

Frailty and Intellectual and Developmental Disabilities: a Scoping Review



Katherine McKenzie, MSc¹, Lynn Martin, PhD², H el ene Ouellette-Kuntz, PhD¹

¹Department of Public Health Sciences, Queen's University, Kingston, ON; ²Department of Health Sciences, Lakehead University, Thunder Bay, ON, Canada

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ABSTRACT

Background

Individuals with intellectual and developmental disabilities (IDD) are both living longer than in previous generations and experiencing premature aging. Improved understanding of frailty in this aging population may inform community supports and avoid negative outcomes.

Methods

The objective of this study was to review the literature on frailty and IDD and determine areas for future research and application. The methodological framework for a scoping review as developed by H. Arksey and L. O'Malley was applied to identify and select original studies published since 2000.

Results

Seventeen studies were identified; these were based on the work of researchers from four research programs. The studies utilized six measures of frailty, including two frailty indices, the VFQ-ID(-R), the frailty phenotype, and the frailty marker. Frailty was equally studied as an outcome and as predictor for other outcomes (e.g., mobility, falls, care intensity, institutionalization, and survival).

Conclusions

There is evidence of a growing interest in the measurement of frailty in aging adults with IDD. As in the general population, frailty in this group is associated with many negative outcomes. While a few measures have emerged, more work is required to replicate results, validate tools, and test the feasibility of applying frailty measures in practice and to inform policy.

Key words: frailty, frail, intellectual disabilities, developmental disabilities, epidemiology

INTRODUCTION

Population aging has become a topic of increasing interest in the last few decades. In 2000, the World Health Organization report brought attention to a particular subset of the aging population: those with intellectual and developmental disabilities (IDD).⁽¹⁾ It noted that "most adults with intellectual [and developmental] disabilities who live past their third decade are likely to survive into old age ... Numerous adults are surviving into late old age, with some surviving to become centenarians" (p.6). The WHO report also noted that as more people with IDD attain older age, increased needs and further disparities in functional impairment, morbidity, and even mortality can result. The report highlights that special considerations must be given to "the long-term consequences of therapeutic interventions—examples are movement disorders that may result from the prolonged use of neuroleptic medications, and bone demineralization that may occur secondary to the chronic use of certain anticonvulsants." (p.7). Evidently, aging adults with IDD are at risk of additional struggles beyond those normally expected at increased age. In Canada, persons with (IDD) represent between 1–3% of the population.⁽²⁾ In the province of Ontario, it is projected that the number of seniors (65+ years) with IDD will almost double from roughly 6,000 to 10,000 by 2021.⁽³⁾

Persons with IDD are higher users of health care, including aging care. In Ontario, adults (18–64 years) with IDD were almost twice as likely to visit the emergency department and to have been hospitalized in the last year as adults without such disabilities.⁽⁴⁾ They are also twice as likely to use home care, and are admitted to long-term care approximately 25 years earlier.⁽⁵⁾ The World Health Organization report mentioned above urged governments and the academic community to "undertake a course of research that will help further expand knowledge of the ageing process among older adults with intellectual [and developmental] disability[ies]" (p.20).⁽¹⁾

Frailty has become an important construct in relation to aging. In the general population, it has been shown to be a predictor of adverse outcomes, such as institutionalization, falls, and death.^(6–8) Despite efforts, a consensus still has not been

reached on the definition of frailty.⁽⁹⁾ Some components of definitions have high agreement: frailty is a clinical syndrome different from a disability and reflects an increase in vulnerability to stressors;⁽⁹⁾ it is multidimensional, with cognitive, social, psychological and environmental aspects, in addition to the traditional biological and physical factors;^(9,10) and it is correlated with age.⁽¹¹⁾

Many measures of frailty have been developed over the past couple of decades,^(12,13) including the frailty phenotype,⁽¹⁴⁾ the frailty index,⁽⁶⁾ the Study of Osteoporotic Fractures (SOF) index,⁽¹⁵⁾ the Fatigue, Resistance, Ambulation, Illness and Loss of weight (FRAIL) index,⁽¹⁶⁾ the Tilburg Frailty Indicator,⁽¹⁷⁾ PRISMA-7,⁽¹⁸⁾ Groningen Frailty Indicator,⁽¹⁹⁾ and the Edmonton Frailty Scale (EFS),⁽²⁰⁾ among others. The frailty phenotype, developed by Fried et al.,⁽¹⁴⁾ describes frailty as the presence of at least three of five key symptoms (weight loss, weakness, poor endurance and exhaustion, low physical activity, and slowness), and pre-frailty as the presence of two of these features. The frailty index, described by Rockwood et al.,⁽⁶⁾ is a quantitative measure of nonspecific accumulation of deficits, operationalized as the ratio of health deficits present to health deficits measured. The health deficits could be signs, symptoms, diseases, disabilities, or lab measurements.

A systematic review by de Vries *et al.*⁽²¹⁾ described twenty frailty outcome instruments and reported their inclusion of essential frailty factors: nutritional status, physical activity, mobility, strength, energy (physical domain), cognition, mood (psychological domain), and social relations/social support (social domain). While both the frailty index and the frailty phenotype can predict adverse outcomes,^(8,22-25) de Vries and colleagues report that only the frailty index can capture all eight frailty factors. Additionally, the frailty index does not have a predefined list of items, which allows for the inclusion of deficits that are appropriate to a specific population.^(26,27)

It has been recognized that not all measures of health suitable for the general population will produce accurate results among persons with IDD,⁽²⁸⁾ and frailty is likely one that needs reconsideration in this population. In particular, individuals with IDD have pre-existing and life-long conditions which may incorrectly appear as age-related deficits. These conditions may include reduced mobility, seizure disorders, limited daily functioning, or sensory impairments;⁽²⁹⁾ these contribute to multi-morbidity experienced by this population.⁽³⁰⁾ By definition, frailty is the “consequence of age-related decline in many physiological systems”,⁽³¹⁾ and as a result, a measure of frailty must be positively correlated with age to be valid.^(27,32) While pre-existing deficits may contribute to increased vulnerability or increased risk of adverse outcomes, they would not directly contribute to frailty. It is therefore important to re-evaluate both the conceptualization and measurement of frailty to ensure they emphasize change and vulnerability to stressors.⁽³³⁾

After over two decades of researching frailty in the general population, discussions regarding using frailty measures

to improve health policy and clinical practice have begun to emerge.⁽³⁴⁻³⁸⁾ This is a goal shared by the IDD community,^(39,40) however, a better understanding of the current measures used to assess and monitor frailty among community-dwelling adults with IDD is needed. Early identification of frailty in this vulnerable subset of the aging population has the potential to provide an opportunity to put in place needed community supports, which may help to reduce the number of inappropriate admissions to long-term care or institutionalization (re-institutionalization for some).

METHODS

The aim of this scoping review was to understand frailty and its measurement in IDD, and to determine the next steps of research and implementation. Arksey and O'Malley's⁽⁴¹⁾ methodological framework for developing a scoping review was applied. This framework has five stages: (1) identifying the research question; (2) identifying relevant studies; (3) study selection; (4) charting the data; (5) collating, summarizing and reporting the results.

To identify relevant studies, the following databases were searched for peer-reviewed articles: Medline, PsycINFO, EMBASE, CINAHL, Cochrane Library, EBM Reviews, and Joanna Briggs Institute.

The focus was on articles published between 2000 and December 31st, 2015. This period corresponds to the landmark publication of the WHO review of aging and IDD.⁽¹⁾ Terms related to IDD and frailty were used in all searches, though these sometimes differed slightly between databases. Appendix 1 presents the search strategy for EMBASE, as an example.

Targeted searches of the grey literature (i.e., unpublished material) were also conducted. Here, websites of groups/agencies concerned with aging/older adults with intellectual and developmental disabilities were reviewed (e.g., Ontario Partnership on Aging and Developmental Disabilities) and key authors were contacted.

To be eligible for review, articles were required to meet the following criteria: be published in English or French, present or discuss a measure of frailty, include adults 50 years of age or older, and present original research. Review articles were excluded.

Studies were first screened, using titles and abstracts, to select the ones that met the eligibility criteria. If full-text articles were available, these were retrieved. All three authors reviewed studies using the inclusion and exclusion criteria. Agreement of at least two members determined whether an article was included.

The full-text articles were read by all three authors. A tracking and review form was used by one of the authors (KM) to collate reviews and identify factors related to frailty, descriptors, themes, and methods used. All three authors discussed and agreed upon common themes and limitations across studies. The information extracted from studies in-

cluded population characteristics (i.e., study setting, age, sex, level of intellectual disability) frailty measures, purpose of the frailty measurement, key findings, and the authors' suggested next steps.

RESULTS

Study Selection

Figure 1 shows the number of studies screened, assessed for eligibility and included in the review. Excluded studies were ones that did not focus on persons with IDD (e.g., mental illness, mild cognitive impairment) ($n = 195$) or on frailty (e.g., specific conditions that may be related to frailty, such as limitations in activities of daily living, oral health issues, medications, infections, sarcopenia) ($n = 47$) per se.

Seventeen ($n = 17$) publications of original research published between 2000 and December 31st, 2015 were retrieved that measured frailty in aging adults with IDD (Table 1). These 17 studies present the work of four research groups from the Netherlands ($n = 11$),^(39,42-51) Austria ($n = 2$),^(28,33) Canada ($n = 3$),^(5,40,52) and Taiwan ($n = 1$).⁽⁵³⁾ This includes grey literature (e.g., theses or dissertations),^(42,47,52) although some has since been published in peer-reviewed journals.^(54,55)

Study Setting

The 11 publications from The Netherlands were from the Healthy Ageing with Intellectual Disability (HA-ID) study;

these analyzed data from a cohort of 1,050 individuals from care providers in The Netherlands providing specialized support (e.g., ambulatory support, day care, residential settings).^(39,42-51) The investigators from Austria, as part of the POMONA II project “Health Indicators for People with Intellectual Disabilities”, recruited volunteers from eight health areas across the country.^(28,33) One Canadian study accessed administratively held health data of individuals with and without IDD in Ontario;⁽⁵⁾ the other two Canadian studies accessed data of individuals with IDD who were accessing provincially funded home care in Ontario.^(28,33) The study from Taiwan recruited individuals from the voluntary registry, “the Republic of China Foundation for Persons with Down Syndrome”.⁽⁵³⁾

Age

The HA-ID study included individuals aged 50 years and older.^(49,56) Most of the studies using data from the HA-ID cohort reported a mean age of approximately 62 years (SD = 8 years).^(39,42,43,46,47,51) In the articles from the POMONA II project, 190 adults over the age of 18 years were included (mean = 41 years; range = 18–76 years).^(28,33)

The Canadian studies used large, population-level cohorts of adults aged 18–99 years.^(5,40,52) In the largest cohort ($n = 51,138$), approximately half of individuals were between the age of 18 and 44 years.⁽⁵⁾ In a cohort of 7,863 individuals assessed for home care, the median age was 57 years old,⁽⁴⁰⁾ while in an analysis of a subset of this cohort ($n = 3,034$), the mean age was 54 ± 17 years.⁽⁵²⁾

The study using data from the cross-sectional survey “Healthy Aging Initiatives for Persons with an Intellectual Disability in Taiwan: A Social Ecological Approach (II) People with Down syndrome” analyzed data from 216 individuals over 15 years of age.⁽⁵³⁾ Only 10% were over the age of 30 years.

Sex

All studies included both sexes in analyses. Approximately 50–52% of the HA-ID cohort,⁽⁵⁶⁾ and the various cohort subsets,^(39,42-51) were men. Similarly, 52% of the sample of individuals in the Austrian studies were men.^(28,33) The studies from Canada represented males and females less equally, with men representing between 48% and 57% of the cohorts.^(5,40,52) Approximately 62% of the sample in the studies by Lin et al.^(53,57) in Taiwan was male.

Level of IDD

Studies included wide ranges of levels of IDD from borderline and mild cognitive impairment to severe and profound impairment. In the HA-ID studies, approximately a quarter of the cohort had borderline or mild IDD, while a quarter had severe or profound IDD.^(39,42-51) In Brehmer

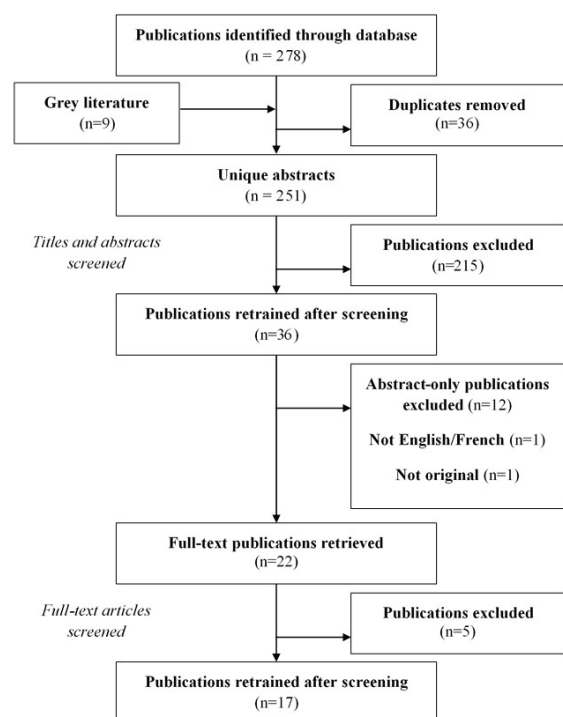


FIGURE 1. Flowchart of search results

TABLE 1.
Studies included in scoping review

Reference	Author (Year)	Study	Country	Study Population	Sample Size	Age (yrs)	Frailty Definition	Role of Frailty
(46)	Schoufour et al. (2015)	HA-ID	NL	Receiving care and support services	982	50+	HA-ID frailty index	Predictor
(48)	Schoufour et al. (2015)	HA-ID	NL	Receiving care and support services	632	50+	HA-ID frailty index	Predictor
(47)	Schoufour et al. (2015)	HA-ID	NL	Receiving care and support services	757	50+	HA-ID frailty index	Predictor
(42)	Schoufour, Echteld & Evenhuis (2015)	HA-ID	NL	Receiving care and support services	838	50+	HA-ID frailty index; HA-ID frailty phenotype	Predictor
(45)	Schoufour et al. (2014)	HA-ID	NL	Receiving care and support services	1050	50+	HA-ID frailty index	Predictor
(43)	Schoufour et al. (2014)	HA-ID	NL	Receiving care and support services	1047	50+	HA-ID frailty index	Outcome
(44)	Schoufour et al. (2014)	HA-ID	NL	Receiving care and support services	676	50+	HA-ID frailty index	Predictor
(39)	Schoufour et al. (2013)	HA-ID	NL	Receiving care and support services	1050	50+	HA-ID frailty index	Outcome
(51)	Zaal et al. (2013)	HA-ID	NL	Receiving care and support services	600	50+	HA-ID frailty index	Outcome
(50)	Bastiaanse et al. (2012)	HA-ID	NL	Receiving care and support services	728	50+	HA-ID frailty phenotype	Outcome
(49)	Evenhuis et al. (2012)	HA-ID	NL	Receiving care and support services	1050	50+	HA-ID frailty phenotype	Outcome
(28)	Brehmer-Rinderer et al. (2013)	POMONA II	AT	All (institution, group, with family, alone)	147	20-72	VFQ-ID-R	Outcome
(33)	Brehmer & Weber (2010)	POMONA II	AT	All (institution, group, with family, alone)	190	18-76	VFQ-ID	Outcome
(53)	Lin et al. (2015)	Healthy Aging Initiatives for Persons with an Intellectual Disability in Taiwan	TW	From voluntary registry	216	15+	Not specified	Outcome
(52)	McKenzie, Ouellette-Kuntz, & Martin (2015)	H-CARDD	CA	Assessed for home care	3034	18-99	42-item frailty index	Predictor
(40)	McKenzie, Ouellette-Kuntz, & Martin (2015)	H-CARDD	CA	Assessed for home care	7863	18-99	42-item frailty index	Outcome
(5)	Ouellette-Kuntz, Martin, & McKenzie (2015)	H-CARDD	CA	Population-level	> 3.3 million	18-99	Frailty Marker	Outcome

NL = Netherlands; AT=Austria; TW=Taiwan; CA=Canada; VFQ-ID(R) = Vienna Frailty Questionnaire for persons with Intellectual Disability (Revised); H-CARDD= Health-Care Access Research and Developmental Disabilities.

and Weber's Austrian study,⁽³³⁾ 78% of individuals could speak for themselves and had low support needs, while the remaining 22% were non-verbal and had high support needs. In the Canadian studies of home care recipients, approximately half of the cohort had moderate, severe or very severe cognitive impairment.^(40,52) The sample from Taiwan included approximately 59% of individuals with severe or profound IDD.^(53,57)

Each research program utilized a different method of determining level of IDD. The HA-ID study used scores from psychologists or test assistants, who ascertained levels based on available IQ tests, Vineland scores, and social emotional development.⁽³⁹⁾ The Canadian studies of home care users used the Cognitive Performance Scale as a proxy for level of IDD^(40,52) or provided no information on the level.⁽⁵⁾ Lin *et al.*⁽⁵³⁾ did not describe how level of IDD was established.

Frailty Measures

Four distinct measures of frailty were identified. Half of the studies ($n = 9$) utilized the frailty index developed by the HA-ID study investigators,^(39,42-48,51) based on the accumulation of deficits approach. Two studies used the accumulation of deficits approach to develop a frailty index based on data from the Resident Assessment Instrument- Home Care (RAI-HC).^(40,52) Three studies applied the frailty phenotype method.^(42,50,58) Two studies used the Vienna Frailty Questionnaire for Persons with Intellectual Disabilities (VFQ-ID),⁽³³⁾ and its revision (VFQ-ID-R),⁽²⁸⁾ which uses a modified accumulation of deficits approach. One study did not specify how frailty had been measured.⁽⁵³⁾ Lastly, one study measured frailty using a marker derived from the John Hopkins University Adjusted Clinical Group (ACG) System,⁽⁵⁾ which classifies a person as frail if he/she has at least one of 81 diagnostic codes.⁽⁵⁹⁾

Most studies categorized individuals based on their frailty score. The VFQ-ID-R used three levels: frail, pre-frail, and non-frail. Similarly, the frailty phenotype measure also required categorization (frail, pre-frail, and robust/non-frail). The Johns Hopkins frailty marker gives a dichotomous outcome (yes/no).⁽⁵⁾

The frailty index, however, was used differently across studies. In the development of the HA-ID frailty index,⁽³⁹⁾ and in subsequent analyses,^(44,45,47,48,51) it was reported as a continuous variable. Other analyses categorized the frailty index values, including as a dichotomous variable,⁽⁴³⁾ with three levels,⁽⁴²⁾ with four levels,⁽⁵¹⁾ and with five levels.⁽⁴⁶⁾ The McKenzie *et al.* study⁽⁴⁰⁾ used categorization to characterize individuals as frail, pre-frail and non-frail.

Key Findings

Frailty as an Outcome

Several studies viewed frailty as an outcome of interest,^(5,28,33,39,40,43,49-51,53) and reported associations with age, sex, level of intellectual disability, living situation, and other factors.

Age was consistently reported to be associated with frailty, regardless of the method of measuring and categorizing frailty.^(5,28,39,40,43,49) Most studies reported that sex was not associated with frailty,^(28,33,39,43,49,53) although two studies reported that women had greater odds of frailty, independent of age and other characteristics.^(5,40)

Most studies evaluated the effect of IDD on frailty. Brehmer and Weber⁽³³⁾ found that those with mild or moderate IDD were more frail, while studies published from the HA-ID cohort consistently reported that adults with severe/profound IDD were most likely to be frail,^(39,43,49) regardless of the measure. Comparing a population of adults with IDD to adults without IDD, the Ouellette-Kuntz *et al.* study⁽⁵⁾ noted that those with IDD were approximately three times more likely to be frail. Without controlling for covariates, the study described a protective effect of cognitive impairment against frailty; however, this was reversed after controlling for age, gender, caregiver status, living situation, and other individual characteristics.⁽⁴⁰⁾

There was no agreement with regard to the association between living situation and frailty. In the Austrian study, there was an insignificant increase in risk of frailty in individuals living in institutionalized settings, compared to living alone or with family, while living in an institution or a community-based group home was not significantly associated with increased frailty in the Dutch study.⁽⁴⁹⁾ The Canadian study found that compared to living alone, living with family (other than spouse and/or children) or living in a group home, was associated with reduced frailty.⁽⁴⁰⁾

Only two studies compared frailty levels in urban versus rural groups, neither of which were significant.⁽³³⁾ Specific diagnoses were also studied in relation to frailty—the presence of dementia,⁽⁴⁹⁾ Down syndrome,^(39,43,49) mental illness or addiction,⁽⁵⁾ and sarcopenia⁽⁵⁰⁾—were positively associated with frailty. Frailty was also associated with relevant or potentially relevant prescription errors.⁽⁵¹⁾

Frailty as a Predictor

Eight studies used baseline measures of frailty to predict future age-related outcomes, including (instrumental) activities of daily living,^(45,53) mobility,^(42,45) co-morbidity,⁽⁴⁸⁾ falls,⁽⁴⁸⁾ fractures,⁽⁴⁸⁾ medication use,⁽⁴⁸⁾ biochemical markers,⁽⁴⁷⁾ care intensity,⁽⁴⁴⁾ hospitalizations,⁽⁴⁸⁾ admission to long-term care,⁽⁵²⁾ and survival.^(42,46,52)

Frailty significantly reduced survival in the community in one four-year follow-up study,⁽⁵²⁾ and in another study with a three-year follow-up, over 60% of the frailest individuals died.⁽⁴⁶⁾ Frailty was significantly correlated with a decrease in functioning at follow-up, independent of baseline functioning and individual characteristics;⁽⁴⁵⁾ and increases in future medication use.⁽⁴⁸⁾ Frailty was more closely associated with survival and future functional status than age.^(45,46) High levels of frailty were significantly associated with increased care intensity, including institutionalization.^(44,52) Frailty was also correlated to some biochemical markers, which

indicated inflammation, anemia, metabolic issues, and poor renal functioning.⁽⁴⁷⁾

Fraility was not predictive of all outcomes examined. After adjusting for sex, age, level of IDD, presence of Down syndrome, and history of outcomes, frailty was not significantly associated with hospitalization, fractures, or falls.⁽³⁸⁾

Next Steps

This review also aimed to understand the status of the use of frailty measures in the field of IDD. Currently, the identified measures are used in the context of research, often in stages of development.^(28,29,33) As such, the next step is to consider how frailty measures could be applied in practice and policy. This requires a discussion of the limitations in existing studies, and the best strategy for implementing frailty measures in policy and practice. Many of the studies identified in this review suggested future steps.

Study Limitations

The current body of research on frailty in the field of IDD is limited. A few are prospective cohort studies that face the issue of drop-out and loss-to-follow-up. These longitudinal studies do not repeatedly collect frailty measurements,^(44,45,46,52) and so the extent to which frailty status changes over time is unknown. Brehmer and Weber⁽³³⁾ noted the importance of and need for longitudinal research to understand how frailty develops over time.

Samples with overrepresentations of individuals living in care settings or receiving care, rather than in the community, could bias estimates of frailty and increases the likelihood of uncontrolled confounding. Both the HA-ID and the home care studies relied on samples identified through care centres and databases, potentially limiting the generalizability of results to those with higher medical needs. Within the subset of persons with IDD who have higher levels of disability or medical care needs, it may be difficult to obtain some measures needed for the assessment of frailty—for example, walking or answering questions.⁽⁴⁹⁾ Additional work is needed to identify appropriate and reliable methods of measuring frailty among those with higher levels of disability.

Implementation

Some authors view the use of a frailty measure as a method of better describing and identifying individuals with IDD experiencing aging.^(42,48) However, it is acknowledged that validation studies must be completed prior to implementation into practice. While the HA-ID frailty index has good predictive validity (e.g., mortality, higher care intensity),^(44,46) the results have not been repeated with a different population or with a shorter version better suited for clinical situations. If cut-off scores are used, as they likely would be clinically, these scores should be validated as well.^(44,46)

To validate a measure of frailty, however, the purpose of the measure must be known. For example, individuals

receiving care may be assessed for frailty to either prevent future decline or to address current issues causing frailty.⁽⁴²⁾ Some have proposed that the most independent, and least-frail, may benefit the most from interventions.⁽⁴³⁾ Frailty, they proposed, should be stopped in the relatively fit subgroup of the aging population, to alleviate the burden of early frailty.^(43,44,53) Doing so would promote healthy aging and functioning, and may provide the most observable benefit.⁽⁴⁵⁾ This hypothesis is based on the theory that frailty may be reversible only in its early stages.^(9,60)

No studies in this review identified a known intervention for the population of adults with IDD, which would be necessary to justify a screening program.⁽⁶¹⁾

Others have suggested frailty could be used to evaluate interventions, or monitor its progression.^(46,48)

DISCUSSION

Advocates for the improvement of the health status of older adults with IDD have repeatedly acknowledged the uniqueness of the aging process in this population,⁽⁶²⁻⁶⁶⁾ and expressed the need for a better understanding of aging with IDD. Traditionally, shorter life expectancies,⁽⁶⁷⁻⁶⁹⁾ early diagnosis of dementia,^(57,69,70) and a higher prevalence of various co-morbidities⁽⁷¹⁻⁷⁴⁾ have been cited as evidence of premature aging. A report prepared in 2001 by the collaboration of the Aging Special Interest Research Group of the International Association for the Scientific Study of Intellectual Disabilities (IASSID) and the World Health Organization listed the development and evaluation of interdisciplinary interventions for complicated conditions as a high research priority.⁽⁷⁵⁾ Over a decade later, the development of frailty measures for this population has occurred; though the literature review spanned the last 15 years, all articles retrieved were published in the last 5 years.

The emerging research focused on IDD and frailty highlights how health indicators focused on physical conditions do not appropriately reflect the age-related risk of vulnerability in this population.^(63,70) Individuals with IDD often have lifelong conditions and it is the deterioration, not the presence, of these conditions that indicate worsening well-being.^(49,56)

In the literature on the general aging population, upwards of 20 unique measures of frailty exist;^(13,21) however, variations of two instruments (i.e., the frailty phenotype and the frailty index) are most frequently found in the literature. Given the desire for a validated and multi-item measure applicable to adults with IDD, both the frailty phenotype of Fried et al.⁽¹⁴⁾ and the “accumulation of deficit” frailty indices of Rockwood et al.⁽⁶⁾ have been studied in this population. The HA-ID study used both approaches, and reported a preference for the frailty index. The frailty index has the flexibility to include items of all domains of health.

In adults with IDD, frailty has been studied in relation to age-related outcomes, including falls, fractures, health care utilization, functional status, and mortality. It is important to note that these outcomes have largely not been repeated across

studies, and are the results of only three research studies. The dearth of research in this area is a limitation of this review. In addition, we also note that the authors (KM, LM, HOK) wrote three of the seventeen articles.

While their samples had broad ranges of age and level of IDD, consideration should be given to other factors that could limit generalizability, such as the type of care individuals are receiving (e.g., home care, group home, institutions) and jurisdictional policies towards aging care. In addition, this scoping review identified discrepancies between the risk of frailty based on sex, level of IDD, and living situation; however, many analyses did not adjust for relevant factors, including age. The threats to both internal and external validity present in the identified studies suggest that further research should be conducted. The current studies must be repeated in other populations to assess the replicability of results. This could include in other jurisdictions, care settings, and age groups. Future studies of frailty in this population should also seek to further determine relevant determinants of frailty, using methodology to adjust for covariates.

Many authors proposed validation studies as appropriate future steps; however, consideration should be given to the purpose of measuring frailty before further work is conducted. A measure to inform policy decisions may be vastly different than a measure applied in clinical practice. Within the clinical scope, a frailty screening tool will demand different characteristics than will a diagnostic tool. This includes appropriate levels of sensitivity and specificity, and predictions of outcomes of interest.

If there is a desire to use a frailty measure beyond describing a population's characteristics, consideration of the feasibility of preventing, modifying, or treating frailty, as well as the potential harms of a frailty designation, is warranted. Before a measure of frailty is implemented into clinical practice, research should determine the degree to which frailty indicators add to individual-level prediction of the onset of disability, or other meaningful outcomes, beyond readily available indicators, including age, sex, and co-morbidities.⁽³⁶⁾ In the general population, reviews^(76,77) and ongoing randomized control trials^(78,79) have been published that explore possible methods to reduce frailty (e.g., rehabilitation, exercise). Interventions to address frailty in adults with IDD have not been widely explored, although the need to focus on improving quality of life of older adults with IDD has been previously raised.^(75,80) The potential for unintended consequences of labelling someone as frail should also be considered.

CONCLUSION

The past five years have indicated a new interest in the measurement of frailty in aging adults with IDD. Similar to the general population, frailty in the population of adults with IDD is significantly associated with many age-related outcomes. While a few measures have emerged, more work is required to replicate results, validate tools, and test the feasibility of applying frailty measures in settings beyond research studies.

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CONFLICT OF INTEREST DISCLOSURES

The authors declare that no conflicts of interest exist.

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Correspondence to: H el ene Ouellette-Kuntz, PhD, Department of Public Health Sciences, Queen's University & Ongwanada, 191 Portsmouth Avenue, Kingston, ON K7M 8A6, Canada
E-mail: helene.kuntz@queensu.ca

APPENDICES

APPENDIX 1. Search strategy example.

<i>Eligibility Criteria</i>	<i>Search Term</i>
Intellectual and developmental disabilities	Intellectual impairment
	Mental deficiency
	Multiple malformation syndrome
	Metabolic encephalopathy
	Congenital hypothyroidism
	Mental patient
	Autism
	Developmental disorder
	Learning disability
Frailty	Frail