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PREVALENCE OF FRAILTY SYNDROME AND ITS PREDICTORS IN THE POPULATION OF PEOPLE WITH INTELLECTUAL DISABILITY COMPARED TO THE GENERAL POPULATION: A SYSTEMATIC REVIEW

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Abstract

The aim of the systematic review was to evaluate the prevalence of the frailty syndrome (FS) and to identify its predictors in people with intellectual (ID) and developmental (DD) disabilities. It was assumed that in people with ID in adulthood period and early elderly period (20-60 years old) the ageing process (lower mobility in joints, frequent falls, the incidence of the coexist diseases and disability) occurs earlier and more frequent than in general population. The methodology of this systematic review was planned according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. A search of electronic databases (PubMed, EBSCO, MEDLINE) was conducted to identify all studies on the incidence of the FS in people with intellectual disabilities from 2010 to 2022 based on the physical and functional status. In contrary to the general population, the assessment of the prevalence of FS among people with ID and DD was performed mostly with multivariant model. Age, gender, coexist diseases, mobility impairment in everyday life activities and Down syndrome were identified as the most frequent predictors of FS among the studied populations. In conclusion, the prevalence of FS among people with ID was diverse (9-27%) and included the following variables: coexisting diseases, coupled disability, intelligence quotient, everyday life activities, dwelling place. Moreover FS was found to occur significantly earlier (from 10 to 25 years) than in general population. Future studies should include the assessment of FS based on both phenotype and multivariant models.

Keywords

Aging; elderly; older adults; frailty.

1 INTRODUCTION

The number of older adults is growing worldwide. The increase in the size of this social group leads to a higher prevalence of geriatric conditions in the population. One of the syndromes characteristic of older adulthood is frailty syndrome (FS) (Mendiratta, Latif, 2021). The word frailty comes from the French word *frêle*, which means: fragile, weak, or delicate (Diaz et al., 2015). The term FS is usually considered a syndrome of weakness, frailty, or depletion of reserves. It is " *a multidimensional syndrome of loss of homeostatic reserve (energy, physical, and mental abilities) that promotes the accumulation of deficits, increasing the patient's vulnerability and risk of adverse medical consequences* " (Clegg et al., 2013; Rajabali, Rolfson, Bagshaw, 2016). It is often associated with age. However, the functional status of a person depends primarily on the physiological resources of the body.

FS is a multidimensional geriatric syndrome associated with many adverse consequences including falls (impaired mobility and self-control), the need for care and support for activities of daily living, and higher mortality. The condition also places a significant burden on the health care system (Clegg et al., 2013). Etiological factors of FS include social (poverty, loneliness, low education level) and psychological (depression) determinants, nutrition (malnutrition), polypragmasia, other diseases and their complications (cancer,

endocrine disorders, dementia), and low physical activity (Di Ciaula, Portincasa, 2020; Mendiratta, Latif, 2021). FS is a condition that can be stopped and reversed (Morley et al., 2013).

FS is not an inevitable part of the aging process and does not apply only to older adults, i.e. those over 60 years of age. It also affects younger patients with chronic diseases and cognitive dysfunctions (Bagshaw et al., 2014; Wleklik et al. 2020). A study by Goldfarb, Sheppard, Afilalo (2015) indicated that one in ten older adults develops FS symptoms. In Europe, an estimated 17% of older adults suffer from FS, while its prevalence increases with age, especially among women (Oresanya, Lyons, Finlayson, 2014).

The available scientific literature points to various methods of assessing FS, which focus on two frailty models: one-dimensional (phenotypic) and multidimensional (Pilotto et al., 2020). The phenotypic model identifies frailty based on the presence of at least three of five symptoms: *(1) unintentional weight loss, (2) exhaustion, (3) low level of physical activity, (4) slow walking speed, and (5) muscle weakness* (Fried et al., 2001). The multidimensional model assesses frailty based on functional, sensory, and clinical deficits (Pilotto et al., 2020; Rockwood, Mitnitski, 2007). Both cross-sectional observations and longitudinal studies have been used to search for the gold standard for estimating FS regardless of the assessment model. According to Ding, Kuha, Murphy (2017), the longitudinal study design is effective for identifying factors that predict the occurrence of FS. Among other things, the following factors to be predictive of FS include: (*1) chronic disease, (2) elevated blood pressure, (3) elevated inflammatory markers, (4) abnormal blood parameters identifying risk for cardiometabolic diseases, (5) unfavorable changes in body physique and composition characteristics (anthropometric measurements), (6) physical activity below levels recommended for the patient's age, (7) cognitive and/or depressive disorders, and (8) poor social support and polypragmasia* (Ding et al., 2017; Veronese et al., 2017; Wleklik et al., 2020).

In this context, attention should be paid to people with disabilities, especially intellectual disabilities (ID). Like that of the general population, their life expectancy is extending, and their health problems associated with aging are comparable. It has also been shown that people with ID over the age of 50 had similar symptoms to those of frailty syndrome in older adults over the age of 75 in the general population (Schoufour et al., 2013). Furthermore, studies involving people with ID have indicated that FS symptoms tend to occur earlier and are more severe than in people in the general population, which is associated with earlier mortality (Evenhuis et al., 2012; Schoufour, Echteld, Evenhuis, 2015b).

At the same time, scientific studies conducted on both younger and older populations of adults with ID have demonstrated that these groups are characterized by reduced daily physical activity and reduced body mobility (Celebańska, Gawlik 2013; Chow, Choi, Huang, 2018; Gawlik, Zwierzchowska, Celebańska, 2018; Gawlik et al., 2016; Hsieh et al., 2017). This group also showed a higher prevalence of abnormal blood parameters and body composition components, which are factors for identifying the risk of cardiometabolic diseases (Gawlik et al.,2018; Zwierzchowska et al., 2021). Undoubtedly, this is a reason for the use of prevention in the form of increasing and long-term use of pharmaceuticals, which have therapeutic and supportive effects but long-term selective use can lead to damage in other organs and body systems and also the acceleration of involutional changes (Brehmer, Weber, 2010; Schoufour et al., 2015b).

Previous one-dimensional (phenotypic) and multivariate studies conducted on individuals with ID confirm that Down syndrome, comorbid dementia trait syndrome, profound ID (IQ ≤35), age, and motor disabilities are significantly associated with FS (Evenhuis et al., 2012; Evenhuis, Schoufour, Echteld, 2013; Schoufour et al., 2013).

However, identifying FS in people with ID is much more difficult since intellectual disability has a constitutional origin, which consequently always leads to morphofunctional disorders of varying severity with coexisting physical pathologies and lifestyles. At the same time, studying populations with ID using the longitudinal design is not only hampered by

the duration of the research process but also has limitations that stem from ID itself. The internal compensatory mechanisms that take place are individually differentiated due to the morphofunctional characteristics and pathologies of the patients, which significantly limits the identification of symptoms and estimation of the FS scale.

The past decade has seen a significant increase in the interest of researchers in identifying factors for FS in people with ID. Although many hypotheses have been proposed to date, the problem still has not been fully clarified. Given these scientific reports, there is a need for deeper analyses explaining FS and its predictors in the group of people with intellectual disabilities, which will allow the implementation of preventive measures against the occurrence of FS in this group.

2 THE AIM OF STUDY

The aim of this study was to assess the prevalence and identify predictors of FS in a group of people with intellectual and/or developmental disabilities based on a systematic review. It was assumed that in the group of people with ID in adulthood and early older adulthood (20-60 years), the aging processes, manifested by reduced mobility, more frequent falls, comorbidities, and disabilities are more likely to occur at a younger age than in the general adult population.

3 METHODS

Study design

The methodology of this systematic review was planned according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (equator-network.org).

Inclusion and exclusion criteria

In this systematic review, inclusion criteria were (a) cross-sectional study, (b) people with intellectual and/or developmental disabilities <=18 years old, (c) males and/or females, (d) cross-sectional study and/or cohort study, and/or longitudinal study. The exclusion criteria were as follows: (a) no data on the prevalence of FS, (b) no data on the method used for identification of FS, (c) poor methodological design, and (d) full-text not in English.

Literature search

A search in electronic databases (PubMed, EBSCO, MEDLINE) was conducted by three authors (DC, EG, BR) to identify all studies on the prevalence of frailty syndrome in people with intellectual disabilities from 2010 to 2022. The following methods were used: (a) data mining, and (b) data discovery and classification. As a prerequisite, all studies were performed on populations of people with disabilities including both adults and adolescents. Search terms were combined by Boolean logic (AND/OR) in PubMed, EBSCO and MEDLINE databases.

The search was undertaken using two keyword combinations in English with the assumed hierarchy of their importance: ‘frailty syndrome’, and ‘intellectual disability’. Furthermore, three authors (DC, EG, BR) with expertise in people with intellectual disabilities and frailty syndrome reviewed the reference lists of the included studies and screened Google Scholar to find additional studies. The corresponding authors of the selected publications were also contacted directly if the crucial data were not available in the original articles.

Methodological Quality of Included Studies (Risk of bias)

The Joanna Briggs Institute (JBI) Critical Appraisal Checklist for the analytical cross-sectional study was used to assess **t**he methodological quality of the included studies (Ma et al., 2020). The JBI is known as the newest and the most preferred tool for assessing the methodological quality (risk of bias) of analytical cross-sectional studies [24]. The checklist consists of 8 questions (see Table 1). Each study was read and scored ‘Yes’, ‘No’, ‘Unsure’, or ‘Not applicable’. If the criterion was fulfilled, a ‘Yes’ was assigned to the article, which simultaneously received a score of one, whereas if the criterion was not

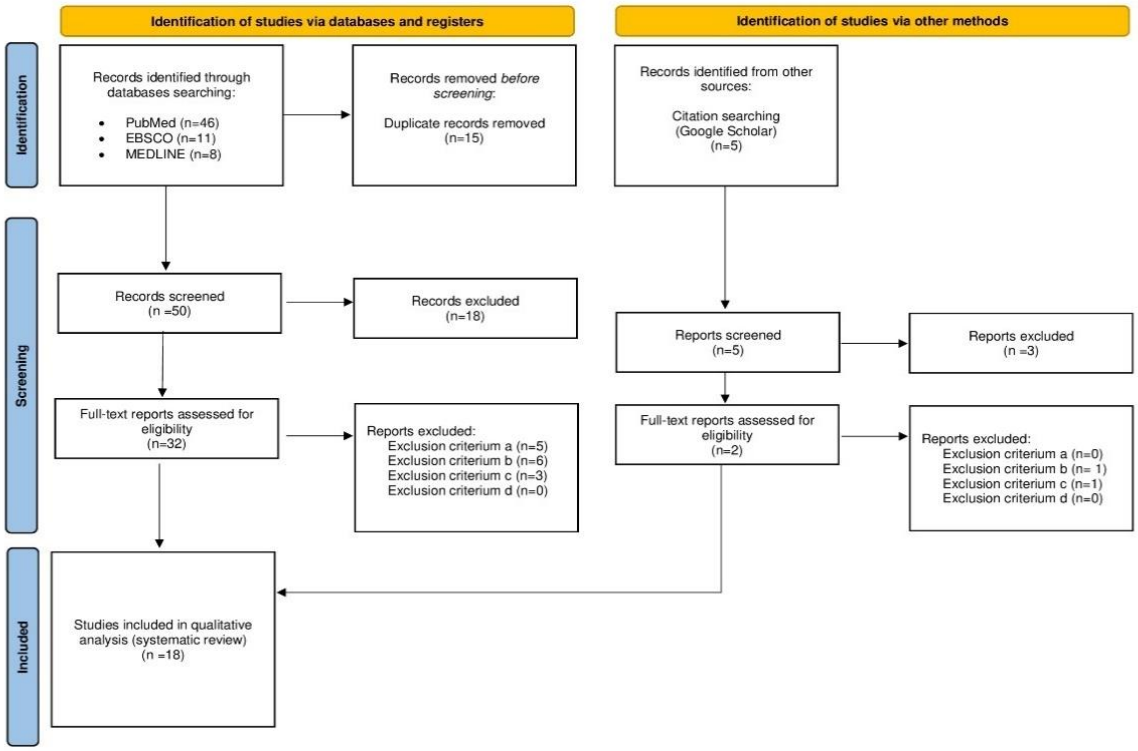
fulfilled, a 'No', 'Unclear', or 'Not applicable' was assigned to the article, and the article received a zero score. Each study was read and ranked by three independent investigators (DC, EG, BR). Furthermore, an independent co-author (AZ) was designated to resolve all discrepancies that could occur among investigators during the assessment. The sum of the awarded points (out of a possible 8 points) indicated the methodological quality (risk of bias), with the higher values representing better quality in the included publications.

4 RESULTS

Study selection and characteristics

The flow of the systematic review is presented in Figure 1.

Figure 1. PRISMA flow diagram detailing the study inclusion process (equator-network.org).



Thirty-two full-text articles were assessed to determine eligibility, while eighteen studies met the inclusion criteria and were subjected to detailed analysis and assessment of their methodological quality (see Table 1).

Over three-fourths of the reports that were assessed for their methodological quality were considered to have 8/8 points of eligibility to be included in the systematic review. Two publications (Brehmer et al., 2010, Brehmer-Rinderer et al., 2013) were considered to have 7/8 points of eligibility. The initial agreement of the three independent investigators (DC, EG, BR) was 90%. All discrepancies among the investigators were resolved by the expert evaluation by an independent co-author (AZ).

Eighteen full-text articles were finally included in the systematic review (see Tab. 2-3).

Table 1 The assessment of the methodological quality of the included studies (risk of bias) using the JBI method for analytical cross-sectional study and cohort study.

No.	Author	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Sum
1.	Brehmer, Weber (2010)	Y	Y	Y	U	Y	Y	Y	Y	7/8
2.	Evenhuis et al. (2012)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
3.	Brehmer-Rinderer et al. (2013)	Y	Y	Y	U	Y	Y	Y	Y	7/8
4.	Schoufour et al. (2013)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
5.	Evenhuis (2014)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
6.	Schoufour et al. (2014)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
7.	Schoufour et al. (2015a)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
8.	Schoufour et al. (2015b)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
9.	Schoufour et al. (2015c)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
10.	McKenzie, Ouellette-Kuntz, Martin (2015)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
11.	Schoufour et al. (2016)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
12.	Schoufour, Echteld, Evenhuis (2017)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
13.	Martin, McKenzie, Ouellette-Kuntz (2018)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
14.	Ouellette-Kuntz, Martin, McKenzie (2018)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
15.	Lee, Ouellette-Kuntz, Martin (2019)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
16.	O'Connell et al. (2020)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
17.	Schoufour et al. (2022)	Y	Y	Y	Y	Y	Y	Y	Y	8/8
18.	Lin, Tseng (2022)	Y	Y	Y	Y	Y	Y	Y	Y	8/8

Q1- Were the criteria for inclusion in the sample clearly defined?; Q2- Were the study subjects and the setting described in detail?;Q3- Was the exposure measured in a valid and reliable way?; Q4- Were objective, standard criteria used for measurement of the condition?;Q5- Were confounding factors identified?;Q6- Were strategies to deal with confounding factors stated?;Q7- Were the outcomes measured in a valid and reliable way?;Q8- Was appropriate statistical analysis used?; Y-yes; N-No; U-unsure; NA-not applicable

Table 2 The summary of the studies from 2010 to 2022 evaluating the prevalence of FS among people with intellectual disabilities

Author	**Study group**	**The prevalence of FS (one-dimensional model)**	**FS characteristics (multi-dimensional model)**
Brehmer, Weber (2010)	nP=190/ age;18-76/ ID; IQ (from 69 to <25)	**Not applicable**	FS = 27% (>50 years old) FS = 9% (two criteria) PFO = 12%
Evenhuis et al. (2012)	nP= 848/ age; >50/ ID; IQ (from 69 to <25)	FS= 13% (50-64 years old) FS =18% (>65 years old) FS =21% (>70 years old) PFO = 60%	**Not applicable**
Brehmer-Rinderer et al. (2013)	nP = 147/ age; 20-72/ ID; IQ (from 69 to <25)	**Not applicable**	FS= 17.7% PFO= 17.7%
Schoufour et al. (2013)	nP= 1050/ age; >50/ ID; IQ (from 69 to <25)	**Not applicable**	FI: 0.27±0.13
Evenhuis (2014)	nP = 1050/ age;50-94/ ID; IQ (from 69 to <25)	**Not applicable**	**Not applicable**
Schoufour et al. (2014)	nP=703/ age; 50>80/ ID; IQ (from 69 to <25)	**Not applicable**	FI=0.27 FI= 0.26 (3 years later) FI=0.32 (CG)
Schoufour et al. (2015a)	nP=632/ age;50>80 ID; IQ (from 69 to <25)	**Not applicable**	FI=0.27±0.13

Schoufour et al. (2015b)	nP=982 age;50>90/ ID; IQ (from 69 to <25)	**Not applicable**	FI=0.27±0.13
Schoufour et al. (2015c)	nP=982/ age; 50-93/ ID; IQ (from 69 to <25)	**Not applicable**	FI= ≥ 0.30 FS=38.5% PFO= 28.4%
McKenzie et al. (2015)	nP=7863 (participants with developmental disabilities)/ age; 18-99	**Not applicable**	FI =0.22±0,13 FS= 26.9% PFO = 21.3%
Schoufour et al. (2016)	nP=757/ age; >50/ ID; IQ (from 70 to <25)	**Not applicable**	FI=0.28±0.12
Schoufour et al. (2017)	nP=818/ age; >50/ ID; IQ (from 70 to <25)	FS= 13.3% PFO=59.7%	FI=(0.22±0.13) FS=25.2% PFO=38.8%
Martin et al. (2018)	nP=2893/(participants with developmental disabilities)/ age;18-99/ ID; IQ (from 70 to <25)	**Not applicable**	FS= 16.8% PFO =16.2%
Ouellette-Kuntz et al. (2018)	nP=5074 /(participants with developmental disabilities)/ age; 18-99	**Not applicable**	FI= 0.17±0.12
Lee et al. (2019)	nP=170/(participants with developmental disabilities)/ age; 19.8-86.4	**Not applicable**	FI=0-0,58 (The scores were derived only from these 13 items questionnaire)
O'Connell et al. (2020)	nP=570/ age; 44-60+/ ID; IQ (from 70 to <25	FS=18.1% PFO=64.0%	**Not applicable**

Schoufour et al. (2022)	nP=982/ age; >50/ ID; IQ (from 69 to <25)	**Not applicable**	FS= 29.4% PFO=41.7%
Lin, Tseng (2022)	nP=85/ age; 40+ ID; IQ (from 69 to <25)	**Not applicable**	FS=23.5% FS=20.0% (9 months later) PFO=68.3% PFO=70.6% (9 months later)

nP – number of participants; FS – frailty syndrome; PFO – possible frailty onset, prefrail; FI - frailty Index CG – control group

Table 3 The summary of the studies from 2010 to 2022 evaluating the predictors of FS among people with intellectual disabilities based on the physical and functional status

Author	**Research model & tool**		**Identified FS predictors**	**FS: the main findings**	
	Phenotype model	**Multivariant model**		**Phenotype model**	**Multivariant model**
Brehmer, Weber (2010)	**Not applicable**	Vienna Frailty Questionnaire for Persons with ID	↑pharmaceuticals intake, memory difficulties, nervous and/or anxiety behavior and/or anxiety, fear of failing ↓ general health status, muscular strength, joint mobility ↓ cognitive abilities, vision, disturbances in social relationships	**Not applicable**	No statistical significance between genders, FS and ID.
Evenhuis et al. (2012)	Physical activity (pedometer), conditional tests (grip strength - Jamar hand grip dynamometer, comfortable walking speed, poor endurance or exhaustion - "lacks energy" on the Anxiety, Depression and Mood Scale.	Psychiatric assessment of depression and IQ	↑ age, mobility impairment, Down syndrome, dementia, ID	FS correlated with dementia and physical disability	The high prevalence of frailty and motor disabilities in the group aged 50 to 64 suggests frailty before age 50.
Brehmer-Rinderer et al. (2013)	**Not applicable**	Vienna Frailty Questionnaire for Persons with ID (VFQ-ID)	↑ mobility impairment in activities of daily living	**Not applicable**	All four frailty domains (social, physical, psychological, and cognitive) of the VFQ-ID were a reliable measure of frailty and age-related changes in persons with ID.

Schoufour et al. (2013)	**Not applicable**	FI Questionnaire	↑ age, ID level, Down syndrome, residence in nursing homes	**Not applicable**	Older adults with ID aged 50 years and over already accumulate as many deficits as older adults without ID aged 70 and above (early aging).
Evenhuis (2014)	laboratory tests, pedometer, conditional tests, dynamometer	FI Questionnaire, nutrition assessment, psychological assessment (questionnaire) psychiatric interview, observation, sleep assessment,	↑ polypragmasia, ID level, Down syndrome, ↓ mobility impairment in activities of daily living, independence	↑ depression ↑ sleep disorders ↑ cardiovascular diseases (can lead to early weakness)	The correlation between ID and FS was (r=0.94±p< 0.001)
Schoufour et al. (2014)	laboratory tests pedometer, conditional tests, observation,	FI Questionnaire, nutrition questionnaire, psychiatric questionnaire and interview, life quality questionnaire, IQ test, medical data assessment,	↑ mobility impairment in activities of daily living, ↓ joint mobility	FS was associated with physical disability and mortality.	In 84%, FI was not observed at the end of the study. FI at the beginning of the study correlated with the risk of deterioration and/or death (RR 1, 24, 95% CI 1.04-1.49).
Schoufour et al. (2015a)	**Not applicable**	FI Questionnaire	↑pharmaceuticals intake, comorbidities	**Not applicable**	Frailty is related to decreased health status. Frailty has serious consequences in older adults with ID (mortality, increased care intensity, deterioration in independence and mobility|).
Schoufour et al. (2015b)	**Not applicable**	FI Questionnaire	↑ mobility impairment in activities of daily living, pharmaceuticals intake, health care ↓ joint mobility -mortality	**Not applicable**	People with ID became weaker earlier than those in the general population.
Schoufour et al. (2015c)	**Not applicable**	FI Questionnaire	↑ age, ID level, Down syndrome, mobility impairment in activities of daily living, comorbidities	**Not applicable**	In 37.6% of participants, an improvement or deterioration of FI was observed. ID and comorbidities were the main predictors of changes of the FI.

McKenzie et al. (2015)	**Not applicable**	Resident Assessment Instrument – Home Care	↑ age, impairment, comorbidities	**Not applicable**	Premature aging has frequently been reported in adults with intellectual disabilities and Down syndrome.
Schoufour et al. (2016)	**Not applicable**	FI Questionnaire Blood samples tests	↑ age, ID level, residence in nursing homes, Alzheimer's disease, numbness, inflammation of the IL-6 and CRP, anemia, metabolic markers (glucose, cholesterol, albumin), and renal function	**Not applicable**	Frailty is associated with the current inflammation and nutritional status. Biochemical measurements can allow for the early identification of weak individuals with ID.
Schoufour et al. (2017)	physical activity (pedometer), laboratory tests (hand grip tester), conditional tests, exhaustion (the Anxiety, Depression, and Mood Scale)	FI Questionnaire, FP assessment, nutrition questionnaire, psychiatric questionnaire, health questionnaire	↑ bad health status, mostly females, loneliness, residence in nursing homes	Those who were weakened and frail were more likely to die, by 2.04 and 4.20 times, respectively.	Those who were weakened and frail were more likely to die, by 2.27 and 10.3 times, respectively.
Martin et al. (2018)	**Not applicable**	FI Questionnaire	↑ age, comorbidities, Down syndrome, mostly females, residence in nursing homes	**Not applicable**	Being pre-frail at baseline was associated with an increase in the risk of worsening or death (RR 1.24, 95% CI 1.04–1.49).
Ouellette-Kuntz et al. (2018)	**Not applicable**	Resident Assessment Instrument for Home Care	↑ age, Down syndrome, mostly females, residence in nursing homes, mobility impairment in activities of daily living, independence	**Not applicable**	Frailty should be monitored from the age of 40 years, those with Down syndrome, and those who live in group homes.
Lee et al. (2019)	**Not applicable**	The Home Care-IDD FI	↑ age, developmental disability level, residential conditions, hospitalization, comorbidities	**Not applicable**	Frailty predicts adverse outcomes and is more prevalent among adults with ID and IDD. Client charts should capture key information needed to measure frailty as knowledge of frailty status could improve care planning and facilitate personalized care.

O'Connell et al. (2020)	Modified Fried's Frailty Phenotype	Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing (IDS-TILDA);	↑ age, ID level, residence in nursing homes, polypharmacy	Alzheimer's disease or dementia was found to be statistically significantly associated with frailty status	We identified an association between excessive polypharmacy and frailty status.
Schoufour et al. (2022)	**Not applicable**	ID-FI Short Form FI-Questionnaire	**Not applicable**	**Not applicable**	A good agreement between the full and short forms in dividing the participants into the frailty categories.
Lin, Tseng 2022	**Not applicable**	FI variables, Barthel rate, number of falls and hospitalizations, types of comorbidities	↑ ID level, comorbidities, mobility impairment in activities of daily living	**Not applicable**	Pre-frail condition is more commonly detected than frail condition in pre-maturely aging adults with ID. Adults with pre-frail or frail conditions possibly experience change over a short-term period and are associated with comorbidities and disabilities

FS – frailty syndrome; FI - Frailty Index; IDD - intellectual disabilities and Down syndrome; ↑ - increase; ↓ - decrease

5 DISCUSSIONS

Frailty syndrome has been an object of interest to researchers for many years, especially in terms of finding ways to identify this condition and estimating the strength and direction of change (Clegg et al., 2013; Di Ciaula, Portincasa, 2020; Mendiratta, Latif, 2021; Morley et al., 2013). Recent cohort, prospective, and longitudinal studies of the non-disabled population in middle and late adulthood have indicated several important predictors that identify frailty syndrome (Fried et al., 2001; Searle et al., 2008). Our study confirmed these predictors, especially with regard to the age of the subjects, comorbidities, and/or physical disabilities (Evenhuis et al., 2012; McKenzie et al., 2015; O'Connell et al., 2020; Schoufour et al., 2013; Schoufour et al., 2015a; Schoufour et al., 2015b). However, for people with ID, the predictive variable of age is indicated by most authors as a factor that manifests itself significantly earlier in explaining FS (Evenhuis et al., 2012; Schoufour et al., 2015a; Schoufour et al., 2015b).

In studies of the general population, according to the criteria of the Cardiovascular Health Study (CHS), FS has been diagnosed when the subject developed at least three of the following 5 criteria: weight loss, poor grip strength, low walking speed, low physical activity, poor endurance, and exhaustion. Such a one-dimensional (phenotypic) model was used in studies by Evenhuis et al. (2012), Schoufour et al. (2017), and O'Connell et al. (2020) and confirmed the effectiveness of FS identification in a group with ID. At the same time, they emphasized the importance of comorbid developmental, motor, and intellectual disabilities to the manifestation and aggravation of the syndrome's symptoms. On the other hand, when one or two of the above-mentioned criteria occurred, we observed that, as is the case in the general population, the subjects were classified as unstable individuals with possible frailty onset (PFO), which was found only in studies by Evenhuis et al. (2012), Schoufour et al. (2017); O'Connell et al. (2020). Such a one-dimensional (phenotypic) model is currently most commonly used as a reliable way to diagnose FS in the general population. However, in the available literature of the last decade, we failed to find more studies using a phenotypic model in a population with ID, which is an important limitation for inferring from our study of phenotypic predictors that determine FS in this group.

Evenhuis (2014) also pointed out the importance of the age criterion in identifying FS, which is consistent with findings published in previous studies such as those by Schoufour et al. (2014), Schoufour et al. (2015b), while several authors indicated on gender criterion (female) (Martin et al., 2018; Ouellette-Kuntz et al., 2018; Schoufour et al., 2017). This conclusion was one of the most common findings in the manuscripts we analyzed regardless of the FS identification model used (McKenzie et al., 2015; Lee et al., 2019; Martin et al., 2018; O'Connell et al., 2020; Ouellette-Kuntz et al., 2018; Schoufour et al., 2013, 2015c, 2016, 2017).

The definition of FS includes both phenotypic and functional criteria, which have been described as a multidimensional way of identifying the syndrome. Researchers who created criteria for assessing FS in the general population relied on four domains (social, cognitive, psychological, and physical), recommending that a minimum of 30 to 40 assessed criteria should be met in order for the FS assessment index to be reliable (Searle et al., 2008). In the analyzed studies, this was the way which has been most popular among researchers who used psychological and social tests and interview questionnaires, without taking into account objective laboratory tests (Brehmer, Weber, 2010; Brehmer-Rinderer et al., 2013; Lee et al., 2019; Lin, Tseng 2022; Martin et al., 2018; McKenzie et al., 2015; Ouellette-Kuntz et al., 2018; Schoufour et al., 2013, 2015a. 2015b, 2015c, 2016, 2022). On the other hand, in general population studies, a complementary way of identifying FS using one-dimensional (phenotypic) and multivariate models has been more often employed to identify FS, which provides a predictive picture of the subject (Fried et al. 2001).

Therefore, the number of criteria is an important element of evaluation, since predictive accuracy is one of two types of criterion validation, the other being validation using a gold standard (Streiner, Norman, 2003). Given that there is no gold standard for assessing FS,

predictive validation is an important method in any approach to the operationalization of frailty syndrome (Searle et al., 2008). It was predictive validation that was also our goal of the systematic review of the available literature in the field of FS problems in the population of people with ID. At the same time, the systematic review using Prisma methodology is an indirect way of verifying the prediction of the prevalence of FS in the group of people with ID rather than developing criteria for assessing FS. Studies presented here (see Tab. 2) have mostly discussed a diagnosis of deficits and daily functional status of participants with ID, which made it possible to capture the occurrence of this status in significantly different (by ID, age, comorbid disabilities, social status (place of residence - nursing homes) groups of participants. However, with the lack of longitudinal studies on the populations with ID and, at the same time, its large internal variation in cross-sectional studies, it is impossible to clearly describe the predictors of FS.

6 CONCLUSIONS

1. The frailty syndrome in the population with ID occurs significantly earlier (from 10 to 25 years), and varies (9-27%) due to variables such as comorbid disabilities, and/or comorbid diseases, IQ, lifestyle including daily physical activity, and place of residence. Moreover, the frequency of the prevalence of FI varies from 0.17-0.58.

2. Limitations of the study include a large internal variation of the groups of participants (ID, developmental disabilities, age, and different methodological approaches of the researchers) and a small number of complementary studies using two models. Therefore, future research should focus on the standardization of methodologies and implementation of both one-dimensional and multidimensional models into research to infer FS.

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AUTHOR CONTRIBUTIONS

DC was the major contributor in writing and editing the original article. DC, EG and BR screened the databases, reviewed the list of the included studies and scanned the reference lists to find additional studies, contacted directly the corresponding author, if the crucial data were not available in the original articles and read and evaluated the methodological quality of the selected studies. AZ conceptualized the purpose and hypothesis of the study, wrote the discussion section, was designated to resolve all discrepancies that could occur among investigators during the risk of bias assessment and supervised during the study. The tables were prepared by DC and ZK. ZK was responsible for formatting, writing the review and editing. All authors read and approved the final version of the manuscript.

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