AGING WITH INTELLECTUAL AND DEVELOPMENTAL DISABILITIES: THE EFFECT OF FRAILTY AND HEALTH INSTABILITY IN HOME CARE USERS ON ADMISSION TO LONG-TERM CARE

by

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Abstract

BACKGROUND: An increasingly large proportion of adults with intellectual and developmental disabilities are frail. Measuring frailty is challenging, and generic measures of frailty inaccurately identify frailty in this population. In the general population, frailty reflects an increased risk of outcomes, including admission to long-term care. Institutionalization of adults with intellectual and developmental disabilities may be a reflection of high levels of frailty.

OBJECTIVES: 1) To develop a frailty index for home care users with intellectual and developmental disabilities using Ontario standardized clinical assessment data, 2) to describe the prevalence of frailty among these adults, and 3) to determine the association between frailty and admission to long-term care.

METHODS: A frailty index was developed using published criteria to select items from the Resident Assessment Instrument-Home Care. Assessments of 7,863 individuals with intellectual and developmental disabilities in Ontario (aged 18-99 years) provided the data. A comparison measure of health instability, the Changes in Health, End-stage disease, Signs and Symptoms scale, identified individuals as stable or unstable. Using a subset of this sample (n=3,034) retrospectively followed over four years, survival analyses determined differences in rates of admission to long-term care according to frailty and instability.

RESULTS: The frailty index combined 42 deficits from five domains of health (physiological, psychological, cognitive, social and service use). Almost half of the cohort was pre-frail (21%) or frail (27%). In the 4-year follow-up, 22% of the sample was admitted to long-term care. Adjusting for age, sex, rural status, caregiver status, living situation, and cognition level, at any given time, frail individuals were twice as likely to be admitted to long-term care compared to non-frail individuals.

CONCLUSIONS: The frailty index is developed from items currently incorporated into home care assessments, and can predict institutionalization in adults with intellectual and

developmental disabilities. Further research should evaluate the feasibility of implementing this measure.

Co-Authorship

This thesis describes the work done by Katherine McKenzie under the direction of Dr. Hélène Ouellette-Kuntz and Dr. Lynn Martin. Data linkage for both manuscripts was completed by Ms. Marlo Whitehead, Institute for Clinical Evaluative Sciences Senior Programmer and Biostatistician.

Manuscript 1: Using an accumulation of deficits approach to measure frailty. The idea of developing a measure of frailty for adults with intellectual and developmental disabilities was Dr. Ouellette-Kuntz's and Dr. Lynn Martin's. The writing of the manuscript, statistical analysis, and interpretation of results were completed by Katherine McKenzie, with editorial feedback provided by Dr. Ouellette-Kuntz and Dr. Martin.

Manuscript 2: Prediction of institutionalization. The idea of determining the risk of institutionalization was Dr. Ouellette-Kuntz's and Dr. Lynn Martin's. The writing of the manuscript, statistical analysis, and interpretation of results were completing by Katherine McKenzie, with editorial feedback provided Dr. Ouellette-Kuntz and Dr. Martin.

All other chapters were written by Katherine McKenzie, with feedback and advice provided by Dr. Ouellette-Kuntz and Dr. Martin.

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List of Abbreviations

ADL: Activities of daily living

AIC: Akaike information criterion

ALC: Alternate level of care

AUC: Area under the receiver operating characteristic curve

CAP: Clinical Assessment Protocol

CCAC: Community Care Access Centre

CHESS: Changes in Health, End-stage disease, Signs and Symptoms

CI: Confidence interval

CIF: Cumulative incidence function

CPS: Cognitive Performance Scale

CSHR: Cause-specific hazard ratio

FI: Frailty index

IADL: Instrumental activities of daily living

ICES: Institute for Clinical Evaluative Sciences

IDD: Intellectual and developmental disabilities

KM: Kaplan-Meier

LTC: Long-term care

MOHLTC: Ministry of Health and Long-Term Care

OR: Odds ratio

RAI-HC: Resident Assessment Instrument- Home Care

ROC: Receiver operating characteristic (curve)

SD: Standard deviation

SDHR: Subdistribution hazard ratio

WHO: World Health Organization

Chapter 1

Introduction

1.1 General Overview

Frailty is an appealing method to understand the risk of poor health and mortality in aging adults [1]. Despite the numerous measures of frailty available [2,3], few are appropriate for use in a population of adults with intellectual and developmental disabilities (IDD) [4]. Individuals with IDD face lifelong disabilities that can be mistakenly viewed as a consequence of frailty [4]; rather, changes in health-related factors better indicate risk and vulnerabilities.

While frailty measures exist for the population of persons with IDD [5,6], wide-scale application of the measures is limited by the use of specific questionnaires and medical examinations. Access to routine home care assessments of adults with IDD from a large, Ontario-based cohort provides the opportunity to develop a frailty measure that could, if validated, be implemented as a prognostic tool in service delivery practices [7].

A better understanding of the frequency and consequences of frailty is important for policy and provision of health care services, especially in a population already disadvantaged [4,8]. Individuals with IDD, who comprise almost 1% of Canadians [9,10], face increased risks for adverse health outcomes likely due to genetic predispositions, unfavourable social circumstances, less frequent use of generic health services, and environments that support inactivity and poor lifestyle choices [11]. These risk factors may contribute to the premature, or accelerated, aging often observed, but poorly understood, in adults with IDD [12]. Early frailty might also contribute to early aging and the subsequent use of age-related health care services.

In the general population, frailty is a strong predictor of admission to long-term care (LTC) [13–17]. Postponing, or preventing completely, the transition to a LTC home corresponds with wishes of the general public [18,19] and caregivers of persons with IDD [20]. Given the efforts over the past few decades to deinstitutionalize all persons with IDD, culminating in 2009 with the closure of the last institution in Ontario, it is crucial to evaluate how individuals with IDD are coping and aging in the community. In particular, there is interest from policy makers, families and communities to better understand the risk of re-institutionalization of younger adults into LTC homes intended for seniors [21,22].

1.2 Empirical Objectives

Given the need to better understand frailty and its implications in this population, an IDD-specific measure to identify those at risk of health decline and institutionalization is warranted. The three main objectives of this thesis were:

(1) to create a frailty index (FI) for adults with IDD using Ontario standardized clinical assessment data,

(2) to describe the prevalence of frailty among adults with IDD using home care services in Ontario, and

(3) to calculate the association between both a measure of health instability and a measure of frailty (FI) and admission to LTC care, among adults with IDD receiving home care services in Ontario.

1.3 Thesis Organization

The thesis conforms to the framework provided by the *General Forms of Theses* which is outlined by the School of Graduate Studies at Queen's University [23]. The second chapter of this thesis presents background literature for this thesis. The third chapter is Manuscript 1 which addresses objectives 1 and 2. This manuscript will be submitted to the *BMC Population Health Metrics.* The fourth chapter, Manuscript 2, addresses objective 3. This manuscript will be submitted to the *Journal of Intellectual Disability Research.* Chapter 5 provides an overall discussion of the results and a conclusion.

Additional documentation describing the methods and additional results are found in the Appendices, including the statement of approval from the Queen’s Health Sciences & Affiliated Teaching Hospitals Research Ethics Board. The thesis title has since been modified.

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Chapter 2

Literature Review

2.1 Aging with Intellectual and Developmental Disabilities

The size of the population of adults with intellectual and developmental disabilities (IDD) has been steadily increasing, with administrative prevalence in Canada now reaching almost 1 per 100 individuals [1]. The growing number can largely be attributed to an increasing lifespan. Historically, individuals with IDD have had limited longevity [2,3], likely due to genetic predispositions, unfavourable social circumstances, less frequent use of generic health services, and environments that supported inactivity and poor lifestyle choices [4]. However, steady improvements to the promotion, management and delivery of care to persons with IDD have resulted in life extension [4–6], with more adults with IDD living into their seventies and eighties [7].

As with the general population, the population of adults with IDD is aging, however aging occurs earlier in life and may not be considered healthy aging [6,8–10]. Facing the "double jeopardy" of old age and IDD [11], these adults experience the same conditions as the general aging population, compounded by the additional risks associated with IDD. Adults with IDD face increased risks for conditions including: sensory impairments, obesity, epilepsy [12], fall related injuries [13], oral health conditions, gastrointestinal issues, and musculoskeletal conditions [14]. These issues present themselves far sooner in adults with IDD compared to the general population. Consequently, there is a higher use of health care services and the potential for reduced quality of life [15]. Given the premature aging and specific health care needs of this population, it would be insufficient for both health care providers and researchers to use age as the primary identifier of risk of health decline and future care needs [16,17].

2.2 Aging At Home

In Ontario, individuals with IDD can access services and support from the Ministry of Health and Long-Term Care (MOHLTC) [18]. MOHLTC's Community Care Access Centres (CCACs) determine eligibility and coordinate providers for home care services [19]. A wide range of services are provided [20], including visiting health professionals, who help manage health conditions, either temporarily or on an ongoing basis. Health professionals include nurses, physiotherapists, occupational therapists, speech-language pathologists, social workers and dieticians. Personal support workers can help with personal hygiene and routine activities of daily living (ADLs), such as dressing, eating, and toileting. Homemaking services are available to help individuals receiving personal support to do routine household chores, for example: cleaning, paying bills, preparing meals, doing laundry, and caring for children. Home care also includes community support services, such as day programs, friendly visiting, meal deliveries and transportation services, among many others. To receive home care, an individual must require at least one professional service in the home and be insured under the Ontario Health Insurance Plan (OHIP) [21].

Home care is a quickly expanding segment of Canada's health care system. Romanow's (2002) description of home care as the "next essential service" [22] corresponds with the wishes of older adults to remain at home [23,24], while costing the health care system significantly less money [25]. Studies indicate that home care helps to maintain independence, improve quality of life [26], and lessen health care utilization [27].

Individuals with IDD can receive home care while living in their own home, family home, supported living arrangements (e.g., group home, semi-independent living), or specialized residences for those with additional needs (such as mental health services). Despite the comprehensive supports available, long-term care (LTC) services remain necessary for some individuals whose needs cannot be met in the community.

2.3 Long-Term Care

There are various residential options that provide accommodation and care for adults transitioning from the community to an institution. Terminology varies between provinces and can be conflicting [25]; in Ontario, three options exist [28]. Retirement homes are privately owned rental residences and are meant generally for those who need minimal to moderate support with daily activities. Assisted living housing, or supportive living, is government-funded, and has 24-hr personal care and support. LTC provides the same care as assisted living, in addition to assistance with ADLs, 24-hour nursing care, and specialized care that can accommodate various health and safety needs are available. Individuals must be over the age of 18 years to be accepted into these homes.

Many factors and conditions contribute to institutionalization. A meta-analysis of predictors of nursing home admissions in the U.S. reported a variety of significant factors from a pooled analysis [29]. In the general population, medical conditions such as high blood pressure, cancer, diabetes and strokes were predictive of admissions. In addition, prior hospitalization, recent falls, living alone, low annual income and the unavailability of informal caregivers contributed to the risk of admission to nursing homes [29]. Maxwell *et al*.'s (2013) study of residents of assisted living facilities in Alberta reported significantly increased risk for admission to LTC for older residents, and those with poor social relationships, low involvement in activities, cognitive and/or functional impairment, recent falls, hospital visits and severe bladder incontinence [30].

Health services research frequently concentrates on risks of institutionalization in the general population; however, fewer studies have discussed risk of institutionalization in adults with IDD.

2.4 IDD and Long-Term Care

The de-institutionalization of adults with IDD is an international goal [31]. In 1987, Ontario made the decision to close all of its residential institutions for individuals with IDD, which resulted in the transfer of individuals to the community by the final closure in March of 2009 [32,33].

While Ontario no longer operates institutions for adults with IDD, many view the growing number of adults with complex needs moving to residential care facilities as an act of re-institutionalization [34]. Individuals with IDD, family members, and service providers generally view placement in a LTC facility as one of the least desirable plans for the future [11,35,36]. Despite reluctance from these key stakeholders, adults with IDD continue to be admitted to LTC [37,38]. Even when LTC is intended as a temporary solution in a period of crisis, once placed, it can be difficult for people to move out [37]. In one study, “temporary” residents stayed an average of 1.9 years in LTC [37]. Of the 69 adults (of a total of 253) who were discharged from LTC in a 5-year period, only five were discharged to independent accommodation or back to their families [37]. Persons with IDD living in LTC homes are much younger than other residents [39], have restricted access to developmental services, have difficulty fitting in with other residents, and have limited activity outside of the home [37].

Canadian policies toward deinstitutionalization, specifically for adults with IDD, aim to follow the principles of community inclusion, choice making, and the improvement of quality of life [40]. Canada has significantly invested in strategies to help older Canadians age successfully in the community. Considering that adults with IDD continue to be misplaced in LTC homes [41,42], early identification of increased risk could offer opportunities to prevent or delay admission.

In the Canadian general population, the literature has established frailty as a significant risk factor for admission to LTC, reporting increased risk in frail seniors, regardless of the measure or care setting [30,43–46]. However, the concept is neither consistently defined nor consistently measured. It is even less well studied among individuals with IDD, who, while likely to have many of the same risk factors for admission, may experience frailty differently [47].

2.5 Frailty, Health Instability, & Disability

The growing size of the aging population and the subsequent disproportionately high use of health care lead to the exploration of "geriatric syndromes" to identify conditions associated with increased vulnerability. In a systematic review of the literature, Wang, Shamliyan, Talley, Ramakrishnan, & Kane (2013) identified eight such syndromes: multi-morbidity, cognitive impairment, frailty, disability, sarcopenia, malnutrition, impaired homeostasis, and chronic inflammation. It was reported that frailty had the strongest associations with future institutionalization [48].

Two geriatric syndromes, frailty and health instability, in the context of disability, have the potential to identify individuals at risk of negative health outcomes.

2.5.1 Frailty

Systematic reviews have presented the utility of frailty measures as prognostic tools for adverse outcomes [49,50], however researchers have not agreed on a universally-applicable definition and operationalization [51,52], despite a large consensus meeting [53]. The initial definitions of frailty focused on biological functions. Buchner and Wagner (1992) described frailty as "a state of reduced physiological reserve associated with increased susceptibility of disability" (page 2). This "reserve" refers to redundant mechanisms in physiology that can compensate for normal age- or disease-related changes. While gradual decreases in reserve are expected in the aging process, they are accelerated with frailty [51]. Given its biological basis, some research has explored biological markers, such as interleukin-6 [55] or cholesterol [56], or single-item tests, such as grip strength [57], to indicate frailty. However, single-item markers and tests are not comprehensive measures of frailty [49], and clusters of clinical manifestations of frailty are likely greater risk factors for adverse outcomes [52].

A systematic review of frailty outcome measurements concluded that eight risk factors are most important to the construct of frailty: nutritional status, physical activity, mobility, strength, energy, cognition, mood and social contacts and support [49]. This supports the alternative perspective that is now widely accepted: frailty is a multidimensional state of vulnerability [58], with cognitive, social, psychological, and environmental aspects in addition to the traditional biological ones.

Two multi-dimensional approaches to measure frailty are frequently described in the literature. The model developed for the Cardiovascular Health Study [59] led to the creation of the frailty phenotype approach. Five characteristics of frailty were identified: weight loss, weakness, poor endurance and exhaustion, low physical activity and slowness. The presence of at least three of these criteria identified an individual as frail.

The second method is the accumulation of deficits approach, introduced by Rockwood *et al*. (2006), which provides the methods to develop a frailty index (FI) [44]. This model proposes that the more health deficits an individual has, the frailer he/she will be. The emphasis is on the proportion of deficits present, of the total measured, rather than the contributions of specific deficits. In the accumulation of deficits model, deficits can refer to signs, symptoms, disabilities, diseases or abnormal laboratory measurements. Unlike the frailty phenotype approach, this model considers a wide variety of factors and can be applied to a range of datasets, even those that may not have been designed to measure frailty [60]. De Vries *et al.* (2011) examined 20 frailty instruments and determined that the accumulation of deficits approach is the most inclusive of all the potential factors leading to frailty [49].

Various studies have applied multi-dimensional measures of frailty to predict adverse outcomes in the general population. A prospective cohort study using data from the Longitudinal Aging Study-Amsterdam determined the relationship between frailty and the risk of residential or nursing home admissions in the general, Dutch population [24]**.** At baseline, frailty was operationalized using nine criteria: body weight, peak expiratory flow, cognition, vision and hearing ability, incontinence, sense of mastery, depressive symptoms and physical activity. It was reported that the risk of admission was 1.73 times higher in the frail group compared to the non-frail group (95% CI: 1.19-2.50), controlling for age, sex, income, partner status, care received, number of chronic diseases, and functional limitation.

Researchers have studied the relationship between frailty and death and/or institutionalization, using interRAI instruments in samples of the Canadian general population. Armstrong *et al*. (2010) used survival analyses to predict how well three measures could predict adverse outcomes, defined as institutionalization or death, within a year of assessment in a large sample (n=23,952) of home care clients from eight Ontario Community Care Access Centre (CCACs) [43]. One measure was a FI based on the Resident Assessment Instrument-Home Care (RAI-HC), created with Searle *et al*.'s (2008) criteria, which provides guidelines for creating an index using relevant and available variables [60]. Comparing the frailest with the least frail, hazard ratios were well over 1.0 (HR= 1.93, 95% CI: 1.79-2.08). Being female and being younger were protective against adverse outcomes.

In a study of 1,066 residents of assisted living (AL) and supportive housing in Alberta (aged 65 or older), Hogan *et al*. (2012) reported an adjusted HR of 4.14 (95% C.I.: 1.87-9.14) using a FI to predict LTC placement within one year, using data from the interRAI-AL tool [45]. While individuals using assisted living services likely have higher needs, this relationship might indicate a similar association in an Ontario home care setting.

Using the accumulation of deficits approach to determine the risk of institutionalization, Jones *et al.* (2005) obtained a hazard ratio of 1.20 (95% C.I.: 1.10-1.32) in their analysis of 2,305 community and residential living adults (≥65 years old) in the 1996-1997 wave of the Canadian Study of Healthy Aging (CSHA-2), adjusted for age, sex and education [46].

With a FI of 36 deficits, Drubbel *et al.* (2013) reported an increase in risk of adverse outcomes (including institutionalization and death) of 17% per additional deficit (HR=1.17, 95% CI: 1.13-1.21), adjusted for age, sex, and consultation gap (number of days since last interaction with a general practitioner), in a sample of 1,679 primary care patients in the Netherlands (≥60 years old) [61].

Rockwood, Song, and Mitnitski (2011) used a 42-item FI to determine risk of admission to LTC in community-based individuals aged 15-102 years old [62]. The median time to admission was 61.2 months for those who were least frail, and 34.2 months for those who were most frail (log-rank test, $p<0.001$). In a different cohort of 70-year olds (n=966), Rockwood *et al*. (2006) observed a significant difference in rates of admission to LTC: those in the two frailest quartiles had higher rates compared to those in the lowest quartiles [44].

Data from the Canadian Community Health Survey (CCHS) provided items for the creation of a FI for community-dwelling seniors aged 65 and older [63]. With a sample size of 268,520, the authors reported significant associations between their FI and admission to LTC facilities and mortality.

These studies suggest that frailty measures, and specifically those derived from the accumulation of deficits approach, are suitable measures for predicting institutionalization in a Canadian setting.

2.5.2 Health Instability

Health instability is viewed as an observable outcome of frailty, on the pathway to adverse outcomes [64] (Figure 2.1). Subsequently, health instability can be regarded as a more severe state than frailty. Researchers almost exclusively apply the Changes in Health, End-stage disease, Signs and Symptoms (CHESS) scale to establish the presence of health instability; it is specifically designed to identify decline in functioning [64] and is derived using data from the interRAI suite of instruments in different health care settings. This scale (ranging from 0 to 5) considers factors including vomiting, dehydration, weight loss, shortness of breath, edema, decline in cognition and decline in ADLs.

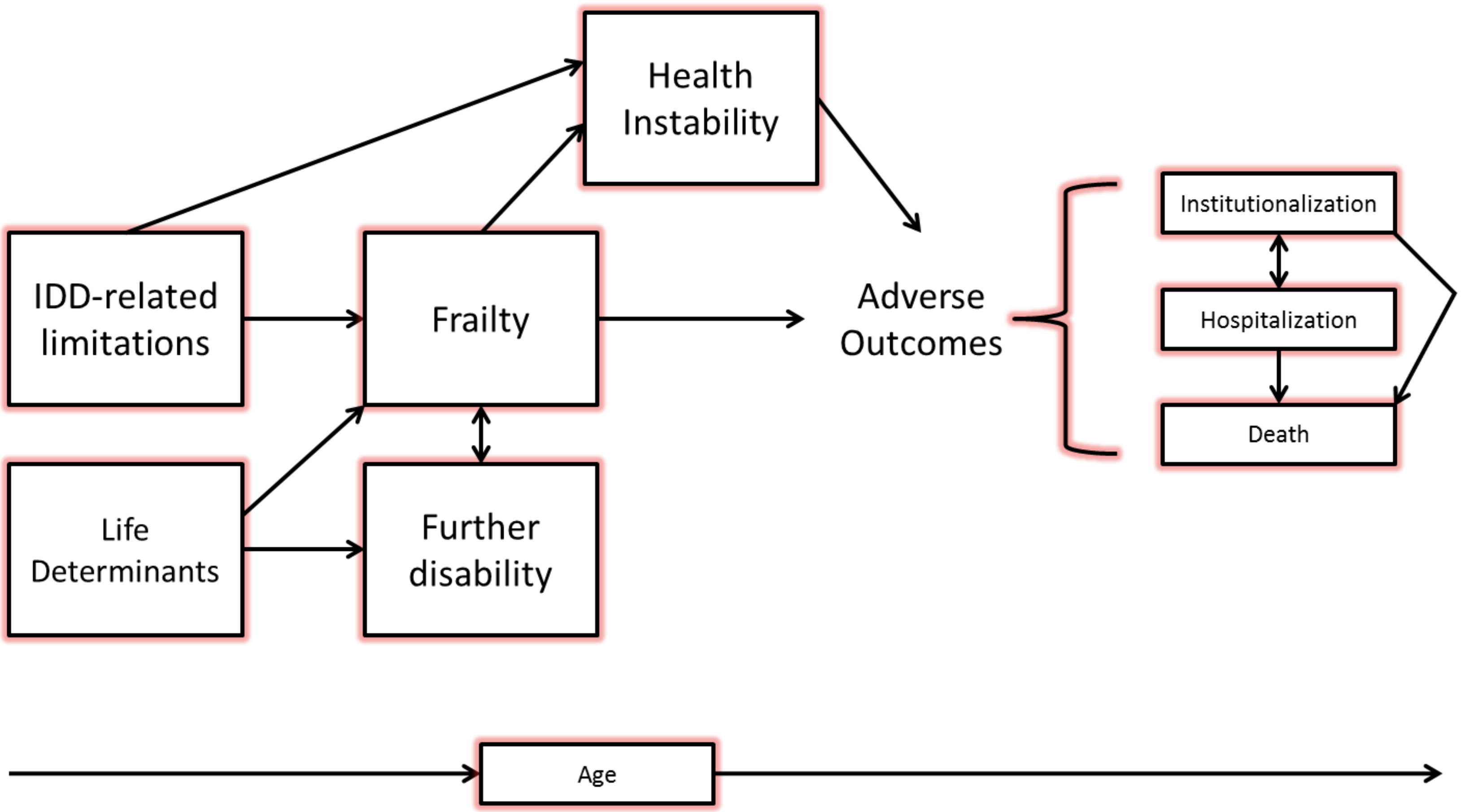
Armstrong *et al*. (2010) and Hogan *et al*. (2012) also applied the CHESS to their studies of home care and assisted living users, respectively. In home care users, health instability was predictive of adverse outcomes (a combination of institutionalization and death) (HR= 2.32, 95% CI: 2.15-2.51) [43]. Comparing the ability of the CHESS and the FI to predict LTC placement, Hogan *et al*. (2012) reported no significant difference in predictive accuracy between the two measures, however the CHESS had a slightly weaker association (HR=1.87, 95% C.I.: 1.39-2.50) [45]. Maxwell *et al*., 2013 reported a hazard ratio of 2.26 (95% CI: 1.49-3.41) to compare the risk of institutionalization between adults with high instability compared to no instability residing in assisted living facilities in Alberta, Canada [30].

2.5.3 Disability

The term disability (physical, cognitive or a combination of both) is sometimes used interchangeably with frailty [65]. Frailty and disability are two common clinical characteristics of aging adults, and although there is some overlap in their definitions, they are distinct constructs [66,67]. The World Health Organization (WHO) broadly defines a disability as "any restriction or lack (resulting from impairment) of ability to perform an activity in the manner or within the range considered normal for a human being" (page 28) [68]. In contrast, frailty is indicative of a state of vulnerability to stressors and a loss of functioning.

Generally, it is accepted that frailty is a precursor to disability [69]. Stressors develop, either progressively (e.g., with disease), or acutely (e.g., with a fracture), and disability ensues, evidenced by mobility loss or impaired (instrumental) activities of daily living (IADL). Frailty is not necessary for disability, and individuals do not cease being frail with the development of disabilities [69].

Figure 2.1. Conceptual framework to describe the theoretical associations between disability, frailty, health instability and adverse outcomes



Individuals with IDD have pre-existing and life-long disabilities, which incorrectly appear as deficits in many frailty measures. This forces the re-evaluation of the early definitions of frailty to emphasize change and vulnerability to new stressors, rather than a recognition of pre-existing limitations [70].

A measure designed specifically for adults with IDD could address this issue. It is accepted in health research that not all instruments that are suitable for the general population will produce accurate results in the population of adults with IDD [71]. For example, many studies have reported that individuals with Down syndrome are at a higher risk for Alzheimer's disease, and have acknowledged the difficulties with the diagnostic process, due to large intra-individual variation and unknown baseline cognitive function [72]. For a study of individuals with IDD, it is important that the chosen frailty measure be considerate of the differentiation between disability and frailty by measuring changes in health and functional status rather than states of health. In addition, the limited utility of self-report measures in the population with IDD eliminates many other identified measures (e.g. the Tilburg Frailty Indicator [73]) [49]. An adequate measure must be accommodating of caregiver respondents and dependent individuals with limited communicative ability.

2.5.3.1 Measures of frailty and health instability in adults with IDD

As of June 2015, only three research groups have published research exploring frailty in aging adults with IDD: the Healthy Ageing with Intellectual Disability (HA-ID) study, the Health Care Access Research and Developmental Disabilities (H-CARDD) team, and the POMONA II study research group.

Evenhuis, Hermans, Hilgenkamp, Bastiaanse, & Echteld, (2012) applied the frailty phenotype approach to individuals from the HA-ID study [74]. Individuals (n=848) had borderline to profound IDD, were 50 years of age or older, were receiving care services in the Netherlands and had at least three criteria (symptoms) assessed. The authors reported that 13% of their cohort was frail (at least three symptoms present), and 60% was pre-frail (1 or 2 symptoms present). In addition, they reported that age was positively associated with frailty: individuals aged 65 years and older were 1.7 times more likely to be frail compared to their younger counterparts. At 11%, the prevalence of frailty among the younger (<65 years) group was similar to the published prevalence estimates in the general Dutch population of adults over the age of 65 years. This supports the hypothesis that individuals with IDD experience frailty earlier. This study had some limitations. Those who were not able to participate in all the assessments (n=202) were more likely to have severe IDD and motor disability. In addition, by using the frailty phenotype approach, the frailty measurement only captured physical symptoms, and subsequently, associations between motor disability and frailty were strong: persons using walking aids or wheelchairs were 9.8 times more likely to be frail than independent walkers. This strong association is likely attributable to longstanding motor disability, rather a decline in health.

In Ontario, home care users (aged 18-99 years) had a high prevalence of frailty (8.9%) compared to a sample of adults without IDD also 18 to 99 years old (3.1%) when measured using Johns Hopkins Frailty Marker [39]. Adults with IDD aged 18-24 had similar frailty proportions compared to adults without IDD at age 60-64 years, corroborating Evenhuis *et al*.'s (2012) findings of early frailty. The Frailty Marker operationalized frailty by counting the number of (unknown) medical diagnoses. This measure may be inappropriate for persons with IDD, given the presence of life-long deficits and the omission of social, psychological, cognitive and environmental conditions.

Recognizing the importance of a frailty measure designed for individuals with IDD, Brehmer & Weber (2010) used the POMONA II study to create a new frailty measure, the Vienna Frailty Questionnaire for persons with Intellectual Disabilities (VFQ-ID) [75], revised a few years later to become the VFQ-ID-R [71]. Their measure focuses on changes in specific aspects of health, rather than the existence of life-long deficits. Combining the accumulation of deficits approach

[70] and the frailty phenotype approach [59], the authors developed a list of 34 symptoms for the questionnaire, which were categorized into four domains: physical, psychological, cognitive and social. To be frail, an individual had to have (1) symptoms present in at least three domains, and (2) a minimum of six symptoms in total. Persons were characterized as "pre-frail" if only one of the two criteria was met. Of a sample of 147 Austrian adults with IDD (aged 20-72 years), they reported that 17.7% of their sample were frail, 17.7% were pre-frail and the remainder of the sample was non-frail [71]. The Vienna Frailty Questionnaire is more multi-dimensional than the HA-ID measure [74], however a small sample size limits the generalizability of findings. The necessity of a questionnaire limits large-scale usage, due to the inclusion of items not routinely collected.

Schoufour *et al.* (2013) applied the accumulation of deficits approach to the HA-ID cohort (n=932) of elderly adults (≥ 50 years) using formal care in the Netherlands, with at least 30 deficits measured [47]. They reported that individuals with IDD had similar rates of frailty at age 50 as did the European general population at age 70. This suggests that individuals with IDD accumulate deficits about 20 years sooner than the general population. This sample had a mean frailty score of 0.27 (SD=0.13) and an upper limit of 0.69 (on a continuum ranging from 0, not frail, to 1, frail).

Using the HA-ID FI, Schoufour, Evenhuis, & Echteld (2014) found significant associations between high levels of frailty and increased care intensity and cost: the odds of a higher level of care increased by almost 10% per increase in 0.1 of the FI (approximately 2 deficits) [76]. Frailty was also associated with higher medication use and several co-morbid conditions (e.g. nervous system, gastro-intestinal and metabolic conditions), but not to falls, fractures, or hospitalizations [77]. Frailty was also significantly associated with limited daily functioning [78].

The accumulation of deficits approach, following predetermined criteria [60], is likely the most appropriate method of observing frailty in aging adults with IDD. The accumulation of deficits approach selects appropriate health-related deficits from available datasets, which offers the opportunity to include items relevant to the health of adults with IDD.

Hirdes, Poss, Mitchell, Korngut, & Heckman (2014) applied the CHESS to a subgroup of home care service users including Ontario residents [79]. While the cohort was not IDD-specific, inclusion into the cohort required at least one neurological condition, representing a specialized subset of service users. Hirdes *et al*. (2014) reported an adjusted hazard ratio of 18.80 (95% CI: 17.40-20.32) comparing survival of home care users with the highest CHESS score to those with the lowest score. The use of the CHESS to characterize adults with IDD has not been widely reported. In an analysis of Ontario home care users with IDD (18-99 years), the prevalence of health instability was 27.6%, which was lower than the overall prevalence in home care users without IDD (37.9%) [9]. The pattern reversed after age 70, where home care users with IDD had a higher prevalence of health instability [39].

2.6 Risk Factors for Frailty and Time to Admission to LTC

The transition from community-based accommodation to a LTC facility is complex, and many factors can contribute to this decision [29,80]. Considering the multi-dimensional nature of frailty, numerous elements also influence the manifestation of frailty. Therefore, to properly assess the relationship between frailty (and health instability) and admission to LTC, careful consideration of multiple potential covariates is warranted. A review of the literature revealed the following potential covariates: age [51,81], sex [24,44,50,63,82–84], rural status [30,85–87], caregiver status [88,89], living situation [24,29,88,90], residential care history [29], cognition level [5,29,91], ethnicity [59,92,93] and income [29,92–94].

By the design of the FI, frailty is associated with age [51]. A systematic review of 36 prospective cohort studies identified strong evidence for age as a predictor of admission to LTC in the general population [81]. Young adults with IDD are admitted to LTC, which may diminish, although not remove, the relationship between age and institutionalization.

It is almost universally observed that women, compared to men, are frailer [24,50,63,82,83]. Differences can be large: the prevalence of frailty in Canadian seniors (65+ years) is estimated to be 19% in men, and 27% in women [63]. Age likely confounds this relationship because women live longer than men. In the population of adults with IDD, women have a reduced life expectancy [5,95], and so the difference between sexes may be less pronounced. Only one study of adults with IDD has reported adjusted risk ratios for sex, noting no significant differences in frailty profiles between sexes [96]. Even though women are frailer, in general, men are more susceptible to the effects of deficit accumulation compared to women, and suffer higher rates of institutionalization and mortality [24,44,83,84].

Canadians with IDD living in rural areas are older, poorer and use primary care less frequently compared to their urban counterparts [85]. In Ontario, rural areas offer fewer home and community-based services [86]. Song, Macknight, Latta, Mitnitski, and Rockwood (2007) found greater proportions of frail Canadians over the age of 80 in rural areas compared to urban areas. However, urban areas have more access to LTC, and thus greater proportions of the population may be in LTC, despite being less frail [30,87].

Studies recognize high caregiver burden or distress as a risk factor for LTC placement in the general population [88,89]. It is hypothesized that the association between frailty and rates of admission to LTC will be reduced when caregivers are experiencing high levels of burden or distress. In this scenario, the transition to LTC will be more dependent on the caregiver than on the individual with IDD [35]. Caregivers who are not distressed may cope better with frail and vulnerable individuals, thus prolonging community-based living.

In the general population, adults living alone are less likely to be frail [92,98], although this may be untrue for individuals in need of home care services. Doran *et al*. (2009) noted the combination of living alone and declining physical function increased the risk of adverse outcomes in home care clients. Adults with IDD who live alone may have fewer ADL impairments and be more independent, but when frail, may be more at risk for admission to LTC [29,90]. The presence of an informal caregiver, such as a spouse, child, or parent, may postpone LTC [24].

The relatively large proportion of individuals with IDD residing in institutions (e.g. LTC homes, psychiatric hospitals) [39,99] has raised concerns about re-institutionalization [34]. It is hypothesized that if an individual has previously resided in a residential care facility [29], he/she may be more likely to return to an institution if the factors driving the first stay are not removed. These factors could include medical needs and frailty-related conditions. Limited research has discussed residential care history as a potential covariate.

Schoufour *et al*. (2013) reported higher frailty scores for individuals with more severe intellectual disability compared to individuals with borderline intellectual disability. Low cognition levels (high levels of IDD) are associated with both low life expectancy and greater institutionalization [5,29], in addition to various health conditions (e.g. cancer, gastrointestinal, heart, hematological, or neurological problems, visual and hearing impairments) [91].

Ethnicity is also an independent predictor of both frailty [92] and institutionalization [29]. A pooled hazard ratio from a meta-analysis of predictors of nursing home admissions in community-based individuals reported a significant increase in risk for white individuals, compared to all other ethnicities [29]. Evidence of an association between race and frailty is less strong. Some report that individuals who are white, compared to other ethnicities, are more likely to be frail [92], while others report opposite associations [59], or no association [93]. Frailty may also be associated with lower income [92–94] and admission to LTC [29], although these findings are not consistently reported [24,81,87].

Measure of risk for institutionalization exist and help to inform decision-making and care planning. In Ontario, the Resident Assessment Instrument-Home Care (RAI-HC) is used to assess all home care users, and it has Clinical Assessment Protocols (CAPs) embedded within it to identify individuals at risk for outcomes including abuse, functional decline or LTC placement [100]. When CAPs are triggered, case managers receive interpretations and potential interventions to improve home care planning [100,101]. The Institutional Risk CAP detects individuals with high risk of institutionalization, and consequently, these individuals receive modified care plans [102,103]. These modifications include increased assistance with activities of daily living and efforts to minimize risks of falling [103]. Individuals with this CAP may be able to tolerate living in the community longer than individuals without the extra care [86].

2.7 Conclusion

The accumulation of deficits approach, which develops a FI, is the most appropriate method for the study of frailty; however an IDD-specific FI has yet to be developed for use in the Canadian health care system. Research shows that frailty in individuals with IDD is experienced at a younger age, compared to the general population, and until now, no studies have considered the association between frailty and rates of admission to LTC, while considering relevant covariates. Results from this thesis have the potential to impact Ontario's health policy, by providing new knowledge on health status and service utilization for persons with IDD.

2.8 References

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Chapter 3

Using an accumulation of deficits approach to measure frailty in a population of home care users with intellectual and developmental disabilities

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Abstract

BACKGROUND: The aging population of adults with intellectual and developmental disabilities (IDD) is growing. In the general aging population, frailty is commonly used to predict adverse health outcomes, including hospital use, death, and admission to long-term care. However, existing frailty measures are less appropriate for aging persons with IDD, given their pre-existing conditions and limitations. An accumulation of deficits approach, which is now widely used to describe frailty in the general population, may be more suitable for persons with IDD. Frailty measures specific to persons with IDD have not been widely studied.

METHODS: Using pre-determined criteria, a frailty index (FI) specific to persons with IDD was developed based on items in the Resident Assessment Instrument - Home Care (RAI-HC), and using the assessments of 7,863 individuals with IDD in Ontario (aged 18-99 years) admitted to home care between April 1st, 2006 and March 31st, 2014. FI scores were derived by dividing deficits present by deficits measured, and categorized into meaningful strata using stratum-specific likelihood ratios. A multinomial logistic regression model identified associations between frailty and individual characteristics.

RESULTS: The resulting FI is comprised of 42 deficits across five domains (physiological, psychological, cognitive, social and service use). The mean FI score was 0.22 (SD=0.13), equivalent to 9 deficits. Over half of the cohort was non-frail (FI score < 0.21), while the remaining were either pre-frail (21%, FI score between 0.21 and 0.30) or frail (27%, FI score > 0.30). Controlling for individual characteristics, women were more likely to be frail compared to men (OR=1.39, 95% CI: 1.23-1.56). Individuals who were frail were significantly more likely to have a caregiver who was unable to continuing caring (OR=1.86, 95% CI: 1.55-2.22) or feeling distressed (OR=1.54, 95% CI: 1.30-1.83). Living with family members was significantly protective of frailty (OR=0.35, 95% CI: 0.29-0.41), compared to living alone.

CONCLUSIONS: Using the FI to identify frailty in adults with IDD is feasible and can be incorporated into existing home care assessments. This could offer case managers assistance in identifying at-risk individuals. Future analyses should evaluate this measure's ability to predict future adverse outcomes.

Keywords: frailty, intellectual disability, developmental disability, accumulation of deficits, home care, RAI-HC, aging

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3.1 Background

Population aging and its associated challenges have been repeatedly reported by policy makers and health researchers across the globe [1]. Much of the aging research has focused on the general population; the subset with intellectual and developmental disabilities (IDD) has received relatively little attention [2].

Almost 1% of the population has an IDD [3,4], which the World Health Organization defines as "a group of developmental conditions characterized by significant impairment of cognitive functions, which are associated with limitations of learning, adaptive behaviour and skills" (page 177) [5]. These conditions originate before the age of 18 years [6], with the expectation of lifelong disability.The risk of health problems is greater among persons with IDD [7], and these contribute to the widely held notion that this group experiences premature aging, where 50 years may be considered "old" [8–12].

While many illnesses and functional impairments become increasingly prevalent with age, age by itself is an insensitive and non-specific predictor of health vulnerabilities [13,14]. Frailty is frequently identified as an effective measure of health and vulnerability [13,15]. While a consistently applied definition does not exist in the literature, frailty is generally viewed as age-related decline associated with higher risk of adverse health outcomes [16]. However, few studies have measured frailty in adults with IDD. A review of the literature revealed the existence of two research groups explicitly measuring frailty in this population, each using a different approach.

Brehmer and Weber (2010) created a frailty measure, the Vienna Frailty Questionnaire for Persons with Intellectual Disabilities – Revised (VFQ-ID-R) [17,18], which captures changes in 34 symptoms across four domains (physical, psychological, cognitive, and social). The presence of frailty is indicated if: (1) symptoms are present in at least three domains, and (2) a minimum of six symptoms are identified overall. Persons are described as "pre-frail" if only one of these criteria is met. In their sample of 147 Austrian adults with IDD aged 20-72 years, 17.7% were frail, 17.7% were pre-frail, and the remainder was non-frail [17]. The small sample size, however, hampers the generalizability of results. In addition, the difficulty in applying the components in the VFQ-ID-R to other studies or databases further limits its use.

In the Netherlands, Fried *et al.*'s (2001) five frailty phenotype symptoms (weight loss, weakness, poor endurance and exhaustion, low physical activity and slowness [19]) were measured in 848 individuals with IDD over 50 in the Healthy Aging and Intellectual Disabilities (HA-ID) study cohort [20]. Here, persons were frail if at least three of the five symptoms were present, and pre-frail if one or two were present. Evenhuis *et al.* (2012) reported that 13% of their cohort was frail and 60% was pre-frail. Approximately 11% of their sample under the age of 65 years was considered frail, which is similar to the published prevalence estimates of frailty in the general Dutch population aged 65 years or older. The findings support the hypothesis that persons with IDD experience frailty earlier than the general population [20].

The frailty phenotype approach to measuring frailty is limited in that it focuses on physical limitations, which are known to be more common in individuals with IDD regardless of age [10]. Others have noted that the strong influence of low mobility on frailty phenotype scores may lead to misclassification in the general population [21]; this may also lead to misclassification among those with IDD [10], who have higher rates of mobility limitations across the lifespan.

In a follow-up study, the HA-ID research group used an accumulation of deficits approach [22] to measure frailty in 982 adults 50 years or more using formal care in the Netherlands [10]. The accumulation of deficits approach emphasizes the proportion of health deficits a person has, rather than the presence of specific symptoms. The mean frailty score was 0.27 (SD=0.13), with an upper limit of 0.69 (on a continuum ranging from 0 to 1, with a higher score representing frailty), and approximately 66% of the sample was frail (score above 0.20). They also found that individuals with IDD had similar rates of frailty at age 50 as did the European population at age

70, again supporting previous work showing that persons with IDD are frail at earlier ages than in the general population.

While these are the only studies reporting on frailty measures developed specifically for populations of persons with IDD, generic frailty measurements have also been applied to this population. For example, in Ontario, Canada, the Frailty Marker, derived from the Johns Hopkins University Adjusted Clinical Group (ACG) System, is used to identify frailty in population-based studies relying on administrative data [23]. This marker categorizes individuals as frail based on 81 diagnostic codes. As the actual diagnostic codes are unknown (i.e., not published), it is suspected that the Frailty Marker relies on the presence of specific medical conditions and fails to capture the multiple domains required for a well-balanced frailty measure. As such, it may not be the most appropriate measure of frailty, which limits its use for both individual-level and population-level health service planning – especially among persons with IDD.

The objective of this study is to describe the process of applying the accumulation of deficits approach to develop a frailty index (FI) based on administratively held clinical data for use among community-dwelling adults with IDD receiving home-based health care services (i.e., home care), and to identify individual characteristics associated with frailty. Using associations with 1-year admission to long-term care (LTC), appropriate cut-off scores for the newly developed FI are determined, and the prevalence of frailty in this population is described.

3.2 Methods

3.2.1 Design/Participants

This study is part of a larger program of research focused on Health-Care Access Research and Developmental Disabilities (H-CARDD) (see www.hcardd.ca). In this study, the cohort includes 7,863 individuals living in Ontario in 2009/10, identified in one or more administrative health datasets (such as physician claims or hospital visits) as per a previous study [4,23] (see Appendix A) and who had at least one home care assessment between April 1st, 2006 and March 31st, 2014.

Individuals were between the ages of 18 and 99 years in 2009/10 and at their first home care assessment. The study protocol was reviewed and approved by the institutional review board at the Queen's University Health Sciences Research Ethics Board and Sunnybrook Health Sciences Centre, Toronto, Canada (see Appendix B).

In the study cohort, 52% are female, and 17% are living in a rural area (Table 3.1). The mean age of the sample at the first home care assessment is 56.2 years (median=57 years). Twenty-two percent (22%) of individuals reportedly lived alone, 17% lived with a spouse and/or child(ren), and 35% lived with other family members. A further quarter (25%) of individuals lived in a group setting with non-relatives (e.g. a group home).

3.2.2 Data Linkage

The RAI-HC database contains the RAI-HC assessments provided by the Home Care Reporting System (HCRS). These data are stored with interRAI Canada and shared with the Institute for Clinical Evaluative Sciences (ICES). Data included the earliest home care assessment occurring in the study period for each cohort member. The Registered Persons Database (RPDB) contains information on all Ontario persons eligible for health coverage. For this study, it provided age (in years). The date of admission to LTC was retrieved from the Continuing Care Reporting System for Long-Term Care (CCRS-LTC) database, which provides demographic and clinical information about individuals receiving care in LTC homes. All accessed data had been de-identified using methods to ensure confidentiality and privacy. Using a patient identifier, known as the ICES Key Number (IKN), information for the cohort from three databases was linked.

Table 3.1. Cohort characteristics

	n	%
Sex		
Female	4105	52.2%
Male	3758	47.8%
Rural Status (n=7,834)		
Rural	1337	17.1%
Urban	6497	82.9%
Caregiver Inability		
Yes	1006	12.8%
No	6857	87.2%
Caregiver Unsatisfied		
Yes	254	3.2%
No	7609	96.8%
Caregiver Distress		
Yes	1270	16.2%
No	6593	83.8%
Living Situation (n=7,862)		
Lives alone	1733	22.0%
Lives with Spouse and/or Child(ren)	1337	17.0%
Lives with Other Family	2769	35.2%
Lives in Group Setting	2023	25.7%
Residential Care History (n=7,862)		
Yes	647	8.2%
No	7215	91.8%
Cognition Level (CPS) (n=7,861)		
Intact (0)	1086	13.8%
Borderline Intact (1)	1232	15.7%
Mild Impairment (2)	1416	18.0%
Moderate+ Impairment (3+)	4127	52.5%
CAP (Institutional Risk)		
Triggered	965	12.3%
Not Triggered	6898	87.7%

3.2.3 Dataset Creation

In Ontario, home care services are provided by the Ministry of Health and Long-Term Care's fourteen regional Community Care Access Centres, who determine eligibility and coordinate providers. Services provided include visiting health professionals, help from personal support workers, homemaking services and community support services. Case managers use the Resident Assessment Instrument- Home Care (RAI-HC) [24] as a needs assessment tool to assess those receiving, or about to receive, home care. InterRAI is an international collaboration that serves to collect and interpret data on health and social outcomes. The interRAI assessments, including the RAI-HC, have been implemented as routine and standardized measures in various care settings by several jurisdictions internationally, including several Canadian provinces [25]. The RAI-HC assessment captures information related to demographic characteristics, home environment, functioning, health, medications, informal support, and formal health services. With respect to demographic characteristics, the current study used age, sex, living situation (i.e., alone, with a spouse and/or child(ren), with other family, or in a group setting with non-relatives), residential care history (i.e., lived in a residential care facility in the last 5 years), and rural status . Following the Statistics Canada definition, rural status was defined as living in a "location not included in a [Canadian census metropolitan area or census agglomeration], living in an urban centre of fewer than 10,000, or living in a rural area" (p. 157) [26]. Rural status was determined from postal codes identified at time of assessment.

Following Brehmer and Weber (2010)'s categorization of frailty domains, the health-related items of interest were categorized into cognitive, physiological, psychological, and social domains. An additional domain of "service use" captures other service-related items [10,27]. The items for each domain were all selected from information available in the RAI-HC. Twenty-seven items related to cognitive patterns (e.g. memory loss, delirium), communication (e.g. ability to understand others), and practical skills (i.e., instrumental activities of daily living; e.g. help needed with ordinary housework, managing finances, shopping) were used to inform on the person's cognitive functioning. One-hundred fourteen items were available to assess physiological health, such as: hearing, vision, bladder and bowel continence, health conditions (e.g. diarrhea, shortness of breath), nutritional status (e.g. morbid obesity, insufficient fluid intake), dental and oral status (e.g. chewing problems), skin conditions (e.g. pressure ulcers, wound care required), medical diagnoses (e.g. hypertension, Alzheimer's disease, hip fracture, diabetes), and medications (e.g. use of anxiolytics, medication compliance). Psychological status was informed by seventeen items related to mood (e.g. feelings of sadness or depression, repetitive anxious complaints) and behaviours (e.g. wandering, verbal abuse). Items from the social domain included social isolation, withdrawal from activities of interest, and five other items.

In addition to these domains, items indicating home environment (e.g. difficulty accessing rooms in house, inhabitable heating/cooling) and service utilization (e.g. recent hospital admissions, unmet treatment goals) were used. Two global health status indicators were also accessed: self-reported health (asking if the individual feels he/she is in poor health) and the presence of diseases or conditions that make cognition, activities of daily living (ADL), mood, or behaviour patterns unstable.

Three variables related to informal support were included in this study: (1) caregiver is unable to continue caring ("caregiver inability"), (2) caregiver is unsatisfied with support from family and friends ("caregiver unsatisfied"), and (3) caregiver expresses feelings of distress, anger or depression ("caregiver distress").

In addition to individual items, two measures embedded in the RAI-HC were included. Clinical Assessment Protocols (CAPs) identify common risks for individuals using home care, such as abuse, functional decline or LTC placement [28]. Algorithms using some RAI-HC items trigger CAPs, which then offer interpretations and potential interventions for case managers to include in home care planning [28,29]. The Institutional Risk CAP is triggered for individuals with a high risk of institutionalization and suggests ways of remaining in the community [30]. The second embedded measure is the Cognitive Performance Scale (CPS), which provides the cognition level and characterizes individuals on a scale from 0 (intact cognition) to 6 (very severe impairment) [31]. Individuals with CPS scores ≥3 (i.e., moderate or worse cognitive impairment) were grouped into one category [32,33].

3.2.4 Analysis

3.2.4.1 Selection of health deficits

The RAI-HC assessment provided 180 deficit variables. Previous work has suggested that deficits can be signs, symptoms, disabilities, diseases or abnormal laboratory measurements [22].The criteria for selecting variables, published first by Searle *et al.* (2008) and then modified by Schoufour *et al.* (2013) for persons with IDD, are described.

First, each deficit must be positively correlated with age. This was done by calculating Spearman's correlation coefficients (r_s) between each deficit (ordinal variables) and age (as a continuous variable). Deficits that were not significantly and positively correlated with age were excluded, using a cut-off of r_s=0.05 ($p<0.0001$). Second, deficits that were too saturated were excluded to prevent ceiling effects, using a prevalence cut-off of 80%. For variables that were not dichotomous, deficits were considered present if any limitation existed. Third, the deficit must be associated with health status, which was determined using Chi-square tests (included if $p<0.05$) for association. Fourth, a wide range of health aspects should be included in the FI. A review of the literature was conducted to determine if the deficits appeared to cover different aspects of health, including all five domains of health [18]. Searle *et al.'s* (2008) final criterion related to use of identical items over time is not relevant to this study, which assesses frailty at a single point in time [22].

Schoufour *et al.* (2013) developed further inclusion criteria for the FI that are appropriate for persons with IDD [10]. If a deficit variable has missing data for greater than 30% of individuals, it should be excluded. Second, deficits were considered uncommon and excluded if prevalent in fewer than 5% of individuals, to prevent floor effects. However, wherever possible, related variables were grouped to form multi-item deficits with a sufficient prevalence. Schoufour *et al.'s* (2013) criteria also suggested reducing the number of variables if they appear repetitive. Variables were grouped into twenty-seven categories, and correlations between remaining variables within categories were determined. Variables that were very highly correlated ($r > 0.9$) were either grouped into a multi-item deficit, or only the item with the highest correlation with age was included.

The list of excluded variables was screened by experts (the authors) for deficits unexpectedly omitted. This current study added one further stipulation. In an attempt to identify a change in deficits, which is crucial to capturing frailty [14,15], deficits were grouped whenever possible to create a "decline" variable to ensure that the FI included recent deficits, rather than long-standing functioning, health, or behaviour patterns.

3.2.4.2 Calculation of the continuous FI

Most variables were ordinal or dichotomous. Variables were recoded, if necessary, to scores of 0 (deficit not present), 0.5 (intermediate deficit), or 1 (full deficit present). One continuous variable ("falls frequency") was recoded into an ordinal variable as 0, 0.25, 0.5, 0.75, and 1.0. The rescaling of deficits was congruent with previous publications [34,35], although some expert judgment was required. Variables were not weighted, therefore all selected deficits contributed to the final FI score equally [36].

A FI score was calculated for each individual by dividing the sum of the deficit scores by the number of deficits measured, to create continuous values between 0 (no deficits present) and 1 (all deficits present).

3.2.4.3 Categorizing the FI

The FI can be informative as a continuous variable to describe and contrast populations' distributions of vulnerability. However, to ease comparisons, the FI is often categorized into meaningful groups though cut-offs have not been consistently applied across studies [34]. Hoover *et al.* (2013) reported methods to validate cut-off points for the FI, using stratum-specific likelihood ratios (SSLRs), to distinguish between frail and non-frail seniors and to identify "natural" ranges of frailty associated with different risks of adverse outcomes. SSLRs represent the likelihood that individuals in a specific frailty group (stratum) will experience an event (admission to LTC in 1-year follow-up) relative to their likelihood of not experiencing an event [34,37]. Using a subset of the cohort (n=7,115) with a home care assessment between April 1st, 2006 and March 31st, 2013, SSLRs were calculated. SSLRs are independent of the population prevalence [34], and are less susceptible to spectrum bias (i.e. change in measure characteristics due to a different mix of severity) than a single cut-off. This process helps to ensure that lower and higher scores are correctly assigned to their own corresponding group [38].

The ten stratum cut-off points identified by Hoover *et al.* (2013) were used in this study. Strata were collapsed if there was an insufficient number of events or non-events, or if 95% confidence intervals clearly overlapped [39]. Confidence intervals were calculated using equations presented by Peirce and Cornell (1992). This process ensured that strata were significantly different from each other.

3.2.4.4 Statistical analysis

The mean, standard deviation, and the maximum and minimum scores for the continuous FI are reported. A histogram shows the distribution of FI scores. Goodness-of-fit tests (e.g. the Cramer von- Mises test) assessed whether the distribution fit a Weibull or gamma distribution [40,41]. The mean FI score per year was estimated by calculating the regression coefficient β [10], and the relationship between the upper limit of the FI and age was determined by plotting the 99th percentile of each 10-year age group. The slopes of these scores can indicate the presence of an age-invariant sub-maximal limit to the FI, demonstrating that even with advancing age, no further deficits are accumulated.

Bivariate multinomial logistic regression models were completed to calculate odds ratios and 95% confidence intervals to report the odds of frailty (pre-frail or frail compared to non-frail), by individual characteristics. These groups included age (per 10 year increase), sex, rural status, caregiver status variables (i.e. caregiver inability, caregiver unsatisfied, caregiver distress), living situation, residential care history, cognition level and the Institutional Risk CAP.

An adjusted multinomial logistic regression model was developed to determine adjusted odds ratios of frailty (pre-frail or frail compared to non-frail) and individual characteristics (listed above). Using backwards elimination to select significant covariates, at a significance level of $\alpha=0.05$ using the Wald test, the model retained significant covariates.

An analysis of variance (ANOVA) was completed to compare frailty groups by age (continuous). The correlation between frailty and self-reported health was determined. Bivariate logistic regression models were also developed for each individual deficit in the FI and 1-year admission to LTC; odds ratios and 95% confidence intervals were determined.

All tests were two-sided tests, with an alpha value of 0.05, unless otherwise stated, to indicate statistical significance. All analyses were done using SAS Enterprise Guide version 6.1.

3.3 Results

3.3.1 Development of the FI

3.3.1.1 Health deficits

The resulting FI includes 42 deficits across five domains (Appendix C): physiological (n=29, where 7 are related to ADLs and 10 to disease diagnoses), cognitive (n=4), psychological (n=3), social (n=3), and service use (n=2).

Only variables that met all of the criteria described were included as deficits in the FI (Appendix D provides the results of all items). One-hundred twenty-five deficits, such as smoking daily, bladder incontinence and multiple sclerosis, were excluded due to poor or negative correlation with age. Only one item was excluded due to a high prevalence (poor cognitive skills for decision-making), however sixty-seven variables did not meet the prevalence criterion (e.g. malnutrition, renal failure, hallucinations, vision decline). One variable, an instrumental activity of daily living (phone use), was excluded because over 30% of the cohort had missing information or the activity did not occur.

Several items were pooled to create combined variables, including a general circulatory disease deficit that contained congestive heart failure, irregularly irregular pulse and peripheral vascular disease items. In addition, seven activities of daily living (ADL) variables were each combined with an ADL decline variable to create new items.

Some variables were highly correlated (e.g. pain intensity and pain frequency) (Appendix E) and only one (i.e. pain frequency) was retained in the FI. No variables were deemed to be excluded inappropriately, due to the grouping of similar variables.

No participants had fewer than 39 variables measured: 85% had complete data for all 42 deficit variables included, while the remaining 15% were missing three deficits. The group with no missing deficits was compared to the group with missing deficits, and there were significant differences in frailty profiles (χ^2=997, p<0.0001). The group missing deficits were significantly more likely to be in the frail group (63%) compared to the group with no missing deficits (20%). The only deficits contributing to missing data were from the social domain.

The FI was more strongly associated with admission to LTC after 1-year (OR=4.45, 95% CI: 3.82-5.19) compared to the 42 individual deficits (Appendix F). Nine deficits were not significantly associated with admission to LTC. The odds ratios of four deficits, all ADL decline items, were not significantly different than the FI's odd ratio.

3.3.1.2 Characteristics of continuous FI

The mean score of the FI was 0.22, with a standard deviation of 0.13, and a range of 0 to 0.77. Forty-eight (0.6%) individuals were in a "zero state", with no deficits present at all. The 99th percentile of the FI was 0.56, which is below the theoretical maximum of 1.0. The slope of age and the 99th percentile of the FI was significant (r=0.49, (<0.0001), even in the subset of the cohort over age 50 years (r=0.55, p<0.0001) (Figure 3.1).

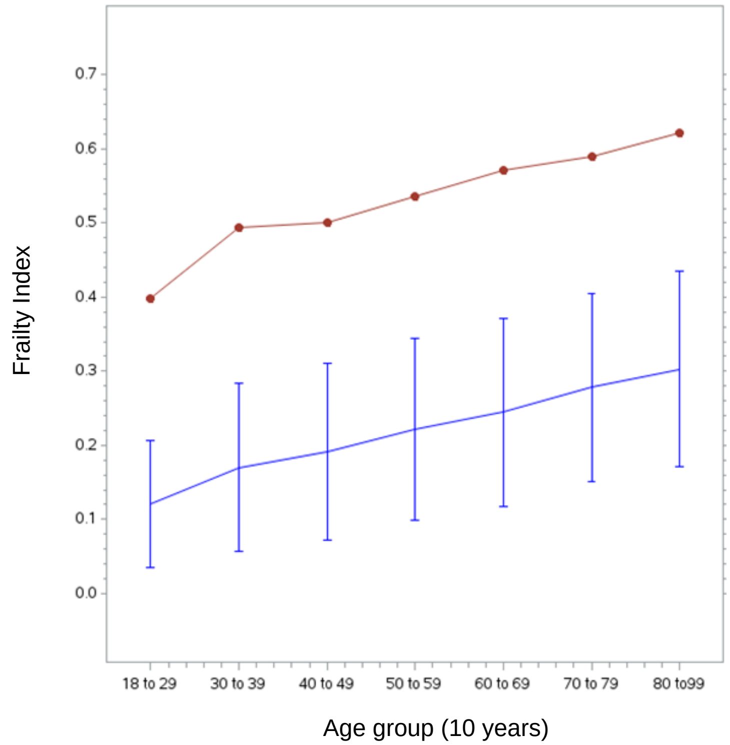

Figure 3.1. FI versus age plot: Average FI (bottom) with standard error bars, and 99th percentile of the FI (top), against 10-year age groups

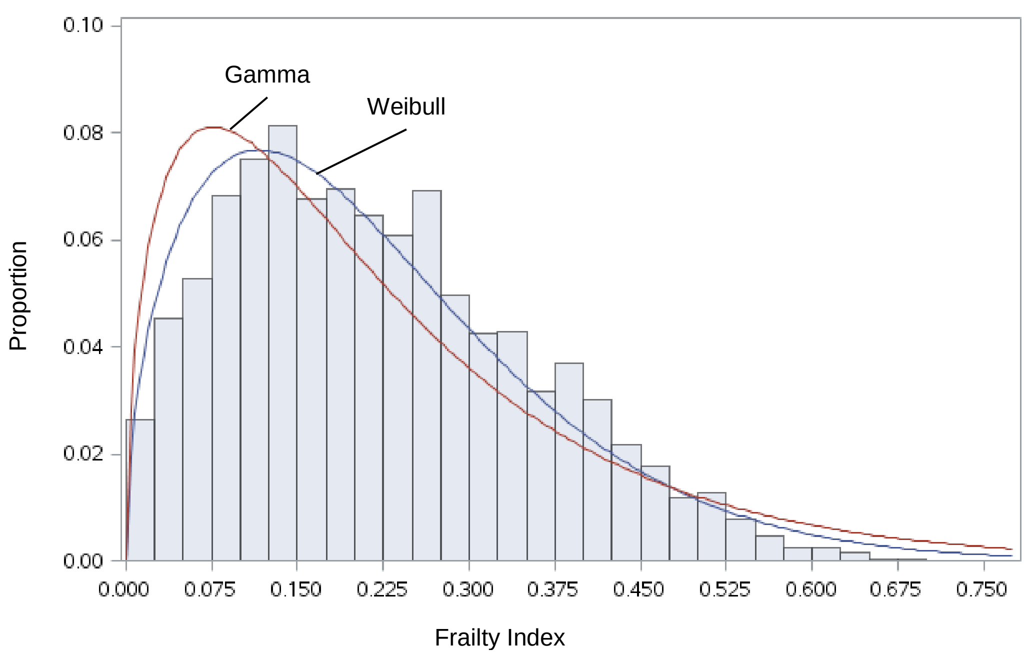

Figure 3.2. Distribution of the FI with Weibull and gamma distribution curves

For each increase of 1 year, the average change in mean of FI was +0.003 (+0.016 on a log scale), indicated by the average slope of deficit accumulation. The index had a right-skewed distribution (skewness value= 0.612), although the FI distribution was significantly different than both the gamma distribution ($p<0.001$) and the Weibull distribution ($p=0.010$) (Figure 3.2).

3.3.1.3 Stratum-specific likelihood ratios

Initially, analyses used ten strata to categorize the FI, however not all strata had significantly different SSLRs when considering 1-year admission to LTC as the outcome (Table 3.2). Strata were combined to increase the power to detect differences between groups. The >0.10 to ≤0.21 stratum was significantly different than the previous stratum, identifying two distinct groups with likelihood ratios less than 1.0, which indicates a reduced risk of an adverse outcome. These strata were combined into a single, non-frail group (≤0.21).

Further categorization suggests three frailty categories with significantly different risks of experiencing an event: non-frail (≤0.21), pre-frail (>0.21 to ≤0.30), and frail (>0.30). The frail group had a significantly higher likelihood of 1-year admission (SSLR=2.21, 95% CI: 2.05-2.38), while the pre-frail group had neither an increased nor decreased likelihood.

Table 3.2. Stratum-specific likelihood ratios of 1-year admission to LTC, with 10 strata and 3 strata, and 95% confidence intervals (n=7,115)

FI Strata	N	$SSLR_{10}$	95% CI Lower	95% CI Upper	$SSLR_3$	95% CI Lower	95% CI Upper
≤ 0.03	196	0.24	0.12	0.47			
> 0.03 to ≤ 0.10	1199	0.29	0.23	0.37	0.50	0.45	0.55
> 0.10 to ≤ 0.21	2301	0.63	0.56	0.71			
> 0.21 to ≤ 0.23	367	0.98	0.74	1.29			
> 0.23 to ≤ 0.25	445	1.02	0.79	1.30	1.05	0.93	1.19
> 0.25 to ≤ 0.27	260	0.95	0.68	1.33			
> 0.27 to ≤ 0.3	447	1.21	0.96	1.52			
> 0.30 to ≤ 0.35	601	1.68	1.41	2.01			
> 0.35 to < 0.45	844	2.14	1.87	2.45	2.21	2.05	2.38
≥ 0.45	455	3.21	2.69	3.84			

3.3.2 Prevalence of Frailty

Over half (51.8%) of the cohort was non-frail at the time of their first home care assessment. The remaining individuals were pre-frail (21.3%) or frail (26.9%) (Table 3.3).

Table 3.3. Cohort characteristics by FI group

	Non-frail		Pre-frail		Frail	
Overall	4073	51.8%	1676	21.3%	2114	26.9%
Sex						
Female	1939	47.6%	924	55.1%	1242	58.8%
Male	2134	52.4%	752	44.9%	872	41.2%
Rural Status (n=7,834)						
Rural	681	16.7%	270	16.2%	386	18.4%
Urban	3386	83.3%	1399	83.8%	1712	81.6%
Institutional Risk CAP						
Triggered	161	4.0%	237	14.1%	567	26.8%
Not Triggered	3912	96.0%	1439	85.9%	1547	73.2%
Caregiver Inability						
Yes	450	11.0%	189	11.3%	367	17.4%
No	3623	89.0%	1487	89.7%	1747	82.6%
Caregiver Unsatisfied						
Yes	124	3.0%	57	3.4%	73	3.5%
No	3949	97.0%	1619	96.6%	2041	96.5%
Caregiver Distress						
Yes	578	14.2%	291	17.4%	401	19.0%
No	3495	85.8%	1385	82.6%	1713	81.0%
Living Situation (n=7,862)						
Lives Alone	680	16.7%	486	29.0%	567	26.8%
Lives with spouse and/or child(ren)	432	10.6%	386	23.0%	519	24.6%
Lives with Other Family	1894	46.5%	427	25.5%	448	21.2%
Lives in Group Setting	1067	26.2%	376	22.5%	580	27.4%
Residential Care Facility (n=7,862)						
Yes	342	8.4%	132	7.9%	173	8.2%
No	3731	91.6%	1543	92.1%	1941	91.8%
Cognition Level (CPS) (n=7,861)						
Intact (0)	564	13.8%	276	16.5%	246	11.6%
Borderline Intact (1)	692	17.0%	268	16.0%	272	12.9%
Mild Impairment (2)	631	15.5%	360	21.5%	425	20.1%
Moderate+ Impairment (3+)	2186	53.7%	771	46.0%	1170	55.4%

The mean age was higher in the frail group (64.5 years) compared to the non-frail group (50.0 years) ($p<0.001$). Frailty was positively correlated with poor self-reported health ($r_s=0.21$, $p<0.001$). Women were significantly more likely to be pre-frail (OR=1.30, 95% CI: 1.25-1.35) and frail (OR=1.47, 95% CI: 1.42-1.53) compared to men (Table 3.4). Living in a rural area, rather than an urban area, did not significantly change the odds of being pre-frail or frail over non-frail ($p>0.05$). A prior admission to a residential care facility was significantly associated with reduced odds of being pre-frail or frail (OR=0.60, 95% CI: 0.47-0.76). Individuals who triggered the Institutional Risk CAP were much more likely to be frail compared to those who did not (OR=9.19, 95% CI: 7.47-11.31). This relationship between triggering a CAP and being pre-frail was also strong (OR=4.32, 95% CI: 3.45-5.42) (Table 3.4).

Living with a spouse and/or child(ren) was not associated with being frail ($p>0.05$) compared to living alone. Conversely, living with other family members or living in a group setting was protective against frailty (Family: OR=0.35, 95% CI: 0.29-0.41, Group Setting: OR=0.60, 95% CI: 0.47-0.76) compared to living in alone (Table 3.4).

Caregivers who reported that they were unable to continue caring had higher odds of caring for an individual who was frail compared to caregivers who did not report that they were unable to care (OR=1.86, 95% CI: 1.55-2.22). Being pre-frail did not appear to be associated with these caregiver feelings (OR=1.13, 95% CI: 0.93-1.37). Caregiver distress, however, was associated with increased odds of the individual with IDD being frail (OR=1.54, 95% CI: 1.30-1.83), and pre-frail (Table 3.4). Individuals with caregivers who were unsatisfied by the support from other informal caregivers were not at increased risk of frailty compared to those who were satisfied.

Table 3.4. Unadjusted and adjusted odds ratios of frailty and individual characteristics from bivariate logistic regression models and a multinomial logistic regression model

Independent Variable	Pre-frail vs. Non-frail		Frail vs. Non-frail	
	Unadjusted ORs (95% CI)	Adjusted ORs (95% CI)	Unadjusted ORs. (95% CI)	Adjusted ORs (95% CI)
Age (per 10 year increase)	**1.42 (1.38-1.47)**	**1.30 (1.25-1.35)**	**1.63 (1.57-1.68)**	**1.47 (1.42-1.53)**
Sex (Female vs. Male)	**1.35 (1.21-1.52)**	**1.22 (1.08-1.38)**	**1.57 (1.40-1.74)**	**1.39 (1.23-1.56)**
Rural Status (Rural vs. Urban)	0.96 (0.82-1.12)	-	1.12 (0.98-1.29)	-
Caregiver Inability (Yes vs. No)	1.02 (0.86-1.23)	1.13 (0.93-1.37)	**1.69 (1.46-1.96)**	**1.86 (1.55-2.22)**
Caregiver Unsatisfied (Yes vs. No)	1.12 (0.82-1.54)	-	1.14 (0.85-1.53)	-
Caregiver Distress (Yes vs. No)	**1.27 (1.09-1.48)**	**1.53 (1.29-1.83)**	**1.42 (1.23-1.63)**	**1.54 (1.30-1.83)**
Living Situation (ref. group = Lives Alone)				
Lives with Spouse and/or Child(ren)	**1.25 (1.04-1.50)**	0.99 (0.82-1.19)	**1.44 (1.22-1.70)**	0.90 (0.75-1.09)
Lives with Other Family	**0.32 (0.27-0.37)**	**0.43 (0.36-0.51)**	**0.28 (0.24-0.33)**	**0.35 (0.29-0.41)**
Lives in Group Home	**0.49 (0.42-0.58)**	**0.53 (0.44-0.63)**	**0.65 (0.56-0.76)**	**0.59 (0.49-0.70)**
Residential Care History (Yes vs. No)	0.93 (0.76-1.15)	0.81 (0.64-1.02)	0.97 (0.80-1.18)	**0.60 (0.47-0.76)**
Cognition Level (CPS) (ref. group= Intact)				
Borderline Intact (1)	**0.79 (0.65-0.97)**	0.94 (0.76-1.17)	0.90 (0.73-1.11)	1.08 (0.87-1.36)
Mild Impairment (2)	1.12 (0.96-1.42)	1.18 (0.96-1.45)	**1.54 (1.27-1.87)**	**1.41 (1.14-1.75)**
Moderate+ Impairment (3+)	**1.23 (1.04-1.45)**	0.98 (0.81-1.18)	**0.72 (0.61-0.85)**	**1.46 (1.20-1.77)**
Institutional Risk CAP (Triggered vs. Not)	**4.00 (3.25-4.93)**	**4.32 (3.45-5.42)**	**8.91 (7.40-10.71)**	**9.19 (7.47-11.31)**

Bolded values indicate significant at α=0.05

3.4 Discussion

3.4.1 Characteristics of the FI

This study applied an accumulation of deficits approach using deficits specific to a population of home care users with IDD. Frailty indices used in the general population may fail to identify items that are pertinent to this population because of a failure to account for life-long disabilities [10].

A FI was created using 42 RAI-HC variables. An index with a minimum of 30-40 deficits is reliable enough to predict adverse outcomes [42]. In the general population, mean FI scores derived using similar methods but different data sources and populations vary significantly: from 0.068 in a study of community-dwelling individuals aged 15-102 years [13], to 0.32 in an acute-care population of adults over 70 years old [35]. In Schoufour *et al.*'s (2013) study of older adults with IDD living in the community ($\geq$50 years), the mean frailty score was 0.27. The mean FI of the current study of 18 to 99 year olds was lower than that for Schoufour *et al.*'s (2013) older population: a mean of 0.22 was calculated.

The FI had right-skewed distribution [10,22,40]. Mitnitski et al. (2001) reported that in populations that are "well", the FI should follow a gamma distribution. Gamma distributions, and similar distributions (e.g. log-normal, Weibull), are often used for survival analysis and in frailty models [41]. The distribution of the FI in the current study followed neither a gamma nor a Weibull distribution, but it was right-skewed.

Another characteristic typically discussed is the presence of a sub-maximal, age-invariant limit. Typically, this limit is roughly two-thirds the maximum frailty score of 1.0 [22]. Sub-maximal limits indicate that the measure has no ceiling effects [43], which occur when a measure has a distinct upper limit for possible scores, and many participants are near this limit [44]. From a clinical perspective, a sub-maximal limit relates to the notion that an individual can be as frail as they could be, without experiencing every deficit or illness possible. This study did not find a sub-maximal limit where the slope of age and the 99th percentiles of the FI became negligible. However, the maximum score reached was 0.77, indicating that very few individuals accumulated more than 30 deficits and survived in the community. While few individuals have high scores, typically even fewer individuals are in the zero state (i.e. a score of 0) as measured by a FI [45], which was also observed in the current study (<1%).

The last characteristic of frailty indices frequently reported is the mean rate of deficit accumulation per year increase in age. While this rate varies, it is generally around 0.03 deficits per year on a log scale [42]. This rate is almost twice as high as the rate observed in the current study (0.016). Initially, this appears to give evidence against the hypothesis of premature aging in IDD. However, premature aging is not necessarily equivalent to fast aging. It could be that individuals with IDD begin accumulating deficits sooner, but more gradually. Rockwood *et al.*'s (2011) relatively younger population of individuals (mean age 44 years) observed a slope of the line relating to the log mean of the FI of 0.029, almost double the slope observed in the current study. Unlike Rockwood *et al.* (2011)'s findings, where a mean score of 0.21 was reached at 70 years of age, a mean score of 0.21 was reached in the mid-forties.

3.4.2 Validation of the FI

Other research using this approach have validated the resulting measures of frailty across countries [42], ages [13], and health care settings [35,46,47], each potentially including different deficits following the same definition of frailty. However, as different and new deficits were selected for the current study, there is a need to consider and acknowledge the validity of the measure.

Rockwood *et al.* (2000) specified that a measure of frailty should have: content validity (e.g. multisystem impairment, instability, changes over time, an allowance for heterogeneity within a population), construct validity (e.g. a positive association with aging), and criterion validity (e.g. a positive association with risk of adverse outcomes). Using this definition, the FI derived in this study appears to meet all the aspects defined.

To ensure that the measure considered multi-system impairment, deficits were categorized into five general domains (physiological, psychological, cognitive, social and service use) following guidelines in the literature. The number of dimensions of frailty vary [48–50], however Brehmer and Weber's (2010) categorization complies with the notion that frailty should include the environmental and social context of a person [14]. The FI created in the current study has deficits in all domains.

The definition of frailty also includes an element of instability. The derivation process ensured that the FI included items that captured a recent status change or decline wherever possible. The way a health deficit develops can be as critical, or even more critical, than the presence of the deficit itself [14,51]. This may be especially true in persons with IDD, who likely have lower baseline health status due to life-long disabilities and higher predispositions to various health conditions [45].

Construct validity refers to whether the measure corresponds to similar measures and constructs [52]. Frailty should have some, although not perfect, correlation with age, co-morbidity and self-rated health [53]. In addition to a significant, positive association with age, the FI was positively correlated with poor self-reported health. A measure of co-morbidity was not included in the available data, but would have strengthened the construct validity of the FI.

Criterion validity is necessary to test a new measure against a reference. The measure should be compared to either a gold-standard test or evaluated based on its predictive ability. As there is no gold standard for frailty, a measure's ability to predict adverse outcomes is the best method of validation. Adverse outcomes independently associated with frailty include, but are not limited to, mortality [54,55], hospitalization [19] or falls [56,57]. In this study's adjusted model, the FI is significantly associated with age and admission to LTC. More work is required to understand the relationship between the FI and adverse outcomes, while considering relevant confounders.

In addition to assessing the validity of the FI, attempts were made to validate cut-off points for the FI. This study identified three frailty categories that can be used to distinguish between risk groups among persons with IDD. Defining three distinct groups, rather than four or five as in some other studies, may better distinguish between groups in future studies of adults with IDD with smaller sample sizes. While important information can be lost when forcing a single cut-off to create a dichotomous variable [39], three groups retain the potential to identify different risk groups. This was evident in the bivariate relationships presented in this study. For example, the caregiver inability variable had a strong relationship with the frail group, but is not strongly associated with the pre-frail group.

Using the FI, the 0.21 cut-off is widely used to discriminate between fit and frail in both the population with IDD [58] and in the population of Canadian seniors [13,27,34,59]. Three frailty categories were identified. Hogan *et al.* (2012) used similar cut-offs (<0.2, 0.2-0.3, >0.3) and labels. The "pre-frail" categorization is congruent with the literature: pre-frailty is an intermediate status that may indicate an increased risk of adverse outcomes [17,19,60].

Given that this FI specific to persons with IDD incorporates some items not included in previous studies, the potential impact of any given variable on the FI should be considered. Several studies have conducted re-sampling methods similar to "bootstrapping" by randomly selecting 80% of deficits for inclusion in the FI and repeating this process for 1000 iterations. Using this strategy, authors have consistently reported negligible changes to the slope of the FI and age [10,22,35,61] concluding that there is little sensitivity as to which deficits are included in the construction of the FI [35]. In other words, the proportion of accumulated deficits is more important than specific deficits. Given the consistency of these previous findings, this procedure was not repeated.

3.4.3 Descriptive Findings

Females had an increased risk of being pre-frail and frail. On average, women have more deficits than men, although they often tolerate these deficits better. Women have better rates of survival compared to men with the same level of frailty [10,62,63].

Frailty was strongly associated with caregiver status. Frail individuals require more care and daily assistance than pre-frail or non-frail individuals, and this may have some harmful effects on a primary caregiver's well-being. Being pre-frail (rather than frail or non-frail) was not significantly associated with poor caregiver status, suggesting it is not until an individual accumulates roughly 12-13 deficits (FI score ≈ 0.3) that caregivers feel that they are unable to continue to care. Providing care for the frail elderly can lead to physical and mental health problems, and stress or burnout can increase the risk of admission to a LTC home for the person being cared for [64,65]. Service providers should consider the resilience of families requesting services [66] to prevent crises that could lead to a move to a residential care facility. Additional research may wish to investigate if certain frailty deficits are larger contributors to caregiver distress than others.

3.4.4 Strengths and Limitations

Strengths of this study include the use of a standardized assessment instrument, the RAI-HC, as a source of data regarding deficits to create the FI. Items from the RAI-HC have strong correlations with their gold standard equivalents [67,68], high internal consistencies (between 0.6 and 0.8) [69,70] and high inter-rater reliability (mean κ=0.69) [25]. The strength of the RAI-HC in this study is the wide range of items available, which provided deficits for all five domains of interest (physiological, psychological, cognitive, social, and service use). Compared to other datasets used to apply the accumulation of deficits approach to frailty measurement, the RAI-HC provided items that incorporated recent changes in health status, which is especially important among those with IDD.

This study has some limitations. It would have been informative to have had additional demographic information, such as socioeconomic status. In the general population, socioeconomic status is associated with frailty, after controlling for race, age, and co-morbidities [71].

A future study might compare the level of frailty experienced by persons with IDD and in the general population to inform our understanding of aging with IDD. While this FI was created for persons with IDD, by emphasizing change in deficits and issues known to be related to frailty, it could easily be applied to the general population of home care users in Ontario, or elsewhere.

3.5 Conclusion

Measuring frailty among persons with IDD using home care services is feasible. This study has identified health deficits applicable for those with IDD to include in a FI and has presented cut-off points for the FI to distinguish between risk groups.

Premature aging has frequently been reported in adults with IDD; however the increased vulnerabilities that come with aging are rarely quantified. Frailty may be a better way to understand the needs of the young old with IDD. In the general population, caring for elderly citizens is particularly challenging due to the blend of both medical and social problems [14], however adults with IDD face these challenges throughout their lives and these may worsen as they age.

Next steps include applying the FI to predictive models. If the FI is associated with time to adverse events (e.g. admission to LTC), the potential exists to use this measure as a tool in the community.

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Chapter 4

Frailty as a predictor of institutionalization among adults with intellectual and developmental disabilities using a frailty index and the interRAI CHESS Scale

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Abstract

BACKGROUND: Adults with intellectual and developmental disabilities (IDD) become frail earlier and more frequently than the general population, resulting in higher care needs. Despite a shift towards community-based care to meet increasing demands, there is concern that the population of individuals with IDD is at risk for institutionalization, or re-institutionalization, in long-term care homes.

METHODS: A retrospective cohort design was used to follow 3,034 individuals (aged 18 to 99 years) living in Ontario, Canada and assessed with the Resident Assessment Instrument – Home Care. Individuals were characterized as frail using a frailty index specific to persons with IDD, and as unstable with the interRAI Changes in Health, End-stage disease, Signs and Symptoms (CHESS) scale. Survival analyses determined differences in rates of admission to long-term care.

RESULTS: Over a 4-year period, 22% of the sample was admitted to long-term care. Controlling for age, sex, rural status, caregiver status, living situation, and cognition level, frail individuals had rates of admission two times greater than non-frail individuals (HR=2.19, 95% CI: 1.81-2.64). Using the CHESS scale, adults who were unstable had a greater risk of institutionalization compared to those who were stable (HR=1.52, 95% CI: 1.29-1.79).

CONCLUSIONS: The frailty index specific to persons with IDD can predict institutionalization in adults of all ages and provides results comparable to current measures of health vulnerabilities. Future research should assess the feasibility of implementing the frailty index as a routine measure in home care settings.

Keywords: intellectual disabilities, aging, frailty, institutionalization, long-term care, RAI-HC

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4.1 Background

Over the past few decades, the population of persons with intellectual and developmental disabilities (IDD) has been growing larger and older compared to previous generations [1]. The prevalence of IDD among adults is estimated to be almost 1% [2], with some living into their seventies and eighties [3]. Even though the promotion, management and delivery of care are advancing [4–6], persons with IDD continue to face increased risks for adverse health outcomes. Genetic predispositions, unfavourable social circumstances, less frequent use of generic health services, and environments that support inactivity and poor lifestyle choices contribute to the risk [4].

Within the Canadian health care system, there is a growing emphasis is care in the community, and Ontario, Canada's most populous province, has invested significantly in an Aging at Home Strategy, which aims to help seniors "avoid the unnecessary loss of independence and dignity due to premature admission to higher care long-term care homes or hospitals" [7]. Home care is a significant method of achieving this goal, by providing services including visiting health professionals, help from personal support workers, homemaking services and community support services.

As governments' management of services for aging adults has shifted towards community-based options, so has the organization and funding of services aimed specifically at supporting persons with IDD. In Ontario, deinstitutionalization was completed on March 31, 2009, transferring the remaining institutionalized individuals into the community [8,9]. This deinstitutionalization corresponds to Canada's policies for persons with IDD, which are based on principles of community inclusion, choice making, and the improvement of quality of life [10]. However, policy makers, families and communities are concerned about the risk of re-institutionalization of younger adults into long-term care homes intended for seniors [11,12].

It is well-supported that frailty and health instability play large roles in determining admission to LTC for the general population [13–17]. In the population of adults with IDD, there is some evidence that frailty is associated with increased care intensity [18]. Individuals with IDD generally have high rates of frailty [19–21]. Frailty measures for adults with IDD should be sensitive to changes in health, and should reflect factors from multiple domains of health. The objective of this study was to use a frailty index (FI) created for persons with IDD to predict admission to LTC among persons receiving home care services in Ontario, Canada. A measure that identifies individuals at increased risk for LTC could provide the opportunity, given adequate interventions, for frail persons with IDD to maintain independence in the community and improve their quality of life [22].

4.2 Methods

4.2.1 Design

This study is part of a larger program of research focused on Health-Care Access Research and Developmental Disabilities (H-CARDD) (see www.hcardd.ca). A retrospective cohort design was used to follow 3,034 individuals (aged 18 to 99 years) living in Ontario in 2009/10, who had their first home care assessment between April 1st, 2010 and March 31st, 2014, who had complete data for all covariates of interest, and who were not living in a residential care facility. A total of 4,628 person-years were analyzed. Additional details on the H-CARDD cohort and method for identifying persons with IDD are described elsewhere [23–25]. The study protocol was reviewed and approved by the Queen's University Health Sciences Research Ethics Board and the institutional review board at Sunnybrook Health Sciences Centre, Toronto, Canada.

4.2.2 Data Linkage

Three administrative datasets were used: (1) the Home Care Reporting System (HCRS) for RAI-HC data [26]; (2) the Registered Persons Database (RPDB) for age and date of death [27]; and (3) the Continuing Care Reporting System for Long-Term Care (CCRS-LTC) for dates of admission to LTC [28]. These datasets were linked using unique encoded identifiers and analyzed at the Institute for Clinical Evaluative Sciences (ICES).

4.2.3 Dataset Creation/Variables

The Resident Assessment Instrument- Home Care (RAI-HC) [29] was developed by interRAI, an international collaboration that interprets data on health and social outcomes (see www.interrai.org). The RAI-HC assessment instrument is used by trained case managers as part of regular clinical practice in various jurisdictions in Canada and elsewhere [30] to assess the preferences and needs of those receiving or about to receive home care. The RAI-HC includes items relating to demographic characteristics, home environment, functioning, health, medications, informal support, and formal health services. Various studies have reported the reliability for interRAI items; these values consistently meet acceptable levels of agreement [30–33].

4.2.3.1 Frailty

The accumulation of deficits approach [17,34] to measuring frailty has been applied in home care populations [16] and in adults with IDD [19]. This approach is based on the principle that the more deficits (e.g. signs, symptoms, diseases, impairments) a person has, the frailer he/she will be [34]. Using this approach, a FI comprised of forty-two deficits from the RAI-HC was developed specifically for persons with IDD [35] (Chapter 3). FI scores are based on the proportion of deficits accumulated [17]. To facilitate interpretation, the FI was divided into three categories: non-frail (FI≤0.21; roughly 8 or fewer deficits), pre-frail (0.21<FI≤0.30; roughly between 8 and 12 deficits), and frail (>0.30; 12 or more deficits); methods used to develop these categories are described elsewhere [35,36].

4.2.3.2 Health Instability

The CHESS (Changes in Health, End-stage disease, Signs and Symptoms) scale is embedded in the RAI-HC and evaluates health instability and risk for future decline [37]. Scores are calculated by counting symptoms (i.e., vomiting, dehydration, decline in food/fluid intake, weight loss, shortness of breath) to a maximum of 2 points, worsening of decision making, decline in activities of daily living, and the presence of end-stage disease (1 point each); scores range from 0 (stable) to 5 (unstable). CHESS scores were dichotomized using a cut-off score of 2 or more to categorize individuals as unstable [13,38–40]. The CHESS scale has previously been shown to predict institutionalization and mortality among Canadian home care recipients [13,41].

4.2.3.3 Potential Covariates

Several demographic characteristics were included as potential covariates: age, sex, rural status, living situation, and residential care history. Postal status listed at the time of assessment informed rural status, defined as living in a location not included in a Canadian census metropolitan area or census agglomeration, living in an urban centre of fewer than 10,000, or living in a rural area, which is consistent with the Statistics Canada definition [42]. Living situation at time of assessment was grouped as lives alone, with spouse and/or child(ren), with other family, or in a group setting with non-relatives. Residential care history refers to the five years prior to assessment.

Three caregiver status items were also used: (1) caregiver is unable to continue caring ("caregiver inability"), (2) caregiver is unsatisfied with other informal support ("caregiver unsatisfied"), and (3) caregiver expresses feelings of distress, anger or depression ("caregiver distress").

In addition to individual items, two measures embedded in the RAI-HC were included as covariates: the Institutional Risk Clinical Assessment Protocol, and the Cognitive Performance Scale. Clinical Assessment Protocols (CAPs) identify common risks for individuals using home care, such as abuse, functional decline or LTC placement [43]. When CAPs are triggered, interpretations and potential interventions are offered to case managers to better plan home care services [43,44]. The Institutional Risk CAP is triggered for individuals with high risk of institutionalization [39]. The Cognitive Performance Scale (CPS), measures cognition level and characterizes individuals on a scale from 0 (intact cognition) to 3 (moderate or greater impairment) [45]. The CPS is highly correlated with level of IDD [46] and served as a proxy of level of IDD.

Prior to analysis, potential effect modifiers were identified. It was hypothesized that an individual's Institutional Risk CAP score, caregiver status, and/or sex might modify the effect of frailty or health instability on the risk of admission to LTC.

4.2.4 Time Frame and Outcome Measurement

The outcome of interest was the first admission to LTC after an individual's first home care assessment, in the period between April 1st, 2010, and March 31st, 2014. The index point was the date of the individual's first home care assessment and time was measured from this assessment to the date of the event; that is admission to LTC.

Competing risk events (CREs) prevent the possibility of observing the onset of the outcome of interest [47]. In this study, if death occurred before admission to LTC, observation of admission to LTC would be prevented. As such, pre-LTC death was considered as a potential competing risk event.

Individuals were censored if they were alive in the community at the end of the follow-up period (i.e. had neither event). As this analysis used administrative data to identify admissions and deaths across Ontario, it is assumed that the number of individuals lost to follow-up (e.g. left the province, died out of province) is negligible and not associated with the outcome.

4.2.5 Statistical Analysis

4.2.5.1 Time-to-Event Models

Time-to-event analysis measured the relationship between both frailty and health instability with admission to LTC. Models for both frailty and health instability were developed separately. Two approaches to regression modeling were used: one that ignored competing risk events by censoring (cause-specific proportional hazards model, i.e. the Cox model), and one that adjusted for competing risk events (the subdistribution proportional hazards model, i.e. the Fine & Gray model [48]). Efron's method was used to handle ties [49]. These models are further discussed in Appendix G.

To gain a complete understanding of the effect of frailty as well as health instability on the outcome (with the competing risk of death), both modeling approaches were applied [15,50]. This lead to the derivation of four fully adjusted models: frailty and admission to LTC using the Cox model (Model A); health instability using the Cox model (Model B); frailty using the Fine & Gray model (Model C); and health instability using the Fine & Gray model (Model D).

4.2.5.2 Model Assumptions

Both Cox model and Fine & Gray model rely on the key assumption of proportionality [50]; hazards must be proportional (and not overlapping) at all points in time [51]. Individual covariates were tested for non-proportionality by plotting Schoenfeld residuals [52]. A correlation matrix identified no multicollinearity between independent variables. Further details about the regression diagnostics are available in Appendix H.

4.2.5.3 Model Selection

Model selection was performed separately for all four adjusted models. Starting with saturated models, backwards elimination selected significant covariates. The exposure of interest (frailty or health instability), age, and sex were forced into the model; additional covariates were retained if parameter estimates differed from zero at a significance level of $\alpha=0.05$ using the Wald test.

Potential effect modifiers were decided *a priori*. Covariates were considered effect modifiers if the interaction between the covariate and the exposure of interest (frailty or health instability) was significantly associated with admission to LTC, at a level of α=0.05, while adjusting for other main effects in the model.

4.2.5.1 Statistics

Descriptive analyses examined the frequency of independent variables by outcome status. Bivariate models were derived for all independent variables. The multivariate models provided statistics for covariates that were retained during model selection. For all models, the following statistics were generated: cause-specific hazard ratios (CSHRs), 95% confidence intervals, type III p-values from the test of overall differences between levels of categorical variables, and p-values of statistical significance for each level of categorical variables. The level of significance was α =0.05. Analyses were completed with SAS Enterprise Guide v.6.1.

Cumulative incidence of admission to LTC, and the accompanying 95% confidence intervals [53], was calculated for 6 months and 2 years after first home care assessment. Survival in the community was evaluated using Kaplan-Meier (KM) curves (using admission to LTC or death as a combined outcome event). Log-rank tests determined differences between frailty groups and health instability groups. The correlation between the categorized FI and the CHESS is reported.

Supplemental analyses are also included in the appendices. Cumulative incidence functions (CIFs) plots [53], and the accompanying Gray's Test for Equality of CIFs, are included in Appendix I. Logistic regression models to determine predictive accuracy using the area under the receiver operating curve are presented in Appendix J. Also, time-to-event models with pre-LTC death as the outcome of interest are also included in Appendix K.

4.3 Results

4.3.1 Sample Characteristics

Of the 3,034 individuals included, half (50%) were female (Table 4.1). Roughly 18% were living in a rural area. Many individuals lived with other family members (39%), while some lived alone (19 %) or with a spouse and/or child(ren) (12%); almost a third (31%) of individuals lived in a group setting with non-relatives (e.g. a group home). Almost half of the cohort had at least a moderate cognitive impairment (49%), while a quarter had intact, or borderline intact, cognition (25%). Approximately 19% of individuals had a primary caregiver who indicated distress. Almost half of the individuals were pre-frail or frail (49%).

4.3.2 Outcomes

Almost a quarter of the sample was admitted to LTC (n=678; 22%), while 283 (9%) died before admission to LTC, and the remaining 2,073 (68%) were censored. Individuals who were admitted to LTC were more likely to be pre-frail or frail (68%) and unstable (43%) compared to individuals who were censored (40% and 23%, respectively). A similar pattern existed for those who died before admission to LTC: 69% were pre-frail or frail and 45% were unstable. Those who were admitted to LTC were more likely to be older at the time of their home care assessment (mean age: 63.1 ± 14.5 years), as were those who died (mean age: 60.1 ± 16.2 years), compared to those who were censored (mean age: 50.1 ± 16.9 years). The CHESS scale and the FI were moderately correlated (r_s=0.59).

Overall, the cumulative incidence of admission was 13.8% (95% CI: 12.5-15.1) at 6 months and 18.6% (95% CI: 17.2-20.1) by 2 years. Survival in the community was significantly different between frailty groups (χ^2=314.2, p<0.001) (Figure 4.1) and between CHESS groups (χ^2 =148.3, p<0.001) (Figure 4.2).

Table 4.1. Cohort characteristics at first home care assessment by outcome event during 4-year follow-up (n=3,034)

Characteristics	Total Number (% of total)		Outcome Number (% of row total)					
			Censored		Moved to LTC		Died Pre-LTC	
Overall	3034		2073	(68.3%)	678	(22.4%)	283	(9.3%)
Follow-up days								
Mean ± SD	557	± 441	692	± 433	234	± 269	343	± 329
Age								
Mean ± SD	53.9	± 17.3	50.1	± 16.9	63.1	± 14.5	60.1	± 16.2
Sex								
Female	1519	(50.1%)	1014	(66.8%)	377	(24.8%)	128	(8.4%)
Male	1515	(49.9%)	1059	(69.9%)	301	(19.9%)	155	(10.2%)
Rural Status								
Rural	545	(18.0%)	322	(59.1%)	166	(30.4%)	57	(10.5%)
Urban	2489	(82.0%)	1751	(70.3%)	512	(20.6%)	226	(9.1%)
Caregiver Inability								
Yes	423	(13.9%)	245	(57.9%)	153	(36.2%)	25	(5.9%)
No	2611	(86.1%)	1828	(70.0%)	525	(20.1%)	258	(9.9%)
Caregiver Unsatisfied								
Yes	122	(4.0%)	82	(67.2%)	31	(25.4%)	9	(7.4%)
No	2912	(96.0%)	1991	(68.4%)	647	(22.2%)	274	(9.4%)
Caregiver Distress								
Yes	569	(18.8%)	379	(66.6%)	152	(26.7%)	38	(6.7%)
No	2465	(81.2%)	1694	(68.7%)	526	(21.3%)	245	(9.9%)
Living Situation								
Alone	566	(18.7%)	387	(68.4%)	130	(23.0%)	49	(8.7%)
Spouse and/or Child(ren)	351	(11.6%)	214	(61.0%)	80	(22.8%)	57	(16.2%)
Other Family	1170	(38.6%)	875	(74.8%)	219	(18.7%)	76	(6.5%)
Group Setting	947	(31.2%)	597	(63.0%)	249	(26.3%)	101	(10.7%)
Residential Care History								
Yes	257	(8.5%)	157	(61.1%)	75	(29.2%)	25	(9.7%)
No	2777	(91.5%)	1916	(69.0%)	603	(21.7%)	258	(9.3%)
Cognition Level (CPS)								
Intact (0)	326	(10.7%)	261	(80.1%)	34	(10.4%)	31	(9.5%)
Borderline Intact (1)	427	(14.1%)	312	(73.1%)	70	(16.4%)	45	(10.5%)
Mild Impairment (2)	811	(26.7%)	550	(67.8%)	197	(24.3%)	64	(7.9%)
Moderate+ Impairment (3+)	1470	(48.5%)	950	(64.6%)	377	(25.7%)	143	(9.7%)
Institutional Risk CAP								
Triggered	469	(11.6%)	247	(52.7%)	157	(33.5%)	65	(13.9%)
Not Triggered	2565	(88.4%)	1826	(71.2%)	521	(20.3%)	218	(8.5%)
Frailty Index (FI)								
Frail	860	(28.3%)	433	(50.3%)	305	(35.5%)	122	(14.2%)
Pre-frail	625	(20.6%)	399	(63.8%)	153	(24.5%)	73	(11.7%)
Non-frail	1549	(51.1%)	1241	(80.1%)	220	(14.2%)	88	(5.7%)
Health Instability (CHESS)								
Unstable (≥2)	914	(30.1%)	497	(54.4%)	290	(31.7%)	127	(13.9%)
Stable (<2)	2120	(69.9%)	1576	(74.3%)	388	(18.3%)	156	(7.4%)

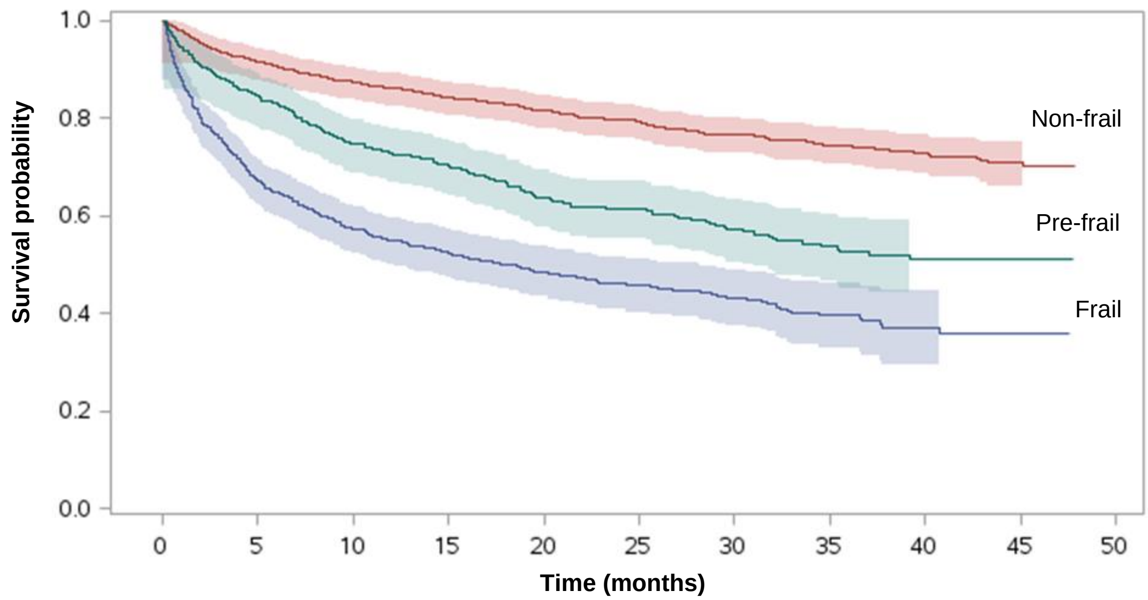

Figure 4.1. KM survival curves comparing survival in the community by FI group

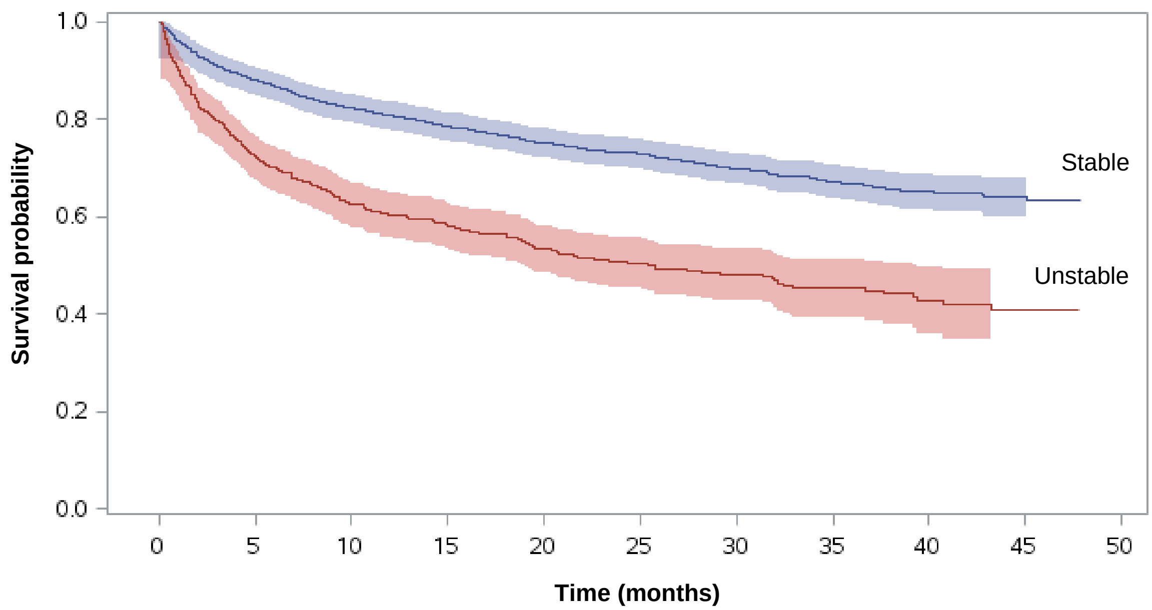

Figure 4.2. KM survival curves comparing survival in the community by CHESS group

Table 4.2 presents the bivariate and multivariate associations between independent variables and admission to LTC. The subdistribution hazard ratios for these associations did not significantly differ from the cause-specific hazard ratios; however they are included in Appendix L for reference. Interactions between frailty group and potential effect modifiers (Institutional Risk CAP, caregiver distress, sex) did not change the hazard ratios in any of the models with admission to LTC as the outcome (data not shown).

4.3.3 Frailty and Admission to LTC

The fully adjusted Cox model of frailty and admission to LTC included: age, sex, rural status, caregiver inability, living situation, and cognition level. Controlling for these independent variables, those who were frail had twice the risk of being admitted to LTC than those who were non-frail (CSHR=2.19, 95% CI: 1.81-2.64, $p<0.001$); the risk was approximately one and a half times higher for those who were pre-frail (CSHR=1.54, 95% CI: 1.25-1.91, $p<0.001$) (Table 4.2). Another strong predictor of admission to LTC was the Cognitive Performance Scale: a moderate or greater impairment was associated with a 2.88 increase in risk of admission (CSHR: 2.88, 95% CI: 2.00-4.14, $p<0.001$) (Table 4.2). Having a caregiver who is unable to continue caring was associated with a two-fold increase in risk of admission (CSHR=2.10, 95% CI: 1.74-2.54, $p<0.001$). An increase in age of 10 years was associated with a 48% increase in risk (CSHR=1.48, 95% CI: 1.40-1.56).

4.3.4 Health Instability

The fully adjusted Cox model of health instability and admission to LTC included: age, sex, rural status, caregiver inability, caregiver distress, living situation, and cognition level. Keeping other predictors constant, the risk of admission was 52% greater for those who were unstable compared to those who were stable (CSHR=1.52, 95% CI: 1.29-1.79, $p<0.001$) (Table 4.2). As in the model with frailty, the strongest predictors were low cognition levels and the presence of a caregiver unable to continue caring activities (Table 4.2).

Table 4.2. Cause-specific hazard ratios (CSHRs) and 95% confidence intervals (CIs) from bivariate and multivariate models of admission to LTC (n=3,034)

Independent Variable	Bivariate Models		Multivariate Model A		Multivariate Model B	
	CSHRs (95% CIs)	**p-value (Type III)**	**CSHRs (95% CIs)**	**p-value (Type III)**	**CSHRs (95% CIs)**	**p-value (Type III)**
Frailty (ref. group=Non-frail)						
Pre-frail	1.92 (1.56-2.36)	<0.001	**1.54 (1.25-1.91)**	<0.001	**-**	-
Frail	3.44 (2.89-4.09)	<0.001	**2.19 (1.81-2.64)**	<0.001	**-**	-
Health Instability (Unstable vs. Stable)	2.08 (1.79-2.42)	<0.001	-	-	**1.52 (1.29-1.79)**	<0.001
Age (per 10 year increase)	**1.48 (1.41-1.55)**	<0.001	**1.48 (1.40-1.56)**	<0.001	**1.53 (1.46-1.61)**	<0.001
Sex (Female vs. Male)	**1.24 (1.06-1.43)**	0.006	1.09 (0.94-1.28)	0.254	1.16 (0.99-1.35)	0.064
Rural Status (Rural vs. Urban)	**1.60 (1.34-1.90)**	<0.001	**1.38 (1.16-1.65)**	<0.001	**1.38 (1.15-1.64)**	<0.001
Caregiver Inability (Yes vs. No)	**2.20 (1.84-2.64)**	<0.001	**2.10 (1.74-2.54)**	<0.001	**2.04 (1.68-2.48)**	<0.001
Caregiver Unsatisfied (Yes vs. No)	1.17 (0.81-1.67)	0.407	-	-	**-**	-
Caregiver Distress (Yes vs. No)	**1.28 (1.07-1.54)**	0.007	**-**	-	**1.22 (1.00-1.48)**	0.045
Living Situation (ref. group = Lives Alone)		(<0.001)		(<0.001)		(<0.001)
Lives with Spouse and/or Child(ren)	0.95 (0.72-1.26)	0.740	**0.63 (0.48-0.84)**	0.002	**0.62 (0.46-0.82)**	0.001
Lives with Other Family	0.74 (0.60-0.92)	0.008	1.17 (0.92-1.49)	0.192	1.09 (0.85-1.38)	0.504
Lives in Group Home	**1.15 (0.94-1.43)**	0.173	1.20 (0.96-1.50)	0.115	1.25 (0.99-1.57)	0.056
Residential Care History (Yes vs. No)	**1.44 (1.13-1.83)**	0.003	-	-	**-**	-
Cognition Level (CPS) (ref. group= Intact)		(<0.001)		(<0.001)		(<0.001)
Borderline Intact (1)	**1.65 (1.10-2.49)**	0.017	**1.72 (1.14-2.60)**	0.010	**1.68 (1.12-2.54)**	0.013
Mild Impairment (2)	**2.80 (1.95-4.03)**	<0.001	**2.22 (1.54-3.20)**	<0.001	**2.19 (1.52-3.17)**	<0.001
Moderate+ Impairment (3+)	**2.82 (1.99-4.01)**	<0.001	**2.88 (2.00-4.14)**	<0.001	**2.91 (2.02-4.20)**	<0.001
Institutional Risk CAP (Triggered vs. Not)	**1.93 (1.62-2.31)**	<0.001	**-**	-	**-**	-

Bold indicates significance at α=0.05;
Model A controls for: frailty, age, sex, rural status, caregiver inability, living situation, and cognition level
Model B controls for: health instability, age, sex, rural status, caregiver inability, caregiver distress, living situation, and cognition level

4.4 Discussion

This study evaluated the relationship between frailty and admission to LTC, in a cohort of persons receiving home care in Ontario, using a FI tailored specifically for adults with IDD. The FI was significantly associated with higher rates of admission to LTC, independent of age, sex, rural status, caregiver inability, living situation, and cognition level. Individuals in the frail group had rates of admission over twice as high as those in the non-frail group. Institutionalization is arguably one of the more severe outcomes of frailty [14], and is frequently used as an outcome to validate FI measures. Studies of Canadian seniors have reported increased risk of LTC admissions with elevated frailty, recording adjusted hazard ratios from 1.2 to 3.3 [13,14,16]. In older adults with IDD, a FI was associated with increases in care intensity and health care costs [18]. The current analysis helps to expand our understanding of frailty in this population by identifying significant associations in a cohort of both young and old adults with IDD (aged 18-99 years), suggesting that frailty is a relevant consideration in younger adults.

Caregiver status was a strong predictor of admission to LTC. In the general population, the RAI-HC has identified 2-8% of caregivers unable to continue caring [54]; in this study, 14% report this inability. Adjusting for other factors, individuals with struggling caregivers had twice the risk of admission at any given time. The presence of a system that is responsive to the needs of families may explain higher rates of admission; as caregivers indicate a struggle, the system is able to secure higher levels of care. Alternatively, leaving transition planning until a crisis emerges (e.g. caregiver no longer able to provide adequate care) can lead to emergency, temporary, or inappropriate placements for adults with IDD, who may meet this sudden upheaval with confusion and distress [55]. Future research should explore the relationship between caregiver status and admissions to LTC among persons with IDD, and the best way to support informal caregivers in the community.

One notable deviation from the findings of other studies is the lack of sex differences in the risk of admission to LTC. Only one of the fully adjusted models (modeling CHESS using the Fine & Gray model, see Appendix L) found a small increase in risk for females. In the general population, some studies report that women are at greater risk of institutionalization [56,57], while others report that men have the higher risk [38,58,59]. We expected to see a significant interaction between frailty and sex, considering that women are understood to tolerate deficits better than men [17]. Our descriptive results suggest that men may be more likely to die before admission to LTC. More research is required to understand the patterns of mortality and service utilization in men and women with IDD.

Chronological age is considered an imprecise predictor of adverse outcomes [60], especially in adults with IDD [61]. A ten-year increase in age contributed roughly to the same risk of admission to LTC as being characterized as pre-frail. However, accumulating at least 12 deficits (frail) is a stronger predictor of admission to LTC than age in this sample of adults with IDD. Using measures, such as the FI, can improve care practices that rely heavily on age as an indicator of functioning or need.

After adjusting for other factors, triggering the Institutional Risk CAP did not alter risk of admission to LTC. This may indicate that home care services are appropriately responding to individuals identified as being most at risk for institutionalization by modifying care plans and offering more care [62]. Alternatively, the Institutional Risk CAP currently in use may not be as sensitive a tool to use among adults with IDD.

The large proportion of individuals with IDD residing in institutions (e.g. LTC homes, psychiatric hospitals) [21,63] has raised concerns about re-institutionalization [64]. However, the current study reports an insignificant association between residential care history and admission to LTC. Future research should confirm these findings by better defining "residential care facility" in assessments. The current variable in the RAI-HIC could refer to multiple care settings (e.g. group home, LTC home).

The CHESS scale was a significantly weaker predictor of admission to LTC in comparison to the FI. To our knowledge, only two studies have directly compared the CHESS scale and a FI, with conflicting findings [13,16]. Beyond the current research group, the CHESS has not been evaluated in a population of adults with IDD; from initial analyses, it was hypothesized that the CHESS scale captures a state of health that is more severe than frailty, and thus does not identify as many individuals as at-risk. However, similar proportions of the cohort were classified as frail and unstable (28% and 30%, respectively), suggesting that, at least in adults with IDD, health instability (as determined by CHESS scores ≥2) is not a subset of frailty, but a different characterization of vulnerability.

Individuals with unstable health as measured by the CHESS (a measure embedded in the RAI-HC assessments) had an increase in risk of admission of 83% compared to stable individuals. This increase in risk is comparable to findings in the general population [13,15,65], although a study of assisted living residents in Alberta, Canada, found a slightly larger hazard ratio using a cut-off of ≥3, rather than ≥2 (HR=2.26, 95% CI: 1.49-3.41) [15]. Consistent cut-off scores would ease comparisons between studies. To determine cut-off points, researchers and knowledge users must determine an acceptable balance between sensitivity and specificity of the measure. Using 2+ as a cut-off, this study achieved a sensitivity of 57% and specificity of 74% (Appendix J). Alternatively, using the full 6-level scale might be more appropriate [41].

Strengths of this study include the use of a large, population-based cohort of adults with IDD. Given that individuals with IDD age prematurely [66–68], including young adults in the cohort was a better reflection of aging across the lifespan. While there is no gold standard for frailty, the FI's strong association with adverse outcomes provides some criterion validity [69], and can be recommended for inclusion in future studies.

This analysis derived hazard ratios using two time-to-event models: the Cox proportional hazards model and the Fine & Gray subdistribution model, to consider competing risks. The Fine & Gray model is proposed to offer better interpretations for decision-makers as it considers multiple outcomes [70]. However, risk estimates were not significantly different between the models, and either hazard ratio can be interpreted.

A few of the study's limitations require acknowledgement. First, by using a baseline measure of frailty to predict a first admission to LTC, we ignored the changes in frailty that could have occurred during the study follow-up. These changes could be due to positive events, such as receipt of home care services, or negative events, such as a trauma (e.g. a fall) or change in informal support (e.g. death of a caregiver). These events may be stronger drivers of the move to LTC [58]. Furthermore, some individuals may have become long-stay patients at hospitals prior to the move to LTC, which could also be considered a form of institutionalization. However, one potential use of the FI is as a screening tool, capable of predicting future outcomes before future events are known [71,72]. On a similar note, another limitation of this study is the unspecified status of individuals after their first admission to LTC; it is not known how soon after admission adults die, or if moving into a LTC facility is a temporary solution to a crisis. Further research should seek to better understand the relationship between frailty and health care services beyond first interaction with both home care and LTC.

Another limitation of this study is the use of administrative data to identify adults with IDD. While the cohort was developed using algorithms to balance sensitivity and specificity [24], it is still possible that some adults in the cohort do not have an IDD, evidenced by the high proportion of the cohort who had "intact" cognition (11%).

Future research should assess the predictive ability in other populations of individuals with IDD, and consider the feasibility of implementing the FI in a jurisdiction using the RAI-HC. These results should be compared to other RAI-HC embedded indicators of institutional risk, such as the Method for Assigning Priority Levels (MAPLe) [73]. In addition, further consideration should be given to the complexity of the transition from community to a LTC facility, which frequently entails hospitalizations and time spent as alternate-level of care (ALC) patients. ALC patients remain in hospitals even after acute hospital services are no longer needed, often because of inadequate community support or unavailable beds in LTC facilities [74]. These patients have higher risk of high levels of functional deficits, complexity, and disease burden [74]. Lastly, a large-scale analysis of caregivers would expand on the strong association noted in this study between measures of caregiver distress and admission to LTC.

Using a measure of frailty, rather than age, may improve care practices. Both the FI and the CHESS can be calculated in the RAI-HC assessments, as well as other interRAI assessment tools. Frailty is a dynamic construct [75], which indicates that an individual can both improve and decline over time. Indeed, frailty is perceived to be potentially modifiable [76]. Intervention strategies are varied, and include optimizing nutritional status [77], improving psychological well-being [78], and participating in high-intensity resistance training [79], among others. Early identification could lead to community-based interventions, and a meta-analysis of 89 trials reported that such interventions postpone admissions to LTC in the general population [80].

Given the emphasis on care in the community of adults with IDD, determining risk factors and adequate predictors of admission to LTC homes could be beneficial for appropriate care planning and service allotment.

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Chapter 5

Discussion

5.1 Study Summary

The purpose of this thesis was to develop a measure of frailty for a population of persons with intellectual and developmental disabilities (IDD) accessing home care services in Ontario and to use this measure to determine the risk of admission to long-term care (LTC) with a retrospective cohort design. Using RAI-HC assessments, available from an administrative database held at the Institute for Clinical Evaluative Sciences (ICES), a 42-item frailty index (FI) was developed. Survival analysis, with consideration of competing risks, determined the risk of admission to LTC.

This chapter will reflect on the key findings of this study, in the context of the current body of literature, review strengths and limitations, and discuss future directions and implications of this research.

5.2 Summary of Major Findings

5.2.1 Development of a FI

Chapter 3 described the process of creating a FI for persons with IDD using the accumulation of deficits approach. The description of the FI is consistent with other derivations [1–5]. The FI developed in this study exhibited previously established characteristics, including a right-skewed distribution, and a significant and positive correlation with age. The mean (0.22 ± 0.13) is in the expected range, considering estimates from similar studies [4,6].

As there is no biological standard to determine a cut-point for the FI, stratum-specific likelihood ratios were compared to predict admission to LTC. This outcome was selected because it was dichotomous, unequivocal and relevant [3]. Three categories of frailty were determined to characterize individuals as non-frail, pre-frail, or frail, which reflects terminology used in the literature [7,8].

5.2.2 Frailty and Health Instability on Risk of Admission

Analyses have presented evidence that both measures of frailty and health instability are significantly predictive of admission to LTC. At any given time, frail individuals were twice as likely to experience an admission to LTC compared to non-frail adults, while unstable, compared to stable, individuals had a 50% increase in risk of admission. These relationships are independent of factors including age, sex, rural status, caregiver status, living situation, and cognition level.

Schoufour *et al.* (2015) also reported a significant association between a FI specific to persons with IDD and institutionalization ($\chi^2 = 23.4$, $p<0.001$), however this finding was not adjusted [9]. To our knowledge, no other study has analyzed frailty's contribution to risk of admission to LTC in a sample of adults with IDD.

Comparing the CHESS and the FI, the FI is a stronger predictor of admission to LTC, although the CHESS was also significantly associated with admission to LTC. In the literature, institutionalization is generally viewed as an outcome associated with both frailty, as measured by frailty indices [7,8,10–12], and health instability, as measured by the CHESS [7,11,13,14], although it is not clear if one measure is superior to the other.

5.2.3 Other Predictors of Admission

This thesis identified several predictors of admission to LTC beyond the exposures of interest. Independent of the individual's age, sex, rural status, living situation, cognition level, and frailty status, at any given time, adults with caregivers unable to continue caring were twice as likely to be admitted to LTC.

Caregivers of adults with IDD are often parents who are aging themselves, and who have been in a caring role for decades [15]. It is estimated that at least one third of adults with IDD are living with a parent over the age of 60 [16]. These aging family caregivers are twice as likely to report psychological ill health [17], more likely to be socially and economically disadvantaged [18,19], and more likely to live with a variety of physical ailments [20]. Among caregivers of home care users in general, those who are unable to continue caring are more likely to provide more informal care but receive fewer formal supports [21]. In the general population, individuals are more likely to be admitted to LTC if their caregivers report unmet needs and poor subjective health [22].

Our measure of a caregiver's inability to continue caring was recorded before, or soon after, the beginning of home care services (i.e., on their first home care assessment). Home care should reduce the burden faced by caregivers, although the strong association with admission to LTC suggests that home care services may not be sufficient to reduce this risk. The finality of the statement "unable to continue" may be indicative of a point of no return, where institutionalization is inevitable. After controlling for age, sex, rural status, caregiver inability, living situation, cognition level, and health instability, caregiver distress, defined as feelings of distress, anger, or depression, provided only a minimal increase in risk of admission (HR= 1.22, 95% CI: 1.00-1.48). It is hypothesized that while caregiver distress is a risk factor, its impact on admission to LTC may be mitigated by the receipt of services. Stronger conclusions are beyond the scope of this thesis.

Cognition level was another independent predictor of admission to LTC. All levels of cognitive impairment provided significant increases in risk compared to having no cognitive impairment. At any given time, individuals with at least a moderate level of impairment were 2.9 times as likely to be admitted to LTC. These results are not surprising, considering that adults with profound IDD have lower life expectancy [23], and make up a larger proportion of the institutionalized population than do adults with mild IDD [23]. Level of IDD is also associated with increased prevalence of various health diagnoses (e.g. cancer, gastrointestinal, heart, hematological, neurological, visual and hearing impairments) [24] and functional limitations [25], which also contribute to increased care needs [26], and consequently, increased risk of admission to LTC.

Future research should quantitatively evaluate the best measure of caregiver stress to predict admission to LTC, and consider possible changes in service allocation specifically to families and caregivers with high levels of stress. In addition, research should seek to better understand the association between level of IDD and admission to LTC.

5.3 Strengths and Limitations

This thesis reported strong associations between frailty and admission to LTC, for adults with IDD receiving home care services, which has never been done in a Canadian study. The identification of vulnerable home care users with IDD could potentially lead to proactive care and increased support. To understand the context of the study, some additional strengths and limitations are discussed.

5.3.1 Internal Validity

5.3.1.1 Selection bias

Studies of aging adults with IDD are often limited by sample sizes of only a couple hundred [27,28]. The use of administrative data eliminated the need for individuals or organizations to volunteer. The cohort acquired a larger sample than would have otherwise been possible and the risk of selection bias was reduced.

Using multiple datasets to define our cohort of individuals with IDD reduced the likelihood of incorrectly excluding or duplicating individuals, creating a representative cohort of individuals [29]. This cohort was also heterogeneous: individuals with IDD have various risks, diagnoses, and medical needs. Treating the population as a single group may dilute important variation.

Adults with IDD do however form a distinct group in terms of social policy and disability services provision in Ontario.

5.3.1.2 Information bias

5.3.1.2.1 Exposure assessment

Individuals in the cohort were not likely misclassified as frail (or unstable) when they were actually non-frail (or stable), or vice versa. The items of the FI and CHESS originated from the RAI-HC, which is a comprehensive and standardized instrument that measures the needs and preferences of adults living in their homes, designed by an international and multi-disciplinary team. Assessors (health care professionals) follow the guidelines of a comprehensive manual, allowing the assessment to be consistently administered. This is supported by a high inter-rater reliability scores [30,31]. International studies have reported strong correlations (minimum 0.75) between RAI-HC scales and their gold standard equivalents [32,33]. Internal consistencies (evaluated by the Cronbach Coefficient) of interRAI scales are between 0.6-0.8 [34]. Case managers doing the assessments ask direct questions to both the caregiver and the home care client, which reduces the limitations imposed by relying only on self-reported information from adults with IDD, or on caregivers who may not be fully informed. Case managers also include clinical data in the assessments. Recall error was likely minimal, as most questions in the RAI-HC ask about events or conditions in the past 3-7 days.

The FI consists of 42 health-related deficits. If a few of these deficits were non-differentially misclassified, it would likely not impact the overall score. As the outcome occurred after the exposure data was recorded, it is unlikely that exposures were differentially recorded. In addition, all data, regardless of the outcome, had the same source (the Home Care Reporting System [35]). As the FI contains many deficits, the FI’s representation of frailty may be diluted by items that represent frailty less strongly.

While the RAI-HC and its embedded scales are validated [34], cut-off points are well-used but infrequently discussed as misrepresenting risk. As the FI specific to persons with IDD was developed in this thesis, and categorization was desired, cut-off scores were chosen that appeared to be the most informative for the objectives. Depending on the use, and the construct of frailty, these classifications may not be suitable. For example, in Rockwood et al.'s (2011) study of 15-102 year olds, the highest frailty group had a cut-off of 0.21 (7% of sample) [6], while Schoufour et al.'s (2015) cohort of older adults with IDD identified a cut-off score of 0.5 to indicate the most frail group (6% of sample). Thus, frailty might be a relative term, dependent on the setting or population.

While the FI was categorized into three groups, the CHESS scale was dichotomized into stable (<2) or unstable (≥2) based on a review of the literature [7,36,37]. It is likely that stronger hazard ratios would have been calculated between those with truly high CHESS scores (e.g. CHESS≥4) and those with the lowest score (CHESS=0), however the power of this analysis would not have been strong, as very few (n=63; 2%) individuals had high scores (CHESS≥4). Future analyses should evaluate the ideal cut-off for the CHESS by maximizing predictive ability in a population with IDD.

5.3.1.2.2 Outcome assessment

By using administrative health data, it is unlikely that many outcomes were misclassified. It is expected that few individuals were lost to follow-up, by emigrating from Ontario. Deaths or admissions to LTC outside of Ontario would not have been captured in the analysis. These are expected to have been uncommon. Furthermore, it is unlikely that these individuals had significantly different frailty profiles compared to individuals who remained in Ontario.

5.3.1.2.3 Covariate assessment

The cohort members' age and sex were from a reliable source (the Registered Persons Database), and misclassification is likely minimal. Rural status may have non-differential misclassification, which may bias risk estimates toward to the null. However, du Plessis, Beshiri, Bollman, & Clemenson (2001), on behalf of Statistics Canada, extensively deliberated the most appropriate definitions of "rural" in a Canadian context [38], and their definition was applied in this study. Ten (0.3%) individuals were excluded from analyses in Chapter 4 due to missing postal code information, however this missing data is expected to be non-differential and too small to affect estimates.

The RAI-HC is a tool not exclusively used for the population of adults with IDD, therefore, some variables are less informative than assessments specific to persons with IDD might be. For example, living situation was categorized as living alone, living with a spouse and/or child(ren), living with other family members, or living in a group setting. In the general population of adults receiving home care, usually senior citizens, these groups are fairly informative. Adults with IDD, however, could be living with parents, grandparents, siblings, or in various styles of group home, all of which may contribute different risks of admission to LTC. There is some possibility for non-differential misclassification in individuals who have multiple living situations or complex arrangements.

In addition, this analysis relied on the Cognitive Performance Scale (CPS) as a proxy for level of IDD [39]. Level of IDD is an important predictor of both frailty [4] and institutionalization risk [23], however it is difficult to accurately capture in administrative data. Almost a quarter of the cohort (753/3,034) had intact or borderline cognition levels, which suggests that cohort includes adults without IDD.

IDD diagnoses are not available in administrative data; however certain diagnoses may be confounding variables in the association between frailty and LTC. For example, adults with Down syndrome experience higher risks for Alzheimer's disease and functional decline [40,41] compared to individuals without Down syndrome. In the general population, dementia is a strong predictor for institutionalization [42,43].

Additionally, this study did not evaluate the influence of home care on frailty improvements. Some individuals may have been frail upon entering home care, but by receiving services, ceased being frail. On the contrary, other individuals may accumulate deficits for years (to a maximum follow-up of 4 years) and it is the frailty status in the months prior to admission to LTC that is the most relevant. Alternatively, a crisis, such as the death of a parent, could be sufficient cause for admission to LTC [44], regardless of level of frailty. Both of these situations would bias the estimate toward the null, thus weakening frailty's accuracy as a measure.

Similarly, individuals begin home care, and enter LTC, for a myriad of reasons, many of which cannot be captured in a standardized assessment tool. For example, some enter home care as a gateway to receiving more intensive services, which is a likely scenario for some of the individuals who were admitted to LTC soon after receiving home care. In another scenario, individuals may have ceased home care services, and instead received alternate level of care (ALC) in a hospital while waiting for LTC. The frail elderly are most at risk for unnecessary hospitalizations due to delays in matching services [45]. These individuals are no longer community-based, and not experiencing the levels of independence and autonomy that can be provided by home care. In this analysis, these individuals would be censored and would not be seen as having an event. While this is not a misclassification, as they did not truly enter a LTC facility, such individuals could bias estimates towards the null.

Despite not knowing the effect of the "in between" period, the intention of this analysis was to develop a measure that could be used as screening tool. With this in mind, it was less practical to use time-dependent exposures or covariates, or limit the sample.

5.3.2 External Validity

The findings from the survival analysis are generalizable to all adults with IDD using home care in Ontario. Findings would also be generalizable to adults with IDD in provinces with similar coverage of home care services, similar sharing of responsibilities between the health care sector and the developmental services sector, and similar levels of access to LTC facilities. Further research should be done before conclusions are made about individuals outside of Ontario.

Roughly 9% of adults with IDD in Ontario received at least one home care assessment between 2007 and 2009 [46]. Home care users have higher needs, and consequently, these results are not generalizable to the population of adults in IDD in general. From a practical perspective, it is unlikely that the data required to tabulate FI scores would be available for individuals who are not accessing health care services.

Often, studies of frailty only examine the elderly (e.g. aged 65+). However, a significant body of research supports that frailty is a construct that begins earlier in the lifespan [6]. This idea is particularly prevalent in the IDD literature, which suggests early aging [15,47–50]. Inclusion of adults aged 18-99 years suggests that findings can be generalized to adults with IDD of all ages.

While the results are likely only generalizable to Ontario home care users with IDD, the FI developed in Chapter 3 is likely to be widely applicable. It was developed for adults with IDD, but followed the theoretical model and considerations presented for the general population. The deficit items included in the FI are available in other RAI assessments. For example, 37 identical, or near-identical, items present in the FI are also available in the MDS 2.0, the interRAI assessment for LTC (which is also mandated for use in Ontario). Future work could estimate differences between populations with and without IDD, or between community-based and institutionalized populations.

5.3.3 Additional Strengths and Limitations

The statistical analysis applied in this study is considerably more in depth compared to other studies investigating the relationship between frailty and admission to LTC in adults with IDD. The findings add to the growing body of literature about aging with IDD [19,51,52]. Survival analyses had not previously been used to evaluate the association between frailty and rates of admission to LTC in this population. This type of analysis, compared to other regression models, has the benefit of determining rates of admission rather than final outcomes. In addition, this study took into consideration the effect of the competing risk of death on the relationship between frailty and admission to LTC, which is not frequently reported in the frailty literature, with a few exceptions [7,14].

5.4 Future Research and Practice Implications

Multiple studies have identified early aging in adults IDD as an important area for research [19,53]. Comprehensive, routine screening assessments are necessary in an aging population that is largely relying on community-based and primary care services [54].

Future studies should apply the new FI specific to persons with IDD in other care settings to replicate and validate the findings of this thesis. Research should further investigate the contribution of caregiver burden and distress, as well as the influence of certain living situations, on the risk of admission to LTC.

While frailty indices repeatedly demonstrate their utility for describing aging populations and identifying associations with adverse outcomes, they may have untapped potential as prognostic tools for use by care providers [55]. Although there is an understanding that the “one diagnosis at a time” approach is not always effective method of care provision, it is a challenge to determine how to apply the existing knowledge of frailty into an assessment tool [6]. To be beneficial, the FI should outperform predictions made by care providers [56]. Before implementation, it should be evaluated for its accuracy (i.e. calibration and discrimination) and its generalizability (i.e. reproducibility and transportability) [56]. Ideally, future validation studies should prospectively test the FI and should consider using repeated assessments over time [56].

However, a measure should be judged not only by its accuracy and generalizability, but by its contribution to decision making and ultimately, its improvement of outcomes [56,57]. While there is an abundance of studies and reviews of frailty and other geriatric syndromes [58–60], fewer studies exist that discuss the impact of knowing about frailty on clinical outcomes [55,61,62].

Ultimately, for a measure to have an impact on clinical outcomes, one's level of frailty needs to be modifiable. Evidence has shown that different interventions can be appropriate, depending on the level of frailty, with the understanding that no single intervention will be suitable for all frail individuals [55]. Exercise plans and changes in support structure have positive effects on physically frail adults in the general population [63], while psychological, social and environmental sources of frailty can also be targeted [61]. Health promotion and disease prevention interventions also have positive effects on numerous frailty-related outcomes in home care clients [64]. The effectiveness of interventions and health promotion activities specifically designed for aging adults with IDD is an area of research to be further explored [50]. Interventions could be evaluated using the RAI-HC [65].

The FI developed for home care users with IDD is likely the most feasible to implement on a large-scale. One limitation commonly reported with other measures is the reliance on information requiring labour-intensive data collection of information not routinely collected [56,66,67], with few relying on administrative data [62]. The FI described in this study uses data from routinely administered assessments, with the potential for a tool embedded in software with real-time estimates of risk [57].

The World Health Organization developed principles to guide population screening [68]. Applying these principles to frailty screening, five criteria must be met: prevalence and relevance to public health; an available sensitive and specific test for detection; detectable early stages; presence of effective interventions; and a net benefit [55]. It is likely that frailty meets all of the criteria; however, further research is required to confirm this.

5.5 Conclusion

Janicki (1994), while considering persons with IDD, defined healthy aging as "an individual retaining his or her capacities to function as independently as possible into old age and promoting the belief that persons who age successfully are able to remain out of institutions, maintain their autonomy and competence in all activities of daily living, and continue to engage in productive endeavors of their own choosing" (p. 146) [69]. Home care is an essential approach to facilitate this notion of healthy aging. The findings from this thesis add to the growing literature of frailty and aging with IDD, and provide the first step required to screen home care users for risk of future institutionalization.

5.6 References

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Appendix A
Definition of IDD

Any individual who had at least one hospital stay, emergency room visit, home care assessment, or LTC assessment, or at least two physician claims where a diagnosis of IDD was noted was included in the cohort [1]. A diagnosis of IDD was based on the International Classification of Diseases codes (ICD-9 and ICD-10). Table A.1 summarizes the list of ICD codes used to define IDD, as listed in the Atlas on the Primary Care of Adults with Developmental Disabilities in Ontario [2]. Codes are consistent with the World Health Organization's definition [3], and not based only on a specific etiology or IQ score cut-off.

Table A.1. ICD codes to define intellectual and developmental disabilities

Code	Label
	ICD-9
299-299.9	Pervasive developmental disorders (e.g. autism)
317-319.99	Mental retardation
758.0-758.39	Chromosomal abnormalities for which is IDD is typically present
758.5	Other conditions due to autosomal anomalies
758.8, 758.89	Other conditions due to chromosome abnormalities (do not include 758.81)
758.9	Conditions due to anomaly of unspecified chromosome
759.5	Tuberous sclerosis
759.81	Other and unspecified congenital abnormalities: Prader-Willi syndrome
759.821	Other and unspecified congenital abnormalities: de Lange syndrome
759.827	Other and unspecified congenital abnormalities: Seckel syndrome
759.828	Other and unspecified congenital abnormalities: Smith-Lemli-Opitz syndrome
759.83	Other and unspecified congenital abnormalities: Fragile X syndrome
759.874	Other and unspecified congenital abnormalities: Beckwith-Wiedemann syndrome
759.875	Other and unspecified congenital abnormalities: Zellweger syndrome
759.89	Other and unspecified congenital abnormalities: other syndrome (ex. Menkes disease, Laurence-Moon-Biedl syndrome, Rubinstein-Taybi syndrome)
760.89	Fetal alcohol syndrome
760.77	Fetal hydantoin syndrome
	ICD-10
F700	Mild mental retardation with the statement of no/ minimal, impairment of behaviour
F701	Mild mental retardation, significant impairment of behaviour requiring attention/treatment
F708	Mild mental retardation, other impairments of behaviour
F709	Mild mental retardation without mention of impairments of behaviour
F710	Moderate mental retardation with the statement of no/minimal impairment of behaviour
F711	Moderate mental retardation, significant impairment of behaviour requiring attention/treatment
F718	Moderate mental retardation, other impairments of behaviour
F719	Moderate mental retardation without mention of impairments of behaviour

Code	Label
	ICD-10 cont'd
F720	Severe mental retardation with the statement of no/minimal impairment of behaviour
F721	Severe mental retardation, significant impairment of behaviour requiring attention/treatment
F728	Severe mental retardation, other impairments of behaviour
F729	Severe mental retardation without mention of impairments of behaviour
F730	Profound mental retardation with the statement of no/minimal impairment of behaviour
F731	Profound mental retardation, significant impairment of behaviour requiring attention/treatment
F738	Profound mental retardation, other impairments of behaviour
F739	Profound mental retardation without mention of impairments of behaviour
F780	Other mental retardation with the statement of no/minimal impairment of behaviour
F781	Other mental retardation, significant impairment of behaviour requiring attention/treatment
F788	Other mental retardation, other impairments of behaviour
F789	Other mental retardation without mention of impairments of behaviour
F790	Unspecified mental retardation with the statement of no/minimal impairment of behaviour
F791	Unspecified mental retardation, significant impairment of behaviour requiring attention/treatment
F798	Unspecified mental retardation, other impairments of behaviour
F799	Unspecified mental retardation without mention of impairments of behaviour
F840	Childhood autism
F841	Atypical autism
F843	Other childhood disintegrative disorder
F844	Overactive disorder associated with mental retardation and stereotyped movements
F845	Asperger's syndrome
F848	Other pervasive developmental disorders
F849	Pervasive developmental disorder, unspecified
Q851	Tuberous sclerosis
Q860	Fetal alcohol syndrome
Q861	Fetal hydantoin syndrome
Q871	Aarskog, Prader-Willi, deLange, Seckel…etc.
Q8723	Rubinstein-Taybi syndrome
Q8731	Sotos syndrome
Q878	Other
Q900-Q939	All Down syndrome types, cri du chat…etc. (except extra marker chromosomes)
Q971	Female with more than three X chromosomes
Q992	Fragile X syndrome
Q998	Other specified chromosome abnormalities

References

1. Lin E, Balogh R, Cobigo V, Ouellette-Kuntz H, Wilton AS, Lunsky Y. Using administrative health data to identify individuals with intellectual and developmental disabilities: a comparison of algorithms. J. Intellect. Disabil. Res. 2013;57:462–77.
2. Lunsky Y, Klein-Geltink JE, Yates EA. Atlas on the Primary Care of Adults with Developmental Disabilities in Ontario. Institute for Clinical Evaluative Sciences; 2013.
3. Salvador-Carulla L, Reed GM, Vaez-Azizi LM, Cooper SA, Martinez-Leal R, Bertelli M, et al. Intellectual developmental disorders: towards a new name, definition and framework for "mental retardation/intellectual disability" in ICD-11. World Psychiatry. 2011;10:175–80.

Appendix B

Ethics Approval

QUEEN'S UNIVERSITY HEALTH SCIENCES & AFFILIATED TEACHING HOSPITALS RESEARCH ETHICS BOARD-DELEGATED REVIEW
November 17, 2014

Ms. Katherine McKenzie
Department of Public Health Sciences
Queen's University

Dear Ms. McKenzie
Study Title: EPID-488-14 Aging with Intellectual Developmental Disorders: The Effect of Frailty and Health Instability in Home Care Recipients on Time to Admission into Long-Term Care
File # 6014151
Co-Investigators: Mrs. H. Ouellette-Kuntz

I am writing to acknowledge receipt of your recent ethics submission. We have examined the protocol for your project (as stated above) and consider it to be ethically acceptable. This approval is valid for one year from the date of the Chair's signature below. This approval will be reported to the Research Ethics Board. Please attend carefully to the following listing of ethics requirements you must fulfill over the course of your study:

Reporting of Amendments: If there are any changes to your study (e.g. consent, protocol, study procedures, etc.), you must submit an amendment to the Research Ethics Board for approval. Please use event form: HSREB Multi-Use Amendment/Full Board Renewal Form associated with your post review file # **6014151** in your Researcher Portal (https://eservices.queensu.ca/romeo_researcher/)

Reporting of Serious Adverse Events: Any unexpected serious adverse event occurring locally must be reported within 2 working days or earlier if required by the study sponsor. All other serious adverse events must be reported within 15 days after becoming aware of the information. Serious Adverse Event forms are located with your post-review file **6014151** in your Researcher Portal (https://eservices.queensu.ca/romeo_researcher/)

Reporting of Complaints: Any complaints made by participants or persons acting on behalf of participants must be reported to the Research Ethics Board within 7 days of becoming aware of the complaint. Note: All documents supplied to participants must have the contact information for the Research Ethics Board.

Annual Renewal: Prior to the expiration of your approval (which is one year from the date of the Chair's signature below), you will be reminded to submit your renewal form along with any new changes or amendments you wish to make to your study. If there have been no major changes to your protocol, your approval may be renewed for another year.

Yours sincerely,

Albert F. Clark.

Chair, Health Sciences Research Ethics Board
November 17, 2014

Investigators please note that if your trial is registered by the sponsor, you must take responsibility to ensure that the registration information is accurate and complete

QUEEN'S UNIVERSITY HEALTH SCIENCES & AFFILIATED TEACHING HOSPITALS RESEARCH ETHICS BOARD

The membership of this Research Ethics Board complies with the membership requirements for Research Ethics Boards and operates in compliance with the Tri-Council Policy Statement; Part C Division 5 of the Food and Drug Regulations, OHRP, and U.S DHHS Code of Federal Regulations Title 45, Part 46 and carries out its functions in a manner consistent with Good Clinical Practices.

Federalwide Assurance Number: #FWA00004184, #IRB00001173

Current 2014 membership of the Queen's University Health Sciences & Affiliated Teaching Hospitals Research Ethics Board:

Dr. A.F. Clark, Emeritus Professor, Department of Biomedical and Molecular Sciences, Queen's University (Chair)

Dr. H. Abdollah, Professor, Department of Medicine, Queen's University

Dr. R. Brison, Professor, Department of Emergency Medicine, Queen's University

Dr. M. Evans, Community Member

Ms. J. Hudacin, Community Member

Mr. D. McNaughton, Community Member

Ms. S. Rohland, Privacy Officer, ICES-Queen's Health Services Research Facility, Research Associate, Division of Cancer Care and Epidemiology, Queen's Cancer Research Institute

Dr. M. Sawhney, Assistant Professor, School of Nursing, Queen's University

Dr. A. Singh, Professor, Department of Psychiatry, Queen's University

Dr. J. Walia, Assistant Professor and Clinical Geneticist, Department of Paediatrics, Queen's University and Kingston General Hospital

Ms. K. Weisbaum, LL.B. and Adjunct Instructor, Department of Family Medicine (Bioethics)

Dr. J. Whiteley, Community Member

Appendix C

Frailty Index Deficits

Table C.1. Frailty index deficits, descriptions, and values

	Deficit Variable	Description	FI Value
		Physiological	
1	Hearing impairment	Hears adequately Minimal difficulty or in special situations only Highly impaired	0.0 0.5 1.0
2	Cataract	Not present Present (indicated by doctor)	0.0 1.0
3	ADL decline: Mobility in bed	Independent or no decline Requires increased set up help, supervision or limited assistance Requires increased extensive assistance, maximal support or completely dependent	0.0 0.5 1.0
4	ADL decline: Transfers/ in-home locomotion	Independent or no decline Requires increased set up help, supervision or limited assistance Requires increased extensive assistance, maximal support or completely dependent	0.0 0.5 1.0
5	ADL decline: Locomotion out of home	Independent or no decline Requires increased set up help, supervision or limited assistance Requires increased extensive assistance, maximal support or completely dependent	0.0 0.5 1.0
6	ADL decline: Dressing body	Independent or no decline Requires increased set up help, supervision or limited assistance Requires increased extensive assistance, maximal support or completely dependent	0.0 0.5 1.0
7	ADL decline: Hygiene and bathing	Independent or no decline Requires increased set up help, supervision or limited assistance Requires increased extensive assistance, maximal support or completely dependent	0.0 0.5 1.0
8	ADL decline: Toilet use	Independent or no decline Requires increased set up help, supervision or limited assistance Requires increased extensive assistance, maximal support or completely dependent	0.0 0.5 1.0
9	ADL decline: Eating	Independent or no decline Requires increased set up help, supervision or limited assistance Requires increased extensive assistance, maximal support or completely dependent	0.0 0.5 1.0
10	Stair climbing	Up and down stairs without help (last 3 days) Up and down stairs with help or not at all (last 3 days)	0.0 1.0
11	Stamina	Leaves home every day (in a typical week) Leaves home 2-6 days (in a typical week) Leaves home once or never (in a typical week)	0.0 0.5 1.0
12	Fall frequency	No falls (last 90 days) 1-2 falls (last 90 days) 3-5 falls (last 90 days) 6-8 falls (last 90 days) 9 or more falls (last 90 days)	0.0 0.25 0.50 0.75 1.0
13	Unsteady gait	No unsteady gait Unsteady gait	0.0 1.0

	Deficit Variable	Description	FI Value
		Physiological cont'd	
14	Stroke	Not present	0.0
		Present (indicated by doctor)	1.0
15	Coronary artery disease	Not present	0.0
		Present (indicated by doctor)	1.0
16	Hypertension	Not present	0.0
		Present (indicated by doctor)	1.0
17	Other circulatory disease	Congestive heart failure, irregularly irregular pulse and peripheral vascular disease not present	0.0
		Congestive heart failure, irregularly irregular pulse or peripheral vascular disease present (indicated by doctor)	1.0
18	Arthritis	Not present	0.0
		Present (indicated by doctor)	1.0
19	Osteoporosis	Not present	0.0
		Present (indicated by doctor)	1.0
20	Specific infection	HIV infection, pneumonia, tuberculosis and urinary tract infection not present	0.0
		HIV infection, pneumonia, tuberculosis or urinary tract infection present (indicated by doctor)	1.0
21	Dementia/ Alzheimer's	Not present	0.0
		Present (indicated by doctor)	1.0
22	Diabetes	Not present	0.0
		Present (indicated by doctor)	1.0
23	Respiratory disease	Emphysema, COPD and asthma not present	0.0
		Emphysema, COPD or asthma present (indicated by doctor)	1.0
24	Worsening of continence	No worsening of bladder incontinence (last 90 days)	0.0
		Worsening of bladder incontinence (last 90 days)	1.0
25	Edema	No edema (last 3 days)	0.0
		Edema (last 3 days)	1.0
26	Shortness of breath	No shortness of breath (last 3 days)	0.0
		Shortness of breath (last 3 days)	1.0
27	Pain frequency	No pain	0.0
		Less than daily	0.5
		At least once daily	1.0
28	Pain disruption	Pain does not disrupt usual activities	0.0
		Pain disrupts usual activities	1.0
29	Number of medications	0-3 different medications taken (last 7 days)	0.0
		4-6 different medications taken (last 7 days)	0.5
		7 or more different medications taken (last 7 days)	1.0
30	Use of antidepressant	Did not take antidepressant (last 7 days)	0.0
		Used antidepressant (last 7 days)	1.0
		Cognitive	
31	Short-term memory loss	Memory OK (appears to recall after 5 minutes)	0.0
		Memory problem (does not appear to recall after 5 minutes)	1.0
32	Worsened decision-making	No worsening of decision making compared to 90 days ago	0.0
		Worsening of decision making compared to 90 days ago	1.0
33	Delirium	No indicator of delirium (last 90 days)	0.0
		Agitated or disoriented such that his/her safety is endangered	1.0
34	Communication decline	No worsening in communication compared to 90 days ago	0.0
		Worsening in communication compared to 90 days ago	1.0

	Deficit Variable	Description	FI Value
		Social	
35	Changes in social activities	No decline in participation in social activities (last 90 days)	0.0
		Decline in participation in social activities (last 90 days)	1.0
36	Social isolation	Never or hardly ever alone, or for about one hour, during the day	0.0
		Alone for long periods of time or all the time	1.0
37	Loneliness	Does not feel lonely	0.0
		Feels lonely	1.0
		Psychological	
38	Mood decline	No worsening of mood indicators (last 90 days)	0.0
		Worsening of mood indicators (last 90 days)	1.0
39	Changes in behaviour	No worsening of behavioural symptoms (last 90 days)	0.0
		Worsening of behavioural symptoms (last 90 days	1.0
40	Fear of falling	No fear of falling preventing outdoor activity	0.0
		Fear of falling limits outdoor activity	1.0
		Service Utilization	
41	Hospital admission	No admission to hospital (with overnight stay) (last 90 days)	0.0
		At least 1 admission to hospital (with overnight stay) (last 90 days)	1.0
42	Change in care needs	No change or improvement in overall self-sufficiency (last 90 days)	0.0
		Deterioration in overall self-sufficiency (last 90 days)	1.0

Appendix D

FI Creation: Deficits Considered for Inclusion

Deficits from the RAI-HC assessment considered for inclusion in the frailty index (FI) are presented alongside the results from the variable selection process in the tables below, by domain. Deficit items with no missing data (-) or had less than 30% missing, with a prevalence greater than 5% and less than 80% (*), a correlation with age greater than r_s= +0.05 (*) and a significant association with health status ($p<0.05$) (*) met the criteria. Bolded items indicate inclusion in the final FI. Italicized subheadings indicate categories within domains.

Table D.1. Potential deficits from the physiological domain

Deficit	Missing (%)	Prevalence (%)		Correlation with Age (r_s)		Association with Health Status (χ^2)	
		Sensory Impairments					
Hearing impairment	**-**	**23.1**	*****	**0.233**	*****	**13.7**	*****
Visual impairment	0.01	25.7	*	-0.010		22.5	*
Visual Limitation/Difficulties	-	1.6		0.043		49.0	*
Vision Decline	-	3.5		0.049		69.0	*
Cataract	**-**	**6.7**	*****	**0.175**	*****	**6.1**	*****
Glaucoma	-	2.6		0.104	*	10.0	*
		Activities of Daily Living					
Help Needed with Mobility in Bed	0.28	19.0	*	-0.089		11.2	*
ADL Decline: Mobility in Bed	**-**	**10.0**	*****	**0.101**	*****	**5.5**	
Help Needed with Transferring	0.15	30.4	*	-0.079		19.3	*
Help Needed with Locomotion in Home	1.18	30.2	*	-0.076		25.3	*
ADL Decline: Transfers/ In-home Locomotion	**-**	**19.3**	*****	**0.145**	*****	**14.5**	*****
Help Needed with Locomotion Out of Home	14.56	36.1	*	-0.130		20.2	*
ADL Decline: Locomotion Out of Home	**-**	**15.5**	*****	**0.096**	*****	**29.0**	*****
Help Needed Dressing Upper Body	0.41	55.9	*	-0.167		76.8	*
Help Needed Dressing Lower Body	0.89	58.5	*	-0.158		53.5	*
ADL Decline: Dressing Body	**-**	**28.6**	*****	**0.170**	*****	**9.8**	*****
Help Needed Eating	0.05	47.7	*	-0.176		91.2	*
ADL Decline: Eating	**-**	**22.2**	*****	**0.137**	*****	**5.2**	*****
Help Needed with Toilet Use	0.53	44.1	*	-0.181		61.6	*
ADL Decline: Toilet Use	**-**	**21.6**	*****	**0.127**	*****	**3.0**	
Help Needed with Hygiene	0.47	62.5	*	-0.193		105.2	*
Help Needed with Bathing	1.78	79.1	*	-0.111		30.1	*
ADL Decline: Hygiene and Bathing	**-**	**38.1**	*****	**0.211**	*****	**58.6**	*****
ADL Decline	-	41.2	*	0.230	*	74.5	*

Deficit	Missing (%)	Prevalence (%)	Correlation with Age (r_s)	Association with Health Status (χ^2)
		Mobility, Gait & Stamina		
Primary Modes of Locomotion Indoors	1.18	39.2 *	0.033	102.9 *
Primary Modes of Locomotion Outdoors	14.56	37.4 *	-0.028	95.1 *
Stair Climbing	**-**	**56.2 ***	**0.087 ***	**28.8 ***
Stamina	**-**	**68.7 ***	**0.196 ***	**59.0 ***
Physical Activity	-	30.4 *	0.015	7.1 *
Capable of Functional Independence (Client's View)	-	16.0 *	0.052 *	39.8 *
Capable of Functional Independence (Caregiver's View)	-	10.3 *	-0.025	83.7 *
Good Prospects of Recovery	-	8.1 *	-0.020	4.9 *
Fall Frequency	**-**	**34.3 ***	**0.142 ***	**78.9 ***
Unsteady Gait	**-**	**49.9 ***	**0.108 ***	**79.9 ***
		Incontinence		
Bladder Incontinence	0.17	46.0 *	-0.061	24.3 *
Worsening of Bladder Incontinence	**-**	**12.3 ***	**0.147 ***	**11.7 ***
Use of bladder device (pads or briefs)	-	39.4 *	-0.013	1.9
Use of bladder device (urinary catheter)	-	3.1	0.033	9.9 *
Bowel Incontinence	0.06	29.5 *	-0.152	19.8 *
		Cardiovascular Diagnoses		
Stroke	**-**	**7.8 ***	**0.172 ***	**7.6 ***
Congestive Heart Failure	-	4.4	0.141 *	39.0 *
Coronary Artery Disease	**-**	**8.7 ***	**0.213 ***	**36.9 ***
Hypertension	**-**	**27.1 ***	**0.338 ***	**61.1 ***
Irregularly Irregular Pulse	-	3.7	0.139 *	34.0 *
Peripheral Vascular Disease	-	2.8	0.066 *	14.0 *
Other Circulatory Disease	**-**	**9.6 ***	**0.187 ***	**61.9 ***
		Neurological Diagnoses		
Alzheimer's Disease	-	5.4 *	0.200 *	8.6 *
Dementia (Not Alzheimer's)	-	11.3 *	0.278 *	6.8 *
Dementia/Alzheimer's	**-**	**16.3 ***	**0.351 ***	**17.3 ***
Head Trauma	-	3.4	-0.039	0.5
Hemiplegia/Hemiparesis	-	2.2	-0.025	0.0
Multiple Sclerosis	-	0.6	-0.039	2.0 *
Parkinsonism	-	2.2	0.117 *	13.0 *
		Musculo-skeletal Diagnoses		
Arthritis	**-**	**23.0 ***	**0.267 ***	**128.0 ***
Hip Fracture	-	2.5	0.093 *	2.0 *
Other Fracture	-	6.9 *	0.073 *	0.3
Osteoporosis	**-**	**10.6 ***	**0.185 ***	**6.4 ***
		Infectious Diagnoses		
HIV Infection	-	0.1	-0.016	4.0 *
Pneumonia	-	3.0	0.004	3.0
Tuberculosis	-	0.0	0.001	4.0
Urinary Tract Infection	-	4.6	0.076 *	20.0 *
Specific Infection	**-**	**7.4 ***	**0.060 ***	**23.1 ***

Deficit	Missing (%)	Prevalence (%)		Correlation with Age (r_s)		Association with Health Status (χ^2)	
Other Diagnoses							
Cancer	-	4.6		0.076	*	9.0	*
Diabetes	**-**	**18.6**	*****	**0.118**	*****	**75.5**	*****
Respiratory Disease	**-**	**11.9**	*****	**0.064**	*****	**97.4**	*****
Renal Failure	-	3.6		0.040		21.0	*
Thyroid Disease	-	13.8	*	0.075	*	0.1	
Preventative Health Measures							
Blood Pressure Taken	4.97	78.5	*	-0.001		0.2	
Influenza Vaccine Received	4.97	57.5	*	0.097	*	0.6	
Colon Screening	4.97	9.9	*	0.067	*	27.6	*
Mammography or Breast Examination Received	4.97	11.2	*	0.062	*	13.7	*
Problem Conditions							
Diarrhea	-	4.6		-0.005		7.6	*
Urination Difficulties	-	4.6		0.070	*	97.0	*
Fever	-	0.5		-0.018		17.0	*
Loss of Appetite	-	5.6	*	0.041		138.0	*
Vomiting	-	1.8		-0.067		30.0	*
Chest Pain	-	2.4		0.024		119.0	*
No Bowel Movement in 3 Days	-	1.2		-0.007		24.0	*
Dizziness/Lightheadedness	-	9.8	*	0.068	*	241.2	*
Edema	**-**	**13.7**	*****	**0.096**	*****	**80.5**	*****
Shortness of Breath	**-**	**12.7**	*****	**0.086**	*****	**261.2**	*****
Pain							
Pain Frequency	**-**	**44.8**	*****	**0.116**	*****	**352.2**	*****
Pain Intensity	-	44.7	*	0.103	*	387.0	*
Pain Disruption	**-**	**20.6**	*****	**0.055**	*****	**436.4**	*****
Pain Character	-	44.7	*	0.114	*	286.9	*
Pain Medication	-	16.0	*	0.018		222.1	*
Lifestyle Habits							
Poor Drinking Habits	-	2.1		0.047		28.0	*
Drinking in Morning or Troubles	-	0.9		0.026		12.0	*
Daily Smoking	-	12.9	*	-0.010		97.0	*
Symptoms of Abuse							
Fearful of Family Member or Caregiver	-	0.8		0.013		7.8	*
Unusually Poor Hygiene	-	2.0		0.026		15.0	*
Unexplained Injuries	-	0.1		0.001		1.0	
Neglected or Abused	-	0.7		0.018		0.3	
Nutritional Status							
Unintended Weight Loss	-	7.0	*	0.048		44.0	*
Severe Malnutrition	-	1.3		0.009		14.0	*
Morbid Obesity	-	6.9	*	-0.089		60.2	*
Irregular Meals	-	3.3		0.001		47.0	*
Decreased Consumption	-	2.4		0.024		33.0	*
Insufficient Fluid	-	1.3		0.008		6.0	*
Enteral Tube Feeding	-	3.1		-0.178		9.0	*
Oral Health							
Swallowing Difficulty	-	18.0	*	-0.148		21.5	*
Chewing Problems	-	11.7	*	*-0.025*		1.9	

Deficit	**Missing (%)**	**Prevalence (%)**		**Correlation with Age (r_s)**		**Association with Health Status (χ^2)**	
Oral Health cont'd							
Dry Mouth While Eating	-	1.3		0.011		23.0	*
Brushing Teeth Difficulty	-	9.9	*	-0.101		12.2	*
Skin Conditions							
Skin Problems	-	26.5	*	-0.004		5.0	*
Pressure Ulcer	-	4.6		0.008		1.4	
Stasis Ulcer	-	1.9		0.024		2.0	
Burns	-	0.2		-0.021		0.9	
Open Lesions	-	2.7		-0.012		3.4	
Skin Tears/Cuts	-	2.6		0.025		0.2	
Surgical Wounds	-	4.4		-0.051		2.8	
Skin Problems	-	7.2	*	0.030		0.7	
History of Resolved Pressure Ulcer	-	7.2	*	-0.031		0.1	
Antibiotics for Ulcer/Wound	-	4.5		-0.006		0.9	
Dressings for Ulcer/Wound	-	8.7	*	-0.038		5.2	*
Surgical Wound Care	-	3.3		-0.047		1.5	
Other Ulcer/Wound Care	-	3.5		-0.015		0.2	
Medications							
Number of Medications	**-**	**76.7**	*****	**0.257**	*****	**90.5**	*****
Use of Antipsychotic/Neuroleptic	-	34.9	*	-0.017		1.6	
Use of Anxiolytic	-	25.5	*	-0.020		27.4	*
Use of Antidepressant	**-**	**31.2**	*****	**0.061**	*****	**74.3**	*****
Use of Hypnotic	-	17.8	*	0.018		14.4	*
Medications Have No Medical Oversight	15.38	2.4		-0.036		2.6	
Poor Compliance/Adherence to Meds	3.6	10.3	*	0.050	*	44.2	*

Table D.2. Potential deficits from the cognitive domain

Deficit	Missing (%)	Prevalence (%)		Correlation with Age (r_s)		Association with Health Status (χ^2)	
Cognitive Patterns							
Short-Term Memory Loss	**-**	**62.0**	*****	**0.082**	*****	**38.9**	*****
Long-Term Memory Loss	-	51.1	*	-0.090		149.5	*
Impaired Cognitive Skills for Daily Decision Making	-	81.1		-0.077		249.0	*
Worsening of Decision Making	**-**	**16.7**	*****	**0.231**	*****	**14.7**	*****
Change in Mental Function	-	3.0		0.051	*	11.0	*
Delirium	**-**	**8.7**	*****	**0.070**	*****	**2.8**	
Making Self Understood	0.03	55.7	*	-0.204		205.6	*
Ability to Understand Others	0.01	56.9	*	-0.159		204.1	*
Communication Decline	**-**	**9.1**	*****	**0.137**	*****	**2.1**	
Instrumental Activities of Daily Living							
Help Needed with Meal Preparation	16.22	74.8	*	-0.102		80.5	*
Difficulty with Meal Preparation	-	91.4		-0.055		75.0	*
Help Needed with Ordinary Housework	16.89	76.7	*	-0.079		20.6	*
Difficulty with Ordinary Housework	-	94.0		-0.024		5.6	
Help Needed with Managing Finances	16.75	71.9	*	-0.147		156.5	*
Difficulty with Managing Finances	-	87.9		-0.106		176.0	*
Help Needed with Managing Medications	19.99	64.2	*	-0.144		155.2	*
Difficulty with Managing Medications	-	81.9		-0.086		142.0	*
Help Needed with Phone Use	30.26	34.3	*	-0.186		161.0	*
Difficulty with Phone Use	-	61.1	*	-0.151		271.6	*
Help Needed with Shopping	20.03	72.4	*	-0.087		37.0	*
Difficulty with Shopping	-	91.8		-0.033		36.0	*
Help Needed with Transportation	20.21	58.9	*	-0.133		46.6	*
Difficulty with Transportation	-	76.7	*	-0.056		39.0	*
Help with Medical Equipment							
Help Needed with Oxygen Equipment	0.28	2.1		-0.001		69.0	*
Help Needed with IV Equipment	-	1.4		-0.009		18.0	*
Help Needed with Catheter Equipment	-	3.6		0.017		5.0	
Help Needed with Ostomy Equipment	-	1.5		-0.030		19.0	*

Table D.3. Potential deficits from the social domain

Deficit	Missing (%)	Prevalence (%)		Correlation with Age (r_s)		Association with Health Status (χ^2)	
Withdrawal from Activities of Interest	-	9.6	*	0.039		94.7	*
Reduced Social Interaction	-	12.7	*	0.027		88.3	*
Not at Ease Interacting with Others	-	12.0	*	-0.064		4.4	*
Openly Expresses Conflict/ Anger	-	17.1	*	-0.037		10.6	*
Changes in Social Activities	**15.38**	**22.8**	*	**0.119**	*	**130.9**	*
Social Isolation	**15.38**	**25.5**	*	**0.196**	*	**95.9**	*
Loneliness	**15.38**	**9.4**	*	**0.079**	*	**194.0**	*

Table D.4. Potential deficits from the psychological domain

Deficit	Missing (%)	Prevalence (%)		Correlation with Age (r_s)		Association with Health Status (χ^2)	
Mood							
Feelings of Sadness or Being Depressed	-	18.4	*	0.033		327.2	*
Persistent Anger with Self or Others	-	16.3	*	-0.026		32.2	*
Expressions of Apparently Unrealistic Fears	-	8.2	*	0.009		90.0	*
Repetitive Health Complaints	-	6.4	*	-0.004		287.0	*
Repetitive Anxious Complains	-	18.6	*	-0.020		94.0	*
Sad, Pained, Worried Facial Expressions	-	20.9	*	0.040		295.3	*
Recurrent Crying or Tearfulness	-	11.5	*	0.003		88.0	*
Mood Decline	**-**	**14.2**	*	**0.053**	*	**101.9**	*
Behaviour Symptoms							
Wandering Behaviour	-	5.2	*	0.011		17.0	*
Verbally Abusive Behavioural Symptoms	-	8.6	*	0.002		0.5	
Physically Abusive Behavioural Symptoms	-	4.5		-0.097		21.0	*
Socially Inappropriate/Disruptive Behavioural Symptoms	-	10.4	*	*-0.126*		32.0	*
Resists Care	-	12.8	*	-0.021		8.1	*
Changes in Behaviour	**-**	**10.1**	*	**0.064**	*	**0.1**	
Other Psychological Symptoms							
Fear of Falling	**-**	**29.3**	*	**0.094**	*	**118.0**	*
Delusions	-	3.8		0.052	*	4.1	*
Hallucinations	-	4.4		0.023		9.3	*
Psychological Disorder		32.6	*	0.012		104.4	*

Table D.5. Potential deficits from the environment/service use domain

Deficit	Missing (%)	Prevalence (%)		Correlation with Age (r_s)		Association with Health Status (χ^2)	
Home Environment							
Hazardous/Uninhabitable Lighting	4.97	0.5		-0.011		17.0	*
Hazardous/Uninhabitable Flooring/Carpet	4.97	1.9		0.009		13.4	*
Hazardous/Uninhabitable Bathroom	-	2.7		-0.008		8.0	*
Hazardous/Uninhabitable Kitchen	-	0.7		0.006		0.0	
Hazardous/Uninhabitable Heating/Cooling	-	0.6		0.017		1.8	
Home Environment Threatens Personal Safety	-	1.8		0.025		10.0	*
Difficulty Accessing Home	-	6.3	*	0.039		28.5	*
Difficulty Accessing Rooms in House	4.97	5.5	*	-0.044		3.7	
Client lives with others	-	9.6	*	0.000		5.0	*
Service Utilization							
Hospital Admissions	**-**	**36.0**	*****	**0.132**	*****	**56.7**	*****
Number of Visits to Emergency	-	21.6	*	0.036		113.2	*
Number of Emergent Care Visits	15.38	5.2	*	-0.005		20.7	*
Unmet Treatment Goals	15.38	9.1	*	0.026		2.1	
Change in Care Needs	**-**	**45.4**	*****	**0.232**	*****	**53.3**	*****
Made Health Trade-Offs	15.38	2.3		-0.023		82.8	*

Appendix E

FI Creation: Item Correlations by Category

The following tables include correlations (with Spearman correlation coefficients) between potential FI items that have already met the criteria presented in Appendix D (a significant, positive correlation with age; a prevalence between 5 and 80%; less than 30% missing data). Items are shown in categories with similar deficits. If a category had only one item meeting the criteria, the category is not presented as no correlations were calculated.

Table E.1. Correlation matrix: Sensory impairment

		1	2
1	**Hearing Impairment**	1.00	0.09
2	**Cataract**		1.00

Table E.2. Correlation matrix: Activities of daily living

		1	2	3	4	5	6	7
1	**ADL Decline: Mobility in Bed**	1.00	0.68	0.29	0.56	0.56	0.61	0.49
2	**ADL Decline: Transfers / In-home Locomotion**		1.00	0.47	0.74	0.69	0.78	0.68
3	**ADL Decline: Locomotion Out of Home**			1.00	0.53	0.47	0.45	0.53
4	**ADL Decline: Dressing Body**				1.00	0.77	0.80	0.83
5	**ADL Decline: Eating**					1.00	0.76	0.73
6	**ADL Decline: Toilet Use**						1.00	0.74
7	**ADL Decline: Hygiene and Bathing**							1.00

Table E.3. Correlation matrix: Mobility, gait, and stamina

		1	2	3	4	5
1	**Stair Climbing**	1.00	0.39	0.08	0.18	0.39
2	**Stamina**		1.00	0.08	0.11	0.24
3	**Capable of Functional Independence (Client's View)**			1.00	0.10	0.07
4	**Fall Frequency**				1.00	0.38
5	**Unsteady Gait**					1.00

Table E.4. Correlation matrix: Cardiovascular diagnoses

		1	2	3	4
1	**Stroke**	1.00	0.14	0.16	0.10
2	**Coronary Artery Disease**		1.00	0.22	0.18
3	**Hypertension**			1.00	0.18
4	**Other Circulatory Disease**				1.00

Table E.5. Correlation matrix: Musclo-skeletal diagnoses

		1	2	3
1	**Arthritis**	1.00	0.04	0.16
2	**Other Fracture**		1.00	0.11
3	**Osteoporosis**			1.00

Table E.6. Correlation matrix: Other diagnoses

		1	2	3
1	**Diabetes**	1.00	0.07	0.04
2	**Respiratory Disease**		1.00	0.04
3	**Thyroid Disease**			1.00

Table E.7. Correlation matrix: Problem conditions

		1	2	3
1	**Dizziness/ Lightheadedness**	1.00	0.10	0.20
2	**Edema**		1.00	0.19
3	**Shortness of Breath**			1.00

Table E.8. Correlation matrix: Preventative health measures

		1	2	3
1	**Influenza Vaccine Received**	1.00	0.15	0.15
2	**Colon Screening**		1.00	0.21
3	**Mammography or Breast Examination Received**			1.00

Table E.9. Correlation matrix: Pain

		1	2	3	4
1	**Pain Frequency**	1.00	0.95	0.61	0.96
2	**Pain Intensity**		1.00	0.61	0.98
3	**Pain Disruption**			1.00	0.57
4	**Pain Character**				1.00

Table E.10. Correlation matrix: Medications

		1	2	3
1	**Number of Medications**	1.00	0.28	0.00
2	**Use of Antidepressants**		1.00	0.03
3	**Poor Compliance/ Adherence to Meds**			1.00

Table E.11. Correlation matrix: Cognitive patterns

		1	2	3	4
1	**Short-Term Memory Loss**	1.00	0.95	0.61	0.96
2	**Worsening of Decision Making**		1.00	0.61	0.98
3	**Delirium**			1.00	0.57
4	**Communication decline**				1.00

Table E.12. Correlation matrix: Social domain

		1	2	3
1	**Changes in Social Activities**	1.00	0.08	0.11
2	**Social Isolation**		1.00	0.28
3	**Loneliness**			1.00

Table E.13. Correlation matrix: Service utilization

		1	2
1	**Hospital Admissions**	1.00	0.35
2	**Change in Care Needs**		1.00

Appendix F

Additional Results: FI Items and 1-Year Admission (Odds Ratios)

Table F.1. Unadjusted odds ratio for 1-year admission to long-term care and the 42 FI items, sorted from strongest to weakest association

FI Item	OR (95% CI)	p-value
FI (Frail vs. Non-Frail)	*4.45 (3.82-5.19)*	*<.0001*
ADL decline: toilet use	3.62 (3.06-4.28)	<.0001
ADL decline: locomotion out of home	3.61 (3.02-4.33)	<.0001
ADL decline: dressing body	3.44 (2.93-4.03)	<.0001
ADL decline: hygiene and bathing	3.41 (2.96-3.92)	<.0001
Change in care needs	3.06 (2.67-3.51)	<.0001
Worsened decision-making	3.02 (2.61-3.50)	<.0001
Communication decline	2.94 (2.46-3.52)	<.0001
ADL decline: mobility in bed	2.82 (2.18-3.65)	<.0001
Worsening of continence	2.75 (2.34-3.24)	<.0001
Dementia/Alzheimer's	2.63 (2.27-3.05)	<.0001
Stamina	2.41 (2.12-2.75)	<.0001
Stair climbing	2.37 (2.05-2.73)	<.0001
Delirium	2.27 (1.88-2.75)	<.0001
Hospital admission	2.00 (1.75-2.27)	<.0001
Number of medications	1.94 (1.70-2.22)	<.0001
Changes in behaviour	1.93 (1.60-2.32)	<.0001
Stroke	1.92 (1.57-2.35)	<.0001
Short-term memory loss	1.91 (1.65-2.20)	<.0001
Unsteady gait	1.83 (1.60-2.09)	<.0001
ADL decline: eating	1.82 (1.35-2.44)	<.0001
ADL decline: transfers/ in-home locomotion	1.76 (1.36-2.27)	<.0001
Specific infection	1.69 (1.36-2.10)	<.0001
Coronary artery disease	1.61 (1.32-1.97)	<.0001
Hypertension	1.56 (1.36-1.79)	<.0001
Mood decline	1.56 (1.32-1.85)	<.0001
Fall frequency	1.56 (1.30-1.87)	<.0001
Change in social activities	1.47 (1.24-1.75)	<.0001
Cataract	1.45 (1.15-1.83)	0.0019
Other circulatory disease	1.44 (1.18-1.76)	0.0003
Osteoporosis	1.42 (1.18-1.72)	0.0003

FI Item	OR (95% CI)	p-value
Fear of falling	1.39 (1.22-1.60)	<.0001
Loneliness	1.32 (1.04-1.68)	0.0227
Edema	1.27 (1.07-1.52)	0.0077
Arthritis	1.15 (0.99-1.34)	0.0612
Diabetes	1.12 (0.95-1.31)	0.187
Pain frequency	1.11 (0.97-1.28)	0.1237
Use of antidepressant	1.09 (0.95-1.25)	0.2138
Shortness of breath	1.06 (0.87-1.28)	0.5671
Hearing impairment	1.05 (0.67-1.64)	0.8448
Social isolation	1.02 (0.86-1.22)	0.8251
Pain disruption	1.01 (0.86-1.18)	0.9417
Respiratory disease	0.91 (0.75-1.12)	0.3841

Appendix G
Rationale for Modeling Strategy

The objective of this study was to determine the association between two measures of vulnerability (frailty and health instability) and admission to long-term care (LTC). Given the availability of event dates, and the interest in understanding differences in community "survival" time, a time-to-event analysis was chosen over logistic regression. This modeling strategy also coincides with the theory that home care can prolong time spent living independently in the community, but may not necessarily prevent admission to LTC entirely [1].

For this study, two types of models were used simultaneously in an attempt to characterize the desired relationship: one that ignores competing risk events by censoring, and one that adjusts for competing risk events. Both approaches are useful, but answer slightly different questions and thus provide different answers [2, 3].

Cox Proportional Hazards Model

In many survival analyses, the proportional cause-specific hazard model (i.e. the Cox model) is the model of choice. It provides an estimate of the exposure's effect on survival after adjusting for other covariates [4]. Cox models, adjusted for covariates, relate to the outcome of interest only, without considering how covariates act on competing risks [2, 5].

Fine & Gray Proportional Subdistribution Hazards Model

Alternatively the Fine & Gray approach models the hazards of the cumulative incidence function, which is the probability subdistribution function of failure from a given event [6].

This method provides the probability of occurrence of a given event by time *t* under the influence of competing risks [2, 5]. The Fine & Gray model is a modified Cox regression, where competing risk observations are kept in the risk set with diminishing weights [7]. For this document the term "Cox model" will refer exclusively to cause-specific hazard models.

Fine & Gray models measure the effect of the covariates on the specific event cumulative probability. This covariate effect may originate from the direct effect of making the outcome of interest more or less likely to happen, or the indirect effect of making the competing risk events more or less likely to occur [2]. Fine & Gray models are useful to evaluate the ultimate effect of frailty on admission to LTC, and can be more informative about the true utility of the measure [2].

When competing risk events are ignored, by censoring observations with death, the estimation of the probability of the event overestimates the true probability [6], and violates the assumption that death is independent of the time to admission to LTC.

Modeling Strategy

It is recommended that in the presence of competing risk events, both Cox proportional hazards models and Fine & Gray subdistribution proportional hazards models be presented simultaneously for a complete understanding of event dynamics [3]. This principle was followed in the current study, leading to four final models, included in Chapter 4:

Model A: Frailty + covariates on admission to LTC (Cox model)

Model B: Health instability + covariates on admission to LTC (Cox model)

Model C: Frailty + covariates on admission to LTC (Fine & Gray model)

Model D: Health instability + covariates on admission to LTC (Fine & Gray model)

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Appendix H
Regression Diagnostics

Methods

Proportional Hazards Assumption

The fundamental assumption of the Cox model is the proportionality of the hazards, which implies that the predictors of the model have a constant impact on the hazard, or risk, over time. In other words, the hazard ratio (either the cause-specific HR or the subdistribution HR) does not depend on time [1]. Schoenfeld residuals compare the observed and expected number of predicted values [2], and are frequently used to observe the proportionality assumption [2]. Sets of residuals were derived for all covariates in the final models. Plots of the residuals versus time were graphed to confirm the presence of zero slopes with no discernable pattern [2].

Also, interaction terms between time and independent variables with parameter estimates significantly different than zero also indicated violation of the proportionality assumption in the multivariate models. Different time functions were considered for inclusion in these interaction terms. The best function of time was determined by comparing the AIC of models with interaction terms of time (t) as s linear function (t), logarithmic functions ($\log_2(t+1)$), or as a piecewise function [2,3]. Global tests of proportionality for Models A-D were performed.

Multicollinearity

As age and frailty are predicted to be highly correlated, it was necessary to check for multicollinearity in the models, which could lead to unstable coefficient estimates and large estimated variances of the regression coefficients [6]. A correlation matrix of the independent variables, using spearman's rank correlation coefficients, was developed.

Non-informative censoring

The assumption of non-informative censoring states that individuals who leave (i.e. are censored from) the study should do so for reasons not unrelated to the study question. As it was hypothesized that censoring would be informative, analyses considered competing risks [4].

Informative censoring was checked by comparing the mean follow-up times of the non-frail and frail groups, and the stable and unstable groups, using a t-test.

Outlying Observations

Deviance residuals plotted against the linear component of the fitted Cox PH models identified potential outliers and observations poorly predicted by the model [2,5]. Deviance residuals are standardized residuals about the mean. Positive residuals indicate that an event occurred sooner than expected, while negative residuals indicate that the event occurred later than expected [2]. Roughly 5% of the sample is acceptable outside of ± 2 deviance scores [2].

Results

Proportional Hazards Assumption

Schoenfeld residual plots detected some independent variables with non-proportional hazards, including: age, frailty, health instability, cognition level, living situation and the Institutional Risk CAP (plots not shown). A global test of proportionality confirmed that hazards are non-proportional in all four models ($p<0.001$ for all models).

Models included significant time-covariate interactions, using the time function $log_2(t+1)$, for age and the Institutional Risk CAP, and using the time function t, for frailty, health instability, cognition level, and living situation (Table H.1).

Multicollinearity

Independent variables were not strongly correlated (Table H.2) and multicollinearity was not a concern in the models in this thesis.

Table H.1. Parameter estimates and standard errors of significant time-covariate interactions included in models A-D

Significant Time-Covariate Interactions	Model A		Model B		Model C		Model D	
	Parameter Estimate	Standard Error	Parameter Estimate	Standard Error	Parameter Estimate	Parameter Estimate	Parameter Estimate	Standard Error
Frail**time***	- 0.00154	0.00036	-	-	- 0.00182	0.00038	-	-
Unstable**time***	-	-	- 0.00123	0.00034	-	-	- 0.00124	0.00033
Age**log_2(time+1)***	0.00496	0.00118	0.00361	0.00123	-	-	0.00353	0.00114
CPS-Mild Impairment**time***	0.00086	0.00031	0.00069	0.00032	0.00078	0.00034	-	-
Lives with Spouse and/or Child(ren)**time***	-	-	0.00096	0.00045	0.00099	0.00044	0.00092	0.00047
Institutional Risk CAP**log_2(time+1)***	0.17999	0.05114	- 1.14176	0.35811	0.19532	0.04797	-	-

Table H.2. Correlation matrix of independent variables (Spearman correlation coefficients)

	Age	Sex	Rural	CAP	CPS	Liv. Sit.	Cg. Inab.	Cg. Unsat.	Cg. Distress	Res. Hist.	FI	CHESS
Age	1	**0.05**	**0.09**	**0.16**	**-0.15**	**-0.10**	-0.02	-0.03	**-0.10**	**0.04**	**0.35**	**0.22**
Sex		1	-0.01	**0.08**	**-0.05**	**-0.07**	-0.03	0.00	-0.01	-0.03	**0.10**	**0.05**
Rural			1	0.03	0.02	-0.01	0.03	0.02	0.02	0.01	**0.04**	**0.05**
CAP				1	**0.18**	**0.14**	0.01	0.02	**0.04**	**0.25**	**0.34**	**0.25**
CPS					1	**0.35**	**0.15**	**0.07**	**0.12**	**0.11**	-0.03	**-0.04**
Living Situation						1	**-0.04**	**-0.05**	**-0.10**	**0.21**	**-0.07**	**-0.11**
Caregiver Inability							1	**0.17**	**0.21**	-0.03	**0.07**	**0.10**
Caregiver Unsatisfied								1	**0.25**	-0.03	0.01	**0.06**
Caregiver Distress									1	**-0.06**	0.03	**0.09**
Residential History										1	**0.06**	0.02
FI											1	**0.59**
CHESS												1

Bold indicates significance at **α**=0.05;

Note: Liv. Sit. = Living situation; Cg. Inab. = Caregiver inability; Cg. Unsat. = Caregiver unsatisified; Cg. Distress= Caregiver distress; Res. Hist.= Residential history

Non-informative censoring

Informative censoring was apparent, as the mean follow-up duration of frail individuals who were censored (597 ± 409 days) was less than the mean follow-up duration for non-frail individuals (729 ± 440 days, $p<0.0001$). Similarly, adults who were censored and unstable (645 ± 411 days) had shorter follow-up durations compared to stable adults (706 ± 438 days, $p=0.006$).

Outlying Observations

Plotting deviance residuals gave no indication of outlying observations in any of the models (e.g. see). Fewer than 5% of observations were outside ±2 deviance residuals (Figure H.1).

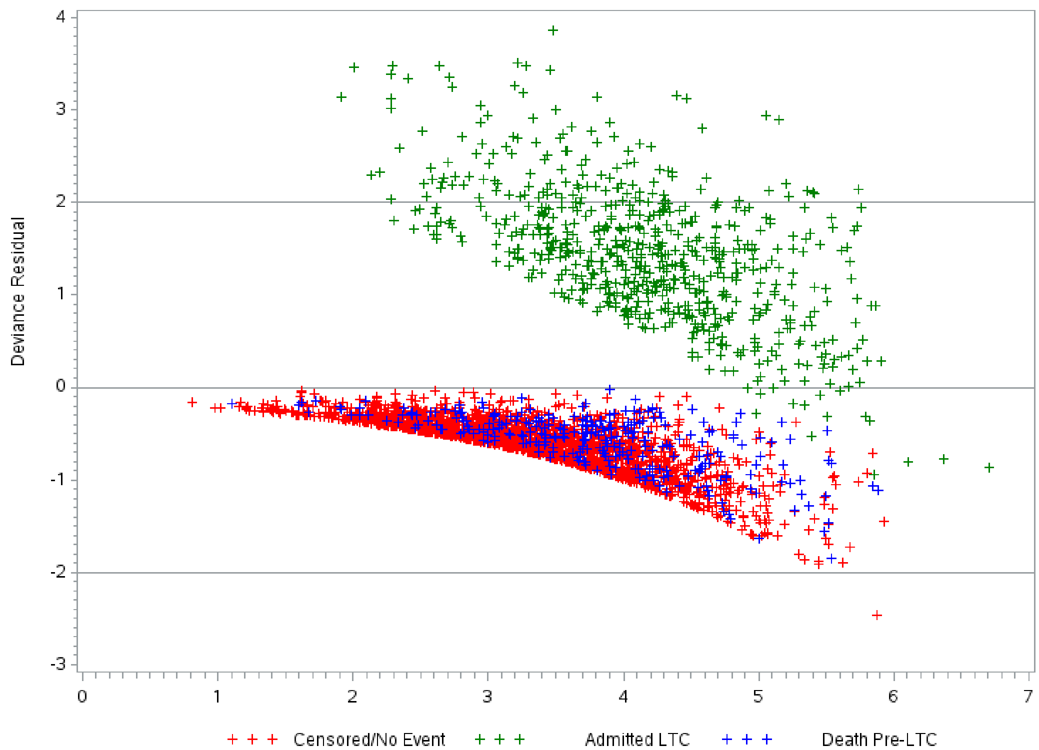

Figure H.1 Testing for outlying observations with deviance residuals

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Appendix I

Additional Analysis: Cumulative Incident Functions

Methods

A non-parametric approach [1] was used to plot the cumulative incidences of admission to LTC over time by frailty and health instability. A SAS macro, %CIF, was applied to estimate these cumulative incidence functions (CIFs), stratified by exposure groups [2]. Differences between frailty as well as health instability groups were tested using Gray's Test for Equality of CIFs [2], provided by the same macro [1].

Results

Gray's test showed statistically significant differences in the cumulative incidences of admission to LTC between frailty ($\chi 2 = 173.0$, $p<0.001$, Figure I.) and health instability ($\chi 2 = 74.2$, $p<0.001$, Figure I.2) groups.

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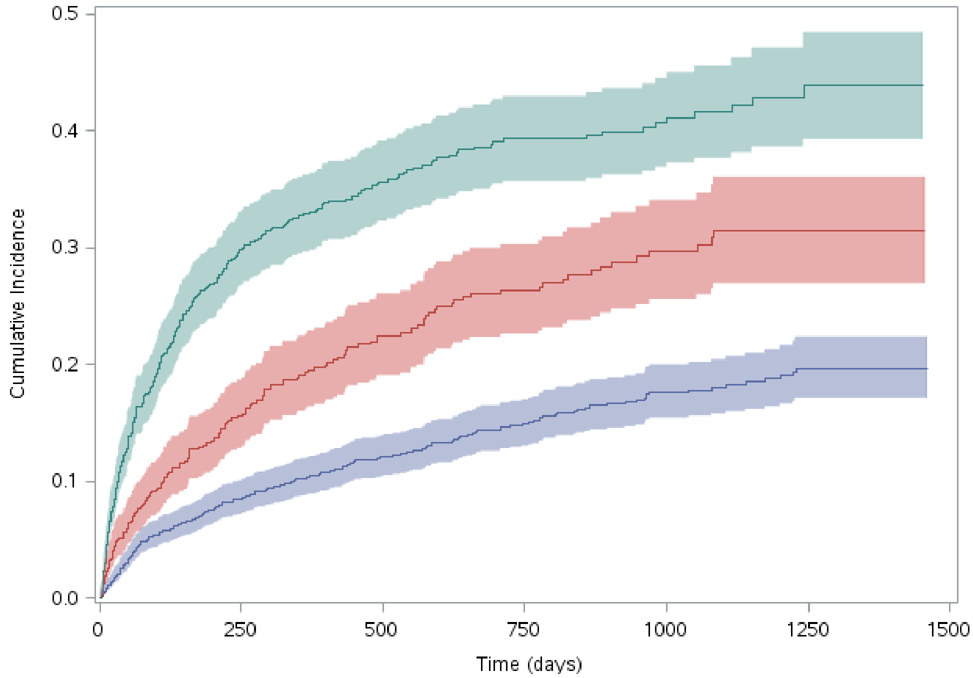

Figure I.1 The cumulative incidence functions (with plotwise confidence limits) for non-frail (bottom), pre-frail (middle) and frail (top) individuals

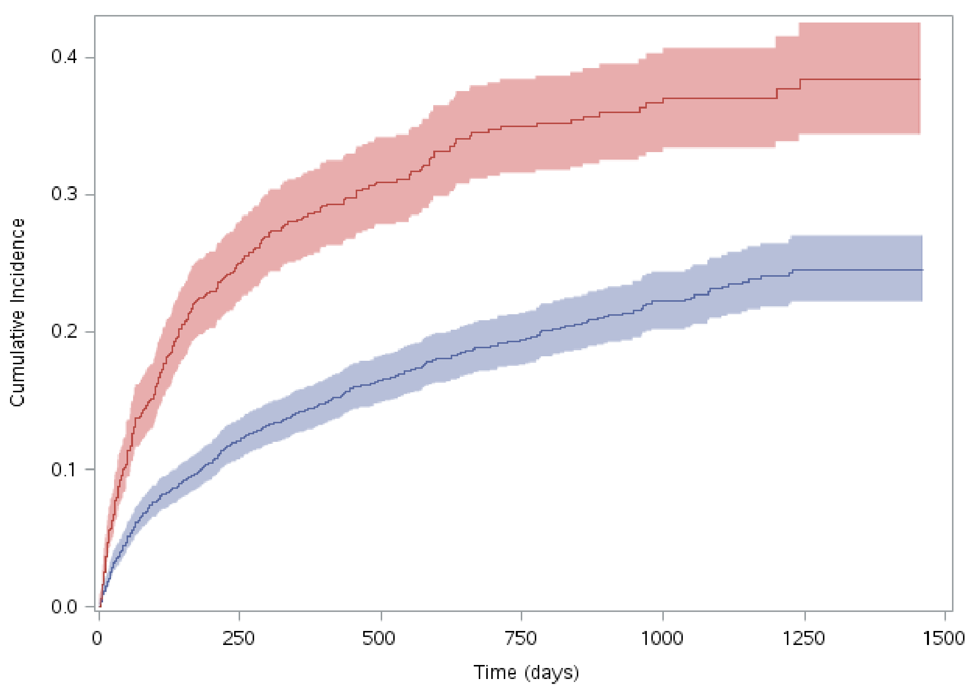

Figure I.2. The cumulative incidence functions (with plotwise confidence limits) for stable (bottom) and unstable (top) individuals

Appendix J
Additional Analysis: Predictive Accuracy

Methods

By categorizing the frailty index (FI), a continuous variable, into three groups (Chapter 3), important information about risk might be lost. To further investigate, the predictive accuracy of the continuous frailty score was compared to the predictive accuracies of four categorized versions of the FI: FI-10 (cut-off points: 0.03, 0.10, 0.21, 0.23, 0.25, 0.27, 0.3, 0.25, 0.45), FI-5 (0.1, 0.21, 0.30, 0.45), FI-3 (0.21, 0.30), and FI-2 (0.30).

To compare the predictive accuracies, logistic regression models were derived using a subset of the cohort (n=2,289) who had a first home care assessment between April 1st, 2010 and March 31st, 2013. The outcome was admission to long-term care (LTC) within one year of follow-up. The areas under the receiver operating characteristics (ROC) curve (AUC) for the models were determined. The AUC of a classifying measure is equal to the probability that the measure will rank a randomly chosen outcome (i.e. admission) higher than a randomly chosen non-outcome (i.e. no admission).

Additional analyses included a second set of comparisons. Three logistic regression models were derived with admission to LTC (within 1 year) as the outcome: age alone, age and the FI (with 3 strata), and age and the CHESS (cut-off point: 2). A ROC curve, plotting the sensitivity and false positive rate of the FI and the CHESS, was created. An additional three models were developed with death before an admission to LTC (within 1-year) as the outcome.

All AUC are presented with 95% confidence intervals. Chi-square tests identified significant differences between estimates of AUC. Sensitivity and specificity of the FI and the CHESS were calculated.

Results

Table J.1 includes the AUC values for models with different variations of the FI. The FI in its continuous form (FI-C) has the largest discriminative ability; however it is not significantly better than the FI in its categorical form with three strata (FI-3).

Figure J.1 compares the FI (with three strata, as in Chapter 4) and the CHESS predictions of 1-year admission to LTC. Table J.2 and Table J.3 compare the AUCs of age-adjusted models, which did not have significantly different predictive abilities, suggesting that neither the FI nor the CHESS scale have better predictive accuracy than age alone when predicting admission to LTC or death one-year following a first home care assessment.

For admission to LTC, the CHESS scale, dichotomized into stable (<2) and unstable (≥2), had a sensitivity of 57% and a specificity of 74%. The frail group (FI>0.30) has a sensitivity of 55% and specificity of 79%, while the pre-frail group had a sensitivity of 77% and a specificity of 81%.

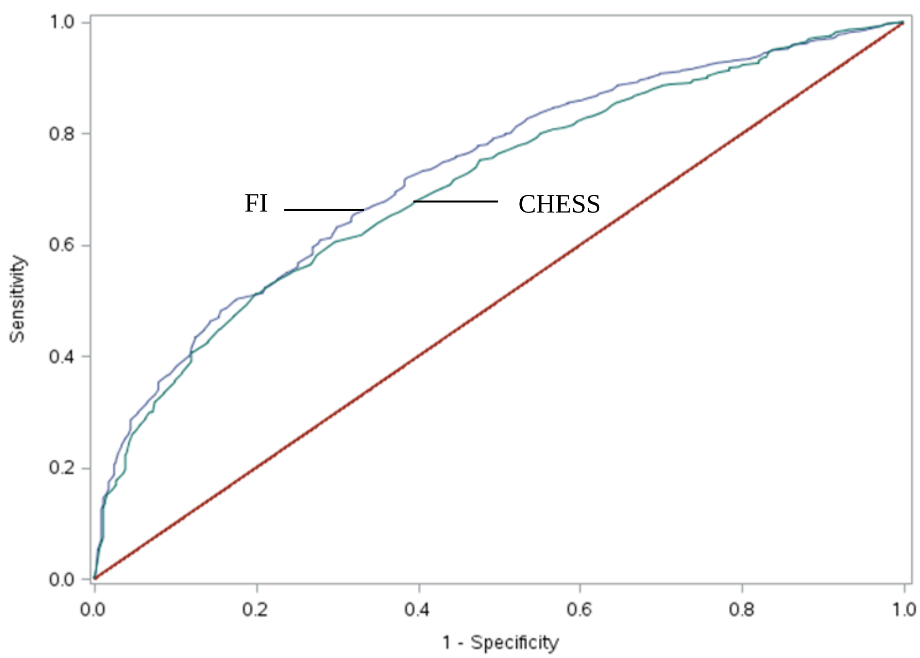

Figure J.1. ROC curve of FI+age and the CHESS+age predicting 1-year admission to LTC

Table J.1. Comparison of areas under the ROC curve (95% CIs) for different frailty and age-adjusted models of 1-year admission to LTC (n=2,289)

Model		Admission to LTC	Contrast p-value			
			1 vs. 2	1 vs. 3	1 vs. 4	1 vs. 5
1	**FI-C**	0.704 (0.678-0.731)	0.467	0.411	0.082	0.001
2	**FI-10**	0.690 (0.663-0.717)				
3	**FI-5**	0.688 (0.662-0.715)				
4	**FI-3**	0.671 (0.644-0.697)				
5	**FI-2**	0.642 (0.619-0.667)				

Table J.2. Area under the ROC curve (95% CIs) for age-adjusted models of 1-year admission to LTC (n=2,289)

Model		Admission to LTC	Contrast p-value		
			1 vs. 2	2 vs. 3	1 vs. 3
1	Age	0.696 (0.671-0.722)	0.067	0.228	0.530
2	Age, FI	0.729 (0.705-0.754)			
3	Age, CHESS	0.708 (0.683-0.733)			

Table J.3. Area under the ROC curve (95% CIs) for age-adjusted models of 1-year death before LTC (n=2,289)

Model		Death Before LTC	Contrast p-value		
			1 vs. 2	2 vs. 3	1 vs. 3
1	Age	0.622 (0.577-0.667)	0.119	0.493	0.380
2	Age, FI	0.671 (0.628-0.714)			
3	Age, CHESS	0.650 (0.606-0.693)			

Appendix K
Additional Analysis: Frailty & Pre-LTC Death

Methods

Frailty is associated with mortality, and the validation of frailty measures often comes from the interpretation of associations with mortality.

In the main analysis (Chapter 4), death before an admission to long-term care (LTC) was considered as a potential competing risk event. To complement these analyses, additional models ascertained hazard ratios for death before LTC. The methods described in Chapter 4 applied to these additional analyses: the cohort included 3,034 individuals with IDD assessed for their first home care admission between April 1st, 2010 and March 31st, 2014. Cox proportional hazard models (bivariate and multivariate).

Results

Hazard ratios are reported in Table K.1. Cox models were adjusted for age, sex, caregiver distress, and the Institutional Risk CAP. The Institutional Risk CAP is an effect modifier: frail individuals who triggered this CAP were not more likely to die before an admission compared to non-frail individuals. However, in individuals who did not trigger the CAP, frailty was a strong predictor of mortality prior to admission to LTC.

Table K.1. Cause-specific hazard ratios (CSHRs) and 95% confidence intervals (CIs) from bivariate and multivariate models for death pre- LTC (n=3,034)

Independent Variable	Bivariate Models			Multivariate Model 1		Multivariate Model 2	
	CSHRs (95% CIs)	p-value (Type III)		CSHRs (95% CIs)	p-value (Type III)	CSHRs (95% CIs)	p-value (Type III)
Frailty (ref. group= Non-frail)		(<0.001)			(<0.001)		
Pre-frail	**2.40 (1.76-3.27)**	<0.001	*CAP*	0.51 (0.22-1.20)	<0.001	-	-
			No CAP	**2.35 (1.67-3.30)**			
Frail	**3.80 (2.89-5.01)**	<0.001	*CAP*	0.96 (0.49-1.87)	<0.001	-	-
			No CAP	**3.37 (2.43-4.68)**			
Health Instability (Unstable vs Stable)	**2.41 (1.90-3.05)**	<0.001	*CAP*	-	-	0.88 (0.54-1.44)	<0.001
			No CAP			**2.51 (1.91-3.30)**	
Age (per 10 year increase)	**1.36 (1.26-1.45)**	<0.001		**1.29 (1.20-1.40)**	<0.001	**1.35 (1.25-1.45)**	<0.001
Sex (Female vs. Male)	0.81 (0.64-1.03)	0.083		**0.65 (0.51-0.83)**		**0.69 (0.54-0.87)**	0.002
Rural Status (Rural vs. Urban)	1.26 (0.94-1.68)	0.123		-	-	-	-
Caregiver Inability (Yes vs. No)	0.78 (0.52-1.17)	0.230		-	-	-	-
Caregiver Unsatisfied (Yes vs. No)	0.82 (0.42-1.59)	0.557		-	-	-	-
Caregiver Distress (Yes vs. No)	**0.70 (0.50-0.98)**	0.039		-	-	-	-
Living Situation (ref. group = Lives Alone)		(<0.001)					
Lives with Spouse and/or Child(ren)	**1.78 (1.22-2.61)**	0.003		-	-	-	-
Lives with Other Family	**0.66 (0.46-0.95)**	0.023		-	-	-	-
Lives in Group Home	1.26 (0.89-1.77)	0.189		-	-	-	-
Residential Care History (Yes vs. No)	1.14 (0.76-1.72)	0.528		-	-	-	-
Cognition Level (CPS) (ref. group= Intact)		0.7093			(0.027)		(0.013)
Borderline Intact (1)	1.18 (0.74-1.86)	0.491		1.30 (0.82-2.05)	0.270	1.34 (0.85-2.12)	0.213
Mild Impairment (2)	1.07 (0.70-1.65)	0.749		0.97 (0.63-1.49)	0.872	0.94 (0.61-1.45)	0.786
Moderate+ Impairment (3+)	1.22 (0.83-1.80)	0.323		1.48 (0.99-2.21)	0.055	**1.51 (1.01-2.26)**	0.044
Institutional Risk CAP (Triggered vs. Not)	**2.03 (1.54-2.68)**	<0.001		-	-	-	

Bold indicates significance at α=0.05; Model 1 controls for: FI group, age, sex, cognition level and Frailty*Institutional Risk CAP; Model 2 controls for: CHESS group, age, sex, cognition level and CHESS*Institutional Risk CAP

Appendix L

Additional Analysis: Subdistribution Hazard Ratios

Table L.1. Subdistribution hazard ratios (SDHRs) and 95% confidence intervals (CIs) from bivariate and multivariate models for admission to LTC (n=3,034)

Independent Variable	Bivariate Models		Multivariate Model C		Multivariate Model D	
	SDHRs (95% CIs)	p-value (Type III)	SDHRs (95% CIs)	p-value (Type III)	SDHRs (95% CIs)	p-value (Type III)
Frailty (ref. group= Non-frail)						
Pre-frail	**1.84 (1.50-2.25)**	<0.001	**1.47 (1.19-1.81)**	<0.001	-	-
Frail	**3.10 (2.61-3.68)**	<0.001	**1.92 (1.58-2.34)**	<0.001	-	-
Health Instability (Unstable vs Stable)	**1.95 (1.67-2.27)**	<0.001	-	-	**1.41 (1.20-1.66)**	<0.001
Age (per 10 year increase)	**1.44 (1.39-1.51)**	<0.001	**1.45 (1.38-1.53)**	<0.001	**1.50 (1.43-1.57)**	<0.001
Sex (Female vs. Male)	**1.25 (1.08-1.46)**	0.004	1.14 (0.98-1.34)	0.061	**1.20 (1.02-1.40)**	0.026
Rural Status (Rural vs. Urban)	**1.56 (1.31-1.86)**	<0.001	**1.34 (1.12-1.60)**	0.001	**1.33 (1.18-1.59)**	0.001
Caregiver Inability (Yes vs. No)	**2.22 (1.85-2.67)**	<0.001	**2.06 (1.69-2.52)**	<0.001	**2.06 (1.68-2.52)**	<0.001
Caregiver Unsatisfied (Yes vs. No)	1.18 (0.82-1.68)	0.407	-	-	-	-
Caregiver Distress (Yes vs. No)	**1.31 (1.09-1.57)**	0.004	**1.23 (1.01-1.50)**	0.038	**1.26 (1.03-1.53)**	0.023
Living Situation (ref. group = Lives Alone)		(<0.001)		(<0.001)		(<0.001)
Lives with Spouse and/or Child(ren)	0.93 (0.71-1.23)	0.616	**0.59 (0.44-0.79)**	<0.001	**0.59 (0.44-0.79)**	<0.001
Lives with Other Family	**0.77 (0.62-0.95)**	0.015	1.13 (0.88-1.45)	0.330	1.08 (0.84-1.38)	0.561
Lives in Group Home	1.14 (0.93-1.41)	0.215	1.17 (0.93-1.48)	0.183	1.20 (0.95-1.52)	0.122
Residential Care History (Yes vs. No)	**1.42 (1.12-1.81)**	0.005	-	-	-	-
Cognition Level (CPS) (ref. group= Intact)		(<0.001)		(<0.001)		(<0.001)
Borderline Intact (1)	**1.63 (1.09-2.46)**	0.019	**1.63 (1.07-2.48)**	0.022	**1.62 (1.07-2.46)**	0.023
Mild Impairment (2)	**2.75 (1.91-3.95)**	<0.001	**2.08 (1.44-3.02)**	<0.001	**2.12 (1.46-3.07)**	<0.001
Moderate+ Impairment (3+)	**2.78 (1.96-3.95)**	<0.001	2.65 (1.82-3.85)	<0.001	**2.74 (1.89-3.98)**	<0.001
Institutional Risk CAP (Triggered vs. Not)	**1.93 (1.62-2.30)**	<0.001	-	-	-	-

Bold indicates significance at **α**=0.05; Model C controls for: FI group, age, sex, rural status, caregiver inability, caregiver distress, living situation, and cognition level; Model D controls for: CHESS group, age, sex, rural status, caregiver inability, caregiver distress, living situation, and cognition level