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ARTICLE

"All doctors should be trained in that": The coproduction and mixed-methods evaluation of an educational toolkit to enable safe, high-quality genetic health care for people with intellectual disability

Iva Strnadová[1,2,3], Manjekah Dunn[4,5], Chloe Molnar[4], Julie Loblinzk Refalo[1,3], Jackie Leach Scully[2], Joanne Danker[1], Michelle Tso[1], Tiffany Qing Lim[4], Yasmin Cathcart-King[4], Karen-Maia Jackaman[1], Sarah Hayes[1], Sierra Angelina Willow[1], Jackie Boyle[6], Jennifer Hansen[1], Skie Sarfaraz[3], Caroline Basckin[1], Celia Halliburton[7], Thulasee Sri Ganeshan[8], Edwina K. Middleton[8], Bronwyn Terrill[9,10,11], Elizabeth Emma Palmer[4,5,*]

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ABSTRACT

Purpose: People with intellectual disability inequitably access high-quality genetic health care. However, they are keen to understand genetic health care and recommend that clinicians need education on delivering more inclusive care and that multimodal genetic health literacy resources should be coproduced.

Methods: Our inclusive research team applied best-practice coproduction principles to deliver a suite of resources, the GeneEQUAL Toolkit. Mixed-methods evaluation, including surveys and focus group/interviews, assessed (1) clinicians' perceived capabilities, motivation, and opportunities for providing inclusive health care for people with intellectual disability before and after exploring the Toolkit; (2) the perceptions and opinions of people with intellectual disability about the Toolkit; (3) the reach of the Toolkit components; and (4) the reflections of people with intellectual disability and clinicians on the coproduction process.

Results: The Toolkit met the expectations and preferences of people with intellectual disability and clinicians, and had a global reach. Coproduction was feasible and judged as critical for the high value of the Toolkit, in motivating clinicians to change their clinical practice and empowering people with intellectual disability.

Conclusion: Coproduction can be successfully applied to improve the engagement of people with intellectual disability, potentially reducing health inequity and improving the safety and quality of genetic health care.

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*Correspondence and requests for materials should be addressed to Elizabeth Emma Palmer, Discipline of Paediatrics and Child Health, School of Clinical Medicine, Faculty of Medicine and Health, Level 8, Bright Alliance Building, Cnr Avoca and High Street, UNSW Sydney, Randwick, NSW 2031, Australia.
Email address: elizabeth.palmer@unsw.edu.au

Affiliations are at the end of the document.

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Introduction

Research has highlighted that health care for the 1.7% of the global population with intellectual disabilities is inequitable and frequently of low quality and unsafe, with a high prevalence of avoidable deaths and trauma.[1] Genetic medicine, that is, genetic counseling, testing, and potentially genetic-based therapies, has the potential to improve physical and mental health outcomes through precision medicine, patient empowerment, better health literacy, and the provision of genetic counseling.[2] Genetic counseling and diagnoses can have a profound impact, but opportunities to translate a diagnosis into tailored health care, appropriate support, peer connections, and reproductive planning are often missed. Our previous research showed that people with intellectual disability are missing out on such opportunities.[3,4] Access to genetic health care services is inequitable, there are numerous barriers in the informed consent process, and, in line with international literature,[5] we heard that people with intellectual disability have a high incidence of trauma, and their genetic health care experiences may be associated with further trauma.[3]

Our inclusive research team comprised coresearchers with intellectual disability, clinicians, and researchers in disability studies, bioethics, and special education. We aim to create a community of shared research practices in which a wide range of stakeholders play roles in knowledge creation. The most important stakeholders are people with intellectual disability. Our research process also incorporates the voices of health services and government representatives, clinicians, parents, and support people, teachers, and other researchers. Our ethos aligns with the fundamental values of coproduction: genuine power-sharing and democratic relationships between the different individuals, groups, and institutions involved.[3] In line with best practice and current guidelines, we also believe health care should be inclusive, which we define as health care that actively adapts its practices to ensure full participation of people with intellectual disability.

In our previous research, a prioritized list of recommendations was generated by people with intellectual disability,[3] with the potential to improve genetic health care and health outcomes for people with intellectual disability. The recommendations were supported by a Multi-Stakeholder Advisory Committee that included clinicians, academics, government representatives, and representatives from self-advocacy and patient organizations. Our program aimed to coproduce an Educational Toolkit in response to the top 3 recommendations: (1) a suite of accessible point-of-care resources for genetic health care education and awareness should be available, (2) resources should be coproduced with people with intellectual disability to improve the accessibility of genetic medicine, and (3) education for clinicians should be tailored to improve knowledge and skills in, and the motivation to apply, best-practice genetics health care for people with intellectual disability.

The Toolkit was developed in New South Wales (NSW), Australia, as a key initiative of the NSW Health Genomics Strategy[6] to improve knowledge, skills, and capabilities in the health care workforce, optimize patient care, and provide safe, cost-effective, equitable, and beneficial genomic health care. The Toolkit was published on the NSW Health Centre for Genetics Education website (www.genetics.edu.au) to maximize accessibility to health care professionals and was linked to the GeneEQUAL website (www.geneequal.com).

We report an evaluation of the Toolkit and coproduction process, addressing the following research questions:

1. What are clinicians' capabilities, motivation levels, and perceived opportunities for providing inclusive health care for people with intellectual disability before and after exploring the Toolkit?
2. What are the perceptions and opinions of people with intellectual disability about the Toolkit?
3. What is the reach of the Toolkit components?
4. What do people with intellectual disability and clinicians think about the coproduction process?

An Easy Read version of this paper is available in the Supplemental Appendix.

Materials and Methods

Coproduction of the Toolkit

The Toolkit was coproduced by a multidisciplinary group of educators, clinicians, trainees, and coresearchers with intellectual disability, following best practices in coproduction outlined by the Co-production in Action guidelines (Figure 1, Supplemental Material).[7] Diverse perspectives shaped the coproduction of the Toolkit, gathered through a series of coproduction workshops with people with intellectual disability, advisory committee meetings with multiple stakeholders, including self-advocate organizations, clinicians, government representatives, organizations supporting people with rare genetic conditions and intellectual disability, and regular meetings of the inclusive research team and with government. The Toolkit was coproduced in NSW and developed and evaluated using a program logic

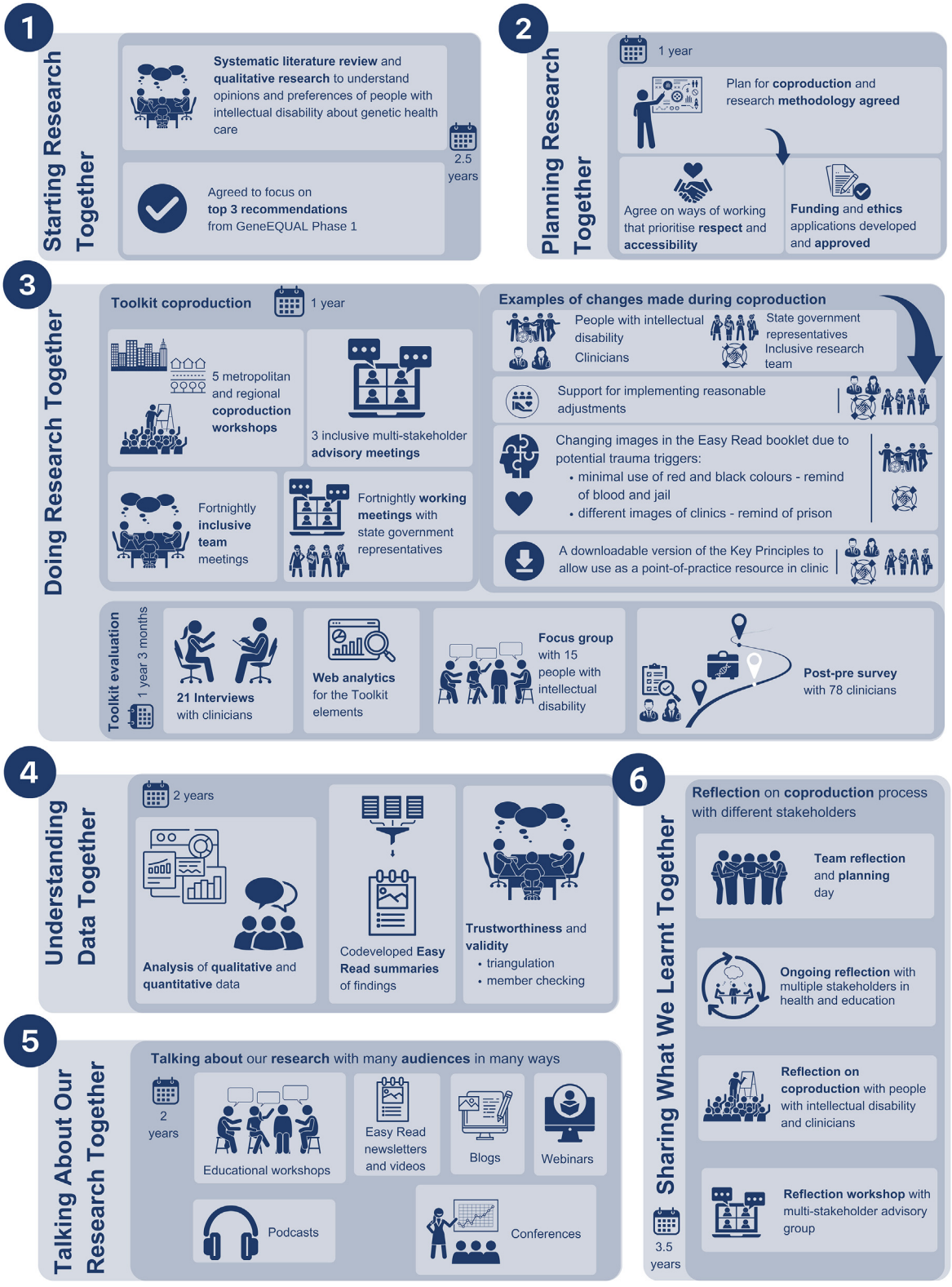

Figure 1 **Overview and timeline of the intervention (coproduction of the Toolkit) and its evaluation.** This image was drawn by the authors using Canva.

model.[8] Toolkit development is reported according to the *Rise2Genomics* reporting standards and SQUIRE 2.0 Reporting Standards for Quality Improvement.[9,10]

The aim of the Toolkit is to support clinicians involved in delivering genetic health care to make reasonable adjustments that ensure patients with intellectual

disability receive inclusive care. The Toolkit includes advice for clinicians (Key Learning Principles), videos contrasting poor practice with inclusive practice, and Easy Read genetics booklets (Figure 2).

An Easy Read report on the coproduction and evaluation of the Toolkit was written for reporting to the funder and dissemination on the team's website (www.geneequal.com), copresenting the Toolkit and reflections on coproduction in multiple formats.

Evaluation of the Toolkit

The Toolkit was evaluated using a mixed-methods study design, including quantitative surveys of clinicians' perceptions, and qualitative focus group/interviews with clinicians and individuals with intellectual disability.

Access and engagement with resources

Google Analytics was obtained for websites hosting the Toolkit from May 31, 2023 to July 18, 2024 including page views, downloads, unique users, and data on visitors' locations. Video views and watch time were obtained from Vimeo Analytics for the same period.

Quantitative methods

The survey was designed with multistakeholder involvement and contained questions adapted from the literature[11,12] and purpose-built questions, including 5-point Likert Scales (Supplemental Material). There were questions about demographics, as well as the quality, content, and perceived impact of the Toolkit. In brief, the survey used the Capability, Opportunity, and Motivation Model of Behavior (COM-B) model within a post-pre design to evaluate the Toolkit's impact on perceived knowledge and confidence, awareness of resources, and understanding of the need for inclusive practice.[13] The COM-B model examines factors that influence behavior and is a useful tool for evaluating behavior-change intervention methods.[13] It is particularly valuable within the public health setting[13] and has been used to identify barriers and facilitators to behavior change in health workers.[14,15]

The survey was completed by clinicians (including those in training and qualified professionals) at a single time point after engaging with any component of the Toolkit. Questions were asked about their perceptions of their capability, opportunity, and motivation to deliver inclusive, person-centered, and respectful genetic questions after (post) and before (pre) interacting with the Toolkit in the same survey. This post-pre design aimed to avoid potential bias about knowledge of inclusive health care before Toolkit engagement and only needed to be completed once, maximizing participant uptake.[16]

The survey included 50 questions and took an average of 10 minutes to complete. There was also an opportunity for participants to respond to open-ended questions at the end of the survey to provide more feedback.

The inclusion criteria were clinicians who were either training or qualified, aged 18 years or older, and who had interacted with the Toolkit. The survey was hosted on a secure online Research Electronic Data Capture (REDCap) platform, hosted and managed by Research Technology Services (University of New South Wales Sydney), and accessible via a publicly available link on the GeneEQUAL and Centre for Genetics Education websites. It was promoted among networks of the GeneEQUAL team and GeneEQUAL Advisory Committee, at conference and workshop presentations, and on social media. The survey was disseminated through the Centre for Genetics Education and GeneEQUAL websites and social media, conference and workshop presentations, and social media.

Participation was voluntary, and participants provided online consent before commencing the survey. In line with Australian competition rules, an incentive to win an e-gift card or iPad upon completion was added to boost recruitment.

Survey data collected from 1 March 2023 to 30 June 2024 were deidentified and analyzed using IBM SPSS Statistics (Version 27.0). Descriptive statistics and frequencies were used to analyze data on demographics, engagement, and overall evaluation. Dependent paired t tests were used to analyze data from the 5-point Likert scale, and Cronbach's alpha was calculated to determine the internal consistency of the survey items related to capability and motivation. It was not possible to determine the internal consistency of the survey items related to opportunity because only 1 question was asked. A statistical significance (P) value of $< .05$ was considered significant. For more details, see Supplemental Materials.

Qualitative methods

Qualitative data were reported following the Standards for Reporting Qualitative Research.[17]

Clinician interviews

Qualitative interviews explored individual perspectives on integrating the Toolkit into clinical practice and their reflections on the value of coproduction.[18,19] Clinician participants were recruited via convenience sampling. Qualified or trained clinicians aged 18 years or older who expressed interest in being contacted for an interview, either directly to a member of the GeneEQUAL team or through the survey between March 1 and October 17, 2023, were emailed. All participants provided written informed consent before the interviews. Participation was voluntary, and those who completed the interview after September 1 were given coffee vouchers.

The interviews were semistructured, conducted by 3 members of the research group, and recorded using Zoom and a second audio-recording device. The interview guide was developed by the inclusive research team to explore

Figure 2 Components and reach of the Toolkit as of July 2024. Permission to show images from the websites were granted by the GeneEQUAL team and NSW Health Centre for Genetics Education.

clinicians' overall impressions of the Toolkit and preferences for future development and was codesigned by the inclusive GeneEQUAL research team informed by a preceding literature review.[4]

Focus group with people with intellectual disability

A semistructured focus group was conducted with people with intellectual disability who had participated in the coproduction process through previous workshops but were seeing the final version of the Toolkit for the first time. Information about the focus group was shared through a grassroots self-advocacy group, and participants were recruited via convenience sampling with Easy Read consent forms and study information provided. Written consent was obtained before the focus group with a coresearcher (J.L.R.) and a researcher with experience in research with people with intellectual disability (I.S.), with whom most participants were familiar.

This focus group explored the thoughts and feedback of people with intellectual disability on the Toolkit and the coproduction process for the Toolkit, following a semi-structured focus group protocol, which was codesigned by the inclusive GeneEQUAL team. The focus group lasted 3 hours and included regular breaks and varied activities to maintain participant engagement. The session was held in person at a self-advocacy organization in metropolitan Sydney, and audio recorded. The focus group was led by an experienced coresearcher (J.L.R.) and supported by researchers with extensive experience in research with people with intellectual disability (I.S.) and genetic health care (E.E.P., J.B., and M.D.). Their expertise and trusted relationships were essential to ensure that this large focus group was accessible and inclusive.

Data analysis

Recording transcripts were deidentified and then analyzed using inductive content analysis.[20] Two researchers

independently open coded 1 interview, and after resolving any differences, 1 researcher open coded all remaining transcripts, and another researcher checked coding accuracy. For the focus group, 2 independent researchers open coded 10% of the transcript to ensure intercoder reliability, and 1 researcher open coded the remainder. Initially open coding 10% of the data enabled the establishment of a robust foundation for the coding framework while ensuring thorough discussion of any discrepancies between coders early in the process. This percentage provided sufficient data to identify emerging patterns while allowing detailed discussions between coders to ensure consistency in the approach.

Separate researchers clustered the codes into categories and themes, providing general descriptions of the phenomena. The themes were discussed and refined by the team as part of researcher triangulation. During this process, different researchers independently coded the data, cross-checked the coding accuracy, and collaboratively refined themes. This approach allows the incorporation of diverse perspectives, including those of coresearchers with intellectual disability, academic researchers, and clinical experts. The choice of researcher triangulation aligned with our coproduction principles and helped ensure that interpretations benefited from multiple viewpoints while maintaining methodological rigor.

Mixed methods

Qualitative data from clinician interviews and quantitative data from the clinician survey were analyzed in parallel and merged for comparison. Interview data were categorized post-analysis according to the COM-B framework to identify areas of confirmation and discordance.[21]

Survey, focus group, and interview participants

Survey

Of the 120 clinicians who consented to participate in the survey, 81 completed at least 1 part, and 74 completed the survey. Participant characteristics are summarized in Supplemental Table 1. Most were female (82%), non-Indigenous (97%), based in Australia (90%), and working in genetics (68%). Ten percent were genetic counselors in training, and 8% were medical students.

Focus group

Fifteen participants with intellectual disability and 1 support person participated in a 3-hour focus group. Participant demographics are summarized in Supplemental Table 2. Seven participants were female, with ages ranging from 23 to 66 years (median 44.5). Three people were identified as Indigenous Oceanians (from Australia or New Zealand); the majority were identified as Oceanians of northwestern European heritage, with 3 individuals identified as Oceanians of southern or eastern European heritage.

Interviews

Of the 32 clinicians offered recruitment, 21 provided informed consent and were interviewed. Participant demographics are summarized in Supplemental Table 3. Seventeen of the participants were female. Twelve identified as Oceanian (from Australia or New Zealand), of which 5 were of northwestern European heritage, and 2 were of Asian heritage. None of the participants identified as Indigenous. Each interview lasted approximately 35 minutes (average 35:19 minutes; range: 21:30-56:08 minutes).

Results

Reach and engagement

The Toolkit had a global reach, with components visited online by over 5000 people from over 53 countries (Figure 2).

Survey

The overall evaluation showed that 96% of the respondents ($n = 71$) agreed or strongly agreed that the Toolkit would improve their quality of practice, and 95% ($n = 70$) agreed or strongly agreed that the Toolkit format was effective (Supplemental Figure 1).

There was a statistically significant improvement in the respondents' reported capability, opportunity, and motivation to deliver inclusive genetic health care after engaging with the Toolkit compared with before engagement (Supplemental Figure 2). The capability subscale consisted of 9 items (before: Cronbach's alpha (α) = 0.946; after: $\alpha = 0.905$) and the motivation subscale consisted of 3 items (before: $\alpha = 0.971$; after: $\alpha = 0.886$) with 1 item for the opportunity subscale.

Interviews and focus group

The focus group involving people with intellectual disability was the accumulation of a series of 5 coproduction workshops (Figure 1) held in metropolitan and regional areas of NSW. Through a series of workshops, the concept of genetics-related health care was extensively discussed, and participants with intellectual disability knew that the Toolkit was about genetics and were very interested in this topic. Furthermore, they understood the concepts included in the Easy Read booklets and videos and found them very relevant to their own health and that of their families.

Analysis of the data from the focus groups identified 3 key themes (Figure 3): (1) health care for people with intellectual disability, (2) feedback on the Toolkit and recommendations for the future, and (3) coproduction in health care for people with intellectual disability. Illustrative quotes are presented in Table 1.

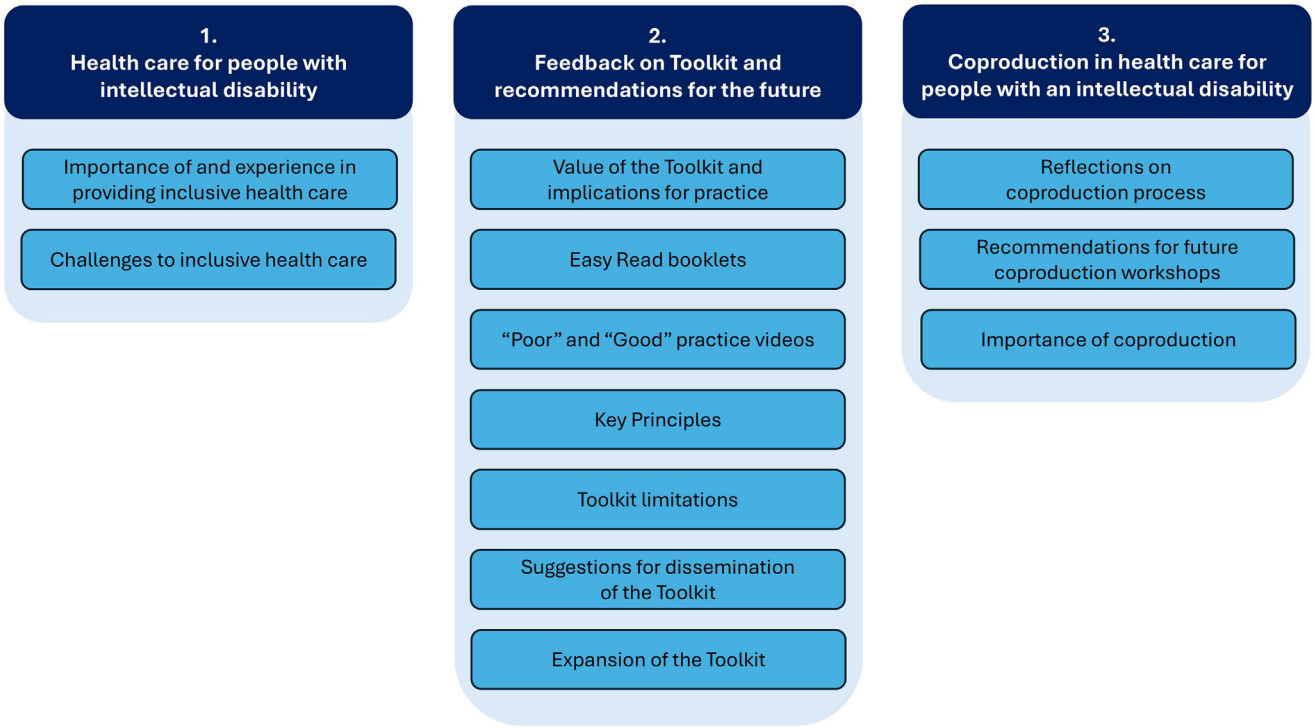

Figure 3 Overarching themes and subthemes from the qualitative evaluation of the Educational Toolkit, including interviews with clinicians and focus groups with people with intellectual disability. Themes are listed in the navy boxes; subthemes are listed in the light blue boxes. This image was drawn by the authors using PowerPoint.

Theme 1: Health care for people with intellectual disability

The importance of inclusive health care was highlighted by clinicians and people with intellectual disability.

Subtheme 1.1: Importance of and experience in providing inclusive health care

Many clinicians (12/21) had experience providing care to people with intellectual disability, and most (18/21) recognized the importance of inclusive practice. Several people with intellectual disability (4/15) also noted the importance of inclusive health care.

Subtheme 1.2: Challenges to inclusive health care

Challenges in providing inclusive health care include insufficient time, limited training, communication difficulties, and obtaining informed consent. Clinicians (6/21) recognized that limited time prevented them from preparing themselves and their patients or providing reasonable adjustments.

Clinicians (11/21) identified limited pre- and postgraduate education in health care for people with intellectual disability and communication principles as significant barriers, while 7 said they had limited knowledge of inclusive health care or relevant legislation.

Providing information in an understandable form was a common challenge (16/21). Five clinicians noted that difficulties in determining an individual's level of understanding reduced their ability to provide information. Clinicians tended to rely on family members, a support person, or past medical records, even though they recognized that this increased the likelihood of the person with intellectual disability being marginalized.

The videos prompted a discussion of personal experiences within the Australian medical system and what people with intellectual disability felt needed to change to ensure more inclusive health care. Discussing the inaccessibility of current health care, participants and a support person shared experiences in which they felt rushed, ignored by health care staff, noticed disrespectful body language, struggled to navigate the health system, or received poor psychological support. People with intellectual disability noted difficulty in complaining about poor health care experiences, including fear of retaliation if they speak up.

Theme 2: Feedback on Toolkit and recommendations for the future

Both the stakeholder groups provided rich feedback on the Toolkit, including how it could be improved.

Subtheme 2.1: Value of the Toolkit and implications for practice

All clinicians had a positive view of the Toolkit, feeling that it targeted both clinicians and people with intellectual disability, contained helpful resources (21/21), and provided an evidence-based framework for changing their own clinical practice, such as making reasonable adjustments and using more inclusive language (16/21). The Toolkit also

Table 1 Illustrative quotes from interviews/focus groups and survey by subtheme

People With Intellectual Disability	Clinicians
Theme 1: Health care for people with intellectual disability	
"I think doctors and nurses don't give enough time… my experience is, you go in there and some of them don't explain things enough to you." Gemma (subtheme 1.2)	"…little equipped…to make those reasonable adjustments…" James (subtheme 1.2)
"But some patients might be scared because they'll think maybe they'll go to them again next time and they'll get treated worse… so they don't complain because they're often afraid to speak up." Gemma (subtheme 1.2)	"…often people are talking around them [the person with intellectual disability] and never at them, or to them…" Milly (subtheme 1.2)
	"…sometimes there's not a power of attorney or a guardian or someone who can provide consent on their behalf, and knows them better than I do, obviously, to make that call." Sophia (subtheme 1.2)
Theme 2: Feedback on Toolkit and recommendations for the future	
"if this becomes part of their training when they first join the post of nurse or doctor, this can be really helpful in guiding a person with intellectual disability." Andrew (subtheme 2.1)	"…as patients begin to understand what kinds of things are okay and are not okay, they're probably going to – hopefully – start using their voice to ask for what they need…" Olivia (subtheme 2.1)
"The way I see how it could be useful for the doctor… it's a guide to talk with them, their patient, to talk to them at, like, the same wavelength as them, in an easy-to-understand conversation." John (subtheme 2.1)	"I'm just really glad that you've done this, because I learnt a lot just by looking at it, and it made me reassess some of my own preconceptions as well, which I didn't realise I had." Sasha (subtheme 2.1)
"It's also good how they're showing pictures of people trying to understand the whole thing, because a lot of people can still find it difficult" Gemma (subtheme 2.2)	"They're [the booklets] fantastic, and.. from my perspective, in my practice, they're perfect. …we don't have anything like this, and they're just what I need. They're really, really helpful." Isabella (subtheme 2.2)
"More empowerment to speak up about situations, when it's about your health, so after seeing that video today, the next time, for example, I see a doctor, it will be like, oh, OK. I'll feel more confident to be able to speak up." Andrew (subtheme 2.3)	"Just by watching the videos, you will actually start to reflect on your own style of, like, communication, and that sort of really kick-starts a series of becoming more and more respectful and more…a more respectful way to communicate with people with intellectual disability…" Charles (subtheme 2.3)
"And family members often don't know how to treat you. They forget that you're an adult half the time, and try to teach you like a child, when you want them to talk to you and treat you like an adult, and let you make decisions for yourself." Gemma (subtheme 2.6)	"So, I think practising trauma-informed care would be an area that I would focus on a bit more going forward." Grace (subtheme 2.4)
"(Create booklets) to teach you, to show your family how to listen to you, and respect your decisions, because with my family, you can tell them whatever you want at that time, and they'll say "yes" to everything, but when you go, it's all different and they'll say, "No, I know better than them!" Gemma (subtheme 2.7)	"…I vaguely knew that this was there, but until we had the talk, I…so I think that, you know, there's not a lot of, you know, there's nothing that's better than actually going out there and talking to people and making it…" Fiona (subtheme 2.6)
	"And if you tell someone – like, if you tell any health professional, "Here's five booklets," you run the risk that they won't look at any of them, whereas if you tell them, "Here's one booklet that might be useful in your practice," then you've got more chance that that will actually be used." Josephine (subtheme 2.6)
Theme 3: Coproduction in health care for people with intellectual disability	
Subtheme 3.1: Reflections on coproduction process	
"(Co-production) benefited us too, in receiving that information about GeneEQUAL and what it's about, and how it can help us." Andrew (subtheme 3.1)	"…it's a really comprehensive toolkit because of the stakeholders that you involved in developing this." Anna (subtheme 3.3)
"We've all been participating as equals for a common purpose." John (subtheme 3.1)	"…as a health professional, it gives me more confidence in the resources to know that these have been trialled, I guess, or have had, you know, valuable input from the people that they are designed for…" Maya (subtheme 3.3)
"Well, what I've learnt over these past consultations, as we've done, is that we can actually, you know, this makes a huge difference to how we access the health system that we needed, and also valuable information that can help us with the future as well." Andrew (subtheme 3.1)	"…the language that we are using has been vetted by people who will be reading it. So, that makes sense that that's who you want to make sure it's clear and understandable and respectful." Emily (subtheme 3.3)
"The benefits of this consultation are just knowing that, as a result of today's GeneEQUAL session, just how much of a benefit and how much will help so many other people into key groups from, yeah, just so many other places." John (subtheme 3.1)	"…and it's always very important to ask about their opinions, because after all, they are the people who will be interacting with…" Charles (subtheme 3.3)

helped clinicians identify the need for systemic changes in the health care system (2/21), with 1 person suggesting that it could help clinicians advocate for change in the workplace.

Seven clinicians believed that the Toolkit would be useful for people with intellectual disability and would help fill a gap in health care resources, and 4 felt that the Toolkit had the potential to empower people with intellectual disability.

People with intellectual disability believed that the Toolkit would be a helpful guide to accessing services and making complaints. Some participants also said that the Toolkit could help clinicians better understand people with intellectual disability (3/15 participants) and that some components could be used by clinicians during an appointment to provide more accessible conversations.

Subtheme 2.2 Easy Read booklets

Seventeen clinicians had viewed at least 1 booklet. Fifteen participants were positive, with 11 commenting that the layout of the booklets contributed to their accessibility.

Fifteen clinicians suggested how the booklets could be integrated into the clinical setting, including sharing booklets with the person with intellectual disability before appointments (10/21). Eleven clinicians discussed using the booklets during the genetics appointment, for example, showing the booklets online (2/21), printing copies (4/21), or annotating the booklets to make them more personal (1/21). Seven clinicians thought that the booklets would be useful to the person with intellectual disability after their appointment. Clinicians commented that booklets could be equally helpful for people with lower health literacy or those who do not speak English as a first language (6/21).

People with intellectual disability reviewed the Easy Read booklet *Tips about genetic health care*. They liked the booklet formatting, felt that the images were appropriate, and contributed to ease of understanding.

Subtheme 2.3 "Poor" and "good" practice videos

All 18 clinicians who viewed the videos thought that they were useful, particularly the contrast between "poor" and "good" practices (15/18). Seven appreciated their realism, although 1 participant found them patronizing, another stated they were unlikely to watch the "poor" practice videos, and 2 were shocked that practice shown in the "poor" practice videos could happen in real life: 1 clinician commented in the survey they "found it hard to believe that anyone could practice so poorly." In stark contrast, people with intellectual disability felt that the "poor" practice videos accurately reflected their experiences in the Australian health care system, whereas the "good" practice videos did not. Only 1 person with an intellectual disability said that her doctor incorporated reasonable adjustments into her health care but added that finding an accessible doctor is very hard.

People with intellectual disability commented that the videos were effective at showing what "poor" and "inclusive" practice is, and empower people with intellectual disability to speak up when they experienced poor health care. A support person attending the focus group reflected that watching the videos helped her recognize disrespectful and inaccessible health care and reconsider how she could support her daughter with intellectual disability.

Subtheme 2.4: Key principles

Because the key principles were designed for use by clinicians, only this group provided feedback. Of the 10 participants who had accessed the key principles, all found them useful, although 7 commented that the third principle, trauma-informed care, was not something they had previously considered within the context of genetics and health care for people with intellectual disability.

Subtheme 2.5: Toolkit limitations

Eleven clinicians discussed the Toolkit's limitations. Four were concerned that clinicians do not have sufficient time to completely explore the Toolkit, while another 4 noted that the Toolkit's resources might not be appropriate for people who could not read, had visual impairments, or had more significant support needs. Some clinicians (7/21) were unsure how to integrate the Toolkit into clinical practice and raised logistical issues, such as how to share the booklets before an appointment (1/21) or over Telehealth (1/21), or resource constraints that would limit their ability to print booklets (3/21). Nevertheless, the Toolkit was seen as a valuable resource for clinicians and people with intellectual disability.

Subtheme 2.6: Suggestions for dissemination of the Toolkit

Effective dissemination is recognized as a requirement for increased engagement. Clinicians suggested engaging with disability support groups (1/21), sharing resources on social media (1/21), distributing hardcopy versions of the resources (2/21), and organizing more educational clinician workshops (3/21).

People with intellectual disability and support people suggested that physical copies of the Toolkit could be made available in clinic waiting rooms. One participant with an intellectual disability commented that many family members do not support people with intellectual disability in making independent decisions.

Subtheme 2.7: Expansion of the Toolkit

Clinicians and people with intellectual disability have suggested that the Toolkit could be expanded to cover more

topics and target a wider audience. They suggest broader ways to improve their implementation in clinical practice.

Fifteen clinicians believed that more educational resources about informed consent would be useful; for example, guidance on making the process as inclusive as possible (3/21) and on determining an individual's capacity for informed consent (2/21). Suggestions for further resources included a checklist on how to practice in an inclusive manner (1/21); a downloadable version of the Key Principles for use as point-of-care resources (3/21); and more educational podcasts, webinars, or videos (5/21). The clinicians believed that the Toolkit would be relevant to all clinicians and students (8/21), including primary care practitioners. Six clinicians commented on the value of additional Toolkit workshops, providing dedicated time to review resources, reflecti on their practice, and consider implementation of the Toolkit. Clinicians and people with intellectual disability highlighted the need to improve clinician education, for example, by incorporating the Toolkit into undergraduate and postgraduate clinician training (4/15 participants; 15/21 clinicians), with several suggesting that it should be a part of mandatory training.

Six clinicians suggested topics for future Easy Read booklets, including (1) genetic testing, including risks and benefits, (2) reproductive rights, (3) medical research, including research pathways for testing, (4) general information about the health system, (5) conversations with family members, and (6) information on specific conditions. Additional topics suggested by people with intellectual disability included (1) disability discrimination in school, (2) the future of genetic health care, (3) participation in genetic research, and (4) resources to support independent health decision making. Clinicians wanted material to support parents and families of children diagnosed with intellectual disability (1/21) and clinical resources in other formats (2/21). Some clinicians (2/21) and people with intellectual disability suggested translating the components of the Toolkit into other languages for people with intellectual disability from non-English-speaking backgrounds. People with intellectual disability recommend creating videos showing the referral process or showing clinicians how to communicate information about medication in an accessible manner.

Theme 3: Coproduction in health care for people with intellectual disability

Subtheme 3.1: Reflections on the coproduction process

One person with intellectual disability commented that participating in coproduction improved their knowledge of health care. Other participants described feeling listened to and treated as equals and enjoying the collaborative effort to improve health care accessibility (4/15). Three participants said the coproduction workshops illustrated how collaborative work can lead to improvements in health care for people with intellectual disability.

Subtheme 3.2: Recommendations for future coproduction workshops

People with intellectual disability made suggestions for improving future coproduction workshops. More written information could be provided about where to seek support, while a joint focus group including people with intellectual disability, support people and clinicians could explore different opinions. However, this has raised the issue of acquiescence and the willingness of people with intellectual disability to speak.

Subtheme 3.3: Importance of coproduction

Most clinicians believed that coproduction was necessary to produce an effective Toolkit (14/21). Five clinicians noted that coproduction allowed them to hear the voices of people with intellectual disability. Three believed that coproduction can be empowering for those involved in the process.

Clinicians acknowledge that coproduction requires careful planning and resources. Seven people noted that limited resources would affect the ability to undertake coproduction (7/21), while 2 thought that it might be challenging to find people who wanted to be involved. It was crucial to ensure that people involved in coproduction were valued and supported throughout the process (3/21), particularly considering the potential for retraumatization (3/21). Overall, the clinicians highly valued that the resources were coproduced, reporting that this increased both the engagement and authenticity of the recommendations.

Mixed-methods findings

The survey findings were overwhelmingly confirmed by qualitative themes. When the interview data were integrated into the COM-B behavior-change model (Figure 4), which guided the development of the survey, it provided additional information about the nature of clinicians' increased perceived capability, opportunity, and motivation to provide inclusive health care.

The clinicians' perceived capability was associated with their trust in the coproduced resources to help them make reasonable adjustments and guide them in using a more inclusive language. They valued the opportunity to implement the Easy Read booklets directly into practice and to advocate for systemic change using an evidence-based Toolkit. Clinicians also reflected on their motivation to model "good" practice after reflecting on good and poor practice and the lived experience of health care by people with intellectual disability. Illustrative quotes are included in the COM-B categories in Figure 4.

Discussion

The mixed-methods evaluation showed that our program met its aims and answered the research questions.

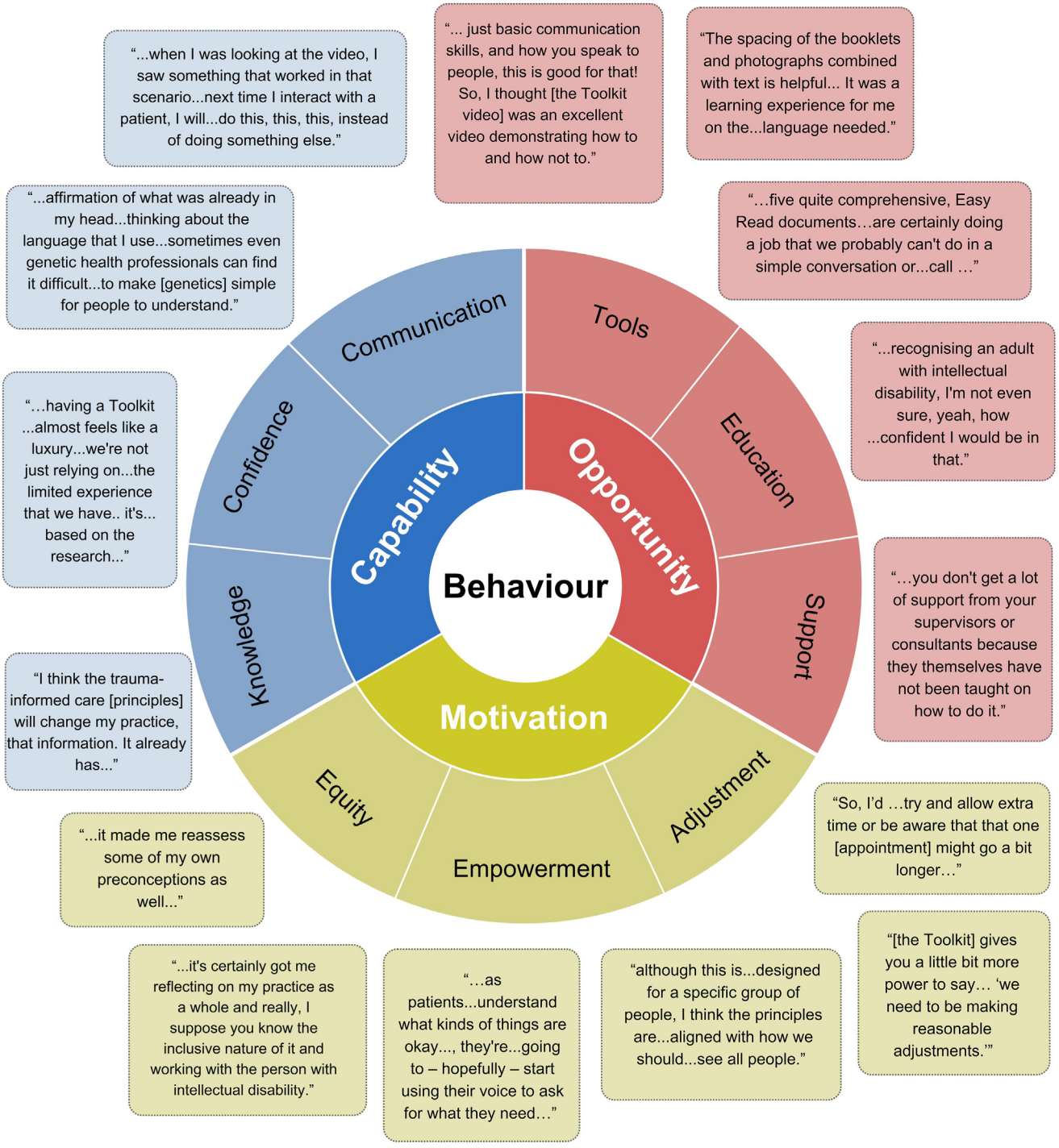

Figure 4 Mixed methods evaluation framework demonstrating how the Toolkit has the potential to improve clinician's practice using the COM-B framework. This image was drawn by the authors using Canva.

(1) Clinicians valued the Toolkit as a high-quality educational resource, with statistically significant improvements in their perceived capability, opportunity, motivation, and behavior to deliver genetic health care; (2) all stakeholders considered the Toolkit included useful point-of-care resources; (3) Toolkit components had a wide global reach; and (4) the coproduction methodology was considered safe and effective.

The coproduction process was a major strength. Co-researchers with intellectual disability were integral at all stages (Figure 1). Iterative feedback from all stakeholders was critical to ensure high-quality resources.

Another strength was that people with intellectual disability acted as coeducators throughout the project.[22,23] The effectiveness of coeducation aligns with recommendations from the World Health Organization that clinicians

have better confidence and communication skills when they learn from people with disability.[24]

Our study was also strengthened by rigorous reflection on potential biases in the research design, data analysis, or interpretation of the findings, including regular meetings with government staff and the advisory committee throughout the project and evaluation.

The need for dedicated education and resources to build clinicians' skills in delivering inclusive health care is supported by numerous other studies.[25-28] There is a gap in studies targeting how to deliver more inclusive genetic health care for people with intellectual disabilty.[29] This publication reported on immediate outcomes (knowledge, skills, confidence, perceived capability, opportunity, and motivation). However, there was some self-reported evidence of more intermediate outcomes (shifts in practice, information seeking, and interest in increasing competence at a systemic level). The multimodal delivery of the Toolkit was designed to reduce the opportunity costs of making reasonable adjustments, as mandated by state health care policy, by providing ready-made materials.

Some clinicians identified areas in which the Toolkit could be expanded or requested to adapt it to their specific practice context. The extensive international reach of the resources suggests the Toolkit could influence practice both nationally and internationally. A recurrent concern is how to extend the reach of the Toolkit to clinicians with the least motivation to change their practice. Exploration of enhanced delivery mechanisms and novel behavior-change approaches, such as peer mentoring within health services, as has recently been introduced in the United Kingdom,[30] will be important, as well as the inclusion of the Toolkit in mandatory training. It was striking that, although people with intellectual disability felt that the "poor" practice videos accurately reflected their experiences, some clinicians felt that they were unrealistic. Others said they had not appreciated the importance of trauma-informed care for individuals with intellectual disability. The recent Australian Royal Commission into Violence, Abuse, Neglect, and Exploitation of People with Disability (2023)[31] explicitly links health care trauma with poor health outcomes, for example, by deterring people from cancer screening. There are similar findings for Indigenous and culturally and linguistically diverse communities. Our findings emphasize the need for ongoing work to address clinicians' awareness of the trauma experienced by people with intellectual disability in genetic health care and health care generally.

This study had some limitations. First, the Toolkit was developed within a state context, and some materials require revisions to make them more relevant across Australia and beyond. Such changes would need to be coproduced with people with intellectual disability and clinicians and thus require further resourcing.

Second, recruitment for the evaluation was voluntary and may have generated a biased sample of those who viewed the Toolkit positively. The poor practice videos demonstrate how health care can be a source of trauma for people with intellectual disability, as has been reported in our previous qualitative research[3] and in the recent Australian Royal Commission into Violence, Abuse, Neglect, and Exploitation of People with Disability (2023).[31] For example, they showed people with intellectual disability being ignored and referred to in derogatory ways, such as being impaired. However, it was clear from the response of several health professionals who reported that the "poor" practice videos were unrealistic or "slightly verging on patronizing" that these clinicians had gaps in understanding about the current health care experiences of people with intellectual disability. By contrast, the videos resonated strongly with the experiences of people with intellectual disability in our focus group. Indeed, when shown the "poor" practice video, 1 support person commented that it showed "the worst aspects of every appointment we've ever had!" This disparity in perspectives highlights that even among clinicians who chose to engage with the Toolkit, there is a lack of appreciation for the neglect and abuse of people with intellectual disability in our health care systems.

Third, the relatively low completion rates of the survey may reflect Australian clinicians' postpandemic burnout or the length of the survey. Nevertheless, sufficient responses were obtained to achieve statistical significance.

Conclusion

Global engagement with the Toolkit indicates that the Toolkit is a valuable suite of resources, filling the gap in equivalent resources elsewhere.[3] Moreover, the evaluation demonstrated that all stakeholders valued the authentic coproduction of the Toolkit. Our study confirms that the coproduction of health resources and clinician education with people with intellectual disability is not just feasible but best practice and has the potential to be effectively applied to different areas of health care.

Our study supports the ongoing need to call out unsafe and low-quality health care for people with intellectual disability and to codesign innovative approaches to urgently address health care trauma. Such focus is needed to ensure that health care provision is truly safe and of high quality for all.

Data Availability

Deidentified data are available upon request. Additional data and materials, such as data collection forms, data extraction, and analysis templates, can be obtained by contacting the corresponding author.

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Author Contributions

Conceptualization: I.S., M.D., C.M., J.L.R., J.L.S., J.D., M.T., T.Q.L., Y.C.-K., K.-M.J., S.H., S.A.W., J.B., J.H., S.S., C.B., C.H., T.S.G., B.T., E.E.P.; Data Curation: I.S., M.D., C.M., J.D., E.E.P.; Formal Analysis: I.S., M.D., C.M., J.D., E.E.P.; Funding Acquisition: I.S., J.L.S., E.E.P.; Investigation: M.D., C.M., J.L.R., S.S.; Methodology: I.S., J.L.R., J.L.S., J.D., S.S., B.T., E.E.P.; Project Administration: I.S., J.L.S., M.T., C.H., T.S.G., E.M., E.E.P.; Resources: K.-M.J., S.A.W., J.B., T.S.G., E.M., B.T.; Supervision: I.S., J.L.S., J.D., B.T., E.E.P.; Validation: I.S., J.L.R., J.L.S., J.D., T.Q.L., Y.C.-K., K.-M.J., S.H., J.B., J.H., S.S., C.B., E.E.P.; Visualization: M.D., C.M., J.L.R., S.A.W., J.B., J.H., S.S., B.T., E.E.P.; Writing-original draft: I.S., M.D., C.M., J.D., J.B., B.T., E.E.P.; Writing-review and editing: I.S., M.D., C.M., J.L.R., J.L.S., J.D., M.T., T.Q.L., Y.C.-K., K.-M.J., S.H., S.A.W., J.B., J.H., S.S., C.B., C.H., T.S.G., E.M., B.T., E.E.P.

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Ethics Declaration

The coproduction and evaluation of the Toolkit were approved by the University of NSW Human Research Ethics and Clinical Trials Governance (HREC HC210342). All participants gave informed consent before participation, which was subsequently archived. All participant information and consent forms were available in Easy Read, with reasonable adjustments made to support the participants in making an informed choice to participate.

Consistent with best practice in inclusive research, we used a continuous (written and verbal) voluntary consent process to seek consent from participants with intellectual disability before and after data collection. All healthy consumers were appropriately remunerated. A safety plan for all participants was developed in view of the recognized potential of the topics to remind people of previous traumatic experiences.

Important note: The National Statement guiding all research in Australia dictates that researchers need to demonstrate how they will assess the 'capacity' of people with intellectual disability to consent.[32] Having worked with people with intellectual disability for decades, we have difficulty using the term 'capacity' in our study documents because it very much aligns with the outdated medical model of disability. In our experience, and based on the latest research literature, there should be a presumption that people with intellectual disability have the 'capacity to consent' to their involvement in research.[32-34] With appropriate supports and accommodations, people with intellectual disability can provide informed consent.

Conflict of Interest

The authors declare no conflicts of interest.

Additional Information

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Affiliations

[1]School of Education, UNSW Sydney, Sydney, NSW, Australia; [2]Disability Innovation Institute, UNSW Sydney, Sydney, NSW, Australia; [3]Self Advocacy Sydney Inc., Sydney, NSW, Australia; [4]Discipline of Paediatrics and Child Health, School of Clinical Medicine, Faculty of Medicine and Health, UNSW Sydney, NSW, Australia; [5]Sydney Children's Hospitals Network, Randwick, NSW, Australia; [6]Genetics of Learning Disability Service, Waratah, NSW, Australia; [7]NSW Ministry of Health, St Leonards, NSW, Australia; [8]Centre for Genetics Education, Health Education and Training Institute, NSW Health, St Leonards, NSW, Australia; [9]Australian Genomics, Melbourne, Vic, Australia; [10]St Vincent's Healthcare Clinical

Campus, School of Clinical Medicine, Faculty of Medicine and Health, UNSW Sydney, NSW, Australia; [11]Garvan Institute of Medical Research, Darlinghurst, NSW, Australia

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