Policy Briefing: Implementing empirical ethics and rights - IDEAS for ensuring disability equity in research

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Introduction

In this policy briefing, we illustrate the development and potential of tools developed from commissioned public health research project where empirical ethics was combined with human rights to elucidate a critical framework for conducting and evaluating Randomised Controlled Trials (RCTs) in public health and disability.[1] Public health research often struggles with conceptualising the different consequences of having lifelong, acquired or fluctuating conditions, as well as integrating how social disadvantage, gender, ethnicity, sexual orientation and life-course mediate those everyday experiences, including experiences of health. However, the potential impact of public health interventions on disadvantaged populations, in terms of ensuring health and flourishing *with* disability is considerable, hence the need for more inclusive and better informed research. Thus, England's major health research funding agency, the National Institute for Health Research - Public Health Research (NIHR-PHR), issued a call for a review of the implications of models and theories of disability for public health research. Our team secured the contract for this scoping review, with inputs from panels of politically and socially active disabled people and their organisations, as well as from public health professionals.[2]

In the United Kingdom (UK), evaluative research has focused on the retrospective effects of policy on health and impact on health inequalities.[3,4] This research, by contrast, concentrated on how the evidence for public health and disability policy was being constructed. RCTs of interventions are viewed as the gold standard of scientific research and evidence informed medicine and are increasingly important in terms of influencing policy decisions. Despite this, there is limited critical theoretical and practical research about how exactly disability and people with disabilities are being integrated in RCTs, their designs and the establishment of research priorities by researchers and commissioners. Previous studies have mostly recorded exclusions of population groups most affected by health inequalities, such as people with intellectual disabilities.[5,6,7] While public health interventions are often presumed to apply to an entire population, exclusions illustrate this is not the case.

Our study began with a scoping of disability theories and models of disability. We identified that human rights models[1] had the most in common with ecological models of public health[8] and public health policy focusing on the social determinants of health.[9] We found that human rights models focused on issues of equity and individual rights within societies, while also advocating legislative, social and structural policy changes for greater environmental inclusion. We then undertook a systematic review of public health intervention research found in the Cochrane Library of Systematic Reviews. We examined 30 research reviews of generic public health interventions and 30 reviews of specific public health interventions focusing on persons with disabilities. We found that disability mainstreaming was not a part of many generic RCTs and much of the specific evaluations focusing on disability were not disability sensitive or inclusive. Of further significance were the ethical and empirical gaps within how public health RCTs were being constructed and evaluated. We found several lacunas, from the exclusion of people and children with various forms of disabilities, inappropriate use of outcome measures, to the total lack of evaluation of disability theory and integration into public health.[1] We thus wanted to find a way in which human rights paradigms could be used to build an ethical bridge to

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improve the inclusivity of public health research, making it relevant to disability experiences across the life-course and more empirically robust.

Public Health, Disability and RCTs

In recent years there has been a renewed interest in reconceptualising the relationship between public health and disability [10, 11, 12] In industrialised countries like the UK, this has been due to technological and medical innovations now enabling people with complex disabilities, diseases and conditions to live longer, the biological and socio-cultural discovery of new forms of disability that can be identified early, and an aging population acquiring disabilities. This has required a public health shift from traditional focus on prevention of disability or its conceptualisation as minority issue, to the promotion of healthy lives and quality of life across the life course.[10] Such a shift also includes an understanding of social participation and inclusion of persons with disabilities, as well as ensuring an identification of environmental barriers to health.[13,14]

At the same time, the public health and disability context linked to health inequalities and social gradients linked to ill-health are still deeply entrenched, with differences in mortality and morbidity between the poorer North and richer South of England.[3] Countries, like England, have also been impacted by the political and social effects of the 2008-2009 economic crisis. The government policy responses in terms of neoliberal austerity measures and social welfare cuts have led to increases of ill-health.[4] This has meant that health inequalities affecting persons with disabilities have been increasing, affecting people with intellectual disabilities and mental health conditions in particular. For instance, Heslop et al.,[15] in their confidential inquiry into premature deaths of people with learning difficulties in the UK, found that men with intellectual disabilities die 13 years earlier and women 20 years earlier than the general population.

The relationship of public health and disability, despite obvious synergies in terms of a paradigm shift towards ensuring diversity across the life-course and a focus on health inequalities[10,2] has always been ethically fraught. The historical association with eugenics in terms of disability prevention, surveillance and incarceration, as well as the complex relationship between the new genetics - and its association with 'soft eugenics' - and the rights of disabled people are often mentioned.[16, 17,18] However, people with disabilities voice more complex and nuanced points about involvement in research, especially noting that they had a 'right' to set agendas and be involved if it was going to lead to medical innovations; more social justice; or improved quality of life.[1,2]

Within public health and disability research, the ethical inclusion and role of persons with disabilities in RCTs has only recently been given attention.[6, 19, 20, 21] Generally speaking, ethical conceptions of 'disability' in public health interventions have tended to valorise non-inclusion, for example, in institutional ethical review boards or governance structures.[22] Most trial-based studies of the general population also often apply various forms of exclusion, for example, by discounting participants with mental health issues or intellectual disabilities.[23] In principle, within RCTs, nobody is excluded per se. However, if no culturally and disability sensitive arrangements are made to understand what inclusion means for differing minority groups, exclusions are offered, become ethical routines or populations are viewed as hard to reach because they do not engage. Asking marginalised groups and in particular, those most affected by health inequalities, such as people with intellectual disabilities, if and how they want to be involved in public health randomised controlled trials is also a fairly recent development.[23,25] While research should be inclusive, research is now conceptualising how even in recruitment and development of an RCT, people's experiences should be captured in an ethically sensitive and socio-culturally meaningful way.

Amongst socially and politically active people with disabilities,[2] there is an understanding of the importance of public health and willingness to be involved in intervention research. However, there are certain qualifications such as ensuring principles of public and patient involvement (PPI) and

inclusion of theories and models of disability to guide research, such as a more social model of human rights.[1] However, some of the most striking findings of our research were the disillusionment with ethics processes and procedures, as well as a lack of trust in not only ethics and human rights but also in research to ensure equity.[1] In addressing these concerns, we developed a theoretical and practical way in which we could combine empirical ethics with human rights to guide and evaluate interventions.

Combining Empirical Ethics with Human Rights

Public health interventions, despite their importance to scientific innovation and influence on national and international policy, have been neglected by bioethics. In medical ethics, the 'empirical- ethical turn'[26, 27] is well known for combining social science empirical research with normative ethics. Yet, according to Salloch et al.[28] empirical-ethical research seems to slot mainly into two different categories, those that offer conceptual accounts and those that use social science empirical methods to understand ethical issues in differing social contexts. We wanted to try and find a third way that was more practically orientated in developing an evaluative tool based on empirical ethics. This would aid researchers to evaluate ethical decision-making, thus uncovering how research as a social practice involving ethics is constructed. We decided to focus on conceptual accounts of how moral decisions have been made within research design, but also in terms of social impact on inequalities after research has occurred. We wanted a way in which we could evaluate the 'trustworthiness'[29] of an intervention which would allow us to develop an empirical ethical tool or guide that would explicitly link ethics to better empirical design but also allow innovation.[30]

Ethical and empirical design could thereby become mutually reinforcing, whereby ethical design leads to empirically robust public health research that addresses disabling health inequalities. A way in which we thought we could do this would be to link empirical-ethical design to international human rights conventions. Historically human rights were not always aligned to disability rights, but influenced by Feldman et al.,[25] we used the United Nations Convention on the Rights of Persons with Disabilities (CRPD)[31] to develop an ethical tool that could help to make public health research more inclusive of disability, and we argue, thus empirically stronger too. The CRPD is an international human rights treaty and innovative legal instrument that also enshrines protected characteristics, such as gender, but assigns disabled people 'rights' which governments and public bodies have duties to uphold.[31] In particular, we felt that Article 3 was inclusive of ethical principles relevant to public health research and committed to the duty of equality, while also emphasising life-course, sexuality, intersectionality and gender. Article 3 of the CRPD specifically notes the following:

> "(a) Respect for inherent dignity, individual autonomy including the freedom to make one's own choices, and independence of persons; (b) Non-discrimination; (c) Full and effective participation and inclusion in society; (d) Respect for difference and acceptance of persons with disabilities as part of human diversity and humanity; (e) Equality of opportunity; (f) Accessibility; (g) Equality between men and women; and (h) Respect for the evolving capacities of children with disabilities and respect for the right of children with disabilities to preserve their identities"[31]

There are methodological issues with implementation of rights-based approaches without conceptualising how rights link to the empirical robustness of RCT design. One way of ensuring empirical robustness but enforcing ethical design would be to link human rights frameworks to those guidelines linked to RCT evaluations such as CONSORT,[32] PRECIS,[33] or RE-AIM.[34] For example, we found that we could practically link five general principles of Article 3 of the CRPD (intersectionality, inclusion, accessibility, dignity and equity) to RE-AIM which evaluates five dimensions of a public health interventions (reach, efficacy, adoption, implementation, and maintenance) for empirical robustness.[1,34] We illustrate this below.

Figure 1: **IDEAS for RE-AIM**

"The **Reach** of an intervention or its representativeness is linked to *Intersectionality* and the kinds of population groups that are typically recruited or neglected in research despite experiencing health inequalities linked to disability; such as women, children, LGTBQ+ people and people from ethnic minority backgrounds or lower-socioeconomic groups. These populations are typically (and stereotypically) framed as 'vulnerable' or 'hard to reach'. Intersectionality should also connect with the diverse nature of the disabling experience and a range of different impairments.

The **Effectiveness** of an intervention is linked to primary and secondary outcome measures and how they impact on who becomes included (or excluded). It also explains and describes how participation in research is viewed and measured. *Inclusion* is important in ensuring the diversity of the disabling experience is captured, which in turn impacts on the use of primary and secondary outcome measures. It also raises questions about how inclusion is defined, negotiated, and measured; and the extent to which participation is an important consideration.

The **Adoption** of the intervention is linked to the setting of the intervention and how well the setting can be accessed. If a design is universal and accessible, it is more likely to be adopted. In this way, *Accessibility* is linked to mainstream adoption and uptake.

The **Implementation** of an intervention or how it is adapted to a particular setting, while generally linked to cost-effectiveness, can also become linked to the *dignity* of participants. This includes participatory design methods, ensuring ethics and consent and ensuring enabling environment. Implementation, therefore, should not be (dis)ableist and modifications need to take account of the potential consequences of disability.

The **Maintenance** of an intervention or its uptake and adaption to real-world settings is linked to issues of equity. *Equity* would address the extent the intervention addresses the social gap, gradient or disadvantage linked to disability."[1]

We then adapted the framework to a simple and accessible guide that researchers as well as people with disabilities could use.

Using IDEAS to guide, evaluate and innovate.

We developed a simplified version of RE-AIM[34] incorporating the CRPD[31] which could be adapted by researchers to quantify ethical scores or develop a critical appraisal tool. The enshrinement of the CRPD[31] in research can in this way, deliver a capacity-building tool by functioning as a link between public health and socially informed understandings of disability. Ensuring the tool also functions as an evaluative guide means that it becomes adaptable to the needs of both interventions to be empirically robust but also stakeholders in assurances of ethical inclusion and social justice. We called the tool IDEAS. See Figure 2.

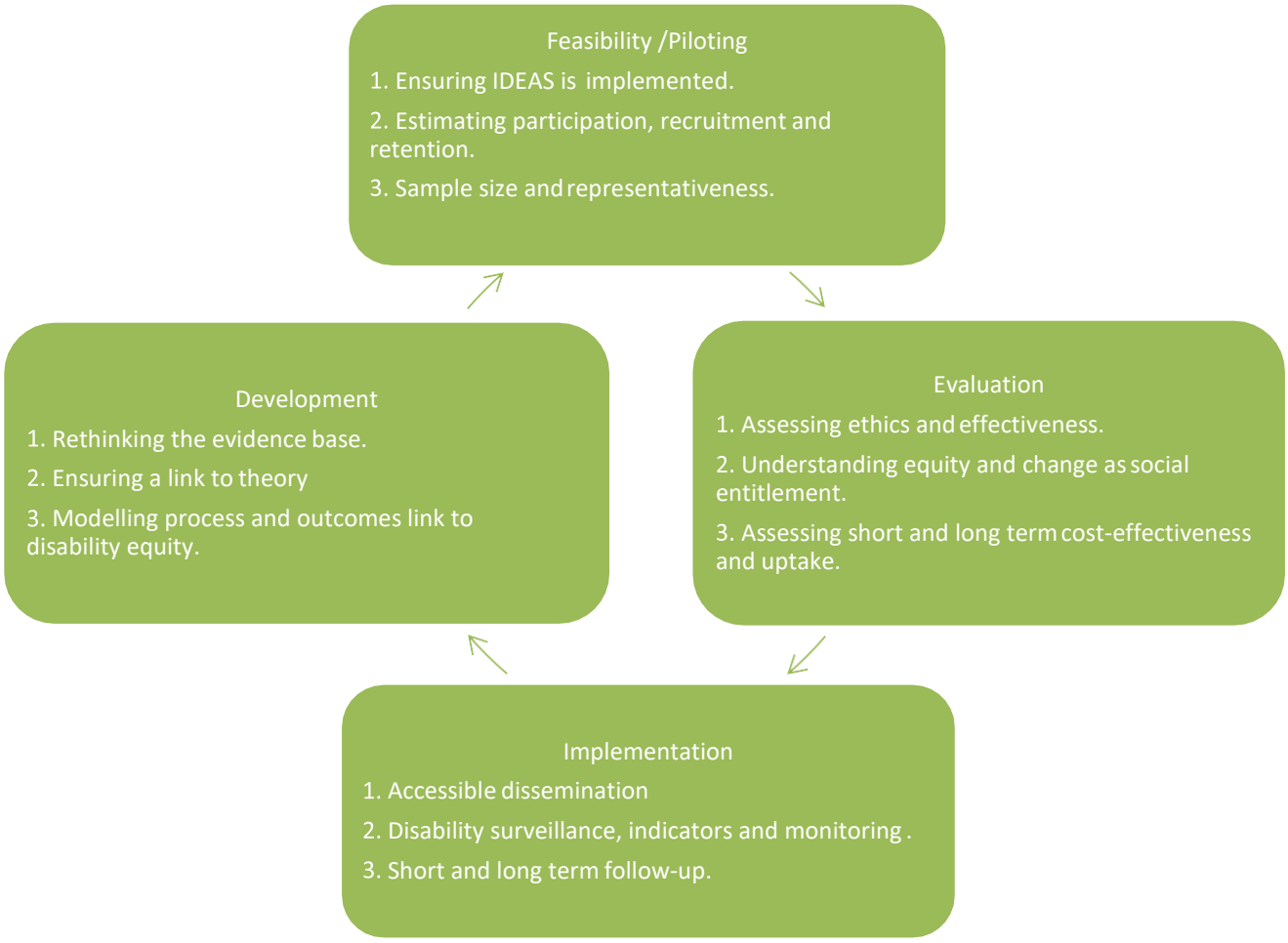
Figure 2: **IDEAS**

RCT	Inclusion	Dignity	Equity	Accessibility	Equity	Inter-Sectionality
	How were people included?	How was dignity ensured?	What kind of short or long term impact will this have in changing inequalities?	Was accessibility thought about?	What kind of short or long term impact will this have in changing inequalities?	Did they involve people with different types of impairments, sexualities, from different ethnic groups, women or children?

The IDEAS guide provides a critical appraisal tool for dealing with the complex ethical and empirical issues that arise when designing and/or evaluating public health interventions. The CRPD establishes the importance of inclusion, in addition to the need to respect dignity, assurances of intersectionality, access and equity when engaging with disability. There are also important legal considerations which should not be overlooked in study designs, which an engagement with human rights can ensure. For example, assurances of inclusion or dignity in the UK could require an engagement with The Mental Capacity Act (2005) which outlines issues like supported decision-making.

The use of the CRPD thus also entails that IDEAS is an international framework and can be adopted globally.[13,14] We illustrate how this could be adapted to the development of all RCTs.[35] For example, in Figure 3, an illustration is given how IDEAS could be integrated into the Medical Research Council guidance for complex interventions.[36]

Figure 3: **IDEAS for MRC guidance**



In the above, IDEAS is integrated in all stages of RCTs from development, piloting and evaluation through to assessing implementation. The MRC guidance (similar to RE-AIM) ensures methodological robustness while IDEAS ethically refines empirical design for greater inclusion and assurances of rights. The potential for using IDEAS in national and international research and developing it further to develop indicators of equity, social determinants of human flourishing or adherence of disability related projects to human rights exists. The tool also illustrates how innovative empirical ethics can be in practice when pragmatically combined with human rights.

Conclusion

Our research suggested that there has been little debate in public health about what ethical inclusion in research looks like or how it should be respectful of disability.[1] We also noted how people with disabilities now felt that they had a right and social need to be involved and co-creating research.[2] Building on these insights, we illustrated one way in which human rights-based models could inform an empirical-ethical critical appraisal tool or evaluative guide. We argue that such guides that could be used to critically engage with current empirical evidence, while also allow researchers to adapt to more inclusive ethical practices that are sensitive to the experiences of disability and expectations of stakeholders.

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