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Is Safety in the Eye of the Beholder? Safeguards in Research with Adults with Intellectual Disability

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Abstract

Human subjects research has a core commitment to participant well-being. This obligation is accentuated for once exploited populations such as adults with intellectual disability. Yet we know little about the public's views on appropriate safeguards for this population. We surveyed adults with intellectual disability, family members and friends, disability service providers, researchers, and Institutional Review Board (IRB) members to compare views on safeguards. We found many points of convergence of views, particularly for decision-making and participation. One trend is that adults with intellectual disability perceive greater safety in being engaged directly in recruitment, and recruitment by specific individuals. Researchers and IRB members need to consider community views to facilitate the safe and respectful inclusion of adults with intellectual disability.

At the core of human subjects research is a commitment to securing the well-being of research participants. This obligation is accentuated for members of populations once subjected to high-harm, low-benefit research, often without their valid consent. People with intellectual disability were among those whose marginalized status was exploited for scientific gain absent concern for their well-being, and whose treatment spurred action to prevent such dynamics from re-occurring (Freedman, 2001; Goldner, Duble, Coleman, & Menikoff, 2015). Many adults with intellectual disability – a lifelong disability associated with low IQ and difficulty with everyday skills – experience less control over their lives and have less experience making decisions (American Association on Intellectual and Developmental Disabilities, 2012; Dybwad & Bersani, 1996). These characteristics affect discussions about research safeguards focused on mitigating potential risks, especially those related to valid informed consent. Although they do note they may experience diminished autonomy, federal regulations do not specify protections for adults with intellectual disability. They are, however, often included in non-binding guidance on populations with questionable consent capacity (National Institutes of Health, 2009; US Department of Health

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and Human Services, 2009). As a result, crafting strategies for securing well-being in research is largely left to the discretion of the scientific community, and beliefs about their vulnerabilities or capacities play important roles in this process.

It is not surprising that there is disagreement on appropriate safeguards. One reason is differential commitments to human rights which favor autonomy and self-determination (K. E. McDonald, Keys, & Henry, 2008; K. E. McDonald & Kidney, 2012; Northway, 2014). For example, noting that informed consent begins with recruitment (L. W. Roberts, Teddy D. Warner, Charles T. Anderson, Megan V. Smithpeter, & Melinda K. Rogers, 2004), some members of the scientific community seek to avoid undue influence on participation decisions by placing recruitment in the hands of people who provide social services to adults with intellectual disability; others counter that researchers can achieve a more balanced and effective recruitment approach and avoid the selective gatekeeping that service providers may deploy (Becker, Roberts, Morrison, & Silver, 2004; Cameron & Murphy, 2007; Cleaver, Ouellette-Kuntz, & Sakar, 2010; Iacono & Murray, 2003; K. E. McDonald, Kidney, Nelms, et al., 2009). With respect to participation decisions, some focus on providing disability accommodations to promote autonomy and valid consent, whereas others err on the side of finding (perhaps assuming) impaired decisional capacity and involving substitute decision-makers as a primary approach (Becker et al., 2004; Dalton & McVilly, 2004; K. E. McDonald, Kidney, Nelms, et al., 2009). Such divergent thinking about risk and protections is also present in research with other populations perceived to be vulnerable (Welch et al., 2015).

Despite the importance of securing participant well-being, there are concerns that an unbalanced focus on safety fosters a protectionist mindset that can threaten human rights and contribute to greater exclusion from research – a situation which can create inequitable access to research and diminish opportunities for people with intellectual disability to benefit from scientific advances (Brooker et al., 2014; Feldman, Bosett, Collet, & Burnham-Riosa, 2013; K. E. McDonald & Kidney, 2012; Northway, 2014). In practice, identifying an appropriate equilibrium between protections and autonomy is a context-specific determination, complicated for populations who have experienced and may be at risk for exploitation (Goldner et al., 2015; Welch et al., 2015). Currently, members of the scientific community largely remain in control of translating broad principles into research practices. It is possible that their perspectives on appropriate safeguards run counter to community preferences which are increasingly informed by human rights and equal opportunity (Dresser, 2014; Dybwad & Bersani, 1996; L. W. Roberts, Teddy D. Warner, et al., 2004).

Our society is at the frontiers of learning about these ideas from adults with intellectual disability, people close to adults with intellectual disability, and disability social service providers. These community stakeholders appear to value the research participation of adults with intellectual disability provided their well-being and feelings of safety are secured (Bailey et al., 2014; Heller, Pederson, & Miller, 1996; K. E. McDonald, 2012; K. E. McDonald, Schwartz, Gibbons, & Olick, 2015a). For example, adults with intellectual disability want to be treated with respect, including receiving honest communications from researchers who recognize their capacities and do not apply undue pressure (K. E. McDonald, 2012; K. E. McDonald, Kidney, & Patka, 2012; K. E. McDonald et al., 2015a).

While these preferences may seem obvious, they are not experiences people with intellectual disability can take for granted. Community members also believe that appropriate safeguards include accommodations to help adults with intellectual disability understand and communicate, opportunities to get to know researchers, and having research conducted in safe and familiar places (K. E. McDonald et al., 2015a). Close family members and friends of adults with intellectual disability and disability social service providers emphasize that researchers should possess intimate familiarity with people with intellectual disability and skills in interacting with them for research purposes. They also see their own role as including proactive steps to keep adults with intellectual disability safe, though some adults with intellectual disability worry that such protective or gatekeeping strategies may actually decrease their safety (K. E. McDonald et al., 2015a). Views toward appropriate safeguards may vary among community members based on their commitments to protecting individuals in the face of real or perceived vulnerability and harm, pushing some toward more restrictive views favoring exclusion from research, particularly research that poses greater risk, and greater limitations on control over participation decisions (Clegg, 1999; K. E. McDonald & Keys, 2008; K. E. McDonald et al., 2015a; Saaltink, MacKinnon, Owen, & Tardif-Williams, 2012).

In the context of evolving rights and opportunities for people with disabilities, scientific innovations, and an increasing emphasis on the value of direct participation of adults with intellectual disability in research, it is essential to re-examine research safeguards, and to include the views of community stakeholder members in these efforts (Dresser, 2014; Northway, 2014). Safeguards instituted in response to prior research misconduct, largely determined by the scientific community and with a protectionist mindset, may not accurately reflect the realities and preferences of citizens today. Indeed, research practices that are *experienced as safe* are essential to earning the public's trust (Dresser, 2014), thereby contributing to a dynamic which may be particularly important to increasing representation in science that can improve lives (K. E. McDonald et al., 2015a). That is, subjective evaluations of safety may be strongly related to motivation to participate in research. Although smaller scale and qualitative **studies** reveal some points of divergence among scientific and community stakeholders, the extent to which these differences may exist is unclear. To examine this question, we quantitatively compare the views of adults with intellectual disability, their close family members and friends, disability social service providers, researchers in the area of intellectual disability, and Institutional Review Board (IRB) members on safeguards in self-report research with adults with intellectual disability. We focus on *perceptions* of safety and seek to identify areas of convergence and divergence, and the influence of these views on research participation decisions. Our focus on self-report research reflects the field's increased emphasis on direct representation in such research and the challenges inherent to assessing risks in self-report research. We anticipate adults with intellectual disability will perceive strategies that promote their agency as safer, and that family members and close friends and IRB members will somewhat favor the strategies that favor safety while restricting agency.

Methods

Participants

Five hundred twelve individuals from 5 stakeholder groups participated in this study: 101 adults with intellectual disability who self-reported their disability, 98 family members and close friends of adults with intellectual disability who have provided them nonpaid support in the past year (more than 80% were family members), 109 professionals who currently provide social services to adults with intellectual disability or have in the past year, 105 researchers who conduct self-report research with adults with intellectual disability or have in the past five years, and 99 IRB members from institutions conducting research with adults with intellectual disability or those who have been members in the last 2 years. Although some individuals fit criteria for more than one group, we privileged professional group membership¹.

To participate, individuals had to be 18 years or older, reside or work in the United States, and have the communication skills to make a participation decision and express opinions with or without accommodations. With accommodations, all individuals who were interested in participating were able to provide consent or assent and answer survey questions; our inclusion criteria thus yielded participants from the population who would be able to participate in the type of research under study.

Instruments

We developed the Project ETHICS survey using a multi-prong approach to survey development (Singleton & Straits, 1999; Willis, 2005): (1) focus groups (K. McDonald & Patka, 2012; K. E. McDonald, Schwartz, Gibbons, & Olick, 2015b) (2) prior research (K. E. McDonald, 2012; K. E. McDonald et al., 2008; K. E. McDonald & Kidney, 2012; K. E. McDonald, Kidney, Kimmel, et al., 2009; Roberts, Hammond, Warner, & Lewis, 2004; Roberts, Warner, & Brody, 2000; Roberts et al., 2002; Roberts, Warner, Hammond, & Dunn, 2006; Rubright, Cary, Karlawish, & Kim, 2011), (3) an Expert Panel, and (4) cognitive interviews. We provided definitions of intellectual disability, self-report research (i.e., research about thoughts, experiences, and behavior), and researcher, encouraged participants to make their best choice even though they did not have information specific to a particular research study (e.g., benefits), and explained how to answer questions. Using two parallel versions, we asked adults with intellectual disability to respond from their personal perspective, and others to respond in reference to adults with intellectual disability. The survey also included questions on harms and benefits in research (K. E. McDonald, Conroy, Olick, & Panel, 2016, 2017), and views toward research, though we focus in this manuscript on the following data:

Safeguards—We asked participants to rate how safe adults with intellectual disability would be for each of 17 safeguards, and how likely adults with intellectual disability would be to take part in research that had each safeguard (on a scale of 1–5, higher numbers

¹There were few differences among subgroups, although professionals with family members or close friends predicted that adults with intellectual disability would be more likely to participate in research with a number of safeguards than their peers without family members or close friends with intellectual disability.

indicated greater perceived safety or likelihood of participating). The safeguards included 5 items about recruitment, 6 items about decision-making, and 6 items about research participation.

Views toward Research and Personal Information—We asked participants to indicate their level of agreement with the statement that adults with intellectual disability should be included in research (on a scale of 1–6, higher levels indicate greater agreement) (K. E. McDonald et al., 2008), how much they trust researchers (on a scale of 1–5, higher levels indicate greater trust) (Rubright et al., 2011), and to provide an array of personal demographic information.

To improve accessibility of the survey, we: (1) included graphic representations for each survey section to convey thematic content, differentiate among sections and show progress towards completion; (2) used plain language and concrete examples; and (3) provided response graphics (e.g., cylinders that were increasingly filled in as values increased, faces with an array of expressions from negative to positive) and named anchors for each extreme value for all scaled items. We also included “do not wish to say” as an option, and asked an open-ended question at the end of each section to capture additional information participants wished to share.

Procedures

We received human subjects approval from our university, and designed all materials, including the survey, with the Expert Panel, which included members from each participant group. We created national sampling frames by searching for groups and organizations by and for adults with intellectual disability, family and friends, and disability service professionals (e.g., social service organizations, advocacy groups, and social or recreational groups). We also created a sampling frame of researchers who had published self-report research with adults with intellectual disability by reviewing 7 English-language journals targeted to research with adults with intellectual disability from 2009 to 2014. We supplemented this initial list with names of researchers who had presented relevant research at one of two national conferences in 2014. We identified IRB Chairpersons through a Freedom of Information Act (FOIA) request, restricting the sampling frame to places with active intellectual disability researchers and asking Chairpersons to forward recruitment materials to their IRB members. We supplemented this list for institutions that provided publicly available IRB member rosters. We identified over 500 groups or organizations, 300 researchers, and 500 IRB members to whom we sent recruitment materials.

We recruited nationally via postal mail letters and flyers, electronic mail, social media, in-person flyer distribution, tablings, and presentations, using a slightly different approach for each participant group. Participants recruited via electronic mail received a pre-notification about the survey, an invitation to participate, a reminder, and a notification that the survey would soon be closing for their participant group (Dillman, Smyth, & Christian, 2009). We collected data for 4 months, closing out participant groups as they reached 100 participants. Most participants learned of the survey directly from us via electronic mail (34.4%) or from having a recruitment email forwarded to them (29.3%). However, adults with intellectual

disability were most likely to learn about the survey from a service provider (52.5%) or family member or friend (19.8%), although 11.9% received an email directly from us. Family members and friends of adults with intellectual disability were also more likely to learn about the survey from other family members and friends (28.6%) or receiving a forwarded email (43.9%).

Participants provided consent or, for those with legal guardians, assent and completed the survey online, with pen-and-paper, in-person, or over the telephone with or without additional supports. In addition to the steps described above for survey items, we also promoted accessibility by providing illustrative graphics in all research materials (e.g., recruitment flyers and consent/assent forms), encouraging participants to take time to make their decision and to discuss their decision with a person of their choosing if they desired, allowing participants to take a break, and providing one-on-one support in-person or over the telephone as requested (e.g., reading material aloud). We emphasized the voluntary nature of participation. When we had direct contact with participants, at no time did we observe anyone demonstrating subtle cues of resistance to participation or who did not understand what they were making a decision about (National Institutes of Health, 2009). All participants received a \$40 gift card to thank them for their participation.

Analysis

We subjected all surveys to a data validation process (e.g., reasonable completion time, verifiable postal address, percent complete, internal consistency of responses, self-reported response quality), retaining only those that met all criteria. We then cleaned the data and prepared the dataset for analysis; less than 1% of data, including responses of “do not wish to say,” were missing. The data were negatively skewed, suggesting that participants perceived all items as relatively safe. For between-group ANOVAs, we examined several variables for inclusion as covariates (e.g., age, gender, education, attitude toward research with adults with intellectual disability, perceptions of trust, relationship to persons with intellectual disability, level of severity of intellectual disability among family members or populations served or researched, student researcher status, involvement in disability advocacy, general views toward research). Because none of these variables emerged as meaningfully associated with outcome variables, we did not retain any as covariates. Given the exploratory nature of the study, we used a cutoff of $p < 0.05$ and Tukey’s HSD test for post-hoc comparisons (Tabachnick & Fidell, 2001). We also conducted all analyses with and without imputed means; analyses yielded identical conclusions. We thematically coded open-ended data, using multiple coders to bolster dependability (Braun & Clarke, 2006).

Results

Sample Characteristics

The total sample included almost three-fourths females who reported an average age of 43.5 years old ($SD = 14.18$). Most participants reported being White (87.1%). Some participants reported having additional disabilities or health conditions; mental health conditions, mobility or physical disabilities and ongoing health conditions were the most prevalent across all groups. Most participants indicated having employment, though fewer adults with

intellectual disability (52.5%) and family and friends (65.3%) did. Over half of participants reported involvement in disability advocacy or rights (56.6%). Participants came from 46 states and the District of Columbia; no state accounted for more than 7% of the total sample.

Nearly one-third (32.7%) of adults with intellectual disability lived alone, over one-fourth (26.7%) lived with a parent or stepparent or another adult family member, 13.9% lived with a partner, 13.9% lived in assisted living or group home, and 11.9% lived with a friend or roommate. Three quarters (74.3%) of adults with intellectual disability had been approved for developmental disability services, and 85.1% received Social Security. About half (46.5%) of adults with intellectual disability reported receiving support taking the survey².

Among family members, about 77% had an immediate family member and 28% had an extended family member with an intellectual disability for whom they provide support. Family members and friends largely believed that the person they support could have completed the survey on their own or with support, though nearly one-quarter did not feel the person could participate. Family members, friends, and service providers indicated supporting people with mild, moderate, and severe intellectual disability; researchers reported conducting research overwhelmingly with individuals with mild and moderate intellectual disability, though just less than one-half also did so with individuals with severe intellectual disability.

Service providers primarily served as direct support or care providers (24.8%) or agency managers or administrators (20.2%), and researchers and IRB members were primarily college or university professors (52%). Among active human subjects researchers and IRB members, 84.9% conducted behavioral or social science research and 17.3% conducted biomedical research. Intellectual disability researchers had been conducting research for an average of 10.19 years ($SD = 9.35$) and conducted an average of 10 studies ($SD = 23.27$). IRB members included voting members (47.5% scientist; 23.2% non-scientist), chairs or other leaders (33.3%), and IRB administrators (5.1%), and have served an average of 8 years ($SD = 5.65$). The majority (60.6%) of IRB members had reviewed applications that included adults with intellectual disability as research participants, and 68.7% have worked or volunteered with adults with intellectual disability. See Table 1 for additional sample characteristics.

Perceptions of Safety

Between Group Comparisons—We performed a one-way between groups ANOVA for each safeguard and found group statistically significant differences in perceptions of safety for 7 of the 17 safeguards (see Table 2). We found group differences for all 5 recruitment-related safeguards: adults with intellectual disability perceived *recruitment by service providers* and *recruitment by researchers* as safer than all other stakeholders groups. Adults

²Compared to those who did receive support to complete the survey, adults with intellectual disability who did not report using support rated the following as safer: researchers ensure understanding of study, give time to think, ask about accommodations, and use respectful language when sharing findings. It is possible that adults with intellectual disability who take surveys without support perceive greater safety in practices that promote their understanding and feelings of respect, though additional research would need to be conducted to further explore the reliability and validity of such findings and their meaning. The only demographic variable that differentiated use or support was education level: those with less education more often used support to complete the survey.

with intellectual disability perceived *recruitment by people with disabilities* as safer than IRB members, and *recruitment by someone they are comfortable with* as safer than all groups except service providers, though service providers did rate the safeguard as safer than IRB members. Overall, adults with intellectual disability rated a variety of recruitment strategies as safe, and safer than other groups perceived the strategies to be.

We found group differences for 1 decision-making safeguard: service providers rated *researchers giving adults with intellectual disability time to think about their participation decision* as safer than family, friends, and IRB members. We found group differences on 2 research participation safeguards: service providers rated *having the choice to have someone they are comfortable with at the study* and *researchers learning how to interact with adults with intellectual disability* as safer than IRB members.

Within Group Comparisons—We also performed a within group ANOVA to determine statistically significant differences in perceptions of safety within participant groups (See Table 3). Generally, adults with intellectual disability perceive being directly involved in recruitment and making their own decisions to be among the safest safeguards. All groups perceived *researchers talking to others before the adult with intellectual disability* as significantly less safe than all other decision-making and research participation safeguards, and adults with intellectual disability perceived it as less safe than the recruitment safeguards. Adults with intellectual disability also perceived *being recruited by someone they are comfortable with* as safer than *being recruited by a family member* or *people with disabilities*, and *having someone they are comfortable with help make their participation decision*. They also perceived *researchers ensuring they are making their own decision* and *reminding them the decision is their choice* as safer than being recruited by people with disabilities. And lastly, they rated *having researchers learn how to interact with adults with intellectual disability* as safer than being recruited by people with disabilities and *having someone they are comfortable with help make their participation decision*.

Family members and friends and service providers perceived *being recruited by researchers* or *people with disabilities* as the least safe means of recruitment, and as the second least safe safeguard. Family members and friends also rated *having someone the person is comfortable with at the study* as safer than most recruitment-related safeguards. Service providers perceived *having researchers ensure the adult with intellectual disability understands the study* and *giving time for them to consider their participation decision* as safer than *having family and friends* or *service providers recruit*. Researchers and IRB members generally perceived recruitment-related safeguards as less safe than decision-making and research participation-related safeguards.

Predicted Likelihood to Participate in Research with each Safeguard

Between Group Comparisons—We performed a one-way between groups ANOVA to identify statistically significant group differences in views about the influence of safeguards on the predicted likelihood of research participation among adults with intellectual disability (see Table 4). Overall, adults with intellectual disability indicated that they would be more likely to participate in research than others thought they would be, particularly IRB

members. We found group differences for 4 recruitment-related safeguards: adults with intellectual disability reported a greater likelihood of participating in research when *recruited by service providers* or *researchers* than all other groups predicted they would, and they were more likely to participate in research when *recruited by others with disabilities* than IRB members thought they would. Family members predicted a greater likelihood of participation if adults with intellectual disability were *recruited by a family member* than IRB members predicted. We also found group differences for 2 research participation safeguards. Adults with intellectual disability indicated greater likelihood of research participation if *researchers keep their personal information private* than IRB members thought they would, and service providers predicted a greater likelihood of research participation than did IRB members. Service providers predicted greater likelihood of research participation if *researchers used respectful language when sharing the findings* than did IRB members. We did not find any group differences for decision-making safeguards.

Within Group Comparisons—We also performed a one-way within group ANOVA to identify statistically significant differences in predicted likelihood of participation among adults with intellectual disability. Adults with intellectual disability indicated some differences in their likelihood to participate based on the safeguard, $F(16, 78) = 5.840$, partial $\eta^2 = .545$. They were more likely to participate in research when *researchers learn how to interact with adults with intellectual disability* or they *were recruited by a service provider* than if they were *recruited by a family member*, and less likely to participate in research where *others are talked to before them* as compared with all other safeguards.

Relationships between Perceptions of Safety and Likelihood to Participate in Research with each Safeguard

We examined the relationship between perceptions of safety and the likelihood of adults with intellectual disability participating in research with each safeguard (see Table 5). Perceptions of safety were positively and moderately to strongly correlated with greater likelihood of participating for all but 1 safeguard ($r = .390$ to $.674$, $p < .01$): The exception was *if recruited by service providers*. After adding trust of researchers, service providers, and family members as control variables for the safeguards related to each respective group, the above relationships held, although the strength of significant relationships decreased ($r = .367$ to $.625$, $p < .01$).

Open-ended Comments

We identified common themes from the open-ended question on safety in research with adults with intellectual disability. In considering issues of safety in research with adults with intellectual disability, disability service providers, family members, researchers and IRB members noted the importance of recognizing the diversity of people with intellectual disability and the heterogeneity of intellectual disability as these factors may influence views on safety, and that issues of safety in research are similar for all populations:

Every adult with intellectual disability is not the same. What one may feel safe with another may not. - Family member

SOME individuals w[ith] disabilities have the skills to have all these feelings and concerns. Some don't... If the clients I serve are non-linguistic I will have a very different point of view than someone who serves high functioning clients. - IRB member

Related, service providers, researchers, and IRB members noted that contextual factors, including the nature of relationships and the aims, methods and risk level of a research study, may modify views on safety and willingness to participate:

Just because someone is a case manager, etc., does not mean trust is automatically given between a person with an intellectual disability and said person. It needs to be someone they feel comfortable with. - Service provider

I typically do not think 'telling' someone about research is unsafe, but again it depends on the context. It can be unsafe if they are told about it in a coercing manner -emphasizing incentive, manipulating, etc. - Researcher

'Safe' depends on the nature of the research, and the process of recruitment must acknowledge the fine line between recruitment and encouragement and coercion. The same things that can make a potential subject feel safe (hearing about a study from family or friends) can easily turn coercive, especially for adults with intellectual disabilities. It's a difficult line. - IRB member

Adults with intellectual disability, family and friends, service providers, and researchers also talked about the importance that researchers have training and experience to facilitate their knowledge of people with intellectual disability. They also emphasized the value of IRB members having some understanding of people with intellectual disability to help promote their inclusion in research:

I think it's very important that people know about disabilities. - Adult with intellectual disability

Give opinions and choices. Know how to work with people and build trust. Those are important skills that will make people want to be a part of a research project. - Adult with intellectual disability

I would hope that people doing research with people who have disabilities have long time experience with people with disabilities, not newly "trained" how to interact. - Service provider

IRBs must be given training so they will not underestimate or exclude people with intellectual disability, and not infantilize or over-protect them simply because they may have a lower IQ. - Researcher

All groups talked about the importance of showing respect towards adults with intellectual disability and providing individualized accommodations to promote their inclusion, especially strategies that may promote their understanding and reduce the potential for undue influence:

To learn to make sure that you can include all people that have ID/DD from with[in] any research study with extra support and or if they need to have an

assistant person to help make sure the[y're] understanding. - Adult with intellectual disability

I think people with intellectual disabilities need a variety of supports and explanation[s] ... The supports should be individualized to the person's needs and desires. - Family member

Ensuring individuals understand that it is their choice and they can stop at any time really fosters an environment in which people choosing to participate are safe. Risk is part of any research study- even low risk studies- but if people know they can st[o]p at any time, they are more likely to be protected. – Researcher

In some cases, family and friends, service providers, and researchers suggested additional safeguards for research with adults with intellectual disability. These included planning for transportation, assuring privacy of data, incorporating enjoyable activities, providing multiple resources related to reporting neglect and/or abuse, greater involvement of support providers in supervising the adult with intellectual disability or assessing their ability to participate, providing detailed information on what participation involves, acknowledging the person's contribution, and allowing to discontinue participation.

Discussion

To address the persistent disparities experienced by adults with intellectual disability and promote their right to self-determination, we must identify ways to safely and respectfully include them directly in research (Iacono & Carling-Jenkins, 2012; K. E. McDonald & Raymaker, 2013). One way to achieve this goal is to improve our ability to provide research protections that are responsive to contemporary community viewpoints, and help participants feel safe in the choice to participate (Dresser, 2014). We compared views among five stakeholder groups on safeguards in self-report research with adults with intellectual disability. We found an array of convergence and divergence of views on recruitment, decision-making and research participation safeguards, and strong evidence that adults with intellectual disability want to be included in scientific pursuits. With only one exception, most stakeholders indicated that the focal safeguards were largely safe. However, stakeholders evidenced notable divergence from one another on the strength of recruitment-related safeguards, which is where the consent process begins. Adults with intellectual disability indicated that learning about a study from someone they trust, service providers, people with disabilities, or researchers was safer than several other groups perceived these recruitment strategies to be. Family members and friends saw recruitment by family members as safer than IRB members. IRB members perceived all recruitment strategies as less safe. In many, although not all, ways these trends resonate with findings from research with other groups who may experience similar dynamics (Edlinger et al., 2010; Karlawish et al., 2008; S. Kim et al., 2009; S. Y. Kim, Kim, McCallum, & Tariot, 2005; S. Y. H. Kim, Cox, & Caine, 2002; L. W. Roberts, K. A. Hammond, et al., 2004; Roberts & Kim, 2014; Laura Weiss Roberts, Teddy D Warner, Charles T Anderson, Megan V Smithpeter, & Melinda K Rogers, 2004; Wendler, Martinez, Fairclough, Sunderland, & Emanuel, 2002).

Conversely, stakeholder groups largely agreed on how safe decision-making safeguards were. Service providers perceive researchers giving adults with intellectual disability time to think about their participation decision as safer than family, friends, and IRB members. There was similar strong support for the perceived safety of participation-related safeguards, with the only exceptions being that service providers perceived giving adults with intellectual disability the choice to have someone they trust with them at the study and researchers learning how to interact with adults with intellectual disability as safer than IRB members. Across all safeguards, adults with intellectual disability indicated greatest perceived safety in being recruited by someone they trust, and having researchers ensure they are making their own decision, remind them that the decision is their choice, and learn how to interact with people with intellectual disability.

In examining predicted likelihood to participate in research, adults with intellectual disability indicated greater interest in taking part in research when recruited by service providers, people with disabilities, or researchers, and when researchers keep their information private and learn how to interact with adults with intellectual disability. These views among adults with intellectual disability were stronger than other stakeholders, particularly IRB members, predicted they would be. For all other safeguards, stakeholder predictions of the interest of adults with intellectual disability in participating in research mirrored those of adults with intellectual disability. There were relatively few differences among adults with intellectual disability in their interest in participating, though they were least likely to indicate interest in participating when others are talked to before them. Not surprisingly, views of safety among adults with intellectual disability were positively and moderately to strongly related to their indicated interest in participating. In open-ended comments, participants noted the importance of additional contextual features such as the diversity of people with disability in terms of their disability and individual preferences and the nature of the study, and emphasized the importance that researchers be skilled in working with people with intellectual disability to promote respectful, accessible interactions.

Although this study is highly innovative, the first of its kind to quantitatively examine these topics and compare findings for five stakeholder groups, there are limitations. First, we focus on self-report research and asked participants to make judgments absent additional information about the study's context and potential participants (e.g., purpose, procedures, benefits); other types of research and additional contextual information may alter views. It is also possible that despite the information provided, some respondents did not correctly identify the focal population (i.e., adults with intellectual disability). Finally, we only studied some safeguards and perceptions of safety; while perceptions are important to feelings of safety, they may not capture all dimensions of safety nor fully account for factors that impact willingness to participate in research.

Best Practices

To bring research policy and practice in line with community views, it is essential to reflect on the implications of this work for how we secure the well-being of adults with intellectual disability in self-report research. This may be especially important when resource limitations

force researchers to prioritize strategies. Our findings highlight the importance of *feelings* of safety in participation decisions, suggesting that safeguards may impact representation in research (Brooker et al., 2014; Feldman et al., 2013; Iacono & Carling-Jenkins, 2012; Northway, 2014).

First, we likely need to adjust recruitment strategies given that adults with intellectual disability emphasize that some approaches are safer than others, and their views of what is safe differs the most from those of the scientific community. There is controversy over which recruitment approaches are the least potentially coercive and most effective (Becker et al., 2004; Cleaver et al., 2010; Iacono & Murray, 2003; K. E. McDonald et al., 2015a). In this study, adults with intellectual disability indicate they may feel largely safe with any method, though they feel safest when recruited by someone they are comfortable with, service providers, and researchers, and are more likely to take part in research when recruited in these ways. Given that valid consent commences with recruitment, researchers and IRB members should be responsive to these views and set conditions for research that enable adults with intellectual disability to feel safe and respected. For example, although some ethics review boards have required researchers to recruit via others (Iacono & Murray, 2003), adults with intellectual disability may prefer that researchers come to their spaces and events (e.g., self-advocacy meetings or recreation activities). They may also prefer researchers inform them directly about opportunities to be in research, perhaps working in collaboration with service providers.

Although researchers and IRB members are concerned about the risk of undue influence (Iacono & Murray, 2003), adults with intellectual disability may be suggesting that concern is misplaced, especially during recruitment. Furthermore, they have reported elsewhere confidence in their ability to decline research participation (K. E. McDonald et al., 2012). Since undue influence can be further addressed during informed consent processes, favoring feelings of safety during recruitment may be help promote participation without compromising voluntariness. However, it is unclear whether adults with intellectual disability perceive feeling safe and feeling pressured as unrelated. Nonetheless, findings suggest recruitment via family members may foster greater opportunity for undue influence or perceived violations of privacy, and point researchers to control recruitment themselves and to work via trusted networks, which may include disability service providers. Researchers may find it effective to enlist disability service providers as allies in the recruitment process, providing them with concrete information about the study so that they can offer necessary and appropriate information about a study to adults with intellectual disability. Situations in which researchers cannot interact directly, at least initially, with adults with intellectual disability (perhaps because of logistics such as distance or organizational limits on sharing client names) may be well-suited to such approaches.

It is possible that the decreased sense of safety adults with intellectual disability feel when someone they are comfortable with helps them make the participation decision points to the place where we ought to be more concerned about the potential for undue influence: Whether it is concern for privacy or the desire to have unwavering control over their decision, we should think carefully about how we proceed in such situations. Advances in human rights based models such as supported decision-making might be worth exploring

(Kohn & Blumenthal, 2014), with the preferences of adults with intellectual disability being solicited and followed absent good reasons to the contrary. This emphasis may be especially important for people with legal guardians, particularly when the adult with intellectual disability desires participation and the legal guardian does not. Second, there is broad agreement that talking to others such as family members or service providers before talking to adults with intellectual disability themselves about their research participation is not a strong safeguard. Although some perceive this as promoting safety, it is likely more widely experienced as paternalistic gatekeeping and should be avoided.

Third, our findings also highlight the importance of researchers maintaining promises of confidentiality. Although doing so can be complicated when researchers interface with individuals providing support to adults with intellectual disability, the presence of a disability label does not diminish the importance of this practice. Researchers must be thus cognizant that discussing participants' data with support providers would violate commitments to confidentiality. IRBs need to understand this dynamic, and may want to seek reassurances that this protection is part of all research protocols, and that any departures from maintaining confidentiality have an appropriate rationale and will be disclosed to participants. In cases where individuals providing support to adults with intellectual disability have access to research data (e.g., when they are helping with communication or understanding during data collection), researchers may want to pursue formal agreements that the support provider will not disclose the information or use it in any way without the permission of the person with intellectual disability. It is possible that these types of potential violations of confidentiality are somewhat unique to adults with intellectual disability who may make use of personal support as an accommodation.

Fourth, our findings also underscore the importance that researchers not only have scientific expertise, but are equally familiar with and skilled in interacting with adults with intellectual disability; and IRBs should specifically consider this information in their review of researcher qualifications. This may include treating the individual as an adult and not a child, and talking directly to the adult with intellectual disability, not to the person providing them support. These results also suggest that adults with intellectual disability benefit from reminders that the participation decision is their own choice. Although researchers may shy away from repeating this information feeling it is unnecessary or awkward, they should be responsive to the community preference for doing so. Collectively, the use of these strategies promotes feelings of safety which in turn foster respect and trust, and ultimately benefit the representation of adults with intellectual disability in science.

Research Agenda

Our findings suggest notable consensus for views on the safety of decision-making and participation safeguards. However, it is possible this broad agreement masks grave divergence of opinion in how precisely to pursue practices such as researchers ensuring the participant understands the study and is making their own participation decision. In fact, we know that although members of the scientific community value these broader concepts, they depart in how to carry them out (K. E. McDonald & Kidney, 2012). We suspect adults with intellectual disability do not support practices that they perceive to be testing, as opposed to

supporting, their understanding and voluntariness, as these may feel childlike and discriminatory. They might support, however, the use of varied, individualized accommodations, supports, and strategies that promote both their understanding and their agency (Heller et al., 1996). Future research can help unpack these possibilities, finding a better balance between safety and respect, and strengthening ethically strong science with adults with intellectual disability. Stakeholder agreement on the importance of the broader concept embodied in the safeguard may be leveraged to build consensus; and these efforts must significantly weigh the preferences of adults with intellectual disability.

Educational Implications

Our findings serve as reminders of the importance of effective training and socialization mechanisms so that researchers and IRB members have the knowledge, skill and attitudes to develop and apply research safeguards that are experienced as safe and respectful by adults with intellectual disability (Lai, Elliott, & Ouellette-Kuntz, 2006; K. E. McDonald et al., 2008). Graduate courses, research apprenticeships, human subjects training, and expectations for transparency in the ethical aspects of our science in grant proposals and dissemination activities can all serve this goal. Moreover, these proactive approaches may mitigate potential conflicts of interests researchers may experience to apply undue influence as they deepen their commitments to acting with integrity, and their knowledge of how to do so. Such strategies may also lessen opportunities for misplaced trust as researchers conduct themselves in community-supported manners. It may be most difficult for IRBs to reflect this capacity; partnerships with experts and diverse membership can help IRBs use informed, valid knowledge in their deliberations. It is also important to help family members, close friends, and disability service providers understand these preferences and the value of particular research practices given their roles in connecting and supporting adults with intellectual disability (Becker et al., 2004; Iacono & Murray, 2003).

Conclusion

Society needs new knowledge to eliminate the health, social, economic, and civic disparities experienced by adults with intellectual disability. Ultimately, sensitivity to the shared humanity of and desire for human rights among adults with intellectual disability may facilitate their greater inclusion in ethical research, without exploitation for scientific gain that stained some research of the past. In pursuing this goal, developing respectful relationships and authentic collaborations with adults with intellectual disability is essential. We hope *Project ETHICS* can make valuable contributions to this work.

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Table 1

Participant demographic characteristics

Variable	Intellectual disability (n = 101)		Family/ friend (n = 98)		Service provider (n = 109)		Disability researcher (n = 105)		IRB Member (n = 99)		Total (N = 512)	
	M (SD)	%	M (SD)	%	M (SD)	%	M (SD)	%	M (SD)	%	M (SD)	%
Age	43.28 (14.66)		44.81 (14.91)		38.85 (13.41)		40.04 (12.89)		51.05 (11.72)		43.50 (14.18)	
Variable	%	%	%	%	%	%	%	%	%	%	%	%
Gender												
Female	49.5		80.6		80.7		85.7		62.6		72.1	
Male	43.6		19.4		19.3		14.3		37.4		26.6	
Other	1.0		0		0		0		0		0.2	
Race *												
Amer. Indian/Alaskan Native	14.9		1.0		1.8		1.0		1.0		3.9	
Asian	0		2.0		3.7		7.6		4.0		3.5	
Black/African American	10.9		6.1		5.5		2.9		2.0		5.5	
Hawaiian/Pacific Islander	1.0		0		0.9		0		0		0.4	
White	81.2		87.8		86.2		88.6		91.9		87.1	
Other	3.0		3.1		0.9		0.9		2.0		2.0	
Latino	2.0		6.1		3.7		2.9		5.1		3.9	
Education												
Never attended school	2.0		0		0		0		0		0.4	
Did not complete high school	21.8		1.0		0		0		0		4.5	
High school diploma/GED	50.5		6.1		5.5		0		0		12.3	
Some college	6.9		14.3		13.8		1.9		2.0		7.8	
Certificate/Associates	7.9		13.3		10.1		1.0		0		6.4	
Bachelor's	0		24.5		35.8		7.6		4.0		14.6	
Graduate degree	0		38.8		32.1		89.5		93.9		50.8	
Disability advocacy	60.4		57.1		72.5		69.5		21.2		56.8	

Note. For items with an asterisk (*), respondents were asked to select all that apply.

Table 2

One-way ANOVA between group differences in perceptions of safety

How safe is it if...	Intellectual disability (n = 101) <i>a</i>	Family/ friend (n = 98) <i>b</i>	Service provider (n = 109) <i>c</i>	Disability researcher (n = 105) <i>d</i>	IRB Member (n = 99) <i>e</i>	F(df), partial η^2
	M (SD)	M (SD)	M (SD)	M (SD)	M (SD)	
<i>Recruitment</i>						
Service provider	4.37 (1.05) ^{b-e}	4.10 (.86) ^a	4.32 (.81) ^a	3.88 (.81) ^a	3.86 (.82) ^a	F(4, 502) = 7.714, η^2 = .058
Family member	4.03 (1.35)	4.14 (.84) ^e	4.31 (.78)	3.90 (.85)	3.77 (.85) ^b	F(4, 503) = 5.051, η^2 = .039
Researcher	4.36 (.98) ^{b-e}	3.63 (.96) ^a	3.89 (.91) ^a	3.86 (.77) ^a	3.80 (.81) ^a	F(4, 504) = 9.307, η^2 = .069
People with disabilities	4.01 (1.20) ^e	3.47 (1.06)	3.94 (.88)	3.75 (.87)	3.41 (1.04) ^a	F(4, 503) = 7.008, η^2 = .053
Someone comfortable	4.64 (.78) ^{b,d,e}	4.17 (.94) ^a	4.42 (.72) ^e	4.20 (.71) ^a	3.98 (.86) ^{a,c}	F(4, 505) = 10.110, η^2 = .074
<i>Decision-making</i>						
Others are talked to first	3.03 (1.65)	3.39 (1.18)	3.50 (1.09)	3.41 (1.07)	3.45 (.99)	F(4, 502) = 2.383, η^2 = .019
Someone comfortable helps make decision	4.02 (1.26)	4.31 (.83)	4.36 (.79)	4.28 (.67)	4.21 (.77)	F(4, 507) = 2.236, η^2 = .017
Researchers ensure understanding of study	4.52 (1.00)	4.37 (.78)	4.62 (.57)	4.52 (.59)	4.48 (.64)	F(4, 505) = 1.627, η^2 = .013
Researchers ensure making own decision	4.51 (.97)	4.33 (.81)	4.56 (.60)	4.47 (.65)	4.37 (.71)	F(4, 506) = 1.686, η^2 = .013
Researchers remind that study is own choice	4.52 (1.04)	4.33 (.79)	4.58 (.67)	4.49 (.59)	4.32 (.75)	F(4, 506) = 2.309, η^2 = .018
Researchers give time to think about study	4.40 (1.05)	4.33 (.77) ^e	4.63 (.59) ^{b,e}	4.47 (.61)	4.30 (.78) ^c	F(4, 505) = 3.182, η^2 = .025
<i>Research Participation</i>						
Researchers keep personal information private	4.52 (1.04)	4.41 (.87)	4.61 (.72)	4.52 (.75)	4.38 (.84)	F(4, 506) = 1.187, η^2 = .009
Choice to have someone comfortable at study	4.46 (1.03)	4.51 (.71)	4.63 (.63) ^e	4.36 (.67)	4.33 (.67) ^c	F(4, 507) = 2.679, η^2 = .021
Researchers ask about accommodations	4.45 (1.02)	4.47 (.74)	4.60 (.58)	4.46 (.64)	4.28 (.78)	F(4, 506) = 2.202, η^2 = .017
Researchers learn how to interact with adult with intellectual disability	4.55 (.88)	4.48 (.66)	4.61 (.54) ^e	4.42 (.60)	4.33 (.64) ^c	F(4, 506) = 2.759, η^2 = .021
Researchers talk about relationship expectations	4.33 (1.02)	4.20 (.89)	4.49 (.73)	4.27 (.72)	4.24 (.76)	F(4, 504) = 1.953, η^2 = .015
Researchers use respectful language when sharing findings	4.47 (1.05)	4.44 (.72)	4.57 (.69)	4.42 (.68)	4.25 (.80)	F(4, 507) = 2.111, η^2 = .016

Table 3

One-way ANOVA within group differences in perceptions of safety

Group	F(df), partial η^2
Adults with intellectual disability	$F(16, 78) = 8.195, \eta^2 = .627$
Family members and friends	$F(16, 80) = 14.739, \eta^2 = .747$
Service providers	$F(16, 92) = 11.164, \eta^2 = .660$
Researchers	$F(16, 85) = 12.793, \eta^2 = .707$
IRB members	$F(16, 82) = 10.780, \eta^2 = .678$

Table 4

One-way ANOVA between group differences in likelihood to participate in research

	Intellectual disability (<i>n</i> = 101) <i>a</i>	Family/ friend (<i>n</i> = 98) <i>b</i>	Service provider (<i>n</i> = 109) <i>c</i>	Disability researcher (<i>n</i> = 105) <i>d</i>	IRB Member (<i>n</i> = 99) <i>e</i>	<i>F</i> (<i>df</i>), η^2
Desire of adult w/intellectual disability to participate if:	<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	<i>M</i> (<i>SD</i>)	
<i>Recruitment</i>						
Service provider	4.51 (.97) ^{<i>b-e</i>}	3.82 (.83) ^{<i>a</i>}	3.92 (.79) ^{<i>a</i>}	3.73 (.75) ^{<i>a</i>}	3.63 (.66) ^{<i>a</i>}	<i>F</i> (4, 505) = 18.627, η^2 = .129
Family member	4.05 (1.31)	4.19 (.74) ^{<i>e</i>}	4.07 (.81)	3.96 (.72)	3.80 (.64) ^{<i>b</i>}	<i>F</i> (4, 504) = 2.795, η^2 = .022
Researcher	4.45 (.95) ^{<i>b-e</i>}	3.48 (.85) ^{<i>a</i>}	3.57 (.84) ^{<i>a</i>}	3.58 (.77) ^{<i>a</i>}	3.34 (.66) ^{<i>a</i>}	<i>F</i> (4, 506) = 28.594, η^2 = .184
People with disabilities	4.22 (1.10) ^{<i>e</i>}	3.92 (.97)	4.11 (.72)	4.07 (.79)	3.87 (.69) ^{<i>a</i>}	<i>F</i> (4, 505) = 2.734, η^2 = .021
Someone comfortable						
<i>Decision-making</i>						
Others are talked to first	3.05 (1.65)	3.34 (1.04)	3.29 (1.18)	3.20 (1.06)	3.16 (.97)	<i>F</i> (4, 501) = 0.847, η^2 = .007
Someone comfortable helps make decision	4.37 (1.01)	4.44 (.66)	4.40 (.63)	4.32 (.70)	4.30 (.71)	<i>F</i> (4, 505) = 0.562, η^2 = .004
Researchers ensure understanding of study	4.51 (.96)	4.33 (.73)	4.42 (.75)	4.28 (.70)	4.28 (.70)	<i>F</i> (4, 505) = 1.069, η^2 = .013
Researchers ensure making own decision	4.56 (.86)	4.41 (.73)	4.55 (.62)	4.49 (.65)	4.35 (.67)	<i>F</i> (4, 505) = 1.624, η^2 = .013
Researchers remind that study is own choice	4.50 (.94)	4.33 (.81)	4.50 (.66)	4.31 (.70)	4.23 (.73)	<i>F</i> (4, 506) = 2.538, η^2 = .020
Researchers give time to think about study	4.48 (.94)	4.28 (.76)	4.50 (.66)	4.34 (.69)	4.22 (.72)	<i>F</i> (4, 507) = 2.570, η^2 = .020
<i>Research Participation</i>						
Researchers keep personal information private	4.55 (1.03) ^{<i>e</i>}	4.37 (.80)	4.49 (.73) ^{<i>e</i>}	4.24 (.78)	4.16 (.83) ^{<i>a,c</i>}	<i>F</i> (4, 505) = 3.882, η^2 = .030
Choice to have someone comfortable at study	4.48 (.97)	4.60 (.57)	4.63 (.56)	4.46 (.62)	4.51 (.60)	<i>F</i> (4, 507) = 1.401, η^2 = .011
Researchers ask about accommodations	4.47 (.95)	4.52 (.66)	4.48 (.63)	4.29 (.70)	4.27 (.70)	<i>F</i> (4, 505) = 2.493, η^2 = .019
Researchers learn how to interact with adult with intellectual disability	4.59 (.87)	4.49 (.66)	4.51 (.66)	4.33 (.76)	4.31 (.68)	<i>F</i> (4, 504) = 2.751, η^2 = .021
Researchers talk about relationship expectations	4.30 (.99)	4.08 (.84)	4.35 (.74)	4.13 (.72)	4.21 (.64)	<i>F</i> (4, 504) = 2.028, η^2 = .016
Researchers use respectful language when sharing findings	4.46 (1.01)	4.50 (.61)	4.60 (.56) ^{<i>e</i>}	4.41 (.68)	4.26 (.75) ^{<i>c</i>}	<i>F</i> (4, 506) = 2.858, η^2 = .022

Note. Italicized superscript indicates a significant difference with reference group (*a* = adult with intellectual disability, *b* = family/friend, *c* = disability service provider, *d* = researcher, *e* = IRB member) at the $p < .05$ level. All *F* statistics are also significant at the $p < .05$ level.

Table 5

Pearson correlation coefficients for perceptions of safety and likelihood to participate

Safeguard	Likelihood to participate
	<i>r</i>
<i>Recruitment</i>	
Service provider	.185, <i>ns</i>
Family member	.674
Researcher	.676
People with disabilities	.598
Someone comfortable	.398
<i>Decision-making</i>	
Others are talked to first	.641
Someone comfortable helps make decision	.528
Researchers ensure understanding of study	.616
Researchers ensure making own decision	.422
Researchers remind that study is own choice	.529
Researchers give time to think about study	.474
<i>Research participation</i>	
Researchers keep personal information private	.429
Choice to have someone comfortable at study	.462
Researchers ask about accommodations	.517
Researchers learn how to interact with adult with intellectual disability	.412
Researchers talk about relationship expectations	.625
Researchers use respectful language when sharing findings	.487

Note. *ns* indicates that the correlation is non-significant. All other correlations are significant at the $p < .01$ level.