# **Nutrition, nutritional state and related conditions** in older adults with intellectual disabilities






Luc Bastiaanse

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## CONTENTS

# Chapter 1

General introduction

As generally known, healthy food intake (together with physical activity) is of great importance for physical and mental health [1]. However, (healthy) food intake was never comprehensively studied on a large scale among older adults with intellectual disabilities. Therefore, this theme was investigated within the 'Healthy Ageing and Intellectual Disability' (HA-ID) study. More precisely, food intake, nutritional status and negative health outcomes due to a poor nutritional status were investigated. In this introduction we will lay out the motivation of this study and the importance of research into healthy food intake and nutritional status within older adults with intellectual disabilities.

## RESEARCH IN OLDER ADULTS WITH INTELLECTUAL DISABILITIES: 'THE HEALTHY AGEING AND INTELLECTUAL DISABILTY STUDY'

In 2008 a consortium was founded, consisting of three intellectual disabilities care provider services (Abrona, Amarant and Ipse de Bruggen) and one academic center (Intellectual Disability Medicine, Department of General Practice, Erasmus University Medical Center Rotterdam). The main goals of this consortium were to increase knowledge on healthy ageing in people with intellectual disabilities by means of scientific research, to increase the expertise and scientific attitude of staff by means of participation in research and continuous education, and innovation of care by means of implementation of research outcomes. The theme of healthy ageing was chosen because life expectancy in people with intellectual disabilities has increased in the last decades [2]. As a consequence, the number of older people with intellectual disabilities is rising [3]. However, health in this population was only studied before on a large scale by using questionnaires or data retrieved from medical records [4, 5], not by using objective tests. An advantage of objective tests over questionnaires is that questionnaires are more vulnerable to different forms of information bias, like recall bias and social desirability bias. Furthermore, in medical files only people with a known diagnosis are counted, leading to a probable underestimation.

The first step in reaching the scientific goals of the consortium has been made by starting a large-scale, cross-sectional epidemiological study, titled 'Healthy Ageing and Intellectual Disability (HA-ID)'. In this study three subthemes were investigated: (1) depression and anxiety, (2) physical activity and fitness and (3) nutritional intake and nutritional state. These subthemes were chosen because they have a strong mutual relationship and influence health and quality of life. In the HA-ID study prevalences of health problems identified within each subtheme were assessed, risk groups were identified and diagnostic tools to assess health problems were selected and evaluated.

At the start of the study, the three care organizations of the consortium provided support or care to 2322 clients aged 50 years and over, which was 10% of the total Dutch client population of the same age receiving formal ID care [6]. In total, 1050 clients participated in the HA-ID study. Measurements in the study involved physical examination, laboratory assessment, swallowing observations, fitness tests, physical activity assessment, sleep assessment by ambulatory accelerometers and depression and anxiety screening. In chapter 2 of this thesis the design, the informed consent procedure and the study population of the HA-ID study are described.

## HEALTHY FOOD AND PHYSICAL ACTIVITY

As mentioned before, healthy food and physical activity are important for physical and mental health. For both themes, global and national recommendations are available to prevent diseases. Regarding healthy food, the Health Council of the Netherlands made recommendations about healthy intake of energy, proteins, fat, carbohydrates and fibers [7, 8], and regarding physical activity, the Dutch standard for Healthy Moving states that every adult should have at least 30 minutes of moderate intensity physical activity, at least five, but preferably all days of the week [9]. When measured by pedometers, 10,000 steps per day are recommended to realize health benefits [10].

In the general population negative associations were shown between food intake and physical activity on the one hand and age on the other hand [11, 12]. With regard to food intake, in the older population in the Netherlands, people consume more unhealthy saturated fatty acids and more salt than recommended, and less wholemeal products, fruit and fish than recommended [11]. Most of the participants in the survey by Ocke et al. were relatively vital and only a small number of older adults with functional disabilities took part. In this latter group, people had a lower energy intake, consumed less protein, vegetables, alcohol, calcium and magnesium, and ran a higher risk of undernutrition.

As mentioned in the previous section, health in older people with intellectual disabilities was never studied before on a large scale by using objective tests. Therefore both food intake (by using food frequency lists) and physical activity (by doing fitness tests and measuring steps using pedometers) were investigated in the HA-ID study. Hilgenkamp et al. have shown that physical activity is low in this population [13]. Among 257 older people with intellectual disabilities, only 43 persons (16.7%) complied with the guidelines of 10,000 steps per day and 99 persons (38.5%) had a sedentary lifestyle (< 5,000 steps per day). Because in this sample persons in a wheelchair and persons with a low walking speed were excluded, these percentages were likely to be a overestimation of the actual physical activity levels in this population [13]. The results

of the assessment of food intake among people with intellectual disabilities, aged 50 years and over, are described in this thesis.

## BACKGROUND MODEL

The background model on which diagnostic measurements, applied in this theme, have been based, is shown in figure 1.

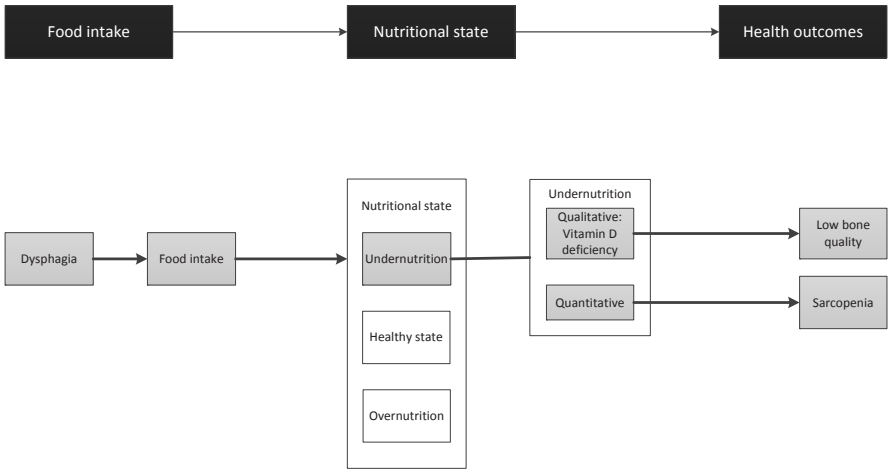


**Figure 1** *Background model of this thesis. All grey boxes are described in this thesis.*

An insufficient food intake leads to a poor nutritional status, which is defined as a deficiency (undernutrition) or excess (overnutrition) in fat, protein and other nutrients, with measurable adverse effects on body size (underweight or overweight) and body function. Both undernutrition/underweight and overnutrition/overweight may lead to negative health outcomes. Because overweight in the HA-ID population has been addressed in the thesis by Channa de Winter, the focus of this thesis will be on undernutrition.

In the next sections food intake, nutritional status, health outcomes, and the diagnostic methods used to measure these issues, are described in more detail.

## FOOD INTAKE

Analysis of food intake data includes both quantity (how much) and quality (what). If the intake of a person is insufficient for a longer time, malnutrition (either undernutrition or overnutrition) may occur. A person can be malnourished when he or she eats or drinks too little (quantity), but a person can also be malnourished when he or she takes unvaried food products (quality). In the latter case people could have a normal weight or even overweight, and nevertheless be undernourished regarding specific macro- or micronutrients. In chapter 3, the intake of food among older people with intellectual disabilities is presented. The focus of this study lied on intake of energy, proteins, fibers and fat, because of the high prevalence of protein energy malnutrition in the general ageing population, the high prevalence of constipation in people with intellectual disabilities (regarding fibers), and the key role of restricted (saturated) fat intake in preventing cardiovascular diseases [14-18]. The study aims were (1) to provide insight in the energy, protein, fat and fiber intake of older adults with intellectual disabilities, (2) to compare the actual dietary intake of older adults with intellectual disabilities to the Dutch national guidelines for healthy dietary intake, and (3) to study the associations of dietary intake of older adults with intellectual disabilities.

Food intake in the general population is on the one hand dependent on factors such as education, individual preference, budget and the supply of food products. On the other hand, disorders and/or diseases may lead to decreased or increased food intake. For example, dysphagia may lead to decreased food intake [19]. Therefore, in the HA-ID study dysphagia was investigated by (1) describing the prevalence of dysphagia within the population with intellectual disabilities aged 50 years and over, (2) determining the associations of dysphagia, and (3) estimating the awareness of dysphagia amongst professionals. The results of this study are described in chapter 4.

## NUTRITIONAL STATUS

To investigate an individual's nutritional status, nutritionists use several measures: (a) anthropometric testing (Body Mass Index [BMI], waist circumference and waist-to-hip ratio), (b) biochemical assessments, (c) food intake analysis and (d) a combination of these three measures [20]. In the HA-ID study all these measures have been used. Regarding anthropometry, the BMI of older people with intellectual disabilities was calculated by dividing their body weight (in kilograms) by squared height (in meters). The criteria of the world Health Organization were used to categorize them into underweight (BMI $<18.5$), normal weight ($18.5 \leq$ BMI $< 25$), overweight ($25 \leq$ BMI $< 30$) and obesity (BMI $\geq 30$) [21]. Out of the 893 participants, 3.0 % had

underweight, 33.1% normal weight, 38.2% had overweight and 25.6 had obesity [22]. In addition to the BMI, overweight and obesity were measured by waist circumference and the waist-to-hip ratio. Prevalences of these last mentioned measurements have been published elsewhere [22].

As mentioned before, in this thesis the focus is on underweight and undernutrition. Unfortunately, there are no hard criteria to operationalize undernutrition [23]. In clinical practice, BMI, waist circumference and food intake analysis are often used by dieticians, and in hospitals screening tools to detect undernutrition are used. The Mini Nutritional Assessment (MNA) is such a screening tool [24]. It is an internationally widely used screening method for chronic undernutrition, validated for use in healthy and frail older adults [25]. The MNA may be suitable for ageing people with intellectual disabilities as well, but it had not been evaluated for this population. Therefore, we have investigated the psychometric properties of the MNA in the HA-ID study. In chapter 5 the feasibility and the reliability of the MNA are described, and in chapter 6 we describe the results of a validation study.

As described above, malnutrition may also occur due to unvaried food intake. In such a case, shortage in macronutrients (fat, proteins, carbohydrates) and/or micronutrients (vitamins and minerals) arise. Vitamin D regulates homeostasis of serum calcium and phosphate and, in turn, the development and maintenance of bone health. A insufficient level of vitamin D increases the risk of osteoporosis and fractures. In the general population, it is known that vitamin D deficiency is highly prevalent, especially in older people [26, 27]. The prevalence of vitamin D deficiency may even be higher in older people with intellectual disabilities, because the frequently used anticonvulsant drugs can negatively influence vitamin D serum levels. Therefore we studied vitamin D deficiency in the HA-ID study. Study aims were (1) to describe the prevalence of vitamin D deficiency in older adults with intellectual disabilities, and (2) to study the associations of vitamin D deficiency. The results of this study are described in chapter 7.

## HEALTH OUTCOMES

A poor nutritional status may lead to a reduced quality of life [28] and increased morbidity and mortality. On the one hand, overnutrition and/or overweight may lead to symptoms and diseases such as diabetes mellitus, cardiovascular diseases, hypertension, metabolic syndrome, chronic lung disease, joint problems, and psychiatric disorders such as depression [29]. Prevalences of these disorders have been studied within the HA-ID study and published elsewhere [30].

In this thesis the consequences of undernutrition among older people with intellectual disabilities will be described. When people are undernourished, bone quality may deteriorate, leaving the bones fragile and vulnerable to fractures [31, 32] due to vitamin D deficiency. Therefore, in the HA-ID study we measured bone quality by using Quantitative UltraSound (QUS). We aimed to (1) determine the prevalence of low bone quality in older adults with intellectual disabilities, and (2) identify the associations of low bone quality, including intake of food (more precisely, intake of calcium) and vitamin D serum levels. The results of this study are presented in chapter 8.

Other negative health outcomes of undernutrition may be sarcopenia [33] and frailty [34]. Sarcopenia is a relatively new described syndrome in scientific literature because of its associations with loss of independence and morbidity in older people in the general population. It has been defined as a syndrome characterised by progressive and generalised loss of skeletal muscle mass and strength. It has not been studied before in older people with intellectual disabilities. Due to the high prevalence of physical inactivity [13] and a high prevalence of (lifelong) mobility impairments [35], sarcopenia may be a serious problem in this population. In chapter 9 the prevalence of sarcopenia and its associated factors (including intake of energy and proteins) are described. In addition to the prevalence and the associations of sarcopenia we also determined the overlap between sarcopenia and frailty. The prevalence of frailty within the HA-ID study is published elsewhere [36].

In the general discussion (chapter 10), we reflect on the results presented in this thesis, comment on the applicability of these results in clinical practice, and provide recommendations for future research.

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# Chapter 2

## Study Healthy Ageing and Intellectual Disabilities: recruitment and design

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## ABSTRACT

Questions encountered in epidemiologic health research in older adults with intellectual disabilities (ID) are how to recruit a large-scale sample of participants and how to measure a range of health variables in such a group. This cross-sectional study into healthy ageing started with founding a consort of three large care providers with a total client population of 2322 clients of 50 years and over, and two academic institutes. This consort made formal agreements about a research infrastructure and chose three research themes: 1. Physical activity and fitness, 2. Nutrition and nutritional state, and 3. Mood and anxiety. Subsequently, preparation was started by carefully reviewing and selecting instruments to measure a wide set of health variables to answer the research questions. Specific demands of these instruments were that they could be executed efficiently and accurately on-site in a large sample of participants and that the burden of these measurements for participants as well as their caregivers was as minimal as possible. Then, preparation was continued by designing and executing a thorough communication plan for clients, legal representatives and staff of the care providers, preceding the informed consent procedure. In this plan, which had a top-down structure, specific attention was given to personally informing and motivating of key stakeholders: the professional caregivers. This preparation led to a recruitment of 1050 participants (45.2%) and to high participation rates in key parts of the assessment. A detailed description is provided about the recruitment and organization and the selected instruments.

## INTRODUCTION

Life expectancy of adults with intellectual disabilities (ID) is lengthening towards that of adults without intellectual disabilities, but daily practice indicates that this ageing is relatively often not a healthy ageing. With a higher risk of motor impairments, sensory impairments and epilepsy since earlier in life, these people are prone to develop multiple physical and mental comorbidities at older age [1-3]. 'Frail patients' (multiple diagnoses, complex medical routines, frequent hospitalisation, functional impairment) [4], requiring individualised managed care, are expected to be highly prevalent in this population. Furthermore, functional deterioration is frequent [5], leading to diagnostic and therapeutic uncertainty, transfers from community-based to central residential settings, and high costs. With these risks in mind, three Dutch care organizations (Abrona, Huis ter Heide; Amarant,Tilburg; Ipse de Bruggen, Zwammerdam) and two academic departments (Intellectual Disability Medicine, Department of General Practice, Erasmus MC in Rotterdam; Center for Human Movement Sciences, UMCG, Groningen) intended to start a large-scale project to study health in older adults with intellectual disabilities in 2006. Inspired by questions of the care organizations themselves (formulated by client panels and staff panels), three themes were chosen: 1. Physical activity and fitness, 2. Nutrition and nutritional state, and 3. Mood and anxiety. These themes cover a substantial impact on health and quality of life and are supposed to have strong mutual relationships, but have hardly been studied in ageing people with ID. The scientific aims of this project were: a. To perform baseline assessments of prevalence rates and secondary health effects for each theme and to identify risk groups. b. To assess mutual relationships between the themes and their underlying concepts. c. To select and evaluate diagnostic tools to assess each theme.

To meet these aims, an observational cross-sectional design was chosen for this multi-centre research project. However, before such a study in this particular and complex target population could be executed, two major obstacles needed to be dealt with.

The first obstacle in the execution of such a study is caused by the specific living circumstances of older adults with ID. Many older adults with ID depend on a care system, involving family and professional caregivers. Lack of involvement, commitment and ultimately support by the care system can be an obstacle to the recruitment of a large, representative sample, as well as to participation in the assessments which would be a part of the study.

The second obstacle is how to measure a range of health characteristics in older adults with ID. In the general population, preventive health checks are used to collect data about certain health characteristics or risk factors, like the Canadian Study of Health and Aging [6], or the Cardiovascular Health Study [7]. This kind of screening is

not applicable to the population of older adults with ID because self-report questionnaires, neuropsychological tests and often physical tests may require a certain level of cognitive and physical abilities which may not be compatible with that of older adults with ID.

Because of such barriers, most published epidemiological research in adults with ID is based on existing (medical) records or registries, or observations of professional caregivers [8-13]. With this method, underrecognition of certain health problems or risk factors is to be expected [14], due to communication difficulties of the participants and lack of suitable diagnostic instruments. Another solution is to limit the number of participants [15]. With this solution, extrapolation of the results is hampered since the number of participants is often limited or narrowed by strict exclusion criteria, thus often underestimating the actual problems in this group [16].

This gives rise to the following research question: How to successfully measure health in older adults with intellectual disabilities in a large, representative sample?

## MATERIALS AND METHODS

Before starting the actual study, measures were taken to ensure optimal circumstances for executing a large-scale study. Therefore, the formation of a consort and description of the base population will be presented first. The method section then proceeds with a detailed description of the selection of instruments and organization of measurements, after which the standard informed consent procedure is described. Subsequently, extra activities undertaken to optimize recruitment will be described, such as extra activities in communication and consent procedures. Inclusion, representativeness and participation are described as main outcome measures.

### Founding a consort

Former research has shown the importance of cooperation and commitment of different management levels to provide the necessary conditions for a successful execution of a large-scale study in the field [17-19]. For this reason, three large care providers and two academic departments joined together in a consort, and preparation of a first large-scale study was started at CEO level in 2006. Formal agreements were made about financing and grant acquisition, responsibilities, communication, project management and infrastructure, involvement of clients and client representatives. Agreement was reached on the following aims of the consort: 1. to increase knowledge on healthy ageing in intellectual disability by means of scientific research, 2. to increase the scientific attitude of staff of care providers by means of participation in research and continuous education, 3. innovation of care by means of implementation

of research outcomes. In the preparatory phase and during the execution of the study, the consort discussed about policy, practical issues, results and future directions on three management levels: CEO-level, level of the boards of directors, and middle-management level, to ensure embedding of and commitment to this project.

The members of this consort cooperated in obtaining a governmental grant for this first research project (granted by the Netherlands Organisation for Health Research and Development, 2007, nr. 57000003).

## Base population

The three involved care providers in the consort mentioned above provided financial and organizational support and gave access to a large population of older adults with intellectual disabilities receiving any type of care or support from these care organizations.

The care organizations are geographically located in different regions of the Netherlands, both in urban and rural areas and all provide care to a broad spectrum of clients, varying in level of intellectual disability, mobility and living arrangements and all including different care settings: central residential settings, community-based homes, day activity centers and supported living. Together they provide care for 8550 persons with intellectual disabilities, which is approximately 10% of the total Dutch client population of specialized care providers [20]. The distribution of clients primarily receiving care (35%) and clients primarily receiving support (65%) is similar as that in the total Dutch client population with ID [20]. Furthermore, the percentage of older adults (50 years and over) in their client population (10%) is similar to that in the total Dutch population with ID [20]. We therefore consider this base population to be representative for the total Dutch client population of older adults with intellectual disabilities.

## Materials

The selection of diagnostic methods had to be performed with great care. A detailed description of the selection process of instruments within each subtheme stretches too far for this paper, but has been published elsewhere [21, 22].

In general, reliability, validity and feasibility in this specific population were important criteria in the selection of instruments.

As far as feasibility is concerned, the instruments had to be applicable in large-scale research, which means they had to be not too time-consuming and suitable for a large part of this heterogeneous population. Where possible, instruments which were also used in the general (older) population were chosen. This enables comparison between this specific population of older adults with intellectual disabilities and the general population. Furthermore, they had to be executable by a large group of professionals,

without high risks of differences between test observers. Due to the on-site nature of the assessments, instruments had to be ambulatory available, and if possible, non-invasive. The costs of the instruments were also an important factor, considering future use in clinical practice.

For the physical fitness tests and the instruments measuring anxiety and depression, a literature search and evaluation of the retrieved instruments did not result in a definite evidence-based choice for an instrument. Expert meetings were used to incorporate the clinical experience of scientific and care professionals in the final choice. In some cases English instruments had to be translated into Dutch and tested for feasibility and reliability, for example the questionnaires for anxiety and for eating disorders. A pilot study in November 2008 was used to evaluate those instruments, as well as the feasibility of the entire set of instruments.

The definite selection of instruments is presented in the Appendix, with a distinction between measurements requiring active involvement of the participant and measurements without active involvement of the participant.

## Procedure

The large-scale nature of an epidemiological study puts three specific demands on the organization of measurements. The organization needs to be efficient, the measures need to be executed accurately and the burden of these measurements for participants as well as their caregivers needs to be as minimal as possible. The burden for participants and their caregivers was considered a central factor in designing the organization of measurements. The feasibility of this organization was also tested in the pilot study and led to minor adjustments in the instruments and organization.

To complete all assessments efficiently, and to comply with one of the aims of the consort as well, the measurements needed to be executed by groups of test administrators, consisting of professionals of the involved care providers. To enhance their commitment and to optimise the organization, they were informed and consulted in an early stage of the study. Their preferences considering planning and location were followed as much as possible, and interference with existing (medical) routines was avoided as much as possible. To enhance efficiency even further, the particularly time-consuming diagnostic process of psychiatric disorders (through expert interviews) was replaced by a two-step model, with a screening for all participants by self-report or informant-report questionnaires, and only a diagnostic interview for those participants who scored above cut-off points on the questionnaires. Cognitive, social and emotional capabilities determined if a participant could be assessed by self-report questionnaires, administered by a trained test assistant in a screening interview. To ensure accurate administration of the assessments in this large group of test administrators, they were

all trained by the researchers themselves or external experts and regularly checked on correct test assessment and scoring during the entire duration of the study.

Professional caregivers of the clients were informed in an early stage of the study, even before the consent procedure had been started. After consent, involved caregivers were consulted about their preferences and suggestions for the organization of the measurement, to increase their collaboration during the assessments. These preferences were used as input for the final schedule of measurements for individual participants. Involvement and cooperation was thus managed by careful communication and organization.

In order to enhance participation during the assessments, we needed to keep the impact for participants and caregivers as low as possible. All diagnostic assessments needed to be organised at settings nearby participants, preferably locations they were familiar with. Furthermore, all assessments needed to be carried out by trained professionals of the health care organizations themselves, who were familiar to most of the participants. We decided that to decrease the burden of participation even further, all assessments needed to be concentrated in a period of two weeks for a participant, and all participants of the same living facility needed to be clustered together in the same two weeks, to decrease the impact for the involved professional caregivers too. The assessment consisted of parts where active involvement of the participant was necessary (i.e. physical examination) and of parts with no need of active involvement of the participant (i.e. questionnaires for professional caregivers), and the advice of the professional caregiver was followed concerning what parts were too stressful for a specific client.

In these two weeks, the emphasis of the assessment was on the first day, with a physical examination and a physical fitness test for the participants, and questionnaires to be completed by the professional caregivers. In the following two weeks the participants carried a pedometer and an accelerometer, and had appointments for a mealtime observation of swallowing and a short interview structured by self-report questionnaires about anxiety and mood and, if consented to, a venipuncture. Only when a participant scored above cut-off points in this screening for anxiety and/or depression, an in-depth diagnostic interview by trained behavioral therapists with client and/or a professional caregiver took place (all assessments described in more detail in the Appendix).

After the assessment on the first day, the participant received a medal, and after the whole two weeks, each participant received a certificate of participation. The professional caregiver received a report with a summary of the results of the assessment, with advice whether to consult a physician or behavioral therapist or not.

## Standard informed consent procedure

We aimed to include all clients aged 50 years or older receiving care or support by one of the three health care organizations (at the 1st of September 2008). No other exclusion criteria were applied. This selection method is likely to result in a very heterogeneous cohort with regard to etiology and disabilities, reflecting the heterogeneity in the actual population of older adults with intellectual disabilities. All eligible clients were invited to participate from November 2008 to July 2010.

Separate consent procedures were followed for clients who were capable of understanding the available information and deciding themselves to participate or not in this research project, and clients who were not capable of doing so. In some health care organizations this distinction was already available from their databases, in others we sought advice from the involved behavioral therapists in this matter, following the guidelines of WGBO [23], the Dutch law that provides in rights and obligations between patient and health care professionals.

For clients who could make their own decision regarding consent for participation, information consisted of an introductory letter, an information booklet and a consent form, all with adjusted texts and pictograms to be easily readable. For clients who were not able to make this decision themselves, their legal representatives were approached, again with an introductory letter, an information booklet and a consent form. In case of doubt or unavailable information about the capability of the clients to decide for themselves, we first approached the legal representatives, giving them the possibility to forward this decision to the clients.

The study would not interfere with routine medical practice. Ethical approval was obtained (number 2008-234) from the Ethics Committee of the Erasmus University Medical Center. The study followed the guidelines of the Declaration of Helsinki [24].

## Optimizing recruitment

- Extra feature in the organizational structure is that this study was executed by PhD students, who were each employed by one of the health care organizations. This resulted in further strengthening of the connection between research and daily practice and at the same time complying with one of the requirements of the grant organization.
- A time period of around six months was reserved for the communication and practical preparation of the measurements. Extra efforts were made to design a detailed communication plan. Previous projects have shown that the success of a study in ID care depends on the commitment of the professionals in the participating health care organization [25]. Informing and motivating all involved professionals as well as different management levels is essential. Furthermore, information should be adapted to the particular professionals who are informed, for

example management versus professional caregivers. Within the three health care organizations, a top-down information route was applied, from top management to the teams of professional caregivers, and this route was extended horizontally to the local ethical committees and client councils. Preceding the study, routine meetings of these groups were used to provide oral and written information. Only after this information route was fully completed, including the level of the professional caregivers, the consent procedure was started.

- Local ethical committees and boards of clients and client representatives of the three involved care organizations were informed as well and they formally consented to this research project. This created support on different levels of the involved care organizations.
- The invitations to the participants were sent in sequential batches, to limit the time between consent and assessment and therewith minimize the loss of participants due to lack of motivation.
- Extra efforts were made to receive responses of all invited participants. Telephone calls were made to announce the sending of the consent materials, and if not returned in time, telephone reminders were made to obtain the missing consent forms. This offered the opportunity to clients and/or their caregiver to ask remaining questions about the study.
- The consent procedure was accompanied by extra information about the possibility to exclude measurements which were too stressful for a specific participant. This took away expected concerns of legal representatives and/or professional caregivers and was therefore an important extra activity in the consent procedure: After consent, intellectual or physical disabilities of various levels were taken into consideration in the actual participation in different parts of the assessment. Furthermore, the advice of the professional caregiver was to be followed concerning which parts of the assessment would be too stressful or not possible to execute for a specific client and thus be omitted. At all times unusual resistance to (parts of) the assessment by the client was leading [26].

## Outcomes

### *Inclusion*

Numbers of clients in the different phases in the consent procedure will be presented, with a detailed description of non-participants.

### *Representativeness*

To determine if the resulting sample would be representative for the base population, we collected administrative data of all clients aged 50 year and over (gender, age, type of living facility and ZZP-score). ZZP (ZorgZwaartePakket) is the Dutch classification

of levels of support, care and/or treatment as a basis for long-term financing [27] (Table 1). ID care and mental health care (MHC) have different ZZP-classifications. A small number of clients may be indicated according to the ZZP classification for mental health care, although having an intellectual disability as well. For clients who participate in day activities within the consort and obtained residential care from other care providers, the ZZP score needed to be collected elsewhere.

To determine representativeness of the included sample, we used Pearson's Chi-square test for independence, with null hypothesis that the participants and non-participants are similar (i.e. that characteristics are not depending on group).

**Table 1** *ZZP Classifications ID care [27]*

| ZZP score | Content of ZZP |
|---|---|
| 1 VG | Residence with minimal support |
| 2 VG | Residence with support |
| 3 VG | Residence with support and care |
| 4 VG | Residence with support and intensive care |
| 5 VG | Residence with support and very intensive care |
| 6 VG | Residence with intensive support, care and regulation of behavior |
| 7 VG | (Enclosed) Residence with very intensive support, care and regulation of behavior |
| Functional indication | Support with no residence (only day care or ambulatory support) |

### *Participation*

To evaluate whether health was successfully measured in this sample of older adults with intellectual disabilities, participation rates are given for four key measurements of the complete health assessment (physical examination, physical fitness test, questionnaires completed by caregivers, interviews). Data on all other measurements will be provided in separate papers concerning those measurements.

## RESULTS

### Inclusion

In figure 1 the results of the recruitment procedure are shown. Although the consent rate (consent/invited) was 1069/2150 (49.7%), the total rate of participants of the total cohort (total number/participants) was 1050/2322 (45.2%).

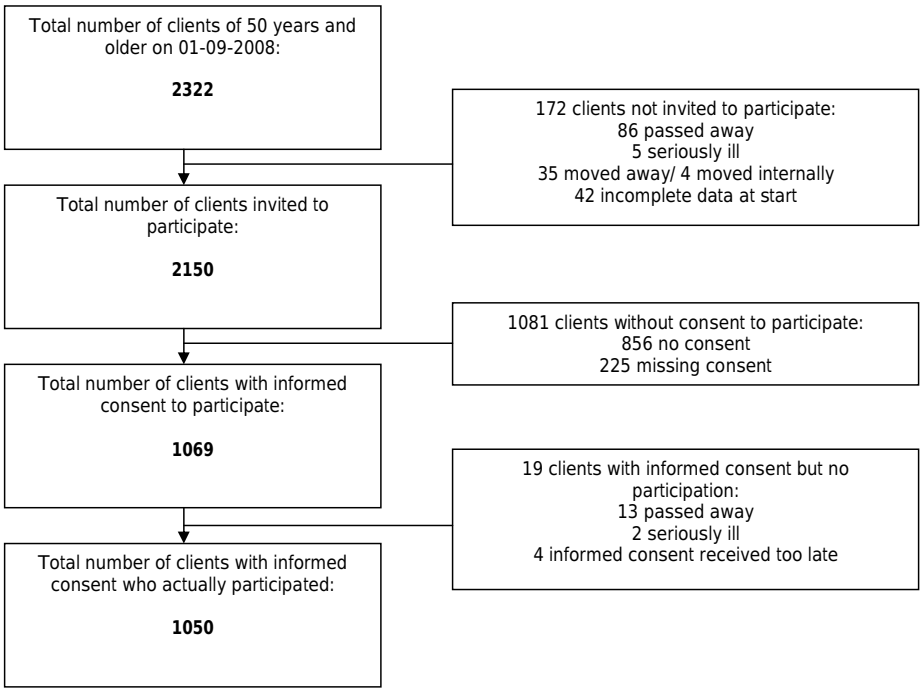


**Figure 1** *Flow chart inclusion of the HA-ID study.*

### Representativeness

In table 2 the numbers are presented for the total population of older adults in all three care organizations, for participants and for non-participants, including the contributing Chi-square terms per category. The categories with the largest deviation from the expected numbers are bold, to show which categories cause the significant differences between both groups. Overall Chi-square statistics are presented in table 3.

### Participation

Participation to parts of the health assessment is presented in table 4.

## CONCLUSION AND DISCUSSION

This paper describes how to successfully include a large sample of older adults with ID and to measure their health. A selection of instruments suitable for large-scale health assessment in this group is presented. Involvement of top and middle management in the entire process and a thorough communication plan (with a focus on key

**Table 2** *Representativeness of the study population*

| | Total population | Participants | | Non-participants | |
|---|---|---|---|---|---|
| | N | N | $(X_o\text{-}X_e)^2/X_e$ | N | $(X_o\text{-}X_e)^2/X_e$ |
| Total | 2322 | 1050 | | 1272 | |
| *Gender* | | | | | |
| Male | 1253 | 539 | 1.34 | 714 | 1.11 |
| Female | 1069 | 511 | **1.58** | 558 | 1.30 |
| *Age* | | | | | |
| 50 – 54 years | 638 | 304 | 0.83 | 334 | 0.69 |
| 55 – 59 years | 605 | 246 | 2.78 | 359 | 2.14 |
| 60 – 64 years | 471 | 224 | 0.57 | 247 | 0.47 |
| 65 – 69 years | 235 | 118 | 1.29 | 117 | 1.06 |
| 70 – 74 years | 181 | 90 | 0.82 | 91 | 0.68 |
| 75 – 79 years | 110 | 47 | 0.15 | 63 | 0.12 |
| 80 – 84 years | 56 | 11 | **8.08** | 45 | **6.66** |
| 85 – 89 years | 19 | 8 | 0.04 | 11 | 0.03 |
| 90 – 94 years | 7 | 2 | 0.45 | 5 | 0.38 |
| *Residential status* | | | | | |
| Central setting | 1159 | 557 | 0.65 | 602 | 0.56 |
| Community-based | 867 | 432 | 2.13 | 435 | 1.85 |
| Independently living with ambulatory support | 192 | 43 | **23.93** | 149 | **20.76** |
| With relatives | 19 | 7 | 0.37 | 12 | 0.32 |
| Unknown | 85 | 11 | | 74 | |
| *Level of care (ZZP-scores)* | | | | | |
| Only day care indication | 21 | 6 | 1.54 | 15 | 1.37 |
| Only indication ambulant care | 125 | 37 | **8.26** | 88 | **7.41** |
| 1 VG | 23 | 12 | 0.11 | 11 | 0.10 |
| 2 VG | 95 | 39 | 0.78 | 56 | 0.69 |
| 3 VG | 308 | 138 | 0.41 | 170 | 0.37 |
| 4 VG | 366 | 207 | **6.64** | 159 | **5.86** |
| 5 VG | 690 | 325 | 0.01 | 365 | 0.01 |
| 6 VG | 202 | 93 | 0.07 | 109 | 0.06 |
| 7 VG | 278 | 142 | 0.84 | 136 | 0.75 |
| MHC ZZP scores | 8 | 2 | 0.85 | 6 | 0.77 |
| Unknown | 206 | 49 | | 157 | |

**Table 3** *Chi-square statistics*

| Characteristic | Chi-square (df) | *p* |
|---|---|---|
| Gender | 5.3 (1) | 0.028 |
| Age | 27.41 (8) | 0.001 |
| Type of living facility | 50.55 (3) | <0.001 |
| Level of care | 41.06(9) | <0.001 |

**Table 4** *Participation to parts of the health assessment*

| Measurement | Participation |
|---|---|
| Physical examination (or part of it) | 90% |
| Physical fitness test (or part of it) | 87% |
| Questionnaires by the caregivers | 94% |
| Interviews participants themselves | 20% |

groups such as professional caregivers) proved of paramount importance to effectively organize this kind of large-scale research projects.

Not documented in this study, but an important factor in recruitment and measurements, was the actual involvement and cooperation of professional caregivers. Feedback from management of all levels in the care organizations, combined with our personal experiences in this process, suggest that the professional caregivers reacted positively to the personal communication and cooperativeness of the researcher to follow their preferences in the organization of measurements, leading to widespread cooperation during the consent procedure as well as the measurements themselves.

The actual percentage of clients with informed consent was 49.7 %. This percentage seems low, but considering the extensive health screening, which could be seen as a burden for the participant, it might be relatively good. In a multi-center study with only an assessment of visual and hearing function, the consent percentage was 61% [19].

The absence of exclusion criteria (except for age) led to a very heterogeneous population. The study population showed significant differences in all categories between participants and non-participants, so it is not a completely representative sample for the total Dutch client population. The significant difference for the category 'gender' was caused by a small overrepresentation of women. For age, the significant difference was caused by an underrepresentation of 80-84 year-olds. This could be explained by the small numbers in the higher age groups, with large consequences for representativeness by small deviations in absolute numbers. Older adults with supported living and often with an indication of ambulant care only, proved hard to reach or to motivate to participate in this study, resulting in an underrepresentation of this group in both the categories 'residential status' and 'ZZP-scores'. One possible explanation might be

that they do not recognise themselves as clients of services for people with ID or do not want to be labelled as 'intellectually disabled'. On the other hand, clients with an indication of residence with support and intensive care are overrepresented. Weighting will have to be applied for the results to be generalised to the complete older adult client population with ID in the Netherlands.

Researchers of earlier large-scale studies in populations with intellectual disabilities have reported a number of obstacles, which were avoided in this study by the carefully prepared communication routes and set-up of assessments [25]. Already in 2004, Evenhuis et al concluded that local coordination, sufficiently supported by the management, was the key factor in a successful organization of an epidemiological study in ID services [19]. Meuwese et al (2005) concluded that it is not possible to organize a large-scale intervention study without the active cooperation of the management to provide sufficient resources and support [17]. Sjoukes et al (2006) studied concept-mapping as a method to effectively introduce complex interventions, but concluded this method alone was not sufficient. This method resulted in actions which were primarily operational and ad hoc, instead of changing strategic policies of the care organizations. This resulted in a lack of motivation of the professional caregivers and the middle management [18]. In our study, involvement of top and middle management was secured in the research infrastructure. Next to management involvement in decision-making and policy strategies, they provided necessary conditions and solutions for problems in the execution of this study.

Next to the involvement of top and middle management, this paper provides a few other take home messages for the infrastructure of a large-scale multi-center study for adults with ID. First of all, good preparation of the organization of measurements is as important as designing the research protocol, and requires just as much effort and time. This preparation consists mainly of writing and executing a thorough communication plan, with specific attention for key stakeholders (i.e. professional caregivers). Involved professionals of any kind within the care organizations need to be informed and trained timely and to enhance cooperation they need to have a say in the organization and planning of the assessments. A more detailed description of the research infrastructure and management of involvement and cooperation will be published elsewhere.

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## Appendix

**Measurements with active involvement of the participant**

| Type | Outcome | Details |
|---|---|---|
| Physical assessment | Height | Seca stadiometer, type 214.<br>Body Mass Index calculated: weight divided by squared height. |
| | Knee height | Formulas Chumlea et al. [28] for calculating body height. |
| | Weight | Digital floor scale (Seca robusta type 813).<br>Body Mass Index calculated: weight divided by squared height. |
| | Fat percentage | Formulas Durnin and Womersly [29] for calculating fat percentage from the sum of four skinfolds: triceps, biceps, subscapular and suprailiacal.<br>Thickness of skinfolds measured with skinfold caliper (Harpenden). |
| | Body circumferences | Flexible tape for hip, waist, calf and upper arm circumference.<br>Waist-to-hip ratio calculated: waist circumferences divided by hip circumference. |
| | Blood pressure | Omron M7. |
| | Ankle-Arm-Index | Omron M7 (arm).<br>Boso classico and 8-MHz Doppler probe (Huntleigh MD II) (ankle).<br>Ankle-arm-index calculated: systolic blood pressure ankle divided by systolic blood pressure arm. |
| | Bone Quality | Ultrasonometer (Lunar Achilles Insight) for measuring bone stiffness calcaneus. |
| Fitness Assessment | Manual dexterity | Box and block test [30]. |
| | Response time | Response time test. |
| | Balance | Berg Balance Scale [31]. |
| | | 5 m walking speed (comfortable and fast). |
| | Muscle strength | Grip strength [32] with Jamar Hand Dynamometer (#5030J1, Sammons Preston Rolyan, USA). |
| | Muscle endurance | 30s Chair stand [33]. |
| | Cardiorespiratory endurance | 10m Incremental shuttle walking test [34].<br>Results of this test recalculated to VO2max [35]. |
| | Flexibility | Extended version of Modified back saver sit and reach test [21,36] |
| Diary | Food intake | 3-day food intake diary |
| Two weeks at home | Rest-activity rhythm | Actiwatch AW 7 (Cambridge Neurotechnologies) |
| | Physical activity | Pedometer (NL-1000, New Lifestyles, Missouri USA) |
| Meal time observation | Swallowing problems | Dysphagia Disorders Survey [37]. |
| Interview (if possible) | Self-report depression | Inventory of Depressive Symptomatology Self Report (IDS-SR) [38]. Phrasing of the questions adapted to people with ID. |
| | Self-report anxiety | Glasgow Anxiety Scale for people with an Intellectual Disability (GAS-ID) [39]. Translated version of the GAS-ID into Dutch. |
| | Self-report anxiety | Hospital Anxiety and Depression Scale (HADS) [40] -anxiety subscale. Phrasing of the questions adapted to people with ID |
| | Social contacts | Checklist about number of contacts with family, friends and peers and visiting leisure-clubs. |
| | Quality of life | Intellectual Disability Quality of Life (IDQOL-16) [41] |

| Measurements with active involvement of the participant | | |
|---|---|---|
| **Type** | **Outcome** | **Details** |
| Interview | Diagnostic interview depression and/ or anxiety | Participants with scores above the preset cut-off scores on one of the depression or anxiety questionnaires further examined by behavioral scientists trained in assessing the PAS-ADD-10 interview with participant or his/her caregiver [42] |
| Venipuncture | Biochemical markers | Fasting plasma levels: glucose, cholesterol, HDL-cholesterol, triglycerides, CRP, Hb and albumin. |
| **Measurements without active involvement of the participant** | | |
| History | Medical files | Checklist for general practitioners or ID-physicians |
| | Psychological files | Checklist for psychologists or behavioral therapists |
| | Dental files | Checklist for dentists |
| Questionnaires professional caregiver | Malnutrition | Mini Nutritional Assessment (MNA) [43]. |
| | Eating disorders | Screening Tool of fEeding Problems (STEP) [44]. Translated version in Dutch. |
| | Gastro-oesophageal reflux disease (GORD). | GORD Questionnaire: a newly developed questionnaire consisting of 50 items involving risk factors and symptoms of gastro-oesophageal reflux disease. |
| | Informant-report depression and anxiety | Anxiety, Depression, And Mood Scale (ADAMS) [45]. Translated version of the ADAMS into Dutch. |
| | Somatic complaints | Somatic complaints subscale of the Symptom Checklist-90 (SCL-90) [46] |
| | Life-events | Checklist Life Events. Newly developed checklist based on other checklists, earlier life event-studies and experience from professionals working with people with ID. |
| | Social outcome | Checklist about number of contacts with family, friends and peers and visiting leisure-clubs. |
| | Cognitive functioning | Dementia questionnaire for people with intellectual disabilities (DMR) [47] |
| | Activities of daily life and mobility | Barthel Index [48] |
| | Instrumental activities of daily Life | Questionnaire based on the Instrumental Activities of Daily Living of Lawton and Brody [49] and the Groningen Activities Restriction Scale [50-51]. |
| | Mobility | Questionnaire based on the Hauser Ambulation Index [52] and the characteristics of the Gross Motor Function Classification Scale [53]. |
| | Physical activity | Questionnaire about the participants' habitual physical activity. |
| Interview | Diagnostic interview depression and/ or anxiety | Participants with scores above the preset cut-off scores on one of the depression or anxiety questionnaires further examined by behavioral scientists trained in assessing the PAS-ADD-10 interview with the caregiver [42]. |

# Chapter 3

## Inadequate dietary intake in older people with intellectual disabilities: results of the HA-ID study

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*Submitted*

## ABSTRACT

In this study, we observed the intake of energy, protein, fat and fiber of 228 older adults with all levels of ID, compared it with Dutch recommendations for healthy dietary intake and identified its associated factors. No participants satisfied all dietary recommendations. The number of participants meeting recommendations for fiber, saturated fat and energy were especially low. High Body Mass Index (BMI) was independently associated with poor energy and protein intake, low BMI with high total fat intake, lower age with poor energy intake, higher age with poor protein intake, and lower levels of ID with poor protein intake. The data provide information for targeting subgroups that may benefit from dietary interventions.

## INTRODUCTION

It is commonly known that older people are at risk for malnutrition, which includes overnourishment and undernourishment. Overnourishment may lead to overweight and obesity, which is highly prevalent in people aged 50 years and over in the Netherlands [1]. On the other hand, undernourishment is also common in the older population, with prevalences varying from 17% in long-term care facilities to 22% in general hospitals [2]. It may lead to adverse conditions such as sarcopenia [3] and frailty [4]. In people with intellectual disabilities (ID) undernourishment is very common as well. In this population undernourishment and/or underweight may be less prevalent than overnourishment and/or overweight, but the prevalence of underweight is high compared to adults in the general population [5, 6]. Individuals with more profound disabilities experience higher rates of undernourishment [7] due to more feeding problems and swallowing problems. Regarding older people with ID, it might be assumed that the prevalence of undernourishment is even higher than in the general ageing population, because of a combination of age-related and ID-related risk factors for undernourishment. Therefore we set up this current study in which malnourishment, or more in general, nutritional status in older people with ID was studied.

To investigate an individual's nutritional status, nutritionists use several measures: (a) anthropometric testing, (b) biochemical assessments, (c) dietary intake analysis and (d) a combination of these three measures [8]. In 2009, Humphries, Traci, & Seekins reviewed the literature of these measures in studies involving adults with ID. The 28 studies they found about anthropometric testing of adults with ID have shown a bimodal distribution of body weights outside the normal range. Both over- and undernutrition were concerns in this population [9]. Published reports on biochemical testing of adults with ID are few and often limited to specific deficiencies such as vitamin D deficiency [10-12], and iron deficiency [13]. In 2008 Ohwada et al. studied albumin levels in 477 individuals with ID. They found a low prevalence of low serum albumin [14].

Studies on nutritional status assessment measured by dietary intake analysis are few as well. Moreover they are difficult to compare, because of different study populations (people living in centralized settings or people living in the community) and different outcome measures (food products, macronutrients or micronutrients), but there is some evidence that the dietary intake of adults with ID who reside in group homes is less adequate compared to individuals who reside in intermediate care facilities [15]. Nevertheless, low intake of fruit and vegetables in people with ID was demonstrated in several studies [16-18], as well as a high intake of fat [17-19] and a low intake of energy [19-22].

In the study we report in this paper, we have focused on dietary intake of adults with ID, aged 50 years and over. We were especially interested in intake of energy and proteins because of the high prevalence of protein energy malnutrition in the general ageing population [23-25], intake of fat because of the key role of restricted (saturated) fat intake in preventing cardiovascular diseases [26], and intake of fiber because of the high prevalence of constipation in people with ID [27], given the essential role of fiber in the development of constipation. Furthermore, we were interested in the associations of dietary intake. In the general population dietary intake is associated with disability, risk factors for cardiovascular disease and chronic diseases, and other health problems [28-30]. The causality of these associations is different. For example, dietary intake is a risk factor for instrumental activities of daily living (IADL) [28], whereas frailty could be a cause or a consequence of a poor dietary intake [31]. All these associations have hardly been studied in older adults with ID.

Therefore the aims of this study were (1) to provide insight in the energy, protein, fat and fiber intake of older adults with ID, (2) to compare the actual dietary intake of older adults with ID to the Dutch national guidelines for healthy dietary intake, and (3) to study the associations of dietary intake of older adults with ID with gender, age, level of ID, residential status, mobility, activities of daily living (ADL), IADL, body mass index (BMI), sarcopenia, frailty, chronic obstructive pulmonary disease (COPD), depression and congestive heart failure.

## METHODS

### Study design

This study was part of a large cross-sectional study in older adults with ID in the Netherlands, called "Healthy Ageing in people with Intellectual Disability" (HA-ID). The HA-ID study was executed by a consortium of three Dutch ID care provider services (Abrona, Amarant and Ipse de Bruggen), in collaboration with Erasmus MC, University Medical Center Rotterdam in the Netherlands. One of the main topics of the study was nutrition and nutritional status. The nutritional status of the participants was measured by anthropometric measures (BMI, waist circumference and hip circumference, sum of four skinfolds) [32], biochemical parameters (Albumin, CRP) and dietary intake. The recruitment and selection process of the HA-ID study has been outlined in detail by Hilgenkamp et al. [33]. This study has been approved by the Medical Ethics Committee of the Erasmus MC, University Medical Center Rotterdam, the Netherlands (MECnr. 2008-234) and by the ethics committees of the participating care provider services.

## Participants

The study population of the HA-ID study consisted of clients aged 50 years and over. Informed consent was obtained from 1069 clients and/or their legal representatives and 1050 clients actually participated in the assessments. Data collection took place between February 2009 and July 2010. The study population was nearly representative for the total Dutch client population aged 50 years and over receiving formal ID care, with a slight overrepresentation of women, and a slight underrepresentation of individuals living independently and individuals aged 80 years and over [33].

For this dietary intake study only a part of the HA-ID study population was studied. Because completion of food frequency lists by professional caregivers and processing the data by dieticians was very time-consuming (one hour per participant) and because of the limited availability of dieticians for this study, filling out the food frequency lists was only feasible for about one quarter of the HA-ID study population. For participants in the period between March 2009 and May 2009 and between April 2010 and June 2010 food frequency lists were collected, resulting in 287 ageing persons with ID participating in this study.

## Dietary intake

During three days (two week days and one weekend day) the food and fluid intake of the participants was recorded by their professional caregivers. To maximize the uniformity of administration of the food frequency lists, caregivers had to read the 'instructions for use of the food frequency lists' before filling in the lists. An example of a completed lists was added to the instructions.

We designed a 3-day dietary record consisting of a printed list of food and fluid products including usual portion sizes (slices, pieces, etc.). The list was divided into breakfast, snacks during the morning, lunch, snacks during the afternoon, dinner and snacks during the evening. Space was left to record foods and fluids not present in the list and nutritional supplements (appendix). Professional caregivers recorded the consumption of their clients immediately after eating or drinking of the participant.

After filling in the food frequency lists by professional caregivers, dieticians checked the lists for completeness and subsequently they processed the intake data using a web program based on nutritional values of Dutch food products [34]. The software converts the food and fluid products into intake data of energy, macronutrients and micronutrients. Data were included in the analyses if the following criteria were met: (1) three completed days, including one weekend day, (2) at least one breakfast or one lunch moment per day completed, (3) all dinner moments completed, (4) at most two eating moments per day missing.

## Energy requirements

The required energy intake was calculated by multiplying the resting energy expenditure (REE) by the physical activity level (PAL value) [35]. For REE, we used the equation proposed by Harris and Benedict (1919), which has been widely used for clinical and research purposes. By using multiple regression analysis Harris and Benedict generated the gender-specific equations, including easily measurable variables such as age, body weight and height:

Men: REE (kilocalories) = 66 + (13.7 x weight) + (5.0 x height) - (6.8 x age).

Women: REE (kilocalories) = 655 + (9.6 x weight) + (1.85 x height) - (4.7 x age).

After calculating the REE, we multiplied it with PAL-values for people with a sedentary / low active lifestyle or with a (moderately) active lifestyle, in accordance with the joint FAO/WHO/UNU expert consultation [35]. The PAL-value for people in a wheelchair was 1.2, for people with a sedentary lifestyle (5000 or less steps per day) 1.4 and for people with a more active lifestyle (> 5000 steps per day) 1.7. Finally, to assess the required energy intake a surcharge of 10% was added in case of known spasticity in accordance with outcomes of Hemingway et al. [36].

## Fat, protein and fiber intake references

The recommended intake of fat is 20 to 40 percent of total energy intake for people with a healthy weight and 20 to 35 percent of total energy intake for people with overweight, according to the dietary reference intakes for the Dutch population [37]. The tolerable upper intake level for saturated fat intake, defined as the level of intake above which there is a chance that adverse effects will occur, is 10 percent of total energy intake [37].

According to the dietary reference intakes for the Dutch population, the recommended dietary allowance for protein is 0.8 g/kg per day, which matches approximately 10 percent of total energy intake. As shown in previous studies, higher intake of protein in ageing people results in increase in muscle mass and beneficial effect on bone quality [38, 39]. For this reason and in accordance with the study of Houston et al. we set the recommended dietary allowance for protein, which is the minimal intake necessary for health reasons, at 1.2 g/kg per day [40] for people aged 65 years and over.

According to the guideline for dietary fiber intake, the optimal fiber intake per day is 35 grams for men aged 51-70 years, 30 grams for men aged 71 years and over, and 25 grams for women aged 51 years and over [41].

## Factors associated with dietary intake

Potential associated factors were chosen, based on associations found in previous studies in the general older population. Gender, age and residential status (living in

group homes in centralized settings or living in group homes in the community) were retrieved from the records of the care providers. General practitioners or specialized physicians for people with ID recorded etiology of ID (Down syndrome yes/ no) and psychologists or behavioral therapists recorded level of ID (mild [IQ > 55], moderate [IQ 35 -55], or severe [IQ < 35] ID).

Professional caregivers were asked to rate the mobility (divided into walking independently, walking with an aid and being wheelchair dependent) of the participants. They administered the Barthel Index [42], a questionnaire for ADL, and the Lawton IADL scale [43]. The Barthel Index consists of eight items with a total score between 0 (completely dependent) and 20 (completely independent). The Lawton IADL scale consists of eight items with a total score ranging from 8 (completely dependent) to 24 (completely independent).

Participants underwent a physical examination by specially trained medical assistants to assess body height and weight. Body height was measured using a Seca stadiometer, type 214, with the participant standing, wearing no shoes. For non-ambulant people, knee height was measured using a non-stretchable flexible tape, and the formula developed by Chumlea et al. [44] was used to calculate body height. Weight was measured using a digital floor scale (Seca robusta type 813), with participants wearing light clothes and no shoes. BMI was calculated by weight divided by squared height.

Sarcopenia (calf circumference, grip strength and comfortable walking speed) and frailty (weight loss, grip strength, comfortable walking speed, physical activity and exhaustion) were defined in accordance with the European Working Group on Sarcopenia in Older People [3] and the Cardiovascular Health Study [4] respectively. Measurements and results have been described in detail by Bastiaanse et al. [45] and Evenhuis et al. [46] respectively.

General practitioners or specialized physicians for people with ID recorded the diagnoses of congestive heart failure and COPD. Participants were also identified as having COPD when they were using inhalation therapy. Depressive symptoms were assessed with the depression subscale of the Anxiety, Depression And Mood Scale (ADAMS) [47]. The depression subscale of the ADAMS scale consists of seven items with a total score ranging from 0 (no depressive symptoms) to 21.

## Statistical analysis

Statistical analyses were performed with IBM SPSS Statistics 20.0. Participants were compared with non-participants within the total HA-ID study population on baseline characteristics (gender, age, level of ID, residential status and mobility) using Pearson's chi-square tests. We compared the actual intake of energy, protein, fat and fiber with the recommended intake and, by using descriptive statistics, percentages

of participants satisfying the recommendations were calculated as well as the number of unsatisfied recommendations for each participant. Independent samples t-tests and Oneway Analyses of Variance (ANOVA) were used to test whether there were differences in the absolute difference between actual and recommended intake for subgroups.

To explore associations between energy intake, protein intake, (total and saturated) fat intake, and fiber intake (dependent variables) and mobility, ADL, IADL, BMI, sarcopenia, frailty, COPD, depression and congestive heart failure (independent variables), univariate regression analyses were performed. If an independent variable showed a significant association ($p < 0.05$) with the dependent variable, it was subsequently entered into a multivariate logistic regression analysis together with participant characteristics (gender, age, level of ID, Down syndrome and residential status). The participant characteristics were added to the regression analyses independently from their association with the dependent variables, because of their potential influence on the other independent variables. Participant characteristics were entered into the equation first; mobility, ADL, IADL and BMI were entered in the second step; thirdly sarcopenia, frailty, COPD, depression and congestive heart failure were entered.

Multicollinearity, which refers to the undesirable linear association between independent variables in one regression model, was checked with Pearson's correlation coefficient. If a reasonably high correlation existed (>0.70), we entered the variables separately in the models, to see if any change in associations with the dependent variable or explaining value of the model occurred.

## RESULTS

### Participants

Due to incomplete food frequency lists, 59 out of 287 participants did not meet the inclusion criteria. They were excluded from data analyses. The patient characteristics of the remaining 228 particpants are presented in Table 1. In comparison with the total HA-ID population, in this dietary intake study men were slightly overrepresented. Moreover, people with severe and profound ID and people living in centralized settings were overrepresented, whereas people with a mild ID and people living in community settings were underrepresented.

### Dietary intake

In Table 2 both the mean actual intake and the mean recommended intake of energy, protein, total fat, saturated fat and fiber are shown. One participant (a women with Down syndrome, aged 62 years, using a wheelchair) had a modified diet based on

**Table 1** *Characteristics of participants (n = 228) and non-participants (n = 822)*

| | Participants | Non-Participants | |
|---|---|---|---|
| | *n* (%) | *n* (%) | $X^2$ |
| *Gender* | | | 5.02* |
| Men | 132 (57.9) | 407 (49.5) | |
| Women | 96 (42.1) | 415 (50.5) | |
| *Level of ID* | | | 28.64** |
| Mild (IQ > 55) | 32 (14.0) | 222 (27.0) | |
| Moderate (IQ 35-55) | 101 (44.3) | 404 (49.1) | |
| Severe (IQ < 35) | 83 (36.4) | 180 (21.9) | |
| Unknown | 12 (5.3) | 16 (1.9) | |
| *Residential status* | | | 79.02** |
| Centralized setting | 182 (79.8) | 375 (45.6) | |
| Community (group homes or independent living) | 46 (20.2) | 430 (52.3) | |
| Unknown | 0 | 17 (2.1) | |
| *Age* | | | 0.08 |
| 50-64 years | 155 (68.0) | 567 (69.0) | |
| 65 years and over | 73 (32.0) | 255 (31.0) | |
| *Mobility* | | | 4.75 |
| Independent | 153 (67.1) | 578 (70.3) | |
| With support | 37 (16.2) | 114 (13.9) | |
| Wheelchair | 32 (14.0) | 75 (9.1) | |
| Unknown | 6 (2.6) | 55 (6.7) | |
| *Down syndrome* | | | 1.73 |
| No | 172 (75.4) | 552 (67.2) | |
| Yes | 28 (12.3) | 121 (14.7) | |
| Unknown | 28 (12.3) | 149 (18.1) | |

** $p < 0.05$ ** $p < 0.01$*

fruitjuices because of excessive mucus production. Due to the modified diet, this participant had an extremely low intake of saturated fat.

Seventeen participants (7.5%) did not meet one criterion for healthy intake, 80 participants (35.1%) did not meet two criteria, 67 participants (29.4%) did not meet three criteria, 53 participants (23.3%) did not meet four criteria and 11 participants (4.8%) did not meet all criteria for healthy intake.

## Dietary intake within subgroups

Subgroups according to level of ID and residential setting did not differ in age, gender, mobility and presence of Down syndrome (data not shown). In Figure 1 the percent-

**Table 2** *Dietary intake and recommended intake of participants per day*

| | | Actual intake | | Recommended intake | | % satisfying recommended intake |
|---|---|---|---|---|---|---|
| | *n* | Mean (SD) | Range | Mean (SD) | Range | |
| Energy (kcal) | 188 | 1727 (407) | 690 – 3349 | 1986 (398) | 1087 – 3532 | 31.4 |
| Protein (g) | 192 | 75.7 (16.3) | 33.3 – 115.0 | 64.4 (18.7) | 30.2 – 146.0 | 69.8 |
| Total Fat (kcal) | 190 | 548 (172) | 182 – 1119 | LL: 346 (81)<br>UL: 642 (171) | LL: 138 – 670<br>UL: 276 – 1339 | 81.6 |
| Total Fat (% energy)[a] | 190 | 31.4 (5.1) | 14.1 – 45.4 | BMI < 25: 20 – 40 % energy<br>BMI ≥ 25: 20 – 35 % energy | | |
| Saturated Fat (kcal) | 228 | 220 (71) | 2 – 500 | 174 (40) | 69 – 335 | 10.5 |
| Saturated Fat (% energy)[b] | 228 | 12.6 (2.4) | 0.1 – 18.9 | 10 % of energy | | |
| Fiber (g)[c] | 228 | 17.8 (4.9) | 3.3 – 36.4 | Men 51-70 years: 35<br>Men ≥ 71 years: 30<br>Women ≥ 50 years: 25 | | 1.8 |

*Note. BMI = Body Mass Index; LL = Lower Limit; UL = Upper Limit*
*[a]The recommended intake for total fat in % of energy is only dependent on body mass index. [b]The recommended intake for saturated fat in % of energy is the same for the whole study population. [c]The recommended intake for fibers is only dependent on gender and age.*

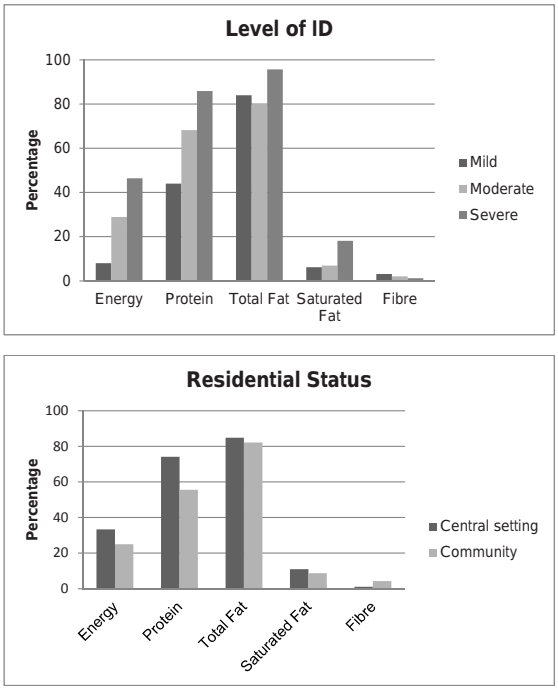


**Figure 1** *Percentages satisfying recommended intake within different subgroups.*

ages of participants satisfying the recommended intake of energy, protein, total fat, saturated fat and fiber are shown. There were significant differences in the category level of ID for energy ($F$ = 11.89, $p$ < 0.01), protein ($F$ = 9.34, $p$ < 0.01) and fat ($F$ = 9.36, $p$ < 0.01 for total fat; $F$ = 3.17, $p$ < 0.05 for saturated fat). There were no significant differences in intake between participants living in the community and participants living in centralized settings.

## Factors associated with dietary intake

Univariate and multivariate associations of the difference between actual and recommended intake of energy, protein, total fat, saturated fat and fiber (dependent variables) and patient characteristics, mobility, ADL, IADL, BMI, sarcopenia, frailty, COPD, depression and congestive heart failure are presented in Table 3. The following groups were significantly associated with a too low energy intake: people with a lower

**Table 3** *Univariate and multivariate independent associations (Odds Ratio) between satisfactory dietary intake and participant characteristics*

| | Energy | | Protein | | Total fat | | Saturated fat | | Fiber | |
|---|---|---|---|---|---|---|---|---|---|---|
| | Uni | Multi | Uni | Multi | Uni | Multi | Uni | Multi | Uni | Multi |
| Gender | | - | | - | | - | | - | | - |
| Age | | 1.14 (1.07-1.21)** | | 0.89 (0.84- 0.96)** | | - | | - | | - |
| Res. St. | | - | | - | | - | | - | | - |
| Down | | - | | - | | - | | - | | - |
| Level of ID | | - | | 2.76 (1.26- 6.05)* | | - | | - | | - |
| ADL | * | - | - | | * | - | ** | - | - | |
| IADL | ** | - | ** | - | * | - | * | - | - | |
| BMI | ** | 0.08 (0.03-0.24)** | ** | 0.19 (0.06- 0.58)** | * | 4.12 (1.10- 15.37)* | - | | - | |
| Mobility | - | | * | - | - | | ** | - | - | |
| Sarcopenia | * | - | - | | - | | ** | - | - | |
| Frailty | - | | - | | - | - | - | | - | |
| Depression | * | - | * | - | - | | - | | - | |
| COPD | - | | * | - | - | | - | | - | |
| CHF | - | | - | | * | - | - | | - | |

*Note. Dashes indicate the univariate or multivariate regression analysis was not significant. Uni = univariate analysis; multi = multivariate analysis; Res. St. = residential status; ID = intellectual disability; ADL = activities of daily living; IADL = instrumental activities of daily living; BMI = body mass index; COPD = chronic obstructive pulmonary disease; CHF = congestive heart failure.*

***** *p < 0.05; ** p < 0.01*

age and people with a higher BMI. The proportion explained variance of this multivariate logistic model (Nagelkerke $R^2$) was 0.49. People with a higher age, people with a less severe level of ID and people with a higher BMI were significantly associated with a too low protein intake. The proportion explained variance of this model (Nagelkerke $R^2$) was 0.44. In the regression model for total fat intake, only people with a lower BMI were significantly associated with a too high total fat intake. The proportion explained variance of this model (Nagelkerke $R^2$) was 0.20. For both saturated fat intake and fiber intake, there were no significant associations.

## DISCUSSION

This study provides a first overview of the dietary intake, measured with food frequency lists by proxy, of older people with ID receiving care varying from ambulatory support to intensive residential care. The results show that most of the participants poorly match the daily recommendations. With 82%, most of the participants have satisfied the recommendations for total fat intake. However, 30% of the participants, especially those with a higher age and a less severe level of ID, eat insufficient amounts of proteins. This is a substantial group of people that is more vulnerable for developing sarcopenia. For energy, saturated fat and fiber intake the percentages of participants meeting the criteria for healthy intake are worrying. Especially the recommendations for fiber intake are hardly met in this population in which constipation is very prevalent.

It is remarkable that people with a higher BMI more often consume insufficient amounts of energy. There may be some participants with a low calorie diet because of their overweight and this may be one explanation for the negative association of energy intake and BMI. Another explanation may be the consumption of snacks (which contain relative large amounts of calories) by people with mild to moderate ID, at times when there is no supervision. In a previous study in the HA-ID population, it was shown that a higher BMI was significantly associated with eating independently and doing groceries independently [32]. Thus, people with a higher BMI have more possibilities to eat without supervision.

The mean energy and protein intake in our study is lower than those in young adults with Down syndrome in Spain (2260 kcal/day [*95% CI* 2137-2383] and 90.7 gram/day [*95% CI* 86.3-95.1] for men and 1909 kcal/day [*95% CI* 1722-2096] and 88.1 gram/day [*95% CI* 82.1-94.1] for women) [48]. However, fiber intake is equal in both populations (20.4 gram/day [*95% CI* 18.8-22.0] for men and 17.0 gram/day *[95% CI* 15.5-18.5] for women). This may reflect relatively more attention for fiber intake in the Netherlands especially for older people in centralized settings with specialized medical care who are prevalent in the current study, although fiber intake for this

group is not sufficient as well. For older people without ID living in the Netherlands there are only data available for people aged 50 to 70 years [49]. In this population the median energy intake (2390 kcal/day for men and 1849 kcal/day for women), the median protein intake (95 gram/day for men and 74 gram/day for women) and the median fiber intake (21 gram/day for men and 19 gram/day for women) are higher compared to our study population. Thus, older people in the general population seem to have a more adequate diet compared to their age equivalents with ID.

The strengths of this study are the systematic and objective data collection. Food frequency lists were immediately completed after eating or drinking of the participants and therefore this type of measures is more accurate than recall methods [50]. Another strength is the processing of data by dieticians, which was necessary because of the enormous variety of food and drink products in the Netherlands.

The most important limitation of this study is the representativeness of the study population, in which people with severe and profound ID and people living in centralized settings were overrepresented. This may reflect difficulties for professional caregivers working with people with ID not living in centralized settings to properly administer the food frequency lists, because, although not significantly different, in the excluded 59 participants there were relatively more people living in the community or living independently (data not shown). Another limitation is the proxy report of dietary intake in our study protocol. To maximize uniformity in administration of food frequency lists, we have chosen only for professional caregivers to fill in the lists. However, the use of proxy reporters is only adequate in case of continuous support. People living in group homes in the community or people living independently (46 participants in our study) do not need continuous support.

Our results show that a worrying percentage of older people with ID can be classified as having an inadequate diet. More longitudinal research is needed for drawing conclusions about causality for some associations. For example, the (univariate) association between (1) energy and protein intake and (2) depression may be two-fold: On the one hand it is known from the literature that one of the symptoms of depression is decreased dietary intake. On the other hand it may also be because people have poor intake, their general condition is reduced and / or their quality of life is diminished which may result in depression. Second, more research is needed for developing an instrument that can reliably measure dietary intake in people with ID who do not have continuous support. So far dieticians have not validated such an instrument because of various barriers, such as problems in communication and problems with memory. For example by developing mobile applications for this population it might be possible to create a new valid instrument to measure dietary intake. An additional advantage of using mobile applications is that it could be less time consuming and less labor-intensive than food frequency lists.

This study has shown that more attention is needed for food intake in daily care for older people with ID. More knowledge about healthy nutrition and the harmful effects of an unhealthy diet is necessary for professional caregivers. Cooking classes for people with ID and their caregivers might be a fun and useful method to start with. In addition, an intervention should be developed with the purpose of improving the nutritional state of older people with ID. Within the HA-ID study, we recently started with the development of such an intervention. By using Intervention Mapping [51], the first step in this process is to consider what the barriers and facilitators are for professionals working with people with ID in the Netherlands for promoting a healthy dietary intake.

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## **Appendix** *Food frequency list*

Name participant: ……. Name caregiver:…… Date: …./…./……..

**Restrictions in food texture and viscosity?**
No
Yes: ground/pureed/liquids restricted

| **Breakfast** | **Amount** |
|---|---|
| Bread white/wheat/whole wheat | ….. Pieces |
| Croissant…… | ….. Pieces |
| Cracker/rusk…….. | ….. Pieces |
| Porridge……. | ….. ml |
| Other namely…… | ….. |
| Diet-margarine/margarine/butter….. | |
| Cheese 20+/30+/48+…….. | ….. Cuts |
| Cold meat namely…… | ….. Cuts |
| Sweet spread namely…… | ….. |
| Other namely……. | ….. |
| Coffee/tea//water……. | ….. ml |
| Sugar…… | ….. Cubes |
| Milk…….. | ….. Splashes |
| Milk skimmed/low-fat/full-cream | ….. ml |
| Buttermilk…… | ….. ml |
| Other namely…….. | ….. |

| **During the morning** | |
|---|---|
| Coffee/tea/water…… | ….. ml |
| Sugar…… | ….. Cubes |
| Milk…… | ….. Splashes |
| Milk skimmed/low-fat/full-cream | ….. ml |
| Buttermilk…… | ….. ml |
| Soda regular/light…… | ….. ml |
| Fruit juice namely……. | ….. ml |
| Other namely……. | ….. |
| Cookie namely…….. | ….. Pieces |
| Fruit namely……... | ….. Pieces |
| Other namely……. | ….. |

| **Lunch** | |
|---|---|
| Bread white/wheat/whole wheat | ….. Pieces |
| Croissant…… | ….. Pieces |
| Cracker/rusk…….. | ….. Pieces |
| Porridge……. | ….. ml |
| Other namely…… | ….. |
| Diet-margarine/margarine/butter….. | |
| Cheese 20+/30+/48+…….. | ….. Cuts |
| Cold meat namely…… | ….. Cuts |
| Sweet spread namely…… | ….. |
| Other namely……. | ….. |
| Coffee/tea//water……. | ….. ml |
| Sugar…… | ….. Cubes |
| Milk…….. | ….. Splashes |
| Milk skimmed/low-fat/full-cream | ….. ml |
| Buttermilk…… | ….. ml |
| Other namely…….. | ….. |

| **During the afternoon** | **Amount** |
|---|---|
| Coffee/tea/water…… | ….. ml |
| Sugar…… | ….. Cubes |
| Milk…… | ….. Splashes |
| Milk skimmed/low-fat/full-cream | ….. ml |
| Buttermilk…… | ….. ml |
| Soda regular/light……. | ….. ml |
| Fruit juice namely……. | ….. ml |
| Other namely……. | ….. |
| Cookie namely…….. | ….. Pieces |
| Fruit namely……... | ….. Pieces |
| Other namely……. | ….. |

| **Dinner** | |
|---|---|
| Starter namely……. | ….. ml |
| Soup namely…… | ….. ml |
| Boiled potatoes…….. | ….. Pieces |
| Gravy…… | ….. Gravy spoon |
| Mash pot namely..……. | ….. Serving spoon |
| Fried potatoes/French fries…. | ….. Serving spoon |
| Rice white/brown…… | ….. Serving spoon |
| Pasta (whole wheat yes/no)…. | ….. Serving spoon |
| Pulse namely…….. | ….. Serving spoon |
| Vegetables namely…… | ….. Serving spoon |
| Raw vegetables namely..…… | ….. Schaaltje |
| Meat namely…… | ….. Gram/Portion |
| Fish namely……. | ….. Gram/Portion |
| Fat for preparation | |
| Olive oil……. | ….. Table spoon |
| Margarine…….. | ….. Table spoon |
| Butter……. | ….. Klontje |
| Dessert namely……. | ….. Dishes |
| Fruit namely……... | ….. Pieces |
| Other namely……. | ….. |

| **During the evening** | |
|---|---|
| Coffee/tea/water…… | ….. ml |
| Sugar…… | ….. Cubes |
| Milk…… | ….. Splashes |
| Milk skimmed/low-fat/full-cream | ….. ml |
| Buttermilk…… | ….. ml |
| Soda regular/light…… | ….. ml |
| Fruit juice namely……. | ….. ml |
| Other namely……. | ….. |
| Cookie namely…….. | ….. Pieces |
| Fruit namely……... | ….. Pieces |
| Other namely……. | ….. |

| **Others** | |
|---|---|
| Cake/pastry namely..…… | ….. Pieces |
| Crisps/snacks namely..…… | ….. |
| Eggs boiled/fried…..…….. | ….. Pieces |
| Liquor namely……..…… | ….. ml |

**Diet products**

| | |
|---|---|
| Thickeners namely……. | ….. number of spoons per cup |
| Nutritional drinks namely…… | ….. bottles per day |
| Others namely…….. | ….. |

Circle the products that the participant has eaten.

# Chapter 4

## Dysphagia in older people with intellectual disabilities: results of the HA-ID study

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*Submitted*

## ABSTRACT

In the general ageing population, dysphagia is a common disorder. However, the prevalence of dysphagia and its contributing factors in older people with intellectual disabilities (ID) are unknown. Therefore, swallowing function was determined in 931 people with borderline to profound ID, aged 50 years and over, by using a standardized mealtime observation, the Dysphagia Disorder Survey (DDS). The prevalence of dysphagia was calculated by using descriptive statistics. To identify the associations of dysphagia, logistic regression

analysis was performed. Finally, the awareness of dysphagia amongst professionals was studied by comparing the results of the DDS with registration of swallowing problems in medical files.

According to the DDS, 51.7% had moderate to severe dysphagia. In 89.5% of the study population with dysphagia, swallowing problems had not been registered in medical files. Higher age, Down syndrome, mobility impairment, feeding with help, and use of benzodiazepines were positively and independently associated with dysphagia. The prevalence of dysphagia in older people with ID is comparable to the prevalence found in nursing home residents. More attention to screening and diagnosing dysphagia is needed to prevent complications.

## INTRODUCTION

Dysphagia is a common disorder within the older general population, which may lead to life-threatening health problems such as choking, aspiration and aspiration pneumonias [1-6]. In a Swedish study among 556 people aged 50 years and over in 1991, it was shown that 35% (95% CI: 31.1% - 39.2%) had difficulties during eating and swallowing [7]. In a more recent study among 634 healthy citizens in the UK aged 69 years and over, 11.4% (95% CI: 9.1% - 13.7%) of the population had symptoms that were indicative of significant oropharyngeal dysphagia [8]. In a frail population, nursing home residents aged 65 years and over, the prevalence of dysphagia was 52.7% (95% CI: 47.7% - 57.5%) [9].

Risk factors for dysphagia have also been studied among ageing people. Among others, higher age, stroke, dementia, more dependent functional status, underweight, and centrally acting medication (antipsychotics, benzodiazepines, and anticonvulsant drugs) are known for their association with dysphagia [6, 9].

With regard to the population of people with intellectual disabilities (ID), published prevalence rates of dysphagia vary from 8.2 % to 99% [10-13]. This wide range is caused by differences in studied samples and methods used to measure dysphagia. In studies among people with more severe levels of ID, higher prevalence rates were found [13].

Until now, there is hardly any literature available that combines the above mentioned high risk populations: the ageing population with ID. This study was aimed at describing the prevalence of dysphagia within the population with ID aged 50 years and over and the correlation between dysphagia and patient characteristics (gender, age, level of ID, Down syndrome, residential status), mobility, spasticity, dependency on feeding, body mass index (BMI), stroke, dementia, and centrally acting medication use. Finally, because of the occurrence of unanticipated swallowing incidents in clinical practice, the awareness of dysphagia amongst professionals was estimated by comparing the study outcomes with registration of swallowing problems in medical files.

## METHODS

### Study design and participants

This study was part of the large cross-sectional study "Healthy Ageing and Intellectual Disability (HA-ID)" [14]. Ethical approval was obtained (number 2008-234) from the Medical Ethics Committee of the Erasmus Medical Center Rotterdam. The study population consisted of clients, aged 50 years and over, of three care provider services in the Netherlands. Informed consent was obtained from 1069 clients and/or their legal

representatives and 1050 clients actually participated in the assessments. The study population was nearly representative for the total Dutch client population aged 50 years and over of the care providers, with a slight overrepresentation of women and a slight underrepresentation of individuals living independently and individuals aged 80 years and over. Information about recruitment and design of this study has been published elsewhere [14].

## Dysphagia

Swallowing function was observed by specifically trained speech and language therapists or dieticians. They used the Dysphagia Disorder Survey (DDS), a standardized mealtime observation developed for people with ID [15]. The DDS is divided in two parts. Part one (seven items) consists of related factors of swallowing, such as consistency of food, and body position. Part two (items 8 -15) is the actual mealtime observation. It assesses signs of the neuromotor and behavioural function of swallowing divided in four phases (preparatory, oral, pharyngeal, and oesophageal phase). The observation is applied to three food consistencies; solid, non-chewable, and liquid. The maximum score of part two is 22 points; it is positively related to more signs of dysphagia.

The outcomes of the second part of the DDS were used for an ordinal dysphagia severity score (DSS) [13]. People without any points on this part of the DDS were categorized as 'no dysphagia' and people with one or more points were categorized as 'mild dysphagia'. Change in the pharyngeal and oesophageal phase is related to aspiration of food, so a positive score on one of these items (13 and 14) was an indication of 'moderate to severe dysphagia'. When tube feeding was used the participant was scored as having 'profound dysphagia'.

The DDS was found to have the strongest clinical utility in children with intellectual disabilities [16]. Its psychometric properties were investigated in 1988. Pearson Correlation Coefficients between DDS sub-scores for Part 1 and Part 2, and the total DDS score on the one hand and the expert diagnosis of dysphagia by clinicians on the other hand ranged from $r = 0.84$ to $r = 0.97$ ($p < 0.01$). Cronbach's Alpha scores for internal consistency of DDS subscales Part 1 and Part 2, and the total DDS were good: 0.82, 0.86, and 0.91 respectively. Inter-rater reliability was determined in a subsequent study. Three pairs of speech-language pathologists participated. Each pair tested 7 subjects. There were eight disagreements in 336 pairs of judgments. This is a 97% agreement rate and it was considered to be adequate [17].

## Associated factors

Information on age, gender, and residential status was collected from the records of the care provider services. Residential status was categorised as centralized setting,

community-based setting, and living independently with ambulatory support or living with relatives. Level of ID was obtained from behavioural therapists' and psychologists' records (mild, moderate or severe). General practitioners and specialized physicians for people with ID recorded aetiology of ID (Down syndrome) and information on dysphagia, spasticity, stroke, and medication. Antipsychotics, anticonvulsant drugs, and benzodiazepines with a long half-life time (>12hrs) were taken into account for this study. Diagnoses of dementia (defined as 'possible or probable dementia') were obtained from the participants' physicians and behavioural specialists, and were only included in the analysis in case of consensus between these professionals.

Professional caregivers were asked about mobility, categorized as walking independently, walking with aids, and mobility in wheelchair. They administered the Barthel Index [18], a questionnaire for activities of daily living (ADL) in which one item concerns dependency on feeding. This item is categorized into dependent, partly dependent, and completely independent.

BMI was calculated by weight divided by squared height [19]. Body height was measured using a Seca stadiometer, type 214, with the participant standing, wearing no shoes. For people who were unable to stand up straight, knee height was measured using a non-stretchable flexible tape. Subsequently, body height was estimated by using equations developed by Chumlea et al [20]. Weight was measured using a digital floor scale (Seca robusta type 813), with participants wearing light clothes and no shoes.

## Statistical analysis

Statistical analyses were performed using Statistical Package for Social Sciences for Windows version 20 (SPSS Inc., Chicago, Il, USA). Prevalence with 95% confidence intervals for mild to profound dysphagia was calculated by using descriptive statistics. Descriptive statistics were also used for comparing the results of the DDS with registration of swallowing problems in medical files.

To explore associations, we divided the participants in two groups. The first group were the participants with no dysphagia and those with mild dysphagia (because for participants in both groups the risk for developing aspiration pneumonia is low) and the second group were the participants with moderate to severe dysphagia and those with profound dysphagia. First, univariate regression analyses were performed (chi square tests for categorical variables and ANOVA tests for continuous variables) with dysphagia (no/mild = 1, moderate/severe = 2) as dependent variable and gender (men = 1, women = 2), age, level of ID (borderline/mild = 1, moderate = 2, severe/profound = 3), residential status (centralized setting = 1, community setting = 2, independent or with relatives = 3), Down syndrome (no = 1, yes = 2), mobility (independent = 1, walking with aids = 2, sitting in a wheelchair = 3), spasticity (no = 1,

yes = 2), dependency on feeding (completely independent = 1; partly dependent = 2; completely dependent = 3), BMI, stroke (no = 1, yes = 2), dementia (no = 1, yes = 2), use of anticonvulsant drugs (no = 1, yes = 2), use of benzodiazepines (no = 1, yes = 2), and use of antipsychotic drugs (no = 1, yes = 2) as independent variables. Second, a logistic regression analysis was performed with determinants that were found to be significantly ($p<0.05$) or nearly significantly ($p<0.10$) associated with dysphagia. Multicollinearity was checked for all independent variables with the variance inflation factor (VIF) of linear regression analysis. VIF values above 10 indicated multicollinearity [21].

## RESULTS

### Prevalence

We obtained a complete DDS form for 931 participants. Reasons for non-participation were informed consent limited to file information (n=12), illness (n=7), refusal (n=57), incomplete observation forms (n=6), deceased (n=1), other reasons (n=36) or unknown (n=12). The mean age of the participants was 61.6 years (range 50-93; SD = 8.05). Other characteristics of the participants are shown in Table 1.

**Table 1** *Characteristics of participants (n = 931)*

| | *n* (%) |
|---|---|
| *Gender* | |
| Men | 473 (50.8) |
| Women | 458 (49.2) |
| *Level of ID* | |
| Mild (IQ > 55) | 229 (24.6) |
| Moderate (IQ 35-55) | 447 (48.0) |
| Severe (IQ < 35) | 235 (25.2) |
| Unknown | 20 (2.1) |
| *Residential status* | |
| Centralized setting | 495 (53.2) |
| Community based setting | 388 (41.7) |
| Living independently | 48 (5.2) |
| *Down syndrome* | |
| No | 690 (74.1) |
| Yes | 134 (14.4) |
| Unknown | 107 (11.5) |

Dysphagia was present in 710 out of 931 participants (77.4%; 95% CI = 74.7% – 80.0%). 235 participants (25.2%; 95% CI = 22.6% - 28.1%) had mild dysphagia, 481 participants (51.7%; 95% CI = 48.5% - 54.9%) had moderate to severe dysphagia and five participants (0.5%; 95% CI = 0.2% - 1.3%) were partly or completely dependent on tube feeding.

Medical files were available for 824 participants, of which 68 (8.3%; 95 CI = 6.6% - 10.3%) had a diagnosis of dysphagia. In 570 out of 637 participants with any level of dysphagia according to the DDS (89.5%; 95% CI = 86.9% - 91.6%), swallowing problems were not registered in the medical files.

### Associated factors

Because of missing values, univariate analyses with dysphagia and the independent variables were performed with different numbers. The outcomes of the univariate analyses showed that dysphagia was significantly associated with age, level of ID, residential status, Down syndrome, mobility, spasticity, feeding dependency, BMI, dementia, stroke, use of anticonvulsant drugs, and use of benzodiazepines (Table 2). All these independent variables were entered in the multivariate logistic model.

Table 3 shows that higher age, Down syndrome, mobility impairment, feeding with help, and use of benzodiazepines were positively associated with moderate to profound dysphagia.

The explained variance of the model ($R^2$ Nagelkerke) was 0.18. VIF values were well below the threshold for multicollinearity.

## DISCUSSION

For the first time, frequency of dysphagia has been investigated in a large population of ageing people with ID which is considered to be representative for the total Dutch population of older clients of formal ID service providers [14]. The prevalence of dysphagia was 77.4% and that of severe dysphagia was nearly 52%. In 89.5% of the participants with dysphagia, swallowing problems were not registered in medical files. Associated independent factors of dysphagia known from the general population were also found in our study: older age, mobility impairment, feeding dependency, and use of benzodiazepines were positively associated with dysphagia. Also people with Down syndrome (DS) were prone to have dysphagia. Swallowing problems have been investigated before in children with DS. For example, a high prevalence of 58% was found in a group of 201 children with DS [22], probably due to reduced oral sensorimotor skills, developmental delays, and generalized hypotonia [23]. As far as we know, only one study by Thacker et al. has previously described a positive association between

**Table 2** *Univariate associations of dysphagia with independent variables*

| | | $X^2$ | *n* | t | *p*-value |
|---|---|---|---|---|---|
| Gender | Male | 473 | 0.02 | | 0.89 |
| | Female | 458 | | | |
| Age | | 931 | | -3.23 | < 0.01 |
| Level of ID | Mild | 229 | 28.32 | | < 0.01 |
| | Moderate | 447 | | | |
| | Severe | 235 | | | |
| Residential setting | Central | 495 | 16.57 | | < 0.01 |
| | Community | 388 | | | |
| | Independent | 48 | | | |
| Down syndrome | No | 690 | 9.79 | | < 0.01 |
| | Yes | 134 | | | |
| Mobility | Independent | 663 | 43.49 | | < 0.01 |
| | Walking aid | 135 | | | |
| | Wheelchair | 96 | | | |
| Spasticity | No | 710 | 23.92 | | < 0.01 |
| | Yes | 92 | | | |
| Feeding dependency | No | 537 | 67.31 | | < 0.01 |
| | Partial | 281 | | | |
| | Yes | 76 | | | |
| Stroke | No | 762 | 5.26 | | 0.02 |
| | Yes | 45 | | | |
| BMI | | 836 | | 3.00 | < 0.01 |
| Dementia | No | 838 | 8.02 | | < 0.01 |
| | Yes | 75 | | | |
| Anticonvulsant drugs | No | 651 | 18.83 | | < 0.01 |
| | Yes | 173 | | | |
| Antipsychotic drugs | No | 524 | 0.43 | | 0.51 |
| | Yes | 300 | | | |
| Benzodiazepines | No | 776 | 9.09 | | < 0.01 |
| | Yes | 48 | | | |

*n* **=** *number of participants; ID = intellectual disability; BMI = body mass index.*

swallowing problems and DS in adults [24]. The current study shows that there is also a positive relation between dysphagia and Down syndrome in old age.

Prevalence of moderate to severe dysphagia within our study population can be compared to that in an average nursing home population [9]. In general, the mean age of people living in a nursing home is higher compared to our study population. Based on the results in the current study, we may conclude that dysphagia is already prevalent in older adults with ID at a relatively young age. This is probably due to the

**Table 3** *Independent associations of dysphagia*

| | *B* | Wald | OR | 95% CI OR | |
|---|---|---|---|---|---|
| | | | | *LL* | *UL* |
| Age** | 0.04 | 10.77 | 1.04 | 1.02 | 1.06 |
| Residential setting | -0.06 | 0.13 | 0.94 | 0.68 | 1.30 |
| Down syndrome** | 0.88 | 12.03 | 2.41 | 1.47 | 3.95 |
| Level of ID | 0.15 | 1.19 | 1.16 | 0.89 | 1.52 |
| BMI | -0.03 | 2.85 | 0.97 | 0.94 | 1.01 |
| Mobility* | 0.34 | 3.94 | 1.41 | 1.00 | 1.97 |
| Dementia | 0.19 | 0.23 | 1.21 | 0.56 | 2.60 |
| Stroke | 0.45 | 1.12 | 1.57 | 0.68 | 3.60 |
| Spasticity | 0.59 | 3.29 | 1.81 | 0.95 | 3.42 |
| Feeding dependency** | 0.48 | 7.88 | 1.61 | 1.16 | 2.24 |
| Anticonvulsant drugs | 0.22 | 0.89 | 1.25 | 0.79 | 1.96 |
| Benzodiazepines* | 1.09 | 5.85 | 2.98 | 1.23 | 7.20 |

*Note. Proportion of variance explained by the model (Nagelkerke $R^2$): 0.18.*
*OR = odds ratio; 95% CI = 95% confidence interval; LL = lower limit; UL = upper limit; ID = intellectual disability; BMI = body mass index.*
** $p < 0.05$; ** $p < 0.01$*

high rate of pre-existing neurological problems in the total group of people with ID. These neurological problems may be an explanation for the low explained variance of the logistic regression model, since most other known risk factors for dysphagia were entered in the model.

The strengths of this study are the large study population and the comprehensive set of predictors. A limitation of this study is its cross-sectional design. Because of this design, we could not investigate the causality of the found associations. Second, the DDS is a screening instrument. To diagnose dysphagia, more invasive tests are needed such as the video-fluoroscopic swallowing study, which is the gold standard. Therefore, the comparison of DDS scores and the medical files should be interpreted with caution.

In conclusion, dysphagia is highly prevalent in older adults with ID. The high prevalence, the low explained variance of the model, and the not so high odds ratios of the significant associated factors, implicate that professionals working with people with ID, must have a proactive attitude against screening, diagnosing, and treating dysphagia in the entire population of people with ID, to prevent life-threatening situations. Professional caregivers working with people with ID should be educated on risk symptoms.

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# Chapter 5

## Feasibility and reliability of the Mini Nutritional Assessment in older adults with intellectual disabilities

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## ABSTRACT

The Mini Nutritional Assessment (MNA) is internationally widely used and validated for use in healthy and frail older adults. However, the psychometric properties of the MNA have not been evaluated for older people with intellectual disabilities (ID). Therefore the feasibility and the reliability of the MNA in older clients of ID care providers were investigated.

The MNA was performed by means of interviews with participants (N = 12) and caregivers (N = 48) and physical assessments of participants (N = 47). Aspects of feasibility were completion of interview, difficulty of answering interview items, duration of interview, and completion of physical assessment. Aspects of reliability were inter-observer reliability between caregivers and between participants and caregivers, test-retest reliability and internal consistency. For inter-observer and test-retest reliability, intraclass correlation coefficients (ICC) were calculated, and for internal consistency Chronbach's alpha.

All participants and caregivers completed the interview part. For 7 out of 12 personally interviewed participants and none of the caregivers, at least 3 out of 15 questions were difficult to answer. Mean duration of the interview was 7 minutes in participants and 4 minutes in caregivers. Physical assessment was successfully performed in 40 participants (85.1%). In the remaining 7 participants (14.9%) missing values were retrieved from the medical records.

ICCs (95% confidence interval) for test-retest and inter-observer reliability between caregivers were good, 0.85 (0.72 – 0.92) and 0.86 (0.74 – 0.92) respectively, but ICC for inter-observer reliability between caregivers and persons with ID was low, 0.03 (-0.51 – 0.59). Internal consistency was 0.61.

We conclude that the MNA is feasible and reliable for older people with ID. Interview data can be reliably obtained through caregivers, but not through people with ID.

## INTRODUCTION

Malnutrition negatively influences quality of life [1] and is associated with social isolation, increased morbidity and increased mortality [2-4]. It is frequent in the general Dutch older population with prevalences of 21.7% in home-care organisations and19.2% in nursing homes [3]. Malnutrition is also a common disorder in people with intellectual disabilities (ID) of all ages [5]. Specific risk factors in this population are impaired mobility, swallowing disorders, gastro-oesophageal reflux disease and polypharmacy [6-8]. Although evidence is currently lacking, it might be assumed that the prevalence of malnutrition in older people with ID is higher than in the general ageing population, because of a combination of age-related and ID-related risk factors for malnutrition.

Apart from clinical diagnostic tests, such as anthropometry, bioelectrical impedance analysis or biochemical markers, several screening instruments are used for detecting malnutrition . The Mini Nutritional Assessment (MNA), developed by Guigoz et al. in 1994 [9] is internationally widely used and validated for use in healthy and frail older adults [10]. It is considered to be a "gold standard" for ambulatory living ageing people and those living in long-term care facilities [11]. It may be suitable for ageing people with ID as well. However, the psychometric properties of the MNA have not been evaluated for this population. Therefore the feasibility and the reliability of the MNA in older clients of ID care providers were investigated.

## METHODS

### Participants

This study was part of a large-scale cross-sectional study among older adults with ID, called "Healthy Ageing in people with Intellectual Disabilities (HA-ID)" [12]. In this study 1050 adults with ID, aged 50 to 93 years, were investigated. All participants were allied to a care provider for people with ID. Both people living in the community (low or medium care and support) and people living in central locations (high care and support) were included.

This study was approved by the Medical Ethics Committee of Erasmus Medical Center, Rotterdam, the Netherlands (MECnr. 2008-234). Informed consent for participation was obtained from the participants themselves or their legal representatives.

Sample size for the present feasibility and reliability study was established according to estimates provided by Walter et al. for reliability studies using intraclass correlation coefficients (ICCs) [13]. To test reliability at a 95% significance level and a power of

80% (β=0.20), a sample of at least 46 cases was required based on two observers with a criterion coefficient value of 0.8 and a true value of 0.9.

### MNA

The full Mini Nutritional Assessment (MNA) involves 6 general (about residential status, psychological problems, mobility, medication and skin ulcers), 4 anthropometric (about weight and height to calculate BMI, arm and calf circumferences, and weight loss), 6 dietary (about number of meals, food and fluid intake, and autonomy of feeding) and 2 subjective (about self-perception of health and nutrition) items. In the general population it can be administered in less than 15 minutes. Three items (Body Mass Index, arm circumference and calf circumference) are assessed by means of a physical assessment. The 15 other items are assessed by means of an interview. If data cannot be obtained through a physical assessment or an interview, it is allowed according to the user guide [14] to use the medical record of the participant for most of the items. The total score (0-30 points) categorizes the results as follows: (1) well-nourished (24 points or more); (2) at risk for malnutrition (17-23.5 points); and (3) malnourished (< 17 points). For persons who are labeled 'at risk' or 'malnourished', referral to a dietitian is recommended for further assessments and nutritional intervention. [10].

In the present study, one trained researcher performed the physical assessment in all participants and interviewed both the participants (if possible) and the professional caregivers. Only participants with a borderline, mild or mild-to-moderate level of ID were interviewed. They had to be able to answer questions about their nutritional status and their health status over the last three months before the interview. For these participants, items were simplified according to the user guide of the MNA [14]. One question (about neuropsychological stress) was excluded from the participant interview because it was not possible to simplify this question without changing its meaning. The interviewed caregivers were required to have been working with the participant for at least three months.

Weight, height and calf and arm circumferences were measured according to the user guide of the MNA [14]. If participants were unable to stand upright, knee height was measured and equations developed by Chumlea et al. were used to calculate body height [15]. For immobile participants, recent weight was retrieved from the medical records.

### Feasibility

The following aspects of feasibility were studied: completion of the interview, difficulty of the items of the interview, duration of the interview and completion of the physical assessment. Feasibility for participants and feasibility for caregivers were separately studied. Criteria for judgment of feasibility for participants were (1) all interwiew items

answered; (2) at least 13 out of 15 (85%) items understood; (3) maximum duration of interview 15 minutes and (4) physical assessment successfully performed or required information successfully retrieved from medical files. For professional caregivers these criteria were: (1) all interwiew items answered; (2) at least 13 out of 15 (85%) items easily answered and (3) maximum duration of interview 15 minutes.

To meet the second criterion for participants, after administration of the interview the researcher asked them if they understood all questions, whereas during the interview, the researcher judged whether the questions were understandable by observing non-verbal signs, such as hesitations during replying and facial expressions of uncertainty. If the participant answered 'no' and/ or the researcher judged negatively, this criterion was not met. To meet the second criterion for caregivers the researcher asked them if it was easy to answer the questions about someone else.

Participants as well as caregivers had to meet all criteria to consider the MNA feasible for each individual. The MNA was considered to be feasible for participants with ID and their caregivers if at least 90% of them met all criteria.

## Reliability

Test-retest reliability of the interview part completed by caregivers, inter-observer reliability between two different caregivers, respectively between participants and their caregivers, and internal consistency were studied.

For test-retest reliability, two weeks following the initial administration of the MNA, the same caregiver was interviewed again. If, according to the caregiver, the clinical status of the participant had changed during this period, data were excluded from the analysis for test-retest reliability.

For inter-observer reliability between caregivers, two caregivers of the same participant were interviewed independently at the same day. For inter-observer reliability between participants and their caregivers the scores of the participants were compared to the scores of the caregivers' first interview.

## Statistical analysis

Statistical analysis was performed with SPSS 15.0

Descriptive statistics were used to express feasibility. For inter-observer and test-retest reliability intraclass correlation coefficients (ICC) including 95% confidence intervals were calculated. For the internal consistency Crohnbach's Alpha was calculated.

The reliability of the interview was categorized as poor (ICC/ alpha ≤ 0.40), moderate (0.41 ≤ ICC/ alpha ≤ 0.60), substantial (0.61≤ ICC/ alpha ≤ 0.79), or excellent (ICC/ alpha ≥ 0.80) [16].

## RESULTS

### Population

The first 48 participants, aged 50 to 89 (mean 64.6 years (SD 9.7)) of the HA-ID study were included in the present study. Sex, residential status and level of ID are described in table 1.

47 out of 48 participants underwent the physical assessment, for one participant there was no permission for this. 12 participants with borderline or mild ID were eligible for the interview. Because most caregivers were responsible for two or more included participants, 16 different caregivers for 48 participants were interviewed as well.

**Table 1** *Characteristics of participants*

| | *n* | % |
|---|---|---|
| *Sex* | | |
| Men | 13 | 27.1 |
| Women | 35 | 72.9 |
| *Level of ID* | | |
| Borderline (IQ 70-85) | 6 | 12.5 |
| Mild (IQ 55-70) | 10 | 20.8 |
| Moderate (IQ 40-55) | 23 | 47.9 |
| Severe (IQ 25-40) | 7 | 14.6 |
| Profound (IQ <25) | 2 | 4.2 |
| *Residential status* | | |
| Independent | 0 | 0 |
| Group home | 14 | 29.2 |
| Central location | 34 | 70.8 |

*n = number of participants*

### Feasibility

All 12 participants answered all items of the interview part of the MNA. Seven of them considered at least 3 out of 15 questions difficult to answer, whereas the researcher judged the questions difficult for all 12 participants. Mean duration of the interview was 7 minutes (range 4 – 12 minutes).

During physical assessment, measurement of calf circumference and upper arm circumference was successful in all cases. For 9 out of 47 participants it was not possible to measure height because they were not able to stand in the correct position. For these participants knee height was measured successfully. For 5 out of 47 participants measuring weight was not possible and two participants refused the measurement. For these seven participants recent weight data from the medical files were used.

All caregivers answered all items of the interview part of the MNA. For all caregivers it was easy to answer at least 13 of the 15 items (85%). It was sometimes hard for them to correctly answer the questions about mealtimes of the participants because of the day-time activities of the participants at other places. Mean duration of the interview was 4 minutes (range 2 - 7 minutes).

In conclusion, the interview part of the MNA is insufficiently feasible for participants with borderline to mild ID and feasible for caregivers. The physical assessment is feasible for people with ID.

### Reliability

For test-retest reliability the mean time between T1 and T2 was 13.8 days (range 12-15 days). For two participants the answers were excluded from the calculation, because the clinical status of the participant had changed. Results of test-retest reliability and inter-observer reliability are shown in table 2.

Both test-retest reliability and inter-observer reliability for caregivers are excellent, but accordance between participants and caregivers is poor. The internal consistency is substantial with Chronbach's alpha 0.61.

**Table 2** *Results of test-retest ant inter-observer reliability*

| Reliability | ICC (95% CI) |
|---|---|
| Test-retest (*n* = 46) | 0.85 (0.72 – 0.92) |
| Inter-observer caregiver 1 – caregiver 2 (*n* = 48) | 0.86 (0.74 – 0.92) |
| Inter-observer caregiver 1 – participant (*n* = 12) | 0.03 (-0.51 – 0.59) |

*n* **=** *number of participants; 95% CI = 95% confidence intervals.*

## DISCUSSION

This study has shown that the MNA is feasible and reliable for older people with ID, if the interview is completed by caregivers. In that case, test-retest and inter-observer reliability are both excellent and comparable to the findings of Bleda et al. in institutionalized elderly people [17].

In this study accordance between caregivers and personally interviewed people with ID is poor (0.03). This outcome is in line with the study of Kaiser et al. [18]. In that study the MNA results of interviewed nursing home residents were compared with the MNA results when administered by nursing staff. Possible explanations of the poor inter-observer reliability between people with ID and their caregivers result may be (1) the comprehensibility of the interview items for participants and (2) the socially accepted answers by participants on items about lifestyle and consumption of food. For example 6 out of 12 participants answered positive on the item about weight loss

during the last three months, whereas only one caregiver confirmed this. In addition, the small group of participants may be the most probable reason. Therefore inter-observer reliability between people with ID and their caregivers needs to be further studied on a larger scale.

A group of people with ID living in small group homes or in a central location were assessed. A large group of independently living older people with borderline and mild ID is missing from the study. This group may be well capable to complete the interview with reliable answers [19].

The internal consistency in this study is substantial, but lower than findings in a previous study form Bleda et al. [17]. In both studies populations were not very large. This may be a possible reason for just a natural variability.

Although the MNA is feasible for older people with ID, if the interview is completed by caregivers, it was sometimes difficult to answer the questions about mealtimes of the participants because of day-time activities of the participants at other places. For a correct assessment of nutritional intake caregivers should try to obtain additional information from other caregivers or relatives of the participant.

Research on validity of the MNA in this population has to be studied right now, because the authors expect that some items (medication use, independently living) are more related to intellectual disabilities instead than to nutritional status.

The authors conclude that the MNA is feasible and reliable for older people with ID and recommend that the MNA should be applied by caregivers. The authors advise to implement the MNA as soon as possible into clinical practice for people with ID to identify malnutrition and to give professionals the opportunity to intervene at an early stage.

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# Chapter 6

## Evaluation of the Mini Nutritional Assessment in older people with intellectual disabilities

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*Submitted*

## ABSTRACT

The Mini Nutritional Assessment (MNA) is a screening tool for malnutrition that might be appropriate for people with intellectual disabilities (ID). Aim of this study was to investigate its construct and criterion validity in older adults with ID.

For this purpose the MNA was completed for 835 persons aged 50 years and over with borderline to profound ID. Construct validity was determined by correlating the individual items and the total score of the MNA with the four parameters of a consensus-based nutritional state index. To investigate criterion validity, tests of sensitivity and specificity were performed to compare the outcomes of the MNA with the consensus-based nutritional state index.

The construct validity was low, with Spearman's correlation coefficients varying from $r = 0.17$ to $r = 0.29$. Compared with the nutritional state index, sensitivity of the MNA was 46.4% and specificity was 60.8%.

In conclusion: The MNA seems to be insufficiently valid for use in people with ID in the current format. This is probably due to proxy bias and a poor content validity. Therefore, for older people with ID, modification of the MNA is needed.

## INTRODUCTION

Life expectancy in people with intellectual disabilities (ID) has increased in the last decades [1]. As a consequence, the number of older people with ID rises [2]. Just like the general population, older people with ID are dealing with age-related conditions. Malnutrition, defined as a nutrient deficiency state of energy, macronutrients (including proteins), or micronutrients, is one of these conditions and is related to several diseases, such as infections [3, 4] and depression [5], prolonged length of hospital stay [6], loss of quality of life [7], and mortality [8]. To investigate malnutrition, nutritionists use several measures [9]. Malnutrition, measured by dietary intake analysis, is prevalent in older people with ID, with a prevalence rate of 30% for protein energy malnutrition [10]. Malnutrition, measured by anthropometric measures, in people with ID may be less prevalent than overnourishment, but the prevalence of underweight is high compared to adults in the general population [11, 12]. Individuals with more profound disabilities experience higher rates of malnutrition [13] due to more feeding problems and swallowing problems.

Because of the high prevalence of malnutrition in people with ID and its associations with high morbidity and mortality, early detection and intervention is important. In clinical practice, several screening instruments are available, of which one is the Mini Nutritional Assessment (MNA), developed by Guigoz, Vellas and Garry [14]. It is internationally widely used and is considered as state of the art for ageing people in the community and in long-term care facilities [15]. Originally, the MNA was validated in more than 600 healthy and frail ageing subjects [14]. Afterwards it was validated for other subgroups [16-19].

The psychometric properties of the MNA have hardly been studied for people with ID [20, 21]. We have recently demonstrated that the MNA is feasible and reliable for older people with ID, with intraclass correlation coefficients of 0.85 for test-retest reliability and 0.86 for inter-observer reliability between two professional caregivers [20]. Tsai, Hsu and Chang reported low correlations between the MNA and various nutrition-related parameters [21].

It is important to specifically validate the MNA for people with ID, because the MNA involves two subjective items (about self-perception of health and nutritional status). Especially for people with a moderate to profound ID it is not possible to answer these items, meaning that these items need to be completed by proxy. Moreover, for people with ID, some of the items do not correlate with malnutrition in the same way as in the general elderly population. For example, 'living independently' is an item of the MNA. However, because of their cognitive disabilities, large numbers of people with ID do not live independently and therefore this item will probably not discriminate very well in this population. We therefore studied the construct and criterion validity of the

MNA by comparing MNA scores with other parameters for nutritional status, in older adults with ID.

## METHODS

### Study design and participants

This study was part of a large cross-sectional study, called "Healthy Ageing and Intellectual Disability (HA-ID)". In the HA-ID study, prevalences of diseases, risk factors and secondary health effects for the subthemes 'nutrition and nutritional state', 'physical activity and fitness' and 'depression and anxiety' were investigated. The recruitment and selection process of this study was outlined in detail by Hilgenkamp et al. [22]. The study population consisted of clients, aged 50 years and over, in a consortium of three Dutch care provider services in the Netherlands, offering various specialized support options, varying from ambulatory support or day care for clients living independently or with family, to residential settings providing more or less intensive care.

Informed consent was obtained from 1069 clients and/or their legal representatives and 1050 clients actually participated in the assessments. The study population is nearly representative for the total client population of the consortium, aged 50 years and over, with a slight overrepresentation of women, and a slight underrepresentation of individuals living independently and individuals aged 80 years and over [22]. This study has been approved by the Medical Ethics Committee of the Erasmus Medical Center Rotterdam, the Netherlands (MECnr. 2008-234) and by the ethics committees of the participating care provider services. The study was conducted in accordance with the guidelines from the Declaration of Helsinki for research involving human subjects.

All people that participated in the HA-ID study, in whom a final MNA score was obtained, were included in the present study.

### MNA

The full Mini Nutritional Assessment involves 6 general items (residential status, stress or acute disease, psychological problems, mobility, medication, and skin ulcers), 4 anthropometric items (Body Mass Index (BMI), arm and calf circumferences, and weight loss), 6 dietary items (appetite, number of meals, protein intake, fruit or vegetables intake, fluid intake, and mode of feeding) and 2 subjective items (self-perception of health and self-perception of nutritional state). It can be completed by health professionals in less than 15 minutes. Three items (BMI, arm circumference and calf circumference) are assessed by means of a physical assessment. The 15 other items are assessed by means of an interview with the patient or his/her caregiver. If data

can not be obtained through a physical assessment or an interview, it is allowed to use the medical record of the participant for most of the items. The total score (0-30 points) categorizes the results as follows: (1) well-nourished (24 points or more); (2) at risk for malnutrition (17-23.5 points); and (3) malnourished (< 17 points).

### Consensus-based nutritional state index

In line with previous validation studies [16, 18, 23] we developed an index, consisting of several nutritional parameters, to calculate the criterion validity of the MNA. The parameters of this nutritional state index have been carefully chosen after literature review and consultation of experts working in the field of nutrition or medical care for people with ID.

We conducted a literature search in the Medline database for scientific evidence of 20 variables measured in the HA-ID study, including anthropometric, biochemical and physical parameters, regarding relationships with nutritional status. When sufficient evidence was shown, this variable was presented to the experts. After review of the literature, six variables were left: serum hemoglobin, serum cholesterol, serum albumin, grip strength, BMI and fat percentage calculated from the sum of four skinfolds (biceps, triceps, subscapular, and suprailiacal). The experts advised to remove serum cholesterol because hypocholesterolemia seems to occur only late in the course of malnutrition, and therefore the value of cholesterol as part of a nutritional state index is limited. Because it was not possible to measure suprailiacal skinfolds for participants in a wheelchair and we did not want to exclude that high-risk group, fat percentage was eliminated as well. Thus finally four variables were left in the consensus-based nutritional state index for this validation-study: serum hemoglobin, serum albumin, grip strength, and BMI. Grip strength was defined in accordance with the criteria of the Cardiovascular Health Study, with cut-off points of 29 kilograms for men with a BMI ≤ 24, 30 kilograms for men with a BMI between 24.1 and 28, and 32 kilograms for men with a BMI > 28, and 17 for women with a BMI ≤ 23, 17.3 for women with a BMI between 23.1 and 26, 18 kilograms for women with a BMI between 26.1 and 29, and 21 kilograms for women with a BMI > 29 [24]. The cut-off points for BMI were 20 kg/m² for participants aged 65 years and over, and < 18.5 kg/m² for participants aged 50-64 years [25, 26]. A low level of serum hemoglobin was defined as ≤ 8.0 mmol/L for men and ≤ 7.4 mmol/L for women and a low level of serum albumin as < 35 g/L. Because serum albumin is also an acute phase protein, people with a C-reactive protein ≥ 10mg/l were excluded for this part of the study.

In a next step, we assigned weights to the variable scores: together with the experts, we agreed that BMI and serum albumin are more important aspects of malnourishment than grip strength and serum hemoglobin, because we suspected that the validity of our grip strength data may not be optimal, especially in people with a more severe

ID (because of non-cooperation or lack of understanding), and serum hemoglobin is a multifactorial condition [27]. Thus, in case of low BMI and serum albumin scores, the participant was assigned one point for each positive score and in case of low grip strength and/or low serum hemoglobin the participant was assigned one point. Thus, the minimum score of the nutritional state index was zero and the maximum score three points. Participants were considered malnourished with a score of two or three points, at risk of malnutrition with a score of one point, and well-nourished with a score of zero points.

## Data collection

Gender, age and residential status were collected from the records of the care providers. Residential status was divided into living in group homes in central settings, living in group homes in the community, independent living and living with relatives. Psychologists or behavioral therapists recorded level of ID (divided into borderline, mild, moderate, severe or profound ID). Professional caregivers, who were familiar with the participant for at least three months, were asked about the participants' mobility (divided into walking independently, walking with aid and wheelchair dependent). They also completed the interview items of the MNA. The anthropometric items of the MNA (weight and height [to calculate BMI], and calf and arm circumference) were measured by trained medical assistants measured according to the user guide of the MNA [28]. If participants were unable to stand upright, knee height was measured; equations developed by Chumlea, Roche and Steinbaugh [29] were used to calculate body height. For immobile participants, recent weight was retrieved from the medical records

For grip strength the participants squeezed a Jamar Hand Dynamometer (Sammons Preston Rolyan, USA) to their maximum ability in seated position, according to the recommendations of the American Society of Hand Therapists [30]. The best result of three attempts for both the left and the right hand (with a one-minute pause between attempts), was recorded. The test instructors squeezed a rubber ball as a visual example to explain the purpose of the grip strength test and they had to be convinced that the participant squeezed with maximal effort; otherwise they did not record a test result.

A fasting blood sample was taken from participants who separately consented to venipuncture. From the blood sample serum albumin and serum hemoglobin were assessed.

## Statistical analysis

Statistical analyses were performed with SPSS 20. A non-response analysis of the participants with a valid MNA score but without completed data for the criterion validity of

the MNA was conducted using a $\chi^2$-test for gender, age, level of ID, residential status, mobility and nutritional state according to the MNA (divided into well-nourished, at risk for malnutrition and malnourished).

To investigate construct validity, Spearman correlation analyses were performed to assess the relations between the individual items and the total score of the MNA on the one hand, and the parameters of the consensus-based nutritional state index on the other hand. To investigate criterion validity, tests of sensitivity and specificity were performed to compare the outcomes of the MNA with the consensus-based nutritional state index. Sensitivity was defined as the number of participants found to be malnourished or at risk for malnutrition according to both the MNA and the nutritional state index divided by the total number of participants identified as malnourished or at risk for malnutrition by the index. Specificity was defined as the number of participants found to be well-nourished according to both the MNA and the nutritional state index divided by the total number of participants classified as well-nourished by the index.

## RESULTS

### Participants

The MNA was completed for 835 participants. Thirty-one (3.7%) were categorized as malnourished, 375 (44.9%) as at risk for malnutrition and 429 (51.4%) as well-nourished. The mean age of this group was 61.6 years (SD 7.8 years, range 50 – 93

**Table 1** *Characteristics of participants (n = 835)*

| | *n* | % |
|---|---|---|
| *Gender* | | |
| Men | 428 | 51.3 |
| Women | 407 | 48.7 |
| *Level of ID* | | |
| Borderline (IQ 70-85) | 30 | 3.6 |
| Mild (IQ 55-70) | 184 | 22.0 |
| Moderate (IQ 40-55) | 402 | 48.1 |
| Severe (IQ 25-40) | 129 | 15.4 |
| Profound (IQ <25) | 69 | 8.3 |
| Unknown | 21 | 2.5 |
| *Level of mobility* | | |
| Walks independently | 648 | 77.6 |
| Walks with support | 121 | 14.5 |
| Wheelchair | 66 | 7.9 |

*n = number of participants.*

years). Sex, level of ID, and level of mobility of the participants are described in Table 1.

For 464/835 (55.6%) complete data of the consensus-based nutritional index were available. Seventy-four of them had a C-reactive protein of 10 mg/l or higher; these participants were excluded for this part of the validation study. This means that 390 participants were included to test criterion validity. This group had a similar gender and age distribution as the excluded group (n = 445), but included significantly more persons with borderline and mild ID ($\chi^2 = 70.8$, $p < 0.01$), and less persons using a wheelchair ($\chi^2 = 16.7$, $p < 0.01$). In the included group, malnutrition, according to the MNA, was significantly less prevalent (6 vs. 25; $\chi^2 = 17.0$, $p < 0.01$) compared to the excluded group. Thus the included group was a less disabled and healthier subgroup of the study population.

## Construct and criterion validity

Total MNA scores were significantly and positively correlated with BMI, grip strength, hemoglobin and albumin (Table 2), but correlations were all small. Five of the individual items of the MNA (BMI, mode of feeding, consideration of health status, mid-arm circumference and calf circumference) and the total score of the consensus-based nutritional state index were also significantly, but weakly associated (Table 3). The total score of the MNA and the total score of the nutritional state index were also significantly correlated, but this relationship was small as well (r = -0.14).

Nutritional state of the participants according to the MNA and the consensus-based nutritional state index is presented in Table 4. Sensitivity of the MNA to detect malnourished participants or participants at risk for malnourishment was 46.4% (95% CI: 40.0% – 53.0%) and specificity was 60.8% (95% CI: 53.3% - 67.9%).

**Table 2** *Correlation between individual items of the consensus-based nutritional state index and the total score of the MNA*

| | *n* | *r* |
|---|---|---|
| BMI | 835 | 0.21* |
| Albumin | 520 | 0.19* |
| Hemoglobin | 623 | 0.29* |
| Grip strength | 655 | 0.17* |

*n* **=** *number of participants; r = Spearman correlation coefficient.*
***** *p < 0.01*

**Table 3** *Correlation between individual items of the MNA and the total score of the consensus-based nutritional state index*

| | *n* | *r* |
|---|---|---|
| BMI | 390 | -0.13* |
| Mode of feeding | 390 | -0.17* |
| Consideration of health status | 390 | -0.17* |
| Mid-arm circumference | 390 | -0.14* |
| Calf circumference | 390 | -0.16* |

*n = number of participants; r = Spearman correlation coefficient.*
** p < 0.01*

**Table 4** *Nutritional state of the participants according to the MNA and the consensus-based nutritional state inde.*

| | | Consensus-based nutritional state index | | | Total |
|---|---|---|---|---|---|
| | | Well-nourished | At risk | Malnourished | |
| MNA | Well-nourished | 104 | 118 | 2 | 224 |
| | At risk | 65 | 91 | 4 | 160 |
| | Malnourished | 0 | 5 | 1 | 6 |
| | Total | 169 | 214 | 7 | 390 |

## DISCUSSION

This is the first study into validity of the Mini Nutritional Assessment in a large near-representative older population with intellectual disabilities. The results show that sensitivity and specificity are unsatisfactory and that construct validity is low [31].

Previous studies show higher sensitivity levels, although it depends on the setting. In municipal care (service buildings, retirement homes, and nursing homes) sensitivity was 96% [23], but in acute care hospitals a sensitivity of 57% was found [18]. The moderate sensitivity found by Azad et al. was explained by the scoring system of the MNA, giving less importance to weight loss, which is prevalent in hospitalized patients [18]. That result is in line with the conclusions made by Sieber [15] who argued that the MNA is state of the art for ageing people in the community and in long-term care facilities but not for people in the acute care setting.

The low levels of sensitivity and specificity in our study could be attributed to the population and to the composition of the consensus-based nutritional state index. The MNA was originally not developed for people with ID. Although most factors contributing to malnutrition in people with ID are also common in people without ID, there might be disorders that are highly prevalent in the ID population and related to malnutrition, such as gastroesophageal reflux disease [32, 33] and dysphagia [34] that are not adequately covered in the MNA. Furthermore, the MNA consists of 18

items related to malnutrition in the elderly, but in people with ID some of these items, specifically living circumstances and mode of feeding, could be more related to ID than to malnutrition. This might lead to more subjects with a diagnosis of malnutrition. In fact, the content validity of the MNA seems to be poor for people with ID because of the above mentioned reasons and the two subjective items in the MNA that are very hard to answer by proxy or by people with ID themselves.

With regard to the consensus-based nutritional state index, grip strength measurements may have been hampered by participants' lack of motivation or understanding, which is frequently observed in people with severe ID. Moreover, there is no international consensus on the cut-off value for underweight according to the BMI in people aged 65 years and over. Applied values differ from 18.5 to 25 kg/m² [35-37]. In our study the cut-off value was 20 kg/m². If we would have chosen a higher cut-off value for underweight in this study, then more participants would have been malnourished according to the nutritional state index. However, the effect on sensitivity and specificity would probably be small, because the nutritional state index consists of multiple variables. Thirdly, low hemoglobin and to a lesser extent low albumin may occur in malnutrition, but can be caused by other disorders or diseases as well. In other words, both measurements lack specificity.

In order to counter the lack of specificity in the instruments, we chose a combination of indicators from several categories (anthropometry, biochemical parameters and physical function), as was recommended before [38]. Additional strong points in our study are the stepwise process of defining our index (a literature review followed by consultation of experts), the thorough evaluation of the MNA by testing different forms of validity, and the adequate sample sizes.

A limitation of our study may be 'proxy effect'. The MNA has originally been developed for completion by patients themselves. In our study population of people with ID, self-report was not possible or reliable [20], so professional caregivers of the participants were interviewed. Most of the items of the MNA are objective, and for those items answers from proxy caregivers are reliable. However, the subjective items of the MNA are more difficult to answer for these persons. Another limitation is the use of a consensus-based nutritional state index to investigate criterion validity. It would have been better to use an extended nutrition assessment done by clinical experts as a gold standard. But in a large study with 835 participants with ID, such expert assessments are not feasible because of the investment of time and lack of experienced physicians trained in both nutrition and medical care for people with ID.

In conclusion: the MNA is insufficiently valid for use in people with ID in the current format. This information is crucial for all practitioners who consider the use of the MNA to detect malnutrition in people with ID. Modification of the MNA is needed. ID-specific disorders related to malnutrition and level of ID could be added and some items could

be better adapted to people with ID. After this modification, a new validation study with an extended nutritional assessment done by clinical experts as a gold standard is necessary as well. Responsiveness of the new instrument should also be studied in order to test the ability of the MNA to detect true change.

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# Chapter 7

## Observed vitamin D deficiency variations in older adults with intellectual disabilities

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*Submitted*

## ABSTRACT

Because life expectancy of people with intellectual disabilities (ID) is increasing gradually and sufficient serum levels of vitamin D are important for multiple health issues, we were interested in the prevalence of vitamin D deficiency and its associations in this population.

In this cross-sectional study, serum was obtained from 618 persons with borderline to profound ID aged 50 years and over. The prevalence rates of vitamin D deficiency and severe deficiency (serum $25(OH)D_3$ level below 50 nmol/l and 25 nmol respectively) were calculated by using descriptive statistics. To identify its associations, logistic regression analyses were performed with patient characteristics, mobility, mean number of steps per day, number of falls, grip strength, body mass index (BMI), bone quality, depressive symptoms, major depression, dementia, anticonvulsant drug use, serum calcium levels and suppletion of vitamin D.

The prevalence of vitamin D deficiency was 42% and that of severe deficiency was 9%. Vitamin D suppletion was being routinely provided to 45% of the population. This group had significantly higher mean vitamin D serum levels than those not using suppletion. Apart from suppletion, vitamin D deficiency was significantly associated with lower serum calcium levels and higher BMI-scores. We recommend general practitioners and specialized physicians for people with ID to be aware of the necessity of routine vitamin D substitution in all adults with ID aged 50 years and over. The association of BMI with vitamin D deficiency stresses even more the relevance of healthy food intake and physical activity in this population.

## INTRODUCTION

Life expectancy of people with intellectual disabilities (ID) is increasing gradually, including groups with neurological comorbidities such as motor impairments and epilepsy [1]. Because both old age and anticonvulsant drug use, which is very prevalent in people with ID, are negatively associated with vitamin D serum levels, and by consequence bone quality, we were interested whether there is an increased risk of vitamin D deficiency in this population.

Maintaining recommended vitamin D levels is important for multiple health issues. Apart from the significant role of vitamin D in bone metabolism [2], it is also associated with obesity, blood pressure, orthostatic hypotension, cardiovascular disease and cerebrovascular disease [3-11]. Its association with muscle weakness and balance [12, 13] together with orthostatic hypotension [4], might explain the link of vitamin D deficiency with falls [14-16]. Other possible associations are autoimmune disease [17], depression [18, 19], dementia [20, 21] and cancer risk (prostate, colon, breast) [22].

Various factors make older people more susceptible to vitamin D deficiency: diminished production in the skin [23], less sunlight exposure [24], decreased dietary intake, impaired intestinal absorption, and impaired hydroxylation in the liver and kidneys [25]. Indeed, high prevalence rates of vitamin D deficiency were identified in healthy Spanish postmenopausal women (70% < 50 nmol/l ) [26], elderly community-dwelling people in 11 European countries (36% of males and 47% of females < 30 nmol/l) [27], and Dutch nursing home residents (98% < 50 nmol/l; 77% < 30 nmol/l) [28].

The risk may even be higher in older people with ID, because anticonvulsant use may negatively influence vitamin D levels as well, due to increasing vitamin D catabolism by inducing liver enzymes [11, 29]. So far, published studies of vitamin D status in people with ID are limited to selected adult subgroups with a high risk of epilepsy. In two file studies of 280 Australian and 138 Finnish institutionalised adults with ID, vitamin D serum levels lower than 50 nmol/l were found in 57% [30] and 78% [31] of the sample. In a small observational study, 20 out of 23 Dutch adults with ID and severe generalised cerebral palsy had serum vitamin D levels below 25 nmol/l [32]. In these publications, data obtained in older participants were not separately reported.

Because of the complete lack of valid epidemiologic information on vitamin D status in older persons with ID, we studied the prevalence and associations of vitamin D deficiency in a large Dutch older population with intellectual disabilities.

## METHODS

### Study design

This cross-sectional study was part of the 'Healthy Ageing and Intellectual Disability' (HA-ID) study, a large epidemiological study conducted in a consortium of three large intellectual disability care provider services in the Netherlands. Detailed information on design, population and diagnostic assessments has been published separately [33]. Ethical approval was provided by the Medical Ethics Committee of the Erasmus University Medical Center (MEC 2008-234) and by local ethics committees of the participating ID care services. Informed consent was obtained from all participants and/or their legal representatives.

### Study population

We included 1050 clients aged 50 years and over. The study population was nearly representative for the total older Dutch client population receiving formal ID care, with a slight overrepresentation of females, and a slight underrepresentation of individuals living independently and individuals aged 80 years and over [33]. Consent for drawing blood samples was separately asked, and obtained from 618 participants.

### Laboratory measurements

Serum samples were drawn in Becton Dickinson collection tubes, centrifuged and stored at -80°C. Albumin and calcium levels were determined on a Beckman Coulter DxC analyser. Corrected calcium serum levels were calculated with the following formula: corrected calcium (mmol/l) = calcium serum level + ((40 - albumin serum level in g/l ) x 0.02). Low serum calcium level was defined as a level below 2.10 mmol/L.

Vitamin D3 (25-OH vitamin D) measurements were performed using an electrochemiluminescence immunoassay (ECLIA) with polyclonal antibodies from Roche on a Cobas E module (Roche Diagnostics®, Penzberg, Germany). The between-run coefficient of variation was 4.6% at 53 nmol/L and 9.9% at 28 nmol/L.

Vitamin D deficiency was defined as $25(OH)D_3$ level < 50 nmol/l, and severe deficiency as $25(OH)D_3$ level below 25 nmol/l [11].

### Associations

Associations of vitamin D level with the following general participant characteristics were assessed: gender, age, residential status, severity of ID, and Down syndrome. Gender, age and residential status were collected from the records of the care provider services. Residential status was classified as 'centralized setting' (primarily care), 'community-based group home' (primarily support), 'ambulatory support' (living in-

dependently or with family, participating in daycare or getting outreaching support). Psychologists or behavioural therapists recorded level of ID; severity of ID had been classified as borderline (IQ 70-80), mild (IQ 55-70), moderate (IQ 35-55), severe (IQ 25-35) or profound (IQ<25). General practitioners and specialized physicians for people with ID recorded aetiology of ID (Down syndrome yes /no).

Associations with the following variables that may influence vitamin D levels were studied: vitamin D supplementation, anticonvulsant drug use, body mass index (BMI), mobility, and physical activity. Information on vitamin D supplementation and anti-convulsant drug use was retrieved from medical files at baseline. BMI was calculated as weight (kg) divided by squared height (m). Because sunlight exposure had not been addressed, mobility and physical activity were used as substitute variables. Mobility was classified as 'independent mobility', 'mobility with aids', and 'wheelchair dependent'. Physical activity was measured as mean number of steps per day during at least 4 days with the NL-1000 pedometer (New Lifestyles, Missouri, USA). Dietary intake, another factor that may influence vitamin D levels, has only been analysed in a subsample of the HA-ID population and could therefore not be included in this study.

The following associations with variables that may be influenced by low vitamin D serum levels were studied: bone quality, serum corrected calcium level, muscle strength and by consequence falls, depressive symptoms, major depression and dementia.

Bone quality was assessed by ultrasound measurement of the heel bone (Lunar Achilles type Insight, GE Healthcare, Clinical systems Ultrasound, Hoevelaken, The Netherlands). Grip strength, as a valid measure of muscle strength, was measured with a Jamar Hand Dynamometer (Sammons Preston Rolyan, USA). The best result of three attempts for both hands was recorded. Information on falls during the past 3 months was obtained from the caring staff. Symptoms of depression were measured with the Inventory of Depressive Symptomatology Self Report (IDS-SR) in participants capable of self-report, and in the other participants with the Dutch informant-report Signalizing Depression List for people with Intellectual Disabilities (SDL-ID). A diagnosis of major depression was made with the Psychiatric Assessment Schedule for Adults with a Developmental Disability (PAS-ADD), a semi-structured diagnostic interview that has been validated against expert psychiatric diagnosis. Dementia was defined as 'possible or probable dementia'. Diagnoses were obtained from the participants' own physicians and behavioural specialists, and were only included in the analysis in case of consensus between these professionals.

## Statistical analysis

Statistical analysis was performed using Statistical Package for Social Sciences for Windows version 20 (SPSS Inc. , Chicago, Il, USA).

Differences between participant characteristics in the current study population (N=618) and the total HA-ID study population (N=1050) were checked with Chi-square tests. The same procedure was followed to analyse differences between participants using vitamin D suppletion and participants without vitamin D suppletion.

Prevalence rates with 95% confidence intervals were calculated for vitamin D deficiency and severe deficiency as well as for low corrected serum calcium levels by using descriptive statistics. The correlation of vitamin D and corrected calcium serum level was assessed with Pearson correlation coefficient.

Chi square tests for categorical variables and point biserial correlations for continuous variables were performed, to explore associations between vitamin D deficiency (no = 0, yes = 1) as dependent variable and gender (men = 0, women = 1), age, severity of ID (borderline/mild = 1, moderate = 2, severe/profound = 3), residential status (centralized setting = 1, community setting = 2, independent or with relatives = 3) , Down syndrome (no = 0, yes = 1), mobility (independent = 1, walking with aids = 2, wheelchair dependent = 3), mean number of steps, number of falls (no falls = 1, one or two falls = 2, three or more falls = 3), grip strength, BMI, bone quality, depressive symptoms (no = 0, yes = 1), major depression (no = 0, yes = 1), dementia (no = 0, yes = 1), use of anticonvulsant drugs (no = 0, yes = 1), serum calcium levels, and vitamin D suppletion (no = 0, yes = 1), as independent variables. In order to determine the independent contribution of these factors to the variance of vitamin D deficiency, a multiple logistic regression analysis was performed with determinants that were found to be at least nearly significantly ($p<0.10$) associated with vitamin D deficiency in the univariate analyses. Multicollinearity was checked for all independent variables with the variance inflation factor (VIF) of linear regression analysis. VIF values above 10 indicated multicollinearity [34].

## RESULTS

### Study population

Participant characteristics of the group that consented to blood sampling (N=618) are presented in Table 1. Mean age was 61.7 years (range 50-92). The last column shows that the study population and the total HA-ID population differed significantly with respect to gender and residential status. For gender, this did not influence further outcomes of the study, because the prevalence of vitamin D deficiency was not significantly different between males and females ($X^2 = 0.35$, $p = 0.55$). But, this does not apply to residential status, because of differences in prevalence of vitamin D deficiency between subgroups.

**Table 1** *Participant characteristics (n = 618) and comparison with the total HA-ID population*

| | | *n* | % | X² |
|---|---|---|---|---|
| Gender | Male | 334 | 54 | 4.16* |
| | Female | 284 | 46 | |
| Age | 50-59 | 283 | 46 | 0.82 |
| | 60-69 | 222 | 36 | |
| | 70-79 | 98 | 16 | |
| | 80 and over | 15 | 2 | |
| Residential status | Centralized setting | 356 | 58 | 14.41** |
| | Community setting | 237 | 38 | |
| | Independent | 25 | 4 | |
| Severity of ID | Mild | 140 | 23 | 2.89 |
| | Moderate | 308 | 50 | |
| | Severe | 162 | 26 | |
| | Unknown | 8 | 1 | |
| Down Syndrome | Yes | 86 | 14 | 0.55 |
| | No | 473 | 77 | |
| | Unknown | 59 | 10 | |

*ID = intellectual disability*
***** = $p < 0.05$; ** = $p < 0.01$*

**Table 2** *Comparison of patient characteristics of participants with and without suppletion*

| | | No Suppletion | Suppletion | X² |
|---|---|---|---|---|
| Gender | Male | 204 | 130 | 10.83** |
| | Female | 135 | 149 | |
| Age | 50-59 | 129 | 154 | 23.34** |
| | 60-69 | 149 | 73 | |
| | 70-79 | 53 | 45 | |
| | 80 and over | 8 | 7 | |
| Residential status | Central setting | 109 | 247 | 200.85** |
| | Community setting | 205 | 32 | |
| | Independent | 25 | 0 | |
| Severity of ID | Mild | 109 | 31 | 41.35** |
| | Moderate | 158 | 150 | |
| | Severe | 69 | 93 | |
| Down Syndrome | No | 261 | 212 | 17.14** |
| | Yes | 26 | 60 | |

*ID = intellectual disability*
***** = $p < 0.05$; ** = $p < 0.01$*

Two hundred and seventy-nine participants (45%) were reported to receive routine vitamin D supplementation; 151 used 400 IU/day and 128 used 800 IU/day. Eleven participants who used a multivitamin preparation with a vitamin D dose lower than 400 IU/day, were added to the non-suppletion group in the analysis. Comparing the groups of 'using suppletion' with 'no suppletion', Table 2 shows that there were relatively more women, more people who live in a centralized setting, more people with Down syndrome, and more people with a more severe level of ID in the group with vitamin D suppletion.

## Vitamin D status

Vitamin D deficiency (< 50 nmol/l) was identified in 257/618 participants (42%; 95% confidence interval (CI) 36% - 48%), and 55 had a severe deficiency (< 25 nmol/l) (9%; 95% CI 7% -11%). Low corrected serum calcium levels (< 2.10 mmol/l) were found in 36/585 participants (6%; 95% CI 5% - 8%). Vitamin D serum level was positively correlated with the corrected serum calcium level ($r = 0.24$; $p < 0.001$). Mean vitamin D serum level in the 36 participants with low corrected calcium levels was 49.3 nmol/l (range 15-132, SD 30.6).

Participants receiving vitamin D supplementation had significantly higher mean vitamin D serum levels (85.4 nmol/l in case of dose 400 IU/day and 85.8 nmol/l in case of dose 800 IU/day, $p = 0.99$) than participants who did not take supplementation (45.0 nmol/l; $p < 0.001$).

## Associations

Because of missing values, univariate analyses with vitamin D serum levels and the independent variables were performed with different numbers. Especially physical activity and the PAS-ADD interview to diagnose major depression could not be established in the majority of participants (Table 3). For physical activity it was largely due to unreliable pedometer results because of a walking speed lower than 3.2 km/h and limited understanding or non-cooperation [35]. For the diagnosis of major depression it was due to our study protocol, in which only the participants with a score above the cut-off of the IDS-SR or the SDL-ID were further examined with the PAS-ADD interview [36].

Table 3 shows that age, severity of ID, residential setting, mobility, mean number of steps, BMI, bone quality, anticonvulsant drug use, serum calcium level, and vitamin D suppletion were significantly associated with vitamin D serum levels. Because of the small amount of participants in physical activity, this variable was excluded from the multivariate analysis.

Table 4 shows that serum calcium level and vitamin D suppletion were negatively associated with vitamin D deficiency with an odds ratio (OR) of 0.96 and 0.06 respectively, which means 1.04 and 16.7 times less chance of vitamin D deficiency respectively. BMI was positively associated with vitamin D deficiency (OR = 1.09), so

people with a higher BMI were more likely to have vitamin D deficiency than those with a lower BMI.

The explained variance of the model (Nagelkerke $R^2$) was 0.45. VIF values were well below the threshold for multicollinearity.

**Table 3** *Univariate associations of vitamin D deficiency with independent variables*

| | | *n* | $X^2$ | *r* | *p*-value |
|---|---|---|---|---|---|
| Gender | Male | 334 | 0.35 | | 0.55 |
| | Female | 284 | | | |
| Age | | 618 | | 0.08 | 0.05 |
| Severity of ID | Mild | 140 | 23.32 | | < 0.01 |
| | Moderate | 308 | | | |
| | Severe | 162 | | | |
| Residential setting | Central | 356 | 72.56 | | < 0.01 |
| | Community | 237 | | | |
| | Independent | 25 | | | |
| Down syndrome | No | 473 | 0.83 | | 0.36 |
| | Yes | 86 | | | |
| Mobility | Independent | 432 | 8.06 | | 0.02 |
| | Walking aid | 89 | | | |
| | Wheelchair | 71 | | | |
| Number of steps | | 152 | | -0.19 | 0.02 |
| Number of falls | No falls | 444 | 2.71 | | 0.26 |
| | 1-2 | 112 | | | |
| | ≥3 | 37 | | | |
| Grip strength | | 441 | | 0.04 | 0.45 |
| BMI | | 552 | | 0.10 | 0.02 |
| Bone quality | | 502 | | 0.09 | 0.04 |
| Depressive symptoms | No | 484 | 0.54 | | 0.46 |
| | Yes | 105 | | | |
| Major depression | No | 136 | 1.64 | | 0.20 |
| | Yes | 23 | | | |
| Dementia | No | 483 | 1.22 | | 0.27 |
| | Yes | 56 | | | |
| Anticonvulsant drugs | No | 491 | 18.53 | | < 0.01 |
| | Yes | 127 | | | |
| Serum calcium level | | 585 | | -0.23 | < 0.01 |
| Vitamin D suppletion | No | 339 | 210.79 | | < 0.01 |
| | Yes | 279 | | | |

*n* **=** *number of participants; r = Pearson correlation coefficient; ID = intellectual disability; BMI = body mass index.*

**Table 4** *Independent associations of vitamin D deficiency*

| | *B* | Wald | OR | 95% CI OR | |
|---|---|---|---|---|---|
| | | | | *LL* | *UL* |
| Age | 0.02 | 1.83 | 1.02 | 0.99 | 1.06 |
| Severity of ID | 0.11 | 0.33 | 1.12 | 0.77 | 1.62 |
| Residential setting | 0.04 | 0.03 | 1.04 | 0.64 | 1.70 |
| Mobility | 0.17 | 0.43 | 1.19 | 0.71 | 1.99 |
| BMI* | 0.09 | 9.56 | 1.09 | 1.03 | 1.15 |
| Bone quality | -0.15 | 2.69 | 0.86 | 0.72 | 1.03 |
| Anticonvulsant drug use | -0.19 | 0.24 | 0.83 | 0.39 | 1.76 |
| Serum calcium level* | -0.04 | 7.91 | 0.96 | 0.93 | 0.99 |
| Vitamin D suppletion* | -2.88 | 72.86 | 0.06 | 0.03 | 0.11 |

*OR = odds ratio; 95% CI = 95% confidence interval; LL = lower limit; UL = upper limit; ID = intellectual disability; BMI = body mass index.*
*** =** $p < 0.05$

## DISCUSSION

This is the first large-scale epidemiological study of vitamin D status in 618 clients aged 50 years and over of Dutch formal intellectual disability services. Vitamin D suppletion was being routinely provided to 45% of the population. This group had significantly higher mean vitamin D serum levels than those not using suppletion; doses of 400 IU/day and 800 IU/day were equally effective. The prevalence of vitamin D deficiency was 42% and that of severe deficiency was 9%. Different from what we expected, these prevalence rates were lower than published prevalence rates in general older populations [26-28]. This may be explained by differences in suppletion [26] and age [27, 28]. Nevertheless, a prevalence of 42%, in spite of routine suppletion regimens in part of the settings, is unnecessarily high.

The found multivariate correlations with BMI, serum calcium level, and vitamin D suppletion were to be expected. Indeed, obesity reduces serum vitamin D levels, probably due to decreased bioavailability of vitamin D from cutaneous and dietary sources because of its deposition in body fat compartments [37, 38]. To our surprise, anticonvulsant drug use was not correlated with vitamin D deficiency in the multivariate regression analysis, in spite of the fact that in none of the settings, anticonvulsants were specifically considered an indication for routine suppletion. In the univariate analysis bone quality is positively associated, whereas in the multivariate regression analysis it is negatively associated with vitamin D deficiency. This is due to the confounding effect of suppletion, which is given in a subpopulation consisting of relatively more women, more people who live in a centralized setting, more people with Down syndrome, and more people with a more severe level of ID.

The strengths of this study are the large study population and the comprehensive set of predictors. Therefore, results of this study may be extrapolated to the entire older ID population receiving specialized care, although the underrepresentation of people living in community settings or living independently (who had a higher risk of vitamin D deficiency) should be taken into account.

A limitation of the present study is that we chose physical activity and mobility as substitutes for sun exposure. We are aware that this is debatable because the unknown correlation between physical activity / mobility and being outdoors. Nevertheless, physical activity was correlated with vitamin D deficiency in this study.

We conclude that general practitioners should be aware of the necessity of routine vitamin D substitution in all adults with ID aged 50 years and over. Prior to the current analysis, we have identified increased risks of obesity, a sedentary lifestyle, and low bone quality in the HA-ID study population, as compared to the general Dutch population [35, 39, 40]. The univariate or multivariate associations of these conditions with vitamin D deficiency, as found in the current study, stresses even more the relevance of healthy food intake and physical activity in this population, that is liable to early geriatric frailty [41, 42].

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# Chapter 8

## Bone quality in older adults with intellectual disabilities

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## ABSTRACT

Osteoporosis is a progressive bone disease leading to an increase in fracture risk. It has hardly been studied on a large scale in older people with intellectual disabilities (ID). In this study 768 persons with ID aged 50 years and over, were measured with quantitative ultrasound to determine the prevalence of low bone quality in this group. Also the associations of low bone quality with patient characteristics, mobility, physical activity, BMI, prior fractures, anticonvulsant drug use, intake of calcium and vitamin D3 levels were studied. The prevalence of low bone quality was 43.9%. Low bone quality was positively associated with female gender, age, more severe level of ID, mobility impairment and anticonvulsant drug use and negatively with BMI. In clinical practice, people with ID who are at risk for low bone quality should periodically be screened for osteoporosis and physicians should give them advice about nutritional supplements and lifestyle.

## INTRODUCTION

Osteoporosis is a disease characterised by low bone mineral density (BMD), leading to an increase in fracture risk and as a consequence an increased risk of pain, deformity, loss of mobility and independence. In the general population, osteoporosis is particularly common in older people. In the Netherlands the prevalence of osteoporosis increases from 1.1% among men aged 45 to 64 years to 8.6% among men aged 75 years and older and from 8.5% among women aged 45 to 64 years to 42.3% among women aged 75 years and older [1]. In people with intellectual disabilities (ID) osteoporosis is highly prevalent as well. Prevalence rates vary up to 78.5% depending on the population of interest [2-9]. Regarding people with ID of higher age, it can be assumed that the prevalence of osteoporosis is even higher than in the general older population, because of a combination of age-related and ID-related risk factors for this disease. Remarkably, osteoporosis has hardly been studied on a large scale in this specific population. Therefore, in this study the prevalence of osteoporosis and its associated factors in older people with ID were investigated.

In the general population a low vitamin D serum level, chronic malnutrition and physical inactivity are associated with a low BMD, apart from known risk factors in the ID-population [10]. The risk factors for the latter group have been studied in different study populations. In adults with ID living in a residential facility, female sex, immobility, use of anticonvulsant drugs and a more severe level of ID were found to be risk factors for low BMD [11]. Leslie et al. confirmed immobility as an associated factor for osteoporosis in institutionalized adults with ID, together with a low body mass index (BMI) and prior fractures [6]. In a study among 94 young adults with ID living in the community, Down syndrome (DS) was associated with low BMD [2]. However, information about risk factors in the heterogeneous group of older people with ID living inside or outside the institutions is currently lacking.

In clinical practice, osteoporosis is diagnosed using dual energy X-ray absorptiometry (DXA) [12]. However, in large-scale research, this technique may not be regarded as feasible for people with ID, because DXA-scan equipment is often located in hospitals and people have to lie completely still to prevent artifacts. Therefore, quantitative ultrasound (QUS) measurement of the heel bone has been used in previous studies among people with ID to identify people with a high fracture risk [11, 13]. Advantages of QUS are its lack of radiation and its portability. On the other hand, it is not possible to directly measure BMD with the QUS device. Instead, speed of sound (SOS) and broadband ultrasound attenuation (BUA) are measured, which both provide data on other indicators of bone quality. Both variables are found to be predictive of fracture risk [14]. Some QUS devices provide additional variables like a stiffness index, which is a parameter derived from a linear combination of BUA and SOS. The QUS is feasible

for people with ID. In a study by Mergler et al. QUS measurements were successfully performed in 94.7% and induced barely or no stress in 90.4% of people with ID living in residential care [15].

To obtain insight into bone quality in older adults with ID, we set up the current study. Aims of this study were (1) to determine the prevalence of low bone quality in older adults with ID, and (2) to identify the association of low bone quality with gender, age, level of ID, residential status, Down syndrome, mobility, physical activity, BMI, prior fractures, anticonvulsant drug use, intake of calcium and vitamin D3 serum levels.

## METHODS

### Study design and participants

This study was part of the large cross-sectional study "Healthy Ageing in people with Intellectual Disability" (HA-ID). Details about design, recruitment and diagnostic methods have been presented elsewhere [16]. The study population consisted of adults aged 50 years and over, who receive support or care from three Dutch care provider services (Abrona, Amarant and Ipse de Bruggen). Informed consent was obtained from 1069 older adults with ID or their legal representatives and 1050 persons actually participated in the assessments. For collection of blood a separate consent procedure was followed. The study population was nearly representative for the total Dutch client population aged 50 years and over receiving formal ID care, with a slight overrepresentation of women, and a slight underrepresentation of individuals living independently and individuals aged 80 years and over [16]. Ethical approval was provided by the Medical Ethical Committee of the Erasmus MC, University Medical Centre Rotterdam (MEC 2008-234) and by the ethical committees of the participating care provider services. The study adheres to the Declaration of Helsinki for research involving human subjects.

### Bone quality

Bone quality of the heel bone was measured using the Lunar Achilles (type Insight, GE Medical Systems Europe, Diegem, Belgium), a portable device that measures ultrasound variables to provide a clinical measure called Stiffness Index. The Stiffness Index indicates risk of osteoporotic fractures comparable to bone mineral density as measured by DXA. Stiffness Index results expressed as T-scores are used to assist physicians in the diagnosis of osteoporosis in the same way as are T-scores obtained by DXA [17]. The Lunar Achilles device uses built-in reference values based on age and gender, obtained from healthy German adults, to calculate the T-scores, expressed in standard deviation (*SD*) units, of the stiffness index. These T-scores result from the

comparison of the participants' bone status with the average peak value in healthy young people. In accordance with the guideline for prevention of fractures of the Dutch college of general practitioners [18], T-scores obtained by DXA equal to or lower than – 2.5 *SD* indicate treatment with bisfosfonates. In this study we used the same cut-off score for T-scores obtained by the Lunar Achilles.

The ultrasound measurement was part of a broad physical assessment, carried out by specially trained medical assistants. To minimize the burden for participants, people in a wheelchair were measured in their own wheelchair. Sometimes this was not possible due to the wheelchair designs. Measurements started with positioning the participant in either a chair or their wheelchair in front of the QUS device with bare feet. After thoroughly spraying the ankle with alcohol, one foot was placed in the device. Then the two membranes of the Lunar Achilles on either side of the ankle automatically filled with lukewarm water, enabling the transducers on both sides of the ankle to transmit and receive the ultrasound signal. If possible, bone status of both feet was determined. The lowest T-score was used for analysis. When the ultrasound signal does not reach the receiving transducer, for example in case of insufficient spraying with alcohol or in case of insufficient water levels in the membranes, the lunar device displays the result "out of range". This can also occur when bone mass is either extremely high, and the signal cannot pass through the bone, or when bone mass is extremely low and therefore not measurable. After obtaining an out of range result, the measurement procedure was repeated after checking the water level of the membranes and thoroughly spraying with alcohol a second time. The measurement was abandoned after a second failure.

## Associated factors

Information on age, gender and residential status was collected through the care provider services. Residential status was categorised as centralized setting, community-based setting and living independently with ambulatory support. Level of ID was obtained from behavioural therapists' and psychologists' records (mild, moderate or severe). Information on aetiology of ID (Down syndrome yes or no), use of anti-convulsant drugs and fractures in the past five years were retrieved from medical records. Professional caregivers were asked about mobility, categorized as walking independently, walking with aids and mobility in wheelchair.

Physical activity was assessed with pedometers (NL-1000, New Lifestyles, Missouri, USA) worn for at least four days [19]. BMI was calculated by weight divided by squared height [20]. Body height was measured using a Seca stadiometer, type 214, with the participant standing, wearing no shoes. For people who were unable to stand up straight, knee height was measured using a non-stretchable flexible tape. Subsequently, body height was estimated by using equations developed by Chumlea et al. [21]. Weight was

measured using a digital floor scale (Seca robusta type 813), with participants wearing light clothes and no shoes. For people who were unable to leave their wheelchairs, the most recent weight according to their medical record was used to calculate the BMI. Intake of calcium was assessed by food frequency lists. During three days, including one day in the weekend, food and fluid of the participants was recorded by their professional caregivers. Dieticians processed the intake data using a web program based on nutritional values of Dutch food products [22]. The software uses the information on the food and fluid products to estimate intake data of energy, macronutrients and micronutrients. Blood was collected from participants to measure serum levels of vitamin D3 by using an electrochemiluminescence immunoassay (ECLIA) from Roche on a Cobas E module (Roche Diagnostics, Penzberg, Germany). The between-run coefficient of variation was 4.6% at 53 nmol/L and 9.9% at 28 nmol/L.

## Statistical analysis

Statistical analyses were performed using Statistical Package for Social Sciences for Windows version 20 (SPSS Inc., Chicago, Il, USA).

Participants in the current study sample were compared with non-participants within the total HA-ID study population on baseline characteristics (gender, age, level of ID, residential status and Down syndrome) using Pearson's chi-square tests.

Prevalence rates of low bone quality (T-score ≤ -2.5) with 95% confidence intervals for the total study population and for subgroups according to gender, age and Down syndrome were calculated by using descriptive statistics. Kruskal-Wallis tests were used to calculate differences between mean T-scores in subgroups according to level of ID and residential setting.

To explore associations between low bone quality (dependent variables) and gender (men = 0, women = 1), age, level of ID, residential status, Down syndrome (no = 0, yes = 1), mobility, BMI, fractures in the past five years (no = 0, yes = 1), anticonvulsant drug use (no = 0, yes = 1), vitamin D3 serum levels and physical activity (independent variables), univariate regression analyses were performed. For the following independent variables two dummy variables were used: moderate and mild ID (1) compared to mild ID (0) for level of ID, community-based and centralized setting (1) compared to independently living (0) for residential status, and walking with support and wheelchair-dependence (1) compared to people who are able to walk independently (0) for mobility. If an independent variable showed a significant association ($p < 0.05$) with the dependent variable, it was subsequently entered into a multivariate logistic regression analysis. Multicollinearity, the undesirable linear association between independent variables, was checked for all independent variables with the variance inflation factor (VIF) of linear regression analysis. VIF values above 10 indicated multicollinearity [23].

## RESULTS

### Participants

For 768 out of 1050 participants in the total sample, a valid result for bone quality was obtained. Reasons for missing results were physical disability ($n$ = 63; 22%), too anxious ($n$ = 37; 13%), "out of range" results ($n$ = 37; 13%), logistical problems ($n$ = 25; 9%), errors in data input ($n$ = 4; 1%), and unknown ($n$ = 116; 41%). Characteristics of participants and non-participants are presented in Table 1. The excluded group ($n$ = 282) had a similar gender, age and Down syndrome distribution as the included group, but in the participants-group more persons with mild and moderate ID ($X^2$ = 25.71, $p$ < 0.01), and more persons living in the community or living independently ($X^2$ = 28.76, $p$ < 0.01) were included.

**Table 1** *Characteristics of participants (n = 768) and non-participants (n = 282)*

| | Participants | Non-Participants | |
|---|---|---|---|
| | *n* (%) | *n* (%) | $X^2$ |
| *Gender* | | | 0.21 |
| Men | 398 (51.8) | 141 (50.0) | |
| Women | 370 (48.2) | 141 (50.0) | |
| *Age* | | | 3.19 |
| 50-59 years | 372 (48.4) | 121 (42.9) | |
| 60-69 years | 267 (34.8) | 103 (36.5) | |
| 70-79 years | 112 (14.6) | 50 (17.7) | |
| 80 years and over | 17 (2.2) | 8 (2.8) | |
| *Level of ID* | | | 25.71** |
| Mild (IQ > 55) | 207 (27.0) | 47 (16.7) | |
| Moderate (IQ 35-55) | 380 (49.5) | 126 (44.7) | |
| Severe (IQ < 35) | 164 (21.4) | 99 (35.1) | |
| Unknown | 17 (2.2) | 10 (3.5) | |
| *Residential status* | | | 28.76** |
| Centralized setting | 369 (48.0) | 188 (66.7) | |
| Community based setting | 353 (46.0) | 84 (29.8) | |
| Living independently | 46 (6.0) | 10 (3.5) | |
| *Down syndrome* | | | 0.89 |
| No | 570 (74.2) | 201 (71.3) | |
| Yes | 104 (13.5) | 45 (16.0) | |
| Unknown | 94 (12.2) | 36 (12.8) | |

** $p$ < 0.05; ** $p$ < 0.01*

## Prevalence of low bone quality

Out of the 768 participants, 337 (43.9%; 95% CI = 40.4 – 47.4) had a T-score ≤ 2.5. The prevalence of low bone quality was significantly higher in women (49.5%; 95% CI = 44.4 – 54.5) compared to men (38.7%; CI = 34.0 – 43.6) and in older people compared to younger people ($X^2 = 9.73$, $p = 0.02$), but there was no significant difference in prevalence rates between participants with Down syndrome and participants with other causes of ID ($X^2 = 1.20$, $p = 0.27$). Because of the significant differences between participants and non-participants in level of ID and residential setting, we compared the T-scores for the three subgroups of each variable. Regarding level of ID, the Kruskal-Wallis test revealed a significant difference in T-scores. The median score for participants with a mild ID was -1.70, for participants with a moderate ID -2.30, and for participants with a severe ID -2.75 ($p$ <0.01). There was also a significant difference in T-scores for participants in different residential settings. The median score for participants in centralized settings was -2.50, for participants in community-based setting -2.10, and for participants living independently -1.40 ($p$ <0.01).

## Associations of low bone quality

Because of missing values, univariate analyses with low bone quality and the independent variables were performed with different numbers. Especially physical activity and intake of calcium could not be established in the majority of participants. For physical activity it was largely due to unreliable pedometer results because of a walking speed lower than 3.2 km/h ($n$ = 256 in the total HA-ID study population) and limited understanding or non-cooperation [24]. For intake of calcium the reason for missing values was the large amount of time it took to administer the food frequency lists for both caregivers and dieticians. Therefore these frequency lists were filled out for only a part of the study population [25].

The outcomes of the univariate analyses are presented in Table 2. Independent variables included in the multivariate logistic regression model were gender, age, moderate ID, severe ID, living in a community-based setting, living in a centralized setting, walking with walking aid, wheelchair dependency, BMI, prior fractures, use of anticonvulsant drugs and vitamin D3 serum level. The regression model is shown in Table 3. The proportion of variance explained by the model (Nagelkerke $R^2$) was 0.23. Being female, higher age, having a moderate ID, having a severe ID, sitting in a wheelchair and using anticonsulvant drugs were positively associated with low bone quality with an odds ratio (OR) of 2.37, 1.03, 2.49, 2.67, 3.25 and 2.17 respectively. BMI was negatively associated with low bone quality (OR = 0.94), meaning that people with a lower BMI were more likely to have a low bone quality than those with a higher BMI. VIF values were well below the threshold for multicollinearity.

**Table 2** *Univariate associations of bone quality with independent variables*

| Determinant | *n* | $X^2$ | *R* | *p*-value |
|---|---|---|---|---|
| Gender | 768 | 8.59 | | < 0.01 |
| Age | 768 | | -0.10 | < 0.01 |
| Level of ID | 751 | 24.34 | | < 0.01 |
| Residential status | 768 | 23.67 | | < 0.01 |
| Down syndrome | 674 | 1.20 | | 0.27 |
| Mobility | 736 | 35.08 | | < 0.01 |
| BMI | 745 | | 0.13 | < 0.01 |
| Prior fractures | 643 | 8.84 | | < 0.01 |
| Anticonvulsant drug use | 677 | 18.44 | | < 0.01 |
| Physical activity | 234 | | 0.09 | 0.16 |
| Intake of calcium | 158 | | 0.03 | 0.72 |
| Vitamin D3 | 501 | | 0.10 | 0.03 |

**Table 3** *Independent associations of low bone quality*

| | *B* | Wald | OR | 95% CI OR | |
|---|---|---|---|---|---|
| | | | | *LL* | *UL* |
| Female gender** | 0.86 | 11.62 | 2.37 | 1.44 | 3.88 |
| Age* | 0.03 | 4.06 | 1.03 | 1.00 | 1.06 |
| Moderate ID** | 0.91 | 8.83 | 2.49 | 1.36 | 4.54 |
| Severe ID** | 0.98 | 7.17 | 2.67 | 1.30 | 5.48 |
| Community setting | 1.20 | 1.22 | 3.31 | 0.40 | 27.55 |
| Centralized setting | 1.79 | 2.73 | 5.97 | 0.72 | 49.74 |
| Walking aid | 0.51 | 2.40 | 1.66 | 0.87 | 3.14 |
| Wheelchair* | 1.18 | 3.96 | 3.25 | 1.02 | 10.39 |
| BMI* | -0.07 | 5.63 | 0.94 | 0.89 | 0.99 |
| Prior fractures | 0.56 | 2.41 | 1.76 | 0.86 | 3.58 |
| Anticonvulsant drugs** | 0.77 | 6.76 | 2.17 | 1.21 | 3.88 |
| Vitamin D3 | 0.00 | 0.10 | 1.00 | 0.99 | 1.01 |

*Note. Proportion of variance explained by the model (Nagelkerke $R^2$): 0.23.*
*OR = odds ratio; 95% CI = 95% confidence interval; LL = lower limit; UL = upper limit; ID = intellectual disability; BMI = body mass index.*
** p < 0.05; ** p < 0.01*

## DISCUSSION

This study provides insight into prevalence and associated factors of low bone quality in older adults with intellectual disabilities. We found that low bone quality was highly prevalent in both older men and older women with ID, with prevalence rates of 39%

and 50% respectively. Prevalence in the total sample was 44%. The study sample consisted of relatively few participants with a severe ID. Because these participants particularly had a higher risk of low bone quality, the prevalence rates found in this study may be an underestimation of the actual prevalence rates. Besides being female and having a moderate or severe ID, also people with a higher age, people using anticonvulsant drugs, people in a wheelchair and people with a lower BMI had a higher risk of low bone quality.

High prevalence rates of low bone quality and/ or osteoporosis in different groups of people with ID were previously described. Jaffe et al. found low T-scores in 16 out of 33 men (48.5%; 95% CI 32.5 – 64.8 ) and in 53 out of 79 women (67.1%; 95% CI 56.2 – 76.5) aged 60 years and over living in residential facilities [11]. These rates seem higher than in our study population (for men 39.2%; 95% CI 32.9 – 45.8, and for women 55.9% ; 95% CI 48.5 – 62.9), but due to their smaller sample size, the studies' confidence intervals for both men and women overlap. In a much younger population of 298 people with ID living in the community, osteoporosis was present in 17.1% (95% CI 13.3 – 21.8) [9]. This prevalence is much lower than we found in our study, but it is not remarkable because of their population characteristics. We stress that, compared to the general population, the total prevalence we found in our study is the same as the prevalence of osteoporosis in women aged 75 years and over in the Netherlands [1]. However, the latter prevalence is likely to be an underestimation as well, because in the study by van der Linden et al people were not screened for osteoporosis; only people with a known diagnosis of osteoporosis were counted.

In the study by Zylstra et al. three predictors of osteoporosis were identified: older age, Caucasian race and non-ambulance [9]. In our study race was not included in the analyses because we didn't have information about it. However, in line with the above-mentioned study, age and level of ambulation were significantly associated with low bone quality. The other predictors identified in our study, were previously prescribed as well in studies among people with and without ID [5, 10, 11]. It is remarkable that in most of these studies the DXA-scan was used to measure bone quality. Although the QUS measurement has not been validated to determine low bone quality in people with ID, the overlap in independent predictors in DXA-studies and QUS-studies may be indicative of its adequate construct validity in this population.

The strengths of this study are the large study population and the comprehensive set of predictors. Therefore, results of this study may be extrapolated to the entire older ID population receiving specialized care, although the underrepresentation of people living in centralized settings and people with severe ID should be taken into account.

There are also some limitations of the present study. One limitation is that the prevalence rates of low bone quality that were found in this study are based on ultrasound

measurements of the heel bone. With the QUS device, SOS and BUA are measured, which are both indicators of bone quality. With the DXA-scan another indicator of bone quality is measured: bone mineral density. It is known that there is no perfect correlation between SOS and BUA measurements on the one hand and BMD measurements on the other hand [26, 27], making studies based on measurements of bone mineral density by DXA difficult to compare with our results. Second, as mentioned above, quantitative ultrasound has not been validated in people with ID. To that end, criterion validity should be tested by looking at the predictive value of QUS for future fractures among people within this population. At this moment, within the HA-ID research group we are preparing such a study.

In conclusion, because of the high prevalence of low bone quality in older adults with ID, general practitioners and specialized physicians for people with ID need to pay attention to prevention of osteoporosis and prevention of fractures, meaning that people with ID who are at risk for low bone quality should periodically be screened for osteoporosis. Factors that require attention are anticonvulsant drug use, poor mobility and low BMI. In addition, the usual guidelines for the prevention of fractures should be followed (sufficient intake of vitamin D and calcium and spending sufficient time outside).

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# Chapter 9

## Prevalence and associated factors of sarcopenia in older adults with intellectual disabilities

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## ABSTRACT

Sarcopenia is defined as a syndrome characterized by progressive and generalized loss of skeletal muscle mass and strength. It has hardly been studied in older people with intellectual disabilities (ID). In this study 884 persons with borderline to profound ID aged 50 years and over, were investigated to determine the prevalence of sarcopenia in this group. To identify the associations of sarcopenia, logistic regression analyses were performed with patient characteristics, mobility, physical activity, intake of energy and proteins, body mass index (BMI) and levels of CRP, albumin and vitamin D in serum. The prevalence of sarcopenia was 14.3% in the total group. In the age group 50 – 64 years prevalence was 12.7%. Sarcopenia was positively associated with mobility impairment and inflammation and negatively with BMI. The next thing to do is collecting longitudinal data to study the relation between sarcopenia and negative outcomes in older people with ID.

## INTRODUCTION

Sarcopenia was originally described in the late 1980s as the age-related loss of skeletal muscle mass. Parallel to the increasing knowledge, the definition of sarcopenia has evolved [1]. Nowadays, combining both muscle mass and muscle function to define sarcopenia is the recommended approach. In 2009 the European Working Group on Sarcopenia in Older People (EWGSOP) defined sarcopenia as a syndrome characterised by progressive and generalised loss of skeletal muscle mass and strength with a risk of adverse outcomes such as physical disability [2].

The underlying pathophysiological pathways of sarcopenia are complex. There are several internal and external processes that contribute to its development [3]. With regard to internal processes, inflammation (expressed as increased levels of C-reactive protein), reductions of anabolic hormones, accumulation of free radicals and increases of apoptotic activities play a central role in sarcopenia [3-5]. External processes leading to sarcopenia are disuse (including immobility and physical inactivity) and a deficient intake of energy and proteins [3, 6, 7].

Prevalences of sarcopenia vary between 3% and 52% depending on the study population, the criteria used to define sarcopenia and the instruments used to measure it [8]. In subsequent studies in the same population, it was shown that prevalences of sarcopenia increased from 12% when people were aged 60 to 70 years to nearly 30% when they were older than 80 years [9, 10].

In most studies, men are more likely to be sarcopenic than women [11-15]. People with a low body mass index (BMI) [13], or with low serum vitamin D levels [16] or low serum albumin levels [17, 18] are also at greater risk for sarcopenia.

In the general population sarcopenia is a risk factor for frailty [19, 20]. Both sarcopenia and frailty are highly relevant conditions with regard to functionality and independence in old age [11, 20-23]. It is known that these conditions overlap; most frail older people exhibit sarcopenia and some older people with sarcopenia are also frail [20]. Looking at the criteria for sarcopenia in accordance with the EWGSOP [2] and the criteria for frailty in accordance with the Cardiovascular Health Study (CHS) [24], this overlap can partly be explained by the similarity in parameters for both muscle strength and muscle performance.

In people with intellectual disabilities (ID) sarcopenia has hardly been studied before. As far as we know, the only study in this population was executed by Carmeli, Imam and Merrick [25]. They studied the correlation between sarcopenia and physical ability in a small group of healthy, older participants with a relatively high level of independency [25]. They found that loss of muscle mass and loss of muscle strength were associated with early onset of physical and functional decline.

We expected that sarcopenia is frequent in older people with ID, due to high levels of physical inactivity [26] and a high prevalence of (lifelong) mobility impairments [27]. Therefore, we set up a study into sarcopenia, in accordance with the EWGSOP-criteria, in a nearly-representative older client population of Dutch ID care provider services. The aims of the current study were (1) to determine the prevalence of sarcopenia in older adults with ID, (2) to identify the association of sarcopenia with participant characteristics (gender, age, level of ID, Down syndrome, residential status) and mobility, physical activity, intake of energy and proteins, BMI and levels of C-reactive protein (CRP), albumin and vitamin D in serum, and (3) to determine the overlap between sarcopenia and frailty in accordance with the CHS-criteria in this group.

## METHODS

### Study design and participants

This study was part of a large cross-sectional study, called "Healthy Ageing in people with Intellectual Disability" (HA-ID). The recruitment and selection process of the HA-ID study has been outlined in detail by Hilgenkamp et al. [28]. The study population consisted of clients, aged 50 years and over, in three Dutch care provider services in the Netherlands. Informed consent was obtained from 1069 clients, aged 50 years and over, and/or their legal representatives and 1050 clients actually participated in the assessments. The study population was nearly representative for the total client population aged 50 years and over of the care providers, with a slight overrepresentation of women, and a slight underrepresentation of individuals living independently and individuals aged 80 years and over [28]. This study has been approved by the Medical Ethics Committee of the Erasmus Medical Center Rotterdam, the Netherlands (MECnr. 2008-234) and by the ethics committees of the participating care provider services. The study was conducted in accordance with the guidelines from the Declaration of Helsinki for research involving human subjects.

### Sarcopenia

The criteria of the EWGSOP [2] were used to define sarcopenia. These criteria comprise muscle mass, muscle strength and muscle performance. Presarcopenia was defined as having low muscle mass and normal muscle function (strength and performance); sarcopenia as having low muscle mass combined with low muscle strength or low muscle performance; and severe sarcopenia as having low muscle mass, low muscle strength and low muscle performance. The criteria were operationalized as follows:

- For muscle mass, calf circumference (CC) was measured. This parameter has been considered to be the most sensitive anthropometric measure of muscle mass in

the elderly by the World Health Organization [29]. It has been shown that a cut-off value of 31 cm may serve as an indicator for sarcopenia and that it is associated with disability and self-reported physical function [30]. CC was measured using a standard anthropometric tape with the participant in standing position. The tape was wrapped around the calf of the non-dominant leg at the widest part to obtain the maximal circumference. Subcutaneous tissues were not compressed.

- Grip strength [31] was measured to determine muscle strength. For grip strength participants squeezed a Jamar Hand Dynamometer (Sammons Preston Rolyan, USA) to their maximum ability in seated position, in accordance with the recommendations of the American Society of Hand Therapists [32]. The best result of three attempts for both the left and the right hand (with a one-minute pause between attempts) was recorded. The test instructors had to be convinced the participants squeezed with maximal effort; otherwise they did not record a test result. Low muscle strength was defined as lower than 30 kilograms for men and 20 kilograms for women in accordance with Laurentani et al. [23].
- To determine muscle performance, comfortable walking speed was measured over a distance of 5 meters (after 3 meters for acceleration). Three attempts were averaged to get the participant's result. The participants had to walk the distance without someone walking alongside or physically supporting them, to avoid influence on comfortable speed. Walking aids were allowed. A cut-off point of 0.8 m/s was chosen because several studies showed adverse outcomes of mobility, ADL and mortality in people with a walking speed lower than 0.8 m/s [33].

Because not all participants were able to participate in all assessments, criteria could be missing. If it was possible to categorize participants into 'no sarcopenia', 'presarcopenia', 'sarcopenia' or 'severe sarcopenia', they were included. At least a valid result for calf circumference was needed to meet this requirement.

### Factors associated with sarcopenia

Gender, age and residential status were collected from the records of the care provider services. Residential status was divided into living with relatives, independent living, living in group homes in the community and living in group homes in central settings. General practitioners and specialized physicians for people with ID recorded aetiology of ID (Down syndrome yes /no) and psychologists or behavioural therapists recorded level of ID (borderline, mild, moderate, severe or profound ID).

Professional caregivers were asked about mobility (divided into walking independently, walking with aid and wheelchair dependent). Participants underwent a physical examination by specially trained medical assistants to assess body height and weight. Body height was measured using a Seca stadiometer, type 214, with the participant standing, wearing no shoes. For non-ambulant people, knee height was measured us-

ing a non-stretchable flexible tape, and the formula developed by Chumlea, Roche and Steinbaugh [34] was used to calculate body height. Weight was measured using a digital floor scale (Seca robusta type 813), with participants wearing light clothes and no shoes. BMI was calculated by weight divided by squared height.

Physical activity was assessed with pedometers (NL-1000, New Lifestyles, Missouri, USA) worn for at least four days [35]. Participants with a comfortable walking speed of 3.2 km/h or more in at least one of three recordings were invited to wear a pedometer. The pedometer was attached to the belt worn by the participant, halfway between the umbilicus and the side of the body. The professional caregiver was instructed to record the number of steps, distance and activity in minutes in a diary every evening.

In blood samples serum levels of albumin, CRP and vitamin D were assessed. Intake of energy and proteins was assessed by food intake diaries. During three days, including one day in the weekend, professional caregivers had to register the whole intake of food and fluid of the participants. Dieticians entered the products into a web program based on nutritional values of Dutch food products [36]. This program converted the food and fluid products into energy and protein intake and other macronutrients and micronutrients.

## Frailty

The criteria of the Cardiovascular Health Study were used for the definition of frailty [24]. Participants with at least three of the following five criteria were defined as frail: weight loss, poor grip strength, slow walking speed, low physical activity, and poor endurance or exhaustion. The criteria were operationalized as follows:

- Weight loss: asked as an item of the Mini Nutritional Assessment (MNA) [37]. In this screening tool for malnutrition weight loss during the past three months is asked on a four point rating scale. Losses over 3 kg were scored.
- Grip strength: measured with the Jamar Hand Dynamometer. Poor grip strength was defined in accordance with CHS criteria [24]. For men with BMI ≤ 24, between 24.1 and 28, and > 28 the cut-off points were 29, 30 and 32 kilograms respectively and for women with BMI ≤ 23, between 23.1 and 26, between 26.1 and 29, and > 29 these cut-off points were 17, 17.3, 18 and 21 kilograms respectively.
- Comfortable walking speed: measured by the average of three recordings of the time to complete a distance of 5 metres. Slow walking speed was defined in accordance with the CHS criteria, with cut-off points of 0.65 m/s for men ≤ 1.73 meters and women ≤ 1.59 meters and 0.76 m/s for men > 1.73 meters and women > 1.59 meters [24]. All participants in a wheelchair and all participants who could not engage in walking speed assessment due to physical limitations were scored as 'slow walking speed' as well.
- Physical activity: tested with pedometers. All participants walking less than 5000 steps/day (sedentary lifestyle) were scored positive on low physical activity, as

were all participants in a wheelchair and all participants who could not engage in walking speed assessment due to physical limitations.

- Exhaustion: asked as the item 'lacks energy' of the Anxiety, Depression And Mood Scale (ADAMS) [38]. This item has a four point rating scale. The 'no problem' and 'mild problem' answers were recoded into 'no' and the 'moderate problem' and 'severe problem' answers into 'yes'.

## Statistical analysis

Statistical analyses were performed with IBM SPSS Statistics 17.0. Prevalence rates and 95% confidence intervals (CI) of presarcopenia, sarcopenia and severe sarcopenia were calculated, by using descriptive analyses. For further analyses, no sarcopenia and presarcopenia were combined into 'no sarcopenia' and sarcopenia and severe sarcopenia into 'sarcopenia'.

A non-response analysis of the participants with consent to participate but without completed data for sarcopenia was conducted using a $X^2$-test for gender, age, level of ID, residential status and mobility. The same procedure was followed to analyze differences between participants with sarcopenia and participants without sarcopenia.

To explore associations between sarcopenia (dependent variable) and gender, age, level of ID, Down syndrome, residential status, mobility, physical activity, intake of energy, intake of proteins, BMI, serum level of CRP, serum level of albumin and serum level of vitamin D (independent variables), univariate logistic regression analyses were performed. If an independent variable showed a significant association ($p < 0.05$) with the dependent variable, it was subsequently entered into a multivariate logistic regression analysis. Due to the small amount of participants in physical activity, intake of energy and intake of proteins, these variables were excluded from the multivariate analysis.

All independent variables were continuous, except for gender (men = 0, women = 1), level of ID (borderline = 0, mild = 1, moderate = 3, severe = 4, profound = 5), Down syndrome (no Down syndrome = 0, Down syndrome = 1), residential status (group homes in central setting = 0, group homes in community = 1, independent living = 2, with relatives = 3) and mobility. For mobility two dummy variables were used: walking with support and wheelchair-dependent, both compared to people who are able to walk independently (walk independently = 0, walking with support or wheelchair-dependent = 1). In the multivariate model patient characteristics (age, gender, level of ID, Down syndrome and residential status) were entered into the equation first, after which mobility, BMI, CRP, albumin and vitamin D were entered simultaneously. Multicollinearity was checked for all independent variables with the variance inflation factor (VIF) of linear regression analysis. VIF values above 10 indicated multicollinearity [39].

To explore the relationship between frailty and sarcopenia we used a $X^2$-test.

## RESULTS

### Participants

For 929 out of 1050 participants in the total cohort, a valid result for CC was obtained. Due to missing data for muscle strength or muscle performance 45 persons with a low CC could not be categorized into presarcopenia, sarcopenia or severe sarcopenia and

**Table 1** *Characteristics of participants (n = 884)*

| | *n* | % |
|---|---|---|
| *Gender* | | |
| Men | 450 | 50.9 |
| Women | 434 | 49.1 |
| *Level of ID* | | |
| Borderline (IQ 70-85) | 30 | 3.4 |
| Mild (IQ 55-70) | 201 | 22.7 |
| Moderate (IQ 40-55) | 437 | 49.4 |
| Severe (IQ 25-40) | 132 | 14.9 |
| Profound (IQ <25) | 62 | 7.0 |
| Unknown | 22 | 2.5 |
| *Residential status* | | |
| Group homes, central setting | 441 | 49.9 |
| Group homes, community | 393 | 44.5 |
| Independent living | 43 | 4.9 |
| Living with relatives | 7 | 0.8 |
| *Age* | | |
| 50-59 years | 411 | 46.5 |
| 60-69 years | 311 | 35.2 |
| 70-79 years | 143 | 16.2 |
| 80 years and over | 19 | 2.1 |
| 50-64 years | 597 | 67.5 |
| 65 years and over | 287 | 32.5 |
| *Down syndrome* | | |
| No | 616 | 69.7 |
| Yes | 111 | 12.6 |
| Unknown | 157 | 17.8 |
| *Mobility* | | |
| Independent | 641 | 72.5 |
| With support | 123 | 13.9 |
| Wheelchair | 82 | 9.3 |
| Unknown | 38 | 4.3 |

therefore they were excluded. Of the remaining 884 participants, data of 103 persons on comfortable walking speed were missing, largely due to physical impairments, and data of 173 persons on grip strength were missing, largely due to limited understanding or non-cooperation.

The excluded group ($n$ = 166) had a similar gender and age distribution as the included group (data not shown), but included significantly more persons with severe and profound ID ($X^2 = 36.3$, $p < 0.01$), more persons living in group homes in central settings ($X^2 = 23.5$, $p < 0.01$) and more persons in a wheelchair ($X^2 = 11.6$, $p < 0.01$). In the included group 111 persons had Down syndrome. Other characteristics of the participants are presented in table 1.

## Prevalence of sarcopenia

Out of the 884 participants, 734 (83.0%) had no sarcopenia (95% CI = 80.4-85.4%), 24 (2.7%; 95% CI = 1.8-4.0%) had pre-sarcopenia, 80 (9.1%; 95% CI = 7.3-11.1%) had sarcopenia and 46 (5.2%; 95% CI = 3.9-6.9%) had severe sarcopenia, resulting in a total prevalence of 14.3% (95% CI = 12.1-16.7%) of sarcopenia. Muscle mass was scored as low in 150/884 participants (17.0%), muscle strength in 349/711 (49.1%), and muscle performance in 321/781 (41.1%). Differences in gender, age, level of ID, residential status, Down syndrome and mobility between the non-sarcopenic and the sarcopenic participants are presented in table 2. In the sarcopenic group participants were older, more participants were living in group homes in central settings, more participants had a profound ID and more persons were wheelchair-dependent compared to the group without sarcopenia.

In the age group 50-64 years, prevalence of sarcopenia was 12.7% (95% CI = 10.3-15.7%). In the sarcopenic group of this age 38.4% of the participants were dependent on a wheelchair versus 3.4% in the non-sarcopenic group. In the age group 65 years and over, prevalence of sarcopenia was 17.4% (95% CI = 13.5-22.2%). In this age group 36.7% of the sarcopenic people were dependent on a wheelchair versus 8.3% in people without sarcopenia.

## Associations of sarcopenia

Because of missing values, univariate analyses with sarcopenia and the independent variables were performed with different numbers. These numbers were 884 for gender, age and residential status; 862 for level of ID; 846 for mobility; 844 for BMI; 647 for serum level of albumin; 646 for serum level of CRP; 586 for serum level of vitamin D; 252 for physical activity; and 225 for intake of energy and proteins. Especially physical activity and intake of energy and proteins could not be established in a majority of participants. For physical activity it was largely due to unreliable pedometer results because of a walking speed lower than 3.2 km/h ($n$ = 256 in the total HA-ID study

**Table 2** *Comparison of patient characteristics of sarcopenic and non-sarcopenic participants*

| | No sarcopenia | | Sarcopenia | | |
|---|---|---|---|---|---|
| | *n* (%) | $(X_o-X_e)^2/X_e$ | *n* (%) | $(X_o-X_e)^2/X_e$ | $X^2$ |
| *Gender* | | | | | 0.05 |
| Male | 387 (51.1) | 0.00 | 63 (50.0) | 0.02 | |
| Female | 371 (48.9) | 0.00 | 63 (50.0) | 0.02 | |
| *Age* | | | | | 13.9** |
| 50-59 years | 360 (47.5) | 0.16 | 51 (40.5) | 0.99 | |
| 60-69 years | 271 (35.8) | 0.07 | 40 (31.7) | 0.42 | |
| 70-79 years | 113 (14.9) | 0.75 | 30 (23.8) | 4.52 | |
| 80-89 years | 13 (1.7) | 0.04 | 3 (2.4) | 0.21 | |
| 90-94 years | 1 (0.1) | 0.98 | 2 (1.6) | 6.40 | |
| *Residential status* | | | | | 34.9** |
| Central setting | 348 (45.9) | 2.40 | 93 (73.8) | 14.40 | |
| Community-based | 362 (47.8) | 1.85 | 31 (24.6) | 11.16 | |
| Independent | 42 (5.5) | 0.70 | 1 (0.8) | 4.26 | |
| With relatives | 6 (0.8) | 0.00 | 1 (0.8) | 0.00 | |
| *Level of ID* | | | | | 51.9** |
| Borderline | 28 (3.7) | 0.23 | 2 (1.6) | 1.31 | |
| Mild | 186 (24.5) | 1.16 | 15 (11.9) | 6.83 | |
| Moderate | 376 (49.6) | 0.02 | 61 (48.4) | 0.09 | |
| Severe | 112 (14.8) | 0.01 | 20 (15.9) | 0.04 | |
| Profound | 35 (4.6) | 6.11 | 27 (21.4) | 36.00 | |
| Unknown | 21 (2.8) | | 1 (0.8) | | |
| *Mobility* | | | | | 140.6** |
| Independent | 590 (77.8) | 1.99 | 51 (40.5) | 28.31 | |
| With support | 98 (12.9) | 7.53 | 25 (19.8) | 0.47 | |
| Wheelchair | 36 (4.8) | 33.66 | 46 (36.5) | 60.02 | |
| Unknown | 34 (4.5) | | 4 (3.2) | | |

** $p < 0.01$

population) and limited understanding or non-cooperation ($n$ = 204) [26]. For intake of energy and proteins it had to do with the large amount of time it took to administer the food intake diaries for both caregivers and dieticians. Therefore these diaries were filled out for only a part of the study population.

The outcomes of the univariate analyses showed that sarcopenia was not significantly associated with gender ($p$ = 0.83), Down syndrome ($p$ = 0.29), protein intake ($p$ = 0.07), energy intake ($p$ = 0.19) and physical activity ($p$ = 0.90). Eight out of 252 participants who successfully wore a pedometer to measure physical activity were sar-

copenic. Independent variables included in the multivariate logistic regression model were age, residential status, level of ID, BMI, walking with walking aid, wheelchair dependency, serum albumin, serum CRP and serum vitamin D. The regression model is shown in table 3. The proportion of variance explained by the model ($R^2$) was 0.34.

Walking with walking aid, wheelchair dependency and to a lesser extent CRP were positively associated with sarcopenia with an odds ratio (OR) of 3.96, 34.23 and 1.01 respectively. BMI was negatively associated with sarcopenia (OR = 0.82), so people with a lower BMI were more likely to be sarcopenic than those with a higher BMI.

**Table 3** *Independent associations of sarcopenia*

| | *B* | Wald | OR | 95%CI OR | |
|---|---|---|---|---|---|
| | | | | *LL* | *UL* |
| Age[a] | 0.03 | 2.31 | 1.03 | 0.99 | 1.07 |
| Residential status[a] | -0.19 | 0.33 | 0.83 | 0.44 | 1.56 |
| Level of ID[a] | -0.16 | 0.66 | 0.85 | 0.58 | 1.26 |
| BMI[b]** | -0.20 | 25.70 | 0.82 | 0.76 | 0.88 |
| Walking aid[b]** | 1.38 | 14.10 | 3.96 | 1.93 | 8.11 |
| Wheelchair[b]** | 3.53 | 46.49 | 34.23 | 12.40 | 94.51 |
| Serum albumin[b] | -0.03 | 0.41 | 0.97 | 0.88 | 1.07 |
| Serum CRP[b]* | 0.01 | 4.52 | 1.01 | 1.00 | 1.03 |
| Serum vitamin D[b] | 0.002 | 0.21 | 1.00 | 0.99 | 1.01 |

*Note. Proportion of variance explained by the model ($R^2$ Nagelkerke): 0.34.*
*The Wald test is used to test the statistical significance of each coefficient (B) in the model.*
*OR = odds ratio; 95% CI = 95% confidence interval: LL = lower limit; UL = upper limit.*
[a] *Entered in step 1;* [b] *Entered in step 2.*
***** *p < 0.05; ** p < 0.01*

## Overlap sarcopenia and frailty

Data on both sarcopenia and frailty were available in 798 participants. The overlap between sarcopenia and frailty is presented in table 4. Out of 122 sarcopenic and 676 non-sarcopenic participants, 33 (27.0%) and 64 (9.5%) persons were frail respectively ($X^2 = 28.3$, $p < 0.01$). The other way around, 33 out of 97 (34.0%) frail people

**Table 4** *Overlap between sarcopenia and frailty (n = 798)*

| | Sarcopenic<br>*n* (%) | Non-sarcopenic<br>*n* (%) | Total<br>*n* (%) |
|---|---|---|---|
| Frail | 33 (27.0) | 64 (9.5) | 97 (12.2) |
| Non-frail | 89 (73.0) | 612 (90.5) | 701 (87.8) |
| Total | 122 (100) | 676 (100) | 798 (100) |

had sarcopenia. In the total group of frail people, calf circumference was low in 35.1%, grip strength in 90.5%, and walking speed in 89.0%.

## DISCUSSION

This is the first study into prevalence and associated factors of sarcopenia in a large near-representative older population with intellectual disabilities. The prevalence of sarcopenia in this group is 14.3%. Sarcopenia is positively associated with mobility impairment and inflammation and negatively with BMI. Sarcopenia and frailty partly overlap, but less than we expected based on the similarity in criteria. With a prevalence of 12.7%, sarcopenia is already remarkably present in the age group 50 – 64 years. One possible explanation for early sarcopenia is the high percentage of people who are wheelchair-dependent (38.4% in the sarcopenic group vs. 3.4% in the non-sarcopenic group). It may be hypothesized that in the population with ID sarcopenia is a disuse-related condition as well as an age-related condition, although with the current set of independent variables only 34% of the variance in sarcopenia is explained.

The EWGSOP criteria for sarcopenia that we used in this study, originate from 2009 [2]. Since then, hardly any prevalence studies were published using these criteria, hampering comparison with prevalences in the general population. Landi et al. [40] investigated sarcopenia in nursing homes, using the same methods as we used for muscle strength and muscle function; muscle mass was measured by bioelectrical impedance analysis (BIA). A prevalence of 32.8% (95% CI = 25.1-41.5) was found in 122 persons aged 70 years and over. Although our methods do not completely overlap, this prevalence is much higher than in the subgroup aged 65 years and over in our population. However, the prevalence we found is likely to be an underestimation, because the participating group was functionally more able.

In contrast with previous studies, we did not find an association of sarcopenia with physical activity. This may be explained by the fact that only people with a walking speed of at least 3.2 km/h were included for pedometer assessment. Because this is the more functionally able group of the study [26], it is not surprising that only 8 out of 252 persons within this group were sarcopenic.

The low overlap between sarcopenia and frailty warrants consideration. We were not surprised that frailty was more related to functional measurements than to anthropometric measurements. Indeed, the CHS-criteria consist of functional measures in particular. Moreover, the low percentage of frail people with a low muscle mass (35.1%) is in line with results from the study of Barzilay et al. [41]. On cross-sectional analysis, they found a positive association between frailty and obesity due to an increase in fat mass.

The strengths of this study are the large, nearly representative study population. Therefore, results of this study may be extrapolated to the whole older ID population receiving specialized care in the Netherlands, although the underrepresentation of people living in central settings, people with severe and profound ID and people in a wheelchair in the for this study included group should be taken into account. Secondly, the use of international criteria to define sarcopenia enables future comparison with prevalences and associations of sarcopenia in other populations.

A limitation of the present study is that calf circumference was used to determine muscle mass. It is known that anthropometric measures are vulnerable to error, especially in older and obese people due to changes in fat deposits and loss of skin elasticity [2]. Therefore dual energy x-ray absorptiometry (DEXA), computed tomography (CT), magnetic resonance imaging (MRI), and bioelectric impedance (BIA) are recommended [42, 43]. However in large-scale research, these techniques may not be regarded as feasible for people with ID, equally to nursing home residents [44]. CT, MRI, and DEXA can not be used, because they are expensive and located in hospitals. With regard to BIA, equations for calculation of skeletal mass in people with ID are not available, despite different body height and fat distribution for example in people with Down syndrome.

Secondly, accurate walking speed and hand grip measurements may have been hampered by functional impairment, which is frequent in people with ID. For both measurements, it is essential that the participant is able to cooperate. To advance optimal measurements, in our study professionals working with people with ID were trained to measure both grip strength and walking speed. A rubber ball to squeeze in, was used as a visual example to explain the purpose of the grip strength measurement.

As mentioned above, in our sample a low BMI and impaired mobility were associated with sarcopenia. In this respect, the most important option for prevention and treatment in people with ID includes resistance training programmes and nutritional intervention as described in a review by Rolland, Dupuy, Abellan van Kan, Gillette and Vellas [45].

Within the HA-ID research group, we are interested in several concepts predicting (un)healthy ageing. Besides sarcopenia and frailty [46] in accordance with CHS-criteria [24], we are now investigating the frailty index [47], multimorbidity, and polypharmacy in the same study population. This exploratory study into sarcopenia is a first step to a model in which causal relationships between these concepts are elaborated in order to gain insight into the relevance of each concept for people with ID. The next thing to do is collecting longitudinal data to study the relation between sarcopenia and negative outcomes in older people with ID.

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# Chapter 10

General discussion

This thesis describes studies on nutrition and nutritional status in a large sample of older adults with intellectual disabilities (ID) in the Netherlands. The focus lies on underweight and undernutrition and the factors associated with these unhealthy conditions. After an overview of the main findings, we will reflect on these findings. Subsequently, the methodological issues of this study are discussed. Finally, recommendations for clinical practice and future research will be presented.

## MAIN FINDINGS

We have shown that the Mini Nutritional Assessment (MNA) is feasible and reliable for use among older people with intellectual disabilities. However, its construct validity and criterion validity are low. As a result, we were not able to study undernutrition in the HA-ID population.

Alternatively, we investigated food intake within our study. We have shown that in a group of 228 older people with intellectual disabilities, a majority poorly meets the daily recommendations for healthy food intake. Especially the recommendations for saturated fat and fibre intake are hardly met in this population, making these persons more vulnerable for developing cardiovascular diseases and constipation, respectively. Almost one third of the examined group ate insufficient amounts of proteins as well. It is commonly known that in the general population, most people do not meet these criteria as well, but compared to the dietary intake of older people with ID aged 50 to 70 years, the intake in the general older population is better [1].

Regarding micronutrients, we have studied vitamin D deficiency. We found a high prevalence of 42%, in spite of routine supplementation regimens in part of the settings, making these people prone to develop low bone quality. The prevalence of low bone quality was 43.9%. To our surprise, bone quality was not correlated with vitamin D deficiency. This was most likely caused by the confounding effect of supplementation, which was given in a subpopulation consisting of relatively more women, more people in a wheelchair, and more people with a more severe level of intellectual disabilities, which are all risk factors for low bone quality. Also higher age, anticonvulsant use and low BMI were associated with low bone quality.

Another negative consequence of insufficient food intake, especially protein intake (together with low physical activity), may be sarcopenia (increased loss of muscle mass). We were the first to perform an epidemiological study of this condition in older adults with intellectual disabilities. We have shown that sarcopenia appears to be present at a relatively young age in older people with intellectual disabilities, with a prevalence of 12.7% in the age group of 50-64 years. As was to be expected, especially people using a wheelchair are at risk of developing sarcopenia (odds ratio

34.2), although we have to be careful interpreting the causality of this relationship due to the cross-sectional design of our study.

## REFLECTION

Within the HA-ID research group, we have studied sarcopenia and frailty [2], which are both conditions predicting (un)healthy ageing in the general population [3, 4]. Regarding frailty, a frequently used physically oriented operationalization has been developed for the American Cardiovascular Health Study, and is based on the presence of weight loss, poor grip strength, slow walking speed, low physical activity, and poor endurance or exhaustion [4]. In the population with ID, both sarcopenia and frailty are present at a relatively young age [2]. Unhealthy food intake may play an important role in developing early sarcopenia and frailty. Indeed, in case of undernutrition, muscle mass and muscle function decrease and as a consequence, frailty may arise. Therefore, healthy food should be high on the agenda of policymakers.

However, in people with intellectual disabilities, there are a lot of barriers to achieve healthy food intake. In a yet unpublished focus group interview study among Dutch dieticians working with people with intellectual disabilities, we found that the following barriers make it difficult to improve dietary intake: lack of knowledge regarding healthy food among professional caregivers, a low budget hampering use of diverse food products, a shortage of professional caregivers hampering preparation of diverse meals, lack of an explicit vision of the management regarding food supply and education. To improve dietary intake, a multi-level approach is therefore very important.

A single widely accepted diagnostic operationalization of malnutrition is currently lacking [5]. Malnutrition is defined as 'a nutritional status involving a deficiency (undernutrition) or excess (overnutrition) for energy, protein, and other nutrients that have measurable adverse effects on body composition (body shape, size and composition) and body function, and have negative clinical implications' [6]. This broad definition implies that malnutrition may be caused by many factors that may vary in severity and cause. It is therefore not surprising that malnutrition has been operationalized in various ways in scientific literature [7-9]. In the HA-ID study we have chosen for the MNA to detect undernutrition, but, unfortunately, the MNA appeared insufficiently valid in this population. Therefore, screening for malnutrition among (older) adults with intellectual disabilities is not easily possible at this moment, hampering timely detection of this condition. Because of the negative consequences of malnutrition, the development of a valid screening tool for this population is urgent. In the paragraph 'recommendations for research' we discuss this issue in more detail.

## METHODOLOGICAL ISSUES

### Representativeness of the study population

Older people with a mild intellectual disability not known to formal ID services were not included in the HA-ID study. Consequently, the results of the HA-ID study are only generalizable to older people with ID receiving formal care and/or support. Furthermore, after the informed consent procedure we had a slight overrepresentation of women, and a slight underrepresentation of individuals living independently within the HA-ID study population. The consequences for generalizability of the results would probably be small, because gender and living situation were mostly not significantly associated with the main outcomes.

Due to selection bias, people with severe and profound ID and people living in centralised settings were overrepresented in the group of 228 participants who were included in the dietary intake study. The consequences for the representativeness of our data are hard to define. On the one hand, it was shown in the HA-ID study that people living relatively independently (doing groceries themselves, preparing meals themselves) were more at risk for being overweight [10]. Therefore, quality of dietary intake in the total group of clients of ID care might even be worse. On the other hand, it is known that people with severe ID are more often suffering from conditions like dysphagia and gastroesophageal reflux disease [11]. This may lead to an unhealthy food intake and therefore, the results of the dietary intake study might be an overestimation of the problem of unhealthy intake. Probably due to the small sample size of the dietary intake study, we did not find the expected associations between intake of calcium and low bone quality and between intake of proteins and energy, and sarcopenia. Therefore, we recommend to study these associations in larger populations, especially regarding sarcopenia, because of the minimal evidence-based knowledge of sarcopenia in people with ID so far.

### Methodological issues regarding the diagnostic methods

Before the HA-ID study started, most large-scale epidemiological studies among people with ID were based on questionnaires and medical files [12, 13]. In this study, we chose to use objective tests. For some assessments we have chosen for instruments which are not considered to be gold standard. To measure bone quality, we have used quantitative ultrasound (QUS) of the heel bone and for muscle mass, we have measured calf circumference, because measurements of bone quality by Dual-energy X-ray Absorptiometry (DXA) and muscle mass by DXA or Bioelectrical Impedance Analysis (BIA), which are considered to be the gold standard, are expensive and require hospital visits. Therefore, they are less applicable in large-scale research.

Indeed, both QUS and calf circumference are recommended in scientific literature for epidemiologic research [14-17].

A severe setback to the success of this study was the fact, that both the MNA and QUS of the heel bone appeared invalid in the current population. After ample consideration, we had chosen these methods because of their on-site applicability, good feasibility in people with ID, and good validity in general population studies. The lack of validity of the MNA was discussed above. We have only recently established that low bone quality as measured by QUS significantly predicts fractures during a 3-year follow-up of the study population in the univariate analysis. However, in the multivariate analysis, its effect was overshadowed by the effects of antiepileptic drug use and past fractures [18]. These findings have implications for the recommendations regarding screening and prevention, which will be described in the next paragraph.

## IMPLICATIONS AND RECOMMENDATIONS FOR CLINICAL PRACTICE

### Nutrition and nutritional state

Because many people with ID depend on others for their food, it is not always possible for them to improve the intake themselves. Therefore, policy makers, together with dieticians and physicians, should pay more attention to food supply and food abundance. In the near future the responsibility for the care for relatively independently living persons with ID will be changed from government-based to municipality-based. At the same time, the budget for care will decrease, meaning that for people with a mild and moderate ID in particular, support will be reduced. As a consequence, more people with ID will be responsible for doing groceries and/or preparing meals themselves. In the HA-ID study we have shown that those people are prone to develop obesity [10]. Dieticians should be involved to prevent obesity in this high-risk population.

In addition, more focus should be on education about healthy nutrition and the harmful effects of an unhealthy diet among professional caregivers working with people with ID and people with ID themselves. It is important for both groups to look critically at the intake of snacks, because both caregivers and people with ID are often unaware of the unhealthy effects of these products.

Finally, in previous research, several studies into food interventions among children have shown that a multidisciplinary approach is important to improve nutritional status. Interventions addressing education, changing behavior, healthier food intake (more fruit and vegetables), and a better food supply at the same time, are most successful [19-21]. Because of the similarity between children and people with intel-

lectual disabilities regarding dependency on others for food, we recommend to act similarly in care provider services for people with intellectual disabilities.

### Associated factors of a poor nutritional state

Because it has been shown in this thesis that conditions related to a poor nutritional status (dysphagia, low bone quality and sarcopenia) are common in older people with intellectual disabilities, and because these diseases are related to negative consequences regarding independence and/or health, prevention and/or proactive screening seems to be important. In the next paragraphs prevention and screening of each condition will be discussed.

### Dysphagia

We found that fifty-two percent of the HA-ID study population are suffering from moderate to severe dysphagia, not restricted to people with severe motor impairment and people with dementia. It is even more alarming that in almost 90% of the participants with dysphagia identified by a standardized mealtime observation, this diagnosis was not mentioned in his/her medical file. Therefore, in the HA-ID consortium, at this moment we are preparing a care improvement project aimed at recognition and diagnosis of dysphagia and prevention of choking. Education of professional caregivers will play a key role in this project. Moreover, we recommend screening risk groups for dysphagia (age ≥ 70 years, Down syndrome, mobility impairment, feeding with help, use of benzodiazepines), in line with the general population, where screening for dysphagia is also limited to risk groups (i.e. stroke patients, patients with Parkinson disease or other progressive neurological diseases, patients with dementia) [22]. Screening can be done by using the Dysphagia Disorder Survey (DDS), because of its good feasibility and reliability in people with ID. However, to administer the DDS, a standardised training that results in certification is necessary. At this moment, there are insufficient certified speech therapists in the Netherlands. Therefore, in order to guarantee expert screening of dysphagia in people with ID in the future, the availability of sufficient trainers should be arranged by means of train-the-trainer courses by the Dutch professional association of speech therapists for people with intellectual disabilities.

### Low bone quality

In the general population, screening for osteoporosis is not recommended because the effectiveness of bisphosphonates to prevent fractures in this population has not been established, and because the reliability of available diagnostic methods for screening is insufficient [23]. Secondary prevention of fractures in the general population is only indicated in people with an increased fracture risk [24]. An increased fracture risk is

based on previous fractures and/or a positive score in a risk table (age ≥ 60 years, BMI < 20 kg/m2 , two or more falls in the past year, a parent with a hip fracture [24]).

We have shown that the prevalence of low bone quality in older people with ID is high, as measured by QUS. However, as mentioned above, we have recently found that the validity of QUS for predicting fractures in people with ID is insufficient [18]. Therefore, in our opinion, at this moment, QUS cannot be recommended for screening of low bone quality in people with ID. Currently, our opinion is that addition of antiepileptic drug use and longstanding mobility impairment to the risk table in the guideline of the Dutch College of general practitioners [24] might be sufficient to identify people with ID with a high fracture risk. In general, more attention to primary and secondary prevention of low bone quality in this population is of paramount importance. In line with general population recommendations [25], every person with an intellectual disability aged 50 years and over should take vitamin D supplementation. An intake of 4 dairy products per day is recommended as well, as is exposure of sunlight during at least 30 minutes a day.

### Sarcopenia

Recommended prevention of sarcopenia usually consists of nutritional interventions (calories and proteins) and resistance training programs to improve muscle mass. Moreover, these interventions are likely to be beneficial as well for prevention of associated conditions, like osteoporosis [26, 27].

In addition to prevention (or treatment) of sarcopenia, we recommend to start with monitoring muscle mass, muscle strength and muscle performance in all older people with intellectual disabilities [28]. Because, in line with national guidelines [29-31], most of them should be checked annually anyway, because of either cardiovascular risk factors (central obesity 46%, hypertension 53%, diabetes mellitus 14%, hypercholesterolaemia 23%, metabolic syndrome 45% [10, 32]), or epilepsy (22% [33]), or antipsychotic drug use (29% [33]), measurements of muscle mass (calf circumference), strength (grip strength) and performance (5-meter walk test) could be combined with this check-up. In case of obvious decline in muscle mass, strength or performance, extra protein supplementation (e.g. β-hydroxy-β-methylbutyrate) could be added to the diet of the individual [34].

## RECOMMENDATIONS FOR RESEARCH

### Food intake

According to Dutch national guidelines, the recommended dietary allowance for protein, which is the minimal intake necessary for health reasons, is 1.2 g/kg per day

for people aged 65 years and over and 0.8 g/kg per day for people aged 50-64 years [35, 36]. However, we have shown that sarcopenia risk is already present in the age group 50-64 years, meaning that this group should increase their protein intake, too. Therefore, we recommend to set up a randomized controlled trial to study the health effects of a higher protein intake (combined with resistance training programs) in people with intellectual disabilities aged 50-64 years.

### Nutritional state

We have shown that the MNA is insufficiently valid for use in people with ID. Modification of the MNA specifically for this population seems a logical first step. ID-specific disorders related to malnutrition and level of ID could be added, whereas some items could be adapted. Subsequently, a new validation study with a nutritional assessment performed by clinical experts as a gold standard should be done.

### Health issues related to a poor nutritional state

We have recommendations for further research regarding dysphagia, low bone quality and sarcopenia. Firstly, we recommend more research regarding the validity of the Dysphagia Disorder Survey (DDS). The DDS was developed specifically for screening of adults with intellectual disabilities for dysphagia. However, it is unknown whether the DDS is suitable to identify persons with a high risk of aspiration, in particular silent aspiration. Therefore, we recommend to study the sensitivity and specificity of the DDS, by comparing its outcomes with videofluoroscopic swallowing assessment, which is the gold standard for diagnosing aspiration.

Secondly, we have recently found that bone quality as measured by QUS was not an independent predictor for fracture occurrence [18]. However, in this validation study, people using a wheelchair were underrepresented and the follow-up was relatively short. Moreover, the study population was relatively small (547 participants [18]). Because of the feasibility of the QUS of the heel bone, the disadvantages of DXA for people with mobility impairment (lying completely still), we recommend setting up a validation study for the QUS measurement in a large representative population of older people with ID with a follow-up of at least 5-10 years. After validation of the QUS among people with ID, its role in clinical practice should be considered again.

Thirdly, in the general population, sarcopenia is associated with negative health outcomes such as physical disability and morbidity [3, 37]. At this moment, that association has never been studied in people with ID. Because of the relatively high prevalence of sarcopenia in people with ID aged 50-64 years, we recommend longitudinal research to study the relation between sarcopenia and negative outcomes in that population. Furthermore, in people with ID, sarcopenia may be present at a young age due to mobility impairment. The pathophysiology of sarcopenia in this specific group

may be different from sarcopenia among older persons in the general population. Indeed, increased levels of inflammatory cytokines are seen in older people in the general population. These cytokines are directly associated with muscle mass loss and muscle strength reduction in that population [38]. It is uncertain whether inflammation plays also a role in sarcopenia among people with lifelong mobility impairment. Therefore, we recommend fundamental research towards better understanding the pathophysiology of sarcopenia in people with ID and mobility impairment.

## PROMOTING HEALTHY FOOD INTAKE

In general, this thesis shows that both a poor nutritional status, especially underweight, and mobility impairment are linked to several age-related disorders and diseases. Therefore, people with ID and a low BMI should periodically be assessed by general practitioners or physicians for people with ID, by dieticians and other professional therapists. Secondly, it has been shown in this thesis that the nutritional intake of older people with ID is insufficient. Therefore, a national campaign and/or care improvement projects should be launched to promote healthy food intake among people with ID. Dietary interventions, based on a multi-modal approach, should be implemented in both living facilities and day activity centres. Because of positive outcomes in a previous study on social inclusion through unified sports (players with and without intellectual disability in the same team [39]), individuals with ID could be coupled to a peer without an intellectual disability to gain or to lose weight.

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# Summary

# SUMMARY

## Chapter 1 General introduction

In 2008 a consortium was founded, consisting of three care provider services for people with intellectual disabilities (Abrona, Amarant and Ipse de Bruggen) and the department of intellectual disability medicine of the ErasmusMC, University Medical Center Rotterdam. The main goals of this consortium were to increase knowledge on healthy ageing in people with intellectual disabilities by means of scientific research, to increase the expertise and scientific attitude of staff by means of participation in research and continuous education and innovation of care by means of implementation of research outcomes. The theme of healthy ageing was chosen because life expectancy in people with intellectual disabilities has increased in the last decades. As a consequence, the number of older people with intellectual disabilities has risen. However, health in this population was never studied before on a large scale.

The first step in reaching the scientific goals of the consortium has been made by starting a large-scale, cross-sectional epidemiological study, titled 'Healthy Ageing and Intellectual Disabilities (HA-ID)'. In this study three subthemes were investigated: (1) depression and anxiety, (2) physical activity and fitness and (3) nutrition and nutritional status. After a description of the design and the recruitment of the HA-ID study, this thesis presents results of the theme nutrition and nutritional state. More precisely, food intake, nutritional state and negative health outcomes due to undernutrition are described.

## Chapter 2 Study Healthy Ageing and Intellectual Disabilities: recruitment and design

The cross-sectional study 'Healthy Ageing and Intellectual Disabilities (HA-ID)' started in 2008 to increase knowledge on health in older people with intellectual disabilities. Preparation of the study was started by carefully reviewing and selecting instruments to measure a wide set of health variables to answer the research questions on three themes. Specific demands of these instruments were that they could be executed efficiently and accurately on-site in a large sample of participants and that the burden of these measurements for participants as well as their caregivers was as minimal as possible. Subsequently, preparation was continued by designing and executing a thorough communication plan for clients, legal representatives and staff of the care providers, preceding the informed consent procedure. In this plan, which had a top-down structure, specific attention was given to personally informing and motivating the professional caregivers. This preparation led to a recruitment of 1050 out of 2322 participants (45.2%) and to high participation rates in key parts of the assessment.

A detailed description is provided about the recruitment and organization and the selected instruments.

### Chapter 3 Inadequate dietary intake in older people with intellectual disabilities: results of the HA-ID study

For the subtheme 'nutrition and nutritional state' we have measured food intake of the participants of the HA-ID study. We were especially interested in intake of energy and proteins because of the high prevalence of protein energy malnutrition in the general ageing population, intake of fat because of the key role of restricted (saturated) fat intake in preventing cardiovascular diseases, and intake of fiber because of the high prevalence of constipation in people with ID, given the essential role of fiber in the development of constipation. The actual dietary intake of the participants was compared with recommendations for healthy food intake in accordance with national guidelines. To measure food intake a 3-day food frequency list consisting of a printed list of food and fluid products including usual portion sizes was designed. Because completion of food frequency lists by professional caregivers and processing the data by dieticians was very time-consuming, filling out the food frequency lists was only feasible for 228 participants of the HA-ID study population.

The food frequency lists have shown that most of the participants poorly match the daily recommendations. With 82%, most of the participants have satisfied the recommendations for total fat intake. However, 30% of the participants, especially those with a higher age and a less severe level of ID, eat insufficient amounts of proteins, and 98% eat insufficient amounts of fiber. Compared to the general population in the Netherlands aged 50 to 70 years, the median energy, protein and fibre intake of older people with ID are lower. Therefore, healthy food should be high on the agenda of policymakers and more knowledge about healthy nutrition and the harmful effects of an unhealthy diet is necessary for professional caregivers and people with intellectual disabilities.

### Chapter 4 Dysphagia in older people with intellectual disabilities: results of the HA-ID study

We have studied swallowing disorders (dysphagia) in the HA-ID study, because it may lead to decreased food intake. We have investigated the prevalence of dysphagia within the population of older people with ID and its associations with gender, age, level of ID, Down syndrome, residential status, mobility, spasticity, dependency on feeding, body mass index, stroke, dementia, and centrally acting medication use. Because of the occurrence of unanticipated swallowing incidents in clinical practice, the awareness of dysphagia amongst professionals was also investigated by comparing the study outcomes with registration of swallowing disorders in medical files.

Swallowing function was observed by specifically trained speech and language therapists or dieticians. They used the Dysphagia Disorder Survey (DDS), a standardized mealtime observation developed for people with ID. According to the DDS, 52% of the 931 participants had moderate to severe dysphagia. This prevalence is comparable to the prevalence found in nursing home residents. Dysphagia was associated with older age, mobility impairment, feeding dependency, Down syndrome, and use of benzodiazepines. In 89.5% of the study population with dysphagia, swallowing problems had not been registered in medical files, meaning that recognition of dysphagia in clinical practice is poor. Therefore, professional caregivers working with people with ID should be educated on risk symptoms and more attention is needed for screening risk groups for dysphagia.

### Chapter 5 Feasibility and reliability of the Mini Nutritional Assessment (MNA) in older adults with intellectual disabilities

Although evidence is currently lacking, it might be assumed that the prevalence of malnutrition in older people with ID is higher than in the general ageing population, because of a combination of age-related and ID-related risk factors for malnutrition. In the general population, several screening instruments are used for detecting malnutrition. The Mini nutritional assessment (MNA), is internationally widely used and validated for use in healthy and frail older adults. It may be suitable for ageing people with ID as well. However, the psychometric properties of the MNA have not been evaluated for this population. Therefore, we investigated the feasibility and the reliability of the MNA in older clients of ID care providers. 48 caregivers of participants with all levels of ID and 12 participants with a mild ID were interviewed and 47 participants underwent a physical examination. To test feasibility an evaluation form was administered by caregivers and to test reliability intraclass correlation coefficients (ICC) for test-retest and inter-observer reliability were calculated.

The evaluation forms have shown that it was easy for caregivers to answer at least 13 of the 15 items (85%) of the MNA. Sometimes it was hard for them to correctly answer the questions about mealtimes of the participants because of the day-time activities of the participants at other places. The mean duration of the interview was 4 minutes. ICCs for test-retest and inter-observer reliability between caregivers were good, 0.85 and 0.86 respectively, but ICC for inter-observer reliability between caregivers and persons with intellectual disabilities was low, 0.03. Therefore, we have concluded that the MNA is feasible and reliable for older people with intellectual disabilities. Interview data can be reliably obtained through caregivers, but not through people with ID.

## Chapter 6 Evaluation of the Mini Nutritional Assessment in older people with intellectual disabilities

After studying the feasibility and the reliability of the MNA, we aimed to investigate its construct and criterion validity in older adults with intellectual disabilities. For this purpose the MNA was completed for 835 persons aged 50 years and over with borderline to profound ID. Construct validity was determined by correlating the individual items and the total score of the MNA with the four parameters of a consensus-based nutritional state index. To investigate criterion validity, tests of sensitivity and specificity were performed to compare the outcomes of the MNA with the consensus-based nutritional state index. The construct validity was low, with Spearman's correlation coefficients varying from $r = 0.17$ to $r = 0.29$. Compared with the nutritional state index, sensitivity of the MNA was 46.4% and specificity was 60.8%. We have conclude that the MNA seems to be insufficiently valid for use in people with ID in the current format. Therefore, for older people with ID, modification of the MNA is needed. ID-specific disorders related to malnutrition and level of ID could be added and some items could be better adapted to people with ID. After this modification, a new validation study with an extended nutritional assessment done by clinical experts as a gold standard is necessary.

## Chapter 7 Observed vitamin D deficiency variations in older adults with intellectual disabilities

Because life expectancy of people with intellectual disabilities is increasing gradually and sufficient serum levels of vitamin D are important for multiple age-related health issues, we were interested in the prevalence of vitamin D deficiency and its associations in this population. Therefore, serum was obtained from 618 persons with borderline to profound intellectual disabilities aged 50 years and over. The prevalence rates of vitamin D deficiency and severe deficiency (serum 25(OH)D3 level below 50 nmol/l and 25 nmol respectively) were studied. To identify its associations, logistic regression analyses were performed with gender, age, level of intellectual disability, residential status, Down syndrome, mobility, mean number of steps per day, number of falls, grip strength, body mass index, bone quality, depressive symptoms, major depression, dementia, anticonvulsant drug use, serum calcium levels and suppletion of vitamin D. We found a prevalence rate of 42% for vitamin D deficiency and a prevalence rate of 9% for severe deficiency. Vitamin D suppletion was being routinely provided to 45% of the population. This group had significantly higher mean vitamin D serum levels than those not using suppletion. Apart from suppletion, vitamin D deficiency was significantly associated with lower serum calcium levels and higher BMI-scores. Because of the high prevalence rate of vitamin D deficiency, routine vitamin D substitution in all adults with intellectual disabilities aged 50 years and over is

necessary. The association of BMI with vitamin D deficiency stresses even more the relevance of healthy food intake and physical activity in this population.

### Chapter 8 Bone quality in older adults with intellectual disabilities

Although osteoporosis is a progressive bone disease leading to increased risk of fractures, it has rarely been investigated on a large scale in older people with intellectual disabilities. In the HA-ID study, the stiffness index of the heel bone, as a measure for bone quality, was assessed by using quantitative ultrasound. 768 persons with borderline to profound intellectual disabilities, aged 50 years and over were measured. Also the associations of low bone quality were investigated: gender, age, level of intellectual disability, residential status, Down syndrome, mobility, physical activity, body mass index, prior fractures, anticonvulsant drug use, intake of calcium, and vitamin D serum levels. The prevalence of low bone quality was 43.9%. Low bone quality was positively associated with female gender, age, more severe level of intellectual disability, mobility impairment, and anticonvulsant drug use, and negatively with body mass index. Although quantitative ultrasound measurement has not been validated to determine low bone quality in people with intellectual disabilities, the overlap in independent predictors in studies in the general population may be indicative of its adequate construct validity in people with intellectual disabilities. In clinical practice, people with intellectual disabilities who are at risk for low bone quality should periodically be screened for osteoporosis and be given advice about nutritional supplements and appropriate lifestyle.

### Chapter 9 Prevalence and associated factors of sarcopenia in older adults with intellectual disabilities

Sarcopenia is defined as a syndrome characterised by progressive and generalised loss of skeletal muscle mass and strength. It has not been studied before in older people with intellectual disabilities. Because of its association with negative health outcomes, such as loss of independence, in the general older population, the prevalence of sarcopenia and its associated factors were investigated in the HA-ID study. 884 persons with borderline to profound intellectual disabilities aged 50 years and over, were investigated. Calf circumference was measured to determine muscle mass, grip strength to determine muscle strength, and walking speed to determine muscle performance. To identify the associations of sarcopenia, logistic regression analyses were performed with gender, age, level of intellectual disability, residential status, Down syndrome, mobility, physical activity, intake of energy and proteins, body mass index and levels of CRP, albumin and vitamin D in serum. The prevalence of sarcopenia was 14.3% in the total group. In the age group 50–64 years prevalence was 12.7%. Sarcopenia was positively associated with mobility impairment and inflammation and

negatively with body mass index. This first study into the prevalence of sarcopenia in older people with intellectual disabilities stresses the need to collect longitudinal data to study the relation between sarcopenia and negative outcomes in older people with intellectual disabilities.

### Chapter 10 General discussion

Despite some methodological issues, this thesis shows that the nutritional intake of older people with ID is insufficient, making these persons more vulnerable for developing diseases. Unhealthy food intake may play an important role in developing early sarcopenia and frailty, which are both conditions predicting (un)healthy ageing in the general population. Therefore, healthy food should be high on the agenda of policymakers.

Conditions related to a poor nutritional status, like dysphagia, low bone quality and sarcopenia are common in older people with ID, especially those with a low BMI or mobility impairment. Therefore, more attention is needed to prevention of and/or screening for these conditions in clinical practice.

The Mini Nutritional Assessment, an internationally widely used screening tool for malnutrition, appeared insufficiently valid in people with ID. Therefore, screening for malnutrition in this population is not easily possible at this moment, hampering timely detection of this condition. Because of the negative consequences of malnutrition, the development of a valid screening tool for this population is urgent.

# Samenvatting

# SAMENVATTING

## Hoofdstuk 1 Algemene inleiding

In 2008 werd een consortium opgericht, bestaande uit drie zorgaanbieders voor mensen met een verstandelijke beperking (Abrona, Amarant en Ipse de Bruggen) en de afdeling Geneeskunde voor Verstandelijk Gehandicapten van het Erasmus Medisch Centrum Rotterdam. De belangrijkste doelstellingen van dit consortium waren: (1) kennis vermeerderen van de gezondheid van ouderen met een verstandelijke beperking door middel van wetenschappelijk onderzoek, (2) verhogen van expertise en wetenschappelijke houding van medewerkers van zorgorganisaties door middel van deelname aan wetenschappelijk onderzoek en (3) innovatie van de zorg door middel van implementatie van onderzoeksresultaten. Als centraal thema werd 'gezond ouder worden' gekozen omdat de levensverwachting van mensen met een verstandelijke beperking de laatste decennia is toegenomen met als gevolg dat het aantal oudere mensen met een verstandelijke beperking is gestegen. Echter, gezondheid in deze populatie werd nooit op grote schaal onderzocht.

De eerste stap in het bereiken van de wetenschappelijke doelstellingen van het consortium is gemaakt door het starten van een grootschalige epidemiologische studie, getiteld 'Gezond OUDer met een verstandelijke beperking (GOUD)'. In deze studie werden drie subthema's onderzocht: (1) depressie en angst, (2) fysieke activiteit en fitheid en (3) voeding en voedingstoestand. Na een beschrijving van de opzet van de GOUD studie, zijn in dit proefschrift de resultaten van het thema voeding en voedingstoestand beschreven. In detail worden voedselinname, voedingstoestand en negatieve gezondheidseffecten als gevolg van ondervoeding beschreven.

## Hoofdstuk 2 'Gezond OUDer met een verstandelijke beperking': opzet van de studie

De epidemiologische studie 'Gezond OUDer met een verstandelijke beperking (GOUD)' is in 2008 gestart met als doel om meer kennis te verkrijgen over de gezondheid van ouderen met een verstandelijke beperking. Ter voorbereiding op het onderzoek werden bestaande meetinstrumenten zorgvuldig geëvalueerd en geselecteerd om een brede reeks van gezondheidsvariabelen te kunnen meten. De meetinstrumenten moesten voldoen aan de volgende eisen: (1) efficiënt en nauwkeurig, (2) uitvoering mogelijk op locatie bij het grootste deel van de deelnemers en (3) minimale belasting voor deelnemers en hun begeleiders. De volgende stap in de voorbereiding was het opstellen en uitvoeren van een gedegen communicatieplan voor cliënten, wettelijk vertegenwoordigers en medewerkers van de zorgaanbieders. In dit communicatieplan, dat een top-down structuur had, werd specifiek aandacht besteed aan het persoonlijk informeren en motiveren van de woonbegeleiders van de deelnemers. Dit alles heeft

geleid tot een inclusie van 1050 van de in totaal 2322 deelnemers (45,2%) en een hoog deelnemerspercentage in de verschillende delen van het onderzoek. In dit hoofdstuk wordt een gedetailleerde beschrijving gegeven over de gekozen meetinstrumenten, de werving van de deelnemers en de organisatie van de GOUD-studie.

### Hoofdstuk 3 Onvoldoende voedselinname door oudere mensen met een verstandelijke beperking

Voor het subthema 'voeding en voedingstoestand' hebben we de voedselinname van de deelnemers van de GOUD-studie gemeten. We waren vooral geïnteresseerd in de inname van energie en eiwitten vanwege de hoge prevalentie van ondervoeding in de groep ouderen zonder verstandelijke beperking, de inname van vet vanwege de essentiële rol van verzadigd vet bij het ontstaan van hart- en vaatziekten en de inname van vezels, gezien de essentiële rol van vezels in de ontwikkeling van obstipatie en het veelvuldig voorkomen van deze aandoening bij ouderen met een verstandelijke beperking. De werkelijke voedselinname van de deelnemers werd vergeleken met de aanbevelingen voor gezonde voedselinname volgens richtlijnen van de Gezondheidsraad. Voedselinname werd gemeten door het invullen van een 3-daagse voedselfrequentielijst bestaande uit een lijst van eet- en drinkproducten en gebruikelijke portiegroottes. Omdat het invullen van de voedselfrequentielijsten door begeleiders en de verwerking van de gegevens door diëtisten zeer tijdrovend was, bleek het invullen van de voedselfrequentielijsten slechts haalbaar voor 228 deelnemers van de GOUD-populatie.

De voedselfrequentielijsten hebben aangetoond dat de meeste deelnemers niet voldoen aan de aanbevolen dagelijkse hoeveelheden. Het merendeel van de deelnemers (82%) voldoet weliswaar aan de aanbevelingen voor totale vetinname, maar slechts 30% van de deelnemers, vooral die met een hogere leeftijd en een minder ernstige mate van verstandelijke beperking, eet onvoldoende eiwitten en 98% van de deelnemers eet onvoldoende vezels. In vergelijking met de algemene bevolking in Nederland van 50 tot 70 jaar is de inname van energie, eiwitten en vezels door oudere mensen met een verstandelijke beperking lager. Daarom moet gezonde voeding hoog op de agenda komen van beleidsmedewerkers en managers van zorgorganisaties en moet er meer aandacht komen voor kennisvermeerdering over gezonde voeding en de schadelijke effecten van ongezonde voeding voor begeleiders van mensen met een verstandelijke beperking en mensen met een verstandelijke beperking zelf.

### Hoofdstuk 4 Dysfagie bij oudere mensen met een verstandelijke beperking

In de GOUD-studie hebben we slikstoornissen onderzocht omdat slikstoornissen gerelateerd zijn aan verminderde voedselinname en ondervoeding. Zowel de prevalentie van

slikstoornissen als diens associaties met geslacht, leeftijd, niveau van verstandelijke beperking, syndroom van Down, woonsetting, mobiliteit, spasticiteit, afhankelijkheid van anderen met betrekking tot eten geven, body mass index, beroerte, dementie en gebruik van antipsychotica, anti-epileptica en/of langwerkende benzodiazepines zijn onderzocht. Ook is het herkennen van slikstoornissen door professionele zorgverleners onderzocht door het vergelijken van de aanwezigheid van slikstoornissen onder de deelnemers van de GOUD-studie met de registratie ervan in de medische dossiers van de deelnemers.

Slikstoornissen werden onderzocht door speciaal opgeleide logopedisten en diëtisten. Als meetinstrument gebruikten zij de Dysphagia Disorder Survey (DDS), een gestandaardiseerde maaltijdobservatie ontwikkeld voor mensen met een verstandelijke beperking. Volgens de DDS heeft 52% van de 931 deelnemers een matige tot ernstige slikstoornis. Dit percentage is vergelijkbaar met de prevalentie van slikstoornissen onder verpleeghuisbewoners. Slikstoornissen zijn geassocieerd met oudere leeftijd, beperkte mobiliteit, afhankelijkheid van anderen met betrekking tot eten geven, het syndroom van Down en het gebruik van langwerkende benzodiazepines. In 89.5% van de studiepopulatie met dysfagie stonden slikstoornissen niet geregistreerd in hun medische dossier, duidend op een ondermaatse herkenning van slikstoornissen in de praktijk. Daarom is er meer aandacht nodig voor scholing van begeleiders op dit gebied. Vanwege de hoge prevalentie van slikstoornissen bij mensen met een verstandelijke beperking wordt daarnaast ook screening van risicogroepen aanbevolen.

### Hoofdstuk 5 Toepasbaarheid en betrouwbaarheid van de Mini Nutritional Assessment bij mensen met een verstandelijke beperking

Hoewel wetenschappelijk bewijs ontbreekt, mag verondersteld worden dat de prevalentie van ondervoeding bij oudere mensen met een verstandelijke beperking hoger is dan in de algemene populatie ouderen, als gevolg van een combinatie van aan leeftijd gerelateerde en aan verstandelijke beperking gerelateerde risicofactoren voor ondervoeding. In de algemene bevolking worden verschillende screeningsinstrumenten gebruikt voor het opsporen van ondervoeding. De Mini Nutritional Assessment (MNA) is zo'n instrument. Het wordt internationaal veel gebruikt en het is gevalideerd voor gebruik in gezonde en kwetsbare oudere populaties. De MNA zou ook geschikt kunnen zijn voor gebruik bij ouderen met een verstandelijke beperking. De psychometrische eigenschappen zijn echter niet eerder onderzocht in deze populatie. Daarom hebben wij onderzoek gedaan naar de toepasbaarheid en de betrouwbaarheid van de MNA bij oudere cliënten die zorg ontvangen van zorgorganisaties voor mensen met een verstandelijke beperking. 48 begeleiders en 12 cliënten met een lichte tot matige verstandelijke beperking werden hiervoor geïnterviewd en 47 cliënten ondergingen een lichamelijk onderzoek. Om de toepasbaarheid te testen werd een evaluatieformulier

ingevuld door begeleiders en voor de betrouwbaarheid werden intraclass correlatiecoëfficiënten (ICC) voor de test-hertest en inter-observer betrouwbaarheid berekend.

De evaluatieformulieren hebben aangetoond dat het gemakkelijk was voor begeleiders om de vragen van de MNA te beantwoorden. Soms was het weliswaar moeilijk voor hen om de vragen over de maaltijden van de deelnemers te beantwoorden vanwege de dagbesteding van de deelnemers op andere plaatsen, maar allemaal vonden ze dat ze tenminste 13 van de 15 items betrouwbaar konden beantwoorden. De gemiddelde duur van het interview was 4 minuten. De MNA is dus toepasbaar gebleken. Voor wat betreft betrouwbaarheid waren de ICC's voor test-hertest en inter-observer betrouwbaarheid tussen twee zorgverleners goed, 0,85 en 0,86 respectievelijk. De ICC voor inter-observer betrouwbaarheid tussen zorgverleners en mensen met een verstandelijke beperking was laag (0,03). Daarom hebben we geconcludeerd dat de MNA betrouwbaar is voor gebruik in een populatie van oudere mensen met een verstandelijke beperking. De items moeten echter wel ingevuld worden door begeleiders, niet door de mensen met een verstandelijke beperking zelf.

## Hoofdstuk 6 Validiteit van de MNA bij oudere mensen met een verstandelijke beperking

Na het bestuderen van de toepasbaarheid en de betrouwbaarheid van de MNA, hebben we de constructvaliditeit en de criteriumvaliditeit bij ouderen met een verstandelijke beperking onderzocht. Hiervoor werd de MNA ingevuld door begeleiders van 835 personen van 50 jaar en ouder met een lichte tot zeer ernstige verstandelijke beperking. De constructvaliditeit werd bepaald door het correleren van de individuele items en de totale score van de MNA met de vier parameters van een op consensus gebaseerde voedingstoestand-index. De criteriumvaliditeit werd bepaald door sensitiviteit en specificiteit te berekenen door de uitkomsten van de MNA af te zetten tegen de uitkomsten van dezelfde op consensus gebaseerde voedingstoestand-index.

De constructvaliditeit was laag, met Spearman's correlatie coëfficiënten variërend van r = 0,17 tot r = 0,29. De sensitiviteit van de MNA was 46,4% en de specificiteit 60,8%. We moeten daarom concluderen dat de validiteit van de MNA in zijn huidige format onvoldoende is voor gebruik bij mensen met een verstandelijke beperking. Voor (oudere) mensen met een verstandelijke beperking is dus een aanpassing van de MNA noodzakelijk. Aandoeningen specifiek voor mensen met een verstandelijke beperking die gerelateerd zijn aan ondervoeding en het niveau van de verstandelijke beperking zouden aan de items van de MNA kunnen worden toegevoegd zodat deze screeningslijst beter aansluit bij de doelgroep. Uiteraard moet, voorafgaand aan toepassing in de praktijk, een nieuwe versie van de MNA gevalideerd worden waarbij de MNA wordt afgezet tegen een uitgebreide evaluatie van de voedingstoestand door experts.

## Hoofdstuk 7 Prevalentie en gerelateerde factoren van vitamine D deficiëntie bij oudere mensen met een verstandelijke beperking

Omdat de levensverwachting van mensen met een verstandelijke beperking geleidelijk toeneemt en omdat vitamine D belangrijk is voor diverse leeftijdsgerelateerde aandoeningen, hebben we de prevalentie van vitamine D deficiëntie en ernstige vitamine D deficiëntie (serum 25(OH)D3 niveau lager dan 50 nmol/l, respectievelijk 25 nmol/l) in de GOUD-populatie onderzocht. Hiervoor werd bloed afgenomen bij 618 personen met een verstandelijke beperking van 50 jaar en ouder. Ook werden de mogelijke associaties van vitamine D deficiëntie (geslacht, leeftijd, mate van verstandelijke beperking, woonsetting, syndroom van Down, mobiliteit, gemiddeld aantal stappen per dag, aantal valincidenten, knijpkracht, body mass index, botkwaliteit, depressieve symptomen, depressie, dementie, anti-epileptica gebruik, serum calcium en vitamine D suppletie) onderzocht middels logistische regressie.

We vonden een prevalentie van 42% voor vitamine D deficiëntie en een prevalentie van 9% voor ernstige vitamine D deficiëntie. 45% van de onderzochte personen nam vitamine D suppletie. Deze groep had een significant hogere vitamine D spiegel vergeleken met de groep zonder suppletie. Naast suppletie, was vitamine D deficiëntie significant geassocieerd met lagere calciumspiegels en hogere BMI-waardes.

Deze studie heeft de noodzaak voor routinematig suppleren van vitamine D bij ouderen met een verstandelijke beperking aangetoond. De associatie met hoge BMI-waardes benadrukt nog meer het belang van een gezonde voedselinname en voldoende beweging in deze populatie.

## Hoofdstuk 8 Botkwaliteit bij ouderen met een verstandelijke beperking

Hoewel osteoporose een progressieve aandoening is die leidt tot een verhoogd risico op fracturen, is deze aandoening zelden op grote schaal onderzocht bij oudere mensen met een verstandelijke beperking. In de GOUD-studie hebben we botkwaliteit onderzocht door middel van kwantitatieve echografie van het hielbot. 768 personen met een lichte tot zeer ernstige verstandelijke beperking van 50 jaar en ouder hebben meegedaan aan deze deelstudie. Behalve de botkwaliteit zijn ook de mogelijke associaties van lage botkwaliteit onderzocht: geslacht, leeftijd, mate van verstandelijke beperking, woonsetting, syndroom van Down, mobiliteit, lichamelijke activiteit, body mass index, eerdere fracturen, anti-epileptica gebruik, inname van calcium en vitamine D spiegels.

De prevalentie van lage botkwaliteit was 43,9%. Lage botkwaliteit was positief geassocieerd met vrouwelijk geslacht, leeftijd, ernstiger mate van verstandelijke beperking, beperkte mobiliteit en anti-epileptica gebruik, en negatief geassocieerd met body mass index. Hoewel kwantitatieve echografie niet is gevalideerd voor lage

botkwaliteit bij mensen met een verstandelijke beperking, kan de overlap met onafhankelijke voorspellers in studies in de algemene bevolking een indicatie zijn voor adequate constructvaliditeit bij mensen met een verstandelijke beperking. Voor de klinische praktijk wordt aanbevolen om mensen met een verstandelijke beperking met een verhoogd risico op lage botkwaliteit regelmatig te screenen. Daarnaast moeten zij adviezen krijgen over een gezonde leefstijl en eventuele calcium- en vitamine D supplementen.

### Hoofdstuk 9 Prevalentie en gerelateerde factoren van sarcopenie bij oudere mensen met een verstandelijke beperking

Sarcopenie wordt gedefinieerd als een syndroom dat gekenmerkt wordt door een progressief en gegeneraliseerd verlies van spiermassa en spierkracht. Het is niet eerder onderzocht bij ouderen met een verstandelijke beperking. Vanwege de associatie met negatieve gevolgen voor de gezondheid in de algemene oudere bevolking, zoals het verlies van zelfstandigheid, hebben we de prevalentie van sarcopenie en diens associaties onderzocht in de GOUD-studie. Aan dit deelonderzoek hebben 884 personen van de GOUD-populatie meegedaan. Bij hen werd de kuitomtrek gemeten als maat voor spiermassa, de knijpkracht bepaald als maat voor spierkracht en de loopsnelheid gemeten als maat voor spierfunctie. Logistische regressie werd gebruikt om de associaties tussen sarcopenie enerzijds en geslacht, leeftijd, mate van verstandelijke beperking, woonsetting, syndroom van Down, mobiliteit, lichamelijke activiteit, inname van energie en eiwitten, body mass index en serumspiegels van CRP, albumine en vitamine D anderzijds, te onderzoeken.

De prevalentie van sarcopenie was 14,3% in de totale groep. In de leeftijdsgroep 50-64 jaar was de prevalentie 12,7%. Sarcopenie was positief geassocieerd met beperkte mobiliteit en ontsteking en negatief geassocieerd met body mass index. Voor vervolgonderzoek zou het zinvol zijn longitudinale gegevens te verzamelen bij ouderen mensen met een verstandelijke beperking om de relatie tussen sarcopenie en negatieve gezondheidseffecten te bestuderen voor deze doelgroep.

### Hoofdstuk 10 Algemene discussie

Dit proefschrift laat zien dat de voedselinname van oudere mensen met een verstandelijke beperking onvoldoende is, waardoor zij kwetsbaarder zijn voor het ontwikkelen van ziekten. Ongezonde voedselinname kan een belangrijke rol spelen bij de ontwikkeling van vroege sarcopenie en kwetsbaarheid, die beide voorspellers zijn van (on)gezond ouder worden in de algemene bevolking. Daarom moet gezonde voeding hoog op de agenda komen te staan van managers en beleidsmakers in de verstandelijke gehandicapten sector.

Aandoeningen gerelateerd aan een slechte voedingstoestand, zoals dysfagie, lage botkwaliteit en sarcopenie komen vaak voor bij oudere mensen met een verstandelijke beperking, vooral die met een lage BMI of beperkte mobiliteit. Daarom is er meer aandacht nodig voor preventie en/of screening van deze aandoeningen in de klinische praktijk.

De Mini Nutritional Assessment, een internationaal veel gebruikt screeningsinstrument voor ondervoeding, blijkt niet valide voor toepassing bij mensen met een verstandelijke beperking. Daarom is screening op ondervoeding op dit moment niet goed mogelijk in deze populatie. Dit belemmert tijdige detectie van deze aandoening. Omdat ondervoeding kan leiden tot allerlei gezondheidskwalen is ontwikkeling van een geldig meetinstrument voor deze populatie zeer belangrijk.

# Dankwoord

## DANKWOORD

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En tenslotte natuurlijk Mariëtte, Emma en Pim. Jullie zijn mijn alles. Emma en Pim, jullie hebben voor heel veel afleiding gezorgd in deze drukke tijd en wat voor een afleiding! Ik ben echt superblij met jullie om wie jullie zijn. Mar, ik heb behoorlijk wat flexibiliteit van je gevraagd de afgelopen jaren. En die flexibiliteit heb je ook altijd getoond. Het is heerlijk om een stabiele thuisbasis te hebben. Ik hou van je. XXX.

# About the author

## CURRICULUM VITAE

Luc Bastiaanse is op 17 juli 1974 geboren in Bergen op Zoom. Hij is opgegroeid in Halsteren en volgde zijn middelbare schoolopleiding aan het Mollerlyceum in Bergen op Zoom. In 1992 behaalde hij zijn VWO-diploma en aansluitend is hij geneeskunde gaan studeren aan de Erasmus Universiteit in Rotterdam. Zijn afstudeeronderzoek heeft hij gedaan op de afdeling medische microbiologie naar het vóórkomen van cryptosporidium-infecties bij mensen met een HIV-infectie. In 1999 behaalde hij zijn artsexamen. Hierna is hij als arts-assistent gaan werken in het Drechtsteden ziekenhuis, locatie Refaja in Dordrecht op de afdelingen chirurgie en gynaecologie in het kader van de tropenopleiding. In 2001 maakte hij de overstap naar de medische zorg voor mensen met een verstandelijke beperking. Ter voorbereiding op de opleiding voor Artsen voor Verstandelijk Gehandicapten (AVG) aan het Erasmus Medisch Centrum (Erasmus MC) in Rotterdam heeft hij nog een paar maanden gewerkt op de afdeling neurologie van het Holy ziekenhuis in Vlaardingen. In oktober 2001 is hij bij Ipse de Bruggen in Nootdorp gaan werken, van 2001 tot 2004 als AVG in opleiding (opleider Joop van den Berg) en vanaf december 2004 tot heden als AVG. Behalve bij Ipse de Bruggen heeft hij in 2005-2006 op detacheringsbasis gewerkt bij Humanitas DMH in Barendrecht en in 2010-2011 bij Orion Camphill in Rotterdam. Van 2004 tot 2010 bekleedde hij een bestuursfunctie bij de Nederlandse Vereniging van Artsen voor Verstandelijk Gehandicapten (NVAVG).

Behalve als praktiserend AVG werkt Luc vanuit Ipse de Bruggen sinds 2008 ook als onderzoeker aan het onderzoek 'Gezond OUDer met een verstandelijke beperking (GOUD)'. Dit onderzoek is een samenwerkingsverband tussen drie zorgaanbieders (Abrona, Amarant en Ipse de Bruggen) en het Erasmus MC. Hij heeft zich toegelegd op het subthema voeding en voedingstoestand. De resultaten van dit onderzoek staan beschreven in dit proefschrift. Voortbordurend op dit onderzoek, houdt hij zich op dit moment bezig met het zorgverbeteringsproject slikstoornissen binnen het GOUD-consortium waarvoor een subsidie van het Fonds Verstandelijk Gehandicapten is toegekend.

Luc is getrouwd met Mariëtte Schenkel en samen hebben ze twee kinderen: Emma (2005) en Pim (2010). Zij wonen in Barendrecht.

## PhD PORTFOLIO

| | | Workload (ECTS) |
|---|---|---|
| *General courses* | | |
| - BROK ('Basiscursus Regelgeving en Organisatie van Klinisch onderzoek') | 2011 | 1.0 |
| - Biomedical English Writing and Communication | 2009 | 4.0 |
| *Specific courses (NIHES Research School)* | | |
| - Regression Analysis for Clinicians | 2012 | 1.9 |
| - Basic principles of Epidemiology | 2009 | 0.7 |
| - Introduction to Data-analysis | 2009 | 1.0 |
| *Workshops and presentations* | | |
| - Workshop GOUD-symposium | 2014 | 1.0 |
| - NVAVG studiedag '(onder)zoekt en je zult vinden' (workshop) | 2014 | 1.0 |
| - Congress 'Bewegen in de langdurige zorg' (oral presentation) | 2013 | 1.0 |
| - Congress 'Focus op onderzoek' (oral presentation) | 2011 | 1.0 |
| - Landelijke dag DVG (oral presentation) | 2011 | 1.0 |
| - Studiedag nvFVG (oral presentation) | 2011 | 1.0 |
| - NVAVG studiedag 'De oudere VG: een kwetsbare balans' (oral presentation) | 2011 | 1.0 |
| *(Inter)national conferences* | | |
| - 8th European Congress of EUGMS, Brussel (poster presentation) | 2012 | 1.0 |
| - 14th World Congress of IASSID, Halifax (2 oral presentations) | 2012 | 3.0 |
| - 7th European Congress of EUGMS, Malaga (poster presentation) | 2011 | 1.0 |
| - 3rd European Congress of IASSID, Rome (2 oral presentations) | 2010 | 3.0 |
| - Roundtable SIRGAID IASSID, Prato (oral presentation) | 2010 | 2.0 |
| - Roundtable SIRGAID IASSID, Edinburgh (poster presentation) | 2009 | 1.0 |
| *Lecturing* | | |
| - Intellectual Disability Medicine, ErasmusMC (3 lectures) | 2009 – 2012 | 1.8 |
| *Supervising* | | |
| - 5 Master Medical Science student research projects | 2008 – 2014 | 7.3 |
| - 1 Master Dietetics student research project | 2009 | 1.5 |

# Publications

## PUBLICATIONS

Bastiaanse LP, Mergler S, Evenhuis HM, Echteld MA (2014). Bone quality in older adults with intellectual disabilities. *Research in Developmental Disabilities, 35 (9): 1927-1933.*

De Winter CF, Bastiaanse LP, Kranendonk SE, Hilgenkamp TIM, Evenhuis HM, Echteld MA (2013). Peripheral arterial disease in older people with intellectual disability in The Netherlands using the ankle-brachial index: results of the HA-ID study. *Research in Developmental Disabilities, 34 (5): 1663-1668.*

Bastiaanse LP, Vlasveld G, Penning C, Evenhuis HM (2012). Feasibility and reliability of the Mini Nutritional Assessment (MNA) in older adults with intellectual disabilities. *Journal of Nutrition, Health and Aging, 16 (9): 759-762.*

Evenhuis HM, Hermans H, Hilgenkamp TIM, Bastiaanse LP, Echteld MA (2012). Kwetsbaarheid bij ouderen met een verstandelijke handicap. *Nederlands Tijdschrift voor Geneeskunde, 156: A4808.*

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Evenhuis HM, Hermans H, Hilgenkamp TIM, Bastiaanse LP, Echteld MA (2012). Frailty and disability in older adults with intellectual disabilities: results from the healthy ageing and intellectual disability study. *Journal of the American Geriatrics Society, 60 (5): 934-938.*

De Winter CF, Bastiaanse LP, Hilgenkamp TIM, Evenhuis HM, Echteld MA (2012). Overweight and obesity in older people with intellectual disability. *Research in Developmental Disabilities, 33 (2): 398-405.*

Hilgenkamp TIM, Bastiaanse LP, Hermans H, Penning C, van Wijck R, Evenhuis HM (2011). Study healthy ageing and intellectual disabilities: recruitment and design. *Research in Developmental Disabilities, 32 (3): 1097-1106.*

Evenhuis HM, Bastiaanse LP, Kuijpers B, Louisse AC, Theunissen M, Mergler S (2009). Gebruik van inhalatiecorticosteroïden door volwassen patiënten in zorg van de AVG. *Tijdschrift voor Artsen voor Verstandelijk Gehandicapten, 27 (1): 12-13.*

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