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Informant-based assessment instruments for dementia and their measurement properties in persons with intellectual disability: systematic review protocol

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ABSTRACT

Introduction Persons with intellectual disability (ID) are at a higher risk of developing dementia than persons without ID, with an expected earlier onset. Assessment methods for the general population cannot be applied for persons with ID due to their pre-existing intellectual and functional impairments. As there is no agreed-upon measure to assess dementia in persons with ID, multiple instruments for this purpose have been developed and adapted in the past decades. This review aimed to identify all available informant-based instruments for the assessment of dementia in persons with ID, to evaluate and compare them according to their measurement properties, and to provide a recommendation for the most suitable instruments. Additionally, an overview of the amount and quality of research on these instruments will be provided.

Methods and analysis This review will be conducted and reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement. We will adhere to the Consensus-based Standards for the Selection of Health Measurement Instruments (COSMIN) guidelines and use a set of characteristics developed for assessment instruments for persons with ID, the Characteristics of Assessment Instruments for Psychiatric Disorders in Persons with Intellectual Developmental Disorders. Two comprehensive, systematic literature searches will be applied in 10 international databases, including ASSIA, CINAHL, Cochrane Library, ERIC, MEDLINE, PsycINFO, Scopus, Web of Science, OpenGrey and ProQuest Dissertations and Theses Global. Risk of bias and quality assessment will be done according to COSMIN guidelines. We will apply the modified Grading of Recommendations, Assessment, Development and Evaluation approach to rate the overall quality of the available evidence.

Ethics and dissemination No ethics statement is needed for this study. The results will be submitted to a peer-reviewed journal and will be presented at international conferences.

Strengths and limitations of this study

- This review follows the most up-to-date standards for conducting systematic reviews on assessment instruments, the Preferred Reporting Items for Systematic Reviews and Meta-Analyses and Consensus-based Standards for the Selection of Health Measurement Instruments guidelines, and additionally uses the Characteristics of Assessment Instruments for Psychiatric Disorders in Persons with Intellectual Developmental Disorders, a system especially developed for evaluating assessment instruments for psychiatric disorders in persons with intellectual disability.
- Two very comprehensive consecutive search strategies will be applied in a total of 10 international databases, including grey and unpublished literature.
- We use no language restrictions to minimise language bias.
- We include only informant-based instruments assessing dementia in our evaluation and exclude direct cognitive tests.
- Due to expected heterogeneity in studies, a quantitative pooling of psychometric data will probably not be possible.

INTRODUCTION

Intellectual disability (ID) is characterised by limitations in intellectual functioning (IQ<70) and in adaptive behaviour originating in the developmental phase of an individual.[1] It is also known as intellectual developmental disorder in the *Diagnostic and Statistical Manual of Mental Disorders 5* (DSM-5)[2] and Disorders of Intellectual Development in the *11th Revision of the International Classification of Diseases* (ICD-11).[3] Prevalence of ID is hard to establish since in many countries, no official records of persons with ID exist.[4] In large meta-analyses and reviews, the worldwide prevalence of ID is estimated to range from 1.0% to 3.3%.[5–7]

Persons with ID are at the same or at higher risk of developing dementia than persons

without ID.[8–10] Yet, due to their limitations in intellectual functioning, it is often hard to recognise dementia in this population, especially at an early stage. Well-evaluated assessment and screening instruments for the general population, such as the frequently used Mini-Mental State Examination,[11] are not suitable for persons with ID due to their pre-existing disabilities.[12 13] Diagnostic overshadowing[14 15] makes it difficult to distinguish symptoms linked to the pre-existing disability from symptoms caused by the onset of dementia. Additionally, the presentation of dementia in persons with ID can differ from the presentation in persons without ID, with behavioural symptoms and personality changes being more frequent and probably earlier in the course of the illness, especially in persons with Down syndrome (DS).[16 17] To reliably detect dementia in persons with ID, it is recommended to compare a baseline assessment with periodic reassessments.[18–20] Most dementia assessment methods for persons with ID rely on informant-based measures. The respondent of these instruments should be a person who knows the respective person with ID very well, for instance, a family member or care staff. In contrast to direct tests of cognitive functioning, informant-based instruments can be applied for all persons with ID, irrespective of their intellectual and functional capacity.

Early recognition of dementia is particularly important to start early interventions, to plan for the future and to get adequate support for family carers or care staff.[21–23] Not being able to recognise early signs of dementia constitutes a disadvantage for persons with ID and contradicts the Convention on the Rights of Persons with Disabilities by the United Nations (UN-CRPD).[24] Article 25 and 26 of the UN-CRPD require states parties to ensure that persons with disabilities can get the 'highest attainable standard of health without discrimination on the basis of disability'.[24]

There are several tools and screening instruments in use for the early recognition of dementia in persons with ID.[13 25] These instruments can be placed into one of three categories: medical tests (eg, fMRI and gene markers), direct cognitive tests and informant-based scales, which are also called observer-rated scales. In this review, we focus solely on informant-based scales, which include observer-reported outcome measures, as well as clinician-reported outcome measures.[26]

One systematic review found 114 instruments and four test-batteries that have been used to assess dementia in persons with ID. However, some of these instruments have never been designed or adapted to be used in persons with ID, or even to assess dementia.[13] Although there are already some reviews summarising tools and screening instruments in use for assessing dementia in persons with ID,[13 25 27 28] no systematic review on measurement properties using up-to-date guidelines for review conduction and psychometric evaluation has been conducted so far. We want to provide an inventory of available informant-based instruments and their measurement properties. This should help clinicians and research in choosing the adequate instrument for their respective purpose. Our review adds to the existing body of knowledge by using a very inclusive systematic search of the literature and, most importantly, by providing a systematic evaluation of informant-based dementia assessment instruments following up-to-date guidelines.

For each instrument, we will systematically summarise the amount and quality of available evaluation studies, depicting which measurement properties have been evaluated to what extent, and which measurement properties have not been sufficiently evaluated, yet.

The objectives of this systematic review were (1) to identify informant-based instruments suitable for the assessment of dementia in persons with ID, (2) to provide a systematic overview of descriptive aspects for each instrument (eg, respondent requirements and response format), (3) to provide a systematic overview of the amount and quality of available research for each instrument and each measurement property, and (4) to provide a recommendation for the most suitable instruments based on all information collected.

METHODS AND ANALYSIS

This review will be conducted and reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement.[29] The review protocol has been developed using the PRISMA guidelines for protocols (Preferred Reporting Items for Systematic Reviews and Meta-Analyses Protocols).[30 31] We will adhere to the Consensus-based Standards for the Selection of Health Measurement Instruments (COSMIN) guidelines[32] and complement them with a set of characteristics especially developed for assessment instruments for persons with ID, the Characteristics of Assessment Instruments for Psychiatric Disorders in Persons with Intellectual Developmental Disorders (CAPs-IDD).[33] The systematic review has been registered with the International Prospective Register of Systematic Reviews (PROSPERO) with registration number CRD42020181773. If amendments to the protocol are needed, we will register these in PROSPERO, including date and rationale. In the final publication of our results, any amendments to the protocol will be depicted and explained.

Search strategy

Two systematic searches will be applied consecutively and carried out between May 2020 and August 2020. The first search should provide an inventory of available informant-based assessment instruments for dementia in persons with ID. The goal of the second search is to locate evaluation studies for each instrument found in the first search. Figures 1 and 2 depict our search strategies using PRISMA flowcharts.

First search

To identify instruments, we will search in ten international electronic databases, including ASSIA, CINAHL,

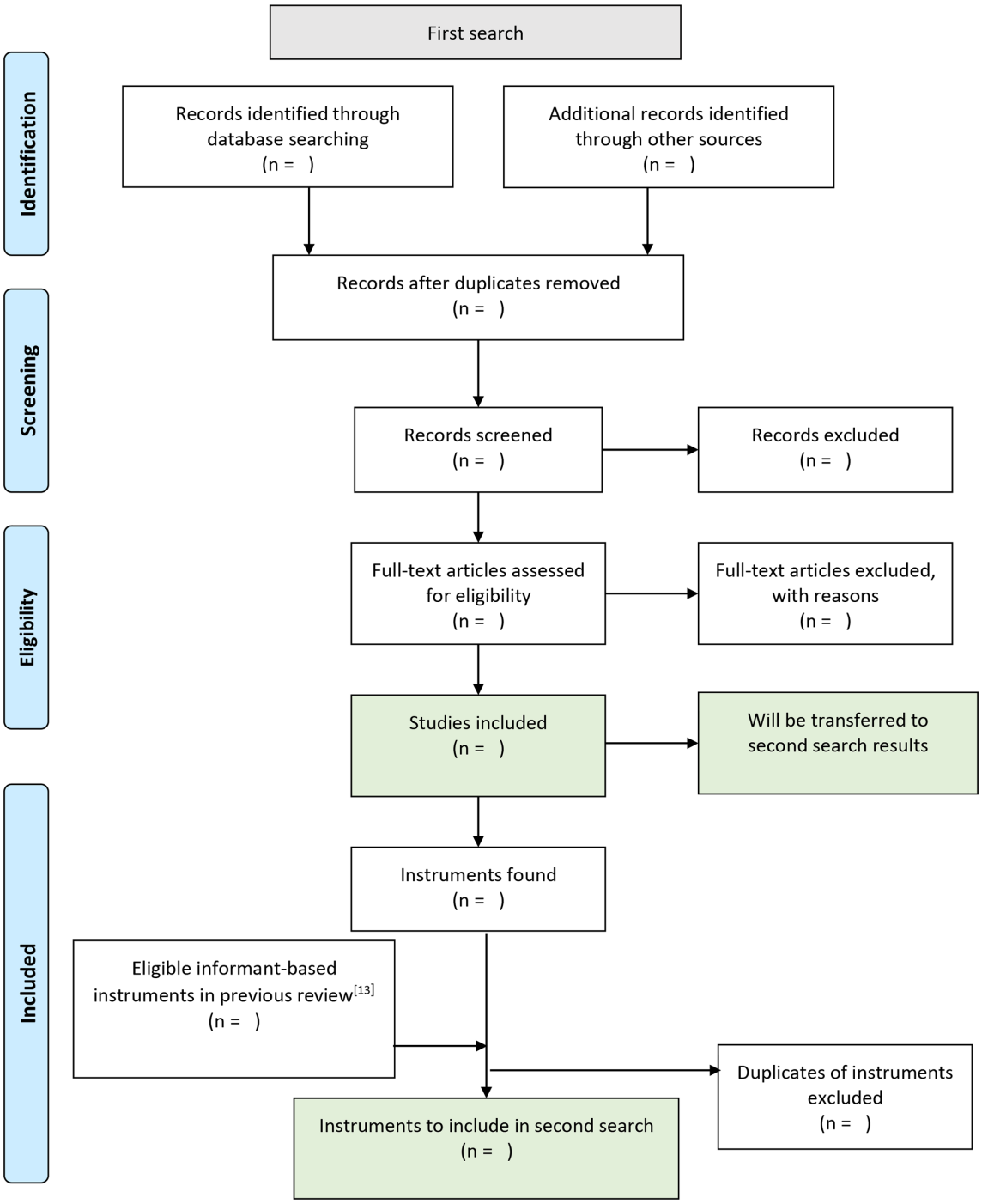

Figure 1 Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart of the first search.

Cochrane Library, ERIC, MEDLINE, PsycINFO, Scopus, Web of Science, OpenGrey and ProQuest Dissertations and Theses Global. The search strategy is described in table 1 and depicted in detail in the online supplemental file. It will include various terms for the (1) output of interest, (2) construct of interest and (3) the specified population. As persons with DS are very prone to develop dementia, this subgroup of persons with ID is included in our search strategy. We will use a limit on the timespan of publication in the first search, not including publications before the year 2012. Instruments published up to the year of 2012 are summarised in a previous systematic review.[13] This review used a very inclusive search strategy and listed all assessment instruments that have been used to assess dementia in persons with ID. We will examine the total of 114 dementia assessment instruments listed in the review of 2013 and include those instruments that are in line with our inclusion criteria.

Inclusion criteria for the first search will be as follows: (1) studies need to focus on assessing dementia in persons with ID; (2) description of the development or evaluation of an informant-based instrument for the assessment of dementia; (3) and this instrument has to be especially developed or adapted for persons with ID.

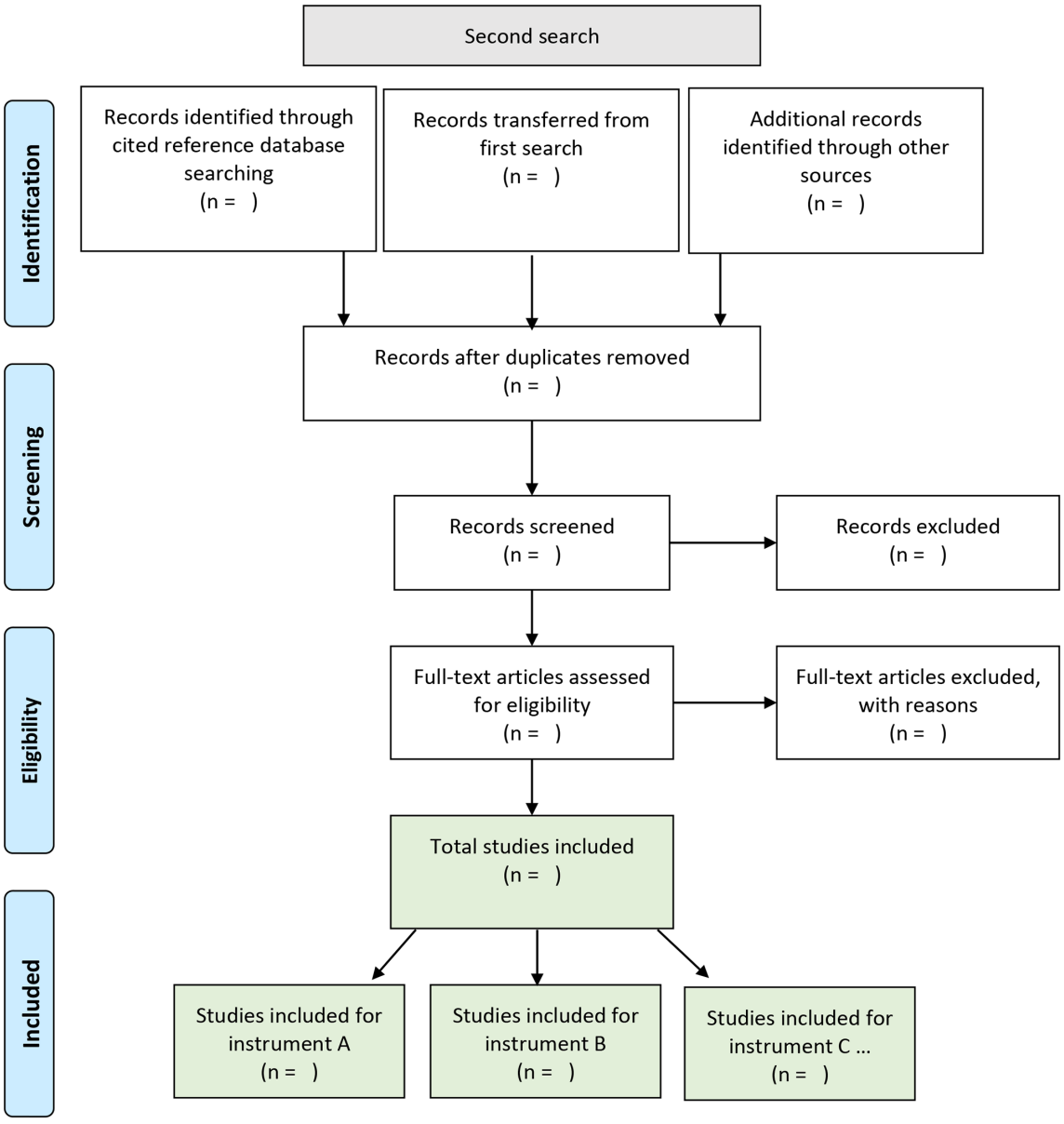

Figure 2 Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart of the second search.

Exclusion criteria include (1) classification systems like ICD-11 and DSM-5, and (2) scales including dementia, but focusing on a broader spectrum of disorders for screening purposes or differential diagnosis, such as the Psychiatric Assessment Schedule for Adult with Developmental Disability.[34]

Second search

Once we have identified the instruments, we will conduct a search by citation strategy using the initial publications of each instrument as a reference point. This search strategy was chosen on the assumption that a paper evaluating an instrument would surely cite the initial publication of the respective instrument. The papers used as reference points will also be included in the further appraisal of the literature. For published papers, we will use five international databases allowing a search by citation strategy, including ERIC, PsycInfo, MEDLINE, Scopus and Web of Science. For published manuals not listed in at least one of the five databases, we will use Google Scholar. Additionally, all records fulfilling the inclusion and exclusion criteria of the first search will be transferred and examined in the second search.

The following inclusion criterion will be used in the second search: studies need to describe an evaluation of the respective instrument in persons with ID. Exclusion criteria comprise (1) the use of the respective instrument primarily for other investigations not related to an evaluation of the instrument or (2) or the study being a review on assessment instruments, not providing novel information.

To further include grey and unpublished literature in both searches, we will apply an invisible college approach, contacting authors in the field for information or articles on this topic, and we will follow up on meeting abstracts.

Table 1 Search strategy for the first search

	(1) Output	(2) Construct	(3) Population
Search terms	Assessment instruments	Dementia	Intellectual disability
Synonyms	assessment; diagnostic; diagnosis; screening; instrument; tool; measurement; questionnaire; psychometrics; scale; interview	dementia; Alzheimer's disease	Intellectual disability; learning disability; intellectual developmental disorder; trisomy 21, Down syndrome
Combined and truncated	assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview*	dement* OR Alzheimer*	((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*)
Example search string for Scopus	TITLE-ABS-KEY ((assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview*) AND (dement* OR alzheimer*) AND (((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*))) AND PUBYEAR >2011		

Full texts of reviews on assessment instruments identified in the course of the two searches will be screened for possible further studies to include. References of papers meeting the inclusion criteria will be hand-searched. We will re-run both searches before the final analyses to include the most recent publications.

For study selection, one reviewer will exclude duplicates. All remaining records will be screened and reviewed for eligibility by two team members independently, that is, blinded to each other's decisions. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in discussion.

Data extraction

The first search will result in a list of instruments. Data extracted will be the names of the instruments and information on their initial publications. In the second search, we will extract evaluation data of instruments, that is, measurement properties and characteristics as listed in the COSMIN checklists and the CAPs-IDD. For each characteristic/property extracted, we will record the study design and sample characteristics, including sample size, gender distribution, age distribution, aetiology of ID and country (language) in which the instrument was evaluated. We will include all studies, irrespective of their design.

The extraction of all relevant data will be done via standardised and piloted Excel spreadsheets (version 16.0) by two team members independently. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in discussion. If data necessary for coding are missing in a study, we will contact the respective study authors for this information.

Risk of bias and quality assessment

Quality and risk of bias will be assessed on study level (for each measurement property), on outcome level (for each assessment instrument) and on an aggregated outcome level, applying the Grading of Recommendations, Assessment, Development and Evaluation (GRADE) approach. We will combine the COSMIN checklists[35–37] with the CAPs-IDD,[33] a comprehensive tool specifically developed for the evaluation of assessment instruments for psychiatric disorders in persons with ID. The CAPs-IDD consists of two parts: (1) conceptual and measurement model (including descriptive aspects of instruments, eg, respondent requirements and theoretical foundation) and (2) psychometric properties. We will only use the first part as the second part is more comprehensively covered by the COSMIN checklists.

All ratings will be done by two reviewers independently. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in the discussion. Initial inter-rater agreement will be determined using percentage agreement calculated in R.[38]

As to publication bias, we assume that evaluation results not in favour of the respective instruments are likely to be under-reported. This may be partly due to evaluations being frequently done and published by the developers of the respective instrument. We will address this by including grey literature and by discussing this aspect in the interpretation of our results.

Strategy for data synthesis

A narrative synthesis will be conducted. Assessment instruments will be presented in a table along with descriptive aspects according to CAPs-IDD, and their measurement properties and quality ratings according to the COSMIN checklists. Quantitative data pooling will probably not be possible. This is due to an expected limited number of

studies evaluating the same property (eg, internal consistency) for an instrument and an expected heterogeneity in the population studied (eg, severity of ID, persons with DS vs persons with ID of other aetiology). However, if applicable, we will calculate pooled estimates and 95% CIs using R.[38]

Analysis of subgroups

We define persons with DS/trisomy 21 as a special subgroup, as they are more often affected by Alzheimer's dementia, with a suspected earlier onset.[16] We will group instruments according to their intended use, and studies according to their participants in four clusters: (1) persons with ID, including persons with DS; (2) only persons with DS; (3) only persons with ID, not including DS; and (4) aetiology of ID not specified. For the fourth cluster, we will contact study authors to determine aetiology of ID in the respective sample or for the respective instrument. We will then allocate each study or instrument to the first three clusters according to the information provided by the authors. If no information is provided, the respective study or instrument remains in cluster 4.

Confidence in cumulative evidence

The modified GRADE approach as suggested by the COSMIN guidelines[32] will be applied to grade the quality of the evidence.

Data management

We will use ZOTERO for saving records and managing and storing literature, including managing duplicates. For extracting data and recording decisions on quality ratings, we will use standardised and piloted Excel spreadsheets.

Patient and public involvement

This research was done without patient involvement due to limited resources.

DISCUSSION

This review will summarise measurement properties of available informant-based assessment instruments for persons with ID and give an overview of the quality of each instrument and the quality of available evaluation studies. For each instrument, we will depict which psychometric properties are evaluated to what extent and which properties need further evaluation in future research. This will be the first systematic review of dementia assessment instruments for persons with ID using PRISMA and COSMIN guidelines, as well as applying the ID-specific criteria of the CAPs-IDD.

Our work will highlight gaps in research on these instruments, thus setting the ground for more effective research in the future. The results of this review will inform researchers and clinicians of the quality of available instruments to assess dementia in persons with ID and guide them in choosing an adequate instrument. This will hopefully contribute to an improvement of dementia assessment in persons with ID and a better, earlier and more adequate provision of healthcare services, as demanded by the UN-CRPD.[24]

ETHICS AND DISSEMINATION

No ethics statement is needed for this study. The results of this systematic review will be submitted for publication to a leading peer-reviewed journal, and presented at international conferences and congresses in the fields of ID, ageing and dementia.

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Contributors ELZ conceived the study, drafted the protocol and is the guarantor of the review. SK, IZ and FF contributed to the study design and drafting of the protocol. ELZ, SK and KW designed and tested the search strategy. FF and IZ tested quality rating tools and software options. All authors read and approved the final protocol.

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Competing interests None declared.

Patient consent for publication Not required.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement Data sharing is not applicable as no datasets are generated and/or analysed for this study. No data were generated or analysed for this study.

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