Letter from Bristol

INCLUDING VULNERABLE PEOPLE IN RESEARCH: THE CASE OF INTELLECTUAL DISABILITIES

People with intellectual disability (ID) are a vulnerable group with a considerably reduced ability to understand new or complex information, to learn new skills and to cope independently—which started during the developmental period with a lasting effect. Between 2% and 3% of the population have IDs.[1] People with ID often face extensive health inequalities, a range of mental and physical problems and early mortality.[2] Apart from these personal costs, ID is also associated with long-term responsibilities on carers and families. Yet people with ID are generally not included in medical research.[3] This is unfortunate because apart from advancing knowledge about the health and care for people with ID, such research may also benefit the understanding of issues relevant to the wider population. For example, a major advance in the understanding of dementia occurred following identification of the link between ApoE lipoprotein on chromosome 21 and dementia in Down syndrome.[4] Moreover, research can have a place in allowing those whom society tends to ignore to make potentially great contributions.[5]

To plan and run health services well, it is important to know the needs of this population. For example, we need to know the prevalence of ID and incidence of comorbid conditions.[3] But those with ID are not a homogeneous group, approximately 25% may have an identifiable genetic cause but 30%–50% are of unknown cause.[1] If we do not have a clear picture of what treatments work best with whom then we risk our care being subpar.[6] Even where research exists, it does not always get translated into clinical practice and guidelines.[7]

Barriers to inclusion

Paternalistic attitudes to people with ID can be a major barrier. There can be instances of active exclusion of those with ID from opportunities to participate in research.[3] Doctors and carers, including families are often reluctant to involve those with ID under their care in research—fears around ability to consent being an important factor.[8] The UK Mental Capacity Act 2005 provides a framework to guide researchers in involving people who cannot give consent in medical research.

It is important to acknowledge that people with ID are a vulnerable group and there have been instances of human rights abuses. One historical example of misuse of research in this population was the infecting with hepatitis of 'mentally defective' children in Willowbrook State School in the USA in the early 1960s to study disease course.[3] The UN Universal Declaration of Human Rights and international ethical guidelines for research now stand to provide safeguards and protect vulnerable populations. On the other hand, an assumption of lack of capacity[9] may create an unnecessary barrier to involvement of this population in research. People with ID can give valid informed consent to many issues if the information is presented in an accessible way and where they cannot consent, legislation such as the Mental Capacity Act can be used as a guide.[10]

The ethics of conducting research involving people with ID is complex. Questions asked by ethical approval committees in the UK include whether the research question could involve other participants, what are the risks versus benefits and can these be justified?[11] Sometimes important safeguards to protect the vulnerable might actually deter researchers to work with people with ID. It is important to consider whether it is ethical to allow a group to *not* be included in research when there is a clear clinical need and a paucity of research evidence to date.

Research often involves narrow eligibility criteria. However, not investigating those who have physical and mental health comorbid conditions means that those who are excluded from research are those who may provide the most telling information. Moreover, illness classifications themselves can sometimes present a hindrance to cleanly applying research findings to whole populations. Comorbid illness rates in those with ID have been put at between 5.7% and 47%.[3] Such imprecise statistics are perhaps due to small study sample sizes, high study drop-out rates leading to low statistical power studies and different subcategories of ID being used for different studies and so limiting comparability and reliability.

There is a difficulty in recruiting and retaining people with ID into medical studies.[3] This is often a socially isolated and disadvantaged group and one which has coping as best possible with daily living as a higher priority than attending study follow-up appointments. This along with carer reticence can further limit opportunities for involvement. This all can contribute to high drop-out/lost-to-follow-up rates and to small sample study sizes.

Practicalities of research also need due consideration. Research into ID often involves adjustments to methods that might be used in the general population. Reasonable adjustments must be made to enable participants to understand. For example, study questionnaires for depression may have to be modified to be made accessible to those with a lower reading age or to include pictorial representation. Even then, their validity as an indicator of depressive symptoms may be questionable as this population may present with unusual symptoms such as behavioural problems.

Finally, carrying out research in this population may entail greater requirements in terms of time and costs, which may be a hindrance to research.[12] Funding opportunities are often limited and there are a relatively small number of researchers in the field. As such, ID research often seems not to be treated as a priority.

Ways forward

Complexity should not be a barrier to research. It is possible to conduct randomized controlled trials involving participants with ID.[3,13,14] However, there is a need to make reasonable adjustments for including people with ID—a fact that must be embraced rather than side-stepped.

Reading the reflections on drop-out rates in two studies—Nicholson *et al*.[15] and Turk *et al*.[16]—the importance is noted of keeping in frequent contact with participants, being flexible around their life commitments and trying to make the research relevant to them. Information given early on to participants about the nature of, not only the specific study, but of the whole trial process—provided in various, more accessible formats—will be needed. This, along with direct face-to-face or phone contact, has been shown to help.[17] Greater collaboration between researchers and ID representative groups might enable greater uniformity in approaches to ethical trial design and implementation.[5]

Policy-makers should strive to create and maintain accurate statistical datasets for their populations. Highlighting health inequality to the general public, interest groups and potential funders will be important. In the UK, a public body has been created—the 'Improving Health and Lives: Learning Disability Observatory' (IHAL)—to look into health inequalities and fund research as well as encourage local community doctors (general practitioners) to report on the quality of care those with ID receive. It has also recommended a national auditing scheme to monitor for comorbid illness.[18] Initiatives like this could allow for more inclusive, systematic and methodologically consistent research. Formulating systematic protocols and good-practice research guidance which can be widely applied to this group may make starting and following ethically approvable trials easier.

In conclusion, it is possible to carry out high-quality research with vulnerable groups such as people with IDs but collaborative approaches at all levels early on in the development process may be necessary. A move away from paternalistic models and a change in our awareness and approach to the problem may be needed so that we can fulfil our duty to these patients and ensure their right to well-evidenced care.

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