

NSW Agency for Clinical Innovation - Intellectual Disability Network Research and Development Sub-Committee

Discussion Paper

GUIDELINES FOR RESEARCH

Background

WestWood Spice was engaged by the NSW Agency for Clinical Innovation (ACI) to assist the research and development (R & D) subcommittee of the Intellectual Disability (ID) Network with a number of tasks associated with furthering its objectives for support for research in the area of intellectual disability and health. One of these tasks was assisting the subcommittee with the development of guidelines on research procedures in the area of intellectual disability.

Aim of this paper

This paper has a number of purposes:

- to contextualise the discussion about guidelines within the broader research framework of the ACI;
- to identify other existing guidelines and statements about research relevant to intellectual disability research;
- to provide an overview of key aspects identified by subcommittee members as relevant to the coverage of such guidelines; and
- to provide links to a number of possible resources.

The ACI Framework for Responsible Governance, Management and Conduct of Research

In July 2013, the ACI published its framework “Responsible Governance, Management and Conduct of Research: An ACI Framework” to ensure that all research undertaken by, on behalf of, or in partnership with the ACI meets the highest ethical, scientific, regulatory and professional standards.¹ As the title indicates, the framework addresses the three broad areas of governance, management and conduct of research. The ambit of the ACI framework is health and medical research.

While the interest of the subcommittee in the development of guidelines on research procedures in the area of intellectual disability may be broader than research which is linked directly to the ACI, any guidelines which are developed could be presented in the context of the ACI framework. This context could be seen to operate at two levels:

- a. At the global level of meeting the common purpose of ensuring appropriate ethical, scientific, regulatory and professional standards in any research which is undertaken; and
- b. At the specific level of direct linkage to individual framework items. In particular, item 3.5 Consumer engagement.

¹ Responsible Governance, Management and Conduct of Research- A Framework p i.

Item 3.5 Consumer engagement is as follows:

“The ACI believes that consumer engagement in research strategy is key to ensuring its relevance and value. As such, the ACI strongly encourages meaningful engagement of consumers in the identification of research questions, development of methods and dissemination of findings in all research it supports.”²

Questions for R & D subcommittee:

1. *What should be the relationship of the proposed guidelines to the ACI research framework?*
2. *Who will use the guidelines?*
3. *Is the focus health and medical research?*

Existing guidelines/ research statements

Existing guidelines which have been identified and which are discussed in this paper are:

1. The International Association for the Scientific Study of Intellectual Disabilities (IASSID)’s Ethical Guidelines for International, Multicentre Research Involving People with Intellectual Disabilities (2004)
2. The Edinburgh Principles and accompanying guidelines (IASSID 2001)
3. Research- Joint position statement of AAIDD and the Arc (2010)
4. Disability Inclusive Research principles (Disability Inclusive Research Collaboration 2012)
5. 2013 Geneva Declaration on Person-centred Health Research

IASSID’s Ethical Guidelines for International, Multicentre Research Involving People with Intellectual Disabilities

This 2004 statement, while prompted in the main by the need for clarity around standards which should apply where research involving people with an intellectual disability crosses national boundaries and involves participants in developing countries, nevertheless provides a comprehensive coverage of issues relevant to the conduct of all research which involves people with intellectual disabilities:

“The statement draws on internationally recognized documents outlining the ethical considerations involved in human research activities. It interprets these documents in the light of the particular needs and interests of people with intellectual disabilities and incorporates international consultation involving researchers from a variety of disciplines.”

The statement makes recommendations about ethics review, research design, consent and the conduct of research. It supports the development of partnerships between researchers and people with intellectual disability and their families, advocates and local communities in developing research projects. It considers that strategies to communicate research findings to participants and their communities are important as well as access to the benefits of the research findings.

² P9 Responsible Governance, Management and Conduct of Research - A Framework

A further concern included in the statement is the wish “to ensure that the needs and priorities of people with intellectual disabilities throughout the world are represented in all research activities designed to advance the health and well-being of the general community.”

The statement recognises the power imbalance that can exist between researchers and participants and calls upon researchers and ethics committees to “work in partnership with people with disabilities, their families and advocates in the development of research goals, questions, strategies, methodologies and information dissemination.” A number of recommendations are made to guide independent ethics decision-making review processes. Approval should only be given where the ethics committee is satisfied that:

- a. *“The proposed research is in the interests of people with intellectual disabilities;*
- b. *The methods proposed are scientifically sound and are both culturally appropriate and legal in the communities in which the research is proposed to be conducted; and*
- c. *Those who are to conduct the research are competent to do so and/or will be supervised by appropriate specialists.”*

While many of the research design consideration recommendations reflect the international focus of the guidelines, common recommendations include ensuring that benefits to research participants are sustainable, that there is post-trial access to any effective treatments and that methods are the least intrusive and involve minimum risk to participants.

With respect to consent, the statement recognises the sometimes difficulty of “informed consent” and proposes the introduction of “process consent” into the research protocol. Other key aspects are:

- assumed capacity to consent for adults, unless established otherwise;
- use of oral, witnessed consent where person is unable to read and sign a consent form;
- caution with the use of a “person responsible” such as spouse, parent, legal guardian etc to provide consent; and
- provision of participant information materials in accessible ways such as visual cues and practical demonstrations of research procedures.

Finally, regarding the conduct of research, recommendations cover aspects such as monitoring and complaints provisions, security and privacy of data, publication and dissemination of findings. This should include access by participants to the findings for example through post-project information sessions and plain language summaries of findings.

Question for R & D subcommittee:

4. *How should the proposed guidelines relate to the 2004 IASSMD guidelines?*
5. *Does the existence of the IASSMD guidelines change the need for separate guidelines?*

The Edinburgh Principles and accompanying guidelines (IASSID 2001)

This document is principally about people with intellectual disabilities who also acquire Alzheimer disease or related dementias and the provision of support to these individuals. However, amongst its four-point approach is the topic of promoting relevant research.

In addition to listing a range of areas³ where research is needed, the document suggests that the research agenda needs to include questions on how to measure and research these issues and the ethical and consent challenges faced by researchers “*such as ‘legal prohibition’ of participation in research, the use of protocols for consent and ethical questions raised around participation in medication research*”.

Questions for R & D subcommittee:

6. *How/ should the proposed guidelines cover areas of intellectual disability and other conditions?*
7. *Does medication research require specific attention?*

Research- Joint position statement of the American Association of Intellectual and Developmental Disabilities (AAIDD) and the Arc (2010)

In the context of recognition that government and private funding is insufficient to support the broad research agenda that includes issues most important to people with intellectual disabilities and their families, the position statement acknowledges the historical lack of input of people with disabilities and their families into research decisions. This includes design, methodology, dissemination, use and evaluation of research.

The solutions proposed in the statement include the following:

- *Advocacy, service provider and professional organisations, government agencies, the research community, and people with intellectual and/or developmental disabilities and their families must work together in defining, evaluating, and promoting a research agenda;*
- *Results of research must be available in multiple formats, easily accessible and understandable for a wide audience, including people with intellectual and/or developmental disabilities and their families;*
- *Stringent scientific and ethical standards must be enforced to ensure efficient and effective use of limited research funds and to prevent exploitation or harm of people with intellectual and/or developmental disabilities and members of their families; and*
- *For all basic and applied research involving persons with intellectual and/or developmental disabilities:*
 - *Specific procedures must be implemented to ensure their full voluntary, informed, initial, and ongoing agreement to participate;*
 - *All research must be conducted by qualified researchers, in adequately monitored settings and reviewed for potential risk and benefit by qualified, competent scientific review boards;*

³ Research areas listed included evaluation of different models of care, conflicts between philosophies of care in ID services and dementia services, health needs, effects on dementia of a range of issues (AOD, HRT, nutrition), patterns of disease variation, system factors, family caregiving, social policy & financing questions.

- *No research may be conducted exclusively on persons with intellectual and/or developmental disabilities unless there is reasonable likelihood that the treatment would address unique intellectual and/or developmental disabilities medical issues or apply differentially to them; and*
- *Persons with intellectual and/or developmental disabilities should not be excluded from research that might benefit them as members of the general population.*

Question for R & D subcommittee:

8. *Should the proposed guidelines incorporate any of the AAIDD Research statement?*

Disability Inclusive Research Principles (Disability Inclusive Research Collaboration 2012)⁴

The Disability Inclusive Research Collaboration⁵ developed the following set of principles to guide disability inclusive research. They were initially used as a quality statement to guide the contribution of speakers, presenters and participants at their 2012 conference (Research Rights: Disability Inclusion Change, 13-14 June 2012 at the University of Sydney):

- i. **Research that is informed by and/or led by people with disability** -The need for research, and its design must be identified and led by people with disability.
- ii. **Ownership** -The research process, its design, management, implementation and findings must be owned by people with disability and their representative organisations.
- iii. **Inclusive and participatory** -The research process, and its methodologies, must ensure that people with disability, about whom and for whom the research is designed, play a central role as researchers and as research participants; and the voice of people with disability is validated as data.
- iv. **Co-presenting**- People with disability must be provided with opportunities to present research findings.
- v. **Materials that are accessible**- Information about the research process, research tools, and research reports, must be provided in ways and in formats that are accessible.
- vi. **A range of types of activities**- Adjustment must be made to the design of research to render research appropriate to the participants and accommodate a variety of approaches (research design reflects the diversity of potential research participants). Good research design must emphasise the need for a variety of approaches to ensure that a diversity of views are researched.

⁴ <http://www.cdds.med.usyd.edu.au/disability-inclusive-research-principles>

⁵ The collaboration includes organisations of and for people with disability, and universities and their research centres, including NSW Council on Intellectual Disability, People with Disability Australia Inc., Women with Disabilities Australia, the Australian Federation of Disability Organisations, People with Disabilities Western Australia, the Social Policy Research Centre at the University of NSW, Griffith University and the University of Sydney through the Centre for Disability Studies.

- vii. **Research that transfers through to real life**- Research by and with people with disability must provide tangible benefits to individuals and the constituency of people with disability, and work toward greater inclusion of people with disability in the community.
- viii. **Re-defining what research is** -Inclusive disability research is part of the universal research endeavour and as such must contribute to ongoing discussions about the role and form of research in general.
- ix. **The right people asking the right questions and getting the right answers"**- Inclusive disability research must be careful to ensure that research questions are relevant and important to people with disability (determined/informed by them), and that answers are sought from the correct sources using the best inclusive methods (identify "right people").
- x. **Consent** -Researchers must apply processes of ethics approval that ensure that people with disability are included in the research as willing and supportive participants.

There are a number of approaches to inclusive research and these can be seen to operate according to the level of participation and role played by individuals with an intellectual disability. A recent presentation by Prof Christine Bigby⁶ categorised these as:

- Advisory
- Collaborative
- Leading and Controlling

The guidelines reproduced above would appear to relate most closely to the “Leading and Controlling” variant of inclusive research.

Question for R & D subcommittee:

9. *How should the proposed guidelines recognise and incorporate inclusive research principles?*

2013 Geneva Declaration on Person-centred Health Research⁷

This declaration emerged from the 6th Geneva conference on Person-centred Medicine (PCM) and the International College of Person-centred Medicine (ICPCM). It expressed the need for both more research in person-centred medicine and for making general health research more person-centred. It describes PCM as encompassing “a wide range of concepts, tasks, technologies and practices which aim to place the whole person in context at the centre and as the goal of clinical practice and public health. While written from the perspective of the whole population, the declaration includes recognition of the following priority: *“studying scientifically the complexity of health, including illness and disability as well as functioning, resilience, resources and wellbeing, plus experiences of health and contributors to health.”*

⁶ See Conceptualising Inclusive Research with People with Intellectual Disability: Paradigm or Method at <http://cds.med.usyd.edu.au/component/search/?searchword=inclusive%20research&searchphrase=all&start=20>

⁷ http://www.imh.org.rs/wp-content/uploads/2013/09/02_2013_Geneva_Declaration_on_PCHR.pdf

Question for R & D subcommittee:

10. Does PCM have a place in the proposed guidelines?

Coverage of the guidelines

Discussions with subcommittee members suggested the following areas as those which have potential resonance in the context of thinking about guidelines to cover research activity about and involving people who have an intellectual disability. These themes are also reflected in the various guidelines and statements discussed in the previous section of this paper.

1. Issues of consent
2. Inclusive research
3. Under-researched groups
 - a. People with more severe levels of intellectual disability
 - b. People with mental health or challenging behaviour
 - c. People with communication challenges

Each of these three issues are discussed separately below, but there is considerable inter-relationship between them, for example, the challenges of securing informed consent acting as a barrier to the inclusion of individuals with less cognitive capacity.

Consent

Ethics committees are used amongst other things, to review issues of consent to participate in research and participant information materials.

The ACI framework requires that before any ACI research commences, that review and approval has been obtained from an appropriate Human Research Ethics Committee (HREC).

The September 2006 issue of the *Journal of Intellectual & Developmental Disability*⁸ included a special “Opinions and Perspectives” section on people with intellectual disabilities as participants in research and the difficulties of informed consent and ethics review. One of the themes of the discussion was the tendency of ethics committees to make “conservative” decisions about research involving persons with intellectual disability. There were mixed views about the attribution of this-concern about protecting vulnerable groups from exploitation and harm, the impact of regulations imposed by research funding bodies or inexperience on the part of some committees in judging risks of harm/potential benefits of research.

McVilly and Dalton (2006) note that in the area of medical research and treatment, there are clearly delineated rules and practices governing proxy consent, but that in the field of social research, surrogate consent is a “persistent and perplexing problem.”⁹

Since the time of the discussion, the United Nations Convention on the Rights of Persons with Disabilities has made explicit the fundamental rights of people with disability to make decisions in all walks of life, including equality of legal rights and access to support to properly exercise those legal

⁸ Vol 31(3)

⁹ P187 Commentary on Iacono (2006) Ethical challenges and complexities of including people with intellectual disability as participants in research” *J IDD* Sept 2006, 31(3) 186-188.

rights¹⁰ (Article 12- Equal recognition before the law). There is an increasing interest in supported decision-making as a more empowering alternative to traditional approaches of guardianship and substitute decision-making.

The NHMRC National Statement on Ethical Conduct in Human Research, 2007 (updated May 2013) contains a specific chapter (Chapter 4.5)¹¹ on People with a Cognitive impairment, an intellectual disability or a mental illness. Guidelines cover research merit and integrity, justice, beneficence and respect. Guidelines under respect deal with issues of consent. However, these do not appear to fully embrace the concept of supported decision-making.

Iacono (2006) explains that consent obtained through a supported decision-making process is the result of a) a presumption of competence and b) allowing significant people in an individual's life to provide support to the person in evaluating the risks and benefits of research participation.¹²

Question for R & D subcommittee:

11. *What should the proposed guidelines say about ethics approvals?*
12. *What relationship should the guidelines have to NHMRC guidelines in Ch 4.5?*
13. *What is the place of "Easy English" participant information sheets?*
14. *What other accessible information provisions are needed?*

Inclusive research

There is considerable interest in securing greater involvement of people with an intellectual disability in research at a number of levels, including:

- As participants and respondents to research questions;
- As advisors, planners and conductors of research.

As detailed earlier in this paper, there is now a specific set of disability inclusive research principles against which the level of inclusiveness of research activity can be measured.

Under-researched groups

McVilly & Dalton (2006) highlight some of the consequences of exclusion of people from research because of a (presumed) lack of decisional capacity. They are:

- problems of generalisability of research findings; and
- further devaluing the person as someone not able to contribute to scientific knowledge.

¹⁰ States Parties shall take appropriate measures to provide access by persons with disabilities to the support they may require in exercising their legal capacity.(Article 12 No.3 UNCRPD).

¹¹ p 65-66.

¹² P177 Iacono, T. Ethical challenges and complexities of including people with intellectual disability as participants in research" J IDD Sept 2006, 31(3) p173-179.

Particular concern was expressed by subcommittee members who were interviewed about the exclusion of particular people from research. This would include people with more severe levels of intellectual disability, those who also have mental health or challenging behaviour and people with communication difficulties.

Considerations of issues such as consent processes and the provision of information at all stages of the research from participant information through to information about research results are relevant here.

Question for R & D subcommittee:

15. What recommendations/guidelines should the subcommittee make to encourage and support greater participation in research by underrepresented groups?

Resource materials

<p>AAIDD and the Arc (2010) Research - Joint position statement of AAIDD and the Arc http://aidd.org/news-policy/policy/position-statements/research#.Ug6iCU9-8dU</p>
<p>ACI Responsible Governance, Management and Conduct of Research An ACI Framework Version 1.3, July 2013 www.act.health.nsw.gov.au</p>
<p>Dalton, A.J., & McVilly, K. (2004) Ethical Guidelines for International, Multicentre Research Involving People with Intellectual Disabilities IASSID Journal of Policy and Practice in Intellectual disabilities Volume 1 Number 2 pp 57-70 June 2004 http://iassid.org/pdf/ethics-guidelines.pdf</p>
<p>Disability Inclusive Research Collaboration (2012) Disability Inclusive Research Principles http://www.cdds.med.usyd.edu.au/disability-inclusive-research-principles</p>
<p>2013 Geneva Declaration on Person-centred health Research http://www.imh.org.rs/wp-content/uploads/2013/09/02_2013_Geneva_Declaration_on_PCHR.pdf</p>
<p>Heller, T., Nelis, T., Collins, S. & Pederson, E.L. (2011) Getting involved in Research and Training: A Guide for Persons with Intellectual Disabilities University of Illinois at Chicago Department of Disability and Human Development http://www.rtcadd.org/resources/Resources/Publications/Getting-Involved-in-Research-2012.pdf Plain English guide for a person with a disability to explain how to participate in research activities (Not Easy English) - includes advice about informed consent.</p>
<p>Meltzer, A. & Fisher, K.R. Including people with disability in SPRC research (2013) SPRC Newsletter article Number 112 https://www.sprc.unsw.edu.au/research/publications/newsletter/including-people-with-disability-in-sprc-research/?utm_source=UNSW+Social+Policy+Research+Centre&utm_campaign=779259847c-UNSW+SPRC+News+Edition+111&utm_medium=email&utm_term=0_6aef7b76ad-779259847c-75620337 Describes a number of inclusive methods being used by SPRC</p>
<p>National Statement on Ethical Conduct in Human Research 2007 (Updated May 2013). The National Health and Medical Research Council, the Australian Research Council and the Australian Vice-Chancellors' Committee. Commonwealth of Australia, Canberra. http://www.nhmrc.gov.au/files_nhmrc/publications/attachments/e72_national_statement_130813.pdf</p>
<p>Wilkinson, H. & Janicki, M.P. The Edinburgh Principles with accompanying guidelines and recommendations IASSID (2001) https://iassid.org/pdf/edinburg-principles.pdf</p>

Abbreviations

The following abbreviations have been used in the discussion paper.

ABBREVIATION	MEANING
AAIDD	American Association on Intellectual and Developmental Disabilities
ACI	Agency for Clinical Innovation
HREC	Human Research Ethics Committee
IASSID	International Association for the Scientific Study of Intellectual Disabilities
ICPCM	International College of Person-centred Medicine
ID	Intellectual disability
NHMRC	National Health and Medical Research Council
NSW	New South Wales
PCM	Person-centred medicine
R & D	Research and development
WWS	WestWood Spice