
Characteristics of Social Frailty among Older Adults with Intellectual Disability

A dissertation submitted to the University of Dublin, Trinity College,
in fulfilment of the requirements for the award of Doctor of Philosophy

Brendan Patrick Dee, BSc, MSc

School of Nursing and Midwifery
Faculty of Health Sciences
University of Dublin
Trinity College

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Declaration

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Summary

Aim

It is increasingly understood that social frailty poses a significant challenge to aging in place for older adults. However, our understanding of this health state is limited to studies involving older adults in the general population. This study aimed to obtain a holistic understanding of the characteristics of social frailty among older adults with intellectual disability including its association with physical frailty and capacity to predict mortality in this population.

Methods

The primary data source was the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing. Social frailty was operationalised according to the accumulation of deficits model to construct a Social Frailty Index. The measurement of physical frailty was based on modified phenotypic criteria including grip strength, vitality, unintended weight loss, timed up and go, and physical activity. Characteristics of a social frailty sub-sample were analysed using descriptive statistics. The association between social frailty and physical frailty was evaluated using Venn diagram. Receiver operating characteristic was used to assess the capacity of social frailty to predict mortality over a three-to-six-year timeframe. Multinomial logistic regression was performed to examine the impact of independent variables on social frailty status and mortality.

Results

Social frailty was associated with advancing age, female gender, severe/profound level of intellectual disability, the presence of Down syndrome, and living in a congregated setting. Physical frailty was more prevalent in comparison to social frailty, while a significant proportion of the study sample had both physical frailty and social frailty. Risk of high levels of social frailty and mortality was associated with advancing age, the presence of Down syndrome, and living in a congregated setting.

Conclusion

Insights obtained may have potentially significant implications for policy and clinical practice. Further research is required to enhance our understanding of social frailty among older adults with intellectual disability.

Contents

Declaration	ii
Summary	iii
Acknowledgements	ix
Presentations and Publications	x
Chapter 1: Introduction	1
1.1 Overview	2
1.2 Structure of Thesis	3
1.3 Background	5
1.3.1 Frailty	5
1.3.2. Social Frailty	5
1.3.3 Relevance of Social Frailty for Older Adults with ID	6
1.4 Research Aim and Objectives	7
1.4.1 Research Question	7
1.4.2 Aim	7
1.4.3 Objectives	7
1.5 Key Definitions	8
1.5.1 Frailty	8
1.5.2 Intellectual Disability	10
1.5.3 Social Determinants of Health	11
1.6 Conclusion	12
Chapter 2: Narrative Review of Social Factors Associated with Frailty Status among Older Adults.....	14
2.1 Introduction	15
2.2 Methods	16
2.3 Findings	23
2.3.1 Age	23
2.3.2 Gender	23
2.3.3 Education	24
2.3.4 Employment and Income	24
2.3.5 Functional and Cognitive Impairment.....	25
2.3.6 Social Connections and Loneliness	26
2.3.7 Lifestyle Behaviours and Religion	28
2.3.8 Social Frailty	28
2.3.9 Common characteristics of social frailty	30

2.4 Conclusion.....	32
Chapter 3: A Systematic Review of Literature Regarding Factors Associated with Frailty Progression among Adults with Intellectual Disability	33
3.1 Introduction.....	34
3.2 Methods.....	34
3.2.1 Search Strategy.....	34
3.2.2 Inclusion and Exclusion Criteria	35
3.2.3 Screening	36
3.2.4 Quality Assessment.....	36
3.2.5 Data Extraction and Analysis.....	36
3.3 Results.....	37
3.3.1 Study Characteristics	38
3.3.2 Outcome Measures	40
3.3.3 Statistical Analysis.....	41
3.4 Outcomes	41
3.4.1 Baseline Frailty Status.....	42
3.4.2 Age.....	42
3.4.3. Gender	42
3.4.4 Down Syndrome.....	43
3.4.5 Group Home Living	43
3.4.6 Functional and Cognitive Impairment.....	43
3.4.7 Use of Nursing and Therapy Services.....	43
3.5 Discussion	44
3.6 Limitations	46
3.7 Conclusion.....	46
Chapter 4: Theoretical Framework	48
4.1 Introduction.....	49
4.2 Frailty Phenotype.....	50
4.3 Accumulation of Deficits	51
4.4 Integral Conceptual Model of Frailty	53
4.5 Conceptual Model of Social Frailty	55
4.6 A Synthesised Model of Frailty	57
4.7 Conclusion.....	58
Chapter 5: Methods	60
5.1. Introduction.....	61
5.2 Study Design	61

5.3 Data Source	62
5.4 Data Collection.....	63
5.4.1 Pre-Interview Questionnaire	63
5.4.2 Computer Assisted Personal Interview	63
5.4.3 Health Fair	64
5.4.4 Impact of COVID-19.....	64
5.5 Study Sample.....	64
5.6 Ethical Considerations	66
5.6.1 Ethical Approval.....	66
5.6.2 Research Principles	67
5.6.2.1 Autonomy.....	67
5.6.2.2 Beneficence and Non-maleficence	68
5.6.2.3 Justice.....	69
5.6.3 Consent	69
5.6.3.1 Disclosure of Information	70
5.6.3.2 Comprehension.....	70
5.6.3.3 Competency.....	70
5.6.3.4 Voluntariness	71
5.6.4 Confidentiality	71
5.7 Measures	73
5.7.1 Social Frailty	73
5.7.2 Physical Frailty.....	78
5.8 Covariates.....	79
5.9 Data Cleaning	80
5.10 Statistical Analysis	80
5.11 Reporting of Findings.....	82
5.12 Conclusion	82
Chapter 6: Development and Validation of a Social Frailty Measure.....	84
6.1. Introduction	85
6.2 Data Suitability	85
6.3 Exploratory Factor Analysis	86
6.4 Confirmatory Factor Analysis	90
6.5 Distribution of the SFI.....	97
6.6 Conclusion	98
Chapter 7: Results.....	100
7.1 Introduction	101

7.2 Prevalence of Social Frailty and Associated Factors	101
7.3 Social Frailty and Physical Frailty	106
7.4 Social Frailty and Mortality.....	107
7.5 Conclusion.....	115
Chapter 8: Discussion and Conclusion	117
8.1. Overview.....	118
8.2 Summary of Research.....	118
8.3 Discussion and Interpretation of Findings.....	119
8.3.1 Construction and Validation of a Social Frailty Measure	119
8.3.2 Prevalence of Social Frailty and Associated Factors.....	124
8.3.3 Relationship Between Social Frailty and Physical Frailty	128
8.3.4 Social Frailty as a Predictor of Mortality	129
8.4 Limitations	131
8.5 Recommendations and Implications	132
8.5.1 Risk Assessment.....	132
8.5.2 Health Promotion.....	133
8.5.3 Policy Development.....	135
8.5.4 Future Research.....	135
8.6 Conclusion.....	137
References	139
Appendix 1: SANRA Guidelines.....	169
Appendix 2: Search Terms	171
Appendix 3: CASP Cohort Study Checklist.....	172
Appendix 4: Data Summary Table	173
Appendix 5: Health Fair Invitation Cover Letter	174
Appendix 6: Health Fair Accessible Information Booklet	176
Appendix 7: Health Fair Appointment Card.....	186
Appendix 8: Modified Frailty Phenotype.....	187

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Presentations and Publications

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2021 World Congress of the International Association for the Scientific Study of Intellectual and Developmental Disabilities, Amsterdam, “Social frailty among older adults with an intellectual disability” Brendan Dee, Roman Romero-Ortuno, Éilish Burke, Philip McCallion, Mary McCarron.

2019 World Congress of the International Association for the Scientific Study of Intellectual and Developmental Disabilities, Glasgow “Early onset of frailty among people with an intellectual disability” Brendan Dee, Éilish Burke, Philip McCallion, Mary McCarron.

Chapter 1: Introduction

1.1 Overview

As in the general population, people with intellectual disability (ID) are now living longer due to medical and social progress (WHO 2001). However, this group have a significantly lower age of death in comparison to the general population (O'Leary *et al.* 2018). In Ireland, people with ID are more likely to die younger and have a higher rate of death in comparison to their non-disabled peers (Doyle *et al.* 2021). Studies have found that people with ID are more likely to become frail and earlier in life (O'Connell *et al.* 2020, Ouellette-Kuntz *et al.* 2017, Ouellette-Kuntz *et al.* 2015, Schoufour *et al.* 2013), increasing their risk of experiencing increased functional dependency (Schoufour *et al.* 2014a), institutionalization (McKenzie *et al.* 2016a, Schoufour *et al.* 2014b) and early mortality (Schoufour *et al.* 2015, McKenzie *et al.* 2015a). However, there is a relative paucity of research on frailty among older adults with ID (McKenzie *et al.* 2016b), with calls for the evidence base to be grown (Ouellette-Kuntz *et al.* 2018).

It has been suggested that new theoretical and practical endeavours are required to address the high prevalence of frailty in ageing populations (Baltes and Smith 2003). In recent years social frailty has been receiving growing attention and traction as a concept. Mounting evidence suggests that social frailty poses a significant challenge to successful ageing. However, older adult populations are highly heterogenous (Mitnitski *et al.* 2017) and therefore the generalisability of these findings to older adults with ID cannot be assumed. The primary aim of this study is to obtain an enhanced understanding of the characteristics of social frailty among older adults with ID. The broad frame of the studies main data source, the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing (IDS-TILDA), has enabled a comprehensive exploration of this health state including its relationship with physical frailty and capacity to predict mortality. These insights may support the design of targeted interventions aiming to delay or prevent social frailty and inform future social and health policy.

1.2 Structure of Thesis

This section provides an overview of the structure of this thesis including a description of the contents of each chapter. In the next section of this chapter, the contextual background of this study is provided. The studies aim and objectives are then discussed, followed by a glossary of key terms defined in the context of this study. In Chapter 2, a narrative review presents evidence on the role of social factors in the development and progression of frailty. This is followed by a systematic literature review on social factors associated with frailty among people with ID in Chapter 3. Based upon a discussion and evaluation of relevant theories, the studies theoretical framework is presented in Chapter 4. This framework provides the basis for the studies hypotheses and choice of research methods, as described in Chapter 5. Chapter 6 reports on the development and validation of the measurement used to operationalise social frailty in the study sample. This is followed by an analysis of the characteristics of social frailty in Chapter 7 in relation to its prevalence and associated factors, relationship with physical frailty, and capacity to predict mortality among older adults with an ID. Finally, Chapter 8 discusses these findings in the context of other studies, with thought given to potential implications for practice and research. Further details in relation to these individual chapters is provided below:

Chapter 1 – Introduction:

This chapter contextualises the study and the research problem being investigated. Key terms framing the focus of this study are defined and an outline of the thesis structure is provided. Finally, study aims, and objectives underpinned by research questions are described.

Chapter 2 – Narrative Review:

This chapter describes the aims, methods and findings of a narrative review used to examine the evidence regarding the association between social factors and frailty status among older adults.

Chapter 3 – Systematic Review:

Following on from the narrative review, this chapter presents a systematic review of literature underpinned by a defined research question in relation to social factors

associated with frailty among older adults with ID. Research gaps and implications of findings for practice and research are discussed.

Chapter 4 – Theoretical Framework:

Selected theories are compared and critically analysed to develop theoretical constructs for this dissertation. Conceptual models which explore physical frailty and social frailty, and hypothesise the relationship between these domains and their contribution to mortality are a central focus.

Chapter 5 – Methods:

This chapter describes the methodological approach of this study. Research aims and objectives are first reiterated, followed by a detailed description of the studies design and sample, ethical considerations, procedures for data collection and analysis, and study limitations.

Chapter 6 – Development and Validation of a Social Frailty Measure:

In this chapter, the development and validation of the measurement used to operationalise social frailty in the study sample are described. Outcomes of the factor analysis undertaken to explore and confirm the latent structures of the instrument are reported. This is followed by an evaluation of the reliability of the measure and distribution of scores in the study sample.

Chapter 7 – Results:

This analytical chapter organises and reports the study's main findings in relation to key objectives. Firstly, findings regarding the prevalence of social frailty and associated factors are presented. The relationship between social frailty and physical frailty is then analysed. Finally, the capacity of social frailty to predict mortality among study participants is examined.

Chapter 8 – Discussion and Conclusion:

This chapter synthesizes and discusses findings in relation to the aim and objectives of this study. Outcomes are examined in the context of research findings from other studies. Emerging patterns and themes are discussed. Finally, reflection on the practical and theoretical implications of this study are considered. Based on study

findings, this chapter presents concluding statements and recommendations for future research, policy, and practice. The contribution of this study to the field of frailty research is discussed.

1.3 Background

1.3.1 Frailty

Frailty has been described as a complex state of increased vulnerability due to cumulative molecular and cellular damage across a range of physiological systems over the lifespan (Walston *et al.* 2018, Clegg and Hassan-Smith 2018, Clegg *et al.* 2013, Cohen 2000). It is considered a means of measuring the combined effects of multiple changes and risks as a person ages (Clegg *et al.* 2013, Rockwood *et al.* 2005, Fried *et al.* 2001). Historically, research on physical frailty has dominated the field (Quach *et al.* 2013). Most of the conceptual definitions of frailty primarily focus on physical problems affecting older people (Fried *et al.* 2001, Buchner and Wagner 1992, Bortz 1993, Campbell and Buchner 1997, Hamerman 1999, Bortz 2002). For instance, Campbell and Buchner (1997) define frailty as:

“A condition or syndrome which results from a multi-system reduction in reserve capacity to the extent that a number of physiological systems are close to, or past, the threshold of symptomatic clinical failure; and as a consequence the frail person is at increased risk of disability and death from minor external stresses.” (p. 315).

1.3.2. Social Frailty

It is increasingly accepted that frailty is a dynamic process that affects and is affected by factors across physical, psychological and social domains (De Vries *et al.* 2011, Gobbens *et al.* 2010a). There is growing evidence that social determinants of health can affect frailty outcomes (Chamberlain *et al.* 2016, Theou *et al.* 2015, Levers *et al.* 2006, Woo 2005, Rockwood 2005). In the past decade, there has been increasing interest in a social conceptualisation of frailty which recognises the outcome of the relationship between the individual and their social environment, rather than the mere presence of physiological impairment (Barrett 2006). This approach recognises the role of social circumstances on health outcomes and access to appropriate interventions and supports (Andrew *et al.* 2018). In comparison to the more well-known concept of physical frailty, social frailty remains widely unexplored. Based on current

research evidence, social frailty appears to be associated with advancing age and has a complex relationship with physical frailty. It has also been found to predict several adverse outcomes including muscle weakness, functional impairment, cognitive impairment, physical frailty, and mortality (Yamada *et al.* 2018, Tsutsumimoto *et al.* 2017, Teo *et al.* 2017, Makizako *et al.* 2015, Makizako *et al.* 2015, Andrew *et al.* 2008).

1.3.3 Relevance of Social Frailty for Older Adults with ID

Social frailty has received increasing attention in recent years due to the relevance of this concept to older adults who are becoming increasingly reliant on their social environment due to policies aimed at reducing formal care supports (Bunt *et al.* 2017). From this perspective, the concept of social frailty may therefore be particularly relevant to older adults with ID. In Ireland, people with ID are being supported to lead “ordinary lives in ordinary places” in line with National policy promoting the de-congregation of care services (HSE 2011). However, current research evidence from IDS-TILDA have revealed that older adults with ID living in Ireland are uniquely exposed to adverse social circumstances which from a social frailty perspective may have an impact on health outcomes.

It has been shown that older adults with ID are more likely to have a low level of education attainment or no formal education at all in comparison to those in general population (McCarron *et al.* 2011, McCausland *et al.* 2016). A majority are unemployed and have lived a life devoid of paid employment (McCausland *et al.* 2020). This population tends to have unmet social needs and impoverished access to healthcare (McGlinchey *et al.* 2019, McCarron *et al.* 2017). In comparison to those in the general population, older adults with ID are significantly less likely to be married or have children of their own (McCarron *et al.* 2014). Many do not live within close proximity to family (McCausland *et al.* 2018) or feel involved in decisions regarding where they live and who they live with (McCarron *et al.* 2011). Furthermore, many older adults with ID in Ireland feel excluded from their community, have difficulties participating in social activities, have fewer friends and experience loneliness (Wormald *et al.* 2019, McGlinchey *et al.* 2019, McCausland *et al.* 2015). A social conceptualisation of frailty may provide an opportunity to explore the combined influence of such social factors, ordinarily studied separately, on health outcomes among older adults with ID. An enhanced understanding of the characteristics of social frailty may support the

development of upstream interventions aimed at preventing or reducing frailty and inform future social and health policy.

1.4 Research Aim and Objectives

This section provides an overview of the studies aim and objectives in line with the main research question.

1.4.1 Research Question

What are the characteristics of social frailty among older adults with ID?

1.4.2 Aim

To obtain a holistic understanding of the characteristics of social frailty among older adults with ID.

1.4.3 Objectives

To address the aim of this study, key objectives include:

- Construct and validate a measurement of social frailty
- Analyse the prevalence of social frailty and associated factors
- Examine the relationship between social frailty and physical frailty
- Assess the ability of social frailty to predict mortality

1.5 Key Definitions

1.5.1 Frailty

The integration of frailty measures in clinical practice is crucial for the development of interventions against age-related conditions in older persons (Subra *et al.* 2012). For the concept of frailty to have practical utility, its theoretical conceptualization must be translatable into an operational definition (Rockwood 2005). There is however a lack of consensus on the specific nature of frailty and no 'gold standard' frailty assessment exists. This applies to both older adults with ID and those in the general population. As a consequence, a plethora of different frailty measures have been developed for clinical and research use (Roppolo *et al.* 2015), with calls for greater homogeneity and simplification of the instruments used (Lozupone 2020). In general, the most common measures adopt either the frailty phenotype (Fried *et al.* 2001) or deficit accumulation model (Mitnitski *et al.* 2001).

The frailty phenotype by Fried *et al.* (2001) describes frailty as a clinical syndrome driven by age-related biologic changes. The underlying assumption of this model is that sarcopenia leads to poor muscle strength, resulting in reduced mobility and physical activity. Lower energy expenditure and nutritional intake causes the person to experience worsening sarcopenia and weight loss (Fried *et al.* 2001). When these changes manifest as clinical signs and symptoms, such as weight loss and decreased energy level, they result in the development of a frail phenotype, which can be concretely measured (Fried *et al.* 2001). Based on the phenotype model, frailty status is determined by the presence of three out of five criteria including shrinking (weight loss, sarcopenia), weakness, exhaustion (poor endurance), slowness, and low activity. Having two of these criteria is indicative of a pre-frailty stage whereby the individual is at a high risk of progressing to frailty. Conversely, the accumulation of deficits model, operationalised as a frailty index (FI) provides a quantitative measurement of frailty. In contrast to phenotypic criteria, the FI emphasises the proportion rather than the nature of deficits present (Rockwood and Mitnitski 2007). The underlying assumption of this model is that the accumulation of unspecified 'deficits' (i.e. health problems or abnormal characteristics) leads to a nonspecific, age-associated vulnerability, or frailty (Mitnitski *et al.* 2001, Rockwood and Mitnitski 2007). The main differences between both approaches are presented in Table 1.1.

Table 1.1 Main differences between phenotype and deficit accumulation

Model	Frailty Phenotype (Fried <i>et al.</i> 2001)	Deficit Accumulation Model (Mitnitski and Rockwood 2001)
Theoretical approach	Frailty is a biologic syndrome characterised by five specific signs or symptoms	Frailty is a state of vulnerability arising from multiple health deficits that accumulate with age
Criteria	Weakness, slowness, low physical activity, exhaustion, unintentional weight loss	Deficits can relate to factors across multiple health domains, including symptoms, morbidities, disabilities and even laboratory abnormalities
Variable type	Categorical (ordinal)	Continuous (with possible ordinal cut-offs)
Application	Meaningful results potentially more relevant to non-disabled older persons	Meaningful results potentially relevant in every individual, independently of functional status

It is also important to note that other frailty measures have been developed for specific use in the ID population such as the Vienna Frailty Questionnaire for Persons with ID (VFQ-ID) (Brehmer and Weber 2010), and its revised version VFQ-ID-R (Brehmer-Rinderer *et al.* 2013). These measures adopt a similar approach to the deficit accumulation model which describes frailty as multisystemic instability involving physiological, psychological, cognitive and social domains. However, in contrast to the deficit accumulation approach, these instruments comprise of specific items (deficits) based on the frailty phenotype (Fried *et al.* 2001) and criteria developed by Rockwood *et al.* (2000). While the inter-rater and retest reliability of the VFQ-ID-R has been reported as good, limitations in relation to the study's sample have been acknowledged. The tools authors acknowledge that further research should be conducted on its use in larger and culturally different populations (Brehmer-Rinderer *et al.* 2013).

1.5.2 Intellectual Disability

Intellectual disability is characterised as significant limitations both in intellectual functioning and in adaptive behaviour as expressed in conceptual, social, and practical adaptive skills which originates before the age of 18 (Schalock *et al.* 2010). This study adopts the definition of ID used by its primary data source, IDS-TILDA. IDS-TILDA recruited its sample from the National Intellectual Disability Database (NIDD) who base their definition of ID on criteria described in the Classification of Mental and Behavioural Disorders (ICD-10) (WHO 1993) (Table 1.2). Levels of intellectual disability described in ICD-10 are based on standardised IQ score of below 70, with sub-classifications of mild ID (IQ score in the range 50-69), Moderate ID (IQ score in the range 35-49), severe ID (IQ score in the range 20-34); or profound ID (IQ score estimated less than 20). Beyond these four main categories of ID, there are two other classifications, “Other Intellectual Disability” and “Unspecified Intellectual Disability”. The former category is used when the assessment is difficult due to other impairments, while the latter is used when there is evidence of intellectual disability but insufficient information to enable categorisation into a sub-classification of ID.

Table 1.2 Levels of Intellectual Disability in ICD-10 (WHO 1993)

Level of ID	Description
Mild	Approximate IQ range of 50 to 69 (in adults, mental age from 9 to under 12 years). Likely to result in some learning difficulties in school. Many adults will be able to work and maintain good social relationships and contribute to society.
Moderate	Approximate IQ range of 35 to 49 (in adults, mental age from 6 to under 9 years). Likely to result in marked developmental delays in childhood but most can learn to develop some degree of independence in self-care and acquire adequate communication and academic skills. Adults will need varying degrees of support to live and work in the community.
Severe	Approximate IQ range of 20 to 34 (in adults, mental age from 3 to under 6 years). Likely to result in continuous need of support.
Profound	IQ under 20 (in adults, mental age below 3 years). Results in severe limitation in self-care, continence, communication and mobility.

1.5.3 Social Determinants of Health

This study pursues a social explanation of frailty which considers the role of the social environment on health outcomes. Social determinants of health have been defined as "the conditions in which people are born, grow, live, work and age" and those conditions, in turn, are "shaped by the distribution of money, power and resources" (Marmot *et al.* 2012). Social factors such as childhood experiences, education, employment, work conditions, economic status, housing and environment have been observed to affect access to effective systems of preventing and treating ill-health (Marmot *et al.* 2008). It has been asserted that health outcomes among ageing populations are subject to a complex inter-play between social and medical factors (Andrew *et al.* 2018). For instance, there appears to be a distinct relationship between social engagement and disability (Mendes de Leon *et al.* 2003), while social support and social connectedness have been found to have a protective effect on mortality (Seeman *et al.* 1993)

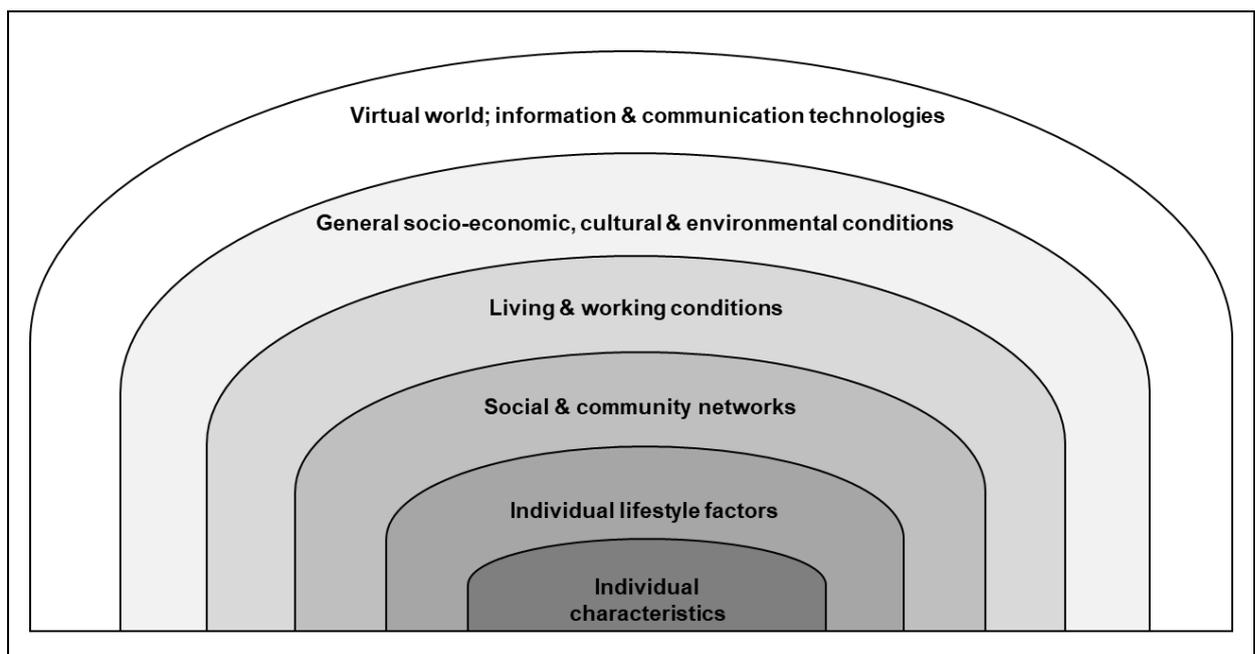


Figure 1.1 Modified determinants of health model, adapted from Rice & Sara (2019)

Numerous models have been used by researchers to explore social determinants of health. Of these, the 'rainbow model' developed by Dahlgren and Whitehead (2007) is the most widely used. This study defines social determinants of health according to a modified version of this model, adapted from Rice and Sara (2019) (Fig. 1.1). This model maps the relationship between the individual, their environment and health. It arranges social determinants of health into different "layers", according to their level of coverage and represents the levels of reality where the phenomena occur. At the centre are individual characteristics which are largely fixed such as age, gender, age, ethnic group, hereditary factors and constitutional factors which may influence a person's health. Surrounding them, are other influences which are theoretically modifiable by policy. The second layer includes individual lifestyle factors, such as smoking habits, alcohol use, and physical activity. The influence of the individual's interaction with their peers and immediate community is represented in the third layer. Next, a person's ability to maintain their health is influenced by living and working conditions, food supply, access to essential goods, education, and welfare services. These are surrounded by mediators of population health including economic, cultural and environmental factors. The final layer, added in the modified version of the model by Rice and Sara (2019), gives recognition to the impact of information and communication technology (ICT) on the determinants of health and its profound implications for the operationalization of all other layers. It should be noted that others have suggested that ICT use and access has particular importance for people with ID in terms of increasing their influence on and participation in society (Werner and Shpigelman 2019, Chiner *et al.* 2017).

1.6 Conclusion

While research on physical frailty has dominated the field, the concept of social frailty offers a new perspective on the importance of wider determinants not often considered medically relevant. Current evidence suggests that social frailty poses a significant challenge to successful ageing among older adult populations. However, much of our current understanding of this state is based upon research involving older adults in the general population. The aim of this study is to obtain a holistic understanding of the characteristics of social frailty among older adults with ID, including its association with physical frailty and capacity to predict mortality. These insights may have potentially significant implications for policy and clinical practice.

Central to the social perspective of frailty being explored in this thesis is the understanding that social factors are associated with changes in frailty status. In the upcoming chapters, research evidence supporting this claim is comprehensively examined. Firstly, in Chapter 2, a narrative review explores social factors associated with frailty among older adults. This is followed by a systematic review in Chapter 3 which has a specific focus on factors associated with changes in frailty status among older adults with ID. Insights yielded from these reviews provide important context for this study, situating it amongst other studies in the field. Furthermore, findings informed the development of this studies theoretical framework and methodology, as described in Chapters 4 and 5 respectively.

Chapter 2: Narrative Review of Social Factors Associated with Frailty Status among Older Adults

2.1 Introduction

Frailty is commonly defined as a disruption of homeostatic mechanisms leading to a state of vulnerability (Rockwood *et al.* 2005). However, as discussed in Chapter 1, the vulnerability of older adults does not appear to be completely explained by a biological perspective (Panza *et al.* 2019). As discussed in Section 1.5.3, there are numerous individual characteristics which may influence health status such as age, gender, ethnic group, hereditary factors, constitutional factors (Marmot *et al.* 2012). Differences in relation to lifestyle factors, living and working conditions, food supply, access to essential goods, education, and welfare services have the potential to affect health outcomes. Furthermore, it is increasingly recognised that economic, cultural and environmental factors impact population health (Rice and Sara 2019, Dahlgren and Whitehead 1991). Indeed, it is becoming increasingly accepted that social determinants of health can affect frailty outcomes (Chamberlain *et al.* 2016, Theou *et al.* 2015, Levers *et al.* 2006, Woo 2005, Rockwood 2005). This has led to the development of a social conceptualisation of frailty which considers the relationship between the individual and their social environment (Andrew *et al.* 2018). It is of utmost importance that research evidence in relation to these topics are comprehensively examined as an initial focus of this dissertation.

The overall aim of this chapter is to present evidence regarding the association between social factors and frailty status among older adults. To provide a broad understanding of the current evidence base, this review focuses on studies involving older adults in the general population. Firstly, social correlates and determinants of frailty are examined with the support of relevant literature. This is followed by a review of emerging research regarding the concept of social frailty which considers the combined influence of social related factors on health outcomes. In line with study objectives, common characteristics of social frailty and approaches to its measurement among older adult populations are analysed. Findings of this chapter frame the focus of the systematic review in chapter 3 which specifically investigates factors associated with frailty development among older adults with ID

2.2 Methods

This review has been conducted in accordance with The Scale for the Assessment of Narrative Review Articles (SANRA) items (Baethge *et al.* 2019) (Appendix 1). SANRA was developed between 2010 and 2017 by three experienced journal editors as a simple and brief quality assessment instrument. The authors of the scale recommend its use in preparing narrative reviews. The version of the SANRA scale used in this narrative review was developed by the authors in 2014 which includes several modifications to simplify the scale and enhance its robustness. Six items form the scale and cover the following topics (1) explanation the review's importance; (2) statement of the aims of the review; (3) description of the literature search; (4) referencing; (5) scientific reasoning; and (6) presentation of relevant and appropriate endpoint data. These items are rated in integers from 0 (low standard) to 2 (high standard), with 1 as an intermediate score. The maximal sum score is 12.

The instructions for the SANRA scale call for transparency about the information sources used in a narrative review. The authors note that to achieve the highest rating on the scale it is not necessary to describe the literature search in as much detail as for a systematic review (e.g., searching multiple databases, including exact descriptions of search history, flow charts), but it is deemed necessary to specify search terms, and the types of literature included. In this review, searches of literature were performed using electronic databases PubMed, PsychINFO and Embase. Search strings were created using the terms 'frailty' (OR 'frail elderly') AND 'social determinants' (OR 'social factors') AND 'social frailty' (OR 'social vulnerability'). Studies were required to meet the following inclusion criteria: (i) be an original scientific article; (ii) provide a definition of frailty; (iii) report at least one individual characteristic associated with frailty status. To ensure the quality of information sources, only peer-reviewed articles were included. No limitation was placed on date of publication. A manual search of references of included studies was performed to identify other relevant articles. This review is limited to articles printed in the English language only. A summary of studies retrieved from the search is provided in Table 2.1.

Table 2.1 Summary of studies included in narrative review

Authors	Study type	Sample population	Social factors related to frailty
Alvarado <i>et al.</i> (2008)	Cross-sectional	10,661 men and women aged ≥60 years living in five large Latin American cities	<ul style="list-style-type: none"> • Adulthood socioeconomic situation • Childhood socioeconomic situation • Perception of income • Marital status
Ament <i>et al.</i> (2014)	Cross-sectional	475 community-dwelling older adults aged ≥70 in the Netherlands	<ul style="list-style-type: none"> • Missing people around • Feeling abandoned • Experiencing emptiness
Andrew <i>et al.</i> (2008b)	Measure construction	3,707 community-dwelling older adults aged 70+ in Canada	<ul style="list-style-type: none"> • Ability to read or write • Satisfaction with income • Education • Experience of warm and trusting relationships • Feeling empowered • Feeling in control of life situation • Home ownership • Leisure activities • Maintaining close relationships • Marital status • Socially oriented activities of daily living • Social support
Andrew and Rockwood (2010)	Measure construction	2,468 community-dwelling older adults aged 70+ in Canada	<ul style="list-style-type: none"> • Ability to read or write • Satisfaction with income • Education • Experience of warm and trusting relationships • Feeling empowered • Feeling in control of life situation • Home ownership • Leisure activities • Maintaining close relationships • Marital status • Social support • Socially oriented activities of daily living

Armstrong <i>et al.</i> (2015)	Measure construction	3,271 older men was derived from the Honolulu-Asia Aging Study (HAAS)	<ul style="list-style-type: none"> • Ability to read or write • Satisfaction with income • Education • Experience of warm and trusting relationships • Feeling empowered • Feeling in control of life situation • Home ownership • Leisure activities • Maintaining close relationships • Marital status • Social support • Socially oriented activities of daily living
Shega <i>et al.</i> (2012)	Measure construction	5,703 individuals aged 65 and older from 36 cities and their surrounding rural areas across Canada	<ul style="list-style-type: none"> • Ability to read or write • Satisfaction with income • Education • Experience of warm and trusting relationships • Feeling empowered • Feeling in control of life situation • Home ownership • Leisure activities • Maintaining close relationships • Marital status • Social support • Socially oriented activities of daily living
Wallace <i>et al.</i> (2015)	Measure construction	18,289 community-dwelling participants 50 years and older from SHARE Wave 1 (2004)	<ul style="list-style-type: none"> • Ability to read or write • Satisfaction with income • Education • Experience of warm and trusting relationships • Feeling empowered • Feeling in control of life situation • Home ownership • Leisure activities • Maintaining close relationships • Marital status

				<ul style="list-style-type: none"> • Socially oriented activities of daily living • Social support
Aranda <i>et al.</i> (2011)	Longitudinal	2,069 community-dwelling Mexican Americans aged 75 years and above		<ul style="list-style-type: none"> • Gender • Cognitive performance • Education • Emotional support • Financial strain • Gender • Nativity • Type of insurance • Neighbourhood composition • Positive affect
Bilotta <i>et al.</i> (2010)	Cross-sectional	239 community-dwelling adults in Italy aged 65 and above		<ul style="list-style-type: none"> • Functional independence • Home and neighbourhood • Religion • Leisure activities
Chamberlain <i>et al.</i> 2016	Longitudinal	16,443 community dwelling older adults aged 60-89 years in Minnesota, USA		<ul style="list-style-type: none"> • Low level of education • Marital status • Concerns from family/ friends about alcohol consumption • Smoking
Cramm and Nieboer (2013)	Cross-sectional	945 community-dwelling people from Rotterdam aged 70 years and above		<ul style="list-style-type: none"> • Gender • Educational level • Marital status • Social cohesion and a sense of belonging within the neighbourhood • Gender
Etman <i>et al.</i> (2012)	Longitudinal	14,424 community-dwelling persons aged ≥55 years in the Netherlands		<ul style="list-style-type: none"> • Gender • Level of education • Marital status • Gender

Garre-Olmo <i>et al.</i> (2013)	Cross-sectional	875 community-dwelling adults in Spain aged >74 years	<ul style="list-style-type: none"> • Living alone • Contact with family and/or friends/ neighbours less than once a week • Lack of a person to help with ADL • Infrequent contact with family • Infrequent contact with friends or neighbours (less than once a week) • Absence of a confidant • Lack of support for daily living during the past three months
Gobbens <i>et al.</i> (2010)	Cross-sectional	Two cohorts of community-dwelling persons aged 75 years and older in the Netherlands (N= 245; N= 234)	<ul style="list-style-type: none"> • Gender • Education • Ethnicity • Income • Influence of life events • Living alone • Living environments • Marital status • Gender • Social support • Social relationships
Gobbens <i>et al.</i> (2012)	Longitudinal	213 community-dwelling people, aged 75 years and older in the Netherlands	<ul style="list-style-type: none"> • Gender • Education • Ethnicity • Income • Influence of life events • Living alone • Living environments • Marital status • Gender • Social support and relationships
Harttgen <i>et al.</i> (2013)	Cross-sectional	180,108 adults aged 50 years and above living in Europe	<ul style="list-style-type: none"> • Education • Income

Herrera-Badilla <i>et al.</i> (2015)	Cross-sectional	927 community-dwelling people in Mexico aged ≥ 70	<ul style="list-style-type: none"> • Age • Gender • Lone living/ Loneliness • Cognitive performance • Disability
Hoogendijk <i>et al.</i> (2016)	Longitudinal	1,115 community dwelling people aged 65 and over in Amsterdam	<ul style="list-style-type: none"> • Emotional and instrumental support • Loneliness
Hsu and Chang (2015)	Longitudinal	2,306 older Taiwanese adults	<ul style="list-style-type: none"> • Gender • Education • Financial satisfaction
Kawano-Soto <i>et al.</i> (2012)	Cross-sectional	927 adults aged 70 and older living in Mexico City	<ul style="list-style-type: none"> • Gender • Care from family member • Education • Financial support • Family/friends in the same neighbourhood • Friends/family to assist in case needed
Lo <i>et al.</i> (2017)	Cross-sectional	923 community-dwelling people in Taiwan aged 65 years	<ul style="list-style-type: none"> • Dietary pattern
Lu <i>et al.</i> (2017)	Longitudinal	4,386 community-dwelling people aged 50 years or above in England	<ul style="list-style-type: none"> • Gender • Employment • Career breaks/ retirement
Makizako <i>et al.</i> (2015)	Longitudinal	4304 community-dwelling older adults aged ≥ 65 years living in Japan	<ul style="list-style-type: none"> • Feeling helpful to friends or family • Going out less frequently compared to last year • Living alone • Talking with someone every day • Visiting friends sometimes
Makizako <i>et al.</i> (2018)	Longitudinal	1226 community-dwelling older adults ≥ 65 years living in Japan	<ul style="list-style-type: none"> • Feeling helpful to friends or family • Going out less frequently compared to last year • Living alone • Talking with someone every day • Visiting friends sometimes
O'Halloran <i>et al.</i> (2018)	Longitudinal	4,908 people aged 58 years and older living in Ireland	<ul style="list-style-type: none"> • Age • Gender • Living alone

			<ul style="list-style-type: none"> • Disability in ADL or IADL • Lower levels of cognitive function
Park <i>et al.</i> (2019)	Cross-sectional	408 older adults (mean age 75 years) in the Aging Study of PyeongChang Rural Area	<ul style="list-style-type: none"> • Feeling helpful to friends or family • Going out less frequently compared with last year • Living alone • Talking with someone every day • Visiting friends sometimes
Theou <i>et al.</i> (2013)	Cross-sectional	36,306 community-dwelling people aged 50 and older from 15 countries participating in SHARE study	<ul style="list-style-type: none"> • GPD per capita • Healthcare expenditure
Tsutsumimoto <i>et al.</i> (2017)	Cross-sectional	4,425 older Japanese people from the National Centre for Geriatrics and Gerontology Study of Geriatric Syndromes	<ul style="list-style-type: none"> • Feeling helpful to friends or family • Going out less frequently compared with last year • Living alone • Talking with someone every day • Visiting friends sometimes
Woo <i>et al.</i> (2005)	Cross-sectional	2,032 older Chinese people aged 70 years and above	<ul style="list-style-type: none"> • Gender • Lifestyle • Socioeconomic status • Social support network

2.3 Findings

A total of 29 papers were obtained from the literature search which included cross-sectional studies (N=14), longitudinal studies (N=10), and studies involving construction of frailty measures (N=5). Based on an analysis of their findings, the following social related factors were identified as being associated with frailty: age; gender; education; employment; income; functional impairment; cognitive impairment, lone living; social relationships; loneliness; lifestyle behaviours; religion; social frailty.

2.3.1 Age

The relationship between frailty status and age is well-documented. It is widely understood that the likelihood of being frail is significantly associated with increasing age. The longitudinal cohort study by Etman *et al.* (2012), which had a sample of 14,424 community-dwelling persons aged ≥ 55 years living in the Netherlands, found that participants aged ≥ 80 years were more likely to experience worsening frailty status in comparison to those aged 55-59 years. Similar findings have been reported in the study by O'Halloran *et al.* (2018), which involved people aged 58 years and older living in Ireland (N=4,908). They found that the prevalence of frailty increased with advancing age across the age groups 58-64 (8%), 65-74 (14% to 33%) and ≥ 75 years (30%). A similar but smaller increase in prevalence in pre-frailty was also observed across these age groups from 28% to 38% to 45% respectively. Numerous other studies have observed older adults to have higher levels of frailty in comparison to younger cohorts (Cramm and Nieboer 2013, Harttgen *et al.* 2013, Jürschik *et al.* 2012, Kawano-Soto *et al.* 2012, Alvarado *et al.* 2008).

2.3.2 Gender

Several studies internationally have found a strong correlation between gender and frailty status. The cross-sectional study by Alvarado *et al.* (2008), which included 10,661 men and women aged ≥ 60 years living in five large Latin American cities, revealed that gender was significantly and consistently associated with frailty after the age of 60 years. There is growing evidence that women are significantly more at-risk of having poorer frailty status in comparison to men. For instance, female participants in the study by Etman *et al.* (2012) had a 1.26-fold higher risk of worsening frailty state compared to their male peers. The cross-sectional study by Woo *et al.* (2005), which had a sample of 2,032 people aged ≥ 70 years in Japan, revealed that frailty levels defined as deficit accumulation were higher among women in comparison to men. Findings by O'Halloran *et al.* (2018) indicate that the prevalence of frailty is almost

twice as high women in comparison to men (22% versus 13%). Similar associations have been widely reported elsewhere (Herrera-Badilla *et al.* 2015, Harttgen *et al.* 2013, Kawano-Soto *et al.* 2012, Jürschik *et al.* 2012).

2.3.3 Education

Level of education attainment appears to be strongly linked to frailty status among older adults. The longitudinal study by Chamberlain *et al.* (2016), which involved community dwelling older adults aged 60-89 years in Minnesota, USA (N=16,443), showed that worsening frailty was associated with having a lower level of educational attainment. O'Halloran *et al.* (2018) revealed that community dwelling older adults in Ireland were three times more likely to have a lower level of educational attainment (29% for primary level versus 10% for third level). Etman *et al.* (2012) observed a higher risk of worsening frailty status among older adults with lower levels of education in comparison to those with higher levels of education. Hoogendijk *et al.* (2016) had similar findings in their study involving community dwelling people aged 65 and over in Amsterdam (N=1,115). They showed that high frailty levels were significantly associated with a lower level of education. These findings are supported by other studies (Cramm and Nieboer 2013, Alvarado *et al.* 2008).

2.3.4 Employment and Income

The role of occupational factors in frailty manifestations among older adults is poorly understood and few studies have explored this relationship. Current research evidence indicates that unfavourable occupational conditions may affect frailty outcomes at older age. In the longitudinal survey study by Lu *et al.* (2017), which included a nationally representative sample of community-dwelling people aged 50 years or above in England (N=4,386), women who were never employed were more likely to have higher FI scores in comparison to men. Type of occupation and career trajectories also appear to be associated with frailty status. In the study by Alvarado *et al.* (2008), the likelihood of being frail was associated with having a manual occupation and being a housewife ($p \leq 0.05$), while Woo *et al.* (2005) found that increasing frailty was associated with non-white collar occupations among men. Lu *et al.* (2017) observed that distinct periods focused on work and family care had a protective effect on frailty levels after the age of 60 years among female participants. Women who took a short break for family care and then undertook part-time work until 59 years had lower FI scores at 60 years in comparison to those who were mostly in full-time work until 59

years. Among male participants, being able to leave paid employment before 65 years if in poor health appeared to slow down increases in frailty after retirement. However, those who exited early from paid work were more likely to have higher FI scores.

Others have reported that frailty levels among older adult populations are correlated with national economic indicators. The study by Theou *et al.* (2013) involved a secondary analysis of data from the Survey of Health, Ageing and Retirement in Europe (SHARE), a longitudinal population-based survey (SHARE 2021). Participants included community-dwelling people aged 50 and older across 15 countries in Europe (N=36,306). Findings revealed that mean FI scores were higher among low-income countries in comparison to high-income countries. However, mean FI scores were negatively correlated with both gross domestic product and health expenditure. Higher income levels appeared to have a protective effect on frailty levels and survival.

The cross-sectional study by Harttgen *et al.* (2013), which also used data from SHARE, explored frailty in older adults aged 50 years and above living in high- and low-income countries across Europe (N=180,108). They found that the level of frailty was distributed along the socioeconomic gradient in both higher and lower income countries with a lower level of income associated with a greater likelihood of being frail. Furthermore, Hsu and Chang (2015) observed that higher financial satisfaction was a protective factor against high frailty levels, while frail participants in the study by Alvarado *et al.* (2008) were more likely to have had impoverished childhoods or economic hardship later in life.

2.3.5 Functional and Cognitive Impairment

Several studies report an association between functional capacity and frailty status among older adults. Findings by Aranda *et al.* (2011) indicate that increasing levels of frailty are associated with higher levels of impaired Activities of Daily Living (ADL) and Instrumental Activities of Daily Living (IADL). Frail participants in the longitudinal study by Chamberlain (2016) who were dependent on assistance from others such as paid support staff, family, and/or friends were more likely to experience adverse frailty trajectories. O'Halloran *et al.* (2018) found that the prevalence of at least one disability in instrumental or basic activities of daily living to be significantly higher among adults living with frailty compared to those with pre-frailty or in robust health. The longitudinal study by Hsu and Chang (2014), which analysed data from individuals aged 64 years

or older in Taiwan (N=3363), revealed that lower levels of ADL difficulty were protective factors against frailty. Similar findings have been replicated elsewhere (Kawano-Soto *et al.* 2012, Bilotta *et al.* 2010).

Cognitive impairment has also been identified as a risk factor for frailty among older adults. In the cross-sectional study by Herrera-Badilla *et al.* (2015), which included a sample of community-dwelling people in Mexico aged ≥ 70 (N= 927), frail participants were more likely to have poorer cognitive performance. Similarly, Aranda *et al.* (2011) observed that higher frailty levels were associated with lower levels of cognitive functioning. The cross-sectional study of 2,069 Mexican American adults aged 75+ years and living in Mexico City by Kawano-Soto *et al.* (2012) found that frailty status was associated with memory loss based on Mini-Mental State Examination (MMSE) scores. O'Halloran *et al.* (2018) assert that frailty may be both a risk factor for and a consequence of decline in cognitive function based on the bi-directional relationship between frailty and cognitive health observed in their study.

2.3.6 Social Connections and Loneliness

There is growing evidence that frailty status among older adults is associated with relationship and lone-living status. The cross-sectional study by Cramm and Nieboer (2013), which involved community-dwelling people aged 70 years and above (N=945), reported that individuals who were frail were more likely to be unmarried. A longitudinal study by O'Halloran and O'Shea (2018) found that higher levels of deficit accumulation frailty were associated with living alone or being widowed. In total, 10% of participants aged 58 years and over were living alone, of whom 23% were living alone with frailty. Among those aged 75 years and over living alone, 44% were frail. Similarly, Jürschik *et al.* (2012) observed a greater likelihood of being frail among participants who were living without a partner (widow/er or single). These findings were replicated in the study by Chamberlain *et al.* (2016), which showed that being married or living with a spouse/ domestic partner was found to be a protective factor for frailty progression. Conversely, one study by Etman *et al.* (2012) found no significant differences in risk of worsening in frailty state in relation to marital status (i.e., married/registered partnership, never married, divorced, widowed).

It is increasingly understood that social cohesion and sense of solidarity among members of a community are associated with frailty status among older adults. A cross-sectional study by Aranda *et al.* (2011) examined the relationship between medical, psychosocial, and neighbourhood factors and increasing frailty among community-dwelling Mexican Americans aged 75 years and above (N=2,069). They found that neighbourhood characteristics had a protective effect on frailty outcomes with increasing levels of frailty being associated with living in a less ethnically dense neighbourhood. Furthermore, Cramm and Nieboer (2013) observed that feeling more secure and having a stronger sense of social cohesion and neighbourhood belonging was associated with lower levels of frailty. This supports earlier work by Woo *et al.* (2005) who reported that having few relatives or neighbours and no or infrequent participation in helping others was associated with higher levels of frailty.

Several studies report that frailty status among older adults is associated with differences in relation to social connections. In the longitudinal study by Hoogendijk *et al.* (2016), older adults who had smaller networks than their non-frail peers were more likely to be pre-frail. Interestingly in the study by Kawano-Soto *et al.* (2012), frailty status was not associated with quality of social networks. Other studies have revealed an association between frailty status and loneliness. Herrera-Badilla *et al.* (2015) found that pre-frail and frail participants were more likely to report loneliness than non-frail participants. Similarly, in the study by Chamberlain *et al.* (2016), frail and pre-frail participants were lonelier in comparison to those identified as robust. Hoogendijk *et al.* (2016) observed that higher levels of frailty were associated with poor social functioning, and with an increase in loneliness over time.

2.3.7 Lifestyle Behaviours and Religion

Several studies report that frailty status among older adults appears to be influenced by smoking and alcohol consumption. In the cross-sectional study by Jürschik *et al.* (2012), involving 640 individuals aged 75 to 105, frail participants were more often smokers and significantly more often alcohol consumers than non-frail participants. Similar findings have been reported by Chamberlain *et al.* (2016). They found that participants aged 60-79 years who had ever smoked and/or have concerns about alcohol consumption were more likely to have a high frailty trajectory. However, this effect was not seen in the oldest cohort aged 80-89 years. Additionally, adverse frailty trajectories defined as deficit accumulation have also been shown to be associated with no or little exercise and abstinence from alcohol (Woo *et al.* 2005). In one study by Lo *et al.* (2017) involving community-dwelling people in Taiwan aged 65 years (N= 923), dietary intake was associated with frailty status. A dietary pattern featuring more phytonutrient-rich plant foods, tea, protein-rich foods, and omega-3-rich deep-sea fish was associated with a reduced prevalence of frailty. Other studies have found that religiosity is significantly associated with frailty status among older adults. The cross-sectional study by Bilotta *et al.* (2010), which involved community-dwelling adults in Italy aged 65 and above (N=239), found that frailty status was associated with an increased level of engagement in religious activities. Similar findings were observed in the study by Woo *et al.* (2005) which revealed that absence in participation of religious activities was predictive of higher frailty levels among women.

2.3.8 Social Frailty

To this point, research evidence on the association between individual social factors and frailty status has been analysed. The following sections examine the findings of studies adopting a social conceptualisation of frailty to investigate the accumulative effect of social factors on health outcomes among older adults. Social frailty has been described in the realm of the overall frailty concept (van Campen 2011, Gobbens *et al.* 2010a), however it is arguably the least explored domain (Bunt *et al.* 2017). Most of the literature is still inconclusive on the nature and scope of social frailty as a concept (Bessa *et al.* 2018) and there are calls for further research to obtain a deeper understanding of the relative importance of specific factors in relation to social frailty.

Although different theories on social needs exist, the conceptual model by Bunt *et al.* (2017) defines social frailty as:

“A continuum of being at risk of losing, or having lost, social resources, general resources, and social activities or abilities that are important for fulfilling one or more basic social needs during the life span” (p. 326).

Among studies analysed in this review, several different approaches to measuring social frailty were adopted. Some assessed social frailty using questionnaires comprising of specific questions about daily social activities, social roles and social relationships (Park *et al.* 2019, Tsutsumimoto *et al.* 2017, Makizako *et al.* 2015). Another questionnaire-based tool, The Tilburg Frailty Indicator (Gobbens *et al.* 2010b), measures social frailty as the presence of at least two of the following criteria: lone living; lack of contacts; and lack of support. Similarly, Garre-Olmo *et al.* (2013) created a social frailty phenotype which measured social frailty as the presence of two or more off the following criteria: living alone, infrequent contact with family (less than once a week), lack of a person to help with ADL, infrequent contact with friends or neighbours (less than once a week), absence of a confidant and lack of support for daily living (during the past 3 months).

The Social Dysfunction Rating Scale (SDRS), developed by Lozupone *et al.* (2018), measures social frailty according to 21 subjective and objective evaluations in relation to personal satisfaction, self-fulfilment and social role performance. In the study by Andrew *et al.* 2008, and in subsequent studies (Armstrong *et al.* 2015, Wallace *et al.* 2014, Shega *et al.* 2012, Andrew and Rockwood 2010), social frailty was measured according to the deficit accumulation model.

All studies constructed frailty indices comprising of self-report variables relating to social factors that could be considered as deficits. In the study by Andrew *et al.* (2008) deficits included in the index related to social support, social engagement, sense of mastery or control over one’s life circumstances, socioeconomic status and social-related IADLs required for community participation and maintaining social ties. A summary of the different factors represented in each of the aforementioned social frailty measures is provided in Table 2.2.

Table 2.2 Comparison of social frailty measures

Measure	Social factors represented	Examples of use
Social Frailty Questionnaire	<ul style="list-style-type: none"> • Feeling helpful to friends or family • Going out less frequently compared to last year • Living alone • Talking with someone every day 	Park <i>et al.</i> (2019), Tsutsumimoto <i>et al.</i> (2017), Makizako <i>et al.</i> (2015)
Tilburg Frailty Indicator	<ul style="list-style-type: none"> • Do you live alone? • Do you sometimes miss having people around you? • Do you receive enough support from other people? 	Gobbens <i>et al.</i> (2010b)
Social Vulnerability Index	<ul style="list-style-type: none"> • Ability to read or write • Satisfaction with income • Education • Experience of warm and trusting relationships • Feeling empowered • Feeling in control of life situation • Home ownership • Leisure activities • Maintaining close relationships • Marital status • Social support • Socially oriented activities of daily living 	Armstrong <i>et al.</i> (2015), Wallace <i>et al.</i> (2014), Shega <i>et al.</i> (2012), Andrew and Rockwood (2010), Andrew <i>et al.</i> (2008)

2.3.9 Common characteristics of social frailty

Few studies have investigated social frailty and therefore our understanding of this health state is relatively limited. However, based on current research evidence, social frailty appears to have distinct characteristics among older adults. As in overall frailty, the prevalence of social frailty appears to increase with age. Tsutsumimoto *et al.* (2017) examined data from community-dwelling adults in Japan aged 65+ (N=4,425). In this study, the prevalence of social frailty was higher according to increased age in

comparison to social pre-frailty. In the longitudinal study by Andrew *et al.* (2008), which involved a secondary analysis of data from community-dwelling older adults aged 70+ in Canada (N=3,707), levels of social frailty increased significantly, albeit weakly, with advancing age. Additionally, female participants were observed to have on average higher index scores compared to men at all ages.

The cross-sectional study by Park *et al.* (2019), which included a sample of community dwelling older adults aged ≥ 65 years in Korea (N= 408), showed that prevalence of social frailty was higher among participants living in urban areas compared to those living in rural areas (20.5% vs 10.2%). Others have reported a greater likelihood of being frail to be associated with having a widowed marital status (Garre-Olmo *et al.* 2013). The longitudinal study by Wallace *et al.* (2014) investigated adverse outcomes associated with social frailty among participants of SHARE aged ≥ 50 years (N= 18,289). They found that higher levels of social frailty predicted mortality and disability. Interestingly, this relationship varied by the social model of the country. Other studies have found that that social frailty is independently associated with physical function and predicts future disability (Tsutsumimoto *et al.* 2017, Makizako *et al.* 2015). Conversely, the cross-sectional study by Ament *et al.* (2014), which involved community-dwelling older adults aged ≥ 70 in the Netherlands (N= 475) showed that in addition to other domains, social frailty had no additional impact on disability, hospital admission or quality of life. Findings by Gobbens *et al.* (2012) indicate that while social frailty is associated with increased contacts with health care professionals over two years, it is not predictive of other adverse outcomes beyond a timeframe of two years. Furthermore, they found that physical frailty was more predictive of adverse outcomes in comparison to social frailty.

Several studies have investigated the relationship between social frailty and other domains of frailty. Andrew *et al.* (2008) found that social frailty and overall frailty defined as deficit accumulation were weakly to moderately correlated with each other, with correlations being higher for women in comparison to men. The observational prospective study by Garre-Olmo *et al.* (2013), which used a sample of 875 community-dwelling adults in Spain aged >74 years, found that almost half of participants identified as socially frail were also mentally or physically frail, while the remaining individuals had social frailty only. A similar association was observed in the cross-sectional study

by Park *et al.* (2019), which included a sample of community dwelling older adults aged ≥ 65 years in Korea (N=408). They showed that social frailty was more prevalent in comparison to physical frailty with the percentage of social frailty alone being significantly higher than that of physical frailty alone or both social and physical frailty. Others have asserted that social frailty may contribute to physical frailty (Makizako *et al.* 2018).

2.4 Conclusion

This review aimed to present evidence regarding the association between social factors and frailty status among older adults in the general population. Based on the literature examined in this review, it seems clear that a broad range of social-related factors, oftentimes ignored from a medical context, are associated with adverse frailty outcomes. Social risk factors for frailty include age, gender, education attainment, employment, financial status, cognitive and functional ability, social relationships, loneliness, lifestyle behaviours and religiosity. Furthermore, studies adopting a social conceptualisation of frailty have shown that a sum of social factors might traduce a socially adverse environment, increasing the odds for elderly to be frail. While still an emerging field of research, there is growing consensus that social frailty is a complex and multidimensional process associated with adverse health outcomes. While social frailty appears to overlap with physical frailty to a degree, it has been shown to independently contribute to mortality. This demonstrates the potential utility of a social frailty measure in identifying individuals with poorer health status not captured by physical frailty alone (Park *et al.* 2018).

This review has provided a broad understanding of social related factors associated with frailty status and the characteristics of social frailty among older adults in the general population. However, these findings need to be interpreted with caution. Older adult populations are highly heterogenous (Mitnitski *et al.* 2017) and therefore the generalisability of these findings to older adults with ID cannot be assumed. In the next chapter, a systematic review addresses a defined research question in relation to factors associated with frailty status among older adults with ID. Insights obtained were used to inform the development of this studies theoretical framework and methodology, described in Chapters 4 and 5 respectively.

**Chapter 3: A Systematic Review of Literature
Regarding Factors Associated with Frailty
Progression among Adults with Intellectual
Disability**

3.1 Introduction

The narrative review in Chapter 2 demonstrated how social factors clearly influence frailty outcomes among older adults in the general population. However, aging populations are highly heterogeneous (Mitnitski *et al.* 2017) and therefore it cannot be assumed that these findings are generalizable to older adults with an ID. This chapter presents a systematic review conducted to identify and critically analyse all available primary research regarding factors associated with the development and progression of frailty in this population. The research question underpinning this review is: “What individual characteristics are associated with changes in frailty status over time among adults with ID?”. In line with the aims and objectives of this study, the influence of social factors on frailty transitions and trajectories is given priority focus. Insights obtained have helped to inform the theoretical framework and methodology underpinning this study, and these are expounded upon in greater detail in Chapters 3 and 4 respectively.

3.2 Methods

This systematic review was conducted using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher *et al.* 2009) to identify studies reporting factors associated with changes in frailty status over time among adults with ID. The review has been registered on PROSPERO (179803, 05/07/2020). A version of this literature review has been previously published (Dee *et al.* 2021).

3.2.1 Search Strategy

Searches of literature were performed up until October 2019 using the electronic databases MEDLINE, CINAHL Complete, APA PsycINFO and Embase. Search strings were created from the research question following preliminary trial-and-error searches. These search strings were combined using the Boolean operators ‘AND’ and ‘OR’. Search terms included a combination of keywords and related controlled vocabulary for “intellectual disability” and “frailty” (Appendix 2). A manual search of references from included articles was also conducted.

3.2.2 Inclusion and Exclusion Criteria

Criteria used to determine the inclusion or exclusion of articles for inclusion in this review are described in Table 3.1. Studies were required to meet the following inclusion criteria: (i) be an original scientific article; (ii) define frailty as deficit accumulation; (iii) longitudinally analyse change in frailty states; (iv) report at least one individual characteristic associated with change in frailty status; (v) include a study sample of people with ID aged ≥ 18 years; (vi) be printed in the English language. Studies were excluded if (i) they did not measure frailty based on the accumulation of deficits; (ii) did not report the instrument used to measure frailty, or (iii) had a cross-sectional design. To ensure the quality of information sources, only peer-reviewed articles were included. No limitation was placed on date of publication

Table 3.1 Eligibility criteria

Criteria	Inclusion	Exclusion
Study design	<ul style="list-style-type: none"> • Longitudinal 	<ul style="list-style-type: none"> • Cross-sectional
Type of article	<ul style="list-style-type: none"> • Peer-reviewed 	<ul style="list-style-type: none"> • Not peer-reviewed
Participants	<ul style="list-style-type: none"> • Intellectual disability • Aged ≥ 18 years 	<ul style="list-style-type: none"> • No intellectual disability • Aged ≤ 18 years
Language	<ul style="list-style-type: none"> • English 	<ul style="list-style-type: none"> • Any other language
Outcomes of interest	<ul style="list-style-type: none"> • Frailty defined as deficit accumulation • Reporting of change in frailty state over time • Individual characteristics associated with change in frailty status 	<ul style="list-style-type: none"> • Frailty measure not based on the accumulation of deficits • No details of frailty measure used reported

To be considered eligible for inclusion in this review, studies were required to meet the following criteria: (i) be an original scientific article; (ii) define frailty as deficit accumulation; (iii) longitudinally analyse change in frailty states; (iv) report at least one individual characteristic associated with change in frailty status; (v) include a study sample of people with ID aged ≥ 18 years; (vi) be printed in the English language. Studies were excluded if (i) they did not measure frailty based on the accumulation of

deficits, (ii) did not report the instrument used to measure frailty, or (iii) had a cross-sectional design. No limitation was placed on date of publication.

3.2.3 Screening

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

3.2.4 Quality Assessment

Following agreement on eligibility, two reviewers independently assessed the methodological quality of each study using the cohort study checklist developed by the Critical Appraisal Skills Programme (CASP 2018). This tool comprises of 14 questions: 1) did the study address a clearly focused issue?; 2) was the cohort recruited in an acceptable way?; 3) was the exposure accurately measured to minimise bias?; 4) was the outcome accurately measured to minimise bias?; 5) have the authors identified all important confounding factors?; 6) have they taken account of the confounding factors in the design and/or analysis?; 7) was the follow up of subjects complete enough?; 8) was the follow up of subjects long enough?; 9) what are the results of this study?; 10) how precise are the results?; 11) do you believe the results?; 12) can the results be applied to the local population?; 13) do the results of this study fit with other available evidence?; 14) what are the implications of this study for practice? Conflicts between the two reviewers were resolved using discussion and consensus.

3.2.5 Data Extraction and Analysis

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

3.3 Results

The PRISMA flow diagram, provided in Figure 3.1 depicts the flow of information through the different phases of this systematic review. Overall, 5420 articles were retrieved from the literature search. These were uploaded to Covidence systematic review software, and all duplicates (n=1600) were removed.

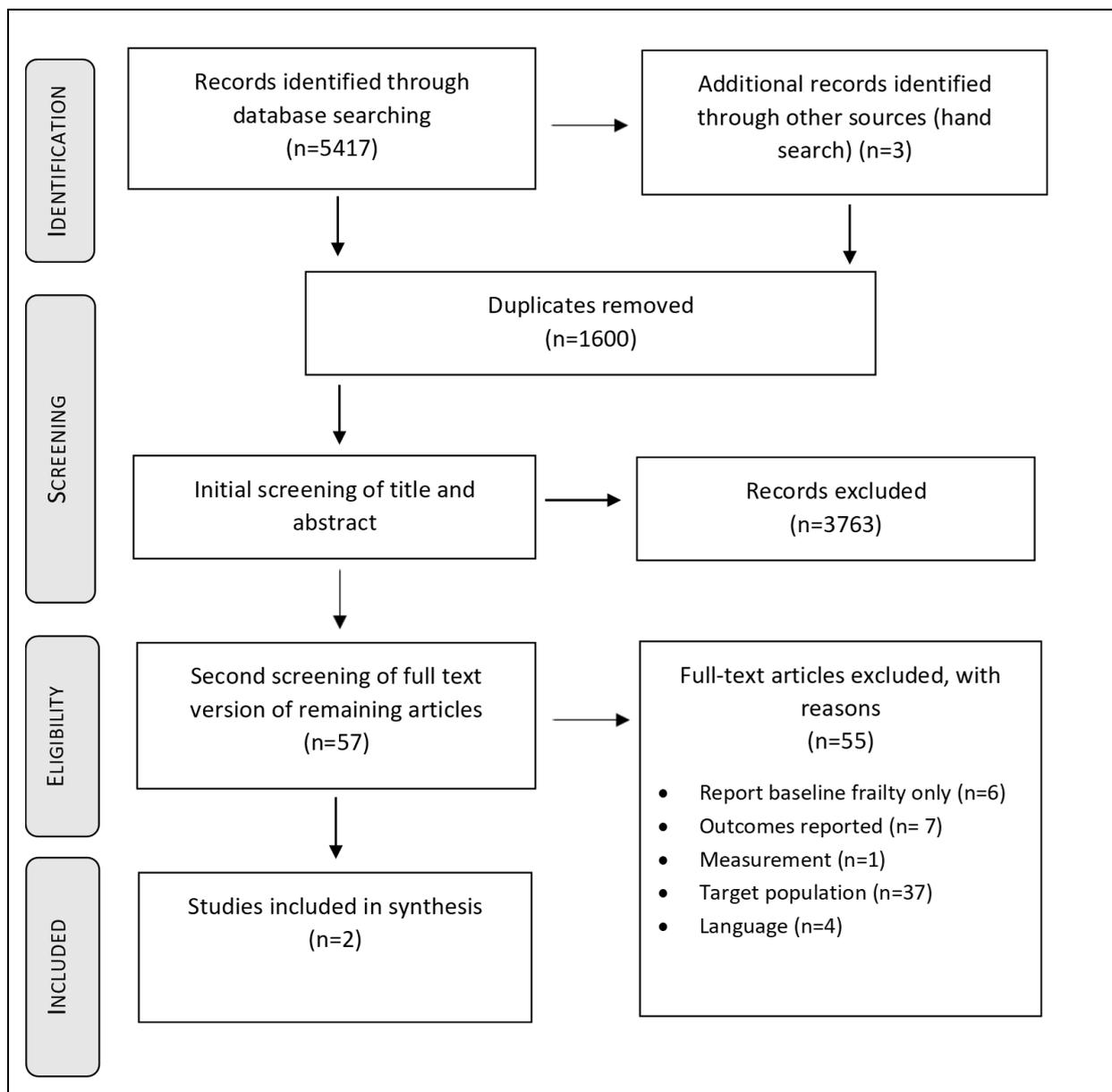


Figure 3.1 PRISMA flow chart of the literature screening process

In the first step of screening, abstract review, 3763 articles were identified as irrelevant. After screening the full text of the remaining 57 articles, further exclusions were agreed based on outcomes reported (n=7), no frailty measure specified (n=1), target population (n=37), language (n=4) or reporting of baseline frailty status only (n=6). No

studies were excluded based on methodological quality following review using the CASP Cohort Study Checklist (Appendix 3). Potential limitations of both studies are discussed in Limitations.

3.3.1 Study Characteristics

In total, two articles (Martin *et al.* 2018, Ouellette-Kuntz *et al.* 2018) qualified for inclusion in this review. These studies provide longitudinal analysis of administratively held clinical data collected from community dwelling adults with ID receiving home care in the same geographical region of Ontario, Canada. Martin *et al.* (2018) explored transitions in frailty status defined as deficit accumulation and the study by Ouellette-Kuntz *et al.* (2018) investigated factors associated with the rate of deficit accumulation over time. They both utilise the same data source, the Resident Assessment Instrument-Home Care (RAI-HC), as described by Hawes (2007). This instrument was developed by interRAI, an international organisation responsible for developing comprehensive clinical assessment systems for persons with disabilities and complex needs. The RAI-HC includes open-text diagnoses for assessors to indicate the presence of any disease/infection that a doctor has indicated is present and affects client's status, requires treatment, or symptom management or a disease that is monitored by a home care professional or is the reason for a hospitalization in the last 90 days (Morris *et al.* 2009). A summary of both studies is provided in Appendix 4.

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

Both studies provide sample characteristics at baseline. Martin *et al.* (2018) report the prevalence of cognitive impairment (52.1%), unsteady gait (47.3%), polypharmacy (i.e. ≥ 7 medications) (36.8%), functional impairment (36.0%), daily pain (24.5%), arthritis (17.6%), Down syndrome (17.2%), group home living (13.9%), dementia/Alzheimer disease (9.2%) and morbid obesity (8.1%). Baseline frailty status in this sample is reported as non-frail (67.0%), pre-frail (16.2%) and frail (16.8%). It was noted that non-frail individuals were younger (mean age 46 years) than those identified as pre-frail (mean 55.8 years) and frail (mean 57.7 years). Individuals who were pre-frail at baseline were more likely to have diabetes (25.2%) and morbid obesity (11.1%). Frail persons were more likely to be female (54.8%), have a diagnosis of Down syndrome (26.7%) and live in a group home setting (22.7%). In comparison to those who were non-frail and pre-frail at baseline, frail individuals were more likely to present with unsteady gait (83.8%), polypharmacy (64.9%), cognition impairment (59.1%), ADL impairment (56.5%), daily pain (51.3%), arthritis (30.2%) and dementia/Alzheimer disease (25.1%). The authors note that slightly under half of all individuals with Down syndrome were pre-frail or frail at baseline.

Oullette-Kuntz *et al.* (2018) report similar baseline characteristics in their sample regarding the presence of group home living (14.5%) cognitive impairment (49.3%), unsteady gait (47.3%), functional impairment (32.1%) and Down syndrome (16.6%). Use of nursing services and therapies at baseline are 17.4% and 13.3% respectively. Unlike the study by Martin *et al.* (2018), baseline characteristics regarding polypharmacy, daily pain, arthritis, dementia/Alzheimer, morbid obesity and frailty status by category are not provided. The mean FI score at first assessment was 0.17. Based on the FI's cut-off points (McKenzie *et al.* 2015a), whereby a FI score of < 0.21 indicates pre-frailty, participants in this study were on average pre-frail at baseline. They also report that individuals accumulated deficits at a rate of 2-3 per year depending on prior frailty level. A post hoc analysis by the authors revealed that between 24.5% (assessment #2: 189/773) and 31.84% (assessment #5: 78/245) of individuals in this study had recently moved to their current residence. In total, 18-19% of those living in non-group homes had a recent move at each follow-up assessment.

3.3.2 Outcome Measures

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

Functional and cognitive ability is assessed by both studies using algorithms embedded in the RAI-HC. In both studies, the Activities of Daily Living Hierarchy (ADLH) scale (Morris *et al.* 1999) was used to identify functional impairment in both studies. This instrument uses four ADL items which include personal hygiene, toilet use, mobility and eating. Scores range from 0 (independent) to 6 (total dependence), with a cut-off of 3 indicating impairment in self-care skills. Impaired cognition was assessed by both studies using the Cognitive Performance Scale (CPS) (Morris *et al.* 1994). This scale is based on items related to decision-making, expression, and short-term memory. Scores range from 0 (intact cognition) to 6 (very severe impairment), with a cut-off point of 3 indicating impaired cognition. In both studies, group home living was defined as: (i) board and care/assisted living/group home; (ii) group setting with non-relatives; and (iii) not living with a primary informal caregiver. The presence of Down syndrome was based on a record of this diagnosis identified in an open-text diagnostic field at any RAI-HC assessment. The study by Oullette-Kuntz *et al.* (2018) reports nursing and therapy use, based on level of use during the week prior to assessment. Therapies refer to physiotherapy, occupational therapy, and speech and language therapy. Services were coded as either received or not received, regardless of duration or intensity of services provided.

3.3.3 Statistical Analysis

Martin *et al.* (2018) report relative risk (RR) of worsening or dying versus staying stable or improving using a modified Poisson regression model with 95% confidence intervals (CIs). Bivariate models were developed which allowed for adjustment for significant confounders (i.e. biological gender, age, Down syndrome diagnosis, moderate or worse cognitive impairment, moderate or worse self-care impairment, residence in a group home, and time in months between baseline and follow-up assessment or death). In contrast, the study by Ouellette-Kuntz *et al.* (2018) used negative binomial regression models which consider the correlation of repeated assessments. This provided mean incidence rates (IR) (i.e., the mean number of deficits accumulated per year) and incidence rate ratios (IRR) with 95% CIs.

3.4 Outcomes

The significance of association between outcome factors and change in frailty state over time was determined by a p-value of ≤ 0.05 (Table 3.2). To minimize potential bias, both significant and non-significant associations are examined in this review.

Table 3.2 Significance of association between factors and change in frailty status

Factor	Significant association? (Yes/No) *	
	Ouellette-Kuntz <i>et al.</i> (2018)	Martin <i>et al.</i> (2018)
Age	Yes	Yes
Gender	No	No
Down syndrome	Yes	Yes
Group home living	Yes	Yes
Functional impairment	Yes	Yes
Cognitive impairment	Yes	No
Use of nursing services	Yes	Not reported
Use of therapies	Yes	Not reported
Baseline frailty status	Not reported	Yes

*Determined by p-value of ≤ 0.05 in any frailty cohort

3.4.1 Baseline Frailty Status

Martin *et al.* (2018) found that baseline frailty status was the largest predictor of worsening frailty or death over a one-year period and remained a significant predictor of this outcome after adjustment for confounding factors. The direction of the association differed by frailty level. Individuals who were pre-frail at baseline had an increased risk (RR 1.24, 95% CI 1.04-1.49, P-value 0.0179), whereas a lower level of risk was observed among those who had a baseline status of frail (RR 0.82, 95% CI 0.69-0.96, P-value 0.0342). Persons who were non-frail at baseline were more likely to remain non-frail over a period of 1 year (84.26%), while others in this group became pre-frail (7.74%), frail (4.08%), or died (3.92%) over this same period. Pre-frail and frail cohorts were 1.76- and 1.63-times more likely to experience this outcome compared to those who were non-frail at baseline. However, improvement in frailty status was also observed among individuals who were initially pre-frail (40%) and frail (20%).

3.4.2 Age

Age was found to be the strongest predictor of the rate of deficit accumulation from 40 years among non-frail and prefrail cohorts, and from 70 years among frail individuals (Oullette-Kuntz *et al.* 2018). In comparison to non-frail and pre-frail participants aged 18-29 years, frail individuals aged 40-49 years accumulated deficits 1.52 and 1.94 times faster respectively. Rate of deficit accumulation was associated with a 2.14-fold increase in deficit accumulation rate after age 70 when compared to those aged 18- to 29-years. Deficits were observed to accumulate at a rate of 2-3 per year, depending on prior frailty level. A 10-year increase in age was significantly associated with worsening frailty or death within one year (RR 1.30, 95% CI 1.24-1.36, P-value <0.0001) (Martin *et al.* 2018).

3.4.3. Gender

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

3.4.4 Down Syndrome

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

3.4.5 Group Home Living

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

3.4.6 Functional and Cognitive Impairment

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

3.4.7 Use of Nursing and Therapy Services

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

3.5 Discussion

Findings suggest that frailty is a dynamic process and bi-directional change in frailty status can occur. In the study by Martin *et al.* (2018), stability in frailty status was the most likely outcome and positive transitions were observed. However, one quarter of study participants who were robust at baseline died within one year (Martin *et al.* 2008). Similar findings have been reported among older adults in the general population (O'Halloran and O'Shea 2018). A 10-year increase in age was associated with an accelerated rate of frailty progression among non-frail and pre-frail individuals aged 40 years and above. This finding suggests the need to assess frailty from at least the age of 40 year in this population. The assessment of frailty may need to be considered earlier in certain 'at-risk' groups such as individuals with Down syndrome. Findings of this review reveal that the presence of this diagnosis is significantly associated with adverse frailty outcomes in a relatively short timeframe. Other cross-sectional studies have reported a high prevalence of frailty in this cohort (Schoufour *et al.* 2014b, Schoufour *et al.* 2013, Evenhuis *et al.* 2012). The assessment of frailty should therefore be a priority consideration for individuals with Down syndrome.

Surprisingly, frail individuals had a lower risk of experiencing worsening frailty or death one year later. As suggested by Martin *et al.* (2018), this may have been due to frailty being more clinically recognisable than pre-frailty, which may have led to care interventions preventing further deterioration. If this is the case, it may be a promising indicator of the short-term positive influence of interventions in managing frailty. To a degree, the study by Ouellette-Kuntz *et al.* (2018) analysed the influence of interventions on frailty outcomes, specifically the use of nursing and therapy services. However, their findings were inconclusive with the provision of these supports being associated with slower frailty progression among pre-frail individuals and accelerated among those who were frail. Targeted interventions for frailty in the ID population and their effect over time should be a priority focus of future research. The direction of change in frailty status was not found to be influenced by gender, with risk of adverse outcomes comparable between men and women. This finding was unexpected given that gender is associated with frailty transition patterns in general older adult populations (Kojima *et al.* 2019). There is however a lack of consensus on the role of gender in frailty among people with ID. Some cross-sectional studies have found that women with ID are more likely to be frail in comparison to men with ID (Ouellette-Kuntz

et al. 2015, McKenzie *et al.* 2015a), while other studies have identified no difference between men and women in this regard (Lin *et al.* 2015, Schoufour *et al.* 2014b, Brehmer-Rinderer *et al.* 2013, Brehmer and Webber 2010, Schoufour *et al.* 2013, Evenhuis *et al.* 2012).

Both studies found that living in a group home was associated with an increased risk of adverse frailty outcomes. These findings conflict with those in the cross-sectional study by McKenzie *et al.* (2015b) which show that living in a group home or living with other family members is associated with a reduced likelihood of being frail. There is however a lack of agreement generally on the association between type of living situation and prevalence risk of frailty among people with ID. For example, risk appears to be comparable between living with a spouse and/or child(ren) and living alone (McKenzie *et al.* 2016a). Other studies have reported no significant increase in risk between living in an institutionalised setting versus community-based home (Evenhuis *et al.* 2012, Brehmer and Webber 2010). Findings by Oullette-Kuntz *et al.* (2018) suggest that the high rate of relocation observed among participants in their study may have been a contributing factor. Further research is required to explore the relationship between living situation and frailty progression in this group.

The influence of functional and cognitive impairment on frailty outcomes among older adults with ID is considered by both studies. Martin *et al.* (2018) found that high functional impairment was associated with worsening frailty or death within one year. However, this outcome was not predicted by high cognitive impairment in any frailty cohort. Neither functional impairment nor cognitive impairment was associated with an accelerated rate in deficit accumulation (Oullette-Kuntz *et al.* 2018). This apparent protective effect may be reflective of an individual reaching the maximum number of deficits that can be tolerated, in this instance 70% of measured deficits in the FI utilised (McKenzie *et al.* 2015a). The potential limit to the number of deficits that can be tolerated by an individual may indicate a point of exhaustion in reserve capacity (Rockwood and Mitnitski 2006).

3.6 Limitations

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

3.7 Conclusion

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

Findings of this review indicate that a wide range of factors appear to influence frailty progression among adults with ID. In addition to increasing age and the presence of Down syndrome, several social-related factors pertaining to functional ability, living situation, and healthcare use were associated with frailty outcomes. A social conceptualisation of frailty recognises that social circumstances may contribute to health outcomes and affect access to appropriate interventions and supports (Andrew *et al.* 2018). Social frailty may provide a novel and additional approach to understanding the accumulative effect of social-related factors on health outcomes among older adults with ID. Insights obtained may support the development of upstream interventions which address social disparities and reduce modifiable

contributors to frailty. These insights may have potentially significant implications for policy and practice, particularly in the context of ongoing efforts to address social disparities experienced by this population. Informed by the findings of this systematic review, the next chapter sets out a theoretical framework showing the conceptual and analytical approach used by the researcher to address the aim and objectives of this study.

Chapter 4: Theoretical Framework

4.1 Introduction

Historically, frailty has been described as a state of vulnerability to poor resolution of homeostasis after a stressor event and because of cumulative decline in many physiological systems during a lifetime (Clegg *et al.* 2013). However, it is increasingly accepted that frailty cannot be explained solely in terms of biological factors. Findings from the literature review in previous chapters support the need to look beyond intrinsic biologic and physiologic parameters of frailty and consider the extrinsic social context in which people are born, live and grow.

In this chapter, a theoretical framework is used to explore key concepts underpinning this study and proposes relationship between them in the context of current literature. Selected theories are compared and critically analysed to develop theoretical constructs for this dissertation. These insights are intended to provide direction to this study and support the interpretation and explanation of research findings later in Chapter 7. The studies aim and objectives are tightly aligned and intricately interwoven into this framework. Therefore, conceptual models which explore physical frailty and social frailty, and hypothesise the relationship between these domains and their contribution to mortality are a central focus. Insights obtained have been used to inform the selection of this studies research methods, described in Chapter 5.

As discussed in Chapter 1, the most widely known and used conceptualisations of frailty over the past two decades have been the phenotype model (Fried *et al.* 2001) and the deficit accumulation model (Mitnitski *et al.* 2001). Since their emergence, several new and novel conceptual models of frailty have been developed. The integral conceptual model by Gobbens *et al.* (2010a) supports a lifespan approach to understanding the development of frailty and identifying opportunities for intervention. More recently, Bunt *et al.* (2017) devised a conceptual model of social frailty which considers how social and general resources, social behaviours and activities, and self-management abilities affect the fulfilment of social needs. In the following sections, these theories are individually defined and discussed in relation to their typical application, limitations and relevance to this study.

4.2 Frailty Phenotype

As discussed in Section 1.5.2, the most widely known and used conceptualisation of frailty is the phenotype model, commonly referred to as physical frailty. Originally developed by Fried *et al.* (2001) at John Hopkins University, the phenotype model identifies frailty as a clinical syndrome driven by age-related biologic changes. This model describes how sarcopenia leads to poor muscle strength, limiting mobility and physical activity. The resultant reduced energy expenditure and nutritional intake causes the person to experience worsening sarcopenia and weight loss. The involvement of the musculoskeletal system is central to this pathway, with loss in functional capacity accelerating the deterioration of other organ systems (Fried *et al.* 2001).

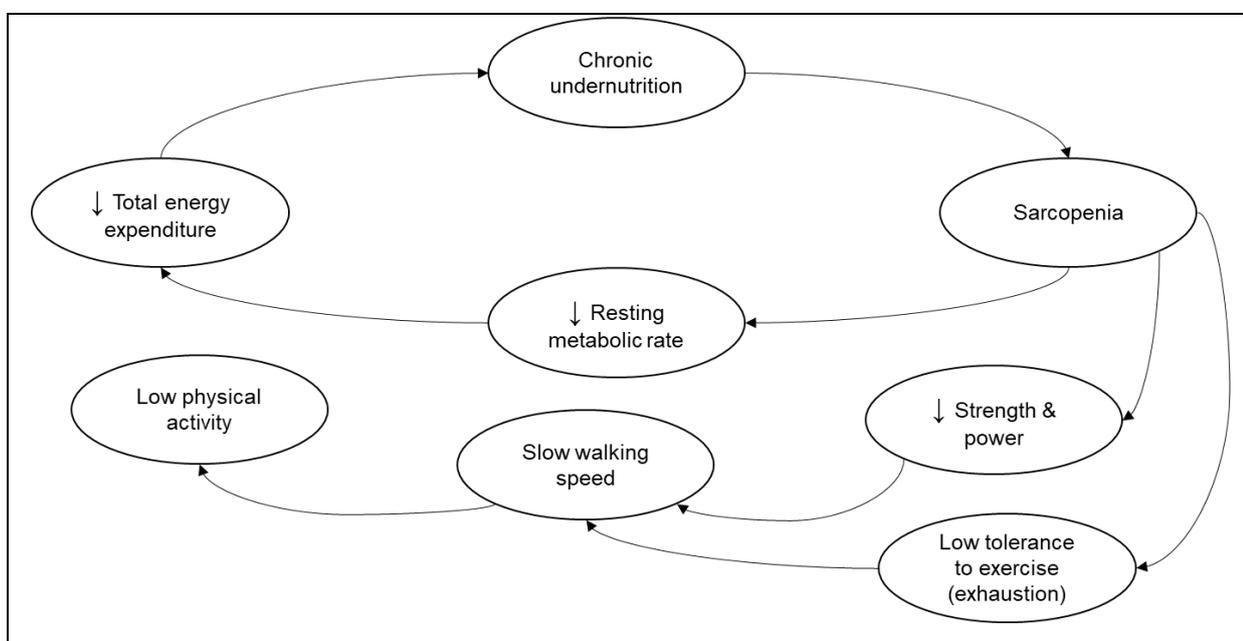


Figure 4.1 Cycle of Frailty, adapted from Fried *et al.* (2001)

Figure 4.1 shows the models underpinning hypothesis of key factors involved in the development of frailty, which combines attributes of body composition, nutrition, and mobility into an explanatory pathophysiologic phenotype. When these changes manifest as clinical signs and symptoms, such as weight loss and decreased energy level, they result in the development of a frail phenotype, which can be concretely measured (Makary *et al.* 2010, Fried *et al.* 2001). Based on the phenotype model, frailty status is determined by the presence of three out of five criteria including shrinking (weight loss, sarcopenia), weakness, exhaustion (poor endurance), slowness, and low activity. Having two of these criteria is indicative of a pre-frailty stage

whereby the individual is at a high risk of progressing to frailty. This phenotype scale was created with the aim of identifying at-risk individuals, thereby providing opportunities for appropriate and timely care interventions aiming to slow or reverse frailty. It has demonstrated capacity to predict adverse outcomes including falls, hospitalisations, disability, and death (Fried *et al.* 2001). The study by O'Connell *et al.* (2020), found that there was a greater prevalence of phenotypic frailty among older adults with ID in comparison to those in the general population, which was associated with excessive polypharmacy (use of ≥ 10 medications).

As it can be applied at first contact without the need for preliminary clinical evaluation, the phenotype is considered a clinical-friendly dichotomous variable useful for informing decisions on the need for adapted care or interventions (Cesari *et al.* 2014). However, a lack of specialist equipment, time and space may prevent the evaluation of gait speed and handgrip strength in a clinical setting (Cesari *et al.* 2014). Additionally, the scales use of sample-specific cut-off points to divide continuously measured variables into dichotomous criteria (e.g., slow gait speed and weak grip strength) may impair its precision (Wu *et al.* 2018). A known limitation of the phenotype scale which also has potential implications for this study is its limited capacity to measure frailty in long-disabled populations, such as people with ID, who oftentimes have lifelong and complex health needs. There is the potential that an individual may be inaccurately labelled as being physically frail due to presence of pre-existing health conditions or disabilities. Therefore, the measurement of frailty among people with ID according to phenotypic criteria may lead to flawed results (Brehmer-Rinderer *et al.* 2013).

4.3 Accumulation of Deficits

A multidimensional approach to conceptualising frailty is the accumulation of deficits model, operationalised as a frailty index (FI), which was developed from the Canadian Health and Aging Study (Rockwood *et al.* 2005). The underlying idea of the model is that accumulation of unspecified 'deficits' (i.e. health problems or abnormal characteristics) leads to a nonspecific, age-associated vulnerability, or frailty (Robinson *et al.* 2015, Mitnitski *et al.* 2001, Rockwood and Mitnitski 2007).

Mitnitski *et al.* (2001) describe deficit accumulation as:

“An example of a macroscopic variable, i.e., one that reflects general properties of aging at the level of the whole organism rather than any given functional deficiency.” (p. 323)

They assert that this provides a proxy measure of aging which reflects severity of illness and proximity to death. The FI can incorporate a diverse range of deficits across physical, psychological and social domains.

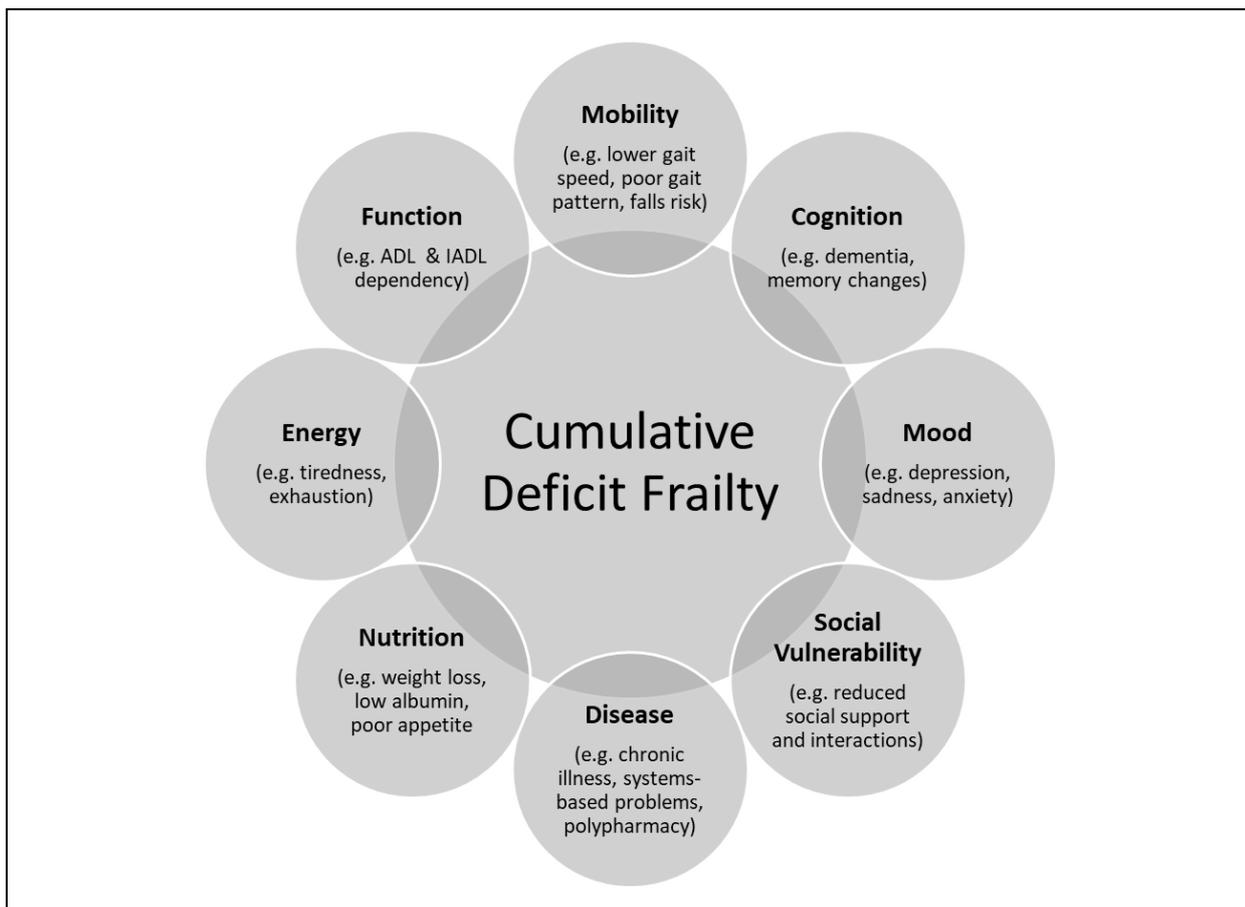


Figure. 4.2 Cumulative Deficit Model of Frailty, adapted from Robinson *et al.* (2015)

As shown in figure 4.2, deficits sustained over a person’s lifetime in relation to medical, social, or functional health can be considered using this model of frailty. Deficit accumulation can also be understood to occur at many levels, from the cellular level to tissues, organisms and complex systems (Lozupone *et al.* 2020). Of pertinent relevance to this dissertation, deficits can accumulate at the social level, pertaining to social environments and circumstances (Armstrong *et al.* 2015, Wallace *et al.* 2015, Shega *et al.* 2012, Andrew and Rockwood 2010, Andrew *et al.* 2008). The main

advantage of using the deficit accumulation model is that it does not include specific criteria, therefore it can be used to measure frailty in specific populations. This provides the opportunity to include issues prevalent in the ID population and utilize routinely collected data from diagnostic questionnaires that have been validated in this group. Despite its reproducibility, the FI may require more cumbersome and labour-intensive assessments which may be more time consuming and therefore uptake of this tool in routine clinical practice is reported to be limited (Kamaruzzaman *et al.* 2010). The development and validation of frailty indices as valid and reliable measures of frailty among older adults with ID has been reported by several studies (McKenzie *et al.* 2016b). It should be noted however that these studies have primarily focused on examining frailty multidimensionally, whereby frailty indices were constructed using items representing deficits across multiple domains. There are no known studies which have operationalised social frailty according to the accumulation of deficits, whereby a social frailty index comprising of only social-related deficits has been used to examine social frailty among older adults with ID.

4.4 Integral Conceptual Model of Frailty

The integral conceptual model of frailty by Gobbens *et al.* (2010a) adopts a lifespan approach to frailty. This model describes a pathway from life course determinants, to frailty, to adverse outcomes, including disability, healthcare utilisation and death. At each stage, interventions aimed at preventing or delaying frailty development can be considered. Transition to less frail clinical states, and even from being frail to non-frail, is possible. Within this framework, a dynamic inter-relationship between the physical, psychological and social domains of frailty is proposed. This model can be used to interpret the findings of the literature reviewed in the previous two chapters. Findings of the narrative review in Chapter 2 demonstrated that frailty status among older adults in the general population is influenced by age, gender, education attainment, employment, financial status, cognitive ability, functional ability, social relationships, loneliness, lifestyle behaviours and religiosity. Similarly, the systematic review in Chapter 3 provides evidence that several factors are associated with frailty transitions and trajectories among older adults with ID including age, presence of Down syndrome, type of residence, healthcare use and access, and functional ability. Based on the underlying hypothesis of the integral conceptual model, life course

determinants such as these may affect physiologic reserve, subsequently leading to frailty development and adverse outcomes (Figure 4.3).

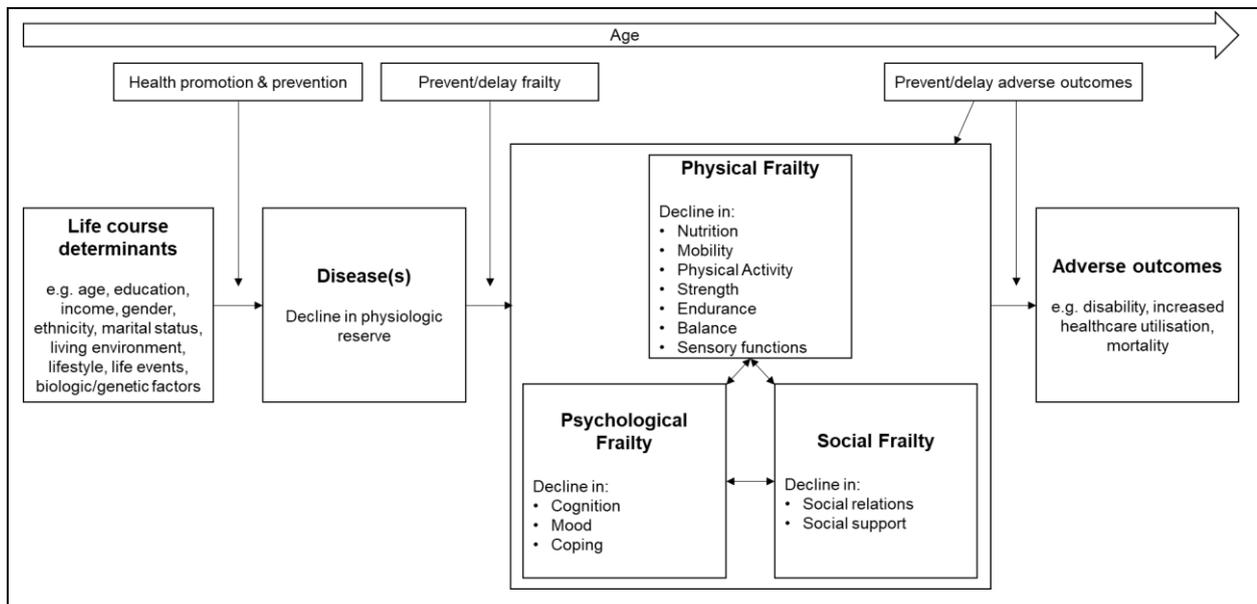


Figure 4.3 Integral Conceptual Model of Frailty, adapted from Gobbens *et al.* (2010a)

The integral conceptual model of frailty has been operationalized as the Tilburg Frailty Indicator (TFI) (Gobbens *et al.* 2010b) which is a self-report questionnaire comprising of two subscales. The first relates to determinants of frailty such as sociodemographic characteristics, life events, lifestyle, satisfaction with home living environment and presence of chronic disease(s). The second section involves the measurement of frailty in terms of its physical, psychological, and social domains. Physical frailty is represented by questions on physical health, unexplained weight loss, difficulty in walking, difficulty in maintaining balance, hearing problems, vision problems, lack of strength in hands, and physical tiredness. Cognition, coping, depression and anxiety symptoms were used to measure psychological frailty. Items in relation to lone living status, social relations and social support represented the social domain. All items are rated dichotomously on a theoretical scale of 0–1, with higher scores indicating a higher level of frailty. Scores for each frailty domain and a total frailty score are produced. The integral conceptual model of frailty has significant relevance to this study given its focus on the inter-relationship between social and physical domains of frailty. In this model, social frailty and physical frailty are shown to be the outcome of life course determinants and/or a decline in physiological reserve. Elements of the physical domain include a wide range of factors including phenotypic criteria and

decline in nutrition, endurance, balance, and sensory functions. The domain of social frailty is presented here as a decline in social relations and supports. The model provides an opportunity to consider how both domains may contribute to each other and to adverse outcomes, such as mortality. Based on findings of studies involving older adults in the general population, there is evidence to suggest that physical frailty and social frailty overlap to a degree, but individually contribute to mortality (Park *et al.* 2019, Andrew *et al.* 2008). While these associations have not been confirmed among older adults with ID, it is plausible to theorise that social frailty and physical frailty may also have a strong inter-relationship in this population, particularly in consideration of this groups unique exposure to social and physical health inequalities (McCarron *et al.* 2014, Lunskey *et al.* 2013, Emerson *et al.* 2012, McCarron *et al.* 2011).

4.5 Conceptual Model of Social Frailty

Social frailty has been described in the realm of the overall frailty concept (Gobbens *et al.* 2010a). However, this remains a largely unexplored concept (Bunt *et al.* 2017, Levers *et al.* 2006). Social frailty recognises that social circumstances may contribute to health outcomes of frail older adults and affect their access to appropriate interventions and supports (Andrew *et al.* 2018). In this context, frailty is not a fixed state defined by the presence of physiological impairment, but rather an outcome of the relationship between the individual and their environment (Barrett 2006). However, most of the literature is still inconclusive on the nature and scope of social frailty as a concept (Bessa *et al.* 2018) and approaches to its measurement remain contentious (Bunt *et al.* 2017). Furthermore, it has been asserted that the lack of theoretical frameworks to guide the conceptualization of social frailty has adversely affected this field of research (Pek 2020). In their seminal study, Bunt *et al.* (2017) evaluated existing insights on social frailty, and structure and synthesize these insights in a scoping review using the social needs concept of Social Production Functions (SPF) (Lindenberg 2013, Ormel *et al.* 1999, Steverink and Lindenberg 2006). SPF theory was first introduced by Lindenberg (Lindenberg 1986, Lindenberg 1991, Lindenberg and Frey 1993). It proposes that social wellbeing is dependent upon the fulfilment of needs for affection, behavioural confirmation and status (Ormel *et al.* 1999). Based on this theory, people produce their own well-being by trying to optimize achievement of universal goals, within the set of resources and constraints they face. SPF theory identifies two ultimate goals that all humans seek to optimize (i.e. physical well-being

and social well-being) and five instrumental goals by which they are achieved (i.e. stimulation, comfort, status, behavioural confirmation, affection). Ormel *et al.* (1999) assert that SPF theory guides research measurement and explanatory models, and it integrates features of contemporary subjective well-being theories. Based on SPF theory, the conceptual model by Bunt *et al.* (2017) (Figure 4.4) describes social frailty as:

“A continuum of being at risk of losing, or having lost, social resources, general resources, and social activities or abilities that are important for fulfilling one or more basic social needs during the life span.” (p. 326)

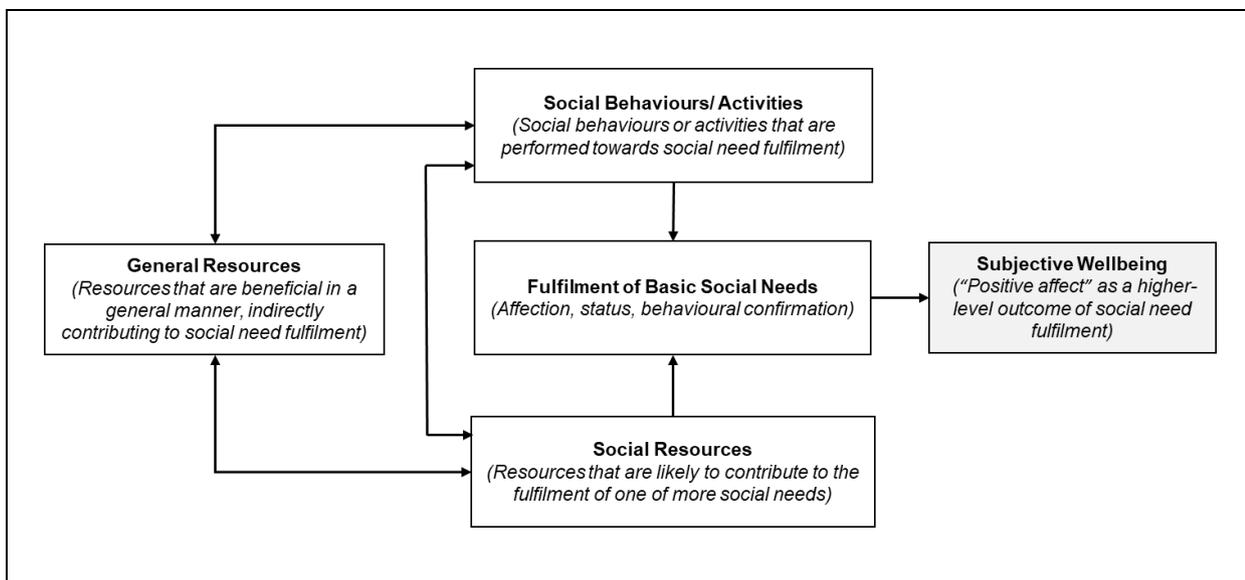


Figure 4.4 Conceptual Model of Social Frailty, adapted from Bunt *et al.* (2017)

The intended aim of the study by Bunt *et al.* (2017) was to develop an integrated conceptualisation of social frailty that can be used to identify research opportunities and inform future practice and policy. Indeed, this model has been adopted by several contemporary studies to obtain an understanding of the dynamics of social frailty among older adults. For instance, the studies by Pek *et al.* (2020) and Nagai *et al.* (2020) used this model to guide the construction of social frailty questionnaires, or scales, to assess social frailty status and predict future incidents of activity limitation. While this model identifies key concepts associated with social frailty including social isolation, loneliness, social networks, social supports, and social participation are identified as relevant to this conceptualisation of frailty, no overarching theoretical framework of social frailty is proposed. Additionally, the authors of the model

acknowledge that factors included in the model, while relevant to understanding the concept of social frailty, were selected without considering their relative weight or abstraction level. Therefore, their relative importance in the context of the overall conceptual model was not considered. There is significant potential for this conceptualisation of social frailty to help guide the identification of factors that should be incorporated in the measurement of this health state. While this conceptualisation of frailty has not been previously operationalised among older adults with ID, it may provide an enhanced understanding of how issues relating to social resources, general resources, and social activities or abilities may lead to a state of social frailty in this population. Furthermore, the ability to identify individuals 'at risk' of being socially frail on the basis of having deficits in relation to these domains may have significant implications from a practice and policy perspective. In practical terms, these insights may be useful for the development of upstream and tailored interventions aimed at preventing or delaying social frailty and associated adverse outcomes (Bunt *et al.* 2017).

4.6 A Synthesised Model of Frailty

The models described in previous sections have been central to frailty research internationally, and as such can be considered as being fundamental to our current understanding of this concept. However their differences in describing frailty perhaps speaks to the lack of consensus regarding its specific nature. In the absence of an overarching theoretical framework, a synthesised model (Figure 4.5) was created which combines key components of models developed by Gobbens *et al.* (2010a), Bunt *et al.* (2017), Mitnitski *et al.* (2001) and Fried *et al.* (2001). As in the integral conceptual model of frailty by Gobbens *et al.* (2010a), a lifespan approach is adopted which recognises the role of life course determinants and aging as foundational to overall frailty development.

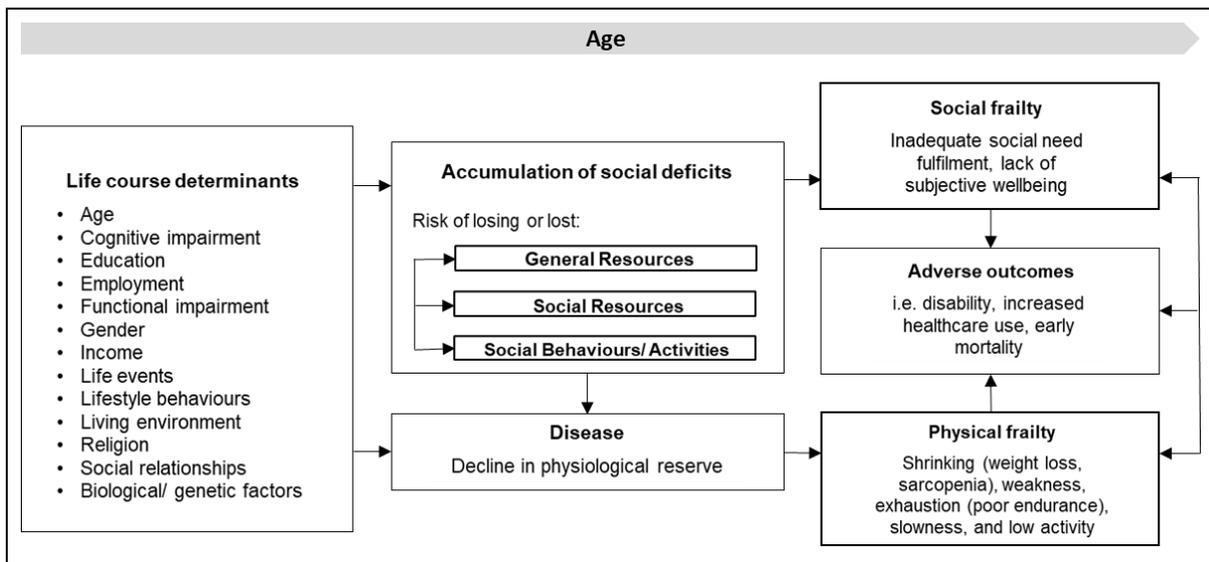


Figure 4.5 Synthesised model of frailty

A key difference in the frailty pathway described in the synthesised model is that these life course determinants not only affect physiological reserve over time leading to phenotypic frailty (Fried *et al.* 2001), but also affect the accumulation of deficits (Mitnitski *et al.* 2001) in relation to general resources, social resources, and social activities and behaviours. In line with the conceptual model of social frailty by Bunt *et al.* (2017), this results in inadequate social need fulfilment and ultimately, lack of subjective wellbeing. It is theorised that the accumulation of these social deficits may also contribute to reduced physiological reserve, compounding the view that there is an implicit relationship between social and physical domains of frailty. Lastly, it is posited that social frailty and physical frailty individually and combinatively contribute towards an increased risk of experiencing adverse health outcomes.

4.7 Conclusion

This chapter presented the theoretical framework being used to explore key concepts underpinning this study and proposes their relationship in the context of current research evidence. Selected conceptual models of frailty developed for clinical and research use were individually defined and discussed in terms of their typical application, limitations and potential relevance to addressing the aim and objectives of this study. Firstly, the phenotype model, which defines frailty in terms of biomedical factors, provides a means of measuring the physical domain of frailty, albeit with the potential risk of misidentifying an individual with intellectual disability as being physically frail due to a pre-existing disability. The integral conceptual model of

developed by Gobbens *et al.* (2010) is useful towards understanding how life course determinants may lead to frailty development, and consequently adverse outcomes such as mortality. The deficit accumulation model (Mitnitski *et al.* 2001) offers a multidimensional perspective which views frailty as non-specific, age-associated vulnerability. This approach to measuring frailty has been validated for use among older adults with ID and holds significant promise as a means of operationalising social frailty in this population. As shown in other studies, it is possible to construct a social frailty index (SFI) comprising of social-related variables (Armstrong *et al.* 2015, Wallace *et al.* 2015, Shega *et al.* 2012, Andrew and Rockwood 2010, Andrew *et al.* 2008). The conceptual model by Bunt *et al.* (2017) provides a useful guide for the selection of candidate index items so that deficits in relation to social resources, general resources, and social activities or abilities required for social need fulfilment are represented. The synthesised model developed integrates these different conceptualisations within a single theoretical construct to guide the development of this studies methods and support the interpretation of findings.

Chapter 5: Methods

5.1. Introduction

As discussed in Chapter 1, the primary aim of this study is to obtain a holistic understanding of the characteristics of social frailty among older adults with ID. To address this aim, key study objectives include construction and validation of a measurement of social frailty, exploring the relationship between social frailty and physical frailty, and analyzing the capacity of social frailty to predict mortality. In this chapter, methodological approaches used by the researcher to address the studies aim and objectives are comprehensively described. Firstly, the design and procedures of this study including its data source, sampling procedures, sample profile and ethical considerations are explained. An in-depth description of measures used to operationalise social frailty and physical frailty in the study sample are then provided. This is followed by an explanation of statistical procedures undertaken as part of this study. Finally, the layout of the results chapter of this dissertation is outlined.

5.2 Study Design

To obtain a holistic understanding of the characteristics of social frailty among older adults with ID, this study has primarily utilised a longitudinal design. This approach involves the collection of data from individuals within a pre-defined group and enables statistical testing to analyse changes over time among individuals or in the group as a whole (Van Belle 2004). A longitudinal design is required to understand how developmental and aging-related changes occur as they allow the direct assessment of within-person change over time (Salkind 2010). The decision to utilise a longitudinal design requires consideration of numerous factors in relation to the statistical testing of data. As described by Edwards (2000) these include (I) the linked nature of the data for an individual, despite separation in time; (II) the co-existence of fixed and dynamic variables; (III) potential for differences in time intervals between data instances, and (IV) the likely presence of missing data. The strategies employed by this study to address these factors are expounded upon in later sections.

5.3 Data Source

The primary data source for this study is IDS-TILDA, which has a nationally representative sample of persons at all levels of ID, and the full range of residential circumstances. IDS-TILDA runs parallel to the Irish Longitudinal Study on Ageing (TILDA), which is an observational study of community-dwelling older adults in Ireland which began in 2006 (Kenny *et al.* 2010). IDS-TILDA is the first longitudinal study of ageing in Europe capable of directly comparing the key determinations of health and well-being of people with ID to the general population (McCarron *et al.* 2017). The ageing of people with ID is not represented in TILDA data, which led to the inception of IDS-TILDA. In contrast to the main TILDA study, which includes people aged 50 years and above, IDS-TILDA includes people aged 40 years and above in recognition of the earlier onset of age associated conditions in the ID population (Doyle *et al.* 2021, O’Leary *et al.* 2018, Lifshitz and Merrick 2004). The main rationale for selecting IDS-TILDA as the primary data source is the studies broad conceptual framework (Figure 5.1), which explores the health, social, economic, and environmental circumstances of older adults with ID as they age (McCarron *et al.* 2017).

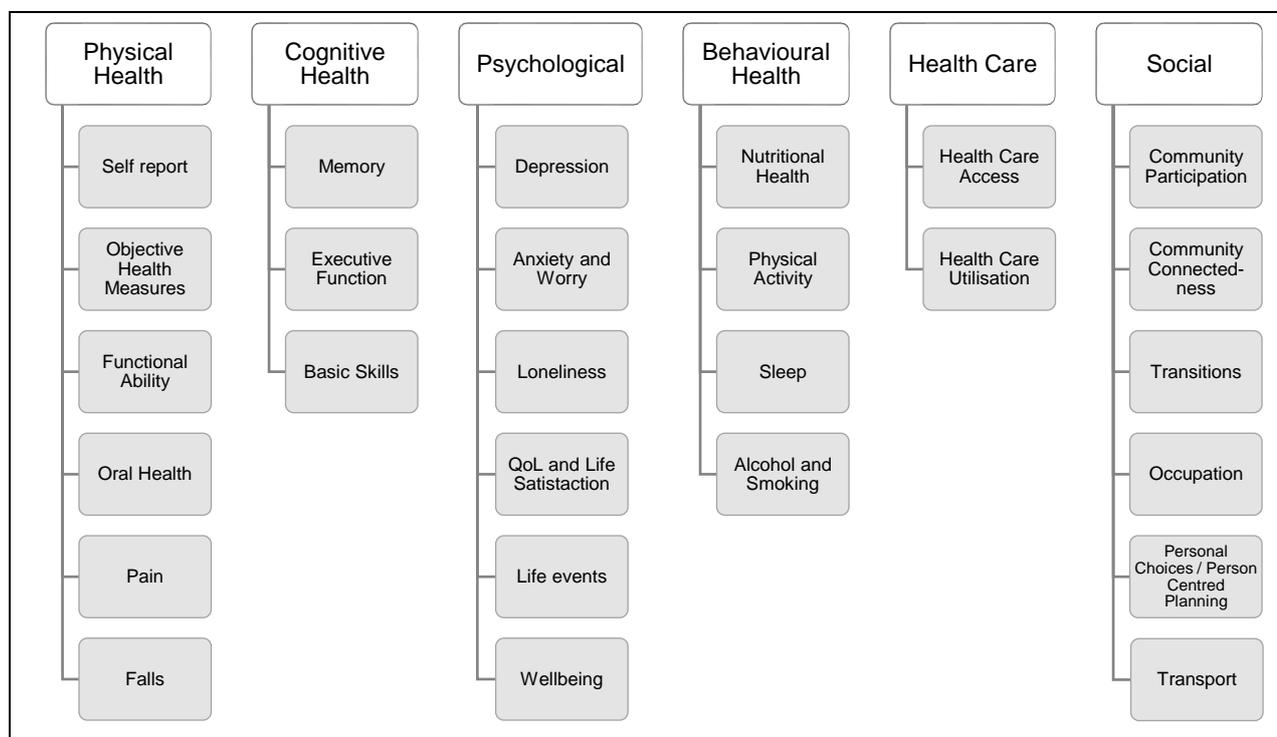


Figure 5.1 IDS TILDA Conceptual Framework

To date, four waves of IDS-TILDA have been completed (McCarron *et al.* 2021, McCarron *et al.* 2017, McCarron *et al.* 2014, McCarron *et al.* 2011), with each wave equating to a three-year data period between data collection. Participants were supported to take part in all components of the study by a person who knows them well (minimum of six months) and by use of proxy respondent to answer on their behalf where required (McCarron *et al.* 2022).

5.4 Data Collection

Data utilized in this study has been obtained from the studies primary data source, IDS-TILDA, as described in section 5.3. Measures used in IDS-TILDA are designed to be comparable to measures of the general older population by TILDA. Some reasonable adjustments were made to accommodate needs specific to the intellectual disability population. This included changes to wording and the use of alternative measures. However, the measures used in IDS-TILDA generally reflect those incorporated within the data collection components of the main TILDA study. Components of the IDS-TILDA data collection protocol utilised by this study include the Pre-Interview Questionnaire (PIQ), Computer Assisted Personal Interview (CAPI), and objective health assessment (referred to as Health Fair).

5.4.1 Pre-Interview Questionnaire

The PIQ questionnaire includes a range of questions covering demographics, health status, healthcare utilisation and medicine usage with some additional detail such as gathering more complete data on dosage and frequency of medications (McCarron *et al.* 2014).

5.4.2 Computer Assisted Personal Interview

CAPI includes detailed questions regarding socio-demographic characteristics, physical health, behavioural health, mental and cognitive health, social participation and social connectedness, health care utilisation, employment, income and life-long learning (McCarron *et al.* 2014).

5.4.3 Health Fair

The Health Fair component was introduced in Wave 2 of IDS-TILDA and involved a series of objective health measures including Timed Up and Go (TUG), grip strength, blood pressure, waist and hip measurement, height, weight, and quantitative heel ultrasound (Burke *et al.* 2020). After being rested in Wave 3, this component was again administered in Wave 4, with new measures introduced including blood drop sample, hair sample, calf circumference, chair to stand assessment, oral health assessment, nutrition assessment, cognitive assessment, foot health assessment, 2-minute step, balance, activPal and pulse wave analysis. The researcher was involved in the piloting and administration of the Wave 4 Health Fair, including on average 2-3 days per week of field work between November 2019 and March 2020. Additionally, the researcher was involved in the entry of Health Fair data (N= 115) into the IDS-TILDA dataset.

5.4.4 Impact of COVID-19

With the onset of the COVID-19 pandemic and the associated lockdown in mid-March 2020, all data collection for Wave 4 of IDS-TILDA, which was ongoing at that time, was suspended. However, ethical approval was obtained in May 2020 to complete Wave 4 data collection by remote interviewing (by phone/video) rather than face-to-face and for the addition of a supplemental COVID-19 survey to investigate the impact of the pandemic and lockdown on the IDS-TILDA participants (McCarron *et al.* 2021).

5.5 Study Sample

The sample for IDS-TILDA, and therefore also that of this study, was drawn randomly from the Irish population of adults aged 40 years and above registered on the National Intellectual Disability Database (NIDD). The NIDD is an administrative database managed by the Health Research Board (HRB) and contains information on people with an intellectual disability in the Republic of Ireland who are registered with a service provider (Kelly *et al.* 2011). For recruitment of the original sample at Wave 1 (McCarron *et al.* 2011), the NIDD dataset contained 26,066 individuals (Kelly *et al.* 2011). From this, the inclusion criterion of being aged 40 years and above was applied and 1,800 individuals were randomly selected by NIDD staff, of which 1,600 were provided information and asked to participate in the study. An invitation pack was then sent to each potential participant with a consent form. Written consent was obtained from 753 individuals (46% response rate), either directly by self-consenting individuals (38%) or by a family member or guardian for those who were unable to self-consent (62%).

Wave 1 of IDS-TILDA, which began in 2007, had a sample population of 753 participants. Wave 2 of IDS-TILDA data collection commenced in 2011 and included participants from Wave 1 who completed at least one component of Wave 2 data collection (PIQ, CAPI or health Fair) (N= 708, 94%). In total, 45 participants from Wave 1 were lost to follow-up in Wave 2 due to death (N= 34) or refusal to participate (n= 11).

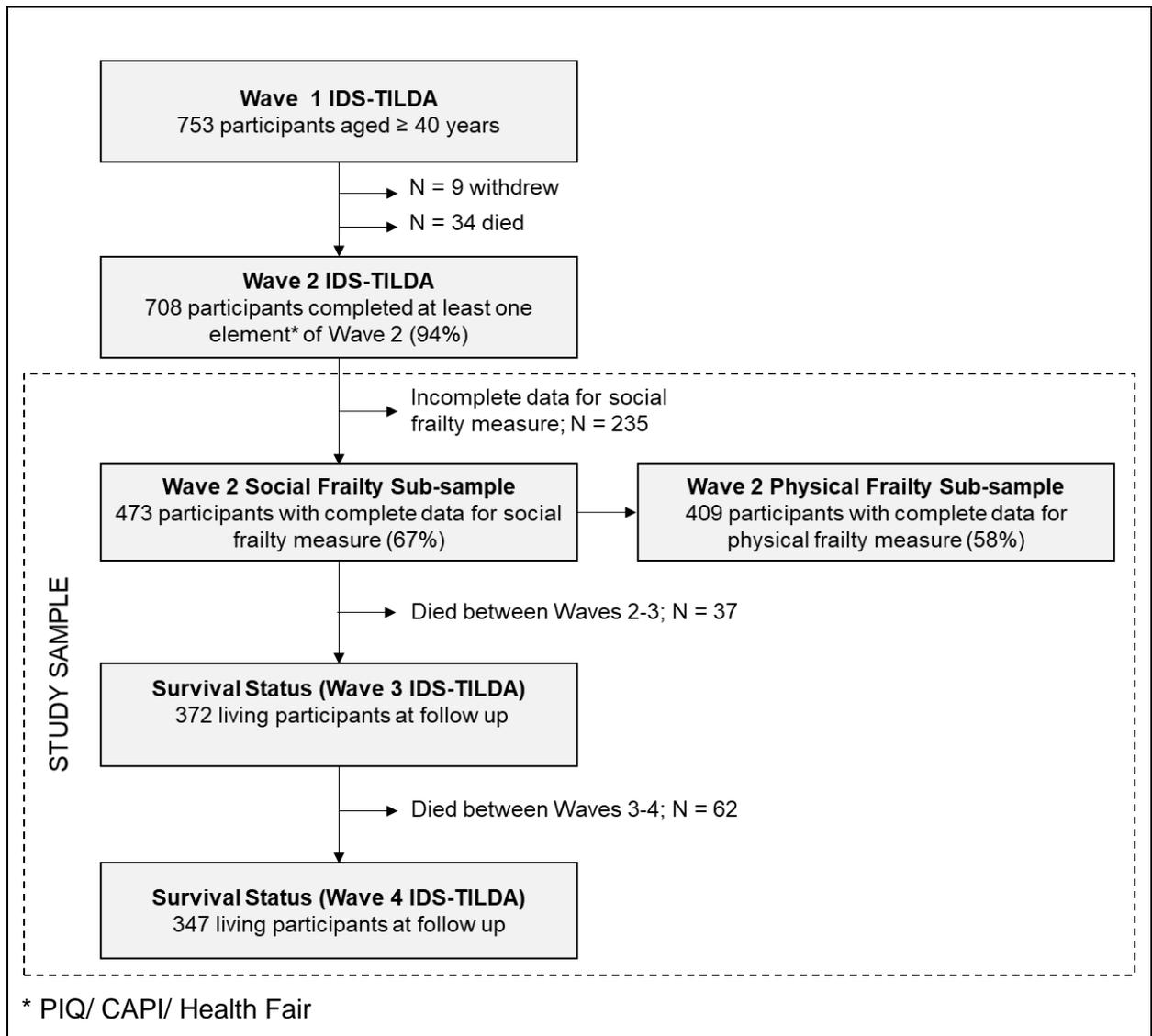


Figure 5.2 Flow chart of sample selection

Questions included in the CAPI and PIQ administered at each wave of the study were screened in relation to their potential suitability as a variable in the social frailty measure developed for this study (see Section 5.7.1). It was found that Wave 2 comprised of the highest number of these items, and on this basis the decision was taken to use Wave 2 of IDS-TILDA as the baseline for this study. While the IDS-TILDA sample was not refreshed in Wave 2, it remained largely representative of the NIDD. 56% of participants were female and 44% were male, with an average age of 56.6 years. Levels of intellectual disability among participants based on ICD-10 criteria (WHO 1993) (see Section 1.5.1) were categorised as mild (22%), moderate (43%), severe/profound (27%) and unknown/unverified (8%). A social frailty sub-sample of IDS-TILDA Wave 2 participants was obtained by including participants with complete data for all variables of 56 variables included in a candidate social frailty index (see Section 5.7.1). Of the total sample for Wave 2 of IDS-TILDA (N=708), 473 participants (66%) provided sufficient data to qualify for inclusion in this study. Mortality rates among participants in the social sub-sample approximately three and six years after baseline was obtained using data collected in Waves 3 and 4 of IDS-TILDA respectively. By the first follow-up interval (Wave 3), 8% (N=37) of participants in the cohort had died. A further 62 participants (13%) died between Wave 3-4 of IDS-TILDA. Figure 5.2 provides an overview of the studies sample selection process.

5.6 Ethical Considerations

5.6.1 Ethical Approval

Ethical approval for all four waves of IDS-TILDA has been granted by the Trinity College Dublin Faculty of Health Sciences Research Ethics Committee. Ethical approval was then obtained from individual service providers supporting the study participant. IDS-TILDA complies with the requirements of the Data Protection Act (DPA) (1988), DPA amendment (2003), General Data Protection Regulations (GDPR) (2018), and Health Research Regulations (HRR) (2018). In accordance with HRR, IDS-TILDA made an application in July 2019 to the Health Research Consent Declaration Committee (HRCDC) for a Consent Declaration in order to continue data processing from previous waves which had been gathered through proxy respondents and to proceed with data gathering for Wave 4 of IDS-TILDA. The Health Research Consent Declaration Committee (HRCDC) granted a full Consent Declaration for the

study in December 2019, facilitating the inclusion of proxy-consented participants. The current study has been granted permission to access data from Waves 1-4 of IDS-TILDA by the Principal Investigators of the study. In advance of data collection, the researcher undertook GDPR training (Research Integrity and Impact in an Open Scholarship Era) (5 ECTS - NCQ Level 9). In adherence with Article 35 of the GDPR, the researcher assisted the IDS-TILDA project team in the completion of Data Protection Impact Assessments (DPIA) to assess the risks posed by this study and to identify suitable controls that minimize these risks. This study aims to comply with local college policy on research practices as required by the Trinity College Dublin Faculty of Health Sciences Research Ethics Committee. Conventions and declarations devised to protect human participants of research, such as the Nuremberg Code (1947), the European Convention on Human Rights (Frowein 1950) and the Declaration of Helsinki (World Medical Association 2013) are recognised.

5.6.2 Research Principles

As per research guidelines provided by the Nurse and Midwifery Board of Ireland (2007), this study adheres to the principles of autonomy, beneficence, non-maleficence and justice (Beauchamp and Childress 2001). The following sections serve to demonstrate how this study has been completed in line with research guidelines provided by the Nurse and Midwifery Board of Ireland (2007) to realise the principles of autonomy, beneficence, non-maleficence and justice (Beauchamp and Childress 2013).

5.6.2.1 Autonomy

Researchers have an ethical obligation to provide accurate and accessible information to participants that will allow them to make an informed decision about participating in a study (Striefel 2001). Participants were supported to make an independent decision on participating in the IDS-TILDA study. An information sharing process was used to obtain the informed consent of study participants. This involved explaining all aspects and proceedings of the study using accessible information. The researcher supported the development of the invitation cover letter (Appendix 5), accessible information booklet (Appendix 6) for the Health Fair component of Wave 4 of IDS-TILDA.

Process consent was obtained by inviting participants to reaffirm their consent at all stages of the study without force or coercion. Participants were informed that they could withdraw consent at any time. Participants and their carers were provided with the opportunity to ask questions. Many people with ID find formal test centres and unfamiliar environments stressful, hence the interviews and health assessments were held in a private location of their choosing, usually at the person's own home or care setting. Scheduling of health assessments and interviews was completed in line with the availability of participants. The researcher was involved in the development of an accessible appointment card (Appendix 7) sent to participants in the Health Fair component of Wave 4 of IDS-TILDA.

5.6.2.2 Beneficence and Non-maleficence

These complementary principles require that research is conducted in a manner that maximises benefits and minimizes risks for participants (Gostin 1991). Research should only be performed by persons with the required level of training and competence (WHO 2011). Objective health measures utilized in this study were administered by field researchers qualified as registered nurses, including the researcher. Eligibility criteria for each element of the health assessment was developed and implemented to safeguard participants. All field researchers were experienced in working with people with ID and received training in sensitive interviewing strategies. An experienced researcher shadowed all new researchers for at least one interview. As the CAPI includes questions on areas such as family, finances, social isolation and quality of life, there is some possibility that the participants may become distressed or express difficulties coping with their circumstances. However, similar questions have been used in other studies, such as the Survey on Lifestyle and Attitudes to Nutrition (Morgan *et al.* 2008) and the English Longitudinal Study of Ageing (Stephoe *et al.* 2013), with limited instances of distress reported. If participants became distressed, field researchers were trained to use redirection techniques and offer to reschedule or stop the interview. Participants were directed to sources of additional support and information such as helplines and websites. A consultation process was undertaken with independent advocates and advocacy groups to review the data protection protocol and help identify components / questions which may cause distress so that possible actions would be determined.

Participants were informed that there were no direct benefits from participating in the study but that they were contributing to the building of empirical evidence on what it is like for people with ID to grow older. However, many participants appeared to enjoy meeting the field researchers and having the opportunity to talk about their health. Health assessment results that were immediately available were shared with participants. This information has the potential to benefit participants by prompting them to adopt a healthier lifestyle, visit their GP or attend health screening. The potential laboriousness of data collection activities, particularly the health assessment, was a key consideration. Efforts were made to reduce the degree of participant burden by time testing all components of the health assessment during the piloting phase. Any component of the health assessment identified as being burdensome due to completion time or difficulty level were reviewed for inclusion. Participants were offered regular breaks during assessments and interviews. To help participants feel comfortable, interviews took place in a location of their choosing, most often in their own home. Where possible, health assessments were administered in a location nearby to the participant to reduce travel distance. IDS-TILDA is guided by a Values Framework that emphasises inclusion and empowerment of people with ID with the ultimate aim to contribute to improving their lives. All aspects of the design, development, delivery, dissemination and governance of IDS-TILDA has involved people with ID, their families, and carers.

5.6.2.3 Justice

Justice involves treating people with respect and without discrimination (Thompson 1987). IDS-TILDA adhered to this ethical principle by ensuring that participants were treated fairly and with impartiality during all stages of the study. The human and citizen rights of participants were respected. Risk of selection bias was eliminated by utilizing random sampling to recruit participants.

5.6.3 Consent

The Assisted Decision-Making (Capacity) Act 2015 was signed into law in Ireland in 2015. This legislation sets out a system of supports for adults who have difficulties with decision-making capacity. The Capacity Act states that everyone is presumed to be able to decide for themselves unless the opposite is shown. However, consent is a complex issue in the field of ID research. Having ID can make a person particularly vulnerable to being misrepresented and misunderstood, leading to superficial consent

discussions and uninformed consent (Biros 2018). This study recognises that valid consent must be informed consent, where the person has enough information to be able to understand what is proposed and the potential risks and benefits. Informed consent involves disclosure of information, comprehension, competency and voluntariness (Beauchamp and Childress 2013). Further details on how the researcher specifically addressed these aspects of informed consent are provided in the following sections.

5.6.3.1 Disclosure of Information

A recruitment pack mailed to participants included an accessible information booklet, cover letter and consent form. Consent forms were completed by participants and/or family members/guardians and returned using stamped addressed envelopes included in the recruitment packs. Study participants were contacted a minimum of one month and a maximum of three months prior to the commencement of each study wave. Informing study participants of research results is often neglected (Iacono 2006). A comprehensive keeping-in-touch strategy is in place to communicate project activities and study findings to participants through the mediums of newsletters, social media and the IDS-TILDA website. Findings immediately available from individual health assessments were disseminated to participants.

5.6.3.2 Comprehension

There can be uncertainty regarding the capacity of people living in congregated or institutional settings to provide informed consent (Dalton and McVilly 2004). IDS-TILDA participants were provided with accessible information which explained how the study would be conducted and how data collected would be used.

5.6.3.3 Competency

For each wave of data collection, an accessible consent form and information booklet were sent to IDS-TILDA participants prior to interview. On the day of interview, informed written consent was obtained from each participant or support person. Competence to consent may vary across time and across tasks (Beauchamp and Childress 2001). Therefore, process consent was obtained prior to commencing each step of the health assessment and interview. Process consent involves the researcher keeping participants informed of any potential dangers, allowing the researcher and participant to make research decisions as a team (Usher and Arthur 1998). Permission

to proceed with the health assessment and interview was reiterated between each section or after each assessment.

5.6.3.4 Voluntariness

The voluntary nature of the consent giving process requires an equal and independent relationship between participants and researchers (Griffin and Balandin 2004). This was demonstrated in this study by efforts taken to ensure a balanced researcher-participant relationship. No researchers had contact with participants outside of the research process. Participants were given the option to withdraw consent verbally or in writing at any time. If a participant chose to withdraw their consent, they would have been removed from all mailing lists and no further attempts would have been made to contact them. Their existing data will remain in the data set and in use under Article 89 of GDPR – i.e., the data will remain in the dataset but will continue to be pseudonymised and subject to the same safeguards and security as other data.

5.6.4 Confidentiality

The privacy of study participants is protected under GDPR (European Parliament 2016). Participant confidentiality was respected at all stages by using a privacy by design approach. Understanding that personal information obtained by the researcher must not lead to the identification of research participants and should not be made available to others without their consent (Polit and Beck 2004), participants were assigned Personal Identification Numbers (PINs) during the sample recruitment process to anonymize data and protect their identity. At no time during the selection or recruitment stage were the contact details of the selected PINs or the list of PINs selected released. The original contact details of other parties involved, for example key workers and family members, were deleted. A user agreement must be completed to access IDS-TILDA data. This data can only be analysed within the IDS-TILDA offices and cannot be taken off site. Any hard copies of participant information, including signed consent forms, are stored in a locked filing cabinet in a locked storage office.

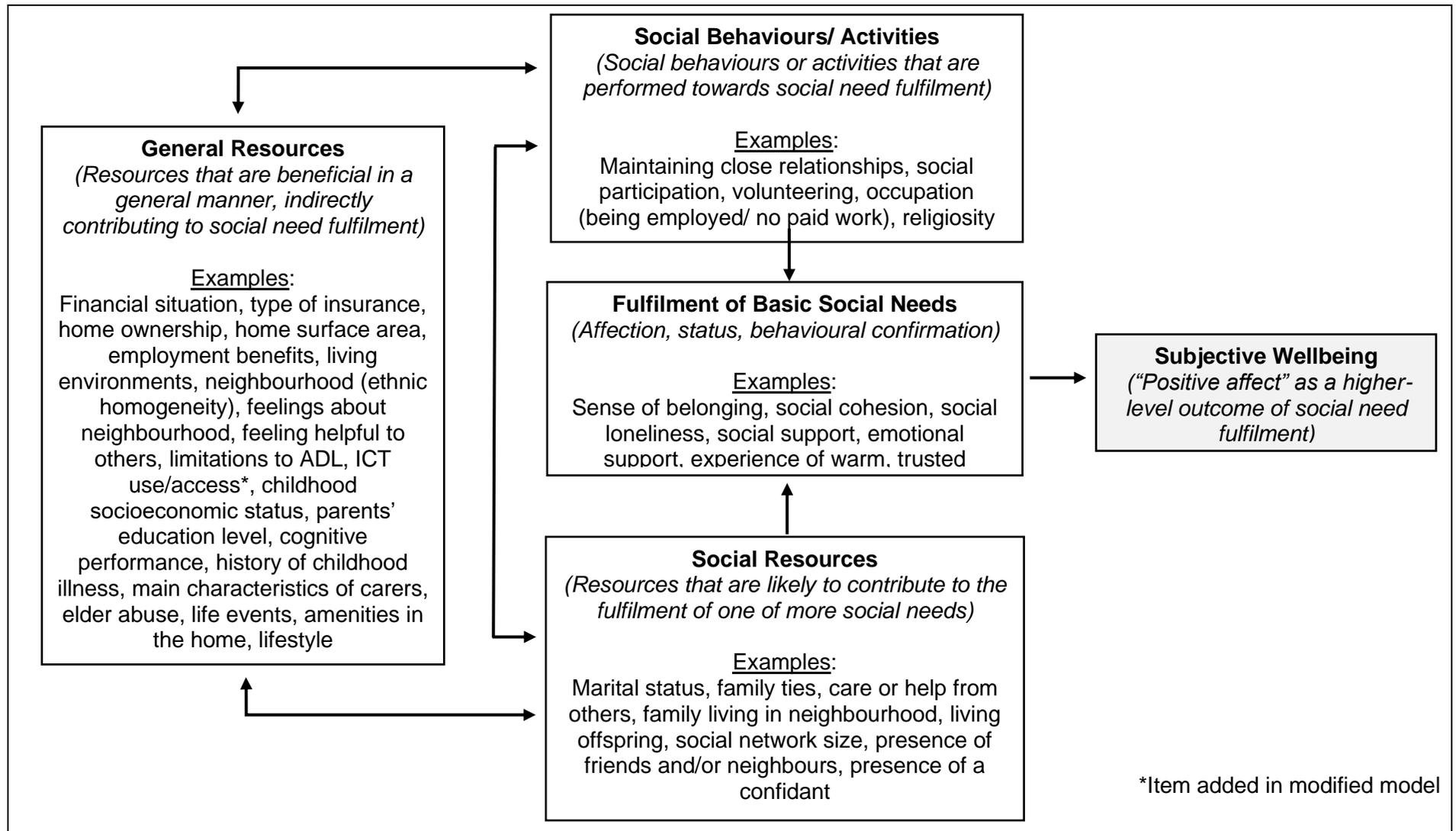


Fig 5.3 Modified Conceptual Model of Social Frailty, adapted from Bunt *et al.* (2017)

5.7 Measures

5.7.1 Social Frailty

Social frailty was operationalised according to a deficit accumulation approach to construct a Social Frailty Index (SFI). Self-report variables that could be considered as social-related health deficits collected in the PIQ and CAPI in Wave 2 of IDS-TILDA were selected for inclusion in the index. The selection of candidate deficits was informed by a theory-guided approach based upon a modified conceptual model of social frailty (Figure 5.3), adapted from Bunt *et al.* (2017). Further to points discussed in Section 1.5.3 regarding social determinants of health, this modified model adopts a broader interpretation of general resources to include variables relating to the use of and access to information and communication technology (ICT). The main rationale for this modification is that ICT use and access has been found to be important to people with ID in terms of supporting their involvement in society (Werner and Shpigelman 2019, Chiner *et al.* 2017).

Following the screening process, a total of 56 variables from IDS-TILDA Wave 4 were identified as candidate items for the SFI (Table 5.1). As many of those included were binary. Firstly, negative wording or scoring among variables were reversed. For each item, a binary score of 0 was assigned if a deficit was absent and 1 if it was present. Intermediate values were applied in equal increments in cases of ordered response categories. For example, an item which had three response categories, such as “How often do you feel lonely?”, received a score of 1 if a deficit was present (e.g. “most of the time”), 0.5 for an intermediate deficit (e.g. “some of the time”) and 0 if no deficit was present (e.g. “hardly ever/never”). Non-response related values (e.g. refused, did not know, no response) were treated as user-missing data. The SFI score was defined as the ratio between existing deficits and the number of evaluated deficits. In other words, SFI scores for each individual participant was calculated by dividing the sum of deficit scores by the total number of deficits considered. This yielded an index with a theoretical range of 0-1. There are no established cut-off points for SFI. In line with the approach utilised by Andrew *et al.* (2008), high SFI scores represented high levels of social frailty.

Table 5.1 Frequencies and Descriptive Statistics for Candidate SFI items (N= 56)

#	Item	Variable	Present (N)	Missing (N)	Missing (%)	Min. Value	Max. Value
1.	Unemployed or looking for work	OC_205	699	9	1.27	0	1
2.	I don't have any family	SC_109	699	9	1.27	0	1
3.	Not a member of any organisation, club or society	SP_311	698	10	1.41	0	1
4.	Single relationship status	B_Marital Status	697	11	1.55	0	1
5.	Do you have any difficulty speaking or making yourself understood when speaking?	PH_31	695	13	1.83	0	1
6.	Level of difficulty with dressing, including putting on shoes and socks	FL_24	694	14	1.97	0	1
7.	Level of difficulty with walking across a room.	FL_29	694	14	1.97	0	1
8.	Level of difficulty with eating	FL_46	694	14	1.97	0	1
9.	Level of difficulty with getting in or out of bed	FL_51	694	14	1.97	0	1
10.	Level of difficulty using the toilet, including getting up or down	FL_56	694	14	1.97	0	1
11.	Doesn't take part in regular physical activity	BH_2101	694	14	1.97	0	1
12.	Level of difficulty with bathing or showering.	FL_38	693	15	2.11	0	1
13.	Change of staff in my home where I live or day service I attend	MH_802	692	16	2.25	0	1
14.	New resident moved into my home	MH_803	692	16	2.25	0	1
15.	Change of my key worker	MH_804	692	16	2.25	0	1
16.	Change at or from work or day service	MH_805	692	16	2.25	0	1
17.	Death of a parent	MH_806	692	16	2.25	0	1
18.	Death of a sibling	MH_807	692	16	2.25	0	1
19.	Death of other relative	MH_808	692	16	2.25	0	1
20.	Death of a friend	MH_809	692	16	2.25	0	1
21.	Death of a pet	MH_810	692	16	2.25	0	1
22.	Major illness of a relative, caregiver or friend	MH_811	692	16	2.25	0	1
23.	Death of a significant other (other than a relative or friend)	MH_812	692	16	2.25	0	1
24.	Moving within service organisation	MH_813	692	16	2.25	0	1

#	Item	Variable	Present (N)	Missing (N)	Missing (%)	Min. Value	Max. Value
25.	Moving from my family home to a service supported home	MH_814	692	16	2.25	0	1
26.	Change in frequency of visits from or to family/friend	MH_815	692	16	2.25	0	1
27.	Major illness or injury	MH_816	692	16	2.25	0	1
28.	Break up of a steady relationship/ Divorce	MH_817	692	16	2.25	0	1
29.	Experience of crime (mugged or burgled)	MH_818	692	16	2.25	0	1
30.	Problems with justice and/or authorities*	MH_819	692	16	2.25	0	0
31.	Has access to a computer/ laptop/ tablet/ smartphone	LE_19	691	17	2.40	0	1
32.	In the last 2 years, did you give any kind of help to your friends, and neighbours	SC_20	691	17	2.40	0	1
33.	In the last 2 years, did your neighbours or friends give you any kind of help?	SC_18	690	18	2.54	0	1
34.	Level of difficulty with making telephone calls (including hearing)	FL_74	681	27	3.81	0	1
35.	Do you experience any difficulties participating in social activities outside your home?	SP_9	680	28	3.95	0	1
36.	Can you follow a conversation with one person (with or without a hearing aid)?	PH_26	676	32	4.51	0	1
37.	Have taken a holiday in Ireland in the last 12 months	SP_103	669	39	5.50	0	1
38.	Have taken a holiday abroad in the last 12 months	SP_104	669	39	5.50	0	1
39.	Have gone on a daytrip or outing in the last 12 months	SP_105	669	39	5.50	0	1
40.	Own a mobile phone	SP_107	669	39	5.50	0	1
41.	Have voted in any recent election	SP_101	669	39	5.50	0	1
42.	Can you follow a conversation with four people (with or without a hearing aid)?	PH_27	620	88	12.42	0	1

#	Item	Variable	Present (N)	Missing (N)	Missing (%)	Min. Value	Max. Value
43.	Can type name on a keyboard	LE_22_Id Name	607	101	14.26	0	1
44.	Can type a letter	LE_22_Letter	607	101	14.26	0	1
45.	Can turn on a computer	LE_22_TurnOn	607	101	14.26	0	1
46.	Can send an e mail	LE_22_Email	607	101	14.26	0	1
47.	Can look up topics of interests on Google	LE_22_Google	607	101	14.26	0	1
48.	Can use social media sites such as Facebook and Twitter	LE_22_Social	607	101	14.26	0	1
49.	Do you experience any difficulty getting around your community?*	SP_11	483	225	31.77	0	1
50.	How important would you say religion is in your life?*	CS_31	360	348	49.15	0	1
51.	Do you ever feel lonely?*	SC_8	355	353	49.85	0	1
52.	Do you have someone with whom you can confide?*	SC_15	344	364	51.41	0	1
53.	Do you find it difficult to make friends?*	SC_12	330	378	53.38	0	1
54.	Do you ever feel left out?*	SC_108	329	379	53.53	0	1
55.	How often do you feel you lack friendship / friends?*	SC_13	313	395	55.79	0	1
56.	Do you ever feel isolated?*	SC_14	300	408	57.62	0	1

Valid N (listwise) = 146**

*Variables deleted

**Complete data for all 56 items

In line with standard procedure for constructing a FI (Searle *et al.* 2008), deficits were considered for inclusion in the SFI if they did not contain too many missing values at item level. This was also required to ensure maximum use of available data without excessive reliance on imputation procedures (Theou *et al.* 2013). Of the initial 56 variables, eight had high percentages of missing values (31-57%) and were subsequently deleted. It should be noted that these missing values were in relation to subjective questions which could only be answered by participants themselves and not by proxy, which may have led to some participants being unable to respond. An additional item (MH_819: Problems with justice and/or authorities) was excluded due to zero variance (all responses had a null score). 15 additional variables were removed following exploratory factor analysis, described in Section 5.11, which provided a 32-item SFI (SFI-32) and a shortened 21-item version (SFI-21). It has been recommended that 30–40 items be included to obtain a stable index capable of accurately predicting adverse outcomes (Searle *et al.* 2008).

There are no established cut-points for social frailty. Other similar studies using a deficit accumulation approach to operationalise social frailty have utilised different approaches to define cut-points, which are required for statistical analysis, discussed in Section 5.10. Armstrong *et al.* (2015) grouped participants using the following cut-points: $0.0 < FI \leq 0.10$ (fittest); $0.10 < FI \leq 0.20$ (at risk); $0.20 < FI \leq 0.30$ (frail); $FI > 0.30$ (frailest). In the study by Wallace *et al.* (2014), social vulnerability was identified as having a score in the highest quartile. Andrew *et al.* (2008) also used quartiles of index scores for their analysis. In a later study by Andrew and Rockwood (2010), participants were grouped into three equal tertiles designating high, intermediate, and low social vulnerability. For the purposes of this study, the latter approach was used to establish tertile cut-points for both social frailty indices (SFI-32 and SFI-21): low (0-.33), intermediate (.33-.66), and high (>.66). Of the 708 participants in Wave 2, 473 (66%) provided full data for the 32-item SFI.

5.7.2 Physical Frailty

Physical frailty among the social frailty sub-sample in Wave 2 was measured according to modified frailty phenotype criteria, adapted from O’Connell *et al.* (2020), as described in Section 1.5.2. The IDS-TILDA protocol did not include the exact criteria according to the phenotypes original definition (Fried *et al.* 2001) and therefore the modified criteria, previously developed by O’Connell *et al.* (2020) for specific use in the IDS-TILDA sample, was utilised in this study. It should be noted that various adaptations of the phenotype have emerged from the literature, primarily due to data available in specific studies (Xue 2011). Several studies report having datasets which did not include Fried’s criteria according to their original definition and therefore used similar data to construct a modified frailty phenotype (e.g., Barreto *et al.* 2012, Macklai *et al.* 2013). Key differences between these criteria and those in Fried’s original definition are shown in Table 5.2.

Table 5.2 Comparison of frailty phenotype criteria, adapted from O’Connell (2020)

Criteria	Original Frailty Phenotype Criteria (Fried <i>et al.</i> 2001)	IDS-TILDA Modified Criteria (O’Connell <i>et al.</i> 2020)
Shrinking	In the last year, have you lost more than 10 pounds unintentionally (i.e., not due to dieting or exercise)? (Yes)	Within the last year, have you lost or gained ten pounds (4.5 kg) or more in weight when you weren't trying to? (Yes – lost weight)
Weakness	Lowest sample quintile (20%) for grip strength, adjusted for gender and body mass index	Lowest sample quintile (20%) for grip strength, adjusted for gender and body mass index
Poor endurance and exhaustion	How often in the last week did you feel this way? (a) I felt that everything I did was an effort; (b) I could not get going. (A moderate amount of the time (3–4 days)/most of the time)	How much of the time during the past 4 weeks did you feel worn out? How much of the time during the past 4 weeks did you feel tired? (All of time/most of the time/a good bit of the time)
Slowness	Walking time/15 feet: slowest 20% (by gender/ height)	Lowest sample quintile (20%) for timed up and go, adjusted for gender and height. Wheelchair users, walking aid users, participants requiring assistance to walk across a room
Low physical activity	Minnesota Leisure Time Activity questionnaire (short version) (Low activity)	International Physical Activity Questionnaire (short version) (Low activity)

In this study, the modified criteria include grip strength, vitality, unintended weight loss, timed up and go and physical activity. Scores from these five criteria were summed and categorised as follows: 0 = robust; 1-2 = pre-frail; ≥ 3 = frail. Individuals missing data in relation to two or more of the frailty criteria were excluded from analysis. Of the 473 participants in the overall social frailty sub-sample, 409 participants (87%) provided sufficient data for a frailty phenotype score to be obtained. Further details in relation to the IDS-TILDA variables used to construct the phenotype index are provided in Appendix 8.

5.8 Covariates

Characteristics of study participants analysed in this study included gender, age range, level of ID, type of residence, and Down Syndrome (as cause of ID). In relation to gender, participants indicated whether they were male (coded as 1) or female (coded 2). Age of participants at date of interview was categorised into tertile cut-off points; <50, 50-64, and 65+. Level of ID is based on reported intelligence quotient (IQ) scores, as described in section 1.5.1. IQ scores were categorised as mild (50-55 to approx. 70), moderate (35-40 to 50-55) and severe/profound (below 35-40). Those with unverified or unknown level of ID (N=42, 8.9%) were excluded from logistic regression analysis, discussed in section 5.10.

Data regarding type of residence among participants was collected at baseline. Type of residence was categorised as independent, with family, community housing, congregated setting or other. Independent residence referred to those living independently or with family. Community housing was defined as homes with small groups of people with ID (<10), based in a community setting with staff support. Congregated setting was defined as where 10 or more people share a single living unit or where the living arrangements are campus based. The presence of Down syndrome was determined by participants or their proxies indicating Down syndrome as cause of ID.

5.9 Data Cleaning

Prior to analysing data, it is recommended that the data is screened for errors (Pallant 2020). The researcher conducted data cleaning using IBM SPSS Statistics 27.0 (IBM Japan, Tokyo, Japan). Firstly, frequencies for each variable were calculated and inspected for values outside the expected range. For categorical variables, this involved checking (i) minimum and maximum values; (ii) the number of valid and missing cases; (iii) cases of individual variables which fell into legitimate categories or had out-of-range values. Continuous variables were screened by checking (i) minimum and maximum values; (ii) the mean scores. Case summaries of each variable in the data file were analysed. After all errors were corrected, the frequencies of all variables were then rechecked.

5.10 Statistical Analysis

The SFI constructed for this study was subjected to factor analysis, which is a statistical procedure for modelling observed variables and their covariance structure in terms of unobserved variables (i.e., factors). Firstly, sampling adequacy and suitability of data for factor analysis were evaluated using Kaiser–Meyer–Olkin (KMO) statistic and the Bartlett test of sphericity respectively. This study included the two main types of factor analysis techniques, exploratory factor analysis (EFA) and confirmatory factor analysis (CFA) (Child 2006). The use of EFA is often preferred in the early stages of scale development (Kelloway 1995). It is considered useful for exploring the possible underlying factor structure of a set of observed variables without imposing a preconceived structure on the outcome (Child 1990). Additionally, it is commonly used as a variable reduction technique to remove items not significantly contributing to the measurement construct and to assess the internal reliability of remaining items. Conversely, CFA is a statistical technique used to verify the factor structure of a set of observed variables, which allows the researcher to examine the expected causal connections between variables and their underlying latent constructs. (Hurley *et al.* 1997). It may be used to evaluate the concept validity and the measurement invariance of testing (Murray *et al.* 2020). Furthermore, both EFA and CFA have been used in studies to construct and validate frailty scales and indices (Kamaruzzaman 2010). It has been argued that the appropriateness of using either EFA or CFA in data analysis is dependent upon the study context, and having a theory a priori is of key importance (Hurley *et al.* 1997). In this study, the findings of factor analysis are interpreted

according to the social conceptual model of frailty (Bunt *et al.* 2017). Firstly, EFA was used to explore the underlying factor structure of the SFI and the relationship among included candidate items. Factor loadings, which represent the correlation between variables and factors (Royce 1963), were then analysed. Following CFA, a further 11 variables with low factor loadings ($<.3$) were removed. The remaining 21 weighted items constituted a short-form SFI (SFI-21). Both the SFI-32 and the SFI-21 are included in the study's statistical analysis. Exploratory data analysis and frequency tables were used to describe the study variables. All reported confidence intervals (CI) are within 95%, and statistical significance level was set at a p-value of 0.05. To evaluate the association of social frailty and physical frailty, the proportion of participants with social frailty alone, physical frailty alone, and both social frailty and physical frailty were visualized using Venn diagram. Data on mortality was obtained at follow-up assessments in IDS-TILDA Wave 3 and 4, approximately three and six years later respectively.

Adopting an approach in Park *et al.* (2019), this study used receiver operating characteristic (ROC) and evaluated the area under the curve (AUC) to test the discriminative capacity of the SFI-32, SFI-21 and frailty phenotype to predict mortality over these timeframes. To determine differences between (a) low frail groups versus high frail groups and (b) intermediate frail groups versus high frailty groups, multinomial regression analyses was performed. This statistical procedure is used to explain the relationship between one nominal dependent variable and one or more independent variables. It is used to model nominal outcome variables, in which the log odds of the outcomes are modelled as a linear combination of the predictor variables (Hosmer and Lemeshow 2000). Multinomial logistic regression was performed to examine the impact of independent variables on frailty status. Adjusted models were used to test the association between frailty status and individual characteristics including presence of Down syndrome, age category, residence type and gender based on the SFI (SFI-32 and SFI-21) and the frailty phenotype. Separate models were fitted using high frailty as the reference group to assess the odds of low and intermediate frailty in the context of each factor. Odds Ratio (OR) was reported as a measure of association between an exposure and an outcome. Associations between social frailty alone (SFI-21 and SFI-32), physical frailty alone (frailty phenotype), or both social frailty and physical frailty combined with mortality were evaluated independently within the same model of

logistic regression analysis. Odds ratio with 95% confidence intervals are reported for expected risk factors including age, gender, Down syndrome, level of ID, and residence type.

5.11 Reporting of Findings

As outlined in Table 5.3, the following two chapters present findings relevant to addressing key study objectives. Firstly, Chapter 6 describes the process undertaken to develop and validate the measurement of social frailty used in this study. Chapter 7 provides a focused description of findings in relation to the prevalence of social frailty and associated factors, the relationship between social frailty and physical frailty, and the capacity of social frailty to predict mortality.

Table 5.3 Layout of Results

Description	Chapter
Development and Validation of a Social Frailty Measure	6
Prevalence of Social Frailty and Associated Factors	7
Social Frailty and Physical Frailty	7
Frailty and Mortality	7

5.12 Conclusion

This chapter has provided a detailed description of the methodological approaches used to address the studies aim and objectives. Additionally, the contribution of the researcher towards the collection and analysis of data has been highlighted. The broad conceptual frame of the studies primary data source, IDS-TILDA, enabled the operationalisation of both social frailty and physical frailty using established conceptual models. For the first time the relationship between these domains will be explored in a sample population of older adults with ID. It will also be possible to examine factors associated with social frailty and the capacity of this health state to predict mortality. Overall, it is intended that these insights should contribute towards a holistic understanding of the characteristics of social frailty among older adults with ID. The following two chapters presents the key results of this study. Firstly, Chapter 6 describes the development and validation of the SFI as a means of operationalising social frailty in the study sample. This is followed by Chapter 7 which presents findings in relation to the prevalence of social frailty and associated factors, the relationship

between social frailty and physical frailty, and the capacity of social frailty to predict mortality.

Chapter 6: Development and Validation of a Social Frailty Measure

6.1. Introduction

An initial objective of this doctoral investigation was to develop and validate a measure to operationalise social frailty in the study sample. Social frailty was operationalised according to the accumulation of deficits to construct a SFI. While several studies in the field of ID research have utilised the deficit accumulation model to measure overall frailty among older adults, this is the first known study to adopt this approach to operationalise social frailty in this population. This chapter provides a comprehensive description of the construction and validation of the SFI used in this study. Firstly, outcomes of the factor analysis undertaken to explore and confirm the latent structures of the SFI are reported. Relationships among subsets of variables are interpreted qualitatively according to the conceptual model of social frailty (Bunt *et al.* 2017). The internal consistency, or “reliability”, of the SFI and its variable subsets are then evaluated using Cronbach's alpha. This is followed by an analysis of the distribution of SFI scores in the study sample.

6.2 Data Suitability

Prior to EFA, the suitability of data for factor analysis was tested using Kaiser-Meyer-Olkin (KMO) Measure of Sampling Adequacy and Bartlett's Test of Sphericity (Kaiser 1974, Dziuban and Shirkey 1974, Kaiser 1970). A KMO value larger than 0.5 (Field 2000, Pallant 2020) is considered sufficient while the Bartlett's Test of Sphericity should be significant ($p < .05$) for factor analysis to be suitable (Tabachnick and Fidell 2007). As shown in Table 6.1, sampling Adequacy was 0.854 and Bartlett's Test of Sphericity was significant (p -value of < 0.001), indicating that the sample was suitable for factor analysis.

Table 6.1 KMO and Bartlett's Test output for SFI-32

Kaiser-Meyer-Olkin Measure of Sampling Adequacy		.854
Bartlett's Test of Sphericity	Approx. Chi-Square	7536.999
	Df	1035
	Sig.	.000

6.3 Exploratory Factor Analysis

Factor analysis was carried out on data of participants with complete data on all 32 items of the SFI, resulting in a study population of 473 complete cases. To ascertain the factor structure of the 32-item SFI, EFA was conducted using Categorical Principal Components Analysis (CATPCA) (Meulman 2004). This is a statistical procedure which simultaneously quantifies categorical variables while reducing the dimensionality of the data (IBM 2021). CATPCA uses optimal scaling process which transforms the category labels into numerical values while the variance accounted for among the quantified variables is maximized (Linting and Van der Kooij 2012). It is commonly used in scale development to uncover the underlying structure of a set of variables (De Witte *et al.* 2013). This approach is suitable for data with mixed measurement levels such that nominal, ordinal or numeric which may not have linear relationship with each other.

Candidate items for the SFI included both nominal and ordinal variables. CATPCA considers user-defined missing values, system-missing values, and values less than 1 as missing. To address this issue, the researcher recoded individual items with values less than 1 to make them non-missing. The optimal scaling level for analysis variables was set to 'nominal' and 'ordinal' for categorical and dichotomous variables respectively. Discretization was adjusted to ensure intermittent values for categorical variables were grouped by rank. The rotation method used was varimax rotation with Kaiser normalization (Kaiser 1958). Varimax is an orthogonal rotation method that minimizes the number of variables that have high loadings on each component and therefore simplifies the interpretation of the components (IBM 2021). Standardised factor loadings for all 32 items in the SFI generated from CATPCA output were examined as part of the EFA. Factor loadings can be described as the correlation of the original variable with its latent factor and are therefore useful in determining the importance of the original variable to the factor (Kamaruzzaman 2010). The relevance of variables with factor loadings greater than 0.3 and 0.6 are considered moderately high and high respectively (Kline 1994). In total, 32 variables of the candidate index had moderately high to high factor loadings and were retained for further analysis. Variables with factor loadings below 0.3 were suppressed according to convention (Kline 1994).

Table 6.2 Factor Loading Coefficients of the SFI-32 after Varimax Rotation (N=473)

ADL		ICT use and access		Communication/ Social participation		Life events	
Level of difficulty with getting in or out of bed	0.920	Can look up topics of interests on Google	0.863	Level of difficulty with making telephone calls (including hearing)	0.667	Change of staff in my home where I live or day service I attend	0.775
Level of difficulty using the toilet, including getting up or down	0.907	Can type a letter	0.843	Do you have any difficulty speaking or making yourself understood when speaking?	0.576	Change of my key worker	0.656
Level of difficulty with walking across a room	0.888	Can send an e mail	0.840	Own a mobile phone	0.572	New resident moved into my home	0.599
Level of difficulty with dressing, including putting on shoes and socks	0.860	Can turn on a computer	0.809	Have voted in any recent election	0.571	Change at or from work or day service	0.524
Level of difficulty with bathing or showering.	0.801	Can type name on a keyboard	0.795	In the last 2 years, did you give any kind of help to your friends, and neighbours	0.560	Change in frequency of visits from or to family/friend	0.431
Level of difficulty with eating	0.750	Can use social media sites such as Facebook and Twitter	0.686	In the last 2 years, did your neighbours or friends give you any kind of help?	0.467	Death of a friend	0.428
Doesn't take part in regular physical activity	0.546	Has access to a computer/ laptop/ tablet/ smartphone	0.304	Not a member of any organisation, club or society	0.428	Major illness or injury	0.347
Do you experience any difficulties participating in social activities outside your home?	0.389			Can you follow a conversation with one person (with or without a hearing aid)?	0.418		
Have taken a holiday in Ireland in the last 12 months	0.343			Can you follow a conversation with four people (with or without a hearing aid)?	0.392		

A model with four components (or factors) provided a stable solution and these were interpreted qualitatively using according to the modified version of the conceptual framework of social frailty (Bunt *et al.* 2017), as described in Chapter 5. Each of these identified latent factors were derived from subsets of variables that correlated strongly with each other and weakly with others in the dataset. Correlations among factors and components provided meaningful theoretical interpretations linking them to the construct of social frailty. It was revealed that the four components comprised of factors in relation to general resources, social resources, and social activities and behaviours, suggesting that they significantly contributed to the SFI construct. These domains were interpreted as “ADL”, “ICT use and access”, “Communication/ social engagement” and “Life events” (Table 6.2).

In the first dimension, “ADL”, six variables regarding basic ADL had high factor loadings including level of difficulty with getting in or out of bed (0.920), level of difficulty using the toilet, including getting up or down (0.907), level of difficulty with walking across a room (0.888), level of difficulty with dressing, including putting on shoes and socks (0.860), level of difficulty with bathing or showering (0.801) and level of difficulty with eating (0.750). High factor loadings in dimension one was also observed among other variables that may be interpreted as ADL-related including physical activity (0.546), difficulties participating in social activities outside the home (0.389) and taking a holiday in Ireland in last 12 months (0.343).

Variables in relation to ICT/use represent the highest loading variables in the second dimension. The majority of these pertained to computer literacy skills including looking up topics of interests on Google (0.863), typing a letter (0.843), sending an email (0.840), turning on a computer (0.809), and typing name on a keyboard (0.795). The use of social media sites (such as Facebook and Twitter) and access to a computer/ laptop/ tablet/ smartphone also had significant loadings (0.686, 0.304). The third component contained variables regarding communication and social engagement. Factors in relation to social engagement included voting in a recent election (0.571); support given/received to/from neighbours (0.467,0.560); membership of an organisation/club/society (0.428). In relation to communication, high loadings were observed for the variables having a mobile phone (0.572), difficulty with making telephone calls (0.667), difficulty speaking or being understood (0.576), being able to

follow a conversation with one person (0.418) and being able to follow a conversation with four people (0.392). Lastly, the fourth dimension, "Life events", comprised of variables in relation to change of staff in home/ day service (0.775), change of key worker (0.656), new resident moved into home (0.599), change at or from work or day service (0.524), change in frequency of visits from or to family/friend (0.431), death of a friend (0.428) and major illness or injury (0.347). Further to EFA, 32 variables were retained for inclusion in the full version SFI, referred to as SFI-32.

Cronbach's alpha (Cronbach 1951) was used to measure internal consistency, or coefficient of reliability, of the SFI-32. Reporting this calculation has become common practice in research when multiple-item measures of a concept or construct are employed (Tavakol and Dennick 2011). Similarly, other studies involving the construction of a frailty index have reported Cronbach's alpha. In this study, Cronbach's alpha was obtained each of the four subsets of variables in the SFI-32 EFA model labelled as "ADL", "ICT use and access", "Communication/Social participation", and "Life events". Additionally, this measure was obtained for the overall 32-item index.

Cronbach's alpha reliability coefficient normally ranges between 0 and 1. However, there is no established lower limit to the coefficient. The closer Cronbach's alpha coefficient is to 1, the greater the internal consistency of the items in the scale. While increasing the value of alpha is partially dependent upon the number of items in the scale, it should be noted that this has diminishing returns. George and Mallery (2003) suggest the following cut-points as a guideline: $<.5$ = Unacceptable; $>.5$ = Poor; $>.6$ = Questionable; $>.7$ = Acceptable; $>.8$ = Good; $>.9$ = Excellent. Cronbach's alpha coefficient was good for "ADL" (.855). and acceptable for "Communication/ Social" (.726), while values were observed to be lower for the remaining components "ICT Use/ Access" (.682) and "Life Events" (.649). Finally, the Cronbach's Alpha for the overall SFI-32 was .844, indicating good reliability.

6.4 Confirmatory Factor Analysis

The four-factor model of the SFI-32 resulting from EFA (Fig. 6.1) was subjected to CFA in order to test the hypothesized connection between observed indicators and components. CFA provides explicit hypothesis testing for factor analytic problems and is therefore considered more theoretically important than EFA (Stevens 1996, Gorsuch 1983).

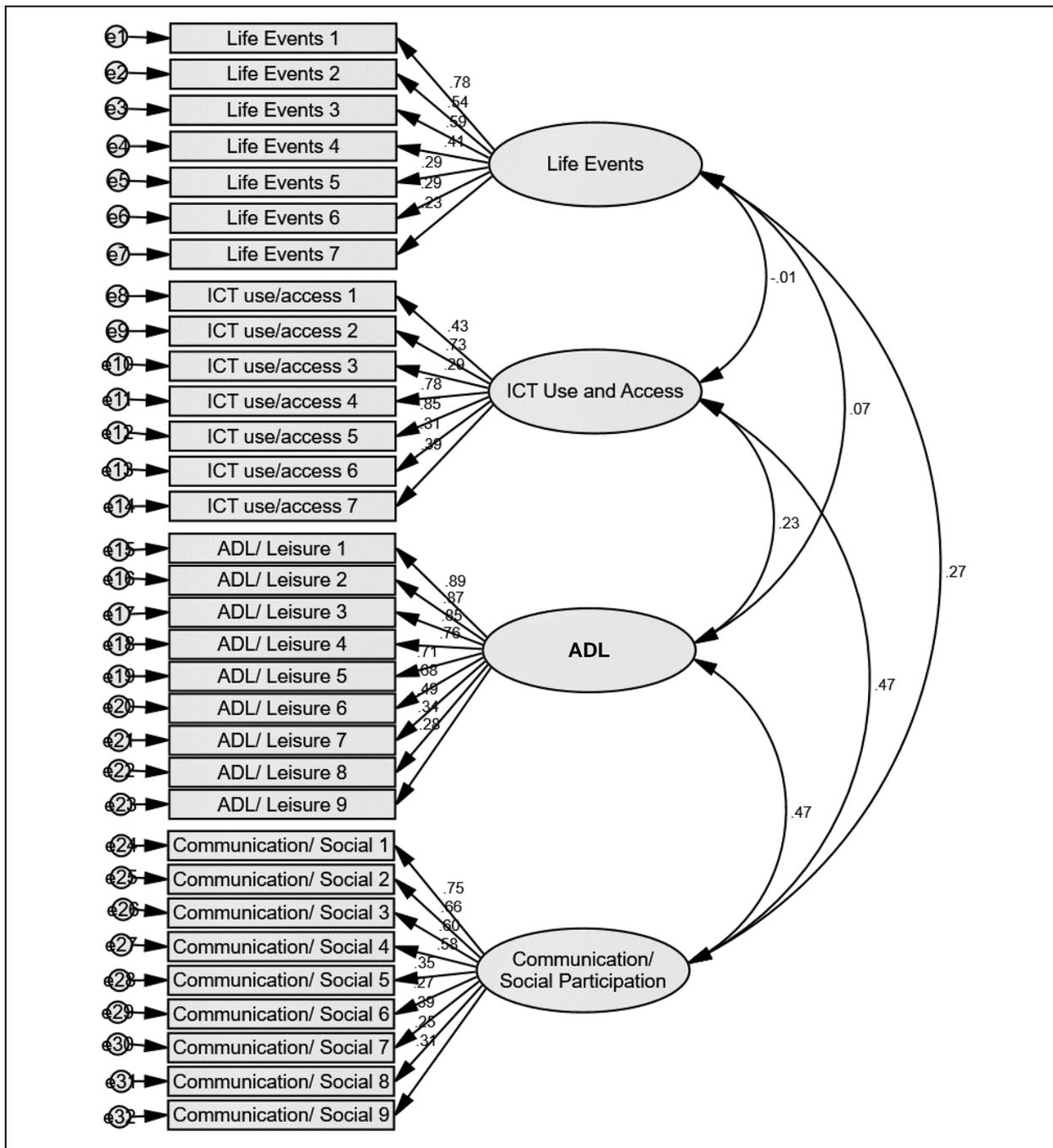


Fig. 6.1 CFA Model of SFI-32 (N=473)

In comparison to the EFA, CFA is theory-driven and aims to determine the ability of a predefined factor model to fit an observed data set (Shek and Yu 2014, Hurley *et al.* 1997). Additionally, CFA reports the model fitness to test hypotheses developed to understand the relationship structure amid set of variables (Harrington 2009). The direction and relationship among variables are articulated on theoretical basis or empirical evidence collected (Brown and Moore 2012). If a decision is made to perform CFA, the next issues to be considered is goodness-of-fit and re-specification of the model, however there is divergent views on what fit statistics tell us about data (Hurley *et al.* 1997).

It has been argued that testing the overall goodness-of-fit of the hypothesized model is essential for understanding how well the model matches the observed data (Orçan 2018) and provides one source of evidence of validity to gauge the significance of a model's misspecifications. Goodness-of-fit of the hypothesized EFA model (SFI-32) was assessed using a variety of model fit indices that examined the connection between the actual data and the conceptual data anticipated from the model (Table 6.3). These can be categorized into two classifications: absolute fit indices and incremental fit indices.

Table 6.3 Model Fit Indices for Hypothesized Model

Fit Index	Scores	Recommended cut-off (P value)
<i>Absolute Fit Measures</i>		
χ^2/df	2.005	<0.05
GFI	.933	≥ 0.90 ; ≥ 0.95
RMSEA	.046	≤ 0.05 ; ≤ 0.1
<i>Incremental Fit Measures</i>		
NFI	.898	≤ 0.90
AGFI	.915	> 0.90
CFI	.946	≥ 0.90
<i>Parsimonious Fit Measures</i>		
PGFI	.739	Highest possible
PNFI	.783	Highest possible

df = degree of freedom

Absolute fit indices directly measure the congruency of theory with the sample selected or observed data of the research. Fit statistics reported include model chi-square (χ^2), Goodness-of-Fit statistic (GFI), and Root Mean Square Error of Approximation (RMSEA) (Han and Johnson 2019). The incremental fit indices, otherwise referred to as comparative or relative fit indices (Miles and Shevlin 2007, McDonald and Ho 2002), are a group of indices that do not use the chi-square in its raw form but compare the chi-square value to a baseline model (Hooper *et al.* 2008). Most commonly, the null model is referred to as baseline model for these indices. Other indices include the Normed Fit Index (NFI), Adjusted Goodness of Fit Index (AGFI), and Comparative Fit Index (CFI).

Parsimonious fit measures comprise of the Parsimony Goodness-of-Fit Index (PGFI) and the Parsimonious Normed Fit Index (PNFI). These indices of the CFA model fit represent the covariation between items and their generalised findings will be adopted by comparing them with cut points scores. Firstly, in relation to absolute fit indices, the χ^2/df measures the accuracy of a model to the actual observed data (Hu and Bentler 1999). A good fitting model fit should provide an insignificant result at a 0.05 threshold (Barrett 2007). As shown in Table 6.3, the obtained value for the CFA model was 2.005, indicating a weak relationship between the hypothesized model and the sample data (Alavi *et al.* 2020). The Goodness-of-Fit statistic (GFI) calculates the proportion of variance that is accounted for by the estimated population covariance (Tabachnick and Fidell 2007). Essentially, it determines the proportion of variability explained by the model. Traditionally GFI cut-off points for good fitting models are considered to be 0.90 or 0.95 (Shevlin and Miles 1998). The value obtained for the CFA model was 0.933, indicating a good fitting model (Hooper *et al.* 2008).

RMSEA assesses how far the hypothesized model is from a perfect model (Xia and Yang 2019). It tests how well a model would fit the populations covariance matrix with unknown but optimally chosen parameter estimates (Byrne 1998). There is a considerable lack of consensus on recommended cut-off points for RMSEA (Hooper *et al.* 2008). MacCallum *et al.* (1996) assert that a value between 0.05 to 0.10 indicates a fair fit, while a value greater than 0.10 suggests a poor fit. Others have asserted that a good fitting model should have a RMSEA value close to 0.06 or 0.07 (Steiger 2007). The obtained value was 0.046, indicating poor concurrency between the hypothesized

and perfect model. Overall, absolute fit measures indicated that the proposed model was not consistent with the perfect model. Values obtained for the incremental fit measures were then considered, beginning with NFI which analyzes the discrepancy between the chi-squared value of the hypothesized model and the chi-squared value of the null model (Bentler and Bonett 1980). The null/independence model is the worst-case scenario as it specifies that all measured variables are uncorrelated (Hubley 2014). A value greater than 0.90 or 0.95 for NFI is considered indicative of a good fitting index (Hooper *et al.* 2008, Hu and Bentler 1999). The value obtained for the hypothesized CFA model was marginally lower at 0.898. As in GFI, AGFI is also a measure of the proportion of variance accounted for by the estimated population covariance. AGFI corrects the GFI, which is affected by the number of indicators of each latent variable. The obtained score of 0.915 suggests a good fit in the context of the recommended cut-off point >0.90 (Hooper *et al.* 2008). The CFI measure analyses the model fit by examining the discrepancy between the data and the hypothesized model, while adjusting for the issues of sample size inherent in the chi-squared test of model fit (Gatignon 2010) and the normed fit index (Bentler 1990). Kline (2005) recommend a cut-off point score of >0.90 for a good fitting index. The score of 0.946 obtained for the hypothesized model was therefore considered to be acceptable.

The final goodness-of-fit indices to be analysed as part of CFA were the Parsimony PGFI and PNFI, developed by Mulaik *et al.* (1989). The PGFI is based upon the GFI by adjusting for loss of degrees of freedom. The PNFI also adjusts for degrees of freedom, however it is based on the NFI (Mulaik *et al.* 1989). Both of these indices penalise for model complexity, resulting in parsimony fit index values that are considerably lower than other goodness-of-fit indices. A more parsimonious model with fewer estimated parameters is considered better than a more complex model (Marsh and Hau 1996). There are no threshold levels recommended for these indices, making them more difficult to interpret. Mulaik *et al.* (1989) note that it is possible to obtain parsimony fit indices within the region of .50. The PGFI and PNFI values obtained for CFA model were 0.739 and 0.783 respectively.

Loading estimates of the four factors in the hypothesized CFA model and their component items were analysed. For convenience, these items are labelled numerically. A cut point of 0.3 was considered significant (Tabachnick and Fidell 2018). Items in the original CFA model had significant scores ranging from 0.30 to 0.91. Factor loadings for ICT Use and Access 3 and 6, ADL 4, 8, 9 and Communication/ Social Participation 5, 6, and 8 had a low loading value ($<.3$) and were subsequently removed. Similarly, lower loading estimation of components 1-4, 5-11, 12-18, and 19-27 determined their exclusion from the model. Items in the original CFA model had loading scores ranging from 0.30 to 0.91 except the factor loadings for ICT Use and Access 3 and 6, ADL 4, 8, 9 and Communication/ Social Participation 5, 6, and 8. As a result, these items were removed from the test for having loading values less than .3. The components 1-4, 5-11, 12-18, and 19-27 were labelled as four categories, namely "Life Event", "ICT Use and Access", "ADL", and "Communication/ Social Participation". The lower loading estimation of these items led to their removal from the model as recommended by Tabachnick and Fidell (2018).

The standardized factor loadings for Life Event 1, 2, 3 and 4 were .798, .522, .598 and .404, respectively. For the second factor, ICT Use and Access, items 2, 4, 5 and 7 had significant loading estimates of .394, .884, .777, .713 and .378 respectively. ICT Use and Access 3 and ICT Use and Access 5 were removed for having factor loading estimates less $<.3$. In relation to the third factor, ADL, items 1, 2, 3, 5, 6 and 7 had high loading estimates of .479, .661, .662, .860, .871, .919 respectively. One item, ADL 4, was excluded due to a low factor loading. The fourth and final factor analysed was Communication/ Social Participation. The factor loading estimates for Communication/ Social Participation 1, 2, 3, 4, 7, and 9 were .300, .378, .571, .605, .651 and .782 respectively. Communication/ Social Participation 5, 6 and 8 were excluded from the analysis. The convergent validity of these items is low and they do not share a high proportion of variance in common (Bessette *et al.* 2018). The cut points score for discriminant validity is 0.850. The standardized factor loadings for Life Event 1, 2, 3 and 4 are .798, .522, .598 and .404, respectively.

Next, the discriminant validity among the four factors in the hypothesized model were evaluated using loading estimates (Table 6.4). The goal of discriminant validity evidence is to be able to discriminate between measures of dissimilar constructs, thereby ensuring that measures of constructs are not highly correlated to each other (Hubley 2014). When looking at the estimations (such as the factor loadings as well as factor correlations), one should consider that only the best-fitting solution makes sense. The parameter estimations may be skewed if the model is not fitting the data well (Kyriazos 2018). By finding correlation between each pair of variables that are of high loading estimates, the discriminant validity can be used to find the correlation between them. A high value represents significant similarity between both variables and suggests they can be treated as one measuring variable. While there is no standard value for discriminant validity, a score of <0.85 suggests that validity likely exists between the two scales (Campbell and Fiske 1959). A result greater than 0.85, however, suggests that the two constructs overlap greatly, and they are likely measuring the same thing, and therefore, discriminant validity between them cannot be claimed (Campbell and Fiske 1959).

Table 6.4 Correlation matrix of hypothesized model

Domains			Estimate
Life Event	<-->	ICT Use and Access	-.018
Life Event	<-->	ADL	.047
Life Event	<-->	Communication/ Social Participation	.254
ICT Use and Access	<-->	ADL	.216
ICT Use and Access	<-->	Communication/ Social Participation	.486
ADL	<-->	Communication/ Social Participation	.406

As shown in Table 6.4, no significant correlation between the four factors of the hypothesized model was observed. This finding indicates that the variables in the model are measuring different aspects but are interrelated to some extent. A correlation value less than 0.2 is considered to have little practical significance (Taylor 1990). Life Event does not hold significant correlation with ICT Use and Access because of very weak correlation value (-0.018). Similarly, Life Event had a weak correlation with the factor ADL (-0.047). Correlation between Life Event and Communication/Social Participation is also not strong with a correlation value of 0.254. The factors ICT Use and Access and ADL were observed to have low correlation

(0.216). ICT Use and Access and Communication/Social Participation were found to be positively and moderately correlated owing to the correlation value 0.486. Similarly, ADL and Communication/Social Participation also appeared to have moderate correlation (0.406). The new weighted variables (N= 21) obtained from CFA constituted a short-form SFI, referred to as SFI-21. Variables included in this model are described in Table 6.5.

Table 6.5 Variables included in SFI-21 (N= 473)

Factor	Item	Variable	Description
Life events	Life Events 1	MH_802	Change of staff in my home where I live or day service I attend
	Life Events 2	MH_803	New resident moved into my home
	Life Events 3	MH_804	Change of my key worker
	Life Events 4	MH_805	Change at or from work or day service
ICT use and access	ICT use/ access 1	LE_22_Google	Can look up topics of interest on Google
	ICT use/ access 2	LE_22_Letter	Can type a letter
	ICT use/ access 4	LE_22_TurnOn	Can turn on a computer
	ICT use/ access 5	LE_22_IdName	Can type my name on a keyboard
	ICT use/ access 7	LE_19	Access to a computer on a regular basis
ADL	ADL/ Leisure 1	FL_51_R_A	Level of difficulty getting in or out of bed
	ADL/ Leisure 2	FL_56_R_A	Level of difficulty using the toilet
	ADL/ Leisure 3	FL_29_R_A	Level of difficulty walking across a room
	ADL/ Leisure 5	FL_38_R_A	Level of difficulty bathing or showering
	ADL/ Leisure 6	FL_46_R_A	Level of difficult with eating
	ADL/ Leisure 7	BH_2101_R_A	Participation in regular physical activity
	Communication / Social participation	Communication/ Social 1	FL_74_R_A
Communication/ Social 2		PH_31_R_A	Difficulty speaking or making self understood
Communication/ Social 3		SP_107_R_A	Own a mobile phone
Communication/ Social 4		SP_101_R_A	Have voted in any recent election
Communication/ Social 7		SP_311_R_A	Membership of an organisation, club or society
Communication/ Social 9		PH_27_R_A	Can follow a conversation with four people

The hypothesized connection between observed indicators and components of the SFI-21 is outlined in Fig. 6.2. Both the SFI-32 and SFI-21 were included in further statistical analysis. However, the former index is intended to be the primary measure for social frailty on the basis that it meets the criteria of including over 30 index items, as recommended in a standard procedure for creating a frailty index by Searle *et al.* (2008).

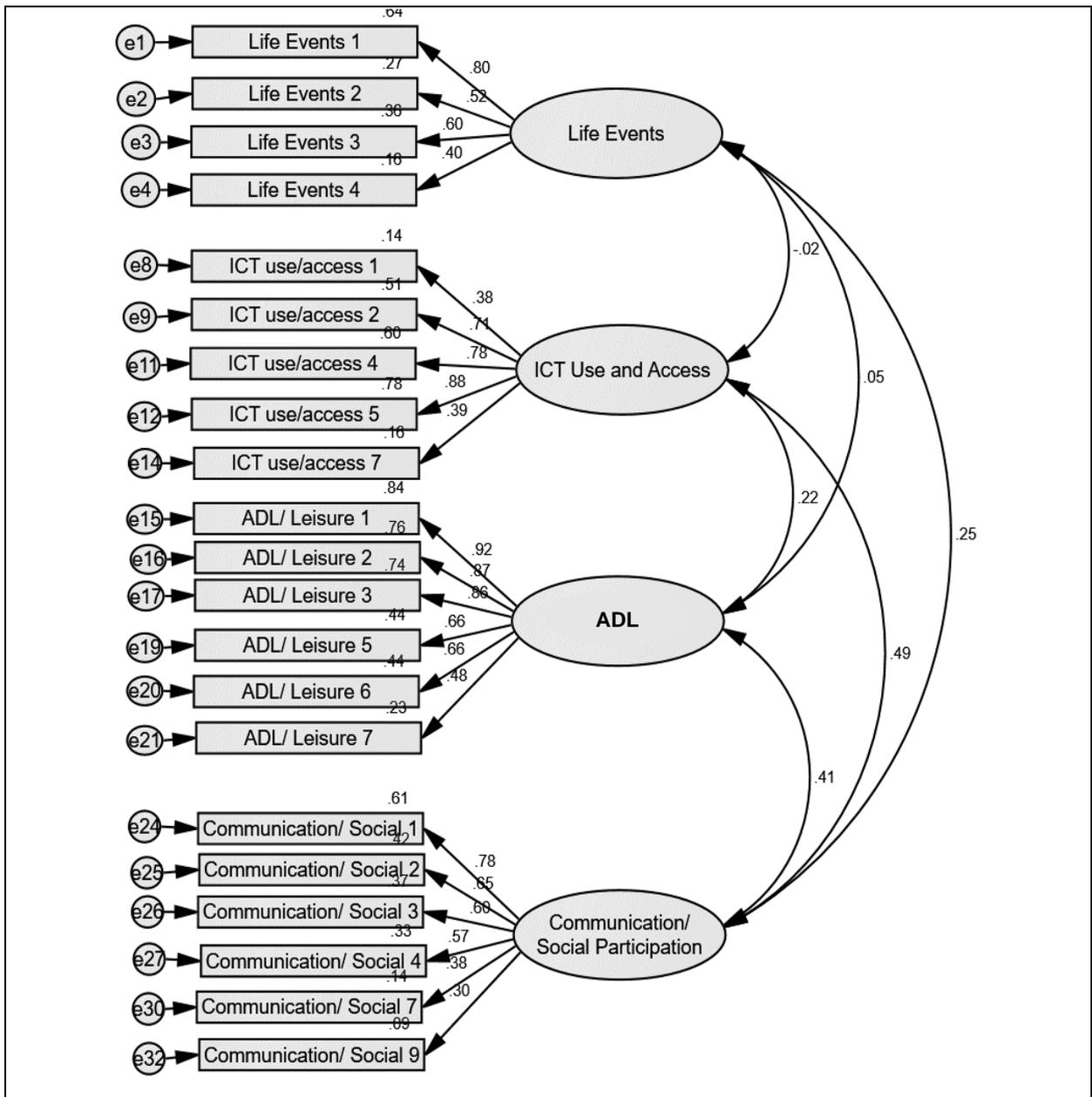


Fig. 6.2 CFA Model of SFI-21 (N=473)

6.5 Distribution of the SFI

Descriptive statistics were generated to assess this normality of distribution of SFI-32 scores. A histogram was first used to display the distribution of the SFI as a single continuous variable. Inspection of the shape of the histogram (Fig 6.3) provides information about the distribution of scores on the continuous variable (Pallant 2020).

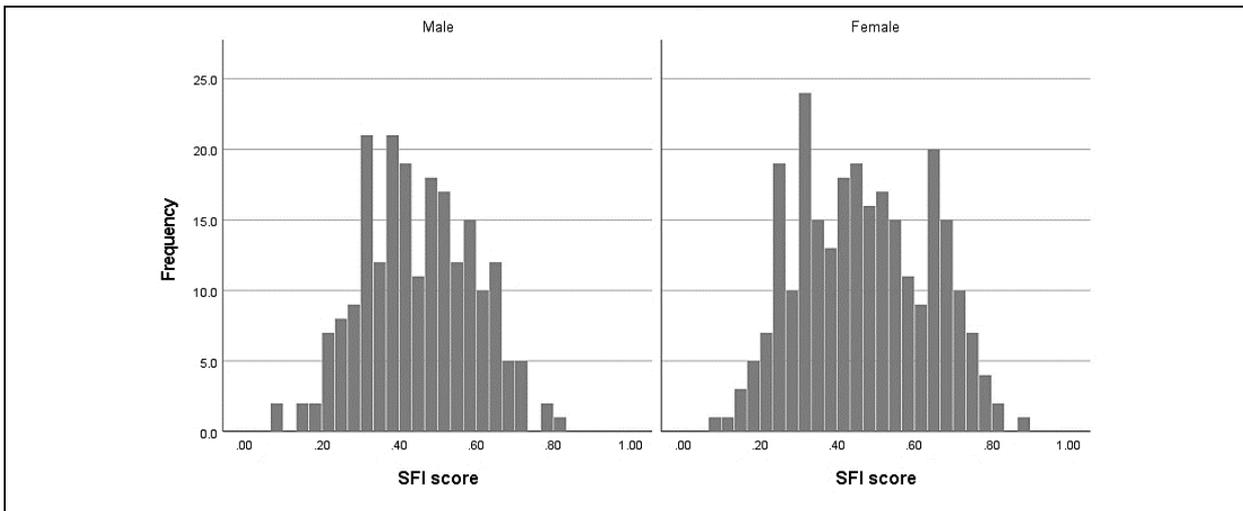


Figure 6.3 Histogram describing parameters of normal Distribution of SFI by gender

SFI scores appear to be relatively normally distributed for both male and female cohorts, with the highest scores occurring in the centre with the rest tapering off to the extremities. Normal distribution is also supported by an inspection of the normal probability plots shown in Fig 6.4. These plot the observed value of each score against the expected value from normal distribution (Chambers *et al.* 1983). The relative straightness of this line indicates normal distribution (Pallant 2020).

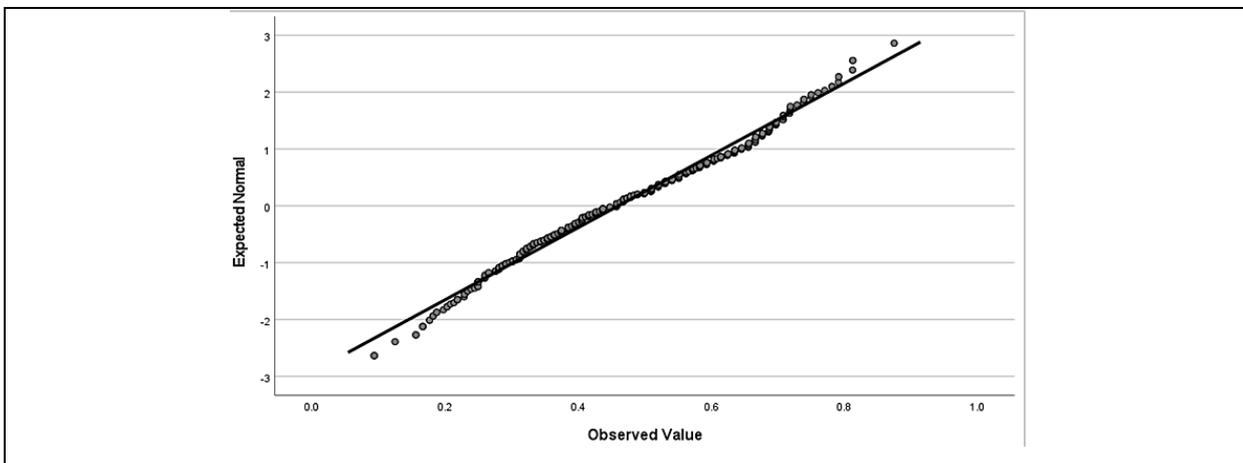


Figure 6.4 Normal probability plots for SFI scores

6.6 Conclusion

This chapter provided a comprehensive description of the construction and validation of the SFI used in this study. The conceptual model of social frailty (Bunt *et al.* 2017) was utilised as an evidence-based means of identifying candidate items for the construction of a SFI. This model was also crucial to interpreting a factor analysis of the SFI, which revealed that this measure could be explained by variable subsets relating to ADL, ICT use and access, communication and social engagement, and life

events. This latent structure was then subjected to CFA in order to test the hypothesized connection between observed indicators and components. Goodness-of-fit of the hypothesized model was assessed using a variety of model fit indices that examined the connection between the actual data and the conceptual data anticipated from the model. Further to confirmatory factor analysis, a short form 21-item index was constructed using weighted items only. Finally, an analysis of descriptive statistics revealed that SFI scores were relatively normally distributed in the study sample. In the next chapter, both the SFI-21 and SFI-32 are utilised in statistical analysis procedures performed to address the remaining key objectives of this study including analysing the prevalence of social frailty and associated factors, examining the relationship between social frailty and physical frailty, and assessing the capacity of social frailty to predict mortality. These findings are discussed in further detail in Chapter 8 (see section 8.3.1). In the next chapter, the SFI measures developed are incorporated in statistical analysis procedures performed to address the remaining aims of this study including analysis of the prevalence of social frailty and associated factors, examination of the relationship between social frailty and physical frailty, and assessing the capacity of social frailty to predict mortality.

Chapter 7: Results

7.1 Introduction

Further to development and validation of a measurement of social frailty, as described in Chapter 6, this chapter presents data forming the basis of this study's investigation of the holistic characteristics of social frailty among older adults with ID. The findings reported address key objectives of this study including the analysis of the prevalence of social frailty and associated factors, examination of the relationship between social frailty and physical frailty, and assessment of the capacity of social frailty to predict mortality. Firstly, an analysis of the characteristics of the study sample is summarized by mean and standard deviation (SD) for continuous variables, and proportions for categorical variables. To evaluate the association between social frailty and physical frailty, the proportion of participants with social frailty alone, physical frailty alone or both social frailty and physical frailty together are visualized with Venn diagram. The individual and combined capacity of these measures to predict mortality after three and six years were analyzed using Receiver Operating Characteristic (ROC). Multinomial logistic regression was performed to examine the impact of independent variables on frailty status. Finally, logistic regression was used to identify mortality risk associated with social frailty alone, physical frailty alone, and social frailty and physical frailty combined after adjusting for age, gender, presence of Down syndrome, and type of residence. Relevant statistical data is presented with the use of tables and figures with commentary by the researcher on the significance of the data shown. A more elaborate commentary is provided in Chapter 7 as part of the analysis and synthesis of these findings.

7.2 Prevalence of Social Frailty and Associated Factors

Of the 708 participants at baseline (IDS-TILDA Wave 2), 473 (66%) provided full data for the 32-item SFI. Characteristics of the social frailty sub-sample are described in Table 7.1. As in the overall IDS-TILDA Wave 2 sample, females were slightly more represented than males (N= 262, 55% vs N= 211, 45%). Age categories represented were <50 (N= 129, 27%), 50-64 (N= 237, 50%) and 65+ (N= 107, 23%), with a mean age of 56 years. Type of residence among participants was independent/ family home (N= 79, 17%), community group home (N= 217, 46%), and residential care (N= 177, 37%). For the SFI-32, no participant had a null score. Scores ranged between .09 and .87 (Mean .46, SD .15). In contrast, a null score was observed for SFI-21, with a maximum score of .90 (Mean .45, SD .18). A total of 409 study participants provided

information for a frailty phenotype score based upon modified criteria developed by O'Connell *et al.* (2020) (see Chapter 5), with scores ranging between 0.00 to 5.0 (Mean 1.45, SD 1.09).

Table 7.1 Characteristics of Social Frailty Sub-Sample (N= 473)

Factor	Interval	N	%
Age (Years)	Min	44	
	Max	85	
	Mean	56.72	
	SD	9.385	
Age Category (Years)	<50	129	27.3
	50-64	237	50.1
	65+	107	22.6
Gender	Male	211	44.6
	Female	262	55.4
Type of residence	Independent/ family home	79	16.7
	Community group home	217	45.9
	Residential care	177	37.4
Level of ID	Mild	110	23.3
	Moderate	209	44.2
	Severe/ Profound	112	23.7
	Unverified/ not known	42	8.9
Cause of ID	Down Syndrome	92	19.5
	Other	381	80.5
SFI-21 Total Score	Mean	.4534	
	Minimum / Maximum	.00 / .90	
	SD	.18	
	Valid N (listwise)	473	
SFI-32 Total Score	Mean	.4603	
	Minimum / Maximum	.09 / .87	
	SD	.15	
	Valid N (listwise)	473	
Frailty Phenotype Total Score	Mean	1.45	
	Minimum / Maximum	.00 / 5.0	
	SD	1.09	
	Valid N (listwise)	409	

The characteristics of participants based on SFI-32 (N= 473) are shown in Table 7.2. Mortality rates are the same as that for the full 32-item index with 47 and 68 participants lost to follow-up due to death between Waves 2-3 and Waves 3-4 respectively. 19 participants with a high tertile SFI-21 score died between Waves 2-3. A further 15 participants in this category died between Waves 3-4. Participants with Down syndrome (N= 92) were represented across all tertile cut points including low (N= 22, 24%), intermediate (N= 55, 60%) and high (N= 15, 16%). Among participants aged <50 (N= 129), 33 (25%) had low social frailty, 87 (68%) had intermediate social frailty, and 9 (7%) had high social frailty. Participants aged 50-64 (N= 237) were represented in the categories of low (N= 83, 35%), intermediate (N= 125, 53%) and high (N= 29, 12%). SFI-21 total scores among participants aged 65+ (N= 107) were low (N= 18, 17%), intermediate (N= 68, 63%), and high (N= 21, 20%). Participants with a residential status of independent/ family or group home (N= 296) were categorised as having low (N= 123, 42%), intermediate (N= 154, 52%) or high (N= 19, 6%) levels of social frailty.

Table 7.2 Characteristics of participants based on SFI-32 only

Factor	Interval	Low	Intermediate	High	Overall	P value
Status at follow-up	Alive Wave 2-3	117	277	32	426	<0.001
	Died Wave 2-3	4	23	20	47	
	Alive Wave 3-4	111	253	41	405	
	Died Wave 3-4	10	47	11	68	
Level of ID	Mild	57	52	1	110	<0.001
	Moderate	49	139	21	209	
	Severe/ Profound	2	81	29	112	
Cause of ID	Down syndrome	20	57	15	92	0.163
	Other	101	243	37	381	
Age Category (Years)	<50	33	87	9	129	<0.001
	50-64	76	135	26	237	
	65+	12	78	17	107	
Residence Type	Indep./Family/ Group Home	103	175	18	296	<0.001
	Residential Care	18	125	34	177	
Gender	Male	51	147	13	211	0.005
	Female	70	153	39	262	

Table 7.3 shows the characteristics of participants based on SFI-21 (N= 473). Mortality rates are the same as that for the full 32-item index with 47 and 68 participants lost to follow-up due to death between Waves 2-3 and Waves 3-4 respectively. 19 participants with a high tertile SFI-21 score died between Waves 2-3. A further 15 participants in this category died between Waves 3-4. Participants with Down syndrome (N= 92) were represented across all tertile cut points including low (N= 22, 24%), intermediate (N= 55, 60%) and high (N= 15, 16%). Among participants aged <50 (N= 129), 33 (25%) had low social frailty, 87 (68%) had intermediate social frailty, and 9 (7%) had high social frailty. Participants aged 50-64 (N= 237) were represented in the categories of low (N= 83, 35%), intermediate (N= 125, 53%) and high (N= 29, 12%). SFI-21 total scores among participants aged 65+ (N= 107) were low (N= 18, 17%), intermediate (N= 68, 63%), and high (N= 21, 20%). Participants with a residential status of independent/ family or group home (N= 296) were categorised as having low (N= 123, 42%), intermediate (N= 154, 52%) or high (N= 19, 6%) levels of social frailty.

Table 7.3 Characteristics of participants based on SFI-21 only

Factor	Interval	Low	Intermediate	High	Overall	P value
Status at follow-up	Alive Wave 2-3	137	242	47	426	
	Died Wave 2-3	4	24	19	47	<0.001
	Alive Wave 3-4	127	227	51	405	
	Died Wave 3-4	14	39	15	68	0.049
Level of ID	Mild	66	43	123	110	
	Moderate	57	129	23	209	
	Severe/ Profound	4	67	41	112	<0.001
Cause of ID	Down syndrome	22	55	15	92	
	Other	119	211	51	381	0.361
Age Category (Years)	<50	40	73	16	129	
	50-64	83	125	29	237	
	65+	18	68	21	107	0.011
Residence Type	Independent/Family/ Group Home	123	154	19	296	
	Residential Care	18	112	47	177	<0.001
Gender	Male	61	132	18	211	
	Female	80	134	48	262	0.004

Among 177 participants living in residential care, 18 (10%) had low levels, 112 (63%) had intermediate levels, and 47 (27%) had high levels. In relation to gender, 211 male participants and 262 female participants provided data for a SFI-21 measure to be obtained. Males were categorised as low (N= 61, 29%), intermediate (N= 132, 63%), and high (N=18, 8%). SFI-21 scores among female participants were low (N= 80, 31%), intermediate (N= 134, 51%) and high (N= 48, 18%). Table 7.4 shows that 37 participants with data for a frailty phenotype score (N= 409) were lost to follow-up due to death between baseline assessment (Wave 2) and the first follow-up interval (Wave 3). A higher mortality rate (N= 62). was observed at final follow-up in Wave 4. In terms of frequency, 18 participants died between Waves 2-3, and a further 14 died between Waves 3-4. Participants with Down syndrome (N= 74) were represented in all three frailty phenotype categories including robust (N=16, 22%), pre-frail (N= 42, 56%), and frail (N= 16, 22%). It was observed that participants aged <50 (N= 107) were physically robust (N=24, 22%), pre-frail (N= 66, 62%), or frail (N= 17, 16%).

Table 7.4 Characteristics of participants based on frailty phenotype only

Factor	Interval	Robust	Pre-frail	Frail	Overall	P value
Status at follow-up	Alive Wave 2-3	86	236	50	372	
	Died Wave 2-3	2	17	18	37	<0.001
	Alive Wave 3-4	76	217	54	347	
	Died Wave 3-4	12	36	14	62	0.389
Level of ID	Mild	28	62	4	94	
	Moderate	41	111	34	186	
	Severe/ Profound	13	56	27	96	<0.001
Cause of ID	Down Syndrome	16	42	16	74	
	Other	72	211	52	335	0.420
Age Category (Years)	<50	24	66	17	107	
	50-64	52	128	28	208	
	65+	12	59	23	94	0.053
Residence Type	Independent/Family/ Group Home	63	156	30	249	
	Residential Care	25	97	38	160	0.002
Gender	Male	45	113	16	174	
	Female	43	140	52	235	0.001

In terms of level of ID, participants with a mild level of ID (N=94) were represented in all frailty phenotype categories including robust (N= 28, 30%), pre-frail (N= 62, 66%), and frail (N= 4, 4%). Among those with a moderate ID level (N= 186), 41 (22%) were robust, 111 (60%) were pre-frail participants, and 34 (18%) were frail. Participants with a severe level of ID (N= 96) were categorised as robust (N= 13, 14%), pre-frail (N= 56,58%), and frail (N= 27, 28%). Among participants aged 50-64 (N= 208), 52 (25%) were robust, 128 (62%) were pre-frail, and 28 (13%) were frail. Participants in the 65+ age category (N= 94) were observed to be robust (N= 12, 13%), pre-frail (N= 59, 63%), or frail (N= 23, 24%). Participants with a residential status of independent/ family or group home (N= 249) were categorised as robust (N= 63, 25%), pre-frail, (N= 156, 63%) or frail (N= 30, 12%). Among participants living in residential care (N= 160), 25 (16%) were robust, 97 (60%) were pre-frail, and 38 (24%) were frail. In relation to gender, 174 male and 235 female participants provided data for a phenotype score. Male participants were represented in all categories including robust (N= 45, 26%), pre-frail (N= 113, 65%), and frail (N= 16, 9%). Female participants were robust (N= 43, 18%), pre-frail (N=140, 60%) or frail (N= 52, 22%).

7.3 Social Frailty and Physical Frailty

To evaluate the association between social frailty and physical frailty, the proportion of participants with social frailty alone, physical frailty alone, and both social frailty and physical frailty together was visualized with Venn diagram (Fig. 7.1). Social frailty is defined as having a SFI-32/ SFI-21 score in the high tertile range (>.66), while physical frailty is measured using the frailty phenotype (≥ 3 = frail).

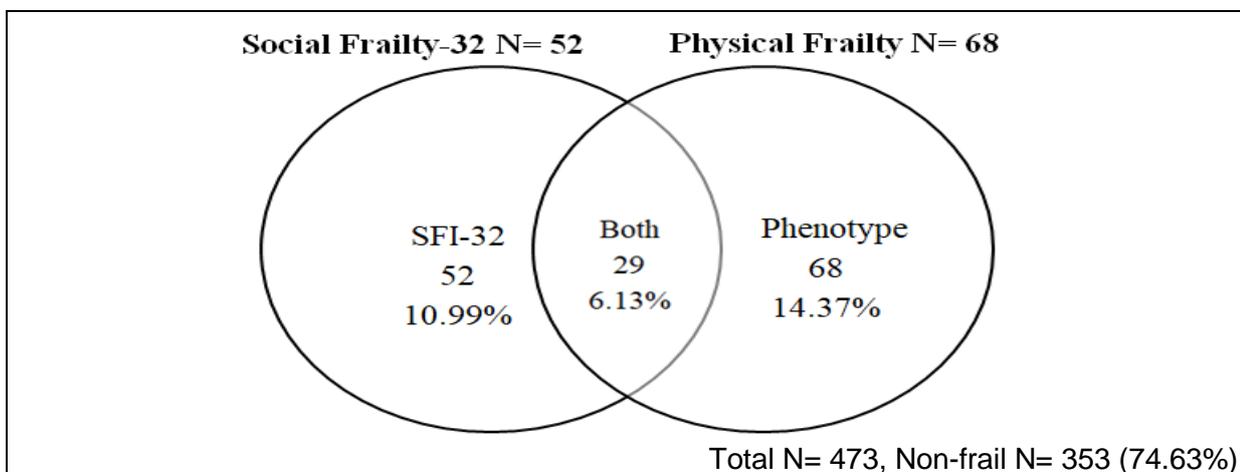


Figure 7.1 Venn diagram of prevalence of social frailty (SFI-32) and physical frailty

The prevalence of physical frailty was found to be higher (N=69, 14.37%.) in comparison to high tertile SFI-32 scores (N=66, 10.99%). A total of 29 participants (6.13%) had a high level of social frailty and were physically frail based on phenotype criteria. Similar findings were obtained with the SFI-21 (Fig. 7.2). The proportion of participants with a high tertile SFI-21 score only was 13.97% (N=66) while 14.37% (N=68) were identified as having physical frailty alone. In total, 6.55% participants (N=31) were identified as having physical frailty alone. In total, 6.55% participants (N=31) were both physically frail and had a tertile score for the SFI-21 score.

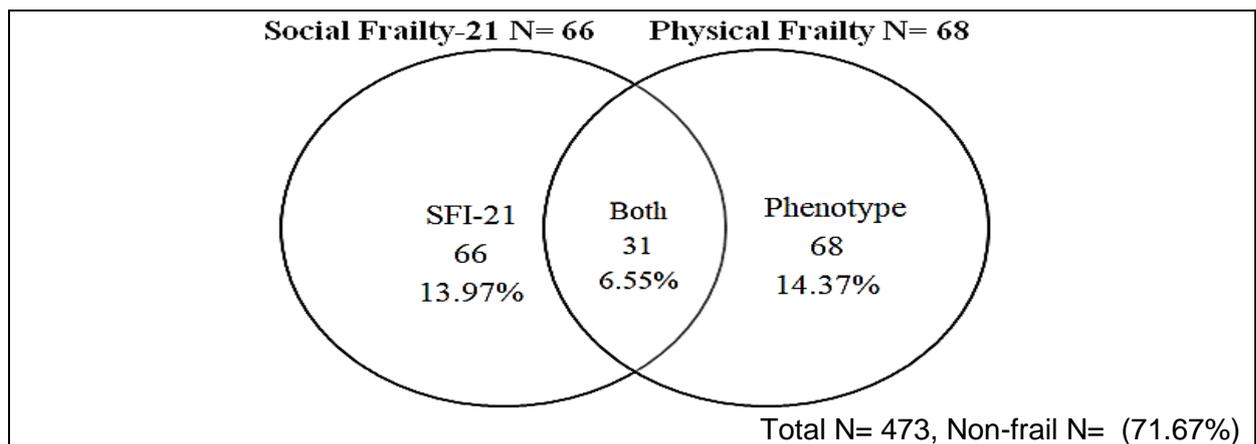


Figure 7.2 Venn diagram of prevalence of social frailty (SFI-21) and physical frailty

7.4 Social Frailty and Mortality

A key objective of this study was to explore the ability of social frailty to predict mortality. Mortality rates among participants in the social sub-sample is based on data collected at follow-up assessments in Waves 3 and 4 of IDS-TILDA, three and six years after baseline respectively (Wave 2). It should be noted that mortality data is based on reported death of participants during sample recruitment or data collection, and their actual date of death was not available to the researcher at the time of this study. Adopting an approach in Park et al. (2019), ROC was used to evaluate AUC to assess the capacity of the SFI-32, SFI-21 and frailty phenotype in predicting mortality approximately three and six years after baseline. ROC curves representing mortality between baseline (IDS-TILDA Wave 2) and first follow-up (IDS-TILDA Wave 3) associated with the continuous scores of these measures are compared in Fig 7.3.

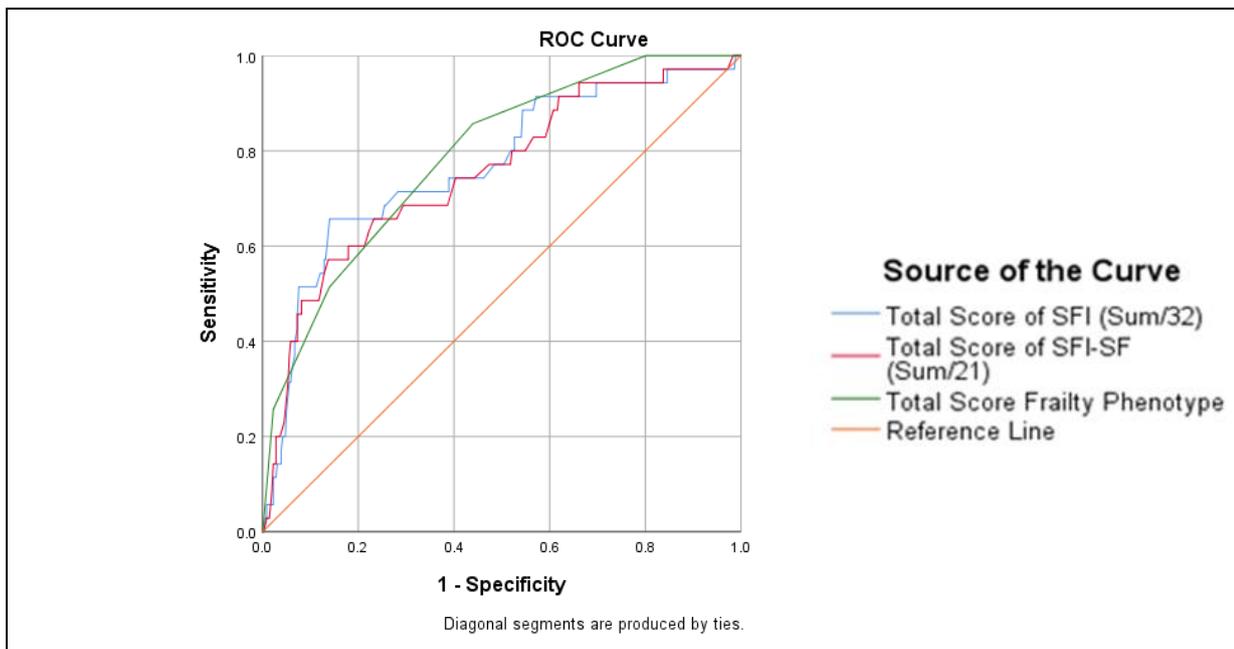


Figure 7.3 ROC analysis of predictive model for mortality (Waves 2-3)

In interpreting the ROC curve, classifiers that give curves closer to the top-left corner indicate a better performance and the closer the curve is in proximity to the 45-degree diagonal of the ROC space, the less accurate the test. ROC curves for SFI-32, SFI-21 and the frailty phenotype are also within close proximity to the reference line and to each other. This indicates that all three frailty measures have good and similar discriminatory capacity to predict mortality over approximately three years (Wave 2 - Wave 3).

Table 7.5 Area Under the Curve in ROC analysis (Waves 2-3)

Test Result Variable(s)	Area
Total Score of SFI (Sum/32)	.768
Total Score of SFI-SF (Sum/21)	.754
Total Score Frailty Phenotype	.788

An analysis of C-statistics (Table 7.5) found that the SFI-32, SFI-21, and frailty phenotype have similar values of 0.742, 0.682 and 0.662 respectively, indicating that there is no statistically significant difference in discrimination ability to predict mortality over an approximate timeframe of three years. Figure 7.4 compares ROC curves representing mortality between first follow-up interval (IDS-TILDA Wave 3) and final follow-up (IDS-TILDA Wave 4) approximately six years associated with the continuous scores of the SFI-32, SFI-21 and frailty phenotype.

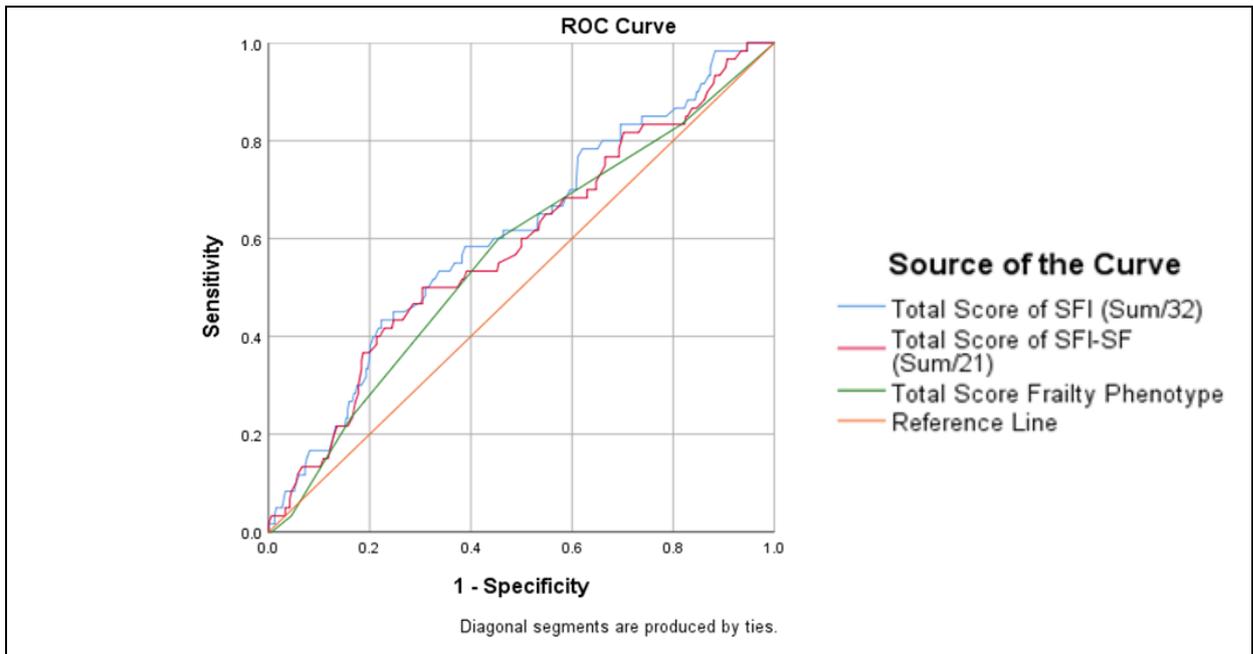


Fig 7.4 ROC analysis of predictive model for mortality (Waves 3-4)

Table 7.6 Area Under the Curve in ROC analysis (Waves 3-4)

Test Result Variable(s)	Area
Total Score of SFI (Sum/32)	.609
Total Score of SFI-SF (Sum/21)	.586
Total Score Frailty Phenotype	.566

As shown in Table 7.6, an analysis of C-statistics found that the SFI-32, SFI-21, and frailty phenotype have similar values of 0.609, 0.586 and 0.566 respectively, indicating that there was no statistically significant difference in the discriminatory capacity of these measures to predict mortality over this timeframe. ROC curves representing mortality between baseline (Wave 2) and all mortality at any follow-up (Wave 3 or Wave 4) associated with the continuous scores of the SFI-32, SFI-21 and frailty phenotype are compared in Fig 7.5.

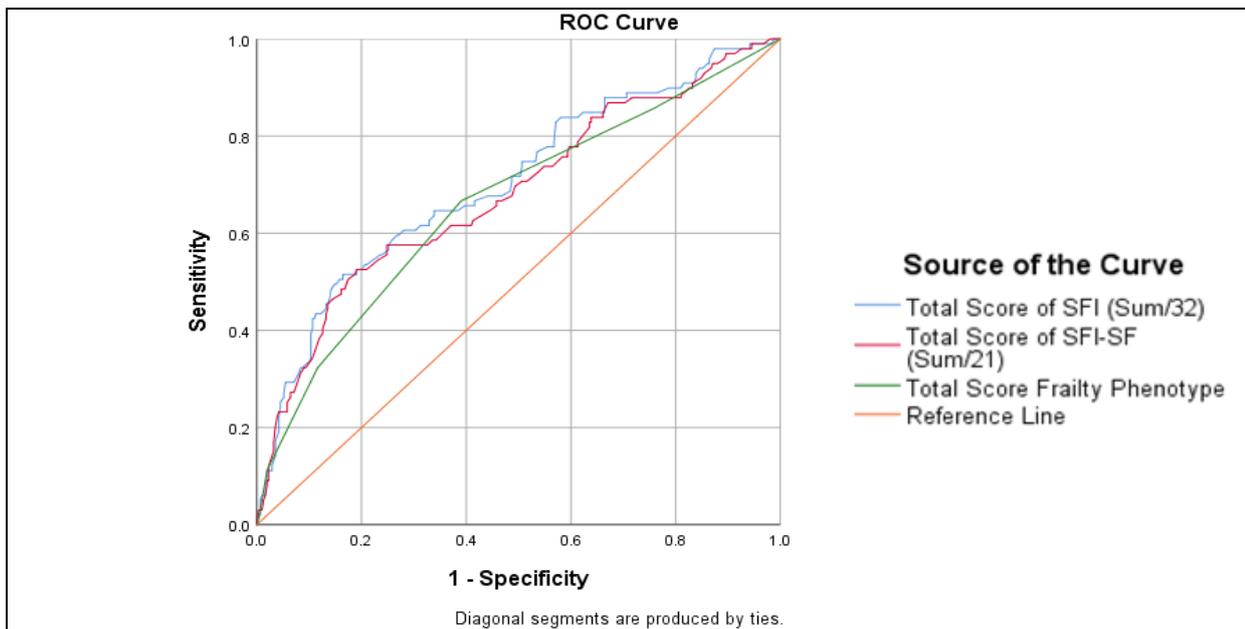


Figure 7.5 ROC analysis of predictive model for mortality (Waves 2-4)

An analysis of C-statistics in Table 7.7 revealed that the SFI-21, SFI-32 and frailty phenotype had similar values of 0.704, 0.682 and 0.662 respectively, indicating that there was no statistically significant difference in the discriminant ability of these measures to predict mortality over six years.

Table 7.7 Area Under the Curve in ROC Analysis (Waves 2-4)

Test Result Variable(s)	Area
Total Score of SFI (Sum/32)	.704
Total Score of SFI-SF (Sum/21)	.682
Total Score Frailty Phenotype	.662

Multinomial logistic regression was performed to examine the impact of independent variables on frailty status. Adjusted models were used to test the association between frailty status and individual characteristics including presence of Down syndrome, age category, residence type and gender based on the SFI-21, SFI-32 and frailty phenotype. Separate models were fitted using high frailty as the reference group to assess the odds of low and intermediate frailty in the context of each factor. For a multinomial logistic model, an outcome group is used as the reference group, and the “coefficients for all other outcome groups describe how the independent variables are related to the probability of being in that outcome group versus the reference group” (UCLA 2021).

Odds Ratio (OR) was reported as a measure of association between an exposure and an outcome (IBM 2021). Exponentiation of the coefficients (Exp(B), reported as Odds Ratio (OR), describe the logistic regression equation using each covariate to predict the log odds of frailty status. Logistic coefficient (B) for each predictor variable for each alternative category of the outcome variable indicates the expected amount of change in the logit for each one unit change in the predictor. The logit is what is being predicted; it is the odds of membership in the category of the outcome variable which has been specified. The closer a logistic coefficient is to zero, the less influence the predictor has in predicting the logit (Starkweather and Moske 2011). By default, Multinomial Logistic Regression uses the last (highest) category level as the reference category for the dependent variable (DV) (IBM 2021). Maximum likelihood estimation was used to evaluate the probability of categorical membership. To address multicollinearity, severe and profound level of ID were linearly combined. Similarly, two categories of residence type, “independent living/ family home” and “community group” were combined due to strong linear relationship.

Table 7.8 Multinomial logistic regression model parameter estimates (SFI-32)

Intervals	Low (OR 95% CI)	Wald	Intermediate (OR 95% CI)	Wald
Down Syndrome	0.271**(0.101-0.726)	6.746	0.407*(0.183-0.901)	4.915
Other Cause of ID	1 (Reference)	.	1 (Reference)	.
<50	14.934**(4.171-53.467)	17.260	2.974*(1.12-7.897)	4.782
50-64	7.04**(2.432-20.381)	12.947	1.55 (0.705-3.408)	1.190
65+	1 (Reference)	.	1 (Reference)	.
Indep./Family/ Group Home	4.232**(1.780-10.061)	10.658	1.631(0.833-3.194)	2.035
Residential Care	1 (Reference)	.	1 (Reference)	.
Male	1.814(0.777-4.236)	1.895	2.676**(1.316-5.444)	7.385
Female	1	.	1	.

Reference category = High; Wald: Wald chi-square test ** OR: Odds Ratio; 95% CI: 95% Confidence Interval; * p < 0.05; ** p < 0.01; *** p < 0.001

Table 7.8 posits the multinomial logistic regression model for SFI-32 with potential risk factors. In this model, Down Syndrome had an OR = 0.271(95% CI 0.101-0.726) for low frail group to high frail group. For the intermediate frail group to high frail group, presence of Down syndrome had OR = 0.407 (95% CI 0.183-0.901). Participants aged <50 were approximately 15 times more likely (OR 14.934, 95% CI 4.171-53.467) to

have low frailty, and approximately three times more likely (OR 2.974 95% CI 1.12-7.897) to have intermediate frailty compared to high frailty. On the other hand, the multivariable-adjusted OR for the age category 50-64 was associated with a sevenfold increase in risk of low frailty (OR 7.04, 95% CI 2.432-20.381), but not intermediate frailty. Compared with participants living in residential care, those living independently/with family or in group home had a fourfold higher risk of having low frailty (OR 4.232, 95% CI 1.780-10.061). In relation to gender, male participants had an increased risk of intermediate frailty (OR 2.676, 95% CI 1.316-5.444) in comparison to female participants, but not with low frailty.

Table 7.9 Multinomial logistic regression model parameter estimates (SFI-21)

Intervals	Low (OR 95% CI)	Wald	Intermediate (OR 95% CI)	Wald
Down Syndrome	0.294*(0.111-0.780)	6.048	0.638 (0.293-1.390)	1.278
Other Cause of ID	1 (Reference)	.	1 (Reference)	.
<50	8.339**(2.701-25.743)	13.600	1.827 (0.771-4.329)	1.877
50-64	5.306**(1.986-14.173)	11.081	1.795 (0.837-3.850)	2.255
65+	1 (Reference)	.	1 (Reference)	.
Indep./Family/ Group Home	8.399**(3.651-19.323)	25.059	2.229**(1.178-4.22)	6.061
Residential Care	1 (Reference)	.	1	.
Male	1.866 (0.849-4.102)	2.409	2.627**(1.372-5.03)	8.497
Female	1	.	1	.

Reference category = High; Wald: Wald chi-square test ** OR: Odds Ratio; 95% CI: 95% Confidence Interval; * p < 0.05; ** p < 0.01; *** p < 0.001

Results of multinomial logistic regression for social frailty based on SFI-21 are shown in Table 7.9. The presence of Down syndrome was associated with a threefold increase in risk of having low frailty (OR 0.294*, 95% CI 0.111-0.780), while there was no statistically significant association with intermediate frailty. Age of participants was associated with high OR value for low frailty only. Participants aged <50 had an eightfold increase of having low frailty compared to high frailty (OR 8.339, 95% CI 2.701-25.743) while participants aged 50-64 had a significant OR value of 5.306 (95% CI 1.986-14.173). In relation to residence type, living independently, with family or in a group Home was associated with significant OR values for low frailty (OR 8.399, 95% CI 3.651-19.323) and high frailty (OR 2.229, 95% CI 1.178-4.22). Additionally, the multivariable-adjusted model found that male participants had 2.6 times (OR 2.627,

95% CI 1.372-5.03) greater risk of intermediate frailty in comparison to female participants, but not with low frailty.

Table 7.10 Multinomial logistic regression model parameter estimates (Phenotype)

Intervals	Robust (OR 95% CI)	Wald	Pre-Frail (OR 95% CI)	Wald
Down Syndrome	0.578 (0.236-1.416)	1.440	0.522 (0.243-1.120)	2.786
Other Cause of ID	1 (Reference)	.	1 (Reference)	.
<50	3.238*(1.113-9.419)	4.652	1.671(0.730-3.822)	1.478
50-64	3.545**(1.39-9.041)	7.019	1.838(0.898-3.761)	2.773
65+	1 (Reference)	.	1 (Reference)	.
Indep./Family/ Group Home	1.578(0.744-3.345)	1.413	1.167(0.633-2.152)	.246
Residential Care	1 (Reference)	.	1 (Reference)	.
Male	3.21**(1.498-6.881)	8.989	2.838**(1.468-5.485)	9.623
Female	1 (Reference)	.	1 (Reference)	.

Reference category = Frail; Wald: Wald chi-square test ** OR: Odds Ratio; 95% CI: 95% Confidence Interval; * p < 0.05; ** p < 0.01; *** p < 0.001

Table 7.10 summarises the multinomial logistic regression model for the frailty phenotype. In contrast to the models previously discussed, no statistically significant values were identified for presence of Down syndrome. Mild level of ID had a significant OR values of 14.502 (95% CI 3.903-53.889) and 7.894 (95% CI 2.463-25.306) for robust and pre-frailty respectively. Moderate ID was associated with robust frailty status (OR 2.923, 95% CI 1.239-6.895), but not pre-frailty. In comparison to participants aged 65+, those aged <50 and 60-64 were 3.2 times (OR 3.238, 95% CI 1.113-9.419) and 3.5 times (OR 3.545, 95% 1.39-9.041) more likely to be robust. However, this association was not observed for pre-frailty status. The multivariable-adjusted OR found no significant values for type of residence in relation to frailty phenotype. Compared to female participants, males had a threefold increase in risk of being robust (OR 3.21, 95% CI 1.498-6.881) and pre-frail (OR 2.838, 95% CI 1.468-5.485).

Finally, logistic regression was used to estimate the OR and 95% CI for the association between mortality and social frailty alone, physical frailty alone, and both social frailty and physical frailty, adjusting for expected associated factors Down syndrome, age category, gender and type of residence (Table 7.11). In the unadjusted model, all three

frailty measures are associated with an increased risk of mortality including SFI-32 (OR 5.922, 95% CI 3.24-10.827), SFI-21 (OR 4.276, 95% CI 2.49-7.342), and frailty phenotype (OR 3.635, 95% CI 2.106-6.275). In the adjusted model with expected risk factors, these measures also had significant OR values of 4.653 (95% CI 2.293-9.442), 3.798 (95% CI 1.947-7.41 and 2.993 (95% CI 1.584-5.655) respectively.

Table 7.11 Logistic regression model for social and physical frailty alone

Intervals	SFI-32 Alone (OR 95% CI)	SFI-21 Alone (OR 95% CI)	Phenotype Alone (OR 95% CI)
Model 1	5.922*** (3.24-10.827)	4.276*** (2.49-7.342)	3.635** (2.106-6.275)
Model 2	4.653***(2.293-9.442)	3.798***(1.947-7.41)	2.993**(1.584-5.655)
Down Syndrome	0.240***(0.129-0.449)	0.222***(0.119-0.412)	0.209***(0.107-0.412)
Age Category	2.452***(1.668-3.603)	2.56***(1.744-3.757)	2.541***(1.679-3.845)
Gender	0.939(0.573-1.539)	0.963(0.589-1.575)	0.918(0.535-1.575)
Residence Type	1.806*(1.073-3.039)	1.672(0.992-2.82)	2.073*(1.186-3.623)

Model 1: Unadjusted in the analysis of each factor; Model 2: Model 1 plus Down syndrome, ID level, age category, gender and residence type; OR: Odds Ratio; CI: Confidence Interval; * p < 0.05; ** p < 0.01; *** p < 0.001

The presence of Down syndrome was found to increase the risk of mortality approximately 0.2 times based on SFI-32 (OR 0.240, 95% CI 0.129-0.449), SFI-21 (OR 0.222, 95% 0.119-0.412) and frailty phenotype (OR 0.209, 95% CI 0.107-0.412). Age category was associated with a 2.5-fold increase in mortality for participants with high tertile scores for the SFI-21 (OR 2.452, 95% CI 1.668-3.603) or SFI-32 (OR 2.56, 95% CI 1.744-3.757), and those categorised as frail according to phenotype criteria (OR 2.541, 95% 1.679-3.845). Additionally, type of residence was associated with the SFI-32 (OR 1.806, 95% CI 1.073-3.039) and frailty phenotype (OR 2.073, 95% CI 1.186-3.623), but not for the SFI-21. No statistically significant values were found for gender.

Table 7.12 posits the relationship between mortality with social frailty (SFI-32 or SFI-21) and physical frailty (frailty phenotype) combined. In the unadjusted model, risk of mortality was increased 4.5 times for SFI-32 and frailty phenotype combined (OR 4.580, 95% CI 2.143-9.798) and 6.4 times for SFI-21 and frailty phenotype combined (OR 6.443, 95% CI 2.871-14.460). After adjustment of expected risk factors, similar associations were found with SFI-32 and frailty phenotype combined (OR 3.917, 95%

CI 1.627-9.431) and SFI-21 and frailty phenotype combined (OR 4.686, 95% CI 1.901-11.55).

Table 7.12 Logistic regression model for social and physical frailty combined

Intervals	SFI-32 and Frailty Phenotype (OR 95% CI)	SFI-21 and Frailty Phenotype (OR 95% CI)
Model 1	4.580** (2.143-9.798)	6.443** (2.871-14.460)
Model 2	3.917**(1.627-9.431)	4.686**(1.901-11.55)
Down Syndrome	0.213**(0.115-0.393)	0.228**(0.123-0.423)
Age	2.512**(1.714-3.681)	2.442**(1.663-3.588)
Gender	0.944(0.578-1.542)	0.951(0.582-1.553)
Residence Type	1.686**(1.142-2.489)	1.70**(1.148-2.516)

Model 1: Unadjusted in the analysis of each factor; Model 2: Model 1 plus Down syndrome, ID level, age category, gender and residence type; OR: Odds Ratio; CI: Confidence Interval; * p < 0.05; ** p < 0.01; *** p < 0.001

Down Syndrome was found to be associated with a 0.2-times increase in risk of mortality based on SFI-32 and frailty phenotype combined (OR 0.213, 95% CI 0.115-0.393) and SFI-21 and frailty phenotype combined (OR 0.228, 95% CI 0.123-0.423). Age of participants was associated with an increased likelihood of mortality for both SFI-32 and frailty phenotype combined (OR 2.512, 95% CI 1.714-3.681) and SFI-21 and frailty phenotype combined (OR 2.442, 95% CI 1.663-3.588). Type of residence was also significantly associated with mortality based on these combined measures, with OR values 1.686 (95% CI 1.142-2.489) and 1.70 (1.148-2.516) respectively. No statistically significant values were found for gender.

7.5 Conclusion

In this chapter, results of statistical analysis performed to address key study objectives have been reported. Social frailty indices and the modified frailty phenotype developed by O'Connell (2020) were used to operationalise social frailty and physical frailty respectively in the study sample. Characteristics of the sample in relation to these measures were analysed using descriptive statistics. Venn Diagram was utilised to evaluate the overlap between social frailty and physical frailty among participants. It was found that there were participants who were socially frail who were not evaluated by physical frailty.

The prevalence of social frailty alone was revealed to be marginally less than physical frailty alone, while social frailty and physical frailty overlapped for a significant proportion. The individual and combined ability of measures for social frailty and physical frailty in predicting mortality three and six years later among participants was analysed using ROC. It was found that individually these measures had good and similar discriminate capacity to predict mortality over this timeframe. Furthermore, capacity to predict mortality improved when these measures were used in combination. Multinomial logistic regression was used to examine the association between frailty status and differences in age, gender, presence of Down syndrome, and type of residence. All independent variables except for gender were shown to be associated with frailty status. Finally, logistic regression was performed to evaluate the association between mortality and social frailty alone, physical frailty alone, and both social frailty and physical frailty combined, adjusting for expected associated factors. High scores in all measures were associated with mortality with an increased risk associated with Down syndrome, age category, and type of residence, but not gender.

In the next chapter, the significance of these findings is explained and interpreted in the context of the literature review and research questions. The potential implications of these insights for research and practice are then considered.

Chapter 8: Discussion and Conclusion

8.1. Overview

The primary aim of this study was to obtain a holistic understanding of the characteristics of social frailty among older adults with ID. To address this aim specific objectives were set including the construction and validation of a social frailty measure, analysis of the prevalence of social frailty and associated factors, examination of the relationship between social frailty and physical frailty, and assessment of the capacity of social frailty to predict mortality. This chapter critically examines the findings presented in Chapters 5 and 6 and considers the extent to which these have addressed study objectives. This is followed by discussion of the potential limitations of the study including characteristics of the study's methodology which may have impacted or influenced the interpretation of findings. The potential clinical and research implications of study findings are then considered in advance of concluding remarks by the researcher.

8.2 Summary of Research

An analysis of descriptive statistics yielded new insights regarding the characteristics of social frailty among older adults with ID. The prevalence of social frailty and associated factors were analyzed. Findings revealed that physical frailty was more prevalent in comparison to social frailty. A significant proportion of the study sample had both physical frailty and social frailty, suggesting an overlap between these health states. These measures were shown to have good and similar predictive capacity for mortality three to six years later, with predictive power maximized when combined. Furthermore, this study revealed that numerous factors are associated with higher levels of social frailty and increased risk of mortality including greater severity of ID, the presence of Down syndrome, and living in a congregated setting. In the following sections, a comprehensive description of findings is interpreted in the context of relevant literature.

8.3 Discussion and Interpretation of Findings

8.3.1 Construction and Validation of a Social Frailty Measure

This study utilised the deficit accumulation model (Mitnitski *et al.* 2001) to develop a SFI comprising of self-report variables relating to social deficits based on the conceptual model of social frailty by Bunt *et al.* (2017). While different approaches to the measurement of social frailty have been developed, the utility of a FI in operationalising this health state has been repeatedly demonstrated (Armstrong *et al.* 2015, Wallace *et al.* 2015, Shega *et al.* 2012, Andrew and Rockwood 2010, Andrew *et al.* 2008). This contrasts with the approach taken by others in measuring this health state according to specific criteria (Park *et al.* 2019, Tsutsumimoto *et al.* 2017, Makizako *et al.* 2015, Gobbens *et al.* 2010, Garre-Olmo *et al.* 2013, Lozupone *et al.* 2018). The main advantage of adopting the deficit accumulation model in this study was that it enabled the construction of a population specific measurement of social frailty using IDS-TILDA data. Additionally, the decision to utilise the deficit accumulation model was also based on its validated use among older adults with ID (McKenzie *et al.* 2017).

This study demonstrated that the conceptual model of social frailty (Bunt *et al.* 2017) can be used to guide the selection of variables for inclusion in a SFI. This differed from the approach taken by other studies which selected deficits based on findings of a literature review and expert consensus (Andrew *et al.* 2008). As in other studies involving the development of a frailty measure, factor analysis was performed as part of the construction and validation of the SFI (Wilmer *et al.* 2019, Lin *et al.* 2016, Zhang *et al.* 2017, Kamaruzzaman 2010). This approach was used to assess the underlying factor structure of the SFI without imposing a preconceived structure on the outcome (Child 1990). This was also useful as a variable reduction technique to remove items not significantly contributing to the measurement constructed and to assess the internal reliability of remaining items.

Further to the EFA, it was revealed that the SFI had four domains which according to the conceptual model of social frailty (Bunt *et al.* 2017) could be qualitatively interpreted as variable subsets relating to ADL, ICT use and access, communication and social participation, and life events. Furthermore, as the first known operationalisation of social frailty among older adults with ID, it is incumbent upon this study to consider the potential relevance of these variables in the lives of older adults with ID. The first domain comprised of variables relating to difficulties in completing ADL including getting in or out of bed, using the toilet, walking across a room, dressing, bathing/showering, and eating. ADL impairment is closely linked to the concept of social frailty given its association with adverse frailty outcomes (O'Halloran *et al.* 2018, Hsu and Chang 2014, Kawano-Soto *et al.* 2012, Aranda *et al.* 2011, Bilotta *et al.* 2010). As shown by the systematic literature review in Chapter 3, difficulties in performing ADL are associated with adverse transitions and trajectories of frailty among older adults with ID (Martin *et al.* 2018, Ouellette-Kuntz *et al.* 2018). It is worth noting that functional impairment is a prevalent issue for many people with ID who may have had fewer opportunities to develop ADL and IADL skills due to living most of their lives in congregated settings (Taylor *et al.* 2005).

Items relating to leisure also had high factor loadings in the first domain including regular physical activity, taking a holiday locally (in Ireland), and difficulties participating in social activities outside the home. It is possible to draw a connection between these items and ADL impairment, whereby the same difficulties affecting the individuals ability to perform ADL may also directly or indirectly affect their ability or opportunities to engage in physical activity, go on a holiday trip, and engage in social activities outside the home. Furthermore, it is important to note that older adults with ID tend to have low levels of physical activity (McCarron *et al.* 2014) and oftentimes have difficulties participating in social activities (McGlinchey *et al.* 2019, McCarron *et al.* 2014). Conceptually, this subset of variables relates strongly to the general resources domain of the conceptual model of social frailty (Bunt *et al.* 2017), which describes ADL and lifestyle as resources that are beneficial in a general manner that indirectly contribute to social need fulfilment. As discussed in Chapter 3, functional impairment has been previously identified as a risk factor for overall frailty among older adults with ID (Martin *et al.* 2018).

The second domain of the SFI identified through EFA related to ICT use and access. Variables included having access to a computer/ laptop/ tablet/ smartphone and being able to turn on a computer, type name on a keyboard, type a letter, send an email, use social media sites, and look up topics of interest online. While not explicitly described in the original conceptual model of social frailty by Bunt *et al.* (2017), the rationale for including these items in the index was that ICT use and access can be viewed as resources beneficial in a general manner that indirectly contribute to social need fulfilment. As discussed in Section 1.5.3, the impact of ICT on the determinants of health and its profound implications for the operationalisation of health determinants has been previously recognised (Rice and Sara 2019). It is becoming increasingly clear that that regular and self-determined use of mobile technology and apps is associated with improved social inclusion among older adults with ID (Martin *et al.* 2021, Murphy *et al.* 2019). As in the general population, many people with ID use ICT to develop and maintain social connections (Martin *et al.* 2021, Chiner *et al.* 2017; Alfredsson Ågren *et al.* 2020). However, low levels of ICT use and access among older adults with ID has been reported elsewhere (Boot *et al.* 2018, McCausland *et al.* 2017, McCarron *et al.* 2017, McCarron *et al.* 2014).

It was interesting to note the significant factor loadings for the item pertaining to having voted in any recent election. The act of voting is a form of social engagement and therefore relates strongly to the concept of social frailty. Previous studies have shown that older adults with ID are less likely to engage in this activity in comparison to those in the general population (McCarron *et al.* 2014, McCarron *et al.* 2011). Furthermore, older adults with ID tend to vote less with advancing age (McCarron *et al.* 2014, McCarron *et al.* 2011). This is in stark contrast to older adults in the general population who ordinarily have high voting turnout, which increases with advancing age (TILDA 2019, Goerres 2007). Also included in the SFI were items in relation to neighbourhood involvement including both giving and receiving help to/from their friends and/or neighbours in the last two years. As discussed in Chapter 2, there is a clear link between neighbourhood involvement and frailty status (Cramm and Nieboer 2013, Aranda *et al.* 2011, Woo *et al.* 2005). Indeed, neighbourhood involvement, the presence of neighbours and receiving help from others have been previously identified as being significantly relevant to the concept of social frailty (Bunt *et al.* 2017).

Items in relation to difficulties speaking or being understood and being able to follow a conversation with others qualified for inclusion in the SFI. It has previously been shown that most people with ID have some form of communication difficulty (Emerson *et al.* 2012). Many individuals with ID do not develop speech or have limited speech and language abilities (Peeters and Gillberg 1999). It has been shown that communication difficulties may be present at all severity of ID. People with mild ID often have superficially adequate speech and language skills, while more subtle communication problems only become apparent through further investigation (Healy and Walsh 2007). On the other hand, people with severe or profound ID oftentimes have limited speech comprehension and are reliant on non-verbal forms of communication (Bellamy *et al.* 2010, Maes *et al.* 2007, Hogg *et al.* 2001). Due to the idiosyncratic nature of their communication and lack of verbal communication, this group are particularly at risk of being misunderstood or misinterpreted by others (Grove *et al.* 1999). Hearing impairment is also prevalent in this population (Emerson 2001) which oftentimes goes unrecognised, leading to exacerbation of communication difficulties (Haveman 2004).

The fourth domain of the SFI, comprised of variables in relation to life events. These items were included in IDS-TILDA as part of a 20-item checklist, adapted from the Life Events Scale developed by Hermans and Evenhuis (2012). Life events can be described as an experience with an ascertainable source and limited duration, which can influence someone's psychological status and change their social and/or physical environment (Coe *et al.* 1999). The accumulation of life events, especially those which are negative, is associated with a higher frequency of increased depressive and anxiety symptoms and major depression (Herman and Evenhuis 2012). There is evidence to suggest that life events may adversely impact mental health in people with ID (Tsakanikos 2006). It is notable that most of the life events included were in relation to a person's social environment. Beginning with change of key worker, this role is ordinarily assigned to a member of the staff team supporting the individual who obtains an enhanced familiarity with the person's needs and liaises between individuals important to their life (Hull and Turton 2014, Whitehouse *et al.* 2000, Greco and Sloper 2004). Current evidence suggests that a change in keyworker is a commonly experienced life event among older adults with ID (McCarron *et al.* 2017, McCarron *et al.* 2014). Similarly, bereavement of a friend has also been shown to be prevalent in this population (McCarron *et al.* 2017, McCarron *et al.* 2014) and has been found to

have an adverse impact on mental health among people with ID (Lyons *et al.* 2000, MacHale 2002).

Change in frequency of visits from family and/or friends was another life event represented in the SFI. Previous studies have shown that many older adults with ID are at a heightened risk of being socially excluded due to having fewer social contacts and lower levels of community participation (McCausland *et al.* 2021, Mithen *et al.* 2015). Therefore, any potential disruption of existing social networks and participation has the potential to have a disproportionately negative impact on their well-being (McCausland *et al.* 2021). Other items with significant factor loadings for this domain of the SFI also have significance for older adults with ID. For instance, changes of staff in home or day service, changes at or from work or day service, and new resident moving into home have been shown to be frequently occurring life events for this group (McCarron *et al.* 2017, McCarron *et al.* 2014).

The inclusion of items relating to day services is particularly pertinent to older adults with ID given their high level of use of these services (McCarron *et al.* 2014). From a social frailty perspective, day services provide opportunities to access a wide range of resources important for achieving social need fulfilment such as social and recreational activities, training, and employment. The inclusion of an item in relation to major illness or injury is particularly noteworthy, as older adults with ID are at an increased risk of experiencing these life events in comparison to those in the general population (McCarron *et al.* 2017, McCarron *et al.* 2014, McCarron *et al.* 2011). Based on guidelines developed by Searle *et al.* (2008), the SFI-32 meets the criteria of having at least 30 items which is required for a stable index capable of accurately predicting adverse outcomes. The number of items included in both social frailty measures, SFI-32 and SFI-21, is comparable to those developed in other studies. For instance, Andrew *et al.* (2008) constructed two indices comprising of 23 and 40 items, while Shega *et al.* (2012) included 39 items in their index. In the study by Wallace *et al.* (2015), two indices were constructed using 32 and 57 items respectively, whereas Armstrong *et al.* (2015) included the largest number of deficits at 58.

As part CFA, component loadings of the EFA model led to the identification of 21 items with high factor loadings. Similar to the approach taken by Kamaruzzaman *et al.* (2010), the latent structure of a CFA model comprising of these 'weighted' loadings was performed with the intention of increasing the reliability of the measure. Analysis of the goodness-of-fit of the CFA model suggested moderate to good internal consistency of the SFI-21. Furthermore, the four domains underpinning the 21-item CFA model did not correlate significantly with one another, indicating a multidimensional measure (Taylor 1990, Campbell and Fiske 1959). It should however be noted that there is a lack consensus regarding how goodness-of-fit indices should be interpreted, with some even calling for their use to be abandoned altogether. Nonetheless, validity of the SFI-21 was reaffirmed by findings that this measure had similar characteristics and discriminant capacity for predicting mortality as an adverse outcome in comparison to full 32-item index. This is an important finding of this study as it suggests that social frailty may be operationalised using a smaller subset of variables.

8.3.2 Prevalence of Social Frailty and Associated Factors

Based on SFI-32 and SFI-21 total scores, high levels of social frailty were present among 17% and 20.5% of study participants respectively. A similar prevalence of social frailty (20.6%) was found in the study by Park *et al.* (2019). However, there is significant diversity in the prevalence of social frailty reported in older adult populations, ranging between three and thirty-six percent while other studies have reported a slightly lower rate ranging from 18–18.4% (Park *et al.* 2019, Makizako *et al.* 2018, Yamada and Arai 2018, Yamada *et al.* 2018, Teo *et al.* 2017, Makizako *et al.* 2015). However, due to the ambiguity regarding the measurement of social frailty, the meaningfulness of comparing prevalence rates is questionable. SFI scores were found to be normally distributed in the study sample, as indicated by the symmetry of the histogram. This accords with the findings of other studies which show that social frailty defined as the accumulation of deficits has a relatively normal distribution among older adults in the general population (Andrew *et al.* 2008, Wallace *et al.* 2014). However, it should be noted that right-skewed distribution of index scores has been observed in previous studies of social frailty (Armstrong *et al.* 2015) and overall frailty (Schoufour *et al.* 2016, Mitnitski *et al.* 2013, Seale *et al.* 2008).

The distribution of social frailty according to the SFI-32 was such that no participant was free of all social deficits, reflecting findings by others (Shega *et al.* 2012, Andrew *et al.* 2008). However, a null score was observed for the 21-item weighted index. Another important finding was that the mean scores for these measures were .46 and .45 respectively, which is almost twice as high as that reported in other studies involving older adults in the general population (Armstrong *et al.* 2015, Shega *et al.* 2012, Andrew *et al.* 2008). This study obtained new insights into the individual characteristics associated with frailty status among older adults with ID. The variables of age range, type of residence, and presence of Down Syndrome were entered in the logistic regression.

Supporting previous findings, individuals with the highest frailty levels tended to be older (Park *et al.* 2019, Armstrong *et al.* 2015, Andrew *et al.* 2008). Participants in the oldest age category of 65+ were more likely to be in the highest tertile for the SFI-32 (16%) in comparison to those aged 50-64 (11%), and under 50 years (9%). Similarly, having an age of 65+ was associated with a high tertile SFI-21 score in contrast to those in the age categories 50-64 (12%) and <50 (7%). Furthermore, findings of logistic regression also support the association between age and social frailty status. Participants in the youngest cohort (aged <50 years) were 15 times more likely to be in the lowest social frailty tertile for SFI-32, and eight times less likely based on SFI-21 ($p < 0.01$). These findings align with those of other studies reporting a significant association between advancing age and overall frailty (O'Halloran *et al.* 2018, Etman *et al.* 2012, Cramm and Nieboer 2013, Harttgen *et al.* 2013, Harttgen *et al.* 2013, Jürschik *et al.* 2012, Kawano-Soto *et al.* 2012, Alvarado *et al.* 2008). It is reasonable to expect that resilience would reduce with advancing age, with the accumulation of deficits having a more profound effect on health status and thereby mortality (Schoufour *et al.* 2017). It is however noteworthy that Makizako *et al.* (2018) reported no distinct differences in frailty status across age cohorts in their study.

As found in previous studies of social frailty (Park *et al.* 2019, Andrew *et al.* 2008), more female participants had higher levels of social frailty in comparison to men according to SFI-32 (15% vs 6%) and SFI-21 (18% vs 8%). It is also interesting to consider these findings in the context of the growing evidence that women are at a heightened risk of having poorer frailty status in comparison to men (O'Halloran *et al.*

2018, Etman *et al.* 2012, Herrera-Badilla *et al.* 2015, Harttgen *et al.* 2013, Woo *et al.* 2005, Kawano-Soto *et al.* 2012, Jürschik *et al.* 2012). The reason for differences in the characterization of social vulnerability based on gender remains unclear and warrants further research. It has however been previously asserted that women may be more susceptible to experiencing adverse frailty outcomes due to a combination of biological, behavioural and social factors (Gordon and Hubbard 2020, Gordan and Hubbard 2018, Hubbard 2015, Gobbens *et al.* 2010a).

This study revealed that participants with high levels of social frailty were more likely to have Down syndrome as cause of ID in comparison to any other aetiology (16% vs 14%). As the first known study to explore the relationship between social frailty and Down syndrome it was not possible to compare findings with those of others. However, it should be noted that in terms of overall frailty, the presence Down syndrome is associated with adverse transitions and trajectories (Martin *et al.* 2018, Ouellette-Kuntz *et al.* 2018). One possible explanation for the observed correlation between Down syndrome and social frailty may be due to there being an overlap between social disparities prevalent in this population and the construct of social frailty used. For instance, it has been repeatedly shown in other studies that the presence of Down syndrome is associated with an increased likelihood of sensory, physical, mental, and cognitive health disparities (McCarron *et al.* 2017, McCarron *et al.* 2014, McCarron *et al.* 2011). These difficulties may affect the individuals ability to communicate, participate in their community, and maintain functional independence – all of which are considered important resources, activities and behaviours for social need fulfilment (Bunt *et al.* 2017). Given that the SFI comprised of items in relation to these factors it is interesting to hypothesize that the accumulation of social related issues prevalent among in this group may have contributed to a high index score.

Another key finding of this study was that several factors associated with social frailty status have unique relevance to older adults with ID. Firstly, level of ID was found to be associated differences in social frailty status. Only one participant with a mild ID was identified as having a high level of social frailty based on either the 21- or 32-item index. Contrastingly, participants with a high tertile score were more likely to have a moderate ID (N=21), or severe/profound ID (N=29) based on SFI-32. This disparity was magnified with the SFI-21, which measured a higher prevalence of social frailty in

the moderate ID (N=23) and severe/profound ID cohorts (N=41). This finding may be explained by the fact that several variables constituting the SFI have unique relevance to this sub-group. For instance, ADL difficulties are typically associated with higher levels of ID. People with a severe or profound ID are more likely to communicate non-verbally which without the necessary supports may impair their interaction with others (Griffith *et al.* 2017, Bellamy *et al.* 2010, Iacono *et al.* 2009, Maes *et al.* 2007, Hogg *et al.* 2001, McLean *et al.* 1999). It has also been shown that older adults with a severe or profound ID are less likely to have purposeful contact with neighbours and opportunities to participate in social activities (McCarron *et al.* 2014, McCarron *et al.* 2011). Furthermore, many older adults with a severe or profound ID reside in congregated settings which places them at an increased risk of lower social participation (McCarron *et al.* 2014, McCarron *et al.* 2011).

In this study it was particularly notable that differences in residence type were associated with social frailty status. Participants living independently, with family, or in a group home were more likely to have a low level of social frailty in comparison to those living in residential care based on SFI-32 (10% vs 35%) and SFI-21 (6% vs 27%). This association was also supported by analysis of the multilinear logistic regression, which showed that this cohort had a four- and eight-times greater odds of having a low level of social frailty based on the 32- and 21-item indices respectively. These results broadly support those of McKenzie *et al.* (2015b) which showed that living in a group home was associated with a reduced likelihood of overall frailty. However, as discussed in Chapter 3, there is a lack of consensus on the role of living situation and prevalence risk of frailty among older adults with ID. In the studies by Oullette-Kuntz *et al.* (2018) and Martin *et al.* (2018) living in a group home environment was associated with poorer frailty outcomes in this population, while others have observed no significant difference in frailty status based on living situation (McKenzie *et al.* 2016a). There is a relative paucity of research on social frailty in institutional settings, but it is reasonable to assume that living in these settings would contribute to social frailty by way of reduced social contact and fewer opportunities for community participation (Andrew *et al.* 2008).

8.3.3 Relationship Between Social Frailty and Physical Frailty

The next objective addressed by this study was the examination of the relationship between social frailty and physical frailty. As described in chapter 4, both domains have been described within the overall frailty concept (Gobbens *et al.* 2010a). However, there is a relative scarcity of research on the association between social and physical frailty. Furthermore, there are no other known studies which have explored the relationship between these health domains specifically in relation to older adults with ID. A significant finding of this study is that despite their use of different criteria, both social frailty and physical frailty appear to be associated with similar characteristics. As in the SFI, phenotypic frailty was more prevalent among those in the 65+ cohort (24%) in comparison to those aged 50-64 (13%) or <50 years (16%). Multinomial logistic regression revealed that physical frailty was also associated with being in the youngest cohort, albeit to a lesser extent.

Another similarity observed was that participants in the highest frailty category were more likely to be female than male (22% vs 9%). Additionally, participants with a diagnosis of Down syndrome had high levels of physical pre-frailty (56%) and frailty (22%). Level of ID also appears to be associated with differences in physical frailty status. Almost one third of study participants with a severe or profound ID identified were identified as physically frail, which was considerably greater than the prevalence risk observed among the moderate ID (18%) and mild ID (4%) cohorts. Furthermore, living in a congregated setting was found to be associated with a two-fold increase in risk of being physically frail in comparison to those living independently, with family, or in a community group home (24% vs 12%). However, type of residence was not found to be significantly associated with a greater odds risk of phenotypic frailty. Findings in relation to the proportion of participants with social frailty alone, physical frailty alone, and both social frailty and physical frailty combined broadly reflected those of other studies involving older adults in the general population. Based on SFI-32 and SFI-21 total scores, high levels of social frailty were present among 17% and 20.5% respectively. In comparison, the proportion of participants with physical frailty was 20.5-21%, depending on the SFI used. This contrasts with the study by Park *et al.* (2019) which found that social frailty was more prevalent than physical frailty (20.6% vs. 16.4%).

As reported by others, this study found that physical and social frailty did not fully overlap (Park *et al.* 2019, Garre-Olmo *et al.* 2013). This finding supports the claim that the main benefit of measuring social frailty is that it may capture a state of vulnerability not evaluated by physical frailty (Park *et al.* 2019). It is interesting to consider how theories underpinning phenotypic frailty (Fried *et al.* 2001) and social frailty (Bunt *et al.* 2017) may be conceptually interlinked in a dynamic and complex relationship. It is plausible that age-related biologic changes characterised by weight loss, sarcopenia, weakness, exhaustion, slowness, and low activity may contribute to or result from lost or a risk of losing resources, activities, and behaviours necessary for social need fulfilment. However, additional longitudinal research is required to establish the causal relationship between social frailty and physical frailty among older adults with ID.

8.3.4 Social Frailty as a Predictor of Mortality

ROC analyses were performed to compare the ability of social frailty and physical frailty to predict mortality over two follow up periods, three and six years after baseline assessment. Supporting the findings of previous studies, social frailty was found to have good discriminative capacity for mortality (Park *et al.* 2019, Andrew *et al.* 2008). Areas under ROC (AUCs) for SFI-21 and SFI-32 were satisfactory and close in magnitude for prediction of mortality. The finding that SFI-21 had similar predictive ability for all-cause mortality as the SFI-32, despite comprising of a smaller subset of items, was another key finding of this study. In the predictive model for mortality after three years, AUC for SFI-32 was 0.76, which was slightly but not significantly lower than the value for the SFI-21 (0.75). Similarly, the full index had a slightly higher AUC value than the shortened version for all-cause mortality over a period of approximately six years (.60 vs .58). Another interesting finding was that AUC value for the frailty phenotype (.78) was greater than both social frailty measures for mortality after three years. Conversely, the frailty phenotype obtained a lower AUC value (.56) for the six-year predictive model, which was slightly lower than that of the SFI-32. This may suggest that, in comparison to the frailty phenotype, the SFI-32 has moderately better detection ability and lower probability of erroneously predicting mortality over a longer timeframe.

It has been asserted that ROC analysis is useful in determining optimal cut-off points of screening tools which may have practical diagnostic significance (Hajian-Tilaki 2013). As discussed in section 5.8.1, there are no established cut-points for social frailty. However, previous studies of social frailty have tended to use tertile or quartile index scores for the purpose of conducting statistical analysis (Wallace *et al.* 2014, Andrew and Rockwood 2010, Andrew *et al.* 2008). As demonstrated by the ROC analysis, using the highest tertile to determine a state of social frailty proved to be a sensitive predictor of mortality. However, it should be noted that this association has not been observed consistently in the literature. For instance, the study by Gobbens *et al.* (2012) found that social frailty did not predict adverse outcomes beyond a timeframe of two years and was less predictive of adverse outcomes in comparison to physical frailty.

Both social frailty and physical frailty were associated with an increased risk of mortality, especially when both criteria were combined. Adjusting for factors including age category, presence of Down syndrome, gender and residence type, a high tertile score based on the SFI-21 and SFI-32 was associated with a four- and five-fold increased risk of this outcome respectively. The risk of mortality was almost five times greater for individuals identified as having both physical frailty and social frailty based on SFI-21. This association was also observed in relation to the 32-item SFI but to a lesser degree with a four-fold increase in the risk of mortality three to six years later. Characteristics associated with social frailty were largely analogous with those for mortality as an adverse outcome of this health state. For both physical frailty and social frailty, participants with Down syndrome were found to have a 0.2-times greater risk of mortality. Age of participants was significantly associated with an increased likelihood of mortality among participants with physical frailty alone, social frailty alone, or both measures combined. Type of residence was found to be significantly associated with physical frailty and social frailty based on SFI-32, which increased when these measures were combined. However, gender was not found to be a risk factor for mortality among socially frail participants.

8.4 Limitations

The findings of this study must be interpreted with caution on account of potential limitations. Firstly, the two social frailty indices constructed for this study are new measures. While findings of this study show that these are robust measures capable of accurately predicting mortality, further research is required to enhance our understanding of their validity and properties. Secondly, the social frailty measures comprised entirely of self-report data which can be less accurate than objective measures (Armstrong *et al.* 2015, Andrew *et al.* 2008). It is therefore possible that some individuals may have under- or over-reported their level of social frailty. However, there is also the view that self-perceived frailty may be more relevant to a person's subjective quality of life and health in comparison to objective measures (Andrew *et al.* 2008).

Factor analysis is considered a useful means of exploring the possible underlying factor structure of a set of observed variables and removing items not significantly contributing to the measurement construct (Child 1990). While this is considered a traditional approach to constructing and validating frailty scales and indices (Kamaruzzaman 2010), there are others who argue the arbitrariness of using factor analysis, such as the ability of the researcher to specify the number of dimensions to be discovered (Andrew *et al.* 2008). Furthermore, it has been asserted that the reductionist approach of including fewer variables based on interpretation of factor analysis is counterintuitive to the deficit accumulation approach which aims to measure frailty status according to the number rather than the nature of deficits present (Shega *et al.* 2012, Andrew *et al.* 2008). It should be noted that several variables which were theoretically significant to the social need fulfilment domain of the social frailty concept were excluded from the index due to high numbers of missing cases. These were in relation to subjective questions regarding feelings of loneliness, exclusion, isolation, and lack friendship that could only be answered by participants themselves and not by a proxy, which would have prevented some participants in responding. Consequently, the domain of social need fulfilment is not specifically represented in either SFI, which may be interpreted as an incomplete representation of Bunt's model. However, there is great potential to collect this subjective data in future waves of IDS-TILDA.

Lastly, a possible limitation of this study was the utilisation of phenotypic criteria to measure physical frailty in the sample population. As discussed in section 5.8.2, the reliability of Fried's phenotype in being able to accurately measure physical frailty in the presence of pre-existing health conditions or disabilities may be limited (Brehmer-Rinderer *et al.* 2013). Subsequently, it is possible that some participants in this study were labelled as being physically frail when they were not.

8.5 Recommendations and Implications

8.5.1 Risk Assessment

This study operationalised social frailty according to the deficit accumulation model (Mitnitski *et al.* 2001) while the conceptual model of social frailty (Bunt *et al.* 2017) was effectively used as an evidence-based approach to select variables from the IDS-TILDA dataset for inclusion in a SFI. Based upon findings of the statistical analysis, the SFI can capture a state of vulnerability not captured by the frailty phenotype. Furthermore, the SFI has demonstrated capacity in accurately predicting mortality risk over a timeframe of three to six years. While additional research is required to replicate these findings in other ID populations with different ethnic and cultural backgrounds, it is anticipated that the SFI would perform well as a means of predicting health outcomes and for informing the development of targeted person-focused interventions.

A SFI may provide important information about the survival prediction of older adults with ID over long follow up periods and therefore serve as a prognostic tool for determining risk of mortality as an adverse outcome in this population. However, for the incorporation of any additional assessment, particular emphasis should be placed on minimizing any clinical burden. It should be noted that there is significant potential for service providers supporting individuals with ID to develop similar population-specific indices using routinely collected data. A SFI could feasibly be incorporated as a risk stratification tool to identify socially frail individuals and to support the incitement of targeted interventions aiming to reduce or delay adverse outcomes. It may also be useful to include this measure in annual health assessments and advance care planning to inform future care decisions. It can be argued that a measurement of social frailty which utilises self-report data may be more practical to undertake in practice compared to the administration of objective measures required for a phenotypic measurement which typically requires specialist equipment, time and space (Cesari *et*

al. 2014). Based on study findings, and as recommended by Searle *et al.* (2008), it is advisable to include at least 30 deficits in a SFI. Additionally, this study revealed that a SFI comprising of a smaller subset of variables had similar and good discriminant capacity to predict mortality. As asserted by Kamaruzzaman *et al.* (2010), the use of such a weighted index may translate as an easy and minimally invasive measure to perform in practice. It is however not intended that a SFI should be used as a replacement for assessing risk or as a substitute for other valid frailty assessments. Moreover, the findings of this study indicate that capacity to predict mortality may in fact be enhanced when both of these measures are used together.

While further research is required to understand transitions and trajectories of social frailty over time, it has been previously found that overall frailty status has the potential to deteriorate and/or improve over time (Ouellette-Kuntz *et al.* 2018, Martin *et al.* 2018). Therefore, the continuous and regular assessment of social frailty may be warranted, particularly among 'at-risk' sub-groups identified in this study such as women, individuals aged 65 years and above, individuals with Down syndrome, individuals with severe/ profound ID, and those living in congregated settings. When an individual is identified as being pre-frail or frail, intersectoral collaboration is needed to review existing care arrangements and develop responsive action plans that address the persons individual needs (Xie *et al.* 2008).

8.5.2 Health Promotion

Despite the increased longevity being experienced by people with an ID in recent decades, there is growing concern regarding their predisposition to age-related health decline (Ouellette-Kuntz *et al.* 2015, McCarron *et al.* 2015). There has subsequently been a renewed focus on how best to support frail adults with ID to age successfully in the community (Ouellette-Kuntz *et al.* 2018). Findings of this study suggest that social frailty poses a significant barrier to successful ageing for this population. While further research is required, insights obtained by this study in relation to the characteristics of social frailty may be useful for the development of targeted health promotion interventions aiming to positively influence the health behaviours of individuals and communities as well as the living and working conditions that influence their health. The World Health Organization (WHO) defines health promotion as the process of enabling people to increase control over and to improve their health (WHO 1984). It has been established that providing individuals with information, health

education and life skills training may assist them in making independent and informed decisions conducive to their optimal health (WHO 1986). The topic of health promotion warrants specific consideration due to the fact that people with ID tend to have less involvement in health promotion activities in comparison to those in the general population (Ouellette-Kuntz 2005). Furthermore, there is a relative paucity of health promotion programs designed specifically for this population (An *et al.* 2018).

Based upon the integral conceptual model of frailty (Gobbens *et al.* 2010a) discussed in Section 4.4, life course determinants may adversely affect physiologic reserve, subsequently leading to frailty development and adverse outcomes. This study has found that there are many potential risk factors for social frailty prevalence and mortality as an adverse outcome, including advanced age, female gender, Down syndrome, severe/ profound ID, and living in a congregated setting. Creating a greater level of awareness among people with an ID and their carers regarding these risk factors for social frailty may support a preventative approach to managing this health state.

Furthermore, it is possible to descry further opportunities for health promotion activities by examining the items comprising the social frailty measures developed for this study. Further to the factor analysis of the SFI, it is clear that many social-related factors significantly contribute to social frailty scores, namely ADL, communication, social participation, life events, and ICT use. As discussed in Section 8.3.1, older adults with ID are significantly more likely than their non-disabled peers to experience functional impairment, communication difficulties, lack of community participation, adverse life events, and lack of ICT access. Therefore, it would be reasonable to assume that health promotion actions which directly address these social-related health disparities would positively contribute to lower levels of social frailty. This hypothesis fits well with the conceptual model of social frailty by Bunt *et al.* (2017) which is underpinned by the hypothesis that these are important general resources, social resources, and social activities and behaviours for achieving social need fulfilment.

Given that frailty is a complex and multidimensional concept, there is a risk of focusing on distinct deficits, rather than on the person as a whole and what he/she wants (Ouellette-Kuntz *et al.* 2018). It is a recommendation of this study that any actions aiming to address social frailty should be delivered using a person-centred approach which respects the persons preferences and their right to make decisions regarding their care.

8.5.3 Policy Development

It is also important to consider the possible implications of this study for future policy development. As noted by others, the approach of obtaining a “holistic quantification” of social frailty has great potential relevance for health and social policy due to this health state being linked to multiple clinical and social consequences (Buckinx *et al.* 2015, Andrew *et al.* 2008). The continuous nature of the SFI offers great potential to monitor individual trajectories over time. From a public health perspective, the capacity to measure social frailty at a population level using routinely collected data may assist in informing future social and health policy development. This study highlighted that differences in relation to living situation were associated with social frailty status among older adults with an ID. Participants who were residing in a congregated setting were shown to be significantly more likely to have a higher level of social frailty in comparison to those living independently, with family or in a group home setting. This finding is interesting in the context of current healthcare policy in Ireland calling for the deinstitutionalisation of care for people with ID (HSE 2011). Indeed, the desire to live independently in the community for as long as possible has been widely expressed by individuals with ID and their carers (Bigby 2010). In this spirit, future health and social policy aiming to address social frailty among older adults with an ID should consider environmental accommodations, in-home healthcare options, and supportive technologies which facilitate independent living in the community (Vasunilashorn *et al.* 2012).

8.5.4 Future Research

There is a relative paucity of frailty research in the field of ID and just three teams internationally have conducted research on this topic to date (Ouellette-Kuntz *et al.* 2019). This study has contributed new knowledge by pursuing a novel approach to operationalising frailty based on a social conceptualisation. This has enabled a nuanced exploration of the social domain of frailty which focuses on the relationship

between the individual and their social environment. The deficit accumulation model (Mitnitski *et al.* 2001) and the conceptual model of social frailty (Bunt *et al.* 2017) are established conceptual models in the field of frailty research. However, this is the first known study to utilize these models to construct a social frailty measure for specific use among older adults with ID. As discussed in the opening chapter, several studies have adopted the deficit accumulation model to operationalise overall frailty among older adults with ID (Schoufour *et al.* 2015a, Schoufour *et al.* 2015b, McKenzie *et al.* 2015). However, there are no other known studies which have used the deficit accumulation model to construct a measure of social frailty in this population. While others have utilized social frailty scales and questionnaires with pre-defined criteria (Park *et al.* 2019, Tsutsumimoto *et al.* 2017, Makizako *et al.* 2015), a key advantage of adopting the deficit accumulation approach in this study was that it enabled for social frailty to be measured using non-specific criteria, thereby optimising the use of the available data. Furthermore, the application of the conceptual model of social frailty to identify items for inclusion in the SFI is seemingly unique to this study, though several other studies have utilised Bunts' model as a theory guided approach to develop social frailty scales and questionnaire-based measures (Nagai *et al.* 2020, Pek *et al.* 2020). Nevertheless, further research is needed to test the performance of the SFI and characteristics associated with high levels of social frailty.

Consideration should be given to affirming the findings of this study in other ID populations from different geographical locations. It is plausible that differences in ethnicity and social class experienced by older adult ID populations living in other countries may present with different social frailty characteristics. Additionally, the social conceptualisation of frailty used in this study may have implications for younger cohorts in the ID population. While this study found that social frailty levels tended to be highest among the oldest age cohort, we do not yet understand the characteristics of social frailty in younger age. In line with a lifespan approach to frailty development, it can be assumed that the accumulation of social deficits can occur earlier in life. Indeed, it is possible to visualise younger aged individuals with ID experiencing adverse social circumstance and environments as a result of losing or being at risk of losing general resources, social resources, and social activities and behaviours (Bunt *et al.* 2017). The potential to identify social frailty at a younger age may present opportunities for early intervention aimed at delaying or preventing its progression, and thereby reduce

the risk of experiencing associated adverse outcomes. The testing of these assumptions should be prioritised in future research. Additionally, this study posits a strong relationship between the physical and social domains of frailty. However, it was not possible to establish the causal relationship between these health states. Furthermore, it remains unclear if factors such as age, gender, ID level, presence of Down syndrome, and type of residence are associated with changes in frailty status over time. Obtaining these insights was beyond the scope of this study and requires further longitudinal research using a different study design involving follow-up assessment of social and physical frailty status.

Research focusing on social vulnerability in this population is perhaps timely given current events surrounding the COVID-19 pandemic. Social frailty may provide an enhanced understanding of the social health impact of the COVID-19 pandemic (Lozupone *et al.* 2020), which is currently ongoing at time of writing. From a social frailty perspective, the reduced ability to engage in and access social, leisure and work activities resulting from restrictive public health measures may contribute to contribute to adverse health outcomes and impair access to appropriate interventions and supports. Moreover, it has been asserted that the adverse social health impact of these public health measures may be even more pronounced among people with ID due to having communication difficulties, smaller social networks, fewer social supports, and no support of partners or children of their own (McCarron 2020).

8.6 Conclusion

This study accomplished key objectives with the aim of obtaining an enhanced understanding of social frailty among older adults with ID. By applying seminal concepts in the field of frailty research, it was possible to construct a measurement of social frailty using self-report variables. A state of vulnerability not captured by frailty phenotype criteria was found to be highly prevalent and associated with several expected risk factors including older age, female gender, Down syndrome, severe/profound ID and living in a congregated setting. As in the general population, social frailty was found to overlap to a degree with physical frailty but individually contributed to mortality over a three- to six-year timeframe. Given the paucity of research on social frailty there is a clear need for further research to arrive at a more precise and differentiated understanding of the concept with regard to people with ID.

Nonetheless, it can be concluded from study findings that a social conceptualisation of frailty is highly relevant to understanding the complex and changing needs of this increasingly aging population. Coinciding with greater longevity are efforts to support people with ID to transition from institutional care settings to community-based homes and provide greater opportunities for community participation and social engagement. Subsequently, people with ID are developing more varied relationships within their families and in the wider community while also becoming increasingly reliant upon informal support to live independently.

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Appendices

Appendix 1: SANRA Guidelines

Scale for the Assessment of Narrative Review Articles – SANRA

Please rate the quality of the narrative review article in question, using categories 0–2 on the following scale. For each aspect of quality, please choose the option which best fits your evaluation, using categories 0 and 2 freely to imply general low and high quality. These are not intended to imply the worst or best imaginable quality.

1) Justification of the article's importance for the readership

- The importance is not justified. _____ 0
- The importance is alluded to, but not explicitly justified. _____ 1
- The importance is explicitly justified. _____ 2

2) Statement of concrete aims or formulation of questions

- No aims or questions are formulated. _____ 0
- Aims are formulated generally but not concretely or in terms of clear questions. _____ 1
- One or more concrete aims or questions are formulated. _____ 2

3) Description of the literature search

- The search strategy is not presented. _____ 0
- The literature search is described briefly. _____ 1
- The literature search is described in detail, including search terms and inclusion criteria. _____ 2

4) Referencing

- Key statements are not supported by references. _____ 0
- The referencing of key statements is inconsistent. _____ 1
- Key statements are supported by references. _____ 2

5) Scientific reasoning

(e.g., incorporation of appropriate evidence, such as RCTs in clinical medicine)

- The article's point is not based on appropriate arguments. _____ 0
- Appropriate evidence is introduced selectively. _____ 1
- Appropriate evidence is generally present. _____ 2

6) Appropriate presentation of data

(e.g., absolute vs relative risk; effect sizes without confidence intervals)

- Data are presented inadequately. _____ 0
- Data are often not presented in the most appropriate way. _____ 1
- Relevant outcome data are generally presented appropriately. _____ 2

Sumscore

SANRA – explanations and instructions

This scale is intended to help editors assess the quality of a narrative review article based on formal criteria accessible to the reader. It cannot cover other elements of editorial decision making such as degree of originality, topicality, conflicts of interest or the plausibility, correctness or completeness of the content itself. SANRA is an instrument for editors, authors, and reviewers evaluating individual manuscripts. It may also help editors to document average manuscript quality within their journal and researchers to document the manuscript quality, for example in peer review research. Using only three scoring options, 0, 1 and 2, SANRA is intended to provide a swift and pragmatic sum score for quality, for everyday use with real manuscripts, in a field where established quality standards have previously been lacking. It is not designed as an exact measurement of the quality of all theoretically possible manuscripts. For this reason, the extreme values (0 and 2) should be used relatively freely and not reserved only for perfect or hopeless articles.

We recommend that users test-rate a few manuscripts to familiarize themselves with the scale, before using it on the intended group of manuscripts. Ratings should assess the totality of a manuscript, including the abstract. The following comments clarify how each question is designed to be used.

Item 1 – Justification of the article's importance for the readership

Justification of importance for the readership must be seen in the context of each journal's readership.

Consider how well the manuscript outlines the clinical problem and highlights unanswered questions or evidence gaps – thoroughly (2), superficially (1), or not at all (0).

Item 2 – Statement of concrete/specific aims or formulation of questions

A good paper will propose one or more specific aims or questions which will be dealt with or topics which will be reviewed.

Please rate whether this has been done thoroughly and clearly (2), vaguely or unclearly (1), or not at all (0).

Item 3 – Description of the literature search

A convincing narrative review will be transparent about the sources of information on which the text is based. Please rate the degree to which you think this has been achieved. To achieve a rating of 2, it is not necessary to describe the literature search in as much detail as for a systematic review (searching multiple databases, including exact descriptions of search history, flowcharts, etc.), but it is necessary to specify search terms, and the types of literature included. A manuscript which only refers briefly to its literature search would score 1, while one not mentioning its methods would score 0.

Item 4 – Referencing

No manuscript references all statements. However, those that are essential for the arguments of the manuscript – “key statements” – should be backed by references in all or almost all cases. Exceptions could reasonably be made for rating purposes where a key statement has uncontroversial face-validity, such as “Diabetes is among the commonest causes of chronic morbidity worldwide.” Please rate the completeness of referencing: for most or all relevant key statements (2), inconsistently (1), sporadically (0).

Item 5 – Scientific reasoning

The item describes the quality of the scientific point made. A convincing narrative review presents evidence for key arguments. It should mention study design (randomized controlled trial, qualitative study, etc), and where available, levels of evidence. Please rate whether you feel this has been done thoroughly (2), superficially (1), or hardly at all (0). Unlike item 6, which is concerned with the selection and presentation of concrete outcome data, this item relates to the use of evidence and of types of evidence in the manuscript's arguments.

Item 6 – Appropriate presentation of data:

This item describes the correct presentation of data central to the article's argument. Which data are considered relevant varies from field to field. In some areas relevant data would be absolute rather than relative risks or clinical versus surrogate or intermediate end-points. These outcomes must be presented correctly. For example, it is appropriate that effect sizes are accompanied by confidence intervals. Please rate how far the paper achieves this – thoroughly (2), partially (1), or hardly at all (0). Unlike item 5, which relates to the use of evidence and of types of evidence in the manuscript's arguments, this item is concerned with the selection and presentation of concrete outcome data.

Appendix 2: Search Terms

Medline
<p>MH “Intellectual Disability” OR MH “Developmental Disabilities”) OR TI “Intellectual Disability” OR TI “Disabilities, Intellectual” OR TI “Intellectual Disabilities” OR TI “Mental Retardation” OR TI “Retardation, Mental” OR TI “Disability, Intellectual” OR TI “Mental Retardation, Psychosocial” OR TI “Mental Retardations, Psychosocial” OR TI “Psychosocial Mental Retardation” OR TI “Psychosocial Mental Retardations” OR TI “Retardation, Psychosocial Mental” OR TI “Retardations, Psychosocial Mental” OR TI “Deficiency, Mental” OR TI “Deficiencies, Mental” OR TI “Mental Deficiencies” OR TI “Mental Deficiency” OR TI “Idiocy” OR TI “Cognitive Impairment” OR TI “Cognitive Impairments” OR TI “Learning Disability” OR TI “Learning Disabilities” OR TI “Mentally Handicapped” OR TI “Mental Handicap” OR TI “Intellectual Development Disorder” OR TI “Intellectual Development Disorders” OR TI “Intellectual impairment” OR TI “Intellectual Impairments” OR AB “Intellectual Disability” OR AB “Disabilities, Intellectual” OR AB “Intellectual Disabilities” OR AB “Mental Retardation” OR AB “Retardation, Mental” OR AB “Disability, Intellectual” OR AB “Mental Retardation, Psychosocial” OR AB “Mental Retardations, Psychosocial” OR AB “Psychosocial Mental Retardation” OR AB “Psychosocial Mental Retardations” OR AB “Retardation, Psychosocial Mental” OR AB “Retardations, Psychosocial Mental” OR AB “Deficiency, Mental” OR AB “Deficiencies, Mental” OR AB “Mental Deficiencies” OR AB “Mental Deficiency” OR AB “Idiocy” OR AB “Cognitive Impairment” OR AB “Cognitive Impairments” OR AB “Learning Disability” OR AB “Learning Disabilities” OR AB “Mentally Handicapped” OR AB “Mental Handicap” OR AB “Intellectual Development Disorder” OR AB “Intellectual Development Disorders” OR AB “Intellectual impairment” OR AB “Intellectual Impairments” AND MH “Frailty” OR TI “Frailty” OR TI “Frail” OR TI “Frailness” OR TI “Frail Elderly” OR TI “Frailty Syndrome” OR AB “Frailty” OR AB “Frail” OR AB “Frailness” OR AB “Frail Elderly” OR AB “Frailty Syndrome”</p>

Appendix 3: CASP Cohort Study Checklist

Question	Martin <i>et al.</i> (2017)	Ouellette-Kuntz <i>et al.</i> (2018)
Are the results of the study valid?	1	1
Was the cohort recruited in an acceptable way?	1	1
Was the exposure accurately measured to minimise bias?	1	1
Was the outcome accurately measured to minimise bias?	1	1
Have the authors identified all important confounding factors?	1	1
Have they taken account of the confounding factors in design/ analysis?	1	1
Was the follow up of subjects complete enough?	3	3
Was the follow up of subjects long enough?	3	3
What were the results?
How precise are the results
Do you believe the results?	1	1
Will the results help locally?	3	3
Do the results fit with other available evidence?	1	3
[are there] implications of this study for practice?	1	1

Appendix 4: Data Summary Table

Studies	Study type	Location	Sample size	Sample description	Frailty measure	Statistical analysis	Characteristics reported	Main findings
Martin et al. (2018)	Retrospective cohort	Canada	2,893	Community based home care users aged 18-99 years	FI	Relative Risk (RR), p-Value, CI (95%),	Age Gender Presence of Down Syndrome Group home living Functional ability Cognitive ability Baseline frailty status	Worsening frailty or death associated with increasing age, presence of Down syndrome, group home living, pre-frail and frail baseline frailty status and functional impairment.
Ouellette-Kuntz et al. (2018)	Prospective cohort	Canada	5,074	Community based home care users aged 18-99 years	FI	Incidence Rate (IR), Incident Rate Ratio (IRR), p-Value, CI (95%)	Age Gender Presence of Down Syndrome Group home living Functional ability Cognitive ability Nursing services/therapies use	Accelerated rate of deficit accumulation associated with increasing age, presence of Down syndrome and group home living. Influence of nursing services/therapies use varied depending on frailty status.

Appendix 5: Health Fair Invitation Cover Letter



Intellectual Disability Supplement to TILDA
School of Nursing and Midwifery, Trinity College
Dublin



DEAR PARTICIPANT



Thank you for taking part in the Health Fair of Wave 4 of the IDS-TILDA study.



This will help us learn more about people's health as they grow older.

The Health Fair has more assessments this time.

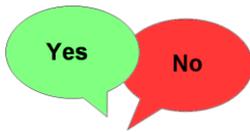
Please read the information booklet enclosed to find out about the assessments



The full health assessment will take about 90 minutes

The day before your appointment, write down everything you eat and drink. Don't forget to bring it with you.





On the day of your appointment we will ask your permission to do the assessments. It is your choice



IMPORTANT

Please do not wear tights as the bone strength assessment will be on your bare foot.



If you have any questions please call me on xxxxxxxx or 01 8963187.

Yours sincerely,

Brendan

Brendan Dee

Intellectual Disability Supplement to TILDA

Appendix 6: Health Fair Accessible Information Booklet



The Intellectual Disability Supplement to
The Irish Longitudinal Study on Ageing
(IDS-TILDA)

IDS-TILDA Wave 4 Happy Healthy Ageing Health Fair

Accessible Information Booklet



Trinity College Dublin
Coláiste na Tríonóide, Baile Átha Cliath
The University of Dublin



IDS-TILDA

Working to Make Ireland the Best Place
to Grow Old

Introduction



The Intellectual Disability Supplement to
The Irish Longitudinal Study on Ageing
(IDS-TILDA)

IDS-TILDA is a study about people who are growing older with an intellectual disability in Ireland.



We need you to help us learn more about people's health and how it can change as they grow older.

**Happy Healthy
Ageing
Health Fair**

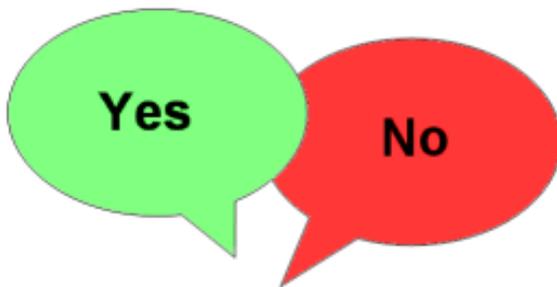
We are going to hold a health fair as part of this study. At the Health Fair we will do some health assessments.



This will help us to learn about people's health as they grow older.



We will first ask for your permission to do the health assessment.



You do not have to take part if you do not want to. It is your choice.

General Information



We will help you feel as comfortable as possible.



If there are any of the assessments you cannot do that is OK, you can do as many as you are able to.



We will arrange the appointment to suit you. Sometimes we will hold the health fair in one place and ask you to come to see us.

The Health Assessments



Blood Pressure

We will take your blood pressure.



Grip Strength

We will measure your grip. This helps us find out how strong you are.



Height

We will find out how tall you are. If you cannot stand up, we will measure your arm instead.



Weight

We will find out your weight.



Hip and Waist Size

We will measure your waist and hips.



Calf Size

We will measure your calf.



Foot Health

We will ask you some questions about your feet.



Bone Strength

We will measure how strong your bones are.



Nutrition

We will ask you questions about what you eat and drink.



Mouth Health

We will count your teeth and check your mouth.



We will prick your little finger to take small drops of blood.



Blood Drop



Sit to chair stand

We will see how steady you are on your feet. We will ask you to stand up and sit down 5 times. We will see how long it takes you to do this.



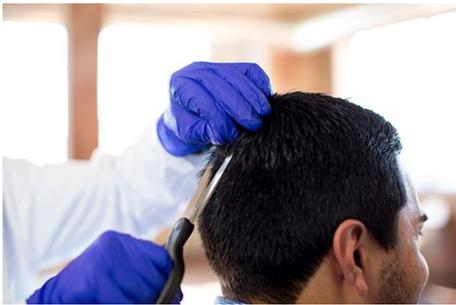
Brain health

We will ask you to do some activities that will tell us about how you remember things and how you work things out.



Activity Levels

We will ask you if you would like to do the physical activity assessment. For this, you would have to wear a small monitor on your leg for a week.



Hair Sample

We will ask you for a small piece of your hair. This will tell us about some of the hormones in your body.



Remember you do not have to do any of the assessments if you are not able to or you do not want to, it is your choice.



If you have any questions you can contact:
Brendan

Mobile Number: _____

Appendix 7: Health Fair Appointment Card



Health Fair Appointment IDS-TILDA Wave 4



Picture of
researcher

Name _____ will visit you:

Date: _____

Time: _____

Venue: _____



The Health Fair will take around 90 minutes to complete

If you want more information, please call Brendan Dee on:
Mobile number: _____

Appendix 8: Modified Frailty Phenotype

(adapted from O'Connell et al. (2020))

Characteristic	IDS-TILDA variable
Weight loss (unintentional)	Within the last year, have you lost or gained ten pounds (4.5 kg) or more in weight when you weren't trying to? No = 0; Yes = 1
Weakness	Please indicate the level of difficulty, if any, you have with lifting or carrying weights over 10 pounds/ 5 kilos, like a heavy bag of groceries: No difficulty/some difficulty = 0 A lot of difficulty/cannot do at all = 1 Grip strength: lowest 20% (by gender, body mass index): <u>Cut-off for men:</u> Overweight/Obese ≤ 16.6 kg Normal ≤ 12.8 kg Underweight ≤ 1 kg <u>Cut-off for women:</u> Overweight/Obese ≤ 12 kg Normal ≤ 10 kg Underweight Cut-off ≤ 2 kg
Poor endurance/ exhaustion	How much of the time during the past 4 weeks did you feel full of pep? No = 1; Yes = 0 How much of the time during the past 4 weeks did you have a lot of energy? No = 1; Yes = 0 How much of the time during the past 4 weeks did you feel worn out? No = 0; Yes = 1 How much of the time during the past 4 weeks did you feel tired? No = 0; Yes = 1

Slowness

Please indicate the level of difficulty, if any, you have with climbing one flight of stairs without resting.

No difficulty = 0

Some difficulty/a lot of difficulty/cannot do at all = 1

Timed Up and Go: slowest 20% (by gender, height):

<u>Men</u>	<u>Cut-off</u>
Height \leq 173 cm	\geq 21.56 seconds
Height $>$ 173 cm	\geq 16.94 seconds

<u>Women</u>	<u>Cut-off</u>
Height \leq 159 cm	\geq 22.75 seconds
Height $>$ 159 cm	\geq 20.43 seconds

Low activity

International Physical Activity Questionnaire (IPAQ)

Presence of Frailty:

- Frail: \geq 3 criteria present
 - Prefrail: 1 or 2 criteria present
 - Robust: 0 criteria present
-