



## American Academy of Developmental Medicine and Dentistry

# Developmental Medicine and Dentistry Reviews & Reports



**Message from the Editor:** In this issue of “Reviews and Reports,” (R&R) in addition to my editorial comments, I have invited a clinician who is familiar with clinical research in people with intellectual disabilities to comment. Dr. Glick has spent a significant portion of his professional career caring for children and adults with disabilities as well as investigating biological and biochemical phenomenon that may have unique presentations in people with disabilities. He notes:

*This issue of R&R explores the role of research in the lives of people with disabilities. Taylor Spencer offers four potential benefits of such efforts, and the discussion warrants serious deliberation despite the complex and controversial nature of the subject. Considering we now know—medically speaking—that children are not just little adults, that women are not like men, and that older people are not like middle-aged people, it is time to recognize that people with*

*disabilities may not be like their age- and gender-matched peers. Rather, they are prone to their own unique manifestations of unique diseases. Meaningful, appropriately conducted research may help bridge the enormous gap in everyday management expertise that now exists*

—Norris R. Glick, MD

Taylor proposes using somewhat different research methodology for people with intellectual disabilities. This may reflect the difficulties of engaging in this kind of research rather than a clear need to actually change research methodology. However, the importance of seeing the individual separate from a larger group as well as part of a larger group is a key concept that should be considered in any research design. And while Taylor focuses on social research, the same can be said of basic research investigating genetics, biochemistry, and neurobiology.

—Steven G. Zelenski, D.O., Ph.D. Co-Editor, R&R

# The Role of Research in the Lives of People with Disabilities: Too Little Too Late?

TAYLOR SPENCER

## Research: What have we lost and what are we losing?

As a medical and public health student, I have researched quality of life in older children and young adults with disabilities. In its third year, however, the research was questioned by a fellow medical student who had been asked to help obtain consent from volunteers and their parents at a summer camp. The medical student replied that she was uncomfortable,

*“...most importantly having to do with the kids. I feel that attempting to get them in the study today...will jeopardize the relationships that we’ve spent all week building. I don’t want them to feel as though the only reason I came to camp this week was to collect data ...!”*

The idea that research trivializes personal relationships established during the week at camp is very damaging, if true, but may not be unexpected. The

roots of criticism of social research on disability can be traced back to the 1960s<sup>2</sup> and then later with Hunt’s critique as an often-cited description of victimization by research. As a resident with disabilities of Le Court Cheshire Home, Hunt became disillusioned by the “unbiased social scientists” who followed their own agenda while ignoring the residents they consulted<sup>3</sup>. Qualitative research interviewing individuals with disabilities indicates a persistence of this perception,

and the suspicion that “academic researchers do not approach a project from a neutral, objective position but are situated within constructed and historically-rooted discourses of knowledge and power” with predetermined agendas that may do little to benefit their subjects<sup>4</sup>. Oliver goes so far as to condemn it as “a rip-off that has done little if anything to confront the social oppression and isolation experienced by disabled people or to initiate policies, which have made significant improvement in the quality of their lives<sup>5</sup>.” These thought-provoking opinions challenge research. Is research defensible?

The following are thoughts about the nature of my study, about public health research, and research with youth with disabilities. The dialogue on the topic is probably most developed for those with intellectual disabilities, with the extra issues such as guardianship, but can be extended to a discussion of all disabilities research. It is an important conversation to have with those who are the most significant—youth, families, and all interested stakeholders—because the only effective way to understand the experience and make improvements is with the active involvement of all affected.

### **1. Research can show that the healthcare community takes interest in individuals with disabilities.**

Thoughts of research often conjure up

images of pharmaceutical projects funded by big corporate sponsors or guided by government agencies. In a culture largely guided by financial gains, it is not surprising that disabilities research is limited. It is not a “popular” research topic like HIV or cancer. And private companies, including the pharmaceutical industry, have little to gain in a “niche” market that may not benefit from drug treatments. Of the president’s proposed \$2.4 trillion budget for 2005, only \$107 million had been designated for the National Institute for Disability and Rehabilitation Research (NIDRR)—about one half of one thousandth of a percent—while the National Institute of Child Health and Human Development received \$1.281 billion and the National Institutes of Health, as a whole, was proposed funding at \$28.773 billion<sup>6</sup>. Although there is a considerable amount of money, only a small piece is for disabilities research.

While disabilities research does not need to be constrained by financial interests, we must also be cautious of other motivations which ultimately will not benefit people with disabilities. Such research can be plagued by the academic greed of researchers who focus more on their own advancement than their subjects. Mohan describes a system of “systematized selfishness” in which the oppressed group is “mined” for information without being given anything in return<sup>7</sup>. New models for the research

process and interactions seem to be taking steps in addressing this concern.

A small select group of researchers shares a genuine interest in the health of those with disabilities, and awareness of its significance is growing. Healthy People 2010, the set of national health objectives for this decade, is the first time the initiative specifically addresses the range of physical and cognitive disabilities. Chapter 6 entitled “Disability and Secondary Conditions” establishes goals of promoting the health of people with disabilities, preventing secondary conditions, and eliminating disparities between people with and without disabilities. In fact, elimination of health disparities for people with disabilities (and others) is one of the program’s two explicit goals<sup>8</sup>.

The research project which was questioned by the other medical student is part of a long term project. The project consists of survey and interview data assessing the relationship between functional independence, family dynamics, and quality of life. Additionally, it has expanded to include qualitative research on the transition from adolescence to adulthood and pediatric to adult health-care. Each year, medical or dental students participate on the research team, in part to foster their education. Training of public health professionals is an identified need<sup>9</sup>, and while little more than half of US public health schools (60 percent),

*continued on page 80*

## **INSTRUCTIONS**

For CME Credit read the editorial at the top of page 78 and the article that follows and complete the Content Test and CME Evaluation Form at the end. Please read “Information and Instructions” following the article.

Specific learning objectives for this CME activity (please refer to general objectives).

Upon completion of the reading of this article the learner will be able to:

1. Appreciate the importance of research in providing optimum care to people with disabilities

2. Describe the challenges that continue to interfere with research for people with neurodevelopmental/intellectual disability creating a significant health care disparity.

3. Describe key factors that might be changed to enable research with people with disabilities.

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*continued from page 79*

offer graduate-level courses focused on disability, less than 15 percent have graduate-level tracks or concentrations in disabilities<sup>10</sup>. Reflecting the need for greater training by the medical field for disabilities, Canadian psychiatric residents have little interest or comfort with intellectual disabilities<sup>11</sup>. Research is an opportunity for healthcare to show an interest in individuals with disabilities and address an individual physician's discomfort with physical or cognitive disabilities. Although financial resources are limited, it does not mean that the interests and concerns of the medical community and students should be. To overlook or otherwise marginalize the population of those with disabilities cannot be justified.

## 2. Research can allow for a voice

While conducting the study, I noticed that participants were truly receptive and supportive. Perhaps the fact that research was occurring at all was encouraging. They eagerly awaited results. A major reason those with disabilities distrust research is the feeling that they do not get a return on their involvement<sup>4</sup>. Research of this nature provides the participants with a voice, to be counted for something that might amount to more than individual voices, and a chance to share their concerns. Individuals with disabilities have stories to share about their healthcare. Too often, they are subject to a healthcare system that doesn't listen to them or understand them, that doesn't meet their needs, and that ultimately drives them away<sup>12</sup>. In interviews with young adults with disabilities, participants have been incredibly open with their experiences, their hopes, and lessons. They want validation that their voice is heard.

Qualitative data amongst those actively involved in the disability movement shows their appreciation for active involvement in research as "a platform from where disabled people can speak for themselves, to seek the services and

support they want, to explicitly influence social policy and fight for disabled rights<sup>4</sup>." The participants in research are no different from anyone else—when they see an opportunity to be heard and make a difference on an issue that matters to them, they tend to feel empowered. Healthcare in general—not just for those with disabilities—could learn from these stories. To do so, actively engaged participants drive successful research that is more than averages and correlations and capture aspects of their greater experiences. Everyone need not choose to take the opportunity, but we all lose when a would-be voice is ignored.

Ultimately, this opportunity to speak out is supported by the emerging paradigm for disabilities research. There is a preference for less rigid research methods, with open qualitative data providing the opportunity to add the individual's voice and context with greater independence from researcher manipulation. The disabilities studies literature also values empowerment, seeking positive change in the individual through participation. Both these ends are not simply a consequence of the research topic, but also the research strategies, including a strong emphasis on including those with disabilities in consultant, partner, or even lead researcher roles. Exposure and involvement in research can be an educational and empowering tool for subject or caregiver. For those able to engage in the discussion, it raises awareness of literature often reserved for academics "in the know," breaks down barriers between academia and the "real world," and develops a sense of ownership. Although many are at first intimidated, participation benefits the participant as much as the researcher by developing and refining their voice<sup>4</sup>.

## 3. Research can have a broader goal in mind

I will admit it: there is selfish motivation in my research. I chose to do something I would enjoy. I wanted to work with indi-

viduals with disabilities because of my own experience as a summer camp counselor. And I am eager to benefit my research population.

My advisor and I have discussed the possible outcomes for our study. This research is designed to clarify the nature of health-related quality of life, with the broader purpose of benefiting individuals with disabilities in today's world. I was excited to realize that, no matter the results, we could contribute meaningful answers to the literature.

Stone and Priestly include the surrender of claims to objectivity in their core principles for a reformulated research strategy. Instead, they endorse overt political commitment to the struggle for self-emancipation, removal of disabling barriers, and practical benefit to the disabled subjects<sup>13</sup>. "Action-led" research explicitly aims to use research to guide actions that change social relationships<sup>4</sup>. This is fundamental in the social model of disablement, which identifies disability as a consequence of society's organization and responses rather than a direct consequence of physical impairment (as it is in the World Health Organization's International Classification of Impairments, Disabilities and Handicaps)<sup>2</sup>. The Union of the Physically Impaired Against Segregation (UPIAS), formed in Britain after the uproar over the research at Le Court Cheshire Home, were early supporters of this model and focused on "overcoming the disabilities which are imposed on top of our physical impairments by the way this society is organized to exclude us<sup>14</sup>." Given this context, research on disabilities is not significant for any potential to change the disabled subject, seen in normalization theory's efforts to reduce stigmatization and devaluation<sup>15</sup>. Rather, it is the potential to change social organization that gives meaning to "emancipatory research<sup>4</sup>."

Disabilities research is particularly demonstrative of this potential for research. Perhaps research in general should be more open to its broader appli-

cations, rather than trying to ignore them. It might be better if researchers and healthcare professionals were a bit less uncomfortable with the interplay between their professional obligations and their personal motivations, such as the desire to benefit their research population. Cold objectivity in research may not be the best answer and is not a prerequisite for academically rigorous work. Disabilities research allows for research with a heart.

#### 4. Research identifies areas for improvement and leads to more questions

Research projects only capture a fragment of a broad and complex subject matter. My research will not definitively answer all questions regarding quality of life or the transition process, for example. And no one project can stimulate or redirect a broader research agenda. But research is not worthwhile simply for the extent to which it understands a phenomenon. It is worthwhile because of the context. There is inadequate research that describes how we can improve the quality of life for people living with disabilities, and because of the frequent difficulty in communication, many assumptions exist. In fact, the potential is boundless. Each study raises new meaningful questions, as much as it answers others. Research stimulates discussion, promotes new research, and changes policies that may benefit individuals with disabilities. If interest is raised through the results and by including others in the process, this may benefit society. New services arise only when there are strong supportive voices<sup>16</sup>, and these voices need something to discuss.

Much of this discussion-generating potential depends on the strength of the data. Strong data serves as the foundation for more strong data. Some researchers anticipate a standardized disability status indicator to allow for comparisons within or between those with disabilities and who are non-disabled, or between differ-

ent conditions. This might promote a better understanding of issues but requires subject participation throughout the process and at various levels. “Interventions targeting people with disabilities can then be justified” as reliable evidence highlights needs<sup>12</sup>. Currently, there are few large epidemiological studies of the health of adults with intellectual disability, for example, despite valid concerns of poor health<sup>17, 18</sup>.

The ability to identify new issues and ask relevant questions is strengthened by involvement of those with disabilities 19 more than through the research alone. Exclusive research is “conducted by a (or team of) non-disabled or disabled researcher(s)” whereas inclusive includes both groups on the same team<sup>4</sup>. This involvement of researchers with disabilities would mean the individual with disabilities enters a “third-space” between researcher and researched, academic and activist<sup>20</sup>. They provide context, identify concerns as “expert knowers”<sup>2</sup>, and escape the dominant power relationships. They give research an extra degree of flexibility to pursue new and meaningful directions. The emancipatory potential of such a position, by shaping the discussion on the research and guiding future directions, can be profound.

Disabilities research is subject to discomfort and uncertainty. This is a combination of a perception of research as cold and self-serving, treating participants as “guinea pigs,” and a misperception of individuals with physical or cognitive disabilities as fragile or needing protection. In response to this sentiment, the “Common Rule” (Federal Policy for the Protection of Human Subjects) can be applied to assure compliance by research institutions, establish requirements for informed consent, and outline the elements of Institutional Review Boards (IRBs)<sup>21</sup>. The Common Rule labels “handicapped or mentally disabled persons” as a vulnerable category of subjects and states “consideration shall be given to the inclusion [in

the IRB] of one or more individuals who are knowledgeable about and experienced in working with these subjects<sup>21</sup>.” But the biases of the medical community may be apparent in the IRB review process—evidence indicates that such reviews are not objective<sup>22</sup>, and that physicians are significantly more likely to have a negative prognosis for individuals with cognitive disabilities.<sup>23,24,25</sup> It is reasonable, although unproven, to assume that this extends to physical disabilities and that, in fact, they may obscure the differences between physical and cognitive functioning. IRBs may be further discouraged by court cases such as Maryland’s *Grimes v Kennedy Krieger Institute, Inc* which ruled “otherwise healthy children should not be the subjects of non-therapeutic research that has the potential to be harmful to the child...consent of parents can never relieve the researcher of this duty<sup>26</sup>.” Requiring absolutely no potential for harm of any degree when guardians provide consent becomes very restricting to the research process. The case of *TD vs. NY* also found that the state’s Office of Mental Health can not issue regulations allowing experiments to be performed on patients in psychiatric facilities without the patients’ knowledge or consent violate the federal and state constitutions, state statutes and the common law<sup>27</sup>. These cases are cited as evidence of the legal and judicial barriers to some research. Perceptions of vulnerability have led to insufficient research on populations with disabilities—it is thought to be not worth the risks (or the modifications needed to avoid risks), and therefore an emphasis on the principle of non-maleficence (avoiding harm) over the principle of justice.

But neglect is not a suitable substitute for protection from harm. Research may be a powerful tool to change policies that may improve quality of life. And quite often, individuals with disabilities are excited to participate in this process.

*continued on page 82*

continued from page 81

Vulnerable individuals in general—defined by economic, social, psychological, physical health, or child maltreatment status—had a stronger emotional reaction to research but were not less willing to continue to participate in a lengthy and intrusive in-person interview and blood sample collection<sup>28</sup>. If willing, they should be afforded the opportunity to participate. But it can often be difficult to recruit within this select population, regardless of their willingness. A three-tier process of negotiating access has been described, involving first the permission of various organizations and ethics committees, second with professionals who facilitate access, and third with the individuals with disabilities themselves<sup>19</sup>. This

can be overwhelming and could vary significantly based on the social framework around the individual and the relative strengths of the three tiers. Recruiting those with intellectual disabilities, in particular, can be further complicated by “frequent need for substituted decision making, the occasional limited literacy of both person and caregiver, and organizational gate-keeping practices<sup>29</sup>.” Even in IRB-approved research, these difficulties can deter positive research and can make neglecting such research an easier alternative, regardless of the wishes of the study population<sup>29,30</sup>.

When these barriers are overcome, research itself is not a “magic potion.” But when pursued in an enlightened way, its role can be profound. You may

lose the deeper understanding when you fail to see those with physical or cognitive disabilities on an individual level. But you also lose something when you are not able to connect them to a larger system or understand the role and experience of the collective in all its complexities. To avoid research (and to avoid providing the opportunity to actively participate) out of fear, no matter how well-intentioned, is a disservice to those we hope to serve. •

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