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SYMPOSIUM

Scientific Reasons for Including Persons with Disabilities in Clinical and Translational Diabetes Research

Shirley M. Moore, R.N., Ph.D., FAAN

Abstract

Despite disparities in health problems and outcomes, people with disabilities are underrepresented in diabetes research. This results in a lack of evidence-based knowledge regarding best approaches in caring for this population. This article addresses the need for research that includes people with disabilities and describes the common reasons persons with disabilities are not included in research, including scientists' concerns regarding threats to a study's internal validity and cost. Arguments are provided as to how involving people with disabilities in research will improve our science and reduce disparities in this population. In addition to the ethical reasons for including persons with disabilities in research, the ability to generalize study findings to this population and thus speed our development and translation of this knowledge for use by clinicians is discussed. The bias in study conclusions that arise from study samples that do not include persons with disabilities and its possible effect on care delivery are presented. Two strategies that researchers can use to increase the inclusion of persons with disabilities in research are described: (1) Universal Design of Research and (2) intervention optimization study designs. Universal Design of Research includes research design processes such as the use of multisensory formats for recruiting participants, approaches to designing and presenting research instruments and interventions, and methods of data collection to promote the inclusion of participants with a wide range of abilities in research studies. Intervention optimization study designs offer an efficient way for scientists to rapidly build the most potent interventions for a wide range of people, including those with disabilities participating in mainstream research.

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The Need for Diabetes Research That Includes More Persons with Disabilities

It is an interesting paradox that a population that could benefit the most from diabetes research, persons with diabetes who have disabilities, is often excluded from research because of these disabilities. Optimal delivery of health care is based on evidence of what is most effective, for whom, and under what conditions. However, studies to produce evidence about diabetes care are often not designed to be accessible for people with disabilities.

Author Affiliation: Frances Payne Bolton School of Nursing, Case Western Reserve University, Cleveland, Ohio

Abbreviations: (MOST) multiphase optimization strategy, (SMART) sequential multiple assignment randomized trial, (UD) universal design, (UDR) universal design of research

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Corresponding Author: Shirley M. Moore, R.N., Ph.D., FAAN, Frances Payne Bolton School of Nursing, Case Western Reserve University, Cleveland, OH 44106-4904; email address smm8@case.edu

This results in a large proportion of people with diabetes who are not included in research studies and whose needs cannot be addressed adequately using the conclusions drawn from these studies. Thus, we are left with gaps in knowledge, or perhaps even distorted knowledge, about the needs of persons with diabetes who have disabilities. In this article, the need for diabetes research that includes people with disabilities is described, including reasons why involving people with disabilities in research will improve our science, and how diabetes research can be designed to enhance our knowledge about people with diabetes who have disabilities.

There is a high incidence of disability among the nearly 20 million people in the United States who have diagnosed diabetes.[1] For example, the number of people with diabetes who report visual impairment (trouble seeing even with glasses or contact lenses) is 3.8 million,[1] and as many as 54% of people with diabetes report hearing loss.[2,3] In 2009, the age-adjusted percentage of adults with diabetes reporting a mobility limitation was 34.0% for walking a quarter mile, 26.9% for climbing up 10 steps, and 43.1% for stooping, bending, or kneeling.[1] Hospitalization rates among people with diabetes also reflect the burden of disability in this population. In 2006, the age-adjusted rate for hospitalizations for nontraumatic lower extremity amputation per 1000 diabetic population was 3.5.[1] Also, in 2007, the number of hospital discharges among people with diabetes was 84,000 for peripheral artery disease, 113,000 for lower limb ulcers, and 75,000 for neuropathy.[1] Chronic illnesses associated with varying degrees of disability, such as arthritis and cardiovascular disease, also occur with great frequency in persons with diabetes.[1] Additionally, individuals with disabilities are more likely than people without disabilities to report poorer overall health, less access to adequate health care, smoking, and physical inactivity.[4,5]

Multimorbidity or co-occurrence of disability limitations and/or medical diagnoses in persons with diabetes presents a unique set of challenges not only in clinical practice, but also in research. First, because of their multimorbidity, persons with diabetes who have disabilities have been typically excluded from various types of clinical studies. For example, disabilities such as sight or hearing impairments have erroneously been considered by researchers to prevent individuals from giving consent to participate in a research study. The absence of these individuals from studies has severely limited the generalizability of the findings and left a substantial gap in our knowledge relative to the effectiveness of various therapies and interventions for this population. Second, the lack of clinical data on this population has channeled our research efforts to observational studies, which rely heavily on secondary data, including survey, clinical assessment, and administrative databases. While each of these data sources is useful in its own way, the fact that these databases represent conceptual silos has hindered our ability to use them in diabetes research to a great extent. Additionally, knowledge gained from only observational studies greatly restricts our evidence for best intervention approaches in care delivery. Together, these limitations have left us unable to gain a good understanding of the disparities in diabetes-related outcomes that are associated with diabetes multimorbidity and disability status.

Reasons Why Scientists Do Not Include More People with Disabilities in Research

Several reasons exist for why people with disabilities are excluded from participation in mainstream research studies. These reasons can be categorized as scientific concerns and feasibility concerns. A major scientific reason is that scientists are concerned about maintaining the standardization of their research protocols to preserve the internal validity of the study. Scientists often are unclear about the extent to which a protocol can be altered to accommodate a participant's functional limitation, such as use of an alternative data collection method or intervention delivery approach, and still assure the internal validity of the study. Another reason given by scientists for not including persons with disabilities in mainstream research is that the number of people with disabilities who participate in any one study would be very few, thus presenting analytic challenges and the inability to draw statistically sound conclusions that would apply to that group. Fortunately, new study designs have emerged that allow for subgroup analyses with small numbers of participants in a subgroup category within a larger trial.[6] These approaches are described later.

Many of the feasibility concerns for not including people with disabilities in research stem from inaccurate conceptions regarding the abilities of people with disabilities to participate in research. The *Healthy People 2020*[7] discussion on disabilities attributes the paucity of research to the common view among health care professionals that disabilities are end points in health research, rather than seeing them as simply facts of life for these individuals. There is a lack of recognition that people with disabilities have many abilities and have ways of successfully navigating their way through their daily activities, which could include participation in research studies.

Another feasibility issue is how to promote access to research studies for people with disabilities. Investigators may not know what recruitment approaches are needed to help people with disabilities become aware of a research study opportunity. For example, special outreach activities and recruitment materials may be needed to help persons who are blind or deaf learn about a research study. Investigators may also be concerned that a person with a disability may not be able to meet the study requirements to participate at a particular time and place for data collection or administration of the study protocol. Study exclusion criteria frequently contain statements regarding the ability to participate in a standard protocol, such as inability to read or inability to come to the treatment center. Additionally, as mentioned earlier, investigators may think that people with sight or hearing impairments would be unable to give informed consent, thus precluding these populations from participating in research.

Another common reason that researchers do not include people with disabilities in research is the view that the cost of including them is prohibitive. Investigators frequently believe that expensive equipment is needed to put research materials into formats that can be accessed by persons with disabilities, and often research staff lack knowledge in designing research materials and procedures that can be used by people with disabilities. Most of the scientific community lack knowledge of low-cost community resources and contemporary approaches that are available to assist research teams adapt their study protocols in ways that would create access to research opportunities for persons with disabilities. For example, consultation with local community agencies serving persons with sight or hearing impairments often results in allaying investigator concerns about safety and may provide access to a vast range of equipment that can be loaned to investigators to assist persons with disabilities to participate in a particular study protocol.

Unfortunately, it is not possible to determine the number of people with disabilities who are excluded from research because of disabilities. Albeit unknowingly, scientists' current research approaches regarding inclusion and exclusion criteria, recruitment methods, intervention delivery modes, and data collection promote systematic exclusion of persons with disabilities from mainstream research. This results in biases in our findings and a lack of understanding of how to improve the health of this large segment of our population. Just as we have made adaptations in our research approaches for different ethnic and age groups, similar adaptations are needed to accommodate persons with disabilities in research.

Reasons Why Including Persons with Disabilities in Research Will Improve Our Science and Reduce Disparities

More Generalizable Scientific Findings

The significance of a study, in part, depends on the types of people to which the findings can be applied. External validity refers to the extent to which one can generalize the study findings beyond the sample. Thus, the inclusion of persons with disabilities in diabetes studies can increase the external validity of diabetes studies and, hence, their significance. Given the high incidence of disability in persons with diabetes, the inclusion of persons with disabilities in diabetes research will provide study samples that are more representative of the population. Generalizable knowledge produced in these studies will improve the usefulness of the research findings to clinicians and researchers alike. More practical clinical trials are needed that use exclusion criteria that reflect only characteristics necessary to ensure representativeness to the ultimate target population (versus strict exclusion criteria to ensure only those who will benefit most are included).[8] Such studies should be designed to address effectiveness in subpopulations and to compare clinically meaningful alternatives. They do not need to be extremely large or expensive.

Less Bias in Our Scientific Conclusions

More empirical knowledge regarding persons with diabetes who have disabilities is needed. Exclusion of a large proportion of the population in diabetes research creates a biased sample, which in turn may produce biased results. When clinicians apply research findings to a population that were not included in the studies that produced those findings, then ineffective, and perhaps even harmful, care may result. The lack of evidence-based interventions for persons with disabilities may be contributing to the disparities found in their health. Diabetes studies should include a description of patient participation rates and include the participation rate of persons with disabilities. Such descriptions will assist potential users of the study findings to assess the possible biases in the research results.

Greater Speed in the Use of New Knowledge

Inclusion of persons with disabilities in diabetes research will accelerate the rate of discovery of new knowledge for this population. Such studies will provide information

about what is effective for persons with disabilities, and under what conditions, thereby increasing the uptake of new knowledge by clinicians who care for this population. By increasing the reach of the study (including people with disabilities), translation of knowledge of interventions found to be effective is speeded up.[9,10] To improve uptake of the findings, studies should include a description of the adaptations made to the intervention to fit particular subpopulations, such as persons with disabilities, and documentation of the accommodations of the intervention to patient level of ability. Measurement of potential harms of the intervention and measures of the training and skills needed to deliver the interventions also are needed. However, such descriptions in research reports can be given only if persons with disabilities were included in the research. The inclusion of more people with disabilities in diabetes research will not only accelerate the rate of discovery of scientific knowledge regarding diabetes management in persons with disabilities, but will also increase translation of this knowledge by increasing the reach, effectiveness, adoption, implementation, and maintenance of new interventions that are found to be efficacious.

Ethical Reasons

There are also several ethical reasons for researchers to include persons with disabilities in research. The principle of justice requires a fair and equitable distribution of benefits. It cannot be considered fair when people with disabilities are excluded from the benefits of knowing if and how current health research applies to them. Also, persons with disabilities should have the benefits of access to research participation, e.g., investigational drugs and protocols, frequent monitoring, and payment for participation. Lastly, people with disability should have equitable access to the beneficial feelings of altruism that come from contributing to the common good by participating in research.

How to Include Persons with Disabilities in Diabetes Research

There are two major strategies that researchers can use to increase the number of persons with disabilities included in research. First, to promote greater access to research by persons with disabilities, research approaches can be designed to be consistent with the principles of universal design (UD). This includes the UD of recruitment methods, consenting processes, intervention delivery, and measurement and data collection methods. The second way to increase the number of persons with disabilities included in research is to use intervention optimization study designs to build and test the most potent interventions to produce optical outcomes for each participant in a study, including those with disabilities.

Use of Universal Design of Research

A concept that has received much attention in recent years is UD.[11] Universal design is defined as the design of products, environments, and services to be effectively and efficiently used by persons with a wide range of abilities to the greatest extent possible, without adaptation or specialized design.[12–14] It is also referred to as *design for all, inclusive design,* and *barrier-free design*.[15] The goal of UD is to create designs that can be used by as many individuals as possible in typical populations. Universal design shifts the focus of the design from an individual average user to a typical population of users who have a wide range of abilities. Universal design includes more individuals in a group that can easily use that design. In the application of UD, consideration of the human factors that may limit the usefulness of particular products and services at the design phase permits eventual use by persons with a wide range of abilities of products without needing special adaptations.

Several authors[16–18] have advocated that the concept of UD be applied to research to promote its inclusiveness. Harness and colleagues[17] describe the use of UD for making health electronic assessment instruments accessible to all persons, including those with disabilities. The authors point out that as more and more studies use and test information technology and electronic approaches for care delivery and research methodology, it is important that these approaches be designed with the needs of a broad set of users in mind, including those with disabilities. Williams[19] has described the use of UD in diabetes care, including the use of technology for care delivery by health care professionals and in the self-management of health by persons with diabetes.

Williams and Moore[16] have put forth a set of guidelines for the Universal Design of Research (UDR). Based on the principles of UD of learning (provide multiple means of representation, provide multiple means of action and expression, and provide multiple means of engagement),[20] the guidelines for UDR address the design of research processes so that all people can be included as potential participants, to the greatest extent possible, without the need for adaptation or specialized design. Elements of UDR include the use of multisensory formats for recruiting participants, approaches to designing and presenting

research instruments and interventions, and methods of data collection to promote the inclusion of participants with a wide range of abilities in research studies.

A laboratory has been established at Case Western Reserve University in Cleveland, Ohio, to provide resources to investigators about the design of their research methods, procedures, and interventions to facilitate fuller inclusion of persons with disabilities in mainstream research. The FIND Lab (Full INclusion of Persons with Disabilities in Self-Management Research) provides consultation, equipment loan, and hands-on workstations where investigators can design research projects, intervention materials, and data collection approaches to accommodate persons with disabilities in their research. More information about the FIND Lab can be accessed at *http://fpb.case.edu/FINDLab/index.shtm*.

Use of Intervention Optimization Study Designs

The need for a standardized or fixed intervention in randomized trials is often given as the reason for not including people with functional limitations in research studies. Researchers believe that functional limitations may impede a study participant's ability to engage in the intervention protocol and/or cause the participant to respond differently to the intervention. The fixed-intervention approach assumes that all individuals in a study will respond to all components of an intervention in the same way. However, we know that people respond differently to the same intervention, even when only persons without disabilities are included in the study. In contrast to the fixed-intervention approach is a new approach to behavioral intervention design, the adaptive intervention,[6,21,22] which does not assume that all subjects will respond similarly to all components of an intervention. Thus, the adaptive intervention design can be a useful approach to support the inclusion of people representing a wide range of abilities in a study.

An adaptive intervention is an intervention in which participant-specific modifications to the intervention are built into the intervention protocol based on explicit decision rules that are developed *a priori*. These decision rules link characteristics of an individual or environment with specific level and types of program components.[21] For example, in an exercise study, a blind person could engage in strength training in which lighter weights, more reps, and a caring spotter are part of a predetermined adapted protocol. In this example, *level of sight impairment* is the tailoring variable, and a decision rule regarding how the exercise routine will be adapted for any person entering the study who has severe sight impairment, such as blindness, is built into the study protocol in a prespecified way. To assure the internal validity of the study, use of such variables must be limited to the intervention arm(s) and to the specific purpose of modifying an *individual* participant's intervention based on observed values,[22] in this case, blind or not blind. The primary aim is to maximize the strength of the intervention for that individual. This approach acknowledges that the varying intervention needs of individuals may not be met by using a single uniform composition and dose. Strict adherence to the decision rules assures the internal validity of the study and its replicability.

There also are a set of new intervention optimization designs that can be used by researchers to empirically determine the best set of tailoring variables and decision rules to be used in an adaptive intervention protocol. These include the sequential multiple assignment randomized trial (SMART)[6] and the multiphase optimization strategy (MOST)[6,23] study designs. The SMART and MOST methods of intervention development use a series of small randomized trials and factorial designs to identify the most potent components to include in an intervention program.[24,25] These components are determined by characteristics of a particular segment of the sample to respond to it. The use of SMART and MOST methods can be an efficient way to rapidly build the most potent interventions for a wide range of people, including those with disabilities participating in mainstream diabetes research.

Conclusion

People with disabilities are an increasingly large segment of the population, and it is important that they are appropriately represented in diabetes research. Inclusion of persons with disabilities in diabetes research will reduce the bias in our study samples and increase the population to whom we can generalize the findings, thus strengthening our evidence base. Given the disparities in health outcomes for persons with disabilities, accelerating the rate of discovery of knowledge regarding interventions that are efficacious for this population is imperative.

Most of the reasons for lack of inclusion of persons with disabilities in research arise from misconceptions and a lack of knowledge about how to create access to research and research methods for persons with varying levels of ability and how to design study protocols that are robust in maintaining study internal validity when adaptations in protocols are needed to accommodate a wide range

of people. It is recommended that researchers use the guidelines for UDR and intervention optimizations study designs as strategies to promote the inclusion of persons with disabilities in mainstream research. These new research approaches should diminish the concerns of scientists regarding the trade-off between internal and external validity when including persons with and without disabilities in research.

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