Appendix 1: Systematic Review Protocol

1. Title

Equitable and Accessible Informed Health care Consent Process for People with Intellectual Disability: A Systematic Literature Review

2. Registration

PROSPERO registration number: CRD42021290548

3. Authors

Please contact the primary investigator.

4. Amendments

Any amendments to this protocol will be dated and described in this section. A rationale will be provided for each amendment. Changes will not be incorporated to the protocol detailed below.

Amendment 19th June, 2023.

Two co-researchers with intellectual disability who are integral members of the inclusive research team and self-advocate leaders will be involved in this review and included as co-authors. They will be involved in sense-checking findings, ensuring findings match lived experiences of people with intellectual disability, review the wording of findings, co-producing an Easy Read summary, and discussion research dissemination. This is in line with best practices of co-production research for intellectual disability (1). Multiple workshop meetings will be held to discuss findings and make amendments to reflect changes.

5. Support

There were no financial supports or sponsors of this systematic literature review.

6. Rationale

People with intellectual disability face multiple barriers to equitable health care that result in poorer health outcomes. (2, 3). The accessibility to health services have improved for the population without intellectual disability, with a focus on patient autonomy and shared decision-making. However, there are ongoing access barriers for people with intellectual disability (4) and there is evidence that poor informed consent practices for medical interventions contributes to further health inequity (5, 6). Informed consent is a fundamental component of bodily autonomy and a protected patient right (7), yet people with intellectual disability continue to be excluded from the medical decision-making process and are not provided the opportunity of proper informed consent (8).

In order to improve the informed consent process for people with intellectual disability, it is crucial that a deeper understanding of the factors that contribute to their informed consent is first established. There is currently no systematic review examining the barriers and enablers of informed consent for people with an intellectual disability. The view of consent has also changed in the last three decades, transitioning from one focused on the ‘assessment of capacity’ (which addresses the primary question of ‘How do we determine whether a person

with intellectual disability has capacity for informed consent?') towards a view that emphasises the reasonable adjustment of the informed consent process to support people with intellectual disability (i.e., addressing the question of 'How do we best support informed consent for a person with intellectual disability?'). For health systems and professionals to best support autonomy and choice for people with intellectual disability, we must understand the status of informed consent and factors that currently affect this process.

7. Objectives (PICO)

The objective of this study is to examine the literature systematically to identify barriers and enablers of the informed consent process for medical interventions for people with intellectual disability.

Participants

We will include studies that examine relevant stakeholders in health care for people with intellectual disability, including people with intellectual disability themselves, their carers, support people, health professionals (doctors, nurses, midwives, general practitioners, allied health staff), other professionals that are involved in the care of people with intellectual disability (including group home staff), or any other people with previous experience with the informed consent process for medical interventions for a person(s) with intellectual disability (including academics or researchers). For studies with mixed participant groups, studies were included if the majority (>50%) of participants were relevant stakeholders. The majority of participants in the studies must also be adults (18 years or older).

Interest/intervention

We will include studies that examine the factors that influence informed consent process. In other words, it will include studies that examine the factors that aid or obstruct an individual with capacity when making decisions. It will not include proxy consent for individuals without capacity.

Context:

Informed consent was examined in the context of any medical/health care intervention. A health intervention was defined as any activity undertaken that aimed to treat, prevent, reduce the severity of, or change the natural progression of a disease process, or to restore function lost by that disease process (9). Studies will not be included if they examine consent for research or consent for non-medical decisions.

8. Eligibility criteria

Included articles must:

1. Examine the informed consent process for a health care intervention from people with intellectual disability.

2. Have collected most of its data (more than 50%) from relevant stakeholders, including adults with intellectual disability, families or carers of a person with intellectual disability, and professionals who engage with people with intellectual disability.
3. Report empirical data from primary research methodology.
4. Be published in a peer-reviewed journal after January 1990.
5. Be available in English.

Included studies must have a focus on informed consent either by including this as a primary objective, research question, or as a significant finding (i.e., outlined in an independent heading). Studies were excluded if informed consent was only intermittently mentioned throughout the text, or if an effect on informed consent was a secondary or partial finding.

9. Information sources

Search strategies will be developed using subject headings and search terms related to 1) informed consent, 2) intellectual disability, and 3) health care, and will be combined using the 'AND' Boolean operator. Synonyms will be combined using 'OR'. Truncated search terms will be used to search titles and key words to capture all possible relevant studies.

We will search Embase, MEDLINE, PsychINFO, PubMed, SCOPUS, Web of Science, and CINAHL as the chosen electronic databases. This search will be limited to studies available in English, and studies published from January 1st 1990 onwards. The search was limited to studies published from 1990 onwards as the current understanding of consent has changed over time, and literature published prior to 1990 is based on an outdated view of consent and capacity.

To ensure full capture of relevant studies, we will use an ancestral search of identified articles and other relevant literature reviews. We also will hand-search three major journals relevant to intellectual disability research: the British Journal of Learning Disabilities, Journal of Intellectual Disabilities, and Journal of Intellectual Disability Research. These journals will be searched from 1990 January onwards.

10. Search strategy

The search will include studies that are qualitative, quantitative, and mixed-methods, and include studies published from 1st January 1990 onwards, inclusive. The specific search strategy will initially be developed in conjunction with a Librarian with previous experience in systematic review search strategies, and will then be reviewed by other researchers within the team.

An initial search strategy was developed in Embase, and then adapted to apply to other electronic databases. The Embase search strategy is below.

1. Intellectual impairment/
2. Learning disorder/
3. Mental deficiency/

4. ("Intellectual* disab*" or "Learning disab*" or "Learning difficult*" or "Developmental* disab*" or "Cognitive* disab*" or "Cognitive* impair*" or "Mental* retard*" or "Mental* Handicap*" or "Developmental* Delay*").kw,ti.
5. Informed consent/
6. Decision making/ or clinical decision making/ or medical decision making/ or patient decision making/ or shared decision making/
7. ("decision mak*" or assent or consent* or autonomy).kw,ti.
8. Exp health service/
9. ("health care" or "health care" or "health service" or "clinical practice" or "medical care" or "procedure" or "interven*" or "treat*" or "management").kw,ti.
10. 1 or 2 or 3 or 4
11. 5 or 6 or 7
12. 8 or 9
13. 10 and 11 and 12

11. Study records

Data management

Results from the search strategy will be imported into Covidence Software, an online program that facilitates streamlining of the screening process with multiple reviewers simultaneously. Duplicates will be automatically removed, and the remainder will be screened on the title and abstract based on the inclusion and exclusion criteria agreed upon.

An inter-reviewer reliability test will first be completed of a minimum 10% of the imported studies to refine the inclusion/exclusion criteria and ensure reliability between independent screeners.

Selection process

Two independent researchers will screen the titles and abstracts identified from electronic databases, ancestral searches, and hand searches of journals. These studies will be screened based on the agreed inclusion/exclusion criteria. The articles that meet these criteria will then undergo additional screening of the full text to determine if they meet the full inclusion criteria. Where unsure, additional information will be sought through the article's Appendix or Supplementary material, and through direct contact with authors if necessary to determine eligibility. Reasons will be provided for any studies that are excluded.

Any conflicts will be identified and resolved with a third independent researcher and open discussion. If there is ongoing conflict, then a fourth independent researcher will be involved.

Data extraction

Standardised analytical forms will be developed and applied to extract data from the included studies. These forms will be developed with input from two different researchers with different expertise to facilitate a multidisciplinary approach to data extraction, and will be

piloted with an initial study before being applied to other included articles. Two independent researchers will extract the data independently using the standardised forms for each eligible study. To ensure consistency, the two researchers will first complete data extraction for at least 10% of articles collaboratively with open discussion, and clarify any specific conflicts and establish consistency between them. A Cohen-kappa score will be calculated to ensure adequate agreeability. If this is insufficient then further clarification about the inclusion and exclusion criteria will be completed between the researchers and the inter-researcher reliability will be re-assessed after this.

Data extracted will include demographic information (sample size, age, gender, country, year, authors, study design, methodology, reported outcomes, and quality scores). In the case of any uncertainties or missing information, data was sought from Appendices, supplementary material, or through direct contact with authors. Specific sources of bias will also be extracted such as participant recruitment, involvement of gatekeepers, inclusiveness of the study design, accessibility of the study, reflexivity of researchers, and other limitations identified by authors.

12. Data variables

For all studies, we will extract data on the participant demographics (sample size, age range, gender), the category of stakeholder (e.g., people with intellectual disability, carers, support people, health professionals), study design, data analytical methods, and main outcomes. Any details on methodology or outcomes that are omitted or have insufficient detail will also be extracted.

For quantitative studies, we will additionally extract any controls used, participant demographics (number, age range, gender), and effect size calculations if available. For qualitative studies, we will additionally extract the key themes and findings of the studies. For mixed-method studies, both qualitative and quantitative additional data variables will be extracted.

13. Outcomes and prioritization

The primary outcome will be any factors that support the process of informed consent (enablers), or any factors that impede the process of informed consent (barriers). This includes factors affecting any relevant stakeholder (including a person with intellectual disability, health professional carer/support person, or any other person involved in informed consent for a person with intellectual disability) as well as systemic factors.

14. Risk of bias

Bias will be assessed at the study-level by two independent researchers during the data extraction phase. The QualSyst tool will be used to assess the quality of each study due to its ability to assess both qualitative and quantitative studies, and the use of standardised criteria allowing accurate replication. The tool includes domains evaluating bias such as method of participant recruitment, clarity of inclusion criteria, any blinding, reflexivity of researchers, selective reporting of outcomes, estimation of variance, controlling of confounding factors, and use of verification procedures.

For each domain within the QualSyst tool, two independent researchers will assess each study and provide a score. Any conflicts will be resolved with a third researcher and with

discussion. The risk of bias will be considered for each study when interpreting the results and outcomes in data analysis. Studies will be excluded if the quality score is below the determined threshold. Mixed-method studies will have both qualitative and quantitative scores calculated and the higher score used. The quality threshold will be between 0.55 – 0.75 and will be determined depending on the spread of quality scores of the included studies, and the number that would be excluded at each threshold.

15. Data synthesis

If the included studies are homogenous in study design and measured outcomes, meta-analysis will be completed. However, it is anticipated that there will be significant heterogeneity to studies given the literature on intellectual disability is primarily qualitative and mixed-methods in nature.

In the case of study heterogeneity, an inductive thematic analysis approach will be used. This will completed using the six-phase method described by Braun and Clarke (10, 11) consisting of familiarization with data, generation of initial codes, searching for themes, reviewing themes, defining themes, and then writing up the findings. Synthesised themes will then be discussed with two other independent researchers. Where possible, related themes will be grouped together into larger umbrella themes.

The frequency of identified themes and subthemes will also be calculated and summarised in a table to provide an objective measure of the significance of a theme. It will be assumed that factors that are commonly identified multiple times in the included studies are more likely to be significant factors affecting the process of informed consent. Each major theme will be synthesised and summarised based on the frequency of the theme in the studies and the quality of the study, considering any limitations or risks of bias.

16. Meta-biases

To assess meta-bias for studies that involved people with intellectual disability as participants, each study that includes this group as participants will be evaluated on their inclusiveness, selection criteria, use of gatekeepers, and specifically if only people with ‘mild’ intellectual disability are included. These are common limitations that contribute to biases in the intellectual disability literature (12-14). The use of advisory groups or other methods of co-design to reduce bias will also be evaluated for all studies, particularly those involving people with intellectual disability, as this can influence the validity of study findings (15).

Part of the QualSyst quality assessment includes assessments on reflexivity and validation procedures to reduce biases in qualitative studies, and this will also be considered in evaluating meta-biases present in the qualitative intellectual disability literature included in this review. The quality assessment will also assess for any missing reported outcomes, small sample size, and whether the conclusions match results described. Triangulating results between different stakeholders (i.e., people with intellectual disability, different health professionals, support workers, family members) will also be a part of validating the results of the studies and the possibility of reporting bias in studies.

Another challenge in intellectual disability is the heterogeneity of terminology. The search strategy will be deliberately broad and involve multiple methods (including ancestral and hand-searching) to improve the likelihood that relevant studies will be identified.

17. Confidence in cumulative evidence

The quality of studies will be assessed using the QualSyst tool which provides assessments on study design, sample size, outcome reporting, and potential for biases, and any low-quality studies will be excluded (based on a determined quality threshold such as 0.55). Each study will be evaluated by two independent researchers for other limitations and any further biases as part of the standardised analytical form used in the data extraction phase. The GRADE tool will also be used to assess the quality of evidence (16, 17), and will also consider the frequency a particular theme or subtheme is mentioned in the literature. These evaluations will be applied in data analysis for each individual study when evaluating the themes identified, and will all be considered when synthesising the review's findings to determine the overall significance of each subtheme. High quality findings would be themes or subthemes that are supported by multiple studies, identified by multiple different stakeholders (i.e., confirmed by triangulation), or arise from studies that have inclusive study design with high quality scores and minimal biases.

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