Published in final edited form as:

Appl Nurs Res. 2015 May ; 28(2): 202–205. doi:10.1016/j.apnr.2014.10.002.

Implementation challenges in end-of-life research with adults with intellectual and developmental disabilities

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Abstract

Although the 4 million+ people in the U.S. with an intellectual or developmental disability (I/DD) experience close to the same life expectancy as those in the general population, end-of-life research including these individuals is lacking and can be difficult to implement. As will be described in this paper, it is possible to overcome barriers to successfully include people with I/DD in end-of-life research. In this paper, the implementation challenges, feasibility, and implications for successful end-of-life research with individuals with I/DD using focus groups are described. Individuals with I/DD were able to discuss their experiences and views about end-of-life care. However, while people with I/DD made valuable contributions to the focus groups, there were several modifications needed in order to execute this study. In order to gain a complete picture of end-of-life care for people with I/DD, it is imperative to include them in research to the best of their ability. By anticipating issues related to recruitment, the consent process, setting, and support needs of participants, focus groups can be successfully implemented.

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Keywords

Terminal Care; Intellectual Disability; Research Design

Introduction

The 4.7 million people in the U.S. who have an intellectual or developmental disability (I/DD) (Braddock, Hemp, & Rizzolo, 2009, p. 1) often experience more complex health problems (Hogg & Tuffrey-Wijne, 2008; Hogg, Juhlberg, & Lambe, 2007; Krahn, Hammond, & Turner, 2006; Strydom, Hassiotis, King, & Livingston, 2008) and require greater health services than the general population (Hogg, et al, 2007). Many adults with mild to moderate I/DD have an equivalent life expectancy (Bittles, et al., 2002) and often experience the same diseases related to aging as those in the general population (Haverman, et al., 2010). However, end-of-life research that includes the perspectives of people with I/DD remains limited (Tuffrey-Wijne, Hogg, & Curfs, 2007), with most research involving only the perspectives of staff (Todd, 2013; Wagemans, et al, 2013; Wiese, Stancliffe, Balandin, Howarth, & Dew, 2012) and is difficult to implement. People with I/DD are considered vulnerable because their capacity to consent to research may be affected by their disability, and historically, people with I/DD have been exploited in human subjects research. It is often difficult to access vulnerable populations because they are appropriately protected from exploitation through institutional regulations (Chiang, Keatinge, & Williams, 2001). Despite the potential barriers, it is imperative to include people with I/DD in end-of-life research. The objective of this article is to describe the implementation challenges, feasibility, and implications for successful end-of-life research with individuals with I/DD using a focus group method. Specifically, issues surrounding participant access and recruitment, the consent process, levels of participant protection, communication and understanding and setting will be described. Including participants with I/DD in research is critical and this study illustrates that by successfully overcoming barriers, such research is both feasible and scientifically rewarding.

Focus Group Methodology

Several scholars in the area of I/DD and EOL have advocated for the use of qualitative methodology in order to conduct research with people with I/DD, whose perspectives often are not represented (Andre-Barron, Strydom, & Hassiotis, 2008; Kaehne & O'Connell, 2010; Llewellyn & Northway, 2008; Tuffrey-Wijne, 2003). This informed, inductive approach is an appropriate first-step methodology to investigate a phenomenon with little empirical documentation (Patton, 1990). Focus groups were used for this research because this methodology has been successfully utilized in both end-of-life research (Aspinal, Hughes, Dunckley, & Addington-Hall, 2006) and in research with people with I/DD (Andre-Barron, et al, 2008; Kaehne & O'Connell, 2010) and may be preferable to surveys for individuals with literacy issues (Boyden, Esscopri, Ogi, Brennan & Kalsy-Lillico, 2009). This approach also facilitates group synergy which allows investigators to gather a comprehensive range of experiences and can yield more information than individual interviews alone (Krueger & Casey, 2000).

Implementation Issues

Participant Access and Recruitment

Accessing and recruiting people with I/DD can be challenging for a number of reasons: (1) this population has been exploited in the past and agencies and families are appropriately protective; (2) investigators must rely on agencies, often underfunded and understaffed, for providing access to and aiding with recruitment; and (3) traditional methods for access and recruitment, such as flyers or media ads, may be ineffective because of lack of access to media and literacy difficulties in this population. The research team's previous experiences with the community agencies expedited gaining access to individuals with I/DD and enabled the focus groups to be conducted at the agencies. In addition, all of the research staff had prior experience working with end-of-life research and/or training in working with people with I/DD which minimized the need for training in conducting sensitive research with a vulnerable population.

In order to minimally disrupt clients' participation in their day programs, agency staff asked that the research team not come in to meet and screen possible client participants. While this method of recruitment relies on uncompensated agency staff taking time out of their hectic schedules in order to facilitate recruitment, in many ways it provided an additional protection against coercion since staff were able to inform participants of the study and participants could feel free to decline. Agency staff did not enroll participants, but facilitated recruitment by approaching individuals that they believed met the eligibility requirements. Having agency staff approach clients reduced coercion on behalf of the client to participate and avoided acquiescence toward a new person the client may wish to please. Clients were eligible to participate if they had experienced the death of someone close to them within the last two years, but not earlier than 3 months ago, were emotionally ready to discuss their loss, and had the ability to verbally articulate their thoughts on death and dying in a group setting. The timeline of 3 months to 2 years was initially conceived to yield more current, and therefore, perhaps more reliable memories of events. Having such a restricted timeline proved extremely challenging in finding eligible participants, so after the first focus group, when participants relayed events from 15 or more years in the past, participants were included if they were able to recall events and feelings surrounding the death of their relative/friend.

Human Subject Protection Issues

The staff was explicitly asked not to consult any records or perform any assessments because this would be considered "engaging in research". According to our Institutional Review Board (IRB), when agencies are engaged in research they must obtain their own Federal Wide Assurance through the National Institutes of Health, which would have authorized them to establish their own IRB. None of the agencies had a need or desire to establish their own IRBs, so this step seemed too burdensome to them in order to help us screen research participants. It would also have required the agency staff to complete human subjects training. Provided the agency staff did not consult any client records or perform any client assessments, they could help in recruiting.

Consent Process

Major challenges in the consent process included assessing clients' capacity to consent, and assuring that the process was free of coercion and acquiescence. Prior to study approval, the IRB requested detailed descriptions of how IQ and capacity to consent would be assessed. Initial capacity was presumed if the client was his or her own guardian, and understanding was further assessed during the consent process by asking open ended questions. For clients who had guardians, the consent of the guardian was first obtained, followed by assent from the client. In addition, agency staff were asked to identify, using their working knowledge only, clients whom they believed met the inclusion criteria. In order to use vernacular familiar to the IRB, the investigators described the potential client participants as having "moderate intellectual disability". However, within the disability community the term "support needs" is preferred rather than using functional level. The broad functional category of "moderate" was used with the understanding that individuals with greater support needs would not be selected by agency staff given the requisite cognitive and communication abilities needed to participate.

The PI had a concern that the template language of the consent forms could be indelicate to the potential participants; the PI wanted to avoid the phrase "You are being invited to participate in this research because you have an intellectual disability..." The title on the consent form was "End-of-Life Care for People with Intellectual and Developmental Disabilities", but the template sentence was re-written to say "We are asking you to participate to hear your views on end-of-life care." There were also references to being a user of an agency serving people with intellectual/developmental disabilities rather than using a label of intellectual disability.

In order to minimize the potential for coercion and acquiescence, the consent process consisted of several phases. Agency staff initially approached potential participants and if the clients agreed to learn more about the study, they were given a copy of the consent form and study information sheet. In cases in which participants were not their own guardians, agency staff contacted the guardians and once consent was obtained, client participants were contacted for assent. Both the consent forms and information sheets used less technical language than the standard IRB template consent form and the information sheet additionally used pictures. According to Krueger and Casey (2000), a minimum of four participants are needed for a focus group. Thus, once there were at least four clients expressing interest in participating, a focus group was scheduled at the agency. As a second step, prior to the start of the focus group, the research team met face-to-face with each client in order to read the consent or assent form aloud and answer questions. This occurred during the first 10–15 minutes of the focus group. Once all questions were addressed and forms signed, the moderator reviewed the study, discussed the importance of not revealing to others what was said in the group, and reinforced the clients' rights to refuse to answer any questions and withdraw from the study at any time. Ideally, more time should have been allotted for the group since the consent process took at least 20 minutes.

Levels of Participant Protection

The research team built in six procedural levels of protection to ensure the focus group experience would not be distressing, and to minimize participants' distress if it occurred. First, the agency personnel were asked to select only individuals they believed would be emotionally ready to discuss end-of-life issues without acute distress. Next, participants were informed of the purpose of the focus group and the nature of the questions. Third, the research team monitored the group for signs of distress, such as tears or agitated behaviors and planned to manage distress with compassion and discretion. For example, if clients became visibly sad, their emotions would be acknowledged and the moderator would ask if they needed to take a break. Having multiple team members (PI, PD and RA) present in the focus group added a fourth level of protection, as they could co-facilitate and/or monitor the groups for signs of distress. The choice to have multiple team members present was also made in order to minimize disruption to the group if a team member needed to attend to a distressed participant. A fifth level of protection was, with the client's permission, to inform the appropriate agency personnel if someone became distressed during the session. In staying within the research role, the research team cannot provide therapy for distressed participants. Thus, it was explicitly discussed with and approved by each agency prior to recruitment that the agency staff would provide any follow up or provide any referrals to mental health resources if needed. (Of note, one agency declined to allow clients to participate in the study because of concern that participation could provoke complicated grief.) However, none of the participants exhibited distress during any of the focus groups. For the final step, the moderator would check in with everyone at the end of the focus group to provide closure and make sure everyone was okay.

Communication and Understanding

Individuals with intellectual disability may have difficulties with literacy and in understanding abstract conceptual issues, such as death. The first issue was whether or not clients could comprehend the abstract concepts associated with end of life, such as death, dying, advance care planning, and grief, and be able to express their thoughts about those concepts. The research team hired an experienced moderator who was able to probe to get information and keep clients focused on the questions at hand. Some clients were not capable of expressing their thoughts or tracking the conversation, but most were.

Despite careful planning, a few days prior to the first focus group, the research team was informed that two of the participants would need assistance with eating lunch, and there were two aides who could provide the assistance. The PI contacted the IRB with this new information, submitted an amendment that included having the aides sign a confidentiality agreement that would allow them to remain in the room during the focus group, and stressed the urgency of a response given the effort to recruit and schedule the focus group. Within two days, an expedited review of the amendment occurred and the confidentiality agreement form was approved. Anecdotally, it was fortuitous that one of the aides was in the room as she was able to tell us what the client she was assisting was saying since no one else in the room could understand the client.

The second communication issue is related to capturing the discussion verbatim. Traditionally, focus groups are audio recorded and transcribed at a later time. This method was not ideal for this study because some individuals with I/DD have articulation issues (for example, the client who no one but her aide could understand) which makes it difficult to discern what they are saying from an audio recording. To ensure clear and accurate transcripts, Computer-Assisted Remote Transcription (CART) was used. A transcriptionist (or “live writer”) from an agency providing CART was present during the focus groups. This transcriber captured 95% or more of the conversation and the cost was comparable to traditional off-site transcription. The transcriptionist and agency were in full compliance with HIPAA and removed proper nouns and identifiable data before returning the transcripts. Prior to the first focus group, the PI spoke with the transcriptionist regarding her experience with people with I/DD. Since she had none, the PI gave her some background, discussed parameters of the focus group, and alerted her to some behaviors, like slight choking or regurgitation that might occur while clients were eating. Later the transcriptionist voiced her appreciation for the “heads up” as she was prepared for the difficulties some clients had while eating, and was especially alert to articulation problems.

Setting

In order facilitate participation all of the groups were conducted within the agency setting that was both convenient and familiar to clients. In an attempt to minimally disrupt participant’s workshop activities, groups were scheduled during the lunch hour. Dietary restrictions, such as diabetes or food allergies were considered when selecting the meal, which consisted of sandwiches. Many of the individuals with I/DD were in wheelchairs, so the room had to have enough accessible space to accommodate them. Another consideration was the need to cool the room during the summer months. The available room was large and was cooled by a window air conditioner. There was concern that the voices of the participants may not be heard with the air conditioner running; in addition, each focus group was digitally recorded using two separate recorders as a quality measure to compare the accuracy of the transcripts, and the noise of the air conditioner might obliterate the recording. The research team arrived early enough to run the air conditioner, then it was turned off during the focus group meeting, and turned back on once the session ended.

Feasibility Issues

Although most participants could follow the discussion, describe their own experiences, and give their opinions, there were a few who were unable or unwilling to participate. There was one participant who frequently interrupted wanting to tell a story or talk about something else and would leave and re-enter the room a few times; one client only wanted to discuss the deceased entertainer Michael Jackson; and one other client rested her head on the table and remained silent during the session. There were a few other clients who had disruptive behaviors at times, but made substantive contributions to the discussion, and the moderator was able to maintain the flow of the session. Demographic information was collected from each client at the time the consent/assent forms were discussed, and the inability to provide information was one indicator that the client may not be able to participate. However, we did not exclude anyone once they agreed to participate, and they were given lunch and a \$30

Target gift card. For future studies, though, it may be helpful to have a way to screen clients for their ability to participate, both in terms of their conceptual abilities but also their communication. Given our IRB view that screening involves “engagement in research” of agency staff, the research team would need access to do the screening in a way that would be least disruptive to the agency activities.

For this particular study, only one agency was used to recruit clients for focus groups. Given that useful data were obtained, and the participants expressed their appreciation for the opportunity to talk about the topic of death, focus groups can be an appropriate method for end-of life research in this population. Despite one agency declining to allow their clients to participate, the research team believes it could be cathartic for the clients, if topics of death, dying, loss, and grief are not ordinarily discussed. A balance should be found between the paternalism of protecting clients and the dignity of risk in permitting clients to participate.

Conclusions and Implications for Research

In order to understand the end-of-life care experiences for people with I/DD, it is important to include them in research. The focus group method is a useful tool for learning the perspectives of people with IDD on end-of-life care and gives them an opportunity to inform research while supporting one another by sharing their experiences. Whereas focus groups do present some unique challenges when working with people with I/DD, this method is also quite powerful. It may be possible that methods used with the general population in doing advance care planning for one’s own death, such as Living Wills or Durable Power of Attorney for Health Care could be adapted for use with people with I/DD. Further research may identify best practices in EOL care. Careful attention to recruitment, the consent process, setting, and support needs is necessary for a respectful and successful focus group session.

Acknowledgments

This publication was made possible by the National Institutes of Health (NIH), National Institute of Nursing Research (NINR) P30 grant NR010680 (Drs. Diana Wilkie and Karen Kavanaugh, co-PIs). Its contents are solely the responsibility of the authors and do not necessarily represent the official views of the NINR. Thank you to Drs. Diana Wilkie, Karen Kavanaugh, and Tamar Heller for their assistance in conceptualizing and implementing the research project.

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