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Preferred outcome measures in treatments for challenging behaviour in individuals with intellectual disabilities: Results of an inclusive Delphi method

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Abstract

Background: Interventions for challenging behaviours in individuals with intellectual disabilities benefit from outcome monitoring that takes clients' preferences into account. We determined clients' and representatives' preferred outcome domains and measures to secure their involvement in treatment decisions for challenging behaviours.

Method: We used an inclusive Delphi method. A focus group of individuals with mild intellectual disabilities and representatives of those with moderate and severe intellectual disabilities prepared the first round by assisting us in collecting possible outcomes. Panels of individuals with intellectual disabilities and representatives were composed to achieve consensus on instruments for preferred outcome domains.

Results: Preferred outcome domains were behaviour, side-effects of psychotropic drugs, quality of life, daily functioning, caregiver burden and family quality of life. Corresponding outcome measures included self-report, interview and proxy-scales, including spoken versions.

Conclusion: Including the preferred domains on outcomes of interventions for challenging behaviours is recommended. Research on corresponding outcome measures is necessary.

KEYWORDS

challenging behaviours, clients' perspectives, inclusive Delphi method, intellectual disability, outcome measures

1 | BACKGROUND

In people with intellectual disabilities the prevalence of challenging behaviours varies, depending on setting and study-population from approximately 20% in community populations (Bowring et al., 2017) to 80% in populations with multiple and profound disabilities (Poppes et al., 2010). Challenging behaviours include externalising (verbal and physical aggression, aggressive-destructive behaviours, inappropriate

(sexual) behaviour, disruptive behaviour, hyperactivity, and irritability) and internalising behaviours (withdrawn behaviour, self-injurious behaviour, stereotypic behaviour, and lethargy). The presence of challenging behaviours is a risk factor for mental and physical health problems, psychotropic drug use (especially antipsychotics) and lower quality of life (QoL) (Balboni et al., 2020; Koch et al., 2015; Ramerman et al., 2018).

Causes of challenging behaviours may lie in biological, psychological, and environmental factors; assessment and treatment should thus be multidisciplinary. Treatment is preferably

Pieter J. Hoekstra and Annelies de Bildt have equal contribution.

non-pharmacological as the effectiveness of psychotropic medication for challenging behaviour has not been proven and harmful side-effects frequently occur (Embregts et al., 2019; NICE guideline, 2015). Systematic monitoring of the effects of pharmacological and non-pharmacological treatments on behaviour and mental functioning and their side-effects is necessary, preferably with standardised scales to increase the validity and comparability of treatment outcomes over time and between interventions. Such monitoring allows for a careful weighing of the advantages and disadvantages of treatments (Embregts et al., 2019; NICE guideline, 2015). This will likely improve shared decision making on which intervention to choose, proceed with or terminate.

Perspectives of clients, their representatives and professionals may differ regarding the need for intervention. Therefore, involvement of all stakeholders in decision making around interventions, including (de)prescribing of psychotropic drugs is necessary (Shankar et al., 2019). Indeed, individuals with intellectual disabilities and their representatives would like to become more involved in decisions regarding the treatment of challenging behaviours, including medication use (Crossley & Withers, 2009; Sheehan et al., 2018; Sheehan et al., 2019). Knowledge about their preferences and perspectives on relevant outcome domains and corresponding measures for the treatment of challenging behaviour may be helpful to increase clients' and representatives' involvement in decision making regarding pharmacological and non-pharmacological treatments.

To secure the active involvement of individuals with intellectual disabilities and representatives in a Delphi study, certain competencies of participants and researchers as described for collaborative inclusive research may be helpful (Embregts et al., 2018), for example, listening and giving feedback as part of communication competencies. Also, to ensure fruitful collaboration between individuals with and without intellectual disabilities, certain adaptations should be made, for example, allowing people with intellectual disabilities enough time to prepare for meetings and study of materials. Other recommendations on how to involve individuals with intellectual disabilities in inclusive research, depending on study design and setting have been provided (Frankena et al., 2019). For example, in a collaborative study design researchers should pay attention to facilitate the process by providing accessible information and to make the process worthwhile on the personal level through gaining new experiences and feeling responsible.

The main aim of this study was to determine outcome domains and corresponding outcome measures to evaluate interventions for challenging behaviours (including psychotropic drug prescriptions and non-pharmacological interventions) based on the perspectives of individuals with intellectual disabilities and their representatives. A second aim was to evaluate the quality of our collaboration with participants with and without intellectual disabilities in this inclusive project by evaluating adherence to the recommendations for inclusive research as provided by Embregts et al. (2018) and Frankena et al. (2019).

2 | METHODS

2.1 | Ethics

All participants had given informed consent to participate in the project.

The study was approved by the Medical Ethics Review Board of the University Medical Center Groningen (UMCG RR number 201900160). All other Dutch data protection rules and research codes of conducts were adhered to as applicable.

2.2 | Design

We used a Delphi method for determining outcomes that individuals with intellectual disabilities and their representatives perceive as important for interventions regarding challenging behaviours.

A Delphi method is a standardised way to collect opinions, views, and preferences of experts and/or stakeholders on certain topics and to achieve consensus on statements on those topics. The method includes an iterative procedure aimed to reach group agreement by asking the experts or stakeholders on the topic (i.e., the respondents and forming a Delphi panel) to independently (often online) answer questions or respond to statements. Statements or questions are adapted between consecutive rounds based on all the respondents' reactions to the previous round. Usually, and as we did in this study, for the first round a questionnaire is prepared based on literature review and experts' opinions on the Delphi topic. Mostly, in three to four rounds consensus is reached. Recommendations for the number of participants in a Delphi panel vary from 10 to 15 when participants are stakeholders and/or their background is homogeneous to 50 when qualifications differ (Hsu & Sandford, 2007).

The Delphi method has been used successfully with participants with intellectual disabilities (Bonell et al., 2012; Salgado et al., 2018).

2.3 | Participants

Participants were individuals with a mild intellectual disability and representatives of individuals with intellectual disabilities. Representatives could be legal representatives, parents, brothers, sisters or other family members, or advocates affiliated to interest groups or organisations.

2.4 | Procedures

The study took place from January 2019 until April 2021. In this inclusive research project, the first Delphi round was prepared in a collaboration of researchers and individuals with and without intellectual disabilities. Therefore, the first two authors composed a focus group, consisting of experts by experience with a mild intellectual disability

and representatives of individuals with moderate, severe and profound intellectual disabilities who (had) received treatments for challenging behaviour and (had) used psychotropic drugs. The aim was to determine the outcome domains of interest and corresponding measures, and to develop a questionnaire with statements on the outcome measures covering the domains. Additionally, a Delphi panel was composed to respond to statements about preferred outcome domains and measures, consisting of the focus group and additional members recruited by the focus group members. The panel was split in a panel of representatives (panel 1) and a panel of participants with intellectual disabilities (panel 2). The focus group and Delphi panels were led by the first two authors of whom the first is next to researcher an experienced clinician in the field.

Focus group and panel members were recruited from a large interest group of people with intellectual disabilities in the Netherlands and from clients' councils of Dutch organisations and interest groups of representatives for individuals with intellectual disabilities.

2.4.1 | Adaptations to facilitate inclusive research

A coach, specialised in guiding experts by experience with intellectual disabilities and developing accessible materials, was available to advise us on the accessibility of materials used in our study, to provide support during focus group meetings, and to assist the participants with intellectual disabilities in understanding the process and the content. Participants with intellectual disabilities could ask the second author or their own caregiver for help when they experienced difficulties in completing questionnaires and/or participating in the focus group. Caregivers were instructed that their clients should autonomously respond to questions and statements. Participants could also use spoken versions of the treatment outcome measures and Delphi statements.

All information materials and questionnaires we used in this study were available in accessible form by adaptation of the language, adding of pictures and adaptation of Likert scales (five-points scale and use of emoticons).

We organised separate information sessions for the focus group participants with and without intellectual disabilities.

Last, to monitor the impact of the collaboration, we asked feedback about potential negative experiences of participants with and without intellectual disabilities in taking part in our inclusive study in evaluation questionnaires.

All participants received a fee and travel cost compensation if they had attended face to face meetings and gift vouchers for taking part in the Delphi rounds.

2.4.2 | Focus group

Written information about challenging behaviour, medication use, effects and side-effects of psychotropic drugs, quality of life, and

caregiver burden as potential domains of interest and the measuring of these concepts as potential outcome measures covering these domains was composed by the first author and the coach. The information was provided 2 weeks before a meeting should take place to the focus group members to allow them to prepare the meetings. For participants with intellectual disabilities, the materials had been edited in an accessible easy-to-read form.

Two whole-day focus group meetings took place. Both days contained explanation on the provided materials by (invited) researchers who were experts in these fields, and discussion of participants' opinions and experiences. This took place in plenary sessions and in subgroup sessions for participants with and without intellectual disabilities. Participants also discussed their preferences on outcome domains, and they could add or remove domains according to their experiences and preferences. Additionally, in the first meeting invited speakers provided information on research teams that included experts by experience with and without intellectual disabilities (Sergeant et al., 2021), and the second meeting provided information on participating in a Delphi process.

Furthermore, participants reviewed the treatment outcome measures presented in the information materials for each of the domains. Participants could indicate their preferred method of treatment outcome measuring (self-report, proxy-, and/or professionals' report), and whether they perceived the existing scales sufficient or whether they thought new ones should be constructed to cover more precisely what they thought to be important aspects of the domains. In case participants would prefer a new instrument, the subjects/items on such questionnaire would be determined at the end of the second meeting. Then, a new easy-to-read-draft questionnaire would be composed by the authors, to be completed by the main caregiver of the client, meant as a base for the first Delphi round, in addition to the other selected outcome measures. In case existing instruments would be preferred, the instruments from the information materials, which were reviewed by the focus group members and covered the preferred domains would be presented in the first round. In case outcome domains were added by the participants, the first author would select additional instruments from the literature covering these domains, using the same criteria as described below (2.5), and add these to the first round.

After each focus group meeting, in order to evaluate whether the project was conducted according to the recommendations for collaborative inclusive research (Embregts et al., 2018), all the focus group members completed questionnaires, which we had constructed for this project (see supplement). The questionnaires also contained open text fields for comments. Agreement with the statements in the questionnaires was rated at 5-point Likert scales (range: totally disagree-totally agree). The first statements evaluated the opinions of the focus group participants on the quality of collaboration of the focus group in general. The other statements evaluated the collaboration on discussing and selecting the domains and measures in the first and second focus group meeting, respectively.

2.4.3 | Delphi panels

The two panels (one with representatives and one with participants with intellectual disabilities) independently aimed to achieve consensus on the preferred outcome domains for the evaluation of the effects of treatments for challenging behaviour and its corresponding outcome measures. To increase the homogeneity of the background of the participants in the panel regarding knowledge, those participants who had not been part of the focus group also received the information materials on the outcome domains and measurements.

Participants were asked to respond to statements on the suitability and clarity of the newly constructed draft questionnaire and existing outcome measures that covered the preferred domains. Respondents could indicate their level of agreement with each statement on Likert scales, ranging from totally disagree to totally agree. We initially aimed to use a nine-point Likert scale. Because of concerns of the coach and their own feedback that a nine-point scale could be too difficult for participants with intellectual disabilities we included the possibility to replace the nine-point Likert scale by a seven-point or five-point Likert scale.

Participants were also asked to indicate in open text fields whether domains or items should be added, adjusted or removed. Between consecutive rounds, statements were changed according to participants' responses in previous rounds until consensus was achieved.

We also assessed the opinions of participants of the panels on the Delphi method by adding open questions to the last round.

2.5 | Potential outcome domains and measures

The outcome domains which were outlined in the information materials and presented in the focus group meetings were based on a selection by the first author of domains often used in mental health-care for outcome monitoring. These included treatment effectiveness, safety of treatments, QoL and patients' and carers' experiences (Chester, 2019; Roe, 2015). Regarding carers' experiences, caregiver-burden was also included. Additionally, she selected instruments to measure the domains behaviour, side-effects and QoL based on a systematic review of diagnostic instruments, scales and questionnaires from the Dutch guidelines on problem behaviour in adults with intellectual disabilities (Embregts et al., 2019) and a selection of the Dutch & Flemish Association of Psychiatrists (<https://www.tijdschriftvoorpsychiatrie.nl/meetinstrumenten>). Given the applicability in clinical practice availability in Dutch language, validity and reliability and user-friendliness were main criteria to select instruments.

Below follows an explanation of the selected instruments by domain.

Domain Behaviour/effectiveness of treatment

The Aberrant Behaviour Checklist (ABC) (Aman et al., 1985) and Behaviour Problem Inventory for people with profound and multiple

disabilities (BPI-PIMD) (Lambrechts et al., 2009) are generic, widely used, validated informant-based scales. The Brief Symptom Inventory (BSI) is a 53-item self-report generic questionnaire on mental symptoms, which is reliable for use in the Netherlands in patients with mild intellectual disabilities (Wieland et al., 2012).

Domain Side-effects psychotropic drugs/safety of treatment.

The Matson Evaluation of Drug Side-effects Scale (MEDS) (Matson et al., 1998) is a comprehensive interview scale validated for the use in people with intellectual disabilities. A Dutch version is available.

The Liverpool University Neuroleptic Side Effect Rating Scale (LUNERS) (van Strien et al., 2015) is a user-friendly self-report scale, but may be less suitable for people with mild intellectual disabilities, because of supposed difficulties in understanding of the wording and the length of the scale. Hence, for those patients the Subjects Response to Antipsychotics short version (SRA-34) (Lako, 2013) may be a better choice. Both instruments are validated and available in Dutch language.

Domain QoL

The Personal Outcomes Scale (POS) (Claes et al., 2010) is a user-friendly, comprehensive, and validated scale, available as interview/self-report and interview/proxy with subscales on mental and physical wellbeing. The Caregivers Concern- Quality of Life Scale (CC-QoLs) (Unwin & Deb, 2014) includes two subscales: one on concerns of the caregiver about the physical and mental health and quality of daily care of the person they are responsible for, and the other on QoL of that person. Because it is a proxy-scale that does not take much time to complete it was translated in Dutch in a back-and-forth procedure with the authors' permission.

2.6 | Analyses

2.6.1 | Delphi method

As criterion for sufficient agreement on statements between participants in the two Delphi Panels we required that >70% of participants rated >3 on 5-point Likert scales (van Hecke, 2015), and >70% of participants rated >4 on 7-point Likert scales (Walsh et al., 2018). To analyse the decision on consensus on the 9-points Likert scales, we used three criteria of the appropriateness method by the Research AND Development (RAND) Corporation and the University of California at Los Angeles (UCLA). These were the median rating, the inter-percentile range (IPR) and the inter-percentile range adjusted for symmetry (IPRAS). The median was used to indicate central tendency. IPRAS was calculated to measure the level of dispersion of the ratings. IPRAS is the threshold beyond which the IPR for a particular item indicates disagreement (Fitch, 2001; Zuidema et al., 2015). Consensus was achieved for statements with a median between 7 and 9, and on which there was agreement when the IPRAS was controlled for.

2.6.2 | Quality of the inclusive research

We calculated frequencies of the scores on the questionnaires for evaluating the quality of conducting inclusive research.

3 | RESULTS

3.1 | Participants

The focus group consisted of three individuals with a mild intellectual disability and experience with challenging behaviour/psychotropic drug use and 10 representatives of individuals with intellectual disabilities who experienced challenging behaviour and (had) used psychotropic drugs. They recruited five additional participants with intellectual disabilities and five without for the Delphi-panel from their networks. However, one participant with an intellectual disability and two without withdrew before the start of the Delphi-rounds. Additionally, two participants with intellectual disabilities and one without withdrew during the Delphi rounds. Lack of time, too much burden or burden of care for their relative were most mentioned reasons for withdrawal.

Therefore, the Delphi panel consisted of five participants with intellectual disabilities and 12 representatives. Most representatives were parents and affiliated to clients' councils. All participants with intellectual disabilities made use of living facilities and care from service providers. In the Supplementary Table 1 more characteristics are shown.

Especially recruitment of individuals with a mild intellectual disability was difficult, sometimes because their paid caregivers thought taking part in inclusive research would be too burdensome for their clients and did not allow us to ask them to participate.

Some participants with intellectual disabilities asked technical assistance when completing the questionnaires from the coach, the second author or their caregivers. Therefore, an extra session for explanation was organised led by the second author and coach. Yet, all participants independently responded to the questions and statements.

3.2 | Preferred outcome domains and outcome measures

In discussions during the focus group meetings participants and researchers the various outcome domains as provided in the information materials were defined as:

- Challenging behaviour is any behaviour that someone displays that is a challenge for others to manage and/or puts the person or others at risks.
- Psychotropic drugs are drugs that affect behaviour, mood, thoughts or perception; in this study limited to prescription of licenced medication.

- Quality of life is the standard of health, comfort, and happiness experienced by an individual or group. In this study we used the eight domains of quality of life as described by Claes (Claes et al., 2010) as a guideline.

- Caregiver burden is the strain or load borne by a person who cares for a chronically ill, disabled, or elderly person.

Participants judged that the severity and kind of challenging behaviour, side-effects of psychotropic drugs and quality of life were all important outcome domains for the treatment of challenging behaviour. They added the domain 'Daily Functioning' as an indicator for the negative consequences of the challenging behaviour itself and the side-effects of treatments as important from patients' perspectives. They added the domain 'Family Quality of Life' next to caregiver-burden as an indicator for the negative consequences of the challenging behaviour for siblings or peers in the person's living group as important from clients' and representatives' perspectives.

These domains were defined as:

- Daily functioning or every day functioning refers to an individuals' abilities to (more or less) autonomously perform basic and instrumental activities of daily living. In this study self-care as well as social functioning and participation are included in the daily functioning of an individual with intellectual disability.
- Family quality of life is the degree to which individuals experience their own quality of life within the family context.

Regarding the outcome measures, initially focus group members preferred to newly construct an individually tailored outcome measure to monitor treatment effects covering all preferred outcome domains. Therefore, we constructed an easy-to-read-draft questionnaire with items covering the determined domains, which should be completed by the main caregiver. Focus group members judged statements on the suitability and clarity of the items on a nine-point Likert scale. Participants with intellectual disabilities indicated that completing a nine-point scale was too difficult for them, and they needed on-site support. Although there was consensus regarding the clarity of the draft-questionnaire, comments on the suitability were mixed and inconclusive. Moreover, participants with intellectual disabilities indicated they preferred a self-report scale instead of a questionnaire to be completed by their main caregivers. Furthermore, we compared the draft with existing scales covering similar domains and concluded that there was (too) much overlap. Last, we doubted the face validity of the draft-questionnaire. Thus, instead of developing a new scale, the Delphi-panels were presented with existing scales covering the preferred domains with statements on the suitability and clarity of these instruments. For the domain Daily Functioning the first author selected the World Health Organisation Disability Assessment Schedule (WHODAS) (World Health Organization, 2010/Rijks Instituut voor Volksgezondheid en Milieu, 2018). This instrument is available in Dutch language as a proxy, self-report and interview instrument, and in short and comprehensive form (12 and 36 questions, respectively) (Federici et al., 2017), covering the domain Daily functioning, including

TABLE 1 Preferred outcome scales and recommendations for the evaluation of treatments for challenging behaviour

Delphi panel ^a	Behaviour	Side-effects psychotropic drugs	Quality of life	Daily functioning	Caregiver burden/family quality of life
Delphi panel 1	ABC recommendation: The BPI-PIMD could be added for individuals with severe and multiple disabilities.	MEDS recommendations: The LUNSERS could be added when antipsychotics are prescribed as monotherapy in patients with mild intellectual disabilities. A spoken word version should then be provided. To be completed in consultation with a doctor or nurse	POS	WHODAS-12	No scale available recommendation: Research must be conducted concerning how to measure the burden of caregivers of persons with intellectual disabilities and challenging behaviours
Delphi panel 2	BSI recommendations: A spoken word version should be provided. Ask your mentor/supervisor to help complete it.	SRA-34 recommendations: A spoken word version should be provided. Complete the scale by yourself and discuss it with your doctor or complete the scale together with your doctor or nurse.	POS recommendation: The POS must be completed in a conversation with a mentor/supervisor in order get a good idea of what someone with intellectual disabilities thinks and feels.	WHODAS-12 recommendation: The WHODAS must be completed in a conversation with a mentor/supervisor, to get a good idea of what someone with intellectual disabilities is capable of.	

Abbreviations: ABC, aberrant behaviour checklist; BPI-PIMD, behaviour problem inventory for people with profound and multiple disabilities; BSI, brief symptom inventory; MEDS, Matson evaluation of drug side-effects scale; LUNSERS, Liverpool University Neuroleptic Side Effect Rating Scale; SRA-34, subjects response to antipsychotics short version; POS, personal outcomes scale; WHODAS, World Health Organisation Disability Assessment Schedule. ^aAn inclusive Delphi study of representatives of individuals with intellectual disabilities (panel 1) and individuals with mild intellectual disabilities (panel 2).

the physical, mental and social functioning. We could not find an appropriate instrument to measure 'Family Quality of Life' in Dutch intellectual disability care neither for the domain Caregiver burden.

The preferred instruments for the outcome domains are presented in Table 1.

In the first Delphi round, we included a statement on whether the CC-QoLs part one could be used as an indicator for the domain Caregiver-burden.

We used a five-point Likert scale for panel 2 as participants with intellectual disabilities had indicated that a nine- and seven-point scale was too difficult for them and for panel 1 a seven-point-scale in round 1 and nine-point-scales in the other rounds. When agreement on a statement was achieved, this statement was omitted for judgement in the next round. Statements with no agreement were adapted according to comments of the participants.

Based on participants' comments while responding to statements, both panels stressed the importance of careful discussion of clients experiencing potential side-effects of psychotropic drugs in a consult with the nurse or doctor and the availability of spoken versions of the self-report scales for patients with mild intellectual disabilities.

Participants indicated that an instrument on the measuring of the domain 'Caregiver burden' should be developed. Additionally, they recommended further research into the value of the WHODAS-12 among people with intellectual disabilities.

3.3 | Delphi rounds

Figure 1 displays the number of Delphi rounds and number of respondents by round for the representatives (panel 1) and for participants with mild intellectual disabilities (panel 2), respectively. Panel 1 reached consensus on the set of outcome measures after four rounds and panel 2 after three rounds. Participants in panel 2 needed an extra online session led by the second author and coach for explanation on completing the questionnaires. Some of the participants needed support by their own caregivers, for example, technical assistance in answering a questionnaire online. Caregivers were instructed to be careful not to influence the opinions of participants.

3.3.1 | Quality of the inclusive research

Table 2 shows the ratings of the focus group participants regarding their judgement of the quality of the inclusive research collaboration on five themes: mutual relationships, communication, collaboration, awareness of skills and needs and awareness of impact of participation (see supplement for the evaluation questionnaires).

Most participants agreed with statements on a range of quality indicators needed for good inclusive research collaboration.

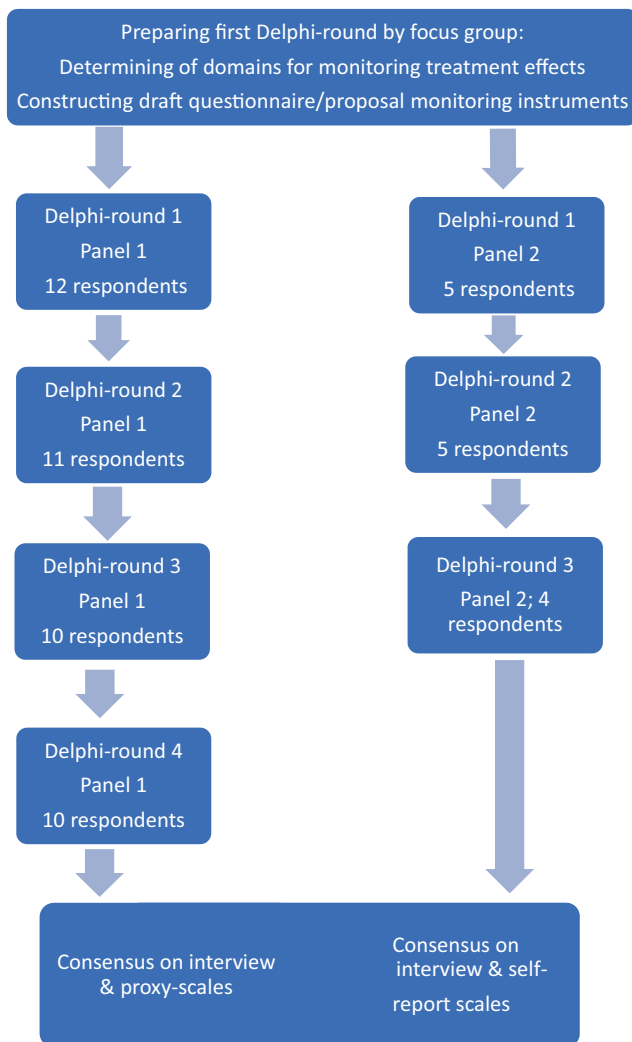


FIGURE 1 Results of the Delphi rounds in an inclusive Delphi method study in achieving consensus on a set of monitoring instruments for the treatment of challenging behaviour in individuals with intellectual disabilities. Panel 1: Twelve representatives of individuals with intellectual disabilities. Panel 2: Five individuals with intellectual disabilities

For some quality indicators the ratings of participants with intellectual disabilities seemed somewhat lower compared to those without intellectual disabilities, especially on the satisfaction regarding researchers' arranging sufficient equipment of meeting location. This is illustrated in Figure 2.

The open questions on 'Outcomes of inclusive research' revealed that representatives as well as participants with intellectual disabilities had gained profits at the personal and professional level. They enjoyed taking part in the study and felt taken seriously, they took responsibility and had gained new research skills and experiences. Also, there were profits for research and healthcare. The Delphi method was appreciated as a method for use in inclusive research. Yet some participants in the Delphi panel, both those with and without intellectual disabilities, who were not part of the focus group, experienced some difficulties in understanding and carrying out the

process. Participants valued the topic of this study as important for healthcare, because the views of patients with intellectual disabilities and their representatives on the treatment of challenging behaviours were accounted for.

4 | DISCUSSION

In this inclusive Delphi method study a group of individuals with mild intellectual disabilities and representatives of individuals with intellectual disabilities determined their preferred outcome domains for monitoring treatments for challenging behaviour. 'Behaviour', 'Side-effects of psychotropic drugs', 'Quality of life', 'Daily Functioning' and 'Caregiver Burden' were all perceived as relevant domains.

It is important to include all these domains when discussing outcomes of treatments for challenging behaviour with clients and their representatives in clinical practice. Moreover, when setting treatment goals, use of these outcome domains will already offer opportunities for clients and their representatives to better weigh the advantages and disadvantages of treatments for challenging behaviour. Including and discussing these domains may thus improve the quality of shared decision making in choosing and evaluating treatments, especially when there is need for psychotropic drug prescription. Also, since in current clinical practice defining treatment goals and monitoring are often limited to specific symptoms and not evaluated more broadly (Carlson et al., 2019), use of all selected domains as treatment outcomes may help to broaden the perspective on treatment goals of professionals, clients, their representatives, and other stakeholders. Perspectives of clients on treatment goals may otherwise remain unknown and/or differences from those of clinicians may become clearer (Chester et al., 2019). Use of all outcome domains will add to securing the patients' perspective on outcomes for treatment (Sheehan et al., 2019). Last, covering all domains will offer opportunities for research based on patient records and for benchmarking between different care providers (Kelly et al., 2021).

The findings in our study regarding the preferred domains are in line with the domains found in the study of Gore et al. (2020). That study identified relevant outcomes of Positive Behaviour Support from the perspective of individuals with intellectual disabilities themselves, caregivers and systems/organisations. The identified domains included individuals' health, emotional wellbeing, participation, and relationships with others, caregivers' emotional wellbeing and team-working. However, the authors recommended to further explore domains at the individual level and measurements to capture the identified domains. The results of the present study may contribute to this exploration, by focusing on outcome domains and potential outcome measures from the perspectives of patients and their representatives.

In the Delphi rounds consensus was achieved for two sets of existing instruments for patients with mild intellectual disabilities and for representatives of those with moderate, severe and profound disabilities, respectively (see Table 1). The participants preferred self-report for individuals with mild intellectual disabilities, and proxy-scales for primary caregivers of those with more severe intellectual

TABLE 2 Judgement of participants with five-point Likert scale^a regarding the achievement of good quality collaboration^b in the Delphi study involving individuals with and without intellectual disabilities

Quality indicators within categories of competencies	Totally disagree	Disagree	Neutral	Agree	Totally agree
A mutual relationship has been built					
Getting to know each other went well (1 item)			22%	22%	56%
Participants felt free to talk about their experiences and thoughts (7 items)			3%	60%	37%
Participants felt supported to talk freely about their experiences and thoughts (6 items)			42%	33%	25%
Communicating					
Listening, understanding, and taking part in discussions went well (9 items)			6%	54%	40%
Accessible written and verbal information were well transferred (12 items)		1%	15%	40%	44%
Achieving a collaboration in which everyone can contribute					
Participants have learned about how to conduct inclusive research (2 items)			16%	73%	11%
Participants have learned about the research topics and to give one's opinion on these went well (15 items)		1%	13%	43%	43%
Practical aspects: We allowed enough time to prepare the meetings (2 items)			17%	39%	44%
Practical aspects: the program and aim of the meetings were clear (4 items)		3%	3%	70%	24%
Practical aspects: Facilitating practicalities went well (2 items)			6%	47%	41%
Being aware of skills and developmental needs of participants					
Addressing the needs for training and support, and ability to deploy knowledge and to reflect (7 items)			8%	67%	25%
Being aware of the impact of participating in research					
We addressed possibly intimidating effect of participating by providing participants support to express themselves in discussions (5 items)			50%	32%	18%

Note: See also supplement for explanation on questionnaires with quality indicators.

^aTotally disagree-disagree-neutral-agree-totally agree.

^bEmbregts et al., 2018. Table does not include all indicators of competencies.

disabilities. They also recommended that the self-report scales should also be provided in spoken versions. However, it must be noted that usability, cost and psychometric properties are important to incorporate in final recommendations, and we did not investigate either of these. We do know that the SRA-34, MEDS, POS and WHODAS-12 are best completed in an interview with a trained clinician, which indicates higher costs than a self-report questionnaire. Furthermore, the psychometric properties of the self-report LUNERS, when used as a proxy-scale should be investigated. Unfortunately, the domain Caregiver burden is not covered by the scales that we had selected. Existing scales, for example, the Zarit-Burden Interview scale for caregivers of patients with dementia (Seng et al., 2010) or Burden Scale for Family Caregivers (Yu et al., 2020) could probably be adapted for use in populations with intellectual disabilities. Finally, since the focus group indicated the need for an individualised tailored instrument, the use of the Goal Attainment Scale (GAS) should be considered (Jones

et al., 2006). This individualised instrument could be used as proxy or as self-report scale, allowing for the tailoring of a concrete treatment goal, and could be added to the selected scales to monitor treatment goals.

4.1 | Quality of inclusive research

Our evaluation questionnaires indicated that the quality of the collaboration with individuals with intellectual disabilities and representatives in the focus group was generally good and the adaptations to compensate for the different qualifications of the participants in the Delphi panel were therefore sufficient. The focus group meetings provided ample opportunities to develop and harness participants' competencies that are needed to conduct inclusive research. Participants felt responsible to contribute to this research project and felt taken seriously as experts by experience as well as co-researchers. Our

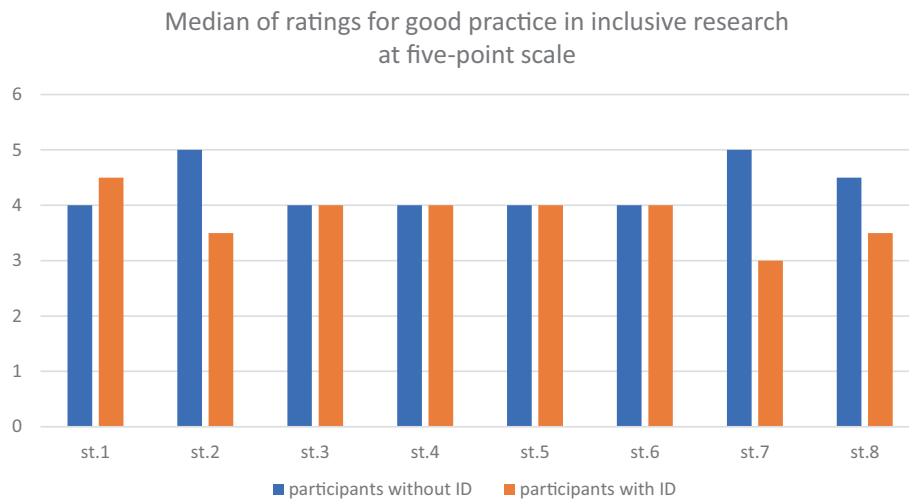


FIGURE 2 Median of ratings at a 5-point Likert scale^a for agreement with statements (st.) on complying with competencies needed for good practicing inclusive research of participants with and without intellectual disability (ID) during two meetings. Indicated are statements on competencies^b within the category 'Achieving a collaboration in which everyone can contribute' which may differently be experienced by participants with intellectual disabilities and those without. See also Table 2. st.1, enough time for reading information to prepare meeting 1; st.2, enough time for reading information to prepare meeting 2; st.3, the program of meeting 1 was clear; st.4, the aim of meeting 1 was clear; st. 5: The program of meeting 2 was clear; st.6, the aim of meeting 2 was clear; st.7, the practicalities organised for meeting 1 were good; st.8, the practicalities organised for meeting 2 were good. ^aTotally disagree (1)-disagree (2)-neutral (3)-agree (4)-totally agree (5). ^bEmbregts et al. (2018). Figure does not include all competencies

design resembled the iterative procedures and focus group meetings advised for participatory research (Bigby et al., 2014).

The responses of participants to the open questions in the last Delphi round also show that the design and methods of using a focus group and Delphi-method were sufficiently tailored to address the study questions (Frankena et al., 2015). Yet, although all participants had experience in the topic of the Delphi rounds as recommended (Hsu & Sandford, 2007), some participants who did not participate in the focus group meetings found it hard to respond to the statements. This finding corroborates the recommendation to organise meetings (Bigby et al., 2014) and indicates that enhancement of the expertise in the Delphi topic and -technique and exchange of experiences and discussion are important.

4.2 | Limitations

A limitation of this study is the small size of the Delphi panel. Although recruitment by the focus group resulted in enough participants for the panel in total, and although we tried to compensate for different qualifications of participants with and without intellectual disabilities by paying attention to the recommendations for inclusive research, we had to split the panel into two groups who were offered different statements. Also, responses in the Delphi rounds were not always complete, and we do not have a complete picture of participants' characteristics. Moreover, most participants in panel 1 were highly motivated family members and advocates of individuals with intellectual disabilities. Therefore, the results of the Delphi-rounds may show shortcomings in generalisability.

Another limitation is that the results concerned only clients' and representatives' perspectives. Other stakeholder's perspectives should be accounted for when choosing a definite set of outcome domains and measures in treatments of challenging behaviours. Moreover, since the discussions in the focus group were led by the first two authors, they may have influenced the opinions of the clients and representatives. Also, the content of the information materials and the outcome measures were selected by the first author which may have influenced participants' choice of domains and outcome measures. We tried to avoid this by following the recommendations for collaborative research by creating a mutual relationship between all the participants and the researchers and a collaboration in which everyone could contribute.

A final limitation is that our evaluation questionnaires that assessed the quality of the inclusive research collaboration were constructed by ourselves. We do not know their validity or reliability.

5 | CONCLUSION AND RECOMMENDATIONS

Outcome domains and measures for the treatment of challenging behaviour in people with intellectual disabilities should consider the perspectives of patients and their representatives. According to our study, preferred outcome domains were 'Behaviour,' 'Side-effects of psychotropic drugs,' 'Quality of Life,' 'Daily Functioning' and 'Caregiver Burden'. Preferences regarding instruments for the monitoring of treatment effects within these domains were self-report scales for clients with mild intellectual disabilities and

proxy-scales for those with moderate, severe and profound intellectual disabilities. For self-report scales spoken versions should be available. Some of the selected instruments are available for direct use in this population, others should be adapted and/or validated or explored.

The Delphi method was appreciated by the participants and feasible in inclusive collaborative research. Organising preparing information meetings on the Delphi topics and method is recommended to enhance expertise and allow for fruitful exchange and discussion.

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CONFLICT OF INTEREST

No conflicts of interest have been declared.

DATA AVAILABILITY STATEMENT

Data of this study is available from the first author upon request (research.cvbp@ggzdrenthe.nl).

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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