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Systematic Review

Informed Consent or Assent Strategies for Research With Individuals With Deafblindness or Dual Sensory Impairment: A Scoping Review

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KEYWORDS
Deaf-blind disorders;
Disabled persons;
Hearing disorders;
Informed consent;
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Abstract *Objective:* To synthesize evidence on existing informed consent/assent strategies and processes that enable the participation of individuazls with deafblindness or dual sensory impairment in research.
Data Sources: Five scientific databases (PubMed, MEDLINE, Cumulative Index to Nursing and Allied Health, Web of Science, and PsycINFO) and other sources such as Google Scholar, *Journal of Visual Impairment and Blindness*, and *British Journal of Visual Impairment* were hand-searched from January 2015 until July 2020.
Study Selection: Studies were selected using *a priori* inclusion criteria of sensory and cognitive disabilities and focused on consent/assent strategies and processes in research within this population. Articles related to the medical or sexual consent processes were excluded.
Data Extraction: An Excel spreadsheet was used to extract data from the eligible sources. Discrepancies were resolved in discussion with team members.
Data Synthesis: A total of 2163 sources were screened, and 16 articles were included in the review. Seven sources only examined consent strategies, whereas the remaining 8 included a combination of consent/assent and dissent strategies. Using thematic analysis, 3 key themes emerged: consent/assent strategies, researcher capacity, and capacity to consent tools. Key identified strategies included the accessibility of the consent/assent process, building

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List of abbreviations: LMIC, low-middle-income country; PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analysis.
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relationships with participants and caregivers, identifying behavioral cues, and communication training for researchers.
Conclusions: Despite the absence of literature on consent/assent strategies within the population with deafblindness, the review found promising strategies applied to individuals with other cognitive or sensory disabilities that researchers can adopt. Researchers are encouraged to use best practices in creating an inclusive research environment to include individuals with deafblindness.

Deafblindness or dual sensory impairment is a distinct disability with varying combinations of vision and hearing impairment affecting communication, mobility, and access to information.[1] Individuals with deafblindness can be categorized into 3 types: (1) congenital deafblindness that includes individuals born with both hearing and vision impairment; (2) acquired deafblindness whereby individuals acquire sensory loss through accidents or illnesses or are born with a single sensory loss and acquire the other impairment; and (3) age-related deafblindness (or dual sensory loss) in older adults, resulting from changes to vision and hearing.[2-4] Globally, 0.2%-2% of the population live with some form of deafblindness, with the actual number expected to be much higher because this disability is poorly understood and often misdiagnosed.[5] This population is more likely to face depression, health issues, and social exclusion.[6-8] Given the complex nature of deafblindness, researchers often face challenges in the implementation of informed consent and data collection because of issues in communicating with participants.[9,10] This may lead researchers to exclude those with deafblindness from participating in research.[5]

Historically, Research Ethics Codes, such as the Declaration of Helsinki, the Nuremberg Code, and the Belmont Report, were developed to ensure there are strong ethical standards when involving human participants in medical research.[11-14] However, none of these ethics codes discuss the process of conducting research with individuals with disabilities nor provide any guidelines to obtain informed consent. In Canada, the *Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans — TCPS 2,2018*, a joint policy by the Canadian Institutes of Health Research, the Natural Sciences and Engineering Research Council of Canada, and the Social Sciences and Humanities Research Council of Canada, provides guidance relevant to inclusion in research of specific groups such as women, children, older persons, and those who lack the capacity to decide whether to participate.[15] These policies or ethics codes only provide general guidelines rather than focus on how to apply them in practice, particularly when working with individuals with complex disabilities. Currently, there are no evidence-based guidelines or peer-reviewed literature to support researchers in obtaining consent from individuals with deafblindness. As a result, researchers may get discouraged and place individuals with deafblindness in the study exclusion criteria. Given that the number of individuals with deafblindness is expected to grow in the next decade,[8,16-18] research involving this population is critical to better support and design services unique to their needs. This review determined gaps in the literature on obtaining informed consent/assent when conducting research with the population with deafblindness and summarized various strategies available with other similar disabilities. The purpose is to assist researchers in adopting these strategies when conducting research with individuals with deafblindness.

Methods

In accordance with Arksey and O'Malley's methodological framework,[19] a scoping review was conducted based on the following 6 stages: identifying the research question; identifying relevant studies; study selection; charting the data; collating, summarizing, and reporting the results; and consuling with stakeholders. A scoping review is defined as a form of knowledge synthesis that highlights gaps in the literature and provides key evidence on a specific topic.[20,21] Given the early stage of development in this field of research, the team chose to conduct a scoping review over other forms of review to synthesize and map the existing literature on informed consent/assent process and strategies that could be relevant to the population with deafblindness.

This review, in addition to studies on deafblindness, also included studies in populations other than deafblindness, such as dementia, intellectual or developmental disabilities, and hearing or vision impairment. We anticipated finding very few studies on informed consent in individuals only with deafblindness. Furthermore, individuals with congenital or acquired deafblindness often have multiple disabilities, such as cerebral palsy, autism, or intellectual disabilities.[2,22-24] Hence, we decided to broaden the scope of our search to include studies on cognitive and sensory disabilities whose practices could be applied in research with deafblindness. Likewise, individuals with age-related deafblindness are more likely to experience cognitive decline than those without sensory impairment.[25-27] For example, up to 90% of individuals with mild or moderate dementia also live with hearing loss, and 30% report vision impairments.[28] As recommended by the Joanna Briggs Institute Manual for Evidence Synthesis, the population, concept, and context were defined.[21] This review uses the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) extension for Scoping Review reporting guidelines.[20]

Identifying the research question

The review was guided by the research question, "What are the existing informed consent/assent practices to enable the participation of individuals with deafblindness in

research?" This review defined the population *individuals with deafblindness* as those that have "combined loss of hearing and vision to such an extent that neither hearing nor vision can be used as a means of accessing information to participate and be included in the community."[29(p2)] The concept of *consent* is defined as an ongoing process whereby an individual that has the capacity and understanding to make an informed decision voluntarily accepts without coercion to participate in research that benefits them.[30-32] *Assent* is defined as when a legally authorized representative is required to make the decision for an individual to participate in the study.[30,33] The individual is too young to provide consent but mature enough to understand the risks and benefits associated with the research.[30,33] On the other hand, *dissent* is defined as verbal or nonverbal behavioral cues signaling an unwillingness to participate in the research.[34] Such cues include verbally saying *no* or behaviors indicating a lack of cooperation, wanting to leave, or signs of distress.[34] The spatial context for this review are communities around the world where research policies and practices to support those living with deafblindness and other cognitive and sensory disabilities exist. The temporal context was restricted to articles published in the last 5 years because the researchers' intents were to identify emerging evidence and most current consent practices.

Identifying relevant studies

A search strategy was created in discussion with a librarian from Ryerson University (supplemental appendix S1, available online only at http://www.archives-pmr.org/). The electronic databases PubMed, MEDLINE, PsycINFO, Cumulative Index to Nursing and Allied Health, and Web of Science were searched for the time period from January 2015-July 2020 in accordance with the PRISMA extension for Scoping Review (supplemental appendix S2, available online only at http://www.archives-pmr.org/). Because the research team was interested in synthesizing the current context, the time frame was restricted for studies from 2015 onward. Moreover, most research related to deafblindness and Alzheimer disease was published in the last 5 years, making this the most relevant period. To ensure comprehensiveness, the team conducted a search on Google Scholar for the first 100 hits (see supplemental appendix S1). Additionally, the *Journal of Visual Impairment & Blindness* and the *British Journal of Visual Impairment* were hand-searched for articles published from January 2015 to July 2020 because they may contain studies that have not been indexed electronically and may include relevant articles in the field of sensory impairments. Through the process of snowballing, the reference lists of included articles were screened to capture any additional studies.[35] Results from the search were imported into Mendeley Desktop Version 1.19.6,[a] a reference manager, and exported to Covidence,[b] an online screening tool, to remove duplicates.

Study selection

Studies were screened by 3 authors (A.P., R.M., A.J.) at both the title/abstract level and full-text level based on the exclusion and inclusion criteria, using Covidence (table 1). Articles selected for full-text analysis were reviewed and recorded into a Microsoft Excel 2016[c] spreadsheet. During the 2-stage screening, 2 authors (A.P., R.M.) reviewed the compiled list of sources, and 2 authors (A.J., W.W.) resolved conflicts in study selection. Figure 1 provides the PRISMA flow chart providing details on identification, screening, eligibility, and inclusion.

Charting the data

Based on the study objectives, a data charting template was mutually created by authors (A.P., A.J., R.M.) in consultation with the senior author (W.W.). The spreadsheet contained a set of descriptors including author's names, year of study, journal title, location, population, terminology used for consent/assent, focus of the article, aim of the study, methodology used, consent strategies, assent strategies, key findings, implications, and any other significant information. The first author (A.P.) extracted the data and the other authors (A.J., R.M.) were consulted to review the extraction. A consensus meeting was held to discuss any discrepancies and to confirm study inclusions.

Collating, summarizing, and reporting the results

The findings were analyzed using descriptive numerical summary and qualitative thematic analysis[19] and are presented

Table 1 Inclusion and exclusion criteria

Inclusion Criteria	Exclusion Criteria
Studies related to persons with deafblindness and other sensory and cognitive disabilities, such as vision impairment, hearing impairment, Alzheimer disease, dementia, developmental disabilities, or intellectual disabilities	Studies related to populations without disabilities or other forms of disabilities, such as physical or psychiatric disabilities, or focusing on genetic research
Studies related to qualitative or quantitative research consent and assent strategies and processes	Studies that focus on consent for medical (ie, end of life), clinical trials, or sexual purposes
Studies that were published 2015 and onward	Studies examining only the capacity of an individual with a disability to consent
	Full text in a language other than English

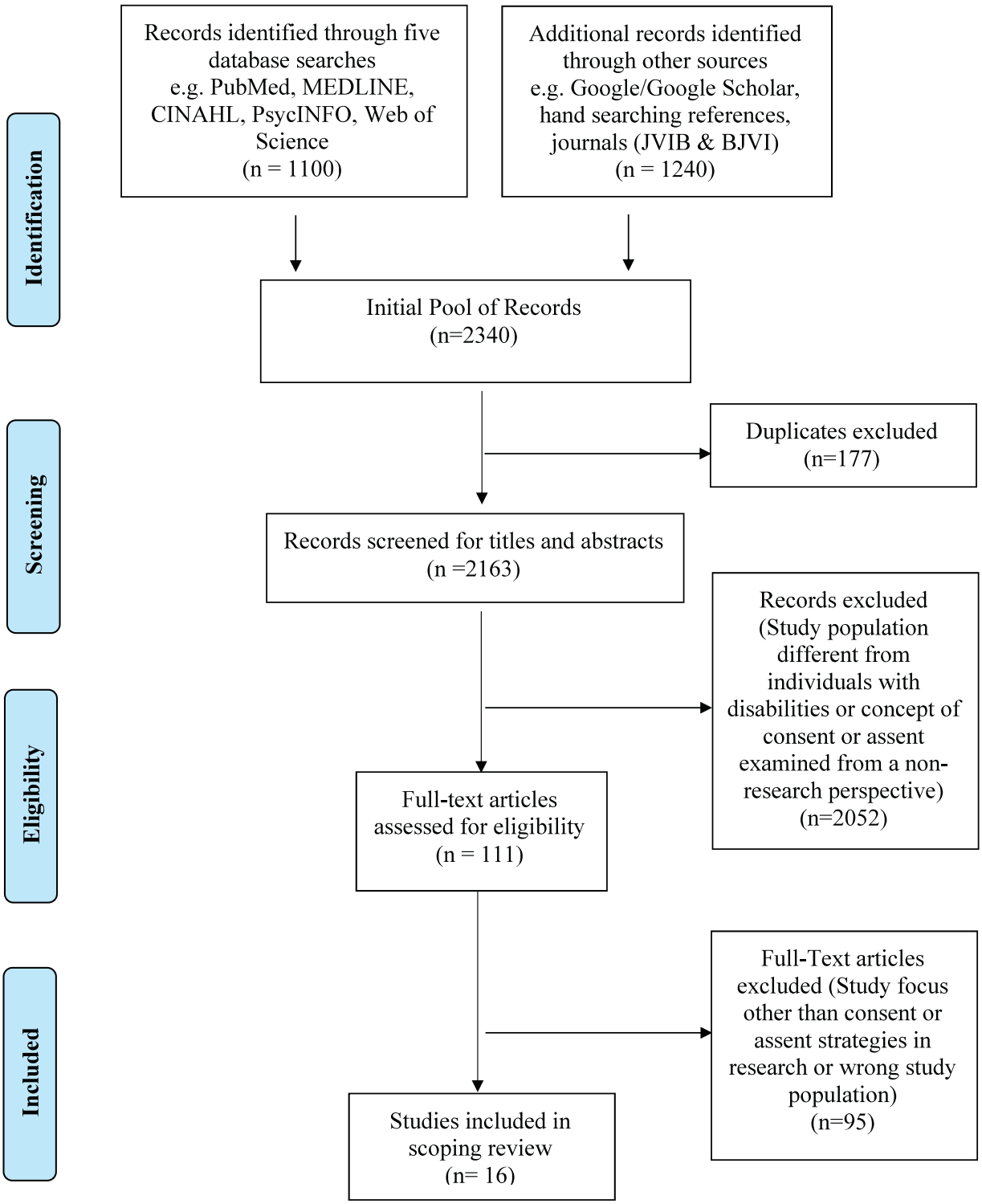

Fig 1 PRISMA flow diagram. Abbreviations: BJVI, British Journal of Visual Impairment; CINAHL, Cumulative Index to Nursing and Allied Health; JVIB, Journal of Visual Impairment and Blindness.

in tables and figures. The extracted data were coded for words, and key themes were identified using Braun and Clarke's[36] principle of thematic analysis by the first author (A.P.) and confirmed by the team. The findings of each eligible source were discussed to understand the scope, implications, and limitations for future research. After a discussion, the authors determined the best approach to present the identified themes and strategies and then divided each strategy by age because of the diversity in age range within study populations.

Consultation with stakeholders

The research team chose to conduct the sixth optional stage and meet with stakeholders to discuss possible consent strategies for research in individuals with deafblindness. However, because of the coronavirus disease 2019 pandemic, it was not feasible to hold consultations with stakeholders of the partner community organization. In the future, there are plans to consult with stakeholders once the pandemic situation improves.

Results

Of the 2340 sources identified, 1100 articles were extracted from 5 bibliographic databases; 1240 were from Google/Google Scholar, hand-searching references, and journals. Based on the eligibility criteria (see table 1), 16 articles were accepted for the review. Figure 1 provides details about the identified, screened, and eligible sources.

Study characteristics

Of the 16 eligible studies, the majority were empirical (n=9), followed by reviews (n=6) and a guide (n=1). The empirical studies varied in methodology (qualitative or mixed), design (eg, randomized control studies, cross-sectional, case studies, reflections), and data collection techniques (eg, interviews, focus groups, observations). Table 2 presents details on the study descriptors. Other than 1 study that was conducted in a low-middle-income country (LMIC), the remaining studies were conducted in high-income countries within Europe, Oceania, and North America. Most were from Australia (n=7), followed by Ireland (n=4), the United Kingdom (n=2), Canada (n=1), the United States (n=1), and India (n=1) (fig 2). Interestingly, the year 2017 had the highest number of published articles (n=5), with the remaining years at only 2 or 3 articles. Approximately half of the sources focused on intellectual disabilities (n=7), followed by dementia (n=3), combination of cognitive and sensory disabilities (n=2), deafblindness (n=1), autism (n=1), complex communication needs (n=1), and cognitive impairment (n=1). Seven studies specifically included the age of participants, ranging from 6-86 years old, whereas others used terms such as children, adults, or older adults. Only 1 study included the population with deafblindness to examine and address the challenges of conducting qualitative research in individuals with congenital and acquired deafblindness.[9]

Major themes from qualitative synthesis

After qualitative synthesis, 4 key strategies in the consent/assent process emerged (fig 3). They include accessibility of the consent/assent process; relationship building between researchers, participants, and caregivers; behavioral cues of participants; and training for communication strategies and assistive devices for researchers. To present our findings, the applicability of these strategies has been divided into 3 age groups: children/adolescents aged 0-19 years[53]; working age adults aged 20-59 years[54]; and older adults aged ≥60.[54] Table 3 provides an overview of suggested strategies applicable to each age group per theme. For each strategy, the authors discuss the tools and adaptations explained by identified sources, then classify the studies according to each age group.

Accessibility of consent/assent process

A key strategy in obtaining consent in individuals with disabilities was ensuring the accessibility of the consent process. Studies reported the use of plain language, using pictures, and verbally reading the forms to ensure understanding of the research.[9,38-42,44,46,49-52] Two studies included adults with autism and intellectual disabilities that designed or reviewed consent forms for younger participants of the same disability.[44,46] The authors explained that consent forms should be tailored to the needs of each individual, such as having more text with fewer pictures or using tactile methods over visual tools.[40,46,50] These strategies can be further divided into each age group.

Children

Including pictures of the researchers when obtaining assent from children with vision or hearing impairment, cognitive disabilities, and/or autism, was recommended.[40,49] One source used a DVD featuring children and young people with cognitive, autism, or physical disabilities to inform potential participants and their parents about the research activities they will be partaking in.[49] The DVD included children using "Talking Mats," an interactive communication tool that can help users express their perspectives and feelings more clearly.[49] In addition, researchers can use objects for communication, such as a toy that children can communicate with. One study used a doll to ask participants, "If this doll had a disability like you, what do you think he would wish for to make his life better?"[40] Additionally, having thumbs up and down stickers, drawing, and/or cameras and tape recorders were recommended.[40] The communication needs of children were met giving preference to tactile methods rather than visual tools.[40] Another technique involved role playing to provide an overview of the consent process to children/youth participants. In the consent process as part of a play, researchers provided alternative endings to show participants that they can make their own choice, and any choice they make will be respected equally.[41]

Working-age adults

Similar to work with children, plain language and clearly worded information sheets were used. These forms included bullet points, pictures, and repeated information that was read out loud.[38,39,50] One study with adults with intellectual disabilities included other adults with intellectual disabilities to review information sheets and make recommendations.[46] Participants were also given a choice of 2 documents: one with more text and fewer pictures and vice versa.[46] One study involving individuals with deafblindness asked participants for their preferred accessible format, such as if they wanted a large-print, braille, or accessible word file.[9]

Table 2 Characteristics of records included in the review

Author	Location	Type of Disability	Study Design	Participants (n)	Age Group of Participants	Study Focus
Beattie et al[37]	Australia	Dementia	Descriptive, cross-sectional	392	Older adults	Informed consent process of individuals with dementia
Dorozenko et al[38]	Australia	Intellectual disability	Qualitative	Not applicable	Working age adults	Participation of individuals with intellectual disabilities in research
Ho et al[39]	Australia	Intellectual disability	Qualitative	40	Working age adults	Informed consent process in research with people with intellectual disabilities
Jenkin et al[40]	Australia		Guide	Not applicable	Children	Consent and assent strategies for children with disability in research
Musicka-Williams[41]	Australia	Intellectual disability	Qualitative	Not applicable	Children	Assent strategies of adolescents with intellectual disability in research
Taylor & Balandin[42]	Australia	Complex communication needs	Literature review	Not applicable	Does not specify	Ethical strategies to include those with complex communication needs in research
Wark et al[43]	Australia	Intellectual disability	Descriptive case study	10	Older adults	Consent processes in research with individuals with intellectual disabilities
Cascio et al[44]	Canada	Autism	Literature review	Not applicable	Children and working age adults	Involvement and decision-making process of individuals with autism in research
Jaiswal et al[45]	India	Deafblind	Case study	16	Working age adults	Challenges in obtaining informed consent and participation of individuals with deafblindness in research
Carey & Griffiths[46]	Ireland	Intellectual disability	Qualitative	12	Working age adults	Decision-making process of adults with intellectual disabilities
Doody[47]	Ireland	Intellectual disability	Literature review	Not applicable	Does not specify	Consent and inclusion process of people with intellectual disability in research
Murphy et al[48]	Ireland	Dementia	Literature review and randomized controlled trial	304	Does not specify	Consent strategies for inclusion of persons with dementia in research
McNeilly et al[49]	Ireland	Cognitive impairment, autism physical disability	Mixed methods approach	18	Children and working age adults	Recruitment, consent, and confidentiality issues in research with those with disabilities
Haines[50]	United Kingdom	Intellectual disability	Case study	5	Working age adults	Recruitment and inclusion in the consent process in individuals that lack capacity in research
Hampson & Morris[51]	United Kingdom	Dementia	Literature review	Not applicable	Does not specify	Inclusion of people with dementia in research through consent processes
Prusaczyk et al[52]	United States	Cognitively impaired	Literature review	Not applicable	Older adults	Gaps in the informed consent process in those with cognitive impairment in social work research

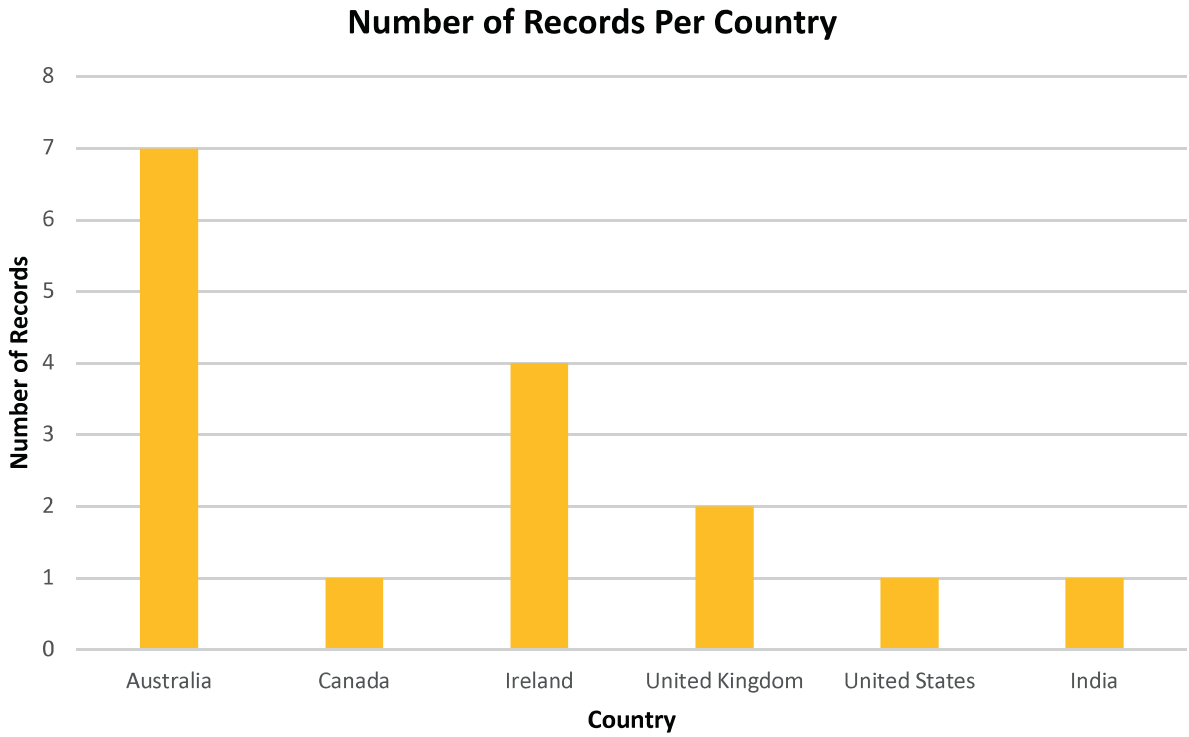

Fig 2 Number of records per country.

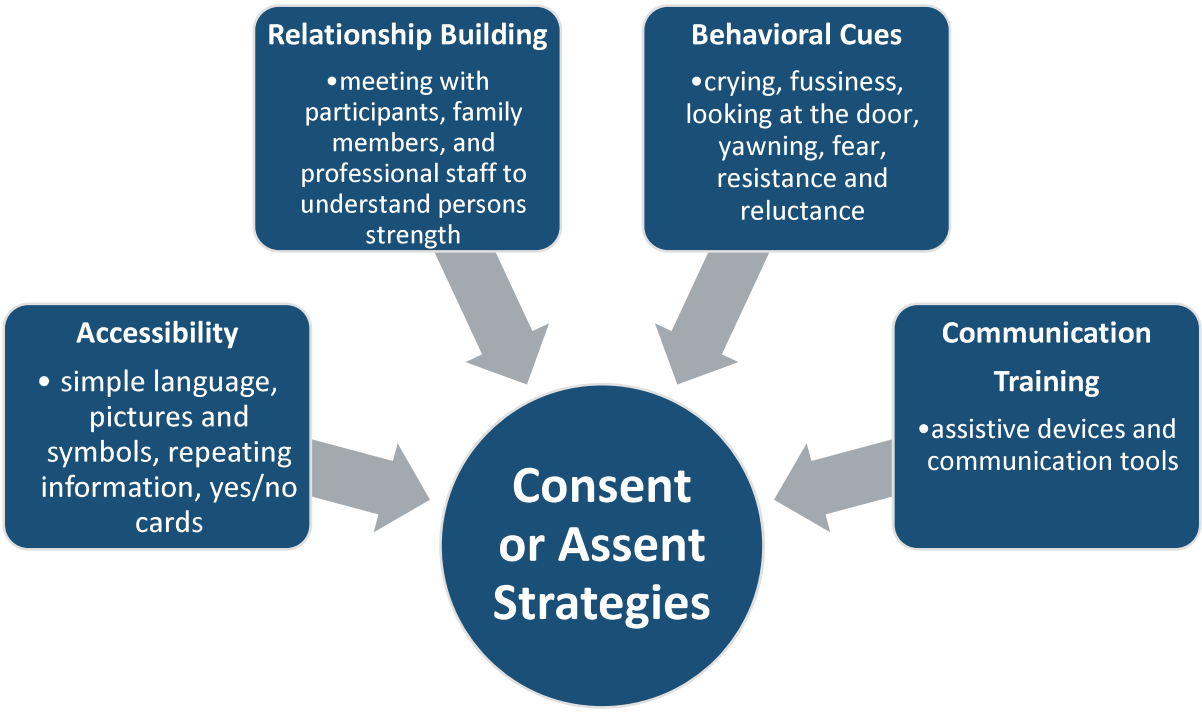

Fig 3 Consent or assent strategies for researchers.

Older adults

Similar to the 2 other younger groups, it was suggested that using simple, plain language with pictures or verbally communicating the information would be helpful.[43,52] One study used yes/no cards and sign language to evaluate the participant's ability to provide consent.[43]

Relationship building between researchers, participants, and caregivers

When working with individuals with intellectual disabilities or dementia, a common strategy identified across studies was researchers building relationships with participants and

Table 3 Suggestive strategies for individuals with sensory and cognitive disabilities across age groups

Strategies	Children Ages 0-19 y	Working Age Adults Ages 20-59 y	Older Adults Age ≥60 y
Accessibility	Simple language, pictures and symbols, pictures of researchers, cameras, drawing, tape recorders using a doll that children can communicate with, or thumbs up/down stickers. Adults with disabilities make a consent form for children with disabilities. Use of role playing to act out the consent process scenario. Showing a DVD film of the research activities that will take place.	Plain language and clearly worded information sheets. Forms consist of bullet points, pictures, and information repeated and verbally communicated. Providing a choice of preferred documentation such as more text and fewer pictures and vice versa; braille; large print.	Using a yes/no card and sign language to evaluate consent. Providing corrective feedback to help improve comprehension.
Relationship building	Meet with the child multiple times. Engage in free play and include tools that will be used in the study during these visits.	Share some vulnerability to show value and genuine interest. Can help the researcher understand the capacity of the participant and willingness to participate in the study. Including family members or professional staff to help communicate information and ensure consent.	Spending time with the participant to understand limitations with language and to create strategies or cues tailored to them.
Nonverbal cues	Dissent: constantly looking at the door, lack of eye contact with the researcher, signs of boredom such as yawning, fussiness, silence, crying, lack of cooperation, refusal, resistance, fear, asking questions such as, "When will I be done?" or saying "I'm tired." Providing a stop card for participants that can be used to indicate withdrawal. Using a talking mat that has a goodbye symbol to indicate withdrawal.	Facial expressions of participants and asking individuals familiar with the participant to help interpret the meaning of bodily cues.	Meeting over a long period of time to understand body cues, signs of discomfort, or uncertainty.
Communication training	Nothing specific to this group.	Nothing specific to this group.	Training for assistive devices and communication tools being used in the study, as well as in interpreting behavioral cues.

their caretakers. Researchers established relationships by scheduling multiple visits before the study and using interactive tools to develop a better understanding of the participant's communication style, build rapport, and develop trust.[9,37-41,44,46-48] Researchers engaged with caretakers or family members to gain a better understanding of each participant's needs.[46] Building relationships with people close to the study participant can help with assessing consent by helping interpret facial expressions or behavioral cues as a sign of engagement or dissent.[9,39,50] One researcher attempted to develop a relationship with participants through voluntarily being vulnerable and available to participants to chat and help in their life to address the power imbalance that typically exists between researchers and participants.[38] Some of the studies reported tips or strategies relevant to the specific age groups.

Children

For this group, it was recommended that researchers schedule multiple visits to build trust and rapport, gain familiarity with the participant's communication style, and aim to ease anxiety in participants.[40,41] These visits should include free play using tools that will be used in the research study to create a stronger rapport and understand the communication method of the child.[40] In addition, parents of children with hearing or communication impairments acted as interpreters when conducting a study; however, it was important for researchers to affirm their role and ensure that they did not speak on behalf of the child.[40]

Working-age adults

In this group, each meeting helped researchers to understand the capacity of the participant and their willingness to participate in the study.[39] One researcher involved family members and staff in helping share information with participants and in obtaining consent.[9]

Older adults

For older adults, some authors recommended spending time with the individual to determine the person's strengths and limitations with language and to understand the person's situation to create strategies or cues tailored toward them for the study.[48]

Behavioral cues of participants

Researchers may observe participants for behavioral cues and signs of dissent that can alert disengagement from the study. These signs include fussiness, lack of cooperation, resistance, fear, facial expressions, sounds, or signs of discomfort.[39,40,43,44,46,49-51] On the contrary, signs of engagement included recurrent attempts to grab test materials, refusing to leave the research site after the session was over, smiling, and staying the entire length of the study session.[39,44] To best identify a participant's interest or dissent in a study, researchers asked individuals close to the participant, such as family members or therapists.[39,43,50]

Behavioral cues are helpful yet subjective, and researchers must use their best judgment. For instance, in 1 study the participant kept repeating, "I don't think we will be there" when the researcher was scheduling a meeting at the participant's house. The researcher later realized the participant was uncomfortable conducting the study at the participant's house and preferred to conduct the study at another location.[46] Initially, the researcher was unsure if this was a sign of dissent and relied on previous positive meetings with the participant before providing an alternative option.[46]

With respect to the various age categories, signs of engagement or dissent may include the following:

Children

Behavioral signs such as constantly looking at the door, lack of eye contact with the researcher, signs of boredom such as yawning, fussiness, fear, resistance, silence, crying, and lack of cooperation can signal a child's refusal to a study.[40,44] On the other hand, verbal cues can be explicit or indirect. The guide explained comments such as "I'm tired" or "When will I be done?" or "I want to go the toilet" are examples of the child's withdrawal from the study.[40] Tools, including using a stop card, can be given to indicate withdrawal from the session.[44] Another study allowed participants to use the goodbye symbol on the Talking Mat as a sign of withdrawal from the study.[49]

Working-age adults

In this group, facial expressions of participants or body language were other examples of indicating dissent. Researchers discussed the body cues with individuals familiar with the participant to determine if it is a sign of dissent.[39,50] The authors explained that when an individual did something, made a certain sound, or had a specific facial expression, it can imply 1 of many emotions. Furthermore, individuals may verbally consent to the study, but if their body language appeared not to be engaged or interested, then it was up to the researcher to use their discretion and exclude them from the study.[50] Researchers can also ask questions such as, "Is it ok with you if I meet you again?" to ensure consent was provided throughout the entire study.[46]

Older adults

For older adults, meeting with individuals with dementia over an extended period of time was significant in helping researchers understand participant's body cues.[51] One study involved 3 researchers who observed the participant during the consent process for signs of dissent, such as discomfort and uncertainty.[43] At the end of the informed consent stage, the researcher's notes were tallied, reviewed by a fourth individual, and discussed in the case of a disagreement.[43]

Communication training

To understand behavioral cues and how communication technologies work, researchers received training ahead of the study.[43,48] One source began training researchers 6 weeks before the actual data collection phase,[43] whereas another study provided a 2-day training program.[48] One of the studies was conducting research in individuals with dementia and included research assistants that were trained nurses who had experience working with individuals with dementia.[48] None of the strategies were specific to children or working-age adults. However, for older adults, there was training for participants' assistive technologies, communication tools being used in the study such as pictorial cards, and pictures of the study participant either happy or sad.[43] Furthermore, training was provided to detect and interpret behavioral cues and to determine positive or negative facial expressions, physical posture, and hand gestures of participants.[43]

Discussion

The purpose of this review was to synthesize the research literature concerning strategies that researchers can use to obtain informed consent/assent when working with individuals with deafblindness. Strategies such as the accessibility of the consent process, relationship building, behavioral cues of participants, and training related to participant

communication style/methods or assistive technologies are helpful in obtaining consent. When researchers meet with participants prior to the study, they will develop a comprehension of the participant's communication style and be able to recognize behavioral cues that can signal dissent. Many of these strategies are applicable across all age groups.

Paucity of research in deafblindness

There were no articles identified in vision and hearing impairments regarding consent strategies for nonclinical research studies, showing that the lack of literature including the population with deafblindness is not an outlier. The lack of studies from LMICs can be attributed to only including studies in English, lack of recognition of deafblindness as a distinct disability, and accessibility of health resources to individuals with deafblindness in LMICs.[5] The 2018 World Federation of the Deafblind report found only 36% of individuals' needs from LMICs were adequately covered by their national health system, compared with 55% in high-income countries.[5] The disproportion in accessibility and health care funding within LMICs for individuals with deafblindness make it difficult for researchers to study this population, leading to a significant gap in evidence.

To address this, we suggest that future researchers collaborate with organizations that serve individuals with deafblindness for support. This review found 1 researcher's study related to participation of individuals with deafblindness in India was supported by professional staff/interpreters of a research partner organization to help determine the most suitable format of consent form for all participants.[9] Despite disadvantages in health systems, the strategies identified here are still applicable and can be adapted to the contextual variations of available resources. We recommend conducting studies in LMIC where there is a higher prevalence of individuals with disabilities such as deafblindness.[55] The experiences from LMIC studies will be significant to indicate if there are other strategies to obtain consent/assent unique to the circumstances in these countries. Furthermore, researchers including participants with deafblindness in the future are encouraged to document their consent process by including a detailed description of steps taken. In addition, by 2050, it is expected 16% of the global population will be 65 years and older, compared with only 9% in 2019.[56] Given this expected increase, researchers will require consent strategies that are suitable to older adults with deafblindness or other comorbidities (eg, dementia) in their studies. Additionally, individuals with congenital or acquired deafblindness are aging as well. This unique population will be important to understand because their experiences will be different from those who become deafblind at an older age.

Use of capacity tools/assessments

Some studies focused on the capacity of participants with a disability to provide consent or through a proxy. A common practice is using capacity tools such as the Mini-Mental State Examination or the MacArthur Competence Assessment Tool for Clinical Research.[48,57] These assessments can also be helpful in determining an individual's communication challenges but should not be used to make them ineligible without adaptations. Studies have deemed participants illegible to consent because they did not pass these assessments.[58,59] It is recommended, instead of merely determining an individual's capacity, that researchers alter their methods to include participants with disabilities. On the contrary, in the study by Wark et al., the researchers created a 7-step process to determine the consent capacity of individuals with intellectual disabilities.[43] They ensured each participant had an assistant they were familiar with to verbally ask the questions and used support tools to help with understanding the questions. The researchers also observed participants to identify any bodily cues indicating dissent. By including support tools, they were able to obtain consent from 7 of the 10 participants, showing that, with the right adaptations, most individuals have the capability to consent. Therefore, we recommend that future studies adapt this type of capacity assessment before excluding individuals.

In addition, researchers have used assistive technologies, such as refreshable braille devices and Skype audio channel, to obtain consent from individuals with deafblindness.[45,60] Another study reported the development of a specialized communication device that involves individuals with deafblindness wearing a glove and the hearing/sighted communication partner holding a keypad. The partner can transmit messages to sensors in the glove, and the individual with deafblindness receives the message as vibrations on the palm.[61] Given that 2 main challenges of including the population with deafblindness in research are access to information and communication, future research can use technology to help mitigate these challenges and assist with communicating directly to participants.

Researcher capacity

The key to informed consent is that participants understand the information being conveyed to them, meaning researchers must consider the best way to transfer the information to facilitate access and understanding. For example, written documentation may be supplemented with audio or visual aids. When language barriers necessitate the assistance of an intermediary for communication between the researcher and participants, an intervenor (one who "facilitates the interaction of the person who is deafblind with other people and the environment"[29(p3)]) or sign language expert should be selected.[16] Future studies need to adapt to the communication style of their participants and determine the best tools or support personnel to ensure informed consent can be obtained.

In addition, more time will be required because researchers have to repeat information and read out loud at a slower pace. For instance, Beattie et al. found when obtaining consent from a proxy for individuals with dementia, obtaining consent took about 20 minutes more in individuals with dementia.[37] Another study gave participants 2 weeks to review the study documents and provided additional time for participants to understand and answer questions.[9] Researchers need to allocate time for receiving training on using assistive technologies, understanding a participant's communication style, designing accessible consent forms,

and, in some cases, designing forms that are individualized to each participant.[9,37] One study scheduled training for the project 6 weeks before the data collection phase.[43] We suggest that researchers who include individuals with deafblindness allocate additional time to ensure their staff can receive adequate training and adhere to each individual's communication style.

Significant disability event

The year 2017 saw the highest number of published articles (n=5) compared with other years with only 2 or 3 articles published per year. This may be in response to the 2016 Social Forum's theme of promotion of full and equal enjoyment of all human rights and fundamental freedoms by all persons with disabilities.[62] Statistics highlighted in the forum found persons with disabilities have the worst living conditions and lower participation rates in public affairs compared with other groups.[62] This observation may have sparked greater interest in researchers to study individuals with disabilities, leading to a higher number of studies published in 2017 compared to other years.

Study limitations

Because of the limited amount of research conducted with individuals living with deafblindness, and specifically within areas related to consent/assent strategies in research, the scope of disabilities had to be broadened to include other disabilities. However, given that we were able to identify strategies to obtain consent among individuals with sensory and cognitive disabilities that are frequently diagnosed among individuals with deafblindness, this study will be helpful for future research in obtaining consent from a population with deafblindness. In addition, this review only reported on articles published from 2015 onwards, thereby excluding possible relevant literature published before 2015. This review was focused on obtaining the most current, legally relevant literature available and identifying regulations and guidelines that are applicable to present time and advancements. Only studies with a full text available in English were included. Lastly, because this article was a scoping review, the methodological rigor and quality of the studies was not evaluated.

Conclusions

Despite growing momentum on inclusion, equality, accessibility, and full and effective individual participation in society as outlined in the United Nations Convention of Rights of Persons with Disabilities, major gaps remain in guidelines to help researchers obtain informed consent/assent from individuals with deafblindness. This review is the first of its kind to provide an overview of such strategies and to help bridge the gap between researchers and individuals with deafblindness during the research process. These findings are of significance to disability and rehabilitation researchers, organizations, and individuals with deafblindness because they can significantly advance their inclusion in research and strengthen evidence on the issues individuals with deafblindness face. Although strategies to obtain consent from the population with deafblindness remain unclear, this review encourages researchers to alter and adapt the strategies identified and to create an inclusive research environment that can help improve the lives of individuals with deafblindness. We recommend that policymakers create ethics codes that will guide researchers conducting studies with individuals living with deafblindness to ensure informed consent is obtained directly from the participant. With the growing population across the world, the voices of millions of individuals with deafblindness are unheard in research, even though they have the potential to contribute toward the improvement of their quality of life.

Suppliers

a. Mendeley Desktop Version 1.19.6; Mendeley.
b. Covidence; Veritas Health Innovation.
c. Microsoft Excel 2016; Microsoft Corporation.

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