Factors associated with the progression of deficit accumulation frailty among adults with an intellectual disability: a systematic review revealing research gaps

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Abstract

Background: People with an intellectual disability are more likely to experience frailty earlier in life and with greater severity compared to the general population. There is growing consideration of determinants of frailty and identifying factors which may influence the change in frailty status over time. The objective of this review was to investigate factors associated with the progression of frailty over time among adults with an intellectual disability.

Methods: A systematic review of literature was conducted using PRISMA guidelines to identify studies reporting factors associated with transitions and trajectories in deficit accumulation frailty among adults with an intellectual disability. The following eligibility criteria was used: defined frailty as deficit accumulation; longitudinal design; reported at least one individual characteristic associated with change in frailty status; sample was people with an intellectual disability aged ≥18 years; English language. No limitation on publication date was applied. Studies which did not measure frailty according to deficit accumulation, did not report the frailty measure used, or had a cross-sectional design were excluded. Selected studies were assessed for quality using the Critical Appraisal Skills Programme (CASP) framework.

Results: In total, two studies qualified for inclusion in this review. Findings revealed that frailty defined as deficit accumulation is a dynamic process and improvements are possible. Changes in the
direction of frailty states over time among adults with an intellectual disability may be influenced by several factors including baseline frailty status, age, the presence of Down syndrome, functional ability, cognitive ability, living in a group home, and the use of nursing services and therapies.

Conclusions: There is a relative paucity of research on frailty among adults with an intellectual disability and the evidence base must be grown. Exploration of the social domain of frailty in this group should be a priority of future research.

PROSPERO registration: 179803 (05/07/2020)

Keywords
intellectual disability, ID, frailty, deficit accumulation, aging
Introduction
As in the general population, people with an intellectual disability (ID) are now living longer because of medical and social progress (McCarron et al., 2011; WHO, 2000). However, older adults with an ID are at an increased risk of early mortality (Lifshitz & Merrick, 2004; McCarron et al., 2015; Stax et al., 2010). They are also more likely to become frail earlier in life in comparison to those in the general population (O’Connell et al., 2020; Ouellette-Kuntz et al., 2017; Ouellette-Kuntz et al., 2015; Schoufour et al., 2013). Current evidence suggests that frailty among older adults with an ID is associated with several adverse outcomes including increased functional dependency (Schoufour et al., 2014a), institutionalization (McKenzie et al., 2016a; Schoufour et al., 2014b) and premature death (McKenzie et al., 2015a; Schoufour et al., 2015).

In recent decades, frailty has been receiving growing attention in the medical literature in response to the unexplained vulnerable state of older adults commonly observed by health care providers (Walston et al., 2018). It provides a means of measuring the combined effects of multiple changes and risks as a person ages (Clegg et al., 2013; Fried et al., 2001; Rockwood et al., 2005). There is however a lack of consensus on the specific nature of frailty and no ‘gold standard’ assessment for frailty exists. Subsequently, a plethora of different frailty measurement tools have been developed for clinical and research use (Roppolo et al., 2015). Generally, these are based on the frailty phenotype (Fried et al., 2001) or the deficit accumulation model (Mitnitski et al., 2001).

The phenotype model describes frailty as a biologic syndrome characterised by the absence or presence of specific signs or symptoms (Fried et al., 2001). This measurement of frailty is considered a clinical-friendly dichotomous variable useful to informing decisions on the need for adapted care or interventions (Cesari et al., 2014). However, this approach does not consider underlying health conditions, as may be present among people with an ID, possibly leading to flawed results in this population (Brehmer-Rinderer et al., 2013). Conversely, the deficit accumulation model, which is operationalised using a Frailty Index (FI), stresses the dynamic biopsychosocial nature of the frailty concept. In contrast to the frailty phenotype, a FI emphasises the proportion rather the nature of deficits present (Rockwood & Mitnitski, 2007). The underlying idea is that the more ‘deficits’ or problems an individual has or accumulates, the less ability they have to recover from a stressful event such as an illness or accident. Among older adults in general the FI has demonstrated robustness in measuring change over time across multiple domains (Dent et al., 2016; de Vries et al., 2011) and has been validated for use in the ID population (McKenzie et al., 2016b).

Regardless of approach, there is a current focus on recognising and describing the progression of frailty and identifying factors which may predict the direction of change in frailty states over time (O’Caoimh et al., 2018). Research with a FI in the general older adult population has shown that there is an apparent natural rate of transition or deterioration in deficit accumulation across the lifespan (Rockwood et al., 2011) which may be influenced by a wide range of physical, behavioural, psychological, cognitive and social health factors (Andrew & Rockwood, 2007; Chamberlain et al., 2016; Gobbens et al., 2010; Levers et al., 2006; Theou et al., 2015; Woo et al., 2005). However, ageing populations are highly heterogeneous (Mitnitski et al., 2017) and therefore the generalizability of these findings to people with an ID cannot be assumed. People with an ID are more likely to have complex health needs and significant health inequalities in comparison to those in the general population (Lunsky et al., 2013; McCarron et al., 2011; McCarron et al., 2014).

The objective of this review is to identify and critically analyse all available longitudinal research regarding factors associated with the development and progression of deficit accumulation frailty among adults with an ID. The research question underpinning this review is: “among adults with an ID, what individual characteristics are associated with changes over time in frailty status defined as deficit accumulation?”

Methods
A systematic review of literature was conducted using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Dee, 2021; Moher et al., 2009) to identify studies relevant to the research question. The review has been registered on PROSPERO (179803, 05/07/2020).

Search strategy
Searches of literature were performed up until October 2019 using the electronic databases MEDLINE, CINAHL Complete, APA PsycINFO and Embase. Search strings were created from the research question following preliminary trial-and-error searches. These search strings were combined using the Boolean operators ‘AND’ or ‘OR’. Search terms included a combination of keywords and related controlled vocabulary for “intellectual disability” and “frailty” (see extended data for full search strategy (Dee, 2021). A manual search of references from included articles was also conducted.

Eligibility criteria
To be considered eligible for inclusion in this review, studies were required to meet the following criteria: (i) be an original scientific article; (ii) define frailty as deficit accumulation; (iii) longitudinally analyse change in frailty states; (iv) report at least one individual characteristic associated with change in frailty status; (v) include a study sample of people with an ID aged ≥18 years; (vi) be printed in the English language. Studies were excluded if (i) they did not measure frailty based on the accumulation of deficits, (ii) did not report the instrument used to measure frailty, or (iii) had a cross-sectional design. No limitation was placed on date of publication.

Screening
The first and second author (BD and EB) independently followed each stage of the screening process and confirmed with each other after completing each step. A third author was available to settle any disparities. The screening process
included: 1) an initial screening of abstracts and titles of articles; 2) a screening of the full text of each article with the full eligibility criteria.

Quality assessment
Following agreement on eligibility, two authors (BD and EB) independently assessed the methodological quality of each study using the cohort study checklist developed by the Critical Appraisal Skills Programme (CASP, 2021). This tool comprises of 14 questions: 1) did the study address a clearly focused issue?; 2) was the cohort recruited in an acceptable way?; 3) was the exposure accurately measured to minimise bias?; 4) was the outcome accurately measured to minimise bias?; 5) have the authors identified all important confounding factors?; 6) have they taken account of the confounding factors in the design and/or analysis?; 7) was the follow up of subjects complete enough?; 8) was the follow up of subjects long enough?; 9) what are the results of this study?; 10) how precise are the results?; 11) do you believe the results?; 12) can the results be applied to the local population?; 13) do the results of this study fit with other available evidence?; 14) what are the implications of this study for practice? Conflicts between the two reviewers were resolved using discussion and consensus.

Data extraction and analysis
A standardised extraction table was used to extract data items from studies that were eligible for inclusion and of sufficient methodological quality. Data was extracted by one reviewer (BD) and cross-checked for accuracy by a second reviewer (EB). Conflicts between the two reviewers would be resolved using discussion and consensus. Data was extracted using the following headings: study type, study location, sample size, sample characteristics, frailty measure, statistical analysis, and findings. Due to the methodological heterogeneity of included studies it was not appropriate to carry out meta-analysis. Findings are presented using narrative synthesis and tabular presentation.

Results
Overall, 5420 articles were retrieved from the literature search. These were uploaded to Covidence systematic review software and all duplicates (n=1600) were removed. In the first step of screening, abstract review, 3763 articles were identified as irrelevant. After screening the full-text of the remaining 57 articles, further exclusions were agreed on the basis of outcomes reported (n=7), no frailty measure specified (n=1), target population (n=37), language (n=4) or reporting of baseline frailty status only (n=6). The PRISMA flow diagram, provided in Figure 1, depicts the flow of information through the different phases of this systematic review. No studies were excluded based on methodological quality following review using the CASP Cohort Study Checklist. Potential limitations of both studies are explained in the Discussion.

Study characteristics
In total, two articles (Martin et al., 2018; Ouellette-Kuntz et al., 2018) qualified for inclusion in this review. These studies provide a longitudinal analysis of administratively held clinical data collected from community dwelling adults with an ID receiving home care in the same geographical region of Ontario, Canada. Martin et al. (2018) explored transitions in frailty status defined as deficit accumulation and the study by Ouellette-Kuntz et al. (2018) investigated factors associated with the rate of deficit accumulation over time. Both studies utilised the same data source, the Resident Assessment Instrument-Home Care (RAI-HC), as described by Hawes et al. (2007). This instrument was developed by interRAI, an international organisation responsible for developing comprehensive clinical assessment systems for persons with disabilities and complex needs. The RAI-HC includes open-text diagnoses for assessors to indicate the presence of any “disease/infection[s] that [a] doctor has indicated is present and affects client’s status, requires treatment, or symptom management … [or a] disease [that] is monitored by a home care professional or is the reason for a hospitalization in last 90 days” (Morris et al., 2009).

Ouellette-Kuntz et al. (2018) had the largest sample of either study (n=5074). They used data collected through repeated follow-up assessments over a period of 3 days to 11.81 years (mean 3.69 years). Significant drop-out rates are reported between these follow-up assessments (n=5074 at T1, n=3693 at T2, n=2813 at T3, n=2183 at T4, n=1734 at T5) due to death or participants stopping to contribute data following their final or discharge assessment. In comparison, Martin et al. (2018) had a smaller sample of 2,893. In total, 21 participants of this study were excluded due to missing data on residential status. Data was collected at baseline and follow-up after 6–12 months (mean 8.3 months). There is a near equal representation of male and female participants in both studies and a mean age of approximately 50 years.

Frailty defined as deficit accumulation is operationalised in both studies using a 42-item FI developed by McKenzie et al. (2015a). This FI was developed with RAI-HC data using standardised procedures (Searle et al., 2008) adapted for use in the ID population (Schoufour et al., 2013). Deficits across multiple domains including physiological (n=29), cognitive (n=4), psychological (n=3), social (n=3), and service use (n=2) are incorporated. Participants in both studies are categorised as non-frail, pre-frail, or frail. These categories represent significantly different risks of experiencing adverse outcomes including institutionalization (McKenzie et al., 2016a) and early mortality (McKenzie et al., 2015b).

Statistical analysis
Martin et al. (2018) report relative risk (RR) of worsening or dying versus staying stable or improving using a modified Poisson regression model with 95% confidence intervals (CIs). Bivariate models were developed which allowed for adjustment for significant confounders (i.e. biological sex, age, Down syndrome diagnosis, moderate or worse cognitive impairment, moderate or worse self-care impairment, residence in a group home, and time in months between baseline and follow-up assessment or death. In contrast, the study by Ouellette-Kuntz et al. (2018) used negative binomial regression models which consider the correlation of repeated assessments. This provided mean incidence rates (IR) (i.e., the mean number of deficits accumulated per year) and incidence rate ratios (IRR) with 95% CIs.
Outcomes
The significance of association between outcome factors and change in frailty state over time was determined by a p-value of ≤ 0.05 (Table 1). To minimize potential bias, both significant and non-significant associations are examined in this review.

Baseline frailty status. Martin et al. (2018) found that baseline frailty status was the largest predictor of worsening frailty or death over a one-year period and remained a significant predictor of this outcome after adjustment for confounding factors. The direction of the association differed by frailty level. Individuals who were pre-frail at baseline had an increased risk (RR 1.24, 95% CI 1.04-1.49, P-value 0.0179), whereas a lower level of risk was observed among those who had a baseline status of frail (RR 0.82, 95% CI 0.69-0.96, P-value 0.0342). Persons who were non-frail at baseline were more likely to remain non-frail over a period of one year (84.26%), while others in this group became pre-frail (7.74%), frail (4.08%), or died (3.92%) over this same period. Pre-frail and frail cohorts were 1.76- and 1.63-times more likely respectively to experience this outcome compared to those who were non-frail at baseline. However, improvement in frailty status was also observed among individuals who were initially pre-frail (40%) and frail (20%).

Age. Age was found to be the strongest predictor of the rate of deficit accumulation from 40 years among non-frail and pre-frail cohorts, and from 70 years among frail individuals (Ouellette-Kuntz et al., 2018). In comparison to non-frail and pre-frail participants aged 18–29 years, frail individuals aged 40–49 years accumulated deficits 1.52 and 1.94 times faster respectively. Rate of deficit accumulation was associated with a 2.14-fold increase in deficit accumulation rate after age 70 when compared to those aged 18- to 29-years. Deficits were observed to accumulate at a rate of 2–3 per year, depending on

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**Figure 1. Flow chart of the literature screening process.**
prior frailty level. A 10-year increase in age was significantly associated with worsening frailty or death within one year (RR 1.30, 95% CI 1.24–1.36, P-value <0.0001) (Martin et al., 2018).

**Biological sex.** Neither study found biological sex on its own to be associated with change in frailty status. The rate of deficit accumulation was comparable for men and women (Ouellette-Kuntz et al., 2018), as was the risk of worsening frailty or death (RR 1.01, 95% CI 0.88–1.16, P-value 0.8457) (Martin et al., 2018).

**Down syndrome.** The presence of Down syndrome was significantly associated with an increased risk of worsening frailty or death (RR 1.48, 95% CI 1.25–1.75, P-value <0.0001) (Martin et al., 2018). Ouellette-Kuntz et al. (2018) showed that rate of deficit accumulation was significantly higher in this cohort with deficits accumulating 1.45–2.09 times faster in comparison to those without this diagnosis.

**Group home living.** Living in a group home was predictive of an increased risk of worsening FI scores or death within 12 months (RR 1.19, 95% CI 1.02–1.40, P-value 0.0358) (Martin et al., 2018). Ouellette-Kuntz et al. (2018) found that non-frail and pre-frail participants living in group homes accumulated deficits 1.2–1.3 times faster than those residing in a non-group home living arrangement. However, this effect was not observed in the frail cohort.

**Cognitive and functional impairment.** Using an adjusted model controlling for confounding factors, Martin et al. (2018) found that functional impairment was associated with an increased risk of worsening or dying (RR 1.24, 95% CI 1.05–1.47, P-value 0.0126). While it was not associated with change in deficit accumulation rate among non-frail and pre-frail cohorts, functional impairment did predict a 1.39-times slower rate of deficit accumulation among frail individuals (Ouellette-Kuntz et al., 2018). Cognitive impairment was not associated with worsening frailty or death (RR 0.96, 95% 0.82–1.13, P-value 0.6026) (Martin et al., 2018). In the study by Ouellette-Kuntz et al. (2018), cognitive impairment was associated with a slower rate of deficit accumulation among non-frail (1.22 times slower) and frail cohorts (1.45 times slower), while no association was identified among pre-frail individuals.

**Use of nursing and therapy services.** The impact of using nursing and therapy services on deficit accumulation was different depending on the individual’s frailty status. Their provision was associated with deficits being accumulated 1.16- and 1.41-times faster, respectively, among non-frail individuals, while 1.61- and 1.59-times slower respectively in the frail cohort. No impact was observed among those identified as pre-frail (Ouellette-Kuntz et al., 2018).

### Discussion

This systematic review has revealed that frailty transitions are more likely to occur among those who were pre-frail or frail at baseline. In the study by Martin et al. (2018), stability in frailty status was the most likely outcome and positive transitions were observed. However, one quarter of study participants who were robust at baseline died within one year. Similar findings have been reported among older adults in the general population (O’Halloran & O’Shea, 2018). Surprisingly, frail individuals had a lower risk of experiencing worsening frailty or death one year later. A potential explanation offered by Martin et al. (2018) is that frailty may be more easily recognizable than pre-frailty, which may have led to the incitement of care actions which prevented or delayed further deterioration in FI scores. If this is the case, it may be a promising indicator of the short-term positive influence of interventions in managing frailty.

### Table 1. Significance of association between factors and change in frailty status.

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Additionally, a 10-year increase in age was associated with an accelerated rate of frailty progression among non-frail and pre-frail individuals aged 40 years and above. While further research is required to explore changes in frailty status over a longer timeframe, these findings suggest the need for regular frailty assessments from at least the age of 40 years among people with an ID, and possibly earlier among certain ‘at risk’ sub-groups, such as people with Down syndrome. This review revealed that having a diagnosis of Down Syndrome was associated with an increased risk of adverse transitions and trajectories in frailty status over time. It should also be noted that other studies report a high prevalence of frailty in this cohort (Evenhuis et al., 2012; Schoufour et al., 2014b; Schoufour et al., 2013). On this basis, the regular and early assessment of frailty should be a priority consideration for individuals with Down syndrome.

Living in a group home was significantly associated with deterioration in frailty levels over time among non-frail and pre-frail individuals. These findings conflict with those by McKenzie et al. (2015b) which show that living in a group home or living with other family members is associated with a reduced likelihood of being frail. However, there is a lack of agreement generally on the association between type of living situation and prevalence risk of frailty among people with an ID. For example, risk appears to be comparable between living with a spouse and/or child(ren) and living alone (McKenzie et al., 2016a). Other studies have reported no significant increase in risk between living in an institutionalised setting versus community-based home (Brehmer & Webber, 2010; Evenhuis et al., 2012). Ouellette-Kuntz et al. (2018) suggest that the high rate of relocation observed among participants in their study may have been a contributing factor. It is interesting to consider the relationship between living situation and frailty progression in the context of ongoing efforts internationally to support people with an ID to transition from institutional to community-based services (Mansell & Beadle-Brown, 2010). Further research is required to explore the relationship between living situation and frailty progression in this group.

The impact of nursing and therapy services on rate of deficit accumulation over time was inconclusive with its provision associated with frailty progression being slower among pre-frail individuals and accelerated among those who were frail. It is not yet known to what extent lifestyle and behaviour factors affect changes in frailty states over time among people with an ID. In the general population, changes in lifestyle and behaviour have the potential to reduce deficit accumulation (Fallah et al., 2011; Hubbard et al., 2009; Woo et al., 2005) due to their role in enhancing a person’s ability to recover from stressor events (Romero-Ortuno & Forsyth, 2018). Interventions such as reducing obesity and sedentary behaviour, promoting physical activity, and smoking cessation have been shown to reduce frailty incidence and progression (Niederstrasser et al., 2019). Targeted interventions for frailty in the ID population and their effect over time should be a priority focus of future research.

The direction of change in frailty status was not found to be influenced by biological sex, with risk of adverse outcomes comparable between men and women. This finding was unexpected given that biological sex is associated with frailty transition patterns in general older adult populations (Kojima et al., 2019). There is however a lack of consensus on the role of biological sex in frailty among people with an ID. While some research has found that women with an ID are more likely to be frail in comparison to men with an ID (McKenzie et al., 2015a; Ouellette-Kuntz et al., 2015), other studies have identified no difference between men and women in this regard (Brehmer & Webber, 2010; Brehmer-Rinderer et al., 2013; Evenhuis et al., 2012; Lin et al., 2015; Schoufour et al., 2013; Schoufour et al., 2014c).

Functional impairment among frail individuals was associated with worsening frailty or death within one year of baseline. This outcome was not predicted by high cognitive impairment in any frailty cohort. Neither functional impairment nor cognitive impairment was associated with an accelerated rate in deficit accumulation. Ouellette-Kuntz et al. (2018) suggest that this apparent protective effect may be reflective of an individual reaching the maximum number of deficits that can be tolerated, in this instance 70% of measured deficits in the FI utilised (McKenzie et al., 2015a). The potential limit to the number of deficits that can be tolerated by an individual may indicate a point of exhaustion in reserve capacity (Rockwood & Mitnitski, 2006).

Findings analysed in this review should be interpreted with caution. Firstly, both studies used samples located in the same geographical location of Canada. Potential cultural differences may have implications for the applicability of findings to people with an ID residing in other regions. Secondly, study samples exclusively included home-care users. Therefore, individuals with an ID not in receipt of home care, possibly due to having more intensive support needs, were not represented. Finally, while the definition constructed for ‘group home’ in these studies is reported as having good face validity following expert panel review (Martin et al., 2018), mis-categorisation of individuals as living in a group home when they did not may have occurred.

Conclusions
This review has shown that frailty is a dynamic process and bi-directional change in frailty status can occur over a relatively short period of time. The utility of the deficit accumulation approach in exploring transitions and trajectories of frailty among older adults with an ID was also demonstrated. The small number of studies which qualified for inclusion in this review reflects the relative paucity of frailty research in the field of ID and highlights the need for the knowledge base to be grown. Furthermore, the influence of baseline frailty status and the use of nursing services and therapies were reported by only one study, while there is a lack of consensus on the impact of cognitive impairment. Further research is required to understand the association between these factors and frailty outcomes among older adults with an ID.

Findings of this review indicate that a wide range of factors appear to influence frailty progression among older adults
with an ID. In addition to increasing age and the presence of Down syndrome, several social-related factors pertaining to functional ability, living situation, and healthcare use were associated with frailty outcomes. There is a social conceptualisation of frailty which recognises that social circumstances may contribute to health outcomes and affect access to appropriate interventions and supports (Andrew et al., 2018). This approach views frailty as an outcome of the relationship between the individual and their environment (Barrett, 2006). Social frailty may provide a novel and additional approach to understanding the accumulative effect of social-related factors on health outcomes among older adults with an ID. Insights obtained may support the development of upstream interventions which address social disparities, and reduce modifiable contributors to frailty. This is another area for further research.

Data availability
Underlying data
All data underlying the results are available as part of the article and no additional source data are required.

Extended data

This project contains the following extended data:
- Supplementary File 2. Appendix 1_Search strategy.docx
- Supplementary File 3. Figure 1. PRISMA flow diagram.docx

Reporting guidelines

Data are available under the terms of the Creative Commons Attribution 4.0 International license (CC-BY 4.0).

References


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