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# Is fatness or fitness key for survival in older adults with intellectual disabilities?

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**Abstract**

**Background:** Overweight/obesity and poor physical fitness are two prevalent lifestyle-related problems in older adults with intellectual disabilities, which each require a different approach. To improve healthy ageing, we assessed whether fatness or fitness is more important for survival in older adults with intellectual disabilities.

**Methods:** In the HA-ID study, we measured obesity and fitness of 874 older adults with intellectual disabilities ( $61.4 \pm 7.8$  years). All-cause mortality was assessed over a 5-year follow-up period.

**Results:** Fitness, but not obesity, was significantly related to survival (HR range of 0.17–0.22). People who were unfit were 3.58 (95% CI = 1.72–7.46) to 4.59 (95% CI = 1.97–10.68) times more likely to die within the follow-up period than people who were fit, regardless of obesity.

**Conclusion:** This was the first study to show that being fit is more important for survival than fatness in older adults with intellectual disabilities. The emphasis should, therefore, shift from weight reduction to improving physical fitness.

**KEYWORDS**

ageing, developmental disabilities, mortality, physical fitness, weight

## 1 | INTRODUCTION

Over the past decades the obesity epidemic has been a major health risk worldwide. From 1975 to 2016, the prevalence of obesity nearly tripled, with 39% of the adult population of the world being overweight and 13% being obese, as estimated by the World Health Organization (WHO) (World Health Organization, 2018). Being overweight and obese are important risk factors for cardiovascular diseases, diabetes, musculoskeletal disorders, cancers and mortality (Flegal, Kit, Orpana, & Graubard, 2013; Guh et al., 2009; Pischon et al., 2008; World Health Organization, 2018).

Being overweight and obese is also highly prevalent in older adults with intellectual disabilities (38.2% overweight, 25.6% obese based on body mass index [BMI]). These prevalence rates are even higher than the already high prevalence rates in the

general older population (41.2% overweight, 9.6% obese) (de Winter, Bastiaanse, Hilgenkamp, Evenhuis, & Ehteld, 2012). In the general population, the relationship between obesity (often referred to as fatness within the literature about this topic) and survival has been consistently demonstrated (Flegal et al., 2013; Pischon et al., 2008). However, due to a lack of longitudinal population studies this has not yet been studied in older adults with intellectual disabilities. We, therefore, do not know how important obesity is for survival in this population. Insight in this relationship can inform the decision making whether or not we should focus our efforts on reducing overweight and obesity to support healthy ageing and survival.

Even though the independent effects of obesity on survival are established in the general population, studies also show that physical fitness and activity may be even more important determinants

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for survival (Barry et al., 2014; Gaesser, Tucker, Jarrett, & Angadi, 2015; Yerrakalva, Mullis, & Mant, 2015). This is also referred to as the “fat but fit” theory. Fitness (mostly assessed as cardiorespiratory fitness) may act as a confounder or effect modifier on the relationship between obesity and survival. A meta-analysis showed that individuals who were fit and overweight or obese had similar mortality risks as those who were fit with normal weight (Barry et al., 2014). However, individuals who were unfit had twice the mortality risk, regardless of their BMI (Barry et al., 2014). This might also be an explanation for the “obesity paradox,” which represents the findings that being overweight and obese (BMI between 25 and 35) is actually beneficial for survival in older adults in the general population (Flegal et al., 2013; Yerrakalva et al., 2015). Therefore, being overweight or obese does not seem to automatically increase one's mortality risk, while a higher level of fitness seems to substantially lower the negative effects of overweight/obesity on survival. One possible mechanism is that being fit results in favourable cardiovascular and metabolic health, thereby maintaining a healthy cardiometabolic profile while being obese, also referred to as “healthy obesity” (Lavie, De Schutter, & Milani, 2015; Phillips et al., 2013; Wildman, 2009).

The negative consequences of obesity may thus be reduced by sufficient physical fitness and activity levels. Unfortunately, older adults with intellectual disabilities also have very low physical fitness and activity levels (Hilgenkamp, Reis, van Wijck, & Evenhuis, 2012a, 2012b; Oppewal, Hilgenkamp, van Wijck, & Evenhuis, 2013). We previously found that these low fitness levels negatively affect survival in older adults with intellectual disabilities. The physical fitness components manual dexterity, visual reaction time, balance, comfortable and fast gait speed, grip strength and cardiorespiratory fitness were independently predictive for survival (Oppewal & Hilgenkamp, 2019). However, we do not know whether obesity negatively affects survival, and whether fitness or obesity is more important with regard to survival. Results from the general population cannot be generalized to people with intellectual disabilities, because of the comorbidities often present in people with intellectual disabilities and the lifelong poor physical activity and fitness levels of people with intellectual disabilities in comparison to the general population. Only a few studies have investigated cross-sectional associations between fitness and obesity, focusing on adolescents with intellectual disabilities. These studies showed that obesity measures were associated with physical fitness in adolescents with intellectual disabilities (Foley, Harvey, Chun, & Kim, 2008; Salaun & Berthouze-Aranda, 2012), but not in adolescents with Down syndrome (Izquierdo-Gomez, Martinez-Gomez, Fernhall, Sanz, & Veiga, 2016). Due to the limited number of longitudinal studies in this population and none including older adults with intellectual disabilities, no study so far has been able to investigate the relationship with survival in people with intellectual disabilities. If the “fat but fit” theory also holds for older adults with intellectual disabilities, focusing on improving physical fitness may be a better strategy to improve healthy ageing and survival than focusing on weight reduction.

### What is already known about this subject

- Obesity and low physical fitness levels are highly prevalent in older adults with intellectual disabilities.
- In the general population fatness and fitness are related to survival and fitness seems to be more important for survival.
- Even though it is unknown whether fatness or fitness is more important for survival in people with intellectual disabilities there is a large emphasis on weight reduction.

### What this study adds

- People who were unfit had a four times higher mortality risk than people who were fit regardless of their fatness.
- The current emphasis on weight reduction should be shifted to improving physical fitness in older adults with intellectual disabilities to improve and support healthy aging in this population

This would be important since the literature currently reports about four times more weight reduction studies than interventions to improve physical fitness.

Therefore, our aim is to assess the relationship between fatness and survival and to assess whether fatness or fitness is more important for survival in older adults with intellectual disabilities.

## 2 | METHODS

### 2.1 | Study design and participants

This study was part of the Healthy Ageing and Intellectual Disability (HA-ID) study, a prospective cohort study regarding the health of older adults with intellectual disabilities ( $\geq 50$  years), conducted by three Dutch care organizations and the Chair of Intellectual Disability Medicine at the Erasmus MC, University Medical Center Rotterdam, the Netherlands. In 2008, all 2,322 clients aged 50 years and over receiving care and support from one of the care organizations were invited to participate. Of these, 1,050 clients or legal representatives provided informed consent, resulting in a near-representative sample. More details about the study are described elsewhere (Hilgenkamp et al., 2011). Participants of whom at least one obesity measurement was available were included in this study. No other inclusion criteria were applied. Participants who were underweight ( $\text{BMI} < 18.5 \text{ kg/m}^2$ ,  $n = 19$  [2.1%]) were excluded from this analysis because being underweight exhibit its own opposite risk profile compared to being overweight. Participants with severe or profound intellectual disabilities and wheelchair users had more difficulties in

performing the fitness measurements and were underrepresented in this study compared to the total HA-ID study sample (Hilgenkamp, van Wijck, & Evenhuis, 2013; de Winter et al., 2012). Baseline data were collected between November 2008 and July 2010. Follow-up data on all-cause mortality were collected up to March 2015.

The Medical Ethics Committee of the Erasmus MC, University Medical Center Rotterdam approved this study (MEC 2008–234 and MEC 2011–309). This study was conducted according to the guidelines of the Declaration of Helsinki (Helsinki, 2013).

## 2.2 | Measurements

### 2.2.1 | Personal characteristics

Characteristics that may act as covariates with regard to obesity, fitness and survival (Yerrakalva et al., 2015) were collected at baseline: age and sex from electronic administrative systems, level of intellectual disabilities from the psychologists' and behavioural therapists' files (borderline = IQ of 70–80, mild = IQ of 55–70, moderate = IQ of 35–55, severe = IQ of 25–35 and profound = IQ <25) and smoking (yes/no) and alcohol consumption (no, 1–2 or  $\geq 3$  glasses per day) from the professional caregivers. From the medical files, we collected the presence of Down syndrome, cardiometabolic diseases and cardiovascular medication use. Cardiometabolic diseases were considered present if the participant had one of the following diagnoses: heart failure, valve abnormalities, coronary heart disease, heart rate disorder, hypertension, hypercholesterolemia, intermittent claudication, stroke and diabetes mellitus type 2. Cardiovascular medication use was classified as using one or multiple medications with the first level code C, according to the Anatomical Therapeutic Chemical (ATC) classification system (WHO Collaborating Centre for Drugs Statistics Methodology).

### 2.2.2 | Obesity

Obesity was measured with BMI, waist circumference (WC), waist-to-hip ratio (WHR) and body fat percentage (fat%).

BMI was classified as normal (18.5–24.99 kg/m<sup>2</sup>), overweight (25–29.99 kg/m<sup>2</sup>) and obese ( $\geq 30$  kg/m<sup>2</sup>) (American College of Sports Medicine, 2018; WHO, 1995).

WC was measured in standing position with the arms resting at the sides, over the unclothed abdomen at the narrowest point between the costal margin and iliac crest (American College of Sports Medicine, 2018). WC was classified as normal (<94 cm for males, <80 cm for females), overweight ( $\geq 94$  cm for males,  $\geq 80$  cm for females) and obese ( $\geq 102$  cm for males,  $\geq 88$  cm for females) (WHO, 1995).

To calculate WHR, hip circumference was additionally measured over light clothing at the level of the widest diameter around the buttocks. WHR was calculated by dividing WC through hip circumference and classified as normal (<0.90 for males, <0.80 for females), overweight ( $\geq 0.90$  for males,  $\geq 0.80$  for females) and obese ( $\geq 1.00$  for males,  $\geq 0.88$  for females) (WHO, 1995).

Fat% was based on the sum of the triceps, biceps, subscapular and suprailiacal skinfolds. Body density was calculated from the sum of the skinfolds (Visser equation (Visser, van den Heuvel, & Deurenberg, 1994)) and then, fat% was calculated from body density (Siri's equation (Siri, 1961)). We did not divide fat% in categories, because there is no consensus with regard to cut-off values (American College of Sports Medicine, 2018).

### 2.2.3 | Physical fitness

As an indicator of fitness, we used comfortable gait speed (CGS) over a 5-meter distance (Bohannon, 1997). Previous research demonstrated that CGS is especially relevant in older populations, as slow gait speed is an important risk factor for health outcomes such as disability, falls and mortality (Abellan van Kan et al., 2009; Oppewal, Hilgenkamp, van Wijck, Schoufour, & Evenhuis, 2014, 2015; Oppewal & Hilgenkamp, 2019). Participants walked an 11-meter walkway, including 3 meters for acceleration, 5 meters of timed comfortable walking and 3 meters for deceleration. The average of three walks was the result (m/s). Participants walked without someone walking alongside or physically supporting them to avoid influencing the speed. Walking aids were allowed. Validity and reliability of this test in the general population was good (Abellan van Kan et al., 2009; Connelly, Stevenson, & Vandervoort, 1996; Cooper, Kuh, & Hardy, 2010; Steffen, Hacker, & Mollinger, 2002; Steffen & Seney, 2008), as well as test-retest reliability in older adults with intellectual disabilities (Hilgenkamp, Reis, et al., 2012; Hilgenkamp Wijck, & Evenhuis, 2012a). A cut-off of 1.0 m/s was used to divide participants into fit or unfit. This speed is often used to predict survival, with older adults who walk faster than 1.0 m/s generally having a better survival (Abellan van Kan et al., 2009).

### 2.2.4 | All-cause mortality

Data on all-cause mortality were collected over a 5-year ( $4.7 \pm 1.7$  years, 0–6.3 years) follow-up period. The administrative services identified deceased participants and the time of death and checked whether all remaining participants were still registered at the care organizations. If not, they provided us with the date of deregistration.

## 2.3 | Statistical analyses

Descriptive statistics were calculated for the total study sample and the following subgroups: survived, deceased and deregistered. Differences in personal characteristics between participants who deceased and those who were either deregistered or survived were analysed with independent *t* tests (continuous variables) and Chi-squared tests (categorical variables). Differences in obesity and fitness between those who survived and those who deceased were also analysed with independent *t* tests and Chi-squared tests.

**TABLE 1** Baseline personal characteristics for the total study sample and the subgroups survived, deceased and deregistered

	Total, n = 874 (100%)	Survived, n = 680 (77.8%)	Deceased, n = 147 (16.8%)	Deregistered, n = 47 (5.4%)
Age, mean (SD) <sup>a</sup> , No. (% of row)	61.4 ± 7.8	60.8 ± 7.4**	64.7 ± 8.9*	59.6 ± 6.0
50–59 year	414 (100%)	346 (83.6%)	45 (10.9%)	23 (5.6%)
60–69 year	311 (100%)	228 (73.3%)	61 (19.6%)	22 (7.1%)
70–79 year	134 (100%)	98 (73.1%)	34 (25.4%)	2 (1.5%)
80+ year	15 (100%)	8 (53.3%)	7 (46.7%)	0
Sex, No. (% of row)				
Female	434 (100%)	339 (78.1%)	67 (15.4%)	28 (6.5%)
Male	440 (100%)	341 (77.5%)	80 (18.2%)	19 (4.3%)
Level of intellectual disabilities, No. (% of row)				
Borderline	30 (100%)	26 (86.7%)	2 (6.7%)	2 (6.7%)
Mild	190 (100%)	153 (80.5%)	27 (14.2%)	10 (5.3%)
Moderate	428 (100%)	326 (76.2%)	75 (17.5%)	27 (6.3%)
Severe	139 (100%)	111 (79.9%)	23 (16.5%)	5 (3.6%)
Profound	65 (100%)	49 (75.4%)	15 (23.1%)	1 (1.5%)
Unknown	22 (100%)	15 (68.2%)	5 (22.7%)	2 (9.1%)
Down syndrome, No. (% of row)				
No	602 (100%)	491 (81.6%)**	88 (14.6%)	23 (3.8%)
Yes	122 (100%)	75 (61.5%)	36 (29.5%)	11 (9.0%)
Unknown	150 (100%)	114 (76.0%)	23 (15.3%)	13 (8.7%)
Smoking, No. (% of row)				
No	657 (100%)	517 (78.7%)	106 (16.1%)	34 (5.2%)
Yes	172 (100%)	132 (76.7%)	36 (20.9%)	4 (2.3%)
Alcohol use, No. (% of row)				
No	695 (100%)	550 (79.1%)	114 (16.4%)	31 (4.5%)
1–2 glasses per day	123 (100%)	90 (73.2%)	26 (21.1%)	7 (5.7%)
≥3 glasses per day	11 (100%)	9 (81.8%)	2 (18.2%)	0
Cardiometabolic diseases, No. (% of row)				
No	456 (100%)	373 (81.8%)**	61 (13.4%)	22 (4.8%)
Yes	307 (100%)	221 (72.0%)	73 (23.8%)	13 (4.2%)
Heart failure	31 (100%)	11 (35.5%)	20 (64.5%)	0
Valve abnormalities	54 (100%)	30 (55.6%)	21 (38.9%)	3 (5.6%)
Coronary heart disease	21 (100%)	10 (47.6%)	10 (47.6%)	1 (4.8%)
Heart rate disorder	22 (100%)	13 (59.1%)	7 (31.8%)	2 (9.1%)
Hypertension	170 (100%)	132 (77.6%)	31 (18.2%)	7 (4.1%)
Hypercholesterolemia	84 (100%)	71 (84.5%)	11 (13.1%)	2 (2.4%)
Intermittent claudication	16 (100%)	11 (68.8%)	5 (31.3%)	0
Stroke	42 (100%)	29 (32.7%)	12 (28.6%)	1 (2.4%)
Diabetes mellitus type 2	60 (100%)	45 (75.0%)	14 (23.3%)	1 (1.7%)
Cardiometabolic medications, No. (% of row)				
No	616 (100%)	482 (78.2%)	97 (15.7%)	37 (6.0%)
Yes	230 (100%)	175 (76.1%)	47 (20.4%)	8 (3.5%)

Abbreviations: n, number of participants; SD, standard deviation.

<sup>a</sup>Age at time of inclusion in study.\*Indicating a significant difference between deregistered and deceased participants,  $p < .001$ .\*\*Indicating a significant difference between survived and deceased participants,  $p < .001$ .

**TABLE 2** Baseline obesity and fitness measures for the total study sample and the subgroups survived and deceased

	Total	Survived	Deceased
<b>Obesity measures</b>			
<b>BMI</b>	<i>n</i> = 874	<i>n</i> = 680	<i>n</i> = 147
Mean (SD)	27.5 ± 5.1	27.3 ± 4.8	27.5 ± 5.7
No. (% of row)			
Normal weight	304 (100%)	237 (78.0%)	52 (17.1%)
Overweight	341 (100%)	274 (80.4%)	52 (15.2%)
Obese	229 (100%)	169 (73.8%)	43 (18.8%)
<b>Waist circumference</b>	<i>n</i> = 850	<i>n</i> = 669	<i>n</i> = 135
Mean (SD)	94.6 ± 12.9	94.3 ± 12.6	95.7 ± 14.6
No. (% of row)			
Normal	266 (100%)	212 (79.7%)	40 (15.0%)
Overweight	184 (100%)	146 (79.3%)	32 (17.4%)
Obese	400 (100%)	311 (77.8%)	63 (15.8%)
<b>Waist to hip ratio</b>	<i>n</i> = 805	<i>n</i> = 646	<i>n</i> = 113
Mean (SD)	0.92 ± 0.09	0.93 ± 0.1	0.92 ± 0.08
No. (% of row)			
Normal	116 (100%)	86 (74.1%)	23 (19.8%)
Overweight	298 (100%)	241 (80.9%)	40 (13.4%)
Obese	391 (100%)	319 (81.6%)	50 (12.8%)
<b>Body fat %</b>	<i>n</i> = 648	<i>n</i> = 523	<i>n</i> = 88
Mean (SD)	37.8 ± 7.1	37.8 ± 7.0	36.6 ± 7.5
<b>Fitness measure</b>			
<b>Comfortable gait speed</b>	<i>n</i> = 678	<i>n</i> = 555	<i>n</i> = 84
Mean (SD)	0.98 ± 0.34	1.00 ± 0.34*	0.83 ± 0.31
No. (% of row)			
Fit	324 (100%)	284 (87.7%)*	22 (6.8%)
Unfit	354 (100%)	271 (76.6%)	62 (17.5%)

Abbreviations: BMI, Body mass index divided in normal (18.5–24.99 kg/m<sup>2</sup>), overweight (25–29.99 kg/m<sup>2</sup>) and obese (≥30 kg/m<sup>2</sup>); waist circumference divided in normal (<94 cm for males, <80 cm for females), overweight (≥94 cm for males, ≥80 cm for females) and obese (≥102 cm for males, ≥88 cm for females); waist to hip ratio divided in normal (<0.90 for males, <0.80 for females), overweight (≥0.90 for males, ≥0.80 for females) and obese (≥1.00 for males, ≥0.88 for females); *n*, number of participants; SD, standard deviation.

\*Indicating a significant difference between survived and deceased participants, *p* < .001.

The relationship between obesity and survival was analysed with survival analyses, with Log-rank tests and Cox proportional hazard models. Participants lost to follow-up were censored (observation ended before death occurred) on the date of deregistration or at the end of the study, whichever came first. The scaled Schoenfeld residuals and plotting  $\beta(t)$  for the variables against time were used to assess the proportional hazard assumption. To evaluate the risk of informative censoring, the characteristics of those lost to follow-up have been analysed previously (Schoufour, Mitnitski, Rockwood, Evenhuis, & Ehteld, 2015). The assumptions of proportional hazards and non-informative censoring were sufficiently met.

Cox proportional hazard models were used to assess the predictive value of each obesity measure (both as a continuous and

categorical variable) for survival (model 1). Next, Cox proportional hazard models adjusted for age, sex, level of intellectual disabilities, Down syndrome, smoking behaviour, alcohol consumption, presence of cardiometabolic diseases and cardiovascular medication use were calculated (model 2). Finally, Cox proportional hazard models adjusted for the covariates in model 2 plus CGS were calculated (model 3).

Kaplan–Meier curves were used to compare mortality rates stratified by obesity and fitness, with four groups: (a) CGS ≥ 1.0 m/s and BMI 18.5–29.99 kg/m<sup>2</sup> (fit and no obesity), (b) CGS ≥ 1.0 m/s and BMI ≥ 30 kg/m<sup>2</sup> (fit and obesity), (c) CGS < 1.0 m/s and BMI 18.5–29.99 kg/m<sup>2</sup> (unfit and no obesity), (d) CGS < 1.0 m/s and BMI ≥ 30 kg/m<sup>2</sup> (unfit and obesity). The Log-rank test was used to assess whether there was a difference in survival between groups. A Cox proportional hazard model was used to estimate the hazard ratios (HR) adjusted for the covariates.

### 3 | RESULTS

#### 3.1 | Baseline characteristics

Of the 874 participants of whom at least one obesity measure was available, 147 (16.8%) died and 47 (5.4%) were deregistered (Table 1). Deregistered participants were significantly younger (*t* = 4.5, *p* < .001) than those who died. Participants who survived were significantly younger (*t* = 5.0, *p* < .001) and had less often Down syndrome ( $\chi^2$  = 19.0, *p* < .001) and cardiometabolic diseases ( $\chi^2$  = 13.5, *p* < .001) than those who died.

Compared to participants who died, participants who survived walked significantly faster (*t* = −4.4, *p* < .001) and were more often categorized as fit (51.2% versus 26.1%;  $\chi^2$  = 18.2, *p* < .001). No differences were seen in obesity (Table 2).

#### 3.2 | 5-year survival

In the simple Cox models, only being obese based on WHR was significantly related to survival (*HR* = 0.52, 95% *CI* = 0.28–0.97; Table 3, model 1), however, not after adjustment for covariates (Table 3, model 2). None of the other obesity measures were related to survival.

After adding fitness to the model (Table 3, model 3), fitness was significantly related to survival. One 0.1 m/s (0.22 mph) increase in CGS (m/s) resulted in a 7.8%–8.3% lower mortality risk (*HR* ranged 0.17–0.22 across models).

Looking at the covariates, older age (*HR* range of 1.04–1.05) and smoking (*HR* range of 2.33–2.69) consistently showed a higher mortality risk. Having Down syndrome (*HR* range of 2.44–2.59) and cardiometabolic diseases (*HR* range of 1.93–1.98) resulted in higher mortality risk, although not consistent across models. Down syndrome was not a significant covariate in the model with fat% as the variable of interest and having cardiometabolic diseases was not a significant covariate in the models with WHR and fat% as the variables of interests.

**TABLE 3** Results of the Cox proportional hazard models for each obesity measure, with fitness (CGS) as a covariate

Outcome measure	Model 1, B (SE)	HR (95% CI)	Model 2, B (SE)	HR (95% CI)	Model 3, B (SE)	HR (95% CI)
BMI continuous (n = 541)	-0.01 (0.03)	0.99 (0.94–1.04)	-0.003 (0.03)	1.00 (0.94–1.05)	-0.01 (0.03)	0.99 (0.94–1.05)
+Comfortable gait speed	-	-	-	-	-1.58 (0.43)**	0.21 (0.09–0.48)
BMI categorical (n = 541)						
Normal		1 (reference)		1 (reference)		1 (reference)
Overweight	-0.40 (0.29)	0.67 (0.38–1.19)	-0.44 (0.31)	0.64 (0.35–1.17)	-0.39 (0.31)	0.68 (0.37–1.23)
Obese	-0.07 (0.29)	0.93 (0.53–1.64)	0.03 (0.33)	1.03 (0.55–1.96)	-0.02 (0.33)	0.98 (0.52–1.86)
+Comfortable gait speed	-	-	-	-	-1.53 (0.43)**	0.22 (0.09–0.51)
Waist circumference continuous (n = 532)	-0.002 (0.01)	1.00 (0.98–1.02)	-0.001 (0.01)	1.00 (0.98–1.02)	-0.001 (0.01)	1.00 (0.98–1.02)
+Comfortable gait speed	-	-	-	-	-1.69 (0.44)**	0.19 (0.08–0.44)
Waist circumference categorical (n = 532)						
Normal		1 (reference)		1 (reference)		1 (reference)
Overweight	-0.10 (0.34)	0.90 (0.47–1.74)	-0.19 (0.36)	0.83 (0.41–1.66)	-0.25 (0.36)	0.78 (0.39–1.56)
Obese	-0.28 (0.28)	0.76 (0.44–1.31)	-0.20 (0.33)	0.82 (0.43–1.56)	-0.24 (0.34)	0.79 (0.41–1.53)
+Comfortable gait speed	-	-	-	-	-1.71 (0.40)**	0.18 (0.08–0.43)
Waist to hip ratio continuous (n = 531)	-0.29 (1.30)	0.75 (0.06–9.43)	-1.18 (1.66)	0.31 (0.01–7.86)	-0.91 (1.71)	0.40 (0.01–11.41)
+Comfortable gait speed	-	-	-	-	-1.63 (0.44)**	0.20 (0.08–0.47)
Waist to hip ratio categorical (n = 531)						
Normal		1 (reference)		1 (reference)		1 (reference)
Overweight	-0.63 (0.34)	0.53 (0.28–1.03)	-0.54 (0.35)	0.58 (0.29–1.15)	-0.45 (0.35)	0.64 (0.32–1.27)
Obese	-0.66 (0.32)*	0.52 (0.28–0.97)	-0.56 (0.34)	0.58 (0.29–1.11)	-0.52 (0.35)	0.60 (0.30–1.18)
+Comfortable gait speed	-	-	-	-	-1.60 (0.45)**	0.20 (0.08–0.48)
Body fat % continuous (n = 435)	-0.03 (0.02)	0.97 (0.93–1.01)	-0.10 (0.05)	0.91 (0.82–1.00)	-0.09 (0.05)	0.91 (0.83–1.01)
+Comfortable gait speed	-	-	-	-	-1.80 (0.52)**	0.17 (0.06–0.46)

Note: Model 1: Simple Cox proportional hazard model for each obesity measure, with fitness (CGS) as a covariate.

Model 2: Multiple Cox proportional hazard model for each obesity measure, adjusted for age, sex, level of intellectual disability, presence of Down syndrome, smoking, alcohol use, presence of cardiometabolic diseases, use of cardiovascular medication.

Model 3: Multiple Cox proportional hazard model for each obesity measure, adjusted for covariate listed in model 2 plus comfortable gait speed (m/s).

Abbreviations: B, Beta coefficient; BMI, Body mass index divided in normal (18.5–24.99 kg/m<sup>2</sup>), overweight (25–29.99 kg/m<sup>2</sup>) and obese (≥30 kg/m<sup>2</sup>); waist circumference divided in normal (<94 cm for males, <80 cm for females), overweight (≥94 cm for males, ≥80 cm for females) and obese (≥102 cm for males, ≥88 cm for females); waist to hip ratio divided in normal (<0.90 for males, <0.80 for females), overweight (≥0.90 for males, ≥0.80 for females) and obese (≥1.00 for males, ≥0.88 for females); CI, Confidence interval; HR, Hazard ratio; n, number of participants; SE, Standard error.

\**p* < .05.

\*\**p* < .001.

### 3.3 | Analyses stratified by obesity and fitness

Table 4 shows the distribution across the groups stratified by obesity and fitness, with the smallest groups being “fit and obesity” (12.5%), followed by “unfit and obesity” (15.2%). Figure 1 shows

the survival curves, with higher survival rates in the fit than in unfit participants, regardless of obesity ( $\chi^2 = 18.82$ , *p* < .001). People who were unfit, regardless of being obese or not, were 3.6 to 4.6 times more likely to die within the 5-year follow-up period than the people who were fit.



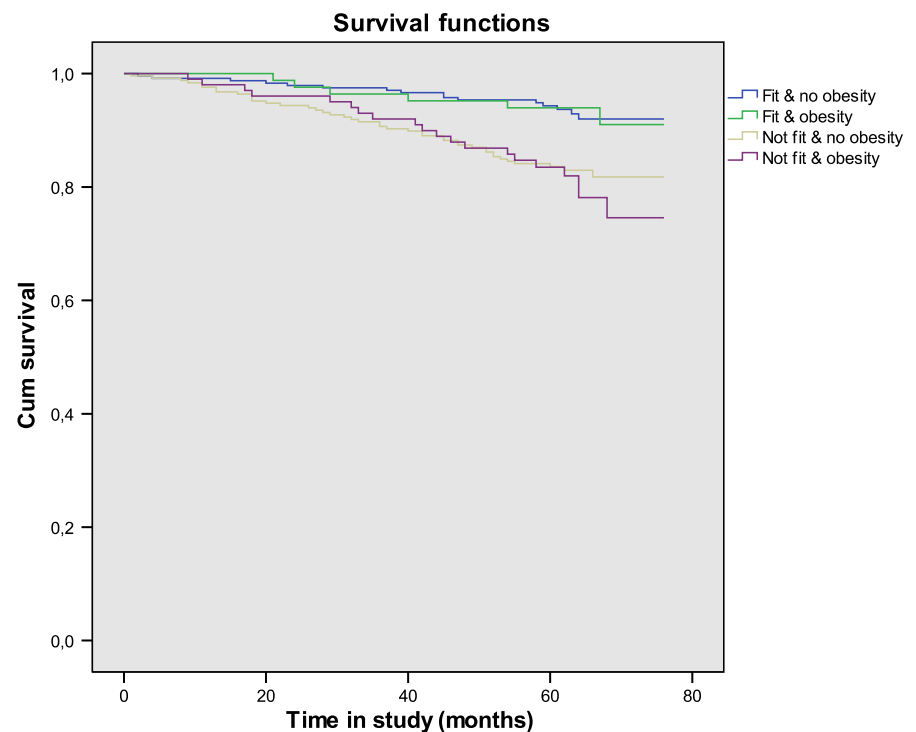
**TABLE 4** Distribution of participants across obesity and fitness groups and results of the Cox proportional hazard model

Obesity and fitness groups	n (%)	B (SE)	HR (95% CI)
Fit & no obesity	239 (35.3%)		1 (reference)
Fit & obesity	85 (12.5%)	0.20 (0.62)	1.22 (0.36–4.07)
Unfit & no obesity	251 (37.0%)	1.28 (0.37) <sup>*</sup>	3.58 (1.72–7.46)
Unfit & obesity	103 (15.2%)	1.52 (0.43) <sup>*</sup>	4.59 (1.97–10.68)

Abbreviations: B, Beta coefficient; CI, Confidence interval; HR, Hazard ratio; n, number of participants; SE, Standard error.

<sup>\*</sup> $p < .001$ .

**FIGURE 1** Kaplan–Meier survival curves according to obesity (BMI 18.5–29.99 kg/m<sup>2</sup> and BMI  $\geq$  30 kg/m<sup>2</sup>) and fitness (CGS < 1.0 m/s and CGS > 1.0 m/s). BMI, Body mass index; CGS, comfortable gait speed



## 4 | DISCUSSION

This is the first study to examine the relationship between fatness and survival and whether fatness or fitness is more important for survival of older adults with intellectual disabilities. Being obese based on WHR was the only measure of obesity that was significantly related to survival, however, this relationship disappeared after adjusting for covariates. We saw that older adults who were unfit were 3.6 to 4.6 times more likely to die within the 5-year follow-up period than older adults with intellectual disabilities who were fit, regardless of being obese or not. Fitness, therefore, seems to be more important for survival than any of the obesity measures and focusing on improving fitness may be a better strategy for healthy ageing and increasing survival than focusing on weight reduction.

In the general population it was also found that physical fitness is a more powerful predictor for survival than fatness. However, we found that older adults with intellectual disabilities who were unfit had four times ( $HR$  of 3.58 [95% CI = 1.72–7.46] to 4.59 [95% CI = 1.97–10.68]) the mortality risk than those who were fit,

regardless of being obese or not. This seems to be higher than the twofold risk ( $HR$  range of 2.14–2.46) seen in the general adult population (Barry et al., 2014). This confounding role of fitness is often raised as one of the potential contributors to the “obesity paradox,” referring to that being overweight and obese (BMI between 25 and 35) is actually beneficial for survival (Flegal et al., 2013; Yerrakalva et al., 2015). In fit people, excess adipose tissue may have a role in protective cardiovascular and metabolic mechanisms by having larger coronary arteries and reduced systemic inflammation (Yerrakalva et al., 2015). In addition, physical activity and exercise have positive effects on cardiovascular and metabolic health, such as more favourable glucose and insulin metabolism, reduced blood pressure, more favourable cholesterol levels and increased anti-inflammatory markers, independent of (changes in) overweight/obesity status (Gaesser, Angadi, & Sawyer, 2011; Gaesser et al., 2015). Exercise can also reduce visceral adipose tissue, ectopic fat and hepatic fat without weight loss (Gaesser et al., 2015). A methodological issue often raised in the discussion about the obesity paradox is the use of BMI as an obesity measure, because BMI is

affected by general weight gain/loss and is not sensitive for reductions in for example visceral adipose tissue. Also, a high BMI in fit people may reflect a high amount of lean tissue mass instead of fat mass. However, similar “fat but fit” results have been found with other measures such as WC, WHR and fat% (Gaesser et al., 2015; Yerrakalva et al., 2015).

Based on our results, we question whether weight reduction is the most beneficial strategy to try to improve health and survival of the ageing population with intellectual disabilities. As has been suggested for the general population, focusing on improving physical fitness, rather than reducing body weight may be a more beneficial strategy (Gaesser et al., 2015; Lavie et al., 2015). Weight reduction is very hard to accomplish for any individual, as it requires a lifestyle change and not a single intervention (Bray, Fruhbeck, Ryan, & Wilding, 2016). A recent review showed that successful interventions to reduce weight are scarce in adults with intellectual disabilities (Harris, Melville, Murray, & Hankey, 2018). In a meta-analysis, current weight loss interventions did not show a clinical meaningful effect and were not superior to no treatment (Harris et al., 2018). Changing the focus from weight loss to improving physical fitness and activity may, therefore, be a more successful strategy. Physical exercise interventions have been successful in improving physical fitness in people with intellectual disabilities of all ages (Bartlo & Klein, 2011; Heller, McCubbin, Drum, & Peterson, 2011; van Schijndel-Speet, Evenhuis, van Wijck, van Montfort, & Echteld, 2017; Shin & Park, 2012) while also demonstrating positive effects on cardiometabolic factors (van Schijndel-Speet et al., 2017). However, motivating people with intellectual disabilities to become and stay active is also challenging, because they experience health problems and physiological, cognitive organizational and environmental barriers (Rimmer & Marques, 2012; Willems, Hilgenkamp, Havik, Waninge, & Melville, 2017). We, therefore, need to concentrate our efforts towards improving our understanding of the best ways to increase physical activity and fitness in this population and obtaining optimal health benefits. Therefore, we should not only focus on older adults, but also on all ages. Starting with an active lifestyle early in life is important for a healthy life and healthy ageing.

Strengths of this study are the multiple obesity measures used, the long follow-up period and the large study sample. Furthermore, this was the first study to address the fitness-fatness question in older adults with intellectual disabilities, thereby providing important new knowledge for this ageing population. However, there are some limitations that need to be taken into account. The HA-ID study had a near-representative study sample, but adults with no or very little registered support were underrepresented (Hilgenkamp et al., 2011). The 47 deregistered participants were significantly younger, which could have been selective and related to time of death, causing a selective bias. People with severe or profound intellectual disabilities were underrepresented in the measurements and wheelchair users were not included in the CGS measures (Hilgenkamp et al., 2013; de Winter et al., 2012).

Finally, we used CGS as a measure for fitness. Physical fitness is a construct comprised of health and skill-related components (American College of Sports Medicine, 2018), of which cardiorespiratory fitness, gait speed and muscular strength and endurance are most often studied with regard to health. In the studies in the general population, cardiorespiratory fitness was most often used as the fitness measure. However, because to date, we have no suitable field test to measure cardiorespiratory fitness in older adults with intellectual disabilities we used gait speed instead, in line with previous research in the general older population (Woo, Yu, & Yau, 2013). For future studies, it would be interesting to see what the confounding role of the other physical fitness components is with regard to fatness and survival.

The current study focused on survival, expressed as being alive after our 5-year follow-up period. In addition to survival, the quality of life during those years is just as or even more important. Looking at other health outcomes in addition to survival will provide more insight in the importance of fatness and fitness for specific health aspects in older adults with intellectual disabilities. Including other comorbid conditions associated with obesity, such as respiratory disorders, musculoskeletal conditions and cardiometabolic diseases, will also provide relevant additional information about the impact on health and quality of life.

In this study, we examined obesity and fitness at a single time point. As a next step, we aim to assess how changes in obesity and fitness over time relate to survival. In the general population, improved fitness reduces mortality risk independent of changes in fatness and changes in fitness have been shown to be a better predictor for survival than changes in fatness (Gaesser et al., 2015; Lavie et al., 2015). Results of the current study also need to be replicated in other populations of people with intellectual disabilities to establish a firm body of evidence on which lifestyle and health care decisions can be made.

In conclusion, fitness was a more powerful predictor for survival than fatness in older adults with intellectual disabilities. Fitness, therefore, seems to be more important for a longer life than fatness among older adults with intellectual disabilities and maybe even more so than in the general population. The large emphasis put on reducing weight of people with intellectual disabilities is, therefore, not supported by our results and shifting the emphasis to improving physical fitness in older adults with intellectual disabilities is necessary to improve and support healthy ageing in this population.

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## CONFLICT OF INTEREST

The authors declared no conflicts of interest.

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