use in Pediatric Psychopharmacology Research* published in two recent issues of the *Psychopharmacology Bulletin* (Rapoport & Conners, 1985; Reatig, 1985) are also especially helpful.

Common Methodological Problems

Design and Analysis Issues

Sprague and Werry (1971) in their classic critical review of psychopharmacology in mental retardation list six minimum design requirements: (a) placebo control, (b) random assignment of treatments and/or subjects, (c) double blind evaluation, (d) multiple standardized doses, (e) standardized (reliable and valid) evaluations, and (f) appropriate statistical analysis. The past decade has witnessed a substantial improvement in the design of behavioral pharmacotherapy studies, especially with high-prevalence/low-severity disorders like attention deficit disorders and conduct disorders. However, these requirements are more difficult to meet with low-prevalence/high-severity disorders like self-injurious behavior, aggression, destruction, and disruptive behavior. Chassan (1979) notes that in such populations one can anticipate the following problems: (a) lack of adequate sample size, (b) extreme heterogeneity of subjects, (c) subject attrition, (d) idiosyncratic all-or-none response by some subjects, (e) lack of specificity of treatment effects for individual subjects, (f) ethics of placebo groups or wait lists, and (g) difficulty of maintaining double-blind conditions, for example, when side effects are observed.

The previously mentioned difficulties will often influence the decision to use single-subject or group designs. Group designs are still by far the most prevalent designs used, especially in pediatric psychopharmacology. They are especially useful in detecting small but statistically reliable effects. However, when the subject population is so heterogeneous, it is questionable whether measures of central tendency in the group fairly represent the behavior of individuals. Indeed, even with the safest and most reliable drugs we use, it is still very difficult to predict who will respond to the drug and who will not. Single-subject designs with their repeated measurement techniques are much closer to the clinically accepted methods of titrating the effective dose of a drug. However, they take longer and are usually more arduous than group studies. Their external validity must come through replication across subjects. Treatment orders can be randomized or counterbalanced, so that sta-

tistics can be performed, although these are usually less powerful nonparametric statistics. To compensate for lack of sensitivity of instruments and lack of statistical power, studies often use a multimodal within-subjects crossover design strategy, involving repeated measurements from different domains, for example physiological measures, laboratory measures, and measures from the natural environment. Such a strategy assures that one "will find something," but it also increases the risk of Type I errors. It is still rare to find a group study in this area that takes into careful account a multivariate strategy, which assesses statistical power, sensitivity of measures, and confounding covariates such as side effects, caregiver practices, attribution bias inherent in drug trials, and ascertainment bias due to subject selection. It is equally rare to find single-subject designs that include an exploratory phase, then a confirmatory phase with repeated replications across treatments and subjects. Much work remains to be done on precision and accuracy in studies of behavioral pharmacotherapy. Accuracy has to do with whether the correct response is being assessed or predicted; precision has to do with how often it can be done reliably, so that replication can occur. These must be common goals of single-subject and group design strategies if both analysis and intervention are to be successful.

Pharmacological Issues

There are a number of pharmacological considerations that are likely to affect behavioral assessment in pharmacotherapy.

Dose response is certainly one of the most important parameters to consider. Some psychotropic drugs, for example, the stimulants, have a relatively stable dose-effect range, whereas others, for example, the neuroleptics, have a highly variable dose-effect range. There may be many pharmacological reasons for this variability, for example, the amount of metabolism of the drug by the liver, whether its sites of action are primarily in the peripheral or central nervous system, how easily it passes the blood–brain barrier, and the like (Briant, 1978).

Different doses of the same drug may well affect independent classes of behavior (e.g., learning versus social) differently (Sprague & Sleator, 1977). Low doses may synergize positively with behavioral intervention, whereas a high dose may have negative effects (Campbell *et al.*, 1978).

The type of drug and behavior plus the risk of side effects due to high doses may well influence the choice of a fixed versus titrated dose of the drug. For instance, the Campbell *et al.* (Campbell *et al.*, 1978; Campbell *et al.*, 1982) study previously cited concerned the effects of

haloperidol, a potent neuroleptic, with autistic preschoolers in an inpatient setting. They argued that, if they had used a fixed rather than a titrated dose of haloperidol, they would likely have failed to find a good response in a large proportion of their very limited sample of 40 children. Their point seems well taken.

Drug history (acute versus chronic administration) will very likely affect the interpretation of a study. Most psychotropic drugs have known serious side effects that are differentially affected, depending on whether the subject is drug-free or has been on the medication for years.

Drug metabolism often interacts with a behavioral response. Children differ greatly from adults in their response to different drugs due to rapid growth changes, neuroendocrine changes, and the like. There are substantial differences in the pharmacokinetics of children versus adults due to different drug absorption, distribution, renal excretion, and metabolism differences (Briant, 1978). Drug metabolism considerations may affect choice of route of administration, for example, oral versus intravenous, because the latter bypasses absorption by the stomach and metabolism by the liver. It may also affect order of treatment effects and length of washout periods between treatments. For instance, it generally takes a stimulant less than 48 hours to wash out of the blood, whereas neuroleptics may take weeks or months (Gualtieri & Hawk, 1980). Thus, repeated time-series designs, for example, ABAB, are probably confounded with irreversible period and carryover effects if a neuroleptic is used, unless each treatment period is at least a month long, whereas such a design might be achieved with a stimulant in a few weeks.

Drug–drug interactions are very common among psychotropic drugs. They add such a dimension of uncertainty and complexity to most studies as to be rarely recommended (Gualtieri & Hawk, 1980). A common problem still found in the literature is to do drug trials while a subject is receiving seizure medication. Most seizure medications have psychoactive effects that interact copiously with other psychotropic drugs (Aman, 1984). Such studies must always be interpreted with extreme caution.

Drug metabolism can also result in pharmacodynamic effects. For instance, it is not uncommon to be clinically titrating the dose of a psychotropic drug and to observe an all-or-none effect, for example no effect versus somnolence, at the next dose level. Such effects may be due to metabolic events distal to the targeted receptor site in the brain, for example, absorption by the liver. Some psychotropic drugs metabolize into other psychoactive compounds that are several times more potent than the parent drug (Lewis *et al.*, 1986). There is still much to be learned about the metabolism of most psychotropic drugs before we have a good

theoretical rationale for selecting a particular drug at the right dose for the appropriate person in a given setting for a particular occasion. The state of the science of neuropharmacology is advancing rapidly in this direction.

Behavioral Issues

Rojahn and Schroeder (1983) have discussed extensively observational assessment strategies, coding systems, data analysis, and the assessment of accuracy and reliability of behavioral assessment in mental retardation. The present section will not repeat this general textbook information but only touch upon those issues specific to behavioral pharmacotherapy.

Types of Measures Used

Walls, Werner, Bacon, and Zane (1977) compiled a list of 166 different behavioral checklists used in mental retardation. Rapoport and Conners (1985) compiled over 75 instruments for use in pediatric psychopharmacology research. Although this is a very useful compendium, only a small subset of these measures has been used in behavioral pharmacotherapy with persons who have developmental disabilities. Fortunately, Aman and White (1986) have performed an excellent critical review of these instruments used to measure drug change in developmental disabilities, which is highly recommended for an in-depth coverage of this topic. The three major types of measures are direct behavioral observations, rating scale techniques, and measures of learning and cognition. Other less frequently used techniques are mechanical tranducers, for example, actometers, rockometers, and the like; observations in a structured clinic setting, for example, in a playroom or during IQ testing; and motor performance measures. More recently, standard neuropsychological test batteries have been used or developed that are aimed at specific hypothesized neuropsychological substrates affected by a particular drug in children with a particular diagnosis. Such batteries usually contain a mix of standardized rating scales, for example, the Conners Teachers Rating Scale (Conners, 1969) as a valid and reliable measure of intensity; direct observations in the natural environment, to assess clinical validity; laboratory learning and performance measures, to provide an index whose sensitivity can be manipulated; and checklists, for the negative side effects expected depending upon the drug used.

By far the most used and powerful rating scale with higher func-

tioning clients who have low severity disorders, like attention deficits and conduct disorders, is the Conners Teachers Rating Scales (Conners, 1969) and its several recent revisions. Until recently, there has been no comparable rating scale for lower functioning clients. Aman, Singh, Stewart, and Field (1985) have published a 58-item scale constructed similar to the Conners scale but for low-functioning clients, The Aberrant Behavior Checklist, which appears to have good psychometric properties. How powerful a measure it turns out to be will require its wider use in future research. But at present, it would appear that we now have some good standardized rating scales of maladaptive behavior, and the use of unstandardized *ad hoc* rating scales requires further justification. Some of the advantages of rating scales are that they are cheap, quick, and allow observers to aggregate observations over time and settings, so that they can be filled out by primary caregivers without much training, and are based on standardized normative data. Disadvantages tend to be a high degree of subjectivity, lower reliability across raters and time, and a susceptibility to rater set, "halo effects," repeated measures effects, and statistical regression effects.

Direct observational instruments tend to be developed empirically and tailored to the specific needs of a study. They have rarely been subjected to standardization. There is no single particularly popular observational instrument used in behavioral pharmacotherapy studies. Aman and White (1986) list the following advantages to direct observations: objectivity, repeated measurement, usually independent of the skills or compliance of the subject observed, tailored to the needs of the study. Disadvantages listed are labor intensiveness, requiring trained observers, lack of sensitivity, reactivity to observer presence. However, there is a considerable body of evidence (Kazdin, 1977) that observer presence usually habituates with repeated exposure to the observer.

Measures of learning and cognition in higher functioning clients have involved measures of sustained attention, for example, the Continuous Performance Test (Sykes, Douglas, & Morganstern, 1972); impulsivity, the Matching Familiar Figures Test (Kagan, 1966); short-term memory, Rie and Rie (1977); paired-associate learning, Helper, Wilcot, and Garfield (1963); and measures of intelligence or achievement, Gittelman-Klein and Klein (1976). Some of these measures, especially IQ and Achievement, have been used with lower functioning clients (Aman, 1984). Other measures have been work productivity (Christensen, 1975; Marholin, Touchette, & Stewart, 1979) and a number of operant tasks, for example fixed ratio performance, (Poling & Breuning, 1983); discrimination learning sets (White & Aman, 1985); delayed matching-to-sample (Wysocki, Fuqua, Davis, & Breuning, 1981); and

repeated acquisition of stimulus chains (Walker, 1982). By and large, the exquisitely sensitive laboratory measures of animal behavior pharmacology, such as repeated acquisition of stimulus chains, have made a slow entry into human studies. This area shows great promise for development in the future (Johnston, *et al.*, 1983; Poling, Picker, & Hall-Johnson, 1983).

The use of mechanical transducers, for example stabilometric cushion (Christensen, 1975) and rockometers (Hollis, 1968), have the advantages of objectivity and good quantifiability, but the disadvantage is that they require cooperation of the subject (see Pfadt & Tryon, 1983, for review). Observations in structured standard settings, for example, playroom with mother present or absent in a clinic setting, show promise as a rather uncontaminated measure, but their ecological validity in the developmentally disabled population remains to be established. The same is true of tests of motor performance (Aman & White, 1986).

Drug–Behavior Interactions

From a behavioral point of view, drugs may interact in several ways to affect learning and performance: They may act as discriminative stimuli, as in the phenomenon of *state-dependent learning* (Kinsbourne, 1985); they may affect the central nervous system directly, as do the major tranquilizers, tricyclic antidepressants, and central stimulants; they may act as reinforcing consequences, as in the case of addiction; they may potentiate behavior by modifying the effectiveness of behavioral contingencies, as in the case of analgesics, narcotic antagonists, and minor tranquilizers; or they may act in combinations of any or all of the mentioned ways. On the other hand, "schedule-induced" effects, drug effects that are modified by the schedule of reinforcement the subject is receiving, and base-rate dependency effects, that is, where the magnitude and direction of a drug effect is contingent upon the rate of the target behavior at baseline, also have been demonstrated in higher functioning children with methylphenidate (Hicks, Gualtieri, Mayo, Schroeder, & Lipton, 1984) and in lower functioning adults with haloperidol (Aman, Teehan, White, Turbott, & Vaithianathan, 1986). Drug–behavior interactions in both directions in human behavioral pharmacotherapy seem to be an especially fruitful direction for behavior analysis in developmental disabilities.

Compliance with Drug Regimens

Gadow (1982) has documented the difficulties of compliance with drug regimens in the public schools. We have had similar problems with

developmentally disabled persons in community residential facilities. Compliance problems are not nearly so bad in public residential facilities because they are more closely regulated by state and federal standards.

Reactivity

Whalen and Henker (1976) have poignantly demonstrated how children with attention deficits who are receiving methylphenidate falsely attribute some of their behaviors to taking the drug. Client reactivity has not been documented so well with lower functioning subjects. However, Schroeder and Gualtieri (1985) have documented that staff rate subjects who have tardive dyskinesia as having more serious behavior problems. Direct observations also show that staff offer more noncontingent positive attention to subjects who have been withdrawn from chronic neuroleptics irrespective of their appropriate behavior. Aman and Singh (1986) have described how bias for or against the use of psychotropic drugs has affected the interpretation of some drug studies.

Side Effects

Table 1 which has been abstracted from Werry (1978) summarizes the main short-term and long-term side effects for the most commonly used medications in persons with developmental disabilities. These effects are considered toxic to the degree that they interfere with or limit the subject's capacity to function within his or her setting or constitute a hazard to his or her physical well-being. It should be noted that the evidence for toxic effects is based mostly on observations of behavior. A clear theoretical model of neurotoxicity due to drug administration that has been supported by neurobiological evidence there is not really available. We know very little about the toxicokinetics of most of these drugs, the cumulative effects of chronic doses, or the persistence of their damage to the central nervous system except from symptoms that have been observed. Behavior toxicologists with sensitive measurement techniques, such as performance on operant reinforcement schedules, could make a substantial contribution to our knowledge in this area.

Theoretical Issues

By now it should be clear that it is no simple task to conduct good behavioral pharmacotherapy research in the developmentally disabled

Table 1. Potential Side Effects of Psychotropic Drugs Used with Children[a]

Psychotropic drugs	Potential side effects	
	Short-term	Long-term
Stimulants Sympathomimetic amines (and others)—amphetamine, dextroamphetamine, methamphetamine, methylphenidate, pemoline Xanthines—caffeine Acetylcholine analogs—deanol	Decreased appetite, loss of weight, abdominal pain, insomnia, headaches	Depressed growth rate, anorexia, drug dependence, cardiovascular effects
Antipsychotics (major tranquilizers) Phenothiazines—chlorpromazine, thioridazine, trifluoperazine Butyrophenones—haloperidol Thioxanthenes—thiothixene, chlorpothixene	Extrapyramidal effects, drowsiness, dry mouth, blurred vision, nasal congestion, constipation, urinary retention, behavior and mood	Tardive dyskinesia, weight gain, jaundice, lenticular opacities
Antidepressants Trycylics—imipramine, disipramine, amitriptyline	Dry mouth, drowsiness, lethargy, tremors, appetite disturbance, nausea, sweating, weight loss, seizure threshold lowered, cardiotoxicity	Depressed growth rate, cardiovascular effects, dyskinesia
Antianxiety and sedatives Barbiturates—phenobarbital Benzodiazepines—diazepam, oxazepam, chlordiazepoxide, nitrazepam Propanediols—meprobamate Antihistamines—diphenhydramine, hydroxyzine	Drowsiness, disinhibition, neurotoxic skin reactions, incoordination	Drug dependence, withdrawal symptoms, withdrawal seizures
Antimanics Lithium	Nausea, diarrhea, muscular weakness, tremor, blurred vision, drowsiness, polyuria, polydipsia	Hypothyroidism, diabeteslike syndromes

[a] Adapted from Werry, 1978.

population. The heterogeneity of the clients and the commitment to the labor-intensive effort required are a challenge to the resourcefulness of the investigators.

A major difficulty is that the practice of pharmacotherapy in this population is data-driven and not theory-driven. The choice of drug and dose is still based mostly on clinical experience or trial and error. Yet we know from animal research that several of these drugs have a well-understood neuropharmacological basis that may predict differential response to a drug.

There has been renewed interest in diagnosing mental illness such as schizophrenia (Eaton & Menolascino, 1982) and depression (Sovner & Hurley, 1983) in developmental disabilities. Behavior analysts have generally not espoused diagnostic classificatory schemes, but it now appears that such efforts may be useful for behavioral pharmacotherapy in developmental disabilities. Such efforts should be supported (Schroeder, 1985), in that they may lead us to more specifically testable neuropharmacological hypotheses.

Neuropharmacologically Based Models for Behavioral Pharmacotherapy

The need for neuropharmacologically based models for behavioral pharmacotherapy is growing. A good example is self-injurious behavior (SIB) in the Lesch-Nyhan syndrome. We will treat this example in detail for illustrative purposes. There are many other examples like it in the literature.

Recent interest in the biological bases of SIB (Baumeister, Frye, & Schroeder, 1984; Cataldo & Harris, 1982; Schroeder, 1984) suggest that Lesch-Nyhan syndrome might provide a neurobiological model for conceptualizing some types of SIB. Lesch-Nyhan syndrome is a sex-linked disorder of purine metabolism in which the child demonstrates spasticity, choreoathetosis, possible mental retardation, elevated urine uric acid, self-mutilation, and aggressive behaviors. Mutilation, especially biting of the oral structures and fingers, is most common; this mutilation does cause pain, and the child may beg for restraints to prevent further injury. Patients can cause such severe self-mutilation so that the mouth orifice is totally deformed or the fingers lost. Lesch-Nyhan syndrome represents one of the first conditions with a demonstrated biochemical defect in which very specific abnormal behaviors are described. Although the serum uric acid level may be elevated, replacement therapy by reduction of the uric acid level with administration of Allopurinol to the patient does not alter the neurologic or behavioral phenomena.

Thus, control of uric acid level is not a contributing factor in controlling the behavior.

Nyhan, Johnson, Kaufman, and Jones (1980) conceptualize self-biting in Lesch-Nyhan patients as the result of imbalance in serotonergic, dopaminergic, and adrenergic mechanisms in the brain. Nine patients were treated with 5-hydroxytryptophan, a serotonin precursor, in combination with a peripheral decarboxylase inhibitor, carbidopa, and with imipramine to prevent its rapid excretion. Most patients had a dramatic decrease in self-biting in which they could be left free of restraints for several hours for the first time in years. However, within 1 to 3 months, each patient developed tolerance to the drug, and the pharmacological effect could not be produced again even a year later. The effect apparently was not due to a serotonin deficiency that may have been corrected by 5-HTP but by the temporary restoration of balance between these interacting neurotransmitter systems.

In an even more dramatic demonstration, Lloyd *et al.* (1981), in a postmortem examination of the brains of three Lesch-Nyhan patients, found profound alterations in basal ganglia dopaminergic neurons. All indexes of dopamine function in these brain regions were decreased, resulting in an estimated 65% to 90% functional loss of dopamine terminals. Such a hugh loss would result in dopaminergic supersensitivity, a compensatory action caused by a long-term decrease in the availability of dopamine at its receptor sites. The result may be an increased responsiveness of the cells containing dopamine receptors to exogenous dopamine or dopaminergic drugs.

An animal research model (Breese *et al.*, 1984) suggests that a key factor for SIB may be selective neonatal dopamine depletion in the nigrostriatal pathways of the basal ganglia. Autopsy studies of adult Lesch-Nyhan patients also indicate a significant depletion of dopamine and increased serotonin in the striatum and related brain areas (Lloyd *et al.*, 1981), just as has been seen in rats in which brain dopaminergic fibers were destroyed when the animal was 3 days of age (Breese *et al.*, 1985). A study by Gualtieri, Breese, Schroeder, & Keppel (1986) suggests that the dopamine antagonist, fluphenazine, is effective with a wide variety of SIB topographies.

Breese *et al.* (1985) have proposed that *dopamine depletion at an early age is related to later production of SIB*, a symptom not seen in rats when the destruction occurred during adulthood. These workers found that if young rats (aged 5 days) with dopamine depletion, by administering 6-hydroxydopamine (6-OHDA), were later challenged with L-DOPA, self-biting resulted, whereas this did not occur with animals receiving the same lesion later in life. This result suggests that *later* severe chronic

SIB is related to *early* dopamine depletion. Such an interpretation is supported indirectly by prevalence studies of SIB (Baumeister & Rollings, 1976), which suggest that severity of SIB is related to early onset and chronicity.

The neonatal dopamine depletion hypothesis does not explain why replacement therapy with L-DOPA, a dopamine agonist, or treatment with thioridazine, haloperidol, and chlorpromazine, dopamine antagonists, fail to decrease SIB. Perhaps the reason is that adaptive changes occur in different populations of dopamine receptors. Two different classes of dopamine-binding sites or receptors, D1 receptors and D2 receptors, have been distinguished on the basis of their differential sensitivity to certain dopamine agonists and antagonists. According to current terminology, D-1 receptors are associated with dopamine sensitive adenylate cyclase and are sensitive to micromolar concentrations of butyrophenone neuroleptics. In contrast, D-2 receptors are not linked to dopamine-stimulated adenylate cyclase and are sensitive to nanomolar concentrations of most neuroleptics. The dopaminergic supersensitivity observed following unilateral destruction of the nigrostriatal pathway in adult rats has been linked to increased concentrations of D-2 receptors (Seeman, 1981), but little is known about the mechanisms responsible for supersensitivity in neonatally 6-OHDA-treated rats. Dopamine sensitive adenylate cyclase increases rapidly in the rat striatum between birth and 7 days of age, whereas there is little change in haloperidol binding during this time (Pardo, Creese, Burt, & Synder, 1977). This period of rapid development of D-1 receptors corresponds to the age at which 6-OHDA lesions of dopamine containing neurons increase susceptibility to SIB in the rat (Breese *et al.*, 1985).

Goldstein, Anderson, Reuben, and Dancis (1985) showed a dramatic effect of fluphenazine, a D1 and D2 receptor blocker, on a 20-month-old Lesch-Nyhan syndrome child, whereas it showed only a modest effect on a 15-year-old Lesch-Nyhan case. The explanation for failure of the drug for the second case was that it "may be the result of behavioral overlay after years of self-mutilation" (p. 339). Thus, a drug-by-environmental interaction is hypothesized to account for this apparent age-related effect. Recent clinical trials by our group (Gualtieri *et al.*, 1986) have shown dramatic effects of fluphenazine on SIB patients who do not have Lesch-Nyhan syndrome. This is in marked contrast to our previous work using D2 receptor blockers. Thioridazine and haloperidol showed inconsistent effects on SIB (Lewis *et al.*, 1986; Schroeder & Gualtieri, 1985). Such data implicate the D1 receptor in this response and suggest that the SIB is due to release of endogenous dopamine.

The choice of fluphenazine over haloperidol, a neuroleptic with a

very similar behavioral profile, would not have been made without the neuropharmacologically based theoretical hypothesis. For years, clinicians have been experimenting unsuccessfully with neuroleptics to control SIB. An overly strong behavioral bias has likely caused behavior analysts in this field to overlook potent biological analyses that could have prevented much fruitless searching and advanced our understanding of the disorder. The original paper on Lesch-Nyhan syndrome (Lesch & Nyhan, 1964) had actually been published *before* the early pioneering paper of Lovaas, Freitag, Gold, and Kassorla (1965) on behavior modification of SIB, to which most modern day behavioral analyses of SIB can be traced. As Cataldo and Harris (1982) have noted, it is time for behavioral analysts to become more sophisticated in recent developments in the neurosciences and to be aware of the biological bases of the phenomena with which they are dealing.

ACKNOWLEDGMENT. I wish to acknowledge Grants HD 03110, HD 20980, MCH Project 916 for support during the writing of this chapter.

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CHAPTER 10

Pharmacotherapy in Developmental Disabilities

Methodological Issues

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It is a pleasure to discuss a chapter that so admirably sets forth the major issues in behavioral assessment of pharmacotherapy with the developmentally disabled. Schroeder (Chapter 9) presents, in highly compact form, a number of insights that have been painstakingly achieved during the relatively brief history of pharmacotherapy of the developmentally disabled child. The chapter highlights the importance of bridging the gaps between the "two cultures" of behavior and neuropharmacology, at both the methodologic and conceptual levels. The interplay between the behaviorally oriented clinician, the basic neuropharmacologist and neurobiologist, the animal pharmacologist, and the child psychiatrist is evident in this chapter and undoubtedly reflects a point of view nurtured by the interaction of these disciplines in a clinical-academic setting. The paper provides a useful outline of both methodological and conceptual issues, some of which I will consider further.

The choice of experimental design for studying drug–behavior interactions is sometimes seen as a contest between single-case designs—primarily the realm of the behavioral psychologist—and group designs that have been more common in psychiatric drug testing programs.

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Apart from the parochialism associated with traditions within professional disciplines, the scientific issues here are complex, and the reader is referred to the excellent discussion of these issues in the volume edited by Kazdin and Tuma (1982). One of the dilemmas can be seen in Schroeder's statement that the external validity of single-case designs "must come through replication across subjects." However, when one replicates a single-case design across subjects, it then takes on the rules of inference associated with group designs. One resolution of the dilemma is to accept the fact that replication is relevant to the problem of generalization to a *class* of subjects, an entirely different issue than the question of whether a drug has a causal role in removing symptoms in a *particular* patient. Although the latter is still a problem of scientific inference and therefore requires appropriate controls, the internal validity of the experiment does not stand or fall on the number of cases involved. The purview of the scientist may flexibly shift between highly limited and specific domains of phenomena (e.g., a single child), where the question is something like, "Does this drug influence this behavior in this particular child?", to the domain of processes and behaviors associated with many children who fall under a given class (e.g., children with Down's syndrome). The single case may provide hypotheses for the group design or merely be the basis of a replication from another single case. The reverse process, though painfully absent in the literature, is a meaningful one, of testing out generalizations derived from group studies on particular individuals within the larger group.

It may also be useful to consider single-case designs and group designs as a continuum rather than a dichotomy. For example, we pointed out elsewhere (Conners & Wells, 1982) that the multiple-baseline-across-subjects design (or "staggered start" approach), which avoids the problems associated with reversal designs, has been underutilized in psychopharmacological research. In this design, several subjects are given a treatment following a baseline measurement, but in staggered order, thus allowing the inference that changes in behavior occur in a time-locked fashion to the onset of treatment. But what happens in such a design when the effect is true of only 3 of 4 patients? Or 2 of 6? It is obvious that one can take two stances. Either one begins to invoke the binomial theorem and calculate the associated probabilities or views each case as a valid experiment in its own right, with the effect being true (i.e., nonchance) in some individuals and not true in others. Both approaches are valid but for entirely different goals. Such experiments represent neither single-case studies nor group studies but an intermediate category with some advantages and disadvantages from the two extremes of the spectrum. This design, incidentally, would ap-

pear to be particularly appropriate for the study of developmentally disabled children sharing a common classroom, where withholding of a treatment may not be possible and where the introduction of a novel treatment might be expected to generate social carryover effects.

As pointed out by Schroeder, variations in dose response between children of the same age and apparent type of disorder, as well as variations within children across different classes of behavior, are familiar and vexing issues. Although there are indeed a number of possible metabolic reasons for such wide variation, one explanation that has received little attention has to do with the measuring instruments used to define dose-response functions. For example, the inference that stimulants affect learning and social behaviors at different dose levels (Sprague & Sleator, 1977) is based upon the presumption that teacher ratings (social behavior) are as equally reliable and drug-sensitive as cued recall in the Sternberg paradigm. Not only are these measures different in reliability, thus affecting the ease with which drug effects might be detected at a given dose, but they also are quite different in the ease with which they can be precisely aligned with the peak of drug action.

Time-action effects may be a more significant problem than dose effects, particularly with short-acting stimulants. We have shown (Solanto & Conners, 1982) that type of task, task difficulty, dose, and time all interact with respect to autonomic, motor, and cognitive behaviors measured concurrently. Indeed, Pelham (1983) has made a convincing case that the entire literature on the effect of stimulants on academic behaviors is flawed by failure to attend to such issues as time-action curves and lack of drug-sensitive measures. Academic behaviors are, of course, among the most important targets for pharmacotherapy of developmentally disabled children, both in terms of measures of progress and possible interfering side effects. Careful definition of target behaviors, work products, and subskills, with close attention to the time and dose-response parameters of drug therapy, are mandatory in children being treated within educational settings. Here also, a carefully thought-out single-case design may be the most sensible way for *clinical* management of multiple behavioral and academic problems (see Wells, Conners, Imber, & Delamater, 1981, for an example of clinical decision making in the context of single-case study on an inpatient service).

The most important contribution of Schroeder's chapter is a clear statement of the need for theoretically guided psychopharmacologic studies. He suggests that one way to help achieve this end is to pay greater attention to diagnosis, presumably because diagnoses contain "surplus meaning" in terms of etiology or mechanism. In this respect, the recent efforts of DSM-III (*Diagnostic and Statistical Manual of the*

American Psychiatric Association, 3rd ed.) to refine diagnostic categories for children are welcome. For example, the search for continuities between adult diagnoses where important pharmacologic treatments have emerged (such as the use of lithium in manic-depressive disorders) has inspired a renewed interest in major depressive disorders in children and the application of antidepressant drug therapies.

On the other hand, DSM-III diagnoses, in addition to considerable problems with reliability (especially for subcategories), have been notably ineffectual in advancing childhood psychopharmacology so far (Rapoport, 1986). It may well be the case that the committee-based, nonempirical rules for diagnosis are actually a hindrance. Because the diagnostic rules are not operationalized, they leave open the possibility of using a wide variety of methods to define research samples, which may actually agree with each other in name only. For example, one response to DSM-III has been the further development of a variety of structured interviews, each of which differs in the specific content and methods of ascertainment (Orvaschel, 1986). The development of brief structured interviews in order to ascertain prevalence figures for mental disorders in children (Costello, Edelbrock, Dulcan, Kalas, Kessler, & Klaric, 1984) has fostered the notion among some researchers that information from parent and/or child interviews are valid as well as sufficient sources of information regarding diagnosis. This assumption is of course completely untested. The failure to align biologic (including pharmacologic) variables with diagnoses may thus represent a failure, not of theory, but of diagnostic paradigms. The whole question of the extent to which there are stable, etiologically unique syndromes of child behavior has perhaps been prejudged. In this respect, the alternative approach of studying specific *behaviors*, which is the hallmark of both animal and human behavioral neuropharmacology, may represent a more productive strategy, at least until there are empirically validated categories with clear operational guidelines.

In this regard, Schroeder's choice of self-injurious behavior (SIB) is instructive. Lesch-Nyhan patients represent a relatively well-defined disorder with a known genetic defect that shares with other disorders, such as childhood autism, a pattern of self-injurious behavior. The neuropharmacologic hypothesis for the treatment of SIB is based upon the specific behavior pattern, not upon the diagnosis of Lesch-Nyhan syndrome. Although having a specific diagnostic entity to test the hypothesis has a number of advantages, in terms of other parameters of experimental investigation, the choice of a dopamine D1 blocker to regulate SIB has implications beyond the specific diagnostic entity with which SIB is perhaps most dramatically associated. There may be other im-

portant diagnostic entities in which D1 dopamine receptors are defective and that share the pattern of SIB with Lesch-Nyhan patients. Similarly, drugs to treat aggressive, antisocial behavior may eventually be better understood in terms of some common neurobiologic abnormality (such as defective serotonin metabolism) without regard to the "diagnosis" of such categories as unsocialized aggressive or socialized aggressive conduct disorder. Naturally, this is not a call for a step backward to the state of psychiatric research before any valid diagnostic categories were defined, but it is rather a caution that further progress must rest upon empirically validated syndromes developed from clear operational rules.

As a researcher who has been identified with the use of global rating scales and a somewhat "dimensionalist" approach to the problem of identifying subject samples, I hasten to add that there are considerable dangers in *any* single method for selecting subjects for pharmacologic study, whether by the use of rating scales, mechanical devices, or behavioral time sampling or interval sampling. Method variance has remained one of the largest sources of error in almost all multimethod studies. In this respect, Schroeder's caution that multimethod studies should be employed with appropriate controls for Type I errors is very well taken.

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CHAPTER 11

The Role of Neuropsychological Assessment in Behavioral Medicine with the Developmentally Disabled

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As behavioral treatment procedures gain acceptance as viable contributors to medical and rehabilitative care, behavioral psychologists are becoming increasingly involved in the evaluation and treatment of children and adults suffering from medical and neurological disorders (Pinkerton, Hughes, & Wenrich, 1982; Russo & Varni, 1982). Behavioral interventions are now being used to alleviate various conditions once regarded as involuntary, organic, and under the exclusive domain of the medical profession. Behavioral medicine as a technology has developed means by which individuals can modify their physiological functions. Consequently, the recent growth of behavioral medicine and behavioral assessment methods has focused on both environmental and organismic variables.

At the same time, the role of neuropsychology is increasingly being recognized as important in the development and implementation of behavioral management programs with a brain-injured population (Horton & Miller, 1985). For example, research and practice in clinical neuropsychology (Diller, Buxbaum, & Chiotelis, 1972; Diller & Gordon, 1981; Golden, 1981; Ince, 1980) has emphasized various behavioral strategies

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when working with the neurologically impaired. These authors have acknowledged the importance of considering both the environmental events that sustain and maintain problematic behaviors in organically impaired individuals as well as the cognitive/behavioral consequences of their cerebral injuries. Clinical neuropsychologists have been active in studying the behavioral expression of localized and diffuse brain dysfunction for many years and more recently in the planning and implementation of effective rehabilitation/treatment programs. Those clinicians with training and experience in both behavioral therapy and clinical neuropsychology have begun to see the value of integrating these two approaches. Wood (1982) has described the importance of utilizing data from neuropsychological assessment when developing rehabilitation programs with a closed head-injury population, and Horton and Wedding (1984) have similarly viewed the integration of these two approaches as crucial when dealing with a brain-injured population. Professional groups have been formed in an attempt to interface these two approaches. Horton (1979) offers the following definition of behavioral neuropsychology, a new special interest group established by the Association for the Advancement of Behavior Therapy (AABT):

> Essentially, Behavioral Neuropsychology may be defined as the application of behavior therapy techniques to problems of organically impaired individuals while using neuropsychological assessment and intervention perspective. This treatment methodology suggests that inclusion of data from neuropsychological assessment strategies would be helpful in the formulation of hypotheses regarding antecedent conditions (external and internal) for observed phenomena of psychopathology. That is, a neuropsychological perspective will significantly enhance the ability of the behavior therapist to make accurate discriminations as to the etiology of patient behaviors. Moreover, the formulation of a cogent plan of therapeutic intervention and its skillful implementation could, in certain cases, be facilitated by an analysis of behavior deficits implicating impairment of higher cortical functioning. (Horton, 1979, p. 20)

The clinical alliance between behavioral theory and neuropsychology has slowly gained acceptance. Historically, however, early behavioral psychologists emphasized only observable stimulus response relationships and rejected the need to explore underlying psychological processes or physiological mechanisms (Skinner, 1950). They were concerned with objective and observable events and rejected mentalistic formulations that were considered empirically untestable (Hefferline, 1962). In contrast, Kanfer and Saslow's (1969) SORKC model of behavioral assessment (stimulus, organismic, response, contingencies of reinforcement, and consequences) has offered an alternative conceptual approach that stresses the importance of considering the organismic (0)

status of the individual along with those environmental contingencies that serve to maintain and sustain behavioral responses. Organismic variables from this perspective include autonomic activities, organic conditions, as well as central nervous system functioning. The importance of the central nervous system in determining the development and expression of psychological and physiological functioning has long been recognized. Consequently, behavioral assessment procedures may not be complete or properly understood unless the involvement of specific brain system functions are considered, particularly in the treatment of physical and developmental disabilities (Melin, Sjoden, & James, 1983). For example, neuropsychological deficits associated with a disability can affect one's response to antecedent events by eliciting or precipitating various target behaviors, or affect consequent events by influencing the effectiveness of various reinforcers or the behaviors associated with attaining them. Unfortunately, organismic or neuropsychological variables have frequently been neglected in traditional behavioral assessments, despite the fact that they can exert a significant influence on behavior (Cuvo & Davis, 1983; Mash & Terdal, 1981; Nelson & Hayes, 1979). In this regard, the status of cognitive or neuropsychological functions can be central to behavioral assessment procedures with the developmentally disabled by providing necessary information pertaining to the application of various intervention strategies. The purpose of this chapter is therefore to illustrate how neuropsychological assessments can contribute to the planning, implementation, and evaluation of various behavioral medicine treatment programs with the developmentally disabled.

Neuropsychological Assessment

Lezak (1983) defines *clinical neuropsychology* as an applied science that is concerned with the behavioral expression of brain dysfunction. Simply stated, it is the study of brain–behavior relationships. Neuropsychological assessment techniques are utilized to provide valuable information regarding the functional organization of psychological processes in the brain through extensive assessment of language, perception, memory, learning, problem solving, and adaptation. From this assessment, it is then possible to assist in the diagnosis of neurological conditions, establish the current cognitive status of an individual whose neurological diagnosis has already been verified, design and coordinate remedial/rehabilitation treatment programs, and monitor progress from

treatment and/or recovery (Walsh, 1978). In this sense, neuropsychological techniques can provide unique and comprehensive information about an individual's cognitive functioning that surpasses the range and extent of routine psychometric assessments of intelligence alone.

Early neuropsychologists conceptualized brain damage as a unitary homogeneous entity and attempted to differentiate organic conditions from other functional impairments using single tests of "brain damage." However, with the growth of research in neuropsychology, it became increasingly clear that various neurological disorders presented with very different symptoms and sequelae. Cognitive deficits after brain damage are diverse and represent a complex interaction between premorbid cerebral organization of brain functions, brain development, and the site of lesion or damage (Chadwick & Rutter, 1983; Goodglass & Kaplan, 1979). Consequently, the notion that one measure can distinguish those with and without brain damage was generally abandoned, and neuropsychological assessment procedures were developed that included a battery of tests designed to assess a wide range of brain functions and cognitive abilities (Reitan & Davison, 1974).

The Halstead-Reitan Neuropsychological Battery emerged as a quantitative approach to neuropsychological assessment and exemplifies the most popular and most utilized quantitative neuropsychological test battery in the United States. It is comprised of a group of related tasks that rely on scores, derived indexes, and the relationship of numerical scores to known patterns of diagnostic categories. The core tests and allied procedures from this battery include the category test, the tactual performance test (TPT), the finger tapping test (FT), the speech sounds perception test, the rhythm test, the strength of grip test, the trail making test (TMT), the Reitan-Klove sensory perceptual examination, the Reitan-Indiana aphasia screening examination, and the lateral dominance test. The scores from these tests are combined to compute the Halstead Impairment index, which ranges from 0 to 1.0, with 0.5 and above indicating brain damage. The major advantage of this approach is the standardization of scores that are valid and reliable and the ability to use this data in comparing diagnostic groups and in monitoring an individual's progress following treatment and/or recovery (Reitan & Davison, 1974). However, the derivation of an impairment index continued the basic premise of a "cutoff score" between brain-damaged and non-brain-damaged individuals (Walsh, 1985). As such, the major drawback of the quantitative approach is that it lacks specificity and does not differentiate precisely those cognitive functions that are responsible for performances on tasks that are complex and multidimensional (Lezak, 1983). For example, the TPT is considered one of the

most sensitive indicators of brain damage in the Halstead-Reitan Neuropsychological Battery. Essentially, the patient must integrate simple and complex tactile stimuli while blindfolded using primarily kinesthetic feedback and visual imagery. It requires a multitude of abilities for successful completion including motor-sensory integration, nonverbal problem-solving strategies, higher order concept formation, sustained attention and concentration, cognitive flexibility, and memory, without exhausting all the contributing elements. Patients who make 51 or more errors on this test are considered to be functioning in the "impaired range." However, it is important to note that low scores on this task may be due to disruption of any of the functions or combination of functions mentioned before. This highlights the shortcomings of neuropsychological methods that rely solely on quantitative information. Different cognitive abilities alone or in combination can affect test outcomes in different ways, making it difficult to determine which function or combination of functions contributed to lower scores on individual tasks. Walsh (1985) states that "there may be different determinents of failure in individual cases even where there is a comparable level of score" (p. 14). The insensitivity of quantitative data to detect individual differences created an interest in developing a more individualized approach to neuropsychological assessment.

A rather different use of neuropsychological tests emerged from the work of the Russian neuropsychologist, A. R. Luria, based on the qualitative aspects of an individual's test performance. This approach to neuropsychological assessment involves the theoretical understanding that the brain is comprised of many complex cognitive and behavioral functional systems that are made up of many elementary components or functions. In order to understand the cerebral organization of the brain or basic brain–behavior relationships, one must examine and identify the most discrete elements of functioning to account for failure or success on any complex multidimensional task (Luria, 1973). In this theory, every functional system requires the interactions or integration of many cerebral areas of the brain. These different areas then make separate and unique contributions to the overall system of cognitive functioning. When damage effects any part of the system, the entire system can be altered or changed. The goal of assessment from this perspective is not a quantitative measurement of abilities or deficits but a qualitative analysis and description of the patient's use of underlying cognitive components or abilities. Particular emphasis is paid to the qualitative aspects of how a task is performed and not only to the absolute level of performance. For example, patients who fail on constructional tasks are typically said to have visuospatial deficits that are associated pri-

marily with right hemispheric dysfunction. However, constructional abilities rely on a number of cognitive skills that involve a variety of brain system functions including perceptual, spatial, planning, and motor abilities. Patients with right hemispheric dysfunction often employ a part-oriented approach to constructional material and demonstrate little appreciation of the contour or organizing principles of the figural information that they are copying. In contrast, patients with left hemispheric disorders appear to have difficulty appreciating the individual elements of a design despite their ability to preserve the major configurational features. Those patients with frontal dysfunction tend to have difficulty with the analytic and problem-solving aspects of the solution, despite their preserved visuospatial abilities (Milberg, Hebben, & Kaplan, 1985; Walsh, 1985; Weintraub & Mesulam, 1985). By carefully observing the strategy employed by the patient while performing constructional tasks, valuable information can be gained that can implicate impairments in a variety of cortical and subcortical locations. As such, only by eliminating contributing functional processes to the execution of complex multidimensional tasks can an accurate and detailed description of an individual's cognitive deficits and strengths be understood.

The individually centered Boston Process Approach (Milberg *et al.*, 1985; Weintraub & Mesulam, 1985) exemplifies the qualitative method of neuropsychological assessment. It relies to some extent on formal psychometric approaches but emphasizes the need to accommodate the assessment to the patient's particular difficulty. The strategy of assessment is to conduct a broad general evaluation of cognitive functioning, identifying areas of difficulty and strength, and pursuing an analysis of these particular areas to confirm clinical hypotheses developed from early observations made in the assessment process. Many original test measures have been modified in this approach to facilitate the collection of data about an individual's cognitive state. However, these modifications do not interfere with the standard administration of the tests so that reference can be made, whenever possible, to established norms against which the patient's performance can be judged. For example, the Arithmetic subtest of the WISC-R or WAIS-R measures a patient's ability to perform basic mental calculations. However, a variety of contributing factors may impair a patient's performance on this task and must be appropriately ruled out to conclude a basic calculation deficit. This can be achieved by modifying the administration of the subtest but only after standard administrating methods have been met. A patient who cannot perform computations mentally may adequately solve the same problem when allowed to use paper and pencil. This suggests that

the failure on mental calculations was the result of impaired concentration and not an impairment in fundamental computational skills. In addition, patients with spatial difficulties may misalign numbers while working visually so that calculation errors are made even though the numerical operations employed are correct (Weintraub & Mesulam, 1985). Because many published tests provide insufficient comprehensive information about cognitive or behavioral functions, procedural modifications can be made to any neuropsychological test in the assessment process to investigate clinical hypotheses more comprehensively. For example some of the modifications, to name a few, include taking verbatim accounts of the patient's verbal answers on any verbal task; making detailed accounts of the patient's performance on visuospatial tasks; testing limits whenever possible by simplifying the demands on the patient's responses (such as offering the patient a multiple choice format from which to choose) after an established point of failure has been reached; using considerable encouragement to push the patient beyond minimal responses or consistent claims that they "don't know"; and expanding time limits on timed tasks to allow the patient additional time to complete the solution, and thus, examine separately the effects of response slowing or inertia from the actual loss or inability to perform the task (Milberg *et al.*, 1985). It is important to note that the qualitative approach not only attempts to localize and lateralize lesion sites but attempts to understand the individual's cognitive style from an information-processing point of view. This kind of analysis can be related to specific areas of cerebral dysfunction but can also facilitate the planning, implementation, and treatment of the patient.

The qualitative and quantitative methods of neuropsychological assessment represent two divergent views within the field. However, the integration of these approaches can provide two important components of contribution to the whole evaluation process. Precise quantification of functioning can be used as a baseline reference to assist in measuring progress during and after treatment, whereas descriptive observations can sort out the nature and pattern of specific cognitive deficits and strengths (Lezak, 1983).

Neuropsychological Assessment with the Developmentally Disabled

With the growing interest in utilizing neuropsychological assessment techniques with a variety of clinical populations, neuropsychol-

ogists are now using neuropsychological approaches with the developmentally disabled (Gordon, 1977; McCaffrey & Isaac, 1985; Slomka & Tarter, 1984). Although there are some inherent limitations with this approach, it does provide an interesting clinical framework from which to better understand how the functional integrity of the brain affects the performance of the developmentally disabled on different cognitive and behavioral tasks. Historically, the diagnosis and classification of mental retardation has been based on the medical model that primarily emphasized the etiological determinants of the syndrome. Although there was little agreement about the etiology of retardation because of the variations in causal factors, the classification system generally represented some form of central nervous system dysfunction as being responsible for the condition. Within this context, traditional psychometric testing was used by psychologists for educational planning, placement, and classification. The concept of mental retardation, much like neuropsychology's early concept of brain damage, was based on the belief that it was a homogeneous entity, regardless of etiology, and, as such, psychologists working with the developmentally disabled primarily relied on single tests of general intelligence in an attempt to identify and classify the mentally retarded (Boll & Barth, 1981). However, the classification system and assessment process had little utility for treatment. More recently, the classification and definition of mental retardation has shifted to a more behavioral perspective that emphasizes the functional capacities of the individual regardless of the biological or etiological determinants. The proper diagnosis of mental retardation now requires the patient to evince subaverage intellectual functioning as well as impaired adaptive behavior, with onset before 18 years of age (Cuvo & Davis, 1983; Grossman, 1977). Subaverage intelligence is defined as an IQ score of more than two standard deviations below the mean on standardized tests of intelligence, whereas adaptive behavior is measured less formally by adaptive behavior scales and is best used for planning and programming rather than classification (Lesak, 1983). Although testing of intelligence and adaptive behavior are necessary for the diagnosis of mental retardation, the information obtained can still be of limited value for developing treatment programs. Tests of general intelligence may serve as quantitative indexes of a patient's intellectual functioning, but, like single tests of brain damage, may not be sensitive enough to differentiate specific cognitive deficits and strengths in individuals who are mentally compromised. In this sense, individuals with similar IQ scores may have little more in common than the test scores themselves (Boll & Barth, 1981). Measures of adaptive behavior, on the other hand, are more functional for behavioral programming, but the number and

range of items within each behavioral domain are often too restrictive to be useful for developing appropriate treatment programs (Cuvo & Davis, 1983). Moreover, the items are often chosen, not for their programmatic value, but for their ability to discriminate between age groups. Because neuropsychology is the study of brain–behavior relationships, it might provide a different and more useful perspective to the current diagnostic and classificatory system of mental retardation by considering and integrating both the medical and behavioral models. To the extent that neuropsychological procedures measure cerebral pathology and a wider range of cognitive and behavioral functions than routine tests of intelligence or adaptive behaviors, they can be useful for describing the specific functional capacities of the mentally retarded as well as the involvement of central nervous system dysfunction, and in doing so, provide more comprehensive information for rehabilitation and/or treatment.

The application of neuropsychological techniques to the assessment of the mentally retarded can provide more valid and reliable descriptive information regarding specific cognitive deficits and strengths than most traditional psychometric measures. In fact, psychometric tests of intelligence may at times represent an inaccurate or incomplete picture of the patient's cognitive functioning. For example, patient's suffering from decreased attention and/or low motivation may demonstrate significant deficits on virtually all of subtests of the WISC-R or WAIS-R that inevitably would result in an overall lowered IQ. However, it would be a mistake to conclude that these patients are suffering from multiple bilateral lesions or diffuse cognitive deficits based simply on the pattern of inter- and intrasubtest scatter. Similarly, individuals suffering from neuromuscular disorders will inevitably perform poorly on the timed performance subtests of the WISC-R or WAIS-R, resulting in a lowered Performance IQ and the possible misinterpretation of a primary perceptual motor deficit. As mentioned previously, neuropsychological assessment procedures afford the opportunity to identify and specify those cognitive factors that are responsible for success and failure on individual tasks. Thus, although psychometric approaches may demonstrate cognitive strengths and weaknesses from subaverage IQ scores and low subtest scores, neuropsychological testing can determine the specific perceptual, attentional, memory, language, and motor/sensory functions that are responsible for the patient's overall level of cognitive functioning (Slomka & Tarter, 1984).

Unfortunately, little direct research has been conducted on the applicability of neuropsychological assessment procedures with the developmentally disabled. Despite the theoretical rationale for conducting

such research, the application of these procedures with the mentally retarded is quite limited. Disturbances of gross and fine motor activity and coordination, imperceptions of sensory-perceptual material, deficits in the production and comprehension of oral and written language, impairments of attention and concentration, dysfunction of higher level cognitive skills, disinhibition in interpersonal relations, poor impulse control, and problems with encoding, storing, and retrieving new information have all been reported to varying degrees (Arnold, Yule, & Martin, 1985; Benton, 1970; Detterman, 1979; McCaffrey & Isaac, 1985; Teeter, 1983; Thompson & O'Quinn, 1979). However, although retardation may be associated with a number of behavioral patterns and cognitive inefficiencies, there are considerable individual differences that may manifest themselves differently across diagnostic and etiological categories. McCaffrey and Isaac (1985), in a study of adults who are mentally retarded, suggest that signs of frontal lobe dysfunction may be the common denominator in all forms of mild to moderate mental retardation. Intact frontal lobe functioning is important for the organization of ongoing behavioral responses and the overall operation of higher level cognitive skills (Luria, 1973). It is not surprising that if frontal lobe functioning is impaired, deficits in the planning, regulation, and execution of simple behavioral tasks would be observed. In contrast, for the severely and profoundly retarded or multiply handicapped individual, a complete neuropsychological evaluation may not be feasible (Slomka & Tarter, 1984). In these individuals, it may not be possible to identify selective deficits within the context of severely depressed performances across modalities. Severe and profound mental retardation is characterized by widespread and diffuse neurophysiological dysfunction that may preclude the identification of specific or discrete cognitive impairments. Consequently, under these circumstances, a traditional behavioral or cognitive assessment may be the only way to obtain valuable information regarding the individual's specific needs and capacities. Despite these limitations, there continues to be a need for systematic comparative studies of cognitive functioning in the developmentally disabled in which specific neuropsychological skills are assessed rather than the global, ill-defined abilities evaluated by intelligence tests alone.

Integration of Neuropsychology and Behavioral Medicine

The role of clinical neuropsychological assessment is to provide accurate and reliable descriptive information regarding brain–behavior re-

lationships for the purpose of specifying behaviors that may serve as appropriate targets in academic remediation and/or treatment (Rourke, Bakker, Fisk, & Strang 1983; Slomka & Tater, 1984). Behavioral medicine, on the other hand, focuses on the employment of behavioral techniques in the therapeutic intervention and/or amelioration of problems associated with health and illness (Pinkerton *et al.*, 1982; Russo & Varni, 1982). In this regard, from a behavioral perspective, physiological, behavioral, as well as environmental factors need to be considered in appreciating the etiology and maintenance of various medical disorders. As such, neuropsychological assessment techniques can form a productive alliance with behavioral medicine in the treatment of the developmentally disabled by contributing to the planning, implementation, and evaluation of individual treatment programs.

The assessment of an individual's neuropsychological strengths and weaknesses can provide important information pertaining to the therapeutic plan by determining the methods of treatment that might be employed to circumvent a particular deficit or by identifying those sensory-processing modalities that appear most intact. As mentioned previously, therapeutic strategies and techniques need to be adapted to suit the specific needs and capacities of the developmentally disabled individual. Such information may also be of value to other members of the treatment team (i.e., physicians, nurses, therapists, and teachers) in that they may use this information to modify their approach to the patient based on their knowledge of his or her neuropsychological strengths and weaknesses. For example, the individual who exhibits subtle impairments to language functioning will probably not respond well to verbal instructions or rewards, whereas individuals with deficits within the visuospatial realm may benefit from procedures and directions that are presented in a highly verbal and concrete fashion (Rouke, *et al.*, 1983). The effectiveness of various behavioral treatment programs can be enhanced if they include procedures that utilize the patient's neuropsychological strengths to compensate for his or her deficiencies.

Before formulating a treatment plan, neuropsychological assessment techniques can be used to determine the modality of treatment from which the patient can best profit. Several neuropsychological functions should be considered in this regard: (a) patients ability to sustain, select, and divide their attention visually and auditorily, (b) their ability to perceive, scan, organize and integrate visuospatial information, (c) their capacity to perceive, decode, and express linguistic information, (d) their capacity to form concepts, to generate problem-solving strategies, to discern cause and effect relationships, and to benefit from positive and negative feedback, and (e) their ability to encode, store, and

retrieve new information effectively. With the data generated from a complete neuropsychological assessment, therapies can then be adjusted within the limits of the developmentally disabled's cognitive strengths and weaknesses. For example, a patient's inability to sustain and maintain attention for appreciable periods of time can hinder the effectiveness and benefit of any therapeutic program. Consequently, therapy might be more profitable when treatment is conducted in minimally distracting environments where sessions are short but frequent. In addition, because the ability to divide one's attention requires processing information from multiple inputs, the functional integrety of both visual and auditory modalities should be assessed. Patients often respond to one sensory mode better than others, and the presentation of instructions and procedures can be modified to accommodate the patient's perceptive strengths and weaknesses. Language-impaired patients may maximally benefit from therapy that utilizes visual stimuli, whereas visually impaired patients may show greatest improvement when information is presented via auditory channels. When considering memory dysfunction, it is useful to distinguish encoding and storage difficulties from problems with retrieval, although impairments at any of these levels can result in poor memory skills. The pairing of new information with overlearned, familiar concepts can enhance the encoding of material necessary for the implementation of various treatment procedures. Additionally, frequent cuing, repetition, and rehearsal can improve one's ability to store and recall new information, whereas presenting instructions that must be remembered as part of the therapeutic regimen in a multiple choice or recognition format may facilitate retrieval of instructions and procedures that are important to the therapeutic process. Finally, visual imagery can be used as a compensatory technique with patients suffering from memory difficulties associated with their dysfunctional verbal/language skills. Considerable research will be needed to determine the effectiveness of neuropsychological assessments in distinguishing the optimal modalities or conditions for learning as well as their specific therapeutic applications with the developmentally disabled.

Biofeedback and relaxation training have frequently been used in various combinations to treat a variety of health-related problems (Pinkerton *et al.*, 1982). Biofeedback is a therapeutic procedure wherein one can learn to modify or control one's own autonomic activity through electronic information about his or her physiological responses, whereas relaxation training is generally used to achieve a lowered state of arousal. Biofeedback is often used in conjunction with other behavioral techniques, including relaxation training, assertiveness training, systematic

desensitization, self-monitoring procedures, and instruction in coping strategies (Masek, Spirito, & Fentress, 1984). However, research on the application of biofeedback procedures and relaxation techniques with the mentally retarded has been extremely limited. Several studies that have attempted to utilize relaxation training with the mentally retarded concluded that these individuals were able to learn to relax without undue difficulty (Harvey, Karan, Bhargava, & Morehouse, 1978; Peck, 1977). Unfortunately, these studies utilized multifaceted treatment programs, making it difficult to isolate the effects of relaxation on the patient's behavioral gain. In addition, a number of problems associated with the relaxation treatment were identified, such as communicating relaxation states via hand signals, learning the order of practice for muscle groups, and remembering to relax before imaging. Although it has been common to assume that mentally retarded individuals are more difficult to teach biofeedback and relaxation than a normal IQ population, neuropsychological assessment techniques can be used to help determine which procedure or combination of procedures would be most efficacious. For example, if a developmentally disabled individual experiences difficulty with both visual and auditory perception, essentially two sources of input have been eliminated. Feedback might then be more helpful in this situation if it is provided to other sensory fields. In other cases, various aids can be used (i.e., tape recorders or portable biofeedback equipment) to compensate for memory deficits that might interfere with home practice or learning relaxation skills. Additionally, attentionally disordered patients might benefit more from concrete, step-by-step approaches to relaxation, such as progressive muscle relaxation, rather than passive imagery techniques that can be self-distracting (Masek *et al.*, 1984). Some attentionally disordered patients may not find the typical biofeedback display inherently reinforcing or engaging, and modifications in the display can enhance the mentally retarded individual's interest and motivation. The exact procedures or combination of procedures will vary, according to specific patient characteristics and need. The specific nature of a patient's neuropsychological strengths and deficiencies can be very important when considering the therapeutic approach and mode of behavioral treatment.

Another important contribution of neuropsychological assessment techniques with the developmentally disabled is that of monitoring fluctuations in cognitive abilities over time. Quantitative data generated from neuropsychological assessment procedures can be used to detect change or trends associated with cognitive recovery or decline (Chadwick & Rutter, 1983). The relative sensitivity of neuropsychological measurements make them well suited for following the course of many neu-

rological conditions. One obvious example of the application of neuropsychological procedures in monitoring cognitive change in patients with neurological or medical disorders comes from research with epilepsy. Epilepsy is a complex neurological condition that is characterized by recurrent disturbances of cerebral electrical activity that can result in a variety of intellectual, perceptual, and affective disorders. It is not a unitary impairment but is rather diverse in character in terms of etiology, frequency, and types of behavioral manifestations. Hartlage and Telzrow (1984) report epileptic seizures represent abnormal brain functioning that may exist independently of some underlying organic process or can result from a variety of neurological conditions (lesions, trauma, perinatal injuries, infections, and metabolic disorders). Developmental disabilities and epilepsy are often related and frequently occur in the same individual (Slomka & Tarter, 1984). The incidence of mental retardation is three to four times higher in patients with seizure disorders than in the general population (Thompson & O'Quinn, 1979). Whitehouse (1971) reported 57% of those with epilepsy have an IQ of 89 or below. Repeated and uncontrolled seizure activity has been associated with progressive neurological and cognitive decline (Dodson, Prensky, Devivo, Goldring, & Dodge, 1976). Continuous poorly controlled seizures can present many problems for the developmentally disabled epileptic, including mental deterioration, physical injury from falling, and dulled alertness (Detterman, 1979). In addition, lapses of attention and concentration may be due to subclinical minor seizures that are not overtly obvious. Unless the person is monitired continually by EEG, the seizure activity may go unnoticed. Periodic assessment of neuropsychological functions can be used under these circumstances as an adjunct to traditional medical care by not only identifying areas of cerebral dysfunction but by monitoring cognitive changes over time. Reevaluation at regular intervals of those individuals with seizure-related cognitive deficits can provide considerable information to the physician that may assist in the diagnosis and decision to implement and/or evaluate the medication regimen.

Repeated neuropsychological testing at regular intervals cannot only provide important information regarding a patient's changing neurological condition but can monitor his or her response to various medical/behavioral interventions as well. For example, a relatively large number of developmentally disabled individuals are being maintained on one or more anticonvulsant medications for prophylactic and/or therapeutic reasons (Hartlage, 1981). Although anticonvulsant medications have been most helpful in treating various seizure disorders, toxic levels of these drugs can impair a variety of cognitive functions (Mathews &

Harley, 1975). New techniques for determining serum anticonvulsant levels, such as gas-liquid chromatography, have greatly improved the ability of physicians to detect the toxic effects of seizure control medications (Hartlage & Telzrow, 1984). However, the effects of anticonvulsant medication on cognitive functioning can be more reliably monitored, utilizing the quantitative aspects of neuropsychological assessment techniques. Lezak (1983) reports that neuropsychological data are probably the most sensitive indexes of the extent to which medications enhance or compromise a patient's mental efficiency. Tests involving fine motor control and integration, reaction time, visual and/or auditory attention and concentration, mental flexibility, and short-term memory have been shown to vary as a function of serum levels of anticonvulsant medication (Hartlage, 1981; Novelly, Schwartz, Mattson, & Cramer, 1986). Additionally, dramatic improvements on these cognitive measures have been reported when medication was initiated with patients who had previously had an undiagnosed seizure disorder (Rourke, Bakker, Fisk, & Strang 1983). This highlights the need to detect various neuropsychological changes in terms of recovery or deterioration and to reevaluate at regular intervals those individuals whose medication regimen may contribute to a variety of changes in their cognitive functions. In this regard, information gained from neuropsychological assessments can be integrated with traditional medical monitoring techniques, thereby providing a model of interdisciplinary collaboration in the diagnosis and treatment of medical/neurological disorders. Continued research is needed to study the effects of anticonvulsant medication on various neuropsychological measures and to determine their effects on new learning.

Conclusions

The integration of behavioral and neuropsychological data can provide important information regarding the treatment of the developmentally disabled. Neuropsychological assessment techniques can be used as part of the diagnostic process and can help guide the methods and strategies of intervention that can be utilized with this population. The needs to differentiate various neuropsychological functions, to reevaluate at regular intervals, and to monitor cognitive changes over time have been highlighted. Cognitive dysfunction can contribute to a variety of behavioral and medical disorders, allowing neuropsychology and behavioral medicine to form a productive partnership aimed at incorpo-

rating information from both perspectives in formulating and implementing appropriate treatment plans.

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CHAPTER 12

The Utility of Neuropsychological Assessments of Mentally Retarded Individuals

Barry F. Skoff

The preceding chapter by Fischer provides a firm foundation for conceptualizing how neuropsychology can be integrated into behavioral treatment programs for developmentally disabled individuals. He describes current approaches to neuropsychological assessment and suggests a number of important ways in which neuropsychological procedures can be used in treatment planning, behavior management, and treatment monitoring and assessment. I would like to elaborate on some of Fischer's points, in particular the question of "organicity" and qualitative versus quantitative approaches to neuropsychological assessment, and then present a brain-based model of cognitive functioning. Finally, I would like to suggest some ways that neuropsychological assessments can be advantageously used to guide clinical, and in particular behavioral, interventions in mentally retarded individuals.

The Question of "Organicity"

Fischer discusses some of the ways neuropsychology has evolved and its more recent integration with behavioral psychology. Neuropsy-

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chology's beginnings were closely associated with neurology and the diagnosis of brain damage. This led to the question of "organicity": whether someone's behavior was secondary to his or her brain injury. We now recognize the limitations of this concept. For one thing, brain damage results from a variety of sources and expresses itself in a multitude of complex ways. Therefore a single test for brain damage or a simple diagnosis of organicity is no longer perceived as feasible or useful. The question of organicity also carried with it an implicit prescription for treatment: An environmentally determined behavior is susceptible to behavioral interventions, whereas an organically based one is not. In his article, Fischer points out how advances in behavioral medicine have altered this view. We now know that all complex behaviors are based in central nervous system functioning but are also shaped by environmental factors. Furthermore, even previously identified "organic" disorders, such as epilepsy, can be amenable to behavioral interventions. Despite these changing views, many professionals in other fields and caregivers who work with developmentally disabled individuals continue to see neuropsychology as merely an adjunct to neurological diagnoses and to view behavior as either organically based or environmentally determined.

The use of neuropsychology as merely a tool in the diagnosis of brain damage obscures its potentially more important contribution in understanding and working with mentally retarded individuals. Neuropsychological assessment can provide a much more comprehensive and useful description of a mentally retarded individual's cognitive functioning than traditional psychometric testing methods. Psychological reports dealing with mentally retarded clients are frequently criticized for not providing practical or relevant information. A recent survey of staff members working in community mental retardation programs found that they preferred data regarding clients' academic and vocational achievement and adaptive skills (VandeCreek & Smith, 1983). Such information was considered more valuable for habilitative planning than descriptions of clients' personality dynamics. Das's (1984) delineation of what good psychological testing should encompass suggests where traditional approaches fall short. He lists three components of a useful evaluation: (a) assessment of competence, (b) delineation of processes underlying competence, and (c) recommendation for remediation based on (a) and (b). Processes involve a particular method of doing something and are differentiated from abilities, which involve a particular skill or capacity. Traditional psychological testing, with its focus on level of functioning and emphasis on final score, only addresses competence. Such tests, and descriptors such as IQ, not only do not delineate un-

derlying processes, but, as Fischer and others (Milberg, Hebben, & Kaplan, 1986; Weintraub & Mesulam, 1985) point out, may even obscure them.

Qualitative versus Quantitative Approaches

Neuropsychological assessment, in particular the more qualitative approach with its emphasis on *how* a subject goes about doing a task (Christensen, 1975); Goodglass & Kaplan, 1979; Luria, 1966) is especially well suited to meet the three requirements of a good psychological evaluation described by Das. The process approach to neuropsychological assessment recognizes that any single test may involve multiple different abilities and processes, whereas, at the same time, different tests may utilize the same process (Das, 1984; Milberg *et al.*, 1986). A careful analysis of the individual's approach and the overall pattern of successes and failures can reveal not only level of functioning (i.e., competence) but underlying abilities and processes as well. As such, the information obtained will be more directly related to adaptive behavior and more easily translated into prescriptive recommendations concerning educational, vocational, or behavioral management programming.

In his article, Fischer points out that quantitative measures of neuropsychological functioning can be used to monitor and document the effects of various behavioral and psychopharmacological interventions and changes in CNS functioning over time. Unfortunately, appropriate quantitative neuropsychological test batteries for use with mentally retarded (MR) individuals are not currently available. The quantitative neuropsychological batteries that do exist were not developed for, or appropriately normed on, MR populations. Consequently, use of these batteries with MR individuals often result in homogeneously low scores, which are insufficient to reveal the individuals' cognitive profile. In addition, their inflexible administration particularly penalizes MR individuals and may result in incorrect conclusions regarding an individual's abilities and deficits (Delis & Kaplan, 1983; Spiers, 1981). The more qualitative approach, with its emphasis on individual differences and relative (i.e., personal) cognitive strengths and weaknesses, is thus better suited to an MR population. There is no reason, however, that qualitative and quantitative approaches cannot eventually be integrated (Goldberg & Costa, 1986; Goldberg, Bilder, & Jaeger, 1987).

A Neuropsychological Model

The neuropsychological approach discussed here clearly owes a lot to cognitive psychology and information-processing theory in its attempt to analyze complex skills and activities into more elementary component abilities and processes. But neuropsychological assessment is also grounded in our knowledge of the organization of the brain and brain–behavior relationships. Most current neuropsychological models of brain functioning differentiate information processing from executive functions (Lezack, 1983). The former, information processing or coding processes (Das, 1984), involve *what* someone can do, or *how much* they know. Executive functions, on the other hand, involve *how* (*when* and where) or *whether* a person goes about doing something. Coding and executive processes are interrelated and interdependent (Das, 1984). In Luria's (1973) model of neuropsychology, coding is the responsibility of occipitoparietal and frontotemporal areas, whereas the frontal region of the brain controls executive functions. Understanding and assessing coding processes and executive functions in mentally retarded individuals can better inform any clinical, and in particular, behavioral intervention.

Coding Processes

Das and his colleagues (Das, Kirby, & Jarman, 1975, 1979), based on Luria's (1973) work, have described two types of coding processes that are involved in receiving, analyzing, and synthesizing information and in its subsequent retrieval. Simultaneous integration involves the synthesis of elements into a coherent whole. Successive (or sequential) information processing involves temporally or successively ordered sequences. This delineation of two main styles of information processing is similar to the findings of other brain researchers, who use labels such as serial or time-dependent (i.e., successive) versus holistic, unitary, or time-independent (i.e., simultaneous) (Bogen, 1969; Springer & Deutsch, 1981). These two modes of processing information are not affected by the form of input—visual information may be processed successively, and auditory information can be processed simultaneously. Most complex, everyday tasks, such as reading, require the combination of both for efficient execution. The type of coding used or emphasized on any particular task according to Das (1984) depends on (a) an individual's competence and habitual preferred mode of processing, (b) an individual's training and experience, and (c) the specific task demands.

Assessing a mentally retarded individual's capacity for simultaneous versus successive processing may help to provide a more parsimonious explanation of his or her performance. Furthermore, understanding an individual's relative competence in, and use of, simultaneous and successive processing can lead to more specific recommendations for educational, vocational, and behavioral programming (Das *et al.*, 1979; Gunnison, 1984; Kaufman & Kaufman, 1983). For example, an appropriate question for a psychologist working with MR clients might be why an individual may be having difficulty learning a particular task, such as sweeping the floor, and how best to instruct the individual. Neuropsychological testing may reveal that the client has a relative deficit in simultaneous processing, which prevents him or her from assessing the cleanliness of the floor as a whole. At the same time, he or she may be able to learn to sweep in a systematic sequence such that the entire floor is eventually covered. An analysis of an individual's abilities and information-processing style as applied to a specific task bears a similarity to the behaviorists' "task analysis." Incorporating information from neuropsychological testing when formulating a task analysis is one way that neuropsychology might interface with behavioral psychology.

Executive Functions

Potentially more significant for understanding and working with mentally retarded individuals are the so-called executive processes that are necessary for organizing and using coding skills in order to accomplish a set goal or task. Executive functions are metacognitive in the sense that they operate on more elementary cognitive abilities (Bransford, Sherwood, Vye, & Rieser, 1986; Wong, 1986). As such, executive processes can be difficult to conceptualize, label, or assess because they are less concrete than, for example, one's language or visual-motor skills. One way of viewing executive functions is as those abilities necessary for goal formulation, planning, inhibition of impulsive responses, the establishing and maintaining of a specific problem-solving or mental response set, and the use of feedback from the environment and one's performance in order to shift sets when necessary, that is, self-monitoring and self-regulation (Lezack, 1983; Stuss & Benson, 1984).

The idea that it is deficits in these executive processes that distinguish mentally retarded individuals, more so than other processing deficits, is certainly not new, although it is not always stated as such.

Cruickshank and Qualtere (1950) spoke of lack of "autocritical attitude" as differentiating mentally retarded from normal children on tests of intelligence. A number of individuals have described deficits in strategy utilization and memory in the mentally retarded (Brown, 1974; Friedman, Krupski, Dawson, & Rosenberg, 1977; Kopp, Krakow, & Johnson, 1983). While back in the 1940s, Kounin (1941a,b; also see Budoff & Pagell, 1968) proposed the concept of "rigidity" to explain "feeble-mindedness." What distinguishes the current concept of "executive functions" is its basis in neuroscience and our better (though still far from optimal) ability to assess these functions on neuropsychological evaluations (Lezack, 1983; Goldberg *et al.*, 1987; Wong, 1987). In a more recent discussion of mental retardation and executive processes Das *et al.* (1979) commented that "the retarded child is backward in the decision-making and planning processes which determine how information is encoded and the task executed" (p. 92). A related conclusion was drawn by McCaffrey and Isaacs (1985) who found that impairments on neuropsychological tests presumed to assess frontal lobe systems (responsible for executive functions and planning in Luria's system) was the one commonality in a group of mentally retarded adults. We have recently found, in a group of mildly, moderately, and severely retarded adults, that as IQ level decreases, performance on tests of executive functioning deteriorates (Skoff & Son, 1987).

Executive functioning is closely related to performance in a wide variety of areas, due to the hierarchical nature of executive processes over more elementary abilities and skills. Thus impaired executive functioning can have far-reaching effects but can be overlooked or misinterpreted. For example, apparent perceptual deficits on psychological testing may actually reflect impaired executive processes (Skoff, 1986; Skoff & Furbish, 1986). Using a multiple-choice matching task, we presented a target figure (one of the Weschler Memory Scale (1974) Visual Reproduction Figures) at the top of a vertical array of five other figures. The subject's task was to choose the figure that matched the model at the top. We found that many mildly to moderately retarded adults failed at this task, a task similar to other nonmotor, nonmemory, nonverbal tasks that propose to assess "simple" perception. However, when the choices were presented separately, one by one alongside the model and the subjects asked whether they were the same or different, many of these same individuals succeeded. We concluded that what could have been interpreted as a perceptual deficit in fact was due to impaired executive functioning. This finding has important implications for educational and vocational recommendations. Such individuals may be capable of many tasks demanding perceptual skills but only if such tasks

are structured for them in such a way as to compensate for their poor self-monitoring and self-regulation.

An appreciation of the role of executive deficits in mental retardation may be vital in understanding many otherwise perplexing phenomena. Uneven or inconsistent performance often confuses and frustrates both the client and staff attempting to work with him or her. It is precisely the individual's ability to know how and when to use a particular skill that is affected by the executive deficits typically found in mentally retarded individuals and that frequently results in inconsistent performance. Understanding this may help staff and clients to formulate more realistic expectations and may lead to better ways of structuring the client's routines. A related problem involves mentally retarded individuals' frequent trouble in adapting to new situations and in shifting jobs or tasks within a job. Difficulty in shifting mental set is again an executive deficit, which is likely the result of impaired brain functioning (McCaffrey & Isaacs, 1985) and may not be alterable by training (Das *et al.*, 1979). In both these cases, it is suggested that, although many skills may be taught mentally retarded clients, they may only be consistently performed in familiar settings. Duncan (1986), in his discussion of patients with frontal lobe damage, notes that although their ability to use active, controlled processes in the organization of their behavior is impaired, there is a concomitant greater reliance on passive, automatic processes. He concludes that these individuals can be best taught new skills by the repetition of familiar sequences of action in familiar settings. A similar conclusion can be drawn about many mentally retarded individuals.

The concept of executive functions is clearly related to adaptive behavior and independent functioning. Lezack (1983, p. 507) states that executive functions "are necessary for appropriate, socially responsible, and effectively self-serving adult conduct." Snart, O'Grady, and Das (1982) suggest that one's ability to judge what is appropriate and inappropriate behavior and to assess the social context of a situation, as well as one's own needs and goals before responding, in essence, to plan before acting, are the basis for adaptive behavior. An individual's behavior is generally determined by a combination of environmental variables and "internal" processes. The more a person lacks the executive processes described here, the more he or she becomes a "slave to the environment." Indeed, Lhermitte (1986) has proposed that the executive deficits resulting from frontal lobe damage essentially result in an "environmental dependency syndrome."

This suggests why behavioral management may be so important and useful with mentally retarded clients, because they are much more

dependent on environmental, as opposed to internal, controls. This is especially true as IQ level decreases and executive deficits increase. It is common, for example, to hear reports of behavior problems in one setting (e.g., at home) but not in another (e.g., a workshop). Neuropsychological testing can help determine an individual's ability to internalize rules of behavior and to assess the amount of supervision that might be necessary. Recommendations for counseling or therapy might be more appropriate for those clients with some capacity for verbal self-regulation, whereas behavioral interventions may be better suited to those individuals without such capacity.

Summary

Neuropsychological assessment, with its basis in brain–behavior relationships and emphasis on cognitive abilities, has much to contribute to the field of developmental disabilities and clinical interventions with mentally retarded individuals. This is especially true of the qualitative process approach to neuropsychological assessment, with its focus on individual differences and flexible procedures. Neuropsychological evaluations may result in better matching clients to appropriate educational, vocational, or residential programs. Suggestions for breaking down or restructuring specific tasks can ensue. A more comprehensive description of a person's cognitive strengths and weaknesses may help staff to better understand a mentally retarded client's inconsistent abilities that often confuse or frustrate staff and client. More accurate predictions concerning an individual's potential for independent living or competitive employment may be feasible, especially with a better understanding and assessment of executive functions. Behavior changes and maladaptive behaviors may also be better understood and dealt with. Finally, in all these areas, behavioral programming and interventions can be better informed and are more likely to succeed, given an appreciation of the individual's neuropsychological status.

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PART III

Educative Interventions

CHAPTER 13

Behavioral Medicine Approaches to the Prevention of Mental Retardation

Jack W. Finney and Patrick C. Friman

Mental retardation is a devastating handicap. Its cost to society in economic terms is enormous, and the costs to the family in emotional terms are immeasurable. Mental retardation is not rare: Approximately 3% of the general population have IQs below 70 (Milunsky, 1975). Several specific causes for retardation have been identified, and knowledge of these causes provides opportunities for the development of preventive approaches.

The psychological literature devoted to treatment issues for children with mental retardation is vast and diverse, but very little literature is devoted to preventive issues, and an even smaller portion is devoted to primary prevention. The limited development of a preventive approach is disturbing in light of estimates that half of the cases of retardation may be preventable (Litch, 1980). Currently, the literature discussing prevention from a behavioral or psychological perspective is primarily focused on early enrichment of the child's environment to promote optimum cognitive development (e.g., Ramey, Sparling,

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Bryant, & Wasik, 1982). One reason for the limited promotion of other preventive issues could be the great diversity of causes of mental retardation.

Mental retardation can be caused genetically. Over 2,000 genetic disorders have been identified, including phenylketonuria (PKU), an inborn error of metabolism (McKusick, 1971). Retardation can also be caused through human errors of consumption *in utero*, among which is fetal alcohol syndrome, or through a human error of consumption during childhood, an example of which is lead poisoning. Finally, mental retardation can result from severe injuries such as head trauma. Because of the overwhelming number of causes of mental retardation, its prevention cannot be approached on a collective scale. That is, mental retardation as a specific disorder cannot be prevented, but mental retardation due to some specific causes can be prevented.

Partly due to the growing influence of collaboration by behavioral and medical scientists, strategies for prevention are receiving greater application in all of medicine. Two major preventive approaches of behavioral medicine—risk factor interventions for disease prevention and compliance enhancement interventions for regimens to reduce the severity of health problems (Masek, Epstein, & Russo, 1981)—are relevant for the prevention of mental retardation.

Four etiologies of mental retardation representative of the breadth of its known causes and amenable to behavioral medicine approaches to prevention include retardation due to PKU and fetal alcohol syndrome, retardation and intellectual deficiencies resulting from elevated lead absorption, and mental retardation caused by severe injuries in childhood. The behavioral medicine and prevention literature concerning these four etiologies of retardation is limited. A lack of data, however, need not inhibit a biobehavioral plan for prevention (Friman & Christophersen, 1986). In fact, for many causes (e.g., fetal alcohol syndrome), data on the benefit of preventive programs are not available. Thus, each etiology must be examined in terms of epidemiology, etiological considerations, developmental impact, and previously attempted preventive interventions, in order to elucidate suggestions for further prevention research.

Fetal Alcohol Syndrome

Epidemiology

Alcohol is a teratogen. Heavy drinking during pregnancy adversely affects fetal growth and development. Alcohol crosses the placental bar-

rier, and experimental evidence suggests that it interferes with organogenesis and growth and development throughout gestation. Alcohol, like many teratogens, produces a spectrum of effects, the most profound of which results in the fetal alcohol syndrome (FAS). The exact number of children with FAS is unknown, but estimates range from 1 in every 750 to 1,000 live births (Nathan, 1985). Some degree of fetal alcohol effects occurs in 1 out of every 300 to 400 live births (Hanson & Streissguth, 1978). Thus, alcohol ingestion during pregnancy may be the major cause of preventable mental deficiency.

Etiology

Although alcohol ingestion during pregnancy is the major cause of FAS, the amount of alcohol necessary to induce morphogenesis and the degree to which other risk factors contribute to the syndrome have not been determined. The severity of intellectual deficiency is usually correlated to the severity of the physical effects of FAS, and both increase with the chronicity of the mother's alcoholism. Mothers in the earlier phases of alcoholism have infants whose symptoms are less dramatic than long-term alcoholic mothers (Graham, 1983). Former alcoholic mothers who do not drink at all during pregnancy are not at high risk for producing offspring with FAS. Yet mothers who drink even small amounts of alcohol regularly during pregnancy are at risk for delivering abnormal infants. In a study of 900 pregnant women, those who drank more than 100 g of alcohol a week (about nine drinks) were twice as likely to deliver small-for-gestational-age babies as women who drank only 50 g a week (Wright *et al.*, 1983). Thus, although there are several unknowns (e.g., exact amount of alcohol, critical or sensitive periods during gestation), justification for reducing or eliminating alcohol intake during pregnancy is increasingly supported.

One confounding factor in determining the etiological variables for FAS is the life-style of heavy drinking mothers. Maladaptive dietary habits and adjunctive addictive behaviors (e.g., cigarette smoking, drug use) could contribute to the problems of the fetus. Although the effects of various life-style factors are extremely difficult to determine experimentally, the research suggests that alcohol is the primary variable of concern, and any reduction in intake during the pregnancy increases the probability of prevention of fetal alcohol effects (Sokol, Miller, & Reed, 1980). The reduction of heavy drinking to moderate drinking does not eliminate the possibility of fetal alcohol effects but does in fact reduce the probability of FAS.

Developmental Impact of FAS

FAS involves a triad of symptoms that include facial deformity, prenatal and postnatal growth deficiency, and central nervous system dysfunction. Other fetal alcohol effects that are not primary for the syndrome but that are frequently observed include cardiac murmurs, myopia, cleft palate, retarded tooth development, hydrocephalus, and meningomyelocele (Clarren & Smith, 1978).

Facial Deformities

Although severe cases of FAS can mimic other syndromes (e.g., Cornelia de Lange syndrome, Noonan syndrome), the facial deformities in FAS are highly characteristic and are used to distinguish the victims as a class (Bureau of Alcohol, Tobacco, and Firearms, 1979). The deformities include midface deficiencies, diminished or absent philtrums, flat nasal bridges, and short palpebral fissures (Graham, 1983). Overall, the facial features, particularly the eyes, have an appearance of being unfinished or underdeveloped. Of the facial deformities, the shortened palpebral fissures are considered to be the most important for diagnosis. In a major study of the syndrome, 59 subjects (91%) had this symptom (Clarren & Smith, 1978).

Growth Deficiency

Growth deficiency associated with FAS begins prenatally, and postnatal catch-up growth is uncommon. FAS children are usually below the third percentile for height, weight, and head circumference; and failure to thrive is common (Graham, 1983). Heavy-drinking mothers who reduce their intake during the second and third trimesters often have infants with less growth retardation.

Mental Retardation

The most significant and least reversible aspect of FAS is central nervous system dysfunction. The prevailing hypothesis is that alcohol retards fetal brain growth, just as it retards fetal eye, face, and body growth. Research on intellectual development in FAS shows a variety of individual IQ scores with an average of just below 70 (mildly retarded). The continuum of alcohol's effects on intellectual functioning in the exposed child ranges from hyperactivity, learning difficulties, and

communication disorders on the milder end to serious developmental brain disorders and severe mental handicap.

Identification of Drinking Status

Approximately 18 million individuals in the United States are heavy drinkers. The evidence suggests that heavy drinking is escalating among women and is beginning to match that of men (Abel, 1980). Estimates of the proportion of women who drink heavily during pregnancy range from 2% to 13% (Sokol, 1981). Women in their primary childbearing years (18 to 34) account for nearly one-fifth of the nation's "heavy drinkers." Unfortunately, identification of pregnant women who drink heavily is almost completely dependent on the mother's self-report (Landesman-Dwyer, 1982).

The use of self-report data introduces a degree of uncertainty concerning the identification of problem drinkers. Human recall is often incomplete, and many women may—inadvertently or not—underreport their drinking habits. Furthermore, heavy drinking is a behavior with a negative valence and, therefore, is not one that is likely to be reported accurately. Given the possibility of adverse effects from even relatively low levels of drinking, however, problem drinking during pregnancy does not necessarily mean heavy drinking. It can mean regular, moderate drinking or at least one occasion where more than 5 ounces of alcohol (e.g., five mixed drinks) are consumed. Thus, during the history, the physician's assessment could take the form of one question, "Do you drink?" A positive response to that question should be followed by questions about regular and episodic drinking amounts. If the mother regularly drinks one or more ounces of alcohol daily or has on any occasion, during or immediately before the pregnancy, drunk five or more ounces of alcohol, she should be targeted for some level of intervention.

More elaborate assessments are available in the form of questionnaires. Rosett and Weiner (1981) present a widely used drinking questionnaire that involves 10 simple questions that target the kind and the amount of alcohol the mother typically consumes. The questionnaire could be completed in the waiting room and discussed during a prenatal visit.

Assessments for FAS should target only regular, moderate, or heavy drinking, and episodic heavy drinking during pregnancy. The epidemiologic evidence linking low levels of drinking and fetal alcohol effects is weak, and its exaggeration could lessen the credibility of the well-

established link between higher levels of drinking and teratologic outcomes (Rosett & Weiner, 1982).

To increase the effectiveness of early identification programs, the health community may need to focus on problem drinking in the fertile female population rather than just the pregnant population. If a physician ascertains that a woman of childbearing age is a moderate to heavy drinker, that information should be charted so that it can be addressed when and if the woman becomes pregnant.

Preventive Approaches for FAS

Although impossible to calculate exactly, New York State has estimated that babies affected by alcohol in a single year will cost the state $155 million for lifetime care (Blume, 1981). Estimates from the state of South Dakota for the yearly cost of care for a child with FAS range from $10,000 to $30,000 (Stanage, Gregg, & Massa, 1983). Obviously, the costs to society of FAS in dollars alone—not to mention the emotional costs to the child and family—suggest that any effective preventive effort is likely to be cost-effective. Not many preventive programs have yet been implemented and, similarly, few data are available to evaluate their effectiveness. Preventive programs primarily involve health education for the public, professionals, and expectant or potentially expectant mothers, and individual counseling for drinking mothers.

Health Education for the Public

Increased public awareness of the risks that alcohol presents to the fetus is clearly needed. A Seattle survey showed that over one-third of the citizens surveyed believed that consumption of three or more drinks daily was safe during pregnancy (Little, Streissguth, & Guzinski, 1980). This finding is particularly striking because Seattle has been a major center for the study of FAS, and a significant amount of media attention has been given to FAS in the Seattle area. A sample community has been targeted to provide every resident with information about the risks of drinking during pregnancy. The primary techniques are media coverage and distribution of informational pamphlets. The goal of the program is to increase the number of referrals of women with drinking problems to appropriate health care providers (Little *et al.*, 1980). If the prospective study shows that the health education approach increases referrals, similar programs should be established around the country.

Health Education for Professionals

Health professionals need to be knowledgeable about the effects of alcohol on the outcome of pregnancy. A primary method of reaching licensed professionals could be inclusion of FAS information in continuing education workshops that are usually mandatory for maintaining certification or licensure. Professionals could also help to change the legal status of alcohol abuse during pregnancy, which could correspond to the change that has occurred with child abuse in recent years. When a mother drinks to excess during pregnancy, she places the fetus at great physical risk. Drunkenness during pregnancy has been characterized as a form of intrauterine child abuse (Stanage *et al.*, 1983). If the legal status of alcohol abuse during pregnancy were changed, adjunctive programs could be incorporated to assist professionals in identifying and referring abusing mothers for services. The goal of formalizing the offense would not be to punish the drinking mother but rather to arrange for her to receive treatment for her problem drinking in order to protect her baby.

Health Education for Expectant Women

Information about the dangers alcohol poses for the fetus should be routinely delivered to all pregnant women during prenatal care. Similarly, women of childbearing age should be familiar with these hazards. The education of professionals, using the most recent and rigorously obtained scientific findings, will help to ensure that women receive accurate information. Behavioral interventions, however, will be necessary to increase the rate at which such information is disseminated across the country. Ideally, information about alcohol should be included in all health education classes for high school students, all prenatal classes for expectant parents, and all parenting classes. Research could focus on increases in the number of services that deliver this type of information to women.

Counseling

Counseling for heavy-drinking mothers should begin as soon as alcohol abuse is identified (Rosett & Weiner, 1984). The mother with a drinking problem may have diminished concern for herself. In counseling with pregnant mothers, however, the provider may be able to take advantage of the mother's increased concern for her infant. The goal for the treatment of heavy drinking should be the attainment and maintenance of abstinence throughout the pregnancy. One obstacle to

abstinence is the patient's despair over her ability to stop drinking. A supportive and encouraging attitude from the provider can decrease this despair and increase the likelihood of success (Chappel & Schnoll, 1977), whereas a negative or accusatory attitude from the provider may increase despair and imperil treatment (Rosett & Weiner, 1984). Thus, frequent supportive contact between the mother and the provider will be necessary in most cases, with inpatient treatment being a necessary alternative only in severe cases.

Social support could also increase the cost-effectiveness and the success of counseling (Janis, 1983). The drinking mother is likely to either be insular or involved socially with individuals who support her drinking. In either case, establishing a network of social support for sobriety could help to weaken previous behavior chains that led to drinking and to establish new chains that lead to sobriety. Previous clinical reports indicate that the involvement of family members can contribute to the support necessary for abstinence (Rosett & Weiner, 1984).

Phenylketonuria

Epidemiology

Phenylketonuria (PKU) results from an inborn error of metabolism, and mental retardation is its most serious outcome. The *Nelson Textbook of Pediatrics* describes the classic case of PKU as involving (a) elevated levels of phenylalanine in the blood; (b) normal or reduced plasma tyrosine levels; (c) increased phenylalanine metabolites in the urine; and (d) an inability to tolerate an oral challenge of phenylalanine (Morrow & Auerbach, 1983). The incidence of PKU in the United States is 1:15,000 with the distribution between males and females being equal (Nyhan, 1984).

Phenylalanine is an essential amino acid present in all natural proteins. The basic defect resulting in PKU is the failure of the enzyme phenylalanine hydroxylase to convert phenylalanine to tyrosine. As a result, phenylalanine accumulates in the blood at abnormal levels and causes injury to the brain. The inborn error is transmitted by an autosomal recessive gene; approximately 1 in 60 persons is an asymptomatic heterozygous carrier (Centerwall & Centerwall, 1972; Morrow & Auerbach, 1983).

Developmental Outcome

Rarely will a person with untreated PKU have normal mental development. The untreated child with PKU is normal at birth but begins

to show the features of slowed brain development as early as 4 months. The classic clinical picture involves moderate to severe mental retardation accompanied by erratic behavior patterns. The children are usually blonder than their unaffected siblings, have blue eyes, skin problems, a "musty" odor, and microcephaly. About 80% have abnormal EEG patterns, and approximately one-third have seizures (Centerwall & Centerwall, 1972; Nyhan, 1984).

Detection

Screening programs for PKU are well established, and virtually every infant born in developed countries is tested for the disease (Nyhan, 1984). In fact, the widespread screenings have produced an understanding of PKU and a body of data that is more extensive than that available for any other inborn error of metabolism (Levy, 1973; Nyhan, 1984). The screening most commonly used involves the blood test developed by Guthrie and Susi (1963). A serum concentration of 20 mg/dl is usually required for a diagnosis of classic PKU (Morrow & Auerbach, 1983; Nyhan, 1984).

Preventing Retardation Due to PKU

The primary way to prevent mental retardation from PKU is to control the dietary intake of phenylalanine. Bickel pioneered dietary treatment of PKU by limiting dietary phenylalanine (Bickel, Gerrard, & Hickmans, 1954). During the 1960s, experimental evidence accumulated, showing that early dietary management beginning in the first month of life resulted in normal developmental progress. Preventive dietary treatment for PKU is now well established. Several versions of the diet are available, but they all are focused on the same general goals: providing enough proteins and other essential nutrients to ensure optimal growth and development while maintaining a reasonable concentration of phenylalanine in the blood; a reasonable concentration range from 3 to 15 mg/dl, and an optimal range between 4 and 8 mg/dl (Guttler, 1984; Nyhan, 1984; Rohr, Levy, & Shih, 1985).

One problem with the goals of dietary interventions is that all proteins in commonly available foods contain 5% to 6% phenylalanine, and it is virtually impossible to devise a diet from natural foods that lowers phenylalanine and provides essential nutrients. Therefore, PKU diets use synthetic foods that provide amino acids but minimal phenylalanine (e.g., Lofenalac, Meade Johnson) or no phenylalanine (e.g., Phenyl-Free, Meade Johnson).

A potential problem with the PKU diet is phenylalanine deficiency (Rohr *et al.*, 1985). Phenylalanine is an essential amino acid and thus, a phenylalanine-free diet must be supplemented with low protein dietary supplements such as fruits, vegetables, and synthetic foods in order to maintain essential serum phenylalanine levels. The necessity of supplements for the special diet complicates the process for the parent but also allows for greater dietary variations for the child. One way to decrease the complications and to increase dietary variations is to provide lists of acceptable foods and sample menus to parents (Anderson, Kennedy, & Acosta, 1985; Centerwall & Centerwall, 1972).

How long the children with PKU should remain on the diet has varied as the research on dietary interventions has accumulated. Early literature has suggested that deficits in growth and development are less obvious after age 3 (Centerwall & Centerwall, 1972) but that children should be kept on the diet until age 5 (Hanley & Linsano, 1973). More recent findings indicate that children should not be taken off the diet until after the age of 8 (Holtzman, Kronmal, Van Doorninck, Azen, & Koch, 1986). Furthermore, the evidence shows that the development problems occurring as a result of early discontinuance are multimodal involving social (Matthews, Barabas, Cusak, & Ferrari, 1986) as well as intellectual domains (Holtzman *et al.*, 1986). Thus conservatism appears to be the best preventive policy with respect to discontinuing the PKU diet.

Programmed Instruction

Diets for normal children can lead to difficulty and misunderstanding (Finney, 1986). The rigor and restrictions of the diet for PKU children have a high potential for misunderstanding. Although parents of children receive health education, many parents are unlikely to understand the intricacies of managing PKU or the exigencies of its dietary management. Misunderstanding and/or ignorance of PKU and its preventive regimen could lead to noncompliance with the regimen by the parent and delayed developmental outcome for the child. Thus, better teaching techniques are necessary to improve the understanding of, and compliance with, dietary requirements for PKU (Acosta, Wenz, & Williamson, 1978).

Programmed instruction, an educational method that provides individualized instruction with immediate reinforcement for successful progress (Skinner, 1968), has potential usefulness in nutrition education for parents of PKU children. A programmed instruction manual has been successfully tested with 10 mothers of children with PKU (Marino, 1980). Pre- and posttest scores showed that the manual significantly improved

the mother's knowledge of PKU and its dietary management. Consumer satisfaction measures also showed that the mothers' reactions to the manual were favorable. This study was preliminary and provided only measures of knowledge, not compliance with the PKU diet or developmental outcome of children on the diet. Future research should document further needs and benefits of improved educational programs for maintaining the PKU dietary regimen.

Managing Feeding Problems

One problem with the PKU diet is the limited variety of foods available for the child; the diet is drab, monotonous, and unappetizing (Schild, 1979). Despite its monotony, most parents and children adapt to the diet (Centerwall & Centerwall, 1972; Schild, 1979). For some children, however, dietary compliance is a problem and, thus, retardation is a risk. For these families, simple behavior management could have preventive benefits.

Feeding problems are not uncommon in the normal population, and the technology that has been applied to their remediation can be applied to feeding problems of children on the PKU diet. A first concern is early intervention. Many parents report feeding problems only after they have become severe (Forsyth, Leventhal, & McCarthy, 1985). With the early introduction of the PKU diet, problems that might occur as a result of a later, dramatic change in diet when the child is older are prevented. But as children are exposed to the relatively rich and varied diets of their peers, parents, and siblings, they may react adversely. The first of these reactions should be reported to the pediatric provider. At this point, the provider should consider an intervention involving simple behavior management.

A well-developed technology is available for managing mealtime problems (Finney, 1986). Space does not allow review of that technology in detail, but a few salient points should be addressed. First, parents should be instructed how to reward and discipline mealtime behaviors, depending on the appropriateness of the behavior. Second, parents should be instructed to model and shape appropriate mealtime behavior. Third, parents should use the variety available within the diet for increased compliance. The key is to use preferred foods (which will probably not be the synthetic foods) to reinforce consumption of the nonpreferred but necessary foods. Fourth, all behavior management instructions should be written in a handout form for continued reference by the parents at home (Finney, 1986)).

Social Context of Mealtimes

All children develop food preferences or nonpreferences. The key to the successful PKU diet is for the child to prefer the special diet foods. The social context in which foods are served can significantly influence a child's preference for foods (Finney, 1986). The literature shows that children who observe their peers eating a nonpreferred food will develop a preference for that food (Birch, 1980). Other studies show that nonpreferred foods that are used as a reward or whose delivery is paired with adult attention become preferred foods (Birch, 1981; Birch, Zimmerman, & Hind, 1980). Thus, observational learning can determine the development of food preferences.

Several suggestions for promoting a mealtime context that is conducive to dietary compliance can be made:

1. Make meals a pleasant family time.
2. Eat as a family as early and often as possible.
3. Have the parent eat some of the dietary supplement often.
4. Avoid coercion.
5. Respond only to food intake, not to comments about food.

Feeding, an important issue for all children, is extremely important for children with PKU. The implementation of behavior management strategies, combined with improvements in the social context of meals, could make the dietary substitutions necessary for PKU children less problematic for the family and therefore decrease the possibility of dietary noncompliance.

Maternal PKU

Sufficient evidence now exists to show that adult women with PKU have an elevated risk for producing non-PKU children with mental retardation and multiple congenital anomalies (Schild, 1979). In fact, current projections suggest an incipient rebound of mental retardation due to maternal PKU (Kirkman, 1982). Unfortunately, special dietary therapy initiated during the pregnancy has not proven effective for preventing the problem (Lenke & Levy, 1980). In order to be truly preventive, the dietary intervention may have to be initiated at or before conception (Bush, 1985). Such an intervention presents a major problem in that both health professionals and women with PKU of childbearing age need to be informed of the risk associated with pregnancy and the need for dietary intervention prior to conception.

Because women with PKU will have been off the special diet for

many years, they must face problems relearning the diet's requirements and its adverse reactions. One possibility for a behavioral intervention involves teaching young women with PKU (e.g., young adolescents) the diet so that they will be more familiar with it during their childbearing years. Simply being on the diet presents another problem. The change in diet for the woman with PKU may cause nausea and could lead to noncompliance (Bush, 1985). Therefore, frequent contact by the mother with her medical provider and the establishment of social support for compliance with the provider's recommendations are critical. Overall, maternal PKU has had little investigation, and research in this area could lead to prevention of the predicted rebound in mental retardation due to dietary intake in pregnant women with PKU.

Other Genetic Disorders

Numerous other genetically determined metabolic and endocrine disorders associated with retardation have been identified, the effective treatment of which will prevent retardation (Moser, 1982). The preventive interventions constitute excellent examples of the interface of the medical and behavioral sciences. Supplying a missing metabolite in diseases such as hypothyroidism is one such example. Diagnosing the disease and supplying the metabolite is the responsibility of medical practitioners, but assuring compliance with the daily regimen necessary for full preventive effect is a behavioral issue.

Depleting the storage of a toxic substance is another preventive intervention. In Wilson's disease, copper accumulates in various organs throughout the body, and its storage causes multiple pathological symptoms, including mental retardation. Wilson's disease is an example of a disease that causes CNS damage that can be reversed through specific therapy (Moser, 1982), in this case typically involving regular ingestion of penicillamine. Identifying the disease and supplying the penicillamine are medical problems, but maintaining compliance with the regimen, which can last many years, requires a behavioral medicine solution.

A final example is supplying a vitamin co-factor for a vitamin-responsive disease, such as maple syrup disease. This type of disease is rare but important because a child with a vitamin-responsive form of maple syrup disease can be maintained in good health with a daily dose of thiamine (Moser, 1982). Again, ensuring that the parent will continue to give the thiamine, and that the child will take it, is a preventive problem for the behavioral sciences.

Lead Poisoning

Epidemiology

Lead poisoning is a significant problem in the United States. Over 675,000 children between the ages of 6 months and 5 years have low-level asymptomatic lead absorption (blood-lead concentrations greater than 30 μg/ml of blood) (Mahaffey, Annest, Roberts, & Murphy, 1982). A far greater number of children have lower but elevated blood-lead concentrations from environmental exposure.

Lead is a common environmental pollutant in industrialized nations. The most common lead threats to children are lead-based paint and emissions from automobiles that use leaded gasoline. The primary way that children absorb lead is from mouthing and pica that result in ingestion of house dust, actual eating of chips of old, flaking paint, and ingestion of food, water, and dirt that contain high amounts of lead (Charney, Sayre, & Coulter, 1980). Children who reside in older housing containing lead-based paint are susceptible to lead absorption through common, developmental behaviors that all children exhibit. Children in impoverished settings mouth objects and body parts significantly more often than children in enriched settings with toys and adult attention, and it is not uncommon for children in impoverished settings to ingest flaking paint chips (Madden, Russo, & Cataldo, 1980b). Children from low-income families may be exposed to an impoverished environment, thereby placing them at greater risk of mouthing and pica that result in an increasing body lead burden.

Developmental Impact

It is known that high blood lead concentrations can result in significant intellectual and attentional deficits. At severe concentrations (>70 μg/dl) severe encephalopathy and death can occur, although mortality and severe encephalopathy due to acute lead poisoning are now rare. The presence of "mild" CNS symptoms such as irritability and clumsiness (Pueschel, Kopito, & Schwachman, 1972) and poor classroom performance (Needleman *et al.*, 1979) suggest that even low levels of lead may adversely affect children. However, there have been conflicting findings about the effects of low blood lead concentrations on impairment of intelligence (Ernhart, Landa, & Schell, 1981; Needleman, Leviton, & Bellinger, 1982).

The debate about the effects of low lead levels is unresolved. However, a thorough review of the effects of excessive lead burden concludes

that, although studies have not proved that lower blood-lead concentrations impair children's intellectual abilities, it is safer and scientifically valid to assume that low levels of lead poisoning are harmful and should be prevented when possible (Rutter, 1980; Rutter & Jones, 1983).

Reducing the incidence of lead poisoning—one cause of encephalopathy, mental retardation, and intellectual deficits—is one way of preventing mental retardation. Although lead poisoning is not a major cause of intellectual disabilities, it is a significant threat to a large number of children, particularly those who live in poor, urban areas. Reducing lead poisoning is also a preventive effort that can be advanced by behavioral science interventions.

Preventive Interventions to Reduce Lead Poisoning

Significant progress has been made through legislation to reduce the amount of lead introduced into the environment by industrial and other wastes. However, a considerable number of children will be exposed to increased lead for many years. Three efforts that have been pursued to reduce children's exposure to and absorption of lead will be reviewed. These measures are representative of environmental and behavioral science efforts to prevent developmental disabilities due to lead absorption.

Removing Lead Hazards

Removal of lead sources from the environment is the primary method of eliminating initial and recurrent increases in blood lead concentrations. Federal acts such as the Clean Air Act and the Lead-Based Paint Poisoning Prevention Act have helped in reducing ambient lead in the environment and in preventing the continued use of lead-based paint (Farfel, 1985). Sources such as lead-based paint are very common in old inner-city housing, where low-income residents are often forced to reside due to the low rents associated with decrepit housing.

Abatement of lead paint hazards by sanding or burning the lead-based paint or by covering the painted walls with paneling or dry wall materials is required by law when home lead hazards are reported to a health department. Due to a lack of personnel and funds, problems occur with enforcement of abatement procedures, and state and city legal requirements are often not sufficient for truly removing the risk of lead poisoning (Chisolm & O'Hara, 1982); some abatement programs only remove the lead-based paint up to 4 feet above the floor. Therefore, substantial risks for lead absorption can remain after abatement of older

housing. Two approaches to supplement abatement procedures have some promise for reducing the risk of lead poisoning for specific populations and, thereby, for advancing prevention of development disabilities: reducing the amount of house dust and reducing mouthing and pica.

Reducing Exposure to House Dust

Dust-control techniques offer some promise for reducing high lead levels in children who must continue to reside in housing with lead hazards. The techniques include concentrated home cleaning of areas where lead-containing dust accumulates, keeping children away from areas in the home where leaded dust exists, and regular handwashing before meals and at bedtime. A dust-control program was provided to an experimental group, whose blood-lead values over a 12-month follow-up were compared with a control group who did not receive the dust-control intervention (Charney, Kessler, Farfel, & Jackson, 1983). When intensive dust-control cleaning every 2 weeks was provided for identified "high-lead" areas (e.g., windowsills and floors around windowsills), blood-lead levels for children in the experimental group were significantly reduced, whereas only a minor reduction occurred for control group children. The effect was strongest for children with high blood-lead concentrations; children with blood-lead levels around 30 to 35 μg/ml did not show a significant reduction in lead levels from the dust-control techniques.

Parents are routinely instructed in dust-control techniques during visits to lead poisoning screening and treatment clinics. They may, however, be unlikely to maintain the rigorous wet mopping and wiping regimen that is required to achieve dust control, especially in the absence of feedback concerning the efficacy of their cleaning habits. Interventions for enhancing parental compliance with short- and long-term recommendations have been developed (e.g., Finney, Friman, Rapoff, & Christopherson, 1985; Masek & Jankel, 1982). Development of training and compliance interventions for home dust-control techniques would benefit families who are forced to reside in older, inner-city housing. Training programs are needed because it is unlikely that federal and state financial support can be used to provide cleaning services for families who must continue to reside in the risky housing. But the implications are clear: Dust control techniques can make a house with lead hazards more habitable, but practical limitations of dust-control techniques indicate that only removal of lead hazards—or removal of fam-

ilies from hazardous housing—can truly protect children from exposure to lead.

Reducing Mouthing and Pica of Children Exposed to Lead Dust

Children who have been treated for increased lead levels must often return to houses where only partial—or no—abatement has occurred. Children may also be exposed to lead hazards during visits with relatives and friends whose homes may also contain lead-based paint. In these cases, mouthing and pica can place children at continued risk for ingestion of lead-containing dust or actual paint chips. Thus, for some children, interventions to reduce mouthing and pica may help prevent occurrences—or recurrences—of elevated blood-lead levels.

Mouthing and pica are normal developmental behaviors for young children, and parents are usually counseled about the lack of need for interventions to reduce or eliminate these oral habits during the preschool years. The risk of lead absorption, however, argues for teaching children to replace mouthing with other ways of interacting with the environment. Oral habits have been shown to be responsive to environmental stimuli, such as increases in mouthing and pica in impoverished settings and reductions in frequency when adult attention or playmates are present (Madden *et al.*, 1980b). Such environmental responsiveness suggests that these behaviors are good targets for direct behavioral intervention.

Two studies explored behavioral interventions for eliminating pica and mouthing in children with lead poisoning (Finney, Russo, & Cataldo, 1982; Madden, Russo, & Cataldo, 1980a). The treatment approach incorporated several behavioral procedures for reducing pica and mouthing behavior. Discrimination and correspondence training was provided to teach children to not eat inedible items (e.g., toys, simulated paint chips, household objects) and to refrain from mouthing body parts that would transmit house dust (e.g., hands, clothing). A differential reinforcement procedure was implemented to increase the duration of play without occurrence of mouthing or pica. And, for some children, an overcorrection procedure, brief toothbrushing contingent on placing objects or body parts to the mouth, was necessary to eliminate mouthing.

Pica and mouthing can be controlled when intensive behavioral procedures are conducted by trained therapists. Scientific validation of the "protective" benefits of reduced pica and mouthing for controlling lead absorption must occur before large-scale programs are developed. Application of the procedures to large groups of children who may be

at risk for lead absorption has yet to be demonstrated. These preliminary studies suggest the potential for controlling pica and mouthing behavior, but the practical issues of implementing behavioral procedures as a public health intervention must be solved. A relatively simple technique, the contingent application of bitter nail polish, reduces oral habits and may be practical for use in pediatric primary care (Friman, Barone, & Christophersen, 1986). The extension of this research to children at risk for lead poisoning is needed.

Temporary Preventive Solutions?

Elevated lead levels are a threat to children's development and intellectual growth. Prevention of lead absorption is one way of reducing this threat. Removal of lead hazards will ultimately remove this developmental threat. This solution, however, will take decades to achieve. Controlling ingestion of lead-containing house dust and reducing mouthing and pica behavior are supplementary prevention activities that have great potential. A firm link between the effectiveness of these techniques and a subsequent reduction in developmental impairment, however, will be needed to justify continued concentration of resources to these preventive solutions.

Motor Vehicle Trauma

Epidemiology

Accidental injuries are common in this country: Approximately 75 million persons in the United States receive medical attention for injuries each year, and injuries are the most common cause of death in humans until the age of 40 (Cone, 1988). For children, accidents cause more deaths and disabilities than the six leading pediatric diseases combined (Pless, 1978). Of the available causes of accidents, the motor vehicle is the greatest cause of death and serious injury. From 1970 to 1980, over 10,000 children under the age of 5 were killed in automobile accidents (Sanders, 1982). An estimated 1 out of every 40 children born in the United States will die in a motor vehicle accident, and 1 out of 20 will be seriously injured (Christophersen & Sosland-Edelman, 1983). Thus, injury associated with motor vehicles is a leading health concern for children.

Etiology

The extent to which injury leads to mental retardation has not been fully studied. Yet it has been linked to mental retardation and developmental disability in several reviews (Carter, 1970; Cone, 1988). Furthermore, head injury is the leading cause of acquired cerebral dysfunction in children, and motor vehicle crashes are the leading cause of head injury (Annegers, Grabow, Kurland, & Laws, 1979; Levin & Eisenberg, 1979). Therefore, preventing motor vehicle trauma is another primary method of preventing cerebral impairment and mental retardation.

Motor vehicle injuries themselves are multiply determined but can usually be linked in some way to carelessness. They are the result of reckless driving, driving under the influence of alcohol or other drugs, fatigue, excessive speed, heedless reactions to changing road conditions, and many other violations of legislated driving codes. Improving the skill and care with which individuals operate motor vehicles is an abiding concern for many human services and health disciplines. The available technology for and preventive benefits of improvements in cautious motor vehicle usage are too large a topic for this review. But many injuries that result from motor vehicle accidents could be prevented through proper passenger restraint. The literature on proper restraint usage is also too voluminous for a comprehensive review here. An appropriate section of that literature, preventing vehicular trauma to infants and young children, will be reviewed.

Assessment

Vehicular trauma results in morbidity and mortality, and the use of child restraints can prevent serious injury, but documenting that children are being properly restrained is difficult. A primary source of assessment data is the survey. Survey data, however, are not reliable indicators of car restraint usage (Barone, Sosland-Edelman, & Christophersen, 1985). Parents who use car seats accurately report their usage, but parents who do not use seats often falsely say that they do. In a study using direct observation of car-seat usage after an intervention, only 11% of parents were observed using seats (Reisinger & Williams, 1978). If surveys had been used, the percentage of parents who reported using car seats would probably have been inflated, and the critical need for an additional intervention would have been less evident. When determining the need for or effect of a car safety program, direct observation is the best assessment.

Preventing Motor Vehicle Injuries

Many childhood injuries and deaths in automobiles are preventable through proper use of child restraints. Of the available methods for promoting the purchase and use of car seats, legislation is one of the most powerful. Currently, the trend across the 50 states toward legislating mandatory car seat usage is encouraging. Even after legislation, problems with affordability, appropriate use, and regular use still exist.

Affordability

The available evidence suggests that mothers most at risk for the loss of a child through an accident have many children and limited education (Wicklund, Moss, & Frost, 1984). The evidence also suggests that families with limited resources are less likely to use car seats (Barone *et al.*, 1985). One method of increasing seat usage for families who cannot afford them is a loaner program. Detailed information on how loaner programs can be established is available from various sources (e.g., EarlyRider, Department of Transportation, National Highway Traffic Safety Administration, 400 Seventh Street N.W., Washington DC 20590) and the benefits of loaner programs have been experimentally demonstrated (Christophersen, Sosland-Edelman, & LeClaire, 1985).

Appropriate Use

Merely possessing a car seat does not ensure its appropriate usage; parents must be instructed how to use the seat. The physician's time is very limited, and therefore nurses are probably the most appropriate persons to provide instruction, which can be done in at least two ways. First, prenatal classes, frequently led by nurses, provide a useful forum for teaching appropriate car seat usage. And second, when accompanying the mother at the time of discharge, the nurse can influence how a car seat is used (Christophersen & Sullivan, 1982). If the nurse allows the child to ride unrestrained at discharge from the hospital, the infant usually rides in the mother's lap, a precarious position that has been called the "child crusher." If the nurse instructs the parent in the appropriate use of a car seat, however, as many as two-thirds of the infants will be correctly restrained in car seats (Christophersen & Sullivan, 1982).

Regular Use

The benefit of child car seats is reduced with inconsistent use. Several variables influence the maintenance of car seat usage. Statewide

networks of hospital programs involving doctors, nurses, and volunteers dramatically increase regular car seat usage (Colletti, 1986). This approach appeared to work because the network in the program promoted car seat usage to such a degree that the message was practically impossible for new mothers to avoid.

In more limited programs, other variables can influence regularity of usage. Response requirement influences compliance with health care recommendations (Friman, Finney, Rapoff, & Christophersen, 1986). An integral part of response requirement is effort. If the use of a particular car seat involves increased effort, a parent's use may diminish over time. To avoid decreased usage due to increased effort, providers should sample various models of car seats and identify those that are easiest to use.

Another variable influencing car seat usage concerns the benefits that accrue from regular usage, above and beyond injury prevention. In fact, merely articulating the negative consequences of not using car seats has historically not been successful (Treiber, 1986). Regular use of car seats has many benefits. Car seats can improve child behavior during the car ride (Christophersen, 1977; Christophersen & Gyulay, 1980). Using car seats can reduce driver distraction and thus increase the "enjoyability" of the ride while also decreasing the risk of a distraction-related accident. Car seats can also decrease the likelihood that a child will suffer from motion sickness and vomit during the ride. Finally, a child comfortably restrained in a car seat is more likely to sleep during the ride than one who is allowed to roam free or who sits on the parent's lap. To promote regular seat usage, providers should point out the positive benefits from use when instructing parents (Treiber, 1986).

Motor vehicle accidents, of course, do not just happen to children. They are a major cause of death and disability for adolescents and adults as well. Behavioral medicine approaches can help to prevent such accidents by increasing seat belt usage, decreasing driver use of alcohol, and promoting defensive driving. The technology for reducing risk of disability or death in children from motor vehicle trauma is now available. The mandate for using that technology is clear. Only the widespread application of that technology to children across the country is lacking.

Other Accidents

Many other accidental causes of retardation are available as targets for behavioral medicine. They may not be as predominant as motor vehicle trauma, but their sequelae are just as tragic. Their importance and the key to their prevention can be gauged developmentally. For

small children, brain damage due to anoxia from ingestion of small objects or accidental strangulation is a concern. For adolescents, sports injuries that damage the head and spine can retard development and diminish cognitive function. Finally, for the elderly, brain and spinal trauma due to falls is a primary concern. For some of these causes, programs have been developed and evaluated (e.g., seat belt usage; Geller, Bruff, & Nimmer, 1986) and for others they have not (e.g., falls in the elderly). Clearly, accident prevention at all ages is a primary method of preventing retardation, disability, and death, and it should be a major agenda item for behavioral medicine research.

Behavioral Medicine Contributions

Psychologists, physicians, social workers, nurses, and other health care professionals can contribute to the prevention of mental retardation. Early identification of disorders for which there are regimens that prevent cognitive-intellectual threats, compliance with preventive regimens to reduce the developmental impact of disorders, removal of or protection from environmental hazards such as lead, and programs that teach parents ways to protect their children during routine activities such as car trips are examplars for prevention efforts designed to reduce mental retardation.

Table 1 summarizes techniques that can be used to advance preventive efforts. The public, parents, and persons directly affected by certain risk factors for retardation must be educated about the risk factors and life-style activities that can result in a handicapped child. But imparting of knowledge is rarely enough (Finney & Christophersen, 1984). For health education to be effective, other interventions must usually be paired with information dissemination. Compliance enhancement interventions can be critical for ensuring that long-term regimens are followed that prevent developmental sequelae from genetic disorders. Behavior management strategies can be useful for maintaining children on a difficult, unpleasant, or boring regimen. And environmental changes and legislative actions can contribute to the effectiveness of preventive efforts (Cone, 1988).

Prevention has only recently become a major focus for medicine, psychology, and other health sciences. Therefore, it is not surprising that extensive plans for prevention have not been developed and evaluated. Most effort has been devoted to prevention through dietary management for persons with PKU. Still, many issues remain including pa-

Table 1. Preventive Interventions for Mental Retardation

Technique	Topics
Health education	Dangers of drinking during pregnancy
	Importance of genetic screening programs
	Hazards of lead sources
	Prevalence of child injury
	Importance of child restraint seats
Compliance enhancement	Abstention from drinking during pregnancy
	Maintenance of PKU diet
	Dust-control regimens for homes with lead
	Regular use of child restraints/seat belts
Behavior management	Feeding problems associated with PKU diet
	Eliminating mouthing and pica
	Behavior problems during car travel
Environmental change	Abatement of home lead hazards
Legislative action	Removal or control of lead hazards
	Mandatory child restraint and seat belt use
	Neonatal screening programs for genetic disorders

rental noncompliance with dietary regimens, child feeding problems, and dietary control needed for women with PKU who wish to become pregnant. Less work has been done on motor vehicle trauma and mental retardation and disabilities, but a mandate for passenger safety is developing across the country and studies showing the effectiveness of behavioral interventions for enhancing car safety are increasing. Lead poisoning has been an identified risk to child development for decades, and abatement and lead hazard removal has been targeted with some success. Only recently have initial studies of protecting children who must remain exposed to lead hazards been reported (Charney *et al.*, 1983; Finney *et al.*, 1982), but these studies suggest several possibilities for temporary preventive solutions to lead poisoning. The literature on FAS is in a developmental period. The health community is aware of the threat alcohol presents to the fetus, but increasing the public's awareness is currently an unmet need. Although preventive programs exist, data on the effects of controlling maternal drinking are incomplete (Little *et al.*, 1980), and a recent review of preventing alcoholism is pessimistic (Nathan, 1985).

We have reviewed only a sample of interventions that are possible for the prevention of mental retardation. Combining information obtained from studies of the epidemiology, etiology, and assessment of health and developmental threats with strategies for treatment can result

in an effective plan for prevention (Friman & Christophersen, 1986). The need for further research is evident from a review of the existing literature. The plausibility of a behavioral medicine approach is apparent. An objective of this chapter has been for readers to place mental retardation in a context of prevention. A major success for children will be the development and evaluation of preventive programs that reduce the threat of mental retardation and that allow children an enhanced potential for optimal growth and development.

ACKNOWLEDGMENTS. Preparation of this manuscript was supported in part by grants from Maternal and Child Health (MCJ 243270 and MCH 000917) to the Kennedy Institute. We thank Fred Leebron and Laura Charping for their assistance.

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CHAPTER 14

On Effective Prevention of Mental Retardation

S. M. Pueschel

Introduction

Mental retardation is one of the nation's greatest public health problems. It is estimated that there are over 6 million persons in this country who are mentally retarded, and each year more than 100,000 new cases of retardation occur.

During the past decades, many causes of mental retardation have been uncovered, and many conditions associated with mental retardation can be prevented today.

Fetal Alcohol Syndrome

Although even during biblical times people must have known of the ill effects of alcohol during pregnancy as it is said in Judges 13:7, "behold, thou shalt conceive and bear a son: and now drink no wine or strong drink. . . , " it was only during the past 20 years that the fetal alcohol syndrome was introduced as a distinct entity. In 1968, Lemoine, Harousseau, Borteyru, and Menuet studied 127 children of 69 chronically alcoholic mothers and found an increased prevalence of "odd faces,

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growth retardation, other malformations, and psychomotor disturbances" (p. 476). In 1972, Ulleland called attention to an association between alcoholic mothers and low-birth-weight offspring with growth and developmental delay. Jones, Smith, Ulleland, and Streissguth reported in 1973 a recognizable pattern of multiple congenital anomalies, including craniofacial, limb, cardiovascular defects, and prenatal onset of growth and developmental retardation in eight unrelated children of chronic alcoholic mothers. In another report published in the same year, Jones and Smith (1973) called the disorder "fetal alcohol syndrome." The anomalies described in Jones and co-investigators' first patients included prenatal and postnatal growth deficiency, developmental delay, microcephaly, and short palpebral fissures. A small number of the infants also had other craniofacial anomalies including epicanthal folds, maxillary hypoplasia, cleft palate, anomalies of external genitalia, capillary hemangiomata, and fine motor dysfunction.

Since these reports appeared in the late 1960s and early 1970s, many articles describing further phenotypic, epidemiologic, pathogenetic, and behavioral aspects of fetal alcohol syndrome have been published. Yet, the precise pathogenetic mechanism leading to fetal alcohol syndrome is still not well understood. It is not clear whether alcohol is acting as a direct toxin via its metabolites or whether other factors must be present to produce abnormalities. Other concomitant aspects that have been considered are malnutrition, smoking, drug abuse, vitamin or trace metal deficiency, and hypoglycemia (Ouellette & Rosett, 1976). At the present time, the evidence favors ethanol or some toxic breakdown product as the most likely etiologic agent of the fetal alcohol syndrome (Hanson, Jones, & Smith, 1976).

Since the initial description of this disorder, it has become apparent that there exists a spectrum of effects of alcohol on fetal development. Olegard *et al.* (1979) found 33% of infants of chronic alcoholic women manifested the full-blown fetal alcohol syndrome, a total of 76% had either a full or partial effect, and only 24% of infants were normal. With respect to a moderate amount of alcohol intake during pregnancy, Hanson, Streissguth, and Smith (1978) observed that 11 offspring born to women who drank between 1 and 2 ounces of absolute ethanol daily during the first trimester of pregnancy had features consistent with prenatal effects of alcohol.

Although Finney and Friman pointed out in the preceding chapter that "the epidemiologic evidence linking low levels of drinking and fetal alcohol effect is weak" and that "a Seattle survey showed that over one-third of the citizens surveyed believed that consumption of three or more

drinks daily was safe during pregnancy," it is important to emphasize that it is prudent to abstain from alcohol intake during pregnancy because (a) alcohol has addictive properties, that is, a few occasional drinks may lead to heavy drinking, (b) the variability of the individual's metabolic "detoxification" of alcohol, that is, a small amount of alcohol may be "harmless" in one situation but may have teratogenic effects in another, (c) the difficulties to determine subtle effects of moderate maternal alcohol intake in the offspring, that is, minor dysmorphic features and/or learning disabilities, and (d) guilt and other emotional consequences that may be associated with alcohol intake during pregnancy.

Fetal alcohol syndrome has been reported worldwide, and it occurs in almost every ethnic group and at all socioeconomic levels. The incidence of fetal alcohol syndrome in the population has been estimated at 1 to 3 cases per 1,000 live births. Brandt (1982) indicated that there will be 1,800 to 2,400 new cases of fetal alcohol syndrome and about 36,000 new cases of fetal alcohol effect each year in the United States.

Fetal alcohol syndrome and fetal alcohol effect are, of course, totally preventable. Extensive education campaigns should bring to young females' attention the effect alcohol may have on the unborn child. Although they have heightened the awareness about alcohol in pregnancy, public education campaigns have had little impact on those women who drink heavily and whose children are at greatest risk. Much work needs to be done in the educational efforts so that in the future children will not suffer the adverse effects of chronic alcohol intake of their mothers during pregnancy.

Phenylketonuria

Phenylketonuria was discovered in 1934 by a Norwegian physician. (Folling, 1934). In the early 1960s, screening programs were introduced throughout the United States, and, since then, most infants with phenylketonuria have been identified and treated early in life. Whereas previously individuals with phenylketonuria usually became severely retarded, today, with appropriate dietary measures, they can grow up to become productive citizens.

Since the introduction of screening programs, we have learned of several subtypes of phenylketonuria. If a screening test is positive, the child could have classical phenylketonuria, atypical phenylketonuria, transient hyperphenylalaninemia, dehydropteridine reductase deficiency, or biopterin synthesis deficiency. An increased phenylalanine

blood level in the newborn may also indicate that the mother has phenylketonuria as her phenylalanine is transported across the placenta in increased amounts into the fetus.

Although there may be new methods of treatment in the future, at the present time the only effective treatment for individuals with phenylketonuria is a phenylalanine-restricted diet. In addition to the commercially available formulas, there are other food items, low in protein, that are offered to individuals with phenylketonuria. It is of paramount importance that the diet for the person with phenylketonuria has sufficient phenylalanine that allows adequate protein synthesis and normal brain growth; however, it should not provide too much phenylalanine that may have an adverse effect on various tissues, in particular on the central nervous system.

Dietary therapy, if started early in infancy, prevents mental retardation, neurologic complications (seizures, hypertonia), skin disorders (eczema), and growth retardation. Despite the dramatic benefit from the dietary management, some children with phenylketonuria do not develop in an entirely normal fashion. Many children have visual motor deficits, difficulties with mathematics, and other learning disabilities.

Most children with phenylketonuria adjust well to their special diets. As Finney and Friman indicate, some patients with PKU, however, have difficulties adhering to the phenylalanine-restricted diet, in particular if people in their environment tease them or make derogatory comments relating to their diet. In such circumstances, educational and behavior modification interventions as outlined by Finney and Friman are very useful.

Since Dent (1956) had suggested that high blood phenylalanine concentrations in mothers with phenylketonuria may interfere with central nervous system development of the fetus, many reports on maternal phenylketonuria have been published in the medical literature. These articles indicate that the majority of children born to phenylketonuric mothers untreated during pregnancy are mentally retarded, have congenital anomalies, low birthweight, and are microcephalic (Pueschel, 1985). In a review of the world literature on maternal phenylketonuria, Lenke and Levy (1980) found the frequency of mental retardation in offspring of mothers who had phenylalanine blood concentrations of more than 20 mg/dl was 92%.

Fortunately, the fetal effects of maternal phenylketonuria can be prevented by providing a phenylalanine-restricted diet throughout pregnancy. Optimally, the diet should be introduced before conception, if the phenylalanine-restricted diet had been discontinued previously.

Although there is limited information available on the efficacy of the phenylalanine-restricted diet, preliminary data indicate that, if phenylalanine levels are kept between 2 and 6 mg/dl throughout pregnancy, the fetus most likely will develop normally. In order to determine whether a phenylalanine-restricted diet will protect the fetus and to answer many other questions about maternal phenylketonuria, a collaborative study that includes centers in the United States and Canada has been initiated.

We have conducted research in nonhuman primates in order to shed light on the intrauterine environment of the fetus whose mother has classical phenylketonuria (Pueschel, Boylan, Jackson, Piasecki, 1982; Pueschel, 1985).

It is of importance that young females with phenylketonuria be educated and learn about maternal phenylketonuria. The adolescent girl with phenylketonuria has to know that, if untreated during pregnancy, she may give birth to a mentally retarded child. These young females should be informed that an increased amount of phenylalanine coming from the mother will cross the placenta and may have a damaging effect on the unborn child's brain and that prevention of these adverse effects is possible.

Future research efforts should provide a better understanding of certain pathogenetic mechanisms in maternal phenylketonuria that together with an appropriate educational and counseling program for phenylketonuric females and a well-designed nutritional approach will make it feasible for women with phenylketonuria to be able to bear children safely.

Lead Poisoning

Although lead has been known to have an adverse influence on human physiologic function for many centuries (Felton, 1965), only in the past several decades has lead poisoning in childhood been recognized as a serious public health problem (Pueschel & Fadden, 1975).

In many other countries, legislation concerning lead poisoning was enacted more than 60 years ago, and childhood lead poisoning as it is known in the United States is practically nonexistent in those countries (Pueschel & Fadden, 1975). Not until 1969 were bills introduced in the United States Congress to provide federal assistance to local governments for projects involving the detection and treatment of lead poisoning in childhood. After lengthy public hearings and discussion in

various committees, a compromise bill, the Lead-Based Paint Poisoning Prevention Act, was passed by the House and Senate and signed into law by the President in January 1971. Unfortunately, programs authorized by this act never gained momentum because of the apparent indifference of the administration to childhood lead poisoning and the limited federal appropriations made available.

In the 1970s, many active citizens groups became involved in the movement to eradicate lead poisoning. Subsequently, many states and local communities initiated lead poisoning prevention programs that dealt not only with identification of children with an increased lead burden but also with etiological factors because most children with lead poisoning were found in rundown neighborhoods where peeling paint and broken plaster prevailed (Pueschel, Kopito, & Schwachman, 1972).

Most often, lead poisoning in childhood is due to habitual eating of lead-containing peeling paint and broken plaster. The children are usually between the ages of 1 and 6 years, living in areas where old buildings still have lead-containing paint. Lead poisoning is also associated with pica, that is, the eating of nonfood substances by young children beyond the physiological oral stage of early childhood. These children often lack adequate supervision, and their parents are rarely aware of the harmful effects caused by the ingestion of lead-containing materials. Although lead poisoning has traditionally been felt to be a disease of poverty, primarily occurring in inner-city slums, there are also reports of increased lead burden in children living in rural and suburban areas (Annest, Pirkle, Makuc, Neese, Bayse, & Kovar, 1983).

The problem of childhood lead poisoning is so clearly defined that it would seem that its prevention should be rather straightforward. Yet, it is not, because the problem of lead poisoning is routed in social, educational, economic, medical, technical, and political factors. An effective attack on the total problem of childhood lead poisoning is an exercise in environmental management. It requires the mastering of all resources of the community, including the medical profession, the communications media, those who manage housing, municipal officials, community leaders, and the residents themselves. All must function together as an integrated force to obtain maximum results. The optimal approach to the control of childhood lead poisoning involves three components:

1. The professional community, as well as lay people, needs to be educated of the dangers of lead poisoning and its prevention. Thus public education through all media of communication should emphasize the dangers of lead poisoning in childhood and acquaint the public with the epidemiology and the symptoms of the disease.

2. Children at risk should be located, screened, and treated when necessary.
3. The environments with lead paint must be identified and deleaded.

In addition, the intake and mouthing of nonfood substances, including lead-containing materials, can be successfully treated using intensive behavioral procedures as outlined in the previous chapter by Finney and Friman.

It is important that all children be screened annually for lead poisoning. Also, environmental improvement, that is, deleading of housing units with lead paint, is a most critical element in the prevention program. Moreover, it is paramount that the federal as well as state and local legislature become concerned with lead poisoning. State laws on lead poisoning should require the establishment of a statewide program for the prevention, screening, diagnosis, and treatment of lead poisoning.

Lead poisoning is a manmade disease and, as such, it is one of the truly preventable causes of developmental disabilities. It is imperative that the public, legislators, physicians, and other health care professionals recognize the seriousness of the problem and that aggressive screening, treatment, and environmental control programs be utilized. Future research to better define the risk of increased burden of lead is needed to further our understanding of the role of lead in subtle neurologic dysfunction.

Accident Prevention

Accidents remain the main cause of death in children over 1 year, as well as the cause of much morbidity. According to the National Center for Health Statistics (1981) of the Department of Health and Human Services, each year there are about 37,000 accidental deaths in people under the age of 25. Twenty-three thousand of the 37,000 deaths are due to moving vehicles.

Beside secure vehicle construction, restraining people within automobiles remains a promising way to decrease severe injuries. Passive restraint systems involving automatic seat belts for occupants or air bags that inflate on impact are not as widespread as systems that require some degree of active participation by the occupants. Among the effective active restraints available in the United States, infant seats are made to accommodate children of up to 20 lb or 26 in. in height. Toddler

seats are suitable for larger children up to about 4 years of age, and seat belts should be used for older youngsters and adults.

To date, consumer education has not been particularly effective in preventing automobile accidents. Since 1968, the Physicians for Automotive Safety has attempted to encourage other physicians to stress automobile safety to their patients and parents. A survey reported in 1976 from southern California indicated, that of 192 board-certified pediatricians, only 3% discussed automobile safety at each visit, and only 39% annually (Lieberman, Emmet, & Coulson, 1976). A report from St. Louis, in 1977, indicated that only 5% of parents recalled their physicians mentioning automobile restraints at any time in the past (Simons, 1977).

Attempts at decreasing childhood automobile death by legislation appear more promising; perhaps they are not so dependent on convincing individual parents or other people that automobile restraints are worthwhile. One other legislative approach has been the development of standards for a child automobile restraint device, a process that has taken many years. In 1966, the United States Congress authorized the Department of Transportation to set safety standards for the design of motor vehicles and related equipment. The 1971 standards for child seating systems did not, however, apply to infants unable to sit up without support and, furthermore, did not require that restraint systems be tested in simulated crashes but only that they be shown to withstand static forces (Shelness & Charles, 1975). Since January 1981, the department has required all children restraint systems be dynamically tested in crashes.

There is still an obvious need notably in the United States for more legislative involvement in child automobile safety, including more penalties for drunken drivers and perhaps increases in the minimal age of driving.

There are, of course, many other accidents that contribute to the unfavorable statistics of morbidity and mortality in young children. Many of the accidents lead to disabling conditions, including insults to the central nervous system. Automobile accidents are the most frequent serious accidents of young people, and, to date, the most successful approaches to accident prevention have tended to be legislative.

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CHAPTER 15

Training Parents in Behavioral Medicine Techniques for the Chronic Care of Their Developmentally Disabled Children

Carol Lewis and Ronald S. Drabman

Behavioral medicine involves the application of behavioral techniques to health-related problems. Although the ultimate goal of behavioral medicine may be self-control of health-related behaviors by the child, in many instances it is more practical to use some responsible other, such as a parent, to bring about the behavior change. This is particularly true with special populations such as developmentally disabled children. With these children, behavior change is often most effectively brought about through the use of outside agents, such as parents, nurses, aides, or teachers. When children, parents, or significant others are taught behavioral techniques to control developmentally disabled children's problematic behavior related to health status, this subset of behavior therapy is referred to as behavioral medicine or behavioral pediatrics with developmentally disabled children.

The health-related issues that behavioral techniques can be used to address are varied. They include insuring intake of adequate nutrients

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and prevention of physical problems such as muscle contractions and pressure sores. Behavioral medicine techniques can also be used to help developmentally disabled children reach their potential in physical development. For example, the physical mobility of these children may be increased by teaching them how to use a wheelchair, walk with a walker, crutches, braces, or independently. They may be taught to use various pieces of equipment as aids for communication, eating, bathing, and a number of other self-help skills.

These and other problems within the developmentally disabled population have been addressed through the use of a number of different behavioral medicine techniques. Specific behaviors are taught to parents and children through modeling, praise, feedback, and behavioral rehearsal. Compliance is maintained through reinforcement schedules. Inappropriate behaviors that might affect physical well-being are eliminated through the use of extinction or punishment paradigms.

The behavioral medicine practitioner works alongside the medical staff. The cooperation of the two specialties is very important to the effectiveness of the behavioral medicine intervention. The task of the behavioral medicine practitioner is to help the medical staff achieve their goals for the patient. The medical staff can also provide the behavioral medicine practitioner with extremely important information such as medical contraindications for the use of certain reinforcers and punishers. The behavioral medicine practitioner may also work closely with other professionals such as physical, occupational, recreational, and speech therapists. Cooperation among the specialties increases the likelihood of treatment success.

We briefly review the literature on parent training of their developmentally disabled children in the following areas as they are concerned with health issues: developmental gain, physical therapy, occupational therapy, and pressure sores. Next we look at work aimed at discovering the effective components of packages designed to train parents and the effects of training and behavior change on the families in question. Then the clinical significance and implications of this literature are considered. Important issues in developing such programs are discussed, and future research directions suggested.

Target: Developmental Gain

Parents have been trained to facilitate the developmental progress of their developmentally disabled children. General physical develop-

ment is a very important target for behavioral medicine. Minor, Minor, and Williams (1985) used a participant modeling technique to teach 14 parents to be the primary developmental skill programmers for their developmentally disabled infants. Six months after pretest, the participant modeling group showed a mean developmental gain of 5.16 months. A comparison group of children (sample size of only 5) who received more traditional clinic-based treatment showed a mean developmental gain of 2.03 months.

Developmental education of parents with developmentally disabled children was compared to child management education by Moxley-Haegert and Serbin (1983). Parents in the developmental education group conducted significantly more training sessions with their children, and these children showed significantly more developmental gain than the child management group or a no-education control group.

In summary, these two studies indicate that developmental progress can be facilitated by training parents in developmental skill programming or educating them about appropriate development.

Target: Physical Therapy

Physical therapy is a very important component of the treatment of developmentally disabled children. These children often need to be stretched, taught particular physical skills that they have not developed spontaneously, taught to use wheelchairs, walk, or simply taught to tolerate therapeutic seating arrangements. A small number of studies have looked at the use of behavioral techniques to facilitate these tasks. Only a few have used parents as behavior change agents. However, the other studies potentially could be extended to include parents. Effects of the behavioral medicine techniques are evaluated by looking at such things as changes in the range of motion, number of steps able to walk with or without various aides, and length of time therapeutic seating is tolerated. Physical therapy evaluations generally provide the guidelines for developing the goals of treatment.

Body position is very important for wheelchair-bound developmentally disabled children. A checklist has been developed for monitoring correct position in nonambulatory children who are not able to change position independently (Stephens & Lattimore, 1983). One investigation (Lattimore, Stephens, Favell, & Risley, 1984) showed that the use of this checklist by a supervisor in an institutional setting significantly increased the correct positioning of all target children. Another

experimental condition in which information was provided for direct-care staff through a workshop did not significantly affect the positioning of the children.

In other investigations looking at body position, Grove and his associates (Grove, Dalke, Fredericks, & Crowley, 1975) established correct head position in mentally and physically handicapped children through the use of contingent reinforcement. Biofeedback was used by Woolridge and Russell (1976) to correct the head positioning of a child with cerebral palsy.

Use of adaptive seating devices has been shown to be associated with positive behavioral changes in multihandicapped developmentally disabled children (Hulme, Poor, Schulein, & Pezzino, 1983). Wheelchairs, travel chairs, or strollers with custom-made adaptations were used by the children for 1 or more years to produce greater postural stability. With the equipment, the children went to significantly more new places in the community, spent less time lying down, sat more, became better able to sit upright rather than lean to one side, spent less time in the bedroom, became better able to visually track an object, better able to grasp objects independently, and improved in feeding.

A number of different behavior modification procedures were used by Gouvier and his colleagues (Gouvier *et al.*, 1985) to reduce resistance to ambulation in physically disabled patients. Resistance to ambulation often arises due to fears, such as that of falling. Such fears are reality based in that falls sometimes result in broken bones. Resistance can result in extremely slow progress in ambulation training. Some children are unwilling to proceed with ambulation training at all. Modeling and contingent music successfully increased the ambulation of both patients studied. Progressive desensitization has also been used to reduce the resistance to walking in a rehabilitation setting (DiScipio & Feldman, 1971).

A 5-year-old mentally retarded child with spina bifida was taught to use crutches to walk to all of his classes through the use of contingent root beer and noise (noise was a reinforcer; Horner, 1971). Contingent social reinforcement was used with a 4-year-old child with spina bifida to teach her to ambulate independently with braces and crutches both in physical therapy clinic and at home where parents served as therapists (Manella & Varni, 1981).

Two studies have looked at whether parents can carry out physical therapy exercises effectively at home with their physically handicapped children. Slight to marked changes in the movements of target joints were found after parents were taught simple physical therapy procedures (Rosenbaum, Keane, Drabman, & Robertson, 1981). In another

study (Gross, Eudy, & Drabman, 1984), physical therapists taught parents of three developmentally disabled children procedures to elicit use of a target limb. Parental use of reinforcement (toys, fruit loops, praise) for desired behavior resulted in an improvement in extension of that limb.

Now that it has been shown that parents can conduct physical therapy exercises with their child at home, other issues arise. These include how motivated parents are to perform these tasks and how they felt about doing them. Von Wendt and his colleagues (Von Wendt, Ekenberg, Dagis, & Janlert, 1984) conducted a survey of parental involvement in and problems with physical therapy programs performed at home. They found that parents who were primarily responsible for their children's physical therapy did not feel overstrained by this responsibility. They also felt that they were receiving adequate supervision and emotional support for these activities. In a behavioral medicine framework, the physical therapist would determine what exercises need to be done, for instance, and demonstrate those to the parent and behavioral medicine practitioner. This practitioner might then proceed to spend more time teaching procedures to the parent using behavioral techniques, teach the parent the best way to produce the behavior in the child, and work on issues of both parent and child motivation.

There is some dispute about the frequency of therapy (physical and occupational) necessary to produce maximal benefit in the treatment of developmentally disabled children. Jenkins and Sells (1984) found that children treated with motor therapies one and three times per week showed equivalent gain in gross motor skills. Conversely, Campbell, McInerney, and Cooper (1984) found that children who were provided with opportunities to perform target behaviors throughout the day showed faster motor improvement than when provided the opportunity only during therapy. It may be that differences in frequency of treatment are discernible only when daily and less than daily treatment are compared. Further research is indicated to elucidate the effects of this treatment variable.

The frequency of treatment needs to be determined on an individual basis with guidelines provided by the physical therapist. Practical issues need to be considered, such as how much time the parent in question feels he or she has to devote to working with the child. Several arrangements may need to be tried. Assessment is a very important part of this process. The target skill should be monitored on a regular basis. The pattern of change in this skill should be important in determining the frequency of treatment.

In summary, parents have been trained to use behavioral medicine

techniques effectively to conduct physical therapy exercises with their developmentally disabled children. This literature should be extended to teaching parents to work with their developmentally disabled children on adaptive seating, body positioning, and ambulation. Parents may also be able to use these techniques to deal with other factors such as resistance to and fears of ambulation.

Target: Occupational Therapy

Occupational therapy has much to offer developmentally disabled children in the areas of development of self-help skills such as dressing and feeding and in the training of gross and fine motor skills. Occupational therapists have traditionally given parents activities to perform with their children at home (Anderson & Hinojosa, 1984). The problem addressed is sometimes a skills deficit and sometimes physical limitation. For instance, a child may not have developed the motor skills necessary to feed himself, or he may not have sufficient motor control to use eating utensils independently. In the former case, therapeutic eating may be used to enhance development of the appropriate muscles. In the latter case, aides may be used to maximize the independent eating of the child. Behavioral medicine utilizes functional assessment of the particular problem in order to determine the nature of the problem. This involves reducing the problem to its simplest components and studying these to determine their interrelationships. A variety of different causes can result in similar presenting problems. Functional assessment allows the therapist to determine if the presenting problem is due to a skills deficit, noncompliance, fear, physical limitations, or other etiologies.

An interesting application of self-help skills in occupational therapy is bathing independence. Shillam, Beeman, and Loshin (1983) worked on this skill area with 19 wheelchairbound or bedridden patients in a rehabilitation center. All 19 received some bathing equipment such as tub benches, bathtub chairs, shower chairs, or wheelchair shower-commodes. Flexible hoses, long-handled sponges, and grab bars were also used. All patients showed increases in bathing independence posttherapy. This was statistically significant for the group as a whole. The decrease in bathing assistance required was beneficial both for the patient's self-esteem and because it reduced family or paid-aide time needed for this activity.

In summary, there is only a small literature on training parents in occupational therapy skills to use with their developmentally disabled

children. This is in spite of the fact that parents are traditionally given occupational therapy exercises to use with their children at home.

Target: Pressure Sores

A very serious potential problem for wheelchairbound developmentally disabled children or those wearing splints or braces is pressure sores (also called bed sores, decubitus ulcers, and ischemic ulcers). They result from skin and underlying tissues being traumatized by pressure, shear, or friction (DeLisa & Mikulic, 1985). Healing may be a lengthy process, particularly if the ulcers become infected. Infection may be difficult to diagnose if deep tissue is involved (Sugarman, 1985).

Pressure sores can be prevented through proper care. Once they have developed, treatment may be expensive in terms of time, money, and pain. For example, pressure sore development may result in a wheelchairbound child's having to lay flat on his or her stomach until the sore has healed. Behavioral treatments that address this problem show clearly how behavior change can prevent medical problems.

A number of devices have been suggested to help prevent pressure sores. At least 12 different kinds of beds and mattresses and 8 kinds of wheelchair cushions have been developed (DeLisa & Mikulic, 1985). Once the sores are present, at least 14 physical agents and 38 topical agents have been reported as appropriate treatments (DeLisa & Mikulic, 1985).

Obviously prevention is preferable to treatment. In addition to the special cushions mentioned before, a behavioral program has been developed to prevent development of pressure sores in wheelchairbound individuals. This program involves the patient doing push-ups in the wheelchair at regular intervals (at least one every 10 to 20 minutes) in order to relieve the pressure (Malamet, Dunn, & Davis, 1975). In one study (Malamet, Dunn, & Davis, 1975), providing the patient with information about what he should do did not result in a sufficient number of push-ups to prevent pressure sores. Several devices have been developed to facilitate the execution of these wheelchair push-ups at an effective rate. One such device sounds a 30-second alarm at the end of any 10-minute interval in which a push-up is not done (Malamet, Dunn, & Davis, 1975). The patient may avoid the alarm by doing a push-up which then starts the next interval. The alarm can also be terminated by doing a push-up. Push-ups must be of at least 4 seconds duration. This device also records push-up frequency. It has proven effective in

producing an adequate number of push-ups in at least one study (Malamet, Dunn, & Davis, 1975).

A more recent study (Merbitz, King, Bleiberg, & Grip, 1985) has shown that simply electronically recording frequency of lift-offs yields wide variability in frequency. The relationship between lift-off frequency and pressure sore development also appears to be somewhat variable between individuals (Merbitz *et al.*, 1985). Future research with the developmentally disabled should extend this literature to include using parents as the behavior change agents.

In summary, pressure sore development can be avoided through the application of appropriate behavioral medicine techniques. Several devices have been designed to assist in this process. Reports have not as yet included parents as the behavior change agents in this area. This application should be explored.

Other Issues: Analysis of Treatment Components

Several investigators have begun to look at what variables produce the most effective treatment package for training parents of the developmentally disabled. Even when the problems in question were not health-related, the findings about parent training in general are relevant to this aspect of behavioral medicine. Hudson (1982) used four parent-training conditions in his investigation of factors that facilitate gain in developmentally disabled children. The groups were verbal instruction, verbal instruction plus teaching of behavioral principles, verbal instruction plus the use of modeling and role playing, and a wait-control group. Results indicated that parents need to be taught with techniques that directly shaped their behavior (modeling and role playing) in order to affect their children's behavior.

Baker and his group in the UCLA Project for Developmental Disabilities also looked at the effectiveness of various formats for parent training (Brightman, Baker, Clark & Ambrose, 1982). Parents of 66 moderately to severely retarded children (aged 3–13 years) participated in either group or individual training to reduce behavior problems and teach self-help skills or were assigned to a delayed training control group. After 3 months of training, trained parents knew more about behavior modification and were better at using these principles than control parents. Children of trained parents tended to have reduced behavior problems but showed no differences on self-help skills. Further, there were no differences between parents trained using an individual versus a group format.

The UCLA group has also looked at other important aspects of parent training of developmentally disabled children such as the effect of nonspecific factors on treatment gains and the prediction of treatment outcomes. Baker and Brightman (1984) taught one group of parents behavior modification knowledge and skills and another group advocacy knowledge and skills. After approximately 10 weeks of treatment, each group of parents showed gains but primarily on instruments particular to their training program. This demonstrates the specificity of the effects on treatment gains.

Discriminant analysis has been used to classify parental proficiency at conducting a behavioral program with their developmentally disabled children (Clark & Baker, 1983). Parents of 103 moderately to severely retarded children were classified as either high or low proficient after participating in a 10-week group treatment aimed at increasing their children's self-help skills and reducing their behavior problems. The model correctly classified 76% of the parents. Low proficient parents were of lower socioeconomic status, had expected greater problems in teaching their children, and had less previous experience with behavior modification. At 6-month follow-up, factors predictive of low rate of followthrough were marital status (unmarried), low proficiency at the end of training, and low likelihood of teaching their child prior to the program.

Remediation for parents showing low proficiency at the end of training was conducted by Baker and McCurry (1984). These parents were taught skills in a school-based group format supplemented by live modeling, observation of teaching, individual supervised teaching, and videotaped feedback (Brightman, Ambrose, & Baker, 1980). There were significant treatment gains, but maintenance of these gains over 6 months varied according to teaching proficiency at the end of treatment. Skill levels were low at all points, however, compared to the sample used to develop the classification formula (Clark & Baker, 1983).

Family Effects

Most of the parent training research to date, health-related and not, has dealt with mothers. One study, however, has looked at transfer of training effects from a mother to a father (Adubato, Adams, & Budd, 1981). Written and verbal instructions, modeling, and feedback were used to teach the mother of a 6-year-old developmentally disabled boy how to increase her son's independent dressing skills. The mother was

then instructed to teach her husband what she had learned. Results showed that both parents used the procedures, and their child's behavior showed improvement. Training generalized somewhat to nontarget behaviors and maintained over 2 years.

Another study (Sandler, Coren, & Thurman, 1983) taught mothers behavior modification skills to use with their developmentally disabled children but assessed both mothers' and father's knowledge about these procedures, skill in using them, and attitudes toward themselves, their spouse, and their developmentally disabled child. Mothers gained knowledge and skill. Fathers did not. Further, as the child's behavior improved, mothers' attitudes tended to become more positive, whereas fathers' attitudes tended to become more negative.

The interactive nature of family involvement in therapeutic endeavors and the needs and emotions of the family members was also noted by Bricker and Casuso (1979). They found that training parents to perform occupational therapy tasks with their children did not produce the desired effects until the needs of the family were addressed. In another study (Tyler, Kogan, & Turner, 1974), mothers showed more negative behaviors after they began performing occupational therapy exercises at home with their cerebral palsy children.

These studies indicate the intricate connection between family involvement in child treatment and the emotional status of the family. Although it appears that one parent can teach his or her spouse the behavioral skills he or she has learned, the literature indicates that he or she may need to be specifically instructed to do so. Whether parents use these skills is related at least in part to their emotional status, particularly with regard to their developmentally disabled child. Chronic care for a child with special problems takes its toll on primary caretakers such as parents. At some level, they may resent their developmentally disabled child for reasons like requiring so much care, not being the "perfect" baby they expected, or draining their financial resources. These feelings, whether or not acknowledged, are often accompanied by feelings of guilt for not being a completely selfless parent able to give whatever the child needs.

Generally, all family relationships are affected by the presence of the developmentally disabled child. Spouses may not devote much attention to each other or to siblings of the developmentally disabled child. Family members may not have the skills to cope with the feelings they experience. These sorts of family-related issues should be carefully assessed before any family members are involved in the behavioral medicine treatment. A mother may feel that she just does not have 3 hours a week to devote to physical therapy exercises with her child. Or, due

to her own emotional status, she may attempt to work with her child on the exercise to the exclusion of activities with her spouse and other children. The sources of emotional support available to the family member recruited for training and the feelings of all family members about the time this will require of the parent are extremely important issues. By considering these issues prior to training, one reduces the likelihood of program failure or sabotage.

Clinical Significance of Parent Training Research to Date

The parent training literature has a number of implications for behavioral medicine/chronic care of developmentally disabled children. The research has shown that many parents trained in behavioral techniques can effectively provide care and treatment for their developmentally disabled children at home with professional supervision. This care includes not only shaping appropriate/acceptable behaviors but also facilitating developmental progress, skill acquisition, and preventing physical problems.

The time required by professional staff to provide these services for developmentally disabled children can be quite extensive as well as expensive. Training parents can reduce these costs. It also may allow the child to receive needed therapy on a daily basis, an arrangement that might not otherwise be feasible. For example, a child might receive gait training or work on fine motor skills each day rather than the often once-a-week outpatient clinic visit. This may facilitate the attainment of the child's potential. It is certainly in the spirit of Public Law 94-142.

Within the traditional professional staff-based treatment model, developmentally disabled children receive only limited services. They may also require periodic hospitalization in order to have extensive services provided directly by professional staff. Effective parent training provides the opportunity for developmentally disabled children to receive more frequent treatment by parents at home under the supervision of professionals. Therefore, it has the potential of reducing the number or duration of those costly, extensive hospitalizations.

Another important advantage of training caretakers in chronic care/behavioral medicine procedures with the developmentally disabled is getting the caretakers involved in spending productive time with the children. Parents of developmentally disabled children may sometimes feel inadequate and as if their children can only "get what they need" from professionals. Further involvement in their children's care and

therapy provides opportunities for parent–child interactions that might not otherwise present themselves. This is especially true for children who are extremely delayed and unresponsive.

Caretakers will also be able to see improvement and progress in their children as they work with them on a daily basis (i.e., as misbehavior improves, skills develop, language improves). Small bits of progress that may not be noticed by the various professionals occasionally seeing the child might be salient and noticed by the caretaker. This positive feedback will be reinforcing to the caretaker and may increase motivation to both work and interact with the child.

As primary caretakers of developmentally disabled children become responsible for more aspects of the chronic care of their children, new issues will present themselves. For example, specific therapeutic interventions can be used throughout the day or be limited to circumscribed periods. It may be that the child shows maximum benefit from procedures used throughout the day for language, self-help skills, and physical development. However, the primary caretaker may be overwhelmed by the amount of work this requires.

Feelings of guilt about providing as much opportunity as possible for their disabled child's skill development may also play a role in caretaker decisions concerning the amount of intervention to offer. Additionally, the caregiver may believe that because techniques to deal with problem behaviors are to be used consistently throughout the day, other interventions should be daylong as well. Other factors that might affect the amount of time the caretaker can provide are whether the caregiver (a) has other obligations on his or her time such as a job; (b) is solely responsible for the child's care or shares it (i.e., with a spouse); (c) is also caring for other children.

These issues must be handled on an individual basis. A schedule should be developed that will provide the necessary treatment for the child within the context of the other demands and stresses on the system. The scheduling issue should be negotiated. Realistic demands and appropriate expectations for outcome should be discussed. A parent who is overwhelmed will not be much help to his or her developmentally disabled child.

Implications of Parent-Based Interventions on the Medical Components of Chronic Care

Parent-based interventions for developmentally disabled children could affect a number of the medical aspects of chronic care. First, child

behavior problems may reduce the effectiveness of medical interventions. Parent-based interventions could decrease the problem behaviors, thereby increasing efficacy of the medical intervention. For example, a child cannot obtain the maximum benefit from an arm splint if he or she constantly removes it. Parent-based interventions could decrease the child's frequency of removing the splint, thereby optimizing its effect.

Parent-based behavior management of developmentally disabled children can also facilitate cooperation of the child in therapeutic activities such as physical therapy, conducted either at home or in the clinic. It is difficult to make therapeutic progress with a child who is fighting, screaming, or involved in constant self-stimulatory behavior.

Physical and occupational therapy exercises supervised by a parent may facilitate ambulation, use of arms and legs, and development of both gross and fine motor skills. Therapist-prescribed exercises might also reduce contractions of muscle groups due to the inactivity of those groups. This could facilitate the availability of the muscles for use in such activities as transferring from bed to wheelchair. Additionally, appropriately trained parents can help prevent other problems common to developmentally disabled children such as pressure sores from misuse of adaptive equipment.

Important Issues To Consider in Developing Parent-Based Interventions

An extremely important issue to consider when developing parent-based treatment programs is the ability of the parent to understand the program. Programs should be simplified and tailored to the particular parent's level of understanding. Parents should be trained until they can perform tasks to an appropriate criterion level. Their knowledge and skills should be assessed regularly to ensure that programs once learned continue to be used appropriately. Some parents may require quite frequent booster training sessions.

Parental motivation to carry out treatment programs should also be carefully evaluated. Parents may be unable to carry out some treatment programs due to work or other family demands. They may also feel themselves incompetent to carry out treatment. Alternately, they may believe that developmentally disabled children, especially those who also have a chronic illness, should not be forced to change their behavior regardless of the potential positive or negative consequences. Parents

may be attached to the medical model of treatment whereby all or most of treatment is provided by professional staff. These issues must be resolved if parent training is to be successful. The task of the behavioral medicine practitioner may be to work to alter the parents' attitudes prior to attempting programming.

Parent training programs should provide parents with several bits of information. They should instruct parents about the developmental status of their child. Appropriate expectations should be fostered about the course of development and treatment. Parents should also be educated about what skills generally are necessary before other skills can develop. For example, crawling precedes walking.

Reinforcers and punishing agents for use in parent-based interventions with developmentally disabled children should be chosen carefully. Sometimes there are medical contraindications that dictate against use of particular agents as reinforcers or punishers. The behavioral medicine practitioner must be very careful not to recommend use of any such agents. Potential disasters can be averted by checking out possible reinforcers or punishers with medical staff before program implementation. For example, the feeding status of the child should be considered when selecting food items for reinforcers. It is important to know whether the child enjoys eating, can chew, and what texture of food he or she can currently manipulate.

It is also very important to consider the particular disabilities of a child when developing a parent-based behavioral program. For example, visually impaired children should always be told what is being done and touched as much as possible. Textured toys may be very effective reinforcers for them. Other children may be producing so much self-stimulatory behavior that external reinforcers are initially ineffective. The type and extent of disability of each individual child should be considered in order to provide parents with the most effective program.

In some situations parent training may not be the treatment of choice. These include situations in which the parents are not willing/able to carry out the program even after repeated attempts to present it in its most simplified form. Parents who prove unreliable in either record keeping or maintaining contact with professional personnel supervising them should not be used as behavior change agents. Parents should also not be used as behavior change agents when they are experiencing such high levels of stress that either the program is not carried out as it needs to be or there is any potential for child abuse. The child's physical and emotional well-being should be considered and protected at all times.

In situations such as those mentioned, treatment should be con-

ducted by the professional staff. Limited professional contact is preferable to misuse of treatment techniques by caregivers.

Future Research Directions

Continued research is necessary to further elucidate which methods most effectively train parents to provide care and treatment for their developmentally disabled children. Another important research direction involves training parents in behavioral techniques applied to aspects of physical therapy, occupational therapy, and chronic care (bed sores) in which they have not as yet been used as behavior change agents.

Further research should be done to determine the frequency with which parents should be asked to perform therapeutic tasks with their developmentally disabled children. Although some research has commented on this issue (Campbell *et al.*, 1984; Jenkins & Sells, 1984) results are inconclusive. Frequency issues are important not only in considering efficacy of treatment but also in terms of parental motivation. For example, are parents more likely to perform physical therapy exercises with their child if asked to do so one time per day versus two to three times per day?

Parent compliance with home treatment programs for their developmentally disabled children is another area that should be further investigated. Once parents are taught how to reduce hitting, work with their child on fine motor skills, language, or walking, how well do they carry out these programs? Both caregiver competence and compliance need further research as they can have major impacts on efficacy of treatment. Caregiver (i.e., parent) and system (i.e., family) variables that affect competence and/or motivation to carry out programs also need further study. Generalization and maintenance of treatment gains should be investigated.

Aspects of interventions that caregivers are better able and more likely to carry out should be determined. If these can be found, then professional time and effort would best be spent teaching caregivers these aspects of behavioral medicine/chronic management. In this way, staff frustration concerning programs not carried out would be reduced. This could result in a better working relationship between caregivers and professional staff. Optimal frequency of parent–staff contact in parent-based programs should also be investigated.

Research should be conducted to assess the relative effectiveness of clinic/institution-based interventions and home-based interventions

for developmentally disabled children. Given similar programs, can children who are taught skills by their parents at home progress at the same rate as children who receive all their services directly from professionals? A comparison should also be made of the long-term cost-effectiveness of the two types of programs.

Quality of parent–child interactions should also be assessed as home treatment programs are prescribed and studied. Both parent attitudes toward their children and child attitudes toward their parents should be assessed before, during, and after parent-based interventions. These attitudes may not only affect family life but also treatment effectiveness. Research is needed to determine the most successful ways of changing parental attitudes to help them accept and actively participate in behavioral medicine techniques with their developmentally disabled children.

One last area deserving further research is the particular types of developmentally disabled children for which particular parent-based interventions are most effective. Child factors such as developmental status (physical and cognitive), type of disability, and severity and extent of disability are some of the factors that should be explored. Their effect on parental compliance should also be examined.

Conclusions

Parents and other caretakers can be trained in the effective use of behavioral medicine techniques to use with their developmentally disabled children. Parents have been trained to reduce their developmentally disabled children's behavior problems and facilitate acquisition of self-help skills and language, cognitive and physical development. Research has yet to elucidate the most effective treatment package for parent-training, although work in this area has begun.

Parent and family factors conducive to program carrythrough have also begun to be studied, but much more needs to be done. Of particular relevance are the factors that determine parental motivation to continue long-term management and treatment of their developmentally disabled children. Also, comparison of the long-term cost-effectiveness of home versus clinic/institution-based treatment and management of developmentally disabled children should have serious implications for the future.

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CHAPTER 16

Behavioral Parent Training for Families of Developmentally Disabled Children

A Behavioral Medicine Perspective

Karen S. Budd

Because of the family's integral role in the care and treatment of developmentally disabled children, strengthening the family has become an important part of habilitation (Simeonsson & Simeonsson, 1981). Behavioral parent training is one major strategy for strengthening the family by modifying problematic child behaviors, teaching basic skills, and enhancing family coping and interaction patterns (Altman & Mira, 1983; Baker, 1976; Budd & Fabry, 1986; O'Dell, 1985). The pressing needs of handicapped children often call for several types of family services, such as counseling and support, respite care, educational intervention for the child, and training in behavior management. Thus, as Kaiser and Fox (1986) commented, it is not a question of choosing behavioral parent training over other possible interventions but rather of placing parent training in the context of family needs and maximizing its effectiveness in meeting those needs.

The focus of this volume on behavioral medicine with develop-

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mentally disabled persons provides an opportunity to examine the interface of behavioral medicine and behavioral parent training as applied to families of handicapped children. One might ask what a behavioral medicine perspective has to offer the field of parent training and, conversely, what parent training has to offer behavioral medicine. Lewis and Drabman (Chapter 15, this volume) addressed these questions (primarily the latter one) by reviewing research on parents' application of behavioral techniques to control the health-related behaviors of developmentally disabled children. They described parent-mediated interventions for a variety of physical impairments, self-care deficits, and specialized health needs of handicapped children. Their review highlighted several issues implicit in treatment of chronic health-related disorders, including the need for an interdisciplinary approach, individualized analysis in the selection of intervention components, careful monitoring of skill acquisition and compliance with treatment regimens, and attention to family stressors and emotional status. Lewis and Drabman pointed out the economic and programmatic advantages of training parents as chronic care providers, suggested guidelines for judging the appropriateness of parent involvement in particular cases, and outlined areas in need of further research.

In sum, Lewis and Drabman's chapter conveyed that behavioral parent training has much to offer in the treatment of health-related behaviors in developmentally disabled children. I heartily endorse their conclusions, and my own comments will support this point of view. In addition, I believe it may be fruitful to consider the related issue of what a behavioral medicine perspective has to offer the field of parent training. In the ensuing discussion, a conceptualization of behavioral medicine will be offered and examined with regard to its implications for parent (and family) participation in the care and treatment of developmentally disabled children. Suggestions will be proposed of areas where attention to family factors could pay off in advancing the goals of behavioral medicine and behavioral parent training.

A Behavioral Medicine Perspective

Lewis and Drabman defined behavioral medicine as a subset of behavior therapy involved with the application of behavioral techniques to health-related problems. Whereas this disciplinary perspective includes much of what behavioral medicine practitioners do to treat health problems, it fails to capture the diversity of variables contributing to a

functional analysis of health-related behavior. Behavioral medicine has been conceptualized (e.g., Russo & Varni, 1982; Schwartz, 1982; Schwartz & Weiss, 1978) as a comprehensive, multidimensional, and interdisciplinary scientific framework for the study of health and illness. Its biopsychosocial framework derives from a recognition that disease processes involve multiple causes, interaction patterns, and effects, and that considerable individual variation exists in the expression of disease (Russo & Budd, 1987). There is evidence that distal events and systems factors can play an integral role in health and illness despite the indirect nature of their relationship with overt behavioral symptoms (Russo, Tarbell, & Follansbee, in press). The biopsychosocial perspective of behavioral medicine shares many conceptual features (e.g., multilevel analysis, bidirectional effects) with ecological and systems analyses proposed for use in family treatment programs (e.g., Kaiser & Fox, 1986; Patterson, 1982; Wahler & Hann, 1986), with the major distinction relating to the attention paid to health-related variables.

In this broader conceptualization of behavioral medicine, many aspects of family relationships can be seen as potentially relevant to the care and treatment of a developmentally disabled child. Prenatal, perinatal, and postnatal factors, early parent–child bonding, family reactions to the initial diagnosis of developmental disability, parent teaching and disciplinary practices, family attitudes and expectations toward the child's handicaps, family beliefs about and practices in coping with illness and abnormality, emotional stressors impinging on the family, and the effects over time of the child's chronic care needs are a few examples of potentially relevant variables. Likewise, several aspects of the developmentally disabled child (health status, nature and extent of disabilities, behavior problems, adaptive skills) influence the family's role as caretaker and treatment agent.

In the ensuing paragraphs, a brief discussion is offered of six areas in which a developmentally disabled child poses special challenges to the family. These areas, drawn from clinical experiences with families in health settings and the writings of other professionals, illustrate potential targets for parent training from a behavioral medicine perspective. They are not meant to be an exhaustive list but rather as a sample of intersecting concerns between behavioral medicine and behavioral parent training.

Recognizing and Understanding the Child's Condition

Although some developmental disabilities (such as Down's syndrome) are diagnosable at birth, many conditions (such as autism, sen-

sory impairments, or brain damage from seizures) become apparent only after months or years of life. Even when a child's physical and/or neurological abnormalities are known, the medical, cognitive, and behavioral sequelae usually cannot be specified with precision. Thus, it often becomes difficult to identify problems and to distinguish between medical and behavioral disorders. Gourash (1986) observed that medical symptoms can masquerade as behavioral disorders, as when deafness appears as unresponsiveness, gastroesophageal reflux as vomiting, decreased pain sensitivity as self-abuse, or psychomotor seizures or brain tumors as any number of unusual behaviors. Likewise, behavioral disorders such as pseudoseizures, skin rashes from repetitive acts, and abdominal distention from air swallowing have misleading symptoms that at first suggest a medical origin (Gourash, 1986). Some developmentally disabled children also show atypical sleeping patterns, strong reactions to particular sensations, hyperactivity, or other aberrant behaviors for unexplained reasons.

Parents bear the brunt of caretaking consequences prior to accurate identification of these conditions, particularly when the management techniques they try are not effective. Many parents relate stories of intense frustration and uncertainty in attempting to deal with behaviors in their handicapped children they do not understand and cannot control. The preponderance of behavioral parent training has been directed at teaching parents to manage already identified problems of children; however, services also could be directed at teaching prompt recognition and reporting of behaviors that may need professional intervention. Schroeder, Gordon, Kanoy, and Routh (1983) described a call-in service for parents in a pediatric psychology practice designed to screen parent concerns and provide immediate suggestions for management as well as to facilitate clinic appointments for problems that merited further examination. For families of children with known or suspected developmental disabilities, parent education and access to consultation could be useful strategies to improve the initial recognition of problems, which in turn should expedite accurate diagnosis and effective intervention.

Providing Care of the Child's Special Needs

Once the extent of developmental problems affecting a child is evident, family members also can encounter difficulty balancing the multiple needs of the child. Examples come to mind of children who appear

to be overly protected (such as being restricted in school, play, or stimulation activities) in order to avoid possible dangers as well as children (especially those with severe motor impairments or highly impulsive behavior) who need more stimulation or supervision than they receive in the home. These situations may result from a lack of full understanding of the child's problems by the parents, the need for alternative care services, or the need for help in identifying safe means of care that still foster the child's opportunities to develop independence. Behavioral parent training can be a vehicle for teaching parents to analyze the child's overall needs in relation to available resources and to problem-solve solutions (Blechman, 1985). A frequent variant of parent training, in which services are provided in the natural environment, is well suited to the task of balancing multiple family needs and teaching problem-solving strategies.

As Lewis and Drabman document in their review, parent-mediated interventions have provided therapy, taught skills, and facilitated adaptive equipment use with handicapped children. When children have special medical needs, parents receive instruction from medical personnel in carrying out health care procedures (e.g., provision of nutritional intake for children on nonoral tube feedings, care of ostomies, and management of seizures). Typically these procedures are taught informally to parents, without systematic evaluation of parents' ability or confidence in applying them in the home. Recent research with various other pediatric health conditions (e.g., hemophilia, diabetes, preparation for hospitalization) has shown that parents can be trained as health care agents (Carney, Schechter, & Davis, 1983; Sergis-Deavenport & Varni, 1983; Zastowny, Kirschenbaum, & Meng, 1986). Parents' proficiency in learning to implement medical procedures with developmentally disabled children represents an area where behavioral parent training research could help to extend behavioral medicine efforts in treatment.

Managing Behavior Problems

Another concern facing some families of handicapped children relates to the prevalence of behavior problems. The likelihood that developmentally disabled children will display behavior problems is considerably higher than for normally developing children. In a review of epidemiological studies, Schroeder, Mulick, and Schroeder (1979) reported prevalence rates for behavioral disturbances (e.g., physical aggression, temper tantrums, self-injury, stereotypic responses, and

noncompliance) of 20% to 40% for retarded persons compared to around 5% for the general population.

Although the causes of serious behavior problems in developmentally disabled children are not well understood, there is little argument about the distressing effects of the behaviors on the family. Budd and Fabry (1986) remarked that parents often report feeling discouraged, exhausted, or emotionally distant from their child as a result of contending perpetually with disruptive and oppositional behavior. Professionals have commented on the negative self-perceptions held by family members, who interpret the handicapped child's deviant behavior as an indication of their own inadequacy (Heifetz, 1977; Paul & Porter, 1981). Without a normative reference for developmental or behavioral expectations, families are at risk to demand too much or too little of the handicapped child and to lack confidence and consistency in their own disciplinary practices.

A considerable body of behavioral parent training research has demonstrated that parents can modify a host of problematic behaviors of developmentally disabled children. Interventions have been directed at noncompliance, tantrums, sleep disturbances, high activity rates, aggression, self-injury, stereotypic behavior, pica, rumination, and other deviant responses (Altman & Mira, 1983; Baker, 1976; Budd & Fabry, 1986). However, the chronicity of developmental disorders can take its toll on families over time, such that they may need outside help with behavioral management as the disabled person becomes older, during times of family stress, or as an intermittent respite from caretaking demands. Most behavioral parent training programs have been directed at short-term interventions with young handicapped children. It would be fruitful to extend applications to the family's management of behavior problems across the life span of developmentally disabled persons.

Supporting Youth's Self-Management and Adaptation

Whereas the preceding sections focus on the provision of direct care and treatment by parents, many developmentally disabled children will make sufficient gains that they can gradually take on some responsibility for their own care. Parents often have difficulty determining when and how to reduce their assistance and how much responsibility to expect of handicapped youth. Parent–child learning histories also play a role in maintaining established practices even when a youth may be capable of more independent behavior. As adolescence approaches, issues of

sexuality, peer relationships, vocational pursuits, alternative living arrangements, guardianship, and even parenthood arise, and parents may well feel ill-equipped to make informed decisions (Greenspan & Budd, 1986). Little parent training research has been directed at assisting parents in shaping more independent skills over time or in approaching issues of adulthood. These topics would be fruitful areas for interdisciplinary study with rehabilitation specialists, attorneys, social service providers, and physicians, who also deal with families of developmentally disabled persons in regard to these issues.

Coping with Medical Problems and Deterioration

The alternative side of the life-span perspective cited before concerns those youths whose medical conditions worsen over time, leading to repeated hospitalizations, physical weakness, decreased functioning, or death. For some developmental disabilities (including metabolic disorders such as Tay-Sachs or Hurler syndrome), a pattern of deterioration can be predicted with certainty, whereas other conditions (such as Down's syndrome and seizure disorders) entail increased medical risk for specific health problems without a uniform pattern. Families endure emotional, physical, and economic stress related to the developmentally disabled youth's chronic health problems, and few services are available to assist families through these periods of difficulty. Recent research with children who have cancer, cystic fibrosis, or other life-threatening conditions has identified several areas of family need and helpful approaches to assisting families (Katz & Jay, 1984; Koocher, 1984). This literature is relevant to those who work with families of developmentally disabled youth undergoing similar experiences. The cumulative effects of chronic deterioration and strategies to assist families in dealing with the patterns over time are areas in need of professional attention.

Balancing Personal and Family Priorities with Children's Needs

Throughout this discussion, the extensive needs of the developmentally disabled child are regarded as a given. Yet what is not as explicitly stated is the coexisting needs of other family members that compete for the emotions, energies, and resources of the family. Several writers (Harris, 1982; Skrtic, Summers, Brotherson, & Turnbull, 1984; Wahler & Hann, 1986) have advocated that a systems approach toward

handicapped children and their families is needed to conceptualize the multidimensional nature of family interactions and their patterns over time. Lewis and Drabman (in this volume) also discuss areas of failure in behavioral parent training programs when families were under high emotional stress or showed motivational difficulties. Researchers in parent training are increasingly taking ecological and systems factors into account in planning and evaluating interventions (Dumas & Wahler, 1983; Kaiser & Fox, 1986), which reflects a modification in clinical and experimental tactics based on the pivotal role played by broader family issues.

In summary, six specialized concerns facing families of a developmentally disabled child have been reviewed. By considering these areas from a behavioral medicine perspective, several potential areas of parent training research were identified. These include strategies to help parents in initial identification of developmentally disabling conditions and their sequelae, matching overall child needs with resources through family problem solving, implementing specialized medical or health care procedures, fostering increased youth independence and planning for adulthood, dealing with chronic health problems and deterioration of functioning, and maintaining family integrity while meeting the child's needs. Active involvement of parents as health care agents for developmentally disabled children has direct benefits for enhancing behavioral medicine goals. It appears that the fields of behavioral medicine and parent training have much to offer each other in the study and the service of developmentally disabled children.

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CHAPTER 17

Obesity in the Developmentally Disabled

Jennifer E. Burkhart, Robert A. Fox, and Anthony F. Rotatori

Obesity, which refers to an excess of subcutaneous fat (Craddock, 1978; Craig, 1969), represents one of the most prevalent and refractory health problems occurring today (Brownell & Stunkard, 1978). In the professional area, new books on obesity are appearing with increasing regularity. Journals devoted specifically to this topic, for example, *International Journal of Obesity, Journal of Obesity and Weight Regulation*) along with numerous convention papers and workshops also add to the burgeoning professional literature. In the public area, a literal deluge of material awaits the interested individual in the form of popular trade books, over-the-counter drugs, weight loss clinics, health spas, and physical fitness centers. However, in spite of the continued focus by professionals and lay persons on the issues of overweight and obesity, as late as 1977 at a conference, "Obesity and the American Public," investigators were professing their ignorance about the etiology and appropriate treatment of obesity (Kolaka, 1977). This ignorance is even more apparent in the field of developmental disabilities (DD), where a

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paucity of relevant information is available. The purpose of this chapter is to present what is known about obesity in DD individuals as well as pertinent data obtained for non-DD persons. Also, where appropriate, suggestions will be made about what we yet need to learn about this complicated area.

Prevalence

According to the National Center for Health Statistics, over 29 million Americans, between the ages of 20 and 74, are obese (Abraham, 1983). A number of variables are reported in the literature that are associated with a greater prevalence of obesity among certain subgroups. For instance, the prevalence of obesity is greater in certain ethnic groups, increases with age, and is negatively correlated with higher socioeconomic status (Stunkard, 1975). This inverse relationship to socioeconomic status is particularly prevalent in women (Goldblatt, Moore, & Stunkard, 1965) and occurs as early as childhood (Stunkard, d'Acquili, & Fox, 1972; Whitelaw, 1971). Similarly, American black females, although generally thinner than their white counterparts through adolescence, have been found to be heavier through adulthood (Garn & Clark, 1975) regardless of income (Bray, 1979).

Prevalence studies among the mentally retarded population reflects a pattern similar to the general population and suggests that the general cause(s) may not be unlike those within the nonretarded population (Fox & Rotatori, 1982). Consistent with research on the general population, recent research in the mentally retarded (MR)/DD population has revealed a greater incidence of obesity among women than men (Emery, Watson, Watson, Thompson, & Biderman, 1985; Fox & Rotatori, 1982; Wallen & Roszkowski, 1980) and a tendency for the prevalence of obesity to increase with age in both sexes (Fox & Rotatori, 1982), in contrast to earlier studies that suggested a greater incidence of obesity among retarded males than females (Polednak & Auliffe, 1976). These investigators found an incidence of 20.4% in males and 17% for females in a sample of 161 retarded, whereas Fox and Rotatori (1982) found that obesity was represented in 16% of the males and 25% of the females in their sample of 1,152 mentally retarded individuals. The difference in these studies might be attributed to the nature of the populations studied because the first study focused on an institutional population, whereas results from the second study are drawn from various residential settings.

A positive correlation between IQ and obesity has been reported in the MR/DD population for both sexes, with a higher proportion of obesity represented among moderately and mildly retarded individuals (Fox & Rotatori, 1982; Wallen & Roszkowski, 1980). These findings are in contrast to an earlier study by Kreze, Zelina, Zuhas, and Garbara (1974) that suggested an inverse relationship between IQ and obesity for women and a similar but less pronounced trend for men. However, their study included primarily average and above-average subjects with only eight subjects representing the low IQ sample (IQs less than 70), making extension of their findings to the retarded population unwarranted. In a more recent study, Emery *et al.* (1985) found a nonlinear relationship between level of retardation and incidence of obesity as evidenced by the representation of mean weights from lowest to highest in the order of profound, borderline, moderate, severe, and mild. The relationship between IQ and obesity might be attributed to the greater incidence of eating disorders associated with profoundly retarded individuals because the degree of retardation is positively correlated with the presence of a number of physical ailments (Ross, 1974). These include difficulties in chewing and swallowing (Martin, 1973) or resistance to certain foods or textures (Linschied, Oliver, Beyer, & Palmer, 1978) as well as potential abnormalities in metabolism (Crome & Stern, 1972). On the other hand, higher functioning individuals (e.g., borderline) may be more likely to initiate and carry out physical activities that burn off calories. Those individuals falling between these extremes (e.g., severe, moderate, and mild levels of mental retardation) may be more susceptible to becoming obese. Again, the multifactorial component of obesity is a logical explanation for the basis of these findings.

Wallen and Roszkowski (1980) not only found a greater incidence of overweight females in their study but found comparatively more underweight males than would be expected, given their representation in the total sample of 149 institutionalized retarded adults. To account for these findings, the authors suggested that the interaction between sex and degree of mental retardation may be associated with syndromes in which weight is a clinical feature such as Prader-Willi, Down's, Carpenter, Laurence-Moon-Biedl, and Cohen syndromes. However, in both the Wallen and Roszkowski (1980) and Fox and Rotatori (1982) studies, specific syndromes associated with mental retardation (e.g., Down's syndrome) did not account for the differences among obese and nonobese in sex, age, and mental retardation level.

Recently, Fox, Hartney, Rotatori, and Kurpiers (1985) completed an obesity incidence study with 337 MR children enrolled in a large, urban, public school system. Obesity was identified in over 22% of the sample

and was not related to sex, age, race, or mental retardation level. Based on limited comparative data (DuRant, Martin, Linder, & Weston, 1980), obesity appears to occur more frequently in children who are retarded than nonretarded children.

Social and Health Hazards of Obesity

An obese condition is associated with a variety of both social and health hazards. Individuals who are obese are likely to be the objects of discrimination and unacceptance as evidence by Canning and Mayer's (1966) finding of differential college acceptance rates between obese and nonobese students in spite of identical high-school scores. Additional disadvantages include impaired social and job opportunities, difficulty in obtaining clothes, as well as increased cost of clothing (Craddock, 1978).

The numerous health risks associated with obesity are well-known. Several research studies have revealed an association between obesity and such medical conditions as hypertension, coronary artery disease, hyperlipidemia, diabetes mellitus, carbohydrate intolerance, pulmonary and renal problems, slower physical reactions, complications during pregnancy, and higher perinatal mortality (Bray, 1976; Dawber, 1980; Kannel & Gorden, 1979; Van Itallie, 1979). Other reported health risks include increased incidence of varicose veins, arthritis, gallstone and gall bladder disease, liver damage leading to cirrhosis, and numerous complications of surgery and anesthesia (Agras & Werne, 1981). Data from the Manitoba (Rabkin, Mathewson, & Hsu, 1977), Provident (Blair & Haines, 1966), and Framingham Heart (Hubert, Feinleib, McNamara, & Castelli, 1983) studies indicate that the duration of an obese condition largely influences longevity. Overweight individuals, particularly those who are overweight at younger ages, tend to die earlier than average-weight individuals (Simopoulos & Van Itallie, 1984).

Of particular concern for professionals working with DD individuals are not only additional health risks associated with obesity in a population often plagued with physical anomalies, but also the potential for increased social rejection in light of continued efforts for normalization. Overweight may decrease the likelihood of exposure to situations that enhance development and reduce access to experiences, involving social and recreational activities with normal peers (Chumlea & Cronk, 1981). The physical discomforts associated with this condition are more likely to be overlooked or manifested through behavior problems (e.g., aggres-

sion, self-injury, tantrums) in this population, particularly in those individuals who have severe communication deficits.

Clinical Significance of Research

Etiology

A number of variables that alone or in combination are believed to be major factors in the etiology of obesity include inheritance (Bray, 1979; Withers, 1964), excessive caloric intake (Sims *et al.*, 1968), and insufficient activity level (Chirico & Stunkard, 1960; Mayer, 1955). Work in this area suggests that obesity is a complex condition with multiple precursors and consequently with no simple treatment (Agras & Werne, 1981). This problem is confounded by the growing belief that obesity is not one condition but a class of several subtypes (Agras & Werne, 1981).

Although genetic factors have been cited as strongly influencing an individual's predisposition toward obesity (Bray, 1979), there is evidence to suggest that environmental factors may be more influential (Garn & Clark, 1976; Hartz, Rimm, & Griefer, 1977; Schenker, Fisichelli, & Lang, 1974). The clinical subtypes of obesity that are currently recognized include genetic, endocrine, hypothalamic, and drug-induced obese conditions (Mayer, 1957). Genetic obesity includes simple obesity (e.g., a multiplicity of genes) and is associated with rare hereditary diseases such as Cohen, Laurence-Moon-Biedl, Prader-Willi, and Alstrom syndromes (which are also associated with mental retardation), Von Gierke's disease, and hyperostosis frontalis interna. Hypothalamic obesity subtypes include Frohlich syndrome, Cushing syndrome, gynandrism, hypogonadism, hyperinsulinism, thyroid deficiency, and Stein-Leventhal syndrome. Drug-induced obesity includes those associated with phenothiazines, oestrogens, and tricyclic antidepressants. In addition to these conditions, anomalies occur in fat distribution in the form of lipomatosis and lipophilia (e.g., exaggerated fat deposits) that are also considered genetically linked and can be removed surgically (Craddock, 1978).

Genetic Disorders

The study of obesity in the retarded population is particularly difficult due to the strong association between mental retardation and specific genetic disorders that are also associated with obesity. Although these syndromes are rare, they deserve special consideration in order

to clarify their clinical significance and identify effective treatment procedures.

Obesity is often reported as one of the problems associated with Down's syndrome (Pueschel, 1980; Smith & Wilson, 1973) that tends to occur between 2 and 6 years of age in this population and that may persist up to about 12 to 13 years of age (Chumlea & Cronk, 1981) or through adulthood. This problem has been traced to linear growth abnormalities (Cronk, 1978; Rarick & Seefeldt, 1974; Roche, 1965) and delayed skeletal maturation (Roche, 1965). For instance, Chumlea and Cronk (1981) found that even though some children with Down's syndrome may have less than normal weights and lengths at birth, as well as smaller stature throughout early development (up to two standard deviations below the mean at age 3), they eventually catch up to norm levels of growth during adolescence. The specific problems of measuring obesity in this group was recently reported by Cronk, Chumlea, and Roche (1985) who found that values on age-specific weight/stature charts were unreliable for children with Down's syndrome because the values would fall below the 15th percentile due to skeletal/stature differences.

Cohen syndrome, like Down's syndrome, is another genetic disorder associated with mental retardation that is typified by an unusual course of obesity (e.g., truncal obesity) after 5 years of age (Doyard & Mattei, 1984). This syndrome, first reported by Cohen and colleagues (Cohen, Hall, Smith, Graham, & Kampert, 1973) is a distinct clinical entity also characterized by delayed puberty, kypotonia, short stature (Goecke, Majewski, Kauther, & Sterzel, 1982; Kousseff, 1981), and, in some cases, cardiovascular anomalies (Friedman & Sack, 1982). The combination of mental retardation, obesity, and limb and ocular anomalies found in Cohen syndrome links it to other well-known conditions such as Prader-Willi syndrome (Hall & Smith, 1972) and Biedl-Bardet syndrome (Bauman & Hogan, 1973).

Individuals with Prader-Willi syndrome (Prader, Labhart, & Willi, 1956) present a particularly challenging problem for clinicians due to the complexity of this disorder. A number of neurologic and endocrinologic problems are associated with this syndrome, including increased incidence of diabetes mellitus, hypomentia, hypotonia, and short stature (Groover, 1978). Children with this disorder generally exhibit marked increase in appetite in the latter part of the first or early part of the second years of life (Groover, 1978). This insatiable desire to ingest food is attributed to an abnormality of the hypothalamus (Prader *et al.*, 1956). Extremely obese Prader-Willi syndrome individuals are also characterized by temporal melatonin rhythm (i.e., periodic secretion of the hormone Acetyl-5-methoxytryptamine from the pineal gland that causes

lightening of the skin and is believed to suppress gonadal function) similar to other exogenously obese individuals (Tamarkin, Abastillas, Chen, McNemar, & Sidbury, 1982) and development of a hypoventilation syndrome (i.e., reduced ventilation relative to carbon dioxide production within the air sacs of the lungs) between approximately 4 and 12 years of age (Bye, Vines, & Fronzek, 1983).

Certain characteristics associated with Down's, Cohen, and Prader-Willi syndromes such as hypotonia and limb and skeletal anomalies deserve special consideration as potential common denominators for obesity, particularly in relation to their unusual developmental course. Hypotonia and associated hyperactivity have been linked to obesity in Down's syndrome (Cowie, 1970; Diamond & Moon, 1961). Similarly, the relationship between obesity and short stature has been reported in a genetic sex-(X)linked condition by several investigators (Borjeson, Forssman, & Lehmann, 1962; Sengers, Hamel, Otten, Van Gils, & DePagter, 1985; Young & Hughes, 1982). More than likely, the etiology of overweight among individuals with these syndromes is multifaceted. For instance, metabolic disorders in the form of thyroid hypofunction (Pearse, Reiss, & Suwalski, 1963; Sare, Ruvalcalba, & Kelley, 1978), carbohydrate dysfunction (Milunsky, Lowry, Rubenstein, & Wright, 1968), and abnormal Na/K atpase activity (McCoy & Enns, 1978) associated with Down's syndrome may contribute to and interact with an obese condition. Similarly, both Laurence-Moon-Biedl-Bardet and Prader-Willi syndromes are associated with increased incidence of diabetes mellitus (Groover, 1978) that in itself may be related to overweight. The influence on obesity of the various neurologic and endocrinologic problems associated with Prader-Willi syndrome are not fully understood.

Environmental Correlates

In addition to the genetic association between mental retardation and obesity is the contribution of environmental variables. For instance, poor nutrition that is prevalent in this population and most often cited as resulting in malnutrition (Crome & Stern, 1972) and underweight (Wallen & Roszkowski, 1980) may be a valid concern in regards to obesity, particularly in relation to institutional diets that are characteristically high in starch and carbohydrates. For instance, Cronk *et al.* (1985) found a greater incidence of overweight in institutionalized children with Down's syndrome than those reared at home. These results were attributed to diet, activity, and general level of stimulation. A genetic predisposition for an obese condition coupled with environmental factors such as sedentary life-style and poor diet are likely ingredients for

the development of an obese condition in these individuals. Smaller body size, motor problems and late maturation, which would all tend to affect caloric requirements, have also been cited as contributing to an obese condition.

Maintenance

Closely related to etiological variables are factors that contribute to the maintenance of obesity and overweight.

Personality Patterns

There are a number of personality and affective characteristics that have been implicated in relation to an obese condition, including anxiety, self-esteem, neuroticism, and depression (Atkinson & Ringuette, 1967; Clancy, 1965; Held & Snow, 1972; Leckie & Withers, 1967; Levitt & Fellner, 1965; Stunkard, 1967; Werkman & Greenberg, 1967). There appears to be little consistency in personality patterns unique to obese individuals, and the question remains as to whether any personality characteristics or attitudes associated with obesity are primary or secondary to the development of obesity. One of the earliest psychological studies on obesity suggested that stress or anxiety states lead to overeating (Bruch, 1973). Although there are studies to support increased eating patterns under stressful situations in both normal-weight (Schacter, Goldman, & Gordon, 1968) and obese individuals (McKenna, 1972), the bulk of the literature does not support the theory that food consumption reduces anxiety (Leon, 1974). Direct tests of this hypothesis in which caloric intake was measured after laboratory-induced stress have produced equivocal results (Meyer & Pudel, 1972). Consistent with research in the general population, Fox, Burkhart, and Rotatori (1984) found no relationship between personality variables in mentally retarded adults as measured by anxiety and self-concept scales and an obese condition.

Eating Habits

Another speculation is that obese individuals have faulty eating habits that lead to excessive food consumption (Stunkard & Kaplan, 1977; Wooley, Wooley, & Dyrenforth, 1979). These poor habits include rapid eating, insufficient chewing, taking large bites, and eating several times per day. This hypothesis, first proposed by Ferster, Nurnberger, and Levitt (1962) and researched in both children and adults, suggests

that obese individuals eat more quickly and chew their food less than nonobese individuals (Adams, Ferguson, Stunkard & Agras, 1978; Hill & McCutcheon, 1975; Marston, London, Cooper, & Cohen, 1975). Other studies in this area have generally reported either no evidence of an obese eating style (Adams, Ferguson, Stunkard, & Agras, 1978; Stunkard, Coll, Lindquist, & Meyers, 1980) or conflicting results (Stunkard & Kaplan, 1977). An expanded version of this hypothesis was originally proposed by Schachter (1968) who suggested that eating behavior in obese individuals is under external stimulus control. This hypothesis has prompted a number of well-controlled studies, also with conflicting results (Levitz, 1973). However, one factor that appears to reliably distinguish obese and nonobese individuals is greater preference for and responsiveness toward food palatability (Haskin & Van Itallie, 1965; Nisbett, 1972; Wooley & Wooley, 1975; Wooley, Wooley, & Woods, 1975), leading to the conclusion that the obese will choose and eat highly palatable foods. A related finding is that obese individuals may be more sensitive to visually prominent food cues (Johnson, 1974; Ross, 1974).

Fox, Burkhart, and Rotatori (1983a) evaluated mentally retarded adults on a variety of eating disorders measured during a lunch period in a workshop setting but found no difference between obese and nonobese subjects in eating rate, total meal time, or caloric intake. However, the wide variability in eating behavior found between the two groups suggested that, although eating style may not be a valid construct for obesity in this population, eating patterns may contribute to an obese condition in some individuals. Other eating variables such as daily snacks and weekly caloric intake require further research in order to delineate their contribution to overweight and obesity.

Activity Patterns

Closely related to research on the imbalance between food intake and energy expenditure is research related to differing activity patterns in obese and nonobese individuals. This research, which has received increased attention in recent years, is based on a theory of energy imbalance. That is, when energy input exceeds energy expenditure, overweight or obesity may result. Although relatively few studies have been conducted in this area, preliminary research suggests that in naturalistic settings, obese individuals exert less energy and are less active than nonobese individuals (Bullen, Reed, & Mayer, 1964; Dean & Garabedian, 1979; Meyers, Stunkard, Coll, & Cooke, 1980; Stefanik, Heald, & Mayer, 1959).

Activity level may be an important contribution to the sex differ-

ences found in obese individuals and those individuals who have obesity in association with specific genetic disorders characterized by hypotonia. For instance, the available research suggests that males tend to be generally more active than females (Garfield, 1963), which may be important to understanding the greater prevalence of obesity among females (Fox & Rotatori, 1982) as well as the greater prevalence of underweight in males (Wallen & Roszkowski, 1980) in the retarded population. Similarly, hypotonia and hypoactivity may interact with activity level to contribute to an obese condition in specific subgroups of the retarded population (e.g., Down's, Cohen, Prader-Willi syndromes).

Several researchers have suggested that inactivity may contribute more to an obese condition than overeating (Johnson, Burke, & Mayer, 1956; Stefanick *et al.*, 1959). Support for the role of activity in an obese condition among mentally retarded individuals was provided by Fox, Burkhart, and Rotatori (1984) who found that a submaximal cardiovascular endurance test successfully discriminated obese and nonobese retarded individuals.

Treatment

Behavioral

Closely related to research on the etiology and maintenance of obesity are investigations aimed at assessing the effectiveness of treatment procedures for reducing obesity. Since the development of behavioral treatment procedures for obesity (Stuart, 1967), well over 100 clinical and research articles have appeared in the literature (Abramson, 1977). The essential components of the behavioral approaches include self-monitoring (Joachim, 1977), external reinforcement (Foxx, 1972), stimulus control (Foreyt & Parks, 1975), and control of eating behavior (Fisher, 1986; Rotatori, 1978). Energy expenditure (Altman, Bondy, & Hirsch, 1978; Buford, 1975; Gumaer & Simon, 1979; Heiman, 1978), nutritional education (Rotatori & Fox, 1980), and cognitive restructuring (Rotatori, Fox, & Switzky, 1979) have also been applied in treating obesity in both general (see review by Stunkard, 1982), and retarded populations (see review by Burkhart, Fox, & Rotatori, 1985).

In spite of the extensive efforts to treat obesity among the general population, there has been comparatively little treatment research in the MR/DD population. Rotatori and colleagues devised a comprehensive weight reduction program for retarded individuals that involves a number of components including (a) increased self-awareness of overeating patterns and body weight; (b) reduction of food cues; (c) reduction of

eating rates; (d) increased activity level; and (e) reduction of caloric intake (Rotatori & Fox, 1981). This treatment package has veen validated through a number of studies on retarded children (Rotatori, Parish, & Freagon, 1979), adolescents (Rotatori & Switzky, 1979; Rotatori, Fox, & Switzky, 1979), and adults (Rotatori, 1978; Rotatori, Switzky, & Fox, 1981). A streamlined version of this program has also proven successful for retarded adults (Fox, Haniotes, & Rotatori, 1984; Fox, Rosenberg, & Rotatori, 1985).

Future research is needed, however, to investigate the individual and combined contribution of the components of this package (Rotatori, Fox, & Mauser, 1981). At present, the key components for success appear to be involvement of significant others, routine homework with feedback, and maintenance training sessions. The importance of significant others has been substantiated through previous research (Fox, Rosenberg, & Rotatori, 1985; Jackson & Thorbecke, 1982). Further support has been provided recently through a similar program involving nutritional changes, a simple activity program, and parental cooperation successfully carried out in a school for mentally retarded children (Nelson, Catchings, & Pendleton, 1985).

Exercise programs are increasingly being considered a crucial component of weight reduction programs (Craddock, 1973; Stuart & Davis, 1972). The beneficial effects of exercise in mentally retarded individuals was recently demonstrated in a study by Schurrer, Weltman, and Brammell (1985) who found that subjects who attended exercise sessions on an average of 3.1 times per week and covered an average distance of 9.0 km/week, lost an average of 3.6 kg of weight and increased maximal oxygen consumption by 43%. Nardella, Sulzbacher, and Worthington-Roberts (1983) reported a study that assessed activity level on the weight of Prader-Willi subjects who were placed on a weight reduction program that combined exercise and 1,000-calorie-per-day diet. Results revealed a significant correlation between activity level and weight loss. A major drawback to exercise is the extreme effort involved and the implied requirement that a change in life-style occur. The subsequent dropout rate of nearly 25% (Wing & Jeffrey, 1979) far exceeds that of other weight loss procedures. Still, little is known about the long-term benefits of weight loss through exercise.

The dropout rate in the normal population for traditional outpatient treatment (e.g. diet, medication, psychotherapy) has reportedly been as high as 25% to 75% (see Stunkard & McLaren-Hume, 1959), whereas most behavioral programs report rates of 15% or less. Although there are currently no specific rates available from the literature on dropout in the mentally retarded population, it is likely that these rates are low

because these individuals require close supervision, subsequently ensuring greater attendance and followthrough.

Two continued problems persist in the treatment literature for nonretarded and retarded populations: (a) high interindividual variability in weight loss during a program's implementation and (b) poor long-term maintenance of weight loss once treatment is over. Regarding the first problem, Fox *et al.* (1985b) reported weight changes ranging from +5 to −11 lb for mentally retarded adults following 10 weeks of treatment. According to Stunkard's (1982) review of the literature on obesity, weight loss with behavior therapy in the normal population averages 11.2 lb with variability ranging from 0.8 to 17.2 lb. Wing and Jeffrey's (1979) review of the literature revealed that the weight losses reported from behavior therapy research do not differ greatly from those reported using other types of treatments. In terms of long-term maintenance issues, Fox *et al.* (1984b) found only 37.5% of their MR adult sample ($n = 16$) maintained weight loss 1 year after treatment had been terminated. These findings are not inconsistent with those from the normal population. According to their review containing a 5-year follow-up of treatment for obesity, Stunkard and Penick (1979) found that most patients following both traditional and behavioral treatment continued to lose weight during the first year but regained it across the following 4 years. These issues must be addressed in future treatment investigations.

Because of this problem, Rotatori, Zinkgraf, Matson, Fox, Sexton, and Wade (1986) investigated the effect of two weight reduction maintenance strategies for 13 moderately/mildly retarded adults after they were first exposed to a 12-week behavior therapy weight reduction program. One maintenance strategy involved posttreatment booster sessions (12 weeks) followed by weekly weigh-ins plus reinforcements for weight loss (40 weeks). This strategy was compared with weekly weigh-ins and reinforcement for weight loss (52 weeks). Interestingly, subjects exposed to the booster sessions continued to lose weight, whereas subjects not exposed to the booster sessions maintained their weight loss. However, a 12-month follow-up check revealed that the long-term success of the treatment and maintenance procedures was not well maintained. It is important to note that there is currently a lack of information in the literature regarding the level of mental retardation diagnosis and responsiveness to weight reduction programs. However, the type of approach used would logically differ across these groups, according to the extent of the problem and the ability of specific individuals to understand the treatment approach. This is an area that requires further research.

Medical

In addition to the behavioral techniques utilized for treating obesity, there are a number of medical procedures that have been tried. These procedures include diets, drugs, and surgery. Although there are numerous diets available, the main focus of all types is caloric restriction. According to a review by Wing and Jeffrey (1979), diets in general are a more effective means of losing weight than previously thought. Diets as treatment have apparently received a negative image due to wide variability in reported weight loss both between and across studies (Jeffrey, Wing, & Stunkard, 1978). Despite reports that dieters have a tendency to return to their previous weight following dramatic alteration in weight (Sims *et al.*, 1968; Wooley & Wooley, 1975), the prognosis for outcome maintenance overall is good (Ost & Gotestam, 1976; Stunkard & Penick, 1978; Wing & Jeffrey, 1979). However, this wide variability in both short-term and long-term treatment effectiveness suggests that one form of treatment alone may not be sufficient for some forms of obesity.

Drugs and surgery, although prescribed less frequently, have also reportedly been successful in treatment of obesity and overweight. Prescriptions for appetite suppressants such as fenfluramine (Ost & Gotestam, 1976) have produced successful results but are generally not preferred due to associated side effects and the potential for addiction. Surgical techniques such as partial gastrectomy (Craddock, 1978), jejunoileal bypass (Corrodi, 1984) and vagotomy (Kral, 1978) have also been used but are generally not considered the treatment of choice except in extreme cases due to the potential of a number of complications. Little data are available on the extent of the use of drugs and surgery for the treatment of obesity in the mentally retarded population. However, in a comparative study, Ost and Gotestam, (1976) found behavioral treatment to be more effective than fenfluramine. Nevertheless, combination treatment using pharmacotherapy and behavior therapy has not been systematically explored. Although surgical procedures have been performed on individuals with Prader-Willi syndrome, this form of treatment has been associated with higher incidence of complications and mortality in these individuals in comparison to similar operations performed on other obese patients.

Clearly the treatment of obesity will be better supported by further research. Unfortunately, many treatments if effective are often successful in achieving only temporary gains, and the issue of weight reduction maintenance continues to require attention.

Issues of Definition and Measurement

The Build Study in 1979, National health surveys (National Health Examination Survey, 1960 to 1962; National Health and Nutrition Survey II, 1976 to 1980—see Simopoulos & Van Itallie, 1984), Obesity Task Force of 1979 (Bray, 1979) and publications by the National Center for Health Statistics (Abraham, Carroll, & Najjar, 1980) have provided normative data on anthropometric indexes of obesity including height, weight, and skinfold thickness as well as biochemical indexes and their relationship to health and longevity. Among the persistent problems uncovered by these efforts and relevant to any population studies was widespread confusion about the terminology used to define overweight and obese conditions as well as measurement techniques for classification for these conditions. These problems have led to publication of data that are difficult to interpret and/or compare to other studies (see Simopoulos & Van Itallie, 1984). The terms *overweight* and *obesity* though often used interchangeably are not identical (see Fox, Burkhart, & Rotatori, 1983b). Overweight refers to an excess in body weight relative to standards for height (Bray, 1979), whereas obesity refers to excess subcutaneous, nonessential fat (Craig, 1969).

Subscapular skinfold measurement has been recommended as a fairly reliable and more direct index of obesity than relative weight. Skinfold thickness may be preferable over relative weight measures on the basis of two points: (a) it has been found to be significantly related to the presence of obesity that is associated with increased risk of myocardial infarction (Hubert *et al.*, 1983) and (b) there appears to be less error variance in classification of obesity using this technique (Fox, Rotatori, & Burkhart, 1983b). The greater accuracy of skinfold thickness might be attributed to the fact that weight represents a different measure of body mass in each sex (Burkhart *et al.*, 1985). For instance, excess weight occurs from muscularity more often in men (Simopoulos & Van Itallie, 1984), and extreme fluctuations in body fluids occur more frequently in women (Wallen & Roszkowski, 1980). The terminology and measurement standards for overweight and obesity must be clarified and cited appropriately in scientific reports in order to make meaningful comparison.

Relationship between Behavioral and Medical Aspects of Obesity

Special consideration should be given to assessing the interaction between physiological variables that may contribute to an obese con-

dition and those variables that are targeted for treatment. For instance, Wooley, Wooley, and Turner (1975) found that salivary responses to dessert were greater 1 hour after a meal eaten quickly. Similarly, Hallbert (1973) found longer small intestines in obese individuals that would lead to faster food absorption and could thus help explain differences in appetite as well as differential treatment effectiveness between obese and nonobese individuals.

Another important consideration in the treatment of obesity is the potential interaction between neurologic and endocrinologic systems that are involved in coordinating and integrating the operations of other body systems as well as adjusting to meet the demands of environmental change. Further research is needed to delineate the contribution of these systems, particularly in relation to individuals with Prader-Willi syndrome.

Weight reduction procedures used in the MR/DD population present additional risks due to the frequent association and treatment of preexisting physical (e.g., skeletal abnormalities, congenital heart disease), neurological (e.g., seizures, cerebral palsy), physiological (e.g., metabolic disorders), and psychological (e.g., anxiety, depression, psychosis) conditions also prevalent in this population that can be contraindications for use of some treatments. For instance, the health risks of surgery and dieting already confounded by an obese condition are increased in individuals (specifically those with Down's and Prader-Willi syndromes) who have physical anomalies in the form of heart conditions, diabetes, and hypertension. Cardiac dysfunction is also a potentially lethal complication of rapid weight loss among obese dieters (Van Itallie & Yang, 1984). Similarly, medications such as anticonvulsants, neuroleptics, and antidepressants have chemical properties that can interact with appetite suppressants. Medication and nutrition can also be affected by each other to result in erratic responses in patients (Smith & Bidlack, 1984).

Attempts to restrict caloric intake among Prader-Willi patients have often been unsuccessful due to complications associated with food obsessions (attributed to hypothalamic dysfunction) and behavioral outbursts (e.g., oppositionalism, temper tantrums). Surgery used to treat obesity in these patients has also met with limited success. For instance, Fonkalsrud and Bray (1981) reported an initial weight loss of 29 kg in a 17-year-old boy with Prader-Willi syndrome treated by a truncal vagotomy. However, within 11 months, the patient regained 29 kg of weight due to almost continuous ingestion of small quantities of food. Surgical procedures of Prader-Willi patients are also associated with an even greater incidence of complications and mortality than other obese in-

dividuals (Fonkalsrud & Bray, 1981). Another complication in treating Prader-Willi syndrome is the attenuated pancreatic polypeptide (PP) response to low protein meal stimulation (Zipf, Dorisio, Cataland, & Dixon, 1983), making weight reduction programs involving special diets particularly difficult. This functional deficiency to PP response appears to be related to glucose and insulin imbalance. Insulin has been shown to have a direct effect on satiety centers in animals (Debons, Krimsky, & Froh, 1970) and produces increased appetite in individuals in poor nutritional state (see Craddock, 1978). Successful treatment in children with Prader-Willi syndrome has included weight reduction and cardiac failure regimen (Bye *et al.*, 1983) as well as behavioral treatment (Altman *et al.*, 1978).

Individuals included in weight reduction programs should be screened for preexisting pathologic conditions that may lead to medical complications or morbidity. Such individuals should be followed closely by medical professionals with particular attention given toward physical variables that either enhance or hinder treatment.

Summary and Directions for Further Research

Although research in the area of obesity with the mentally retarded has expanded since the first review by Staugaitis (1978), continued research is greatly needed. At the basic level, research is needed in appropriately defining and classifying obesity across all populations so that results can be interpreted clearly and adequate comparisons made. Further investigations are also needed to identify variables that contribute to the development and maintenance of obesity, including eating and activity patterns, caloric intake, metabolic factors, and biomedical correlates (Rotatori, Fox, Matson, Mehta, Bader, & Lopuch, 1986).

Additional research will be required to identify the types of obesity associated with specific diseases at different life stages such as upper trunk obesity with diabetes, fat cell number and hypertension in early adulthood and fat cell size and hypertension in middle age (Simopoulos & Van Itallie, 1984). Attention must also be given to factors that predispose an individual toward an obese condition. Important among these factors are short stature hypotonia and unusual trends in motor development, specifically among children with Down's and Prader-Willi syndromes who tend to acquire motor milestones at slower rates than the normal population.

Research is needed to delineate factors that are critical for preven-

tion and successful treatment outcomes across populations and obesity subgroups. Because little is known about methods for maintaining weight control, some form of maintenance control should be instituted from the beginning of treatment. Currently successful treatments involve strategies to change eating and activity patterns, increase self-control, encourage development of support systems (Burkhart *et al.*, 1985), and provide nutritional information (Rotatori, Fox, Litton, & Wade, 1985). The problems of high interindividual variability revealed in weight loss studies and the inconsistency in long-term maintenance continue to require attention in the research area. Further attention should also be given to the specific clinical groups among the mentally retarded population that are characterized by an obese condition (e.g., Down's, Prader-Willi syndromes). Finally, *children* with MR/DD require study. The characteristics of the obese retarded child as well as possible familial factors (e.g., obese parents) are two examples where investigations are needed. Also, preventative intervention efforts in the schools for retarded children may prove useful.

Medical and Behavioral Evaluation and Treatment

The identification and treatment of obesity is a responsibility shared by all individuals who are concerned with the development and habilitation of the MR/DD person. The role of parents and significant others is particularly important because many obese mentally retarded individuals are seen inconsistently in medical and mental health facilities. Early identification and treatment of obesity poses a continuing problem for behavioral medicine. Unless objectives for identification are translated into specific forms of intervention, early labeling will be a meaningless exercise.

The physician's primary responsibilities in the evaluation and treatment of obesity include (a) correct identification of inheritable disorders that contribute to an obese condition and may compromise treatment effectiveness; (b) identification of physical and medical complications that need concurrent treatment or monitoring; and (c) input and follow-up to address physical/medical issues related to obesity. The role of the behavioral psychologist will be to address behavioral, self-control, and motivational aspects in the treatment of the obese patient to maximize treatment effectiveness in all settings and maintenance of weight loss.

Interdisciplinary Evaluation

Mentally retarded individuals present innumerable combinations of functional disabilities, organic impairments, learning, and social deficits.

Certainly, an obese condition complicates this clinical picture. There are few situations in behavioral medicine in which the consideration of multiple factors is more relevant than obesity in the mentally retarded. With mentally retarded individuals, the complexity of the problem generally exceeds the resolving power of any one discipline.

An important component of the treatment of obese mentally retarded individuals is the maximization of the individual's overall potential and, as such, involves an interdisciplinary approach. Each discipline involved in an interdisciplinary process has the primary responsibility of evaluating the individual from the perspective of his or her own specific skills and, in turn, identifying problems and objectives for intervention and treatment within one's specialized area.

Behavioral treatment should be part of the total plan for any intervention developed by an interdisciplinary team. The thrust of managing obesity in the mentally retarded is the production of behavioral change through prescriptive procedures that clearly specify the objectives for change.

The interdisciplinary team conference should establish a priority ranking among problems and objectives, determine interactional effects among approaches, and develop a program that specifies objectives and establishes responsibility for management. At all levels (e.g., identification, treatment, follow-up), an individual familiar with weight loss programs should be responsible for integrating all components of the interdisciplinary process to address the prioritized needs of the obese patient. In general, the interdisciplinary treatment and habilitation of obese retarded individuals should be undertaken in the context of a strong behavioral support program if functional gains are to be made and maintenance achieved.

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CHAPTER 18

Clinical-Research Issues in the Treatment of Obesity in the Developmentally Disabled

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In the preceding chapter Burkhart, Fox, and Rotatori have provided a comprehensive review of a number of aspects of obesity. Conclusions from a growing body of research and theory have been integrated into a concise, informative discussion, and the coverage of prevalence, health risks, etiology, and treatment options is impressive. As a result, the role of discussant is made more difficult: the areas in which one can expand on the work of Burkhart, Fox, and Rotatori are limited by the scope of their scholarly effort. Therefore, the focus of this article is two-fold. The first intent is to comment briefly on the general conclusions of the preceding chapter. The second is to further highlight general clinical research strategies for assessment and treatment of obesity; this is accomplished through a discussion of problems associated with Prader-Willi syndrome.

State of the Science

Obesity continues to represent a major health problem (Coates & Thoreson, 1978; Woodall & Epstein, 1983; Wooley, Wooley, & Dyren-

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forth, 1979). Despite the growth of awareness in the public at large, obesity in the developmentally disabled has not been a primary concern among researchers and clinicians. Burkhart, Fox, and Rotatori correctly referred to a paucity of available information, and their article would be considerably limited if its scope had been restricted to the developmentally disabled population. Indeed, collectively, these authors have accounted for a large portion of the available literature in the area, including prevalence (Burkhart, Fox, & Rotatori, 1985; Fox, Hartney, Rotatori, & Kurpiers, 1985; Fox & Rotatori, 1982), the effect of personality variables (Fox, Burkhart, & Rotatori, 1984), faulty eating habits (Fox, Burkhart, & Rotatori, 1983), and behavioral treatments (Fox, Haniotes, & Rotatori, 1984; Rotatori, Fox, & Switzky, 1979; Rotatori, Switzky, & Fox, 1981).

The primary conclusion of Burkhart, Fox, and Rotatori that more research is needed on obesity in the developmentally disabled is not difficult to agree with. As they have indicated, work is needed on a broad spectrum of related areas, by researchers from a different range of disciplines. The following section illustrates clinical research efforts to date in one area of obesity. Obesity secondary to Prader-Willi syndrome is only one of many clinical subtypes of obesity, but the integrated efforts of clinical researchers in this area may have generality to the larger population of developmentally disabled clients.

Prader-Willi Syndrome

Characteristics

As noted by Burkhart, Fox, and Rotatori, Prader-Willi syndrome (Prader, Labhart, & Willi, 1956) can be a complex, challenging, and refractory disorder. The syndrome is characterized by failure-to-thrive and hypotonia during infancy, hyperphagia, extreme obesity, mental retardation, dysmorphic facies, small hands and feet, and hypogonadism (Dunn, 1968; Holm, 1981). Hyperphagia is usually the most problematic symptom, resulting in obesity, which can lead to severe cardiorespiratory complications (Bianchine, Stambler, & MacGregor, 1971). The cause of the hyperphagia remains unclear. Hypothalamic dysfunction has been investigated (Hall & Smith, 1972; Zellweger & Schneider, 1968), as has the possibility of neurotransmitter imbalances (Kryiakides, Silverstone, Jeffcoat, & Laurence, 1980).

Treatments and Limitations

Medical treatments have consisted of gastric bypass (Soper, Mason, Printer, & Zellweger, 1975), small intestinal bypass (Randolph, Weintraub, & Rigg, 1974), appetite suppressing drugs, jaw wiring (Bergsma, 1979), and neurotransmitter manipulation (Bray *et al.*, 1983).

Behavioral researchers have attempted to modify mealtime behaviors (Doleys & Gordon, 1980; Marshall, Elder, O'Bosky, Wallace, & Liberman, 1979) and weight gain (Altman, Bondy, & Hirsch, 1978; Thompson, Kodluboy, & Heston, 1980). However, many investigators have reported failure to maintain long-term weight loss (Evans, 1964; Laurence, 1967; Marshall *et al.*, 1981). Reports of successful long-term maintenance have utilized environments where close supervision and stringent control were possible (Altman *et al.*, 1978; Holm & Pipes, 1976; Pipes, 1981; Pipes & Holm, 1973; Thompson *et al.*, 1980).

A primary factor in the lack of success in unstructured settings is the covert nature of foraging for food and food consumption. Individuals with Prader-Willi syndrome are predominantly functioning in the mildly retarded range (Holm, 1981) and have usually developed extensive, sophisticated repertoires for obtaining and consuming food covertly. As a result, most food theft goes undetected and can only be inferred from continued weight gain (Coplin, Hine, & Gormican, 1976).

A study by Page, Finney, Parrish, and Iwata (1983) was designed to address the problem of covert consumption of unauthorized food items. One male (11 years old) and one female (8 years old) had been admitted to an inpatient hospital unit for weight loss and treatment of food theft. Interdisciplinary evaluation and subsequent treatment were provided by behavioral psychology, child life, nursing, nutrition, pediatrics, and social work. Because the hospital was equipped with treatment rooms adjoined by one-way mirrors, covert observation could be arranged. Subjects were observed in two different settings: alone in a small room and in a larger room with other patients and staff present. The rooms were stocked with books, magazines, and other materials as well as a supply of inventoried food items. In baseline, subjects were taken to a room and then left there for 15-minute observation sessions. With both subjects, food theft and consumption were observed to occur, at rates as high as 2.6 per minute with the male and 8.4 per minute with the female. Treatment consisted of a token economy and differential reinforcement of other behavior (DRO) in which tokens were awarded contingent on progressively longer intervals of no food theft, and any occurrence of theft resulted in token response cost. Treatment was effective in eliminating theft under experimental conditions; the DRO interval

was increased from an initial value of 10-seconds to 40-minutes at the time of discharge. However, subsequent to discharge, both subjects resumed food theft after 1 to 2 months and began gaining weight.

In a subsequent study, Page, Stanley, Richman, Deal, and Iwata (1983) treated a 28-year-old woman with Prader-Willi syndrome. Observation methods and initial treatment strategies replicated Page, Finney, Parrish, & Iwata (1983); an additional exercise component was also included in treatment. More importantly, the client was discharged to a group home whose staff consistently implemented program contingencies in the home environment. As a result, a total loss of 37 kg (81 lb) was achieved over a 22-month period.

The results of both studies may be important to the treatment of obesity in Prader-Willi syndrome in two respects. First, covert food theft and consumption were reliably measured. Although the use of one-way mirrors is obviously not plausible in many settings, a variation of the procedures used in the Page *et al.* studies might have utility. For example, the placement of inventoried food items at strategic locations in the home, school, or any setting would allow for assessment and treatment of theft similar to those described by Page *et al.* Access to information on food theft will be a necessary component of comprehensive treatment planning with the many clients whose obesity is related to inappropriate consumption. Second, food theft and inappropriate consumption were effectively eliminated through straightforward contingency management.

Implications for Future Research

The results of the Page *et al.* studies may also have generality to the treatment of obesity in developmental disabilities other than Prader-Willi syndrome. First, discrete behavioral responses directly related to obesity can be identified, monitored, and modified. As Mann (1972) pointed out, obesity is the result of a variety of different responses: mealtime behavior, between-meal consumption, exercise, and general activity level. As a result, the development of effective treatment strategies might be expected to proceed most profitably by initially identifying and measuring a class of relevant behaviors. Although slightly different in focus than the method of task analysis that is common in effective acquisition programs for the developmentally disabled, the identification of relevant component behaviors may have similar utility in treating obesity in this population. The extreme alternative of placing contingencies only on body weight might be successful with normal and higher functioning individuals (Mann, 1972), but attention to covert inappro-

priate consumption (see the Page *et al.*, studies), stimulus control techniques, exercise, weight charts, and other strategies (Aragona, Cassady, & Drabman, 1975) can only enhance clinical outcome and early intervention efforts (Keesges *et al.*, 1983).

Second, hyperphagia and obesity can be effectively treated in a genetic disorder predisposed to their chronic occurrence. These results, in conjunction with previous research in the area, suggest that obesity and hyperphagia with possible organic etiologies can be treated with behavioral interventions. This finding may have relevance to the other types of genetic obesities outlined by Burkhart, Fox, and Rotatori as well as obesity related to hypothalamic and endocrinic dysfunction. Hyperphagia and obesity with organic etiology are, in fact, amenable to environmental control in some cases.

Third, as Burkhart, Fox, and Rotatori pointed out, the active involvement of parents or other caregivers is critical to continued maintenance of treatment gains (Aragona, Cassady, & Drabman, 1975; Fox, Rosenberg, & Rotatori, 1985). Long-term weight loss can be compromised when parents are unable or unwilling to implement simple contingency management programs in the home environment but can sometimes be successfully managed when responsible caregivers play an active role in monitoring and management of program contingencies. It seems clear that effective weight loss programs for the developmentally disabled can only benefit from the application of the existing parent training technology (cf. Bernal, Kinnert, & Schultz, 1980; Forehand & Atkeson, 1977).

Conclusions

Obesity in the developmentally disabled is similar to other disorders that may have behavioral and/or organic components, as well as medical implications. Effective treatment must proceed from a comprehensive interdisciplinary evaluation. Where behavioral components are identified, behavior analysis must be undertaken to identify the occurrence of individual component responses contributing to ongoing weight gain. Subsequent treatment in the form of contingency management can be expected to impact relevant target behaviors and effect changes in body weight even where obesity may have organic etiology. The necessity of integrating behavioral interventions into a comprehensive interdisciplinary treatment plan is obvious. And the incorporation of systematic caregiver training and frequent follow-up contact is critical. Finally, additional research is necessary to determine methods of effectively ap-

plying the previously mentioned considerations to the clinical exigencies of applied treatment programs.

ACKNOWLEDGMENTS. Appreciation is extended to Jack W. Finney for his comments on an earlier draft of the manuscript. This project was supported in part by MCH Grant 00917-15-0 from the Department of Health and Human Services. Reprint requests may be addressed to Terry J. Page, The Kennedy Institute, 707 North Broadway, Baltimore, MD 21205.

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