Emotional and Behavioral Health in Persons with Mental Retardation/Developmental Disabilities: Research Challenges and Opportunities

November 29-December 1, 2001
Rockville, MD

Workshop Executive Summary

Individuals with mental retardation and developmental disabilities are estimated to be 3-4 times more likely than those in the general population to experience an emotional, behavioral, or psychiatric disorder. Recent advances in a number of fields and disciplines – including the neurosciences, genetics, psychopharmacology, developmental neuropsychiatry, psychology, and education – show promise for improving the treatment and lives of those with mental retardation and developmental disabilities. **Despite advances in each of these areas, mental retardation is frequently a criterion for exclusion from research studies**. **Enrollment of individuals with mental retardation in research protocols addressing emotional and behavioral disorders has been limited, issues of informed consent persist, and more researchers with an interest and expertise in this population are needed.**

To address these issues, a two-and-a-half-day Workshop was convened by the National Institute of Neurological Disorders and Stroke (NINDS), the National Institute of Child Health and Human Development (NICHD), the National Institute of Mental Health (NIMH), the National Institutes of Health (NIH) Office of Rare Diseases, and the Joseph P. Kennedy Jr. Foundation.

The Workshop, entitled "Emotional and Behavioral Health in Persons with Mental Retardation/Developmental Disabilities: Research Challenges and Opportunities" drew 97 participants, from academic research institutions, government agencies, service providers, and consumer advocacy organizations. (For a copy of the meeting Agenda and participant list, visit the NINDS web site at http://www.ninds.nih.gov/news_and_events/proceedings.htm.)

The Workshop was designed to identify barriers to the inclusion of people with mental retardation and developmental disabilities in federally funded research in the United States. The goal was to define ways to increase inclusion of people with mental retardation in research in order to promote evidenced-based treatment for this population.

The Workshop was held several days prior to a Surgeon General's Conference on Health Disparities and Mental Retardation, December 5-6, 2001. Results of the Workshop were presented publicly there for the first time. (To view webcasts of the Workshop and the Surgeon General's Conference, visit the NIH web site at http://videocast.nih.gov and click on "Past Events," then "Conferences.")

Opening Remarks and Overview Presentations

The meeting began with welcoming messages from leaders of the sponsoring NIH Institutes:

- Audrey Penn, MD, Acting Director of NINDS noted that three NIH Institutes were represented, but there easily could have been a dozen, given the breadth of the subject matter.

She said NINDS neuroscientists share with their NIH colleagues an interest in the developmental neurobiology, genetics, and cognitive aspects of syndromes associated with mental retardation, and she expressed eagerness to review all that Workshop participants brought to the table.

- Duane Alexander, MD, Director of NICHD, said that the conference provided an opportunity to take stock of what research has been done, what has been learned, what has been learned but has not been applied, and what important issues remain to be addressed. He said the speaker roster read like an "Olympics of champion investigators" in this field, and he looked forward to reviewing and implementing the recommendations that emerged from the Workshop.
- Richard Nakamura, PhD, Deputy Director of NIMH, said that the emotional and behavioral health of those with mental retardation and developmental disabilities has been a neglected subject. He said NIMH recognizes that it has a special duty to help foster research that will advance the health and lives of those with mental retardation and developmental disabilities, and the Institute aims to fully honor that commitment.

Eunice Kennedy Shriver, Executive Vice President of the Kennedy Foundation, issued a call to action to the group to work to ensure that the more than seven million people in the United States with mental retardation and developmental disabilities have the mental health supports and services they need throughout their lives. She noted that:

- Some 20-30 percent of people with mental retardation and developmental disabilities suffer from a psychiatric disorder, yet services are organized as if people have either mental retardation or mental illness—but not both.
- People with all levels of disability experience a full range of mental health challenges, yet 39 percent of psychiatrists admit they prefer not to treat people with mental retardation and developmental disabilities.
- Children with mental retardation and developmental disabilities who are depressed are less likely to get the help they need in schools, but unless their behavior is disruptive, they are likely to be ignored or removed from class for poor performance.

"To improve mental health supports to every American who has mental retardation, and their families, we must focus on research and training," she said. "But over the next few days, do not forget that people need services!"

Following the opening remarks, leaders in the field provided overviews on research opportunities in genetics, neuroimaging, brain plasticity, and psychosocial issues. Subsequent overview presentations provided background in six key areas related to the emotional and behavioral health of individuals with mental retardation: (1) epidemiology, (2) diagnosis/assessment, (3) interventions, (4) ethical issues, (5) research design, and (6) research training needs.

Working Groups met later to identify critical issues in each of these areas and craft recommendations, which were presented to the full group of Workshop participants for discussion. Recommendations from the Working Groups are being reviewed by the three sponsoring NIH Institutes (NINDS, NICHD, NIMH) and appropriate action will be taken to move toward their implementation.

Working Group Discussions

Each of the Working Groups met on the second day of the Workshop to consider a number of questions, including:

Ethical Considerations – How should "consent" and "assent" apply to research involving individuals with mental retardation and developmental disabilities, particularly research with no immediate therapeutic benefit? Should the standard for surrogate decision-making regarding the participation in research (both when to permit it and the standards for its exercise) be the same for children and adults where there are questions about their ability to make decisions?

Research Training Needs – What are the best approaches to establishing new researchers with an interest in the emotional and behavioral health of people with mental retardation and developmental disabilities? Are there useful models from other areas of biomedical research that can be used?

Epidemiology –What epidemiologic evidence is needed to provide accurate measures of the prevalence and incidence of behavioral, emotional, and psychiatric problems; to identify risk and protective factors; and to establish the natural history of these conditions? Which designs, research strategies, and approaches should be considered?

Diagnosis and Assessment – What are the appropriate instruments needed to diagnose and formulate treatment for individuals with mental retardation and developmental disabilities? How can current diagnostic and assessment methods be made more applicable to individuals with mental retardation and developmental disabilities?

Interventions Research – What are the most pressing needs of people with mental retardation and developmental disabilities and co-occurring emotional and behavioral disturbances? How might interventions research be most effectively carried out in hospitals, schools, and community settings?

Research Design – What are the most pertinent research designs to conduct research involving people with mental retardation and developmental disabilities and co-occurring emotional and behavioral disturbances? What steps should be taken to adapt FDA guidelines for clinical medication trials in individuals with mental retardation and developmental disabilities?

Working Group Recommendations

Ethical Considerations

The Working Group on Ethical Considerations identified a wide variety of ethical and legal issues surrounding the participation of people with mental retardation and developmental disabilities in research protocols. ***The members affirmed that well designed non-therapeutic research serves the interests of individuals with mental retardation and developmental disabilities. When there is no effective treatment available, it is in the interest of people with***

mental retardation and developmental disabilities to participate in well-designed placebo-controlled trials. The alternative is to not to receive treatment for disabling conditions, or to receive treatments where safety and efficacy have not been studied, often resulting in harm without compensating benefit. Any determinations about an individual's capacity to decide whether to participate in research should be based on his or her functional ability, not on IQ score or the simple label of mental retardation and developmental disabilities.

Critical Issues

- **The "Common Rule" governing the participation of human subjects in federally sponsored research does not adequately address the inclusion of individuals with mental retardation and developmental disabilities.** It remains unclear whether separate provisions are necessary for individuals with mental retardation and developmental disabilities, mental illness, or impaired decisional capacity.
- **The "minimal risk rule" guiding participation in non-therapeutic research is ambiguous and is being interpreted widely by institutional review boards (IRBs).** Considerable variability is apparent in how IRBs determine the risk level in "ordinary" life, what medical tests are viewed as "routine," and whether such factors should be indexed to people who are healthy or those who are ill.
- **The current understanding of informed consent and assent frequently does not take into consideration the varied capacities of individuals with mental retardation and developmental disabilities to make decisions.** Decisional capacity in adults with mental retardation and developmental disabilities often is assumed to be global, across all decision-making tasks, which is contrary to available evidence.
- **Individuals with mental retardation and developmental disabilities may be more or less vulnerable to undue influence and/or coercion depending on such factors as their residential setting or the absence or presence of an appropriate advocate**.
- **Fear of federal sanctions is restraining IRBs from approving ethically acceptable research, particularly in vulnerable populations.** Similarly IRB members and investigators express growing concerns about litigation. Punitive sanctions are frequently levied by federal oversight bodies for regulatory compliance infractions that are not related to harm to research subjects.
- **Lack of clarity in guidelines may impede genetic testing of individual with mental retardation and developmental disabilities.** There is no consensus on whether or when family members should be considered research subjects if mentioned in the course of a genetic family history. The authority of surrogate decision-makers to consent for genetic research on adults with decisional incapacity with no direct therapeutic benefit to the subject remains unclear.

Recommendations

- **The Institute of Medicine should convene a meeting of all stakeholders to focus on problems in the Common Rule with regard to individuals with mental retardation and developmental disabilities, mental illness, or impaired decisional capacity in federally sponsored research.**
- **This group should address the minimal risk rule as it applies to individuals with mental retardation and developmental disabilities who lack capacity to make decisions regarding research participation.** The group should also address federal guidelines that

affect genetic family histories – specifically, whether a family history that was obtained as part of a research study necessarily makes a family member a "research subject." This group should also address when non-therapeutic genetic testing for research purposes is permissible for individuals with mental retardation and developmental disabilities.

- **Further analysis is needed concerning issues of assent and dissent in individuals with mental retardation and developmental disabilities.** Individuals with mental retardation and developmental disabilities who have the capacity to consent should be given the widest latitude to consent or refuse to participate in research. If an individual with mental retardation and developmental disabilities appears unwilling to participate in research, dissent should generally be honored, even for procedures of minimal risk. Analysis is needed about the relevance of objection in individuals with limited language ability, including infants and severely impaired older children and adults.
- **Appropriate tools for the assessment of decisional capacity in individuals with mental retardation and developmental disabilities need to be developed.** Further research is needed on techniques to enhance the decision-making capabilities of individuals with mental retardation and developmental disabilities.
- **Continued discussion is needed on the legal authority of surrogate decision makers to authorize participation in research that does not offer the prospect of direct benefit to the subject.** Because decisional capacity is situational in individuals with mental retardation and developmental disabilities, appointment of a surrogate for all decision-making may be inappropriate. Absent state law to the contrary, the authority of a surrogate to authorize research that does not have the prospect of direct benefit to the person with mental retardation and developmental disabilities should be clarified.
- **Additional discussion is needed on the role of the residential setting and associated factors in research protections.** Issues of home ownership/control and contracting for services within the residential setting are a necessary part of the discussion.
- **Federal oversight bodies, such as the Office of Human Research Protection (OHRP) and the National Human Research Protections Advisory Committee (NHRPAC), should review the appropriateness of punitive sanctions for infractions that have little apparent relevance to protection of research subjects.**

Research Training Needs

The Working Group on Research Training Needs emphasized that the training of investigators with an interest in the emotional and behavioral health of individuals with mental retardation and developmental disabilities should draw on knowledge in a wide variety of disciplines. Efforts should be collaborative, emphasizing training, research, and clinical work – with a particular focus on translational research that bridges basic science and more applied investigations. Initiatives to train more researchers in this area face a number of impediments, including stigma (for example, the notion that individuals with mental retardation and developmental disabilities are "difficult to treat") research in this area is frequently viewed as less prestigious or "real" compared with other fields; and disciplinary "silos" maintain barriers to the cross-disciplinary collaboration necessary in this research and essential to attracting new investigators to the field.

Critical Issues

- **There is no clear home for grants supporting research on the emotional and behavioral health of individuals with mental retardation and developmental disabilities.** Individual Research Career Development (K) awards may come too late in medical careers and provide insufficient research and salary support in the face of significant debt. Research into the emotional and behavioral health of people with mental retardation and developmental disabilities is not well regarded or understood by funding review groups.
- **Research training opportunities can come too late in clinical training.** Opportunities can be provided at the undergraduate level, during graduate/medical training, and during the residency/post-doctoral fellowship period. Junior faculty need forums to present their work, get feedback, and build networks. Trainees at all levels lack sufficient exposure to individuals with mental retardation and developmental disabilities and co-occurring emotional and behavioral problems.
- **Junior faculty face pressure to publish discrete inquiries in which they retain lead authorship.** Tenure committees frequently do not assign significant value to participation in large, interdisciplinary investigations characteristic of work in this area.
- **There is insufficient funding for seasoned investigators to mentor those in training or young investigators.** Not all mentors are "good" mentors; it is difficult to get good trainees to the right mentor.

Recommendations

- **Encourage a variety of training pathways and mechanisms for different individuals and disciplines.** Systematically evaluate all current research training mechanisms, seeking feedback from junior trainees who have successfully entered research careers and those who have chosen other career paths.
- **Consider which other disciplines should be at the table.** Special education; speech, language, and communication; social work; nursing; and occupational therapy are among the fields that could contribute to this discussion.
- **Look to program models in other areas to develop new training mechanisms.** Work group members highlighted the NINDS Neurological Sciences Academic Development Award and the NICHD Pediatric Scientist Development Training Program as examples.
- **Augment existing support and seek new avenues to channel support.** Increase stipend and research dollar funding for training MD clinical investigators (the NIH T32 program). Add other training funds to University Centers for Excellence in Developmental Disabilities, UCDDs (formerly known as the University Affiliated Programs or UAPs) and other centers where research on mental retardation and developmental disabilities is performed. .Craft a specific request for applications (RFA) to encourage research on the emotional and behavioral health of people with mental retardation and developmental disabilities. Develop new support for research training during medical residency and the post-residency fellowship years.
- **Create an NIH inter-institute coordinating group to promote research and training on the emotional and behavioral health of people with mental retardation and developmental disabilities.** Designate a program officer at each relevant Institute to shepherd research in this area. Create special review groups for research training proposals and develop intramural programs for research on the emotional and behavioral health of individuals with mental retardation and developmental disabilities.

- **Create training centers of excellence in mental retardation and developmental disabilities research to promote the value of training efforts.**
- **Support and augment existing research training networks that could expand training of researchers in mental retardation and developmental disabilities.** These include the UCDDs, the Mental Retardation Research Centers funded by NICHD, as well as the Leadership Education in Neurodevelopmental and Related Disabilities program (LEND) funded by the Maternal Child Health Bureau.
- **Create topical career development clubs for young investigators.** Use these as a vehicle to encourage early career mentoring, network building, and as a source of information about available funding opportunities.
- **Create new categories of special individual research career development awards ("special K's") for investigators at various stages of their careers.** Craft a special-K RFA for researchers interested in the emotional and behavioral health of people with mental retardation and developmental disabilities.
- **Seek private foundation partnerships.**
- **Develop a plan to monitor progress from the outside.** The Institute of Medicine represents one venue with experience in charting change and setting priorities.

Epidemiology

The Working Group on Epidemiology noted that epidemiology is the study of the distribution and dynamics of health and illness in human populations. An important aspect of epidemiology is research on the functioning of health services. Epidemiology is more than simply counting how many individuals are affected and measuring prevalence and demography. Epidemiology can teach us about the nature and scope of mental retardation/developmental disabilities and the associated behavioral, emotional, and psychiatric problems. Modern experimental approaches to epidemiology allow the study of causative processes, factors that influence the course of the disorder, and service needs. Epidemiological methods often are combined with other methods (for example, psychosocial and neurobiological measures). Epidemiological studies can disclose individual developmental trajectories and the influences that shape those trajectories. Some of these influences promote risk; others provide protection and promote resiliency.

Although we have an approximate understanding of the prevalence of emotional and behavioral disturbances in individuals with mental retardation, there are almost no studies on service utilization. We must ask which new data are needed in order to design and conduct research aimed at improving the functioning of persons with mental retardation?

Critical Issues

- **There is a lack of adequate data on risk and protective factors for mental illness in people at different developmental stages, including preschool, school entry, school-to-work transition, and aging.**
- **New opportunities are available to study the interaction between genes and the environment.** It is expected that variations in the heritability of behavioral or cognitive traits will be observed across different social levels and environmental conditions.
- **Issues in sampling and measuring behavior and related characteristics, including adaptative behavior, are critically important.**

- **Ensuring adequate sample sizes is paramount.** For example, many individuals with mild mental retardation may be difficult to locate beyond the school years. In addition, recruiting adequate numbers of individuals with more rare neurogenetic syndromes may be difficult.
- **There is a need to examine behavioral features, emotional problems, and psychiatric diagnoses in groups with specific neurodevelopmental syndromes, including Down syndrome, Fragile X, and Prader Willi syndrome.**
- **Systems research studies are needed, including investigations focusing on state systems, residential units, group homes, supported living settings, and prisons.** Understudied areas include: (1) factors leading to the success or failure of de-institutionalization and (2) the probable increase in use of medications during the last decade.

Recommendations

- **Conduct longitudinal studies to examine key life-stage transitions regarding risk and protective factors.** Comparisons should be made of such factors as time of diagnosis of the disorder, residential status, and family functioning. Although the Working Group focused primarily on concurrent longitudinal designs in its recommendations, the group also recognized the utility of case-control designs for behavioral genetics research and non-concurrent longitudinal designs that make use of record linkages.
- **Use informative samples and innovative research designs for behavioral genetic research, including twin pairs studied from birth and informative family and sibling studies.**
- **Develop and refine an array of measures of outcomes and hypothesized risk and protective factors.**
- **Ensure that those with mild mental retardation or rare neurogenetic disorders are sampled adequately.** Exploit opportunities to piggyback on existing surveys, such as the CDC health risk behavior surveys and the National Health Interview Survey.
- **Conduct syndrome-specific cohort studies to: ascertain specific vulnerabilities to common disorders; inform analysis of interactions between genes, brain, and behavior; and improve understanding of functioning.**
- **Conduct system research studies that target state systems, schools, residential units, group homes, supported living settings, and prisons.**
- **Consider convening another workshop to address the need for systems research studies.**

Diagnosis and Assessment

The Working Group on Diagnosis and Assessment noted that psychiatric disorders have a profound effect on the well being of people with mental retardation and developmental disabilities. The prevalence of emotional and behavioral disorders among this group is greater than in the general population, and the consequences of such disorders are more severe, through their impact on careers (both family and professional care-providers), and because they can prevent those affected with these disorders from being included in family and community life. As a result, the cost to the nation is great.

A major impediment to our ability to treat these disorders effectively in people with mental retardation and developmental disabilities is our inability to recognize mental health problems in these individuals. Assessment and diagnosis of emotional and behavioral disturbance is particularly difficult due to intellectual, adaptive, and verbal impairments that limit reliability or

reporting, and the presence of organic or environmental factors that either produce or exacerbate the specific forms of pathologic behavior. Receipt of services is also frequently linked to the diagnosis of a specific condition or the severity of functional impairment related to it.

As a consequence of difficulties with diagnosis and assessment, persons with developmental disorders may not receive effective treatments, the treatment they receive may actually be harmful, and they may be denied important services. Additionally, the lack of diagnostic tools holds back basic research into the etiology of mental retardation, without which we cannot expect to develop effective rational therapies.

Critical Needs

- **Research aimed at developing appropriate diagnostic procedures applicable to individuals with mental retardation and developmental disabilities.**
- **Investigations to evaluate the clinical utility of assessment procedures in treatment trials.**
- **Studies to identify the biological and environmental determinants of emotional and behavioral disorders in individuals with mental retardation and developmental disabilities.**
- **Research to determine whether differences in the expression of emotional disorders are due to cognitive or functional impairment.** The complex interrelationship between the expression of emotional disorders and cognitive and functional impairment places additional demands on the adequacy of assessment procedures.

Recommendations

- **Conduct research to assess the validity and reliability of adaptations of standard diagnostic and assessment strategies to accommodate people with mental retardation and developmental disabilities.**
- **Develop direct observation instruments to identify overt characteristics of emotional disturbance and their environmental correlates.**
- **Undertake studies to evaluate the tolerability of the diagnostic and assessment process for people with mental retardation and developmental disabilities and their families and care providers.**
- **Develop techniques to assess the impact of psychosocial stressors in the lives of people with mental retardation and developmental disabilities and to integrate this knowledge with diagnostic protocols, treatment strategies and service systems.** It is equally important to determine protective factors and methods to quantify an individual's resilience.
- **Undertake efforts to characterize the phenotypic diversity of mental retardation and developmental disabilities.** Develop appropriate animal models to explore how genetic abnormalities give rise to mental retardation. In addition, complete neuropsychological assessments of mental retardation and developmental disabilities to determine the pattern of cognitive disabilities and competencies and associated behavioral abnormalities present in various forms of mental retardation and developmental disabilities.
- **Conduct prospective studies to describe the developmental trajectory of behavior and skill acquisition in genetic disorders or syndromes associated with mental retardation and developmental disabilities.** While it is often assumed that the pattern of abilities and

impairments will be maintained throughout development, available evidence suggests this is not so.

- **Carry out research aimed to uncover how the manifestations of particular emotional and behavioral disorders and the response to treatment may vary as a function of cognitive or functional disability or developmental level.**

Interventions Research

The Working Group on Interventions Research said that advances in the fields of genetics, neuroimaging, brain plasticity, behavioral sciences, and education show promise in generating effective interventions to improve the emotional and behavioral health of individuals with mental retardation and developmental disabilities. The group noted that there are a number of behavioral and psychosocial treatments that have been well researched in the general population, but have not been tested for their efficacy in individuals with mental retardation and developmental disabilities. Similarly, newer and safer medications are available for a range of emotional and behavioral disorders, yet few rigorous tests of their efficacy in individuals with mental retardation and developmental disabilities have been performed. These and other challenges present critical opportunities for well-designed intervention studies in people with mental retardation and developmental disabilities.

Critical Opportunities

- **In genetics, characterization of specific behavioral phenotypes associated with varied genetic etiologies of mental retardation and developmental disabilities show promise in allowing researchers to craft and target interventions for specific disorders or behavioral disturbances.** Research identifying genes that code for new target proteins and the development of drugs that target those proteins could lead to better focused pharmacologic treatment strategies.
- **In neuroimaging, scientists have begun to document the effects of environment and experience on the brains of individuals with mental retardation and developmental disabilities using a variety of imaging techniques.**
- **Research on the plasticity of the brain, much of it accomplished in animal models, has illustrated the profound effects of early experience on brain development and development of aberrant behavior.** Research has shown that the consequences of central nervous system damage, even as a result of genetic mutations, can be ameliorated by complex environments. Evidence is also accumulating that neural regeneration, in at least some areas of the brain, is possible across the life span.
- **In psychosocial research, functional analysis of behavior has been shown essential for prescribing effective behavioral treatments for people with mental retardation and developmental disabilities who display severe behavior disorders.** Communication training has been shown to reduce the occurrence of emotional and behavioral problems. Intensive early educational interventions produce long-term gains in social and intellectual functioning in some at-risk populations**.**

Recommendations

- **In genetics, assess the differential effects of genetic etiology on treatment outcomes.**

Develop psychotropic drugs selective for newly identified target proteins and test in animal models and controlled clinical trials. Test the effects of experiential/behavioral manipulations on gene expression.

- **Use neuroimaging as a technique for assessing treatment outcome (along with behavioral and clinical outcomes).** Use neuroimaging to establish likely mechanisms of treatment effects.
- **In neuroplasticity research, test theory-based interventions for specific populations, using an array of new technologies to document central nervous system changes as well as behavioral outcomes.** Evaluate importance of age, timing, and intensity of intervention on neuroplasticity. Utilize appropriate animal models to test neuroplasticity associated with developmental insults to the central nervous system.
- **In psychosocial research, evaluate the impact of using functional analysis to test the efficacy of different treatment techniques and in a wider variety of emotional and behavioral disorders.** Test the efficacy of intensive early intervention and communication training to prevent or reduce emotional or behavioral disorders in children with mental retardation and developmental disabilities.
- **Assess the effects of combining pharmacological treatments with behavioral, psychosocial, and educational interventions and/or natural supports.**
- **Develop innovative research designs to address the potential confounds of ongoing treatments and co-occurring conditions.**
- **Urge revision of Food and Drug Administration drug approval standards to include alternative research designs.**
- **Engage consumers and family members in design, implementation, and evaluation of interventions.**
- **Create a federal task force to develop and implement an interdisciplinary clinical research network.** Fund meetings to create coalitions of researchers, advocates, and service providers.
- **Place funding priority on testing promising interventions.** Review portfolio of NIH funded studies that address prevention or treatment of emotional and behavioral disorders. Determine the feasibility of supplemental funding to add participants with mental retardation and developmental disabilities.

Research Design

The Working Group on Research Design noted that the primary purpose of research design is the production of unbiased and efficient data that can be used to serve the needs of all partners in the research enterprise. The group affirmed that while a multiplicity of research designs and analyses can be valid for addressing specific problems and questions in individuals with mental retardation and developmental disabilities, the randomized, controlled, blinded protocol remains the "gold standard" against which other research designs should be judged. In current practice, research on individuals with mental retardation and developmental disabilities frequently relies on single-subject case reports, which do not uniformly produce optimal, valid answers. Through more informed decision-making about research designs and their fit to research questions, the state of knowledge about the emotional and behavioral health of individuals with mental retardation and developmental disabilities could be improved.

Recommendations

- **It should be the normal expectation that individuals with mental retardation and developmental disabilities will be included in federally funded research.** The investigator must assume the responsibility for justifying any exclusionary rules. Exclusions should be based on the functional and safety features of the experiments or interventions being studied, not sweeping criteria, such as "IQ less than 70," that frequently exclude many individuals with mental retardation and developmental disabilities from research.
- **Research designs that emphasize our understanding of "for which person" and "under what circumstances" should be emphasized in research on mental retardation and developmental disabilities.** These research designs are preferred to models that only yield estimates of group means, such as ANOVA and regression models.
- **Multi-site, multi-collaborator designs should be the norm, not the exception.** These designs need to be utilized creatively and not limited to single protocols that are simply replicated at all sites.
- **Research designs used to assess the outcomes of interventions in individuals with mental retardation and developmental disabilities should include measures that assess functional and clinical improvement.** In general, a single variable, or a small number of variables, should be pre-specified as the outcome variable of interest.
- **Longitudinal follow-up is critical for a full understanding of the consequences of an intervention.** Longitudinal follow-up should be pre-planned and examine both the primary outcome variable and other potential mediating variables.
- **Creating an archive of complete and well-documented data sets is essential for the development of cumulative science.** Federally supported researchers should expect to make their data available to the research community after an appropriate delay for publication of the study's primary outcomes.
- **For clinical studies designed to elaborate new therapeutic applicability of a drug, early intervention with the FDA is essential.**

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