

## Commentary

# Determining Economic Factors That Matter to People With Intellectual and Developmental Disabilities and Their Caregivers: A Process Framework

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**Background:** The 2020–2029 strategic plan for the Patient-Centered Outcomes Research Trust Fund calls for addressing data infrastructure gaps that are critical for studying issues around intellectual and developmental disabilities (I/DD). Specifically, the plan calls for data collection on economic factors that affect person-centered approaches to health care decision-making. Among people with I/DD and their caregivers, such economic factors may include financial costs of care, decreased opportunities for leisure and recreation, income losses associated with caregiving, and foregone opportunities for skill acquisition or other human capital investments.

**Objective:** This commentary supports responsiveness to the Patient-Centered Outcomes Research Trust Fund (PCORTF) calls by conceptualizing and operationalizing a framework for identifying preferences on economic factors that are relevant to people with I/DD and their caregivers.

**Main Arguments:** The framework outlined in this commentary addresses barriers to data collection that hinder measure development in the study of I/DD. This work is significant and timely given the continued movement to integrate and maintain people with I/DD within communities and recent methodological advances for eliciting preferences among people with I/DD.

**Relevance to the Special Issue:** Readers will be introduced to a framework for building data capacity in the study of economic outcomes among a population that is a high research priority for federal funding agencies. This commentary aims to be useful to researchers in planning, developing, and initiating projects in this area.

**Key Words:** economic outcomes, independent living, intellectual disability, person-centered care

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In their 2020–2029 strategic plan, the US Department of Health and Human Services, Patient-Centered Outcomes Research Trust Fund (PCORTF) calls for building data capacity to inform person-centered approaches to health care decision-making and for closing data infrastructure gaps that affect populations with intellectual and developmental disabilities (I/DD).<sup>1</sup> Aligned with the PCORTF strategic plan and calls in this *Medical Care* special issue, this commentary notes that developing the data infrastructure to enhance the exploration of economic factors in health care decision-making is vitally important. While relevant to all consumers, such efforts are essential for people with I/DD. In the United States, costs associated with medical and nonmedical supports are increasingly borne by people with I/DD, their families, and caregivers.<sup>2</sup> Examples of economic factors relevant to health care decision-making include financial costs (eg, those associated with transportation, housing, and food), reduced opportunities for recreation and leisure, income losses associated with caregiving, and diminished opportunities for skill acquisition or other human capital investments. In support of the PCORTF call, the following commentary offers a process framework for identifying relevant economic factors that affect health care decision-making regarding people with I/DD. The framework is organized into 7 steps that are built to address gaps, challenges, and available solutions relevant to engaging in this work.

### IDENTIFYING BARRIERS TO COLLECTING DATA RELEVANT TO PEOPLE WITH INTELLECTUAL AND DEVELOPMENTAL DISABILITIES

Conceptual, communicative, social, and behavioral challenges are often present among people with I/DD,<sup>3</sup> which

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may foster a misperception that people with I/DD are unable to collaborate effectively in health care research. This is evidenced by gaps in data and in data collection efforts. In a systematic review from 2014, an analysis of 300 randomly selected randomized control and clinical trials, reported in 6 highly ranked medical journals, found that only 2% of studies included people with intellectual disabilities. However, modest adjustments, such as simplified consent and assent processes or allowing assistance from support professionals, friends, or family, could have facilitated their participation in as much as 70% of the studies.<sup>4</sup> One strategy that has emerged to address these gaps in research about people with I/DD utilizes proxy reports from caregivers where caregivers speak for people with I/DD.<sup>5</sup> While caregivers, including family members and direct support professionals, are often essential to adequate provision of services,<sup>6,7</sup> exclusive reliance on such proxy reporting may violate principles of person-centeredness if proxy reports do not fully reflect the preferences of people with I/DD.<sup>5,8</sup> By utilizing appropriate context and methods (some of which is detailed, later, in this commentary), researchers can go beyond exclusive reliance on proxy reporting by using strategies that facilitate research partnerships that includes people with I/DD.

### **BUILDING ON MOVEMENTS TOWARD INCLUSION OF PEOPLE WITH INTELLECTUAL AND DEVELOPMENTAL DISABILITIES AND THE ROLE OF CAREGIVERS**

In addition to typically being excluded from health care research, people with I/DD carry a history of disparate treatment and restricted access to the community.<sup>3,9</sup> This has been well-documented in reports of maltreatment in large congregant settings such as educational facilities, jails and prisons, residential centers, and other institutions.<sup>9</sup> In a movement towards community integration and participation, public policy efforts in the United States have facilitated the downsizing and closing of many large congregate institutions traditionally used to house and treat people with I/DD (ie, deinstitutionalization).<sup>2</sup> Anchored in the Americans with Disabilities Act of 1990, the US Supreme Court affirmed the right to community living for people with disabilities in *Olmstead v. L.C.* (1999).<sup>10</sup> Legislative efforts contained within the Individuals with Disabilities Education Act (IDEA) and the Developmental Disability Assistance and Bill of Rights Act of 2000 have emphasized the need for less restrictive solutions for people with I/DD and have advanced efforts toward inclusion.<sup>11,12</sup> Taken together, these policy developments have supported a movement toward increased inclusion of people with I/DD in community life.

These movements have led to drastic demographic and living arrangement changes pertaining to people with I/DD. In the United States from 1998 to 2018, the percentage of people with I/DD using long-term supports and services and living with a family member, foster parent, or within their own home increased from 65% to 81%, while the number of people with I/DD who received long-term supports and services doubled.<sup>2</sup> With these changes, the perspectives of people with I/DD and their caregivers regarding the allocation

of scarce resources, such as time and money, has grown in importance. Decisions that were previously the responsibility of an institution (such as housing, meals, leisure time, and transportation) have, in many cases, shifted to people with I/DD themselves, family members, state agencies, providers, and community partners, among others. In response, the federal government has devoted additional funding to support community placements, such as incentivizing waivers to state Medicaid plans that include the provision of additional services, such as case management, caregiver respite care, skill development, private duty nursing, and other “Home and Community Based Services.”<sup>13</sup> To study the impact of these policy changes and to facilitate person-centered approaches to health care decision-making, there is a critical need for measures of economic factors that matter to people with I/DD and their caregivers.

### **UTILIZING AVAILABLE METHODS FOR ELICITING PREFERENCES FROM PEOPLE WITH INTELLECTUAL AND DEVELOPMENTAL DISABILITIES**

Given the importance of understanding the economic factors that influence health care decision-making, researchers interested in partnering alongside people with I/DD should look to adapt preference elicitation methods that can accommodate the practical challenges of engaging this population. The following highlights a few strategies to that end. Hollomotz<sup>14</sup> suggests that challenges to partnering in research with people with intellectual disabilities can be overcome by training researchers to utilize tailored communication strategies, such as developing visual vignettes to describe scenarios and through structuring questions directly (ie, without abstractions). This work can be informed by working with psychologists, allied health professionals (such as speech-language pathologists), and educators who have expertise in evoking and appreciating communication by people with I/DD.

Another useful strategy is called “dyadic interviewing,” which accommodates the role of caregivers in the lives of people with I/DD. Dyadic interviewing is a qualitative research method that is commonly used in dementia research.<sup>15</sup> In this method, an individual with I/DD and their primary caregiver form a “dyad” which works iteratively with a researcher who solicits input from the person with I/DD individually and in collaboration with the caregiver.<sup>16</sup> This may include separate and combined interviews with the dyad members to elicit rich qualitative data that reflects the experiences of the people with I/DD, the experiences of caregivers, and the shared experiences of both.

Dyadic interviewing and adapted interview processes, as described above, can then be combined with a quantitative method for eliciting preferences, such as Best-Worst Scaling (BWS). BWS, a survey-based approach, has grown in popularity among health economists as an effective and efficient way for participants to demonstrate their preferences among sets of options (ie, economic choice sets).<sup>17</sup> In BWS, a choice set often contains a list of potential economic outcomes. The simplest form of BWS is the object case, where each

CHOICES	BEST (Select One)	WORST (Select One)
Missing <b>4 hours</b> of time when I wasn't working and could be with my friends and family.	○	○
Traveling <b>60 minutes</b> to see the doctor.	○	○
Missing <b>4 hours</b> from my job.	○	○
Having to get <b>2 new</b> doctors.	○	○

**FIGURE 1.** Example of a Best-Worst Scaling “Choice Set” (Survey Item). Presents a set of economic outcomes for eliciting preferences on economic factors relevant to people with intellectual and developmental disabilities and caregivers.

choice set typically contains <5 economic outcomes (see Fig. 1 which has 4 outcomes in the “choice set”). The respondent identifies the “worst” outcome among the set along with the “best” outcome (Fig. 1).

A series of different choice sets are shuffled through for each respondent. Researchers can then analyze the responses from all of the survey participants across the varied combinations of choice sets to infer, among a specified population, a ranking of the outcomes in a way that suggests preferences. The BWS object case is a good candidate for eliciting preferences from people with I/DD. It places lower cognitive demands on respondents because a limited number of options are presented in each choice set and rankings can be inferred by the researcher without respondents needing to see every combination of possible choices.<sup>17</sup> This version of BWS can thus ensure that respondent cognition constraints or other contextual factors do not bias results.<sup>17</sup> The BWS object case also allows for a simplified analytic approach and interpretation (compared with more analytically complex preference elicitation methods, such as traditional discrete choice experiments),<sup>18</sup> which may be useful in communicating and disseminating findings to stakeholders, including people with I/DD.<sup>19,20</sup>

### A PROCESS FRAMEWORK FOR DETERMINING ECONOMIC FACTORS THAT MATTER

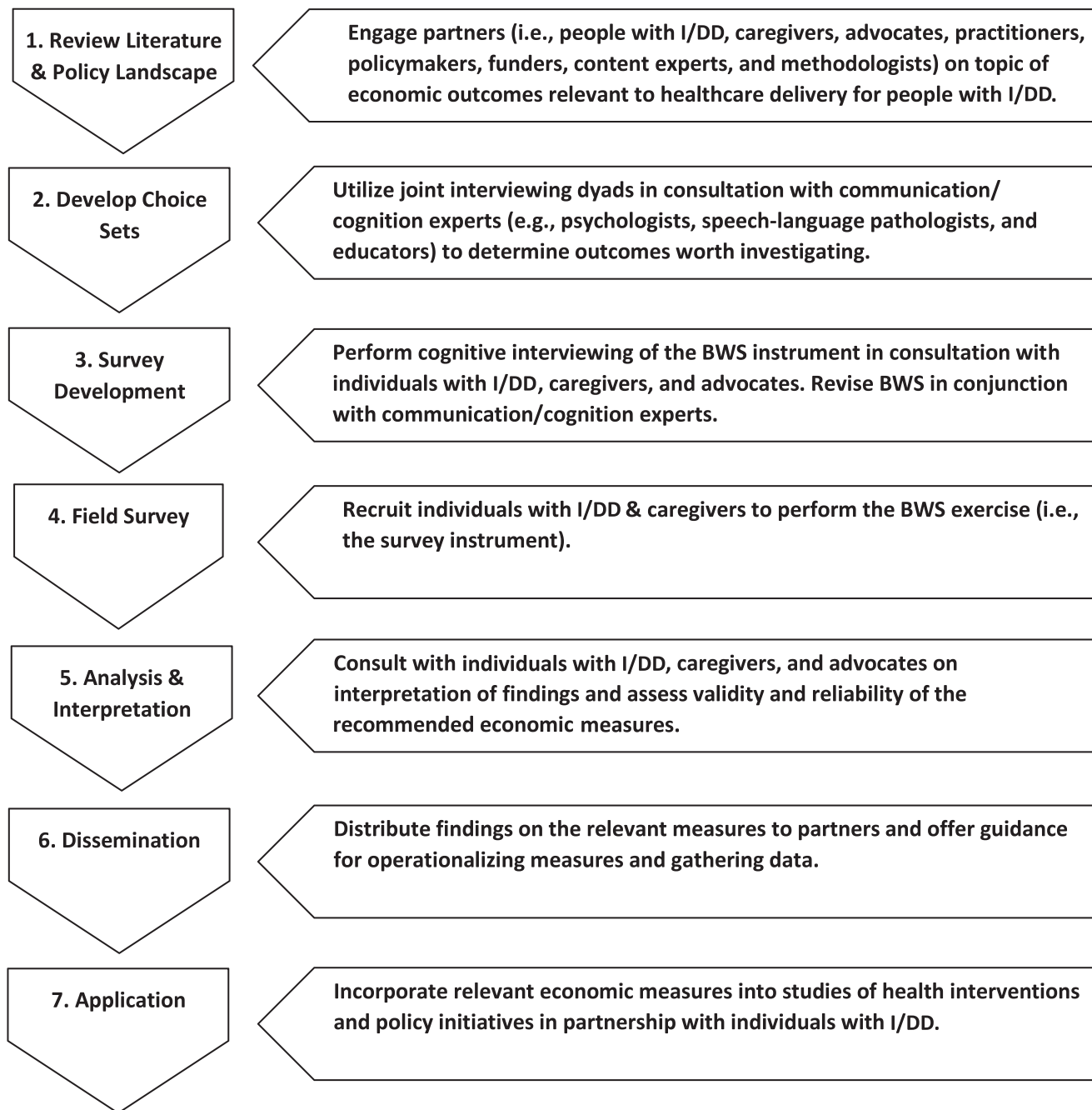
Given these gaps, barriers, and potential solutions, the following provides a framework to determining economic factors relevant to health care decision-making affecting people with I/DD and their caregivers. The 7 steps of the framework offer a strategy for integrating the modified methods in the previous section (ie, adapted interview procedures and dyadic interviewing) into a BWS instrument. Illustrated in Figure 2, the framework includes considerations from the formation of the project to the dissemination of findings and ultimately, including the

identified economic factors in studies of health care and policy initiatives.

In step 1, researchers review the relevant literature and policy mechanisms. The relevance of economic factors will likely differ across subpopulations of people with I/DD and could depend on household income, age of the respondent, social norms, clinical conditions, and service needs, among other factors. In this crucial first step, stakeholders, including people with I/DD, caregivers, practitioners, and content experts, should be engaged for advisement purposes. It is important to recruit these stakeholders as project advisors because they will be able to provide important input throughout the research process, including contributions such as assessing study procedures for person-centeredness, reviewing preliminary findings for face validity, and providing access to networks for disseminating findings. For step 2, researchers apply modified interview methods (eg, tailored communication strategies and dyadic interviewing) in consultation with the relevant population and research questions identified in step 1. The goal in step 2 is to identify relevant domains and potential economic outcomes for analysis and inform the subsequent development of the BWS survey instrument. Within the third step, researchers would utilize the information from the reviews (step 1) and interviews (step 2) to draft the BWS questionnaire and then assess it for usability and validity by reviewing it with people with I/DD and caregivers through a survey assessment strategy known as “cognitive interviewing.”<sup>21</sup>

Step 4 includes fielding the survey among people who belong to the relevant study population and meet eligibility criteria given the specific research question of the project. In step 5, results are analyzed by using regression or, alternatively, by calculating relative frequencies of specific choices to rank economic factors by their relative importance.<sup>19,20</sup> Stakeholders identified in step 1 should then be consulted throughout as findings are reviewed and interpreted. In step 6, findings can then be shared with larger stakeholder audiences. Ultimately, in step 7, findings

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**FIGURE 2.** Process framework for determining economic factors that matter. Presents 7 steps for incorporating knowledge about the gaps, problems, and potential solutions for determining relevant economic factors to health care decision-making among people with intellectual and developmental disabilities (I/DD) and caregivers. The figure also includes strategies for applying findings to health care and policy evaluative studies. BWS indicates Best-Worst Scaling.

can be included as part of studies that include economic outcomes that are relevant to people with I/DD.

**RECOGNIZING A CRITICAL MOMENT FOR ACTION**

Now is a critical time to initiate studies into economic factors that influence the health care decision-making of

people with I/DD and caregivers. To date, this has been a relatively unaddressed area. As a result, there is virtually no peer-reviewed literature on determining economic outcomes that are important to people with I/DD.<sup>22</sup> Valid and inclusive outcome measure development is essential for evaluating the impact of health care, social services, and associated policies. By adopting person-centered approaches to

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understanding factors that affect health care decision-making, researchers have the opportunity to influence and evaluate strategies to mitigate caregiver burnout, to allocate more equitable distributions of resources, and to enhance the well-being of people with I/DD.<sup>7</sup> With the current calls for research and associated opportunities to engage in this work, the above considerations are offered to researchers who are planning, developing, and initiating projects in this area.

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