University of Alberta

Exploring the involvement of adults with Autism Spectrum Disorder (ASD) in developing ASD research

by

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To my friends Amira and Rahim

ABSTRACT

Participatory research (PR) is a research approach which involves partnerships between researchers and those affected by the issues under study. These partnerships lead to the production of research that is relevant and applicable to those involved in the process. Despite the potential value of this approach, few studies have involved individuals with Autism Spectrum Disorder (ASD) or other neurodevelopmental disorders in the development of research. This thesis includes two studies: a scoping review examining the involvement of individuals with ASD and other neurodevelopmental disorders in development of research, and a qualitative study conducted with adults with ASD exploring their preferred process of ASD research involvement. These studies revealed different perspectives among researchers and adults with ASD toward research involvement which need to be resolved in order to develop genuine partnerships of trust and research productivity.

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> "It is both a perplexing and an exciting new world that we enter today — and it should be supremely reassuring and inspiring to all of us that we can enter it together." -- His Highness the Aga Khan IV

I attribute my growth as a researcher to the mentorship of many exceptional individuals who challenged me to deepen my understanding of complex ideas, who reminded me when I struggled with writing to go back to my outline because clear writing is a product of clear thinking, and who were actively engaged in helping me to develop as both a scholar and a person. Collectively, these individuals made possible an environment where I could do my best work.

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CHAPTER 1

INTRODUCTION

This paper-based thesis includes two research papers; one of these has been submitted for publication, and the other is being prepared for journal submission. These papers are based on my graduate research (M.Sc. in Medical Sciences), which focused on examining the involvement of adults with Autism Spectrum Disorder (ASD) in shaping ASD research. This introductory chapter contextualizes my graduate research by providing background literature on ASD, presenting my personal research interests, and outlining each chapter's contribution to my thesis work.

Background

Participatory research (PR) refers to a collection of research approaches which emphasize the participation of those intended to be beneficiaries, users and stakeholders of the research (hereinafter referred to as 'end-users') in its development (Cargo & Mercer, 2008; Cornwall & Jewkes, 1995; Jagosh et al., 2012). End-users may include people affected by the issues under study (e.g., patients, caregivers) and/or those who apply research findings (e.g., policy makers) (Jagosh et al., 2012; Oliver et al., 2004). Depending on the context and setting, these end-users may work with researchers to (a) determine research questions, (b) decide on methodology, (c) participate in data collection and tool development, (d) interpret the findings, and/or (e) contribute to the dissemination of research results.

Researchers have used PR approaches for several decades in an effort "to help improve social and economic conditions, to effect change, and to reduce the distrust of the people being studied" (Hall, 1981; Macaulay et al., 1999, p. 745). Although there are different terms for the various PR approaches, including 'action research' and 'community based participatory research', each recognizes the value of engaging end-users in the construction of research and not solely as research subjects (Cargo et al., 2008; Macaulay et al., 1999). The specific implementation of a particular PR approach is dependent on the setting in which the study takes place, the type of end-users involved, and goals or values are driving a researcher to take a PR approach, rather than explicitly defined variations in the approach (Abma, Nierse, & Widdershoven, 2009; Cargo & Mercer, 2008; Cornwall & Jewkes, 1995; McKevitt, Fudge, & Wolfe, 2009). Accordingly, health researchers have employed PR approaches in a wide range of contexts, from including those affected by breast cancer (Yonas et al., 2006) and diabetes (Giachello et al., 2003), and with those from various racial and ethnic groups including African American (Marcus et al., 2004) and First Nations communities (Macaulay et al., 2007; Royle & Oliver, 2004; Viswanathan et al., 2004).

These partnerships have generated recruitment capacity; contextualized interventions to promote external validity; and have incorporated culturally centered knowledge in seeking to address the mistrust between academia and communities (Israel, Schulz, Parker, & Becker, 1998; Jagosh et al., 2012; Voegtle, 2008). However, working with end-users in such cases also poses significant challenges. “Communication problems and differing goals, values and work styles,” as Voegtle (2008) notes, “can produce tension[s]” between members of a developing partnership, and “the process is time-consuming” (Voegtle, 2008). Such challenges may be especially relevant as PR designs are applied to the population that is the focus of this thesis, individuals (and particularly adults) living with autism spectrum disorder (ASD).

First formally described in the 1940s, ASD refers to a range of complex neurodevelopmental disorders which are characterized by social impairments, communication difficulties, and repetitive patterns of behavior (Howlin & Moss, 2012). Currently, the estimated prevalence of ASD is 1 in 91 births (King & Bearman, 2009). Although ASDs persist throughout life, the focus of research has largely remained on childhood (Bailey, 2012), and research advances have centered around diagnosis, causation, and treatment approaches, particularly during childhood (Howlin & Moss, 2012). A disproportionately small number of articles in the academic literature have examined prognosis, outcomes, effective interventions, or real-life concerns presently impacting autistic adults including research on employment, social relationships, physical and mental health and quality of life (Howlin & Moss, 2012; Robertson, 2010) .

This disparity can be explained in part by political and social factors. Historically, ASD activism has been driven by parents who succeeded in securing significant funds for research on specialist educational provision, and research on early diagnosis and intensive early intervention. In response to the paucity of research for adults, individuals with ASD have sought greater influence in the how autism research is conducted and how findings are used (Nicolaidis et al., 2011). Additionally, through the growing role of funding organizations founded by families that include individuals with ASD (e.g., Autistica, Autism Speaks, Simons Foundation), individuals with ASD are developing a greater voice.

The purpose of my graduate research was to examine the state of participatory research partnerships in ASD research and to better understand the preferences of adults with ASD concerning involvement with ASD research. To achieve these goals, we developed a methodological approach that included a scoping review and a grounded theory study. Scoping reviews have been described as a process of mapping the existing literature or evidence base (Armstrong, Hall, Doyle, & Waters, 2011). As opposed to a systematic review which "focuses on a well-defined question where appropriate study designs can be identified in advance", a scoping review "addresses broader topics where many different designs might be applicable" (Arksey & O'Malley, 2005, p. 20). Given the variation in PR approaches and the limited number of PR studies involving individuals with ASD, we broadened the search criteria and sought to characterize the depth and breadth of research involvement of persons with neurodevelopmental disorders in published PR partnerships. Grounded theory methodology was used to understand the preferred process of adults with ASD toward research involvement. Grounded theory is one of many qualitative methodologies used to generate an in-depth understanding of interrelationships and the component parts of a phenomenon under study. Drawing from symbolic interactionism, this methodology best fit with our research question given its strength to systematically generate a conceptual description, grounded in data, about the dynamic process of the participants' preferences concerning involvement in autism research (Strauss & Corbin, 1990).

Personal Interest

I am grateful to have grown up with two close friends on the autism spectrum. In addition to exemplifying pure affection and loyalty, my friends also sparked my curiosity about the different ways brains develop, and the impact of this diversity on people's lives. As I continue to learn more about each of their special qualities and gifts every day, I wonder if the wider community has allowed itself to be as enriched by their uniqueness as well. In seeking to understand how the research community can draw from the insights and experiences of those with ASD, a path toward empowerment and inclusiveness can be forged.

Outline of Thesis

My thesis consists of two complementary papers. Chapter 2 presents a scoping review of literature examining participatory research partnerships involving individuals with ASD and other neurodevelopmental disorders. This review informed my qualitative study, which is presented in Chapter 3, and was my primary thesis project. In this study, I explored the preferences of adults with

ASD living in Edmonton, Alberta, concerning their potential involvement in the ASD research process. My thesis concludes with Chapter 4, Conclusions, which are based on the findings of my graduate work and considers how the scoping review and qualitative study provide complementary perspectives on the application of PR methods to involving adults with ASD. Lastly, Appendix A includes the search strategy for the MEDLINE® database used for the scoping review, Appendix B includes ethics documents (study poster and information sheet), and Appendix C includes the data collection tools I used to conduct my qualitative research project.

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CHAPTER 2

Title: Assessing the Influence of Participatory Research Partnerships on the Research Process and Outcomes in Autism Spectrum Disorder Research: A Scoping Review

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Abstract

Background

Participatory research (PR) is an approach that aims to increase the relevance and broaden the implementation of health research by involving those affected by the outcomes of health studies (end-users). Few studies within the field of autism spectrum disorders (ASD) research, however, have involved autistic individuals as co-researchers—that is, as end-users who participate in decision-making throughout the research process.

Objectives

To (1) identify and characterize published PR partnerships between academic researchers and individuals with ASD or other neurodevelopmental disorders, and (2) examine the influence of PR partnerships on the research process and reported study outcomes.

Methods

A search of MEDLINE®, EBSCO, EMBASE, ERIC and PsycINFO, and a review of grey literature (reference lists from included studies and web searches) was conducted. One reviewer completed independent screening of study abstracts and extracted the data. A second reviewer checked for completeness and accuracy.

Results

Six studies were identified that described PR partnerships between academic researchers and individuals with ASD or other neurodevelopmental disorders. One study examined PR partnerships between academic researchers and adults with ASD, and five studies examined participatory research partnerships with adults with other neurodevelopmental disorders (i.e., intellectual disability). A comparative analysis of the studies revealed two key themes: (1) variations in the PR design of the studies and (2) limitations in the reporting of the depth of the co-researcher's involvement across studies. Both themes are related to potential limitations in the application and generalizability of the findings

Conclusion

Within the studies identified by this review, gaps were identified in the reporting of the process of PR partnerships. The use of evaluative frameworks for such PR studies is needed to determine the potential benefits of PR partnerships within this population.

Introduction

The growing emphasis on participatory research (PR) over the past decade reflects the belief that research developed through active partnerships between researchers and those affected by the issues being studied (hereinafter referred to as 'end-users') leads to higher-quality research that is more relevant and applicable to all involved in the process (Boote, Telford, & Cooper, 2002; Cargo & Mercer, 2008; Israel, Schulz, Parker, & Becker, 1998; Macaulay et al., 2011). This trend also reflects the impact of increased accountability to publicly supported research funders: researchers are now often expected to specify how end-users will be involved in the research study (Boote et al., 2002; Cargo & Mercer, 2008). Unlike conventional research paradigms, where decision-making rests solely with researchers (Macaulay, 2007), PR methods encompass a range of approaches which engage end-users directly in the research process. Arguably, end-users' real-world knowledge and lived experiences can complement the researchers' theoretical and methodological expertise, thereby enhancing the process and products of research (Cargo & Mercer, 2008; Viswanathan et al., 2004). The implementation of PR approaches is generally determined by the preferences of the researcher and/or the parameters of the study, rather than by explicitly defined variations in the approach (Abma, Nierse, & Widdershoven, 2009; Cornwall & Jewkes, 1995; McKevitt, Fudge, & Wolfe, 2009). At one end of the spectrum, end-users may only be involved in one stage of the research study as consultants or collaborators. However, in its fullest expression, the researcher-end-user partnership involves collaborative decision-making throughout the research process as a co-researcher, from developing the question to collecting, analyzing and interpreting the data; developing conclusions; and disseminating the results (Green & Mercer, 2001; Macaulay et al., 2007; Oliver et al., 2004).

Community based participatory research (CBPR) is one of the most widely recognized PR approaches with a growing number of applications, particularly in communities defined by geographic, racial and/or ethnic group (Cargo & Mercer, 2008). CBPR partnerships have also been undertaken in response to a number of health conditions in diverse populations including those affected by breast cancer (Yonas et al., 2006) and diabetes (Giachello et al., 2003) generating recruitment capacity and contextualizing interventions to promote external validity. These partnerships have also incorporated culturally centered knowledge in seeking to address the mistrust between academia and communities with those from various racial and ethnic groups including African American (Marcus et al., 2004) and First Nations communities (Macaulay et al., 2007; Royle & Oliver, 2004; Viswanathan et al., 2004). As noted in Chapter 1, working with end-users can have distinct challenges; despite the benefits of CBPR, as Voegtle (2008) observes, “communication problems and differing goals, values, and work styles” can produce tensions between members of a developing partnership, and the “process is time-consuming” (p.4).

Although PR partnerships may have been conducted with a variety of stakeholders within the neurodevelopmental field, including parents and caregivers, as yet, relatively few PR studies have involved people with autism spectrum disorders (ASD) or other neurodevelopmental disorders as research partners or co-researchers (Gilbert, 2004; Pellicano & Stears, 2011; Walmsley, 2004). In addition, no comprehensive review has been conducted to characterize how the participation of persons with ASD or other neurodevelopmental disorders influences the processes and outcomes of research. Neurodevelopmental disorders encompass a broad range of impairments associated primarily with the growth and development of the brain or central nervous system. They include intellectual disabilities (ID), attention-deficit/hyperactivity disorder, autism spectrum disorders, and learning disabilities. Collectively, individuals with ASD and other neurodevelopmental disorders experience varying degrees of challenges with speech and language, social relationships, motor skills, behavior, memory, and learning, that have implications for PR designs. Although such challenges may limit the degree to which people can be involved in certain activities (e.g., for some individuals, tasks that require a high degree of abstract thinking), it has been demonstrated that people with neurodevelopmental disorders (in some contexts, with support) can participate in the development of research in many if not all stages (Gilbert, 2004; Walmsley, 2004).

In light of the strength of the ASD advocacy community and the growing role of funding organizations founded by families that include individuals with ASD (e.g., Autistica, Autism Speaks, Simons Foundation), it is important to consider the potential impacts of PR partnerships that allow people with ASD to become more fully engaged in guiding and developing ASD research. Such initiatives may increase the relevance and applicability of this research. Accordingly, the aims of this scoping review were to (1) identify and characterize published PR partnerships between academic researchers and individuals with ASD or other neurodevelopmental disorders, and (2) examine the influence of PR partnerships on the research processes and outcomes reported in primary literature.

Methods

This scoping review was conducted to characterize the depth and breadth of research involvement of persons with neurodevelopmental disorders in published PR partnerships, using the methodological approach defined by Arksey and O’Malley (2005). Accordingly, this review followed five steps:

1. Development of Research Questions

Three questions guided the selection of relevant sources for this review: (a) What is the nature of the research activity concerning PR focused on neurodevelopmental disorders? (b) What is the breadth and depth of PR partnerships which have been formed between researchers and individuals with neurodevelopmental disorders? (c) How are the processes and outcomes of PR partnerships between researchers and individuals with neurodevelopmental disorders reported?

2. Identifying Relevant Publications

To ensure comprehensiveness in identifying relevant primary studies, literature was sourced through electronic databases and reference lists of relevant literature (Arksey & O'Malley, 2005). We located publications in multiple steps, first targeting electronic literature databases. Once relevant material was selected from each of these sources, we searched relevant websites, URLs and reference lists of key studies to increase our capture of relevant material.

Preliminary search terms were developed by the research team to reflect a number of core concepts pertinent to PR. These related to characterizing specific PR approaches that have been utilized, the role of individuals with neurodevelopmental disorders, and the outcomes of PR partnerships. To cover the breadth of the PR field, the following terms were included along with their synonyms: community-based PR, action research, inclusive research, participatory action research (PAR), participative evaluation, and emancipatory evaluation (Jagosh et al., 2011). Theoretical and conceptual work that provides an important backdrop to the scoping review is referenced throughout this paper; however, only primary studies of PR conducted with individuals with neurodevelopmental disorders are reported in the findings.

Scoping reviews generally consult only a few main health research databases for literature; as such, we searched five electronic databases: MEDLINE®, EBSCO, EMBASE, ERIC and PsycINFO. This selection was designed to cover health, psychology and education literature. The final search, undertaken with the help of a health science librarian, was implemented in September 2012. In all databases, terms related to participatory research/research participation/advocacy were combined with terms related to autism/ neurodevelopmental disorders/brain injury. For each set of terms, both controlled vocabulary and text words were utilized. No restrictions on publication date were applied. Duplicate references were filtered out as each subsequent database was searched. Appendix 1 contains the search strategy for the MEDLINE® database; identical or slightly variant versions were employed in the remaining databases. A review of grey literature was also conducted which included web searches, reference lists, and conversations with stakeholders and authors via email.

3. Screening and Selection of Publications

The screening process of studies was guided by four criteria. Included studies were required to have: (1) involved adults with neurodevelopmental disorders as research partners, (2) been reported in English, (3) reported or referred to primary empirical research, and (4) contained a description of how PR partnerships were established and implemented, and/or described the outcomes of

PR partnerships on neurodevelopmental disorder research, and/or described barriers and/or facilitators to the establishment and implementation of PR partnerships (Jagosh et al., 2011).

With the help of a multidisciplinary research team, one reviewer completed independent screening of study abstracts and extracted the data. A second reviewer checked for completeness and accuracy. Given that aim of the study was to examine PR partnerships implemented and reported in research, articles which were not primary studies, such as news items, letters, editorials, book reviews and articles appearing in newsletters or magazines, were excluded.

4. Organizing Publications

Included studies were reviewed and summarized using a framework, adapted from Oliver et al. (2004). Accordingly, descriptive study characteristics were collated in a detailed Microsoft Excel spreadsheet (version 14.0.6112.5000, Microsoft Corporation, Santa Rosa, CA, USA). These characteristics included the country of the first author, the year the study was published, the study population, the study’s aim, the method of involving end-users, and key findings including PR-related outcomes, processes, contexts, challenges, as well as barriers and facilitators.

5. Reporting the Results

A narrative and descriptive account was generated for each reported PR partnership. A comparative analysis focusing on the context for the study, the breadth and depth of the co-researcher’s involvement throughout the research process, and the outcomes attributed to the co-researcher’s involvement, was conducted. Main findings were merged into two broader themes which served as a guide for organizing the discussion: (1) variations in the PR design of the studies and (2) limitations in the reporting of the depth of the co-researcher's involvement across studies.

Results

Figure 2-1 provides an overview of the search and retrieval process. Our search strategy yielded 1,431 citations for initial screening. An additional 10 citations from web-site searching and reference lists were reviewed. After duplicates were removed, 636 unique citations remained. Of these, 586 abstracts were excluded because they did not describe PR in individuals with ASD or other neurodevelopmental disorders, yielding a total of 60 papers. Of these, 54 articles were excluded because they did not report or refer to empirical research (n=37), or discuss how PR partnerships were implemented, or the outcomes of partnerships (n=17). Thus, six studies met inclusion criteria and were included in this review (see Figure 2-1). The included studies were published between 2006 and 2012: three in Australia or New Zealand, two in the United States, and one in the Netherlands. Five studies described PR partnerships with individuals with ID and one study focused on a study population with ASD. The involvement of co-researcher(s) in specified stages of the study development within included studies is summarized in Table 2-1. The context, depth, breadth and outcomes of each of the six studies are described below.

Bigby and Frawley (2010). These authors reported on the study, “Making Life Good in the Community,” which examined factors involved in achieving positive outcomes for people with a disability transitioning from an institutional care model to a group home. The study comprised qualitative and quantitative components. Data were collected through ethnography and PAR using interviews and surveys. The study team included a co-researcher with an ID to work on a portion of the study that sought to understand how to support people with an ID living in group homes.

While it was intended that the co-researcher would be integrally involved as part of the research team, no expectations were reported regarding this role or how the co-researcher would be supported. The researchers initially treated the co-researcher “as [they would] any other new relatively inexperienced research assistant” (Bigby & Frawley, 2010, p. 56) and provided him with training on research methodology and included him in meetings (Bigby & Frawley, 2010). A mentor identified as having both research skills and experience working with people with an ID was hired to provide practical support to the co-researcher. As the study unfolded, the co-researcher and mentor were contracted to manage field visits to group homes; collect, record and transcribe observations about the comfort of different houses; analyze data; and present findings of the study at various conferences. While the authors noted that conference presentations delivered by the co-researcher enhanced the impact and authenticity of dissemination efforts, no formal evaluation was conducted.

Upon study completion, the research team evaluated the process of involving the co-researcher as part of the research team using Walmsley and Johnson's (2003) framework of inclusive research. Because the co-researcher was hired a year into the study and after funding for the study had been awarded, he had no influence on shaping the research questions, methodology, or study design. The study authors noted that the short timelines imposed by grant funding cycles prevented the co-researcher from maintaining a degree of control over the study, although they also acknowledged concerns about maintaining rigor, and their group's lack of experience in participatory approaches. The authors also noted that the co-researcher's perspectives were influenced by significant input from the research mentor. As part of the study, during the data collection stage, the co-researcher contributed to a report summarizing his observations of studied group homes. While the research mentor felt that the ideas captured in the report were a product of a collaborative partnership, the unique intellectual contribution of the co-researcher was not delineated. Based on the components outlined, the authors concluded that the study did not conform to an ideal type of inclusive research as it was neither initiated directly by people with ID, nor owned by them, nor reflective of their interests and experience (Bigby & Frawley, 2010).

Timmons et al. (2011). These authors reported on a study aimed at identifying factors that influenced the employment-related decision-making of individuals with intellectual and other developmental disabilities, and examined whether individual's preferences were consistent with existing employment options and choices. A methodological goal of the study was to fully involve an individual with an ID as a co-researcher (Timmons, Hall, Bose, Wolfe, & Winsor, 2011).

After orienting the co-researcher to the study, the research team developed a training and support plan to integrate her as part of the larger team. With the support of a research mentor, the co-researcher contributed to various stages of the study. She assisted with recruiting study participants and contributed to data collection by developing the consent procedure and conducting face-to-face interviews with participants alongside one of the study authors. In addition, the co-researcher participated in data analysis by coding transcripts and creating memos with members of the research team. The authors did not address the extent to which the co-researcher's involvement was supported by the research mentor nor how her specific perspectives were integrated into the overall interpretation of study findings.

Conder, Milner and Mirfin-Veitch (2011). This paper examined a mixed methods study seeking to develop a quality-of-life questionnaire for individuals with ID. The study authors recognized that people who use services are most knowledgeable about how the services work. Using the participatory action framework, they involved six co-workers with an ID in focus groups identifying quality-of-life indicators for the questionnaire. They also involved the co-workers in other stages of the research (Conder, Milner, & Mirfin-Veitch, 2011) such as training co-researchers in research methods and orienting them to the chosen quality-of-life framework. In addition to conducting focus groups, co-researchers were involved in selecting a name for the project, reviewing the consent process with each participant, analyzing data, developing the questionnaire, and disseminating findings by way of a report of the study and plain language summary. Given the range of tasks led by the co-researchers, a support team provided practical assistance including booking flights and accommodations for travel to collect data for the study, arranging meetings to answer any questions, and ensuring that the co-researchers were comfortable with their research roles.

Because the co-researchers were involved in the study after the terms of reference, timelines, and budget had been agreed upon, they did not have the opportunity to inform the action steps or the quality-of-life framework selected by the research team. A tight study time frame also limited the research team's ability to act on suggestions articulated by the co-researchers. Consequently, both the co-researchers and other members of the research team acknowledged that the capacity for participation in the study was less for the co-researchers. The study authors noted that the co-researchers contributed to team discussions, particularly in the latter stages of the project, and that their ideas influenced the final product. However, the authors do not provide specific examples of intellectual contributions made by the co-researchers, nor how the co-researchers' ideas contributed to the overall interpretation of the data. The study authors concluded that the team achieved their goal of having individuals with IDs author a quality-of-life questionnaire, thus ensuring that individuals for whom the instrument was designed would be involved in its development.

McVilly et al. (2006). These authors conducted a large, multi method study that, in part, examined the friendship experiences and aspirations of adults with an ID (McVilly et al., 2006). While the study was not intentionally established within a PR framework, the authors discuss many characteristics of PR that were included in the study's design—researchers were considered to be experts on the technical process of research, whereas self-advocate co-researchers and study participants were considered to be experts on the subject of friendship.

Self-advocates were initially involved in formulating the topic of inquiry and later took part in the review and analysis of the data. The data analysis stage involved an expert group of 11 self-advocates with an ID who reviewed findings from three phases of data collection (McVilly, Stancliffe, Parmenter, & Burton-Smith, 2006). The study authors noted that the dynamics of the expert group provided an environment in which participants could debate among themselves issues of relevance to them, rather than entering into a "dyadic discourse with a researcher and consequently needing to negotiate the power imbalances inherent in such a discourse and the potential influence of this power imbalance on the findings" (McVilly et al., 2006, p. 705). Many of the recommendations for action and further research were from the expert group's review of study findings.

The study authors noted that the expert group enabled the research team to "assess, evaluate and refine findings based on the views and priorities of people with intellectual disabilities themselves" (McVilly et al., 2006, p. 706). By following this process, the authors mentioned, they could ensure their own perspective did not exert too much influence over the outcomes of the study, and test the validity of the research team’s interpretation of the findings using the collective expertise of a group of adults with IDs. The authors concluded that convening the expert panel was an effective way to review and analyze data and ensure that panel members “had the last say on the topic of inquiry” (McVilly et al., 2006, p. 706). They also concluded that engaging self-advocates in the expert review group was crucial to the overall investigation and contributed to the depth, breadth and rigor of the research process.

Nierse and Abma (2011). These authors reported on a study that aimed to develop a research agenda for people with an ID. Recognizing that current research did not incorporate the needs of people with IDs or their parents, the project involved two adults with a mild ID and the mother of a child with severe ID, who participated in the study as partners on the research team. A responsive methodology was used in the agenda-setting process. In the context of this methodology, specific research activities were not determined beforehand, but were developed in consultation with the stakeholders. This allowed the research team to adapt to the issues that emerge from the subjective experiences and stories of stakeholders (Nierse & Abma, 2011). The decision to involve research partners meant that academic researchers “did not act as experts, but as supporters and assistants of the research partners” (Nierse & Abma, 2011, p. 413). However, the research partners were not involved in identifying the research question (funding was secured prior to their involvement), but rather, were involved in data collection and in the analysis and dissemination of the results. The research partners recruited participants through personal networks, prepared topic lists for interviews, interviewed participants, moderated or co-moderated focus groups, analyzed transcripts, and participated in the construction of the study's questionnaire.

The study authors reported that "the active involvement of people with IDs as research partners was important for the entire process," and that it resulted in "co-ownership of the research agenda and empowered both people with ID, who reported benefits, such as improved knowledge and skills, self-esteem and meaningfulness" (Nierse & Abma, 2011, p. 419). The research team, however, did not pursue a formal evaluation which might have supported this claim. The study authors also noted that a particular strength among the research partners that enhanced research outcomes was that they were able to engage people in conversation, feel "more empathy," encourage "much more authentic" responses from participants, and help "participants to share their intimate voice" (Nierse & Abma, 2011, p. 419). However, the authors did not provide details (e.g., specific criteria or measures) about how these processes were evaluated. Although the authors noted that adults with ID and parents prioritized research differently, no distinction regarding their involvement in the research process was made.

Nicolaidis et al. (2011). This article reported on a collaborative initiative to build an academic-community partnership that uses Community Based Participatory Research (CBPR) to improve the lives of people with ASD. The misalignment between researchers' priorities and those of the autistic community and the exclusion of individuals with ASD from the research process led to the development of the Academic Autistic Spectrum Partnership in Research and

Education (AASPIRE). AASPIRE investigates the health care experiences and well-being of autistic adults and is composed of health services disability researchers, self-advocates, health care providers, disability service professionals, and family members (Nicolaidis et al., 2011).

Nicolaidis et al. (2011) reported that the philosophy espoused by AASPIRE was that academic researchers, individuals with autism, and other community partners (e.g. family members) bring unique and complementary professional and personal expertise and thus should serve as equal partners throughout the entire research process. Community partners ensure that the research is “respectful, accessible and socially relevant,” and academic researchers ensure that research is “scientifically sound and academically relevant, that the work has the proper rigor, and that it advances academic goals” (Nicolaidis et al., 2011, p. 145). Members of AASPIRE pursue research projects which align with priorities areas identified by individuals with ASD and family members. Furthermore, individuals with ASD are actively involved throughout the development of study and contribute to “designing protocols, developing and adapting instruments and consent materials, recruiting participants, collecting and analyzing data, and disseminating findings” (Nicolaidis et al., 2011, p. 145).

Individuals with ASD and family members also “inform the group about potentially offensive or unclear language or assumptions,” and are involved in selecting and adapting study instruments (protocols, recruitment and consent materials, and interview guides) to ensure they are accessible to participants. (Nicolaidis et al., 2011, p. 146). The collaborative approach offers strategies for conducting CBPR with autistic self-advocates. These strategies are designed to integrate text-based online media and consensus building processes to “successfully equalize power and accommodate the diverse communication and collaboration needs” of self-advocates (Nicolaidis et al., 2011, p. 143).

Discussion

This scoping review revealed that researchers are implementing PR approaches in a wide range of contexts, using a variety of methodologies. Of the six studies identified in a literature search that documented PR partnerships between academic researchers and individuals with ASD or other neurodevelopmental disorders, five studies described PR partnerships with individuals with IDs, and one study was focused on a study population with ASD. Four of the studies were mixed-method by design, and two studies were qualitative. Five of the partnerships were described as initiated by the research team and one paper described a partnership that was co-initiated by researchers and individuals with neurodevelopmental disorders. The extent of the co-researcher’s involvement varied among the studies. In all but two studies, the co-researcher was recruited after the study was funded and the research question and methodology had been selected; as a result, the co-researcher had less opportunity to inform the study design (Table 2-1). In such cases, co-researchers were primarily involved in assisting researchers to gain access to certain populations and carrying out tasks determined by study authors. In five of the studies, co-researchers were involved in analyzing data. In only one study did the co-researchers share authorship with researchers in the papers describing partnerships. The number of co-researchers involved in partnerships in studies ranged from one to eleven.

A comparative analysis of the studies revealed two key themes, both related to potential limitations in the use and generalizability of the findings: (1) variations in the PR design of the studies and (2) limitations in the reporting of the depth of the co-researcher's involvement across studies. As concerns variation in PR design across studies, there were differences in the extent to which co-researchers were involved in the development of the studies and a lack of clarity regarding the role of the co-researcher as a research partner. The limitations in reporting and evaluation included a lack of clarity on how multiple perspectives were integrated into decision making and a lack of formal evaluation of partnerships formed with co-researchers and how they were developed.

Variations in Participatory Research (PR) Design

As demonstrated by this scoping review, the extent of end-user involvement in PR studies involving individuals with neurodevelopmental disorders varied. Given the spectrum of involvement that defines PR, the basis for evaluating whether a study is truly participatory is unclear. In only one of the studies included in this review were co-researchers involved in informing study methodology (Table 2-1). Yet, the potential for end-users to enhance the process and outcomes of research by being involved in selecting methods has been reported in the broader PR literature. More specifically, the participation of end-users in methods selection has been reported to increase the validity of a study by increasing the depth and variety of data collected, establishing congruence between a research question and a local reality, and adapting study methodology to specific contexts (Macaulay et al., 1999; Sandoval et al., 2012; Viswanathan et al., 2004). Furthermore, in regard to the rationale for variations in approaches to involving co-researchers in the stages of research, little justification was offered beyond expediency, convenience, and logistic challenges. Increasing the responsibility for researchers to specify how co-researchers will be involved throughout a study in funding applications may help to prevent researchers from citing "funding already in place" as a rationale for not involving end-users from the outset.

Another theme that emerged in this review is the question of what differentiates a 'co-researcher' from a research assistant. More specifically, in five of the studies, where the co-researchers were assigned tasks by researchers, the boundary was not clearly drawn in terms of what is considered PR and what might be considered hiring someone with a disability to assist with a study. Whereas a research assistant may carry out technical aspects of a study, previous literature on PR (Jagosh et al., 2011; Macaulay et al., 1999) suggest that a co-researcher, as part of the investigative team, should be involved in framing questions, interpreting results, and putting them into context. With the exception of two studies, there was an emphasis on the co-researcher's involvement in operational tasks (e.g., data collection) rather than in higher order aspects of the research process (e.g., analyzing data), which speaks to a hierarchy within the research team and a narrow view of 'participation'. Wider use of evaluative tools including the framework we used to assess the breadth, depth and outcomes of PR partnerships included in this review (Figure 2-2) may provide a fuller understanding of the degree to which the co-researcher is contributing to every step ultimately needed to determine how genuine the partnership is (Blackstock, Kelly, & Horsey, 2007).

Limitations in Reporting the Depth of Co-researchers' Involvement

There were varying efforts across the studies to involve co-researchers in the research process (Table 2-1); however, it was generally not made explicit how the multiple perspectives from 'around the table', including that of the co-researcher, were integrated into study decision making. With the exception of one study, it was unclear how dissenting views were integrated into an overall perspective and how consensus was reached in decision making throughout the course of the studies. Because of this lack of reporting, the unique intellectual contributions of the co-researchers at various stages of the research studies were not explicitly acknowledged. Moreover, this review revealed a lack of clarity in the extent to which a co-researcher influenced the overall development of a study, or informed conclusions or contributed to a research study's outcomes.

In general, the influence of PR partnerships on the research process and study outcomes was rarely addressed. The process of interpreting data and the dynamics of working in a group were narrowly reported in the literature. As such, this review calls for future research in this field to more specifically evaluate the contributions of co-researchers with ASD or other neurodevelopmental disorders, and for the creation and testing of tools to assess participatory structures of research engagement for this population. Ultimately, this research may contribute to improvement in research outcomes, but that remains to be demonstrated.

Limitations of the Study

There were some limitations to this scoping review. Ultimately, our findings are limited by the information available in the published literature, and in particular how the roles of co-researchers were described and evaluated. Only one study included a co-researcher as a study author. As a result, missing from our review, and another limitation, are data on the perspectives and experiences of the PR partnership as described by co-researchers. Given the communication delays common in ASD, further exploration is warranted addressing how to support such individuals in articulating their perspective on research so as to potentially improve and/or optimize PR partnerships in the field.

Conclusions

Several studies over the past decades have attempted to involve individuals with neurodevelopmental disorders as co-researchers. Although this inclusion of end-users in research is appealing from an empowerment perspective, this review revealed gaps in how the process and outcomes of such PR partnerships are reported. More specifically, further clarity on the extent to which co-researchers were involved in the development of the study, the roles and responsibilities they undertook relative to other research partners, how their perspectives were integrated into decision making, and how the partnerships were evaluated, is called for. This review calls for the wider use of evaluative frameworks in PR studies within this population as a key to understanding how people with ASD and other neurodevelopmental disorders contribute to research teams and projects and how their involvement is linked to tangible research outcomes.

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Figure 2-1

Flow Diagram

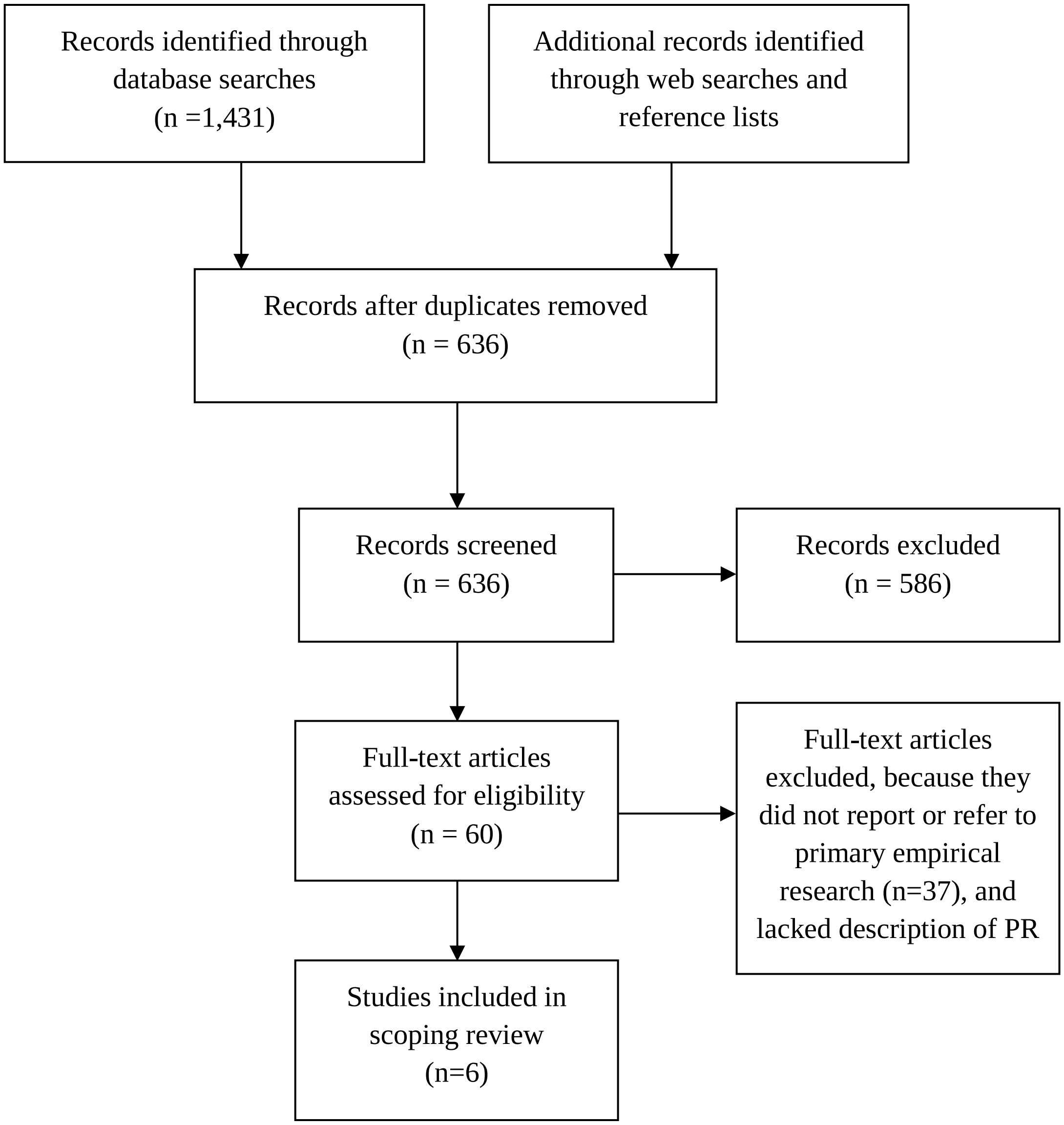

Figure 2-2

Framework for Assessing Breadth, Depth, and Outcomes of Participatory Research Partnerships with Individuals with Autism Spectrum Disorder (ASD) and Other Neurodevelopmental Disorders

(1) Context
  a. Study purpose
  b. Study method
  c. PR purpose
  d. PR method
  e. Was PR required or inspired?
  f. What theory underpinned involvement as described by reports' authors?
  g. Did end-users share authorship?

(2) Breadth
  a. Which stages of the research did end-users participate in?
    i. Identifying or setting the research questions
    ii. Setting the methodology
    iii. Collecting data
    iv. Analyzing data
    v. Uptake or dissemination of research findings
  b. How were end-users involved in each stage of research?

(3) Depth
  a. Which end-users were involved?
  b. How were end-users involved?
  c. What was the direction of the approach/what was the degree of consumer involvement?
    i. Researcher- or consumer-initiated
    ii. Consumers consulted
    iii. Consumer collaborators
    iv. Consumers in control
  d. What were the forums for exchange?
    i. Committee membership
    ii. Written consultation
    iii. Focus groups
    iv. Public meetings
  e. What methods were used for decision-making?

(4) PR Outcomes
  a. How did PR partnerships influence the research process and outcomes?
  b. What were the intended outcomes of PR?
  c. What outcomes were achieved
  d. What outcomes were not specified?
  e. How did involvement from individuals with neurodevelopmental disorders change research outcomes (vs. influence the world views/perspectives of the investigators)?
  f. Which barriers to, and facilitators of, meaningful participation by consumers in PR partnerships are described?

Table 2-1

Summary of Breadth of Participatory Research Studies Involving Individuals with Autism Spectrum Disorder (ASD) and Other Neurodevelopmental Disorders

Authors	Identifying research question	Informing study methodology and methods	Collecting data	Analyzing data	Disseminating research findings
Bigby and Frawley (2010)			✓	✓	✓
Timmons et al. (2011)			✓	✓	
Conder et al. (2011)			✓	✓	✓
McVilly et al. (2006)	✓			✓	
Nierse and Abma (2011)			✓	✓	✓
Nicolaidis et al. (2011)	✓	✓	✓	✓	✓

Note. Checkmarks indicate the involvement of co-researcher(s) in specified stage of the study development.

Table 2-2 *Summary of Studies Addressing Participatory Research (PR) Partnerships with Individuals with Autism Spectrum Disorder (ASD) and Other Neurodevelopmental Disorders*

Paper; year; country; subject	Study Purpose	Rationale for PR Method	Breadth of PR	Depth of PR	PR Outcomes
Bigby and Frawley; 2010; Australia; co-researcher with an intellectual disability	To examine how to support individuals with intellectual disabilities living in group homes so that they can lead fulfilling lives.	Action research allowed for reflective process and immediate analysis of ideas; full research team contributed reflections.	With the support of a mentor, co-researcher participated in collecting and analyzing data and disseminating research findings.	Co-researcher and research mentor hired for one year to work on funded-project. Co-researcher participated in four meetings with research team, and three meetings with research team and mentor.	Conference presentations delivered by the co-researcher enhanced the impact and authenticity of dissemination efforts.
Timmons et al.; 2011; USA; co-researcher with an intellectual disability	To understand what factors influence employment-related decision-making among individuals with intellectual and developmental disabilities and the extent to which their preferences correspond to existing employment options.	Participatory action research allowed for full involvement of co-researcher, reduction of “barriers between researcher and participant” and development of collaborative solutions to problems.	With the support of a project mentor, co-researcher performed various tasks during data collection and analysis.	Co-researcher and research mentor hired for the duration of the project. Co-researcher was involved in the research team’s decision-making.	Outcome of co-researcher’s participation not reported.

Conder et al.; 2011; New Zealand; co-researcher with an intellectual disability	To involve service users in the development of a tool which measures quality of life for people with intellectual disabilities.	Reflective practice allowed for those most affected by the outcomes of research to have ownership over its development, increasing the likelihood that findings would be directly useful.	With the assistance of contracted researchers and a support team, co-researchers led focus groups, assisted in selecting the study title, reviewed the consent process with participants, analyzed data, developed the questionnaire, and disseminated the findings.	The co-researcher and support team were hired for the duration of the project. The co-researcher participated in formal and informal meetings with the research team.	Individuals with intellectual disabilities authored a quality-of-life questionnaire.
McVilly et al.; 2006; Australia; self-advocates with intellectual disabilities	To examine friendship experiences and aspirations of adults with intellectual disabilities.	Participatory research recognized self-advocates as experts in their understanding and researchers as experts on the technical process of research.	Self-advocates were involved in formulating the topic of inquiry and reviewing and analyzing data as part of an expert group.	The researcher invited self-advocates to participate. Self-advocates led the expert panel with support of researcher.	The expert group allowed self-advocates to interpret findings independent of researchers and assured researchers that findings were relevant and contextualized to self-advocates.

Nierse and Abma; 2011; The Netherlands; research partners with intellectual disabilities	To develop a research agenda with people who have intellectual disabilities.	Responsive methodology allowed researchers to adapt to issues that emerged from stakeholders' experiences.	Research partners were actively involved in recruiting participants, data collection, analysis and dissemination of the results.	Research partners were invited to participate by researchers. Research partners met with entire team every two weeks and participated in additional activities and informal meetings with the research team.	Research partners drew authentic responses from study participants during interviews.
Nicolaidis et al.; 2011; self-advocates with autism spectrum disorders	To build an academic-community partnership that uses CBPR to improve the lives of people with ASD.	Community-based participation allowed adults on the spectrum to be included in matters directly affecting them and self-advocates to serve as equal partners in research.	Self-advocates worked alongside the committee in deciding research questions, collecting and analyzing data, and disseminating findings.	To reach consensus, self-advocates were involved in ongoing group decision-making by way of a text-based online medium and a five-finger method to reach consensus.	Self-advocates informed the group about potentially offensive or unclear language or assumptions and ensured that the autistic community considered the research relevant.

CHAPTER 3

Title: Exploring the Process of Research Involvement Preferred by Adults with Autism Spectrum Disorder (ASD)

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Abstract

Background

Participatory research (PR) partnerships are a collaborative approach to research. They seek to increase the role of those affected by the outcomes of health studies (end-users) in the production of the research, with the intent of producing findings which are relevant and applicable. This study sought to determine how adults with ASD would prefer to be involved in ASD research.

Methods

We conducted the study using grounded theory methodology. Purposeful sampling was used to identify and enrol adults with ASD (n=8) from Edmonton, Canada who were willing to discuss their preferences about research involvement and were able to indicate consent to participate. Data were collected via semi-structured individual interviews.

Results

Emerging from the data was a grounded theory that introduced contextual and core concepts that help to explain the preferred process of research involvement for individuals with ASD. Contextual concepts include the motivations of individuals with ASD for wanting to be involved—a desire to gain information they can use, and to contribute to research that responds to the needs of adults with ASD—and the challenges impeding their involvement given that ASD is defined by atypical social interactions and communication. The core concepts include principles of (a) involving individuals with ASD in the planning of the research, (b) recognizing the important ways that both the adult with ASD and the

researcher contribute to the research process, and (c) ensuring that individuals with ASD feel comfortable approaching researchers to offer their input.

Conclusions

The preferred process of research involvement for individuals with ASD is explained by their motivations for taking part in research and the challenges impeding that participation, and by principles of involvement which ultimately lead to the production of research that is useful to individuals with ASD.

Introduction

Autism Spectrum Disorder (ASD) refers to a range of complex neurodevelopmental disorders characterized by social impairments, communication difficulties, and repetitive behaviours. While advances in the science of ASD have excited and challenged researchers, these developments have also generated controversy within the larger ASD community (Pellicano & Stears, 2011). Moreover, ASD stakeholders (adults with ASD, family/informal caregivers, clinicians, researchers, and policymakers) have reported that they feel disconnected from, and even poorly served by, the priorities of researchers (Krahn & Fenton, 2012; Nicolaidis et al., 2011). Studies also suggest that some individuals with ASD reject traditional medical approaches (Offit & Moser, 2009), that policy-makers have been slow in making evidence-based interventions accessible to families affected by ASD (Feinberg & Vacca, 2000), and that individuals with ASD are dissatisfied with attempts to “cure” or “prevent” their ASD without their consent (Bagatell, 2010).

Recent studies have sought to address the above concerns by involving individuals with ASD as partners in the research process (Nicolaidis et al., 2011; Szatmari, 2009). Various approaches include one-time consultation meetings (see www.cairn-site.com/en/conference/2009/cc.html) (Szatmari, 2009), which have generated prioritized ‘research agendas’ but have not led to demonstrable changes in research outcomes or stakeholder involvement. Participatory research (PR) partnerships, a collaborative approach to research, may offer opportunities to change this traditionally limited role of the ASD community in research. PR requires research end-users to work together with researchers to (a) determine research questions, (b) decide on methodology, (c) participate in data collection and tool development, (d) interpret the findings, and (e) contribute to the dissemination of research results (Bowen & Graham, 2013). Such ongoing involvement can yield research findings that are more relevant and applicable to all involved in the process (Cargo & Mercer, 2008; Graham & Tetroe, 2009; Straus, Tetroe, & Graham, 2009).

Despite the potential value of this approach, to our knowledge, there is only one published study describing the application of a PR design with adults with ASD. The Academic Autistic Spectrum Partnership in Research and Education (AASPIRE) is a collaborative initiative to build an academic-community partnership. The partnership is based on Community Based Participatory Research (CBPR) to investigate “the health care experiences and well-being of autistic adults” and is composed of “health services disability researchers, individuals with ASD, health care providers, disability service professionals, and family members" (Nicolaidis et al., 2011, p. 143). Within AASPIRE, academic and community members serve as equal partners in all phases of the research and jointly conduct research studies that address the priorities of community partners (Nicolaidis et al., 2011).

Although the AASPIRE model appears promising, it is important in developing approaches for the implementation of PR with individuals with ASD that we start by understanding their preferences towards being involved. As such, this study sought to understand the role adults with ASD want to have in developing ASD research. More specifically, we sought to understand how adults with ASD describe (1) a preferred process of involvement in the development of research, (2) how they want to be involved in this preferred process, and (3) how they want the research community to interact and engage with them in this process.

Methods

Study Design

A qualitative design, based on a grounded theory approach, was used to generate a theory about the preferences of adults with ASD concerning their involvement in developing research. A qualitative method of inquiry was well suited to this research purpose, as it uses observations and interviews to obtain a deep and rich understanding about a phenomenon. Grounded theory methodology is the qualitative method of choice when the aim of the study is to learn from the participants how to understand processes associated with a substantive situation

(Morse & Field, 1995). The perspectives of the study's participants, adults with ASD, were sought in order to explain the phenomenon based on concepts that emerged from the data rather than through applying existing theoretical ideas or hypotheses to the data (Strauss & Corbin, 1998). This methodology was used to systematically generate a conceptual description, grounded in data, about participants' preferred process of involvement in ASD research.

Participants and Sampling

Purposeful sampling requires the selection of participants with a depth of knowledge and experience related to the phenomenon of interest (Richards & Morse, 2007). Purposeful sampling was used to identify and enrol adults with ASD who were willing to discuss their preferences about research involvement and were able to give indicate consent to participate. Participants were recruited as necessary to capture the variation of key variables (i.e., age, gender, occupation, length of diagnosis, experiences and perspectives of involvement with research) to ensure comprehensiveness of the conceptual description. This theoretical sampling strategy continued until we observed a repetition of findings in the data, thereby achieving data saturation (Morse, 2009).

Recruitment occurred between January 2012 and February 2013, after institutional approval was obtained from the Health Research Ethics Board at the University of Alberta (Edmonton, Canada) and operational approval was obtained from the Glenrose Rehabilitation Hospital (Edmonton, Canada). Potential participants were informed of the study in three ways. First, a familiar service provider informed adults with ASD about the study in person, following group sessions held at the Autism Society of Edmonton and Area (ASEA) offices. ASEA also distributed a flyer about the study via e-mail to participants in the group. Second, clinicians at the Autism Follow-up Clinic and the Autism Research Centre at the Glenrose Rehabilitation Hospital (GRH) informed potential participants about the study. Third, study posters were placed on notice boards at the ASEA and GRH. Adults who were open to receive further information were invited to contact a member of the research team (JJ) to discuss participation and potentially enrol. Posters and information sheets used during recruitment are found in Appendix A. Participation in the clinical programs offered by the main recruitment sources (i.e., the ASEA and GRH) generally required a diagnosis of ASD, which was further confirmed by participant self-report.

Data Collection

Enrolled adults participated in semi-structured individual interviews scheduled at their convenience and held at either the Autism Research Center (GRH) or the Edmonton Clinic Health Academy (University of Alberta) in Edmonton, Canada. At the beginning of each interview, an explanation of the study was provided and informed consent was obtained. Next, demographic data (name, address, telephone number and email address) were obtained from each participant. An interview guide comprising a range of both broad open-ended and specific scenario-based questions allowed participants to share general and specific preferences regarding their possible involvement in ASD research. Probing questions were then used to obtain further explanation or clarification from participants (the interview guide can be found in Appendix B). After the interview was completed, adults were reimbursed for costs incurred in traveling to interview sessions and thanked for their participation in the study. Interviews lasted between 60 and 90 minutes.

Following the interviews, field notes were written, which included a description of the setting (e.g., time of day, interruptions, notable contextual consideration in the interviews) and a running log of methodological decisions made during the interview. Consistent with the grounded theory approach, the interview guide was flexible and was modified throughout the concurrent process of data collection and analysis.

All interviews were digitally recorded and transcribed verbatim into Microsoft Word by a professional transcriptionist. After transcription, identifying information was removed, and a separate electronic file was used to link participant names with study identification numbers. The separate electronic file (de-identified data) and the original transcript were retained separately. Transcripts and field notes were managed using qualitative research computer software *Atlas.ti* 6 (2011, ATLAS.ti Scientific Software Development GmbH; Berlin, Europe). Designed for use in grounded theory studies, the *ATLAS.ti* package allows for basic coding and retrieval of data at the textual level, and more sophisticated analysis at the conceptual level including linking codes to form networks and algorithms (Given, 2008). After the field notes and transcripts had been reviewed, follow-up interviews were conducted with two participants to clarify some of their responses. Following the completion of the study, each study participant was mailed a $25 gift card as an expression of gratitude.

Data Analysis

Consistent with the method of constant comparison, data analysis began upon completion and transcription of the first interview, and continued concurrently with data generation in an ongoing and iterative process (Strauss & Corbin, 1998). In addition, each item of data in transcripts and field notes was compared with every other item of data as it was collected. This approach allowed for the selection of participants to be guided by the emerging analysis and ensured that the conceptual representation developed from data was refined on the basis of additional data obtained from subsequent interviews and analyses (theoretical sampling) (Richards & Morse, 2013). Participants were recruited until saturation was achieved; at this point, concepts were fully developed and no new data contributed to further description, even with the recruitment of additional participants (Bowen, 2008).

Transcripts were checked alongside original recordings in order to ensure accuracy and completeness. Data were subjected to grounded theory tools of open, axial and selective coding, as well as to a repetition of constant comparison and memoing (Strauss & Corbin, 1990). The following steps were employed in this study:

a) Open coding involved analyzing transcripts and field notes line by line and fracturing transcripts into data segments that reflected a particular and unique meaning. These segments, labeled with codes, were compared for similarities and differences and assembled together to form more abstract categories. The justification for assigning particular codes was recorded in memos, as were the properties and dimensions of emerging categories.

b) Axial coding was used to link categories and more abstract concepts to each other by identifying relationships, conditions, and interactions among them. Explanations of the linkages between categories and concepts, and of how they related to the adults' preferences concerning involvement in ASD research, were recorded in memos.

c) Selective coding involved linking categories around a core category, integrating and refining findings, and reviewing, sorting and organizing memos, to form a theoretical construction. During selective coding, decisions were made regarding data saturation and the need for further sampling.

The first two transcripts were independently reviewed and coded by two investigators (JJ and DN) who then met to compare codes. Inter-rater consistency in coding was achieved through discussion and consensus. The remaining six transcripts were coded by one investigator (JJ). Three meetings were held among team members to review and discuss emerging codes, concepts and categories. From this data analysis and team review process, a theoretical model was developed to highlight emergent categories and interconnections. Once all of the transcripts were analyzed, a full conceptual description was derived from the data to highlight the interconnections between categories.

Methodological Rigor

To ensure the integrity of the research process, four aspects of trustworthiness, detailed in Lincoln and Guba's model (1985), were addressed throughout the concurrent data collection and analysis processes.

1. Promoting *credibility* involved ensuring the study's conceptual description represented the participants' preferences. This study included peer-debriefing strategy throughout the study by which the investigation team discussed the emerging themes and personal reactions to the material. These investigators cumulatively bring many years of ASD research, clinical experience, and expertise with qualitative research methods. The team regularly reviewed the study's progress in order to promote credibility.
2. Promoting *transferability* involved using a purposeful and theoretical sampling strategy to identify key informants and satisfy the theoretical needs and comprehensiveness of the conceptual description.
3. We ensured *dependability* by examining whether the study process was consistent over time. Approaches to demonstrating reliability included ensuring a constructive 'fit' between research questions and the grounded theory method (methodological coherence) including the 'fit' of interview guide with Strauss and Corbin's (1990) guide to data analysis. Careful selection of participants through purposeful and then theoretical sampling ensured that an appropriate and sufficient sample was obtained.
4. Finally, *confirmability* was ensured through prolonged engagement including 13 months of data collection and analysis, and the extensive use of memos and consultation with committee members. Techniques of reflexivity and

bracketing (deep reflection and articulation of experiences and perceptions related to the research topic), were used to render explicit any idiosyncratic perspectives and potential biases of the researcher. This approach assisted in identifying, hence mitigating, bias, thus ensuring objectivity and neutrality (Lincoln & Guba, 1985).

Results

Study Participants

The adults with ASD in this study (n=8) lived in Edmonton, Canada with representation of men (n=6) and women (n=2). The adults were between the ages of 19 and 46 years with the majority having been diagnosed for more than two years. Five of the adults were diagnosed at 18 years or older, and four of the adults were employed.

Adults described their preferred process for being involved in developing and implementing ASD research. As part of understanding this process, participants were asked to reflect on the motivations and challenges involved in this participation. In the following sections, findings will be explored, both in terms of contextual concepts related to participating in research and of the core concepts which address specific parameters of the research questions. The concepts depict the preferred process of research involvement for adults with ASD, including the ways in which participants thought that researchers should work with them. Elements in the emergent model and theory are outlined in Table 3-1 and described below.

Contextual Concepts: Motivations for Involvement and Challenges Impeding Involvement in Research

Using research to get information we can use. In addressing the value of participating in developing research, several respondents noted their motivation for being involved. In particular, they noted the usefulness of research knowledge – both as a tool for self-advocacy and as a means of better understanding themselves and their condition. Reflecting on the scope of ASD research, one participant noted that, "*there is a lot, particularly around employment issues and certain housing issues that I feel if I had more information on, I could probably do a better job of advocating for myself.*" This is especially true, he noted, given that "*trying to find organizations that can support me in the ways that I need… is proving to be a real challenge*." Just as important, said another participant, is the potential for gaining other kinds of knowledge: "*To be involved and to have access to the information at the end when it's finished would benefit us because we would learn more of ourselves and of the world.*"

Contributing to research that responds to the needs of adults with ASD. While they recognized and valued personal benefits to their involvement with research, participants also felt that they could help to shape the research agenda. This is crucial, they said, because past ASD research has tended not to focus on areas related to helping adults with ASD overcome the challenges they face. As one participant described, "*autism research doesn't seem to be very responsive to the needs of the actual people with autism.*" Rather, as another participant noted, "*research is focused towards parents, it's focused towards how* *we can stop autism from happening, but less on how we can make life more comfortable or easier or better for the people who have it."*

The perception that research efforts have been diluted over the key issues and priorities of parents instead of adults with ASD, led participants to express feeling like *"just one voice in a large community."* One participant described the complexity of prioritizing research interests:

> *I'm not really sure how to balance the need the parents have of what is the right way of supporting my child at this particular stage, versus adults who are, like, how do I become a member of society that I don't really understand.*

We have our challenges but we also have something to contribute. After identifying the benefits of being involved in research, participants also acknowledged that there would be challenges. More specifically, participants felt that having ASD could make it difficult to participate in research but believed it was still important for them to be involved. As one participant noted:

> *Adults with autism might have difficulty expressing their needs in a day-to-day manner, much less their needs over their lifetime. So I think a lot of [the challenges to being involved in research] would just be inherent in the difficulties that autistic people have.*

Participants recognized that challenges in communication and expression were a significant barrier to participating in research and could impact on the time and resources required.

> *To involve adults would be clunkier, more time-consuming. It would be something that would be more expensive, difficult to organize. So I think, in a way, it complicates things. Although it might be a good idea, it also makes things a little bit more difficult, I think.*

However, a sense of hope and optimism could be found in the insight that these challenges should not prevent people with ASD from participating in research:

> *I don't have the level of communication that would be necessary for that sort of thing. But then I was also thinking probably everybody would think that. So somebody needs to do it, but none of us are suitable, sort of. But yeah, I think, generally, I would be interested, and I'd like to force a lot of other people to be interested, too!*

The value and meaning to be found in the experiences of adults with ASD was seen to outweigh any barriers that existed in engaging them in research, and an individualized approach was favoured: *"To the extent that adults with autism can participate in the process, they should. To the degree that they're competent, and to the degree that they have—that their contributions can be useful"*

Core Concepts: Principles of Involvement

Involve me in the planning. Participants believed they could contribute to the field by helping to shape and plan research studies, but there was variability in perspectives surrounding the process of how and when input could be best sought. Participants felt that research ideas should be generated directly from the breadth of their experiences and life challenges as people with ASD to ensure its immediate relevance:

> *I would say that we want to be involved more in the planning of studies so that we can gain information that will be relevant to us, and possibly, that would happen either through directly participating in the studies that people want to undertake looking at specific issues, for example, employment interviews or housing or those sorts of issues.*

The idea of contributing to the research process at its front end, where research would flow from questions proposed by people with ASD, was echoed by another participant:

> *The best way I could think of would be pretty much right at the start of full-scale study development, with the idea of developing appropriate questions, appropriate understanding of what the situation would be, and using that to advance more detailed research design.*

Others opted for a more strategic approach where they could evaluate questions proposed by researchers so that their input would influence the research that would be pursued:

> *I think probably the most beneficial would be to help refine research topics that are wanting to be looked at by researchers, so that we can gain a better understanding of yes, this as its structured right now will produce beneficial information that we need, or if we looked at these aspects of this topic rather than this, it would produce more beneficial results for us.*

Still others felt that input should best be sought after a plan had been devised by researchers through a carefully chosen focus group for consultation and feedback:

> *Let's say researchers have a few ideas that they think, okay, this is a transition program that could work. Before actually implementing it or conducting research with the junior-high aged students, you might run a focus group with adults saying, if we ran you through a program that offered these things, what would your response be, and then carefully observing reactions to various things.*

Instead of concentrating on driving research ideas, one participant voiced his desire to be involved in the integration and application of insights gleaned at the conclusion of the research process: *"My particular concerns would be not so much about the immediate study itself, but more about how it would—how any useful results or research findings would be sustained, application beyond the immediate study toward the world outside."*

Despite their varied opinions on the timing and nature of their involvement, participants were adamant about the importance of their involvement in proposing research ideas. As one participant expressed, *"I want to be involved just to have input or a vote about where research should be focused."* Another participant reinforced the message that the relevance of research would be improved through the involvement of adults with ASD in planning of studies: "*[our involvement} would make the research more directly applicable to the interests and needs of adults with autism."*

Recognize that adults with ASD and researchers have important roles and both are needed. In devising, planning and implementing research, both the knowledge of the researcher and the experiences of people with ASD were acknowledged as important. A participant described the necessity of both parties in creating a responsive research process:

> *I wouldn’t say that they should necessarily supplant the research role together, because the researcher does have specific skills that the typical adult with autism would not necessarily have. The adults with autism would be able to provide particular experiences from their situations and lived experience to support whatever theoretical knowledge the researcher may already have had.*

In this framework, the necessity of the researcher’s knowledge was emphasized, as was the essential consultative role that can only be played by adult who has lived with ASD. Another participant elaborated on the specific roles that can be played by both parties in optimizing research:

> *The adults with autism would be responsible primarily for helping to refine research content, and the researchers that have the knowledge of how to conduct research studies, how to apply scientific methodology to the issue that the adults identified, would then work with the adults to define a study that could most objectively measure what is most likely a fairly subjective topic.*

Ensure that we feel comfortable approaching researchers, and that they feel comfortable approaching us. Participants expressed a strong desire to interact with researchers. They pointed out, however, that this desire was often frustrated because the research process was inaccessible to them:

> *Right now, I have an understanding that in some circles, research is being conducted into the issues adults are facing, but I don't really know how to gain access to those researchers, how to approach those researchers with the issues that I'm facing. And that, I am finding frustrating, because I know I could contribute in a positive manner to the research; I just don't know how I would gain or go about it.*

One participant favoured an ongoing dialog between researchers and people with ASD to serve as a reciprocal wellspring of research questions and novel insights:

> *I think it would work best if both the researchers and the adults with autism were both able to invite each other to help with things, because then what you will develop is a community where people realize that, okay, over here we've got research that has just told me these strategies work really well for me to help with these issues that I'm having, and at the same time, it's, like, okay, well, I'm running into these issues. Scientifically, how many of my peers are having these issues, how many employers are also experiencing similar issues, and so on.*

This idea was echoed by another participant: *"I think more conversation between researchers and people on the spectrum should take place so that people feel* *engaged in the research process.”* Heightening awareness of already engaged and motivated groups of people with ASD and researchers was seen as critical to ensuring the robust flow of ideas:

> *Researchers need to know that there are autism societies, and then autism societies need to know that there are people all over the place doing all kinds of different research, and if the two groups of people communicated, then everybody would have access to more knowledge and information.*

Discussion

The objectives of this study were threefold: to understand how adults with ASD describe a preferred process of research involvement; to identify how they want to be involved in this process; and to determine how they want the research community to interact and engage them in this process. The preceding section explained the preferred process of involvement for adults with ASD by considering contextual and core concepts formed from the participants’ responses. The section that follows describes the relationships between these categories of concepts and the development of a model to provide a framework for adapting current PR methods to suit the preferences of individuals with ASD.

The preferred process of research involvement for individuals with ASD is represented in an emergent model capturing and illustrating concepts reported by participants (Figure 3-1). A first group, the *contextual* concepts, appear on the left-and right-hand borders of the model, given that they provide the backdrop for the preferred process of involvement. These concepts, which include the motivations of individuals with ASD for wanting to be involved—a desire to gain information they can use, and to contribute to research that responds to the needs of adults with ASD—and the challenges impeding their involvement given that ASD is defined by atypical social interactions and communication. In sum, the contextual concepts suggest that the experiences and understanding of ASD supplied by individuals with the diagnosis are critically important to moving the research agenda forward and in contributing to studies.

The *core* concepts appear in the centre of the matrix. These are concepts that address specific parameters of the research question, including the principles of involving individuals with ASD in the planning of the research, for recognizing the important ways that both the adult with ASD and the researcher contribute to the research process, and for ensuring that individuals with ASD feel comfortable approaching researchers. They are depicted in the diagram in two ways: in a large arrow representing the stages of the research process, and in circular arrows, which represent the partnership between researchers and individuals with ASD. These two ways of co-contributing to the research process require more detailed explanation.

The large arrow, first, captures the key elements of research in which individuals with ASD would like to be involved. They include the steps of planning the study, conducting the study, and determining how findings are beneficial for adults with ASD. The smaller circle on the left of this figure links adults with ASD to the researchers. This represents an ongoing exchange between the two groups involving their collaboration in each of these steps. Participants felt that this process of working together and being integrally involved in the components of research would ultimately lead to the production of research that helps adults with ASD, as depicted on the right side of the model. While participants sought engagement across the stages of research, they also recognized that the goals of the research process will differ between various stakeholders, and that the extent and nature of their involvement may vary relative to the trajectory of the research process (e.g., potential heightened involvement in the conceptual and planning stages and later knowledge translation stage), and depending on the relevance of the research topic to the lives of persons with ASD.

Beyond the focus on the role of individuals with ASD in research, the emergent theory introduced key concepts such as the relationship between adults with ASD and researchers, and their preferred involvement along the course of the study. This theory highlights the importance of the *relationship* between the individuals with ASD and researchers as an important piece of the model. This relationship, at the interface of the research, leads to the production of knowledge, which helps adults face their challenges. However, the end product of the research process extends beyond this relationship. The different perspectives of the researcher and individuals with ASD are thought to be synergistic, ultimately producing research that can be useful to individuals on the spectrum. The ultimate purpose for adults with ASD being involved in research is pragmatic, functional, and goal-oriented, all in the interest of better research. Accordingly, engaging people with ASD in research is about involving them in substantive roles during the planning of the study and in determining how findings are applied.

Adapting Participatory Research (PR) in Current Autism Spectrum Disorder (ASD) Research

The emergent model presented in this study may be used to inform and adapt existing models used to involve individuals with ASD in the development of research. Thus far, only one study (Nicolaidis et al., 2011) has involved adults with ASD in the development of research; it provides an important preliminary opportunity to apply our findings. The study used a CBPR approach, in which researchers, individuals with ASD, and family members, served as equal partners throughout the research process. The collective goal of each partner was to ensure that the research was "respectful, accessible and relevant" (Nicolaidis et al., 2011, p. 145). Individuals on the spectrum were involved in "designing protocols, developing and adapting instruments and consent materials, recruiting participants, collecting and analyzing data, and disseminating findings" (Nicolaidis et al., 2011, p. 145).

While Nicolaidis et al. involved individuals with ASD in every step of the research process, our model suggests that individuals with ASD prefer to be involved in aspects of the research to which they can contribute the most and that are most clearly aligned to their specific goals for being involved. Participants in the current study also recognized that the goals of the research process differ between studies, and that the extent and nature of their involvement may vary relative to the trajectory of the research process, and depending on the relevance of the research topic to the lives of persons with ASD. Participants were keen to contribute to the planning of studies and to determining how the findings of studies would be applied. Thus, as an implication of this research, it is important that individuals with ASD participate in determining how they want to be involved in the process, instead of just being invited to join a prescribed process.

One goal of CBPR is to empower those who have not been empowered by actively engaging them in the research process in order to generate an understanding of the community members' perspectives and needs so as to develop interventions that meet the needs of the community members. Our model suggests that because the ultimate aim of individuals with ASD for being involved in research is to co-produce research that helps them and their peers. Empowerment is not derived solely from being involved in the research process, but rather, from bringing the experiences and understanding of ASD in context to generate ASD research that is ultimately useful to adults with ASD. Therefore, in seeking to empower communities, this model emphasizes the need to identify the ultimate goals of end-users.

Adapting Current PR Approaches in Related Fields

Our model offers insights into the PR approaches that have recently been integrated in studies involving individuals with neurodevelopmental disorders – a more broadly defined group. A review of these studies, as noted above, revealed that the extent of the involvement of co-researchers varied (see Chapter 2). In all but one study, the co-researcher was recruited after the study was funded and the research question and methodology had been selected; as a result, the individual with ASD as co-researcher had less opportunity to inform the study design. In such cases, co-researchers were primarily involved in assisting researchers to gain access to certain populations and carry out tasks determined by study authors. In four of the studies, co-researchers were involved in analyzing data. As concerns the varied PR designs across studies, there were differences in the extent to which co-researchers were involved in the development of the studies, yet consistently, a general lack of clarity as to the role of the co-researcher as a research partner.

Another theme that emerged in the review was the question of what distinguishes a co-researcher's role from that of a research assistant. More specifically, in four of the studies, where the co-researchers were assigned tasks by researchers, the boundary between authentic PR, and merely hiring someone with a disability to be a research assistant, was unclear. While a research assistant may carry out technical aspects of a study, a co-researcher is part of the research team and thus able to assist in framing questions and interpreting results and findings. All but one of the studies placed an emphasis on the co-researcher's involvement in operational tasks (e.g., data collection) rather than in higher order aspects of the research process (e.g., analyzing data), which speaks to a hierarchy within the research team and a narrow view of "participation" (see Chapter 2).

Clearly, then, the preferred process of involvement of individuals with ASD that emerged in this study differed from the roles played by co-researchers in many of the PR studies involving individuals with neurodevelopmental disorders. In several of these projects, researchers tended to view these individuals as assistants, and helping in technical roles. It is interesting that in this study, adults with ASD viewed researchers as having a technical or circumscribed role (in respect to providing methodological expertise) but needing individuals with

ASD to contribute to research questions and studies that were applicable to them. More specifically, participants expressed that a partnership characterized by recognition of the unique skills adults with ASD and researchers contribute to the research process, was required to produce research that benefits adults with ASD.

Study Limitations

The limitations specific to this study are similar to other qualitative approaches. First, in this study we interviewed eight individuals with ASD. Qualitative researchers have recommended sample sizes ranging from 6-30 participants for a grounded theory; as such, the sample size of eight used in this study tends to the lower end. The sample of adults in this study, however, was adequate as sampling continued until categories were saturated. Second, we recruited adults who were willing to discuss their preferences about research involvement and were able to give consent to participate. The range of neurocognitive difference is varied across the spectrum. As such, the implications of this research are not representative of all individuals with an ASD and might be less relevant to individuals with an ASD who are non-verbal or those within the adolescent or childhood demographic, in particular. Third, the depth of participants’ responses varied across interviews. Probing questions were used to obtain further explanation or clarification from participants, and in two cases follow-up interviews were conducted to ensure the completeness of the data.

Conclusion

This study used grounded theory methodology to explore the preferences of individuals with ASD toward research involvement. This process is explained by their motivations for, and challenges impeding, participation in research and by principles of involvement, which ultimately lead to the production of research that is useful to individuals with ASD. The theory generated highlights the importance that adults with ASD place on participating in an ongoing exchange with researchers and being actively involved along the course of a study. This model provides a framework for adapting current PR approaches used to involve individuals with ASD in developing research.

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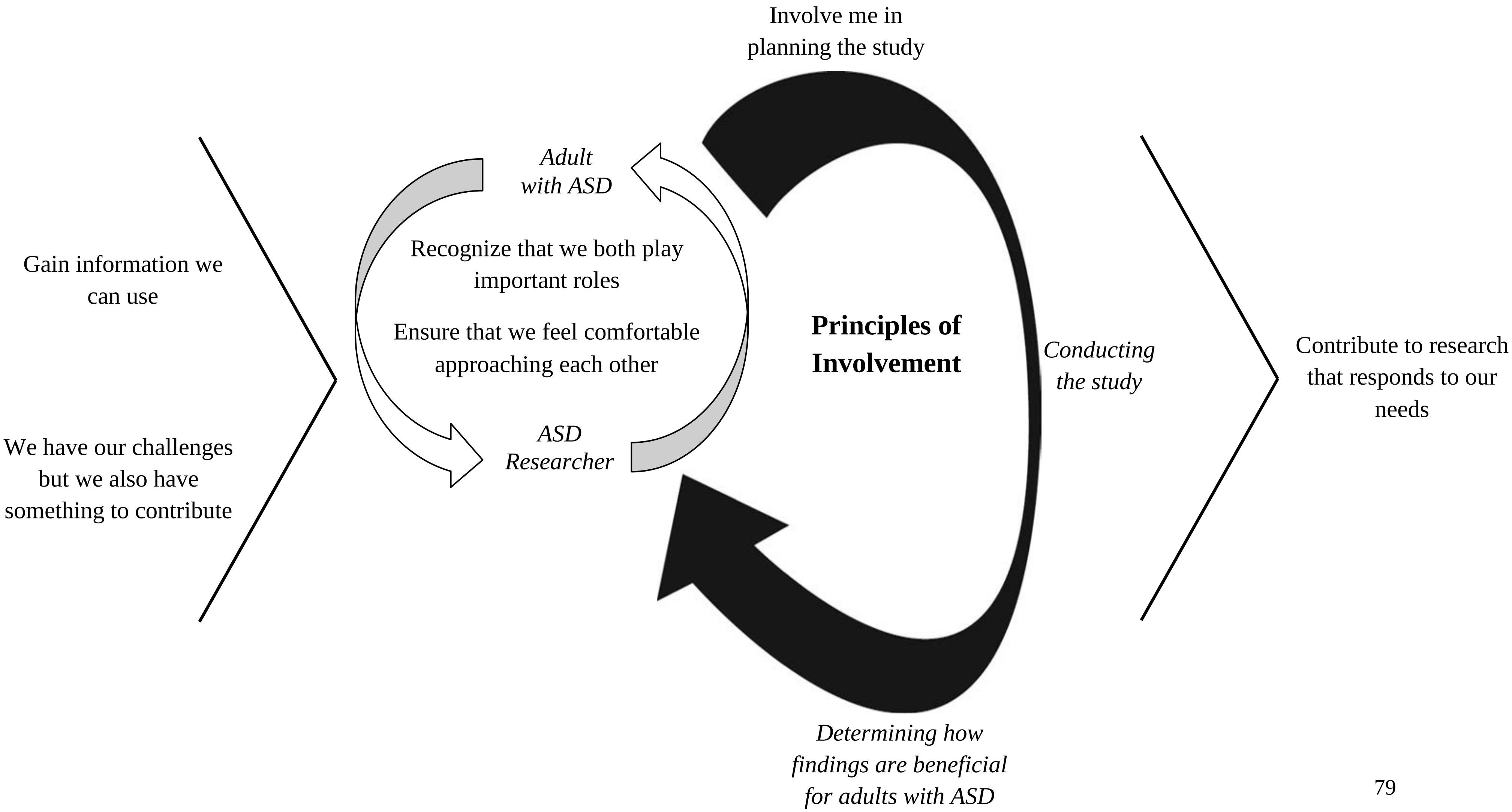
Table 3-1

Grounded Theory Concepts Exploring the Preferred Process of Involvement of Adults with ASD in the Development of Research

Concepts
Contextual Concepts: Motivations for Involvement and Challenges Impeding Involvement in Research
Using research to get information we can use
Contributing to research that responds to the needs of adults with ASD.
We have our challenges but we also have something to contribute.
Core Concepts: Principles of Involvement
Involve me in the planning
Recognize that adults with ASD and researchers have important roles and both are needed
Ensure that we feel comfortable approaching researchers, and that they feel comfortable approaching us.

Figure 3-1

Conceptual Model of the Grounded Theory Explaining the Preferred Process of Research Involvement for Adults with ASD



CHAPTER 4

CONCLUSIONS

Summary of Major Findings

Scoping Review

In the scoping review we sought to identify and characterize published PR partnerships between academic researchers and individuals with ASD or other neurodevelopmental disorders. We also sought to examine the influence of PR partnerships on the research process and reported study outcomes.

Our review identified six studies that described PR partnerships with individuals with neurodevelopmental disorders. Of the six studies, one involved a partnership with individuals with ASD, and five involved individuals with intellectual disabilities. Within these six studies, the PR partnerships employed a variety of methodologies and were implemented in a variety of ways. Our examination of the influence of PR partnerships on the research process and study outcomes revealed two key themes both related to potential limitations in the use and generalizability of findings: (1) variations in PR design across studies, and (2) limitations in the reporting of the depth of the co-researcher's involvement across studies. Concerning the variation in PR design, there were differences in the extent to which co-researchers were involved in the development of the studies as well as a lack of clarity as to the role of the co-researcher as a research partner. The limitations in reporting and evaluation included a lack of clarity on how multiple perspectives were integrated into decision making and a lack of formal evaluation of the partnerships formed with co-researchers and how they were developed.

In general, the influence of PR partnerships on the research process and study outcomes was rarely addressed. The process of interpreting data and the dynamics of working in a group were narrowly reported in the literature. As such, this review calls for future research in this field to more specifically and comprehensively evaluate the contributions of co-researchers with ASD or other neurodevelopmental disorders, and for the creation and testing of tools to assess participatory structures of research engagement for this population. Ultimately, this research may very well contribute to improvement in research outcomes, but that remains to be demonstrated.

Qualitative Study

Given that the scoping review reported mainly on researchers' perspectives on PR partnerships, I sought the perspectives of individuals with ASD using a grounded theory approach. The study had three objectives: to understand (1) how adults with ASD describe a preferred process of research involvement; (2) how they want to be involved in this process; and (3) how they want the research community to interact and engage them in this process.

In an effort to address these questions, participants were asked to reflect on their motivations for involvement -- a desire to gain information they can use, and to contribute to research that responds to the needs of adults with ASD -- and the challenges impeding their involvement. They also identified strategies for involving individuals with ASD in the planning of the research, recognizing the important ways that both the adult with ASD and the researcher can contribute to the research process, and ensuring that individuals with ASD feel comfortable approaching researchers. By examining the relationships between concepts emerging from participant interviews, we developed a model capturing the preferred process of research involvement for individuals with ASD. This model represented core concepts such as the relationship between adults with ASD and researchers, and their preferred involvement over the course of the study. In the preferred process, the different perspectives of researchers and individuals with ASD contribute to synergistic and productive action, all in the interest of better research. The study also revealed that engaging people with ASD in research entails involving them in substantive roles during the planning of the study and determining how findings are applied.

The model we developed in the study also provides a framework for adapting current participatory research methods in ASD, and other neurodevelopmental disorders more broadly, relative to suit the preferences of persons with ASD. An implication of this study is that individuals with ASD should be involved in determining how they want to be involved in the research process, instead of just being invited to join a prescribed process or a CBPR methodology.

Contrasting Study Findings

The scoping review and qualitative study revealed different perspectives between researchers and adults with ASD toward research involvement. Given that only one of the included studies in the scoping review included a co-researcher (an individual with a neurodevelopmental disorder) as a study author, the perspectives on published PR partnerships included in the review, are mainly representative of the academic researchers and not individuals with neurodevelopmental disorders. By contrast, the qualitative study findings in the second study are informed solely by the perspectives of individuals with ASD.

The perspectives captured in these two studies revealed important differences and similarities. In terms of similarity, both groups support the contention that in order to come together, these differences must be understood and negotiated so that the groups ultimately have complimentary expectations of individuals with ASD and researchers. Perspectives differ in two key respects: (a) how participants (individuals with ASD and researchers) view the different research roles, and (b) how they understand the purpose of the research. In the first instance, researchers and adults with ASD maintained different perspectives on the roles that each group should play in research. The scoping review revealed that researchers by and large viewed individuals with neurodevelopmental disorders as providing support around operational aspects of research (such as recruitment, interviewing or dissemination of study findings), working in a sense as research assistants. This was evidenced by the fact that in all but two of the studies, co-researchers were involved after the study was initiated, and therefore individuals with ASD had no input into informing study design. On the other hand, in the qualitative study, adults with ASD viewed researchers in a technical role in that they brought the research tools and methodological expertise required to conduct the study, but needed people with ASD to come up with questions that mattered and to bring an understanding of the real-world context of ASD.

Individuals with ASD and researchers differed in how they viewed the purpose of their relationship. In the studies identified in the scoping review, PR approaches were employed to empower communities that have not been given a voice in research. As such, researchers formed relationships with individuals with ASD as co-researchers in order to empower them through their involvement in research roles and in tasks over the course of the study, primarily during the data collection and analysis stages. In only one study did individuals have the opportunity to inform study methodology, and they participated in identifying the research question in only two studies. However, in the qualitative study, individuals with ASD suggested that empowerment was not achieved just by being involved in research tasks, but by cultivating a partnership with researchers that produces findings that are useful to individuals on the spectrum. As part of this relationship, they preferred to be involved in substantive roles, particularly during the planning of the study and in determining how finding are applied.

These differences revealed in the two studies are instructive because they speak to the differences in worldviews between individuals with ASD and researchers. They also underscore the need for both groups to engage and seek to understand each other’s view of optimal research involvement at the outset in order to develop genuine partnerships of trust and research productivity.

Methodological Limitations

Scoping Review

In conducting the scoping review, we adopted an inclusive selection strategy which requires some justification. The following section explains this in detail, and considers two alternative strategies which might strengthen further efforts.

Initially the focus of the thesis was on PR partnerships with individuals with ASD only. However, given that only one study involving individuals with ASD in such a partnership was located, we decided to broaden the inclusion criteria to include individuals with other neurodevelopmental disorders. In doing so, we were able to draw upon the stronger literature about PR with individuals with intellectual disabilities. On the other hand, a disadvantage of broadening the inclusion criteria was that the findings are less ASD-specific. In spite of this consideration, we feel the limitations identified in the review are relevant to ASD researchers, given that individuals with autism and those with other neurodevelopmental disorders face similar challenges—with language and speech, social relationships, motor skills, behaviour, memory and learning.

Given that our review sought to identify studies where co-researchers were involved in stages during the research process (identifying research questions, informing study methodology, collecting data, analyzing data, and/or disseminating findings), our study was inherently limited to adults as co-researchers. An alternative approach that we could have taken would be to identify studies where children or adolescents with neurodevelopmental disorders were involved in the development of research.

Additionally, given that there is no standard format for how to present information about participatory research, and that many traditional journals do not provide room to discuss all the relevant and important aspects of a PR partnership, it would have been useful to have contacted the authors of the reviewed papers, and the co-researchers who participated in the development of the respective research. These steps likely would have supplemented information and offered detail about the extent of co-researchers involvement and perhaps, offered important commentary about how multiple perspectives were integrated into the overall perspectives.

Qualitative Study

The following section explains two limitations of the grounded theory study. While a relatively small number of participants formed the sample, the number of adults in the study was adequate for data saturation. Moreover, participants represented a fair cross-section on criteria of participants, particularly with regard to age and employment status. Unfortunately, negative cases were not found as all participants were interested in being involved in research. Associated with this concern, we acknowledge that all participants were recruited from a similar context, i.e., through associations with advocacy organizations or through ties with clinical staff.

Finally, the representativeness of participants with regards to the autism spectrum is another potential limitation of the study. Only adults who were willing to discuss their preferences about researcher involvement and who were able to give consent to participate were recruited to participate in the study. Given the range of neurocognitive difference across the autism spectrum, the findings of the study are not representative of all individuals on the spectrum, particularly not individuals who are non-verbal or have significant co-morbid intellectual disabilities.

Implications for Future Research

My graduate research has provided an in-depth understanding of how individuals with ASD and other neurodevelopmental disorders have been involved in PR partnerships to date, and the preferences of adults with ASD regarding research involvement. Findings encourage researchers to consider PR partnerships in order to improve the relevance and applicability of research findings for individuals within this population. Based on the results of my graduate work, the following research areas need to be addressed:

1. *Preferences of ASD researchers towards involving individuals with ASD in the development of research*. While the qualitative study examined the preferences of individuals with ASD, it would be equally valuable to conduct a study examining how ASD researchers prefer individuals with ASD to be involved in research.
2. *Preferences of adolescents with ASD towards research involvement.* Qualitative methodology was useful in gaining understanding of the preferred process of adults with ASD toward research involvement. Another group

facing unique challenges, including the transition between high school and university for example, are adolescents with ASD. As such, I feel it would be valuable to conduct a qualitative study understanding their involvement preferences with regard to research.

3. *Implementing the preferences of individuals with ASD in a research study*. Applying the process which emerged in the grounded theory study would generate additional insights about participatory approaches and would test the model so it could be further refined. This study would also allow researchers to assess whether individuals with ASD and researchers have a similar view on preferred process after having been part of a study that involved such a partnership.
4. *Increasing understanding about the link between involvement of end-users and research outcomes*. In general, the influence of PR partnerships on the research process and study outcomes was rarely addressed. A research study developing metrics for, and ultimately evaluating the outcomes of, the involvement of end-users in the development and implementation of research, would add depth to our understanding of PR approaches in ASD.

Concluding Remarks

My graduate work provides an in-depth understanding of how adults with ASD and other neurodevelopmental disorders have been involved in participatory research partnerships to date, and the preferred process of research involvement for adults with ASD. The first part of my thesis is dedicated to a scoping review which sought to determine what was known in the area (Chapter 2). Findings presented in the review reveal that two key themes both related to potential limitations in the use and generalizability of findings: (1) variations in PR design in studies, and (2) limitations in the reporting about the depth of co-researcher involvement across studies. In Chapter 3, my qualitative study is presented; it provides an understanding of the preferred process of research involvement for adults with ASD. Findings from this study identify a process of research involvement that takes into account motivations for involvement and challenges impeding their involvement, and identifies strategies for involving individuals with ASD in the planning of the research, recognizing the important ways that both the adult with ASD and the researcher contribute to the research process, and ensuring that individuals with ASD feel comfortable approaching researchers. In this study, adults described the importance of being involved in substantive roles throughout the research process and described the importance of relationships with researchers as a means to produce research that is deeply useful to individuals on the spectrum. In conclusion, I feel these two studies will contribute to the overall literature on involving individuals with ASD in participatory research partnerships.

APPENDIX A

Ovid MEDLINE (1946 to Present) Search Strategy

1. Patient Participation/

2. Consumer Participation/

3. Cooperative behavior/

4. Patient advocacy/

5. Self help groups/

6. Community-institutional relations/

7. (self advoca* or ((agenda* or priorit*) adj5 set*)).mp. [mp=title, abstract, original title, name of substance word, subject heading word, protocol supplementary concept, rare disease supplementary concept, unique identifier]

8. or/1-7

9. research.hw. or research*.ti.

10. Researcher-Subject Relations/

11. 9 or 10

12. 8 and 11

13. Community-Based Participatory Research/

14. (participatory action research or participatory research or collaborative inquiry or action research or participatory methodolog*).mp.

15. ((inclusiv* adj5 research) or (emancipatory adj5 research)).mp.

16. 13 or 14 or 15

17. ((patient* or consumer* or participant* or parent or parents or stakeholder* or caregiver* or end user* or client*) adj5 (researcher* or investigator*) adj5 (collaborat* or partner* or consult*)).mp.

18. 12 or 16 or 17

19. exp child development disorders, pervasive/ or developmental disabilities/ or exp learning disorders/ or intellectual disability/ or motor skills disorders/ or stereotypic movement disorder/

20. exp Nervous System Diseases/cn [Congenital]

21. exp Brain Damage, Chronic/

22. exp Language Development Disorders/

23. ((development* or neurodevelopment* or neuromusc* or neuropsychiatr* or intellectual* or language or speech or learning or psychomotor or neuromotor) adj3 (disorder* or delay* or disabil*)).mp.

24. (autis* or asperger* or mental* retard* or down* syndrome* or cerebral palsy or brain injur* or brain damage).mp.

25. or/19-24

26. 18 and 25

APPENDIX B

ETHICS FORMS

PATIENT INFORMATION AND INFORMED CONSENT FORM FOR RESEARCH PARTICIPATION

Research Title: **Exploring consumer involvement in autism research: understanding how adults with autism want to be involved in developing research**

Principal Investigator: Dr. Lonnie Zwaigenbaum, MSc MD FRCSC, (780) 735-8280

Co-Investigators: **Jamil Jivraj, B.Sc.** (780) 984-7526
Dr. David Nicholas
Dr. Amanda Newton

Purpose

In this study, we hope to better understand how adults with autism would like to be involved in the development of autism research.

Background

People with autism offer an important point of view about the way research should be done. We would like to learn more about how adults with autism prefer to be involved in the development of autism research.

Procedures

Survey
We will begin by collecting some information including your age, sex, date of birth, and marital status.

Interviews
You will participate in an interview or small group discussion. During these sessions we will ask you about how you have been involved in autism research, and how you would like to be involved in the planning of future research. These interviews will last about 50 minutes and will be digitally recorded to help us analyze your information. During the interviews, we may take some notes. Most likely, the interviews will take place at the Glenrose Rehabilitation Hospital.

However, if it’s more convenient for you, we can meet at another location (ex. your home, a quiet coffee shop, or an office at the University of Alberta). You will be reimbursed for parking and travel costs.

We will summarize the information you shared with us and invite you to a final session so that you can read the summary of our findings to see if there are any differences or changes needed. The session should last about 45 minutes.

Possible Benefits

This study will help researchers understand the preferences of adults with autism related to being involved in the developing autism research. The long-term goal of this study is to improve the relevance of the research provided to adults with autism.

Possible Risks

By taking part in this study, we do not believe there are any physical risks. However, talking during the sessions may bring up feelings and emotions. Should any issues emerge, and if you desire, we can refer you to appropriate healthcare professionals.

Confidentiality

Confidentiality will be provided to all study participants, as participants will not be identifiable in any published or unpublished results. A master list, accessible only to the principal researcher and co-investigators, will match participant names with identification codes. Study participants will be identified by their codes during all stages of the study. While no names of participants will ever be used in publication, we cannot guarantee that other participants in the group will maintain the confidentiality that is shared. Transcripts and tape recordings will be stored in a locked filing cabinet accessible only to the principal researcher and co-investigators. All the information collected during this study will be kept for a minimum of five years.

Freedom to Withdraw

Your participation in this study is completely voluntary. You may choose not to participate without any negative consequences. You can choose to stop participating in the study at any point in time.

Additional Contacts

If you have any concerns about any part of this study, you may contact the University of Alberta Research Ethics Office at (780) 492-2615. This office has no affiliation with the study investigators.

CONSENT FORM

Title of Project: **Exploring consumer involvement in autism research: Understanding how adults with autism want to be involved in the research process**

Principal Investigator:	Dr. Lonnie Zwaigenbaum	(780) 735-8280
Co-Investigator(s):	Jamil Jivraj	(780) 984-7526
	Dr. David Nicholas	
	Dr. Amanda Newton	

	Yes	**No**
Do you understand that you have been asked to be in a research study?	☐	☐
Have you read and received a copy of the attached Information Sheet?	☐	☐
Do you understand the benefits and risks involved in taking part in this research study?	☐	☐
Have you had an opportunity to ask questions and discuss this study?	☐	☐
Do you understand that you are free to withdraw from the study at any time without having to give a reason and without any negative consequences?	☐	☐
Has the issue of confidentiality been explained to you?	☐	☐
Do you understand who will have access to your information?	☐	☐
Do you agree to be contacted for future research studies?	☐	☐

I agree to take part in this study:

Printed Name of Participant: ____

Signature of Participant: ____

Date: ________________________________

I believe that the person signing this form understands what is involved in the study and voluntarily agrees to participate.

Printed Name of Investigator: _______________________________________

Signature of Investigator or Designee: ___________________________________

Date:__________________________________

ADULTS WITH AUTISM SPECTRUM DISORDER

We want to make autism research more relevant to you

Can you please help us by sharing your experience being involved in autism research?

WHO:	Adults with autism living in Edmonton
WHAT:	A new research study is exploring how adults with autism prefer to be involved in developing autism research.
WHY:	Understanding how adults with autism prefer to be involved in developing autism research may lead to the production of research that is more meaningful and relevant.
WHERE:	Autism Research Center, Glenrose Rehabilitation Hospital Edmonton, AB

QUESTIONS? INTERESTED IN PARTICIPATING?

Please contact Jamil Jivraj (Project Coordinator):
jjivraj@ualberta.ca
780-984-7526

Principal Investigator: Dr. Lonnie Zwaigenbaum
Study Title: Exploring consumer involvement in autism research

APPENDIX C

DATA COLLECTION TOOLS FROM QUALITATIVE STUDY

Interview Guide (Questions and Probes)

Intent: Introduction		
1. Introduction to interviewer and purpose of the interview 2. Review informed consent and ensure copies are with participant 3. Review interview format (expected length, option of declining questions and stopping the interview)		
Intent: To elicit descriptions of processes and experiences		
Open Question	*Follow-up Questions*	*Probes*
I'm interested in your views on autism research - what do you think about it? (How do you feel about autism research?)	What are autism researchers like?	
	Is autism research responsive to the needs of the autism community?	Has autism research made a difference to you? Has autism research done anything for you?
	How could autism research be better?	If you could give advice to researchers to make research more relevant, what would it be and why? How do you think researchers could be most helpful to you?
I would like to know your thoughts on how you think research is developed.	How are research priorities set?	Who determines what the research priorities should be?
	How are research ideas developed?	
	What happens after the research is over?	Tell me more about that.
	Is anything missing from	Do you feel there are

	the research process?	any gaps in how research is developed? Explain.
Would you like to be involved in the development of autism research?	How would you like to involved?	What would make it easier/harder for you to be involved?
		Should parents be involved at the invitation of researchers or should parents approach researchers about being involved?
Researchers in Edmonton want to better understand how to promote language development in toddlers with autism and are interested in working with parents on the development of a research study. Tell me how researchers and parents could work together on developing a research study. *(Backup Scenario)* The school system is struggling with integrating youth with ASD particularly as they enter high school and are trying to generate ideas to develop and evaluate a new program. Tell me how researchers and parents (or adults with autism) could work together on developing a research study.	What would be the most effective way for researchers and parents of individuals with autism to come to the table?	Should parents be involved as individuals or as members of organized groups? Why?
	How could parents of individuals with autism be involved in developing the research project?	What could researchers be responsible for? What could parents be responsible for? What would both groups work together on?
		To what degree should parents be involved?
	What would be different if parents collaborated with researchers in developing the research project rather than researchers working alone?	What are the advantages of parents participate in developing research? What are the disadvantages?
		How important is it for parents to be involved?

AASPIRE is an academic-community partnership composed of health services and disability researchers, autistic self-advocates, health care providers, disability service professionals and family members. They conduct studies about the health care experiences and well-being of autistic adults. In this group, academic and community members serve as equal partners in all phases of research from deciding what to study, designing the study, recruiting participants, collecting and analyzing data, and sharing the findings. I'm interested in knowing what you think about this way of doing research.	What are the strengths of researchers and community members being equal partners throughout the research process? What are the limitations?	
	Is this model the way you see things working ideally?	How might you approach it slightly differently?
	Would like to be involved in developing research in this way?	What would make it easier/harder for you to be involved?
Tell me about your experience participating in autism research.	What was positive/negative about your experiences participating in research?	
	What expectations did you have from the researchers?	Were your expectations met/not met? How did that make you feel?
Intent: Interview Closure		
1. Closing summary and thank-you's 2. Allow time for outstanding participant questions 3. Remind participants of what will happen with the interview content (analysis, confidentiality, use of summary data)		